

**WOMAN-CENTERED CERVICAL SCREENING: IDENTIFYING
WOMEN'S PREFERENCES AND FACTORS RELATED TO THEIR
PREFERENCES IN CERVICAL CANCER SCREENING**

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Abstract

Objectives

This dissertation had two overarching objectives:

1. To determine how stakeholders perceive women's preferences for cervical screening modalities.
2. To understand methods to measure women's cervical screening preferences, to inform the development and testing of a person-centered, evidence-informed approach to preference-elicitation.

Methods

The overarching conceptual framework was the Ottawa Decision Support Framework. The first objective was addressed by interview studies with (1) guideline developers and program managers and (2) health professionals and women considering screening. This was complemented by a systematic review of quantitative, qualitative and mixed-methods studies of women's cervical screening preferences, using the Grading of Recommendations, Assessment, Development, and Evaluation approach to developing preference-based recommendations. This approach was also used in a systematic review of methods to elicit women's preferences, addressing the second objective. These findings led to the development and field testing of a preference-elicitation tool using International Patient Decision Aid Standards criteria, and the development of a protocol for a population-based study of women's preferences.

Results

Objective 1

Experts disagree about whether there is enough evidence to include alternative modalities in cervical screening programs. Women and health care professionals do not recognize that women face a choice to participate in cervical screening. A narrative synthesis of relevant literature presented challenges in aggregating preferences across diverse study objectives, designs, and contexts.

Objective 2

Preference-elicitation approaches for cervical screening are heterogenous in design and rigour. I therefore developed and field tested a tool to elicit women's preferences, which demonstrated that women found the tool helpful to identify their preferences. I then propose a study that uses multiple methods to apply the tool more broadly.

Conclusions

Synthesized preferences data might not be the optimal approach to incorporate preferences into cervical screening guidelines. A tool grounded in shared decision-making can help women identify their informed, values-based screening preferences.

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List of Abbreviations

CHC	Community Health Centre
CIN2+	cervical intraepithelial neoplasia 2 or more severe disease
CCO	Cancer Care Ontario
DNA	deoxyribonucleic acid
GRADE	Grading of Recommendations, Assessment, Development and Evaluation
HCP	health care professional
HIV	human immunodeficiency virus
HPV	human papillomavirus
IARC	International Agency for Research on Cancer
ICMJE	International Committee of Medical Journal Editors
IPDAS	International Patient Decision Aids Standards
LGBTQ	Lesbian, Gay, Bisexual, Trans-sexual, Queer
LHIN	Local Health Integration Network
ODSF	Ottawa Decision Support Framework
OHIP	Ontario Health Insurance Plan
Pap	Papanicolaou
Pap test	Papanicolaou test
PCR	polymerase chain reaction
SOGC	Society of Obstetricians and Gynaecologists of Canada
TFA	Theoretical Framework of Acceptability

Chapter 1: Introduction

Rationale

More than 500,000 women are newly diagnosed with cervical cancer globally each year, of whom the large majority live in low- and middle- income countries (Ferlay et al., 2013).

Worldwide, cervical cancer is the third most commonly diagnosed cancer among females, with an age-standardized incidence rate of approximately 14.0 per 100,000. In Canada, cervical cancer ranks as thirteenth most common cancer among females, with an age-standardized incidence rate of 8.3 cases per 100,000 (Canadian Cancer Society, 2017). In contrast, high-risk regions, such as Eastern Africa, report rates greater than 30 per 100,000, and cervical cancer is the most common cancer among women¹ (Ferlay et al., 2013). Almost seventy years ago, cervical cancer screening using the Papanicolaou (Pap) test (also known as cervical cytology) was introduced to detect cervical intraepithelial neoplasia or more severe disease (CIN2+). Cervical screening programs prompted appropriate and timely management (Cervical Screening Participation, 2015; Rodríguez & Salmerón, 2017; Tota et al., 2017; Canadian Cancer Society, 2016) of cervical pre-cancers and cancers, leading to substantial decreases in cervical cancer incidence and mortality (Canadian Cancer Society, 2016; Schiffman, 2017; Franco et al., 2001). Notably, higher income countries have experienced the benefits of population-based cervical screening programs, but lower income countries that lack organized or widespread opportunistic screening face disproportionate burden of the disease (Rodríguez & Salmerón, 2017).

Persistent infections of human papillomavirus (HPV) play a necessary role in cervical cancer carcinogenesis (Franco et al., 2001). Among over 100 types of HPV, the International

¹ Throughout this dissertation, I use the term *women* to represent individuals who were born with cervixes, although I recognize this term might not feel inclusive to transgender individuals. There is little research and information available on cancer screening among trans- and non-binary individuals, and their unique needs should be considered in cervical screening moving forward.

Agency for Research on Cancer (IARC) categorized twelve types of HPV as “high-risk” or cancer-causing one type as a probable carcinogen, and a further twelve as possible carcinogens (Clifford et al., 2003; Bruni et al., 2017). HPV16 is the most common type of high-risk HPV, and together with HPV18, is responsible for almost three-quarters of cervical cancers (Clifford et al., 2003; Bruni et al., 2017). HPV infections are the most common sexually transmitted viral infections today, though in most cases the infection is transient and does not cause any physical symptoms. Some types of HPV infection are not associated with cancer but may have other clinical consequences, including genital warts (Baseman & Koutsky, 2005; Trottier & Franco, 2006). Most sexually active individuals will have an HPV infection at some point in their life, but only in rare occasions will an infection develop into cancer (Baseman & Koutsky, 2005; Trottier & Franco, 2006).

Cervical cancer and HPV epidemiology.

Cervical cancer and HPV risk factors.

As HPV is a sexually transmitted infection, certain high-risk sexual behaviours increase the risk of acquiring an infection. In particular, an increased number of sexual partners over the lifetime is associated with HPV acquisition (Bruni et al., 2017; Baseman & Koutsky, 2005; Trottier & Franco, 2006). Age at first intercourse has been suggested as an independent causal factor for HPV acquisition, possibly due to cervical cell changes in adolescence though it may also be an indicator for other sexual behaviours (Trottier & Franco, 2006).

Several other factors affect whether HPV infections develop into cervical cancer including smoking, parity (i.e., the number of times a woman has given birth), oral contraceptive use, co-infection with other sexually transmitted infections, and immunosuppression (Bruni et al., 2017; Baseman & Koutsky, 2005; Trottier & Franco, 2006; Dickinson et al., 2013). Current smoking or a history of smoking is positively associated with HPV infection but could be related

to cervical carcinogenesis (Trottier & Franco, 2006). Infection with sexually transmitted bacteria or virus (like HIV) might increase persistence of HPV through inflammation of HPV-infected cells (Trottier & Franco, 2006), or a weakened ability of the body to stave off infection. Additionally, individuals with HIV may have a higher prevalence of other cervical cancer risk factors (e.g. smoking), which compounds the risk of developing cervical cancer (Dickinson et al., 2013; Hernández-Ramírez et al., 2017).

The relationship between cervical cancer and an increase in completed pregnancies could be related to nutritional, traumatic, hormonal, or immunologic factors (Castellsagué & Muñoz, 2003; Muñoz et al., 2002). Additionally, researchers have hypothesized a correlation between persistent HPV infection and oral contraceptive use, though systematic reviews have drawn some conflicting conclusions on the association, often related to the heterogeneity of parity measurement in primary studies (Castellsagué & Muñoz, 2003; Moreno et al., 2002; Green et al., 2003; Gierisch et al., 2013). Overall, it seems that there is an increased risk of cervical cancer with duration of use of oral contraceptives, and but the effect might disappear when use has ceased (Gierisch et al., 2013; International Collaboration of Epidemiological Studies of Cervical Cancer et al., 2007).

Prevalence of HPV.

In Ontario and across Canada, infection with HPV is not reportable, so syntheses of multiple sources are needed to generate local estimates of prevalence. A 2013 systematic review and meta-analysis of eight studies found that the prevalence of any high-risk HPV type in Canada was 27.1% (among 15,188 study participants) (Tricco et al., 2011). Across studies in Canada, 8.6% of the study population had an HPV16 infection, and 3.3% of the study population had an HPV18 infection (Pham et al., 2007). Oncogenic types of cervical HPV infection were more frequently observed in younger (less than 29 years) and older (60 years and older) women

(Pham et al., 2007). While there is little evidence about the distribution of HPV infections by other socio-demographic factors, one group of researchers identified that Indigenous women might have higher rates of infection and increased persistence of infection compared to non-Indigenous communities (Brassard et al., 2012; 2015).

Cervical cancer incidence.

The 2017 Canadian Cancer Statistics report indicated that 1,550 new cases of cervical cancer were expected to be diagnosed in 2017, with an age-standardized incidence rate of 8.3 cases per 100,000 (Canadian Cancer Society, 2017). Almost half of the new diagnoses of cervical cancer will be done in Ontario (projected 710 cases), corresponding to an age-standardized incidence of 9.8 cases per 100,000. The incidence of cervical cancer in Canada plateaued between 2005 and 2010 (Canadian Cancer Society, 2016) with an annual change of 0.5%; a recent analysis indicated a 3.0% decrease in age-standardized incidence between 2009 and 2013 (Canadian Cancer Society, 2017).

Cervical cancer incidence is similar across Canadian provinces (Canadian Cancer Society, 2017; Cervical Screening Participation, 2015) with the highest rates observed in Newfoundland. Certain regions in Canada might experience higher rates of cervical cancer due to a higher prevalence of risk factors (e.g. higher proportion of the population who smokes) and increased barriers to health care (e.g., low attachment to primary care providers). Women in rural and remote areas tend to experience higher incidence of cervical cancer and most cancers in general (Cervical Screening Participation, 2015; Canadian Cancer Society, 2016). In Canada, these disparities are particularly prominent in First Nations, Inuit, and Métis communities who face significantly higher rates of cervical cancer (Chiefs of Ontario et al., 2016).

A recent worldwide analysis showed that cervical cancer accounted for 17.5% of new cancer cases of women between 20 and 39 years (Fidler et al., 2012). In Canada, more than half

of cervical cancers were diagnosed among females under 50 years, with females between 40 and 49 years at the highest risk (Canadian Cancer Society, 2016, 2017; Canadian Cancer Society Statistics, 2018). This was also observed in Ontario (Navaneelan et al., 2018). These age-related patterns reflect the typical epidemiology of HPV infection and subsequent carcinogenesis (Dickinson et al., 2012). A recently-published special edition of Canadian Cancer Statistics (Canadian Cancer Society, 2018) reported that more than 70% of cervical cancers in females aged 18 to 39 years were diagnosed at Stage I, and stage IV diagnoses ranged from 0% in the 18 to 24 age group to 23% in the 70+ years age group.

Cervical cancer mortality and survival.

In 2017, the Canadian Cancer Society reported that females have a 0.2% probability of dying from cervical cancer in their lifetime. In 2017, the projected age-standardized mortality rate was approximately 2.0 per 100,000 females (Canadian Cancer Society, 2017), which has not significantly changed since 2008. The mortality rate ranges from 1.6 per 100,000 in Quebec to 4.0 per 100,000 in Saskatchewan. In Canada, the five-year age-standardized net survival for cervical cancer was 73% which was the sixth highest five-year survival proportion among female cancers (Canadian Cancer Society, 2017).

Cervical Cancer Prevention and Screening

Cervical cancer can be prevented through primary and secondary prevention, specifically HPV vaccination and early detection or cervical cancer screening.

HPV vaccination.

In Canada, there are three vaccines available: the bivalent (2-valent) Cervarix™ protects against two types of high-risk HPV, the quadrivalent (4-valent) Gardasil® protects against four types, and the nonavalent Gardasil®9, the most recent iteration, protects against nine (Public

Health Agency of Canada, 2018). In 2006, Health Canada allocated \$300 million to provinces and territories to establish HPV immunization programs, which can be informed by national evidence-based recommendations. Currently, three vaccines are approved for use in Canada: Gardasil® and Gardasil®9 are approved for use in females aged 9 to 45 years of age, but also for males aged 9–26 years. Cervarix™ is approved for use in females between 9 and 45 years.

The HPV vaccine is now part of the school-based immunization program in Ontario, as in other provinces. In the 2015/2016 school year, Public Health Ontario reported that 71% of eligible children (males and females) received the first dose of the vaccine series (Ontario Agency for Health Protection and Promotion, 2017). Of these “initiators”, 86% completed the two-dose series. Using a methodology that more accurately depicts the vaccination program’s coverage, Public Health Ontario reported that 61% of 13-year old females are *up-to-date* with the HPV vaccine. Ultimately, current estimates of HPV vaccination uptake as of 2016 suggest that the program’s coverage is insufficient to ensure herd immunity (Brisson et al., 2016). A systematic review and meta-analysis showed that most models indicate that with 80% coverage, the HPV vaccine could potentially eliminate HPV 16 and 18 which are responsible for the large majority of cervical cancers after 70 years (Brisson et al., 2016).

Cervical cancer screening.

In Canada, most provinces offer organized, population-based programs for cervical cancer screening. The goal of these programs is to screen an asymptomatic population to detect and treat pre-cancerous lesions, preventing or diagnosing cancer at an earlier stage, improving prognosis and minimizing harmful consequences (Salmi et al., 2016; Miller, 2014; Wilson & Jungner, 1968). Cervical cytology has been the primary screening test in Canadian cervical screening programs for more than 70 years (Canadian Cancer Society, 2016, 2017). Pap tests are used to find cellular abnormalities in a sample of cervical cells. Since their implementation, these

programs have substantially reduced cervical cancer incidence and mortality through early detection of cervical intraepithelial neoplasia or more severe disease (CIN2+), as well as appropriate management (Canadian Cancer Society, 2016; Schiffman, 2017; Franco et al., 2001).

The ultimate goal of any cancer screening is to reduce the mortality of a target cancer or in some cases, reduce all-cause mortality (Saqib et al., 2015; Prasad et al., 2016). Most evidence of benefit with cervical cytology originates from cohort studies or case-control studies (Dickinson et al., 2013). Despite the lack of randomized controlled trials with mortality outcomes, it is widely accepted that cervical cytology is effective at decreasing the mortality from cervical cancer in Canada and similar regions (International Agency for Research on Cancer, 2005). Most trials evaluate the ability of the screening test to detect pre-cancerous lesions or worse, including cancer.

More recently, some guidelines (Tota et al., 2017; Smith et al., 2017; Murphy et al., 2012a) recommend HPV testing as a primary cervical screening test. Such testing detects the presence of high-risk HPV types even before there are any visible changes to the cells. There are several large randomized trials that compare HPV testing to Pap testing for primary cervical screening, although no Canadian trials (or trials in other high-resource settings) have used cervical cancer mortality as an outcome (Dickinson et al., 2013; Ogilvie et al., 2018). A 2017 Cochrane systematic review and meta-analysis concluded that the HPV test performed on cervical samples has increased sensitivity and lower specificity to appropriately detect CIN2+ compared with Pap testing (Koliopoulos et al., 2017). Substantial decreases in abnormal cytology results and CIN2+ diagnoses are expected as a consequence of the implementation of HPV vaccination programs. The reduction of prevalence of cervical lesions in the general population will result in a reduction in the positive predictive value of any cervical screening test

(Tota et al., 2017; Canadian Cancer Society, 2016). As a result, HPV testing might be more favourable given its higher detection rate.

Consistent with Wilson and Jungner's 1968 health screening criteria (Wilson & Jungner, 1968) and new guidance around requirements of a health screening program (Dobrow et al., 2018), the programmatic setting is at least as important as the detection ability of a screening test. In particular, having the appropriate program infrastructure and health system coordination to ensure timely follow-up to positive results is crucial to the success of early detection strategies. In Canada, these programs are currently administered within provincial health care systems. Different decision-makers and guideline developers have judged the evidence on cervical cancer screening differently which has led to provincial differences regarding the type of test recommended, starting/stopping ages, and intervals between tests (Cancerview.ca, 2016).

In 2013, updated guidelines from the Canadian Task Force on Preventive Health Care (Dickinson et al., 2013) recommended that women between 25 and 69 years of age be screened at three-year intervals; these guidelines recommend a later starting age and a longer interval between screens than the previous guidelines. Even though some provincial programs suggest initiation as early as 20 or 21 years, the Task Force argued that cervical screening should not be offered to women younger than 25 years (Dickinson et al., 2017; Krueger et al., 2013). Additionally, due to a lack of “developed” evidence, these recommendations did not address primary screening with HPV testing. Using a 2013 systematic review (Murphy et al., 2012b), Cancer Care Ontario (CCO) has recommended HPV testing for primary screening (Murphy et al., 2012a) for women between 30 and 65 years as part of Ontario's cancer control strategy, which was published even before the Canadian Task Force made their recommendations. Five years following the recommendations, HPV testing has not yet been funded or implemented in Ontario, although the 2017 provincial budget alluded to funding the test in the near future

(Ministry of Finance (Ontario), 2017). The inconsistencies between jurisdictions and respective recommendations are affecting how women view and experience cervical screening, as communicated in a recent editorial (Taylor, 2018).

Most recommendations and evidence syntheses that consider HPV testing as a primary screening test consider HPV testing on samples collected by a health professional (Koliopoulos et al., 2017; Murphy et al., 2012b; Huh et al., 2015). However, an additional option for HPV testing includes a self-collection modality, in which women can use a device (i.e., a brush, swab, lavage, or tampon) to collect their own sample of vaginal cells. A high quality meta-analysis (Arbyn et al., 2014) showed that for PCR-based HPV tests, the sensitivity and specificity for detection of CIN2+ based on self-collected vaginal samples was slightly lower than the sensitivity and specificity based on cervical samples obtained by a health professional. This work complements findings from systematic reviews suggesting that self-sampling is preferred by women who face barriers in accessing the Pap test (Verdoodt et al., 2015; Racey et al., 2013; Nelson et al., 2017). A 2017 systematic review found that, when offered the choice between a self-collection option or a provider-collection option, the majority of women prefer a self-collection option for cervical cancer screening. Interestingly, in a separate analysis of studies that included a third choice of “no preference between collection methods” (versus self-collection or provider-collection), there was no obvious preference for a specific modality (Nelson et al., 2017). Most of the research on the acceptability of HPV self-sampling is limited to samples from the under- or never- screened populations, and in almost all studies, the screening kits have been delivered by mail (Verdoodt et al., 2015). Despite the availability of high quality scientific studies on self-sampling, provincial guidelines in Canada do not suggest that self-sampling for HPV testing is an option for women (Murphy et al., 2012a; Cancerview.ca, 2016). Of the screening modalities – Pap testing, HPV testing collected by a professional, self-collected HPV

testing - no option is definitively superior when considering effectiveness, use of health care resources, and women's preferences.

Cervical Cancer Screening in the Era of HPV Vaccination.

The coverage and impact of vaccination programs, in Ontario and other jurisdictions, has important implications for cervical cancer screening. Most notably, current vaccination coverage is not producing optimal herd effects. Furthermore, simulations suggest that reaching complete elimination of select high-risk types of HPV will take upwards of 70 years, and the immunogenicity duration of the HPV vaccine is not yet confirmed (Canadian Cancer Society, 2016; Giorgi Rossi et al., 2017). As a result, early detection with cervical cancer screening can continue to offer population health benefits, given the continued existence of persistent HPV infection. Additionally, early detection approaches would be expected to reduce the burden of cervical cancers caused by oncogenic HPV types not covered (directly or through cross-protection) by the vaccine. The effects of HPV immunization on cervical cancer risk might not be evident until the immunized cohort reaches 40 years, due to low rates of cervical cancer in the younger age group as well as the etiology of the disease (Canadian Cancer Society, 2016; Giorgi Rossi et al., 2017). As a result, HPV vaccination status could influence how women perceive their risk of cervical cancer and how they wish to participate in screening.

Disparities in Cervical Cancer, Cervical Screening, and HPV Vaccination.

Women's sociodemographic characteristics, including ethnicity, are strongly linked to their risk of morbidity and mortality from cervical cancer, which is correlated with barriers to cervical cancer screening and process of care failures rather than biological differences (Spence et al., 2007; Cancer Care Ontario, 2016; Elit et al., 2013). More than half of Canadian women who are diagnosed with cervical cancer have never participated in cervical cancer screening or in some cases, participated infrequently (Spence et al., 2007). Studies from the United States

indicate that there is up to a 44% relative difference in cancer mortality between white women and black women (Beavis et al., 2017; Yoo et al., 2017); there are no comparable studies in Canada. However, some administrative analyses have demonstrated that First Nations, Inuit, and Metis women have higher mortality rates and lower survival rates (Cancer Care Ontario, 2016), which can be broadly attributed to increased prevalence of risk factors, socioeconomic inequities, communities' mistrust of the mainstream health care system, and the legacy of colonialism (Spence et al., 2007; Cancer Care Ontario, 2016; Elit et al., 2013; Wakewich et al., 2016).

Barriers to participation in HPV vaccination often coincide with barriers to participation in cervical cancer screening (Rodríguez & Salmerón, 2017; Canadian Cancer Society, 2016). Research studies and surveillance data clearly demonstrate that certain communities and populations face barriers when accessing screening. In particular, low-income women (Elit et al., 2013), immigrant women (Lofters et al., 2007), First Nations, Inuit, and Métis women (Cancer Care Ontario, 2016; Withrow et al., 2014), and women who identify as lesbian (Reiter & McRee, 2015) are less likely to participate in organized cervical cancer screening. Low participation in cervical screening or HPV immunization these communities are attributed to socioeconomic inequities, low attachment to primary care providers, a lack of comfort discussing cervical cancer with providers, communities' mistrust of the mainstream health care system, and the legacy of colonialism (Spence et al., 2007; Gesink et al., 2016).

As a result, women who experience barriers to accessing cancer prevention or control strategies face disproportionate risk of cervical cancer. Subsequently, women's personal characteristics and experiences could affect their risk of cervical cancer, how they perceive their risk of cervical cancer, and how they make choices around cervical screening participation.

Introducing multiple options for cervical screening may offer women autonomy in decision-making and is consistent with the shift toward person-centered health care (van Dam et

al., 2013; Sawaya & Kuppermann, 2015; Stacey et al., 2017). Offering women a choice among modalities, including self-sampling for HPV testing, might support groups of women who face barriers to participation in cytology-based provincial programs. This approach raises practical and ethical concerns for health systems. For example, introducing HPV testing technology requires new lab infrastructure that will automate the testing process which will require a substantial investment of resources and significantly reduce the demand for cytology technicians. Some organizations in high-resource settings question whether the investment is worthwhile given that there is a low incidence of cervical cancer and that cytology still has satisfactory performance. To further consider additional modalities such as self-sampling requires decision-makers to make trade-offs with competing health system priorities. As well, health systems must reflect on how offering choice among cervical screening options affects screening uptake and subsequent population health outcomes.

In the era of HPV vaccination, vaccinated cohorts of women might have a differential risk for cervical cancer which might influence their preferred cervical cancer screening strategy. There is little evidence on women's informed preferences in cervical screening (Stacey et al., 2017; Lillie et al., 2014); subsequent implications for population health and resource utilization of implementing preference-based cervical screening, including cost and burden of disease, are unknown. As a result, it is challenging to assess whether offering multiple options to women would affect participation in screening programs, let alone relevant population health outcomes.

Cervical Cancer Screening Preferences

In cervical cancer screening, most Canadian jurisdictions have promoted the Pap test as the only “valid” option because of its observed success in reducing cervical cancer morbidity and mortality (Canadian Cancer Society, 2016; Dickinson et al., 2013; Cancer Care Ontario, 2014; Murphy & Elit, 2013). Experts from Canada and other jurisdictions seem to promote HPV

testing with a similar enthusiasm (Tota et al., 2017). Encouraging all women to participate in early detection with cervical cytology has led to a lower incidence of cervical disease and could continue to contain health system costs by intervening with less intensive treatment earlier. Guideline developers and policy-makers use utilitarian logic to optimize population health and use of system resources; they perceive that women's individual preferences could detract from the population benefit of an organized screening program. This perception is reflected in Canadian cervical cancer screening guidelines as all recommendations lack explicit mention of women's preferences (Cancerview.ca, 2016). Ontario guideline developers even communicated that "initiation of screening based on individual preference. . . is neither reliable, practical nor necessary as a routine" (Murphy & Elit, 2013) in response to the Canadian Preventive Task Force on Preventive Health Care's strong recommendation for Pap testing for women over 30 years. This perspective is somewhat paternalistic as women could have valid reasons for choosing not to participate in cervical screening (Screening, 2005; Schwartz et al., 2004; Stefanek, 2011; Paskett et al., 1995; Sandman et al., 2012).

New approaches to guideline development emphasize the importance of consideration of people's preferences in the creation of evidence-informed, person-centered recommendations (Pieterse & Stiggelbout, 2016; Umscheid, 2009; Légaré et al., 2011; van der Weijden et al., 2010; Biddy et al., 2015; Montori et al., 2013a). In the case of preventive health screening, like cervical screening, most individuals will not experience benefit and there are risks of harms from the screening process or of false positive test results (e.g., anxiety) and false negative results (e.g., missed early intervention). From an equity perspective (Lofters & Vahabi, 2016), an ethical perspective (Snadden, 1992; Jepson et al., 2007), and system performance and population health perspectives (Stacey et al., 2014; Hibbard & Greene, 2013), high quality cervical screening guidelines and practice should incorporate women's preferences.

Given that Ontario and other jurisdictions are currently updating their recommendations to include new modalities – namely HPV testing administered by a professional - for cervical screening (Tota et al., 2017; Murphy et al., 2012a), understanding issues around Ontario women’s preferences for cervical screening modalities is timely and relevant. This doctoral dissertation set out to understand the determinants of women’s cervical screening preferences from the perspectives of multiple stakeholders. With this understanding, I investigated robust methods to elicit women’s preferences among cervical screening modalities. As part of the dissertation, I developed a novel online tool to elicit women’s preferences based on a shared decision-making framework.

Goal and Objectives of the Dissertation

This doctoral dissertation aimed to elicit and describe women’s preferences among cervical screening modalities, to inform person-centered cervical cancer screening guidelines in Ontario, Canada. I identified two overarching research objectives:

1. To determine the perceptions of stakeholders (women, health care providers, system managers, and researchers) in cervical cancer about possible screening options and to understand how women make choices among these options (Chapters 2 to 4).
2. To examine methods to measure women’s preferences in cervical cancer screening, that will either be applied in an Ontario-based study or will inform the development and testing of a tool to elicit Ontario women’s preferences (Chapters 5 to 7).

Organization of the Dissertation

This dissertation is manuscript-based and includes five original manuscripts and a draft protocol for a follow-up study. The current chapter (Chapter 1) presents the rationale for the thesis, the research objectives, and the conceptual frameworks that guided the research. Chapters 2 through 6 present primary studies, and Chapter 7 describes a proposed extension of the work

presented in this dissertation. In Chapters 2 to 4, I aimed to address the first research objective: to understand what is known about women’s preferences in cervical cancer screening by considering the perspectives of guideline developers and program administrators, women considering screening and health care professionals, and then looking at the published literature. The second section of this dissertation addresses the second research objective. In Chapters 5 through 7, I examined approaches to robustly describe or measure women’s preferences for cervical screening, so that I could apply this method in a study of women considering cervical screening in Ontario. Chapter 8 presents an integrated discussion that summarizes and synthesizes the main findings, implications, and final conclusions. Each manuscript contains individual reference lists at the end of the chapter and Appendix F contains a bibliography of all references used throughout this dissertation.

Research ethics board approval was acquired for the studies presented in Chapter 2 (Manuscript 1) from the Ryerson Research Ethics Board (REB 2016-036) and Ottawa Health Science Network Research Ethics Board (Protocol #20160134-01H). Chapters 3 and 6 (Manuscripts 2 and 5 respectively) received approval through amendments made to the Ottawa Health Science Network Research Ethics Board Protocol #20160134-01H. Details are provided in Appendix A.

Table 1-1 presents an overview of the content of the dissertation.

Table 1-1. Thesis Chapter Outline and Alignment to Manuscripts

Chapter	Manuscript	Study Description	Submission Status
1		An outline of the dissertation and description of the current cervical cancer screening context	Not applicable
2	Manuscript #1	A qualitative interview study on barriers and facilitators to implementing alternative cervical cancer screening options, such as self-sampling	Published: Current Oncology, 25(1), Feb 2018
3	Manuscript #2	A qualitative interview study of stakeholders’ perspectives of women’s decisional needs to choose among cervical cancer screening modalities	Published in Global Qualitative

			Nursing Research, July 2018
4	Manuscript #3	A systematic review of quantitative, qualitative, and mixed methods studies that determines the prevalence and determinants of women's informed and values-based cervical screening preferences.	Manuscript #3: Prepared for submission
5	Manuscript #4	A systematic review of qualitative, quantitative, and mixed methods studies of the types of approaches used and the validity of the approaches of preference-elicitation in cervical cancer screening.	Manuscript #4: Prepared for submission
6	Manuscript #5	The development and field testing of a novel online tool to elicit women's cervical screening preferences	Prepared for submission
7	Research proposal	Proposed research project that aims to produce data for preference-based cervical screening guidelines in Ontario.	Potential postdoctoral application
8	Integrated Discussion	Summarizes and integrates the main findings of the dissertation.	

In Chapter 2 (Manuscript 1), I focused specifically on how guideline developers and decision-makers view a self-sampling option, as no guidelines or programs have explicitly addressed it in their recommendations. Considering that other jurisdictions have implemented self-sampling as a method to increase coverage and that, in Canada, a self-sampling kit can be purchased privately, this qualitative interview study aimed to identify the facilitators and barriers to including alternative options, like self-sampling, in Canadian cervical screening programs.

Chapter 3 (Manuscript 2) then built upon Chapter 2 (Manuscript 1) findings by using a shared decision-making framework to determine what Ontario women need to make an informed and values-based choice among modalities: cervical cytology (Pap testing), HPV testing conducted by a professional, self-collected HPV testing, or no screening. Using qualitative interview techniques, Chapter 3 (Manuscript 2) aims to understand the needs of women eligible for cervical screening and health professionals (i.e. providers and program administrators) if multiple options were introduced. I designed this study to highlight the specific information that will support women identifying their informed, valued-based preferences.

As no previous studies have considered women's preferences through a decision-making lens, I examined qualitative, quantitative, and mixed methods literature in Chapter 4 (Manuscript 3) to describe women's informed and values-based cervical screening preferences. Recognizing that guideline developers and decision-makers want robust evidence to support the implementation of alternative screening strategies (Chapter 2, Manuscript 1), I conducted a knowledge synthesis of published and grey literature. The purpose of this study was to understand women's screening preferences, anchored in the study contexts and approaches the preference-elicitation. Unlike previous systematic reviews, this study offered insight into cervical screening preferences described in the qualitative body of research, which could help deepen understanding around why women prefer what they do.

Thus, the first section of this dissertation (Chapters 2 through 4) highlighted important gaps in the body of evidence around women's preferences for cervical screening, particularly in the context of new modalities. In Chapters 5 through 7, I presented two studies and a study protocol to address these gaps. In Chapter 5 (Manuscript 4), I aimed to critically evaluate the approaches used to elicit women's preferences for cervical cancer screening. With this study, I intended to inform guideline developers who are developing preference-sensitive guidelines, the development of which would require robust evidence from preferences and values studies. This study was the first to consider the robustness of methods used to elicit women's preferences and considered diverse study contexts and study designs.

Based on the findings from Chapter 5 (Manuscript 4), as well as the gaps identified in the first section of the dissertation (Chapters 2 through 4), there was a need to develop a novel tool to elicit women's preferences, using a person-centered and evidence-informed approach. Chapter 6 (Manuscript 5) presents the development and field testing of an online preference-elicitation tool tailored to the Ontario cervical cancer screening context, guided by the Ottawa Decision

Support Framework and International Patient Decision Aids Standards quality and evaluation criteria. To my knowledge, this is the first application of a shared decision-making lens to a primary cervical cancer screening decision. Chapter 7 presents a proposal for a population-based study that culminated from Chapters 2 through 6 to achieve the aforementioned research objective. In this proposal, screening stakeholders are engaged to understand how the preference-elicitation tool developed in this dissertation can address the needs of guideline developers and decision-makers. This study is intended to produce relevant and timely information to modernize cervical screening guidelines and practice in Ontario and similar jurisdictions.

Conceptual Frameworks

There are a variety of frameworks that underscore how values clarification and/or preference-elicitation approaches are developed, by outlining important constructs and the relationships between the constructs that lead to preference-elicitation (Pignone et al., 2012; Fagerlin et al., 2013; Witteman et al., 2016a; Witteman et al., 2016b; Pieterse et al., 2013). In this dissertation, I used decision support frameworks to guide the development and assessment of a tool to elicit women's preferences among cervical cancer screening modalities. The Ottawa Decision Support Framework (ODSF) was used to help conceptualize preferences and preference-elicitation in the context of cervical cancer screening. To develop and field test a preference-elicitation tool based on shared decision-making, I used the International Patient Decision Aids Standards (IPDAS) systematic development process and its quality and evaluation criteria. In Chapters 4 and 5, I relied on principles from the evidence-to-recommendation frameworks from Grading of Recommendations, Assessment, Development and Evaluation (GRADE) to assess the findings from a systematic review of the published literature of women's preferences for cervical screening.

Ottawa Decision Support Framework (ODSF).

The Ottawa Decision Support Framework (ODSF) is a widely used framework that offers a conceptual foundation for participating in shared decision-making in practice (O'Connor et al., 1998, 2002). According to this framework, addressing individuals' needs is integral to supporting and achieving high quality decision-making. In particular, exercises to clarify individuals' values related to the decision and examine comprehension of the options will help individuals select their preferred option. A 2016 sub-analysis of a Cochrane systematic review found that decision aids may help patients make screening decisions congruent with their values, but the evidence base is limited due to heterogeneous measures and suboptimal sample size (Munro et al., 2016).

The ODSF outlines the key components of a decision-making process for health or social decisions. This framework contains three overarching elements: decisional needs, decision support, and decision quality (O'Connor et al., 1998, 2002). According to the ODSF, decision support helps individuals through five steps: i) Realizing a decision needs to be made; ii) Information comprehension; iii) Clarifying values and identifying preferences according to these values; iv) Considering resources, including social influences, that affect the decision; and v) Forming an action plan.

The ODSF guided the decisional needs interview process and the subsequent analysis with cervical screening stakeholders in Chapter 3 (Manuscript 2). In Chapters 4 and 5 (Manuscripts 3 and 4, respectively), I used the ODSF conceptualization of preferences (i.e. steps i, ii, and iii) to screen, assess, and organize the preference-elicitation methods included in a systematic review of qualitative, quantitative, and mixed methods studies. I used the ODSF personal decision guide template (O'Connor et al., 2015) to develop the structure of the preference-elicitation tool presented in Chapter 6 (Manuscript 5).

International Patient Decision Aids Standards (IPDAS).

The International Patient Decision Aids Standards (IPDAS) is an international consortium established in 2006 that created a model for developing and certifying high quality tools for decision support (Elwyn et al., 2006). IPDAS have established a checklist of criteria for decision aids in the domains of essential content (providing balanced information, presenting probabilities, clarifying values, and guiding deliberation and communication), development (systematic methods, balanced presentation, up-to-date and transparent evidence, and plain language), and evaluation (informed and values-based decisions). The IPDAS decision aid development process (Kryworuchko et al., 2011; Coulter et al., 2013) and quality and evaluation criteria guided the creation and field testing of an online tool to elicit women's preferences (Chapter 6, Manuscript 5). IPDAS specifies that the development process begin with a protocol that outlines the scope of the project, as well as the establishment of an advisory committee (Coulter et al., 2013; Elwyn et al., 2011). Following the scoping and committee formation, a needs assessment and a review of the relevant evidence rounded out the *content specification* phase. The design phase followed which involved an iterative process of prototype creation and usability testing, first within the project advisory committee, then with individuals not involved in the development process (Coulter et al., 2013; Elwyn et al., 2009, 2011; Joseph-Williams et al., 2014).

An external reviewer used the IPDAS v4.0 - an instrument that was developed to assess the quality of the development process, to examine whether a product meets the minimum criteria for decision aid certification, and to rate the quality of the final preference-elicitation tool (Joseph-Williams et al., 2014). Chapter 6 (Manuscript 5) describes these processes in greater detail.

Grading of Recommendations, Assessment, Development, and Evaluation.

The Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) is a method to appraise the quality of evidence and determine the strength of recommendations in clinical practice guidelines. This approach was developed by a collaboration of methodologists, guideline developers, scientists, clinicians, and other interested members to assist health system decision-makers in assessing and using recommendations in public health practice and policy (Guyatt et al., 2008a, Guyatt et al., 2008b, Guyatt et al., 2008c). The GRADE working group developed the Evidence-to-Decision framework to use a systematic and transparent process to incorporate important criteria into the development of recommendations for practice guidelines and public health practice (Alonso-Coello et al., 2016). To generate a recommendation, stakeholders must consider the balance between desirable and undesirable effects, the certainty of evidence to informing a recommendation, resource utilization and feasibility, implications for system equity, and the values and preferences of stakeholders, including patients or members of the public. A recent set of publications presented a GRADE approach to consider local values and preferences in guideline development, including a method to assess the risk of bias in preference-elicitation primary studies (Yepes-Núñez et al., 2017; Zhang et al., 2017, 2018a, 2018b; Selva et al., 2017). The GRADE approach offers some methodological rigour and structure to integrating individuals' values and preferences into guideline recommendations, although GRADE researchers acknowledge that this approach requires further refinement given the heterogeneity of “preference” definitions. In this dissertation, I intended to apply the GRADE approach to synthesizing preferences data in the systematic reviews (Chapters 4 and 5, Manuscripts 3 and 4, respectively). I contacted two researchers involved with this work so that I could operationalize the risk of bias domains and criteria in these systematic reviews.

Table 1-2 conveys how the conceptual frameworks are used in this dissertation.

Table 1-2. Relationship of Dissertation Content to Systematic Development Process

Chapter	Chapter Title	Conceptual Framework Applications	Objectives	Method
1	Introduction	-	To describe the problem and context	
2	Tailoring cervical cancer screening strategies to hard-to-reach women in Canada: a qualitative study of the potential value of HPV self-sampling	IPDAS - Scoping ODSF and IPDAS - Needs assessment	This qualitative study aimed to explore barriers and facilitators to implementation of HPV self-sampling in Canadian cervical cancer screening programs, from the provider and policy-maker perspective.	Qualitative interview study
3	“They should be asking us”: a qualitative decisional needs assessment for women considering cervical cancer screening	ODSF and IPDAS - Needs assessment	To investigate the decisional needs of women considering cervical cancer screening, and the relevant needs of other cervical screening stakeholders (including health care providers and policy-makers)	Qualitative interview study
4	Women’s preferences in primary cervical cancer screening: a narrative synthesis of qualitative, quantitative, and mixed methods studies	IPDAS - Review the evidence GRADE – Risk of bias criteria	To examine women’s informed, values-based preferences for primary cervical cancer screening, across a variety of study designs and preference-elicitation approaches.	Systematic review
5	Preference-eliciting approaches in cervical cancer screening: A systematic review of qualitative, quantitative, and mixed methods studies	IPDAS - Review the evidence GRADE – Risk of bias criteria	To comprehensively review and critically evaluate approaches that have been used to elicit women’s preferences about primary cervical cancer screening.	Systematic review
6	Development and field testing of a tool to elicit women’s preferences among cervical cancer screening modalities	ODSF – Decision Support IPDAS – Quality and evaluation criteria used in field test with users	To develop and pilot test a decision support tool that elicits preferences of women considering different cervical screening options	Descriptive quantitative study

7	Using multiple methods to understand preference-based cervical cancer screening in Ontario	ODSF – Decision quality and decision action outcomes	To describe a study design to investigate the preferences among cervical cancer screening modalities of a population sample of Ontario women using a validated, online preference-elicitation tool.	Proposed multiple methods study including a qualitative interview study, randomized controlled trial, and population-based study
8	Integrated Discussion		To integrate the dissertation content and identify potential directions for theory, cervical screening policy, and epidemiology	Descriptive synthesis

Author Contributions

According to the International Committee of Medical Journal Editors (www.icmje.org) (ICMJE), authorship should be based on four criteria:

- Substantial contributions to the design, execution, analysis, or interpretation of work;
- Contributions to the drafting or revising of the manuscript;
- Final approval of the version to be published;
- Agreement to be accountable for their respective contributions to the work.

Below I outline the list of authors and their respective contributions for each of the manuscripts and study protocols in this dissertation. All authors meet the ICMJE criteria for authorship.

Chapter 2, Manuscript 1: Strategies to reach marginalized women for cervical cancer screening: A qualitative study of stakeholder perspectives

Authors: Brianne Wood, Aisha Lofters, Mandana Vahabi

Published in: *Current Oncology*, 2018, 25(1)

The interview guide was developed by MV and AL. MV and AL received the CIHR planning grant for work related to this research. I established the analysis plan, conducted all interviews and subsequent analysis, wrote the first draft of the manuscript. All authors provided extensive feedback in the reporting of the results. JL and IDG assisted with the interpretation and presentation of the results, as well as the editing of the manuscript for the thesis chapter. All authors approved the final version of the manuscript.

Chapter 3, Manuscript 2: “They should be asking us”: a qualitative decisional needs assessment for women considering cervical cancer screening

Authors: Brianne Wood, Virginia L. Russell, Ziad El-Khatib, Susan McFaul, Monica Taljaard, Julian Little, Ian D. Graham

Published in: *Global Qualitative Nursing Review*, 5 (July 2018)

I created the study protocol, developed the interview guide, conducted and analyzed the interviews, and wrote the first draft of the manuscript and incorporated revisions. VLR was a second analyst and offered significant intellectual contribution to the final product. JL and IDG contributed to the interpretation of results and made significant editorial feedback on the manuscript. All authors contributed to the study protocol and offered substantial revisions to early drafts of the manuscript. All authors approved the final version of the manuscript.

Chapter 4, Manuscript 3: Women’s preferences in primary cervical cancer screening: a narrative synthesis of a systematic review of qualitative, quantitative, and mixed methods studies

Authors: Brianne Wood, Susan Rogers Van Katwyk, Jesse Elliot, Monica Taljaard, Ziad El-Khatib, Susan McFaul, Ian D. Graham, Julian Little

Prepared for: *BMC Public Health*

I created the study protocol, developed the search strategy, conducted the search of the literature, conducted the screening and data extraction of relevant articles, analyzed and interpreted the data, and wrote the first draft of the manuscript and incorporated suggested revisions. SRVK and JE contributed to the screening, data extraction, analysis and interpretation of the relevant articles, and made important contributions to revisions of the manuscript. IDG, JL, and MT made significant contributions to the interpretation and final presentation of the manuscript. All authors contributed to the study protocol and offered substantial revisions to early drafts of the manuscript. All authors approved the final version of the manuscript.

Chapter 5, Manuscript 4: Preference-eliciting approaches in cervical cancer screening: A systematic review of qualitative, quantitative, and mixed methods studies

Authors: Brianne Wood, Susan Rogers Van Katwyk, Jesse Elliot, Monica Taljaard, Ziad El-Khatib, Susan McFaul, Ian D. Graham, Julian Little

Prepared for: *BMJ Open*

I created the study protocol, developed the search strategy, conducted the search of the literature, conducted the screening and data extraction of relevant articles, analyzed and interpreted the data, and wrote the first draft of the manuscript and incorporated suggested revisions. SRVK and JE contributed to the screening, data extraction, analysis and interpretation of the relevant articles, and made important contributions to revisions of the manuscript. IDG, JL, and MT made significant contributions to the interpretation of the results and final presentation of the manuscript. All authors contributed to the study protocol and offered substantial revisions to early drafts of the manuscript. All authors approved the final version of the manuscript.

Chapter 6, Manuscript 5: Development and field testing of a tool to elicit women's preferences among cervical cancer screening modalities

Authors: Brianne Wood, Monica Taljaard, Ziad El-Khatib, Susan McFaul, Ian D. Graham, Julian Little

Prepared for: *Patient Education and Counselling*

I created the study protocol, led the development of the prototypes and incorporated revisions, developed the analysis plan, led the recruitment of participants, conducted the analysis, and wrote the first draft of the manuscript. All authors contributed significantly to the revised prototypes of the preference-elicitation tool as part of the advisory committee and made important editorial contributions to the final manuscript. All authors approved the final version of the manuscript.

Chapter 7, Potential Postdoctoral Proposal: Can an Online Preference-Elicitation Tool Support Preference-Based Cervical Screening in Ontario: A Protocol for a Multiple Methods Investigation

Authors: Brianne Wood, Julian Little, Ian D. Graham

I created the study protocol and integrated substantial contributions from my supervisors Drs. Little and Graham. I also acknowledge that the thesis advisory committee also provided some input on early drafts of the protocol, which improved the final version.

Framing of Dissertation and Statement of Worldview

I am a PhD Candidate in Epidemiology at the University of Ottawa, in Ottawa, Ontario, and my research focused on women's preferences among available cervical screening options and how to elicit these preferences. My research was funded by a Mitacs Accelerate fellowship

in partnership with Eve Medical, a self-sampling device company based in Toronto, Ontario. Previously, I was part of a team to examine how a self-sampling option affected cervical cancer screening uptake in First Nations communities in Northwestern Ontario. Currently, I am employed as an epidemiologist at the North West LHIN.

I support shared decision-making and personal autonomy of individuals making decisions about health care, in particular women considering cervical cancer screening. I also support evidence-informed medicine and practice in that clinical and/or health system decisions are based on systematic syntheses of high quality research evidence. As presented in this dissertation, I believe that shared decision-making and evidence-informed practice are intricately linked and that both approaches are important for high quality health care.

Introduction to Section 1: Understanding stakeholders' perspectives on women's preferences for cervical screening modalities

In the first section of my thesis (Chapters 2 to 4), I interviewed guideline developers, administrators, health care providers, and women eligible for cervical cancer screening to determine how stakeholders perceive women's preferences for cervical screening options. I also examined the published literature to synthesize what is known about preferences in cervical cancer screening.

Chapter 2: Tailoring Cervical Cancer Screening Strategies to Hard-to-Reach Women in Canada: A Qualitative Study of the Potential Value of HPV Self-Sampling

The work in this chapter was published in the February 2018 issue of Current Oncology (the published article is presented in Appendix B). This chapter contains additional information on the analysis methods, including saturation criteria and context of the research and researcher. I collaborated with two researchers from the University of Toronto and Ryerson University on this study and manuscript. I established the analysis plan, conducted all interviews and subsequent analysis, and wrote the first draft of the manuscript for this study. All authors provided extensive feedback in the reporting of the results and approved the final manuscript. My PhD supervisors from the University of Ottawa assisted with the interpretation and presentation of the results, as well as the editing of the manuscript for this thesis chapter.

Reference: Wood B, Lofters A, Vahabi M. Strategies to reach marginalized women for cervical cancer screening: A qualitative study of stakeholder perspectives. Curr Oncol. 2018; 25(1):e8–16. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/29507490>

Abstract

Background.

Self-sampling for HPV testing has the potential to increase the accessibility and acceptability of cervical cancer screening to marginalized populations, and thus address health inequity in cervical cancer incidence and mortality. Before implementing the offer of an alternative screening modality to under- and never- screened women, the key processes, facilitators, and barriers to inclusion of such an offer in the cervical screening strategy need to be understood.

Methods.

Semi-structured interviews were held with Canadian and international cancer screening health care providers and policy-makers. The interviews were transcribed verbatim and analyzed using directed content analysis. The findings were organized according to Attride-Stirling's basic, organizing, and global themes.

Results.

Data saturation was reached after nineteen interviews. Respondents identified four main categories of interest for developing cervical screening strategies for hard-to-reach women: (1) understanding the disparities in cervical screening and outcomes and the populations who experience them; (2) using the criteria of a successful population screening program to shape optimal strategies to reach marginalized populations; (3) recognizing the need for knowledge creation and translation among researchers, health professionals, and women considering screening; and (4) addressing broader contextual factors that influence screening participation.

Conclusion.

Canadian screening stakeholders should collaborate to identify actionable research questions and make use of existing evidence to implement tailored strategies for vulnerable populations. Respondents highlighted the need to implement change management strategies for women and point-of-care health care providers to encourage successful modernization of cervical cancer screening programs. Researchers and guideline developers should consider women's preferences and the needs of health professionals to implement tailored, women-centered cervical screening strategies.

Introduction

Cervical cancer screening programs using cervical cytology (the Pap test) have substantially reduced incidence and mortality from cervical cancer over the past several decades (1). Despite investment in health promotion initiatives, incidence and mortality have remained unchanged in North American and European countries in the past twenty years (2–5). Demographic shifts (e.g., increased immigration from countries without screening programs), changing social behaviours, limitations of cervical cytology, and suboptimal program delivery could play a role in the plateau of cervical cancer mortality in countries with early detection programs (4,6).

Recent cervical cancer prevention and control innovations target persistent infection of oncogenic human papillomavirus (HPV) - a necessary cause of cervical cancer (7,8) - through HPV vaccination programs that target school-age children before exposure to HPV, and HPV DNA primary screening tests, a test with greater sensitivity and higher positive predictive value than Pap testing (7,9–14). Currently, many jurisdictions, including most Canadian provinces, are considering replacing the Pap test with high-risk HPV testing, which may overcome some of the

performance limitations of cytology, particularly the low positive predictive value in the era of HPV vaccination. While the diagnostic performance of an HPV test is considered superior to traditional cytology (15), a new screening test will not overcome the suboptimal performance of early detection programs. Opportunistic programs and barriers to accessing cervical cancer screening (2,16–19) will still largely affect the impact of early detection programs on cervical cancer mortality. Most concerning is that these methods fail to reach populations who are already marginalized, including newcomers to Canada, Indigenous peoples, and women in rural/remote regions (20–22).

HPV testing can be performed on a vaginal sample that women collect themselves – “self-sampling” - which might be an attractive option to overcome some of the barriers to provider-administered screening (23–27). Though there is high quality evidence internationally to support self-collected sampling in population-based programs, the Netherlands (28), Denmark (29), and Australia (30) are the only organized cervical screening programs that have implemented a self-sampling strategy as part of their population-based program. A recent review (19) found that the evidence indicates HPV testing on self-samples is similar to provider-collected samples, and that it could improve screening rates in some populations.

With such a low implementation rate of self-sampling in cervical cancer early detection programs, there is a gap between the evidence to support a self-sampling option and current policy and practice (31,32). From an equity perspective, self-sampling might increase access to cervical cancer screening for hard-to-reach populations (33), which is important for tailored screening strategies. Understanding the facilitators and barriers to offering self-sampling is an important step for developing equitable, evidence-informed cervical cancer screening programs.

This qualitative study aimed to explore barriers and facilitators to implementation of HPV self-sampling in Canadian cervical cancer screening programs, from the provider and

policy-maker perspectives. This study was part of a larger project that included a symposium on HPV self-sampling in October 2016 (www.equitycancerscreening.ca) that brought together national and international researchers, practicing clinicians, and policy-makers.

Context

BW is a PhD student in Epidemiology at the University of Ottawa who is investigating methods to elicit women's cervical cancer screening preferences. Her research is funded by a Mitacs Accelerate fellowship in partnership with Eve Medical, a self-sampling device company based in Toronto, Ontario. In her previous work, she was part of a team to examine how a self-sampling option affected cervical cancer screening uptake in First Nations communities in Northwestern Ontario. AL is a family physician with St. Michael's Academic Family Health Team and a Scientist with the Li Ka Shing Knowledge Institute. Her research focuses on health equity in cancer screening and prevention. Recently, she has examined the acceptability of self-sampling among Muslim immigrant women. MV is an Associate Professor at Ryerson University, Faculty of Community Services, Daphne Cockwell School of Nursing, whose research interests include chronic disease prevention among women, immigrants, and refugees. MV has partnered with AL to investigate the acceptability of self-sampling for Muslim immigrant women, but previously worked with the Ministry of Health and Long Term Care in Ontario, as a health planner and program manager.

Methods

Data collection.

The team compiled a list of potential stakeholders during a preliminary environmental scan, with an aim to purposively sample individuals who contributed to cervical cancer screening practice or guideline development professionally. Potential screening participants were not included in this study because the research question focused on implementation barriers at the

policy- and practice-level. In the environmental scan, the list of potential invitees included family physicians, gynecologists, colposcopists, researchers, cyto-pathologists, representatives from Ministries of Health, scientists, epidemiologists, guideline developers, cancer screening program managers and directors, and women's health advocacy organizations from Canadian jurisdictions who are contemplating or implementing HPV self-sampling in their cervical cancer screening programs. This list included name, professional position and contact information (usually email), sex, and geographic affiliation of identified stakeholders. A priori, authors expected at least ten interview respondents, to have representation from Canadian regions with and without organized screening programs (34).

One-on-one interviews over the phone, video-conference, or in-person, were conducted by the study research trainee (BW) between May and August 2016. The interviews lasted between 30 to 60 minutes and were audio-recorded. After working through an informed consent process, demographic characteristics, including age, and professional background, were collected through structured questionnaires, administered by the interviewer following consent or completed by the respondent after the interview. Three email or phone reminders were sent to respondents who did not complete the questionnaire. A semi-structured interview guide contained four main questions: a) what are the reasons for the observed cervical cancer disparities among vulnerable populations and how could they be addressed?, b) what are your views in general on the use of alternative cervical cancer screening formats, and HPV self-sampling in particular?, c) what are the knowledge gaps in adopting HPV self-sampling into the cervical cancer screening program in your province or area?, and d) what resources/changes are required for adopting HPV self- sampling as part of screening programs?

Analysis.

Interviews were transcribed verbatim, and the data were then analyzed using an inductive directed content analysis (35), which allows relevant themes and categories of analysis to emerge from the transcribed interviews. Authors organized and managed analytic data using NVivo11 ((NVivo qualitative data analysis Software; QSR International Pty Ltd. Version 11, 2016). BW generated a coding dictionary using the overarching categories outlined in the interview guide, with new concepts and their respective definitions added to the codebook. After initial coding, MV and AL reviewed the transcripts and coding independently, and all authors came to a consensus on the “basic themes”(36) identified in the analysis. The codes were then organized using Attride-Stirling’s thematic network scheme (36), such that the “basic themes” that were inductively identified in the transcripts were organized into organizing themes and global themes. The global themes directly mapped to interview questions. “Saturation” occurred when all authors agreed that no new themes or understanding emerged from the transcript analysis (37,38). After the first ten interviews, authors discussed whether the data was saturated, while considering if representation from another jurisdiction or professional field would add further insight (39). After the tenth interview, authors decided that additional interviews would provide deeper insight into perceptions of the knowledge gaps. Interviews were scheduled in groups of three. Saturation was re-assessed after analyzing each set of three transcribed interviews.

Ethical considerations.

The protocol was approved by the Ryerson University Research Ethics Board (REB 2016-036) and approved for use in BW’s study by the Ottawa Health Science Network Research Ethics Board (Protocol #20160134-01H).

Results

Data saturation was reached (37) after nineteen interviews were conducted - three respondents did not return questionnaires. Six respondents (32%) completed questionnaires during the interview, and ten respondents (53%) decided to send their demographic survey following the interview. At this point, thirty-three interviews had been requested with stakeholders, two of whom refused an interview, citing that it was inconvenient, and nine stakeholders did not respond. Study respondents included fourteen females (74%) and five males (26%). The majority of respondents were located from Central Canada (Quebec, Ontario, Manitoba) (63.1%), followed by Western Canada (15.8%), two international jurisdictions (10.5%), and a single representative from Eastern Canada (Newfoundland, Nova Scotia, Prince Edward Island, New Brunswick) (5.3%) and Northern Canada (Yukon, North West Territories, Nunavut) (5.3%). Socio-demographic questionnaires were completed by 16 of the 19 (85%) respondents. The average age of respondents was 49 years, ranging from 38 years to 64 years. Among survey respondents, 12 (75%) identified European descent, and 4 (25%) identified other ethnic identities. The majority of respondents had training in health administration or policy (62.5%), and the remaining respondents had medical training (37.5%). Respondents worked in hospitals or community health centres (31.3%), government agencies (25%), cancer screening programs (12.5%), or other areas (e.g. research institutes) (31.3%).

