

**Engaging Health Care Professionals in Personalized Medicine:
A Pilot Study Comparing Two Professional Engagement Approaches**

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ABSTRACT

Given the emerging importance of personalized medicine (PM) in primary care, now should be the ideal time for engaging with health care professionals (HCPs), both physicians and nurses, about integrating PM into practice. The question then becomes: what is the most effective way to engage with HCPs about emerging technologies that are not in routine clinical use and which are unfamiliar to many?

The overall aim of this pilot study was to develop and compare two professional engagement (PE) approaches for engaging with HCPs about PM to inform their development and design of a future formal evaluation. The first PE intervention was a structured in-person focus group and the second was an online version, also incorporating an educational component, but without group interaction. The pilot study showed that while participants evaluated both interventions positively, the in-person workshop consistently scored higher; however, recruitment challenges were a major obstacle for this approach.

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CHAPTER 1. INTRODUCTION

1.1 MOTIVATION

Personalized medicine (PM) is the customizing of preventive and therapeutic interventions according to individual variation in risk or treatment response (Chan & Ginsburg, 2011). Proponents believe this will ensure more effective medical interventions at the individual level (Ginsburg & Willard, 2009), promote advances in disease prevention (Haga et al., 2011), reduce harms from adverse drug events (Hughes et al., 2008; Phillips et al., 2001), and lead to more efficient use of health care resources (Teng et al., 2012). However, many are skeptical about unsubstantiated ‘hype’ surrounding PM, raising doubts as to the clinical utility (Dewey et al., 2014) and added value of PM (Haga et al., 2011), as well as questioning the economic feasibility of the PM model (Phillips et al., 2014).

PM is a broad idea that encompasses many different notional applications, including: whole genome sequencing, genetic counseling, direct to consumer testing, pharmacogenomics, companion diagnostics, genomic risk profiling for common complex diseases and family history. The terms ‘stratified medicine’ and ‘precision medicine’ are also used and perhaps more clearly convey the idea that while completely individualized medicine may be unachievable (Nicholls et al., 2014), PM can enable stratified medicine, defined by Trusheim as ‘*Segmenting a patient population into subgroups based on hereditary risk of a disease occurrence, recurrence or likelihood of treatment response, or somatic changes in a tissue, most often a tumor*’ (Trusheim & Berndt, 2007). For a systematic review of the different ways in which the term is used in clinical practice see Schleidgen et al.’s recent work (Schleidgen et al., 2013).

A few PM applications are beginning to transition into clinical practice, particularly in the areas of pharmacogenetics (Caldwell et al., 2011; Kamali & Wynne, 2010; Ridker et al.,

2012) and oncology (Cornetta & Brown, 2013; Santos et al., 2013), but the bulk of research activity is still in the discovery and early technology development phases (Conti et al., 2010). Nevertheless, there is considerable debate surrounding PM, specifically whether genomics should be integrated into mainstream health care.

Although it is impossible to predict the timescale over which clinically useful PM applications will emerge, it is likely that this shift in medicine will to some extent become a genuine reality. The core idea of improving disease prediction or prognostication is not revolutionary, risk stratification systems are widely accepted in medicine (e.g., the Framingham Coronary Heart Disease Model (Kannel et al., 1961)), and informatics advances in business intelligence and analytics will eventually make it practical to apply such approaches more generally. At the same time, the technical developments required for genomic analysis are progressing at a rapid pace, with the ‘\$1,000 genome’ - meaning the sequencing of a person’s entire genome for \$1,000 or less - rapidly becoming a reality (Hayden, 2012; Herper, 2012). At \$1000, the cost will be comparable with other tests ordered in routine clinical applications, making genomic testing a realistic diagnostic option for health care providers (Calzone et al., 2013), and bringing credibility to claims that the *‘Coming decade will bring a steady stream of examples where genomic information is shown to be a valued component of clinical practice’* (Phimister et al., 2012).

To date, genetics in Canada remains embedded in a service delivery model based on genetic counseling and in depth risk assessment by specialists in tertiary centers which is unsustainable as a model for the kind of PM described above. The literature supports the idea that, as evidence-based PM applications become available, primary care physicians (PCP) will play a key role in implementing PM (Bonter et al., 2011). Increasingly the literature also

suggests that health care professionals (HCPs) from all specialties, including nurses and nurse practitioners, will be involved with realizing PM in clinical practice (Calzone et al., 2013), including ordering and interpreting genomic tests. Given the possible emergence of PM in primary care and potential for impact on the Canadian health care system, and knowing that the best time to evaluate a technology is before it comes into routine use (Lehoux, 2006), now should be the ideal time for engaging with both physicians and nurses on the potential integration of PM into clinical practice. The question then becomes: what is the most effective way to engage with HCPs about emerging technologies, such as PM, that are not yet in routine clinical use and which are still unfamiliar to many?

To answer this question, this thesis built upon pilot work by Wilson et al. (Wilson et al., 2009). They used focus groups with community members, PCPs, nurse practitioners, and gastroenterologists, to explore reactions to using a form of PM, genomic profiling, as part of colorectal cancer (CRC) screening for the general population. Measuring multiple genetic variants as a method of risk stratification for CRC is not currently available but it was a useful example for exploring the perspectives of the target population and of the professionals who would implement it. However, this early research raised concerns about the validity of the qualitative methodology, in particular, whether the detailed information about the nature of the technology provided by the group facilitators might have influenced the participants' responses. This led to adoption of a more transparent approach in which defined information was incorporated into a structured qualitative process. Prior to this thesis project, this standardized methodology had been applied in public engagement workshops with community-based participants but not with professional groups (Wilson et al., 2009; Craigie et al., 2012; Nicholls et al., 2012, 2013).

The pilot study conducted in this thesis examined the issues encountered in adapting the public engagement methodology for exercises with health professionals, and evaluated whether structuring it as an online workshop offered a more feasible alternative to the costly and logistically challenging live facilitated workshop format. Potential benefits of the online approach included: eliminating the need for HCPs to be physically present during engagement and to participate at set times; reducing the time needed to participate; and the potential to reach a different demographic than with traditional focus groups.

1.2 THESIS OBJECTIVES

This research was part of a multi-province study based in Ottawa, Ontario to develop effective methodologies for public and health professional engagement surrounding emerging genomic technologies. The overall aim of this pilot study was to develop and compare two professional engagement approaches on the topic of PM in health care to inform their further development and the design of a future formal evaluation. This is a category 1 thesis in that it is a complete research project, including data collection, designed to evaluate a methodology; the thesis also includes components of a category 3 thesis in that it involves instrument development designed to evaluate a methodological question (i.e. how best to engage HCPs about emerging technologies?) The specific objectives of the study were as follows:

1. Develop a structured workshop for HCPs for engagement research on the topic of emerging applications in genomics.
2. Use the content developed for the structured workshop as a basis to develop an online workshop feasible for replication and dissemination, accounting for the target audience and their expectations and experiences with online interventions.

3. Compare the in-person and online workshop in terms of: a) user rating and satisfaction measures; b) the quantity of data collected; and c) logistical considerations, such as feasibility, ease of recruitment, and cost.
4. Describe substantive findings about genomics based on HCPs' opinions about how to integrate PM into primary care and identify issues they perceive as salient to successful adoption and implementation of PM.
5. Identify factors that may facilitate as well as challenge professional engagement with HCPs and inform future efforts to develop best practices for engagement which will be evaluated through a future formal trial.

1.3 SIGNIFICANCE OF THE RESEARCH

While some believe that PM will bring about a 'paradigm shift' in medicine, there is evidence that implementation and integration into routine health care may be challenging (Cornetta & Brown, 2013; Najafzadeh et al., 2013). Professional acceptance of emerging PM interventions, and of the concept of PM overall, will be a major factor in determining the clinical utility achieved in practice. Engaging professionals at an early stage not only informs existing health technology assessment (HTA) processes, but can also provide useful input into the way a technology itself is shaped and implemented in practice. This requires exploration of the issues well in advance of the introduction of a new technology, which is challenging in the case of emergent technologies like genomics which lack an established evidence base (Conti et al., 2010; Veenstra et al., 2010) and where, in some cases, the clinical applications are as yet hypothetical. In Canada, HTA of genetic technologies is in preliminary stages and has '*evolved as a predominantly informal, ad hoc process*' (Adair et al., 2009).

Previous research suggests that, for new uses of genomic information, engagement

methods must embed educational information to develop a baseline understanding among participants as well as eliciting reactions, opinions, and attitudes (Craigie et al., 2012; Nicholls et al., 2012, 2013). In order to achieve this, engagement methods are required which both promote the validity of the data captured and are also practically and economically feasible. This pilot study directly compares two professional engagement approaches and assesses a range of outcomes which inform an understanding of their validity to clarify the most important attributes for their more widespread use and formal evaluation. By trying to understand how HCPs react to genomic profiling approaches as they might be implemented in practice, and by encouraging discussion about the non-technical aspects and consequences, this pilot study provides evidence which informs how PM could best be implemented (e.g., by whom, with what kind of information or consent, with what supporting services, etc.).

The thesis resulted in findings in two areas, i.e., methodological (preliminary evaluation of the two interventions) and substantive (what professional attitudes and reactions were observed?). These data are significant for: 1) comparing the feasibility and utility of two different approaches to professional engagement; and 2) identifying factors that facilitate and challenge the adoption of genomic risk profiling by HCPs and inform future efforts to understand the utility of genomic risk testing. Given that this was a pilot study, the latter are reported with appropriate cautions.

CHAPTER 2. LITERATURE REVIEW

In the future, if scientific research presents a strong evidence base for PM applications, this could bring about a fundamental change in medicine, in which genomics becomes increasingly embedded in everyday clinical practice. This is the key difference between traditional genetics and PM, the former is about rare disorders and specialized genetic practice, while the latter refers to the application of genomic medicine across many or all specialties and areas of health practice. In order for the potential benefits for health care and population health to be realized, HCPs would need to integrate these new genomic applications and systems into routine practice.

The ACCE framework (Haddow & Palomaki, 2003) is used in the US to evaluate genetic tests, focusing on analytic validity, clinical validity, clinical utility and associated ethical, legal and social implications (see Figure 1); however, evaluations based on the ACCE framework do not include the target populations and considerations of their needs. If health care is aiming for evidence-based use of evidence-based technologies then this requires that the technologies yield valid results and that they are applied appropriately. Lehoux advocates a '*technology-in-practice*' perspective where it is understood that health care technologies do not operate in isolation, but are rather applied as part of a network of interacting health care professionals, patients, and other technologies (Lehoux, 2006).

Based on this view, there is a need to engage with the target population to learn their views before genomic technologies are integrated into clinical practice; this is where the literature review fits in. First, an overview of PM is provided, followed by a summary of the professional engagement literature on engaging about genomics with HCPs. Standard professional engagement methodologies are discussed, forming the rationale for the two

professional engagement approaches selected for comparison in this thesis.

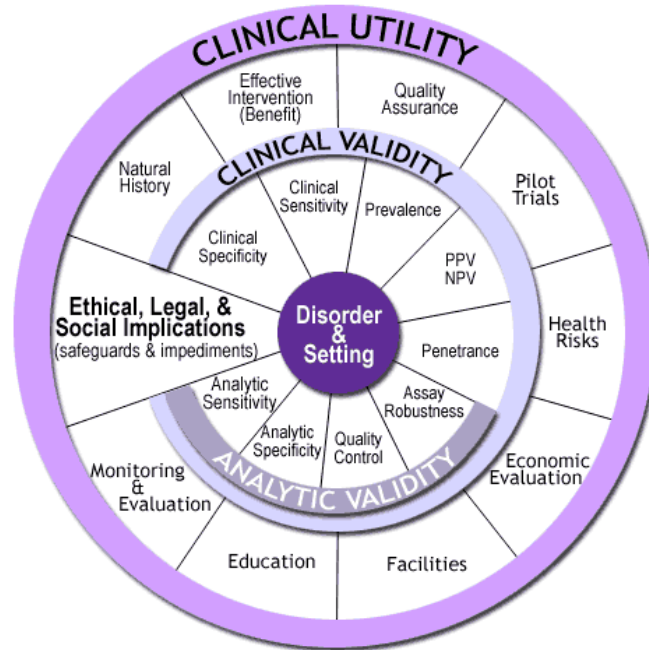


FIGURE 1 ACCE MODEL PROCESS FOR EVALUATING GENETIC TESTS
IMAGE CREDIT: [HTTP://WWW.CDC.GOV/GENOMICS/GTESTING/ACCE/](http://www.cdc.gov/genomics/gtesting/acce/)

2.1 PERSONALIZED MEDICINE

There is a key distinction in the type of information generated between genetic testing for Mendelian disorders and genomic risk profiling for common complex diseases which forms the basis of PM. The former produces a relatively certain prediction of disease, whereas the latter provides risk information through genomic profiling, where genetic variations are linked through statistical associations to common complex diseases (Becker et al., 2011; Haga et al., 2011). Genomic medicine seeks to identify genetic components involved in common complex diseases such as type 2 diabetes, coronary heart disease, cancers, etc. whereas medical genetics (or clinical genetics) focuses on single gene disorders such as Huntington's disease. If a patient's genomic risk information indicates genetic susceptibility to a complex condition this does not mean the patient will necessarily develop the disease, rather it is the '*interaction between genetic and environmental variables that leads to ill health*' (Harvey, 2011). This inherent level of

uncertainty presents new challenges when interpreting results for patients because a positive outcome may not lead to disease development. Despite uncertainty and controversy over how to incorporate tests into routine medical care (Haga et al., 2011), there is increasing patient demand for PM (Adair et al., 2009; Bonter et al., 2011).

Evidence-based genomic tests associated with PM should demonstrate analytic validity, clinical validity, and clinical utility (Secretary's Advisory Committee on Genetic Testing, 2000). As explained by Harvey, analytic validity indicates how well a test measures the property or characteristic it is intended to measure, clinical validity means that the test is able to predict increased disease risk, and clinical utility means that testing leads to an improved health outcome (Harvey, 2011). To date, the evidence base is extremely underdeveloped for PM applications, with many arguing that both the validity and utility of genomic risk information is in doubt (Clarke & Thirlaway, 2011), raising questions as to when and if genomic tests will be ready for integration into patient care.

The motivating goal of PM is to better inform decision making about prevention and treatment. In the ideal scenario, the patient's risk of developing the disease in the first place is reduced or even eliminated. For instance, genomic profiling to improve breast and prostate cancer screening provides a strong motivating example from a public health perspective of the potential clinical utility of genomic risk stratification at the population level (Pashayan & Pharoah, 2012). Increased knowledge of susceptibility could also increase patient awareness and vigilance with respect to disease screening, resulting in earlier diagnosis (Haga et al., 2011) and more successful disease treatment which would be targeted to the patient's genetic profile (McBride, Koehly, Sanderson, & Kaphingst, 2010). However, evidence strongly suggests that knowledge alone does not lead to behavioral change (Ryan, 2009) which raises doubt as to

whether these benefits would actually be realized. While PM has proponents and skeptics, the general sense from the literature is that there is an inevitable force behind PM as a technological shift in medicine, suggesting that it would better to prepare for the dissemination of such applications at this early stage by assessing the clinical and societal possibilities and implications (Becker et al., 2011).

2.2 PROFESSIONAL ENGAGEMENT & PERSONALIZED MEDICINE

When and if personalized medicine applications supported by evidence become available for use in health care, there is a strong case to be made for primary care professionals (PCPs) playing an important role in providing PM: they are the first point of contact for patients (Bonter et al., 2011), it is expected that PM will become increasingly pervasive in daily practices (Ohata et al., 2009), and PCPs have a long-term relationship with patients and families (Burke & Psaty, 2007). Miller et al.'s study of cancer patient expectations of PCPs found that patients expected PCPs be supportive and educated towards genetic testing and to play a role in initial referral and ongoing surveillance (Miller et al., 2010).

Wilson suggests that while PCPs play mainly a referral and supportive role in predictive and pre-dispositional genetic tests, for susceptibility tests for complex disorders their role would logically extend to include test ordering, interpretation and management (Wilson, 2012). Trinidad et al. identified the following new PCP responsibilities associated with integrating PM into primary care: *'recognizing patients who should be referred for genetic testing, ordering and interpreting tests, communicating risk information, promoting prevention strategies, providing advice to patients about the meaning of genetic variations, prescribing drugs and responding to patients seeking information after receiving direct-to-consumer test results'* (Trinidad et al., 2008). While some studies argue that PCPs are ill equipped to manage these new PM related

responsibilities, with limited knowledge of both genetic testing and the skills required to translate results into clinically relevant information, others believe that PCPs will be the gatekeepers for PM (Bonter et al., 2011).

In addition to physicians, recent literature from the nursing field suggests that participation from all levels of the nursing profession will be critical in the successful wide scale implementation of PM in primary care. For example, Bancroft firmly believes in the joint physician/nursing model of genomic care, stating: *‘Primary care practitioners and nurses in particular will be at the forefront of using and interpreting genetic test results for the prevention and treatment of common chronic diseases, with only the more complicated cases referred to a genetics specialist’* (Bancroft, 2013); this includes all nursing professionals and is not limited to advanced practice nurses. Nurses’ roles in clinical practice include but are not limited to assessing family pedigree, interpreting risk factors, offering counseling on genetic testing, and providing education (American Association of Colleges of Nursing, 2008).

Considering both the physician and nursing perspectives, this section summarizes the professional engagement literature relating to PM with the aim of identifying the key points that a professional engagement intervention needs to achieve. Lessons learned from the literature review were subsequently incorporated into the intervention design (see Chapter 3).

PHYSICIAN PERSPECTIVE

In 2008 Scheuner et al. conducted a systematic review of delivering genomic medicine for common chronic adult diseases (Scheuner et al., 2008). They identified numerous challenges, including consumer information need, issues related to delivery of genetic services, and other social barriers. The review concluded that the primary care workforce is unprepared for integration of genomics into practice and that health professionals lack basic knowledge about

genetics. The review recommended that further studies were needed to identify challenges to implementation in clinical practice and to test models for how implementation could be feasibly accomplished. This section presents studies conducted since the publication of the 2008 systematic review.

Haga et al.'s survey of 356 PCPs studied physician attitudes and uptake of genomic risk profiling; all participants were members of a national US health network focused on providing personalized care and had received webinar training from Navigenics. They found that respondent familiarity with genetics was a key predictor of physician ordering behaviour, with 52% of respondents feeling comfortable ordering genetic tests (Haga et al., 2011). Concerns related to clinical utility, risk of long-term/disability or life insurance discrimination, confidentiality, and cost. A limitation of this work is that the results are not widely generalizable because the physicians received discounts and training from Navigenics.

A recent large scale American survey of academic family physicians administered by the Council of Academic Family Medicine Educational Research Alliance included a section pertaining to integrating genetic testing in clinical practice (Mainous et al., 2013). The survey focused on physicians' genetic testing practice behaviors and assessment of the value of genetic testing in primary care. The findings indicated that the majority of respondents did not feel knowledgeable about available genetic tests, which is in line with Scheuner et al.'s previous findings. Despite limited knowledge, respondents for the most part believed that genetic testing would have a substantial impact on their clinical practice in the next 5–10 years and that genetic testing should be an important part of the medical curriculum. However, the authors point out that, because the respondents were academic physicians, they may be more informed about genetics than non-teaching physicians.

Bonter et al. recently published the first national survey of Canadian physicians' perceptions relating to PM. They found only 51% of respondents felt there was sufficient evidence to order genetic tests while 50% believed that genetic tests that would be useful were not readily available in Canada (Bonter et al., 2011). Physicians reported many barriers to successful adoption of PM including: limited provider knowledge, lack of clinical guidelines, lack of evidence-based clinical information, cost of tests, patient anxiety, and doubtful clinical utility (e.g. test results would not affect treatment). The survey had a low response rate of 8.3% and it is worthwhile to note that the authors speculated that the topic of PM may not have been considered relevant to many of the target physician population. This highlights the issue of physicians not viewing PM as being relevant to their clinical practice.

Wilson et al.'s exploratory Canadian study of professional views on population level cancer screening reported confusion with respect to the meaning of PM, with professionals overwhelmingly framing 'genomic profiling' as traditional 'genetic testing' (Wilson et al., 2009). In terms of communicating with patients, PCPs believed that there was a need for both specialist knowledge to interpret genomic test results and for counseling to support informed decision-making. They concluded that professionals viewed PM as inconsistent with public health models of practice (e.g. promotion of high uptake, low cost and necessity for ease of implementation).

Using the example of genetic testing to identify and manage susceptibility to type 2 diabetes, Harvey conducted semi-structured interviews with geneticists, diabetologists and PCPs (Harvey, 2011). She found that while geneticists had little interest in expanding their responsibilities to include PM for common complex diseases, PCPs saw PM as another risk assessment tool, similar to taking a family history: *'PM aligns with their role as doctors who identify health risks and help patients to manage those risks through lifestyle modifications'*

(Harvey, 2011). Haga et al also found that PM aligned with the PCPs' roles, given that one of the expected benefits of PM is disease prevention (Haga et al., 2011), and disease prevention is a main component of primary care. Harvey reported three key findings that are contrary to some previous studies: 1) integrating PM into the patient journey would not require reconfiguring existing practice; 2) PCPs recognize the distinction between genetic and genomic medicine; and 3) PCPs are not ambivalent towards genetics education.

Avard and Knoppers conducted focus groups with Australian PCPs to determine their views on integrating genetics into patient management (Avard & Knoppers, 2009). They identified communicating test results with patients as a key area of concern '*especially when the test has a limited ability to predict whether the gene variant will result in disease*'. Participants were most receptive to PM in situations where results could positively impact patient management. As with other studies, they found that PCPs lacked genomics/genetic knowledge and they reported ambivalence on the part of PCPs about integrating genetics into clinical practice.

Recently, the focus group approach was also used in British Columbia, Canada, to investigate physicians' perceptions about the future role of PM and identify factors that influence their decision about using genetic testing in practice (Najafzadeh et al., 2013). Facilitators started the discussion by asking: '*What comes to mind when you hear the term personalized medicine?*', then after hearing participants' responses they provided a working definition of PM; no further educational materials were provided. While the focus group discussion centered around challenges for integrating PM into clinical practice, participants overall indicated that despite these challenges they were interested in using genetic information in their practice but expected to have access to training and clinical guidelines.

NURSING PERSPECTIVE

In addition to physicians, there is an increasing focus on the impact genomics will have on the nursing profession. A large scale consensus effort of nurses in the United States resulted in the development of the Essentials of Genetic and Genomic Nursing (American Nurses Association, 2008) competencies for registered professional nurses; these were updated in 2012 to include competencies for advanced practice nurses including knowledge and clinical performance indicators in two domains: professional responsibilities and professional practice (Tonkin et al., 2011).

As the largest group of health care professionals in Canada, nurses will play a key role in determining how genomic discoveries translate from research settings into clinical practice. Calzone has published extensively in the area of nursing genomics, and she writes: *‘Genomic developments are changing health care. And because nursing is a fundamental provider of health care, genomics is also changing the profession of nursing’* (Calzone et al., 2013). Nurses will be one of the main points of contact for patients about genomics, and are well placed to do so given their traditionally close relationships with both patients and families (Bancroft, 2013). As genomics continues to move away from the specialist referral model there is an increasing need for nursing professionals to have genetic competencies: *‘Nurses have a responsibility to be informed and to inform other healthcare professionals, individuals, families, and communities of the potential benefits and challenges of genomics’* (Calzone et al., 2013). This team advocates for more research targeted at how nurses will deliver genomics in primary care (Calzone et al., 2013); this is important because previous research showed that most nurses view genetics as being only relevant to specialty nurses rather than the entire profession (Calzone & Jenkins, 2011); this echoes the finding of a perceived lack of relevance reported in physician studies

In 2013 Calzone et al. completed an online survey based on a convenience sample of 620

nurses, examining their attitudes and competencies in genetics and genomics (Calzone, Jenkins, & Culp, 2013). While almost all (94%) of respondents felt it was very or somewhat important for nurses to become more educated about genomics, over half (63%) self-reported that their overall genetic/genomic knowledge was only poor or fair; however, 71% intended to learn more about the topic. From a practice perspective, the majority was not confident about providing patients with information about genomic services, including availability, limitations and benefits of services, nor were they confident in facilitating referrals for these services. The authors measured integration of genomics by assessment of family history utilization, 18% of nurses always or often assessed family history; 21% did so occasionally and 60% rarely or never. Overall the survey findings portrayed nurses as being willing and interested in genomics, yet lacking the knowledge, preparation and support required to promote *‘genomic clinical translation and realize the benefits of PM’* (Calzone, Jenkins, & Culp, 2013).

As with physicians, there is consensus that nurses currently have a lack of genomic knowledge and that this limits their ability to translate genomics into clinical practice (Thompson & Brooks, 2011). The Panel on Genetic/Genomic Nursing Competencies is aiming to overcome this lack of knowledge by preparing current and future nurses with essential genetic and genomic competencies (American Nurses Association, 2008). Trained nurses will then supposedly be able to apply evidence-based interventions and expert professional guidelines to improve patient care (Santos et al., 2013), taking this a step further, Badzek et al believe that *‘Failure to integrate genomic competencies into nursing practice may be associated with negative patient outcomes’* (Badzek et al., 2013).

2.3 PROFESSIONAL ENGAGEMENT METHODOLOGIES

Professional engagement is necessary when considering translational issues relating to

genomic technologies in Canada because Canadians do not have a consumer-based health care system and HCPs are generally gatekeepers to the technology. There are numerous engagement methodologies, based on both quantitative and qualitative research designs, of which surveys, focus groups, and individual interviews are the most commonly used for engaging with HCPs (Rowe, 2005). This was also evidenced by the literature review (Section 2.2), where these methodologies were consistently selected for professional engagement relating to PM. This section examines these three general approaches, and their electronic equivalents, and discusses their strengths and weaknesses as methods for engaging with professionals on emergent technologies.

SURVEYS

Surveys are a commonly used quantitative approach to professional engagement in health care and represent an important source of information when implementing new health care programs (Wiebe & Mackay, 2012). While surveys have a long history and can be easier to implement than qualitative approaches (below), evidence suggests that response rates to health care related surveys are declining (Cook et al., 2009), which raises concerns about selection bias. The literature indicates specifically that surveys of both physicians and nurses are problematic. For the 2007 National Physician Survey, Grava-Gubins et al. applied multiple recommended methods for maximizing response rates (including shortening the questionnaire, enhancing marketing, and providing a monetary lottery incentive) but still failed to match the response rate of the 2004 version of the survey (35.9% in 2004 as compared to 31.6% in 2007) (Grava-Gubins & Scott, 2008). Wiebe et al. conducted a fax survey of Canadian physicians and achieved a response rate of 14% (Wiebe & Mackay, 2012); they examined why response rates in clinician surveys were declining and found that many physicians had office policies not to participate in

any surveys. They suggested that this may result in surveys no longer being '*an effective or useful method of collecting information about physicians' knowledge of, attitude toward, and experiences with various medical practices*' (Wiebe & Mackay, 2012). Such policies pose significant challenges for professional engagement with HCPs, especially when considering that physicians already receive a large amount of surveys in the mail (Ziegenfuss et al., 2012), suggesting that sending unsolicited 'blanket mailings' to physicians' offices may be a poor survey recruitment approach.

Increasingly, online questionnaires are used to reduce cost, and achieve faster responses with more complete data collection (Chizawsky et al., 2011). Chizawsky et al. evaluated the feasibility of a web-based survey approach in a nursing population and reported a response rate of 55% (Chizawsky et al., 2011). However, Dykema et al. argue that online surveys may not in fact be cheaper; their evaluation of response rates based on a national web survey of physicians found that substantial financial incentives were needed to encourage physician participation (Dykema et al., 2011). Their findings indicate extremely low response rates of 3.0–6.2% in the group not guaranteed a financial incentive, as compared to 25.4% for those offered \$100.

Regardless of the application domain, the largest challenge with the Internet-based survey approach is to capture and keep the user's attention. Considering that it is estimated that only 17% of page views last more than 4 seconds, this can be a difficult task (Weinreich et al., 2008); strategies to achieve improved response for Web-based surveys include keeping surveys as short as possible and including rich media content.

FOCUS GROUPS

Focus groups are a form of qualitative research where a small group of participants from a target population are brought together to discuss topics preselected by the researcher

(Lakshman et al., 2000). The moderator guides the discussion, sometimes using pre-developed materials that encourage group interaction (Barbour, 2007). The role of the moderator is to guide the discussion without allowing his or her personal views or biases to intrude (Gill et al., 2008) to ensure that all participants have a chance to be heard, and to manage discussion such that no individual dominates. The goal is that the group interaction will generate a greater breadth of data than could be captured from individual interviews (Festervand, 1984; Morgan, 1997); as well as responding to the moderator, participants are encouraged to respond to other participants' comments with the idea that this will help participants 'explore and clarify their views' (Kitzinger, 2006). The quality and validity of the data obtained from focus group research is largely determined by the skills of the moderator (Ayres, 2007), particularly his or her ability to manage diverse group dynamics; this adds a level of complexity in developing a standardized engagement process with generalizable results.

Focus groups are often used in health services research (Barbour, 2007). On the one hand, they may be well suited to exploring issues regarding the likely use of emergent technologies not yet routinely used in clinical practice (Barbour, 2003). On the other hand, this same issue – discussing emergent technologies which are not in any way familiar in routine practice – may demand considerable expertise on the part of the moderator (to also act as educator) or require carefully developed unbiased materials to facilitate adequately informed group discussion. It is important to note that the problem of recruiting HCPs highlighted in the survey section is also applicable to focus groups. For example, to conduct their physician focus groups Najafzadeh's team offered a \$200 honorarium for participation in a 90 minute session and 28 physicians were recruited (Najafzadeh et al., 2013).

With the development of Web 2.0, new approaches to try to capture the benefits of the

group dynamic in a more flexible way have started to appear. For example, electronic participation experiments are an exploratory method of participation where information distribution and participant interaction are provided via the computer; these are termed '*e-focus groups*' (Williams, 2009). Compared to face-to-face focus group participation, electronic participation experiments might be cheaper and easier, and have other advantages: the ability to reach groups, such as younger people, who are traditionally hard to engage (Forcella, 2006); providing greater time for deliberation; and obtaining more candid responses (Williams, 2009).

INTERVIEWS

Interviews are the most commonly used qualitative technique in health care research (Britten, 2006); a semi-structured approach is most often used in health research, with a list of topics combined with open-ended questions that allow the exploration of responses in more detail. Qualitative interviews are considered a powerful method for learning about the subject's '*everyday world*' (Kvale, 2007) and the ability to expand on issues that are important to the interviewee but might not have been considered by the researchers are a strength of the interview methodology (Gill et al., 2008); this may not be possible in focus groups where time constraints and group dynamic mean that the moderator must limit each participant's contributions. As with focus groups, the validity of the data lies with the interviewer, who must not ask leading questions and remain unbiased while steering the interview, probing for clarity and information, and testing responses for consistency.

SUMMARY OF ENGAGEMENT APPROACHES

The similarities and differences between these different engagement approaches are summarized in Table 1, where we compare across four attributes describing how they work in practice: 1) presence or absence of a moderator; 2) extent of group interaction; 3) communication

style (synchronous communication occurs when all participants are present for the event at the same time, asynchronous when not present at the same time); and 4) time available for deliberation.

TABLE 1 ATTRIBUTES OF PUBLIC ENGAGEMENT APPROACHES

ATTRIBUTES	ENGAGEMENT APPROACHES				
	Survey	Online survey	Focus group	e-focus group	Interview
Moderator	No	No	Yes	Possibility of virtual moderator	Yes
Group interaction	None	None	In person	Virtual	None
Communication style	n/a	n/a	Synchronous	Synchronous and/or asynchronous	Synchronous (with moderator)
Time available for deliberation	~30 minutes	~30 minutes	~1 hour	Possibility of longer deliberation	~1-2 hours (variable)

While some argue that the focus group approach is most likely to produce new insights on a topic because of its semi-structured approach and the synergy of the group interaction, some question whether deliberative processes actually lead to improved or different decisions (Abelson et al., 2003). It is possible that a group session may offer too little time for the participants to fully process and deliberate an issue. Sturgis et al. (Sturgis et al., 2010) followed up participants in public engagement exercises and found that attitudes to new topics became more positive over a period of weeks; this may be particularly relevant to genomics which is a new topic for many HCPs.

From a practical viewpoint, focus groups may cost more than surveys or interviews (Abelson et al., 2003), and are challenging to organize because of the need to physically bring together participants at the same time; this may be particularly true for busy HCPs. This raises the argument that an important question to ask is whether the focus group methodology really

does lead to different kinds of insight and engagement data as compared to the survey approach.

Considering economies of scale, the focus group may be more efficient in terms of the moderator's time spent educating participants and offers the advantage of participants learning from each other's views. In a situation like genomics, where the topic is novel and may inspire trepidation in some participants, the focus group dynamic may put participants at their ease instead of being singled out in an interview situation.

2.4 SIGNIFICANCE OF LITERATURE REVIEW FOR INTERVENTION DESIGN

In summary, the literature reveals strong similarities between nurses and physicians in terms of both their views on genomics and challenges for integration into clinical practice. While the evidence indicates that there is interest in becoming more educated, both groups are currently hindered by a lack of knowledge about genomics; this is particularly true for nurses where genetics is not generally incorporated into the nursing curriculum (Canadian Nurses Association, 2005; Chuang et al., 2013). A number of physician surveys reported ambivalence towards genomics, with many viewing genomics as not relevant to their clinical practice, and this finding was echoed in the nursing research.

From the viewpoint of a professional engagement approach, these findings suggest that, as well as an 'evaluation' component with questions to prompt engagement and learn about HCP's views, there is a need to include a component that provides unbiased and evidence-based educational materials and another component that emphasizes relevance to clinical practice. This points to selecting an engagement approach, or combination of approaches from Table 1, which are feasible and suitable for incorporating these three components.

CHAPTER 3. INTERVENTION DESIGN

This chapter presents the professional engagement approach as a complex intervention design. First, the informed basis for the overall design is discussed generally, focusing on the underpinning theories. This is followed by a description of the interacting components of the intervention and the implementation details for both versions of the professional engagement (PE) intervention: the in-person workshop and online version of the workshop. Developed and refined between November 2012 and March 2013, the intervention design was an iterative process influenced by lessons learned during an initial professional engagement exercise presented at the Primary Healthcare Partnership Forum (PriFor) in Newfoundland Canada in November 2012 [<https://www.med.mun.ca/phru/downloads/ConferenceGuide.pdf>].

3.1 PROFESSIONAL ENGAGEMENT AS A COMPLEX INTERVENTION

Healthcare and social intervention may often be considered ‘complex’. According to the UK Medical Research Council (MRC) (Medical Research Council, 2008), a ‘complex intervention’ contains multiple different components which together are thought to contribute to the effectiveness of the overall intervention, but where it may be difficult to separate out the effect of any one component or isolate the ‘active ingredient’ (Campbell et al., 2000; Medical Research Council, 2008). There are often assumptions that the components act synergistically such that each ‘simple’ component is necessary, implying that removal of one would cancel any overall effectiveness. Examples of complex interventions include multifaceted approaches to patient or professional education, for example interventions designed to reduce inappropriate prescribing in primary care (Grant et al., 2014; Nazareth et al., 2002), improve diagnostics (Iliffe et al., 2012), and coordinate case management (Waugh et al., 2013).

The MRC proposed and refined a phased approach for developing complex interventions which parallels the phases of research and evaluation of drugs, from pre-clinical research to post-marketing surveillance (Medical Research Council, 2000, 2008) (see Figure 2). Ideally, for health care interventions, the starting point is equivalent to pre-clinical research: the development of a coherent framework to guide the selection and drawing together of evidence to indicate the kinds of interventions that would be predicted to be most effective. Working through this phase facilitates formal hypothesis development regarding the intervention, and helps identify strategic issues in intervention design and delivery. The phase that follows is modelling the intervention, i.e., identifying the individual components and the mechanisms by which they are thought to influence outcomes, including considering how they relate to and interact with each other. The first experimental phase follows, with exploratory trial(s), where the goal is to describe the constant and variable components and how they would be put together in a replicable approach. This exploratory phase identifies feasibility issues that need to be addressed before a successful randomized control trial can be initiated, including: *'acceptability, compliance, delivery of the intervention, recruitment and retention, and smaller than expected effect sizes that could have been predicted by thorough piloting'* (Eldridge et al., 2004). The evaluation phase also works towards providing evidence for a future feasible evaluation protocol (for a randomized controlled trial or best alternative).

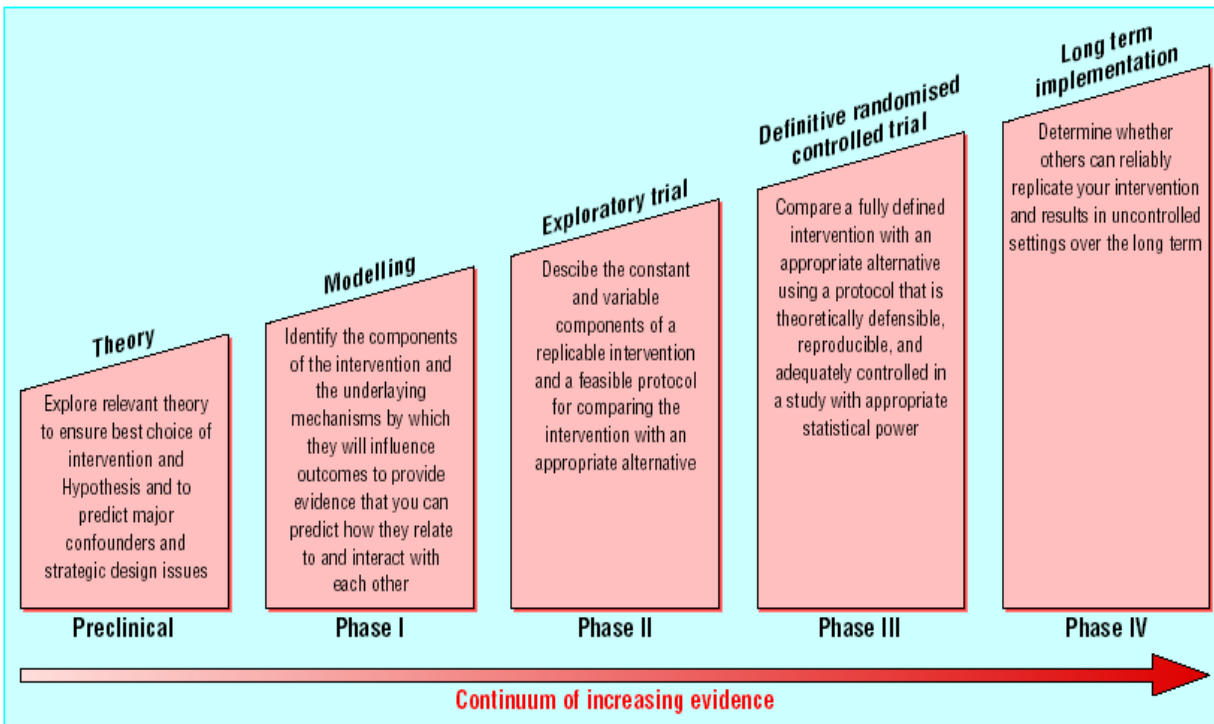


FIGURE 2 MRC'S PHASED APPROACH FOR DEVELOPING COMPLEX INTERVENTIONS

IMAGE CREDIT: [HTTP://WWW.CIHR-IRSC.GC.CA/E/41946.HTML](http://www.cihr-irsc.gc.ca/E/41946.html)

Ideally, once the first three phases of the framework have been completed, the investigators should have developed a theoretically sound, practical, and replicable intervention and be in a position to conduct a definitive evaluation, with an appropriate control intervention, meaningful and relevant outcomes, and with sufficient statistical power.

The complex intervention framework was developed with wide applicability in mind, to be relevant for all kinds of interventions '*with several interacting components such as occur in health service, public health and social policy*' (Medical Research Council, 2008). Craig et al. (Craig et al., 2008) offered thoughts on '*dimensions of complexity*' which include: 1) number of interacting components within the experimental and control interventions; 2) number and difficulty of behaviours required by those delivering or receiving the intervention; 3) number of groups or organizational levels targeted by the intervention; and 4) number and variability of outcomes. After describing the first three stages of the complex intervention framework in the

following sections we will revisit how these complexity dimensions map to the professional engagement intervention developed in this work.

The overarching goal of the two versions of the PE intervention was to engage with health professionals about PM in a valid way. The starting point for developing these interventions was to draw on the key findings from the literature review (Chapter 2) to develop a guiding ‘theory’ which would then inform thinking about important outcomes, selection of specific intervention approaches, and high level design issues. This was followed by the ‘modelling’ phase, to clarify the necessary components of the intervention and work out how these could be implemented. The final part was the ‘exploratory trial’ phase in which the two prototype interventions were evaluated in a pilot manner to gain more experience of their delivery in practice, to inform their further refinement and identify important issues to be considered in the development of a future formal evaluation protocol. Each of the three intervention design phases is described in the following sections.

3.2 ‘PRECLINICAL’ PHASE: GUIDING THEORY

An overarching theme from the literature review was that HCPs, including nurses and physicians, often do not consider genomics/PM as relevant to their current practice and lack training in genomics; this defines an important outcome for a PE intervention and points to developing educational intervention components that promote relevance to practice.

When developing the educational component, exploring the theoretical basis for educating professionals about genetics is especially important because this is anticipatory research lacking an established evidence base. While there is limited research in this area, Farndon & Bennett from the NHS National Genetics Education and Development Centre have published ‘lessons learned’ from their experiences educating health professionals about genetics.

Based on their finding that the majority of non-geneticist HCPs view genetics as being a *'scientific, mostly laboratory based subject, relatively unimportant and not relevant'* (Farndon & Bennett, 2008), they advise genetic educators to provide HCPs with practical clinical scenarios based on examples of common conditions that they would encounter in their daily work to illustrate relevance to practice, stressing that genetic education efforts fail if professionals believe that genetics is *'science'* and *'difficult'* rather than having clinical utility. Korf claims that genetics can be applied in daily clinical practice without an understanding of the underlying science and that genomic pedagogy should be based on presenting clinical cases, focusing on the clinical question being addressed and using a problem solving approach (Korf, 2002). By taking this approach, the focus of educating HCPs about genomics moves towards being able to critically evaluate genomic applications and an understanding of the social implications (Katsanis et al., 2013).

Kirkpatrick's 4 level topology for educational initiatives (Kirkpatrick, 1967) was designed to clarify the kinds of outcomes and evaluations that can be pursued with education. The topology is shown in Table 2; at the bottom level of the topology the educational initiative simply evaluates learners' reactions (e.g. did they enjoy the learning experience?) whereas the top level evaluates whether the initiative creates positive long term change to patient health outcomes. Considering that the literature review revealed that many HCPs are unfamiliar with the concept of genomics, we are focusing the educational goals and subsequent evaluation at levels 1 and 2a. Evaluating whether HCPs have the necessary knowledge and skills (level 2b) to integrate PM into practice or whether the intervention generates a long term change in HCP behavior (level 3), practice (level 4a), or patient outcomes (level 4b) is outside the scope of this thesis given the time and resource limitations.