Four global categories that represent our findings from the interviews map to the interview questions, as illustrated in Table 2-1. Below, we describe the organizing themes by each of the global categories.

Table 2-1. Mapping Interview Questions to Global, Organizing, and Basic Themes

Interview question	Global theme	Organizing themes	Basic themes
What are the reasons for the observed	Understanding the disparities in cervical	<i>Social determinants of health</i>	- Gender, sexuality - Rural, remote, northern

cervical cancer disparities among vulnerable populations and how could they be addressed?	screening and outcomes and the populations who experience them		- Poverty - Histories of trauma - Ethnicity/culture
		<i>Risks specific to cervical cancer</i>	- HPV vaccination status - High-risk sexual behaviour
What are your views in general on the use of alternative cervical cancer screening formats, and HPV self-sampling in particular?	Using the criteria of a successful population screening program to shape optimal strategies to reach marginalized populations	<i>Shift to HPV as primary screening test</i>	- Shift to HPV primary screening - Convenience, efficiency, effectiveness of HPV self-sampling
		<i>Criteria of a high quality, population-based screening program</i>	- Costs - Acceptability of the test - Follow-up coordination - Organized information system - Call/recall system - Quality control
		<i>Change to existing cervical screening program structure</i>	- Incorporating cervical screening with other programs - Comparison with colorectal screening - Community-based, tailored screening program
What are the knowledge gaps in adopting HPV self-sampling into the cervical cancer screening program in your province or area?	Recognizing the need for knowledge creation and translation among researchers, health professionals, and women considering screening	<i>Need for knowledge translation</i>	- Knowledge translation with women eligible for screening - Knowledge translation with providers
		<i>Knowledge creation</i>	- Local knowledge for policy-makers - Areas requiring further research
What resources/changes are required for adopting HPV self-sampling as part of screening	Addressing broader contextual factors that influence screening participation	<i>Health system characteristics</i>	- Health system design and policy-decision-making - Access to health care - Political will
		<i>New perception of cervical cancer prevention</i>	- Stigma of HPV and infectious disease relevance - Bringing men into the discussion

1. Understanding the disparities in cervical screening and outcomes and the populations who experience them.

Social determinants of health.

Respondents understood “vulnerability” through the social determinants of health lens, specifically tailoring the definition of need to their lines of work. Individuals who did not attend cervical screening or faced barriers in accessing screening were considered vulnerable or at increased risk of developing cervical cancer because of socio-demographic issues including rural/remote/Northern geography, ethnicity, education level, gender and sexuality, and income. Different social determinants were seen to be associated with different challenges to accessing screening, suggesting that approaches at the individual-, provider-, and system- level are important for reaching under- or never-screened women.

In a Toronto urban setting, a physician emphasized the unique challenges that individuals face who are marginalized because of their gender or sexuality:

We kind of look at really focused populations, people who are LGBTQ, people who are homeless and people who are new to Canada as our clients. Obviously we see other people who don't fit into those categories. So as a provider in that area working with clients who face barriers to care, that's an area of interest for me in terms of screening.... [Providers need] to be focused more on the LGBT population, I think that in general they face barriers much like the other ones to getting care and a lot of it has to do with either homophobia or transphobic providers. (Respondent 1615)

Five respondents described how the sensitive or invasive nature of a Pap test (through a pelvic exam) particularly marginalizes a subsection of the population – women who have experienced trauma or abuse. A female physician elaborated on some of the

conversations she's had with clients, reframing the communication that cervical screening (via a Pap test) can be empowering for women by taking control of their health.

I've had lots of conversations over the years of women who don't want pelvic exam and they'll talk about abuse. A lot of this very rich conversation of, ok this abuser to power, took away your agency, they took, they gave you PTSD, they created a lot of negative things for you and we talk about, should, is one of the other legacies they are going to give you is cervical cancer because you don't get screened. And we reframe it that by having a pelvic exam you're not letting that abuser continue to have negative harm on your life. (Respondent 1637)

Here, a researcher gives examples of community and personal factors influencing women's cervical cancer screening perceptions and behaviour, intersecting with health system accessibility.

From the perspective of the [First Nation communities] that we are working with, there seem to be a number of structural factors that impact health overall and impact access or lack of access to care, and then a number of factors that are also influenced by culture and social determinants and historical legacy of colonization which was a major part....For many of the women as well there are much bigger struggles that are a focus for people. So food security is a significant issue in some of the communities. Some of the other communities have challenges with substance use and that will impact everything in the family context and make it particularly difficult for women to prioritize their own care. And I think the other major issue that keeps surfacing again and again in our research is a sense around privacy and covering the body which has a cultural aspect to it. (Respondent 1604)

Different levels of risk among potential screenees.

In addition to social determinant risk factors, vulnerable individuals were also characterized by differential risk for cervical cancer, in particular HPV vaccination status. Respondents noted that HPV immunization complicates the advancement of tailored cervical screening programs because it necessitates an overhaul of existing programs. As one respondent noted, there are “too many moving parts” (Respondent 1637) to address all of these changes at once.

2. Using the criteria of a successful population screening program to shape optimal strategies to reach marginalized populations.

Suggested changes and perceptions of alternative cervical screening programs were categorized into three organizing themes: 1) Shift to HPV as primary screening test, 2) Criteria of a high quality, population-based screening program, 3) Change to existing cervical screening program structure.

Shift to HPV as primary screening test.

According to respondents, the shift to using HPV testing as a primary screening test was undoubtedly the most important change needed to population-based cervical cancer screening programs. Respondents were split as to whether provinces need to implement HPV testing in cancer screening programs before offering alternative screening strategies for vulnerable populations, but all felt that the understanding of cervical cancer etiology necessitated the shift to this new diagnostic tool.

We will not be able to implement this [self-sampling] on a larger scale without a paradigm shift to primary HPV testing. So first and foremost that needs to happen.
(Respondent 1664)

And once the testing paradigm is changed, then self-sampling is a shoe-in. It's a natural thing. (Respondent 1645)

Respondents identified challenges with implementing HPV testing in cancer screening programs. Understanding how to sustainably scale up existing knowledge of HPV primary screening strategies was considered part of the problem, though respondents perceived political and financial concerns were the biggest barriers to change:

It's interesting no matter how much evidence you have, no matter the fact it's presented in a guideline, that it's finances that blocks the options. So how do you cross that bridge? You can try making in-roads by chipping away and having smaller pieces, by bringing HPV testing into the colpo [colposcopy] clinic. You can try scaling HPV implementation. (Respondent 1611)

Criteria of a high quality, population-based screening program.

When asked to describe what an alternative cervical screening program, possibly including self-sampling as an option, might look like, most respondents addressed the principles of a successful screening program such as cost of the respective screening strategies, acceptability of the test, organization of the program and follow-up, and the quality of the testing procedure.

[Y]ou offer an alternative test [self-sampling] that costs more and requires a lot more intervention to... for compliance. If we had proven benefits in terms of... of recruiting your population. It's not going to reach the population that we need to reach, and I think people need to understand better what that population characteristics are... (Respondent 1621)

Respondents unanimously believed that fiscal concerns were the biggest barrier to change in the cervical screening programs. Respondents indicated that policy-makers were especially

focused on the “best value” strategy, reflective of the single-payer systems where respondents were based. Respondents noted that alternative cervical screening strategies like self-sampling may go against public health utilitarianism.

I guess I tend toward thinking inclusively, so I, you know, I don't get the Geoffrey Rose [perspective on health screening], do we target, you know, the general population versus the tail that just, I kind of believe that you need to do both like you need to have your general message that you're doing, but if you recognize that there is this tail that message is just not going to reach them or maybe there's a few different tails, right? That and if that's the group where more and more of the cancer burden is going to be, because they're just not getting any screening done, then I think we have a public health imperative to try to address that...I think that's only because there's limited health care dollars and...[S]o we can't all be prevention of one thing to the detriment of all others. (Respondent 1613)

Change to existing cervical screening program structure.

Beyond fundamental elements of a successful population screening program, respondents discussed ways to adapt existing cervical screening programs to meet the needs of women and to accommodate the advancing science. Four respondents mentioned that program managers should look to “model self-sampling after the offer of FOBT [faecal occult blood test]” (Respondent 1699). Many Canadian programs offer an at-home sample collection option for colorectal cancer screening, which could guide how a self-sampling option could be integrated into cervical screening programs. However, respondents noted differences between the epidemiology of cervical cancer and colorectal cancer may limit comparisons.

Many clinician respondents discussed how cervical screening was often paired with other primary care, where practitioners would discuss other health concerns, like prenatal care,

sexually transmitted infection testing, and other cancer screening. These respondents expressed concern that a self-sampling option in cervical screening would minimize these opportunities:

[T]hey [women in northern communities] get their screening because they come in for prenatal care.... But if something else brings them in, that's why we like to see it integrated. You come for your mammogram, and let's check on where your colon check is - this would be for over 50, of course - and what's happening with your cervix.

Whereas under 50, the only screened cancer is cervical cancer. (Respondent 1634)

3. Recognizing the need for knowledge creation and translation among researchers, health professionals, and women considering screening.

Respondents interpreted “knowledge gaps” to mean discrepancies in understanding among women who are eligible to participate in cervical screening, among health professionals who deliver cervical screening, among policy-makers who use research to create guidelines, and among researchers who are trying to understand how to optimize cervical screening programs.

Need for knowledge translation.

Most respondents commented on the need to communicate the change in cervical screening options with women who will face the decision to screen. The connection between HPV and cervical cancer and the self-sampling process were emphasized as important knowledge pieces, as well as the implications of collecting a self-sample (versus a provider-collected sample), the meaning of a positive results, and how to follow up the results.

Respondents also stressed the need for tailoring educational materials to different populations.

I think we have to look at the “who are not getting screened and why are they not getting screened”, and then have some kind of an effort that has to involve champions within the community of people who aren't getting screened. It doesn't help to send out a brief at a

grade 12 level of literacy, we've got to work at the bottom...and answer their questions about why are we doing this and what does it mean to them? (Respondent 1634)

The need for careful communication between cervical screening programs and potential participants is crucial to empower women to make informed decisions about their care.

We're trying to normalize, I guess, HPV infection and make the distinction between HPV and other sexually transmitted infections and you know, reducing the anxiety around having HPV because in fact, you know, administering primary HPV testing means that we're diagnosing women with a sexually transmitted infection [which has] psychological implications [for] women, so really trying to normalize HPV for people. (Respondent 1664)

Practices like self-sampling take health practices out of the clinic, which instilled some anxiety in health care providers who could feel unprepared to appropriately advise or counsel patients, given the several proposed changes to existing cervical screening practice.

From my perspective, some of the takeaways with that were that there needs to be a lot more education of care providers. We shouldn't assume that everybody out practicing is fully up to date even if they're working in the field because there hasn't been a tremendous amount of literature on self-sampling in Canada published. To my knowledge, there are no policy statements yet from SOGC [Society of Obstetricians and Gynaecologists of Canada] or any of the other bodies that might be important in terms of kind of setting the background for people's comfort with it. So I think there does have to be quite a lot of education and that's not just physicians, family physicians who might be doing it or OB-GYNs, but increasingly in the communities that we're in, nurse practitioners and others who provide that level of care do that kind of reproductive care. (Respondent 1604)

Knowledge creation.

There was little consensus among respondents of what the most important knowledge gaps were, as respondents discussed specific unknowns, such as the most appropriate device, as well as more abstract gaps, for example scaling up existing pilot projects.

I think we can look at data from other jurisdictions and say you know, and understand what it looks like. But I think that it still makes sense for us to do a made-in-Canada trial, so look at test properties, and look at the uptake, because some of the studies, the mail-in experience has not been very positive, but they're making results of that. (Respondent 1639)

[W]e think it's [primary self-sampling] is more convenient and it would also be nice to have more information on how women experience self-sampling versus office-based sampling. I mean it would be good if there was some more solid evidence on that. So it's a little bit, I think, it's not very evidence-based, but there is the cost element. (Respondent 1625)

And then I think it's a big step to how do we navigate: should this person get screening in an office versus doing it at home. How does the provider make that decision? Well the patient obviously makes the decision, so I think it's a big change management piece before even having this as a possibility.... So my understanding are these are smaller studies and maybe more rigorous data needs to be available for the province and cancer agencies to act on it. (Respondent 1615)

4. Addressing broader contextual factors that influence screening participation.

New perceptions of cervical cancer screening.

Shifting to an HPV vaccination and testing paradigm requires a shift in mentality toward cervical cancer prevention. This shift, in turn, requires a shift in public perception toward HPV

and cervical cancer, but also in health and political systems values and actions involving cervical cancer screening. Certain societal norms were challenged by respondents who talked about reducing stigma of sexually transmitted infections, including bringing men into prevention conversations, and framing cervical cancer as an infectious disease instead of as a cancer.

[A]s much as we've had this in the cancer screening program, the cancer screening mindset – we probably need to be thinking about it more as an infectious disease and how would we be approaching this as an infectious disease. And that will change a little bit of our thinking. (Respondent 1639)

Existing public health norms were also reinforced by respondents such as the idea that a “good” cervical cancer screening program is one that has high levels of participation, but not necessarily one that allows women to make choices.

Screening needs to be optimally delivered, so we will get a world in which fewer screens detect as many precancerous lesions as possible, and save people from developing cancer later on in life. Best way to do that is to do every five years or so with a provider sample in HPV testing.... You can't force [a change in program] by saying 'let's give options to women'. (Respondent 1645)

Health system characteristics.

Respondents noted that changing how women perceive cervical screening and HPV is important for reducing disparities in cervical cancer screening, but this change needs to be mirrored in the health system and have political support. Barriers to offering alternative cervical screening formats were linked with how health systems are organized and how funding decisions are made, availability and accessibility of primary care, and political will to change.

[W]e look at capacity and we look at our expectations within clinical roles and we inform some policy direction around the provision of health services, especially in rural and isolated remote areas and all those kinds of pieces, right? (Respondent 1621)

Respondents identified cancer control systems as responsible for the lack of progress: a lack of organized recall within their respective cervical screening program was compounded by competing priorities of the broader health system. Respondents were divided on whether an alternative screening modality such as self-sampling could be implemented before all pieces, including an information system, were in place.

So really leveraging off the organized program is really important right, really important and embedding self-collection in that as opposed to sort of an appendage, you know what I mean, like self-collection has to emanate from there ... you've got to embed it in the whole flow of the system and I know it feels really painful to do that right, because systems are so big but if you don't do it, you haven't actually done the women any favours. (Respondent 1637)

Respondents suggested that variation in provincial recommendations and practices were some of the greatest barriers to creating change in their local cervical screening programs.

Really what happened in our province is the pressure usually comes nationally once everyone else has started to do something, and we're about one of the last ones....

[T]hat's when things start changing for us. (Respondent 1688)

Some respondents noted that some of the remote populations struggle to access any health care providers. Additionally, access to a primary care provider that women feel they can trust was emphasized because of the uncomfortable nature of cervical cancer screening tests. Respondents described how broader issues of the health care system directly affect the participation in cervical cancer screening programs.

So in some of the communities the issue is limited access to care. There may be no doctor or nurse practitioner or consistent care provider in the community. There are trust issues often with care providers, especially where there isn't a long kind of history or connection to the community. (Respondent 1604)

Across these four global themes, we did not observe variation in responses by age or gender. Respondents with academic backgrounds were markedly more positive about self-sampling as part of a cancer screening program. Researchers, who were most familiar with the existing body of cervical cancer screening evidence, felt that there was sufficient research to offer a self-sampling modality to hard-to-reach women. Health administrator respondents tended to agree that self-sampling could benefit hard-to-reach women, particularly populations in rural/remote areas or certain cultural backgrounds, but that operationalizing the strategies requires more political support than is currently available. Researchers and health administrators tended toward a utilitarian public health perspective (e.g., "What strategy would benefit the most people?") and reflected on implementing relevant research on self-sampling in their jurisdiction so that it is "optimally delivered".

Clinicians, on the other hand, were more likely to show apprehension about a recommendation of self-sampling in a screening program, suggesting that self-sampling would be appropriate only in specific circumstances (e.g., remote communities, women who have not responded after multiple invitations). These respondents emphasized the importance of a relationship between screening participants and their health care provider and noted that cervical cancer screening is one component of an overall health care plan. Clinician respondents were more likely to draw on patient stories in their responses. Though they seemed to agree with the public health promotion perspective of researcher and administrator respondents, they spoke more about how a shift in strategy would influence their personal practice, and engagement with

women generally.

Discussion

This study is the first qualitative study to focus on policy-maker and health professional perspectives of how HPV self-sampling could fit into existing Canadian cervical screening programs to promote hard-to-reach populations' screening uptake. Seventeen respondents from across Canada and two international stakeholders discussed barriers and facilitators to alternative screening formats, particularly HPV self-sampling, to engage hard-to-reach women with cervical cancer screening, in the Canadian context. Respondents agreed that cervical cancer screening programs need to have tailored strategies to reach marginalized women with early detection opportunities, however, there was disagreement on whether the self-sampling evidence was sufficient to justify implementation in cervical cancer screening programs. More broadly, respondents noted that cervical cancer screening programs must incorporate HPV testing as primary screening before developing tailored strategies, like self-sampling, for hard-to-reach populations.

Clinicians felt that self-sampling might not address all important barriers that vulnerable populations face when accessing cervical cancer screening, particularly poor access to primary health care which is important for following up screening results. Researcher respondents were more enthusiastic about self-sampling from population coverage and cost-effectiveness perspectives, as were policy-makers, though respondents perceived undecided implementation logistics as a barrier to cervical cancer screening program shift. Respondents agreed that the idea of a self-sampling component of a cervical cancer screening program was attractive from a patient empowerment perspective, but knowledge translation strategy was important to engage with women, health care providers, and other cervical cancer screening stakeholders about a change in cervical cancer screening options. The findings from this research suggest that political

priorities and system barriers may be important challenges to implementing tailored screening, like self-sampling, to reach marginalized women.

Though respondents understood that a shift to HPV primary screening represents a major initial investment from governments, most felt that “the science is enough” to warrant the transition from cytology-based cervical cancer screening to a program based on HPV testing. Canadian randomized controlled trials (12,40), and international knowledge syntheses (11,15,27,41–43) also support this shift, which has been stressed in recent editorials (7,44,45), yet some policy-makers feel that the cytology-based programs are “good enough”, echoing the World Health Organization suggestion (46). Researcher respondents were particularly adamant that Canadian cervical cancer screening programs should shift toward primary HPV testing because the increase in sensitivity has the greatest impact on population health and health system spending, in the era of HPV vaccination (7,47). Though not mentioned by interview respondents, some research questions whether this improvement might be exaggerated in the literature because of industry sponsorship of research (48), which suggests that the shift may not be as “urgent” as some authors convey. In Canada, the Ontario government recently allocated funding for HPV testing in primary cervical cancer screening in their 2017 budget, however, the implementation strategy has yet to be disclosed.

A similar disconnect was observed in respondents’ perceptions of the self-sampling evidence base. Some respondents thought that policy-makers needed more rigorous Canadian evidence around target populations and mode of delivery of self-sampling. Most respondents acknowledged that HPV testing on provider-collected cervical samples offered superior sensitivity and specificity to self-collected vaginal samples, aligning with the findings of a 2014 meta-analysis (27). Respondents offered varying perspectives on how population-based screening programs should adapt to the different risks and benefits introduced by this alternative

modality.

Some respondents felt that self-sampling by itself would not effectively reach marginalized communities, and that education/behavioural approaches would address the true barriers that women face with early detection. The literature is similarly divided, such that some authors believe that self-sampling should be an option available to all eligible women (not just non-responders) (49,50), others suggest that self-sampling is appropriate only as a strategy to reach “non-compliant” women (26,51), and yet others argue that self-sampling is not the best way to reach under- or never-screened groups (24,52). Recent evidence syntheses suggest that most women find self-sampling preferable to clinician-collected sampling (25,53). Other systematic reviews found that self-sampling could increase cervical screening coverage in under- or never- screened women (26), though some authors observed similar uptake rates in opt-in scenarios (54).

Clinician respondents stressed the importance of maintaining face-to-face meetings between women and primary care providers, as they suggested a drawback to self-sampling was the loss of an engagement session between a clinician and a patient, which could include discussions on sexually transmitted infections and contraception. A recent study of physician billing claims in Ontario found that increasing the age of initiation for Pap testing significantly reduced the number of chlamydia cases detected in Ontario (55). Because some of the suggested self-sampling strategies affect marginalized women who are at high-risk for sexually transmitted infections, cervical screening strategies that are outside of a clinic may exacerbate other health inequities, by taking away opportunistic screening.

There are some limitations associated with this research study. Though there was diversity in stakeholder perspectives including provincial policy-makers, no representatives from provincial government funding sources agreed to be interviewed. Thus, we could not examine

some of the decision-making factors from the government side that were raised by other respondents. Also, patient perspectives were not included in this study because the research specifically focused on facilitators and barriers to implementing self-sampling in a program. Moving forward, patient advocacy groups will contribute an important voice in developing equitable, patient-centered recommendations. Finally, participants were recruited from across Canada, with two European representatives, implying different jurisdictions, organization of programs, and funding schemes. Most of the programs within Canada are similar enough to warrant comparison of barriers and facilitators of program reform, though some of the barriers that women face to accessing screening may be unique to each jurisdiction. Consistency of the findings was enhanced by discussing recognized themes between three researchers with different academic backgrounds (epidemiology, nursing, medicine). We checked the truthfulness of thematic findings with the broader project group at an in-person meeting in October 2016, which helped challenge our initial assumptions, and also provided detailed verbatim descriptions from interviews to support the themes. Detailed research methods and a priori saturation criteria also helped ensure rigour in this study (38,56).

Conclusions

According to cervical cancer screening health care providers, administrators, and researchers, screening programs must understand the marginalized population who face barriers to accessing screening. Respondents unanimously agreed that shifting to primary HPV testing is the most imminent change in cervical cancer screening programs, and likely required before tailored strategies are implemented. Alternative screening strategies, such as HPV self-sampling, may be appropriate if the implementation satisfies the criteria of successful population screening programs, including acceptability by the population, cost-effectiveness, sufficient sensitivity and specificity, and an organized call/recall program. Additionally, interview respondents

recommended that self-sampling implementation include knowledge translation opportunities for health care providers and the public to ensure appropriate approaches for specific populations and contexts. Broader health system issues, including consistent access to primary care and competing priorities for limited resources, may hinder the implementation of a self-sampling strategy in cervical cancer screening programs. Incorporating the perspectives of potential screening participants is an important next step to creating tailored cervical cancer screening strategies for hard-to-reach women.

Conflicts of interest disclosure.

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Chapter 3: “They Should Be Asking Us”: A Qualitative Decisional Needs Assessment for Women Considering Cervical Cancer Screening

The work in this chapter was accepted for publication in Global Qualitative Nursing Research (the published article is presented in Appendix B). The accepted version contains additional information about relevance to nursing. I created the study protocol, developed the interview guide, conducted and analyzed the interviews, and wrote the first draft of the manuscript and incorporated revisions. My PhD supervisors made substantial contributions to the interpretation of results and made significant editorial feedback on the manuscript. All six co-authors made significant contributions to the study design, manuscript drafts, and approved the final version of the manuscript.

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Abstract

In this study, we examine from multiple perspectives, women's shared decision-making needs when considering cervical screening options: Pap testing, in-clinic human papillomavirus (HPV) testing, self-collected HPV testing, or no screening. The Ottawa Decision Support Framework guided the development of the interview schedule. We conducted semi-structured interviews with seven screen-eligible women and five health care professionals (three health care providers and two health system managers). Women did not perceive that cervical screening involves a "decision", which limited their knowledge of options, risks and benefits. Women and health professionals emphasized how a trusted primary care provider can support women making a choice among cervical screening modalities. Having all cervical screening options recommended and funded was perceived as an important step to facilitate shared decision-making. Supporting women in making preference-based decisions in cervical cancer screening may increase screening among those who do not undergo screening regularly and decrease uptake in women who are over-screened.

High quality health care is based on person-centered and evidence-based philosophies. In particular, health decisions should reflect individuals' preferences among the best available options that are supported by research and available resources (1–4). Shared decision-making provides an opportunity to implement evidence-based practice that accounts for patient preferences, such that when multiple options are available to patients regarding a specific health decision, individuals' personal characteristics and values will influence the best choice for them (3,5). However, the use of shared decision-making in cancer screening is seldom used and such approaches that have been used are diverse (3,6).

Introduction

Traditionally, cancer screening decisions are made through a population health lens in which clinical practice guidelines have been developed, with individual health care providers promoting the recommendations therein. Individuals, on the other hand, are considered compliers if they decide to accept the offer of screening recommended in the guidelines, but usually are not presented the opportunity to make an informed decision about participation (7,8). With new technologies developed to screen for cancer, there are more cancer screening decisions leading to more options presented to individuals and health care professionals (9). Decisions about screening modality, frequency of screening, and starting and stopping age are inherently preference-sensitive, suggesting that individuals and health care professionals could use shared decision-making to identify the optimal option.

The most recent cervical cancer screening guideline recommendations at a national level in Canada (10) and provincial level in Ontario (11,12) are presented in Table 3-1. Guideline developers recommend that the longstanding Papanicolaou (Pap) primary screening test be replaced by testing for high-risk strains of human papillomavirus (HPV), of which have been found to be a causative factor in the development of cervical cancer. Compared to the Pap test, HPV testing of samples obtained by a health care professional offers increased sensitivity, a slightly decreased specificity, and different rates of false positive and false negative test results (13). HPV testing can be conducted on self-collected samples with slightly decreased sensitivity and specificity compared to samples collected by a health care professional (14). These different options are also associated with different recommended intervals between screens (e.g. 3 years for a Pap test, 5-6 years for an HPV test), and different ages to start and stop screening. Recent systematic reviews suggest that most women find self-sampling preferable to clinician-collected

sampling (15–18) because of factors related to ease, ability to conduct the testing without a health care professional or by themselves, privacy, and lack of embarrassment.

Table 3-1. Existing Cervical Cancer Screening Options in Ontario

Option	Guideline recommendation	Funding status
Cervical cytology/Pap test	Recommended by Cancer Care Ontario in interim guidelines Recommended by the Canadian Task Force on Preventive Health Care	Screening according to Cancer Care Ontario guidelines is covered by public insurance
Provider-administered HPV testing	Recommended by Cancer Care Ontario	2017 provincial budget allocated funding toward HPV testing, but not implemented
Self-collected HPV testing	Recognized as an option for HPV testing, no explicit mention in guidelines	No funding; can be privately purchased online
No screening	Not applicable	Not applicable

These three screening modalities (e.g., Pap testing, HPV testing in-clinic, self-collected HPV testing) and the option not to participate in screening are associated with distinct trade-offs between the benefits (e.g. ability to predict cervical cancer risk) and harms (e.g., false positives and false negatives). As these options differ in convenience, privacy, and invasiveness, individuals should actively participate in discussions with their providers to decide their best option. HPV testing modalities also differ from other cancer screening options because of implications of testing for a sexually transmitted virus; stigma surrounding this type of testing may affect how individuals make trade-offs in these decisions. However, provincial and national guidelines have not addressed individuals' values or preferences among these cervical screening options (19), and have not provided explicit guidance for eliciting preferences. In the shared decision-making literature, there are no decision support tools relating to these options (5,9,20).

Evidence-based guidelines use a utilitarian approach to creating population-based recommendations (21). Health care professionals (HCPs) who use these guidelines often feel obligated to promote the recommendations, despite individuals' preferences for alternative options (22–24). The divide between population and individual perspectives may be augmented

for cancer screening programs or policies because of the requirements of a mass screening program (i.e. a defined, long preclinical phase, need for accurate and reliable tests, demonstrable effective early treatment) (25). Shared decision-making processes can reconcile individual and population perspectives for cancer screening by addressing the individual acceptability of tests in the context of HCP values and evidence-informed recommendations (4,26). Therefore, a shared decision-making framework can guide processes to elicit preferences in cancer screening, aligning with person-centered and evidence-informed practice (3,27). To support preference elicitation and shared decision-making in cervical cancer screening, we need to know how stakeholders, including women eligible for screening and HCPs, perceive the decision.

Theoretical Framework

The Ottawa Decision Support Framework (ODSF) (28,29) is a decision support framework incorporating expectancy value, social support, cognitive, and psychology theories (30–37). This framework was validated in previous research involving individuals and families making health and social decisions (38–40). The goal of shared decision-making is to promote a well-rounded decision process rather than a particular action (9), therefore assessment of shared decision-making processes would consider decision-relevant outcomes. Shared decision-making processes are separated into three constructs: decision quality, decision impact, and decision action. Good decision support would assure that individuals feel informed and that they can clearly articulate the characteristics that are most important in making a decision (i.e. decision quality) (28–31,41,42). Additionally, individuals would feel that they are satisfied with the decision process and do not experience decisional conflict (i.e. decision impact) (28,29,41,42). There is no ideal cancer screening action or intended behaviour; instead outcomes focus on making decisions consistent with individuals' preferences and values (5,9,43). At a population-level, a 2017 review found that decision support increased under-use and decreased over-use of

some services, for example prostate cancer screening (5). Patients who are active in decision-making are more likely to have better health outcomes and care experiences, at a potentially lower cost to the system (3,5,44).

According to the ODSF (42), values refer to individuals' informed attitudes about relative desirability or undesirability of the characteristics of a decision's options. Instead of considering values as the underlying ethical, religious, social, or political principles that guide how individuals judge different situations (45), values are associated with how individuals assess the specific attributes of each option. Preferences are constructed from individuals' informed attitudes toward the detailed characteristics of each option, in which the preference is the most-favoured option.

Decision support interventions can address decision quality through five steps: i) Recognition that a decision needs to be made; ii) Understanding information related to the decision; iii) Clarification of values and identification of preferences according to these values; iv) Consideration of resources, including social influences, that affect the decision; and v) Formation of an action plan (9,42).

Research Objective

The objective of this study was to examine, from multiple perspectives, women's needs for shared decision-making when considering available cervical screening options: Pap testing, in-clinic human papillomavirus (HPV) testing, self-collected HPV testing, or no screening. Understanding women's knowledge needs and the values that shape their cervical screening decisions is important for shared decision-making. Because shared decision-making involves bilateral flow of information, nurses, physicians, and professionals (HCPs) who develop or administer cervical cancer screening guidelines can offer alternative perspectives on how women

make decisions in cervical screening, sources of decisional conflict for women, and methods for implementing decision support in practice.

Methods

Using a qualitative, semi-structured interview approach, we interviewed multiple stakeholders, including women and HCPs to determine what women need to identify their preferences among cervical cancer screening modalities, based on a shared decision-making framework. This study was approved by the Ottawa Health Science Network Research Ethics Board (Protocol #20160134-01H). All respondents provided informed consent at the beginning of the interviews.

Participants.

Participants were eligible if they were 18 years or older, had knowledge of the Ontario health care system (as a patient or HCP), and could participate in an English-speaking interview. Study participants included two groups of stakeholders. The first group comprised women who would be eligible to consider participation in the Ontario cervical screening program; previous experience with screening was not required. Screening participants were selected purposively based on age categories (e.g., <35, 35-54, 55+) and ethnicity (e.g., European descent, First Nations/Inuit/Metis, immigrant or another minority population). The second group comprised of health care professionals (HCPs) including primary care providers who provide cervical cancer screening to their patient population (i.e. providers) and non-clinical professionals who develop and administer cervical cancer screening recommendations (i.e. managers). These individuals were purposively selected based on their role within the cervical cancer screening program (e.g., through local policy/management, through provincial guidelines and policy, through direct clinical care). These professional groups play substantially different roles in how cervical cancer screening is delivered but share a common role as “health experts”. In this context, HCPs were

familiar with cervical cancer etiology, population-based cancer screening and guidelines, and the organization of the Ontario health system.

HCPs were recruited by contacting clinics in the Champlain Local Health Integration Network region in Ontario, Canada, and relevant cancer screening agencies. Clinics were purposively based on geographic area (urban and rural) and targeted communities with high proportions of Indigenous or Francophone clients and/or vulnerable populations, including newcomers to Canada, those affected by mental illness, or low-income residents. The research team worked with the clinics and HCPs to identify screen-eligible women who met the aforementioned age and ethnicity criteria. Researcher contact information was also shared with community groups and patient advocacy groups to recruit screen-eligible women. Recruitment continued until data saturation was reached for all guiding categories (further details in Analysis section).

In-person or phone/video interviews were arranged between July 2016 and November 2016. At the beginning of each interview, the project was explained to each respondent, and respondents' questions were answered before obtaining written informed consent. Contact cards were provided to the respondents if they wished to follow up with the interviewer after the interview. Interviews lasted between 30 and 60 minutes and were conducted by Brianne Wood (BW), a researcher trained in the ODSF who was familiar with cervical screening guidelines.

Data collection.

Semi-structured interview guides were developed separately for women and HCPs based on the ODSF and its Population Needs Assessment guide (46) (see Table 3-2 for an overview of the interview guide). The interview guides contained open- and close- ended questions based on these steps, eliciting from screen-eligible women and HCPs what cervical screening decisions women face, what information women may need to make decisions, values that affect their

decision, and the support and resources women may need to make a screening decision among cervical screening modalities. Demographic data on each participant were also collected during the interview. Respondents were asked if they wanted to receive communication about the project, following the interview; those who expressed interest received an email with a high-level summary of the analysis findings. Feedback or comment on the interpretation of the data was invited.

Table 3-2. Overview of Needs Assessment

ODSF Steps to shared decision-making	Guiding questions
1. Recognize the decision to be made	What do you perceive as important decisions for you/women in cervical cancer screening? - Have you considered options in cervical cancer screening modalities? How might you/women feel making decisions about cervical screening? What difficulties might you/they have? What will make the decision difficult for you/women? - Knowledge - Expectations - Values clarity - Support - Resources
2. Information comprehension	What do you see as the main options? What do you see as the main benefits and risks of these options?
3. Values clarification and preference-elicitation	What qualities about these screening options are most important to you? [Potential screening participants only] For example, convenience, process of test, length of time to get results, costs, invasiveness, access, follow-up procedures, sensitivity/specificity
4. Identifying necessary social support and material resources	How will you go about making this decision/support women in making this decision? What will help/hurt the decision-making process? What will help overcome some of these issues? What are the barriers and facilitators to offering options in cervical screening? What options are available to support women in making decisions about cervical cancer screening? What might be the “best” option for a cervical screening decision tool (e.g., Internet, phone app, etc)?

5. Forming an action plan	What do you perceive as implementation facilitators and barriers? [HCPs only] What personal, clinical characteristics are important in decision-making?

Analysis.

The interviewer took field notes during each interview and composed interview summaries afterward, allowing for reflexive analysis (47). All interviews were audio-recorded using a tablet with an external microphone. Digital recordings were transcribed using an intelligent verbatim process, which includes light editing to correct sentences and grammar to improve overall readability. Before interviews were conducted, a coding dictionary was created using the ODSF overarching steps (Table 3-2) as guiding categories (46), and the codebook fields outlined by Guest et al (Guest, Bunce, & Johnson, 2006, p. 64). In particular, each code definition contained a code overview, a full definition, a “when to use” section, a “when not to use” section, and an example of the code. Transcripts were analyzed chronologically during data collection (NVivo qualitative data analysis Software; QSR International Pty Ltd. Version 11, 2016), using deductive content analysis (49,50) with respondents as the unit of analysis. During the analysis, additional codes were developed and added to the codebook, and current code definitions were updated to reflect the nuances presented in the interviews (51). Previous coding was reanalyzed to assess consistency with the new code definition, and a new code was added if necessary. The iterative approach was important to help determine when saturation was reached. A categorization matrix (52) was created to depict which categories were communicated by stakeholder groups, and to indicate when code definitions were added or changed to document saturation. Another member of the study team (Virginia Russell (VR)) – an expert in qualitative research - reviewed the codebook to assure clarity of the definitions. VR independently analyzed each transcript to ensure complete and appropriate coding of important themes. Coded segments

were assessed and refined further to concisely represent the data. Discrepancies in how segments were interpreted were resolved through dialogue between BW and VR.

In this study, we sought to triangulate different stakeholder perspectives (53,54) to gain an in-depth understanding of women's needs to make an informed decision among cervical screening modalities. In particular, we elicited multiple perspectives on women's decisional needs for cervical cancer screening modalities, to understand how women's decisional needs are influenced by individual, provider, and health system factors. We looked for convergence in how ODSF steps were interpreted by stakeholders, to identify opportunities to support women's decisional needs. Any diverging interpretations were explored in further interviews. The sample size was considered adequate once a diverse sample of stakeholders had been recruited (48), and study authors considered that no further interviews would generate insight to help address the research objective (50,55). A category was considered saturated when the code meaning was determined stable for each of the stakeholder groups (50).

Results

Participant characteristics.

In total, seven women eligible for screening and five HCPs were interviewed – three providers and two managers. Of seven screen-eligible respondents, women's ages ranged between 24 and 63 years. Two Indigenous respondents participated, as well as one Francophone respondent, and two women who identify as immigrant or first-generation Canadian. Most screen-eligible respondents had previously had a Pap test. Among HCPs, two nurses and a general practitioner represented clinical providers, and a director of a health centre and a provincial cancer agency director were interviewed as managers. Among the HCPs, one respondent identified as male.

Three screen-eligible respondents who were recruited from the same organization (an Indigenous health organization) were interviewed in a group setting, when they identified at their interview session that they preferred a group interview instead of separate one-on-one interviews. As the interview was the first time that the respondents had met in-person with the interviewer, they communicated that they felt more comfortable discussing some of these sensitive health topics together. One respondent (Woman 1) recruited the two other respondents (Woman 2, Woman 3) and organized the interview space, but did not dominate the interview conversation. There was a large focus on HPV and cervical cancer information in this session, with the discussion exploring opportunities for them to choose among cervical cancer screening options.

The main themes are organized below by the ODSF steps to shared decision-making.

1. Recognition that a decision needs to be made.

Most screen-eligible women had not considered participation (or non-participation) in cervical screening as a decision. Instead, a Pap test was perceived as an obligation for women of a certain age, thus women would not actively choose to avoid a Pap test. Rather, women might defer cervical screening, or they might want to attend screening but face barriers accessing the health care system. When asked about decisions that they had faced with cervical screening, most respondents expressed confusion; they had not considered “no screening” a viable option.

It was just sort of best practice or recommended by the doctor to have it done, so I really didn't give it much thought and just had the procedure. (Woman 6)

I thought it was maybe about the right time to get a Pap test done. So, that was just my reason—like my age. (Woman 7)

Both groups of stakeholder respondents identified that women may prefer a female health care provider, though the choice among options still was not perceived as a decision.

In 2012, Ontario cervical screening guideline extended intervals between screens from annual Pap tests to one every three years, and some respondents - including female HCPs – preferred more frequent screens than recommended. Despite this preference, respondents felt they could not act on their preferred options because they were limited by the provincial guidelines and funding.

OHIP [Ontario Health Insurance Plan] doesn't [sic] cover Pap smears once every 3 years now. For women, not every 6 months . . . that's the law now. (Woman 3)

When it's due, I want it done. And I prefer it actually be done more often than [chuckles] is recommended now, but I'm okay with that. (HCP 3)

Manager HCPs felt strongly that women's options in cervical screening should be governed by guidelines developed by cancer agencies, which was echoed by provider HCPs who currently decide the types of tests to offer women for cervical screening.

So it is confusing to make the choice once there is more of a choice because there's not too much of a choice yet, right? ... Central and national guidelines [are important] because they give individuals as well as primary care providers some framework around the decisions that they make. (HCP 5)

Despite their perceived lack of choice, screen-eligible women wanted to choose between options. Though they wanted to know more about these options, they also wanted to engage with their primary care provider to make a decision. Interestingly, most of the potential screening participants focused on the self-collection option of the test, paying less attention to new diagnostic processes, suggesting that having options that differ in sample collection process as well as diagnostic performance might make decision-making more complex.

I like the idea of having multiple options, but I think if you say that, for example, if I decided I have my Pap test but I still decided to do the HPV test and I realized that it was

positive, then you said well sometimes you can have a virus but it's not a big deal because it's going to go away on its own. ... So I guess it has two sides, like it's good because that would be something convenient. But at the same time, we are not doctors. We don't know those things so maybe it would create more people going to the clinic that we really need. (Woman 5)

2. Understanding information related to the decision.

Most respondents did not know that several options were available for cervical screening in Ontario, or that they faced a decision between in-clinic Pap testing, in-clinic HPV testing, self-collected HPV testing, no screening. A portion of each interview involved communicating what is known about HPV, cervical cancer, and current initiatives for prevention, detection, and treatment of cervical cancer to screen-eligible respondents. Three of the screen-eligible respondents had not heard of HPV previously, though they had previously participated in cervical screening. Except for one woman who had learned about self-collection through work, six screen-eligible respondents had not realized a self-collection option existed.

I think the questions I would ask and stuff, like, oh, how does it work and what's the process it's going to be like? Does it hurt? Especially like for myself when you've never got screened before. I'll probably feel like very anxious and like scared of doing it myself for the first time. (Woman 4)

HCPs were more aware of the connection between HPV and cervical cancer, although three HCPs wanted further information about new cervical screening options, suggesting that more work in knowledge translation with professionals in the health field is needed. A provider argued that these options were hypothetical at this point because HPV testing is not yet funded, so communication opportunities might be premature. As a result, HCPs were concerned about communicating options that were not universally available:

Because so far there isn't that option [for self-collected HPV testing], so there isn't any discussion like that. It's just me explaining you could get cervical cancer. It can be detected before it becomes cancer. This is how it's done and it's recommended to start at age 21. We do it every three years. So I explain that and then they say okay. And they go for it. (HCP 2)

I would imagine there would be some of our clients, patients, who would ask about it [the HPV test] because they would have heard about it, known about it...you know, you really would have to ask them [the clinicians who administer the Pap tests] if they raise it [the options for cervical cancer screening] with women who they know could not pay for the test. (HCP 1)

Though options other than Pap tests are accessible through private payments, clinicians may not engage in knowledge exchange with their clients because the options aren't addressed in the practice guidelines, nor are they funded by the public system. HCPs conveyed a sense of paternalism - that women's decision-making should be contained within the evidence-based guidelines- which coincided with a desire for women to have the autonomy to make informed decisions. Some HCP respondents recognized this contradiction throughout the interview and struggled with balancing autonomy of women versus the population-based goals of a screening program.

I'm a very big believer in clients making choices.... And if it's a bad choice, it's still their choice and they have all the right in the world to make that choice. (HCP 4)

I think most screening programs, not just in Canada but definitely in the U.K. [United Kingdom] and the Nordic countries have moved towards a more balanced message. I do struggle a little bit with that because okay, so I want women to have a balanced message about the benefits and harms of cervical cancer screening, right? (HCP 5)

Both screen-eligible women and HCPs identified that all stakeholders (i.e., women, providers, managers) need more information about the cervical screening modalities, the processes, the risks, and benefits of these options, and how these options appeal to individual women. Women wanted more information so that they could weigh the cervical screening options and consider what aspects of screening are most important to them:

I guess like if you're going to a clinic and an expert is helping you screen it, then I think the information you would want is, like, oh what are you going to do and, like, how does it work in general and the process and like how long or how often does it come back to get screened? Whereas for like, if you're doing a self-test, for me I would want to know, like, more information, I would probably have to like want to reassure myself that I'm doing things correctly. (Woman 4)

3. Clarification of values and identification of preferences according to these values.

Because in Canada we balance, an individual's choice with what's good for the population and what we can afford as a society, right? (HCP 5)

Attributes that respondents thought would affect women's screening decision were organized into three main categories: characteristics of the sample collection process, characteristics of result follow-up and communication, and importance of cervical cancer and early detection. As mentioned previously, women and HCPs tended to focus on the decision between in-clinic cervical screening versus self-collected (e.g., "at-home") cervical screening versus no screening, expressing less concern about a change in diagnostic procedure. (i.e., cytology vs HPV testing).

Screen-eligible women were primarily concerned with sample collection and the convenience among the different modalities. Women's comfort with the sample collection

process often related to prior traumatic or painful screening experiences, and their confidence to collect a sample themselves.

But they're kind of similar, I think the questions I would ask and stuff, like, oh, how does it work and what's the process it's going to be like? Does it hurt? Especially like for myself when you've never got screened before. I'll probably feel like very anxious and like scared of doing it myself for the first time. (Woman 4)

HCPs expressed more concern about the socioeconomic impact of offering cervical screening modalities that require women to pay out-of-pocket and the implications of varying laboratory quality of the screening tests.

So you have to understand that the clientele that we have here are low income is over 50 per cent. We have some low income and our mandate is to serve low income seniors and new immigrants. So it is a huge consideration for them not going forward with that. And a lot of females with low income don't mind coming in for Paps and testing. I'm generalizing, but I don't want to generalize in any way. So it's not about the testing itself and feeling uncomfortable with doing it. It's just about the cost. (HCP 4)

Women and HCPs expressed concern about communication and follow-up of results. Both groups of stakeholders were concerned about privacy when communicating results, noting that current Pap test results are shared via letters from a central provincial agency. The lack of personalization of these letters and potential for ambiguity was worrisome for providers and women, though not mentioned by managers. In the following excerpt, a provider communicates frustration with the current result communication system in Ontario, suggesting that the letters to participants do not clearly articulate what screening results mean, and might cause distress:

Like they [women eligible for screening] know what they would want if they think it through is for their doctor to tell them and to not have to wait for the doctor to say it is

not normal and then tell them what that means and not get this message that it's not normal. Then have to wait for whatever length of time to find out what it means and in the meantime being scared to death of the implications. (HCP 2)

The change in approach to detection of cancer and precursor lesions- from looking for cellular abnormalities with Pap testing to detecting the presence of a high-risk sexually transmitted virus with HPV testing - was considered important because of stigma associated with sexually transmitted infections. Women and HCPs suggested that methods to follow up positive results would require careful communication with women. Unsurprisingly, women were concerned with the privacy and confidentiality of how results, including HPV positivity, would be communicated. The following interaction occurred in the first interview with three women, who had recently learned about the connection between HPV and cervical cancer, and how cancer screening would involve testing for a sexually transmitted infection.

Confidence is the main thing. Because there's a stigma related [to a] sexually transmitted... (Woman 3)

Oh you whore or whatever. (Woman 1)

A moral responsibility to be healthy and participate in healthful behaviours influenced how women contemplated cervical cancer screening options, which was reiterated by HCP and screen-eligible women respondents. The inherent “good” value of cancer screening was emphasized as many respondents described cancer screening participation as healthful. Most respondents ignored the “no screening” option because it was implied that when someone was appropriately informed, they would choose to participate in cervical screening. This conviction that any cervical screening would necessarily improve one's health was also related to certain risk expectations of cervical cancer; women were more enthusiastic about cancer screening when they perceived themselves at a higher risk of developing cancer.

But if it [other option's diagnostic performance] was different or the sensitivity or the results were different between the options, obviously it would be my first deciding factor.

(Woman 7)

A manager HCP agreed that public health enthusiasm for cancer screening could be responsible for an automatic acceptance of screening:

I imagine that's how most women make their decisions and I think a lot of it is probably not evidence based and not balanced and not good quality information. And screening programs used to be part of that because we were like you should be screened no matter what. (HCP 5)

While women and HCPs seemed to identify similar attributes that could influence cervical screening decisions among modalities, they judge them differently. Women focused on decision-making at an individual level and were concerned about personal risks and benefits, while HCPs often spoke of patient populations and collective risks and benefits, reinforcing the population perspective of cancer screening programs.

4. Consideration of resources, including social influences, that affect the decision.

Respondents indicated that primary care providers were most likely to influence women in their decisions among cervical screening modalities. Notably, it seems that screen-eligible respondents who had a consistent primary care provider were more likely to turn to a clinician for input into their decisions. Screen-eligible respondents explained that they rarely involved friends or family in cervical screening decisions, although one HCP noted that women from certain religious groups might prefer to involve their husbands because of the invasive nature of the sample collection:

Anybody's welcome to stay as long as the client wants them to. So we found that East Indian women very uncomfortable with it and need almost that family support around for them. (HCP 4)

Women and HCPs recognized the counselling that primary care providers can offer women in preventive health care. Screen-eligible respondents emphasized the importance of access to a provider whom they trust, explaining that would influence how they or others choose to participate in screening.

I was very lucky because . . . it took no time to have a family doctor first of all. Because one thing I find is that I wouldn't want just any doctor to do the Pap test. (Woman 5)

Screen-eligible women wanted to feel comfortable with the person who is administering the invasive cervical cancer screening; some women mentioned that a lack of familiarity with a provider made self-sampling more appealing. HCPs agreed that cervical cancer screening with the Pap test requires a lot of trust because of the pelvic exam but added that providers help their patients feel supported if abnormalities are found.

More broadly, screen-eligible women "trusted" their primary care provider to offer them any screening tests or preventive health care for which they are eligible. Provider HCPs also noted that their patients will frequently accept cervical cancer screening when they are introduced to it opportunistically. A manager HCP identified that women who are more likely to participate in preventive health care, such as cervical screening, will already have a trusting relationship with a primary care provider; conversely, women who are not attached to a primary care provider are less likely to participate in screening.

[I]t's almost like a bit of a catch-22 because if you're a woman and you're in a position to have that somewhat personal conversation with someone, you're also in a position to be more likely to be screened. (HCP 5)

All respondents communicated that women need some external support to make a decision among screening modalities, with suggestions ranging from educational campaigns to public funding of all options. Some respondents suggested that tailored approaches are needed to reach women from different populations. For example, two Indigenous respondents indicated that they would prefer learning about cervical screening through an online source, and they feel most comfortable accessing health care that is tailored to Indigenous women. Another woman indicated that ‘clear instructions in both languages [French and English]’ were important for communicating the cervical screening options. A manager HCP seconded this bilingual preference and emphasized the importance of structuring support for immigrant and refugee women, as well as women who have experienced sexual trauma.

Most women considering screening felt confident that they could choose their preferred screening modality if they received more information, in the absence of primary care provider input. While discussion groups (e.g., at a public health unit) were considered a satisfactory option by a few women, most women preferred to learn about the options independently through materials they could find online or in a public space, or through a one-on-one conversation with a health care provider. Several of the women mentioned wanting “several sources” of information before deciding on a screening modality:

I would say the website, kind of getting different information from different sources, only because the majority of us have access to like internet. ... I also sometimes look at different experiences... I used to go for different sources because you just want to check the facts and one website might cover something that you never thought about before.

(Woman 4)

5. Formation of an action plan.

They [health care professionals] should be asking us. (Woman 2)

Overall, respondents acknowledged women do not perceive that undergoing cervical cancer screening involves a decision, which women attributed to a lack of awareness and HCPs attributed to the context of a population-based screening program. As new modalities are implemented, screen-eligible women and HCPs identified provider-patient encounters as an optimal environment to have a conversation about screening options. Subsequently, respondents identified that women and primary care providers need support from the cancer screening program to ensure that they have sufficient information and resources to engage in these conversations. HCPs looked to clinical practice guidelines and funding organizations to catalyze this change:

What's available, if we don't have the funding then they [policy-makers] can't recommend it. (HCP 2)

Most respondents emphasized that support from cancer agencies and funding bodies was important to make cervical cancer screening options accessible. Having all options covered under the public insurance plan was important to implicitly establish that the screening options and entire program were trustworthy. For screen-eligible women, public funding would reinforce the notion that governments aren't solely concerned about saving money and that all options were "good". For HCPs, public funding of all options symbolized confidence in the scientific evidence behind these options. Having multiple options available within the organized cancer screening program would allow screen-eligible women to make preference-based decisions in cervical cancer screening, while meeting the objectives of a population-based program.

[I]f it's [the screening modality] well explained, if the price is good... If it's something that's accessible anywhere that anybody can buy anywhere, why not? (Woman 5)

So I think by doing all of these things through an organized screening program and having that structure, you have the opportunity to ensure that women are not over

screened or at least help to make sure women aren't over screened and to try to target unscreened women. (HCP 5)

Discussion

In this qualitative interview study, we observed that screen-eligible women and HCPs do not perceive participation in cervical cancer screening as a health decision, but rather, it is an opportunity or an obligation. Most respondents were reluctant to identify “no screening” as an acceptable option. Recognizing that a decision among cervical screening modalities exists for women is crucial for high quality shared decision-making, according to the Ottawa Decision Support Framework (28,29). Other decisional needs identified in this study include women's knowledge of cervical screening options and associated risks and benefits, and resources to support them in decision-making, most notably a trusting relationship with a primary care provider.

Cancer screening decisions are thought to be inherently preference-sensitive (5,9), though cervical cancer screening has been under-represented in shared decision-making literature (5,9,20). In Canada, Pap testing had long been the only screening option available for early detection of cervical abnormalities and cervical cancer. Consistent with the health messaging by media campaigns, Pap testing might have seemed like the correct option. Even now, as the Pap test is the only publicly funded option among available modalities aside from not screening, respondents asserted that informed women would choose to participate in screening. This public and professional enthusiasm for cancer screening has been discussed previously (56,57) and likely contributes to the perceived imperative to participate in cervical cancer screening.

With regard to eliciting preferences among Pap testing, in-clinic HPV testing, self-collected HPV testing versus no screening, most women and some HCPs were unaware of any type of HPV testing, e.g., modalities that are not funded by the Ontario health system. However,

some organizations, like the United States Preventive Services Task Force (58), already recommend that women make a choice among modalities, though implementation and decision support for individuals is unclear. This study's findings are timely because it seems that awareness and knowledge of the cervical screening options stand out as women's primary decisional needs in cervical screening. Although values played an important role in how women make the decision (e.g., how women make trade-offs between the costs of different options, the convenience of the procedures), screen-eligible women felt confident that they could make the decision if they had more information. However, it is possible that because women have not faced this type of decision-making in cervical cancer screening, that they may miss some of the nuances around the different trade-offs (45). Value clarification methods – strategies to help individuals assess the desirability of attributes of different options - will be important help women identify which option they prefer among cervical screening modalities (42,45).

HCPs worried that multiple options may cause worry or anxiety in women deciding among cervical screening modalities. The HCP concern of decisional conflict may reflect worry that women may not participate with medicine's preferred screening modality (i.e. making the "wrong" choice by selecting a less sensitive option). The belief that there is an ultimate "good" decision suggests that HCPs may have values that differ from their patients. Respondents also believed that if women recognized the decision and were informed of the options, they would choose the option that corresponds with population health objectives. However, it may be challenging for women to act on their preferred option for cervical screening if it deviates from their clinician's preferred option (59). However, providers also face organizational barriers such as performance quality metrics that might impede shared decision-making (6,60).

Some authors have discussed perceived challenges of autonomous decision-making within the context of a population-based cancer screening program (6,23,25,61). In the cervical

screening situation, many HCPs were concerned about offering modalities to patients that were considered inferior to the modalities recommended by clinical practice guidelines. Findings from this study demonstrated an interesting paradox that HCPs and individuals trusted cancer agencies and networks to know the “best” options for cervical screening for individuals, yet, some respondents also expected system-level institutions to prioritize financial or other system prerogatives above an individual’s wellbeing. As test acceptability is a crucial component of a successful screening program (25,62,63), women want confirmation from HCP stakeholders that the options are valid.

One respondent – a HCP – vocalized concern for potential harms with over-diagnosis and overtreatment when women decide to participate in cancer screening, possibly related to their contribution to guideline development. As knowledge of the decision and its options is a key decisional need identified in the interviews, a balanced presentation of risks and benefits for each option is necessary. Two previous systematic review demonstrated that individuals and health professionals tend to overestimate the benefits and underestimate the harms associated with screening (64,65), suggesting that tools to elicit preferences or support shared decision-making need to carefully communicate that cervical screening can directly benefit a small number of women through early detection, and potentially introduce harmful consequences through over-diagnosis and overtreatment (19,25,57). Not only are women’s informed preferences among cervical screening modalities (and other decisions) integral to patient-centered care, but they may lead to more appropriate use of healthcare resources. The 2017 Cochrane review of decision aids (5) found that some options may be under-used and others over-used, which was usually related to inadequate information comprehension. Hence, supporting women in making preference-based decisions in cervical cancer screening may increase screening among those who do not undergo screening regularly, and decrease uptake in women who are over-screened, which is

consistent with utilitarian public health campaigns. This research highlights an opportunity for health professionals to empower women to make autonomous cervical cancer screening decisions.