TABLE 2 KIRKPATRICK'S FOUR LEVELS ON WHICH TO FOCUS EVALUATIONS

[ADAPTED FROM (BARR, FREETH, HAMMICK, KOPPEL, & REEVES, 2000) AS PRESENTED BY (FARNDON & BENNETT, 2008)]

LEVEL	EVALUATION
Level 1	Learner's reactions
Level 2a	Modification of attitudes and perceptions
Level 2b	Acquisition of knowledge and skills
Level 3	Change in behaviour
Level 4a	Change in organisational practice
Level 4b	Benefits to patients, clients, families and communities

Scope of thesis

Another key outcome from the literature is the finding that suggests low response rates to surveys by HCPs; this identifies a strategic design issue driving the choice of professional engagement intervention and suggests that a traditional survey approach may be less likely to be effective than the focus group approach at the other end of the spectrum of attributes from Table 1. Therefore an important question to ask is whether the focus group methodology really does lead to different kinds of insight and engagement data as compared to the survey approach. We chose to evaluate these two engagement approaches in the form of an in-person focus group and online survey because they represent the two ends of the spectrum implied in Table 1 in terms of potential for group interaction, need for a facilitator/moderator, communication style, as well as cost to implement in practice; it was felt that a comparison of their effectiveness would be highly informative.

A further finding from the literature review is that qualitative approaches are considered very appropriate for exploratory research, which we suggest includes attitudes to/experiences with emerging technologies; this finding also positively supports their use from a strategic design

perspective. We considered both focus groups and interviews as possible qualitative approaches; however, individual interviews were not considered practical due to higher costs and the anticipated difficulties in recruiting HCPs; considering economies of scale the focus group approach was chosen as logistically more feasible.

The three key findings identified in this section were used to guide the intervention design and delivery and led to a general intervention approach in which a focus group based methodology is selected for the in-person version of the intervention, and adapted for delivery as an online version. The theory suggests that the methodology should first provide a baseline understanding of genomic concepts, and then apply a case study based approach to promote relevance to clinical practice, following Lehoux's guidance that when considering the role of the physician in health technology it is helpful to define their role '*in the context of clinical work*' (Lehoux, 2006).

3.3 PHASE 1: MODELLING

Having decided to move forward with a focus group type methodology for professional engagement, we looked to previous public engagement work for guidance. Nicholls et al. have already developed and implemented such an approach for the lay public (Nicholls et al., 2013), which itself was based on foundational work by Castle (Castle & Finlay, 2006). We recognized the similarity of purpose of Nicholls et al.'s educational component to our own (promoting baseline understanding and underlining relevance to an aspect of decision making) and concluded that it could be adapted for professional populations.

Briefly, the public engagement approach designed by Nicholls et al. (Nicholls et al., 2013) is as follows. The structured workshop process involves developing standardized information sets which present layers of information and issues to participants in a stepped

manner, providing a *'progressive release of information about genomic risk profiling and its potential implications'* (Nicholls et al., 2013). Using best current evidence and maintaining a neutral approach the layers progress from a general description of PM in set 1, to psychosocial issues which patients might encounter in set 2, to health system and ethical, legal, and social issues in set 3. After each set, the moderator allows time for a facilitated group discussion and data are captured in the form of field notes and through tracker questions that monitor shifts in participants' attitudes towards genomics. The use of pre-developed slides for information provision ensures a standardization of the minimum information provided by the moderator; the information sets are carefully selected to be appropriate for participants with no prior knowledge of genomics or the clinical applications being presented as case studies. The distinction between 'risk tests' [genomic] and 'diagnostic tests' is explained clearly to participants, emphasizing that the genomic case studies are risk tests.

Based on the evidence and considering the intervention and evaluation goals, key differences in approach for applying the workshop framework for professional versus public audiences were identified. The public workshop was designed to provide rapid, targeted education in order to inform deliberation about a topic about which participants had no direct experience, in fact, the specific topics for debate largely represented hypothetical uses of genetics. The key issue for professionals, in contrast, is to recognize that the technical applications may so far be hypothetical but the clinical scenarios in which they might be applied are very familiar. While the literature review revealed that to a large extent HCPs also lack foundational knowledge of genomics, current research on genetics education suggests that information provision is an inadequate educational goal, and that the key task is to convince professionals that the topic matter is relevant to their personal practice.

In response to this thinking, the stepped information provision component of the public engagement workshop was translated into three components in the professional workshop designed to underline relevance and frame deliberation in the context of existing practice: (1) an initial genomic educational component discussing the different visions for applying genomic information in primary care; (2) a selection of genomic screening tests and applications presented as case studies designed to establish linkages with clinical practice and highlight a range of issues in patient and practitioner decision-making (including practical, social, and ethical concerns); and (3) a ‘multi-strand’ data collection component in which the questions also prompt deliberation and engagement. The adapted professional engagement approach is shown in Figure 3; each component’s underlying mechanisms are described in the following sub sections.

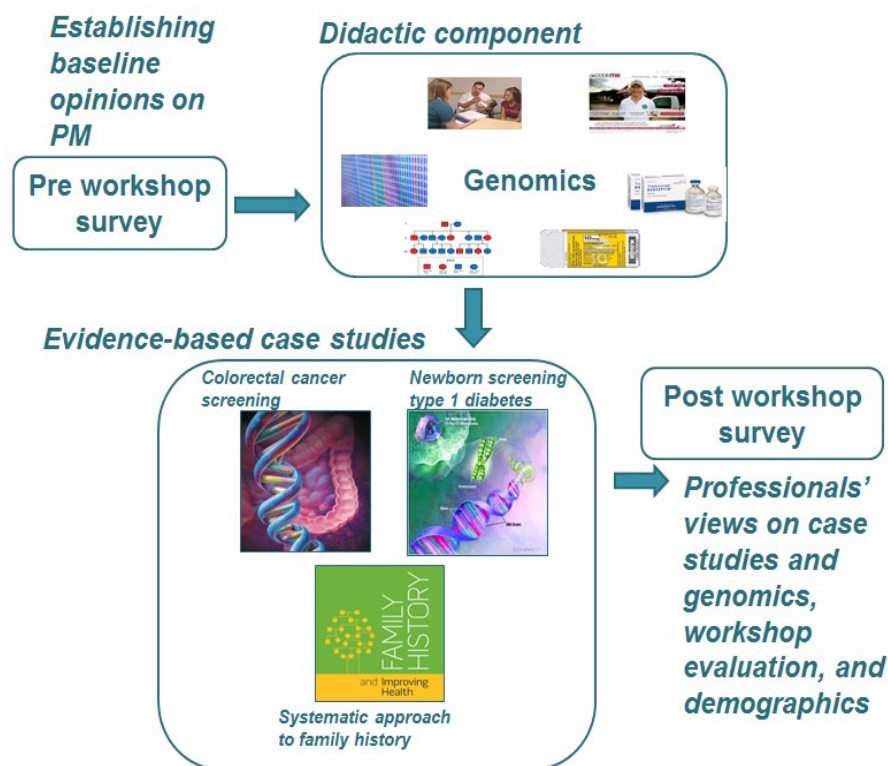


FIGURE 3 PROFESSIONAL ENGAGEMENT APPROACH

COMPONENT #1: EDUCATION

In order to base the educational component on sound educational principles, CC and BW consulted with Robert Parson, a consultant in curriculum development and quality of learning at the University of Ottawa's Centre for University Teaching. Based on his recommendations, we drew on a set of educational principles developed for '*integrated courses for significant learning*' (Fink, 2003). While these principles were developed for course creation, they provided useful guidance relevant to considering the initial design phase of any educational intervention. In addition, lessons learned from the first in-person workshop held at the Primary Healthcare Partnership Forum (PriFor) were applied when applicable.

In Fink's approach, the first step of the initial design phase is to gather information about situational factors; this includes context, nature of the subject and characteristics of the learners and teacher. Table 3 summarizes the situational factors relevant to the intervention, includes lessons learned from the first in-person workshop held at PriFor, and discusses the impact both had on the intervention design.

The next step in Fink's process involves defining the learning goals. The learning goals for the intervention, shown in Table 4, are classified based on Fink's 'Taxonomy of Significant Learning' which includes four different types of learning. By clearly defining the learning objectives, the intervention is designed with a focus on the key ideas, these are the points participants should understand and remember in the future.

TABLE 3 SITUATIONAL FACTORS AND IMPACT ON DESIGN

CONTEXTUAL ELEMENTS (FINK, 2003)	DESCRIPTION	LESSONS LEARNED FROM INITIAL PriFor WORKSHOP	IMPACT ON INTERVENTION DESIGN
Specific context of the learning situation	Approximately 5-10 participants per session.	Very hard to know actual numbers in advance, attendance at PriFor conference was much lower than anticipated.	Focus on expanding recruitment efforts.
	One session lasting ~1 hour (sessions will be repeated for different groups of participants).	Time frame is tight to cover all material.	Limit initial didactic teaching; the focus of the workshop should be on the interactive clinical case studies.
	In person focus group allowing interactive discussion.	Users were very engaged and keen to participate in interactive discussion.	Moderator needs to carefully time discussions for each case study and provide a list of questions that probe deeper into content to elicit participants' views.
	Online version will have a shorter time frame and no interactive component.	Online component might mitigate the challenges associated with recruiting professionals to live focus group type events.	Condense focus groups into shorter online workshop with educational component and case studies lasting no more than 10 minutes.
Nature of the subject	PM is an emerging and controversial topic.	For some, the content will inspire anxiety and trepidation about possibly integrating these ideas into their clinical practice.	Allay fears near the beginning, discuss possible benefits and highlight that PM is another form of risk stratification, which aligns with what HCPs are already doing in their clinical practice.
Characteristics of the learners	Professionals from different backgrounds.	Very heterogeneous group, participants with different levels of knowledge of PM.	Provide enough background for case studies that they can be understood by different clinical backgrounds.
Characteristics of the teacher	Clinical and epidemiological background. Strong knowledge base in PM.	Clinicians were comfortable having a clinician as the moderator. This appeared to be important for establishing credibility.	Emphasize impartiality of moderator, neither strong proponent nor cynic. Present moderator's credentials and background.

TABLE 4 LEARNING GOALS

TYPE OF LEARNING	LEARNING OBJECTIVES	KEY IDEA
Foundational knowledge	Understand the central idea of personalized medicine and how it can be applied for different purposes in clinical practice.	Understand that PM comprises many different applications.
	Recognize several commonly discussed approaches to risk stratification and prediction.	Understand that PM is another form of risk stratification.
Application	Appraise emerging evidence about personalized medicine applications.	Apply critical thinking to evaluate the benefits and challenges of implementing PM in clinical practice.
Integration	Participants relate selected personalized medicine examples to their own practice.	Making connections between workshop material and professional practice.
Human dimensions	Audience learns of other professionals' views during interactive dialogue	Learning from others.

The foundational knowledge section translated into the intervention's initial educational component providing the grounding for the clinical case studies, reflecting best current evidence and striving to be value neutral. First, participants were asked: 'What is genomics in health care?' – To answer this question PM was classified into six categories and an overview of each was provided, highlighting how these applications come together to form the current view of genomics in health care. The categories were: 1) whole genome sequencing, 2) genetic counseling, 3) direct to consumer testing, 4) pharmacogenomics, 5) companion diagnostics, and 6) family history. Lastly, the ACCE framework discussed in Chapter 1 was presented and used to assess the current evidence base for each of these applications.

The application and integration component of the learning are centered on the clinical case studies described in more detail in the following section. The case studies were designed to highlight the salience to clinical practice and raise critical issues about implementation: i.e. Do you think this is a good idea? Do you think your patients would like this? What are the

implications for practice? What would persuade you to use this? What would put you off? These questions are then explored in open discussion during the in-person workshop.

COMPONENT #2: EVIDENCE-BASED CASE STUDIES

Three topics were selected for the clinical case studies: (1) genomic profiling of the general population to improve colorectal cancer risk (CRC) stratification; (2) genomic profiling of newborns to identify infants most susceptible to type 1 diabetes; and (3) a standardized approach to family history taking. These were chosen as timely topics relevant to HCPs, and distinctly different versions of the PM ‘idea’ in clinical practice.

The first two topics represent as yet hypothetical tests that are applicable to two age extremes of the target screening population and are likely to provoke different reactions on the parts of HCPs.

Colorectal cancer is the commonest cancer in relative terms and stage 1 is highly curable with 95% 5-year survival (Canadian Cancer Society, 2011). Unfortunately many people present in later stages where the 5 year survival is much lower. While there is currently no genomic risk profiling test for CRC, there is hope that combining predictive knowledge from individual genetic variants (Dunlop et al., 2012; Hawken et al., 2010) could ‘*provide important information about disease susceptibility*’ (Nicholls et al., 2013) and that enhanced surveillance could be advised for patients with high risk genomic results. This hypothetical test is not cancer detection and it would not replace current fecal occult blood test screening; rather this would be using genomics for better risk stratification.

The incidence of type 1 diabetes is increasing internationally (2-5% annually) with suggestions that rates in young children could double by 2020 (Vehik & Dabelea, 2011). Research into genetic and environmental triggers for type 1 diabetes is active, with many

international studies exploring risk susceptibility testing in infants (Kerruish et al., 2007). There is currently no intervention that would prevent type 1 diabetes; however, because early clinical symptoms are non-specific there is potential clinical utility in identifying children at increased risk, such as providing parental education and promoting enhanced surveillance in primary care. Researchers also suggest that newborn screening to identify infants at elevated genetic risk could help identify environmental triggers and lead to prevention strategies (Hagopian et al., 2006, 2011)

Family history can be considered the ‘first genetic test’, as explained by Chan: *‘Reflecting the complex combination of shared genetic, environmental, and lifestyle factors, a robust family health history can approximate genetic/genomic risk information and integrate it into patient care’* (Chan & Ginsburg, 2011). A more systematic approach to family history taking is relevant to the entire patient population and is already being applied to some extent in selected clinical practices so should be familiar to many HCPs, although the system described in the case study goes beyond what is currently available. The case study presents a standardized approach to family history data collection, incorporating clinical alerts and integration of family history information into the electronic medical record and linkages with information from other family members. This information could then be linked with risk stratification guidelines to alert HCPs to areas of elevated risk, and clinical guidelines could recommend appropriate surveillance and prevention options.

These three topics were extensively explored in the public engagement workshops (Nicholls et al., 2013), which will allow for future comparisons of citizen and professional perspectives on the same topics (although this comparison is outside the scope of this thesis project). For the professional engagement workshop, the public engagement scenarios were

modified to focus on clinical validity and utility; the goal was not to explain the genetic technology behind the testing but rather to discuss how the test might be used in practice. A standardized format was used for presenting all three case studies, described here for the CRC case study.

First, the participants were asked the hypothetical question: ‘What if we could offer patients a genomic test to profile their risk of colorectal cancer?’ Next, a brief overview of the epidemiology of the problem was presented followed by a discussion of how colorectal cancer risk is currently profiled. Then participants were shown a schematic explaining how genomic screening could be integrated and used in clinical practice. The point was made that genomic screening would not replace the current approach but would offer better advance risk stratification and that HCPs could then offer risk reduction advice based on guidelines, available resources, and their patient’s own preferences.

COMPONENT #3: ENGAGEMENT

The third component relates to the engagement portion of the intervention which includes questions that prompt deliberation and engagement; the engagement approach also guided the subsequent evaluation in Phase 2.

The Theoretical Domains Framework (TDF) was used to guide the engagement elements that pertained to level 2a of Kirkpatrick’s educational topology. Level 2a considers HCPs’ attitudes towards the utility of genomics and whether these change over the course of the workshop. We developed the education and engagement components of the intervention cyclically in that considering how we would apply the TDF to engage about HCPs’ attitudes and behaviors towards PM influenced how the information was presented in the case studies.

The TDF is an integrative framework of theories of behavior change; it classifies 14

domains that effect behavior and was designed to assess the barriers and facilitators of a behavior change; it can be used to inform intervention design, for both data collection and qualitative data analysis (Cane et al., 2012). The TDF is well suited to this study, because as highlighted by the literature review, integrating PM into clinical practice will require changes in HCP's behaviors in clinical practice.

Use of the TDF has been shown to elicit more beliefs, barriers and facilitators than in studies not based on theory (Francis et al., 2012); however it requires that domains are interpreted according to the behaviour being studied. Seven of the fourteen TDF domains were identified as being relevant to studying HCPs' behaviors towards integrating PM into primary care: knowledge, skills, social/professional role and identity, beliefs about capabilities, beliefs about consequences, intentions, and environmental context and resources. Table 5 shows how these domains were interpreted in the context of PM and then mapped to level 2a engagement questions. The TDF was also applied to develop coding schemes for the qualitative analysis of textual data (see Chapter 5, Results).

TABLE 5 TDF DOMAIN MAPPING TO SURVEY QUESTIONS

TDF DOMAIN	INTERPRETATION IN THE CONTEXT OF PM	TRANSLATION TO ENGAGEMENT QUESTIONS USED IN THE PE INTERVENTION
Knowledge An awareness of the existence of something	What does genomics mean to HCPs?	This is what genomics in health care means to me <i>(with a list of options provided)</i>
Skills An ability or proficiency acquired through practice	When do HCPs currently take family history? How do HCPs integrate family history taking skills into practice?	In what kinds of situations do you actively ask a patient about his or her family history? <i>(with a list of options provided)</i> What kind of approach do you use to capture family history information? <i>(with a list of options provided)</i>
Social/Professional Role and Identity A coherent set of behaviours	Do HCPs believe ordering and interpreting genomic tests is part of their primary care role	If a properly validated genomic profiling test for [colorectal cancer/type 1 diabetes] became available... I think

TDF DOMAIN	INTERPRETATION IN THE CONTEXT OF PM	TRANSLATION TO ENGAGEMENT QUESTIONS USED IN THE PE INTERVENTION
and displayed personal qualities of an individual in a social or work setting	in patient support? Would patients expect them to provide these services?	most patients would expect me to make them aware of this test
Beliefs about Capabilities Acceptance of the truth, reality, or validity about an ability, talent, or facility that a person can put to constructive use	Do HCPs believe they are capable of ordering and interpreting genomic tests?	If a properly validated genomic profiling test for [colorectal cancer/type 1 diabetes] became available... I think family physicians should be capable of ordering it
Beliefs about Consequences Acceptance of the truth, reality, or validity about outcomes of a behaviour in a given situation	What is HCPs opinion about the clinical utility of genomics in their practice? Do HCPs believe that in this clinical situation better risk stratification would impact their behaviour? Do HCPs believe that in this clinical situation better risk stratification would impact patient care?	In my opinion, genomics will be useful in my clinical practice. Irrespective of the nature of the test, being able to more accurately quantify my patients' risk of [colorectal cancer/type 1 diabetes] would make a difference to my clinical assessment. If a properly validated genomic profiling test for [colorectal cancer/type 1 diabetes] became available, I think it would generally help/harm patients.
Intentions A conscious decision to perform a behaviour or a resolve to act in a certain way	Would HCP be willing to recommend genomic tests to patients?	If a properly validated genomic profiling test for [colorectal cancer/type 1 diabetes] ... I would recommend it to patients.
Environmental Context and Resources Any circumstance of a person's situation or environment that discourages or encourages the development of skills and abilities, independence, social competence, and adaptive behaviour	What resources would facilitate the use of genomic tests in clinical practice? What resources would facilitate the use of standardized family history taking in clinical practice?	If properly validated genomic tests for chronic disease risk or to guide prescribing became available... which of the following would help you work out how to use them in your practice? <i>(with a list of options provided)</i> If a standardized approach to family history taking was recommended, which of the following would help you integrate it into your practice? <i>(with a list of options provided)</i>

3.4 SUMMARY OF THE TWO ENGAGEMENT APPROACHES

IN-PERSON WORKSHOP

This approach took the form of a workshop which could be presented within a conference

setting, continuing medical education, or free-standing. In a plenary format, a facilitator led participants through three components. During Part 1 (~20 minutes), participants received a didactic presentation to establish a baseline knowledge of PM. Part 2 (~40 minutes) was an audience engagement component where a set of scenarios illustrating realistic possibilities for PM applications in general primary care practice settings were presented. Part 3 of the workshop comprised a ‘multi-thread’ data collection exercise to capture participants’ reactions, thoughts, and attitudes towards PM while the workshop progressed. The data were captured in several ways. The first method was through short facilitated discussion pauses following the presentation of each scenario, in which questions designed to elicit participants’ reactions to PM and their immediate assessment of its utility to their own practice were asked. The audience was invited to ask questions and discuss issues which emerge – essentially a brief focus group. Field notes were captured by non-participant observers. The second method was through participants recording, throughout the entire workshop, personal observations, comments and thoughts in an open text format in a booklet. The third method was before and after self-completion surveys designed to elicit a broader set of data on experience with and attitudes towards PM applications. Thus, three types of engagement data were available for analysis 1) content analysis of the field notes; 2) quantitative analysis of the final demographic and attitude survey; and 3) content analysis of the unstructured written comments.

ONLINE WORKSHOP

The online workshop replicated many aspects of the in-person structured workshop, with two important exceptions: there was no moderator and therefore no possibility of clarification or explanation; and there was no group process to prompt deliberation or ‘surface’ ideas which might not occur to an individual. In the absence of a physical moderator, the survey engagement

questions became an integral component of the intervention and were interleaved between the case studies to prompt participant deliberation.

The detailed breakdown of the online workshop was as follows. After completing a short pre-workshop survey (~1 minute), participants watched a video presentation (~7 minutes) providing an overview of genomics in health care. Next, a set of three short videos illustrating realistic possibilities for PM applications in general primary care practice settings was presented (each video lasted approximately 1 minute). Each case study was immediately followed by a set of questions designed to elicit participants' reactions to PM and their immediate assessment of its utility to their own practice. During each video presentation, participants were asked to record personal observations, comments and thoughts in an open text box. Finally, participants were asked to complete a brief post workshop survey designed to elicit a broader set of data on experience with and attitudes towards PM applications, and professional demographic data. All survey questions were identical to the in-person workshop, the only difference being the time within the workshop that the questions were presented; for the in-person version all survey data collection occurred at the end of the workshop whereas for the online version the survey questions were part of the intervention and embedded throughout the content.

The online workshop was designed taking into account the target audience of HCPs and their expectations and experiences with online interventions. Using the same approach made popular by health media experts such as Dr. Mike Evans with his 'Whiteboard Med School' and now well-known video '23 and ½ hours' (Evans, 2014), we used narrated video clips with line drawing animation and text to convey the same key points as the information sets delivered by the workshop facilitator during the in-person workshop. Based on discussion with Dr. Doug Manuel, who has developed widely used online health calculators for life expectancy and salt

intake (Manuel, 2014), we learned that less than half of the people who ‘land’ on a survey actually complete it and to increase completion rates online surveys should be as concise as possible. Because the online intervention involves both an educational and survey component, the time available for education is significantly reduced in the online version, with all videos lasting just over 10 minutes, as compared to over 30 minutes of presentation for the in-person workshop.

The development of the online survey involved the independent production and integration of (a) video animation, (b) audio recording, and (c) the online survey questionnaire. Videos were developed using Video Scribe (<http://www.sparkol.com/>); this software allows non-professionals to create animated videos (also called Whiteboard animation or Fast Drawing) and is a popular way of engaging with audiences online. Figure 4 shows an example of the type 1 diabetes case study, zoomed out to fit all elements.

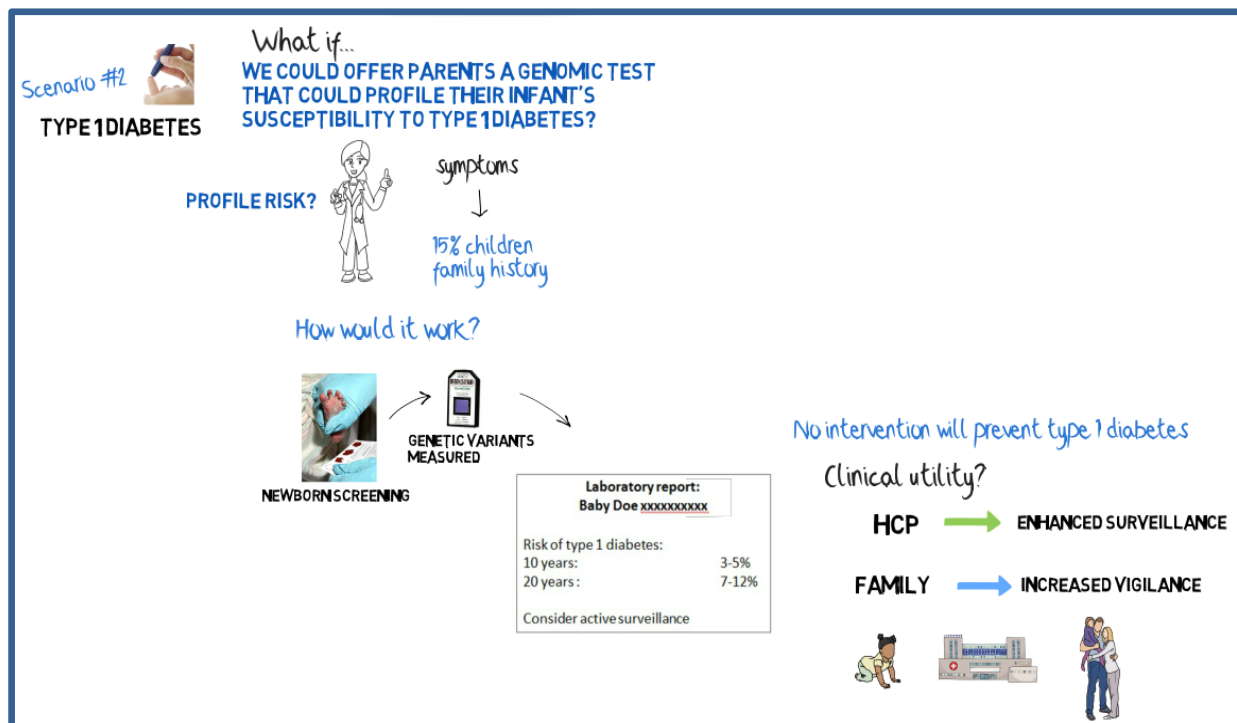


FIGURE 4 ONLINE VERSION OF DIABETES SCENARIO

Table 6 illustrates the mapping between Craig’s dimensions of complexity (Craig et al., 2008) and the PE interventions.

TABLE 6 DIMENSIONS OF COMPLEXITY

DIMENSION OF COMPLEXITY	LINK WITH THESIS INTERVENTION
Number of components and interactions between components within the intervention	The intervention has three interacting components: 1) an initial educational component to establish foundational knowledge; 2) case studies to establish connections with clinical practice and prompt critical thinking about integration; 3) professional engagement, either moderator led or driven by scripted online questions, pertaining to the information presented in 1) and 2).
Number and difficulty of behaviors required by those delivering or receiving the intervention	The person delivering the intervention (i.e. the moderator) acted as both an educator and facilitator. To obtain valid data the moderator had to provide unbiased educational content and prompt discussion while maintaining a neutral perspective. During the online version of the intervention there was no moderator to prompt deliberation and this process had to be replicated as closely as possible through targeted questions embedded in the online content.
Number of groups or organizational levels targeted by the intervention	The intervention targeted a broad group of health care professionals involved in primary care with varying degrees of baseline knowledge about genomics; the target group included: nurses, nursing students, family physicians, residents, and medical students.
Number and variability of outcomes	A range of outcomes, both quantitative and qualitative, were obtained to evaluate the effectiveness of the interventions. Data was also collected pertaining to HCPs’ views on integrating genomics in health care.

3.5 PHASE 2: EXPLORATORY TRIAL

We designed an exploratory trial to gather further insight relevant to the design and delivery of the interventions, to obtain general quantitative estimates of the primary and other outcomes, and to gather data relevant to the design and implementation of a potential future definitive evaluation.

This pilot study used a non-randomized, non-equivalent group design in which participants were allocated to the two interventions in a pragmatic fashion. Neither intervention was considered ‘standard’ because they were both novel. The design was driven by significant

practical constraints, not least the early recognition that some form of randomized design would be impossible to implement within the time and resources available for a master's level thesis.

PARTICIPANTS AND RECRUITMENT

Eligible participants were all physicians and nurses in Canada, including medical students, residents and nursing students. The decision to include nurses was made after the first exploratory workshop, which was the point at which the physician recruitment challenge became clear. We also justified it on the basis of the literature review, which indicated that genomics is increasingly relevant to all nursing practice. Physician and nurse participants were recruited to separate in-person workshops because previous research has indicated that learners may be reluctant to participate in multi-professional groups fearing they would be put in a situation *'where their lack of knowledge may be revealed'* (Burke et al., 2007).

We used recruitment strategies designed to maximize participation. For both arms: 1) dissemination through the investigators' professional networks; 2) using opinion leaders to advertise the study within their own professional groups; and 3) advertising at major professional conferences. In addition, for the in-person intervention, we sought to negotiate the workshop's inclusion in existing educational activities such as courses or conferences, and accreditation of the workshop for professional continuing education credits.

For the in-person intervention, interested participants were informed of the event details by e-mail, and, received the formal letter of invitation, participant information form and consent form either by e-mail in advance or on arrival at the workshop. Recruitment for the in-person workshop was ongoing between November 2012 and February 2014.

For the online intervention, potential participants received an e-mail letter of invitation including the website link, information on the estimated completion time, and an attached

participant information form. Consent was obtained through the survey main page, which specified that participation was voluntary and that participants could choose to stop the workshop at any time. The online workshop was open for completion for a period of 5 months, between January 2014 and May 2014. Section 4.6 presents the recruitment results.

With the exception of a small reimbursement of transportation costs for students attending one in-person workshop, no incentives were provided for participants. Appendix 1 includes all letters of invitation, participant information forms and consent forms for the in-person and online version of the workshop. Appendix 2 contains letters of approval from the Ottawa Health Science Network Research Ethics Board and the Newfoundland Health Research Ethics Authority.

OUTCOMES

There are no standardized outcome measures for public or professional engagement in the published literature. We therefore developed outcomes designed to offer insight into the intervention overall, and aspects of engagement with PM.

The primary outcome was level of agreement, on a five point Likert scale, to the statement: ‘This workshop was an effective way for me to express my views on personalized medicine as it might relate to my own clinical practice [or nursing practice]’. The scale used was: 1=strongly agree, 2=agree, 3=neutral, 4=disagree, and 5=strongly disagree.

The secondary outcomes addressed further questions about the value of the intervention. These items were also presented as extent of agreement on a five point Likert scale with the following statements:

- 1) The workshop was an effective way for me to think about genomics as it pertains to my clinical/nursing practice

- 2) I would participate in a similar workshop in the future
- 3) The workshop raised new ideas
- 4) The workshop provided new information
- 5) The workshop presented information in an unbiased way
- 6) There was enough time for deliberation about the issues

The choice of questions was influenced by the review of the professional engagement literature, and the goal was to focus on attributes that would distinguish participants' evaluation of the two engagement approaches.

ENGAGEMENT DATA

As described above, the design of both versions of the intervention included data collection, which, for clarity, we term 'engagement data'. These items were not developed for the purposes of the evaluation; they were inherent in the engagement design. However, they provide further insight into the performance of the interventions, in that it would be expected, in a completely unbiased comparison, that they would be essentially identical irrespective of the in-person or online approach to the intervention. These items addressed (a) general attitudes to PM as part of personal practice, and (b) specific attitudes to the hypothetical examples presented in the case studies. They were combined into a questionnaire presented in seven sections, as shown in Table 7. To facilitate ease of survey completion the same format was used as much as possible for the colorectal cancer, type 1 diabetes and family history case studies (sections 2-4), where the first question in each section pertained to the overall clinical utility of the test, irrespective of whether the test was based on genomics, with subsequent questions pertaining to the relevant TDF domains identified in Table 5.

TABLE 7 SURVEY COMPONENTS

SECTION	SURVEY COMPONENT
1	Pre-work shop survey – utility of genomics
2	Post-workshop survey – colorectal cancer case study
3	Post-workshop survey – diabetes case study
4	Post-workshop survey – family history case study
5	Post-workshop survey – utility of genomics
6	Workshop evaluation
7	Demographics

Slight changes to the survey wording were made to reflect the different roles of physicians and nurses in patient care. Using the colorectal cancer case study as an example, Table 8 shows the differences in wording between the two versions of the questionnaire. Apart from these differences, the survey questions and format were identical for the nursing and physician versions.

TABLE 8 MODIFICATIONS IN WORDING BETWEEN PHYSICIAN AND NURSING VERSIONS OF THE SURVEY

NURSING VERSION	PHYSICIAN VERSION
Irrespective of the nature of the test, being able to more accurately quantify my patients' risk of colorectal cancer would make a difference to my clinical <u>assessment</u> .	Irrespective of the nature of the test, being able to more accurately quantify a patient's risk of colorectal cancer would make a difference to my clinical <u>decision-making</u> .
If a properly validated genomic profiling test for colorectal cancer became available... I think most patients would expect me to <u>make them aware</u> of this test	If a properly validated genomic profiling test for colorectal cancer became available... I think most of my patients would expect me to <u>recommend it</u>
If a properly validated genomic profiling test for colorectal cancer became available... <u>With training</u> , I think nurses would be capable of <u>supporting patients' informed decision making about ordering and interpreting this test</u>	If a properly validated genomic profiling test for colorectal cancer became available... I think family physicians should be <u>capable of ordering it</u>

NURSING VERSION	PHYSICIAN VERSION
If a properly validated genomic profiling test for colorectal cancer became available...	If a properly validated genomic profiling test for colorectal cancer became available...
<u>If parents asked for my advice</u> , I would recommend this test	<u>I would recommend</u> it to patients

DATA COLLECTION

Quantitative data

A brief pre-intervention survey instrument was developed which captured baseline general attitudes to PM (engagement data). This was administered immediately before the in-person intervention, and at the start of the online intervention. The post-intervention survey instrument captured the full set of engagement data (including repetition of the baseline items), along with the primary and secondary evaluation outcomes, and demographic data.

Qualitative data

Free-text comments were captured in participant workbooks distributed at the beginning of the in-person intervention, accompanied by the facilitator's instruction to note down any comments, thoughts or views as the workshop progressed. For the online intervention, text boxes were inserted at multiple points, inviting participants to offer comments. For the in-person intervention only, non-participant observers captured comments made by participants in field notes. The in-person version of the data collection instrument is included in Appendix 3.

QUANTITATIVE ANALYSIS

The answers to all survey questions were summarized using descriptive statistical techniques. Descriptive statistics were used to describe the baseline characteristics of the study participants and to characterize all structured survey responses using SAS 9.3.

In evaluating the interventions, we wished to determine whether any observed differences in the primary outcome or other evaluation scores might be due to actual differences between the

interventions or attributable to other confounding factors. With the two volunteer samples, we recognized the limitations to internal validity by the non-randomized allocation and anticipated differential distribution of HCPs between intervention groups. Without any intention to generalize results to the target population of Canadian HCPs, we developed an analysis strategy to compare the outcome data for the two intervention arms to determine whether the two samples are similar enough to make useful comparisons about the interventions. To address this problem we planned a multi-step analysis approach:

- Present descriptive comparisons using proportions for the primary outcome and all evaluation scores, and perform the analysis for all HCPs and grouped by profession (physicians and nurses).
- Analyze the evaluation scores statistically using a non-parametric comparison of medians; this was appropriate because the survey questions were based on Likert response items to be analyzed individually (rather than a psychometric scale). Anticipating a small sample size, assumptions about central tendency might not necessarily be applicable. We planned to compare medians using the Mann-Whitney U rank sum test which is appropriate when categories are logically ordered according to magnitude (i.e. Likert-scale responses) (Hollingsworth et al., 2011). We planned the analysis across all HCPs and grouped by HCP with examination of any differences in results.
- Use logistic regression to further explore the effects of confounding; possible confounding variables would include health care profession, age, and years in practice. If appropriate, fit a generalized linear model to further explore the effects of confounding by profession, with evaluation score as the dependent variable and

intervention format (online or in-person) and health care profession (physician or nurse) as the independent variables.

QUALITATIVE ANALYSIS

For the qualitative analysis of the field notes and free text comments, the constant comparison method was used to identify the nature and extent of the issues and themes emerging from the intervention. A line-by-line thematic analysis was done where data were coded in an inductive manner and then grouped into themes through a process of data reduction, as described by Nicholls et al. (Nicholls et al., 2013). The goal was to identify important words and concepts that formed an inclusive set of codes, which were reduced and grouped into key themes and categories, and relationships between themes were identified. The Theoretical Domains Framework (TDF), applied previously to guide the development of the survey instrument, was used as a basis for developing coding schemes for the qualitative analysis. In addition, the themes were reviewed to quantify the number of distinct concepts and categorize the type of concepts that are represented to allow for comparison between the in-person and online version of the intervention.

SAMPLE SIZE

Although definitive sample size calculations are generally not performed in a pilot study, it was decided to approach the evaluation as a superiority trial with the null hypothesis being that the two interventions are not different in terms of the primary outcome and the alternative hypothesis being that they are different. For the purpose of analysis the in-person workshop was considered the standard intervention and the online workshop is the alternative intervention. The minimal clinically important difference (MCID) is then the smallest difference in median value for the primary outcome that is perceived as beneficial and would lead to a change in the choice

of PE intervention.

In the absence of any empirical data relevant to this study, the MCID was determined pragmatically considering end knowledge users, and what a difference in median score might mean in practice. Bearing in mind that the primary outcome was based on a 5 point Likert response item where a difference of 1 point can shift the classification from a positive to neutral response (i.e. 2 to 3) or a neutral to negative response (i.e. 3 to 4), it was decided that a difference of 1 point in the median score would be sufficient to conclude that one intervention is superior to the other. From the knowledge users' perspective, the online intervention might represent an easier, more convenient and less expensive approach to engagement, so it is reasonable that the MCID could be small. The sample size calculation was based on two sided tests because there were no pre-determined hypotheses for the pilot and it was not known which approach would be most acceptable to HCPs. Group sample sizes of 25 per arm achieve 92% power to reject the null hypothesis of equal medians when the population median difference is 1 with a standard deviation for both groups of 1 with a significance level (alpha) of 0.05 using a two-sided Wilcoxon/Mann Whitney U test. Power calculations were performed using SAS's power procedure for the two sample Wilcoxon test.

ETHICS

No personal identifiers were collected for either arm of the intervention. For the in-person version participants were allocated a unique code; the signed consent forms, which include participants' names, were held securely in a separate location. The online survey did not collect internet provider addresses and no identifying information was collected during the consent process. Ethics approval was received from the Ottawa Health Science Network Research Ethics Board and the Newfoundland Health Research Ethics Authority.

CHAPTER 4. EVALUATION RESULTS

This chapter presents the evaluation results divided into five sections. In Section 4.1 we describe the participants and professional demographics. Section 4.2 presents an assessment of the similarities between health care professionals based on the pre-intervention survey data. The primary and supporting outcomes are presented in Section 4.3, followed in Section 4.4 by a comparison of the pre- and post-survey results. Section 4.5 compares the two interventions based on the number of overarching themes identified from the qualitative data. The chapter concludes with a comparison of recruitment and other logistics in Section 4.6.

4.1 PARTICIPANTS & PROFESSIONAL DEMOGRAPHICS

A total of 64 HCPs completed the workshop, with 34 in-person and 30 online participants. Table 9 examines professional demographics overall and by HCP. The mean participant age was 42 years; nurses were younger (mean 39 years) as compared to physicians (mean 48 years). The majority of the sample population was female (83%), driven by the fact that 98% of nurses were female; however, sex was split evenly in the physician sample. The sample represented a mix of experience levels with the largest proportion of participants responding that they had ≥ 20 years in clinical practice (34%) and the next largest proportion choosing '0 to 4' years (22%); years in practice was not significantly different between nurses and physicians. Overall, HCPs served largely urban populations (72%) and the distribution was similar between nurses and physicians. Overwhelmingly, participants had no previous training in genomics (95%). While physicians were slightly more likely to have previous training than nurses (90% as compared to 98%), it is clear that the great majority of the population had no previous genetic training. Table 9 shows that based on our professional demographics, age and sex are the only significant differences between health care professional groups.

TABLE 9 SAMPLE CHARACTERISTICS BY HEALTH CARE PROFESSION

		All HCPs N=64		By HCP				p
				Nurse N=44		Physician N=20		
		n	mean / %	n	mean / %	n	mean / %	
Age		41.5 (SD 13.6)		39.2 (SD 12.8)		47.5 (SD 14.3)		0.0012
Sex*								<.0001
male	10	15.6	0	0.0	10	50.0		
female	53	82.8	43	97.7	10	50.0		
missing	1	1.6	1	2.3	0	0.0		
Years in practice								0.6014
0 to 4	14	21.9	9	20.5	5	25.0		
5 to 9	10	15.6	7	15.9	3	15.0		
10 to 14	10	15.6	8	18.2	2	10.0		
15 to 19	5	7.8	5	11.4	0	0.0		
> =20	22	34.4	13	29.5	9	45.0		
Not in practice	2	3.1	1	2.3	1	5.0		
Missing	1	1.6	1	2.3	0	0.0		
Population								0.9146
Urban	46	71.9	31	70.5	15	75.0		
Small urban (10,000-25,000)	5	7.8	4	9.1	1	5.0		
Rural (<10000)	6	9.4	4	9.1	2	10.0		
Not involved in patient care	5	7.8	4	9.1	1	5.0		
missing	2	3.1	1	2.3	1	5.0		
Special training in genetics								0.2280
yes	3	4.7	1	2.3	2	10.0		
no	61	95.3	43	97.7	18	90.0		
SD: Standard deviation								

Table 10 breaks down HCP by specialty. Most nurses who participated in the workshops classified themselves as registered nurses (71%) and most physicians were primary care physicians (70%). From a training perspective 11% of nurses were students and 20% of physicians were residents.

TABLE 10 HCP SPECIALTIES

Nursing Designation (N=44)	n	%	Physician Designation (N=20)	n	%
Registered Practical Nurse	3	6.8	Primary care physician	14	70.0
Registered Nurse	31	70.5	Specialist physician	2	10.0
Registered nurse, extended class	2	4.5	Resident	4	20.0
Student nurse	5	11.4			
Other (Nurse choice)	2	4.5			
Missing	1	2.3			

4.2 ASSESSMENT OF SIMILARITIES BETWEEN PROFESSIONAL GROUPS

We compared pre-intervention survey results between nurses and physicians in order to make a judgment about whether the evaluation results could be combined over all HCPs in the subsequent analyses (Sections 4.3-4.5).

The pre-intervention survey questions established participants' baseline views on genomics, including: their understanding of PM, their perception of the clinical utility of risk stratification for colorectal cancer and type 1 diabetes regardless of whether these tests were implemented using genomics, and their views on the usefulness of family history in clinical practice.