This study is the first to consider women's decisional needs among multiple cervical screening modalities – Pap testing, in-clinic HPV testing, self-collected HPV testing, and no screening - from the perspectives of women who are eligible for screening as well as health care professionals. However, it has some limitations: some respondents perceived this decision among screening modalities as a hypothetical; sometimes, the way individuals perceive and act in a hypothetical decision may vary from that in real world circumstances (66,67). However, because the decision to screen for cervical cancer is ongoing and the modalities are available, most respondents perceived the decision as real. We determined that twelve respondents provided sufficient data to identify decisional needs using the ODSF, and we will integrate this analysis with the findings of a systematic review to further validate the main categories. Though many studies tend to have larger number of interviews, many do not explain why or how they reached saturation at that sample size (48,50,68). Inquiring about women's decisional needs from women and HCPs, using two independent coders to analyze the ODSF categories, and a clear description of the data collection and analysis methods (55,69) helped to ensure rigour in this project. These results may not be transferable to contexts outside of Ontario, though these findings might apply to other publicly funded healthcare systems where quality metrics and remuneration are determined by adherence to clinical practice guidelines. Additionally, similar decisional needs were identified across a sample of women with diverse socio-demographic backgrounds which suggests that most women may benefit from some form of decision support.

Conclusions

Women and health care professionals did not perceive cervical cancer screening participation as a decision for women to make. When considering available modalities – Pap testing, in-clinic HPV testing, self-collected HPV testing, and no screening – women and health professionals wanted to know more information about these options. Women wanted to make a choice among available options, though health professionals were hesitant to engage in shared decision-making because some of the options are not funded by the public-payer system, and some options might not be consistent with population-based screening goals. Women and health professionals emphasized the importance of a trusting relationship between women considering screening and their primary care providers for shared decision-making. Subsequently, adequate access to primary health care may play a significant role in women’s decision-making process. A tool that addresses some of these decisional needs might support women and their health care providers in cervical screening encounters, which is important for person-centered, evidence-based care. Supporting women in making preference-based decisions in cervical cancer screening might increase screening among those who do not undergo screening regularly, and decrease uptake in women who are over-screened, which is consistent with utilitarian public health campaigns.

Acronyms Used

HCP – health care professional

HPV – human papillomavirus

ODSF – Ottawa Decision Support Framework

Pap – Papanicolaou

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Author contributions.

BW, JL, and IDG conceptualized the study. BW conducted the interviews, analyzed the data, and wrote the first draft of the manuscript. VLR helped to analyze the data and contributed to the first draft of the manuscript. All authors contributed to the interpretation of the data and provided editorial feedback on drafts of the manuscript. All authors read and approved the final manuscript.

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Supplementary materials.

For further information on the data and interpretation of this study, please contact the first author of this study.

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Chapter 4: What are Women's Preferences among Cervical Cancer Screening Options: A Narrative Synthesis of Quantitative, Qualitative, and Mixed-Methods Studies

This chapter will be submitted to BMC Public Health. I created the study protocol, developed the search strategy, conducted the search of the literature, conducted the screening and data extraction of relevant articles, analyzed and interpreted the data, and wrote the first draft of the manuscript and incorporated suggested revisions. The second and third authors contributed to the screening, data extraction, analysis and interpretation of the relevant articles, and made important contributions to revisions of the manuscript. All authors made significant contributions to the study design and/or execution, drafts of the manuscript, and approved the final version of the manuscript.

Abstract

Background.

Guideline developers should consider individuals' preferences among cancer screening options as part of a person-centered approach. Preferences can be elicited at an individual or population level, through interactive processes including interviews or non-interactive measurements like questionnaires. This narrative synthesis describes women's informed, values-based preferences for primary cervical cancer screening, across a variety of study contexts and preference-elicitation approaches.

Methods.

MEDLINE, Embase, PsycINFO, CINAHL, and Cochrane Library were searched from inception to present for qualitative, quantitative, or mixed methods approaches to describing or measuring women's cervical cancer screening preferences. Canadian grey literature sources were also searched for relevant articles. Articles were included if the preference-elicitation approach met the criteria from the Ottawa Decision Support Framework. Methodological quality was assessed using the Mixed Methods Appraisal Tool. Options for the cervical cancer screening decision were extracted in a pre-tested data extraction form, and preferences were described narratively in the context of the study.

Results.

Twenty-eight articles reporting seventeen quantitative, ten qualitative and one mixed methods study were included. Included articles investigated women's preferences regarding modality or location of cervical cancer screening (n=12), participation in cervical cancer screening (n=8), stated preferences among hypothetical options (n=6), screening test type (n=3),

willingness to pay (n=2), and device (n=2). Overall, women's preferences varied by geography, socio-demographic characteristics, and current screening contexts.

Conclusion.

Because of the heterogeneity in the methodological and reporting quality of included studies, it is challenging to determine whether these differences are due to study context or the type of approach used to elicit preferences. This study is the first systematic review to examine women's preferences for cervical cancer screening across qualitative and quantitative study designs, contexts, and decisions. A point-of-care approach to preference-elicitation (e.g., during encounters between women and providers) might capture the dynamic nature of women's cervical screening preferences.

Background

Person-centered, evidence-informed health decisions and processes incorporate individuals' values and preferences, while also considering external factors including research evidence, clinical expertise, and context (1-4). Policy-makers and health care providers recognize the significance of person-centered decision-making, but stakeholders struggle with operationalizing this practice (2,5). Making decisions based on the best available research evidence and personal preferences will encourage individual autonomy in health decision-making and it may improve appropriate use of health care services, reduce costs, and increase participant's satisfaction with the decision (3,6,7). Integrating participants' preferences into cancer screening policies or into cancer screening clinical encounters introduces unique challenges because population screening programs require acceptability by the target population to be "successful" (8-10). Hence, cancer screening preferences that deviate from the aims of cancer screening programs may be perceived as a threat to the effectiveness of a screening program (8,11). Scientific progress and frequently shifting clinical practice guidelines have increased the number and complexity of cancer screening decisions, such that different stakeholders will value the attributes of the decision options differently. Reflecting participants' preferences in cancer screening recommendations and practice is a person-centered and evidence-informed approach to address the increasingly complexity of cancer screening decisions (9,12-14).

New cervical cancer screening options are slowly being implemented in screening programs, resulting from a better understanding of cervical cancer etiology and vaccination programs that target infections of cancer-causing human papillomavirus (HPV) types (15,16). In many contexts, evidence has accumulated that HPV testing on cervical samples is a better way to screen for cervical cancer than cytology performed on cervical samples (often called the Pap test)

(17,18). This has introduced a new paradigm in cervical cancer screening, as individuals who undergo HPV testing are being tested for a sexually transmitted infection (19). HPV testing is more sensitive than the Pap test in detecting women at high risk for cervical disease, especially among women who have received HPV vaccination, but there is a greater risk for women to receive a “false positive”, especially women under 30 years (10,20). Unlike cytology, HPV testing can be performed on self-collected vaginal samples which introduces additional trade-offs for decision-makers to consider. In particular, HPV testing performed on self-collected samples is slightly less sensitive and specific compared to testing on clinician-collected cervical samples (21). However, recent systematic reviews have shown many women prefer to collect their own samples for cervical cancer screening compared to clinician sampling because of comfort, privacy, and convenience (22,23).

Screening participants now face several decisions about whether and how to participate in cervical cancer screening (10,19). For example, potential participants who are contemplating cervical cancer screening might identify what screening test to choose, what age to start, how often to screen. When presented with up-to-date evidence and feeling informed about the options, how individuals value the risks and benefits of the options will shape their preferences and decision-making (4,9,24). Women’s preferences can be integrated into cervical cancer screening through several channels: clinical practice guidelines might incorporate research on population preferences or providers might elicit preferences from women at the point of care through shared decision-making (4,14,24).

We produced two manuscripts from this systematic review (Chapters 4 and 5). In the present manuscript, we describe women’s preferences for primary cervical cancer screening across multiple contexts and study designs, such that the preference-eliciting approaches and realized preferences can be included in future guidelines or in practice. Currently, Canadian

cervical cancer screening guidelines do not incorporate a synthesis of evidence on women's preferences. Existing syntheses on women's cervical screening preferences do not consider the validity or trustworthiness of approaches used to elicit preferences, which leaves guideline developers without the strength of the evidence. This review addresses the gap in evidence by describing the prevalence of cervical screening preferences across diverse study designs, contexts, and preference-elicitation approaches (13,25,26). To provide impact data to guideline developers who look to incorporate high quality data, we describe the risk of bias among preference-elicitation methods.

Methods

The review protocol was registered with PROSPERO (CRD42016035737) and previously published (27). We briefly describe the methods here.

Theoretical framework.

Our theoretical framework was the Ottawa Decision Support Framework (ODSF), a frequently referenced theoretical model for shared decision-making that defines preferences as a person's overall most-favoured option, taking into account the person's knowledge of and attitudes toward the characteristics of the different options (4).

Inclusion and exclusion criteria.

The ODSF criteria for "preferences" (4) was considered in identifying relevant qualitative and quantitative studies. The ODSF define values as an individual's personal attitudes towards certain characteristics (or "attributes") of an option and the probabilities of outcomes. A preference, on the other hand, represents the identified option which is personally most favoured. Studies were included if:

- Participants realized there was a decision to be made (i.e., there were multiple options from which they could select their preferred option);

- There was an information comprehension component that allowed participants to review information about cervical cancer, the screening tests, and the risks and benefits of each option; and,
- There was a values clarification component that considered the person's attitudes to the options.

Studies were included if women were asked about preferences among multiple options for cervical cancer screening. We excluded articles that did not directly ask women about their own preferences. Articles were included only if they examined preferences related to primary cervical cancer screening. Studies were excluded if they examined correlates of cervical cancer screening that were not associated with preferences. Opinion pieces were not included.

Information sources.

Full details of the peer-reviewed search strategy are provided in the previously published protocol (27). MEDLINE, Embase, PsycINFO, CINAHL, and Cochrane Library databases, were searched for all articles from inception to December 2016. Grey literature was identified through ProQuest Dissertations and Theses, relevant Canadian health research/policy databases, and relevant Canadian conference proceedings. Reference lists of systematic reviews were examined for relevant primary studies. Additional references were sought by examining reference lists of non-systematic literature reviews, and through SCOPUS. Fourteen authors were emailed about translations of their articles; one author responded with a translation and another responded with a more recent article published on similar data.

Study selection.

Two of three reviewers (BW, SRVK, JE) independently screened each record and the full text of a potentially eligible study. Mendeley (www.mendeley.com) and Covidence

(www.covidence.org) were used for citation management and full-text screening. Disagreements regarding eligibility of studies were resolved by discussion.

Data extraction.

Study characteristics (e.g., dates, population, location), preference-eliciting approach (e.g., ad-hoc questionnaire, discrete choice experiments), ODSF characteristics (i.e., whether options were communicated, information comprehension, values clarification), outcomes (e.g., what preferences were described in the study), and methodological quality data were extracted using pre-tested forms in DistillerSR. Data were extracted by one reviewer and validated by a second reviewer. Overall study findings were summarized narratively, with consideration of the study context (28). Outcomes were categorized into measurements of validity/reliability and decision-relevant outcomes using the ODSF (e.g., decision quality, decision impact, decision action). Differences were resolved through discussion between the three reviewers.

Quality assessment and risk of bias.

The Mixed Methods Appraisal Tool (MMAT) (29,30) was used to appraise the methodological quality of the eligible studies for this review. The MMAT was selected as a quality appraisal tool for this study because it is easy-to-use and efficient to use, considers the integration of qualitative and quantitative findings, and researchers have demonstrated inter-rater reliability (30, 31).

Two reviewers independently appraised studies included for full text review and came to consensus on the final score. Studies were included in the final review if they scored 2* or greater, according to the scoring scheme (29). The review team determined that excluding low quality studies was the most appropriate method to incorporate risk of bias assessments into the synthesis given the non-aggregative nature of the analysis (32, 33).

To address risk of bias issues specific to preference-eliciting methods, included studies were further assessed using domains outlined by Yepes-Nunez et al. (34). With the guidance of two authors (JYN, YZ), we identified an assessment strategy for these domains, adapting each item to reflect ODSF-relevant features and outcomes. Each criterion could be assessed as “Yes”, “Probably yes (if unclear)”, “Probably no (if unclear)”, “No”, “Not applicable/relevant”. Extra weight was given to the domains “Was the instrument appropriately administered?” (e.g., was there a protocol to administer the instrument?), and “Was the choice of instrument optimal?” because the items in other domains were more concerned with general methodological quality, which was already assessed using the MMAT. Each study then received a preference-specific “risk of bias” rating using the following categories: “Critical”, “High”, “Moderate”, and “Low”. One reviewer (BW) assigned a rating for the preference-specific bias, and a second reviewer (SRVK or JE) checked this rating. Disagreements were resolved through consensus.

Narrative synthesis.

Tabular summaries were created for each decision identified among the included studies, to describe the context of the included studies, the preference-elicitation approach and the quality appraisal scores, and the overarching study findings anchored in the study context (“text-in-context”) (28). In these summary tables, we grouped studies from similar countries and contexts together, identified the options presented to study participants, and ordered the studies by the risk of bias score to help guideline developers identify relevant results. Patterns and themes were identified in qualitative and quantitative studies separately and contrasted in a preliminary synthesis. How study contexts and methodologies influence these findings was assessed (35), and the separate syntheses were integrated to understand similarities, differences, and gaps (36). The preference-elicitation risk of bias scores were presented beside the MMAT scores to contrast the methodological quality of the preference-elicitation approach with the reporting quality of the

study. We used these scores to describe the strength of evidence to facilitate high quality guideline development.

Results

The database searches returned 6,558 results, and 96 grey literature results were identified through additional searching. See Figure 4-1 for the PRISMA diagram of the screening process. After duplicates were removed (n= 4,098), we conducted full text screening of 663 articles, followed by quality appraisal of 131 eligible articles, which led to 28 articles reporting 29 studies being included in the review. The included articles consisted of 17 quantitative (37,38,47–53,39–46), 10 qualitative studies (40,54,63,55–62), and 1 mixed methods study (64), which included a quantitative survey followed by a qualitative focus group study. See Appendix C for a list of articles that were not included in the synthesis.

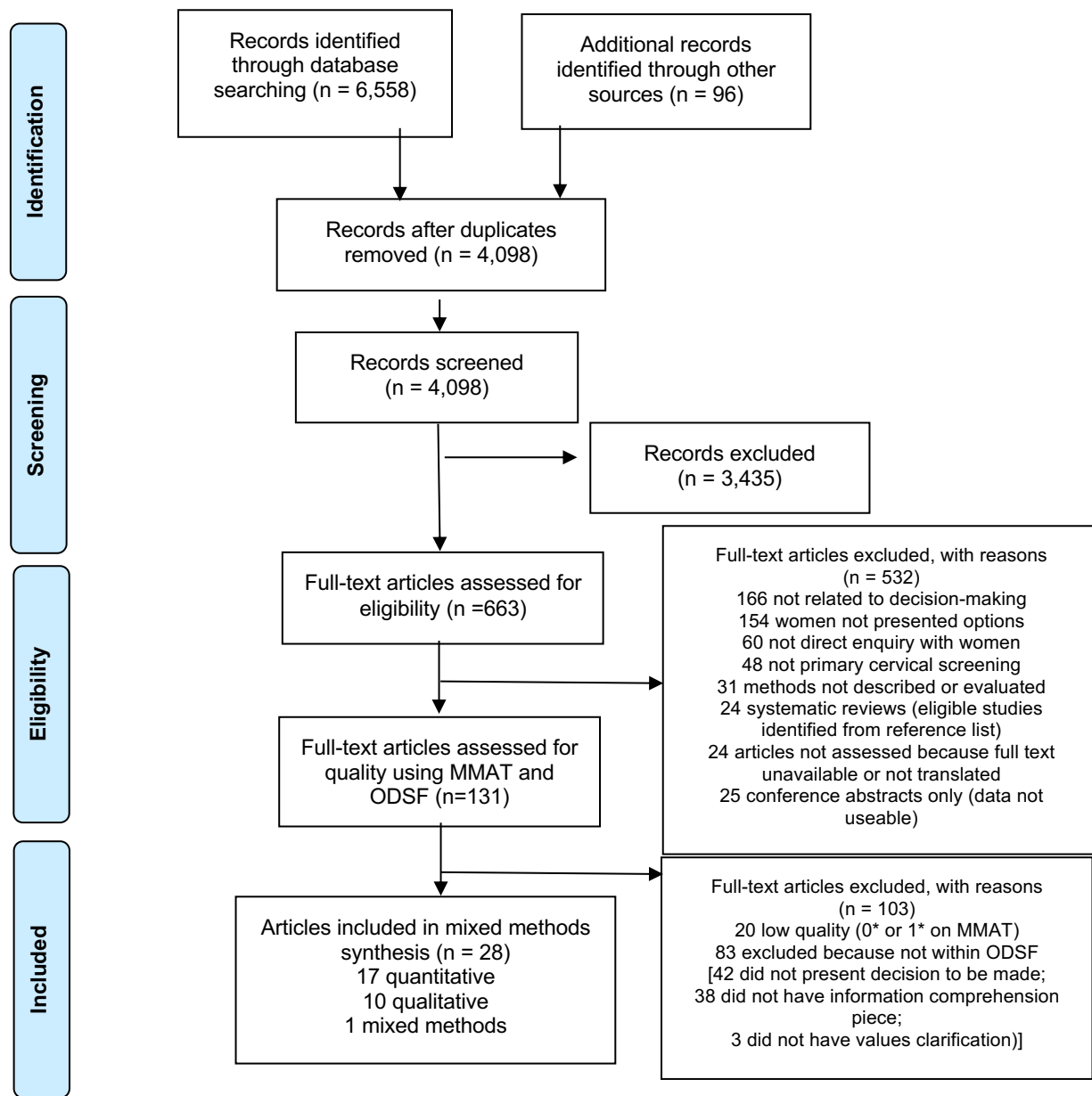


Figure 4-1. PRISMA flow diagram.

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097 For more information, visit www.prisma-statement.org

Eleven studies captured preferences from American populations, eight studies reported on United Kingdom populations, two studies each reported preferences of Canadian and

Zambian populations, and there were single studies from Australia, Cameroon, El Salvador, Ireland, Spain and Sweden.

Among 18 quantitative studies, there were 2 randomized controlled trials (37,38) and 16 non-randomized studies (39,40,49–53,64,41–48). Eleven qualitative focus group studies (47,54–64) were included, of which one study used both focus groups and interviews (37) to elicit preferences from women.

The majority of studies (n=23) focused on adult women, 18 years and older. Three studies considered only women younger than 30 years (46,47,50), and three studies included women solely between 30 and 65 years (39,43,44). Six articles (seven studies) also included women aged 65 years and older (42,44,54,56,58,64). Thirteen articles (fourteen studies) investigated preferences of women in low-income settings or who belonged to a minority or marginalized population (e.g., by ethnicity, living conditions, immigrant status) (39,42–45,48,54,55,57,62–64).

Tables 1 to 6 present the various cervical cancer screening decisions that were reported in the studies. Most articles (n=12) investigated preferences toward a specific modality of cervical screening, which was sometimes conflated with location of screening: HPV self-sampling (also called “home sampling”) versus an in-clinic option (e.g., Pap testing, physician-administered, in-clinic). Eight studies considered women’s preferences about participating in cervical screening and/or frequency of screening tests, in which some studies asked about participation in screening in the context of recommended frequency. Six studies considered women’s stated preferences among hypothetical policy options using a discrete choice or conjoint analysis design. Three studies asked women which type of test (e.g., cytology vs HPV) they would prefer. Other decisions presented include willingness to pay (n=3), and preferred self-collection device (n=2).

Tables 1 through 6 illustrate an overview of the main preferences identified in the included studies.

Key preferences.

Modality/location of screening.

Table 1 presents the text-in-context (28) preferences reported in twelve studies that investigated women' preferred cervical cancer screening modality or location of cervical screening. Six studies based in the United States (43-45,50,54,55) and two studies based in Canada (56,57) investigated women's preferences for a self-collected screening option compared to provider-administered options. Of these eight studies, two (50,54) American studies were considered "low" risk of bias for preference-elicitation. One study examined a questionnaire's validity in eliciting adolescent females' perspectives on self-sampling options compared to clinician testing, and the other used a focus group approach to elicit preferences among low-income women. Despite these different approaches, both studies found that while women may be open to the idea of self-sampling, they are apprehensive about the modality and still prefer to visit with a health care provider for cervical cancer screening. Three other American studies (44,51,53) used questionnaires to elicit preferences of screening modalities among samples of women who frequently face barriers to screening; some women might use a self-sampling option in the future (51), though the studies also conveyed mixed preferences such that respondents did not have a preference. Among North American focus group studies (54-57), respondents indicated that they would prefer to see a healthcare provider for cervical screening over a self-sampling option. However, respondents also noted that a self-sampling option could be useful in some cases. Two studies (60,64) based in the United Kingdom reported similar findings from focus groups with Hindu women living in London, though the studies were scored as "critical" for risk of bias.

A questionnaire study based in El Salvador (39) found that a larger proportion of under-screened women preferred a self-sampling option, compared to provider-collected sampling or no preference. However, when asked about future cervical screening, the majority of respondents indicated a preference for screening at a health centre. Another study in Cameroon (46), also scoring critical for risk of bias, found that respondents preferred physician-collected HPV testing because they perceived it to be more reliable compared to self-sampling.

Overall, there was a wide range of approaches to understand women's preferences among cervical cancer screening modalities, which were found to vary in potential risk of bias among preference-elicitation methods. Women's preferences for specific screening modalities seem to be dependent on their current socio-demographic context, the way that the information was presented and the corresponding options from which women would choose their preference.

Participation in screening/frequency of screening.

Table 2 presents the eight included studies that focused on the decision to participate in cervical cancer screening or intention to participate in screening (38,41,47,53,60–62), with risk of bias scores ranging between low and critical. Four studies – two quantitative and two qualitative studies - were based in the USA (43,46,60,63), a focus group study and a cluster randomized trial were based in the United Kingdom (37,58), one randomized trial was based in Sweden (38), and another focus group study was based in Zambia (62).

An American questionnaire study (43) that had a medium risk of bias investigated women's intention to participate in screening with at-home screening option. In this study, investigators asked Arab and Hispanic women if they would participate in future screening with a home screening option and found that the majority of respondents preferred not to use an at-home screening test in the future. Another American study (46) reported that the majority of female university students who used a decision aid intend to participate in future cervical cancer

screening. While the qualitative studies focused on different population segments, including women living in a homeless respite centre (63) and a representative sample of primary care patients (60), the focus group approach highlighted what women value in relation to their preferences around screening participation. For example, many women recruited from homeless centres indicated that they want to participate in screening but face barriers to accessing safe care, whereas women recruited from general primary care prefer more frequent Pap testing than is recommended.

Two randomized controlled trials, based in Sweden (38) and Wales (37) respectively, investigated women's preferences to participate in cervical cancer screening, as evidenced by their cervical cancer screening behavior. In one trial (38), 29.5% of women who had not been screened in the previous five years chose to get screened within the study period after working with a midwife to overcome barriers, compared to 18.5% of women who did not receive any intervention. In a cluster-randomized trial (37), primary care practices in the intervention arm provided eligible patients with risk perception information about cervical cancer screening, compared to standard care information. In the intervention arm, 44% wished to have annual cervical screening (which authors noted is more frequently than guidelines recommend) compared to 61% of the comparator group. Participants noted that they wanted to participate in cervical cancer screening because they thought it was good for their health (62), believing that more frequent screening reduces their risk of cervical cancer (37,60) A qualitative study (58) conducted in the United Kingdom found that some women may have participated in cervical cancer screening because of social norms and pressure from their health care providers, but not because they had made an informed choice.

The majority of these studies reported that women prefer to participate in cervical cancer screening; researchers used values clarification techniques, both explicit through questionnaires

or implicit in focus groups or interview, to understand and contrast the important factors behind preferences for and against participation in cervical screening. How women value different attributes of cervical cancer screening varied by study location and population, the time that the study was executed, and the specific value statements that were presented to study participants. Additionally, some European studies (37,38,58) question whether participants are making informed decisions to participate in cervical screening.

Stated preferences with discrete choice experiments.

In Table 3, we describe the findings of six stated preference studies that looked at the relative preferences for features of cervical cancer screening tests. These studies were grouped together because the decisions involve hypothetical trade-offs that are usually based on current policy consideration, with an objective to estimate the likelihood of participation in cervical cancer screening. These studies by definition are quantitative, using questionnaires based on a conjoint analysis or discrete choice design.

Two studies (41,53) analyzed responses from slightly different analytic samples of Scottish women, to consider how participants would respond to an explicit “opt-out” option for cervical cancer screening when also offered hypothetical screening scenarios. In these analyses, 27 women (out of 491 (53) and 577 (41) women, 5.5% and 4.7% respectively) indicated that at some point during the questionnaire, they prefer to opt out of cervical cancer screening instead of participating given the hypothetical scenarios. These studies found that women consider the option to screen versus not screen as a separate decision than choosing among screening scenarios. In Wordsworth’s study (41), as well as Arana’s (47) study in a Spanish undergraduate population, participants most valued the screening scenarios that allowed for the shortest time between screens.

Fiebig et al. (49) used a discrete choice experiment to elicit women's preferences among different cytology options, with the majority preferring the standard conventional cytology option in 39.1% of the choice sets, and no testing in 37.3% of the choice sets. The then-new liquid-based cytology test was preferred in 23.7% of the choice sets. Women (and general practitioners who make recommendations for women) in this study placed greater value on cost, false positive rate, and annual screening contributing to preferences against liquid-based cytology.

Two studies (42,51) used conjoint analysis to examine how women value different attributes of cervical cancer screening options, as women made choices among hypothetical cervical cancer screening options. The first study found that Zambian participants preferred, with all other attributes held constant, options that offered: immediate screening results; a female health care provider; returned picture of one's cervix; a urine sample collection; and a close location to one's home. A similar study performed in an American gynecology clinic among 52 patients found that, with all other attributes held constant, participants preferred options that: had the lowest false positive rate; had the lowest false negative rate; returned HPV status; allowed a health care provider to explain the results; more frequent testing; involved a low co-pay option; and, was collected by a health care provider.

Overall, these studies which considered how women make trade-offs among hypothetical policy options demonstrated lower risk of bias, compared to other decisions, as two studies scored low, three studies scored medium, and one study score high. This approaches often integrated several screening decisions, including screening modality and screening test, into a single decision between hypothetical "contextual" options. These studies showed that women in different settings might value similar attributes, including test accuracy or a shorter time between screening, but their ultimate preferences strongly depend on the options that are presented (e.g.,

opting out of screening), and the outcomes associated with these options (e.g., follow-up of an abnormality, risk of cancer).

Type of screening test.

Two qualitative studies (54,59) and one mixed methods study (64) investigated women's attitudes toward HPV testing compared to Pap testing, as presented in Table 4. Although these studies involved diverse study settings spanning eleven years, and were varied in risk of bias, the findings were generally consistent. Researchers found that women expressed anxiety around the possibility of receiving HPV test results in addition to or instead of cytology results for cervical cancer screening, although some women were interested in knowing their HPV status. The stigma of a sexually transmitted infection, and the idea that a positive HPV infection cannot be treated were reasons that women gave for leaning toward a Pap test. These feelings were contrasted with the desire to receive a more detailed risk profile for cervical cancer (54).

Willingness to pay.

Two studies (40,52) examined willingness to pay for Pap tests using payment card techniques, and one study indirectly inquired about women's willingness to pay for cervical cancer screening, using a multiple-choice question in an investigator-developed questionnaire (43). Different study contexts implied different currencies, and different years of study implies different interpretations of costs, hence it is difficult to identify a common theme. A 2001 study of respondents from a Scottish cervical cancer screening program (40) reported a median willingness to pay for a Pap test was £45.00. When examining willingness to pay for unit changes of attributes of cervical cancer screening, analysis showed that women from the aforementioned sample were willing to pay £5.37 for a one percent reduction in the chance of recall. A 2002 American study of obstetrics-gynecology patients (52) found that respondents were willing to increase their pay of a Pap test from a median of \$50 to \$100 with improved

performance of the Pap test, independent of their personal risk of cervical cancer. A more recent American study among Arab and Hispanic women (43) found that 50.1% of participants were willing to pay between \$1 and \$30 for a self-sampling test, whereas 34.7% of participants indicated they would only use the test if free of charge.

Other.

A qualitative study and a mixed methods study (61,64) based in the United Kingdom asked women to consider different self-collection devices; the devices that were preferred by participants were described as “simple”, “easier” to use, and “less intimidating” because of the size. The risk of bias in both studies was scored as “critical”.

Discussion

We identified 131 articles that describe or measure women’s primary cervical cancer screening preferences, although only a small proportion of these studies elicit informed, values-based preferences. Twenty-eight studies that consider cervical cancer screening preferences communicate the decision to select a preference, provide information on the options and their risks and benefits, and clarify values to help women decide among options. This systematic review is the first to critically assess women’s primary cervical cancer screening preferences across multiple contexts and study designs. It is difficult to synthesize or confidently conclude what different groups of women prefer in cervical cancer screening, because a wide range of study methodologies are used to examine context-specific decisions, among varied preference-eliciting approaches. Another report from this systematic review suggests that of the included preference-eliciting articles, there is substantial variation in methodological and reporting quality. In this study, we found that most of the relevant studies of women’s cervical cancer screening preferences investigated preferences among screening modalities (e.g., provider-collected or self-collected), but studies also examined women’s preferences about cervical

screening participation, test type used, time between screenings, willingness to pay, self-collection devices, and other screening characteristics.

Notable themes that emerged include a preference for provider-collected cervical cancer screening compared to self-collected screening, although certain population segments might prefer self-collection. These preferences were associated with trust and confidence in health provider expertise, and high valuation of cervical screening participation; several populations preferred a more frequent screening option. However, there were also examples of populations who preferred self-collected screening that related to convenience, ease, and privacy, or who preferred not to participate in screening at all.

In most cases, cervical cancer screening enthusiasm was correlated with women's desire to "do the best thing for their health", hence the efficacy of the screening test was highly valued. "Efficacy" of screening tests was communicated to respondents as the relationship between screening and subsequent risk of cervical cancer incidence or mortality, highest rate of cancers found (e.g., sensitivity), but also sometimes just as "perceived accuracy". This preference for improved detection is consistent across time and in different countries, which is presumably driving the preference for annual screening, despite respective guideline recommendations. Similarly, a general preference for provider-collected modalities was related to increased confidence in the results, which could be related to perception of improved efficacy, but also because patients rely on clinicians for recommended "best practices". A systematic review of cancer screening stated preferences found that women value sensitivity and efficacy (65) above other cancer screening features, followed by screening process and cost. Preferences for frequent and accurate test technologies illustrate an overall enthusiasm for cervical cancer screening, or cancer screening more broadly, which has been noted in the literature previously (66). The tendency for women to desire a more aggressive screening schedule could be related to an

overestimation of the benefits of screening as well as their actual risk for cancer (8,67). These preferences are consistent with discourses reinforced by cervical cancer screening programs, health care providers, and researchers, namely that women are “at risk” and screening participation is important and normal (68). Constantly changing recommendations (20) and health care provider perspectives (69) may also enhance individual preferences for more frequent, accurate screening.

We observed examples of studies where women’s preferences among cervical cancer screening options contradicted the “general preference” for almost every decision available. Three notable examples of diverging preferences include decisions to “opt out” of cervical cancer screening (38), preferences for cervical cytology over HPV (54,59,64), and preferences for self-collection for cervical cancer screening compared to provider-administered, in-clinic tests (39,43,44,55). In one randomized controlled trial (38), the majority of women – who were considered “non-responders” by the Swedish cervical cancer screening program - revealed a preference to not participate in cervical cancer screening through their screening behaviour. It is worth noting that screening behaviour may not actually reflect women’s preferences, in particular if they did not realize there was a decision (to participate in cervical cancer screening) to be made (4). Additionally, because participation in cancer screening is an ongoing decision - unlike treatment decisions which are often one-time decisions - it is possible that women who chose not to participate in cervical cancer screening during the study time period may have preferred to screen at a later time. The concept that preferences are dynamic is important for future research and implementation of preference-based cervical cancer screening.

Despite evidence suggesting HPV testing’s superior test sensitivity compared with traditional cytology (17) and women’s noted preference for the most sensitive test option, the qualitative reports (54,59,64) included in this review indicate some resistance to HPV testing as a

cervical screening modality. Stigma of a sexually transmitted infection and the lack of “cure” for HPV may have outweighed a perceived benefit of added sensitivity in these cases.

The studies that found that women prefer self-collected screening compared to other modalities aligns with a recent systematic review and meta-analysis that found that the majority of women (65%) expressed a preference for HPV self-sampling compared to clinician-based sampling (22). This review also found that when examining only studies that reported preferences between self-sampling, clinician-sampling, or both (i.e., a choice among three options instead of two), that there was a substantially smaller proportion of women (31%) reporting a self-sampling preference, contradicting an outstanding theme from the present study. Question framing and the attributes presented in a preference-eliciting tool might influence the values and preferences that individuals will report in stated preference experiments for colorectal cancer screening (70,71), which is also likely relevant for cervical cancer screening decisions. It follows that the reliability, validity, and lack of bias in the preference-eliciting approach, and the quality of the study methodology can greatly impact how patient preference studies are presented, interpreted (1,31,72), and used (2) in clinical practice and guidelines.

There are limitations with this systematic review. The heterogeneity of study objectives and designs, preference-eliciting approaches, and population contexts limited our ability to synthesize data across specific decisions for primary cervical cancer screening. However, a narrative presentation of findings is important to guide further studies of women’s preferences, with an emphasis on context in women’s preferences. We excluded a large selection of studies that did not meet the ODSF criteria of “preferences” (i.e., i) recognize decision; ii) information comprehension; iii) clarify values), which could lead to different conclusions using a different conceptualization of preferences. Moving forward, guideline developers and health care providers will prioritize evidence based on informed, values-based preferences, as participant

autonomy embodies the concept person-centered medicine. Studies poorly reported the content and structure of the preference-elicitation approaches; thus, reviewers were liberal about what constitutes information comprehension and values clarification.

Excluding low quality studies from the analysis might affect the external validity of the review findings (33, 34). Given that it was challenging to draw conclusions about women's cervical screening preferences from studies that were considered "robust", it would be nearly impossible to thoroughly assess and summarize studies that lacked methodological rigour (33, 34). As a result, the review team decided to forego a post-synthesis sensitivity analysis given the state of the cervical screening preferences literature.

Communicating the decision, information about the decision's options, risks and benefits, and identifying attributes that are most important to women will support individuals making autonomous choices. Preference data can inform individual cervical cancer screening interactions between health care providers and women and can be aggregated to inform clinical practice guidelines (1,2,9,14,72). However, guideline developers might find that a context-specific synthesis of women's "preferences" for cervical cancer screening does not provide rigorous evidence for high quality recommendations. To account for the dynamic nature of cervical cancer screening preferences, some consideration should be given to a robustly-developed point-of-care preference-elicitation tool. To our knowledge, there are no known existing decision support tools for primary cervical cancer screening in North America.

Conclusion

This study is the first systematic review to examine women's preferences for cervical cancer screening across qualitative and quantitative study designs, contexts, and decisions. Guided by the Ottawa Decision Support Framework, this study considers informed, values-based preferences for screening participation, screening test and modality, frequency, device and

willingness to pay. Overall, women's preferences varied by geography, socio-demographic characteristics, and current cervical cancer screening context. Although some values were generally consistent across study contexts and time, such as a general preference for a provider-collected sample over a self-collected sample or a concern about sexually transmitted infection testing, there were examples of populations with divergent preferences. Because of the wide heterogeneity in the methodological and reporting quality of the included studies, it is challenging to determine whether these differences are due to study context or the type of approach used to elicit preferences. Subsequently, guideline developers might find that even a context-specific synthesis of women's "preferences" for cervical cancer screening does not provide rigorous evidence for high quality recommendations. A robust approach to preference-elicitation in cervical cancer screening is needed, but this review also shows that women's preferences vary by the time and context in which they are captured. In this case, a point-of-care approach to preference-elicitation (e.g., during encounters between women and providers) might capture the dynamic nature of women's cervical screening preferences more accurately than guidelines that incorporate preference prevalence data.

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Table 4-1. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Modality of Cervical Cancer

Screening/Location of Screening (n=12)

Citation	Country	Context	Decision Options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias	MMAT Score
Kahn, 2005 (50)	Ohio, USA	116 women (82% black) from an urban, teen health centre, between 14 and 21 years	Modality: - Self-testing - Clinician testing - No preference	Questionnaire	Pre-examination, 65% of participants preferred clinician testing, 13% of participants preferred self-testing, and 22% reported no preference. Trust of test result, confidence in ability to collect, and perceived effects of testing were significantly related to differences in acceptability between clinician testing and self-testing.	Low	3*
Anhang, 2004 (54)	Massachusetts, USA	Eight focus groups with 48 ethnically diverse, low-income women between the ages of 18 and 81 years	Modality: - In-clinic - Self-sampling	Focus group	Respondents discussed a need for information about HPV transmission, prevention, treatment, and tailored risk profiles. Most women expressed an interest in an HPV test, but not as a substitute for a Pap test. Most women still preferred the physician-collection option for HPV testing because they wanted to make sure that the sample was collected appropriately.	Low	3*
Kidder, 2014 (43)	Florida, California, and District of Columbia, USA	476 Arab and Hispanic women (249 Arab women, 227 Hispanic women), living in the USA, between 30 and 65 years	Modality: - At-home screening test - In-clinic test	Questionnaire	277 (58.2%) of 476 women indicated they would likely participate in cervical screening with an at-home option in the future, every 3 years (43.8% of Arab participants and 74.0% of Hispanic participants). 73.3% of participants (67.5% of Arab participants, 79.7% of Hispanic participants) indicated that they were likely or very likely to use a urine test for cervical screening if it were available.	Medium	2*
Penaranda, 2015 (44)	Texas, USA	110 Hispanic between 30 and	Modality:	Questionnaire	42.7% of participants indicated no preference for either test. Among	High	2*

		65 years (mean age= 48.2 years) living on the USA – Mexico border who accepted an offer to participate in self-sampling	- Definitely prefer Pap; - Somewhat prefer Pap; - Prefer both equally or unsure; - Somewhat prefer self-sampling; - Definitely prefer self-sampling		women who stated a preference, 30% indicated a preference for self-sampling compared to 26% for Pap testing (statistically significant).		
Scarinci, 2013 [55]	Mississippi and Alabama, USA	9 African-American women between 25 and 51 years from the Mississippi Delta were part of a discussion group, that was informed through community participatory research	Modality: - Pap smear vs. self-collected sampling	Focus group	Some women preferred self-sampling because of “privacy and convenience”, while some women indicated they would participate in both because “the doctor’s test is more accurate”. Focus group participants found “in-person” demonstration format, preferring that to a video.	Critical	3*
Anhang, 2005 (45)	New York City, USA	172 low-income, inner-city women living in the USA, primarily Hispanic, over the age of 25	Modality: - Physician-administered at doctor’s office - Self-collected at home - Self-collected at doctor’s office - None	Questionnaire	Of women that declared a preference between self-sampling vs clinician-administered testing, 32.4% preferred self-sampling (no data on how many did not indicate a preference). The survey also asked what scenario would make participants more likely to screen for cervical cancer every year: 47% selected physician-administered test during pelvic exam, 21% said self-collected test at home, 19% said self-collected test at physician’s office before pelvic exam, and 12% said none of the scenarios.	Critical	2*

Barata, 2008 (56)	Ontario, Canada	44 women aged 19 to 64 from 3 distinct settings (semi-rural, medium-sized city, large city) participated in 3 focus groups	Modality: - Pap testing - Self-sampling	Focus group	Respondents appreciated an additional option for cervical screening (self-sampling + Pap testing) but did not want to replace Pap testing. Many women emphasized that a self-sampling option could be "empowering" and particularly relevant for women who face barriers to in-clinic cervical screening (benefits-minus-barriers favouring self-sampling), some women felt that Pap testing was still appropriate (e.g. because a gynecological exam is still useful).	Medium	2*
Howard, 2009 (57)	Ontario, Canada	77 immigrant women who lived in Canada for 1 to 5 years, aged 22 to 69 years participated in 11 focus groups	Modality: - Self-sampling - In-clinic	Focus group	While many respondents perceived some benefits of a self-collection option, most women indicated that they personally would prefer seeing a health provider (though cultural considerations may require this professional to be female).	High	3*
Rosenbaum, 2014 (39)	Paracentral El Salvador	518 women who hadn't been screened in three years were recruited by community health promoters from four rural health units, between 30 and 49 years	Modality: - Self-sampling - Provider-collected sampling - No preference Location: - Home - Clinic - No preference	Questionnaire	After participating in both collection modalities, 38.8% preferred self-sampling, 31.9% of women preferred provider-collected sampling, and 29.3% of women indicated no preference. 37.5% of participants expressed a preference to screen at home in the future, 42.1% preferred to screening at the health centre in the future, and 20.4% had no preference. Among women who preferred self-sampling, privacy/embarrassment, ease, and pain were the three most frequently occurring reasons. Among women who preferred provider-collected sampling, result accuracy, provider experience, and fear of improper sampling were most common concerns. Among women who	High	2*

Berner, 2013 (48)	Yaoundé, Cameroon	243 non-pregnant women in Cameroon between 24 and 65 years (median age= 39 years) who participated in both self-collected and physician-collected HPV testing	Modality: - Self-sampling - Physician-sampling - No preference	Questionnaire	preferred future home screening said privacy, comfort, and time as their main reasons. 29% of women preferred self-sampling, 62% of women preferred physician sampling. The remaining 19/217 women did not answer or indicated no preference. Most women who preferred physician sampling felt that it was more reliable than self-sampling.	Critical	2*
Szarewski, 2009 (61)	England, UK	28 Muslim women were recruited from a community centre to participate in 3 focus groups	- Self- sampling - In-clinic	Focus group	Respondents were enthusiastic toward cervical screening as part of preventive health care. Many respondents were apprehensive toward self-sampling because they didn't trust their ability to properly collect a sample, though they acknowledged that the modality might be useful for some women.	Critical	3*
Cadman, 2015 (64)	England, UK	185 Hindu women living in London returned a survey (92% Indian), 23 Hindu women between 14 and 70 years who were living in London participated in 4 focus groups	Modality: - Self- sampling - In-clinic	Questionnaire + focus group	Preferences were elicited through the focus groups. While positive about HPV self-sampling, many women indicated a preference for participating in cervical cancer screening with their doctor, because they could be confident in having the test correctly. Additionally, when comparing an HPV test of self-sampling versus cytology with a doctor, many women preferred more frequent screens.	Critical/not useable	2*

Table 4-2. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Participation in Screening (Or Intent to Participate) (n=8)

Citation	Country	Context	Decision options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias Preference elicitation	MMAT score
Kidder, 2014 (43)	Florida, California, and District of Columbia, USA	476 Arab and Hispanic women (249 Arab women, 227 Hispanic women), living in the USA, between 30 and 65 years	Intention to screen, if self-test was available: - Very likely to use it if the price was affordable; - Likely to use it if the price was affordable; - Not sure/no opinion; - Unlikely to use it; - Very unlikely to use it	Questionnaire	41.4% of respondents indicated that they would prefer to use an at-home HPV test to screen in the future, and 13.4% of participants did not know/weren't sure. 73.3% of participants (67.5% of Arab participants, 79.7% of Hispanic participants) indicated that they were likely or very likely to use a urine test for cervical screening if it were available.	Medium	2*
Alexander, 2014 (46)	Missouri, USA	568 women aged 18–26 years old seeking care at an urban university student health and wellness clinic	Intention to participate in screening: - Yes - No	“[D]ecision-aid” type intervention + questionnaire	492 (86.3%) of 568 undergraduate women expressed intention to participate in future cervical screening after using a "decision-aid format" intervention. 11% expressed no intention to participate in future cervical cancer screening and 2.7% of women did not answer the question.	High	3*
Wittenberg, 2015 (63)	Massachusetts, USA	8 focus groups with 42 women between 20 and 64 years who were recruited from a homeless medical respite	Participation in screening: - Yes - No	Focus groups + best-to-worst scale	This study did not report women's preferences after using the tool but reported the development. Women reported that screening decisions fall into 6 categories: provider (familiarity and provider gender important), setting (places more accepting of homeless	High	4*

		center, emergency shelters, and a residential substance use treatment program. Pre-tests conducted with 30 women			individuals) test procedure, test results personal fears and barriers, personal motivations. Pretests of the scale indicated that many women interpreted the wording “most important” and “least important” from a personal experience, instead of a population forecast.		
Smith, 2003 (60)	Michigan, USA	8 focus groups with 68 participants between 18 and 65 years who were participating in a risk-based screening trial	Participation in screening: - Yes - No	Focus groups	Respondents do not prefer risk-based screening, which involves reducing the frequency of screening tests. They wanted to have more frequent screening (annual or more frequent), and an early start date (beginning after menses).	Critical	3*
Holloway, 2003 (37)	Wales, UK	1,890 women in North Wales were recruited while waiting for their routine cervical smear (mean age 42 years)	Participation in screening: - Yes - No	Questionnaire	44% in intervention group stated that they wanted screening within 1 year – more frequent than recommendations, compared to 61% in control group (OR=0.51, statistically significant). 7% of 829 women in control group and 5% of 630 women in intervention group participated in screening with shorter than recommended intervals (not statistically significant). Women who undergo a risk perception session are more likely to state a preference for screening intervals aligned with recommendation, but not necessarily attend screening according to recommendations	Low	3*
Jepson, 2007 (58)	Scotland, UK	31 women recruited from the cervical screening program, between 19 and 55 years, among 9 focus groups and 15 interviews (68	Participation in screening: - Yes - No	Focus groups + key informant interviews	Women indicated that they needed more information to make an informed choice for cervical cancer screening. Most respondents (in cervical cancer screening, and other types of cancer screening) did not feel that they had made an informed choice to participate in screening or not. However,	High	3*

		interview participants were recruited to speak about cervical, breast, and colorectal cancer screening)			respondents were not overly concerned about a lack of informed choice.		
Oscarsson, 2008 (38)	Southeast Sweden	133 women in Sweden between 28 and 65 years with diverse screening experience participated in the study group	Participation in screening: - Yes - No	Questionnaire	First author contacted intervention arm to have a personal conversation with non-attendees about reasons for not having cervical cancer screening, communicating risks and benefits. 29.5% of intervention group participated in cervical cancer screening compared with 18.5% in the control group. 175 women (44%) of the study group chose not to participate in the intervention. 120/400 (30%) women in the intervention group felt that they should participate in cervical cancer screening.	Medium	2*
White, 2011 (62)	Lusaka, Zambia	60 women attending primary health care clinic who peer educators identified as eligible for screening, between 18 and 49 years, participated in 6 focus groups	Participation in screening: - Yes - No	Focus group	Respondents participated in cervical screening because they wanted to know if they had cancer, but also to address reproductive health symptoms or concerns. Value of the screening process and supportive attitudes of clinic staff were noted as positive attributes. Discomfort of the screening process and stigma of HIV, sexually transmitted infections and cancer might deter women from screening.	High	2*

Table 4-3. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Stated Preferences Among Hypothetical Policy Options (e.g., Using Discrete Choice Experiments) (n=6)

Citation	Country	Context	Decision options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias Preference elicitation	MMAT score
Fiebig, 2009 (49)	New South Wales, Australia	167 women between 18 and 69 years from with different screening experiences (also examined clinician perspectives, not reported here)	Two hypothetical policy alternatives based on following options (opting out is an option): - Liquid-based cytology - Conventional cytology	Questionnaire based on discrete choice	Two samples of women (previously screened and never-screened) and GPs were asked to weigh cervical cancer screening attributes when making asked which option they would choose or recommend: a standard Pap test, a liquid-based Pap test, or no test. “Women chose no test in 37.3% of choice sets, the standard Pap test, in 39.1% and the liquid-based test in 23.7%. GPs chose no test in 42.8% of choice sets, the standard Pap test in 32.4% and the liquid-based test in 24.7%. 26.3% of women and 22.3% of GPs never chose liquid-based test, suggesting that there were preferences against the liquid-based Pap test.	Low	3*
McCormick, 2008 (51)	(unclear), USA	52 primarily white women between 22 and 78 years who attended an outpatient gynecology clinic participated	Two hypothetical programs (opting out is not an option)	Questionnaire based on conjoint analysis	Among respondents: - A 5% false positive rate was preferred over a 25% false negative rate - Learning HPV status was preferred - Women preferred to have a doctor explain the results - Women preferred to have yearly exams versus every 3 years, or every 5 years . - Women preferred a cheaper co-pay option of \$10 to \$30	Medium	2*

Wordsworth, 2006 (41)	Scotland, UK	577 women eligible for screening in Scotland between 20 and 59 years	Two hypothetical policy alternatives (opting out is an option)	Questionnaire based on discrete choice	-Pap modalities were preferred of self-testing by themselves or with a primary care provider 27 of 577 women indicated a “prefer no screening” response at some point during the questionnaire. The decision to screen or not was taken independently of the programs offered, as indicated by an insignificant inclusive value parameter. This finding suggests that altering the screening characteristics offered to women may not influence uptake.	Low	3*
Ryan, 2004 (53)	Scotland, UK	491 women eligible for screening in Scotland, between ages of 18 and 65	Two hypothetical policy alternatives (opting out is an option)	Questionnaire based on discrete choice	Of 491 responses, 27 individuals indicated at some point that they “prefer no screening” (11 always preferred no screening). The inclusive value parameter is insignificantly different from zero, suggesting that women make a decision about a cervical screening decision based on attributes related to the screening program.	Medium	2*
Arana, 2006 (47)	Gran Canaria, Spain	60 first-year female university students (mean age 22.6 years); compared with 60 female oncologists’ with work experience at a large hospital	Two hypothetical policy alternatives (opting out is an option)	Questionnaire based on discrete choice	Authors were interested in comparing how experienced medical professionals value attributes of cervical cancer screening compared to a sample of female undergraduate students. Participants’ utility is negatively associated with the cost of the program, the time span allowed until a new test for screening is required, the time needed for having the results available, the probability of a false positive and the probability to die of cervical cancer. The largest negative impact on utility is obtained for the extra years to be allowed between the screening tests.	High	2*

Chamot, 2015 (42)	Lusaka, Zambia	208 Zambian women, between 18 and 49 years	Two hypothetical policy alternatives based on following options (opting out is not an option): - Visual screening - HPV screening	Questionnaire based on conjoint analysis	All other attributes held equal, women in this sample preferred the screening options that: - Delivered immediate screening results; - Offered a female nurse (instead of a male nurse); - Provided a picture of their cervix; - Offered urine testing, compared to a vaginal or cervical modality - Were a 10-minute walk or 15-minute bus ride distance from home.	Medium	3*

Table 4-4. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Preferences for Different Screening Tests (e.g., HPV Testing vs Pap Testing) (n=3)

Citation	Country	Context	Decision options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias Preference elicitation	MMAT score
Anhang, 2004 (54)	Massachusetts, USA	Eight focus groups with 48 ethnically diverse, low-income women between the ages of 18 and 81 years	Type of screening test: - HPV test - Pap test	Focus group	Women in these focus groups discussed a need for information about HPV transmission, prevention, treatment, and tailored risk profiles. Most women expressed an interest in an HPV test, but not as a substitute for a Pap test. Most women still preferred the physician-collection option for HPV testing because they wanted to make sure that the sample was collected appropriately.	3*	Low
McRae, 2014 (59)	Ireland, UK	59 women between 17 and 69 years, recruited through primary care and well-women's centres, participated in 10 focus groups	Type of screening test: - HPV test - Pap test	Focus group	Respondents were apprehensive to introduce HPV testing into primary screening framework, because of the psychosocial effect of a positive result and stigma of infection. On a similar note, they were more accepting of having HPV testing as part of colposcopy or following up an abnormal result.	Medium	2*
Cadman, 2015 (64)	England, UK	185 Hindu women living in London returned a survey (92% Indian), 23 Hindu women between 14 and 70 years who were living in London participated in 4 focus groups	Type of screening test: - HPV test - Pap test	Questionnaire + focus group	Respondents indicated that although self-sampling may offer some convenience, they would prefer a doctor collect their sample to be sure it is collected correctly. Respondents mentioned that HPV testing might introduce relationship problems and may cause unnecessary anxiety..	Critical/not useable	2*

Table 4-5. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Willingness to Pay for Cervical Cancer

Screening (n=3)

Citation	Country	Context	Decision options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias Preference elicitation	MMAT score
Raab, 2002 (52)	Pennsylvania, USA	175 women who attended an American urban obstetric-gynecology clinic, aged 18-78 years	Willingness to pay for Pap test (between \$0 and \$1000 USD)	Questionnaire based on payment card (categorical scale)	Respondents indicated an increase median willingness to pay (from \$50 to \$100) as the performance of the new Pap test increased, independent of their individual risk for cervical cancer (no statistically significant differences in mean willingness to pay at different Pap test performance levels).	Medium	2*
Kidder, 2014 (43)	Florida, California, and District of Columbia, USA	476 Arab and Hispanic women (249 Arab women, 227 Hispanic women), living in the USA, between 30 and 65 years	Willingness to pay for a self-screening test (\$0, \$1-\$30, >\$30)	Questionnaire	45.2% of respondents communicated they do not want to use an at-home test to screen for cervical cancer in the future. 41.4% of respondents indicated that they would prefer to use an at-home HPV test to screen and 13.4% of participants did not know/weren't sure. 73.3% of participants (67.5% of Arab participants, 79.7% of Hispanic participants) indicated that they were likely or very likely to use a urine test for cervical screening if it were available.	Medium	2*
Wordsworth, 2001 (40)	Scotland, UK	595 women eligible for screening in Scotland, between 20 and 59 years	Willingness to pay for Pap smear (£0, £5, £10, £15, £20, £25, £30, £35, £40, £50, £60, £70, £80, £90, and £100 or more)	Questionnaire based on payment card (categorical scale)	62 of 582 (11%) of respondents placed zero value on a cervical cancer screening test Mean willingness to pay for cervical cancer screening service was £50.20, and median willingness to pay £45.00.	Medium	2*

Table 4-6. Text-In-Context Preferences, Organized by Country and Risk of Bias. Decision: Other Types of Decisions (n=2)

Citation	Country	Context	Decision options	Method of Eliciting Preferences	Text-in-context summary of preferences	Risk of bias Preference elicitation	MMAT score
Cadman, 2015 (64)	England, UK	185 Hindu women living in London returned a survey (92% Indian), 23 Hindu women between 14 and 70 years who were living in London participated in 4 focus groups	Type of self-sampling device: - Evalyn brush - Dacron swab	Questionnaire + focus group	Comparing between two types of self-sampling devices, the Dacron swab seemed to be more appealing because of its size and simplistic look.	Critical	2*
Szarewski, 2009 (61)	England, UK	28 Muslim women were recruited from a community centre to participate in 3 focus groups	Type of self-sampling device: - Qiagen swab - Pantarhei device	Focus group	The Qiagen device was preferred over the Pantarhei device because it looked “easy”, and “more friendly”. Authors noted that approximately two-thirds of respondents preferred to have a cervical screen completed by a doctor (compared to self-sample). All women indicated a preference for the Qiagen device, and two respondents were open to either option.	Critical	3*

Introduction to Section 2: From understanding perspectives on preferences to improving the measurement of women's preferences for cervical screening

In the first section of my thesis (Chapters 2 to 4), my goal was to understand different perspectives on preferences for cervical cancer screening. I interviewed guideline developers, administrators, health care providers, and women eligible for cervical cancer screening, and examined the research literature to synthesize what is already known about preferences in cervical cancer screening.