UNDERSTANDING OF PERSONALIZED MEDICINE

The first pre-survey question was aimed at understanding what the term PM meant to participants. From a list of 7 options participants were asked to select all that applied, Table 11 shows the breakdown of responses as percentages of total responses – 64 respondents made 227 different selections for an average of 3.5 selections per respondent. Stratifying risk of disease represented 24% of total responses; screening 22%; family history taking 21%; making a treatment or prognostic decision were each 15%; 7% chose the 'other' category. Considering the

responses by health care profession did not change the order of these selections and the distribution of responses was very similar for both groups. For participants who selected ‘other’, responses included: gene testing, tailoring patient education to individual risk, patient centeredness, newborn screening and family planning.

TABLE 11 ‘THIS IS WHAT PERSONALIZED MEDICINE MEANS TO ME’

	All HCPs		By HCP				p (Fisher's)
	responses=227		Nursing		PCP		
			155		72		
	n	%	n	%	n	%	
Taking a family history	47	20.7	34	21.9	13	18.1	0.9191
Screening	50	22.0	36	23.2	14	19.4	
Stratifying risk of disease in a healthy patient	54	23.8	36	23.2	18	25.0	
Making a treatment decision	35	15.4	22	14.2	13	18.1	
Making a prognostic decision	34	15.0	22	14.2	12	16.7	
Other	7	3.1	5	3.2	2	2.8	

CLINICAL UTILITY OF PM

Table 12 shows the response from all HCPs to the question ‘*In my opinion personalized medicine will be useful in my clinical practice*’: 53% of participants strongly agreed or agreed with this statement, 32% were neutral and 15% disagreed or strongly disagreed. Considering nurses’ and physicians’ responses showed that the distributions were very similar across HCP with 55% of nurses and 50% of physicians strongly agreeing or agreeing; 31% and 35% neutral; and 14% of nurses and 15% of physicians disagreeing or strongly disagreeing.

TABLE 12 'IN MY OPINION PERSONALIZED MEDICINE WILL BE USEFUL IN MY CLINICAL PRACTICE'

	All HCPs		By HCP				p (Fisher's)
	N=62		Nursing		PCP		
			N=42		N=20		
	n	%	n	%	n	%	
Strongly agree	13	20.97	10	23.8	3	15.0	0.9037
Agree	20	32.26	13	31.0	7	35.0	
Neutral	20	32.26	13	31.0	7	35.0	
Disagree	7	11.29	5	11.9	2	10.0	
Strongly disagree	2	3.23	1	2.4	1	5.0	

CLINICAL UTILITY OF CRC RISK STRATIFICATION

Table 13 considers responses to the question pertaining to the clinical utility of CRC screening regardless of how the test is implemented: 63% of respondents strongly agreed or agreed; 23% were neutral; and 15% disagreed or strongly disagreed. From the perspective of HCP groups, the distribution of responses was very similar.

TABLE 13 'IRRESPECTIVE OF THE NATURE OF THE TEST, BEING ABLE TO MORE ACCURATELY QUANTIFY A PATIENT'S RISK OF COLORECTAL CANCER WOULD MAKE A DIFFERENCE TO MY CLINICAL DECISION-MAKING.'

	All HCPs		By HCP				p (Fisher's)
	N=62		Nursing		PCP		
			N=42		N=20		
	n	%	n	%	n	%	
Strongly agree	10	16.13	8	19.0	2	10	0.6961
Agree	29	46.77	17	40.5	12	60	
Neutral	14	22.58	10	23.8	4	20	
Disagree	6	9.68	5	11.9	1	5	
Strongly disagree	3	4.84	2	4.8	1	5	

CLINICAL UTILITY OF TYPE I DIABETES RISK STRATIFICATION

Table 14 displays views about the clinical utility of screening for type I diabetes irrespective of the nature of the test. Overall 53% of participants strongly agreed or agreed that there is clinical utility in being able to type 1 diabetes risk; 26% were neutral; and 21% disagreed or strongly disagreed. Responses were very similar by HCP.

TABLE 14 'IRRESPECTIVE OF THE NATURE OF THE TEST, BEING ABLE TO IDENTIFY INFANTS AT HIGHER THAN AVERAGE RISK OF TYPE 1 DIABETES WOULD MAKE A DIFFERENCE TO MY CLINICAL DECISION-MAKING.'

	All HCPs		By HCP				P (Fisher's)
	N=61		Nursing		PCP		
	n	%	N=41	%	N=20	%	
Strongly agree	10	16.4	9	22.0	1	5	0.5136
Agree	22	36.1	14	34.1	8	40	
Neutral	16	26.2	10	24.4	6	30	
Disagree	9	14.8	6	14.6	3	15	
Strongly disagree	4	6.6	2	4.9	2	10	

CLINICAL UTILITY OF FAMILY HISTORY TAKING

Table 15 considers the utility of family history information in clinical practice. Overall 82% of participants strongly agreed or agreed with this statement; 7% were neutral; and 11% disagreed or strongly disagreed. Based on HCP 78% of nurses and 90% of physicians strongly agreed or agreed with this statement; 7% of nurses and 5% of physicians were neutral; and 15% of nurses and 5% of physicians disagreed or strongly disagreed.

TABLE 15 ‘ON THE WHOLE, I FIND FAMILY HISTORY INFORMATION TO BE A USEFUL TOOL IN EVERYDAY PRACTICE’

	All HCPs		By HCP				P (Fisher's)
	N=61		Nursing		PCP		
			41		20		
	n	%	n	%	n	%	
Strongly agree	21	34.4	16	51.6	5	25	0.3595
Agree	29	47.5	16	51.6	13	65	
Neutral	4	6.6	3	9.7	1	5	
Disagree	4	6.6	3	9.7	1	5	
Strongly disagree	3	4.9	3	9.7	0	0	

SUMMARY OF PRE-SURVEY RESULTS

Based on these tentative analyses, the two professional groups looked generally similar in their baseline attitudes. Noting the limitations inherent in this small pilot study, the judgment was made to consider them one population for the purposes of comparing the interventions (Section 4.3-4.5) and presenting the ‘substantive’ findings about genomics (Chapter 5).

4.3 EVALUATION OF PRIMARY OUTCOME AND SUPPORTING OUTCOMES

PRIMARY OUTCOME

The primary outcome was response to the question: ‘This workshop was an effective way for me to express my views on genomics as this area might relate to my clinical/nursing practice.’ Table 16 shows that 79% of in-person participants strongly agreed or agreed with this statement as compared to 62% of online participants; very few people evaluated either intervention negatively. The frequency distributions indicate that the in-person workshop was considered the more effective professional engagement approach based on the primary outcome.

TABLE 16 COMPARISONS ACROSS ALL HCPS FOR PRIMARY OUTCOME: ‘THIS WORKSHOP WAS AN EFFECTIVE WAY FOR ME TO EXPRESS MY VIEWS ON GENOMICS AS THIS AREA MIGHT RELATE TO MY CLINICAL/NURSING PRACTICE.’

	Workshop format				p (Fisher's)
	In-person		Online		
	34		29		
	n	%	n	%	
Strongly agree	15	44.1	3	10.3	0.0091
Agree	12	35.3	15	51.7	
Neutral	6	17.6	9	31.0	
Disagree	0	0.0	2	6.9	
Strongly disagree	1	2.9	0	0.0	

SUPPORTING OUTCOMES

To further compare the interventions we collected data on six other supporting participant satisfaction measures, with the results presented below.

Table 17 shows responses to the question: ‘This workshop was an effective was for me to think about personalized medicine in my own clinical practice’. The results show that 91% of in-person respondents strongly agreed or agreed that the in-person workshop was an effective way for them to think about genomics in clinical practice as compared to 80% for the online workshop. For both interventions the overwhelming majority gave positive rankings; however, the in-person had more positive and fewer neutral responses.

TABLE 17 ‘THIS WORKSHOP WAS AN EFFECTIVE WAS FOR ME TO THINK ABOUT PERSONALIZED MEDICINE IN MY OWN CLINICAL PRACTICE’.

	Workshop format				p (Fisher's)
	In-person		Online		
	34		30		
	n	%	n	%	
Strongly agree	19	55.9	8	26.7	0.0495
Agree	12	35.3	16	53.3	
Neutral	2	5.9	5	16.7	
Disagree	0	0.0	1	3.3	
Strongly disagree	1	2.9	0	0.0	

Table 18 considers whether participants would participate in a similar workshop in the future: While the majority of in-person participants agreed with this statement (74%), only a minority of online participants (48%) strongly agreed or agreed with this statement.

TABLE 18 ‘I WOULD PARTICIPATE IN A SIMILAR WORKSHOP IN THE FUTURE’

	Workshop format				p (Fisher's)
	In-person		Online		
	34		29		
	n	%	n	%	0.0034
Strongly agree	14	41.2	1	3.4	
Agree	11	32.4	13	44.8	
Neutral	6	17.6	11	37.9	
Disagree	2	5.9	3	10.3	
Strongly disagree	1	2.9	1	3.4	

Table 19 shows whether participants felt the workshop raised ideas they had not previously considered; 85% of in-person and 72% of online respondents strongly agreed or agreed with this statement.

TABLE 19 ‘THE WORKSHOP RAISED IDEAS I HAD NOT PREVIOUSLY CONSIDERED’

	Workshop format				p (Fisher's)
	In-person		Online		
	34		29		
	n	%	n	%	
Strongly agree	17	50.0	6	20.7	0.0325
Agree	12	35.3	15	51.7	
Neutral	2	5.9	6	20.7	
Disagree	1	2.9	2	6.9	
Strongly disagree	2	5.9	0	0.0	

Table 20 examines whether participants learned new information during the workshops: 88% of in-person and 79% of online participants strongly agreed or agreed with this statement indicating that while both approaches provided participants with new information again the in-person evaluation was scored higher.

TABLE 20 'I LEARNED NEW INFORMATION DURING THE [ONLINE] WORKSHOP'

	Workshop format				p (Fisher's)
	In-person		Online		
	34		28		
	n	%	n	%	0.0017
Strongly agree	20	58.8	4	14.3	
Agree	10	29.4	18	64.3	
Neutral	3	8.8	4	14.3	
Disagree	0	0.0	1	3.6	
Strongly disagree	1	2.9	1	3.6	

Table 21 considers whether participants felt the workshop presented information in an unbiased way. While the majority for both interventions felt positive about this statement the online group received a somewhat stronger evaluation with 86% of participants strongly agreeing or agreeing as compared to 79% for in-person.

TABLE 21 THE [ONLINE] WORKSHOP PRESENTED INFORMATION IN AN UNBIASED WAY

	Workshop format				p (Fisher's)
	In-person		Online		
	34		29		
	n	%	n	%	0.0141
Strongly agree	14	41.2	4	13.8	
Agree	13	38.2	21	72.4	
Neutral	4	11.8	4	13.8	
Disagree	3	8.8	0	0.0	
Strongly disagree	-		-		

Table 22 examines whether participants felt they had time to think about the issues during the workshop. Here the results were similar with 86% of online and 85% of in-person participants feeling positive about this statement.

TABLE 22 I HAD ENOUGH TIME TO THINK ABOUT THE ISSUES DURING THE [ONLINE] WORKSHOP'

	Workshop format				p (Fisher's)
	In-person		Online		
	34		29		
	n	%	n	%	
Strongly agree	17	50.0	6	20.7	0.0528
Agree	12	35.3	19	65.5	
Neutral	3	8.8	1	3.4	
Disagree	2	5.9	3	10.3	
Strongly disagree					

COMPARISON OF MEDIANS

Table 23 presents the results of a Wilcoxon rank sum test to evaluate the difference in the responses of the point 5-Likert scale for both the primary outcome and the other six evaluation measures (EM) – listed as EM1 through EM6; t-tests based on means are also shown for comparison purposes. Noting that a higher rank indicates a less favorable attitude, for the primary outcome we found a significant effect of intervention (online and in-person mean scores were 38.2 and 26.9 respectively) with $p=0.011$. The online intervention had a higher rank because the actual rank sums were higher as compared to the expected rank sums under the null hypothesis (while the in-person rank sums were lower). The t-test for the primary outcome is also significant at $p=0.02$ with the online group having a mean score of 2.4 as compared to 1.8 for the in-person group.

For the other supporting evaluation measures based on the Wilcoxon rank sum there were significant differences in interventions for EM1, EM2, EM3 and EM4 with all results indicating that the in-person version performed better based on participant rankings; the t-test results are more conservative with only EM2 and EM4 remaining significant.

TABLE 23 WILCOXON RANK SUM TEST AND T-TEST

	Wilcoxon rank sum test					t-test			
	N	sum of score	Expected under Ho	Mean score	Two-Sided Pr > Z	mean	Std. dev.	Pr > F	Pr > t
Primary outcome									
Online	29	1108.50	928	38.224	0.011	2.345	0.769	0.290	0.020
In-person	34	907.50	1088	26.691		1.824	0.937		
EM1: This [online] workshop was an effective way to make me think about genomic medicine in my own clinical practice									
Online	30	1134.00	975	37.800	0.023	1.967	0.765	0.538	0.069
In-person	34	946.00	1105	27.824		1.588	0.857		
EM2: I would participate in a similar [online] workshop in the future									
Online	29	1133.00	928	39.069	0.004	2.655	0.857	0.257	0.007
In-person	34	883.00	1088	25.971		1.971	1.059		
EM3: The [online] workshop raised ideas I had not previously considered									
Online	29	1074.00	928	37.034	0.035	2.138	0.833	0.144	0.172
In-person	34	942.00	1088	27.706		1.794	1.095		
EM4: I learned new information during the [online] workshop									
Online	28	1088.50	882	38.875	0.002	2.179	0.863	0.870	0.011
In-person	34	864.50	1071	25.426		1.588	0.892		
EM5: The [online] workshop presented information in an unbiased way									
Online	29	1009.50	928	34.810	0.221	2.000	0.535	0.003	0.539
In-person	34	1006.50	1088	29.603		1.882	0.946		
EM6: I had enough time to think about the issues during the[online] workshop									
Online	29	1052.50	928	36.293	0.066	2.035	0.823	0.763	0.131
In-person	34	963.50	1088	28.338		1.706	0.871		

Next we restricted the analysis to consider online nurse participant scores versus in-person scores, with the goal of examining whether the observed differences in the primary outcome were due to real differences in the evaluation of the interventions or confounding factors, the most obvious difference being the small number of physicians in the in-person workshop. Table 24 shows that when we control for confounding by health care profession

through stratification the primary outcome remains significant. For the other evaluation measures only EM4 remains significant which is reasonable because of the smaller sample size; the results show the same direction of association as observed in Table 23. Ideally we would have liked to stratify by HCP but the small sample size of physicians who participated in the in-person workshop precluded this.

TABLE 24 WILCOXON RANK SUM TEST CONSIDERING NURSES ONLY

	Wilcoxon rank sum test				
	N	sum of score	Expected under Ho	Mean score	Two-Sided Pr > Z
Primary outcome					
Online	12	342	270	28.458	0.044
In-person	32	649	720	20.266	
EM1					
Online	12	310	270	25.833	0.292
In-person	32	680	720	21.250	
EM2					
Online	11	296	242	26.909	0.121
In-person	32	650	704	20.313	
EM3					
Online	11	281	242	25.500	0.260
In-person	32	666	704	20.797	
EM4					
Online	11	317	242	28.773	0.020
In-person	32	630	704	19.672	
EM5					
Online	12	281	270	23.417	0.768
In-person	32	709	720	22.156	
EM6					
Online	11	270	242	24.545	0.437
In-person	32	676	704	21.125	

REGRESSION MODELING

Based on consultation with a biostatistician (M. Taljaard) we further evaluated the effect of confounding by performing regression modeling. We classified Likert scores as positive (1, 2) or negative (3, 4, 5) and used the Firth method of penalized likelihood because this is appropriate when sample sizes are small and when there is a risk of quasi complete separation of variables

(Allison, 2012). With this type of grouping the sample size was sufficient to examine the effect of the intervention design and health care profession on the odds of having a positive evaluation score. Table 25 shows that online participants had 0.354 times the odds of ranking the primary outcome as positive, compared to in-person participants, after adjusting for health care profession. The analysis of maximum likelihood estimate for the parameter intervention is -1.0396, indicating a negative association between the online intervention and a positive evaluation. While the direction of the association is the same as that reported using the Wilcoxon rank sum test and t-test, the 95% confidence interval [0.093-1.338] is not statistically significant at $\alpha=0.05$.

TABLE 25 LOGISTIC REGRESSION MODEL

Parameter	Estimate	P value, Analysis of Maximum Likelihood Estimates	Odds Ratio	95% Wald Confidence Limits
Intercept	1.6747	0.0372	--	--
Version (Nurses vs. physician)	-0.4041	0.5749	0.668	0.163-2.74
Intervention (Online vs. in person)	-1.0396	0.1257	0.354	0.093-1.338

Because re-grouping the evaluation scores into a dichotomous outcome resulted in loss of information we considered performing logistic regression for ordered categories and refining the grouping as positive (1,2), neutral (3) and negative (4,5); however because this meant that there were only 3 observations in the negative category we did not meet the practical requirement for the cumulative logit model of having at least 10 observations for each category of the dependent variable (Allison, 2012); instead following M Taljaard's advice we next performed linear regression considering the Likert response as a continuous outcome.

Fitting a generalized linear model with evaluation score as the dependent variable and intervention format (online or in-person) and health care profession (physician or nurse) as the

independent variables produced a significant overall F statistic ($p= 0.0428$). The tests of intervention in the Type III sums of squares shows that this term is significant in the model with $p=0.0146$; however health care profession is not significant with $p=0.3347$; the generalized linear model shows that the mean evaluation score will be 0.667 times lower for in-person participants, with lower scores equating to a more positive evaluation:

$$\text{EVALUATION_SCORE} = 2.5097 - 0.667 * \text{INTERVENTION} - 0.2814 * \text{HEALTH_CARE_PROFESSION}$$

Although intervention and health care profession only explain 10% of the variation in evaluation score the model supports our previous results that in-person participants evaluated the intervention more positively and that health care profession was not a confounding variable in this association.

4.4 COMPARISON OF PRE AND POST SURVEY RESULTS

The post workshop survey results are presented first for genomics in general and then for each of the three case studies (these match up with the results from pre-survey data). For each, we compare the pre- and post-survey results by intervention format to see whether participants' views changed over the course of the workshop.

UNDERSTANDING OF PERSONALIZED MEDICINE

Table 26 shows the breakdown of responses as percentages of total responses – 64 respondents made 255 different selections for an average of 4.0 selections per respondent. Overall HCPs the distribution was almost identical to the pre-survey results. Considering the responses by workshop format and by health care profession did not change the order of these selections and the distribution of responses was very similar to the overall category.

TABLE 26 ‘THIS IS WHAT PERSONALIZED MEDICINE MEANS TO ME’

	In person				Online			
	Pre		Post		Pre		Post	
	n	%	n	%	n	%	n	%
Taking a family history	25	19.8	24	17.8	22	21.8	25	20.8
Screening	27	21.4	28	20.7	23	22.8	24	20.0
Stratifying risk of disease in a healthy patient	28	22.2	26	19.3	26	25.7	29	24.2
Making a treatment decision	19	15.1	24	17.8	16	15.8	20	16.7
Making a prognostic decision	20	15.9	24	17.8	14	13.9	21	17.5
Other	7	5.6	9	6.7	0	0	1	0.8

CLINICAL UTILITY OF PM

Table 27 shows a large change in participants’ views about the clinical utility of PM before and after the workshop. Grouping the results into percentage of positive (‘strongly agree’ or ‘agree’), neutral, and negative (‘disagree’) responses shows that there is an increase of 61% to 81% for online participants and an even larger increase of 47% to 74% for in-person participants.

TABLE 27 ‘IN MY OPINION PERSONALIZED MEDICINE WILL BE USEFUL IN MY CLINICAL PRACTICE’

	In person				Online			
	Pre		Post		Pre		Post	
	n	%	n	%				
Strongly agree	10	29.4	9	26.5	3	10.7	5	19.2
Agree	6	17.6	16	47.1	14	50.0	16	61.5
Neutral	11	32.4	9	26.5	9	32.1	4	15.4
Disagree	6	17.6	0	0.0	1	3.6	1	3.8
Strongly disagree	1	2.9	0	0.0	1	3.6	0	0.0

CLINICAL UTILITY OF CRC RISK STRATIFICATION

Table 28 shows the pre- and post-survey views regarding clinical utility of CRC screening. Based on workshop format 69% of online and 58% of in-person participants strongly agreed or agreed with this statement; 23% of online and 15% of in-person participants were neutral; and 8% of online compared to 27% of in-person participants disagreed or strongly disagreed that CRC risk stratification would be clinically useful. There was minimal change in participants' views before and after the workshop. Comparing workshop format, the online participants became more positive after the workshop (65% to 69%) while the in-person participants become slightly more negative (61% to 58%).

TABLE 28 'IRRESPECTIVE OF THE NATURE OF THE TEST, BEING ABLE TO MORE ACCURATELY QUANTIFY A PATIENT'S RISK OF COLORECTAL CANCER WOULD MAKE A DIFFERENCE TO MY CLINICAL DECISION-MAKING.'

	In person				Online			
	Pre		Post		Pre		Post	
	N	%	n	%				
Strongly agree	7	21.2	9	27.3	3	10.3	4	15.4
Agree	13	39.4	10	30.3	16	55.2	14	53.8
Neutral	7	21.2	5	15.2	7	24.1	6	23.1
Disagree	4	12.1	7	21.2	2	6.9	1	3.8
Strongly disagree	2	6.1	2	6.1	1	3.4	1	3.8

CLINICAL UTILITY OF TYPE 1 DIABETES RISK STRATIFICATION

Table 29 shows the pre- and post-survey views regarding clinical utility of type 1 diabetes screening. Based on workshop format 59% of online and 30% of in-person participants strongly agreed or agreed with this statement; 10% of online and 29% of in-person participants were neutral; and 31% of online compared to 41% of in-person participants disagreed or strongly disagreed that type 1 diabetes risk stratification would be clinically useful. The majority of

online participants become more positive towards the idea after the workshop (46% pre to 59% post) while in-person participants became more negative.

TABLE 29 ‘IRRESPECTIVE OF THE NATURE OF THE TEST, BEING ABLE TO IDENTIFY INFANTS AT HIGHER THAN AVERAGE RISK OF TYPE 1 DIABETES WOULD MAKE A DIFFERENCE TO MY CLINICAL DECISION-MAKING.’

	In person				Online			
	Pre		Post		Pre		Post	
	N	%	N	%	N	%	N	%
Strongly agree	8	24.2	6	17.6	2	7.1	3	10.3
Agree	11	33.3	4	11.8	11	39.3	14	48.3
Neutral	7	21.2	10	29.4	9	32.1	3	10.3
Disagree	5	15.2	6	17.6	4	14.3	6	20.7
Strongly disagree	2	6.1	8	23.5	2	7.1	3	10.3

CLINICAL UTILITY OF FAMILY HISTORY TAKING

Table 30 presents pre- and post-survey results regarding the clinical utility of family history information: Online participants were extremely positive with 93% of participants strongly agreeing or agreed with this statement (as compared to 85% for in-person). Across both intervention formats respondents became more positive towards the utility of family history information post workshop; there was a large increase between pre and post for the in-person participants (76% to 85%) – even after the increase this is still not as high as for the online participants who shifted from 89% to 93% positive.

TABLE 30 ‘ON THE WHOLE, I FIND FAMILY HISTORY INFORMATION TO BE A USEFUL TOOL IN EVERYDAY PRACTICE’

	In person				Online			
	Pre		Post		Pre		Post	
	n	%	n	%	n	%	n	%
Strongly agree	13	39.4	22	64.7	8	28.6	11	39.3
Agree	12	36.4	7	20.6	17	60.7	15	53.6
Neutral	2	6.1	2	5.9	2	7.1	1	3.6
Disagree	3	9.1	1	2.9	1	3.6	1	3.6
Strongly disagree	3	9.1	2	5.9	0	0.0	0	0.0

4.5 COMPARISON OF THEMES IDENTIFIED FROM QUALITATIVE DATA

The qualitative data captured participants’ substantive views on genomics, providing greater detail than survey results alone, but also allowing further evaluation of the interventions by comparing the themes emerging in association with each intervention.

As a starting point we used the TDF domains described in Table 5 (Chapter 3) to categorize the qualitative comments. After the first classification attempt we added three additional themes pertaining to integration: integration challenges, integration benefits, and integration solutions; these additions were needed because integration issues are not included in the TDF which is behaviorally based. We also added a theme for terminology surrounding genomics; in total 10 overarching themes were thus identified as shown in Table 31. Overall 223 discrete comments were assigned to these themes; assignment to themes was fairly evenly divided between interventions with 125 online comments as compared to 107 in-person comments (for an average of 4.2 online comments per person as compared to 3.1 in-person comments).

The results show that 78% of the comments are divided between beliefs about consequences (46%) and integration issues (32%); while only 8% of the comments relate to

actual intentions to use the technology, indicating that professionals are still very early in their contemplation of the place of genomics in their practice. Very few HCPs commented that they would or would not use these genomic applications; rather the tone was cautious with a number of comments relating to the need for more evidence. There were no striking differences between the proportion of comments assigned to themes between the online and in-person workshop. Section 5.2 examines these overarching themes in more detail.

TABLE 31 OVERARCHING THEMES FROM QUALITATIVE DATA

Theme	Total comments	% total	Online comments	% online	In-person comments	% in-person
BELIEFS ABOUT CONSEQUENCES - NEGATIVE (DOUBT)	65	28.0	33	26.4	32	29.9
BELIEFS ABOUT CONSEQUENCES - POSITIVE	42	18.1	23	18.4	19	17.8
TERMINOLOGY SURROUNDING PM	6	2.6	3	2.4	3	2.8
INTEGRATION CHALLENGE	47	20.3	22	17.6	25	23.4
INTEGRATION BENEFITS	13	5.6	6	4.8	7	6.5
INTEGRATION SOLUTIONS	15	6.5	10	8	5	4.7
PROFESSIONAL ROLE & IDENTITY	19	8.2	12	9.6	7	6.5
INTENTIONS - towards positive	14	6.0	10	8	4	3.7
INTENTIONS - towards negative	4	1.7	3	2.4	1	0.9
ENVIRONMENTAL CONTEXT AND RESOURCES	7	3.0	3	2.4	4	3.7
TOTAL	232		125		107	

4.6 RECRUITMENT & LOGISTICS

While the number of participants for the in-person and online workshop was similar, the type of professionals who chose to participate in each workshop was quite different. Despite

significant recruitment efforts we were only able to conduct one physician workshop with a total of 2 participating physicians, the remaining 32 in-person participants being nurses. In contrast, 18 physicians participated in the online workshop as compared to 12 nurses. In total 20 physicians and 44 nurses participated in both workshops. We classified our recruitment efforts into three categories: 1) linkages with educational events; 2) recruitment through opinion leaders; and 3) direct contact. This following details our recruitment efforts and logistical considerations for both the in-person and online workshop.

IN-PERSON INTERVENTION

Linkage with educational events

- Unsuccessful negotiation with an Ontario-based professional organization to incorporate the workshop in their semi-annual CME day (contact initially being brokered by an Ottawa based clinician with strong professional ties to the society). This organization was enthusiastic to include the workshop in their CME event, but would not consent to any data collection component.
- Successful negotiation with a professional organization in another province to include the workshop in their annual primary care event. This was well advertised and attracted CME credits, but the programming on the day set it against a vaccination update workshop for which almost all potential participants opted.
- Successful negotiation with a nursing professor (also an opinion leader) from a post-secondary education institution to incorporate the workshop in an educational event for student nurses.
- Successful negotiation with a professional nursing body to incorporate the workshop within an educational day at a local hospital.

Opinion leaders

- A PCP associated with the University of Ottawa contacted a number of groups on our behalf and championed the workshop. No practitioners agreed to participate, most commonly indicating they felt the topic was of little relevance to their practice.
- An advanced practice nurse associated with a post-secondary education institution advertised and supported the workshop; two attempts were required to secure participation.

Direct contact

- CC and BW directly contacted other primary care groups and all respondents declined to participate; the overwhelming sense was that physicians did not have time to participate in professional engagement activities, particularly with no financial or professional incentive offered.

ONLINE INTERVENTION

Opinion leaders

- The nursing opinion leaders involved in the recruitment for the in-person workshop also distributed the online survey link to their professional and educational networks.
- A physician opinion leader sent direct invitations to nurses and physicians associated with a family health team.

Professional networks

- A member of the research team based in another province disseminated the online invitation through list servers for family medicine, rural family medicine and nursing units associated with a post-secondary institution.
- A member of the research team invited family medicine residents participating in a PGY-

3 Enhanced Skills Program at a post-secondary institution to participate in the online intervention.

With Internet based surveys it is common to report the number of useable questionnaires returned rather than the overall response rate. A total of 85 people accessed the online version of the workshop between January-May 2014, and of these, there were 30 completed responses giving a completion rate of 35%. The average survey completion time was 25 minutes. Many people who did not complete the online workshop did so after finishing the initial set of questions and launching the first video, suggesting that the majority of online participants chose not to watch the first seven minute educational video and abandoned the workshop at that point.

COST

We did not include a formal economic evaluation in the pilot study.

The most financially costly element of the in-person workshops was the travel for three investigators to run workshops outside the local area. The costs for Ottawa sessions included survey photocopying, providing light refreshments and parking passes, and paying students to take field notes. The moderators gave their time free of charge. The costs for online sessions included a yearly subscription to Fluid Surveys and Video Scribe, both approximately \$200. The developer (CC) gave her time free of charge.

CHAPTER 5. PRELIMINARY FINDINGS ABOUT GENOMICS

This chapter summarizes the engagement results. Given that this is a pilot study we are not trying to draw definitive conclusions from the engagement data, rather we include it to provide a picture which might apply to Canadian professional populations and allow broad comparison of our findings with the published literature. Based on the fact that there were no major differences between the interventions obviously apparent from Chapter 5, in that both interventions were evaluated positively, this chapter relates an appropriately tentative story about the engagement data, without separating into in-person versus online groups.

5.1 SURVEY ENGAGEMENT DATA

COLORECTAL CANCER CASE STUDY

This set of CRC related questions asked respondents to suppose that a risk stratification test for CRC existed: *‘If a properly validated genomic profiling test for colorectal cancer became available...’* All responses are shown in Table 32. Overall, the majority of respondents felt positively that CRC genomic profiling would benefit patients (71%); that their patients would expect them to recommend CRC profiling (82%); and that they should be capable of ordering this test (78%). However just over half (51%) indicated they would actually recommend such a test, with a large percentage undecided (40%).

TABLE 32 ‘IF A PROPERLY VALIDATED GENOMIC TEST FOR CRC BECAME AVAILABLE...’

Statement	N (%)					N
	SA	A	N	D	SD	
<i>If a properly validated genomic test for CRC became available,</i>						
I think it would generally help patients	24 (38.7)	20 (32.3)	12 (19.4)	6 (19.4)	0 (0)	62
I think most of my patients would expect me to recommend it	20 (32.3)	31 (50.0)	9 (14.5)	1 (1.6)	1 (1.6)	62
I think family physicians should be capable of ordering it	17 (27.0)	32 (50.8)	11 (17.5)	2 (3.2)	1 (.6)	63

Statement	N (%)					N
	SA	A	N	D	SD	
I would recommend it to patients	8 (13.3)	23 (38.3)	24 (40.0)	5 (8.3)	0 (0)	63
SA: Strongly agree; A: Agree; N: Neutral; D: Disagree; SD: Strongly disagree						

To further explore which patient groups HCPs believe should receive CRC risk testing we asked those participants who believed they would recommend this test whether there were any groups of patients for whom they would not recommend it. The results are shown in Table 33 which suggests that, of all respondents who would recommend this test, three quarters would have reservations about offering it to some patient or population groups. The ‘other’ category included responses such as:

- “Patients older than 70 or those with a high mortality risk over the next 5 years.”
- “I would discuss it and come to a decision with the patient (rather than recommending it)”
- “Too little information to answer this question properly - need to know who the test was validated in and how well it predicts risk, etc...”

TABLE 33 PATIENT GROUPS FOR WHOM TEST SHOULD NOT BE RECOMMENDED

Groups for whom test should not be recommended	69 responses	
	n	%
Patients not yet eligible for standard colorectal cancer screening (e.g., under 50)	6	8.7
Patients without a family history of colorectal cancer	12	17.4
Patient who don't actively request this type of test	7	10.1
Patients who are not good at managing their health risks	9	13.0
Patients who are anxious	9	13.0
None (I would recommend it to everyone)	18	26.1
Other	8	11.6

TYPE 1 DIABETES CASE STUDY

The type 1 diabetes questions asked respondents to suppose that a risk stratification test for type 1 diabetes existed: *‘If a properly validated genomic profiling test for type 1 diabetes in newborns became available...’*, results are shown in Table 34. Views as to whether this would

benefit children and parents were almost equally divided between positive, neutral and negative responses; however, the majority of respondents still felt positively that parents would expect them to recommend type 1 diabetes screening for their children (71%) and that they should be capable of ordering these tests (61%). When asked whether they would recommend this test to parents only 23% agreed or strongly agreed, while 40% disagreed or strongly disagreed.

TABLE 34 'IF A PROPERLY VALIDATED GENOMIC PROFILING TEST FOR TYPE 1 DIABETES BECAME AVAILABLE...'

Statement	N (%)					N
	SA	A	N	D	SD	
<i>If a properly validated genomic profiling test for type 1 diabetes in new-borns became available,</i>						
I think knowing the test result would generally help children	11 (17.5)	10 (15.9)	21 (33.3)	7 (27.0)	4 (6.3)	63
I think knowing the test result would generally be beneficial to parents	8 (12.9)	14 (22.6)	19 (30.6)	15 (24.2)	6 (9.7)	62
I think most parents would expect me to recommend this test	16 (26.2)	27 (44.3)	10 (16.4)	5 (8.2)	3 (4.9)	61
I think family physicians should be capable of ordering it	14 (21.9)	25 (39.1)	21 (32.8)	2 (3.1)	2 (3.1)	64
I would recommend this test to parents	8 (13.3)	6 (10.0)	22 (36.7)	16 (26.7)	8 (13.3)	60
SA: Strongly agree; A: Agree; N: Neutral; D: Disagree; SD: Strongly disagree						

As for the CRC example, we asked participants who indicated they would recommend a type 1 diabetes test whether there were any groups of infants for whom they would not recommend it. Table 35 suggests that around 70% would have reservations for at least some infants.

TABLE 35 PATIENT GROUPS FOR WHOM TEST SHOULD NOT BE RECOMMENDED

Groups for whom the test should not be recommended	41 responses	
	n	%
Infants of parents who don't actively request this type of test for their child	6	14.6
Infants where there is no family history of type 1 diabetes	6	14.6
Infants of parents who are not good at managing their own health risks	7	17.1
Infants of parents who are anxious	7	17.1
None (I would recommend it to everyone)	12	29.3
Other	3	7.3

FAMILY HISTORY TAKING CASE STUDY

The set of family history questions ask respondents to consider the statement: ***'If the province promoted a standardized approach to family history taking...'*** Table 36 shows that the majority of respondents believe that this would be beneficial to patients (84%); that their patients would expect them to such a system (67%). Unlike with the previous two scenarios the percentage of respondents who indicated they would actually use such a system was high (69%).

TABLE 36 'IF THE PROVINCE PROMOTED A STANDARDIZED APPROACH TO FAMILY HISTORY TAKING'

Statement	N (%)					N
	SA	A	N	D	SD	
<i>If the province promoted a standardized approach to family history taking...</i>						
I think it would generally help patients	31 (50.8)	20 (32.8)	10 (16.4)	0 (0)	0 (0)	61
I think most of my patients would expect me to use such a system	16 (25.4)	26 (41.3)	19 (30.2)	1 (1.6)	1 (1.6)	63
I would use the system instead of my current approach	20 (31.3)	24 (37.5)	18 (28.1)	1 (1.6)	1 (1.6)	64
SA: Strongly agree; A: Agree; N: Neutral; D: Disagree; SD: Strongly disagree						

Table 37 asks respondents about their current approach to family history taking and shows the percentages of total responses; patients were able to select more than one approach. The results show that 40% of respondents collect family health information at new patient intake; 29% when a patient presents with a new complaint; 25% during periodic health exams; and 6% chose other. For example, one physician explained: *"This depends on the type of visit. If intake I use a more structured tool, but if [patient is] in for a 15 minute visit with a new complaint, I just ask informal family history"*.

TABLE 37 APPROACHES TO FAMILY HISTORY TAKING

Approach to family history taking	143 responses	
	n	%
New patient intake	57	39.9
Periodic health exams	36	25.2
When a patient presents with a new complaint	42	29.4
Other	8	5.6

INTEGRATION SOLUTIONS

Table 38 examines which approaches HCPs believe would help them integrate genomic tests into their clinical practice. For those HCPs who would wish to integrate genomics into their practice the most popular option was targeted education sessions, following closely by specific point of care tools to use within consultation, a colleague within their clinic prepared to become an expert, and easy access to a genetic counselor, the least popular option was Web-based support for answering questions and helping assess patients' risk.

TABLE 38 'WHICH TOOLS WOULD HELP YOU INTEGRATE GENOMIC SCREENING INTO YOUR CLINICAL PRACTICE?'

Integration tools	256 responses	
	n	%
I would not wish to integrate them into my own practice	49	19.1
Specific point of care tools to use within consultation (e.g., guidelines, mobile apps)	52	20.3
Targeted CME/CPD sessions on specific tests	55	21.5
Easy access to a genetic counselor	44	17.2
A colleague in my clinic prepared to become an expert	45	17.6
Web-based support for answering questions and helping me assess patients' risk	10	3.9
Other	1	0.4

Table 39 indicates the approaches HCPs believe would help them integrate a standardized family history taking system into their clinical practice. For those HCPs who would wish to

standardize their approach, the most frequent options were a family history tools linked directly to an electronic medical record, that could be filled in by patients, and could be used by the practitioner him or herself.

TABLE 39 ‘WHICH APPROACH WOULD HELP YOU INTEGRATE A STANDARDIZED APPROACH TO FAMILY HISTORY TAKING INTO YOUR CLINICAL PRACTICE?’

Family History Integration Approaches	164 responses	
	n	%
I would not wish to change my approach to family history taking	3	1.8
A family history tool that my patients could fill in (paper, web-based, mobile app)	45	27.4
A structured family history tool that could be used to gather information in telephone interviews	21	12.8
A family history tool that I could use myself (paper, web-based, mobile app)	42	25.6
A family history tool that linked directly to an electronic medical record	48	29.3
Other	5	3.0

5.2 QUALITATIVE DATA

In this section the qualitative themes identified in Table 31 (Chapter 4) are explored in greater detail; as with the previous section comments are grouped by all HCPs regardless of intervention type. We chose not to include respondent numbers with the qualitative results; because we did not have approval to audio record the focus groups it was not possible for the scribes to link workshop participants (who were not known in advance) with their comments during fast-paced discussion; this was particularly true for the larger nursing workshops.

BELIEFS ABOUT CONSEQUENCES

Participants held a wide range of beliefs about the consequences of genomic testing, both negative and positive. Many expressed negative beliefs concerned whether there was enough evidence of direct patient benefit to justify implementation:

'We need to only be using tools that really make a difference in evaluating risk, otherwise we will get bombarded with so many tools that they won't be taken up into practice.'

'It is important to show direct patient benefit, to justify implementation. Pragmatic studies will be useful to establish whether innovations such as these would be actually reaching patients (research to practice gaps).'

The need to show improved morbidity and mortality and prove test accuracy was particularly evident in the type 1 diabetes scenario, where concerns were expressed about genomic tests increasing parental anxiety, leading to parental hyper-vigilance and increased ER visits, changing the parent/child relationship, and negatively impacting pediatric mental health:

'It would be important to understand the accuracy of genomics testing with respect to diabetes type 1. If the test was very accurate with respect to relative risk, then there may be some value in sharing this information with the parents/family physician in terms of screening and surveillance. On the other hand, if this test required further refinements in terms of accurately predicting the onset of Diabetes type 1, then it would be important consider the possible risks associated with needless worry or concern regarding the potential onset of the disease.'

'The question would be: is there benefit- in terms of morbidity and mortality- from early risk stratification (that may lead to improved surveillance)? If there is no morbidity or mortality benefit, you are just doing harm by having many parents and kids worried.'

'Diabetes is an area where I'm nervous about the motivation. We keep pathologizing it. If we label an infant from the get go, from that moment the child and parent relationship forever changes. How do you quantify the risk there?'

Multiple comments reflected uncertainty that more risk assessment tools were really needed, and doubted the added utility of genomic testing over traditional primary care checkups:

'One could anticipate that regular visits to a family physician would prove equally effective in some cases versus genomic testing.'

'There are more and more risk stratification tools being used in primary care- will these tests really add to the risk prediction for disease?'

Some comments indicted that, even assuming that genomic tests were able to demonstrate clinical validity, there was reason to be skeptical about the value of this information, arguing that it is unlikely that patients identified as having increased risk would actually modify their behaviors. Other comments suggested concern that risk stratification could lead to higher risk patients feeling ‘doomed’ and deciding there was no point in changing their lifestyle behaviors, or conversely lower risk patients feeling ‘immune’ and engaging in riskier activities. Some participants seemed worried that they, as health care professionals, might over interpret a patient’s genomic risk assessment profile or that genomic tests could lead to overly aggressive or even unnecessary interventions:

‘As with many things, my guess is the majority of cancers might still occur in individuals who are at low risk, so what about all those people at average risk? Will we say "your genetic profile doesn't indicate high risk, so we are not going to send you for a colonoscopy, you can only have an FOBT"that would be my concern- over interpretation or reliance on the risk profile and missing people who would have otherwise been screened under the current system. I sound skeptical- I think this information would be helpful- but it needs to be used carefully....’

‘If you get the testing done and it says you have a predisposition the HCP might jump to conclusion that this is what you have – skews the lens of healthcare.’

A sub theme of negative beliefs emerged surrounding ethical and legal issues for the patient and society. Concerns were expressed about genomic information leading to employment and insurance discrimination and violating religious beliefs. There was also skepticism about whether this use of money was warranted within an already limited health care system. On a more philosophical level there was skepticism about where HCPs should ‘draw the line’ in terms of genomic tests, as one participant commented: ‘This is really letting the genie out of the box.’

Many positive statements were also made about genomics, although these comments tended to be less detailed. These included beliefs that improved risk stratification could lead to a

more ‘patient-centered’ approach where HCPs could aid in disease prevention through improved lifestyle advice; this was particularly linked in some cases to the idea that genomic tests could supplement family history information. Many comments reflected participants’ positive views of the proposed standardized approach to family history taking, citing a precise and accurate family history as a key tool in risk reduction.

‘If you could pair genetic information with family history this could give more weight to family history. For example, a 60 year old woman with high cholesterol, knowing her genetics might give an extra push to make lifestyle changes.’

‘Focusing prevention on diseases that have not yet presented, AND being able to really prove who is at "high risk", would be the ultimate advancement in medicine!’