In Chapter 2, I found that guideline developers, health professionals, and researchers consider that self-sampling is a potential strategy for reaching marginalized populations who do not access cervical cancer screening. Of the nineteen interview respondents, most felt that some marginalized populations of women would be more likely to participate in cervical cancer screening if a self-sampling option were made available. However, some policy-makers and researchers were concerned that existing participants (“compliers”) in cervical cancer screening might prefer self-sampling and change their behaviour. A shift from provider-administered screening to self-sampling could have implications on the population-level effectiveness of an organized cervical screening program, as well as participation in preventive practices such as sexually transmitted infection testing. Respondents emphasized a need to appropriately communicate any program changes to providers and the public. This chapter highlighted that guideline developers, health professionals, and researchers have mixed opinions about how the preferences of women and providers can be incorporated into cervical screening programs, if at all.

In Chapter 3, I focused on understanding the information that women need to choose their preferred cervical screening modality. Interview respondents indicated that in the Ontario, women do not perceive that they face a decision for cervical cancer screening. Consequently, if future cervical screening guidelines and practice incorporate women's informed and values-based preferences, women must recognize that there are multiple options available including the option to opt out. Once women recognize that they can choose how to participate in cervical screening, they can explore the risks and benefits of the options to help them identify what characteristics matter most to them. If women in Ontario do not realize that they can choose to participate in cervical cancer screening, then there is need for support such that women can make informed decisions among the available screening modalities. This study identified the content and structure of a tool that would be helpful.

Having a better understanding of how stakeholders perceive women's preferences in cervical screening and their importance to practice and guidelines, I then examined the literature on women's preferences about cervical screening in Chapter 4. Across qualitative, quantitative, and mixed-methods evidence, the wide heterogeneity in objectives and in the methodological and reporting quality of the studies made it challenging to draw any conclusions about women's preferences in cervical cancer screening decisions. Women's preferences among cervical screening options were dependent on the underlying cervical screening context, the study design, and the preference-elicitation approach, including what options were offered and the corresponding information. Guideline developers will face challenges integrating relevant research into cervical screening guidelines, in Ontario or elsewhere, because the research evidence is variable and lacks rigour.

In the first section of the thesis, I learned that women in Ontario do not recognize that they face a decision to participate in cervical cancer screening, and that they can choose among

multiple modalities. To elicit women's informed, values-based preferences, the decision should be explicitly communicated, along with the benefits and risks of each option. I found notable deficiencies in the research literature when trying to synthesize what was known about women's informed preferences among cervical screening options. Women's preferences, as reported in scientific literature, are rooted in the screening context and location, and highly influenced by the method used to elicit the preferences. As a result, it is impossible to extrapolate the "prevalence of preferences" from the current body of literature (Chapter 4) to inform cervical screening guidelines and practice in Ontario, Canada.

Relevant and robust research is needed for the Ontario Cervical Screening Program to integrate women's preferences into the next iteration of cervical screening guidelines. The findings from the first section of this dissertation demonstrate a clear need for rigorous measurement of women's cervical screening preferences specific to the Ontario screening context. In the next section, I focus on methods used to elicit or measure women's cervical screening preferences. Chapter 5 presents an additional analysis of the current body of evidence on cervical screening preferences, specifically considering the robustness of the approaches to elicit preferences. Because previous preference-elicitation approaches were not validated and were embedded in specific geographic and socio-political contexts, I used the Ottawa Decision Support Framework to develop a new online preference-elicitation tool (Chapter 6), which was tested in a real-world setting. In Chapter 7, I propose a post-doctoral project that would rigorously investigate the implementation of this tool in a population-based study.

Chapter 5: Preference-Eliciting Approaches in Cervical Cancer Screening: A Systematic Review of Qualitative, Quantitative, and Mixed Methods Studies

This manuscript is prepared for submission to BMJ Open. I created the study protocol, developed the search strategy, conducted the search of the literature, conducted the screening and data extraction of relevant articles, analyzed and interpreted the data, and wrote the first draft of the manuscript and incorporated suggested revisions. All seven co-authors contributed substantially to study design, data analysis and interpretation, and made significant contributions to the final manuscript, which they approved.

Abstract

Objective.

The objective of this systematic review was to comprehensively review and critically evaluate approaches that have been used to elicit women's preferences about primary cervical cancer screening.

Methods.

MEDLINE, Embase, PsycINFO, CINAHL, and Cochrane Library were searched from inception to December 2016 for research studies describing qualitative, quantitative, or mixed methods approaches to describing or measuring women's screening preferences. Canadian grey literature sources were searched for relevant articles. Articles were included if the preference-eliciting approach met the Ottawa Decision Support Framework criteria. Methodological quality was assessed using the Mixed Methods Appraisal Tool. Risk of bias was further assessed using adapted criteria for studies of preferences and values. Relevant outcomes were summarized within the study context and any preference-eliciting effects were identified.

Results.

Twenty-eight articles reporting seventeen quantitative, ten qualitative and one mixed methods study were included. Investigator-developed questionnaires (n=10) and focus groups (n=10) were the most frequently used preference-elicitation approaches. Other approaches included surveys based on discrete choice experiments (n=4), conjoint analysis (n=2), payment card (n=2), and a best-to-worst scale (n=1). Five studies were considered to be at low risk of bias for preference studies.

Conclusions.

This systematic review found that preference-elicitation approaches for cervical cancer screening were heterogeneous in design and rigour. This study highlights a need for consistent definition of participant preferences and guidance for assessing quality across preference-elicitation approaches.

Keywords: Early Detection of Cancer; Choice Behavior; Decision Making; Patient Preference; Mass Screening; Uterine Cervical Neoplasms

Background

Cancer screening decisions are considered “preference-sensitive” because values will guide how individuals weigh the benefits of early detection options against the respective potential harms, such as invasive procedures, false positives and negatives, or out-of-pocket costs (2,3). Recent advances in technologies and screening approaches have increased the complexity of cancer screening decisions: when to start screening, what technology to use for screening, frequency of screening, and when to stop. In cervical cancer screening, most Canadian jurisdictions have promoted cervical cytology (the Pap test) as the only “valid” option for cervical cancer screening because of its observed success in reducing cervical cancer morbidity and mortality (4–6). Though this perspective is somewhat paternalistic, because women could have valid reasons for choosing not to participate in cervical cytology (7–11), it reflects the utilitarian values of policy-makers and guideline developers. Encouraging women to participate in early detection with cervical cytology might lead to a lower incidence of cervical disease and could contain health system costs by intervening with less intensive treatment earlier. As a result, Canadian cervical cancer screening guidelines reflect population health goals of guideline developers (12,13). Despite the emphasis on person-centered medicine, Canadian cervical cancer screening guidelines do not consider women’s preferences (14). Guidelines currently lack explicit reference to population preferences for cervical cancer screening and do not recommend opportunities for practitioners to elicit individual preferences at the point of care. Collective preferences for cervical cancer screening and the approaches used to elicit them will likely vary from individual preferences and the respective elicitation approaches (15).

Introducing new technologies into the menu of cervical cancer screening options may catalyze a shift toward person-centered cervical cancer screening guidelines and preference-based decision-making for individuals. A new primary cervical cancer screening test – human

papillomavirus (HPV) testing – is now recommended in Ontario, as well as in other jurisdictions, as the optimal method for primary screening (16–18). Though normally administered by a healthcare provider in a clinical setting, HPV testing may be carried out on self-collected samples (19). New primary screening modalities have different benefits and risks that may be valued differently by guideline developers, program administrators, and by women who are contemplating participating in cervical cancer screening. With these new modalities, there are opportunities to integrate women’s preferences for cervical cancer screening into clinical practice guidelines and provider-patient interactions, creating a more person-centered experience.

Previous systematic reviews (3,20) found that there were no randomized controlled trials that examined shared decision making in the cervical cancer screening context. Although RCTs are generally thought to be at the top of the evidence hierarchy, non-experimental studies may describe additional valuable tools for eliciting preferences. Other reviews have considered a more broad range of study designs, but have focused on specific populations (21) or specific elements of preferences (e.g., information comprehension (22)), or have aimed to increase screening (23), which is not consistent with a preference-sensitive approach, or have focused only on quantitative studies (24,25). A 2016 systematic review of stated preference approaches to cancer screening (26) and included only one study that focused on primary cervical cancer screening (27). However, this review did not include a quality appraisal of the included studies. Previous reviews were also limited to quantitative studies, which excludes qualitative approaches like focus groups and Delphi processes that are used to understand what matters to patients (24,25).

The objective of this systematic review was to comprehensively review and critically evaluate approaches that have been used to elicit women’s preferences about primary cervical

cancer screening. We describe approaches to preference-eliciting as they relate to evidence-based decision-making, at the meso-level (e.g., guideline development) that may consider the preferences of the general public, as well as at the micro-level (e.g., patient decision aids), which includes individuals who are facing a decision about cervical cancer screening. Findings from this systematic review might help guideline developers and policy-makers identify and select approaches to understand women's cervical cancer screening preferences. As a result, cervical cancer screening recommendations and practice might be more inclusive of people's preferences. We also identify outcomes that are relevant to eliciting preferences, including decisional outcomes (e.g. satisfaction with decision), health utilization/behaviour outcomes (e.g., uptake of screening), or clinical outcomes (e.g., incidence of cervical abnormalities).

Methods

The review protocol was registered with PROSPERO (CRD42016035737) and published (1).

Theoretical framework.

Our theoretical framework was the Ottawa Decision Support Framework (ODSF), a frequently referenced theoretical model for shared decision-making that defines preferences as a person's overall most-favoured option, taking into account the person's knowledge of and attitudes toward the characteristics of the different options (28).

Inclusion and exclusion criteria.

Studies using qualitative and quantitative approaches to elicit women's cervical cancer screening preferences were eligible for inclusion. Applying the ODSF criteria for "preferences" (28) we included studies reporting that:

- i) Participants realized there was a decision to be made (e.g., there were multiple options from which they could select their preferred option);

- ii) There was an information comprehension component that allowed participants to review information about cervical cancer, the screening tests, and the risks and benefits of each option; and,
- iii) There was a values clarification component that considered the person's attitudes to the options.

The inclusion and exclusion criteria are summarized in Table 5-1.

Table 5-1. Inclusion and Exclusion Criteria for Studies Used to Elicit Women's Preferences in Primary Cervical Cancer Screening.

Inclusion criteria	Exclusion criteria
- Participants: Women aged ≥ 18 years - Assessment of women's primary cervical cancer screening preferences, either current or hypothetical OR development of tool to assess women's primary cervical cancer screening preferences - Investigation of decision-making needs of women who are considering cervical cancer screening - Both qualitative and quantitative study designs	- Interventions targeting health care providers or policy-makers (e.g., program decision-makers) - Reports solely on technical aspects of cervical cancer screening or treatment - Reports of interventions that aim to increase cervical screening uptake - Studies reporting predictors of cervical screening uptake that are not related to a decision-making process - Opinion pieces - Studies that do not adequately report the study design, methods, and interpretation of the research - Studies that do not clearly demonstrate ethical conduct or ethical reporting of research - Studies in languages other than that could not be translated by the research team (i.e. English, French, Danish)

Information sources.

Full details of the peer-reviewed search strategy are provided in (1). MEDLINE, Embase, PsycINFO, CINAHL, and Cochrane Library databases, were searched for all articles from inception to December 2016. Grey literature was identified through ProQuest Dissertations and Theses and relevant Canadian health research/policy databases. For articles not published in English, French, or Danish, study authors were emailed to request a translation. Fourteen authors were emailed about translations of their articles; one author responded with a translation and another responded with a more recent article published on similar data.

Study selection.

Two of three reviewers (BW, SRVK, JE) independently screened each record and the full text of a potentially eligible study. Mendeley (www.mendeley.com) and Covidence (www.covidence.org) were used for citation management and full-text screening. The study team used DistillerSR to conduct methodological quality assessment and data extraction. Data were extracted by one reviewer and checked by a second reviewer. Disagreements regarding eligibility of studies were resolved by discussion.

Data extraction.

Pre-defined, pilot-tested data extraction forms were used to extract data in DistillerSR, with outcomes reported narratively, characterized by the study's context (29). Study characteristics (e.g., dates, population, location), preference-eliciting approach (e.g., ad-hoc questionnaire, discrete choice experiments), ODSF characteristics (i.e., whether options were communicated, information comprehension, values clarification), and outcomes (e.g., how the preference-eliciting approach was assessed) were extracted. Outcomes were categorized into measurements of validity/reliability and decision-relevant outcomes using the ODSF (e.g. decision quality, decision impact, decision action).

For quantitative approaches, validity and reliability were defined by conventional psychometric properties (30,31): validity is the extent to which the elicitation approach accurately measures what it intends to measure; validity refers to how close to the truth the measurements are, and whether the approach is eliciting the preferences it intends to measure. Reliability reflects the extent to which the elicitation approach produces consistent results; ultimately, reliability is concerned with whether the result is replicable. Interpretation of these concepts in qualitative research is less clear (31), as different paradigm assumptions affect what counts as "truth". In our study, "validity" was understood through an application of rigour, also

considered “trustworthiness” of the research report (i.e., “quality”). Reports of steps to verify the consistency of data (e.g. process notes, multiple reviewers) were extracted. Differences were resolved through discussion between the reviewers.

Quality assessment and risk of bias.

The Mixed Methods Appraisal Tool (MMAT) (32,33) was used to appraise the methodological quality of the eligible studies for this review, which were organized into randomized quantitative studies, non-randomized analytic studies, non-randomized descriptive studies, qualitative studies, and mixed methods studies. Two reviewers independently appraised studies included for full text review and came to consensus on the final score. Studies were included in the final review if they scored 2* or greater, according to the scoring scheme (32).

To address risk of bias issues specific to preference-eliciting methods, included studies were further assessed using domains outlined by Yepes-Nunez et al (34), whom we contacted to determine an approach to operationalizing the domains and items identified in their paper. With the guidance of the first two authors of the Yepes-Nunez article, we identified an assessment strategy for these domains, adapting each item to reflect ODSF-relevant features and outcomes. See Appendix C for an example of this adaptation. Each criterion could be assessed as “Yes”, “Probably yes (if unclear)”, “Probably no (if unclear)”, “No”, “Not applicable/relevant”. Extra weight was given to the domains “Was the instrument appropriately administered?” (e.g., was there a protocol to administer the instrument?), and “Was the choice of instrument optimal?” because the items in other domains were more concerned with general methodological quality, which was already assessed using the MMAT. Each study then received a preference-specific “risk of bias” rating using the following categories: “Critical”, “High”, “Moderate”, and “Low”. An example of the assessment tool and the associated scoring mechanism is included in

Appendix C. One reviewer (BW) assigned a rating for the preference-specific bias, and a second reviewer (SRVK or JE) checked this rating. Any disagreements were resolved through consensus.

Analysis.

A tabular summary was created to describe the objectives and design characteristics of the included studies. Evidence from quantitative and qualitative studies were summarized separately. These evidence tables describe the main types of preference-eliciting methods (35) identified in the quantitative and qualitative bodies of literature, respectively, and an assessment of the performance of these approaches. In these summary tables, we identified information comprehension and values clarification components. Any educational or informational material about cervical cancer, HPV, and preference options presented to participants that aimed to increase their understanding was recorded as an “Information comprehension” component. “Values clarification” components were categorized as *explicit* questions that asked participants to indicate how they personally feel about different attributes of the decision, or *implicit* questions that ask for participants’ thoughts on the social, psychological, or physical effects around decision features (28). A text-in-context summary column described the overall preference-eliciting approach in greater detail, and another column reported if and how validity and reliability were established for each preference-eliciting approach.

Behavioural, knowledge, and other impacts of eliciting preferences were categorized using the ODSF domains presented in Lillie’s 2014 systematic review (3), if relevant: Decision quality (includes measures of knowledge, value clarity, or preferred role in decision-making), decision impact (includes decisional conflict, decisional satisfaction, and use of services), and decision action (includes intention to screen, and screening behaviour). Because of philosophical differences between qualitative and quantitative research approaches, we looked for author’s

reports of women's responses and attitudes to the focus group instead of a "measurement" of decision quality, decision impact, or decision action.

We excluded clinical outcomes (e.g., cancer screening abnormalities) because the value of preference-elicitation is to promote or facilitate high quality individual-level decision-making (3), according to ODSF. Therefore, alternative definitions of preferences, including the "relative importance of clinical outcomes" (36) are out of scope for this review.

Results

Description of included studies.

The database searches returned 6,558 results, and 96 grey literature results were identified through additional searching. 663 full-text articles were reviewed for eligibility, and 131 were appraised using the MMAT. The ODSF criteria were applied after quality appraisal, which resulted in 28 unique articles reporting 29 studies: 17 quantitative, 10 qualitative studies, and 1 mixed methods study (i.e. two studies reported in one article: a qualitative study is embedded within a quantitative study). Figure 4-1 (Chapter 4) depicts the results of the screening process using a PRISMA diagram. Three quantitative articles (37–39) used the same discrete choice tool in their approach, but presented unique analyses in each article. Two other articles (40,41) reported preferences from the same sample population. See Appendix C for a list of articles that were not accessed.

Table 5-3 presents the characteristics of included studies (n = 29). Studies were conducted in ten countries – USA (11 studies), UK (8 studies), Canada (2 studies), Zambia (2 studies) and single studies from Australia, Cameroon, El Salvador, Ireland, Spain and Sweden. Among all studies, most (n=12) investigated preferences toward a specific modality (or location) of cervical cancer screening: HPV self-sampling (also called "home sampling") versus an in-clinic option (e.g. Pap testing, physician-administered, in-clinic). Eight studies considered

women's preferences about participating in cervical screening, and six studies presented decisions about hypothetical screening scenarios, which varied by different permutations of screening attributes (e.g. test, time to results, risk of false positives, location). Other decisions presented include type of screening test, willingness to pay, frequency of screening, and device used in self-collected sampling.

Quantitative approaches to preference-elicitation.

Of the 18 quantitative studies assessed, there were 2 randomized controlled trials (42,43), and 16 non-randomized studies (37–40,44–55) (13 analytic and 3 descriptive, according to the MMAT). Table 5-4 (page 207) presents detailed description of quantitative approaches used in preference-elicitation. A wide range of preference-eliciting methods was used, most frequently investigator-developed questionnaires (n = 10), with or without additional materials (e.g. decision aid, risk perception discussion, other educational material). Preference-elicitation questions included a single question with a checkbox, or multiple Likert-scale type questions. Questionnaires were usually paired with non-interactive information comprehension (e.g., static written information), but sometimes questionnaires were used after a decision-aid or an interaction with a health care provider. Values were clarified explicitly and implicitly. These studies most often addressed the acceptability of a novel screening option to women (n = 6).

Conventional “stated preference” approaches included survey tools based on conjoint analysis (n = 2), discrete choice experiments (n = 4), and payment card techniques (n = 2), either online or paper-based. Information was passively communicated to participants as part of the survey tool, though values were explicitly elicited by asking participants to indicate features of cervical screening that were most important to them. Most studies aimed to understand the relative importance of different characteristics of screening options, where the options were often hypothetical.

Most quantitative studies discussed how their findings in the context of future population-based recommendations, although the two randomized trials (42,43) and Alexander's ad-hoc questionnaire with decision aid (50), considered the preferences of individual women.

Qualitative approaches to preference-elicitation.

Eleven qualitative studies were analyzed, all of which used focus groups (41,49,51,56–64) as a method of inquiry, and one study used both focus groups and interviews (61). Table 5-5 (page 218) presents more details about the qualitative approaches used to elicit preferences. Ten of the qualitative studies also reported focus groups as their method of eliciting preferences (41,49,51,57–64), and one study (56) used focus groups to inform the development of a best-to-worst scale preference-eliciting tool. In this article, authors describe the development of a preference-eliciting tool, but do not assess how the best-to-worst scale tool influences individual outcomes. Authors in the latter study indicated that they aimed to identify preferences at a population level, though authors of the remaining qualitative studies did not communicate whether their approach was intended to inform policy or be used in practice.

Risk of bias specific to preference-elicitation.

Overall, five studies scored “Low” in risk of bias for preference-eliciting studies, while nine studies were considered “Medium” risk of bias. Ten other studies were considered to be at “High” risk of bias for preference-eliciting, and five studies were ranked “Critical”. One qualitative and four quantitative studies were classified as “Low” risk of bias according to the preference-eliciting domains. Seven qualitative studies were considered “High” or “Critical” for risk of bias, while eight quantitative studies scored “High” or “Critical”.

Ad-hoc questionnaires were most frequently associated with the greatest risk of bias because there was no evidence of standardized methods used to develop questionnaires, or questionnaires were not validated for the specific health decision. Focus group studies also

presented high risk of bias, which was usually associated with poor reporting of methods and minimal alignment to decision-making. Nine studies were included that used validated stated preference techniques in primary cervical cancer screening, including discrete choice experiments, conjoint analysis, card sorting, payment cards, and best-to-worst scale. These studies varied substantially in rigour and appropriateness. The five studies which presented the lowest risk of bias according to the Yepes-Nunez items (34) include a provider-delivered risk communication strategy (42), two surveys that used discrete choice to compare policy options with an opt-out option (37,53), an investigator- developed questionnaire that was tested for validity and reliability to examine acceptability of self-sampling among young women (54), and focus group discussions to understand important HPV communication priorities (58). Among these studies, the information comprehension component was not clearly articulated.

Validity and reliability of preference-elicitation approaches.

All but one (n=17) of the quantitative studies described the validity or reliability of the preference-eliciting approach. The mixed methods study (49) did not report any techniques to establish validity or reliability in the quantitative or qualitative study. Most often, studies (n=10) (40,42,43,46–48,50,51,53–55) cited the use of scientific literature and/or consultation with professionals to demonstrate validity in their approach. Eight studies (37–39,44,51–54) measured validity or reliability, most frequently observed in stated preference studies through tests of *a priori* model assumptions, or Cronbach's alpha measuring consistency of responses. One study (54) also measured temporal stability using an intraclass correlation coefficient to measure correlation in acceptability scores at different time points, and examined construct validity through exploratory factor analysis. Seven studies (38,40,43,48,51,53,55) conducted pilot tests or field tests prior to implementing in the reported study.

We observed major variability in how authors establish reliability and validity in the qualitative approaches. Only two of the articles (60,62) explicitly mentioned concept or data saturation. Included studies described “maximizing diversity” of their study samples, though most were unclear on what characterized a diverse sample or what criteria was used to assess the completeness of their data (65,66). Three studies pilot tested their preference-eliciting tools (56,58,59). Two studies (56,58) described using literature and/or expert consultation to help develop their preference-eliciting approaches, and four studies (60–63) described specific analytic approaches that intended to enhance the dependability of the research.

Impacts of using a preference-eliciting approach.

Twelve (38,40,42,43,45–48,50–52,55) of the 18 quantitative studies collected data on individual characteristics that we were able to map to ODSF categories, seven of which used ad-hoc questionnaires, two that used conjoint analysis tools, as well as a decision-aid study, payment card study, and one discrete choice survey. Five qualitative approaches (57,62–64) reported findings related to *decision quality* and two qualitative studies reported findings related to *decision impact*; *decision action* characteristics were not reported in any analysis. Most qualitative studies aimed to “understand” or “explore” women’s preferences, informational needs, and attitudes around cervical cancer screening, by incorporating information comprehension and values clarification components in the process. A summary of the impacts of using preference-elicitation are presented in Table 5-2. We were unable to synthesize the impacts within each category due to the heterogeneity in methods to measure or describe relevant outcomes.

Table 5-2. Impacts of Preference-Elicitation on Decision-Making Processes, According to the Ottawa Decision Support Framework.

	Description of concept	Number of studies reporting quantitative outcomes (n)	Examples in quantitative reports (n)	Number of studies reporting qualitative outcomes (n)	Examples in qualitative reports (n)
Decision quality	Desirable effect: to increase knowledge and values clarity	6	Perception of making an informed decision about cervical cancer screening Knowledge scores about cervical cancer and HPV	5	Reflexive interpretation of focus group discussion (values clarity, perceived awareness)
Decision impact	Desirable effect: to increase satisfaction, reduce decisional conflict, and have minimal impact on service utilization	3	Length of time to complete approach Health system utilization (compared to guideline recommendations)	2	Participant satisfaction with approach
Decision action	No desired effect – decisions to be made based on personal values and preferences: screening intention or behaviour	5	Intention to be screened Uptake of screening	0	NA

Discussion

This systematic review is the first study to describe approaches to eliciting women’s preferences for cervical cancer screening, examining the validity and reliability of different approaches, and what effects, if any, result from eliciting preferences. Using a mixed-methods review methodology, this review provides a comprehensive examination of diverse preference-eliciting approaches, with an explicit focus on validity and reliability. We used ODSF domains to reconcile differences in terminology and interpretations around “preferences” in the screening

process and then classify the effects of eliciting preferences. This study is also one of the first reports to operationalize risk of bias domains specific to preference-eliciting (34), and is the first, to our knowledge, to consider preference-elicitation in the context of decision-making.

Overall results.

Two of the most frequently used approaches to elicit women's cervical screening preferences were focus groups and ad-hoc questionnaires. Most questionnaire studies used non-interactive methods to communicate knowledge about options for cervical screening including information sheets and pamphlets. Attitudes toward the options were elicited explicitly as part of the questionnaire, or implicitly during the interview process. Most often, questionnaire studies asked women to consider a hypothetical screening scenario and select an option that was most preferred.

Focus groups were used as a qualitative research methodology as well as a method of eliciting preferences. Semi-structured interview guides elicited women's values implicitly, usually asking women to consider what features of options they might like or dislike, and information about options was communicated interactively (through an educational session with questions) or passively, through pamphlets and information sheets.

Nine studies reported stated preference techniques that used utility-based economic theory, including discrete choice experiments, conjoint analyses, payment card, and best-to-worst scaling approaches. These approaches focused on eliciting the relative importance of the features of different cervical screening options, which corresponds to explicit values clarification according to the ODSF. Information comprehension as part of these stated preference approaches is usually secondary and non-interactive, written into the survey tool. We observed that stated preference approaches were rarely associated with ODSF-relevant outcomes. Stated preference approaches examine how participants make trade-offs among cervical cancer screening options

based on their valuation of different health outcomes and are used to measure the “prevalence” of preferences in a population. ODSF outcomes separates the decision-making process from clinical outcomes.

Cervical cancer screening preferences are rarely addressed in literature on cancer screening preferences. A recent systematic review on stated preferences in cancer screening (26) identified two studies (27,51) related to cervical cancer screening, one that is included in this review (51) and one that was excluded (27) because it relates to follow-up options. In this review, researchers did not conduct a quality assessment of articles or instruments used, and authors aimed to inform health promotion opportunities instead of identifying valid and/or reliable preference-eliciting approaches for guideline developers. Two high quality systematic reviews affirm that there are no randomized controlled trials that investigate the effect of primary cervical screening interventions on decision-making outcomes (3,20). Our broader search strategy allowed us to consider additional studies of strategies to elicit women’s preferences in a decision-making context, but we were challenged to find a well-described approach that was robustly assessed. Data or concept saturation was rarely identified in the qualitative literature, suggesting that context plays a crucial role in our understanding of women’s cervical cancer screening preferences but also that the primary research may not provide a full picture of women’s preferences in cervical cancer screening.

Strengths and limitations.

Strengths of our systematic review include the explicit and broad eligibility criteria, a rigorous, peer-reviewed search strategy, and a detailed, theoretically-driven screening process to identify eligible articles. We also operationalized the recently-published risk of bias criteria for values and preference studies (34), grounded in ODSF (28).

There are some limitations with this review. Notably, reviewers encountered difficulty identifying whether studies elicited preferences because of differences between studies and authors in the wording used to identify “preferences”. The ODSF definition of preferences may not align with other conceptualizations of preferences, such as that used in “revealed preferences” studies, in which participants’ willingness to participate in a behaviour implies that they prefer that behaviour. Most articles reporting cervical cancer screening preferences lacked detailed description of “information comprehension”. This finding is consistent with a recent synthesis (68) which found that reports of patient decision aids – interventions that explicitly aim to improve decision-making encounters between a patient and their care provider – were missing key features.

We operationalized the risk of bias criteria for preference-eliciting instruments as described by Yepes-Nunez (34), but some criteria reflected the economic lens of most stated preference research, and consequently are less relevant for decision making. Items that may be important when investigating how preferences correlate with clinical outcomes are not applicable when examining behavioural, system utilization, or decision-making outcomes. Additionally, these items were not easily applied to qualitative studies; the concepts of *validity* and *reliability* are understood in different terms in qualitative research (e.g. dependability, credibility, trustworthiness), therefore we evaluate robustness differently in qualitative approaches compared to quantitative (31). Hence, the results of our preference-eliciting risk of bias assessment should be interpreted with caution.

Our protocol (1) stated that the GRADE guidelines would be applied to qualify the strength of individual findings for the quantitative and qualitative syntheses separately, using GRADE profiler and the CERQual tool respectively (72–75). However, the heterogeneous nature of the outcomes made the application of the GRADE process inappropriate.

Implications for research and practice.

A lack of research around cervical screening decision-making and preferences could be related to the strong public health enthusiasm for cancer screening generally (8,76–78), and specifically cervical cancer screening (79). Eliciting women's values preferences for primary cervical cancer screening is important for guideline developers tasked with integrating preferences into clinical practice guidelines, either with population level recommendations or an individual level through shared decision-making (28,80); approaches to preference-elicitation should be rigorously developed to ensure validity, acceptability, and neutrality (28).

Conclusion

This mixed-methods systematic review found that preference-elicitation approaches for primary cervical cancer screening were heterogeneous in the design and robustness of the approaches. We used an adapted risk of bias assessment tool for values and preferences studies to assess the credibility of primary study findings. Many studies failed to clearly report on what information was communicated to participants, and most studies used non-validated instruments to ascertain preferences. The risk of bias assessment tool was specifically developed for quantitative studies that associated individual utility with clinical outcomes, but our study also considered decision-making, behavioural, and health system utilization outcomes. Consequently, this study highlights a need for consistent definition of patient preferences, and broader guidance for assessing risk of bias across study designs and important outcomes. A rigorously developed, validated preference-elicitation tool might encourage the consideration of women's preferences in development of cervical cancer screening guidelines and in guiding person-provider encounters in primary cervical screening.

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Conflicts of interest.

BW's PhD research is supported by a Mitacs Accelerate grant with Eve Medical and a University of Ottawa graduate admissions scholarship. The funders did not play a role in the design or reporting of this research. BW has received an educational travel grant from Roche Canada (Research Canada partner) to attend a Research Canada meeting. These companies had no input on the design, conduct, or reporting of the research. For the remaining authors, none were declared.

Ethical considerations.

As denoted in the protocol(1), ethical conduct of primary studies was assessed during the screening stage of this systematic review. No individual-level data was reported.

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Table 5-3. Overview of Included Studies, By Study Type and Region.

Citation	Region, Country	Population and context	MMAT Score (4* = low risk of bias)	Risk of bias rating, specific to preference-eliciting
Quantitative, randomized controlled trials				
Holloway, 2003 [45]	Wales, UK	Cluster RCT of 1,890 women from 29 primary care practices in North Wales were recruited while waiting for their routine cervical smear (mean age 42 years)	3*	Low
Oscarsson, 2008 [46]	Southeast Sweden	133 women in Sweden between 28 and 65 years with diverse screening experience participated in the study group (400 randomized in total)	2*	Medium
Quantitative, non-randomized analytic				
Fiebig, 2009 [50]	New South Wales, Australia	167 women between 18 and 69 years from with different screening experiences (also examined clinician perspectives, not reported here)	3*	Low
Berner, 2013 [49]	Yaoundé, Cameroon	243 non-pregnant women in Cameroon between 24 and 65 years who participated in both self-collected and physician-collected HPV testing	2*	Critical
Rosenbaum, 2014 [54]	Paracentral, El Salvador	518 women who hadn't been screened in three years were recruited by community health promoters from four rural health units	2*	High
Arana, 2006 [47]	Gran Canaria, Spain	60 first-year female university students (mean age 22.6 years); compared with 60 female oncologists' with work experience at a large hospital	2*	High
Ryan, 2004 [42]	Scotland, UK	491 women eligible for screening in Scotland, between ages of 18 and 65	2*	Medium
Wordsworth, 2001 [41]	Scotland, UK	595 women eligible for screening in Scotland, between 20 and 59 years	2*	Medium
Wordsworth, 2006 [35]	Scotland, UK	577 women eligible for screening in Scotland between 20 and 59 years	3*	Low
Alexander, 2014 [48]	Missouri, USA	568 women aged 18–26 years old seeking care at an urban university student health and wellness clinic	3*	High
Kahn, 2005 [51]	Ohio, USA	116 women (82% black) from an urban, teen health centre, between 14 and 21 years*	3*	Low
Kidder, 2014 [55]	Florida, California, and District of Columbia, USA	476 Arab and Hispanic women, living in the USA, between 30 and 65 years	2*	Medium
McCormick, 2008 [46]	(unclear), USA	52 primarily white women between 22 and 78 years who attended an outpatient gynecology clinic participated	2*	Medium

Citation	Region, Country	Population and context	MMAT Score (4* = low risk of bias)	Risk of bias rating, specific to preference-eliciting
Penaranda, 2015 [56]	Texas, USA	110 Hispanic women between 30 and 65 years living on the USA – Mexico border who accepted an offer to participate in self-sampling	2*	High
Raab, 2002 [53]	Pennsylvania, USA	175 women who attended an American urban obstetric-gynecology clinic, aged 18-78 years	2*	Medium
Quantitative, non-randomized descriptive				
Cadman, 2015 [58]	England, UK	185 Hindu women living in London returned a survey, 23 participated in a focus group	2*	Critical/not useable
Anhang, 2005 [57]	Massachusetts, USA	172 low-income, inner-city women living in the USA, primarily Hispanic, over the age of 25	2*	Critical
Chamot, 2015 [43]	Lusaka, Zambia	208 Zambian women, between 18 and 49 years	3*	Medium
Qualitative				
Barata, 2008 [60]	Ontario, Canada	44 women aged 19 to 64 from 3 distinct settings (semi-rural, medium-sized city, large city) participated in 3 focus groups	2*	Medium
Howard, 2009 [61]	Ontario, Canada	77 immigrant women who lived in Canada for 1 to 5 years, aged 22 to 69 years participated in 11 focus groups	3*	High
McRae, 2014 [63]	Ireland	59 women between 17 and 69 years, recruited through primary care and well-women's centres, participated in 10 focus groups,	2*	Medium
Cadman, 2015 [58]	England, UK	23 Hindu women between 14 and 70 years who were living in London participated in 4 focus groups	3*	High
Szarewski, 2009 [65]	England, UK	28 Muslim women were recruited from a community centre to participate in 3 focus groups	3*	Critical
Jepson, 2007 [62]	Scotland, UK	31 women recruited from the cervical screening program, between 19 and 55 years, among 9 focus groups and 15 interviews (68 interview participants were recruited to speak about cervical, breast, and colorectal cancer screening)	3*	Medium
Anhang, 2004 [59]	Massachusetts, USA	Eight focus groups with 48 ethnically diverse, low-income women between the ages of 18 and 81 years	3*	Low
Wittenberg, 2015 [66]	Massachusetts, USA	8 focus groups with 42 women between 20 and 64 years who were recruited from a homeless medical respite center, an outpatient medical clinic, an emergency (i.e., overnight) shelter, a transitional housing program, and a residential substance use treatment program. Pre-tests conducted with 30 women	4*	High

Citation	Region, Country	Population and context	MMAT Score (4* = low risk of bias)	Risk of bias rating, specific to preference-eliciting
Smith, 2003 [64]	Michigan, USA	8 focus groups with 68 participants between 18 and 65 years who were participating in a risk-based screening trial	3*	High
Scarinci, 2013 [67]	Mississippi and Alabama, USA	9 African-American women from the Mississippi Delta were part of a discussion group, that was informed through community participatory research	3*	Critical
White, 2012 [44]	Lusaka, Zambia	60 women attending primary health care clinic who peer educators identified as eligible for screening, between 18 and 49 years, participated in 6 focus groups	2*	High

*Reviewers agreed to include this study even though some of the sample was outside the inclusion criterion of 18 years

Table 5-4. Results of Included Quantitative Studies.

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
Ad-hoc surveys, with or without additional components						
Anhang, 2005 [57]	Questionnaire	Modality and location of cervical screening (physician-administered at doctor's office vs self-collected at home vs self-collected at doctor's office vs none)	Research staff provided information to study participants, including HPV etiology, association with cervical cancer, and instructions for self-collection.	Explicit- Acceptability of self-sampling included questions about features that were "liked or disliked".	Draft survey instruments were pilot-tested among clinic patients for comprehension and content.	Decision quality [Knowledge]
Berner, 2013 [49]	Questionnaire	Modality of screening (self-sampling vs physician-sampling vs no preference)	Information sheet from the screening program	Explicit – part of the questionnaire using 4-point Likert scales: - Pain - Anxiety - Discomfort - Degree of relaxation - Confidence - Embarrassment (3-point scale)	“Acceptability scores were based on the methodology used in previous studies” (p.237) “The coefficient of reliability (consistency) (Cronbach >) was 0.72 for the assessment of physician sampling and 0.53 for self-HPV.” (p. 238)	Decision quality [Knowledge]
Cadman, 2015 [58]	Questionnaire (+ focus group)	Modality of screening (self-sampling vs in-clinic)	Part of focus groups. Participants given pamphlet, discuss what a smear test	Implicit - Part of survey and through focus groups: - Fear - Worry (pain) - Embarrassment	NA	NA

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
		Type of self-sampling device (Evalyn brush vs Dacron swab)	is, why is it done, is it important, for whom.	- Misinformation - Not eligible - Convenience - Importance - Cultural norms - Clarity - Social norms - Other beliefs		
Holloway, 2003 [45]	Risk communication session + questionnaire	Participation in screening (yes vs no) Frequency of screening (next Pap before 12 months or after 12 months)	Brief specific counselling session (~10 minutes) as part of standard smear appointment. Also includes risk communication tools with pictures and numbers.	Explicit - Psychosocial variables in questionnaire: - 5 Likert scales assessing screening-related anxiety/concerns - Spielberg State Anxiety Index - Mental health dimension of SF-36 - Impact of events scale	Risk score previously validated (p. 621), but not risk communication intervention.	Decision quality [Knowledge] Decision impact [Consultation Satisfaction; Psychosocial impact; Appropriate use of health services - timing; Cost of survey] Decision action [participation in screening, measured using national registry]
Kahn, 2005 [51]	Questionnaire	Modality of screening (self-testing versus clinician testing vs no preference)	“Standardized educational protocol about HPV and Pap tests”	Explicit – Survey measured acceptability with 13 items, using 3-point Likert scales: - Believe test result is correct - Trust the test result - Swab is not hard to snert - Proper specimen collection - Specimen collected from right place - Bother by procedure	Content validity measured by expert review and adaptation from previous surveys. (p 409) Test-retest reliability measured intraclass using correlation	NA

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
				- Uncomfortable procedure - Painful procedure - Unpleasant procedure - Embarrassing procedure - Feel in control of one's health - Believe testing is a good thing - Recommend testing to a friend	between baseline survey and final timepoint survey (ICC=0.82). (p 409) Internal consistency measured using Cronbach's alpha. (0.84-0.87 for self testing acceptability). (p 409) Construct validity assessed using factor analysis (p 410) (Mean self testing acceptability was significantly higher for those who indicated self-testing (32.3 vs 29.8))	
Kidder, 2014 [55]	Questionnaire	Intention to screen, if self-test available (Very likely to use it if the price was affordable; Likely to use it if the price was affordable; Not sure/no opinion; Unlikely to use it; Very unlikely to use it)	A one-page handout with information regarding cervical cancer screening and the self-administered HPV test was provided to all participants prior to beginning the survey or interview.	Explicit - Survey examined comfort, confidence, and concerns about using HPV self-sampling using 5-point Likert scale	"Survey and interview questions informed by previous surveys and questionnaires..." (p. 69) "The reliability and validity of this specific survey instrument in its entirety has not	Decision action [intention to screen]

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
		Modality of screening (at-home screening test versus in-clinic test) Willingness to pay (\$0, \$1-\$30, >\$30) Intention to use a urine-based screening test (Very likely to use it if the price was affordable; Likely to use it if the price was affordable; Not sure/no opinion; Unlikely to use it; Very unlikely to use it)			evaluated...”(pp 172-173) “To mitigate inter-rater reliability and maximize consistency, the same interviewer conducted all interviews.” (pp 172-173)	
Oscarsson, 2008 [46]	Questionnaire	Participation in screening (yes vs no)	Part of the interview process	Implicit – part of the interview, based on the investigator-developed Cervical Screening Questionnaire	“Content validity for the CSQ was affirmed by one group of staff members involved in the CCS program, and user-friendliness was affirmed by one group of a convenient sample	Decision quality [value clarity] Decision action [participation cervical screening]

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
					of women ($n = 15$) of various ages, and one group of 20 randomly selected non-attendees.” (p 186)	
Penaranda, 2015 [56]	Questionnaire	Modality (a question on test preference between cervical cytology and self-sampling; response options were 1, definitively prefer Pap; 2, somewhat prefer Pap; 3, prefer both equally or unsure; 4, somewhat prefer self-sampling; and 5, definitively prefer self-sampling)	Bilingual PowerPoint presentations based on the HBM were led by community health workers, using information from focus groups done in this population, literature and self-collection instructions. Women were prompted to participate in cytology if they were due at the end of the session (As self-sampling was for “research purposes”)	Explicit – questionnaire questions used Likert scales to assess preference for test attributes: - ease, - convenience, - pain, - privacy, - embarrassment, - accuracy of test, - confidence that test is properly conducted, - confidence in accuracy of test, - fear of harm.	“Survey items measuring constructs identified as being significant in previous focus groups and existing literature.”(p 324)	Decision action (intention to use self-sampling)
Rosenbaum, 2014 [54]	Questionnaire	Modality (self-sampling, provider-collected sampling, or no preference)	Government-run community education sessions	Implicit – open-ended question in questionnaire	NA	Decision quality [knowledge]

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
		Location (home, clinic, or no preference)				
Alexander, 2014 [48]	“[D]ecision-aid style” intervention + questionnaire	Intention to participate in cervical screening (yes vs no)	Cervical cancer prevention education was provided in a decision aid paper-based format.	Implicit	“The decisional satisfaction survey has been validated by Holmes Rovner and used for many medical decisions.” (p5)	Decision quality (feeling informed) Decision action (intention to screen)
Surveys based on discrete choice experiment						
Arana, 2006 [47]	Discrete choice experiment	Decision between two hypothetical policy alternatives, or opt out	In the questionnaire	Explicit - Attributes include: - Interval between tests - Probability of a false positive - Probability of dying from cancer - Waiting time for the results of the tests - Cost of the test	“These attributes and its levels were based on trial data, which were previously tested and refined in a focus group.” (p 513) “The cost attribute had four levels, which were derived from an open-ended pre-test study.” (p 513) Model assumptions (p. 514)	Decision quality [Ease of completion of survey]
Fiebig, 2009 [50]	Discrete choice experiment	Decision between two hypothetical policy alternatives	In the questionnaire	Explicit – Attributes measured: - Recommended screening interval	“The choice of experimental attributes and levels was informed	NA

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
		(based on context, and liquid-based vs conventional cytology), or opt out		- Familiarity with general practitioner (GP) - GP gender - Time since last test - Recommendation - GP incentive	by a literature review and the current Australian policy context, and tested in a pilot stage (Fiebig & Hall, 2005). (p 1767) Parameter estimates and standard deviations (how they align with expectations) (pp 1770-1771)	
Ryan, 2004 [42]	Discrete choice experiment	Decision between two hypothetical screening alternatives, or opt out	In the questionnaire	Explicit - Attributes of discrete choice experiment: - Time between screens - Time for results - Chance of being recalled - Chance of abnormality - Chance of dying - Cost of smear	“Internally inconsistent responses dropped, and individuals with missing values for their individual characteristics.” (p. 400) Parameter estimate signs matched with a priori expectations. (p. 401)	NA
Wordsworth, 2006 [40]	Discrete choice experiment	Decision between two hypothetical screening alternatives, or opt out	In the questionnaire	Explicit - Attributes of discrete choice experiment: - Time between screens - Time for results	Internal consistency measured using questions where one screening option was clearly “superior” to others (wrong answer	NA

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
				- Chance of being recalled - Chance of abnormality - Chance of dying - Cost of smear	implies failure to understand). If both consistency checks fail, individuals removed from analysis. (p 346). Thirteen attributes were found to be too many in pilot testing; randomly split questionnaire into six and seven questions. (p.345)	
Surveys based on conjoint analysis						
Chamot, 2015 [43]	Conjoint analysis	Hypothetical decision between clinics, based on visual screening and HPV screening characteristics	Trained female interviewers “briefly reviewed and compared” methods for participants, and part of questionnaire	Relative weighting of attributes in conjoint analysis: (1) visual screening or laboratory testing modalities, described solely in terms of visual inspection (yes or no), biological specimen (cervical/ vaginal sample, urine sample, or neither), and method of specimen collection (nurse-collected or self-collected); (2) examiner, defined by their gender and age; (3) option to view a digital picture of one's cervix; (4) travel distance to the clinic; and (5) wait time to receive	The process of survey development involved a review of the literature, consultations with 6 Zambian and foreign experts, and 8 focus group discussions with women and screening personnel. ^{5,6} We also conducted extensive field testing in local languages to ensure that choice questions were relevant and understood by the respondents (p 120)	Decision impact [Time to complete survey]

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
				screening results	“[R]estricted analyses to the 208 women who provided sufficiently reliable responses (root likelihood fit score ≥ 800).” (p 121)	
McCormick, 2008 [52]	Conjoint analysis	Two hypothetical programs (opting out is not an option)	Part of questionnaire	Explicit - Attributes in questionnaire: - frequency of screening - sensitivity of the test - specificity of the test - cost (co-payment) - whether the patient learns her HPV status or not - who performs the test and if a speculum is needed - whether the provider takes time to explain the results	“Pilot run of survey with 15 women allowed for modifications on wording.” (p. 9)	Decision quality (knowledge)
Surveys based on payment card technique						
Raab, 2002 [53]	Payment card	Willingness to pay (between \$0 and \$1000)	Part of questionnaire	Explicit - Willingness to pay out-of-pocket was elicited for the following risk levels: by having the new Pap test, the risk of dying of cervical cancer was reduced from 1:37,000 to (1) 1:50,000, (2) 1:100,000, (3) 1:1,000,000, and	Questionnaire was pilot-tested with 17 female hospital staff, and modified for clarity, brevity, and appropriateness of payment card range. Internal validity measured by association between	NA

Citation	Method of Eliciting Preferences	Decision(s) to be made (options)	Information comprehension	Values clarification	Measurement of validity or reliability	Effects of preference-eliciting approach (grouped by ODSF)
				(4) 0 (definitely would not die of cervical cancer).	willingness to pay and household income (no significant correlation observed). (p. 526) Construct validity measured when willingness to pay increases as perceived benefits increase, or with history of abnormal Pap test result (no significant correlation observed). (p. 526)	
Wordsworth, 2001 [41]	Payment card (categorical scale)	Willingness to pay (£0, £5, £10, £15, £20, £25, £30, £35, £40, £50, £60, £70, £80, £90, and £100 or more)	Part of questionnaire	Explicit - Attributes of discrete choice experiment: - Time between screens - Time for results - Chance of being recalled - Chance of abnormality - Chance of dying	“Payment card was reviewed after pilot study to ensure that a sufficient range of values was being covered” (p. 370) Methodological rigour tested with correlation between willingness to pay and income, age, previous smear status” (p. 370)	Decision quality [Ease of completion of survey] Decision impact [length of time to complete survey]

Table 5-5. Results of Included Qualitative Studies, All of Which Used Focus Groups to Elicit Preferences.

Citation	Additional Method to Elicit Preferences?	Decision(s) to be made (options)	Information comprehension	Values clarification	Description of reliability, validity of approach (e.g. data saturation)	Effects of preference-eliciting approach (grouped by ODSF)
Anhang, 2004 [59]	No	Modality of screening (In-clinic vs self-sampling) Type of screening test (HPV test vs Pap test)	Before discussion, focus group facilitators provided unstructured education about HPV and recommended screening practices	Implicit as part of focus group “Imagine that you have no symptoms, have a normal Pap smear, and receive screening test results that say that you have HPV. How would you feel?”	“Open-ended questions and paragraph messages were formulated based on previously published HPV knowledge research and lay literature on HPV” (p. 317) No criteria for saturation	NA
Barata, 2008 [60]	No	Modality of screening (self-sampling vs in-clinic)	Educational presentation + question period	Implicit - part of focus group (Open-ended questions about beliefs and perceptions of self-sampling)	“Topic guide and educational presentations were pilot-tested with four women and revised slightly before they were used in the focus groups.” (p. 252) No criteria for saturation	NA
Cadman, 2015 [58]	No	Modality of screening (self-sampling vs in-clinic)	Information leaflet + self-collection kit instructions	Implicit - part of focus groups (Open-ended questions about perceived benefits and barriers)	No criteria for saturation	NA

Citation	Additional Method to Elicit Preferences?	Decision(s) to be made (options)	Information comprehension	Values clarification	Description of reliability, validity of approach (e.g. data saturation)	Effects of preference-eliciting approach (grouped by ODSF)
Howard, 2009 [61]	No	Modality of screening (self-sampling vs in-clinic)	Group activity of drawing women's organs on outline of woman's body; video of a doctor explaining a Pap test to a patient.	Implicit - part of focus group (reasons to use self-sampling, reasons to avoid self-sampling)	"Independent coding to ensure consistency..." (p. 366) Authors mentioned common themes between groups, which "suggests saturation of information" (p. 368)	NA
Jepson, 2007 [62]	interviews	Participation in screening (yes vs no)	General information discussion + prompt cards	Implicit - part of focus group (how important the information was, how it might affect decision to get screened or not)	"[C]onstant comparative approach [helped] to ensure the text coded in the themes was well grounded in the data." (p. 893) "Negative and deviant cases were explored ... thereby adding confidence to the results." (p. 893) No criteria for saturation.	Decision quality (knowledge, emerging theme)
McRae, 2014 [63]	No	Type of screening test (HPV test vs Pap test)	Information sheet plus group discussion	Implicit - part of focus groups (discussion on advantages and disadvantages of screening, and psychological impact of different options)	"Ongoing and iterative thematic approach [helped] ensure sufficient depth was reached." (p. 3) "To ensure validity of coding categories and provide analytic rigour, two experienced qualitative researchers	Decision quality (knowledge and value clarity)

Citation	Additional Method to Elicit Preferences?	Decision(s) to be made (options)	Information comprehension	Values clarification	Description of reliability, validity of approach (e.g. data saturation)	Effects of preference-eliciting approach (grouped by ODSF)
					independently reviewed transcripts and agreed on principal themes.” (p. 3) "Groups were organised until conceptual saturation was reached."	
Scarinci, 2013 [67]	No	Modality of screening (Pap smear vs. self-collected sampling)	Two educational sessions held with women over two days	Implicit as part of focus groups: discussion of experiences	“[D]iscussion groups with community health advisors on the best strategies to approach the community... was important to develop a participatory topic guide for the focus groups.” (p. 4) “The validity and reliability of the Health Belief Model is this type of setting and outcomes needs to be addressed in future studies...” (p. 7-8) No criteria for saturation.	Decision quality (value clarity) Decision impact (comfort with process)
Smith, 2003 [64]	No	Participation in screening (yes vs no) Frequency of screening	Focus group facilitator gave orientation to “health screening”	Implicit as part of focus group: “What are your thoughts about Pap smears?” (e.g., effectiveness, advantages and disadvantages, etc)	“After summarizing the results, [authors] returned to the field notes to assess the comprehensiveness and accuracy of data extraction.”	Decision quality (knowledge, via reflexivity)

Citation	Additional Method to Elicit Preferences?	Decision(s) to be made (options)	Information comprehension	Values clarification	Description of reliability, validity of approach (e.g. data saturation)	Effects of preference-eliciting approach (grouped by ODSF)
		(annual vs less frequent) Age range of participation (starting and stopping age)			(p. 205) No criteria for saturation.	
Szarewski, 2009 [65]	No	Modality of screening (in-clinic vs self-collected) Location of screening (family planning clinic, health centre, general practitioner, other) Screening device (Qiagen swab vs the Pantarhei device)	Focus group moderator recapped HPV and cervical cancer information	Implicit as part of focus group: - Fear of results - Worry about pain - Embarrassment - Misinformation - Not eligible - Convenience - Cultural norms - Social norms - Personal beliefs	No criteria for saturation.	Decision quality (knowledge)
White, 2011 [44]	Interviews	Participation in screening (yes vs no)	Lay peer educators	Implicit - part of focus group	No criteria for saturation.	Decision impact (patient satisfaction)

Citation	Additional Method to Elicit Preferences?	Decision(s) to be made (options)	Information comprehension	Values clarification	Description of reliability, validity of approach (e.g. data saturation)	Effects of preference-eliciting approach (grouped by ODSF)
Wittenberg, 2015 [66]	Best-to-worst scaling	Participation in screening (yes vs no)	Best to worst scaling tool: Explanation of the task within the tool	Explicit - Decision factors which women can choose the level of influence (from “biggest” to “smallest” influence) Attribute levels: Provider (Gender, Familiarity, Attitude), Setting (Acceptance, Cost), Test procedure (Timing/convenience, Explanation), Personal fears and barriers (Hygiene, Substance use, Delivery of results/Fear of results), Cross-domain (Support available)	“Qualitative data and clinical expertise were integrated to identify and select an experimental design and to determine attributes for inclusion in preference experiment.” (p. 456) “Prestesting of program functionality was conducted with non-homeless women (research staff).” (p. 459) No criteria for saturation in focus groups.	NA

Chapter 6: Development and Field Testing of a Tool to Elicit Women's Preferences Among Cervical Cancer Screening Modalities

This manuscript is prepared for submission in Patient Education and Counselling. I created the study protocol, led the development of the prototypes of the tool and incorporated revisions, developed the analysis plan, led the recruitment of participants, conducted the analysis, and wrote the first draft of the manuscript. All five co-authors contributed significantly to the revised prototypes of the preference-elicitation tool, design of the field test, made important editorial contributions and approved the final manuscript.

Abstract

Objective.

The objective of the present study is to describe the development and field testing of a preference-elicitation tool for cervical cancer screening, meeting International Patient Decision Aids Standards (IPDAS) quality criteria.

Methods.

We developed a tool designed to elicit women's preferences for cervical cancer screening modalities titled "Cervical cancer screening – it's your choice". The Ottawa Decision Support Framework and IPDAS systematic development process guided the design and we followed IPDAS criteria when conducting a field test in a real-world setting. We evaluated the feasibility, acceptability, balance, and ability to elicit women's informed, values-based preferences.

Results.

Twenty-five women participated in the field test. Participants were aged 20-63 years of age, and identified as predominantly white (88%), living in Northern Ontario (68%), and most had university education (75%). All participants rated the Information sections of the website as "good" or "excellent". Most participants (72%) found the length of the website as "just right" and 100% indicated that they would find the tool useful for making a decision. Over two-thirds (68%) of participants perceived the information in the tool as "balanced". Almost all (92%) of participants scored at least four out of seven on the knowledge quiz, and most participants (84%) selected their preference in an informed, values-based way.

Conclusion.