Other comments drew parallels between genomic testing and current primary care practice, with genomic tests being just another example of patients having access to their own health information:

‘This in some ways is similar to other types of risk stratification that we do like Framingham which guides treatment/testing’

While many negative comments were expressed about the utility of genomics in risk stratification for Type 1 diabetes, an interesting positive comment was that risk stratification could be beneficial by reducing the severity of disease progression:

‘We learned in pathology that the pancreas cells are 80% gone before onset of diabetes becomes obvious, if a genomic test could predict that the patient would develop diabetes before this decline then maybe something could be done.’

TERMINOLOGY SURROUNDING PM

There is debate in the literature about the appropriateness of the term ‘personalized medicine’ with different terms used to describe the use of genomic applications in medicine

(individualized medicine, precision medicine, genomics); opposition to the term ‘personalized medicine’ was also apparent amongst participants. Some comments pertained to ‘personalized medicine’ not being an appropriate term for genomics; some indicated outright offence. Several comments reflected that PM was too ‘broad’ a term and not specific enough; others indicated feelings that the term had been ‘hijacked’ from primary care and that primary care professionals already practiced personalized medicine by using a personalized approach to patient care:

‘I find the term [personalized medicine] problematic if not offensive. It feels like the genetics people have taken it and applied it to their particular approach. The whole point of family medicine is to tailor medicine to the person. The term is so offensive it gets in the way of me actually paying attention.’

‘This is what I think PM is – when you find out a 35 year old woman gave up a baby when she was 16 or that someone’s mother died when they were 6. As family practitioners we bear witness to their life story.’

INTEGRATION

Participants expressed views about how genomic applications would be integrated into primary care; we divided integration comments into three high level themes: integration challenges, integration benefits, and integration solutions.

A sub theme that emerged under ‘integration challenges’ was how PCPs would be able to order, interpret and use genomic information given the inherent complexity of genomic tests. Some comments related concerns about failing to order genomic tests in situations where they would be beneficial (‘should I just screen everyone?’); others pointed out that the complexity of patient care combined with time constraints and financial limitations already makes it challenging to run a patient centered practice and that genomic tests would add to this complexity. Overall, a dominant theme emerged that participants did not feel prepared to incorporate genomics into practice, as reflected in these comments:

'It's really interesting to think of more personalized medicine, but the complexity is going to be hard for primary care specialists to incorporate into everyday practice'

'Family physicians probably won't learn a great deal about genomics until we are told exactly how to use the knowledge - e.g. a certain genomic profile alters decisions regarding screening or treatment.'

Some views indicated a lack of knowledge and formal education as limiting HCPs' ability to interpret genomic results:

'I'm thinking about this from an educational perspective and thinking about how nurses' roles will change as they have access to the data. Health providers need to be well equipped to be able to understand and interpret genomic results and at this point we are not. We need education and tools.'

Many challenges were identified that related to integrating a standardized approach to family history taking into primary practice. There was concern that data captured through a standardized approach, particularly if this was done online with no HCP interaction, would miss clinical details and lack relevance.

'Patients often have no idea what diseases or conditions they have, let alone their family members, and as health care providers we often help piece it together (ie. if a patient says their parent died at the age of 45 suddenly and was a drinker/smoker/obese.....likely myocardial infarction, or at least likely something cardiac. The patient might just say 'died of unknown causes.')

Also sometimes detail helps: for example, a family history of drowning can be indicative of cardiac pathology (arrhythmias) in young patients because the arrhythmia often occurs when the young person is exercising, and has no ability to get help. This kind of "detail" might be missed on a standardized form.'

'Further, can you develop a clinical health history form that can account for all aspects of a person's health? Some level of interaction /interface between the patient and health care practitioner would still be required in order to ensure accuracy and feasibility of the data being added to the health history data base. A healthcare provider would also understand the difference between highly "relevant" vs. "irrelevant" data, acting as a filter of the patient's data and if need be can ask for additional information that may be relevant.'

While apparently receptive to the idea of a standardized approach, many comments indicated a general feeling that HCPs would prefer to use such a system in conjunction with a more traditional approach to family history taking; and that there are situations where this approach would not be appropriate, for example:

‘I’m thinking of clinical situations where FH isn’t relevant and someone could be concerned about privacy, for example the privacy of a teenager. There are lots of benign examples where FH isn’t needed.’

There was also a sense that a standardized approach lacked the ‘human touch’ and that it would be difficult to use such a system to validate social aspects of the patient’s well-being and to understand patient beliefs; views were expressed that family history is about more than just clinical data collection but includes personal history, anecdotes and legends.

‘Family history is a way to open the dialogue and relate what they have observed back to what you see. You have to understand the belief system in the family.’

A major concern related to safeguarding the privacy of family history information (and all genomic results) and the legality of linking family history information between families, and the need for reassurance that this information would not be available to insurance or pharmaceutical companies. Detailed implementation challenges were also raised; for instance once the data was populated how it would be updated and how would the system maintain accuracy in situations where multiple HCPs were adding information?

Comments also related to patient level integration challenges. As well as doubting their own ability to interpret genomic results, comments revealed some worries about whether patients would be able to understand these results and whether all patients would want access to genomic information, particularly for certain cultures and religions. The diabetes scenario raised questions about whether parents have the right to consent to genomic testing for their children or whether

this is a decision that the child should make in adulthood.

Participants also identified many benefits to integration, although, in general, the comments surrounding the more positive aspect of genomics seemed less detailed than those pertaining to the challenges.

Despite identifying numerous integration challenges surrounding a standardized family history taking system, a perception was also expressed that such a system would be more helpful than harmful to patients. While some indicated that their practice was already using an electronic medical record the view was that current EHRs do not make full use of family history information by alerting HCPs at the point of care.

'Systematic health history taking is critical in understanding the chief complaint or diagnosis related to a new admission or routine health examine ... an online system would enable greater accessibility to patient health history'

An interesting idea was proposed that genomics could promote consistency in primary care, with the idea being that genomic screening guidelines could be applied by all HCPs thus standardizing care and eliminating the need for patients to self-advocate for their right to receive tests:

'HCPs need to be on the same page [in regards to guidelines for use with this sort of hypothetical test]. In some cases people have FH but are told that colonoscopy is not needed. We need to follow guidelines for screening but over the years these guidelines change and HCPs don't always keep up. The guidelines have to be evidence based and include clinical reminders. Right now the onus is often on the patient to ask for screening and that isn't right. This would be a more integrated and consistent system.'

The idea of standardized care was also applied to PM addressing diversity in patient populations:

'Inconsistency in practice is a huge problem. For example immigrants often have little idea of their family history. Something like this [test] would help address the diversity in our population, rather than making such broad

categories like: 'are you from the Caribbean?' This would really be personalized.'

Integration costs were also considered in a positive light. Several comments reflected the idea that more targeted prescribing would actually reduce costs and that, rather than viewing genomics negatively, insurance companies might find that genomic information would lead to cost savings.

As well as identifying integration challenges, innovative integration solutions emerged. For standardized family history the point was made that there is a lot of repetition in family history taking and that a standardized approach could use a streamlined checklist and save time. Some suggested that patients could complete forms online prior to their visit, allowing more time for deliberation, and the forms could then be reviewed with their HCP. Several comments reflected the belief that patients would have a right to access information collected through this process and that could be available to both HCPs and patients through mobile technology.

To address the issue of a standardized form missing detail and being overly generic, the idea was proposed that the forms be customizable to better fit individual patients. The proposal was offered that a standardized approach could incorporate and link different types of health information: genomics, family history, lifestyle and environmental factors. Suggestions were also made to link standardized family history information with the EHR, clinical guidelines and alerts for prescribing.

'One idea for integrating personalized medicine is to incorporate any genomic information into the electronic medical record (EMR). For example, for ultra-rapid codeine metabolizers, instead of just having that in the file, it should be somehow in the EMR and linked to the EMR system for prescribing so you get a warning if you try to prescribe codeine. Some EMRs can do this, others can't, but it would be important moving forward to think about systemic ways to incorporate the knowledge, not just advising physicians of a predisposition because that information can get lost in a busy appointment with multiple issues.'

A comment reflected the view that the increased complexity of prescribing would be most effectively managed by including a pharmacist in each health care team.

‘There is so much complexity already in prescribing (i.e. looking for drug interactions, advising patients of possible severe and/or common side effects, assessing the need of a particular medication for a patient, looking at whether one drug or drug class is superior to another drug or drug class). When you add on thinking about a person’s genome in prescribing practices it almost becomes overwhelming. You’d probably need a pharmacist at every primary care group.’

The idea of genomics requiring an extended group of health professionals was also proposed, with suggestions that counselors would need to be part of the health care team; both to help patients complete the standardized forms and to help in interpreting the genomic results.

Comments indicated a sense that HCPs were more comfortable with the idea of integrating standardized family history taking into their practice than genomic tests. Few integration solutions were proposed for genomic tests, although one comment did suggest how they would consider using CRC screening in practice:

‘Because FOBT is non-invasive and already not very sensitive I don’t know that I would decrease the frequency of offering it to patients, based on genomics, but probably would use this information to help guide frequency of colonoscopy, in those patients for whom that is their preferred method of screening.’

PROFESSIONAL ROLE AND IDENTITY

We grouped comments that reflected professionals’ views of their role in genomics under this heading. While few HCPs expressed clear intent as to whether or not they intended to use genomics a number of comments suggested that they considered supporting genomics as part of their role in primary care.

One of the anticipated main roles for professionals would be in ordering and interpreting genomic test results; however this was cause for concern in the case of full genome sequencing:

‘Anytime we order a test, we should be responsible for interpreting the results and passing them on to the patient in an understandable mannerhow would this be possible with full genome sequencing?’

Many comments reflected an apparent belief that family history taking was an important part of their professional role and allowed targeting of patients who were at greater risk; there was a general sense that the hypothetical genomic tests aligned with their current roles in risk stratification and disease prevention.

A view that was repeatedly expressed was that genomics would lead to an increased need for their role in patient education and support, through tailoring patient education to individual risk levels, communicating risk reduction information, and supporting patients and families receiving genomic results. The opinion was expressed that in order to provide patient education at this level HCPs would also require further training and education in order to better understand genomics. While some participants believed that an important part of the HCPs’ role was in supporting patient privacy and protecting access to this information, an alternative view was suggested that in the future patients would be the ‘owners’ of their health information and the HCPs’ role would shift to supporting patient use of this information.

INTENTIONS

In general, comments were lacking about whether participants would or would not incorporate genomics into their practice. A positive intent was more discernible about using standardized family history systems which appeared to be generally considered beneficial to patients; the view was expressed that such a system would likely be used in conjunction with current approaches. Several comments reflected positive intent towards using genomics in CRC screening, indicating that this sort of tool would be particularly useful in cases where the patient had no family history of colorectal cancer. One participant specifically expressed positive intent

to use type 1 diabetes screening, explaining that this would just be adding another condition to newborn screening in Ontario; another stated probable intent but qualified this by stating that the parents would have to receive appropriate counseling to understand test results.

Comments reflecting negative intent were justified by a lack of current evidence and cost benefit analyses. One comment reflected a strong belief that genomic information can be useful but should not be applied to the general population. There was a sense that genomics was not really ‘catching on in Canada’ and was not ready for integration:

‘At this time, I don't think I could use genomic information directly in my critical care practice. Though there might be possibilities for pharmacogenetics and companion diagnostics in the future.’

ENVIRONMENTAL CONTEXT AND RESOURCES

In terms of the resources required, a concern was expressed whether the currently overstretched health care system has the capacity to support genomics in primary care and whether this would take resources away from other screening programs. A strong theme was present that health systems must be accountable for the resources allocated to genetic tests and therapies. The view was expressed that the health care system is already not consistently screening for diseases where screening is directly associated with decreased mortality and morbidity so how would the system support population level genomic screening?

‘Do we have the capacity for this? For example, currently with many childhood cancers we don't even have the capacity to screen properly for secondary cancers that we know these children are at increased risk for. For example, I have seen situations where a young person has a cancer and then 10 years later they develop a cancer that was related to the earlier cancer but we were not screening them.’

Views were also expressed that genomic screening would increase unnecessary primary care visits which has a negative impact at a population level and that emergency department visits and wait times would increase.

CHAPTER 6. DISCUSSION

6.1 THESIS CONTRIBUTIONS

In this thesis project, we developed two interventions for educating and engaging with HCPs about emerging applications in genomics. The first PE intervention was a structured in person ‘focus group type’ workshop and the second was an online version of the same workshop, also incorporating educational and engagement components but without the possibility for group interaction. Comparing these interventions provided methodological research evidence specifically about the ‘effectiveness’ of the two professional engagement approaches. The interventions also offered preliminary evidence about how the target health care professional groups react to new genomics technologies, relevant to health technology assessment in Canada and elsewhere.

The PE interventions were considered ‘complex interventions’ and designed using the complex intervention framework, where the MRC guidelines state: ‘*Best practice is to develop interventions systematically, using the best available evidence and appropriate theory*’ (Medical Research Council, 2008). Following this approach the intervention was based on applicable underpinning theories and frameworks: 1) Fink’s educational principles for developing integrated courses for significant learning were used to guide the development of the intervention’s educational component; 2) Kirkpatrick’s topology for evaluating educational initiative was used to focus the engagement data collection; and 3) the Theoretical Domains Framework was used in developing the professional engagement questions. All intervention components were described in detail following Clark’s suggestion that the individual components of complex interventions have become ‘*researchable aspects*’ in their own right (Clark, 2013) and based on the increasing need for researchers to describe the components of

their complex intervention in publications (Craig et al., 2008; Glasgow & Emmons, 2007).

An important goal of this pilot was to get a sense of whether the underlying theories behind the intervention (Preclinical Phase – Section 3.2), the resulting model components (Phase #1 – Section 3.3), and the subsequent evaluation through an exploratory trial (Phase #2 – Section 3.4) were effective in achieving our goal of evaluating the two intervention approaches and engaging with HCPs. Campbell et al. (Campbell et al., 2007) updated the original linear complex intervention framework (Figure 2, Chapter 3) to a circular model (see Figure 5) where the evidence gained from the first 3 phases feed forward to inform a possible Phase #3 randomized control trial. This chapter summarizes the evidence from our pilot study and presents lessons learned, limitations, and recommendations for a future RCT.

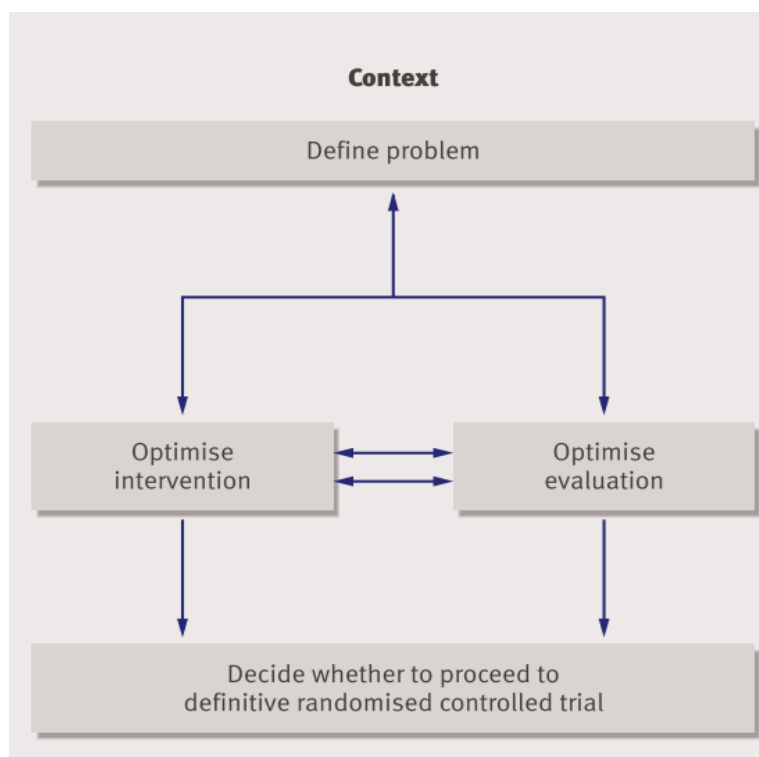


FIGURE 5 CIRCULAR VIEW OF COMPLEX INTERVENTIONS FRAMEWORK (CAMPBELL ET AL., 2007)

6.2 EVIDENCE FROM PRE-CLINICAL AND MODELING PHASE

The intervention design was based on several theoretical constructs that proved to be

effective in the pilot implementation and we recommend maintaining this approach for a future RCT. The appropriateness of Kirkpatrick's topology was evidenced by the fact that most of the qualitative data mapped to the levels we chose to evaluate (learner's reactions and modifications of attitudes and perceptions). The Theoretical Domains Framework was a very effective tool, both for developing the engagement questions which in turn focused the educational aspect of the intervention, and as a means of grouping qualitative data into themes.

The pilot study suggested that the educational component was an effective means of establishing baseline knowledge and creating an environment conducive to informed engagement. While we believe that the educational component was necessary to ensure the validity of the engagement approach, this initial didactic component made recruitment more challenging because we required more of the participant's time; however, we recommend that a future RCT leverage the fact that the workshop provides unbiased and evidence-based genomic education to embed the intervention within well-established continuing medical education and continuing professional development programs. This will require advance planning since the curriculum for these events is set up far in advance of the actual event. We suggest that a future RCT pursue connections we established during this thesis with Robert Parsons to discuss how an online version of CPD/CME programs could be delivered; this would offer an excellent opportunity to compare the two approaches in an even handed way.

While the educational component was effective it did require the moderator to have professional experience in order to establish credibility with the audience. In comparison the online version represents a more standardized approach to education, feasible for replication and wide spread dissemination.

6.3 EVIDENCE FROM EXPLORATORY TRIAL

STUDY DESIGN

By necessity the study design evolved to account for challenges experienced recruiting HCPs to participate in the professional engagement exercises. Ideally, a randomized approach would have been employed where participants would experience both interventions, each intervention focusing on a separate topic in genomics, with random allocation to topic and the order of intervention. This allocation approach would permit individual-level paired data and offer the opportunity to ask direct questions regarding preference for one intervention over the other. Instead, a quasi-experimental design without randomization was selected when it became apparent that it was extremely difficult to recruit HCPs to even one version of the intervention and that expecting participants to participate in both an in-person workshop and online workshop was not logistically feasible given the time and resources available.

RECRUITMENT & PARTICIPATION

Recruitment was an ongoing challenge throughout this project. While we were able to recruit a total of 64 HCPs, despite significant efforts by the investigators and our physician champions (opinion leaders) we were only able to conduct one physician workshop with a total of 2 participating physicians. The main reason cited by physicians for choosing not to participate was a perceived lack of relevance to their clinical practice, which is in line with the findings from the literature review that many HCPs do not perceive genomics as relevant. In other words, it is necessary to find a way to engage physicians in physician engagement research.

There was also an underlying sense that HCPs are disinclined to themselves be research subjects, as evidenced by the reluctance of a professional group to allow data collection, although this may also reflect highly conscientious ‘gate-keeping’ by an organizing committee.

Recruitment for nurses was considerably more successful with 32 in-person nursing participants. In contrast, the online workshop appeared to be more appealing to physicians, with 18 online physician participants as compared to 12 nurses. We suggest that recruitment to the online version was more successful because we eliminated the need for busy HCPs to be physically present at a set time and place.

A further observation made by the research team was that online participants appeared more likely to maintain a ‘professional’ (objective) perspective about genomics as judged by their open comments. The group dynamic during the in-person workshops seemed to foster an environment where participants were more likely to express their views on genomics through a personal lens, often recounting stories of their family experiences with genetics and genetic testing. Considerable skill was required on the moderators’ part to diplomatically redirect participants to consider the issues from the specific viewpoint of a primary care professional.

Our pilot indicates that despite experiencing significant difficulties with recruitment, the majority of HCPs who chose to participate ranked the interventions positively and were very interested in the topic. The challenge is in recruitment, once recruited, participants are highly engaged.

DATA COLLECTION

For the in-person workshop the vast majority of qualitative data was collected from the field notes, very few participants chose to use the writing booklets for free text comments. In contrast, the online text boxes were an effective means of collecting qualitative data; this result was unexpected, we had thought it likely that online participants would not comment in the textboxes and that a lack of qualitative data would be a drawback of the online approach. In reality, while this was the case for some participants, those that did comment often included

more detailed and considered responses than were possible during the in-person workshop. During the in-person workshop the short time frame meant that the moderator had to carefully balance each person's contributions in order to give everyone a chance to participate. The in-person comments built on each other in a way that the online comments could not; this was particularly clear for the type 1 diabetes scenario which presented an ethical dilemma and invoked heated discussion between participants.

OUTCOMES & EVALUATION

Analyzing the pre-survey data showed that nurses and physicians had similar views about genomics at the start of the workshop prompting us to consider these groups as one population for the purposes of the evaluation. This finding suggests that it would be appropriate to include both nurses and physicians in the study population of a future RCT.

The primary outcome considered participants' views on the overall effectiveness of the intervention as an engagement approach; the majority of participants from both groups agreed that the intervention was an effective approach; however, this was a stronger majority for the in-person intervention (79.4% compared to 62%), which was a statistically significant difference.

A similar pattern emerged for several supporting outcomes, where the majority ranked the interventions positively but the in-person version consistently scored higher. This was true for participants' views as to 1) whether the workshop was an effective way for them to think about PM in their nursing/clinical practice (91% in person vs. 80% online for positive results); 2) whether the workshop raised ideas they had not previously considered (85% in person vs. 72% online for positive results); and 3) whether they learned new information during the workshops (88% in person vs. 79% online for positive results).

On the other hand, the online workshop scored higher than the in-person for participants'

perceptions as to whether the workshop presented information in an unbiased way (86% of online vs. 79% of in-person for positive responses) and whether there was time to think about the issues during the workshop (86% of online vs. 85% of in-person for positive responses).