The field test of “Cervical cancer screening – it’s your choice” highlighted the ability of the tool to elicit women’s informed, values-based preferences among available cervical cancer screening modalities. Further research can be done to elicit the distribution of preferences of cervical cancer screening modalities in the Ontario population and to evaluate the tool’s ability to elicit women’s informed, values-based preferences compared to educational materials

Background

Many organized cervical cancer screening programs are considering molecular tests for oncogenic types of human papillomavirus (HPV) as a replacement for the longstanding cervical cytology test (also known as the Pap test) (1,2). Thus, the focus is changing from early detection of cellular changes to testing for sexually transmitted infections that are associated with high-risk of cervical neoplasia (1,3). With an increased sensitivity to detect precancerous lesions compared to the Pap test, but a slight loss of specificity (4), many guidelines recommend HPV testing in part to address an expected decline in performance of Pap testing due to HPV vaccination (2,5,6). HPV testing can be performed on self-collected vaginal samples (1,7), which potentially would enable women who face barriers in accessing cervical cancer screening with a health professional (1,8). Overall, HPV testing of self-collected vaginal samples has slightly lower sensitivity and specificity for cervical precancerous lesions compared to HPV testing of cervical samples collected by a health provider (although this may vary by type of HPV test) (9). The “best option” among these modalities, and the option to opt out of cervical screening all together, is value-dependent: cervical cancer screening stakeholders must make trade-offs between the perceived risks and benefits of each option in selecting the preferred option (3,10,11). Subsequently, women eligible for cervical cancer screening, health care providers, program administrators and guideline developers could vary in how they value the characteristics to each option.

In Ontario and other Canadian jurisdictions, cervical cancer screening recommendations do not explicitly refer to women’s preferences for screening (12,13) despite the potential importance of different values and preferences. Specifically, guidelines do not allude to population estimates of preferences and there are no recommendations for providers to elicit preferences from women at the point-of-care (14,15). To be consistent with the person-centered,

evidence-informed vision of high quality health care systems (1,16,17), cervical cancer screening guidelines and practice would ideally integrate women's informed, values-based preferences. While there has been some research into women's preferences of cervical cancer screening modalities (18,19) the approaches to elicit preferences are heterogeneous in how they are developed and evaluated (Chapter 5). Moreover, it is unclear how cervical cancer screening practice and recommendations can most appropriately incorporate women's preferences. Increasingly, guideline developers are encouraged to present population-based patient preference data in guidelines, though an alternate strategy might involve eliciting personal perspectives during clinical encounters (10,14,20).

Clarifying values and eliciting individuals' informed preferences are an important step in a high quality decision-making process (21,22). Decision support tools, such as decision aids, can facilitate preference-elicitation by providing probabilities on the benefits and risks of the options, then assessing the participant's view on what matters most of them (21,23). To complete a shared decision-making process, an individual works with a health care provider to consider additional resources needed for decision-making, and moves toward a final decision (21,22). The International Patient Decisions Aid Standards (IPDAS) (24) provides a framework for developing, certifying, and evaluating high quality decision aids (25,26), offering specific criteria to design and produce tools that facilitate balanced, informed decision-making. We used these criteria to develop a person-centered and evidence-informed preference-elicitation tool to elicit women's preferences among available cervical cancer screening options (10,25). The tool, once refined and validated, could be used at the individual-level to help individuals understand the decision that they face (Chapter 2; Chapter 3) prior to the encounter with a health professional (1,27,28). This tool could also be used in research surveys to determine the

“prevalence” of different preferences in a population, which could inform clinical practice guideline panels.

The objective of the present study is to describe the development and field testing of this tool using the International Patient Decision Aids Standards (25,29).

Methods.

We developed a tool designed to elicit women’s preferences among cervical cancer screening modalities titled “Cervical cancer screening – it’s your choice”. The ODSF (21,30) and IPDAS systematic development process (25,31) guided the design and we used IPDAS criteria to evaluate this tool in a field test in a real world setting (26,29).

Conceptual frameworks.

Ottawa Decision Support Framework.

The Ottawa Decision Support Framework (ODSF) specifies the key components of a decision-making process for health decisions (30,32). This framework contains three overarching elements: decisional needs, decision support, and decision quality (30,32). According to the ODSF, decision support helps individuals through five steps: i) Realizing a decision needs to be made; ii) Information comprehension; iii) Clarifying values and identifying preferences according to these values; iv) Considering resources, including social influences, that affect the decision; and v) Forming an action plan (21,28,30). We used the ODSF Personal Decision Guide (33) template to structure the tool logically.

Systematic development process.

International Patient Decision Aids Standards.

The International Patient Decision Aid Standards (IPDAS) Collaboration established a checklist of quality criteria for decision aids that includes categories focusing on essential

content (providing balanced information, presenting probabilities, clarifying values, and guiding deliberation and communication), development (systematic methods, balanced presentation, up-to-date and transparent evidence, and plain language), and evaluation (informed and values-based decisions) (24,29). We used the IPDAS decision aid development process (25), specifically considering online tools (31), to guide the development of “Cervical cancer screening – it’s your choice”. Figure 1 conveys an overview of the development process. An external researcher assessed the prototype using the IPDASi v4 prior to field testing (26) [(Appendix D). We have outlined a detailed description of the development process below which is presented in Figure 6-1.

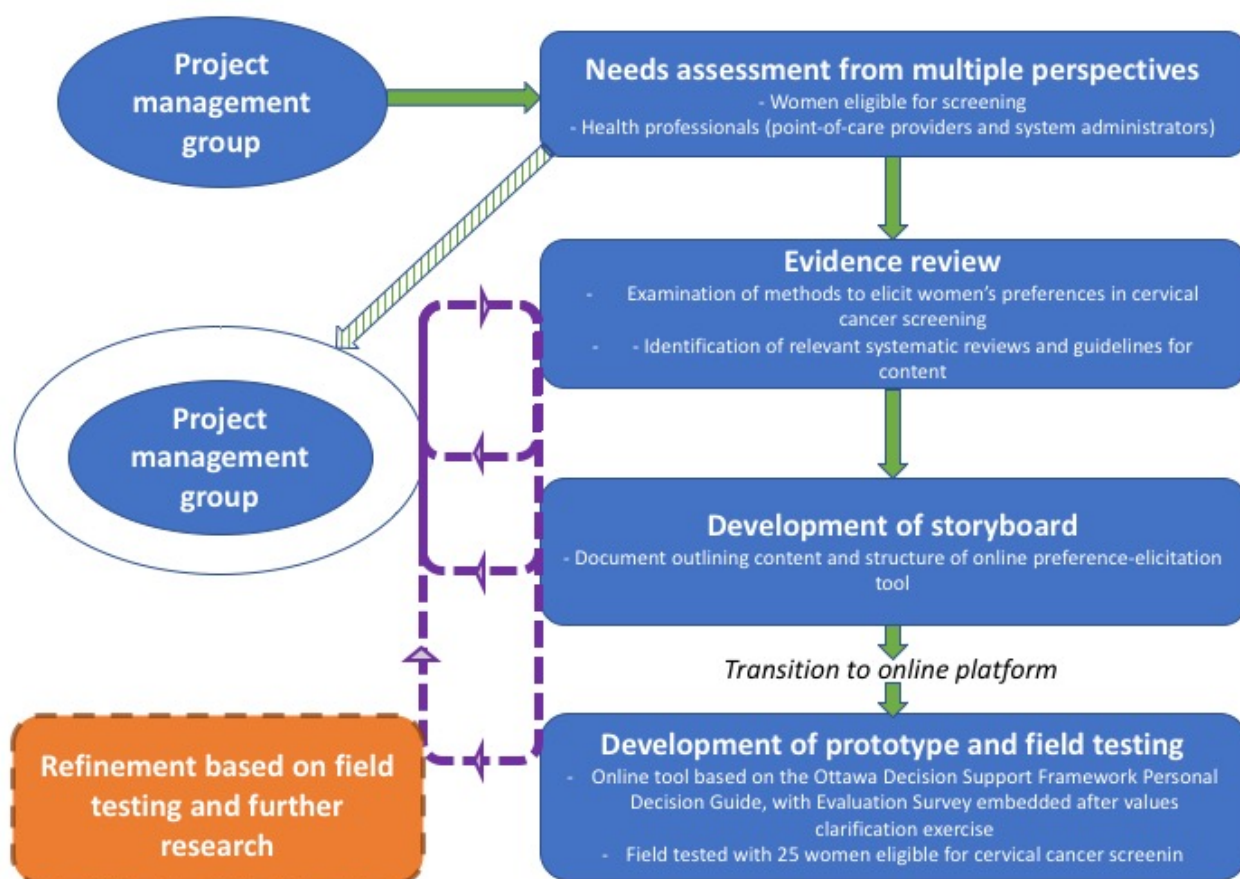


Figure 6-1. Systematic development process of “Cervical cancer screening – it’s your choice”

Project management group establishment and protocol development.

As part of a doctoral dissertation project, a project management group was established in 2015 to oversee the content development, design, and testing of a cervical cancer screening preference-elicitation tool. The six-member core group consisted of an epidemiology research trainee with experience in cervical cancer screening, four researchers with diverse expertise in cervical cancer screening, knowledge translation, epidemiology, and global health, and a regional cervical screening program lead. This group approved the project protocol and were actively engaged in the interpretation of the needs assessment and systematic review, whose findings were incorporated into the tool. The core team communicated at in-person meetings twice per year, and in monthly emails. Three women considering cervical cancer screening, an obstetrician-gynecologist, and a researcher with experience in cancer screening provided feedback on the clarity and overall feel of early prototypes of the tool, which was iteratively integrated into the website by the project lead. Advisors provided feedback to the research team through emails, phone calls, and in-person, one-on-one meetings.

Needs assessment and evidence review.

The needs assessment for this project involved a qualitative investigation of multiple perspectives on what women need to make a decision among cervical cancer screening modalities. Semi-structured interviews were conducted with women considering screening and analyzed using directed content analysis. Briefly, notable findings from the needs assessment study include the need to explicitly acknowledge that women face a decision to participate in cervical screening, particularly emphasizing that multiple modalities exist. While respondents identified that women considering screening and health care providers need more information on the screening options and the respective risks and benefits, respondents noted that additional system-level support would help them make preference-based decisions (Chapter 3).

An initial print version was developed in early 2017 that generally followed the ODSF personal decision guide template (33,34) and incorporated IPDAS content criteria (Table 6-1 presents the relationship between the IPDAS content and development quality criteria). We identified relevant evidence syntheses (4,9) and Ontario-specific resources (35,36) to inform the content of the preference-eliciting tool. We also provided a link to a Cancer Care Ontario tool within the risk assessment section (www.mycanceriq.ca/Cancers/Cervical) to offer a more personalized risk calculation for participants. The structure of the preference-elicitation tool is outlined in Table 6-2. The storyboard was reviewed by the broader project management group, whose feedback culminated in the development of an initial online prototype, a website [www.wix.com] that was web- and mobile-friendly.

Table 6-1. Using IPDAS Quality Criteria in the Development of “Cervical Cancer Screening – It’s Your Choice”.

IPDAS Criterion A: Content	
The patient decision aid provides information about options in sufficient detail for decision making	The tool describes cervical cancer, HPV, and cervical cancer screening. Cervical cancer screening modalities, including the option of not participating, are described in detail. The process, the cost, and provincial recommendations are outlined for each option.
If supporting a screening decision, the patient decision aid: - Describes what test is designed to measure - includes chances of true positives, true negatives, false positives, false negatives - describes possible next steps based on test result - include chances the disease is found with/without screening - describes detection/treatment that would never have cause problems if one was no screened	Differences between what the tests are designed to measure is highlighted in the “Know your options” section. A “What do my results mean” page presents permutations of outcomes depending on screening choice, with deeper detail about the implications of the finding and what suggested next steps are. Potential for overdiagnosis and overtreatment are described on this page. Positive and negative features are described in the “Know your options” section and the risk assessment page. The risk assessment page includes narrative and diagrams that communicate the chances of false positives, false negatives, true positives, and true negatives for each screening option.
The patient decision aid presents probabilities of outcomes in an unbiased and understandable way	Chance of cervical abnormalities, and potential outcomes of screening, are described using numbers and pictures, using 1,000 as the denominator. The pictures are presented side-by-side, so that participants can judge the relative efficacy. Factors that could influence the risk estimates were described in the preamble to the section (e.g., age, vaccination status, health behaviours, etc). Participants were suggested to try Cancer Care Ontario’s risk assessment tool, which provided

The patient decision aid includes methods for clarifying and expressing patients' values	more individualized risk estimates of cervical cancer based on personal characteristics.
The patient decision aid includes structured guidance in deliberation and communication	The tool uses an explicit values clarification method of five-start scales, as presented in the ODSF Personal Decision Guide (31). Characteristics of the options that participants were asked to assess were generated from the needs assessment. In particular, participants were asked to consider how important the following were to their decision: - Understanding risk of cervical cancer - Understanding HPV infection status - Costs of option - Avoid pelvic exam - Chance of false positives - Chance of false negatives
IPDAS Criterion B: Development	
The patient decision aid presents information in a balanced manner	Positive and negative features for each option are identified, and benefits are presented with opportunity to compare potential outcomes among all options.
The patient decision aid has a systematic development process	The systematic development process is described in detail in this paper, including a needs assessment, peer review, and field testing.
The patient decision aid uses up to date scientific evidence that is cited in a reference section or technical document	A reference list is provided, including updated systematic reviews and meta-analyses as well as relevant clinical practice guidelines. Date of publication and projected next update are presented.
The patient decision aid discloses conflicts of interest	The "About the project" section declares that authors have nothing to lose or gain by the preferences identified by participants.
The patient decision aid uses plain language	The tool includes illustrations and text written at approximately eighth grade reading level.
If patient decision aid is internet based: - Provides a step-by-step way to move through the web pages - Allows patients to search for key words - Provide feedback on personal health information that is entered into the patient decision aid - Provides security for personal health information entered into the decision aid - Makes it easy for patients to return to the decision aid after linking to other web pages - Permits printing as a single document	The webpages are enumerated "Steps 1 through 5" to provide clear guidance about how to move through the tool, and participants can return to the home page at any time. Participants can search for key words on most pages, and external websites are embedded in the website, or open in a new window. Information entered into the tool is appropriately secure. Print versions of relevant forms are accessible on the "About the Project" page.

Table 6-2. Structure and Flow of “Cervical Cancer Screening – It’s Your Choice” Website.

Section	Description
Overview	This page that explains the purpose of the tool and outlines each separate page of the tool. Participants can check their eligibility to use the tool on this page. After checking eligibility, the tool guides participants through the steps outlined by the ODSF generic decision support template, using backward and forward arrows. This page explicitly identifies that women eligible for using the tool are faced with a decision among cervical cancer screening options
Step 1: Explore information about cervical cancer and HPV	This page contains information about cervical cancer, HPV, and opportunities to protect from the disease.
Step 2: Know your options	This page contains information about cervical cancer screening generally, and then identifies the processes behind each of the four available modalities. “Step 2” also includes a “What do my results mean” page that presents a matrix of test type and potential results, with suggested next steps identified.
Step 3: Risks and benefits of options	This page contains diagrams illustrating potential outcomes of participating in screening. Participants can choose to visit an external page where they can calculate their cervical cancer risk using Cancer Care Ontario’s My CancerIQ tool (https://www.mycanceriq.ca/Cancers/Cervical).
Step 4: What do you prefer?	“Step 4: What do you prefer” page contains an embedded survey that was developed in Typeform. Based on the ODSF, this survey contains two components: an explicit values clarification exercise and a knowledge quiz that allows the participants to check their understanding of the material. The explicit values clarification exercise used the five-star rating scale that allowed participants to indicate their perceived importance of each decision characteristic. The knowledge quiz used true/false questions to checks participants’ familiarity with the material that was presented in the website. Finally, the tool includes a multiple-choice question that asks participants to select a preference among the available cervical cancer screening options (i.e. Pap test, provider-administered HPV test, self-collected HPV test, no screening, no preference). This question comes with open text comment boxes to allow participants to elaborate on their choice.
Step 5: What did you think?	This page presents the evaluation survey for the pilot study and will not be part of the tool post field testing. Participants were brought to this page after completing the values and preferences survey (“Step 4”). This survey asks participants to provide feedback on acceptability, presentation, and overall usefulness of the online tool. The survey also collects socio-demographic information from participants.
About the project	This page briefly describes the rationale of the project, update timelines, and how the site was developed. There is also a “Frequently asked questions” button that provides summary details on cervical cancer and HPV, the current Ontario cervical cancer screening context, and details about the project. References are provided in this section, as are print-outs that might be helpful for participants to guide conversations with their health care providers.

Field testing of prototype.

In line with the IPDAS systematic development guidance, we planned a field test in order to establish feasibility, acceptability, clarity and balance of presentation, and intended impact for

future use in a study of population preferences. We used the IPDAS Collaboration’s criteria to define our user testing criteria a priori, which included the following constructs: a) feasibility; b) acceptability; c) balanced presentation of information; d) clarity; and e) potential to have desired impact (i.e., women can identify their informed, values-based preferences). Two separate instruments that measured respondent’s knowledge and values clarity, respectively, were used as part of a composite measure (the Multidimensional Measure of Informed Choice) We also included a question that assessed women’s levels of anxiety. A further purpose of field testing analysis was to inform possible revisions to the decision tool before “real-world” testing in a larger population with a more rigorous study design.

Field test evaluation criteria/outcomes.

Table 6-3 presents the evaluation criteria, the associated measures and instruments used. We measured feasibility by tracking completion of processes by participants, such as unique website visits and successful completion of the preferences survey.

Table 6-3. Format and Psychometric Properties of Instruments Used in Field Testing of Preference-Elicitation Tool.

IPDAS Criteria	Scale	Format	Psychometric properties
Feasibility	Process measures: number of completions of survey and number of unique website visitors	Counts	Unknown
Acceptability, Balanced information, Clarity	Ottawa Acceptability Scale (39)	10 multiple choice items scored 1 “very/just right”, and 0 “somewhat/not at all”. Included open-ended comments.	Unknown
Intended impact (informed)	Ottawa Knowledge Scale (43)	7 multiple-choice items scored 1 (correct) or 0 (incorrect or unsure).	Internal consistency coefficients: 0.82, 0.83 Sensitive to change Discriminates between decision aids and usual care

Intended impact (clear values)	Decisional Conflict Scale (44), values clarity subscale	3-item version, using 5-point Likert scale from (i.e. “yes”, “probably yes”, “unsure”, “probably no”, “no”).	Test-retest and Cronbach alpha ≥ 0.78 Correlated to related constructs of knowledge, regret, and discontinuance Discriminates between known groups Sensitive to change Predictive validity
Intended impact	Multidimensional measure of informed choice (45)	Combines knowledge score (Informed – yes/no), values clarity score (values clear – yes/no), and whether preference was stated (did user identify modality in multiple-choice question – yes/no)	Knowledge component scale: alpha =0.68 Attitude component scale: alpha =0.78 Predictive validity: participants who made an MMIC “informed” decision rated their decision informed and high quality 6 weeks later Discriminant validity: lack of association between MMIC and anxiety
Clarity	Perceived Clarity (46)	1 multiple choice question, 4-item responses (“Everything clear”, “Most things clear”, “Some things unclear”, “Many things unclear”)	Unknown
Additional construct	Anxiety	1 question, 5-point Likert scale (“Very anxious”, “somewhat anxious”, “neutral”, “somewhat comfortable”, “very comfortable”)	Unknown

To measure the acceptability of the material and the perceived balance of information, we used the Ottawa Acceptability Scale (37). The evaluation survey included ten validated questions of acceptability, multiple choice and open-ended, of using the tool, the amount and clarity of information, the length of the tool, balance of information, willingness to recommend to others, and overall impression. We also added another question to elicit participant perceptions of clarity (38). Knowledge was assessed using seven question true-false questions about HPV and cervical cancer, using the ODSF Knowledge tool (33,39) (Appendix D contains Values and Preferences Survey). The total knowledge score was calculated as a proportion of correct results.

The values clarity subscale of the Decisional Conflict Scale (40) was used to identify whether participants were clear about what values mattered most to them. Potential to have desired impact was assessed using the Multidimensional Measure of Informed Choice approach, as described by Sepucha in 2013 (41), where informed choice can be measured through a composite score of knowledge, values, and a stated preference.

One additional question was added to the evaluation survey to understand participant anxiety levels as a result of using the tool.

Using IPDAS quality criteria (26,29) along with the scoring schemes of the instruments (42), IPDAS field testing criteria (29) states that field testing is successful when at least two-thirds of users:

- i) Access and use the tool. To determine the feasibility of use in the real-world, we examined the number of unique visitors who accessed the values clarification/preference elicitation exercise versus the number of visitors who used the tool. We also considered qualitative feedback from participants and project advisors.
- ii) Found the tool acceptable. Acceptability was measured in several instances and will be assessed as independent questions. In particular, we report the proportions of participants who:
 - a. rated sections of the website as “Good” or “Excellent”, respectively;
 - b. found the amount of information and the length of the information as “Just right”, respectively
 - c. found the tool useful.
- iii) Perceived a balanced presentation of options, such that the tool presented all options with equal detail.

- iv) Understood the information presented in the tool, which corresponds to a knowledge score of at least 4/7 (57%).
- v) Elicited informed preferences using this tool, consistent with the Multidimensional Measure of Informed Choice.

Ethical conduct.

The field test was approved by the Ottawa Health Science Network Research Ethics Board in February 2018. An informed consent landing page was added to the online tool, such that any visitors to the website would need to read the consent prior to viewing the rest of the material. This page described the background of the study, the study team, and what it meant to be a participant in the study. Specifically, participation involved completing an online session that asked site visitors to review information about cervical screening, indicate their hypothetical preferences, and provide feedback on the information content and the process of eliciting preferences. Surveys were anonymous and meta-data were not collected as part of the study. Visitors either agreed to participate in the study which led them to the Overview page, or declined to participate in the study, which led them to a standalone page that provided information about the study, and email address of the Principal Investigator (PI) if the visitor had any questions about the study.

Sample size.

There is little guidance in sample size calculations for usability and feasibility testing (29,43–45), and most studies justify their sample size based on logistical considerations (42,45–47). The IPDAS systematic development process, an appropriate field test sample size depends on the user testing and feedback elicited (e.g., “alpha testing”) during the iterative development process (25). Because we intend to further evaluate this tool in a larger, more rigorous study, the

study team determined that 25 women would provide sufficient preliminary insight into the delivery mechanism, acceptability, and usefulness of the tool.

Participants and setting.

Aligning with Ontario cervical cancer screening program eligibility criteria, participants included women (identified by sex assigned at birth) aged 21 to 69 years who live in Ontario, Canada. Women who were currently pregnant or identified as immunocompromised were not eligible. The project lead invited participants to view the website through social media posts (Twitter and Facebook) and through blog posts and newsletter distribution with Mitacs partner Eve Medical (a self-sampling device company in Toronto) beginning February 2018. Additionally, health care professional respondents from the needs assessment study were provided with a script to invite eligible patients to use the online tool. Participants were invited to share the outputs of the online tool and their experiences using the tool with their health care providers, although these interactions were outside of the scope of the preference-elicitation field testing.

Data collection and analysis.

Anonymized survey data were collected in the preference-elicitation tool using a web-based survey tool (www.typeform.com) embedded in “Cervical cancer screening – it’s your choice”. The number of unique visits was calculated in the Typeform survey tool was compared to the number of visitors who completed the surveys. Survey response data was downloaded in a spreadsheet, and further data cleaning and analysis were conducted using R Statistical Software 3.4.3. We described each multiple choice and check-box survey response using proportions, medians, and ranges.

A total knowledge score was calculated by summing the number of correct answers, expressing the total score as a proportion of correct answers (39). Participants were considered

“informed” if they answered at least four questions correctly out of a possible seven. Values clarification was assessed using three distinct questions (40), in which the questions were summed, divided by 3, and multiplied by 25 (where a high score reflects lack of clarity). Participants were considered to be clear about their values if their score was 33% or less (40). The Multidimensional Measure of Informed Choice was calculated by summing three dichotomous indicators (i.e. “informed”, “values clarity”, and a stated preference of one of the modalities). Open-text survey responses were analyzed using directed content analysis (48). Comments and suggestions from the survey were triangulated with the quantitative survey responses to summarize the field test findings.

Results

Between February and March 2018, 82 unique visitors accessed the Eligibility tool, 27 unique visitors accessed the first survey “Step 4: What do you prefer?”, and 25 unique visitors had accessed Evaluation survey (“Step 5: Tell us what you think”). We present the survey responses of the 25 women who completed both the Values and Preferences survey and the Evaluation survey.

Figure 6-2 illustrates how 25 site visitors were identified as the field test sample. Eighty-two site visitors indicated that they were female, and all but one confirmed that they were 18 years or older. Eleven site visitors were ineligible to use the tool because they resided outside of Ontario. Five visitors were excluded because they were not sexually active, and ten visitors indicated they had an abnormal Pap result in the previous three years. One woman who reported that she was pregnant and 3 women who indicated that they may be immunocompromised were excluded. Therefore, 51 (62%) of the 82 women who completed the eligibility survey were eligible to use the preference-elicitation tool. There were 27 participants completed the Values and Preferences survey (“Step 4”), and 25 participants who completed the Evaluation survey

(“Step 5”). All participants who started the respective surveys completed the surveys (100% completion rates).

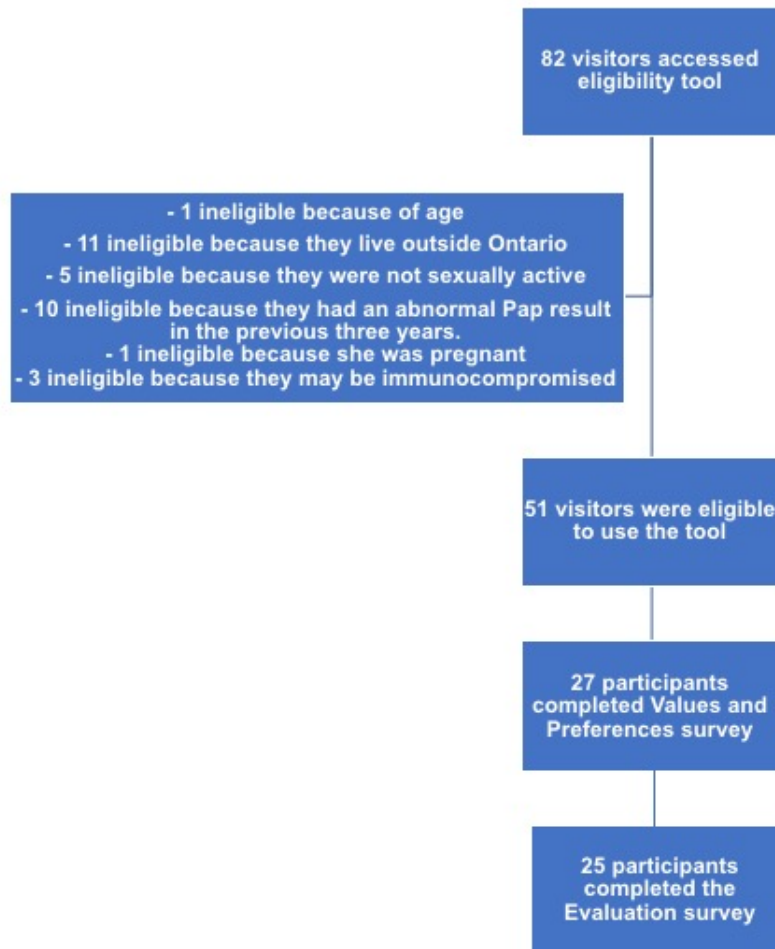


Figure 6-2. Participation in field test of “Cervical cancer screening – it’s your choice” by site visitors.

Socio-demographic characteristics.

The socio-demographic characteristics of field test participants are presented in Table 6-4. Eleven of the participants were younger than 30 years, and three women were aged 50 years or greater. The majority of participants described their ethnic background as White/European descent, and most respondents lived in Northern Ontario, Canada. The women in the sample

were highly educated, with three-quarters of the sample possessing a university degree. Twelve women indicated that they were presently married, and twelve women indicated that they were single, never legally married. All but one participant learned about “Cervical cancer screening – it’s your choice” from a friend or colleague.

Table 6-4. Socio-Demographic Characteristics of Participants Who Completed the Evaluation Survey.

Characteristics	Participants (Total = 25) n (%)
Age (years)*	31 (range: 20, 63)
Geography	
<i>Toronto</i>	3 (12%)
<i>Central/Eastern Ontario</i>	4 (16%)
<i>Northern Ontario</i>	17 (68%)
Ethnicity	
<i>White</i>	22 (88%)
<i>East Asian</i>	1 (4%)
<i>First Nations/Inuit/Metis</i>	1 (4%)
<i>Latin</i>	1 (4%)
Education	
<i>High school diploma or equivalent</i>	2 (8%)
<i>Post-secondary qualification, certificate, or diploma</i>	1 (4%)
<i>College diploma or university certificate</i>	
<i>below Bachelor level</i>	2 (8%)
<i>University degree</i>	20 (75%)
Marital status	
<i>Legally married</i>	12 (44%)
<i>Divorced</i>	1 (4%)
<i>Single, never legally married</i>	12 (44%)
Learned about study	
<i>From a friend or colleague</i>	24 (96%)
<i>Social media</i>	1 (4%)

*median (range)

Summary of field test results.

Revisiting the IPDAS criteria for a successful field test, Table 6-5 presents an overview of the results of the field testing of “Cervical cancer screening - it’s your choice”.

Table 6-5. Summary of Field Testing of Preference-Elicitation Tool Using IPDAS Criteria.

Criteria	Minimum requirement of criteria suggests that two-thirds of participants:	Result n (%)
<i>Website visitors, eligible to use the tool (N=51)</i>		
Feasibility	- Accessed and used the tool.	27 (53%)
<i>Evaluation survey (N = 25)</i>		
Acceptability	- Rated sections of the website as “Good” or “Excellent”.	25 (100%)
	- Rated the amount of information as “Just right”.	18 (72%)
	- Rated the length of the information as “Just right”.	19 (76%)
	- Rated the tool as “useful”.	25 (100%)
	- Felt “Neutral”, “Comfortable”, or “Very Comfortable” after reading the information provided in the tool (compared to “Anxious” or “Very Anxious”).	20 (80%)
Balanced presentation	- Perceived a balanced presentation of options.	17 (68%)
Understood the information presented in the tool	- Scored at least 4 out of 7 on the knowledge quiz.	23 (92%)
Clear values	- Scored 33% or lower on the decisional conflict values subscale.	24 (96%)
Stated preferences	- Were able to identify a preference or lack of preference.	24 (96%)
Ability to identify informed, values-based preferences	- Identified their informed, values-based preferences among cervical cancer screening modalities, based on the Multidimensional Measure of Informed Choice.	21 (84%)
	- Did <i>not</i> feel anxious based on the material in the tool.	20 (80%)
	-	

Feasibility.

Of the 51 site visitors who were eligible to use the preference-elicitation tool, 27 women (53%) completed the Values and Preferences survey (i.e., the interactive portion of the tool).

Acceptability, perceived clarity, and balance (n=25).

Almost half of participants (48%) found “Everything clear” on the website, and the majority of participants ranked the presentation of information on the different sections of website as “Excellent”, and virtually all participants ranked each section of the website as either “Good” or “Excellent”. Though almost one-quarter of participants (24%) found there to be too much information, the majority (76%) found it “just right”. Almost all participants (96%) found it easy to understand their risk of cervical cancer.

Approximately two-thirds of participants (68%) found the material balanced, though one-quarter (24%) of participants found the site slanted toward HPV testing. All participants (100%) indicated that “Cervical cancer screening – it’s your choice” would be useful in making a decision among cervical screening modalities.

Three themes stood out among the qualities that people liked about the online tool: a simple, balanced presentation of complex health information (n=11, 44%); the tool helps empower women and encourages them to be informed to make decisions about cervical cancer screening (n=10, 40%); and, the layout of the website was intuitive and easy-to-follow (n=7, 28%). While 14 participants (56%) did not provide any “suggested improvements”, 3 participants (12%) commented on wanting additional or different diagrams to convey risks of false positives and false negatives. Another 2 participants indicated that the information was too “wordy”, that perhaps visitors can choose to see “more information” rather than immediately being presented with all information by default. One participant (4%) suggested that videos might improve how information is communicated. Another participant suggested that “concrete

next steps” about how to follow up one’s preferences is important for making a useful tool. Participants also mentioned translations, in particular a French translation, for Ontario women (n=2, 8%), and one participant noted that information about symptoms of cervical cancer might be helpful for women contemplating cervical cancer screening.

Ability to identify informed, values-based preferences (n=25).

The Multidimensional Measure of Informed Choice is a composite measure of three indicators: knowledge (informed or not), values clarity (clear or not), and stated preference (made a choice among options or unsure). As presented in Table 6-5, this measure presents the independent elements of the informed choice measure and the proportion of participants who fulfilled each of the criteria. More than 90% of participants satisfied each of the respective criteria of informed choice –comprehended the information, recognized what characteristics were most important to them, and were able to state a preference. Twenty-one participants (84%) met the criteria for making an informed choice (41) among cervical screening modalities.

Clarity of values.

Most characteristics of screening options were rated as important by participants (Table 6-6). Almost all participants rated having a low number of false positives and a low number of false negatives as important or very important (92% each, respectively). Participants were concerned about avoiding out-of-pocket costs (88% ranked this characteristic as important or very important). Only three of the participants (12%) indicated that avoiding a pelvic exam was important to them.

Eleven participants (44%) responded to an open text question about other attributes that might influence their decision. These additional themes were categorized into five themes: enthusiasm and trust in early detection (n=4, 15%); personal experience with abnormalities or cancer (n=2, 8%); consideration about HPV vaccination (n=2, 7%); appropriate primary care

providers (n=1, 4%); and perceived low risk of cervical cancer (n=1, 4%). While some participants were quite eager about the potential to avoid a cancer diagnosis (“I would do anything to avoid”; “best possible outcome”), the participants who indicated a personal or family experience of abnormalities or cancer also suggested that they would “choose to participate” in cancer screening. One participant noted the importance of having the right primary care provider if they were to participate in cervical screening, commenting that they “don't know which providers work well with queer women.” The two participants who mentioned HPV vaccination presented conflicting interpretations: one participant preferred Pap testing because she had not been vaccinated previously because she thought it seemed “more accurate”, and the other participant indicated preferring an HPV test because they had been vaccinated. One participant indicated that cervical screening was not a priority because they’d been in a long-term, monogamous relationship without any previous abnormalities.

Table 6-6. Characteristics that Women Identified as Important or Very Important in Making a Decision Among Cervical Cancer Screening Modalities.

Characteristic	Important or very important (Total = 25) n (%)	Median score, range (higher indicates more important)
Know if you are at a higher risk of cervical cancer	21 (84%)	5 (2, 5)
Know if you have a high-risk HPV infection	19 (76%)	5 (1,5)
Low number of false positives	23 (92%)	4 (2,5)
Low number of false negatives	23 (92%)	5 (2,5)
Avoid paying out-of-pocket costs	22 (88%)	5 (2,5)
Avoid pelvic exam	3 (12%)	2 (1,5)

Based on the values clarity subscale used in the Evaluation survey, all participants were clear about what benefits mattered most to them. One participant (4%) indicated that they felt unsure about the risks that mattered most and which trade-offs were most important. Otherwise,

remaining participants (96%) were clear about which risks matter and how to consider trade-offs between the benefits and the risks.

Knowledge.

Overall, the median number of correct responses was 5 out of a total of 7 questions, ranging from 2 to 7. Fifteen participants (60%) scored 5 or greater on the knowledge quiz. Two participants (8%) scored 2 and 3, respectively, and four participants (15%) correctly answered all questions.

The question-by-question analysis is presented in Table 6-7. Most participants correctly answered that cervical cancer is caused by HPV, that it can be prevented through HPV vaccination, and that cervical cancer screening does not need to happen annually. Participants struggled with the questions asking whether Pap tests provide information about HPV status, and whether the HPV test has the highest rate of true positives.

Table 6-7. Results of Knowledge Quiz, By Question.

Knowledge of cervical cancer screening	Participants who correctly answer (Total = 25) n (%)
Cervical cancer is almost always caused by HPV (TRUE)	21 (84%)
Cervical cancer can be prevented through HPV vaccination (TRUE)	21 (84%)
The Pap test will tell you if you have an HPV infection (FALSE)	10 (40%)
A self-collected sample is worse than a provider (TRUE)	20 (80%)
Cervical cancer screening should be done every year (FALSE)	20 (80%)
Pap testing has the highest rate of true positives (FALSE)	10 (40%)
At any point in time, 20 of every 1,000 women have cervical abnormalities (TRUE)	19 (76%)

Stated preferences.

After working through the information pieces, values clarification exercise, and checking their knowledge, 24 participants (96%) identified a particular modality as their preferred method of cervical cancer screening. Two participants (8%) said that they did not have a preference, and one participant (4%) indicated that they were unsure. Almost half of the participants preferred the Pap test (n=12, 48%), one-third (n=8) of participants preferred in-clinic HPV testing, and two participants (8%) identified a preference for self-sampling. No participants selected “no screening” as their preference.

Unintended consequences.

Twenty participants (80%) indicated that they did not feel anxious after using the tool. Forty-eight percent of participants indicated that they felt at least somewhat comfortable after using the tool.

Discussion

As cervical cancer screening shifts to include HPV testing options, person-centered cervical cancer screening must reflect women’s preferences among the available modalities. We evaluated the feasibility, acceptability, balance, and ability to elicit women’s informed, values-based preferences for cervical screening in a field test of 25 Ontario women using an online preference-elicitation tool. The ODSF (21,30) and IPDAS systematic development process (25,31) and quality and evaluation criteria guided the design and field testing (26,29) of “Cervical cancer screening – it’s your choice”, respectively. Using input from women eligible for screening, health care providers, program representatives, and researchers throughout the development process, the final product aimed to support women to make informed, values-based choices.

Using IPDAS quality criteria, our field test of “Cervical cancer screening – it’s your choice” showed that more than two-thirds of field test participants found the tool to be acceptable and balanced, and the majority of participants made informed, values-based preferences among cervical cancer screening modalities. While most participants were able to move through all steps of the tool (as evidenced by the completion of the survey), only half of eligible website visitors actually worked through the interactive values clarification and knowledge component. This finding could suggest that website visitors were not prepared to work through the entire process or did not consider the preference-elicitation process (or the study) valuable for them. We will need to consider alternative strategies to promote the use of this tool among women, as most participants learned about the tool through word-of-mouth. Active recruitment by health professionals might be the most appropriate delivery method in future evaluation of a broader social media strategy might be appropriate given the community context of the target population. Additionally, involving a health professional in the preference-elicitation process might lead to a higher proportion of eligible women working through the values clarification component of the tool.

A small number of participants felt that the tool contained too much information on the first prototype or felt that the tool was slanted toward HPV testing, though the majority found the presentation was balanced. Several women commented that they appreciated knowing that there were different options available and thought the information would be useful to guide conversations with their health care provider in the future. These findings suggest that further refinements could improve “Cervical cancer screening – it’s your choice”, but the current prototype is a balanced tool that women find acceptable to identify their informed, values-based preferences.

Some researchers have suggested that valid, acceptable preference-elicitation tools might facilitate the integration of women's preferences into cervical cancer screening clinical practice and guidelines (10,14,15,49), because the tool promotes values-based decision-making in an evidence-informed way. "Cervical cancer screening – it's your choice" explicitly acknowledges that women face a decision to undergo cervical cancer screening, and if they choose to participate, they may choose different options because of different valuations of risks and benefits. We found in a qualitative needs assessment that some women do not perceive that undergoing cervical cancer screening involves making a decision (Chapter 3). We have addressed this gap by developing a tool that promotes intentional contemplation to undergo cervical cancer screening while supporting an autonomous, person-centered approach to health care. This tool can be used in future research within cervical cancer screening programs that are transitioning to an HPV-testing population screening approach (13). By providing recent evidence on the cervical screening modalities that are available, women who use this tool can compare the current options independently, prior to a clinical encounter. With a balanced, non-directive design, women might feel more confident to ask for their preferred option in an encounter with a provider, particularly if their preference diverges from the provider's preference (50–52).

Beyond the individual-level, we used a shared decision-making framework to develop an approach that can be used to elicit preferences among a population-based sample. Some experts are wary to apply the population preferences in individual encounters for cancer screening because extrapolating might be inaccurate or ignore individual's autonomy in decision-making (10,53,54). However, many researchers are currently investigating robust methods to elicit and synthesize preferences for use in population-level recommendations (14,15,55,56). "Cervical cancer screening – it's your choice" was designed to inform future person-centered cervical

cancer screening guidelines, and met the quality criteria of an individual-level decision tool. While most women in this sample indicated a preference for cervical screening with a health professional, women were divided between Pap testing and HPV testing. Some women indicated that accuracy was the most important quality of cervical screening, though a small number of women preferred screening based on self-collected samples. The variation in women's values and how they influence their cervical screening preferences will be an important factor for guideline developers to consider when developing person-centered recommendations.

Strengths and limitations.

To our knowledge, this study presents the first preference-elicitation tool involving primary cervical cancer screening that is based in shared decision-making concepts. Developed and field tested according to international standards (IPDAS) and a validated theoretical framework (ODSF), a diverse project advisory team ensured that the first version of “Cervical cancer screening – it’s your choice” was informative, appropriate, and useful. The IPDAS field testing criteria provided guidance about important characteristics of a preference-elicitation tool, focusing on feasibility, acceptability, and the overall objective of eliciting informed, values-based preferences. We used validated instruments from the shared decision-making literature to measure relevant outcomes, although none of the instruments had been applied in a cervical cancer screening context. Feasibility was the most challenging construct to measure, as we relied on counts of visits to the website and completions of using the tool. If assessing a wider distribution of this tool, we will identify additional strategies to engage with women considering screening. More purposeful social media campaigns and engagement with primary care providers might increase the public’s interaction with the tool or produce a more diverse sample. Field test participants were also highly educated and predominantly white. Because of the small, highly

selective sample, a more robust study design and diverse sample will be important to establish validity and reliability of the tool's ability to elicit informed, values-based preferences.

A strength of this preference-elicitation tool includes the online medium, which aligns with Ontario and Canadian focus on eHealth or digital health; this medium promotes accessibility to women outside of a traditional health care setting, such as women eligible for population-based screening. By focusing the website's content on the current Ontario cervical cancer screening context, participants could realistically deliberate the options, and perhaps use the information to guide conversations with their provider in the future. The field test identified some conflicting implications around the feasibility of using this tool in later research or in practice. Only 27 women completed the first survey although 51 unique visitors identified as eligible during the same time frame. As suggested by participants in the field test, further refinements should be made to the tool to allow users to choose how much information they see, identify explicit next steps once they have identified their preferences, and provide additional graphics and media (e.g. videos) to present the options and associated risks.

Conclusion

Field testing of "Cervical cancer screening – it's your choice" highlighted the ability of the tool to elicit women's informed, values-based preferences for available cervical cancer screening modalities. Further research should be done to elicit and abstract the distribution of preferences for cervical cancer screening modalities in a diverse population and to assess the validity and reliability of the tool in eliciting informed, values-based preferences. In particular, further research should investigate how this tool can be used by guideline developers who intend to generate preference-based cervical screening recommendations.

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Competing interests.

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Chapter 7: Preference-Based Cervical Screening in Ontario: Protocol for a Multiple-Methods Investigation of the Prevalence of Women's Preferences

The next step following the field testing of a preference-elicitation tool (Chapter 6) will be to refine it and conduct a study of its effectiveness for eliciting women's informed, values-based preferences among cervical screening modalities. In this chapter, I present a draft protocol for a three-year post-doctoral project in which I propose to work closely with decision-makers and policy-makers of cervical screening in a two-phase study. In the first phase, I propose to refine the preference-elicitation tool after consultation with guideline developer and policy-maker stakeholders. Then I will test the revised tool's effectiveness in a randomized controlled trial. Following the randomized controlled trial, the second phase of the study involves a cross-sectional investigation of women's preferences in three health regions. This study aims to determine what cervical screening modality different groups of women prefer and what personal characteristics are associated with their preferences, using a validated preference-elicitation tool.

Background

Health care providers, guideline developers, and policy-makers have opportunities to and should elicit and incorporate values and preferences of the people whom they serve in multiple ways: i) at an individual level (e.g., using shared decision-making); ii) at a population level (e.g., incorporating population preferences into clinical practice guidelines) (1). Cancer screening decision-making has become increasingly complex for individuals in recent years, as they contemplate whether they should participate in cancer screening for a range of different cancers. If individuals choose to participate in screening for a certain type of cancer, they will face decisions that include when to begin, how frequently to participate, and with what modality? Individuals making these decisions will make trade-offs between potential benefits and harms based on how they value the options. Making decisions about cancer screening is therefore entrenched in personal preferences, which might shift over time (Chapter 4).

After years of encouragement from public health campaigns, women are generally enthusiastic about early detection programs with cervical cytology testing (also known as Pap testing) (2–6). Now, cervical cancer screening programs in upper income countries are beginning to shift their primary screening programs from offering cervical cytology to offering human papillomavirus (HPV) testing (7). With increased sensitivity over the Pap test (8), HPV testing might be better at detecting “true positives”, especially in the HPV vaccination era (9). However, HPV testing also introduces a greater risk of false positives, particularly for younger women (10). While most evidence considers HPV testing on samples collected by a health care provider, HPV testing can be conducted on self-collected samples. This modality might be preferred by some women (11–13), though it has slightly lower sensitivity and specificity than HPV testing of samples collected by a provider (14). Many jurisdictions, including provinces and territories in Canada, are contemplating how to implement HPV testing strategies. Challenges arise in

changing the screening program as guideline developers and program administrators judge issues such as effectiveness and acceptability of different tests in different scenarios of uptake of vaccination, availability of financial and human resources, safety thresholds, and the roles of stakeholders (6). There is little rigorous research on the cervical screening preferences of individuals or populations (15; Chapter 4). Canadian cervical cancer screening recommendations do not explicitly incorporate women's preferences (16), despite a "person-centered" strategy (17,18). Some evidence suggests that robust local data might shift program-centric guidelines to recommendations that are preference-sensitive (Chapter 2), which aligns more closely with person-centered and evidence-informed philosophies. Furthermore, considering women's preferences in cervical screening guidelines might be associated with improved health and system performance outcomes (19,20).

The Ontario Ministry of Finance indicated that the province will fund HPV testing in the near future (21), suggesting that the Ontario Cervical Screening Program will require updated clinical practice guidelines. Integrating women's preferences among available modalities into the recommendations will help create guidelines that are person-centered and evidence-informed (17,18,22,23). Currently, Ontario women do not recognize that they face a decision among cervical cancer screening modalities (Chapter 3), let alone have sufficient information about options to make informed, values-based decisions (24). A recent systematic review (Chapter 4) found that women's cervical cancer screening preferences are widely distributed and dependent on the research study objectives, methods of preference-elicitation, and the screening context. As a result, the Ontario Cervical Screening Program guidelines should consider Ontario women's values and preferences for cervical cancer screening, given the options that the program is contemplating. This project aims to understand *how* can a preference-elicitation tool be used

within an organized program and *what* are the informed, values-based preferences of Ontario women who are eligible for cervical cancer screening?

In order to generate robust evidence to inform person-centered, evidence-informed cervical cancer screening recommendations and practice in Ontario, I propose to conduct a two-phase project. Over three years, I aim to:

1. Understand how Ontario cervical cancer screening guideline developers and relevant stakeholders want or intend to use a preference-elicitation tool.
2. Update the preference-elicitation tool based on current cervical screening context, new scientific evidence, feedback from field-testing, and engagement with stakeholders.
3. Determine the effectiveness of the preference-elicitation tool in a sample of women from a community health centre.
4. Describe the prevalence of cervical screening preferences across a diverse sample of women considering cervical screening.

To my knowledge, this study will be the first population-based study to investigate women's cervical screening preferences using a shared decision-making lens. I will collaborate with Cancer Care Ontario, Canadian Partnership Against Cancer, and Canadian Task Force on Preventive Health Care stakeholders to further refine the design of the tool and learn how the tool might influence preference-based cervical screening in Canada. Building a partnership with decision-maker and policy-maker stakeholders will be important for revising the tool in a way that acknowledges current policy and ensuring that study priorities align with those of the stakeholders. In particular, I will build upon the relationships with researchers, policy-makers, and guideline developers that I established during my PhD to initialize a project advisory

committee. This group will consist of interested members of the stakeholder organizations who will provide oversight to this project and assist with the implementation and dissemination as feasible. As part of the first phase of the project, the project advisory committee will oversee the updates to the preference-elicitation tool and assist with the interpretation of the results of the effectiveness trial.

In the second phase of this project, I will work with the project advisory committee to implement a population-based study to determine the “prevalence” of women’s preferences among cervical screening modalities. While I have proposed the design and outcomes of a cross-sectional study, the project advisory group will ultimately confirm the design of the population-based preference study. The proposed Phase 2 preference study is a suggestion based on my current understanding of the cervical screening context.

Objectives

Phase 1

1. To refine the preference-elicitation tool based on i) an updated literature review; ii) field test findings and feedback from stakeholders; and iii) current policy and health system arrangements.
2. To understand the perspectives of cervical cancer screening policy-makers, decision-makers, and relevant stakeholders in using a preference-elicitation tool to support preference-sensitive cervical cancer screening practice and recommendations.
3. To determine whether the preference-elicitation tool can elicit women’s informed, values-based preferences among a patient population of a community health centre.

Phase 2

4. To characterize the prevalence of women's informed, values-based preferences among available cervical screening options (cervical cytology, provider-administered HPV testing, self-collected HPV testing, no screening) in a sample of Ontario women.
5. To identify correlates of these preferences in diverse Ontario populations using a valid preference-elicitation tool.

Research questions

Phase 1

1. What perspectives do Canadian cervical cancer screening guideline developers have toward an online preference-elicitation tool for cervical cancer screening, and in what ways can the tool be integrated into future cervical screening practice or guidelines? (Objectives 1 and 2)
2. Does using an online preference-elicitation tool increase the proportion of women who can select their preferred cervical screening modality in an informed, values-based way? (Objective 3)

Phase 2

3. What proportions of Ontario women identify the following as their preferred cervical screening modality: i) cervical cytology ii) provider-administered HPV testing; iii) self-collected HPV testing; iv) no screening; v) no preference; or vi) unsure? (Objective 4)
4. What factors are correlated with these preferences (e.g. sociodemographic characteristics, clarity of values, knowledge of options)? (Objective 5)

Outcomes

Phase 1

1. A primary outcome of the qualitative study includes a description of method(s) that Ontario and Canadian guideline developers and relevant stakeholders intend to use to incorporate an online preference-elicitation tool into cervical cancer screening practice or recommendations.
2. Completion of a refined version of the preference-elicitation tool.
3. The effectiveness primary outcome is the proportion of women who identify an informed, values-based preference (using the Multidimensional Measure of Informed Choice) (25,26) among i) those who use the preference-elicitation tool compared to ii) women who access the provincial screening program website. Secondary outcomes will look at the knowledge scores, clarity of values scores among participants. Additionally, I set a feasibility target of >66% completion of the preference-elicitation process by eligible website visitors.

Phase 2

4. A primary outcome of the cross-sectional preferences study includes proportions of women in Ontario who indicate an informed, values-based preference for i) cervical cytology ii) provider-administered HPV testing; iii) self-collected HPV testing; or iv) no screening.
5. A secondary outcome of this study includes description of decision-relevant characteristics and decision outcomes (e.g. knowledge scores, values clarity, preparation for decision-making), and the associations with socio-demographic characteristics.

Theoretical Frameworks

A theoretical framework of acceptability (TFA) (27) will be used to understand how cervical screening guideline developers view the online preference-elicitation tool that aims to support women considering cervical screening. I selected this framework to expand upon some of the concerns raised by health care professionals (Chapters 2 and 3) about women making decisions among multiple cervical screening options. This framework presents acceptability as a multi-faceted concept that consists of seven foundational constructs: affective attitude, burden, perceived effectiveness, ethicality, intervention coherence, opportunity costs, and self-efficacy (27). This framework offers a theoretical foundation for understanding how guideline developers and program managers (or health care professionals and women considering screening) view the preference-elicitation tool. In Phase 1, I propose to develop a semi-structured interview guide according to the seven constructs of acceptability to assess how guideline developers and program managers perceive the online tool before it is used in a larger study. Subsequent revisions to the online tool following the qualitative interviews and conversations with the project advisory board should increase the acceptability of the tool among health system stakeholders.

The Ottawa Decision Support Framework (ODSF) guided the development of the online preference-elicitation tool (28,29) that will be used in this study. According to ODSF, decisional needs affect decision quality which influences actions or behavior. Decision support should therefore address the decisional needs to have an impact on health outcomes, emotions, and use of health services. The ODSF will be used to identify important decisional outcomes for this research project.

Design and Methods

Preference-elicitation tool

In this study, the preference-elicitation tool “Cervical cancer screening – it’s your choice” that was developed as part of my doctoral dissertation (Chapter 6) will be tested and applied in a larger setting. This online tool is based on the ODSF personal decision guide (30). Using this tool, participants can explore information about cervical cancer and HPV, available cervical cancer screening modalities, and the risks and benefits of these modalities. Participants then work through a values clarification exercise and can check their familiarity with the material, before selecting their most-favoured option.

Phase 1

Project advisory committee, tool refinement and qualitative interview study

A research coordinator will be hired within the first month to help execute the research and revise the tool (e.g., literature review, translations). Then, a project advisory committee will be formed by emailing or phoning representatives from provincial cervical cancer screening organizations (such as Cancer Care Ontario), as well as national organizations that including Canadian Partnership Against Cancer and Canadian Task Force on Preventive Health Care. After sharing project information (including the tool) with interested stakeholders, I will arrange an in-person meeting in the first two months to discuss the project objectives and studies. The initial meeting will focus on ways to refine the preference-elicitation tool to appropriately reflect current policy and funding contexts, then discussing the goal and design of the qualitative interview study. After this, monthly meetings (tele- or video- conference) will be arranged to continue the dialogue between stakeholders, discuss study findings, and assess the progress of the project. I will also ensure regular communication over email or through an online collaboration site (www.Slack.com), which will allow sharing of updated prototypes of the

preference-elicitation tool. While coordinating committee meetings, the research coordinator and I will also examine newly published scientific literature to refine the preference-elicitation tool's risk probabilities.

At the initial meeting, I will invite members of the project advisory committee to suggest key informants who could participate in interview study that examines how health system stakeholders (i.e. guideline developers and program administrators) perceive the preference-elicitation tool. I aim to recruit individuals from each organization who can speak to synthesizing evidence for guidelines, making cervical screening policy, and application or dissemination of guidelines. These stakeholders might include, but are not limited to, methodologists, physicians, politicians, and researchers. Once an updated prototype of the preference-elicitation tool is ready, I will conduct one-on-one semi-structured interviews with key informants based the TFA constructs (27). During these interviews, there will be opportunity for key informants to review the prototype of the preference-elicitation tool and offer feedback. Data analysis will occur in parallel with data collection, until the study team/data analysts determine that conceptual and meaning saturation are reached (i.e., no new themes or further understanding of the themes) (31-33). The research coordinator and I will use a content analysis approach (34,35) to deductively code the transcripts according to the TFA (27). Findings will be discussed at a project advisory committee meeting and final revisions to the tool's prototype will be made. A French translation of the tool will be created as well.

The revised tool's ability to elicit women's informed, values-based preferences ("effectiveness") among four cervical screening options (i.e. Pap testing, HPV testing performed by a health professional, self-collected HPV testing, and no screening) will be tested in a randomized controlled trial.

Effectiveness trial

The preference-elicitation tool will be tailored such that randomization occurs simultaneously with the consent process. Site visitors who indicate consent to access the tool will either be taken directly through the preference-elicitation process (“Intervention group”) or directed to the Cancer Care Ontario cervical screening information page (<https://www.cancercareontario.ca/en/types-of-cancer/cervical/screening>) (“Comparator group”).

The Cancer Care Ontario webpage presents guideline recommendations in a text-heavy format, with external links to health care provider websites. By randomizing at the point of website access, participants and study stakeholders are blinded to the allocation scheme.

This tool will be tested in a primary care patient population at the Sandy Hill Community Health Centre (CHC) in the Champlain Local Health Integration Network (LHIN) in Ottawa, Ontario. The Sandy Hill CHC provides comprehensive primary care services to individuals and families living in Central Ottawa, particularly focusing on marginalized populations. Sandy Hill CHC female patients who are eligible to participate in the Ontario Cervical Screening Program and have not had an abnormal Pap test result in the previous three years will be eligible to participate. This study population is appropriate as women typically access cervical screening through a primary care provider or through public health promotion programs. Testing the tool in a sample of women from diverse socio-economic and cultural backgrounds will be important for stakeholders who might be most concerned about the cervical screening preferences of hard-to-reach women. Promoting the preference-elicitation tool in a community-based model of primary health care might also be an acceptable delivery strategy, for women and health care professionals (Chapter 3).

The research coordinator and I will connect with Sandy Hill CHC health professionals who contribute to Health Promotion and Chronic Disease Management programs and the OASIS

program (a program that targets individuals involved with sex work) (36). Women will be invited into the study during regular programming and health promotion activities. Health professionals and managers will also be invited to join the project advisory committee to provide feedback on the execution of the trial or to offer suggestions about publicizing the tool in the next study. The study will be promoted passively through posters at Sandy Hill CHC site and through Community Engagement initiatives led by the CHC.

The primary outcome of the effectiveness trial is the Multi-dimensional Measure of Informed Choice (25,26) which is a composite measure that considers adequate knowledge (>66% on knowledge quiz), clear values (<33 on decisional conflict values subscale), and stated preference (>66%). The Cochrane review of trials of patient decision aids (37) suggests that individuals who use a decision aid are more than twice as likely to have congruence between the chosen option and informed values (relative effect of 2.06). The field test in Chapter 6 suggests that 84% of individuals who use the tool can make informed, values-based decisions. A sample size of 206 (103 per arm) was considered sufficient using a conservative estimate of effect (20% increase in informed, values-based decisions among the intervention group), at 90% power with 95% confidence. With approximately 18,000 residents in Central Ottawa belonging to a CHC and an emphasis on community engagement, it should be feasible to recruit this sample size within six months.

All outcomes will be measured through self-administered questionnaire responses. For the intervention arm, the knowledge and values questionnaire (the evaluation questionnaire) is a step in the preference-elicitation process. For the comparator arm, the questionnaire will appear as a pop-up window after participants are re-directed to the Cancer Care Ontario information page.

The proportion of participants who identified an informed, values-based preference in the intervention arm will be compared to the comparator arm using a two-sample t-test, based on the aforementioned calculation criteria. Means, medians, standard deviations, proportions, and respective 95% confidence intervals will be used to describe the knowledge and values clarity subscales, respectively. Meta-data from website visitors will be collected to assess the feasibility (i.e. the proportion of eligible website visitors who completed the tool). I will meet with the project advisory board monthly throughout the trial to discuss results or work through logistical challenges.

Phase 2

The second phase of this project will require that the preference-elicitation tool effectively elicits women's preferences. Furthermore, the project advisory committee must agree that understanding women's preferences at a population level is important for their roles (e.g., guideline development). In Phase 2, I propose a cross-sectional study to investigate women's preferences for cervical cancer screening in the Champlain LHIN, North West LHIN, and Toronto Central LHIN. These three LHIN regions will be included in this study to maximize variation and increase likelihood of reaching marginalized sub-populations: Champlain LHIN (Francophone, Inuit, some rural communities), Toronto Central LHIN (immigrants, urban setting), and North West LHIN (rural, remote, Northern, First Nations populations). As cancer screening programs are implemented and measured at the LHIN-level, LHINs provide logical boundaries for sampling and implementation purposes. Additionally, we will work with LHINs to engage with their respective Patient and Family Advisory Councils, sub-region clinical leads, and regional cancer screening leads and program managers to identify interested individuals to participate in the project advisory committee. These representatives will be invited to participate in monthly online meetings to discuss design, implementation, analysis, and interpretation

questions throughout the study, from the development of the ethics proposal to the interpretation and dissemination of results.

Sampling and recruitment

The design of this study will be confirmed by the project advisory committee after Phase 1 has completed (approximately 12 months into the project), so the final approach might be modified by stakeholders. I propose to use a quota-sampling approach instead of a population-representative sample to ensure that women from hard-to-reach groups are included in this study. In Canada, the burden of cervical cancer falls in populations of women who have not participated in screening for cervical cancer. First Nations, Inuit, and Métis women (38,39), certain immigrant populations (40,41), and low-income women (42) disproportionately experience barriers to cervical cancer screening. As a result, these populations might be more likely to experience benefits from alternative cervical cancer screening options (41) (Chapter 2).

Women who access health care in these LHINs will be eligible to participate if they are eligible for Ontario Cervical Screening Program and if they have cell phone or internet access. A sample of women will be recruited into the study passively (e.g., mailing lists, posters, and websites) and actively (e.g., with the assistance of local primary care providers). Our socio-demographic strata of interest are: age (i.e., <30 years, 30-49 years, 50-65 years), ethno-cultural (i.e., European descent, First Nations/Inuit/Metis, immigrant, other racialized groups, Francophone), and geographical (i.e. urban, rural). We will use similar sampling and recruitment monitoring strategies reported in a recent Ontario study (43). Email notifications to health and social services, municipalities and city councils, public libraries and various media outlets will be used to promote the study (and its website) to a broad audience of collaborators and potential participants within a one-year recruitment period (43). Using targeted recruitment techniques such as these will allow us to oversample from some marginalized populations. Previous

screening history (i.e., within three years, over three years ago, never, do not know), HPV vaccination status (i.e., yes, no) and previous screening results (i.e., abnormal/positive, normal/negative) will also be collected as clinical characteristics of interest.

Guided by the distribution of preferences across the field test sample (Chapter 6) and previous literature (11,44), we estimated that our prevalence of preferences might be distributed as: 5% not have a preference, 5% were not sure, 5% prefer to opt out, 15% prefer self-sampling, 40% preferred Pap testing, 30% preferred provider-administered HPV testing. Noting that the study sample is a non-probability sample, we estimate that 1400 women should produce a sufficient sample to detect between 5 and 10 percentage point differences in prevalence while accounting for at least one covariate (45,46).

Data analysis

Descriptive analyses of survey questions will present the socio-demographic characteristics, the personal values which participants identified as important in their decision-making process, and their preferred/intended option for cervical screening among the sample population.

To analyze correlates of women's preferences, we will construct separate logistic regression models for each of the six options listed in the preference-elicitation tool, in which the outcome is a preference for the respective modality (yes/no), and the covariates include socio-demographic characteristics (e.g., age, ethnicity, sexuality, LHIN of residence), method of recruitment into study, and decision-making characteristics (e.g., knowledge, values clarity, anxiety scores). First, we will examine bivariate relationships between the preferences and the other covariates. Then we will use stepwise regression techniques to investigate correlation and interaction between covariates. We will also construct a logistic regression with the

Multidimensional Measure of Informed Choice indicator as the dichotomous (yes/no) dependent variable, and we will look for socio-demographic correlates using the aforementioned approach.

Collaboration

This project will involve collaboration between the research team (e.g., those with research affiliations) and different knowledge users including guideline developers, LHIN and regional program managers, health professionals, and members of the general public. By collaborating with people directly involved with guideline development in Phase 1, we expect to facilitate consistent discussion among stakeholders about approaches to use a preference-elicitation tool. A trans-disciplinary project advisory committee will be important for assessing the applicability of a preference-elicitation tool in actual practice. For example, perhaps an effective tool is acceptable to guideline developers to understand preferences in a population but might require adaptation for health care professionals to find it useful. Additionally, an opportunity to share experiences and knowledge will help integrate the study into ongoing public health initiatives.

The regional program managers will have knowledge about their local cervical screening program as well as the provincial perspective through their roles. Having buy-in from these stakeholders from the start of this research will be important for understanding how regional programs work, the differences in the delivery and assessment of cervical screening between organizations and regions, and local community needs. The local perspective will also be supplemented with other health service provider and potential screening participant stakeholders, recruited through LHIN clinical leads and Patient and Family Advisory Councils. These knowledge users will offer their perspectives on how a preference-elicitation tool can be used in a cervical screening program and speak to the potential for preference-elicitation in provider-patient encounters. Collaborating with a diverse group of knowledge users will influence the

study objectives, design, and implementation strategies, and provide different lenses to interpret the study results.

Timeline

Phase 1	
0 – 6 months	Hire research coordinator Engage with guideline organizations about study: establish project advisory committee Establish partnerships with Sandy Hill Community Health Centre and begin discussions with three proposed LHIN partners Develop interview guide according to TFA Examine systematic reviews and scientific literature to update probabilities in preference-elicitation tool Ethics approval for qualitative interview study Begin key informant interviews
6-12 months	Ongoing monthly project advisory committee meetings: Discuss amendments to preference-elicitation tool, progress of effectiveness trial, and study design for cross-sectional study Ethics approval for randomized controlled trial Finish interviews and discuss findings with project advisory committee Finalize prototype of preference-elicitation tool for randomized trial Begin recruitment into effectiveness trial
12-18 months	Ongoing monthly project advisory committee meetings: Discuss interpretation of effectiveness trial results and design of cross-sectional study Complete recruitment of women into effectiveness trial Submit qualitative paper for publication Preliminary results of trial discussed with project advisory committee; study design finalized for cross-sectional study Ethics approval for cross-sectional study Email notifications about cross-sectional study to health and social organizations in three partner LHINs

Phase 2	
18-30 months	Ongoing monthly project advisory committee meetings: Discuss preliminary analysis of preference prevalence discussed with stakeholders, and opportunities to disseminate findings Begin and monitor recruitment into cross-sectional study Implement targeted recruitment strategies if needed to achieve response rates (e.g., health fairs) Complete analysis of effectiveness trial and submit paper for publication Present findings of effectiveness trial and qualitative study at Canadian conference for cancer control
30-36 months	Ongoing monthly project advisory committee meetings: Discuss dissemination materials and identify key messages for knowledge user groups Complete analysis of cross-sectional study and submit paper for publication Prepare reports for partner organizations, and organize webinar conferences to communicate results and identify next steps Share study findings and next steps with health care professionals through Up-to-Date, webinars and teleconferences, and relevant conferences Publicize preference-elicitation tool using email and social media via LHIN patient and family advisory councils, and regional health and social service organizations

Note: Study outputs are bolded

Dissemination

Guideline developers and policy-makers, including Cancer Care Ontario, Canadian Partnership Against Cancer, and the Canadian Task Force on Preventive Health Care, will be involved from the beginning, through the project advisory committee and as interview respondents. These stakeholders have important insight into how a preference-elicitation tool and corresponding research can be used to inform Canadian cervical screening practice. To ensure that the tool is acceptable from their perspectives and that the research is useful for their positions, I wish to involve them from the inception of the project. The primary engagement with

these organizations will occur through monthly meetings with project advisory representatives. The research coordinator and I will produce materials to summarize the results of the studies throughout the project, including reports, presentations, and peer-reviewed publications, using guidance from the project advisory committee. In particular, I will present the effectiveness trial results (and introduce the cross-sectional study) at relevant cancer control research meetings in Year 2 and in peer-reviewed publications. In the final six months of the project, a final report will be written for the guideline organizations that contains the results from the studies. Findings from the entire project will be shared with guideline developers and program administrators through project advisory committee meetings, relevant conferences, and in peer-reviewed publications.

Regional cancer screening program managers will also be engaged early on in this project, to ensure that the studies appropriately consider local needs in the design and execution of the research. Results from the effectiveness trial will be shared as an abstract shared via project advisory committee meetings and distributed through email. I will use webinars and in-person meetings or conferences with LHINs and regional cancer programs to discuss the findings and implications of the cross-sectional study of screening preferences. These meetings will be open to interested health care professionals, patient representatives, and community members, which will allow discussion between stakeholders. Website links to access the project report created for guideline organizations will be shared among this broader group. I will prepare lay summaries and infographics about cervical screening preferences to be distributed through social media channels and the email network that was used to promote participation in the cross-sectional study. These materials will help disseminate study findings and further publicize the preference-elicitation tool.

Impact

The first phase of this study aims to understand how an online preference-elicitation tool might be used to inform preference-sensitive cervical screening guidelines; the tool's effectiveness is then rigorously evaluated in a randomized controlled trial. In the second phase, the tool is implemented in a population-based setting to describe the prevalence of women's preferences for Pap testing, HPV testing by a professional, self-collected HPV testing and no screening. Through the effectiveness trial in the first phase, this project can determine that a preference-elicitation tool grounded in shared decision-making principles can produce robust and valid preferences data for use in guideline development. Findings from the second phase study will provide robust estimates of cervical screening preferences in a diverse population of women eligible for cervical screening. How preferences are distributed in a population is important for cancer screening guideline developers who are interested in developing acceptable, person-centered recommendations. Regional program managers can use this information to make decisions and advocate for policy change that better reflects their region's cervical screening needs. With appropriate evidence, policy-makers might feel more inclined to consider funding strategies that accommodate variation in cervical screening preferences.

Publicizing the preference-elicitation tool through the study might have an impact on health care professionals and women considering cervical screening. By providing high quality information on available cervical screening options, the tool might support women and health care professionals in making preference-based cervical screening decisions during clinical encounters.

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Chapter 8: Integrated Discussion

Overview

Cervical cancer is theoretically preventable, given our understanding that the disease results from persistent infection with a sexually transmitted infection (HPV). In Canada, the burden of cervical cancer has decreased significantly since cervical cytology was introduced in the mid-twentieth century (Tota et al., 2017; Canadian Cancer Society, 2016). Recent introduction of HPV immunization programs in elementary schools across the country are expected to further reduce rates of the disease. The impact of HPV immunization will become clearer as the vaccinated cohort ages, reaching 40 years or older. Given that there is incomplete vaccination uptake, groups of women who were considered ineligible to receive the vaccine, and that the vaccine might provide selective protection (i.e., there is uncertainty about protection against select oncogenic HPV types), early detection programs are still important. However, barriers to participation in cervical cancer control, including lack of access to health care and mistrust of the health care system, have resulted in certain subgroups facing higher rates of cervical cancer (Spence et al., 2007; Cancer Care Ontario, 2016; Elit et al., 2013).

Because of its success, health professionals and the general public are enthusiastic about early detection with cervical screening and cancer screening generally, although awareness of potential risks is growing (Prasad et al., 2016, Carter et al., 2015). For example, several guideline developers have reversed their position on prostate-specific antigen testing as screening for prostate cancer because of the significant risk of health complications and conflicting evidence of benefit. Similarly, some regions are reconsidering mammography screening for breast cancer in light of new information on the potential for overdiagnosis (Biller-Andorno & Jüni, 2014). With the changing landscape for cervical screening, more inclusive eligibility for HPV

immunization, and the introduction of HPV testing, women face new trade-offs between the potential benefits (i.e., improved prognosis) and harms (i.e., overdiagnosis and overtreatment) of cervical screening.

Often, women are targeted by aggressive campaigns for cervical screening, promoting participation as a healthy and “life-saving” (Prasad et al., 2016). With increased emphasis on autonomy in decision-making in recent years, public health institutions are starting to recognize that women’s preferences are an important part of recommendations and practice for screening. Integrating women’s preferences into cervical screening could influence the acceptability and usefulness of the respective recommendations (van der Weijden et al., 2010). Evidence on women’s preferences must be held to similar quality standards as evidence on effectiveness, to ensure that the resulting outputs are high quality. In this dissertation, I aimed to understand how different stakeholders perceive women’s preferences among cervical screening modalities (Chapters 2 through 4). Then I examined robust methods to elicit preferences (Chapters 5 through 7). I conducted this research in hopes of informing the ongoing update of clinical practice guidelines for the Ontario Cervical Screening Program and other Canadian jurisdictions.

In this chapter, I reflect on what I learned through the studies in this dissertation. In Appendix E, I contemplate what I learned more broadly through my experience as a PhD student. I first provide an overview of the findings from each of the studies (Table 8-1). Then I discuss the implications and relevance of this research by considering these findings within the bodies of evidence around methods for preference-elicitation and cancer screening preferences at the individual-level compared to the population-level. Finally, I highlight the implications of this research for different knowledge user groups, the importance for epidemiology knowledge, and opportunities for future research (Tables 8-2 and 8-3). I also examine the implications of this

research on clinical practice and health policies in the context of the changing landscape for cervical screening.

Summary of Findings

Table 8-1 presents a description of the key findings for each of the objectives in this dissertation.

Understanding multiple perspectives on women's preferences for cervical screening

The purpose of the first section of the dissertation (Chapters 2 through 4) was to understand the needs of various stakeholders, including guideline developers and decision-makers, researchers, health care professionals, and Ontario women considering screening, before multiple options are introduced in cervical cancer screening guidelines and practice. In Chapter 2, guideline developers, researchers, and health care professionals had conflicting opinions on whether self-sampling was a feasible or appropriate strategy if HPV testing infrastructure was not already in place. Most respondents raised policy, efficacy, and logistical concerns around self-sampling as a program option, which some researchers and clinicians countered by emphasizing the potential for self-sampling to improve access to cervical screening or to introduce efficiencies.

The second qualitative interview study (Chapter 3) asked women considering screening and health care professionals to identify what information women need to inform their choice between Pap testing, provider-administered HPV testing, self-collected HPV testing, and no screening. After recognizing that there are four available options for cervical screening, women identified that they need more information and an opportunity to discuss the options with their primary provider. Echoing the respondents from Chapter 2, health care professionals in Chapter 3 were apprehensive to offer multiple screening options to women because they felt that women might be overwhelmed by the information or that individuals' preferences might disrupt the effectiveness of an organized program.

In Chapter 4, I examined published research to try to synthesize women's cervical screening preferences from preference-elicitation studies. I found that cervical screening preferences depend on the research objectives of the study, the method to capture the preferences, and the context in which they are captured. Aggregating preferences from the research literature using systematic review techniques (i.e. "the prevalence of preferences") might not be the most appropriate way to capture women's preferences for cervical cancer screening because preferences depend on when, where, and how they were elicited.

Investigating measurement of women's preferences for cervical screening

I considered methods to rigorously describe or measure women's cervical cancer screening preferences in the second section of the dissertation (Chapters 5 through 7), intending to investigate Ontario women's cervical screening preferences. In Chapter 5, I critically evaluated different approaches to describe or measure women's informed, values-based preferences for cervical cancer screening. Using a specific risk of bias assessment tool for preference-elicitation studies, I found that studies widely varied in design and rigour of preference-elicitation approaches. Because studies did not clearly report on the components of their preference-elicitation approach and often lacked validation, published preference-elicitation methods for cervical screening are likely unsuitable for synthesis for clinical practice guidelines. Chapter 5 underscored the need for a rigorously developed and validated preference-elicitation tool for cervical screening.

Chapter 6 presents the development and field testing of a novel internet-based preference-elicitation tool created for the Ontario cervical screening context. Using the International Patient Decision Aids Standards systematic development process and its quality and evaluation criteria, this online tool incorporates the findings from the first section of the dissertation (Chapters 2

through 4). In a field test, most women perceived the tool as balanced, found it acceptable to use, and were able to identify their informed preferences among cervical screening modalities.

After completing the field test in Chapter 6, the tool's effectiveness at eliciting women's informed, values-based preferences should be rigorously test before it can be applied in a population-based study. In Chapter 7, I present a study protocol for a three-year, multiple-methods study that will update the preference-elicitation tool with new scientific evidence while incorporating the acceptability considerations of guideline developers, decision-makers, and policy-makers. The effectiveness of the revised tool will be tested in a randomized controlled trial in a community-based primary care setting before it is applied in a large population-based study to determine Ontario women's preferences.

Table 8-1. Summary of Key Findings from the Dissertation

Key Findings	Study designs, by chapter
<i>Objective: To determine the perceptions of stakeholders (women, health care providers, program managers, and researchers) about possible cervical screening options and to describe decision-making processes among these options.</i>	
Guideline developers, program administrators and researchers acknowledge that there are trade-offs between cervical screening modalities, implying that cervical screening decision is preference-sensitive (Chapter 2).	Chapter 2 – Semi-structured interviews with guideline developers, program managers, and researchers about cervical screening options to reach marginalized women
These stakeholders also have differing perspectives about whether there is sufficient evidence to offer alternative screening options to reach marginalized populations. In particular, there is uncertainty about how under- or never- screened women respond to alternative strategies (like self-sampling) compared to women who have participated.	
Guideline developers, program managers and researchers need information about whether marginalized women would prefer alternative strategies to cervical screening.	
Ontario women and health care professionals do not acknowledge that women face a decision to participate in cervical screening. For women, participating in screening represents healthy behaviour. For health care professionals, cervical screening represents compliance to guidelines and high quality care (Chapter 3).	Chapter 3 – Semi-structured interviews with women considering screening and health care professionals about cervical screening options within the Ontario Cervical Screening Program
Some women and health professionals are unaware that there are multiple options to consider in cervical screening, in particular the option to not participate at all.	
A systematic review of women’s informed, values-based cervical screening preferences indicated that using estimates of preferences from the published literature was unreliable and biased (Chapter 4).	Chapter 4 – Systematic review of qualitative, quantitative, and mixed methods studies of women’s cervical screening preferences
Preferences vary by preference-elicitation method, study objective, design, and screening context. The heterogeneity of methodological and reporting quality of the included studies made it challenging to synthesize preferences across study designs and contexts.	

A synthesis of the published literature cannot contribute reliable data on preferences and values because cervical screening preferences are dependent on study objective, design, and preference-elicitation approach.

This section highlighted a need to identify a robust approach to preference-elicitation and apply it in the Ontario setting.

Objective: To understand methods to measure women's preferences in cervical screening, which will inform the development and testing of a person-centered, evidence-informed approach to elicit women's preferences.

There is variation in how researchers defined and studied women's preferences, highlighting a need for consensus on a definition of preferences and preference-elicitation (**Chapter 5**).

Guideline developers should not rely on published preference-elicitation studies to generate preference-sensitive recommendations because the literature lacks transparency and rigour.

To address the lack of robust preference-elicitation tools, I systematically developed a novel online tool to elicit women's preferences among cervical cancer screening modalities (**Chapter 6**).

In a field test, this tool was found to be acceptable to women considering screening and successfully elicited their informed, values-based preferences.

I propose a multiple methods study to investigate women's preferences across three Ontario health regions to produce robust data on women's informed, values-based cervical screening preferences using an effective preference-elicitation tool (**Chapter 7**).

Chapter 5 – Systematic review of qualitative, quantitative, and mixed methods studies about methods to elicit women's cervical screening preferences

Chapter 6 – Development and field testing of a preference-elicitation tool based on shared decision-making framework and quality criteria

Chapter 7- A multiple methods study proposal to investigate how guideline developers intend to integrate preferences into cervical cancer screening guidelines and practice

Discussion and Implications

Conceptual frameworks in the cervical screening context.

Women's values and preferences can be incorporated into cervical screening at three different levels (Pieterse & Stiggelbout, 2016): at the level of the individual, during clinical encounters between women and their health care providers (micro-level), at the program/institution level, through clinical practice guidelines and protocols (meso-level), and at the system-level, when comparing investments for cervical screening compared to another health system priorities (macro-level). In this dissertation, I used three frameworks to frame research questions, design the studies and interpret results to understand and elicit women's preferences among cervical screening modalities at the micro-level and meso-level. To consider women's preferences at the individual level, the Ottawa Decision Support Framework (ODSF) and International Patient Decision Aids Standards (IPDAS) were integral for conceptualizing preferences for a systematic review (Chapters 4 and 5), investigating how women decide their preferences, and systematically developing and testing a preference-elicitation tool (Chapters 6 and 7). The ODSF and IPDAS criteria have been used in the development and evaluation of decision support tools in other cancer screening settings, including prostate cancer, breast cancer, colorectal cancer, and lung cancer (Lillie et al., 2014; Jimbo et al., 2013; Stacey et al., 2017), but not in a cervical cancer screening context. I integrated the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach into the systematic review protocol (Chapters 4 and 5), so that I could generate useful recommendations for guideline developers based on the preferences and values literature. The GRADE approach is operationalized in some cervical screening guidelines, including those published by the Canadian Task Force on Preventive Health Care (SPOR Evidence Alliance, 2018). However, women's preferences and values have not been previously assessed using the GRADE process, although a

health technology assessment that considers women's preferences for HPV testing is expected in the next two to three years (CADTH, 2017).

The omission of cervical screening from shared decision-making research is not surprising considering that most Canadian guidelines have strongly recommended Pap testing for women 20 or 21 years to 65 years (SPOR Evidence Alliance, 2018). According to the GRADE strength of evidence scores, a strong recommendation implies that careful review with each patient is unnecessary because there is a definite preference for one option and it should apply to all or almost all patients (Neumann et al., 2016). This language seems to discourage the guideline end-users (e.g., health care providers) from eliciting preferences or engaging in shared decision-making with women considering cervical screening. Thus, these apparently rigid recommendations for cervical screening suggest a perspective on the part of guideline developers that screening programs that are based on robust, controlled evidence are necessarily void of preferences and values (Kelly et al., 2015). A main finding of Chapter 2 was that respondents – guideline developers, researchers, and program managers - were particularly concerned about the availability of evidence to support implementation of self-sampling strategies within organized screening programs. The disagreement between respondents in Chapter 2 around the sufficiency of evidence to support alternative strategies indicates variation in values. For example, perhaps the respondents who felt prepared to make recommendations for self-sampling based on the current body of evidence were primarily concerned about equity in health. Interestingly, respondents in Chapter 2 also disagreed about the types of research questions that would fill the knowledge gap (e.g., is more information needed on uptake rates using self-sampling or is more information needed on how to deliver self-sampling?) further emphasizing differences in social perspectives and values among different stakeholders within the health system (Kelly et al., 2015).

When GRADE recommendations imply that discussing health care options with individuals is unnecessary due to a high certainty of evidence for a particular option, guideline developers are dismissing the importance of individuals' needs or desire for autonomous decision-making in their health care (Kelly et al., 2015; Nicholls et al., 2016; Sandman et al., 2012). As observed in Chapter 3, Ontario women and health care providers do not feel that women have a choice to participate in cervical screening. However, knowing that they face a choice and that the modalities are currently available, respondents were enthusiastic about making informed decisions. An individualized approach to preference-elicitation, such as shared decision-making, might be an appropriate way to capture women's values and preferences in cervical screening decisions. However, as discussed in the next section, an individualized "person-centered" approach to cervical screening might appear to diverge from the population-based goals of cancer screening.

A recent selection of papers from the GRADE working group described methods to assess the quality of evidence from the preferences and values literature (Zhang et al., 2017, 2018a, 2018b; Yepes-Nuñez et al., 2017). Authors of this series recognize that individuals who are presented with the same body of evidence may make different choices because of disparate values and preferences (Zhang et al., 2017). Consequently, the authors propose that patients' values and preferences should be integrated into guidelines through systematic synthesis of high quality literature. In Chapters 4 and 5, I applied the suggested risk of bias assessment criteria to studies considered for inclusion in the systematic review of methods to elicit women's preferences for cervical screening. While the strategy to identify and appraise relevant preferences literature was useful, the techniques did not adequately address the heterogeneity in study objectives, designs, or analytic approaches. The heterogeneity of included studies likely reflects variation in values held by the researchers and screening stakeholders in the study

contexts (Kelly et al., 2015; Nicholls et al., 2016). Differences in values were manifested through varying study objectives, study design, preference-elicitation methods, and interpretations. As a result, it was impossible to conclusively describe women's preferences for cervical screening or identify a robust method to elicit women's preferences. This review demonstrated that the published evidence on cervical screening preferences was not sufficiently robust to be used by guideline developers. Thus, I addressed this gap using a systematic and transparent development process outlined by IPDAS and ODSF to create a novel tool that elicits women screening preferences (Chapter 6).

Going forward, guideline developers might want to consider how preference-elicitation through shared decision-making might be the most person-centered and evidence-informed approach to incorporate preferences. Noting that women's cervical screening preferences are time- and context- dependent (Chapter 4), the ODSF suggests that individual-level decision support would ensure that women's values and preferences are at the core of the decision. Given the lack of robust literature around cervical screening preferences, it is challenging to capitalize on new methodologies to synthesize and evaluate preferences data for clinical practice guidelines. In particular, future research should investigate how to make recommendations that balance strength and direction of clinical effectiveness evidence, preferences and values evidence, and evidence on shared decision-making. Further work in this area should consider how to reflect the values of guideline developers and researchers in the syntheses of preferences and values literature (Kelly et al., 2015; Nicholls et al., 2016). The framing of the questions and appraisal of evidence for guideline recommendations depends on the membership of the developer panel (Nicholls, 2016). If guideline developers declared their worldviews during guideline development, guideline panels can begin to address the larger ethical consideration of whose voices are considered "legitimate" (Nicholls, 2016). Cancer screening decisions are, by

definition, preference-sensitive. Acknowledging that different stakeholders might have conflicting preferences might lead to more transparent and careful decision-making in cancer screening at the individual-level and at the population-level. Understanding whether guidelines are rooted in a societal or individual perspective could influence how health policy is made and how its impact is evaluated.

It is particularly important in this changing landscape of cervical screening to determine how women's preferences can be considered. Currently, different judgments of the clinical effectiveness of different screening tests has resulted in varying recommendations for cervical screening across Canada and other high-income regions. Consequently, women's values and preferences will play an important role in shaping how different programs choose and implement their recommendations. Cervical screening guideline developers will face challenges to synthesize preferences and values research literature using the GRADE approach due to heterogeneous evidence. A lack of robust evidence on women's preferences and values should not affect the consideration of preferences in guidelines or in practice more broadly. There are alternative strategies to incorporate women's preferences into cervical screening guidelines (e.g., through guideline panel representation) (Kelly et al., 2015; Montori et al., 2013a). A simple response might be for guideline developers to downgrade a strong recommendation for preference-based decisions like cervical screening to a conditional or weak recommendation when the body of preferences literature is weak. As a result, cervical screening guidelines could present the screening modalities in a way that supports preference-elicitation and shared decision-making at the individual-level. Future guidelines can include tools to support the preference-elicitation and decision-making processes at point-of-care encounters. However, high quality evidence on the effectiveness of these tools in cervical screening contexts, which can be

guided by IPDAS quality and evaluation criteria, will be needed before making broad recommendations about its use.

Cancer screening preferences at the individual-level versus the population-level.

Tensions exist between individual benefits and risks of cancer screening and population benefits and risks of cancer screening. Cervical screening programs look for signs of cervical dysplasia (or worse) among average-risk women. Higher participation in the organized screening program will maximize the effectiveness by detecting a greater number of cases, which can be associated with improved prognosis and lower system costs (Canadian Cancer Society, 2016; Schiffman, 2017). However, cervical screening can also cause physical and psychological harms to the individual and excess costs to the system (and occasionally individual) (Habbema et al., 2017; Salmi et al., 2016; Dobrow et al., 2018). At a population level, the benefits of cervical screening are well-accepted (Canadian Cancer Society, 2016; Schiffman, 2017; Dickinson et al., 2012), although the harms of overdiagnosis (from false positives or incidental findings) and overtreatment are typically understudied or misunderstood (Habbema et al., 2017; Carter et al., 2015). Individuals, on the other hand, face a much lower probability of personal benefit (from early detection) because cervical pre-cancers and cancers are becoming increasingly rare. The potential for harms is similarly increasing because HPV immunization will lead to a decreased positive predictive value of the screening tests. The incongruence between what is best for an individual and what is best for the population reflects different value judgments or preferences.

There are different ways to assess how much cervical screening is worth, which was reflected in how women and health care professionals contemplate cervical screening decisions (Chapters 2, 3, and 6) and the mechanisms behind different preference measurement approaches (Chapters 4 and 5). From the population or system perspective, stakeholders tend to apply a utilitarian philosophy (Kelly et al., 2015; National Collaborating Centre for Healthy Public

Policy, 2016) - the greatest health gain for the greatest number of people. From this perspective, cervical screening is beneficial if the screening program can reduce the burden of cervical cancer in a population in a way that is acceptable to individuals eligible for screening and feasible within a health system. For individuals, participating in cervical screening is important if screening can detect disease such that they have a better prognosis or response to treatment. When individuals place greater value on avoiding the risks of cervical screening such that they choose not to participate, the consequence of this preference contributes to a less efficient, less effective cervical screening program. Focusing on efficiency and effectiveness at the population level might contribute to inequities because an individual might not choose the course of action that aligns with their values (Kelly et al., 2015). For example, some women might recognize that they experience substantial anxiety with a positive screening result, therefore they would prefer to avoid the modality with the greatest probability of false positives even though guidelines recommend the option that is most effective at detecting true positives.

Some researchers suggest that focusing on acceptability of cancer screening – including the tests, the follow-up schedule, and the potential for errors – is key to reconciling the differences between the individual and population perspectives (Salmi et al., 2016; Dobrow et al., 2018; Smith, 2018). A recent systematic review and consensus study (Dobrow et al., 2018) revisited Wilson and Jungner's principles for screening (Wilson & Jungner, 1968). Authors found that a heavier emphasis on acceptability and ethics of screening programs is required, more than acceptability of the screening test itself (e.g., Pap test, HPV test). Potential screening participants (i.e. members of the general public or of high-risk populations) are mentioned briefly among people considered experts as relevant stakeholders to help assess system principles of a screening program (Dobrow et al., 2018). Because acceptability of screening programs is based on value judgments, explicitly incorporating potential participants' preferences into

screening program development and implementation is essential to align person-centered cervical screening with population-based screening goals (Salmi et al., 2016). Beyond balancing these two perspectives, there is an ethical obligation in medicine and in society to ensure that individuals are informed and empowered to make an autonomous decision (Sandman et al., 2012; The Lancet, 2016; Johansson & Brodersen, 2015).

In this dissertation, the population benefits of cervical cancer screening were emphasized by most respondents (i.e., clinical or system-level experts) in Chapter 2 and most health care professional respondents in Chapter 3. Respondents felt that screening strategies that diverge from the current Pap test-based program, including self-sampling for HPV, need to meet the criteria (Smith, 2018; Wilson & Jungner, 1968; Andermann et al., 2008) of a successful population-based screening program. These criteria include “suitable” performance of tests, acceptable and available diagnostic options, and economic feasibility. Many health care professionals felt apprehensive about incorporating individual preferences into cervical cancer screening in case they deviate from the most “effective” test. When contemplating self-sampling as an option in a screening program, respondents considered it inappropriate to offer all women a choice at the point-of-care, because some women might “wrongly” choose to use the less sensitive test (or opt out completely), compromising the population health impact of the screening program (Chapter 2). These sentiments were echoed in part by health care professionals in Chapter 3 who worried about their patients who might miss an early detection opportunity, though most providers felt that if they explained cervical cancer screening to their patients that they would likely choose to participate, particularly if women had a provider whom they trust. Interestingly, during the interviews, some respondents in Chapter 2 and most health care professional respondents in Chapter 3 began to or already recognized potential tension

between individual preferences and behaviors compared to population preferences and behavior in cervical screening.

Women considering cervical screening felt that screening is something that they would *adhere* to, based on their provider's recommendation, instead of actively choosing to receive screening (Chapter 3). Most screen-eligible respondents did not consider how their preferences or behaviors might relate to the overall impact of a population-based program, either through health outcomes or use of publicly-funded resources (in Ontario's publicly-funded health system), although one woman (Chapter 3) commented on the disproportionate use of resources for treating later stages of disease compared to prevention by early detection. Health care professionals might feel pressure to coerce women to choose a cervical screening option that aligns with the traditional system efficiency model (Williams & Carter, 2016). Furthermore, in Ontario and other Canadian settings, the proportion of eligible women participating in cervical screening is used as a quality or performance metric in primary health care, which will also influence point-of-care conversations about screening.

As evidenced in Chapter 3, some women participated in Pap testing – the only cervical screening test funded in Ontario - without deliberation because other options including the option to opt out were not presented. Because we value autonomy in Canadian health systems and society (although perhaps not more than utilitarianism), women eligible for screening should be presented with the opportunity to make preference-based decisions about undergoing cervical screening (Sandman et al., 2012; Williams & Carter, 2016).

This perspective does not necessarily deviate from the population-based perspective mentioned above (Sandman et al., 2012; Salmi et al., 2016; Williams & Carter, 2016), rather it is a core component of cervical screening acceptability. Women should be *informed* about the options that are available to them so that they can assign *value* to the respective harms and

benefits to select their preferences (Salmi et al., 2016; Llewellyn-Thomas & Crump, 2013; Lang et al., 2018). Notably, preference-based, autonomous decisions in cervical cancer screening do not preclude women from engaging with their health care providers or important others throughout the decision-making process (Sandman et al., 2012; Salmi et al., 2016; O'Connor et al., 1998; Pieterse & Stiggelbout, 2016).

The systematic reviews in Chapters 4 and 5 also demonstrated the dichotomy of individual-level and population-level preferences and approaches to preference-elicitation. Researchers tend to think about aggregated preferences from the published literature (e.g. proportions of populations who prefer particular options), to form the basis of guidelines, policy, and funding decisions (Zhang et al., 2017, 2018a, 2018b; Yepes-Nuñez et al., 2017; Montori et al., 2013a). However, for synthesized preferences to be useful, the primary preferences studies must use robust approaches to preference-elicitation.

Yet, it is not clear that aggregated population preferences, like those presented in systematic reviews of stated preference studies (Zhang et al., 2017; Yepes-Nuñez et al., 2017; Mansfield et al., 2016), should form the basis of “preference-based” person-centered guidelines (Lang et al., 2018; Umscheid, 2009; Krahn & Naglie, 2008) because a collective group of people might not adequately represent differences among individuals. Since cervical screening preferences might be context- or time-dependent (Chapter 4), population preferences should not be the only method of incorporating preferences into guidelines and practice (Lang et al., 2018; Krahn & Naglie, 2008). Using effectiveness information from clinical practice guidelines to inform shared decision-making at the point-of-care is one approach that would reflect the dynamic nature of individuals’ preferences (Lang et al., 2018; Umscheid, 2009; Krahn & Naglie, 2008). The preference-elicitation tool developed as part of this dissertation (Chapter 6) could be used as a robust tool to elicit preferences in a population-based study (as proposed in Chapter 7)

but might be useful in point-of-care encounters to support shared decision-making between a primary care provider and women considering screening.

Since Ontario has committed to fund HPV testing in the provincial screening program (Ministry of Finance (Ontario), 2017) and continues to consider a self-sampling option (Chapter 2), understanding women's preferences among cervical cancer screening options is timelier than ever. The preference-elicitation tool created in this dissertation was developed in a systematic and transparent way and can contribute robust evidence of women's preferences to the body of literature. However, there is still a gap in understanding about how to optimally and reliably incorporate women's preferences into cervical cancer screening recommendations and practice. The proposed study protocol in Chapter 7 aims to address this gap.

Contributions to the Advancement of Knowledge, Clinical Practice, and Policy

The studies included in this dissertation offer novel insight into the science of preference-elicitation, future considerations for Ontario cervical cancer screening, and identify areas of further research, as outlined in the following section and corresponding tables. In particular, Table 8-2 presents the key messages from this dissertation by different knowledge user groups, and aligns these messages with the main epidemiologic learnings and opportunities for further research. Table 8-3 provides a high-level summary of the implications of the knowledge generated as part of this dissertation and the identified knowledge gaps.

Table 8-2. Dissertation Key Messages by Stakeholder Groups

	Guideline developers	Policy-makers and program managers	Health care providers	Women considering cervical screening
Key messages	Women's values and preferences are important in cervical screening. Respective guideline recommendations should be softened to conditional or weak in the absence of robust preferences and values data and published evidence. Different cervical screening stakeholders might have different values (i.e. population-based versus person-centered) that could influence decision-making in guideline development.	Preference-based cervical screening has implications on use of system resources, the effectiveness and rates of harm related to screening options, and whether these options create a more equitable screening program. Different cervical screening stakeholders might have different values (i.e. population-based versus person-centered) that could influence decision-making in policy and funding decisions.	There are multiple options available for cervical screening including the option to opt out, and women considering screening might have different preferences. The field test shows that an online preference-elicitation tool is balanced, acceptable to women, and that it can elicit informed, values-based preferences.	An online preference-elicitation tool can support women in recognizing that there are multiple options for cervical screening, communicate important information about the options, and help clarify important values. Different women might have different values and preferences for cervical screening modalities, although all modalities are not equally accessible in different screening programs.
Implications for epidemiology and epidemiologic research	Epidemiologists developing preference-elicitation tools should require systematic development processes and rigorous testing for effectiveness to produce robust evidence that can	Epidemiologists should consider methods to use person-centered outcomes which might be different from population health or health system performance outcomes.	Epidemiologists should compare how health care providers' preferences for screening options differ from the preferences of guideline developers and the preferences of	Epidemiologists should investigate the needs of the target population/knowledge users in the development of a preference-elicitation tool.

	be synthesized in guideline development. Theoretical or conceptual frameworks, including shared decision-making, are useful for guiding the development, evaluation, and implementation of preference-elicitation tools.	These methods are particularly relevant for cancer epidemiology when judging the value of early detection programs (a population-level intervention). Epidemiologists and other researchers should reflect on and make explicit the values that underscore their research questions, choice of methods and outcomes, and interpretation of study results.	individuals considering screening.	
Future research	Future research should rigorously evaluate the preference-elicitation tool's effectiveness at eliciting informed, values-based preferences. Additionally, researchers should study the preferences of populations using "Cervical cancer screening – it's your choice" to provide local evidence on preferences.	Future research should investigate how to assess and synthesize published evidence across varying definitions of preferences and preference-elicitation strategies. Researchers should also investigate how varying definitions of preferences affect the final interpretation of values and preferences data.	Future research should investigate how health care professionals can implement preference-based cervical screening (e.g., can a preference-elicitation tool be used during a clinical encounter), and what supports are needed to do this. Future research should investigate whether health care provider performance can or	Future research should investigate how incorporating women's values and preferences into cervical screening practice affects measures of cervical screening practice and health outcomes (e.g., cancer incidence, mortality, and survival rates). While the course of action is not a decision-relevant outcome, these metrics will be valuable to

		Future research should investigate how quality and performance measures for physicians and health care providers can capture preference-based cancer screening.	should be measured using cancer screening participation. If so, how can metrics capture preference-elicitation processes?	evaluate the population health and health system impacts of individualized cervical screening.

Scientific knowledge and further research.

Shared decision-making interventions often focus on screening decisions because of the inherent “preference-sensitive” nature of early detection (Lillie et al., 2014; Jimbo et al., 2013; Stacey et al., 2017; Lang et al., 2018; Grad et al., 2017). Considering primary cervical cancer screening as a primary decision and then applying shared decision-making principles to develop a preference-elicitation tool, this dissertation advances the epidemiologic knowledge about how preferences data is collected and synthesized. Current guidance (Zhang et al., 2017, 2018a, 2018b; Selva et al., 2017) suggests that preferences reflect how people view the importance of health outcomes, which can be captured through multiple study designs and diverse conceptual frameworks. Recently published search strategies present a broad list of search terms that capture different types of measures (e.g., qualitative or quantitative) (Selva et al., 2017), yet there is still an emphasis on the quantitative stated preferences literature that uses economic utility theory (e.g., discrete choice experiments). The research in this dissertation showed that there is significant overlap between the shared decision-making literature and the preferences and values literature. Importantly, with more advanced understanding of methods to design (Coulter et al., 2013), evaluate (Bouniols et al., 2016; Gärtner et al., 2018), synthesize (Stacey et al., 2017) and implement (Légaré et al., 2018; Grad et al., 2017) shared decision-making research, researchers aiming to generate knowledge in the preferences and values field should look to the shared decision-making literature for insight.

I used concepts specific to shared decision-making and the Ottawa Decision Support Framework to understand what information is needed to elicit women’s preferences (Chapters 2,3, and 6) and to systematically identify relevant studies that address women’s preferences for cervical screening (Chapters 4 and 5). In particular, preference-elicitation required that i) individuals recognize that there is a decision to be made (i.e., that there are multiple options); ii)

individuals feel informed about the decision options, and; iii) individuals can identify what features of the options matter most to them. While some researchers might argue that these concepts are unique to shared decision-making frameworks (Zhang et al., 2017; Selva et al., 2017), the specific and concrete definition of preferences might be most relevant for person-centered and evidence-informed practice and guidelines (Zhang et al., 2017; Umscheid, 2009; Grad et al., 2017; Hoffmann et al., 2014a). Therefore, future research in the values and preferences domain should consider shared decision-making concepts in the design and analysis of primary studies and in syntheses of published literature.

Using a shared decision-making lens to understand preferences across different health decisions highlights gaps in knowledge and potential challenges that could arise. For example, searching for studies where the target population i) recognizes the decision, ii) understands the options, and iii) understands what matters most to them is more restrictive “inclusion” criteria than studies which investigate the relative importance of outcomes. Researchers should identify when a shared decision-making definition of preferences is not appropriate or specific enough, then examine what impact this will have on the understanding of synthesized preferences. Some researchers conflate the concepts of preferences among decision options and acceptability of decision options (Chapters 4 and 5; Zhang, 2017). Theorizing how the concepts preferences and acceptability relate to and differ from each other will be important for developing and evaluating tools to elicit people’s preferences (Sekhon et al, 2017). Operational criteria or materials (e.g., when are participants sufficiently “informed” to identify a preference?) can help researchers, guideline developers, or other knowledge users who are designing interventions or examining values and preferences studies.

Using a shared decision-making lens in preferences studies might indicate support for implementation of shared decision-making in preference-sensitive health decisions. In practice,

shared decision-making can be recommended to address discrepancies between clinical effectiveness recommendations and recommendations based on preferences studies (Zhang et al., 2017; Hoffmann et al., 2014a). Although Canadian physicians are advocating for shared decision-making in preventive health screening (Lang et al., 2018; Grad et al., 2017; Bell et al., 2017), guidelines for most screening programs do not clearly discuss its use (SPOR Evidence Alliance, 2018; Cancerview.ca, 2016; Légaré et al., 2017). Therefore, researchers and guideline developers should explicitly address the evidence (or lack of) on shared decision-making alongside the evidence on clinical effectiveness and values and preferences. A more widespread use of shared decision-making requires continued research and funding of shared decision-making practice and tools, and needs to be accepted by the general public and the health care system (Légaré et al., 2017).

Health services and policy and future research.

In Ontario, cervical screening is organized through a provincial program and delivered by primary care providers. Cervical cancer screening practice, clinical practice guidelines, and quality improvement metrics will likely be updated in the near future to reflect movement toward primary HPV testing (Tota et al., 2017; Ministry of Finance (Ontario), 2017; Murphy et al., 2012a; Ebell et al., 2018; Franco, 2018). Alongside a recent series of papers targeting Canadian primary care practitioners (Lang et al., 2018; Grad et al., 2017; Bell et al., 2017), the findings from Chapters 2 and 3 suggest preference-elicitation and shared decision-making in cancer screening is gaining acceptance among health care providers. Subsequently, health care providers might be more likely to clarify values and preferences of women who are considering cervical cancer screening. Guideline developers and researchers might be more likely to encourage women to make autonomous cervical cancer screening decisions, instead of “nudging” participants to participate (Blumenthal-Barby et al., 2013; Purnell et al., 2015; Ploug

et al., 2012). Cervical screening stakeholders – health care professionals and administrators, researchers, and women considering screening – should acknowledge that participation in cervical screening is a preference-sensitive decision that women face. For cervical screening practice to be evidence-informed and person-centered, women’s values and preferences should be incorporated into decisions at the point-of-care and in guideline development. As previously mentioned, shared decision-making processes could reconcile differences or variation between clinical effectiveness recommendations and recommendations based on women’s preferences and values.

Considering that some of the cervical screening options are still challenging to access in Ontario’s single-payer system (i.e. physicians are gatekeepers for HPV testing, self-sampling for HPV is only available using a privately purchased kit), there are certainly equity implications in the modernization of the provincial screening program. As emphasized by women considering screening and health professionals respondents in Chapters 2 and 3, publicly funding available cervical screening modalities could help women participate in screening that aligns with their preferences. However, this strategy has major implications for system resources, the population effectiveness (and rates of harms) of the different screening modalities, and how these benefits and harms are experienced by various communities of women. Following a study on the prevalence of preferences as suggested in Chapter 7, a simulation study could help researchers to understand the implications of preference-based cervical screening.

Policy-makers and health system administrators will need to accommodate the shift in approach to cervical screening by adjusting quality standards and targets. Currently, “proportion of women attending cervical cancer screening” is considered a measure of quality or performance in health care (e.g., higher proportions of women imply better health care) (Grunfeld et al., 2013; Canadian Partnership Against Cancer, 2012; Cervical Screening

Participation, 2014). Administrators need to consider alternative metrics to communicate to providers and women that informed, preference-based decisions represent ideal cervical cancer screening outcomes rather than high uptake rates. Certainly, if shared decision-making practices become the norm, evaluators might consider more nuanced indicators that consider decision outcomes, such as decision quality.

The information sections for the preference-elicitation tool “Cervical cancer screening – it’s your choice” are still accessible online at <https://bwood096.wixsite.com/screen-preferences>. Before the entire preference-elicitation tool could be useful within the provincial program, it is essential that its effectiveness as a preference-elicitation is rigorously evaluated (as proposed in Chapter 7, Phase 1). While the tool is not actively promoted among women or providers, the website has received approximately 20 new visitors after the field test finished, providing a tangible example of interest by Ontario cervical screening stakeholders.

Table 8-3 summarizes the implications of the knowledge generated by this dissertation, and highlights areas for future research.

Table 8-3. Contributions to Epidemiology Knowledge and Health Services and Policy, Highlighting Future Research Opportunities

Domain	Implications	Future research opportunities
Epidemiology knowledge	Shared decision-making processes offer another preference-elicitation strategy in health care decision-making. Evidence syntheses must consider shared decision-making approaches to preference-elicitation as relevant preferences literature.	Research should investigate how to assess and synthesize across varying definitions of preferences and preference-elicitation strategies. Researchers should also investigate how different theoretical conceptualizations of preferences affect the final interpretation of values and preferences data. Future research should investigate how to make recommendations that balance strength and direction of clinical effectiveness evidence,

		preferences and values evidence, and evidence on shared decision-making. As proposed in Chapter 7, future research should evaluate the tool's effectiveness at eliciting informed, values-based preferences to generate robust cervical screening preferences data.
Health Services and Policy	Recognizing that women's values and preferences are important in cervical screening, respective guideline recommendations should be softened to conditional or weak in the absence of robust preferences and values data. Cervical screening guidelines and most Canadian preventive health screening guidelines should make an explicit statement about shared decision-making within their recommendations. Cancer screening guidelines could be structured to promote shared decision-making at the point-of-care. More evidence-informed tools (e.g., decision aids) are needed for cervical screening guideline users to incorporate preference-elicitation or shared decision-making in their practice. Offering multiple modalities for cervical screening will have implications on use of system resources, the effectiveness and rates of harm related to screening options, and whether these options create a more equitable screening program.	As proposed in Chapter 7, future research should investigate the perspectives of guideline developers and system administrators as to how they intend to incorporate women's cervical screening preferences into updated guidelines and practice. Future research should study the preferences of populations using "Cervical cancer screening – it's your choice" to provide local evidence on preferences. Future research should apply an equity lens when examining the population health and system resource implications of preference-based cervical screening. Future research should investigate how the integration of women's values and preferences into cervical screening practice affects different measures of cervical screening practice. For example, how does preference-informed cervical screening affect: - participation in the cervical screening program, particularly related to the populations who face barriers to participation(equity)? - patient experiences during cervical screening encounters? - system utilization related to cervical screening (e.g., treatment for cervical pre-cancer and cancer)? - relevant health outcomes (cancer incidence, mortality, survival)?

Conclusion

Between 2015 and 2018, I conducted five studies and designed a follow-up study to (i) determine what is known about women's preferences and decision-making processes in cervical cancer screening and, (ii) to understand approaches to rigorously measure women's preferences to ultimately inform Ontario cervical screening guideline developers. Based on a needs assessment and critical evaluation of preference-elicitation approaches, I developed a novel online preference-elicitation tool to support the integration of women's preferences into cervical screening recommendations. A study protocol followed these studies, proposing to use multiple methods to refine the preference-elicitation tool and determine its effectiveness before using it in a population-based study to understand women's preferences among cervical screening modalities and factors related to these preferences. The findings from these studies aim to contribute evidence that supports preference-based cervical screening guideline development in publicly-funded, high-resource cervical screening programs.

An important finding is that women want to make a choice among cervical cancer screening modalities and were able to identify an informed preference among Pap testing, provider-administered HPV testing, self-collected HPV testing, and no screening using the preference-elicitation tool. Most women perceived the tool as balanced, felt that the tool was acceptable, and did not experience anxiety after exploring the information in the tool. While most women who used the tool were enthusiastic about the opportunity to "choose" between options, many wanted the input of their primary care providers before acting on any particular choice. These findings are important for health care providers who might work through preference-elicitation processes when offering cervical cancer screening, particularly in light of transitioning clinical practice guidelines. It is essential that program administrators also consider the preference-sensitive nature of cervical screening decisions in quality and performance

measurement, beyond the crude rates of screening participation that are currently reported. Without consistent execution of “person-centered” or “preference-based” cervical cancer screening, it is unlikely that this shift in philosophy will have an effect on patient or system outcomes.

Guideline developers should also assess the value of shared decision-making for cervical screening, and explicitly address the lack of robust evidence to support subsequent recommendations. In particular, guideline developers can downgrade the strength of the cervical screening recommendations to “conditional” or “weak” to reflect the paucity of high quality preferences research needed for this preference-sensitive decision. Explicit definitions of preferences and preference-elicitation, including those provided in the Ottawa Decision Support Framework, will ensure the approaches used in primary studies on preferences and values are robust and relevant to the screening context. I found in this dissertation that a synthesis of preferences studies is not sufficient considering the context- and time-dependent nature of cervical screening preferences. Future research should examine how to synthesize heterogeneous data from the values and preferences primary studies to support recommendations in clinical practice guidelines. Because more rigour is required to produce valid and reliable evidence on women’s preferences for cervical screening, I propose to conduct an effectiveness trial of the preference-elicitation tool developed in this dissertation.

Researchers, guideline developers, and health care professionals can leverage methods and tools from the shared decision-making body of evidence to incorporate individuals’ preferences into guidelines and cervical screening programs. Going forward, it will be important to explicitly define whose and what values are being considered in the development of guidelines, which will help members of the public and health care providers make judgments about the recommendations.

Appendix A: Ethics Approval

Chapter 2 [Manuscript 1]: Tailoring Cervical Cancer Screening Strategies to Hard-to-Reach Women in Canada: A Qualitative Study of the Potential Value of HPV Self-Sampling

- Ryerson Research Ethics Board (REB 2016-036)
- Letter of permission from primary applicant at Ryerson University
- Ottawa Health Science Network Research Ethics Board (Protocol # 20160134 -01H)

Chapter 3 [Manuscript 2]: “They Should Be Asking Us”: A Qualitative Decisional Needs Assessment for Women Considering Cervical Cancer Screening

- Ottawa Health Science Network Research Ethics Board (Protocol # 20160134 -01H)

Chapter 4 [Manuscript 3]: What are Women’s Preferences among Cervical Cancer Screening Options: A Narrative Synthesis of Quantitative, Qualitative, and Mixed-Methods Studies

- Systematic review – ethics approval not required

Chapter 5 [Manuscript 4]: Preference-eliciting approaches in cervical cancer screening: A systematic review of qualitative, quantitative, and mixed methods studies

- Systematic review – ethics approval not required

Chapter 6 [Manuscript 5]: Development and field testing of a tool to elicit women’s preferences among cervical cancer screening modalities

- Ottawa Health Science Network Research Ethics Board (Protocol # 20160134 -01H)[Amendment]

Chapter 7 [proposal]: Preference-based cervical cancer screening in Ontario: Protocol for a mixed-methods investigation

- Proposed project – ethics approval not required



To: Mandana Vahabi
Nursing

Re: REB 2016-036: Toward developing effective cervical cancer screening programs for
hard-to-reach women

Date: February 8, 2016

Dear Mandana Vahabi,

The review of your protocol REB File REB 2016-036 is now complete. The project has been approved for a one year period. Please note that before proceeding with your project, compliance with other required University approvals/certifications, institutional requirements, or governmental authorizations may be required.

This approval may be extended after one year upon request. Please be advised that if the project is not renewed, approval will expire and no more research involving humans may take place. If this is a funded project, access to research funds may also be affected.

Please note that REB approval policies require that you adhere strictly to the protocol as last reviewed by the REB and that any modifications must be approved by the Board before they can be implemented. Adverse or unexpected events must be reported to the REB as soon as possible with an indication from the Principal Investigator as to how, in the view of the Principal Investigator, these events affect the continuation of the protocol.

Finally, if research subjects are in the care of a health facility, at a school, or other institution or community organization, it is the responsibility of the Principal Investigator to ensure that the ethical guidelines and approvals of those facilities or institutions are obtained and filed with the REB prior to the initiation of any research.

Please quote your REB file number (REB 2016-036) on future correspondence.

Congratulations and best of luck in conducting your research.

Lynn Lavallée, Ph.D.
Chair, Research Ethics Board



Brianne Wood <brianne@eve-medical.com>

Ethics for secondary data analysis

Mandana Vahabi <mvahabi@ryerson.ca>
To: Brianne Wood <brianne@eve-medical.com>
Cc: aisha.lofters@utoronto.ca

Tue, May 3, 2016 at 12:19 PM

Hi Brianne

We have no objection for you to use a few of our interviews for your own research on decision-making for cervical screening. The following paragraph reflects this:

I give Brianne Wood my permission to use interview transcripts from CIHR-funded study "Toward developing effective cervical cancer screening programs for hard-to-reach women". This study has been approved by the Ryerson University Research Ethics Board (REB 2016-036). I understand that Brianne will be using these transcripts for secondary data analysis as part of her PhD dissertation at the University of Ottawa.

Sincerely,

Dr. Mandana Vahabi, Principal Investigator

--

Dr. Mandana Vahabi
Associate professor
Daphne Cockwell School of Nursing (DCSN), Faculty of Community Services,
Co- director, Centre for Global Health and Health Equity, Co-Lead, Diverse, and At-risk
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**Ottawa Health Science Network Research Ethics Board/ Conseil d'éthique de la recherche du
Réseau de science de la santé d'Ottawa**

Civic Box 411 725 Parkdale Avenue, Ottawa, Ontario K1Y 4E9 613-798-5555 ext. 14002 Fax : 613-761-4311
<http://www.ohri.ca/ohsn-reb>

June 29, 2016

Dr. Julian Little
University of Ottawa
Faculty of Medicine
Department of Epidemiology and Community Medicine
451 Smyth Road, Room 3257
Ottawa, ON K1H 8M5

Dear Dr. Little:

Re: Protocol # 20160134-01H Woman-centered cervical screening: Identifying women's preferences and factors related to their preferences in cervical cancer screening

Protocol approval valid until - June 28, 2017

I am pleased to inform you that this protocol underwent delegated review by the Ottawa Health Science Network Research Ethics Board (OHSN-REB) and is approved for the recruitment of English speaking participants only. No changes, amendments or addenda may be made to the protocol or the consent form without the OHSN-REB's review and approval.

Approval is for the following:

- Protocol (version 8) dated February 2016 (Study #2 only)
- English Recruitment Guide/Script (Phone, Email or In-Person), undated, uploaded February 4, 2016
- English Contact Card dated April 18, 2016
- English Interview Guide dated May 12, 2016
- English Consent Form for Healthcare Providers dated June 29, 2016
- English Consent Form for Women dated June 29, 2016

Your request for a French exemption is approved.

The REB no longer requires a 'valid until' date at the bottom of all approved informed consent forms. The consent forms currently approved for use by the REB are listed above.

If the study is to continue beyond the expiry date noted above, a Renewal Form should be submitted to the REB approximately six weeks prior to the current expiry date. If the study has been completed by this date, a Termination Report should be submitted.

The Ottawa Health Science Network Research Ethics Board (OHSN-REB) was created by the merger of both the Ottawa Hospital Research Ethics Board (OHREB) and the Human Research Ethics Board (HREB) for meetings held at the University of Ottawa Heart Institute.

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OHSN-REB complies with the membership requirements and operates in compliance with the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans; the International Conference on Harmonization - Good Clinical Practice: Consolidated Guideline and the provisions of the Personal Health Information Protection Act 2004.

Yours sincerely,

Francine F-A Sarazin, Ph.D., C.Psych.
Vice-Chairperson
Ottawa Health Science Network Research Ethics Board

FFAS/kd



**Ottawa Health Science Network Research Ethics Board/ Conseil d'éthique de la recherche du
Réseau de science de la santé d'Ottawa**

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February 12, 2018

Dr. Julian Little
University of Ottawa
Faculty of Medicine
Department of Epidemiology and Community Medicine
451 Smyth Road, Room 3257
Ottawa, ON K1H 8M5

Dear Dr. Little:

**Re: Protocol # 20160134-01H Woman-centered cervical screening: identifying women's
preferences and factors related to their preferences in cervical
cancer screening**

Thank you the email of February 12, 2018 from Brianne Wood.

The following documents are approved:

- Protocol Amendment Report dated December 1, 2017
- Revised Protocol (Version 8.1) dated February 10, 2018
- Revised English Contact Card/Recruitment Methods dated February 10, 2018
- Revised English Interview Guide dated February 10, 2018
- New English Participant Informed Consent Form- Women dated February 10, 2018

Date of approval: February 12, 2018

Ethical approval remains in effect until June 28, 2018. If the study is to continue beyond the expiry date, a Renewal Form should be submitted to the REB, in hardcopy. All Annual Renewal Reports, regardless of review type (i.e., full board or delegated), must now be submitted according to the full board meeting submission deadlines AND at least 30 days prior to the expiry date of the study to prevent a lapse in approval. If the study is completed by this date, a Termination Report should be submitted.

Appendix B: Published Manuscripts and Permissions

1. Systematic review protocol:

Wood, B., Van Katwyk, S. R., El-Khatib, Z., McFaul, S., Taljaard, M., Wright, E., Graham, I.D., Little, J. (2016). Eliciting women's cervical screening preferences: A mixed methods systematic review protocol. *Systematic Reviews*, 5(1), 136. <https://doi.org/10.1186/s13643-016-0310-9>

2. Permission BioMed Central:

Chapter 2 [Manuscript 1]:

Wood, B., Lofters, A., & Vahabi, M. (2018). Strategies to reach marginalized women for cervical cancer screening: A qualitative study of stakeholder perspectives. *Current Oncology (Toronto, Ont.)*, 25(1), e8–e16. <https://doi.org/10.3747/co.25.3851>

3. Permission from Current Oncology (Multi-med)

Chapter 3 [Manuscript 2]:

Wood, B., Russell V.R., El-Khatib, Z., McFaul, S., Taljaard, M., Wright, E., Little, J., Graham, I.D. (2018). “They should be asking us”: a qualitative decisional needs assessment for women considering cervical cancer screening. *Global Qualitative Nursing Research* [Accepted May 2018].

PROTOCOL

Open Access



Eliciting women's cervical screening preferences: a mixed methods systematic review protocol

Brianne Wood^{1*} , Susan Rogers Van Katwyk¹, Ziad El-Khatib^{2,3}, Susan McFaul⁴, Monica Taljaard^{1,5}, Erica Wright⁶, Ian D. Graham¹ and Julian Little¹

Abstract

Background: With the accumulation of evidence regarding potential harms of cancer screening in recent years, researchers, policy-makers, and the public are becoming more critical of population-based cancer screening. Consequently, a high-quality cancer screening program should consider individuals' values and preferences when determining recommendations. In cervical cancer screening, offering women autonomy is considered a "person-centered" approach to health care services; however, it may impact the effectiveness of the program should women choose to not participate. As part of a larger project to investigate women's cervical screening preferences and correlates of these preferences, this systematic review will capture quantitative and qualitative investigations of women's cervical screening preferences and the methods used to elicit them.

Design and methods: This mixed methods synthesis will use a thematic analysis approach to synthesize qualitative, quantitative, and mixed methods evidence. This protocol describes the methods that will be used in this investigation. A search strategy has been developed with a health librarian and peer reviewed using PRESS. Based on this strategy, five databases and the gray literature will be searched for studies that meet the inclusion criteria. The quality of the included individual studies will be examined using the Mixed Methods Appraisal Tool. Three reviewers will extract data from the primary studies on the tools or instruments used to elicit women's preferences regarding cervical cancer screening, theoretical frameworks used, outcomes measured, the outstanding themes from quantitative and qualitative evidence, and the identified preferences for cervical cancer screening. We will describe the relationships between study results and the study population, "intervention" (e.g., tool or instrument), and context. We will follow the PRISMA reporting guideline. We will compare findings across studies and between study methods (e.g., qualitative versus quantitative study designs). The strength of the synthesized findings will be assessed using the validated GRADE and CERQual tool.

Discussion: This review will inform the development of a tool to elicit women's cervical screening preferences. Understanding the methods used to elicit women's preferences and what is known about women's cervical screening preferences will be useful for guideline developers who wish to incorporate a woman-centered approach specifically for cervical screening guidelines.

Systematic review registration: PROSPERO CRD42016035737

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Background

Health care providers and public health organizations have enthusiastically promoted the importance of cancer screening for several years as some cancer screening programs have been found to reduce mortality from the disease [1, 2]. However, debates about the risks and benefits of cancer screening are occurring in the academic and the public domains [3–5]. For example, several organizations do not recommend population-based prostate-specific antigen testing because the harms outweigh the benefits [6–8], and recent publications question the optimal age range for mammography screening [9, 10]. In some jurisdictions, people are offered choices about what method of colorectal cancer screening they would prefer, though access to these options may be variable [11–14]. While most health professionals agree that personalized cancer screening would benefit their patients, the decision-making process is usually left to clinicians. Often, patients are not presented with a decision between screening options for pragmatic reasons: health systems are organized to evaluate and reimburse clinicians based on specific guideline adherence (which may not include a personalized pathway), clinicians may disagree with personalized recommendations, or certain options may require extra resources [15]. Additionally, health professionals have expressed concern that options may deter individuals from pursuing cancer screening, which could affect cancer rates among the population, although there appears to be no evidence to substantiate this [13, 15, 16]. The benefits of early cancer detection and potential treatment must be weighed against harms such as complications from the screening process (e.g., perforation from colonoscopy), potential for false positives and false negatives, anxiety resulting from receiving abnormal results, and treatment of conditions that may not have developed into a problem, which can vary widely based on a patient's age and health status [15].

Cervical cancer screening has avoided some of the media controversy that has affected breast, prostate, and recently colorectal screening programs because the Papanicolaou (Pap) test has successfully reduced the incidence of invasive cervical cancer and associated mortality [17–19]. In Ontario, Canada, the provincial cancer agency now recommends a new method of screening—human papillomavirus (HPV) testing—because of its increased sensitivity for detecting cervical abnormalities and high negative predictive value for high-grade cervical intra-epithelial neoplasia, compared with Pap testing [20]. Such a change in screening test would require cervical screening stakeholders to make decisions about the age range during which screening should be offered, options for sample collection methods, options for the laboratory testing, screening frequency, and recommended follow-up algorithm. In particular, HPV testing has the option for self-collection, which would allow women to take their own samples without the supervision

of a health care professional, though possibly at a cost of slightly decreased diagnostic performance [21, 22]. This strategy could have implications for access to screening, screening frequency, follow-up algorithms, and how screening results are communicated to women. As for most other health care decisions, women and health care professionals would be expected to make cervical screening choices that maximize the potential for early detection of a disease and minimize harm [23]. Population-level benefits and harms are balanced on a broader scale, looking at reduced disease-specific morbidity and mortality compared to overly intensive screening strategies [24]. Cancer screening decisions such as this one are inherently preference sensitive at both the individual level and program level, as decision-makers are required to understand how they value the benefits and harms for the different elements of the decision [25–27]. Therefore, individuals can identify their preferred choice by considering how they value each option's benefits and risks.

According to the Ottawa Decision Support Framework (ODSF), *preference* refers to the most appealing option of a decision for an individual, which is identified through *values clarification* [28, 29]. Values clarification is a process in which individuals identify the characteristics that matter most to them for a specific health decision. In cancer screening, individuals would compare and contrast what the tests measure, their diagnostic performance, how the tests are performed, follow-up procedures, and the potential consequences of participating in screening, and, subsequently, identify a plan to act on these preferences [30]. In the ODSF, values clarification and preference elicitation are part of shared decision-making processes, in which individuals are supported in these exercises by a decision support person [29]. Llewellyn-Thomas and Crump identified interactive and non-interactive, fine-grained, and coarse-grained approaches to values clarification and preference elicitation for health care decision-making in their 2013 review [29]. As the authors note, there are conceptual and methodological issues between the different values clarification/preference eliciting approaches, and further research is required to determine the appropriate approach based on different subgroups of patients. Considering the evolving and complex evidence in cervical cancer screening, it is imperative that women are supported in these preference-sensitive health care decisions, for individual-level decision-making and through policies that reflect women's preferences.

Systematic reviews have shown that more women may participate in cervical screening if they can self-collect their samples [31, 32]. Other reviews have indicated that women tend to prefer active follow-up of abnormal screening results, predominantly referral to colposcopy, to observation, and predominantly repeated Pap tests [33]. In studies investigating women's views on HPV testing as part of cervical cancer screening [34], some respondents had trouble

interpreting information about cervical cancer screening [33]. This was related to psychosocial burden of positive results, acceptability of the testing process, and women's informational needs [34]. There is weak evidence that women's participation in cancer screening is related to the presentation of the screening information as well as emotional and cognitive responses to the invitation to be screened [35]. While there is a growing body of evidence focusing on women's attitudes and beliefs in cervical screening, it is unclear whether these studies consider women's preferences in terms of making informed decisions, which undermines the potential for improved screening for women. To the best of our knowledge, no systematic review has described or compared methods for eliciting women's cervical screening preferences and the outcomes of these approaches. The proposed review will contribute to an assessment of women's needs in making decisions about primary cervical cancer screening and inform policy-makers and guideline developers about these needs. These findings will be used in an investigation of women's cervical screening preferences in Ontario, Canada.

Aim

The objectives of this systematic review are

- (1) to synthesize and describe what is known about methods used to elicit women's preferences in primary cervical cancer screening;
- (2) to summarize the evidence on women's preferences for cervical cancer screening.

Methods

This protocol has been prepared following Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) guidelines for systematic review protocols [36]. The PRISMA-P checklist is included as Additional file 1. The review protocol has been registered with PROSPERO (CRD42016035737).

Conceptual framework

This systematic review is part of an assessment of women's needs in identifying their preferences and making decisions about cervical cancer screening, taking into account issues laid out in the ODSF [28, 37]. We will apply this framework to conduct a thematic analysis of the synthesis results, looking at determinants of preferences and decision-making, methods used to support decision-making or elicit preferences, and short- and long-term outcomes [28, 29, 37]. In addition to summarizing different techniques of eliciting preferences, we will compare theoretical frameworks used to elicit preferences, processes used to design these techniques, outcomes including construct validity, clinical outcomes, and decision

quality, the identified preferences for cervical cancer screening, and how these preferences are influenced by context.

Eligibility

We will evaluate qualitative and quantitative studies that describe and/or examine methods used to elicit women's cervical screening preferences. These studies will involve women being presented with multiple options for primary cervical cancer screening and identifying the most favored option [29]. We will include studies of women who are presented with options at the point of decision-making, as well as women presented with a hypothetical decision. The criteria for inclusion in this review are summarized in Table 1. For all study designs, we will examine studies in which women are 18 years of age or older and are eligible for cervical cancer screening. We will include articles that are based on direct enquiry with eligible women with no restriction on publication dates or publication status. For articles not published in English that the study team is unable to translate, we will attempt to contact the study authors to see if there are any English translations available. All study designs will be included. Mixed methods studies that satisfy corresponding inclusion and exclusion criteria will also be included (eligibility will be determined separately for the quantitative and qualitative components). We will examine each of the primary studies from any systematic review or meta-analysis for individual inclusion into the review. Opinion pieces will not be included.

Information sources

We will search MEDLINE, Embase, PsycINFO, CINAHL, and Cochrane Library databases, for English articles from

Table 1 Eligibility criteria for studies in systematic review

Inclusion criteria	Exclusion criteria
- Participants: women aged ≥ 18 years	- Interventions targeting health care providers, policy-makers (e.g., program decision-makers), or technical aspects of cervical cancer screening or treatment
- Assessment of women's cervical screening preferences, either current or hypothetical	- Reports of interventions that aim to increase cervical screening uptake in which the content of the intervention was not described or evaluated
- Investigation of decision-making needs of women who are considering cervical cancer screening	- Studies reporting predictors of cervical screening uptake that are not related to a decision-making process
- Both qualitative and quantitative study designs	- Opinion pieces
	- Studies that do not adequately report the study design, methods, and interpretation of the research
	- Studies that do not clearly demonstrate ethical conduct or ethical reporting of research

inception to March 2016. The gray literature will be searched by scanning ProQuest Dissertations and Theses, relevant Canadian health research/policy databases, Canadian health information networks, and known relevant Canadian conference proceedings. Our gray literature search is limited to Canadian resources as the findings from this review will inform an investigation of cervical screening preferences in the Canadian context. A search strategy and its translations to other databases were developed in collaboration with a health sciences librarian at the University of Ottawa and peer reviewed using PRESS [38]. These strategies combined free-text words and subject headings, derived from the following broad topic areas: cervical cancer, early detection of cervical cancer, and patient preferences. The MEDLINE search strategy is appended (Additional file 2). We will search citations of the included studies using SCOPUS to supplement our search of the main bibliographic databases. Reference lists of articles that were selected for full-text review will be scanned for additional relevant material. Subject matter specialists on the review team will be asked to identify further resources for inclusion.

Study identification and data extraction

Two of three reviewers will independently review the studies in three stages: (i) title and abstract screening for inclusion; (ii) full-text screening for inclusion; and (iii) data extraction for eligible studies. All studies which initially appear to meet the inclusion criteria but on inspection of the full-text paper are not eligible will be detailed in a table. We will identify single studies that are reported in multiple publications by juxtaposing the author lists and comparing sample size and setting characteristics. Multiple reports of the same study will be considered for inclusion, but the data will be extracted for single studies from the report deemed the most relevant by the two reviewers. Variation in results reported in multiple publications will be noted as limitations in the appraisal of each study. Characteristics of

excluded studies' together with reasons for their exclusion will be recorded. Disagreements regarding eligibility of studies will be resolved by the third reviewer.

Data will be extracted independently by two of the three reviewers, according to the list in Table 2. Mendeley (www.mendeley.com) and Covidence (www.covidence.org) will be used for citation management, screening, and data extraction. Data will be extracted to preserve the context of the findings, as described by Sandelowski et al. [39] using pre-defined data extraction forms. Descriptive themes of preference eliciting methods and preferred cervical screening options will be developed for each primary study based on the text-in-context results [39]. Differences will be resolved through discussion between the two reviewers, and if necessary, through final consensus with a third reviewer. Customized data extraction sheets will be pilot tested by all reviewers prior to the data extraction phase.

Outcomes

Objective 1: The first objective is to synthesize what is known about methods used to elicit women's preferences in primary cervical cancer screening, including

- Approaches used to elicit women's preferences for screening technologies (e.g., interactive versus non-interactive, coarse-grained versus fine-grained, other);
- Methods used to evaluate the appropriateness of women's preferences (e.g., strength of preferred option, change in decisional conflict, acceptability of option's characteristics, change in knowledge, attitudes, or behavior);
- Reported indicators of effectiveness of methods to elicit preferences (e.g., observed strengths of preferences, observed changes in decisional conflict, observed acceptability of options, observed changes in knowledge, attitudes, or behavior);
- Advantages and disadvantages of the different methods to elicit preferences.

Table 2 Data extraction fields for synthesis

Study details	Eligibility	Study design	Clinical/decision-making issues	Summary
Author, year	Direct enquiry with	Methodological characteristics/	Method/procedure of	Authors' overall
Reference ID	women considering	epidemiological design	preference elicitation (and/	conclusions
Publication type (i.e., full	screening (18+ years)	Population	or values clarification)	Quality assessment
report or abstract)	Preferences elicited	Setting/context	Design process of preference	Reviewer's comments
Country of corresponding	(options presented,	Time period	elicitation/values clarification	
author	women indicate	Method of recruitment	technique(s)	
Sources of funding	most favored option)	Method of data collection	Comparators	
Ethical considerations		Method of data analysis	Results (preferred option, strength	
(approval from committee,		Theoretical frameworks	of preferred option, evaluation	
informed consent, positive		Concept definitions	of preference eliciting technique)	
risk/benefit assessment)			Total number of participants	
			Objective 1: magnitude and	
			significance; study-specific conceptions	
			Objective 2: magnitude and	
			significance; study-specific conceptions	

Objective 2: The second objective is to summarize the evidence on women's preferences for cervical cancer screening, including

- List of options in cervical screening decisions for women;
- Women's preferences for cervical screening (by proportions), by jurisdiction;
- Description of important values for making cervical screening decisions.

Quality assessment/risk of bias

The Mixed Methods Appraisal Tool (MMAT) [40, 41] will be used to appraise the qualitative, quantitative, and mixed methods studies for this review. Two reviewers will independently appraise relevant studies, and disagreement will be resolved through consensus, in discussion with a third reviewer. Studies will be included in the final review if they score 50 % or greater, according to the scoring scheme [40].

The Grading of Recommendations, Assessment, Development and Evaluation (GRADE) guidelines will be used to present a summary of findings and to qualify the strength of individual findings for the quantitative and qualitative syntheses separately, using GRADE profiler and the Confidence in the Evidence from Reviews of Qualitative Evidence (CERQual) tool, respectively [42–45].

Analysis

The quantitative and qualitative evidence will be analyzed separately and then combined in the final stage of analysis, similar to Goldsmith's integrated methodology [46]. We expect considerable heterogeneity between studies and therefore that a statistical meta-analysis would not be appropriate. A tabular summary will be created to describe all of the characteristics of the included studies, as well as key preference eliciting methodologies [47], the advantages and disadvantages of their approaches, and their conclusions regarding women's preferences based on the text-in-context analysis.

Objective 1: We will produce a quantitative evidence table that describes the main types of preference eliciting methods identified in the quantitative body of literature and may also contain relevant evaluative measurements of the different techniques. Methods will be categorized using themes from a textual analysis technique when reviewing the qualitative literature, using the classification as outlined by Llewellyn-Thomas and Crump [29] as an initial reference. A similar evidence summary table of the included qualitative studies will be developed, conveying study characteristics, context, and outstanding themes. In both of these summary thematic tables, we will also include a column that indicates if and how the themes map onto the ODSF concepts.

Objective 2: Women's preferences will be organized into themes using the text-in-context analysis and presented in a table, alongside the options available and values that women identified as important for their decision-making. Quantitative evidence will be synthesized and presented separately from the qualitative evidence.

For both objectives, summary of findings tables will be developed according to the GRADE guidelines (i.e., GRADE and CERQual) for syntheses of the quantitative and qualitative evidence separately. Evidence profiles [46] will be created for each finding to determine recommendations for eliciting women's cervical screening preferences, for each objective. These profiles will be presented in a table that denotes which studies contribute evidence toward a particular theme, the associated type of evidence (i.e., qualitative or quantitative), related quality appraisal scores from the MMAT, the consistency of the finding across the studies (e.g., noting any contradictions or missingness), the applicability of the evidence to the Ontario/Canadian population, and any other factors that might influence the usability of the evidence. Each evidence profile will include overall assessments from the GRADE and CERQual summary of findings tables. Using the categories defined by Goldsmith et al. [46], analytic themes will be graded as "definite" or "suggestive," based on the GRADE and CERQual assessments. Items which were classified as "high" or "moderate" according to GRADE or CERQual will be given a "definite" recommendation, and items classified as "low" and/or "very low" will be denoted "suggestive." This method of synthesis will clarify the important dimensions of eliciting women's preferences for cervical screening, giving equal value to quantitative and qualitative evidence. "Definite" recommendations from the evidence profile will be used to develop a tool to elicit women's screening preferences in practice. "Suggestive" recommendations will be discussed with local stakeholders to determine their relevance for this particular project and can be used to guide further research on women's screening preferences.

Discussion

This study will be the first systematic review synthesizing methods used to elicit preferences for cervical cancer screening. This review constitutes the first part of a needs assessment for the development of a tool to elicit women's preferences for different cervical cancer screening options. The findings of this review will inform a qualitative investigation of women's cervical screening decision-making in Canada.

Strengths and limitations

Recommendations for mixed methods systematic reviews were developed only recently [46, 48–50], and therefore, best practices have not yet been fully established. While

this study will use some validated approaches to synthesizing quantitative and qualitative evidence, it may be challenging to apply the results of this synthesis because of the heterogeneity of the evidence. This mixed methods approach values quantitative and qualitative evidence equally, offering a more complex understanding of how women make decisions about cervical cancer screening.

As with any knowledge synthesis, this review will be subject to particular biases, including publication bias and language bias. This review will be examining the gray literature to attempt to find relevant unpublished work. If there are relevant articles identified that are not published in English, the authors will seek to translate the document internally or will attempt to retrieve a translation by contacting the author. We expect that the heterogeneity in definitions of “preference” and potential reporting biases may complicate the synthesis of across a diverse body of literature. This review will contribute to improved understanding of how best to elicit cancer screening preferences from women, which women, health care providers, and policy-makers may value. A thematic synthesis may not provide the same certainty as a meta-analysis, though an evidence table with descriptive themes is important for comparing quantitative and qualitative evidence and will be useful for generating recommendations from the final aggregation.

This knowledge synthesis will summarize and compare the content and methodologies used to elicit women's preferences of cervical screening. A pooled synthesis will highlight what is known about methods used to elicit women's preferences in cervical screening and their identified preferences across a diverse body of evidence. This information will contribute to the design of a tool to elicit women's preferences for cervical screening for potential use by health professionals in developing woman-centered cervical screening recommendations. These findings will also suggest gaps in the knowledge of women's cervical screening preferences that can be addressed in future research.

Additional files

Additional file 1: PRISMA-P checklist. (PDF 219 kb)

Additional file 2: Search strategy for MEDLINE. (PDF 100 kb)

Abbreviations

CERQual, Confidence in the Evidence from Reviews of Qualitative Evidence; GRADE, Grading of Recommendations, Assessment, Development and Evaluation; HPV, human papillomavirus; MMAT, mixed methods appraisal tool; ODSF, Ottawa Decision Support Framework; Pap, Papanicolaou

Acknowledgements

We would like to acknowledge Mish Boutet at the University of Ottawa for providing useful guidance in developing a final search strategy. We would also like to acknowledge Jesse Elliott—who recently joined the team as a reviewer—for her systematic review expertise.

Funding

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Availability of data and materials

Not applicable.

Authors' contributions

BW conceived the study design and methodology, helped to devise a search strategy, and wrote the first draft of the manuscript. SRVK contributed to the study methodology, helped to construct a data extraction template, helped write the first draft of the manuscript, and revised the later versions. ZEK contributed to the study methodology and helped revise the manuscript. SM contributed to the study design and helped revise the manuscript. MT contributed to the study methodology and helped revise the manuscript. EW led the development of the search strategy, searched the translations, helped to execute the search, and helped revise the manuscript. IDG helped conceive and design this study and helped revise the manuscript. JL helped conceive and design this study and helped revise the manuscript. All authors read and approved the final manuscript.

Competing interests

The authors declare that they have no competing interests.

Ethics approval and consent to participate

There are ethical considerations to consider in the synthesis of primary research studies [51]; however, approval was not required for this analysis. No individual-level data will be presented, and we will exclude studies that do not reflect sufficient ethical conduct.

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Strategies to reach marginalized women for cervical cancer screening: A qualitative study of stakeholder perspectives

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ABSTRACT

Background Self-sampling for human papillomavirus (HPV) has the potential to reach marginalized populations that are underserved for cervical cancer screening. However, before implementing an alternative screening strategy such as self-sampling for under- and never-screened women, the key processes, facilitators, and barriers to reform need to be understood.

Methods A descriptive qualitative study was conducted that involved semi-structured interviews with Canadian and international cancer screening health care providers and policy-makers. Respondents were purposively selected from a list of thirty stakeholders generated through an environmental scan. The interviews were transcribed verbatim and analyzed using directed content analysis.

Results Nineteen stakeholders participated in the interviews. Most respondents thought that self-sampling was an appropriate cervical screening alternative for hard-to-reach populations, as it addressed barriers to cervical screening related to various social determinants of health. All respondents emphasized that transitioning to HPV primary screening would catalyze a policy shift towards self-sampling. Clinician respondents were less enthusiastic about self-sampling strategies since that discouraged women's appointments with primary care providers, because cervical screening offered an opportunity to discuss other preventive health topics. There also was little consensus between respondents on whether the state of evidence was satisfactory to integrate a self-sampling option into policy, or whether more Canadian research was needed.

Conclusion Canadian cervical cancer screening stakeholders should collaborate to identify the knowledge gaps that researchers should address and leverage the existing literature to implement tailored, patient-centred alternative cervical screening strategies. The transition to HPV primary screening would be a key first step in the broad implementation of HPV self-sampling in Canada.

Key Words Cervical cancer, HPV self-sampling, barriers, cervical cancer screening, social determinants of health, vulnerable population

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INTRODUCTION

Cervical cancer screening programs using cervical cytology (the Pap test) have substantially reduced the incidence of cervical cancer and consequent mortality over the past several decades¹. Despite investment in health promotion initiatives, incidence and mortality have remained unchanged in North American and European countries in the past twenty years²⁻⁵. Demographic shifts (e.g., increased immigration from countries without screening programs), changing social behaviours, limitations of cervical cytology,

and suboptimal program delivery could play a role in the plateau of cervical cancer mortality in countries with early detection programs^{4,3}.

Recent cervical cancer prevention and control innovations target persistent infection of oncogenic human papillomavirus (HPV)—a necessary cause of cervical cancer^{6,7}. These include HPV vaccination programs that target school-age girls before exposure to HPV and HPV DNA primary screening test, a test with greater sensitivity and higher positive predictive value than Pap testing^{6,8-13}. Currently, many jurisdictions, including most Canadian

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provinces, are considering replacing the Pap test with HPV testing, which may overcome some of the performance limitations of cytology, particularly the low positive predictive value in the era of HPV vaccination. While the diagnostic performance of an HPV test is considered superior to traditional cytology¹⁴, a new screening test will not overcome the suboptimal performance of early detection programs. Opportunistic programs and barriers to accessing cervical cancer screening^{2,15-18} will still largely affect the impact of early detection programs on cervical cancer mortality. Most concerning is that these methods fail to reach populations that are already marginalized, including newcomers to Canada, Indigenous peoples, and women in rural/remote regions^{16,19,20}.

Human papillomavirus testing can be performed on a vaginal sample that women can collect themselves—"self-sampling"—which might be an attractive option to overcome some of the barriers to provider-administered screening²¹⁻²⁵. Though there is high-quality evidence internationally to support self-collected sampling in population-based programs, only the Netherlands and Australia have implemented a self-sampling strategy as part of their population-based program. A recent review¹⁸ found that the evidence indicates HPV testing on self-samples is clinically comparable with provider-collected samples and that it could improve screening rates in some populations.

With such a low implementation rate of self-sampling in cervical cancer early detection programs, there is a gap between the evidence to support a self-sampling option and current policy and practice²⁶. From an equity perspective, self-sampling might increase access to cervical cancer screening for hard-to-reach populations, which is important for tailored screening strategies. Understanding the facilitators and barriers to offering self-sampling is an important step for developing equitable, evidence-informed cervical cancer screening programs.

This qualitative study aimed to explore barriers and facilitators to implementation of HPV self-sampling in Canadian cervical cancer screening programs, from provider and policy-maker perspectives. This study was part of a larger project that included a symposium on HPV self-sampling in October 2016 (www.equitycancerscreening.ca) that brought together national and international researchers, front-line workers, and policy-makers.

METHODS

Design

A descriptive qualitative study was conducted that involved semi-structured interviews with Canadian and international cancer screening health care providers and policy-makers. This study was part of a larger project that brought together national and international researchers, front-line workers, and policy-makers.

Data collection

A list of potential stakeholders was compiled during a preliminary environmental scan, with an aim to purposively sample relevant stakeholders from across Canada in addition to ensuring diverse professional representation. Family physicians, gynecologists, colposcopists, cyto-pathologists,

researchers, guideline developers, cancer screening program managers and directors, and women's health advocacy organizations were targeted.

One-on-one interviews were conducted by phone or in-person, depending on participants' preferences, by the study research assistant (BW). The interviews lasted between 30 and 60 minutes and were audio-recorded. Demographic characteristics were collected through a brief survey completed by respondents at the beginning of the interview. A semi-structured interview guide contained four main questions: 1) *What are the reasons for the observed cervical cancer disparities among vulnerable populations and how could they be addressed?*, 2) *What are your views in general on the use of alternative cervical cancer screening formats, and HPV self-sampling in particular?*, 3) *What are the knowledge gaps in adopting HPV self-sampling into the cervical cancer screening program in your province or area?*, and 4) *What resources/changes are required for adopting HPV self-sampling as part of screening programs?* The data were considered "saturated" when no new themes or understanding emerged^{27,28}. Interviews were conducted over 4 months, from May to August 2016. The study was approved by the Ryerson University Research Ethics Board.

Analysis

The audio-taped interviews were transcribed verbatim, producing 140 pages of data. Data were then analyzed using an inductive interpretive approach²⁹, which allows relevant themes and categories of analysis to emerge from the transcribed interviews. NVivo11 (NVivo qualitative data analysis Software; QSR International Pty Ltd. Version 11, 2016) was used to organize and manage the data for the analysis. The transcripts were reviewed independently by the research team to identify coding patterns and collectively engage in data interpretation to ensure rigour, auditability, and trustworthiness.

RESULTS

Data saturation was reached after nineteen interviews were conducted with cervical screening stakeholders from Canada, Denmark, and the Netherlands. The study respondents included 14 females and 5 males. Socio-demographic surveys were completed by 16 of the 19 (85%) respondents. Three respondents who participated in the interview failed to return the survey after three follow-up emails. Table 1 depicts the participants' socio-demographic characteristics.

Below, we describe the main themes that emerged, categorized by the four interview questions.

1) What are the reasons for observed cervical cancer screening disparities among vulnerable populations?

Social determinants of health

All respondents understood "vulnerability" through the social determinants of health framework, specifically tailoring the definition of need to their professional role. Individuals who did not attend cervical screening or faced barriers in accessing screening were considered vulnerable or at increased risk of developing cervical cancer because

TABLE 1 Study participants' socio-demographic characteristics

Characteristic	Number of respondents N=16 (%)
Age	
Mean age±SD	48.58
Range	38–64 years
Location of respondent	
Eastern Canada (Newfoundland, Nova Scotia, Prince Edward Island, New Brunswick)	1 (5.3%)
Central Canada (Quebec, Ontario, Manitoba)	12 (63.1%)
Western Canada (Saskatchewan, Alberta, British Columbia)	3 (15.8%)
Northern Canada (Yukon, North West Territories, Nunavut)	1 (5.3%)
International (Europe)	2 (10.5%)
Ethno-racial identity	
White/Caucasian	12 (75%)
Other	4 (25%)
Professional training	
Health administration/Policy analyst (e.g., screening program administrator)	10 (62.5%)
Medicine (e.g., family physician, gynaecologist)	6 (37.5%)
Place of work	
Hospital/community centre	5 (31.3%)
Cancer screening program	2 (12.5%)
Government (ministry of health, public health agency)	4 (25%)
Other (e.g., research institute)	5 (31.3%)

of socio-demographic issues, including rural/remote/Northern geography, ethnicity, education level, gender and sexuality, and income. Different social determinants were associated with different challenges to accessing screening, suggesting that multi-pronged approaches at the individual, provider, and system level are important for reaching under- or never-screened women. Health care resources (e.g., primary care providers) are scarce in rural and remote areas, making it difficult for women to access screening if they desired it. More importantly, certain barriers to screening may compound when they intersect with one another. As one researcher noted, Northern Indigenous communities not only experience challenges with available providers but also mistrust the health care system because of their history of colonization. This means that a provider-administered test may not always be feasible. Women in these communities may not be able to prioritize preventive health because they may be struggling to fulfill basic needs, such as feeding their families. This is captured in the following quote:

“From the perspective of the communities [First Nations] that we are working with, there seem to be a number of structural factors that impact health and access or lack of access to care, and then a number of factors that are also influenced by culture and social determinants and historical legacy of colonization... Food security is a significant issue in some of the communities. Some of the other communities have challenges with sub-

stance use... that makes ... difficult for women to prioritize their own care.” (Subject 1604)

A primary care provider emphasized that providers often struggle to engage individuals who are marginalized because of their gender or sexuality, because the providers are afraid or unsure of how to deliver cervical cancer screening, due to the gendered nature of the test.

“Providers need to focus more on the LGBT population. I think that in general they [LGBT] face barriers much like the other ones to getting care and a lot of it has to do with either homophobia or transphobic providers.” (Subject 1615)

More than a third of the respondents felt that the invasive nature of a Pap test (through a pelvic exam) may also deter participation for women who have experienced trauma or sexual abuse. The following quote shows how a female primary care provider felt that reframing the communication about cervical cancer screening as an empowerment strategy may allow abused women to take control over their own health.

“I’ve had lots of conversations over the years [with sexually abused] women who did not want pelvic exam. We discuss how the abuser took away their agency, caused them PTSD... and they should not allow their abuser to also give them cervical cancer... by having a pelvic exam they are not

letting their abuser to continue having negative harm on their life.” (Subject 1637)

2) Views of alternative cervical screening programs, self-sampling in particular

Perspectives of self-sampling – At the patient level

The majority of the respondents felt that self-sampling would be a great option, not only for women who face barriers in accessing primary care providers, but also for the general population. Respondents noted that while some marginalized groups may not have a primary care provider, or may not feel comfortable during a gynecological exam, the option of self-sampling may be appealing to all women since it provides flexibility.

“If there’s another option like HPV self-sampling for HPV, why wouldn’t we be doing that?” (Subject 1615)

However, some felt strongly that self-sampling should only be offered to women who are otherwise not attending the organized screening program, because the decreased diagnostic performance of self-sampling could negatively impact population health.

“If we want screening that works well and detects lesions in that capacity, you need to have a cervical sample. A self-sample is not a cervical sample it is a vaginal sample.” (Subject 1645)

Perspectives of self-sampling – At the clinician level

Many front-line respondents discussed how cervical cancer screening often provided opportunities for clinicians to engage in discussion about other health concerns, like prenatal care, sexually transmitted infection testing, and other cancer screening. Most expressed concern that a self-sampling option in cervical screening would minimize those opportunities.

“I do think when you actually do a pelvic examination there’s a bunch of other things, [you] can see lesions...you can talk about contraception, you can look for genital warts... vulvar-cancer, vaginal cancer,... so not a big fan of just chucking out the clinician pelvic exam.” (Subject 1637)

These clinicians felt that there needed to be safeguards in place before offering self-sampling, because taking one preventive practice out of the doctor’s office could affect access to a range of preventive health care.

Perspectives of self-sampling – At the system level

At the system level, although most felt that HPV self-sampling could improve system-level efficiencies, many felt that those efficiencies would only be present after “economies of scale” were achieved at the laboratory level. However, a few respondents highlighted that for self-sampling to effectively reach the under-screened population, there would still need to be excessive use of health human

resources, which could increase cost. The need for follow-up algorithms for women who test positive was emphasized. These concerns are reflected in the excerpt below.

“We think that at the end, self-sampling will also turn out to be cheaper than having a smear at the office because you circumvent the GP [general practitioner] route, which also involves some costs. But what implementation strategies are needed to maximize benefits while minimizing harms?” (Subject 1625)

3) What are the knowledge gaps in adopting HPV self-testing into the cervical cancer screening program in your province or area?

Respondents interpreted “knowledge gaps” to mean gaps for health professionals who deliver cervical screening, for policy-makers who use research to create guidelines, and for researchers who are trying to understand how to optimize cervical screening programs.

Knowledge gaps among providers

Practices like self-sampling take health practices out of the clinic, which instilled some anxiety in health care providers who could feel unprepared to appropriately advise or counsel patients, given the several proposed changes to existing cervical screening practice.

“From my perspective, some of the takeaways with that were that there needs to be a lot more education of care providers. We shouldn’t assume that everybody out practicing is fully up to date even if they’re working in the field because there hasn’t been a tremendous amount of literature on self-sampling in Canada published. To my knowledge, there are no policy statements yet from sogc [Society of Obstetricians and Gynecologists of Canada] or any of the other bodies that might be important.... So I think there does have to be quite a lot of education and that’s not just physicians, family physicians who might be doing it or ob-gyns, but increasingly in the communities that we’re in, nurse practitioners and others who provide that level of care...” (Subject 1604)

Scientific knowledge gaps

There was little consensus among respondents of what were the most important scientific knowledge gaps that prevented a shift in cervical screening practice. Examples provided by respondents included specific unknowns, such as the most appropriate device, as well as more complex gaps, for example scaling up pilot projects that test specific strategies in a small, controlled setting. Several respondents acknowledged that while much international research supported the use of self-sampling at a population level, there was little Canada-specific evidence available. Because of Canada’s unique geographies, populations, and multiple health systems, identifying the appropriate self-sampling delivery strategy is vital.

“I think we can look at data from other jurisdictions... But I think that it still makes sense for us to

do a made-in-Canada trial, so look at test properties, uptake, because some of the studies, the mail-in experience has not been very positive..." (Subject 1639)

"...maybe more rigorous data needs to be available for the province and cancer agencies to act on it... like timely follow-up for abnormal results positive results, triage and management guidelines..." (Subject 1615)

4) What resources/changes are required for adopting HPV self-testing as part of screening programs?

When considering how self-sampling could be integrated into current practice, respondents' perspectives were grouped into the following themes:

Introduction of the HPV paradigm in cervical screening

Respondents unanimously agreed that the shift to using HPV testing as a primary screening test was the most important change needed to be made to cervical cancer screening programs in Canada. There was, however, disagreement among respondents as to whether provinces should implement HPV primary screening in cancer screening programs prior to offering self-sampling to vulnerable populations. Respondents were divided as to whether an alternative screening program such as self-sampling could be implemented before all pieces, including laboratories that can handle HPV tests and recall information system and registry, were in place.

"We need the lab system for HPV testing, we need the appropriate engagement mechanisms, both directly for the physician [and] for the patient. So we need a way to say, you know, you're due for your test, you're not due for your test. We need really good information system around that, like that's going to be key." (Subject 1637)

"We will not be able to implement this [self-sampling] on a larger scale without a paradigm shift to primary HPV testing. So first and foremost that needs to happen." (Subject 1664)

"And once the testing paradigm is changed, then self-sampling is a shoe-in. It's a natural thing." (Subject 1645)

A few respondents were open to the idea of integrating self-sampling into a cytology-based program, but program administrators would need to identify an effective outreach strategy to reach the vulnerable populations.

"I think you could put [self-sampling] wherever it fits, I think the issue is how do you get women engaged with it." (Subject 1639)

Several participants highlighted challenges with implementing HPV primary screening in cancer screening programs. Understanding how to scale up existing knowledge of HPV primary screening strategies was part of the

problem, though political and financial concerns were perceived as the biggest barriers to change. A few suggested that politicians may not be as concerned about the health improvements introduced by the HPV test, but would be prone to changing policy if they were convinced that dollars were being saved.

"Their [politicians] concern is mainly the affordability of HPV primary screening, so we need to show them economic (cost/benefit) analysis that shows money can be saved" (Subject 1696)

Several respondents commented on the impact of the transition from cytology to HPV-based screening for cytotechnologists and their labs, stating that the profession stands to lose a substantial number of positions because HPV testing is a primarily automated process. Some other logistic issues with the transition to HPV testing include laboratory capacity to handle HPV tests in the volumes that are expected—most respondents believed that current infrastructure is not conducive to this switch.

Change to existing cervical screening program structure

Most respondents discussed how important it is that alternate screening programs adhere to the principles of a successful screening program, including a desirable cost-benefit and acceptability of the test. Many respondents agreed that program organization, follow-up coordination, and test quality are essential to implement new strategies. Beyond fundamental elements of a successful population screening program, respondents discussed ways to adapt existing cervical screening programs to meet the needs of Canadian women and to accommodate the advancing science.

"If you have an invitation system and you're able to identify women that have actually turned down invitations to screening, you might actually start a self-sampling program for non-responders... So it is really by far the best way, both from an organization point of view and from an HPV laboratory quality control point of view, to start at the same time with self-sampling for non-responders and HPV office-based testing for responders." (Subject 1625)

Four respondents mentioned that program managers should model self-sampling after the FOBT [faecal occult blood test]. Many Canadian FOBT programs offer an at-home sample collection option, which could guide how a HPV self-sampling option could be integrated into cervical screening programs. However, respondents noted that differences between the epidemiology of cervical cancer and colorectal cancer may limit comparisons.

New perceptions of cervical cancer screening

Shifting to an HPV vaccination and testing paradigm requires a shift in mentality toward cervical cancer prevention. Many respondents discussed the need to change social norms around cervical cancer and HPV. Most respondents mentioned how a shift in cervical cancer strategy could

go hand-in-hand with reducing the stigma of sexually transmitted infections. Including men in prevention conversations and framing cervical cancer as an infectious disease instead of as a cancer were suggestions for ways to help change social norms.

“As much as we’ve had this in the cancer screening program...we probably need to be thinking about it more as an infectious disease and how would we be approaching this as an infectious disease.”
(Subject 1639)

Health system characteristics

Respondents noted that changing the social norms around cervical screening and HPV is important for reducing disparities in cervical cancer screening, but this change needs to be mirrored in the health system and gain political support. Barriers to offering alternative cervical screening formats were linked with how health systems are organized and how decisions are made, as well as the availability and accessibility of primary care and the political will of screening funders to change.

Some respondents were critical of their own health systems, many citing the lack of organized recall (e.g., outside of Ontario and British Columbia) and with competing priorities of the health system as being responsible for the lack of progress.

Respondents suggested that the disconnect between federal and provincial policies and practices caused some of the greatest barriers to creating change in their local cervical screening programs.

“Really what happened in our province is the pressure usually comes nationally once everyone else has started to do something, and we’re about one of the last ones.... That’s when things start changing for us.” (Subject 1688)

Responses did not vary by age or gender of respondent, though different perspectives were observed between different professions. Researcher respondents tended to feel more positive about self-sampling and felt confident that the current body of evidence would justify a self-sampling option for hard-to-reach women as part of a cervical screening program. Clinicians, on the other hand, were more likely to show apprehension about a self-sampling option, suggesting that self-sampling could be used if other options were exhausted. These respondents emphasized the importance of a relationship between a screening participant and their health care provider, and that cervical cancer screening is one component of the overall care plan. Policy-maker respondents tended to agree that self-sampling could benefit hard-to-reach women, particularly populations in rural/remote areas or certain cultural backgrounds, but that operationalizing the strategies requires more political support than is currently available.

DISCUSSION

This study is the first Canadian qualitative study that focused on policy-maker and health professional perspectives

of how HPV self-sampling could fit into existing Canadian cervical screening programs to promote hard-to-reach populations’ screening uptake. Seventeen respondents from across Canada and two international stakeholders discussed barriers to and facilitators of alternative screening formats, particularly HPV self-sampling, to engage hard-to-reach women with cervical cancer screening in the Canadian context. Respondents agreed that cervical cancer screening programs need to have tailored strategies to reach marginalized women with early detection opportunities. However, there was disagreement on whether the self-sampling evidence was sufficient to justify implementation in cervical cancer screening programs. More broadly, respondents noted that cervical cancer screening programs must incorporate HPV testing as primary screening before developing tailored strategies, like self-sampling, for hard-to-reach populations.

Clinicians felt that self-sampling might not address all the important barriers that vulnerable populations face when accessing cervical cancer screening, particularly poor access to primary health care, which is important for following up screening results. Researcher respondents were more enthusiastic about self-sampling from population coverage and cost-effectiveness perspectives, as were policy-makers, though respondents perceived undecided implementation logistics as a barrier to changes in a cervical cancer screening program. Respondents agreed that the idea of a self-sampling component of a cervical cancer screening program was attractive from a patient empowerment perspective, but knowledge translation strategy was important to engage with women, health care providers, and other cervical cancer screening stakeholders about a change in cervical cancer screening options. The findings from this research suggest that political priorities and system barriers may be important challenges to implementing tailored screening, like self-sampling, to reach marginalized women. Knowledge-user and researcher respondents in this study agreed on important action points, such as a need for provincial programs to shift to HPV primary screening, though they disagreed on whether self-sampling evidence was sufficient for implementation.

Though respondents understood that a shift to HPV primary screening represents a major initial investment from governments, most felt that “the science is enough” to warrant the transition from cytology-based cervical cancer screening to a program based on HPV testing. Canadian randomized controlled trials^{11,30}, and international knowledge syntheses^{10,14,25,31-33} also support this shift, which has been stressed in recent editorials^{6,34,35}, yet some policy-makers feel that the cytology-based programs are “good enough,” echoing the World Health Organization suggestion³⁶. Researcher respondents were particularly adamant that Canadian cervical cancer screening programs should shift towards primary HPV testing because the increase in sensitivity has the greatest impact on population health and health system spending, in the era of HPV vaccination^{6,37}. Though not mentioned by interview respondents, some research questions whether this improvement might be exaggerated in the literature because of industry sponsorship of research³⁸, which suggests that the shift may not be as “urgent” as some authors convey. In Canada, the Ontario government recently allocated funding for HPV

testing in primary cervical cancer screening in their 2017 budget. However, the implementation strategy has yet to be disclosed.

A similar disconnect was observed in respondents' perceptions of the self-sampling evidence base. Some respondents thought that we needed more rigorous Canadian evidence around target populations and mode of delivery of self-sampling. Most respondents acknowledged that HPV testing on provider-collected cervical samples offered superior sensitivity and specificity to self-collected vaginal samples, aligning with the findings of a high-quality 2014 meta-analysis²⁵. Respondents offered varying perspectives on how population-based screening programs should adapt to the different risks and benefits introduced by this alternative modality.

Some respondents felt that self-sampling by itself would not effectively reach marginalized communities, and that education/behavioural approaches would address the true barriers that women face with early detection. The literature is similarly divided, such that some authors believe that self-sampling should be an option available to all eligible women (not just non-responders)^{39,40}, while others suggest that self-sampling is appropriate only as a strategy to reach "non-compliant" women^{24,41}, and yet others argue that self-sampling is not the best way to reach under- or never-screened groups^{22,42}. Recent evidence syntheses suggest that most women find self-sampling preferable to clinician-collected sampling^{23,43}. Other systematic reviews found that self-sampling could increase cervical screening coverage in under- or never-screened women²⁴, though some authors observed similar uptake rates in opt-in scenarios⁴⁴.

Clinician respondents stressed the importance of maintaining face-to-face meetings between women and primary care providers, as they suggested a drawback to self-sampling was the loss of an engagement session between a clinician and a patient, which could include discussions on sexually transmitted infections and contraception. A recent study of physician billing claims in Ontario found that increasing the age of initiation for Pap testing significantly reduced the number of chlamydia cases detected in Ontario⁴⁵. Because some of the suggested self-sampling strategies affect marginalized women who are at high risk for sexually transmitted infections, cervical screening strategies that are outside of a clinic may exacerbate other health inequities by taking away opportunistic screening.

There are some limitations associated with this research study. Though there was diversity in stakeholder perspectives including provincial policy-makers, no representatives from provincial government funding sources agreed to be interviewed. Thus, we could not examine some of the decision-making factors from the government side that were raised by other respondents. Also, patient perspectives were not included in this study because of the specific research focus on policy change. However, moving forward, patient advocacy groups will represent an important voice in developing equitable, patient-centered practice. Finally, participants were recruited from across Canada (and Europe), implying different jurisdictions, organization of programs, and funding schemes. Most of the programs within Canada are similar enough to warrant

comparison of barriers and facilitators of program reform, though some of the barriers that women face to accessing screening may be unique to each jurisdiction.

CONCLUSION

According to cervical cancer screening health care providers, policy-makers, and researchers, screening programs must understand the marginalized population that faces barriers to accessing screening. After identifying the hard-to-reach populations, alternative screening strategies, including HPV self-sampling, may be appropriate if the implementation satisfies the criteria of successful population screening programs, including acceptability by the population, cost-effectiveness, sufficient sensitivity and specificity, and an organized call/recall program. Additionally, interview respondents recommended that self-sampling implementation include knowledge translation opportunities for researchers, health professionals, and the public to ensure a tailored approach for the specific population and context. To improve health equity, respondents emphasized that collaboration among cervical cancer screening experts will allow stakeholders to leverage existing scientific evidence and identify research that will inform local gaps in cervical cancer screening knowledge. Researchers and policy-makers should continue to engage with health care providers to address their needs, and in the future, incorporate the patient perspective into strategies that are acceptable to diverse populations.

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CONFLICTS OF INTEREST DISCLOSURE

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“They Should Be Asking Us”: A Qualitative Decisional Needs Assessment for Women Considering Cervical Cancer Screening

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Abstract

In this study, we examine from multiple perspectives, women's shared decision-making needs when considering cervical screening options: Pap testing, in-clinic human papillomavirus (HPV) testing, self-collected HPV testing, or no screening. The Ottawa Decision Support Framework guided the development of the interview schedule. We conducted semi-structured interviews with seven screen-eligible women and five health care professionals (three health care providers and two health system managers). Women did not perceive that cervical screening involves a “decision,” which limited their knowledge of options, risks, and benefits. Women and health professionals emphasized how a trusted primary care provider can support women making a choice among cervical screening modalities. Having all cervical screening options recommended and funded was perceived as an important step to facilitate shared decision making. Supporting women in making preference-based decisions in cervical cancer screening may increase screening among those who do not undergo screening regularly and decrease uptake in women who are over-screened.

Keywords

decision making screening and prevention, cancer, women's health, gender, agency, qualitative research, screening, health care

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High-quality health care is based on person-centered and evidence-based philosophies. In particular, health decisions should reflect individuals' preferences among the best available options that are supported by research and available resources (Haynes, Devereaux, & Guyatt, 2002; Hoffmann, Montori, & Del Mar, 2014; Lang et al., 2018; Umscheid, 2009). Shared decision making provides an opportunity to implement evidence-based practice that accounts for patient preferences, such that when multiple options are available to patients regarding a specific health decision, individuals' personal characteristics and values will influence the best choice for them (Hoffmann et al., 2014; Stacey et al., 2017). However, the use of shared decision making in cancer screening is seldom used and such approaches that have been used are diverse (Elston Lafata, Brown, Pignone, Ratliff, & Shay, 2017; Hoffmann et al., 2014).

Traditionally, cancer screening decisions are made through a population health lens in which clinical practice guidelines have been developed, with individual health care providers

promoting the recommendations therein. Individuals, however, are considered compliers if they decide to accept the offer of screening recommended in the guidelines, but usually are not presented the opportunity to make an informed decision about participation (The Lancet, 2016; Sandman, Ganger, Ekman, & Munthe, 2012). With new technologies developed to screen for cancer, there are more cancer screening decisions leading to more options presented to individuals

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and health care professionals (Lillie et al., 2014). Decisions about screening modality, frequency of screening, and starting and stopping age are inherently preference-sensitive, suggesting that individuals and health care professionals could use shared decision making to identify the optimal option.

The most recent cervical cancer screening guidelines at a national level in Canada (Dickinson et al., 2013) and provincial level in Ontario (Murphy et al., 2011; Murphy et al., 2012) recommend that the long-standing Papanicolaou (Pap) primary screening test be replaced by testing for high-risk strains of human papillomavirus (HPV), of which have been found to be a causative factor in the development of cervical cancer. Compared to the Pap test, HPV testing of samples obtained by a health care professional offers increased sensitivity, a slightly decreased specificity, and different rates of false positive and false negative test results (Koliopoulos et al., 2017). HPV testing can be conducted on self-collected samples with slightly decreased sensitivity and specificity compared to samples collected by a health care professional (Arbyn et al., 2014). These different options are also associated with different recommended intervals between screens (e.g., 3 years for a Pap test, 5–6 years for an HPV test), and different ages to start and stop screening. Recent systematic reviews suggest that most women find self-sampling preferable to clinician-collected sampling (Braz et al., 2017; Nelson et al., 2017; Racey, Withrow, & Gesink, 2013; Verdoodt et al., 2015) because of factors related to ease, ability to conduct the testing without a health care professional or by themselves, privacy, and lack of embarrassment.

These three screening modalities (e.g., Pap testing, HPV testing in-clinic, self-collected HPV testing) and the option not to participate in screening are associated with distinct trade-offs between the benefits (e.g., ability to predict cervical cancer risk) and harms (e.g., false positives and false negatives). As these options differ in convenience, privacy, and invasiveness, individuals should actively participate in discussions with their providers to decide their best option. HPV testing modalities also differ from other cancer screening options because of implications of testing for a sexually transmitted virus; stigma surrounding this type of testing may affect how individuals make trade-offs in these decisions. However, provincial and national guidelines have not addressed individuals' values or preferences among these cervical screening options (Sawaya & Kuppermann, 2015), and have not provided explicit guidance for eliciting preferences. Table 1 presents the existing options for cervical cancer screening in Ontario. In the shared decision-making literature, there are no decision support tools relating to these options (Jimbo et al., 2013; Lillie et al., 2014; Stacey et al., 2017).

Evidence-based guidelines use a utilitarian approach to creating population-based recommendations (National Collaborating Centre for Healthy Public Policy, 2016). Health care professionals (HCPs) who use these guidelines often feel obligated to promote the recommendations, despite individuals' preferences for alternative options (Jepson, Hewison,

Thompson, & Weller, 2005; Petsova, Dale, & Fulford, 2006; Trevena, 2009). The divide between population and individual perspectives may be augmented for cancer screening programs or policies because of the requirements of a mass screening program (i.e., a defined, long preclinical phase, need for accurate and reliable tests, demonstrable effective early treatment; Salmi, Coureau, Bailhache, & Mathoulin-Pélissier, 2016). Shared decision-making processes can reconcile individual and population perspectives for cancer screening by addressing the individual acceptability of tests in the context of HCP values and evidence-informed recommendations (Grad et al., 2017; Lang et al., 2018). Therefore, a shared decision-making framework can guide processes to elicit preferences in cancer screening, aligning with person-centered and evidence-informed practice (Hoffmann et al., 2014; Zhang et al., 2017). To support preference elicitation and shared decision making in cervical cancer screening, we need to know how stakeholders, including women eligible for screening and HCPs, perceive the decision.

Theoretical Framework

The Ottawa Decision Support Framework (ODSF; O'Connor, Jacobsen, & Stacey, 2002; O'Connor et al., 1998) is a decision support framework incorporating expectancy value, social support, cognitive, and psychology theories (Ajzen & Fishbein, 1980; Feather, 1982; Fischhoff, Slovic, & Lichtenstein, 1980; Janis & Mann, 1977; Keeney & Raiffa, 1993; Norbeck, 1988; Orem, 1995; Tversky & Kahneman, 1981). This framework was validated in previous research involving individuals and families making health and social decisions (Feenstra, Lawson, Harrison, Boland, & Stacey, 2015; O'Connor et al., 1999; Poirier et al., 2016). The goal of shared decision making is to promote a well-rounded decision process rather than a particular action (Lillie et al., 2014); therefore, assessment of shared decision-making processes would consider decision-relevant outcomes. Shared decision-making processes are separated into three constructs: decision quality, decision impact, and decision action. Good decision support would assure that individuals feel informed and that they can clearly articulate the characteristics that are most important in making a decision (i.e., decision quality; Fischhoff et al., 1980; Llewellyn-Thomas & Crump, 2013; O'Connor, 1995; O'Connor et al., 2002; O'Connor et al., 1998; Tversky & Kahneman, 1981). In addition, individuals would feel that they are satisfied with the decision process and do not experience decisional conflict (i.e., decision impact; Llewellyn-Thomas & Crump, 2013; O'Connor, 1995; O'Connor et al., 2002; O'Connor et al., 1998). There is no ideal cancer screening action or intended behavior; instead outcomes focus on making decisions consistent with individuals' preferences and values (Lillie et al., 2014; Sepucha et al., 2013; Stacey et al., 2017). At a population level, a 2017 review found that decision support increased under-use and decreased over-use of some

services, for example, prostate cancer screening (Stacey et al., 2017). Patients who are active in decision making are more likely to have better health outcomes and care experiences, at a potentially lower cost to the system (Hibbard & Greene, 2013; Hoffmann et al., 2014; Stacey et al., 2017).

Cervical cancer screening promotion and programming often occurs in a multi-disciplinary primary care team environment, following the broader shift in primary care toward patient-centered medical homes (Bryant-Lukosius et al., 2015; Kiran, Kopp, Moineddin, & Glazier, 2015). Because nurses play a key role in facilitating communication between patients and their interprofessional care team, they are well-positioned to support women who are making cervical cancer screening decisions (Légaré et al., 2011; Registered Nurses' Association of Ontario [RNAO], 2015; Stacey, Légaré, Pouliot, Kryworuchko, & Dunn, 2010). Furthermore, nursing professional standards in Ontario highlight the role that nurses play in advocating for patient-centered care (RNAO, 2015), including shared decision-making approaches.

According to the ODSF (Llewellyn-Thomas & Crump, 2013), *values* refer to individuals' informed attitudes about relative desirability or undesirability of the characteristics of a decision's options. Instead of considering values as the underlying ethical, religious, social, or political principles that guide how individuals judge different situations (Pieterse & Stiggelbout, 2016), values are associated with how individuals assess the specific attributes of each option. *Preferences* are constructed from individuals' informed attitudes toward the detailed characteristics of each option, in which the preference is the most-favored option.

Decision support interventions can address decision quality through five steps: (a) Recognition that a decision needs to be made; (b) Understanding information related to the decision; (c) Clarification of values and identification of preferences according to these values; (d) Consideration of resources, including social influences, that affect the decision; and (e) Formation of an action plan (Lillie et al., 2014; Llewellyn-Thomas & Crump, 2013).

Research Objective

The objective of this study was to examine, from multiple perspectives, women's needs for shared decision making when considering available cervical screening options: Pap testing, in-clinic human papillomavirus (HPV) testing, self-collected HPV testing, or no screening. Understanding women's knowledge needs and the values that shape their cervical screening decisions is important for shared decision making. Because shared decision making involves bilateral flow of information, nurses, physicians, and professionals (HCPs) who develop or administer cervical cancer screening guidelines can offer alternative perspectives on how women make decisions in cervical screening, sources of decisional conflict

for women, and methods for implementing decision support in practice.

Method

Using a qualitative, semi-structured interview approach, we interviewed multiple stakeholders, including women and HCPs to determine what women need to identify their preferences among cervical cancer screening modalities, based on a shared decision-making framework. This study was approved by the Ottawa Health Science Network Research Ethics Board (Protocol #20,160,134-01H). All respondents provided informed consent at the beginning of the interviews.

Participants

Participants were eligible if they were 18 years or older, had knowledge of the Ontario health care system (as a patient or HCP), and could participate in an English-speaking interview. Study participants included two groups of stakeholders. The first group comprised women who would be eligible to consider participation in the Ontario cervical screening program; previous experience with screening was not required. Screening participants were selected purposively based on age categories (e.g., <35, 35–54, 55+) and ethnicity (e.g., European descent, First Nations/Inuit/Metis, immigrant or another minority population). The second group comprised of health care professionals (HCPs) including primary care providers who provide cervical cancer screening to their patient population (i.e., *providers*) and non-clinical professionals who develop and administer cervical cancer screening recommendations (i.e., *managers*). These individuals were purposively selected based on their role within the cervical cancer screening program (e.g., through local policy/management, through provincial guidelines and policy, through direct clinical care). These professional groups play substantially different roles in how cervical cancer screening is delivered but share a common role as "health experts." In this context, HCPs were familiar with cervical cancer etiology, population-based cancer screening and guidelines, and the organization of the Ontario health system.

HCPs were recruited by contacting clinics in the Champlain Local Health Integration Network region in Ontario, Canada, and relevant cancer screening agencies. Clinics were purposively based on geographic area (urban and rural) and targeted communities with high proportions of Indigenous or Francophone clients and/or vulnerable populations, including newcomers to Canada, those affected by mental illness, or low-income residents. The research team worked with the clinics and HCPs to identify screen-eligible women who met the aforementioned age and ethnicity criteria. Researcher contact information was also shared with community groups and patient advocacy groups to recruit screen-eligible women.

Table 1. Existing Cervical Cancer Screening Options in Ontario.

Option	Guideline Recommendation	Funding Status
Cervical cytology/Pap test	Recommended by Cancer Care Ontario in interim guidelines Recommended by the Canadian Task Force on Preventive Health Care	Screening according to Cancer Care Ontario guidelines is covered by public insurance
Provider-administered HPV testing	Recommended by Cancer Care Ontario	2017 provincial budget allocated funding toward HPV testing, but not implemented
Self-collected HPV testing	Recognized as an option for HPV testing, no explicit mention in guidelines	No funding; can be privately purchased online
No screening	Not applicable	Not applicable

Note. HPV = human papillomavirus.

Table 2. Overview of Needs Assessment.

ODSF Steps to Shared Decision Making	Guiding Questions
Recognize the decision to be made	What do you perceive as important decisions for you/women in cervical cancer screening? Have you considered options in cervical cancer screening modalities? How might you/women feel making decisions about cervical screening? What difficulties might you/they have? What will make the decision difficult for you/women? Knowledge Expectations Values clarity Support Resources
Information comprehension	What do you see as the main options? What do you see as the main benefits and risks of these options?
Values clarification and preference elicitation	What qualities about these screening options are most important to you? (Potential screening participants only) For example, convenience, process of test, length of time to get results, costs, invasiveness, access, follow-up procedures, sensitivity/specificity
Identifying necessary social support and material resources	What supports do you or other women need to make this decision? What will help/hurt the decision-making process? What will help overcome some of these issues? What are the barriers and facilitators to offering options in cervical screening? What options are available to support women in making decisions about cervical cancer screening? What might be the 'best' option for a cervical screening decision tool (e.g., Internet, phone app, etc.)?
Forming an action plan	What do you perceive as implementation facilitators and barriers? (HCPs only) What personal, clinical characteristics are important in decision making?

Note. ODSF = Ottawa Decision Support Framework; HCP = Health care professionals.

Recruitment continued until data saturation was reached for all guiding categories (further details in Analysis section).

In-person or phone/video interviews were arranged between July 2016 and November 2016. At the beginning of each interview, the project was explained to each respondent, and respondents' questions were answered before obtaining written informed consent. Contact cards were provided to the respondents if they wished to follow-up with the interviewer after the interview. Interviews lasted between 30 and 60 minutes and were conducted by Brianne Wood (BW), a researcher trained in the ODSF who was familiar with cervical screening guidelines.

Data Collection

Semi-structured interview guides were developed separately for women and HCPs based on the ODSF and its Population Needs Assessment guide (Jacobsen, O'Connor, & Stacey, n.d.; see Table 2 for an overview of the interview guide). The interview guides contained open- and close-ended questions based on these steps, eliciting from screen-eligible women and HCPs what cervical screening decisions women face, what information women may need to make decisions, values that affect their decision, and the support and resources women may need to make a screening decision among

cervical screening modalities. Demographic data on each participant were also collected during the interview. Respondents were asked if they wanted to receive communication about the project, following the interview; those who expressed interest received an email with a high-level summary of the analysis findings. Feedback or comment on the interpretation of the data was invited.

Analysis

The interviewer took field notes during each interview and composed interview summaries afterward, allowing for reflexive analysis (Alley, Jackson, & Shakya, 2015). All interviews were audio-recorded using a tablet with an external microphone. Digital recordings were transcribed using an intelligent verbatim process, which includes light editing to correct sentences and grammar to improve overall readability. Before interviews were conducted, a coding dictionary was created using the ODSF overarching steps (Table 2) as guiding categories (Jacobsen et al., n.d.), and the codebook fields outlined by Guest, Bunce, and Johnson (2006, p. 64). In particular, each code definition contained a code overview, a full definition, a "when to use" section, a "when not to use" section, and an example of the code. Transcripts were analyzed chronologically during data collection (NVivo qualitative data analysis Software; QSR International Pty Ltd. Version 11, 2016), using deductive content analysis (Hsieh & Shannon, 2005; Varpio, Ajjawi, Monrouxe, O'Brien, & Rees, 2017) with respondents as the unit of analysis. During the analysis, additional codes were developed and added to the codebook, and current code definitions were updated to reflect the nuances presented in the interviews (Hermink, Kaiser, & Marconi, 2017). Previous coding was reanalyzed to assess consistency with the new code definition, and a new code was added if necessary. The iterative approach was important to help determine when saturation was reached. A categorization matrix (Elo et al., 2014) was created to depict which categories were communicated by stakeholder groups, and to indicate when code definitions were added or changed to document saturation. Another member of the study team (Virginia Russell [VR])—an expert in qualitative research—reviewed the codebook to assure clarity of the definitions. VR independently analyzed each transcript to ensure complete and appropriate coding of important themes. Coded segments were assessed and refined further to concisely represent the data. Discrepancies in how segments were interpreted were resolved through dialogue between BW and VR.

In this study, we sought to triangulate different stakeholder perspectives (Fusch & Ness, 2015; Légaré et al., 2007) to gain an in-depth understanding of women's needs to make an informed decision among cervical screening modalities. In particular, we elicited multiple perspectives on women's decisional needs for cervical cancer screening modalities, to understand how women's decisional needs are influenced by individual, provider, and health system

factors. We looked for convergence in how ODSF steps were interpreted by stakeholders to identify opportunities to support women's decisional needs. Any diverging interpretations were explored in further interviews. The sample size was considered adequate once a diverse sample of stakeholders had been recruited (Guest et al., 2006), and study authors considered that no further interviews would generate insight to help address the research objective (O'Reilly & Parker, 2013; Varpio et al., 2017). A category was considered saturated when the code meaning was determined stable for each of the stakeholder groups (Varpio et al., 2017).

Results

Participant Characteristics

In total, seven women eligible for screening and five HCPs were interviewed—three providers and two managers. Of seven screen-eligible respondents, women's ages ranged between 24 and 63 years. Two Indigenous respondents participated, as well as one Francophone respondent, and two women who identify as immigrant or first-generation Canadian. Most screen-eligible respondents had previously had a Pap test. Among HCPs, two nurses and a general practitioner represented clinical providers, and a director of a health center and a provincial cancer agency director were interviewed as managers. Among the HCPs, one respondent identified as male.

Three screen-eligible respondents who were recruited from the same organization (an Indigenous health organization) were interviewed in a group setting, when they identified at their interview session that they preferred a group interview instead of separate one-on-one interviews. As the interview was the first time that the respondents had met in person with the interviewer, they communicated that they felt more comfortable discussing some of these sensitive health topics together. One respondent (Woman 1) recruited the two other respondents (Woman 2, Woman 3) and organized the interview space, but did not dominate the interview conversation. There was a large focus on HPV and cervical cancer information in this session, with the discussion exploring opportunities for them to choose among cervical cancer screening options.

The main themes are organized below by the ODSF steps to shared decision making.

1. Recognition that a decision needs to be made

Most screen-eligible women had not considered participation (or non-participation) in cervical screening as a *decision*. Instead, a Pap test was perceived as an obligation for women of a certain age, thus women would not actively choose to avoid a Pap test. Rather, women might defer cervical screening, or they might want to attend screening but face

barriers accessing the health care system. When asked about decisions that they had faced with cervical screening, most respondents expressed confusion; they had not considered "no screening" a viable option:

It was just sort of best practice or recommended by the doctor to have it done, so I really didn't give it much thought and just had the procedure. (Woman 6)

I thought it was maybe about the right time to get a Pap test done. So, that was just my reason—like my age. (Woman 7)

Both groups of stakeholder respondents identified that women may prefer a female health care provider, although the choice among options still was not perceived as a decision.

In 2012, Ontario cervical screening guideline extended intervals between screens from annual Pap tests to 1 every 3 years, and some respondents—including female HCPs—preferred more frequent screens than recommended. Despite this preference, respondents felt they could not act on their preferred options because they were limited by the provincial guidelines and funding:

OHP [Ontario Health Insurance Plan] doesn't [sic] cover Pap smears once every 3 years now. For women, not every 6 months ... that's the law now. (Woman 3)

When it's due, I want it done. And I prefer it actually be done more often than [chuckles] is recommended now, but I'm okay with that. (HCP 3)

Manager HCPs felt strongly that women's options in cervical screening should be governed by guidelines developed by cancer agencies, which was echoed by provider HCPs who currently decide the types of tests to offer women for cervical screening:

So it is confusing to make the choice once there is more of a choice because there's not too much of a choice yet, right? ... Central and national guidelines [are important] because they give individuals as well as primary care providers some framework around the decisions that they make. (HCP 5)

Despite their perceived lack of choice, screen-eligible women wanted to choose between options. Although they wanted to know more about these options, they also wanted to engage with their primary care provider to make a decision. Interestingly, most of the potential screening participants focused on the self-collection option of the test, paying less attention to new diagnostic processes, suggesting that having options that differ in sample collection process as well as diagnostic performance might make decision making more complex:

I like the idea of having multiple options, but I think if you say that, for example, if I decided I have my Pap test but I still decided to do the HPV test and I realized that it was positive, then you said well sometimes you can have a virus but it's not a

big deal because it's going to go away on its own. ... So I guess it has two sides, like it's good because that would be something convenient. But at the same time, we are not doctors. We don't know those things so maybe it would create more people going to the clinic that we really need. (Woman 5)

2. Understanding information related to the decision

Most respondents did not know that several options were available for cervical screening in Ontario, or that they faced a decision between in-clinic Pap testing, in-clinic HPV testing, self-collected HPV testing, no screening. A portion of each interview involved communicating what is known about HPV, cervical cancer, and current initiatives for prevention, detection, and treatment of cervical cancer to screen-eligible respondents. Three of the screen-eligible respondents had not heard of HPV previously, although they had previously participated in cervical screening. Except for one woman who had learned about self-collection through work, six screen-eligible respondents had not realized a self-collection option existed:

I think the questions I would ask and stuff, like, oh, how does it work and what's the process it's going to be like? Does it hurt? Especially like for myself when you've never got screened before. I'll probably feel like very anxious and like scared of doing it myself for the first time. (Woman 4)

HCPs were more aware of the connection between HPV and cervical cancer, although three HCPs wanted further information about new cervical screening options, suggesting that more work in knowledge translation with professionals in the health field is needed. A provider argued that these options were hypothetical at this point because HPV testing is not yet funded, so communication opportunities might be premature. As a result, HCPs were concerned about communicating options that were not universally available:

Because so far there isn't that option [for self-collected HPV testing], so there isn't any discussion like that. It's just me explaining you could get cervical cancer. It can be detected before it becomes cancer. This is how it's done and it's recommended to start at age 21. We do it every three years. So I explain that and then they say okay. And they go for it. (HCP 2)

I would imagine there would be some of our clients, patients, who would ask about it [the HPV test] because they would have heard about it, known about it ... you know, you really would have to ask them [the clinicians who administer the Pap tests] if they raise it [the options for cervical cancer screening] with women who they know could not pay for the test. (HCP 1)

Although options other than Pap tests are accessible through private payments, clinicians may not engage in knowledge exchange with their clients because the options are not addressed in the practice guidelines, nor are they funded by the public system. HCPs conveyed a sense

of paternalism—that women's decision making should be contained within the evidence-based guidelines—which coincided with a desire for women to have the autonomy to make informed decisions. Some HCP respondents recognized this contradiction throughout the interview and struggled with balancing autonomy of women versus the population-based goals of a screening program:

'I'm a very big believer in clients making choices . . . And if it's a bad choice, it's still their choice and they have all the right in the world to make that choice.' (HCP 4)

I think most screening programs, not just in Canada but definitely in the U.K. [United Kingdom] and the Nordic countries have moved towards a more balanced message. I do struggle a little bit with that because okay, so I want women to have a balanced message about the benefits and harms of cervical cancer screening, right? (HCP 5)

Both screen-eligible women and HCPs identified that all stakeholders (i.e., women, providers, managers) need more information about the cervical screening modalities, the processes, the risks, and benefits of these options, and how these options appeal to individual women. Women wanted more information so that they could weigh the cervical screening options and consider what aspects of screening are most important to them:

I guess like if you're going to a clinic and an expert is helping you screen it, then I think the information you would want is, like, oh what are you going to do and, like, how does it work in general and the process and like how long or how often does it come back to get screened? Whereas for like, if you're doing a self-test, for me I would want to know, like, more information, I would probably have to like want to reassure myself that I'm doing things correctly. (Woman 4)

3. Clarification of values and identification of preferences according to these values

Because in Canada we balance, an individual's choice with what's good for the population and what we can afford as a society, right? (HCP 5)

Attributes that respondents thought would affect women's screening decision were organized into three main categories: characteristics of the sample collection process, characteristics of result follow-up and communication, and importance of cervical cancer and early detection. As mentioned previously, women and HCPs tended to focus on the decision between in-clinic cervical screening versus self-collected (e.g., "at-home") cervical screening versus no screening, expressing less concern about a change in diagnostic procedure (i.e., cytology vs. HPV testing).

Screen-eligible women were primarily concerned with sample collection and the convenience among the different modalities. Women's comfort with the sample collection pro-

cess often related to prior traumatic or painful screening experiences, and their confidence to collect a sample themselves:

But they're kind of similar, I think the questions I would ask and stuff, like, oh, how does it work and what's the process it's going to be like? Does it hurt? Especially like for myself when you've never got screened before. I'll probably feel like very anxious and like scared of doing it myself for the first time. (Woman 4)

HCPs expressed more concern about the socioeconomic impact of offering cervical screening modalities that require women to pay out-of-pocket and the implications of varying laboratory quality of the screening tests:

So you have to understand that the clientele that we have here are low income is over 50 per cent. We have some low income and our mandate is to serve low income seniors and new immigrants. So it is a huge consideration for them not going forward with that. And a lot of females with low income don't mind coming in for Paps and testing. I'm generalizing, but I don't want to generalize in any way. So it's not about the testing itself and feeling uncomfortable with doing it. It's just about the cost. (HCP 4)

Women and HCPs expressed concern about communication and follow-up of results. Both groups of stakeholders were concerned about privacy when communicating results, noting that current Pap test results are shared via letters from a central provincial agency. The lack of personalization of these letters and potential for ambiguity was worrisome for providers and women, although not mentioned by managers. In the following excerpt, a provider communicates frustration with the current result communication system in Ontario, suggesting that the letters to participants do not clearly articulate what screening results mean, and might cause distress:

Like they [women eligible for screening] know what they would want if they think it through is for their doctor to tell them and to not have to wait for the doctor to say it is not normal and then tell them what that means and not get this message that it's not normal. Then have to wait for whatever length of time to find out what it means and in the meantime being scared to death of the implications. (HCP 2)

The change in approach to detection of cancer and precursor lesions—from looking for cellular abnormalities with Pap testing to detecting the presence of a high-risk sexually transmitted virus with HPV testing—was considered important because of stigma associated with sexually transmitted infections. Women and HCPs suggested that methods to follow-up positive results would require careful communication with women. Unsurprisingly, women were concerned with the privacy and confidentiality of how results, including HPV positivity, would be communicated. The following interaction occurred in the first interview with three women, who had recently learned about the connection between HPV

and cervical cancer, and how cancer screening would involve testing for a sexually transmitted infection:

Confidence is the main thing. Because there's a stigma related [to a] sexually transmitted . . . (Woman 3)

Oh you whore or whatever. (Woman 1)

A moral responsibility to be healthy and participate in healthful behaviors influenced how women contemplated cervical cancer screening options, which was reiterated by HCP and screen-eligible women respondents. The inherent "good" value of cancer screening was emphasized as many respondents described cancer screening participation as healthful. Most respondents ignored the "no screening" option because it was implied that when someone was appropriately informed, they would choose to participate in cervical screening. This conviction that any cervical screening would necessarily improve one's health was also related to certain risk expectations of cervical cancer; women were more enthusiastic about cancer screening when they perceived themselves at a higher risk of developing cancer:

But if it [other option's diagnostic performance] was different or the sensitivity or the results were different between the options, obviously it would be my first deciding factor. (Woman 7)

A manager HCP agreed that public health enthusiasm for cancer screening could be responsible for an automatic acceptance of screening:

I imagine that's how most women make their decisions and I think a lot of it is probably not evidence based and not balanced and not good quality information. And screening programs used to be part of that because we were like you should be screened no matter what. (HCP 5)

While women and HCPs seemed to identify similar attributes that could influence cervical screening decisions among modalities, they judge them differently. Women focused on decision making at an individual level and were concerned about personal risks and benefits, while HCPs often spoke of patient populations and collective risks and benefits, reinforcing the population perspective of cancer screening programs.

4. Consideration of resources, including social influences, that affect the decision

Respondents indicated that primary care providers were most likely to influence women in their decisions among cervical screening modalities. Notably, it seems that screen-eligible respondents who had a consistent primary care provider were more likely to turn to a clinician for input into their decisions. Screen-eligible respondents explained that they rarely involved friends or family in cervical screening

decisions, although one HCP noted that women from certain religious groups might prefer to involve their husbands because of the invasive nature of the sample collection:

Anybody's welcome to stay as long as the client wants them to. So we found that East Indian women very uncomfortable with it and need almost that family support around for them. (HCP 4)

Women and HCPs recognized the counseling that primary care providers can offer women in preventive health care. Screen-eligible respondents emphasized the importance of access to a provider whom they trust, explaining that would influence how they or others choose to participate in screening:

I was very lucky because . . . it took no time to have a family doctor first of all. Because one thing I find is that I wouldn't want just any doctor to do the Pap test. (Woman 5)

Screen-eligible women wanted to feel comfortable with the person who is administering the invasive cervical cancer screening; some women mentioned that a lack of familiarity with a provider made self-sampling more appealing. HCPs agreed that cervical cancer screening with the Pap test requires a lot of trust because of the pelvic exam but added that providers help their patients feel supported if abnormalities are found.

More broadly, screen-eligible women "trusted" their primary care provider to offer them any screening tests or preventive health care for which they are eligible. Provider HCPs also noted that their patients will frequently accept cervical cancer screening when they are introduced to it opportunistically. A manager HCP identified that women who are more likely to participate in preventive health care, such as cervical screening, will already have a trusting relationship with a primary care provider; conversely, women who are not attached to a primary care provider are less likely to participate in screening:

[I]t's almost like a bit of a catch-22 because if you're a woman and you're in a position to have that somewhat personal conversation with someone, you're also in a position to be more likely to be screened. (HCP 5)

All respondents communicated that women need some external support to make a decision among screening modalities, with suggestions ranging from educational campaigns to public funding of all options. Some respondents suggested that tailored approaches are needed to reach women from different populations. For example, two Indigenous respondents indicated that they would prefer learning about cervical screening through an online source, and they feel most comfortable accessing health care that is tailored to Indigenous women. Another woman indicated that "clear instructions in both languages [French and English]" were important for

communicating the cervical screening options. A manager HCP seconded this bilingual preference and emphasized the importance of structuring support for immigrant and refugee women, as well as women who have experienced sexual trauma.

Most women considering screening felt confident that they could choose their preferred screening modality if they received more information, in the absence of primary care provider input. While discussion groups (e.g., at a public health unit) were considered a satisfactory option by a few women, most women preferred to learn about the options independently through materials they could find online or in a public space, or through a one-on-one conversation with a health care provider. Several of the women mentioned wanting "several sources" of information before deciding on a screening modality:

I would say the website, kind of getting different information from different sources, only because the majority of us have access to like internet. . . . I also sometimes look at different experiences . . . I used to go for different sources because you just want to check the facts and one website might cover something that you never thought about before. (Woman 4)

5. Formation of an action plan

They [health care professionals] should be asking us. (Woman 2)

Overall, respondents acknowledged women do not perceive that undergoing cervical cancer screening involves a decision, which women attributed to a lack of awareness and HCPs attributed to the context of a population-based screening program. As new modalities are implemented, screen-eligible women and HCPs identified provider-patient encounters as an optimal environment to have a conversation about screening options. Subsequently, respondents identified that women and primary care providers need support from the cancer screening program to ensure that they have sufficient information and resources to engage in these conversations. HCPs looked to clinical practice guidelines and funding organizations to catalyze this change:

What's available, if we don't have the funding then they [policy-makers] can't recommend it. (HCP 2)

Most respondents emphasized that support from cancer agencies and funding bodies was important to make cervical cancer screening options accessible. Having all options covered under the public insurance plan was important to implicitly establish that the screening options and entire program were trustworthy. For screen-eligible women, public funding would reinforce the notion that governments are not solely concerned about saving money and that all options

were "good." For HCPs, public funding of all options symbolized confidence in the scientific evidence behind these options. Having multiple options available within the organized cancer screening program would allow screen-eligible women to make preference-based decisions in cervical cancer screening, while meeting the objectives of a population-based program:

[I]f it's [the screening modality] well explained, if the price is good . . . If it's something that's accessible anywhere that anybody can buy anywhere, why not? (Woman 5)

So I think by doing all of these things through an organized screening program and having that structure, you have the opportunity to ensure that women are not over screened or at least help to make sure women aren't over screened and to try to target unscreened women (HCP 5)

Discussion

In this qualitative interview study, we observed that screen-eligible women and HCPs do not perceive participation in cervical cancer screening as a health decision, but rather, it is an opportunity or an obligation. Most respondents were reluctant to identify "no screening" as an acceptable option. Recognizing that a decision among cervical screening modalities exists for women is crucial for high-quality shared decision making, according to the Ottawa Decision Support Framework (O'Connor et al., 2002; O'Connor et al., 1998). Other decisional needs identified in this study include women's knowledge of cervical screening options and associated risks and benefits, and resources to support them in decision making, most notably a trusting relationship with a primary care provider.

Cancer screening decisions are thought to be inherently preference-sensitive (Lillie et al., 2014; Stacey et al., 2017), although cervical cancer screening has been under-represented in shared decision-making literature (Jimbo et al., 2013; Lillie et al., 2014; Stacey et al., 2017). In Canada, Pap testing had long been the only screening option available for early detection of cervical abnormalities and cervical cancer. Consistent with the health messaging by media campaigns, Pap testing might have seemed like the *correct* option. Even now, as the Pap test is the only publicly funded option among available modalities aside from not screening, respondents asserted that informed women would choose to participate in screening. This public and professional enthusiasm for cancer screening has been discussed previously (Schwartz, Woloshin, Fowler, & Welch, 2004; Shieh et al., 2016) and likely contributes to the perceived imperative to participate in cervical cancer screening.

With regard to eliciting preferences among Pap testing, in-clinic HPV testing, self-collected HPV testing versus no screening, most women and some HCPs were unaware of

any type of HPV testing, for example, modalities that are not funded by the Ontario health system. However, some organizations, like the United States Preventive Services Task Force (U.S. Preventive Services Task Force, 2017), already recommend that women make a choice among modalities, although implementation and decision support for individuals is unclear. This study's findings are timely because it seems that awareness and knowledge of the cervical screening options stand out as women's primary decisional needs in cervical screening. Although values played an important role in how women make the decision (e.g., how women make trade-offs between the costs of different options, the convenience of the procedures), screen-eligible women felt confident that they could make the decision if they had more information. However, it is possible that because women have not faced this type of decision-making in cervical cancer screening, that they may miss some of the nuances around the different trade-offs (Pieterse & Stiggelbout, 2016). Value clarification methods—strategies to help individuals assess the desirability of attributes of different options—will be important help women identify which option they prefer among cervical screening modalities (Lewellyn-Thomas & Crump, 2013; Pieterse & Stiggelbout, 2016).

HCPs worried that multiple options may cause worry or anxiety in women deciding among cervical screening modalities. The HCP concern of decisional conflict may reflect worry that women may not participate with *medicine's* preferred screening modality (i.e., making the "wrong" choice by selecting a less sensitive option). The belief that there is an ultimate "good" decision suggests that HCPs may have values that differ from their patients. Respondents also believed that if women recognized the decision and were informed of the options, they would choose the option that corresponds with population health objectives. However, it may be challenging for women to act on their preferred option for cervical screening if it deviates from their clinician's preferred option (Adams, Elwyn, Légaré, & Froesch, 2012). Nurses will play a key role in fostering an environment that allows women to learn about their cervical cancer screening options in an unbiased and comprehensive way. By fulfilling the role of decision coaches (Stacey et al., 2012; Stacey et al., 2008), nurses prepare individuals to have conversations with their practitioners about screening decisions (Grad et al., 2017; Lang et al., 2018; RNAO, 2015; Stacey et al., 2008). However, providers also face organizational barriers such as performance quality metrics that might impede shared decision-making (Elston Lafata et al., 2017; Walter & Lewis, 2007).

Some authors have discussed perceived challenges of autonomous decision-making within the context of a population-based cancer screening program (Elston Lafata et al., 2017; Jepson et al., 2005; Salmi et al., 2016; van Dam, Kuipers, Steyerberg, van Leerdam, & de Bontfort, 2013). In the cervical screening situation, many HCPs were concerned

about offering modalities to patients that were considered inferior to the modalities recommended by clinical practice guidelines. Findings from this study demonstrated an interesting paradox that HCPs and individuals trusted cancer agencies and networks to know the "best" options for cervical screening for individuals, yet, some respondents also expected system-level institutions to prioritize financial or other system prerogatives above an individual's wellbeing. As test acceptability is a crucial component of a successful screening program (Dobrow, Hagens, Chafe, Sullivan, & Rabeneck, 2018; Salmi et al., 2016; Wilson & Junger, 1968), women want confirmation from HCP stakeholders that the options are valid.

One respondent—a HCP—vocalized concern for potential harms with over-diagnosis and overtreatment when women decide to participate in cancer screening, possibly related to their contribution to guideline development. As knowledge of the decision and its options is a key decisional need identified in the interviews, a balanced presentation of risks and benefits for each option is necessary. Two previous systematic review demonstrated that individuals and health professionals tend to overestimate the benefits and underestimate the harms associated with screening (Hoffmann & Del Mar, 2014; Hoffmann et al., 2017), suggesting that tools to elicit preferences or support shared decision-making need to carefully communicate that cervical screening can directly benefit a small number of women through early detection, and potentially introduce harmful consequences through over-diagnosis and overtreatment (Salmi et al., 2016; Sawaya & Kuppermann, 2015; Shieh et al., 2016). Not only are women's *informed* preferences among cervical screening modalities (and other decisions) integral to patient-centered care, but they may lead to more appropriate use of health care resources. The 2017 Cochrane review of decision aids (Stacey et al., 2017) found that some options may be under-used and others over-used, which was usually related to inadequate information comprehension. Hence, supporting women in making preference-based decisions in cervical cancer screening may increase screening among those who do not undergo screening regularly, and decrease uptake in women who are over-screened, which is consistent with utilitarian public health campaigns. This research highlights an opportunity for nurses and other health professionals to empower women to make autonomous cervical cancer screening decisions.

This study is the first to consider women's decisional needs among multiple cervical screening modalities—Pap testing, in-clinic HPV testing, self-collected HPV testing, and no screening—from the perspectives of women who are eligible for screening as well as health care professionals. However, it has some limitations: some respondents perceived this decision among screening modalities as a hypothetical; sometimes, the way individuals perceive and act in a hypothetical decision may vary from that in real world circumstances (Feldman-Hall et al., 2012; Kuhberger,

Schulie-Mecklenbeck, & Perner, 2002). However, because the decision to screen for cervical cancer is ongoing and the modalities are available, most respondents perceived the decision as real. We determined that twelve respondents provided sufficient data to identify decisional needs using the ODSF, and we will integrate this analysis with the findings of a systematic review to further validate the main categories. Although many studies tend to have larger number of interviews, many do not explain why or how they reached saturation at that sample size (Guest et al., 2006; Mason, 2010; Varpio et al., 2017). Inquiring about women's decisional needs from women and HCPs, using two independent coders to analyze the ODSF categories, and a clear description of the data collection and analysis methods (Sahu Eto & Kyngäs, 2008; O'Reilly & Parker, 2013) helped to ensure rigor in this project. These results may not be transferable to contexts outside of Ontario, although these findings might apply to other publicly funded health care systems where quality metrics and remuneration are determined by adherence to clinical practice guidelines. In addition, similar decisional needs were identified across a sample of women with diverse socio-demographic backgrounds which suggests that most women may benefit from some form of decision support.

Conclusion

Women and health care professionals did not perceive cervical cancer screening participation as a decision for women to make. When considering available modalities—Pap testing, in-clinic HPV testing, self-collected HPV testing, and no screening—women and health professionals wanted to know more information about these options. Women wanted to make a choice among available options, although health professionals were hesitant to engage in shared decision-making because some of the options are not funded by the public-payer system, and some options might not be consistent with population-based screening goals. Women and health professionals emphasized the importance of a trusting relationship between women considering screening and their primary care providers for shared decision-making. Subsequently, adequate access to primary health care may play a significant role in women's decision-making process. A tool that addresses some of these decisional needs might support women and their health care providers in cervical screening encounters, which is important for person-centered, evidence-based care. Supporting women in making preference-based decisions in cervical cancer screening might increase screening among those who do not undergo screening regularly, and decrease uptake in women who are over-screened, which is consistent with utilitarian public health campaigns. As advocates for person-centered care on interprofessional teams and health promotion settings, nurses are well-positioned to facilitate preference-based decision-making in cervical cancer screening.

Author Contributions

B.W., J.L., and I.D.G. conceptualized the study. B.W. conducted the interviews, analyzed the data, and wrote the first draft of the manuscript. V.L.R. helped to analyze the data and contributed to the first draft of the manuscript. All authors contributed to the interpretation of the data and provided editorial feedback on drafts of the manuscript. All authors read and approved the final manuscript.

Declaration of Conflicting Interests

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Supplementary Materials

For further information on the data and interpretation of this study, please contact the corresponding author of this study.

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Appendix C. Preference-Elicitation Risk of Bias Example and Excluded Articles with Reasons

Example of Risk of Bias for Preference-Eliciting Studies

Domain	Item	Options	Description
Was the instrument appropriately administered?	Was the interviewer trained properly?	Yes; No; Probably yes; Probably no; Not reported; Not applicable	Any mention of interviewer training/education; not applicable if self-administered method (e.g., mailed out decision tool)
	Was there a structured protocol for administering the instrument? (consider internal validity - well-defined study question that includes key PICOTS components)	Yes; No; Probably yes; Probably no; Not reported	This one is about the instruction/users' manual for the administration of measurement instrument. For example, standard gamble as the instrument, we would like to see if the researchers or people who elicit preferences, have been following a standard process to administer the instrument.
	Was a structured pretesting procedure carried out?	Yes; No; Probably yes; Probably no; Not reported; Not applicable	Not applicable if the report is a pilot study, or part of the pre-testing
	Did investigators recruit optimal interviewees?	Yes; No; Probably yes; Probably no; Not reported; Not applicable	Would the interviewees be accepted by the participants (e.g., consider whether the interviewees were male, for a female sensitive topic; community individuals; experts in the field, etc)
	Did authors test for participant understanding?	Yes; No; Probably yes; Probably no; Not reported; Not applicable	Was there any pilot testing? Is there a way to tell whether participants understood the material, in the appropriate context? Not applicable if the report was to test participant understanding
Was the choice of instrument optimal?	Did investigators use a standardized instrument with evidence of validity?	Yes; No; Probably yes; Probably no; Not reported;	Authors may provide information regarding the measurement properties of the instrument they have chosen. Alternatively, they may have chosen an instrument with which assessors are familiar and with widely accepted reliability and validity. A tentative list of generic instruments with accepted

			validity and reliability include: standard gamble, time trade off, visual analogue scale (or feeling thermometers), discrete choice, treatment trade-off, and willingness to pay. Use of these instruments does not, however, guarantee that they have been administered appropriately. If the authors have neither used an instrument with widely accepted satisfactory measurement properties, nor have provided information regarding satisfactory reliability and validity, the risk of bias is likely to be substantial.
	Was the questionnaire used adequately validated for their health condition?	Yes; No; Probably yes; Probably no; Not reported;	Not reported if previously used instrument and development study cited. Look for psychometric properties. "Yes" if study is measuring psychometric properties; "No" if ad hoc questionnaire and no psychometric properties mentioned
	If appropriate, did the authors present probabilities?	Yes; No; Probably yes; Probably no; Not reported; Not applicable	Probabilities of selecting different preferences or attributes. Not applicable if preference-eliciting doesn't lend itself to probabilities
	Were all relevant outcomes included?	Yes; No; Probably yes; Probably no; Not reported;	Consider what a relevant outcome is? Were outcomes defined a priori? Were preferences stated or generated from weighting of values/attributes? Decision Quality, Decision Impact, Decision Action are ODSF relevant outcomes
Did the health state presentations have high verisimilitude?	Did the authors address the literature bearing on the patients' views and experiences of the health conditions?	Yes; No; Probably yes; Probably no; Not reported;	Was the preference-eliciting method evidence-informed? Were references cited about patients' views and experiences, and were these considered in the approach to preference-eliciting? Any theoretical foundations (specific to the condition and patient population)?
	Did investigators justify their choice of health state: patients' own vs hypothetical scenarios?	Yes; No; Probably yes; Probably no; Not reported;	When eliciting values and preferences from patients, were participants clear whether the decision was real or

			hypothetical? If hypothetical, did the paper explain why? If hypothesizing different states of health (e.g. "your risk of disease is this level, how does that affect your values"), does the author justify?
Do results suggest patient understanding was adequate?	Were the health states rated consistently?	Yes;No;Probably yes; Probably no;Not reported; Not applicable	The measurement of preference for health state could be divided into two categories per the nature of the health state assessed: measurement of respondents' own state, which they are experiencing or have experienced, or measurement of described (usually hypothetical) states, which they may or may not experience in the future.
	Did the authors address patient characteristics that may have a strong association with values and preferences?	Yes;No;Probably yes; Probably no;Not reported; Not applicable	Did authors consider/account for patient variables (e.g. age, education, income, previous experiences, etc)? If qualitative, not applicable?

Scoring:

Each study starts with 0 points.

Total sum= +2 for "Yes"; +1 for "Probably yes"; -1 for "Probably no"; -2 for "No"

Final score = Total sum divided by number of items with a response.

Risk of Bias categories:

"Low" > 66%	"Medium" = 33%-66%	"High" = 0%-33%	"Critical" < 0

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Appendix D: External Review of IPDAS Qualifying Criteria for “Cervical Cancer Screening – It’s Your Choice” and Value and Preferences Survey

IPDASi qualifying criteria	
Q1 Describes health condition	Yes – step 1
Q2 Explicitly states the decision	Yes – step 2
Q3 Describes options	Yes – page 1
Q4 Describes positive features of each option	Yes – step 2
Q5 Describes negative features of each option	Yes – step 2
Q6 Describes what it is like to experience consequences of options	Yes – step 2
IPDASi Certification criteria	
C1 Shows the negative and positive features of options in equal detail	Yes – step 2 and step 3
C2 Provides funding source for development	Didn’t see this
C3 Provides citations to evidence used	Yes – “Additional resources”
C4 Provides publication date	Yes – “about the project”
C5 Provides an update policy	Yes – “about the project”
C6 Provides level of uncertainty around outcome probabilities	Yes – step 3
Additional criteria for screening decisions	
C7 Describes what the test is designed to measure	Yes – step 2
C8 Describes next steps if test detects condition	Yes – step 2
C9 Describes next steps if condition not detected	Almost – step 2, the possibility of having a normal test is discussed, but not the steps that would follow.
C10 Describes consequences of detecting condition that would never have occurred if screening not done	You define and describe false positives, but I can’t seem to find a section where the consequences of this are describes.

Cervical screening values and preferences survey (dated December 5, 2017)

Now that you've read about cervical cancer screening options, the following questions will help you see what is most important to you for cervical cancer screening. Finally, we want to hear what you liked and didn't like about this tool, so that we can help other women like you.

These questions will help you consider the risks and the benefits of different cervical cancer screening options that matter most to you. Please show how important these are to you by choosing a number from 1 (not very important to me) to 5 (very important to me).

There are no right or wrong answers.

1. How important is it to you to know if you are at a higher risk for cervical cancer?
Participating in Pap testing, provider-collected HPV testing, or self-collected HPV testing will let you know if you are at a higher risk of cervical cancer.

1*	2*	3*	4*	5*

2. How important is it to you to know if you have a high-risk HPV infection?
Provider-collected HPV testing or self-collected HPV testing will let you know if you have a high-risk HPV infection. You will not know if you choose Pap testing or no screening.

1*	2*	3*	4*	5*

3. How important is it to you to avoid paying out-of-pocket costs for your health care?
In Ontario, it will cost money to participate in provider-collected HPV testing or self-collected HPV testing. In the future, these options might be covered by health insurance.

1*	2*	3*	4*	5*

4. How important is it to you to avoid having a pelvic exam with a health care provider?
There is no pelvic exam involved with self-collected HPV testing and not participating in screening.

1*	2*	3*	4*	5*

5. How important is it for you that your screening test has a LOW number of false positives?
A false positive occurs when your screening test wrongly says your result is "positive" or "abnormal". Provider-collected HPV testing and self-collected HPV testing results are more likely to incorrectly say that you have a "positive" result.

1*	2*	3*	4*	5*

6. How important is it for you that your screening test has a LOW number of false negatives (i.e. that if your screening test says your result is "negative" or "normal", that you truly are NOT at a higher risk for cervical cancer)?
Provider-collected HPV testing and self-collected HPV testing results are more likely to incorrectly say that you have a "positive" result.

1*	2*	3*	4*	5*

7. List any OTHER reason you may choose to participate in cervical cancer screening with a Pap test, an HPV test in-clinic, a self-collected HPV test, or to opt out of screening all together:

8. For the reason(s) you listed above, if any, how important is this to you?

1*	2*	3*	4*	5*

Check Your Knowledge About the Options

Before you tell us your preference, you can check your understanding of the cervical screening options. Don't worry if you get one wrong – this will help us make the information more clear.

1. Cervical cancer is caused by an HPV infection.

- ☐ True
☐ False
☐ I am unsure

--> HPV causes almost 100% of cervical cancers. However, only a small number of people with HPV will develop any type of health problem from it.

2. Cervical cancer can be prevented through HPV vaccination and cervical cancer screening.

- ☐ True
☐ False
☐ I am unsure

→ HPV vaccination will reduce your risk of cervical cancer by protecting you against most high-risk HPV types. Cancer screening will allow a health care provider to offer early treatment if signs of cancer are found. Because of this, cervical cancer is virtually preventable.

3. The Pap test will tell you if you have an HPV infection.

- ☐ True
☐ False
☐ I am unsure

→ The Pap test looks for changes in the cells of the cervix. The HPV test looks for HPV infections.

4. A self-collected HPV sample is worse than an in-clinic HPV test at finding the pre-cancer and cancer signs. (This means that there is a higher chance of a missed "positive").

- ☐ True
☐ False
☐ I am unsure

→ Research has shown that a self-sample HPV test is not quite as "sensitive" as an HPV test done on a doctor-collected sample. This means that there are a fewer number of "true positives" with a self-sampled HPV test, and a larger number of "false negatives"

5. Cervical cancer screening tests should be done every year.

- ☐ True
☐ False
☐ I am unsure

→ According to current Ontario guidelines, Pap tests should be done every 3 years. HPV tests only need to be completed every 5 years.

6. The Pap test has the highest rate of true positives. This means that the Pap test is more likely to report positive when a woman has cervical abnormalities compared to the other screening options.

- ☐ True
- ☐ False
- ☐ I am unsure

→ HPV testing by a doctor is more sensitive at detecting cervical abnormalities than Pap testing. This means that HPV testing by a provider will correctly report positive, most frequently, compared to Pap testing, HPV testing with self-samples, or not screening. Of 20 women who actually have cervical abnormalities, HPV testing (in-clinic) will catch 18 of those women.

7. At any point in time, 20 of every 1,000 women have cervical abnormalities.

- ☐ True
- ☐ False
- ☐ I am unsure

→ A recent summary of available research showed that approximately 20 women for every 1,000 have cervical abnormalities. Most cervical abnormalities will clear up by themselves in 6 to 18 months. Having a cervical abnormality does not mean cancer.

Step 5 Answer Key:

1. True 2. True 3. False 4. True 5. False 6. False 7. True

If you were given the choice to participate in cervical cancer screening right now, what option would you prefer?

- ☐ Pap testing
- ☐ In-clinic HPV testing
- ☐ Self-collected HPV testing
- ☐ No screening
- ☐ I am unsure
- ☐ I do not have a preference

Do you have any comments about why you chose [insert answer from above]?

Now that you've used this online tool [redirects to Acceptability survey]

Appendix E: Learning outcomes for my PhD

Domain	PhD Learning Objectives	Achievements during PhD
Depth and breadth of knowledge	1. Describe principles and advanced methods for causal inference in epidemiology. 2. Identify and describe advanced methods in epidemiologic research in the student's research area. 3. Explain the role of theory in studies within the student's area of research, including its role in study conceptualization, methods, and the interpretation of results. 4. Identify the key components of research funding proposals and processes of peer review.	- My dissertation and coursework in Year 1 of PhD involved in-depth investigation into the principles behind cancer screening as a population-based intervention, and measures of its effectiveness. Cancer screening tests and programs and their relationships to outcomes requires nuanced understanding of epidemiologic biases, study designs, appropriate outcome measures, and etiology of the target cancer (in my case, cervical cancer). A comprehensive exam explicitly challenged my understanding of the impact of cancer screening by requiring me to write a proposal for a study that investigated population health impact of self-collected HPV testing. - My dissertation was grounded in three conceptual frameworks, which guided the study questions, designs, and interpretations. I explicitly addressed the conceptual frameworks and the strengths and limitations of their applications in this dissertation. - I contributed to grant proposals during my PhD for travel funding (presentation at a research conference), as a comprehensive exam question, and as part of my dissertation (Chapter 7).
Application of knowledge	5. Systematically identify current controversies in the student's research area, and	- My dissertation highlights two perceived controversies in the field of cancer screening:

	identify, appraise and synthesize pertinent evidence. 6. Identify important knowledge and evidence gaps in health research; propose and prioritize research questions that respond to these gaps. 7. Apply and interpret theoretical frameworks in epidemiologic research in the student's research area. 8. Develop a research proposal that could be submitted to a peer-review competition of a granting agency. 9. Lead the design and execution of appropriate studies to create new knowledge that addresses pertinent health research questions.	evidence-informed practice versus person-centered practice, and; population-based versus individual-level preferences and decision-making. - For my dissertation, I conducted a critical evaluation of quantitative, qualitative, and mixed methods studies on women's preferences for cervical screening. One of the primary findings of my dissertation was that it was impossible to synthesize relevant literature because of the heterogeneity of the study objectives, design, and quality of the studies. - My thesis proposal works through the identification of knowledge gaps that led to my dissertation studies. While writing my dissertation, I found that there were significant knowledge gaps around robust methods to elicit women's preferences, such that it was impossible to synthesize cervical screening preferences evidence. As a result, my thesis direction slightly shifted to address this gap. - As mentioned above, in my coursework, comprehensive exam, and dissertation, I have turned to theoretical and conceptual frameworks to provide structure in the development and execution of research. - With the mentorship of my supervisors and thesis advisory committee, I led the research presented in this dissertation,

		and tables 8-1 to 8-3 communicate how this research contributed new, original knowledge.
Engaged scholarship, knowledge translation, and communication	10. When appropriate, describe how core components of engaged scholarship apply to the student's own learning and research endeavors. 11. When appropriate, engage stakeholders such as professional, patient, and/or community organizations or individuals as partners in designing, conducting, and implementing research. 12. Design a dissemination and/or implementation plan to facilitate the effective use of evidence in policy, programs, patient care, and/or research. 13. Effectively disseminate and communicate epidemiologic evidence to relevant audiences (clinicians, researchers, policy makers, patients and public) in different written and oral formats, including peer-reviewed journal articles, reports for non-academic audiences, and oral presentations.	- In my thesis proposal, I identified the external stakeholders whose input into my research questions and design would be important. Initially I noted women considering screening, health care providers, and guideline developers. While most of my dissertation research focuses on including these stakeholders as research participants, future research (Chapter 7) highlights more collaboration in the planning and executing of the studies. - My Mitacs Accelerate fellowship promoted engagement with industry partners, Eve Medical. This partnership was important for building a network of industry and policy stakeholders (including Cancer Care Ontario). - During my PhD, in 2015, I began working at a Local Health Integration Network, and have tried to engage with researchers as a health system knowledge user. My role as an Epidemiologist has evolved into a knowledge broker in an attempt to bridge the divide between health systems research and implementation, particularly focused on Northwestern Ontario. I am currently a co-investigator on a Ministry of Health and Long Term Care grant.

		- My thesis proposal and Chapter 7 of my dissertation both include explicit dissemination plans. I have attended conferences, produced peer-reviewed publications, and created presentations for knowledge users and other researchers. - The creation of an online preference-elicitation tool (Chapter 6) is an example of a dissemination tool meant for non-research audiences.
Autonomy, professional capacity, leadership and mentorship	14. Identify and adhere to ethical principles relevant to epidemiologic research that apply locally, nationally, and internationally, including the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS). 15. Adhere to principles of academic integrity according to the policies of the University of Ottawa. 16. Effectively peer-review publications, research funding proposals, and research ethics applications. 17. Acknowledge the strengths and limitations of the student's own work and discipline, of the complexity of knowledge, and of the potential contributions of other interpretations, methods, and disciplines. 18. Appreciate the value of and participate in an interdisciplinary research community in their chosen field of health research.	- I participated in the TCPS-2 training, in my first year of my PhD - Ethical principles such as autonomy, utilitarianism, and paternalism were contemplated and discussed throughout my dissertation. - My professional skills development course in the first year of my PhD helped lay the foundation for academic integrity, specifically for the University of Ottawa. - I participated as a reviewer for the University of Ottawa Medical Journal in my first year of my PhD. I continue to peer review articles for specific journals. As part of my role at the LHIN, I have the opportunity to review funding proposals relevant to regional health systems. - During the final year of my PhD, I was accepted into a fellowship program about the Transdisciplinary Understanding and Training on Research in Primary Health Care. As part of

	19. Further develop leadership and mentorship abilities, including the ability to lead, organize and support teams from various backgrounds to work together to achieve a specific outcome. 20. Effectively teach epidemiologic and public health methods. 21. Commit to proactively seeking new knowledge and being aware of current issues of debate in their chosen field of health research.	this, I am developing a transdisciplinary research network of professionals who are interested in primary health care. - Throughout my PhD, I have been a research tutor (for the undergraduate medical program) and have participated as a lecturer in the Epidemiology Summer School at the University of Ottawa. I aim to apply to Northern Ontario School of Medicine as a lecturer/tutor after I submit my thesis. - During my PhD, I learned that one of the simplest ways for me to learn about new and relevant research is through a Twitter platform. Although I'm trying to establish journal clubs within my organizations to promote discussion about new publications, I learn about new knowledge through Twitter alerts. I continue to use traditional forms of communication as well (through webinars, email communication with relevant journals and organizations, conferences, etc).

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