Despite generally positive evaluations, the online workshop scored much lower than the in-person when it came to participating in a similar workshop in the future, with only the minority of online participants responding positively (48% of online vs. 74% of in-person for positive responses). This is an important indicator from a recruitment perspective, particularly given the challenges we experienced in recruiting HCPs. Given that the online participants had generally evaluated the workshop positively, this also raises the question of whether they interpreted this question differently from the in-person group, perhaps to mean would they receive e-mail invitations for future surveys. We recommend that this outcome measure be revised in a future evaluation to clarify that the participants are not being asked to participate in a future workshop.

After consultation with a biostatistician we performed a series of statistical analyses, restricting the results by professional group and using regression models to control for HCP; the analysis supported the decision to combine HCPs into one category, with all statistical tests indicating that for the primary outcome the in-person workshop performed better than the online workshop after controlling for HCP. While the observed difference in the primary outcome was statistically significant it is important to consider whether this difference is in fact ‘clinically significant’ in light of the recruitment for the in-person workshop described above.

In terms of the choice of outcome scale, no obvious floor or ceiling effects emerged; however, given the relatively small percentage of people who evaluated the interventions negatively it might be advisable to use a seven point Likert scale to better discriminate between

outcomes.

This thesis provided a preliminary qualitative evaluation and presented over-arching themes which were compared between the two interventions; future work would require a more sophisticated approach to quantifying the qualitative data, including ways to measure how long each approach takes to reach saturation. Considering that the pilot indicated that online responses tended to be more detailed, the ability to quantify this level of detail would be another means of comparing interventions.

6.4 EVIDENCE FROM PRELIMINARY FINDINGS ABOUT GENOMICS

Based on the overall positive evaluation for both interventions, we concluded that both interventions were sufficiently ‘valid’ to tentatively group the substantive findings about genomics into a larger set of results combining engagement data from both interventions. Given that this was a pilot study the engagement results can only be considered preliminary findings.

The results showed that majority of respondents felt that CRC risk stratification would help patients (71%) and believed their patients would expect them to recommend such a test (82%), in addition 78% thought they should be capable of ordering this test, suggesting that the majority of HCPs would consider adopting genomic test ordering for CRC risk as part of their professional role and identity. Given these very high percentages it was surprising that just over 50% of HCPs would actually recommend this test to their patients. This suggests a disconnect between HCPs’ perceptions of the clinical utility of genomic tests and their intention to implement these tests in clinical practice.

The type 1 diabetes scenario elicited very different reactions with only 33% of respondents feeling that such a test has clinical utility and a similar percentage believing testing would be beneficial to parents and children. Despite lack of perceived benefit, 71% of HCPs still

believed parents would expect them to recommend this test, suggesting that HCPs may face situations where they are asked to recommend genomic screening tests they do not believe to be useful. Although the numbers were not as high as with the CRC case study, the majority (61%) still believed they should be capable of ordering this test as part of their professional role. Only 23% of HCPs would actually recommend this test and those that would recommend it were less likely to think this test should be available to all patients, considering parental traits such as high levels of anxiety and poor risk management to be potential reasons for withholding testing.

Responses to the family history case study were overwhelmingly positive with 84% of patients feeling positive that this would benefit patients and 67% believing their patients would expect them to implement a standardized system. The majority of participants (69%) indicated they would use such a system instead of their current approach, although several commented that they disagreed with this statement because they would prefer to use a standardized system in conjunction with their current approach.

6.5 STUDY LIMITATIONS

This study was limited by selection bias and a small sample size. Even with significant efforts and help from multiple people recruitment was extremely difficult, suggesting that our aspiration to be early for health technology assessment may have been too ambitious. While the interventions had a strong theoretical grounding and received generally positive evaluations we are forced to consider whether it is feasible to do this sort of professional engagement exercises given the time and money available.

Lower than anticipated recruitment meant that it was not possible to conduct a concurrent RCT with random assignment to intervention format. Instead we used a quasi-experimental study design with two non-concurrent volunteer samples. The quasi-experimental design provided a

practical compromise, taking into account the realities of recruiting professionals for engagement exercises while at the same time providing useful and extensive data. Grouping was performed based on feasibility and convenience; however, without randomization it is unlikely that the two intervention groups will be as similar as if they had been assigned randomly (hence the term ‘non-equivalent groups design’) and allocation bias can be significant threat to internal validity. We conducted a series of statistical analysis to account for the differences in distribution of HCPs between intervention groups and concluded that for the purposes of this pilot study the two samples are similar enough to make useful comparisons about the interventions; however without random sampling these results are not generalizable to the whole population of Canadian primary care professionals. For this reason, the study maintained a hypothesis-generating rather than hypothesis-testing perspective.

6.6 LESSONS LEARNED FOR A FUTURE EVALUATION

An obvious question for a future RCT is whether we would modify the interventions or work from the approaches as delivered in the pilot. Our experience indicates that those who chose to participate in the in-person version ranked the intervention highly and we do not recommend changes to the educational component; however, the in-person data collection could be improved. The pilot showed that writing booklets were not an effective means of capturing participants’ views, obtaining consent to record the engagement discussion would have been helpful and it would be worthwhile to explore innovative technological approaches to capture participants’ free form thoughts. The time allotted for the pilot workshop was short, in a future version it would be helpful if participants could read and sign research forms before the workshop.

The delivery of the online intervention could also be improved. The pilot showed that

while the online evaluations were generally positive only 35% of those who landed on the page actually completed the survey. At this stage the online version was by necessity a prototype developed by CC with minimal resources; while the evaluations indicate that it has promise, the online arm of the intervention in an RCT would need to be of higher quality and professionally produced, given that HCPs are an internet savvy population and sophisticated in how they assess material. Future work could evaluate whether there is benefit in incorporating some form of group dynamic into the online version; a possible suggestion would be to connect with the Evans Health Lab at the University of Ottawa (Evans, 2014).

Given that the pilot indicated that the online workshop represented an easier, more convenient and less expensive approach to engagement, a future RCT could evaluate whether the online intervention was non-inferior to the in-person focus group, which is logistically more difficult to arrange and more demanding of both the participants' and moderator's time. A non-inferiority trial would be applicable here because the purpose is to determine whether the online intervention is no worse than the in-person intervention which is appropriate in situations such as this where the alternative intervention is not necessarily expected to be better than the standard intervention in terms of efficacy, but may be better regarding secondary endpoints such as cost and convenience (Lesaffre, 2008).

A future RCT would need to concentrate on strategies to increase recruitment. Based on discussions with other researchers at the American Society of Human Genetics (ASHG) 63rd Annual Meeting in 2013 we are not alone in experiencing difficulties engaging with professionals about PM; for example, despite offering a substantial financial incentive, researchers from the Veterans Affairs Boston Healthcare System experienced the same challenges recruiting primary care physicians and cardiologists to participate in their survey on

the clinical utility of family history and whole-genome sequencing.

Considering the pilot nature of this study and its inherent limitations neither intervention emerged as being clearly ‘better’. While the in-person version was evaluated more positively by HCPs based on the primary outcome, the online version also received positive evaluations, was logistically more feasible from a recruitment and implementation perspective, was considered to be less biased, and was surprisingly effective at generating qualitative data. If the online version was demonstrated to be at least as effective as the in-person approach in terms of engaging professionals and generating useful data, then it could well be considered to be the approach of choice: it was easier and less expensive to implement and was clearly preferred by physicians. Our experience with poor recruitment for in-person activities is consistent with what is described in the literature. The findings of this pilot study need to be confirmed in a more rigorous RCT with an economic evaluation, but may point the way to a practical approach for health professional engagement activities. The overall positive evaluations and wealth of engagement data obtained from both versions of the intervention support an argument to proceed to a ‘phase 3’ RCT, with the prototype online version re-developed to higher production standards.

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APPENDIX 1: LETTERS OF INVITATION, PARTICIPANT INFORMATION FORM, CONSENT FORM

USING GENOMICS IN HEALTH CARE: NURSING PROFESSIONALS STUDY PARTICIPANT INFORMATION FORM

Principal Investigator: Brenda Wilson

Purpose of the Research

We are exploring nurses' professional perspectives on emerging genomic technologies in health care. The results of this study will be used to develop policy briefs and knowledge translation material for health policy decision makers and providers of health care.

Description of the Research

We have approached you to take part in this study because you are a nursing professional in Canada. This workshop is part of a larger program of research looking at new applications of genomics and related technologies in health care.

Description of Procedures

This study essentially consists of one questionnaire, in which the questions will be asked in stages. The questions are about aspects of actual and hypothetical applications of genomics in routine health care. You do not need specialist genetics knowledge to answer them. The researcher will provide further instructions as necessary. The study should take no more than one and a half hours to complete.

At the end of the questionnaire, we will debrief you further about the purpose of this research.

Length of Study

The study overall is expected to take about 2 years, including several workshops and data analysis. Each participant is asked to attend one study workshop.

Possible Risks

There are no physical or other risks presented by participating in this study. You do not have to answer any questions that make you uncomfortable.

Benefits to Your Participation

There are no benefits to you participating in this study. However, the knowledge we gain is necessary to inform a broader understanding on the part of policy decision makers of nurses' professional reactions to emerging genome based technologies.

Voluntary Participation

You are under no obligation to participate in this study. You may choose to withdraw at any time, including during the workshop, without giving any reason.

Expenses

When you join the study, we allocate you a unique code. Your completed questionnaire has your code, but not your name, on it. Your signed consent form, which includes your name, will be held securely in a separate location. During data collection, analysis and report writing, the Principal Investigator will hold a master list of the participants' names and their codes, separate from the data. If you decide to withdraw from the study, we will use this master list to locate and destroy your questionnaire.

All responses will be entered onto a computer database for analysis, with no personally identifying information. Paper-based data will be kept in a locked cabinet and computer data saved in password-protected files. The Principal Investigator and co-investigators directly involved with the study, and selected research staff, will have access to paper and computer files. Once the study is complete, including analyses and report writing, the codes will be removed from the electronic data, rendering them permanently anonymous.

All personal health information will be kept confidential, unless release is required by law. When the results are analyzed and written in scientific papers, we may include some direct quotes from answers given in the questionnaires. However, they will be presented in a way which cannot be linked to any given participant. The study data will be kept for 15 years after termination of the study and then destroyed. The Ottawa Hospital Research Ethics Board and the Ottawa Health Research Institute may review your relevant study records for audit purposes.

Questions

Dr. Stuart Nicholls is the project Postdoctoral Fellow, and the first point of contact for questions. You are also welcome to contact the Principal Investigator, Dr. Brenda Wilson who will discuss any aspect of the project without any obligation on your part.

If you have any questions regarding your rights as a research subject, you may contact the Chairperson of the Ottawa Hospital Research Ethics Board. The chairperson cannot provide any medical information with regard to this study.

USING GENOMICS IN HEALTH CARE:
HEALTH CARE PROFESSIONALS STUDY
PARTICIPANT INFORMATION FORM

Principal Investigator: Brenda Wilson

Purpose of the Research

We are exploring professional perspectives on emerging genomic technologies in health care. The results of this study will be used to develop policy briefs and knowledge translation material for health policy decision makers and providers of health care.

Description of the Research

We have approached you to take part in this study because you are a practicing health professional or medical student in Canada. This workshop is part of a larger program of research looking at new applications of genomics and related technologies in health care

Description of Procedures

This study essentially consists of one questionnaire, in which the questions will be asked in stages. The questions are about aspects of actual and hypothetical applications of genomics in routine health care. You do not need specialist genetics knowledge to answer them. The researcher will provide further instructions as necessary. The study should take no more than one and a half hours to complete.

At the end of the questionnaire, we will debrief you further about the purpose of this research.

Length of Study

The study overall is expected to take about 2 years, including several workshops and data analysis. Each participant is asked to attend one study workshop.

Possible Risks

There are no physical or other risks presented by participating in this study. You do not have to answer any questions that make you uncomfortable.

Benefits to Your Participation

There are no benefits to you participating in this study. However, the knowledge we gain is necessary to inform a broader understanding on the part of policy decision makers of nurses professional reactions to emerging genome based technologies.

Voluntary Participation

You are under no obligation to participate in this study. You may choose to withdraw at any time, including during the workshop, without giving any reason.

Ethics

When you join the study, we allocate you a unique code. Your completed questionnaire has your code, but not your name, on it. Your signed consent form, which includes your name, will be held securely in a separate location. During data collection, analysis and report writing, the Principal Investigator will hold a master list of the participants' names and their codes, separate from the data. If you decide to withdraw from the study, we will use this master list to locate and destroy your questionnaire.

All responses will be entered onto a computer database for analysis, with no personally identifying information. Paper-based data will be kept in a locked cabinet and computer data saved in password-protected files. The Principal Investigator and co-investigators directly involved with the study, and selected research staff, will have access to paper and computer files. Once the study is complete, including analyses and report writing, the codes will be removed from the electronic data, rendering them permanently anonymous.

All personal health information will be kept confidential, unless release is required by law. When the results are analyzed and written in scientific papers, we may include some direct quotes from answers given in the questionnaires. However, they will be presented in a way which cannot be linked to any given participant. The study data will be kept for 15 years after termination of the study and then destroyed. The Ottawa Hospital Research Ethics Board and the Ottawa Health Research Institute may review your relevant study records for audit purposes.

Questions

Dr. Stuart Nicholls is the project Postdoctoral Fellow, and the first point of contact for questions. You are also welcome to contact the Principal Investigator, Dr. Brenda Wilson who will discuss any aspect of the project without any obligation on your part.

If you have any questions regarding your rights as a research subject, you may contact the Chairperson of the Ottawa Hospital Research Ethics Board at. The chairperson cannot provide any medical information with regard to this study.

USING GENOMICS IN HEALTH CARE:
HEALTH PROFESSIONALS STUDY
CONSENT FORM

I have read this three-page Participant Information Form and this Consent Form and have had an opportunity to ask Dr. Stuart Nicholls, or Dr. Brenda Wilson any questions I may have had about the study.

My questions and/or concerns have been answered to my satisfaction and I agree to my participation in this study. If I decide at a later stage in the study that I would like to withdraw my consent, I may do so at any time.

I will not be identifiable in any publications or presentations resulting from this study. All information will be coded and I will not be identifiable by name. No identifiable information will leave the Ottawa Hospital.

A copy of the Information and Consent Form will be provided to me.

I hereby consent to participate.

Name of participant (Please print)

Signature of participant

Date: _____

Investigator Statement (or Person Explaining the Consent)

I have carefully explained to the research participant the nature of the above research study. To the best of my knowledge, the research participant signing this consent form understands the nature, demands, risks and benefits involved in participating in this study. I acknowledge my responsibility for the care and well being of the above research participant, to respect the rights and wishes of the research participant, and to conduct the study according to applicable Good Clinical Practice guidelines and regulations.

Name of investigator/delegate (Please print)

Signature of investigator/delegate

NURSING LETTER OF INVITATION

[Date]

Dear Nursing Professional

We would like to invite you to participate in a research project on the use of genomic technologies in health care. You have been identified through [name of professional association, department, or other source of referral] as a nursing professional. We are seeking nursing professionals who have no special expertise in genetics to participate in a research workshop exploring reactions to possible applications of genomics in routine health care.

To provide a brief background, DNA-based genomic profiling is a method which combines information on several genetic loci to give an 'integrated' profile of a person's susceptibility to a complex disorder, such as cancer, diabetes, or cardiovascular disease. The routine application of this approach to risk assessment and disease screening in healthy people is some way off. However, there are a number of research groups already proposing the use of genetic information in ways which go well beyond traditional medical genetics, for example screening newborns for familial hypercholesterolaemia as a way of identifying parents who are affected but asymptomatic; or testing young adults for susceptibility to type 2 diabetes.

There are many unexplored issues in considering whether and how such technologies should be introduced into Canadian health services. We would like you to consider participating in a research workshop which has been specially developed to allow exploration of these topics. Attached is a Participant Information Form with more details. A member of our research team [Dr. Stuart Nicholls] will follow up with you in the next week or so to find out if you are interested. If you agree to take part, he will send you information on the date and location of the workshop.

This research is designed to improve the way we provide health services, and we depend on professional participation. Responding positively to this invitation places you under no obligation to the University of Ottawa or The Ottawa Hospital, and does not commit you to actually taking part in the workshop if you change your mind.

If you would like to find out more before Dr. Nicholls contacts you, please call him at _____, or contact me [Brenda Wilson] directly.

Sincerely

Dr. Brenda Wilson
Principal Investigator

PHYSICIAN LETTER OF INVITATION

Dear [name]

We would like to invite you to participate in a research project on the use of genomic technologies in health care. You have been identified through [name of professional association, department, or other source of referral] as a health professional. We are seeking health professionals who have no special expertise in genetics to participate in a research workshop exploring reactions to possible applications of genomics in routine health care.

To provide a brief background, DNA-based genomic profiling is a method which combines information on several genetic loci to give an 'integrated' profile of a person's susceptibility to a complex disorder, such as cancer, diabetes, or cardiovascular disease. The routine application of this approach to risk assessment and disease screening in healthy people is some way off. However, there are a number of research groups already proposing the use of genetic information in ways which go well beyond traditional medical genetics, for example screening newborns for familial hypercholesterolaemia as a way of identifying parents who are affected but asymptomatic; or testing young adults for susceptibility to type 2 diabetes.

There are many unexplored issues in considering whether and how such technologies should be introduced into Canadian health services. We would like you to consider participating in a research workshop which has been specially developed to allow exploration of these topics. Attached is a Participant Information Form with more details. A member of our research team [Dr. Stuart Nicholls] will follow up with you in the next week or so to find out if you are interested. If you agree to take part, he will send you information on the date and location of the workshop.

This research is designed to improve the way we provide health services, and we depend on professional participation. Responding positively to this invitation places you under no obligation to the University of Ottawa or The Ottawa Hospital, and does not commit you to actually taking part in the workshop if you change your mind.

If you would like to find out more before Dr. Nicholls contacts you, please call him at _____, or contact me [Brenda Wilson] directly.

Sincerely

Dr. Brenda Wilson
Principal Investigator

APPENDIX 2: LETTERS OF APPROVAL FROM OTTAWA HEALTH SCIENCE NETWORK RESEARCH ETHICS BOARD AND NEWFOUNDLAND HEALTH RESEARCH ETHICS AUTHORITY



Ottawa Hospital Research Ethics Boards / Conseils d'éthique en recherches

751 Parkdale Avenue Suite 106, Ottawa, Ontario K1Y 1J7 613-798-5555 ext. 14902 Fax: 613-761-4311
<http://www.ohri.ca/ohreb>

Friday, May 22, 2009

Dr. Brenda Wilson
University of Ottawa

Dear Dr. Wilson:

Re: Protocol # 2009275-01H CIHR Emerging Team in Genetics in Screening: Public Engagement Study

Protocol approval valid until - Wednesday, July 22, 2009

Thank you for your e-mail dated May 14, 2009. I am pleased to inform you that this protocol underwent expedited review by the Ottawa Hospital Research Ethics Board (OHREB) and is approved for two months to start recruiting English-speaking participants. No changes, amendments or addenda may be made to the protocol or the consent form without the OHREB's review and approval.

Approval is for the following documentation:

- Research Proposal dated July 14, 2008, received March 27, 2009
- English and French Health Professional Invitation Letter dated April 27, 2009
- English and French Public Invitation Letter dated April 27, 2009
- English Health Professionals Study Participant Information Form dated April 27, 2009
- English Public Information Form dated April 27, 2009

Upon receipt and review of the two French Information Form/Consents, the protocol may be extended to May 21, 2010 (one year from the initial approval date), and the recruitment of French-speaking participants may commence. When submitting the French documentation to the OHREB, confirm that it has been translated or approved by Eric Lepine (email all documentation to Eric at elepine@ohri.ca).

The validation date should be indicated on the bottom of all consent forms and information sheets (see copy attached).

The Ottawa Hospital Research Ethics Board is constituted in accordance with, and operates in compliance with the requirements of the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans; Health Canada Good Clinical Practice: Consolidated Guideline; Part C Division 5 of the Food and Drug Regulations of Health Canada; and the provisions of the Ontario Health Information Protection Act 2004 and its applicable Regulations.

Raphael Saginur, M.D.
Chairman
Ottawa Hospital Research Ethics Board

RS/cb



Ottawa Hospital Research Ethics Boards / Conseils d'éthique en recherches

751 Parkdale Avenue Suite 106, Ottawa, Ontario K1Y 1J7 613-798-5555 ext. 14902 Fax: 613-761-4311
<http://www.ohri.ca/ohreb>

Monday, June 22, 2009

Dr. Brenda Wilson
University of Ottawa



Dear Dr. Wilson:

Re: Protocol # 2009275-01H CIHR Emerging Team in Genetics in Screening: Public Engagement Study

Thank you for your letter dated June 17, 2009. The French Health Professionals Study Participant Information Form dated April 27, 2009 and the French Public Information Form dated April 27, 2009 are approved and the recruitment of French-speaking participants may begin.

The study expiry date has now been extended to May 21, 2010.



Chairman
Ottawa Hospital Research Ethics Board

Encl.

/cb

November 20, 2013

Re: 2009275-01H “CIHR Emerging Team in Genomics in Screening: Public Engagement Study”

Dear Dr Saginur

On May 23, 2013 our amendment was approved to conduct professional engagement workshops as an online survey with embedded videos. I wish to request that this online component of the professional engagement workshop is provided at this stage as an English-only component.

I fully respect, and am committed to, the goal of language inclusivity in health research, and I would point to the evaluation component of the in-person version of this exercise having been developed in French as well as English. The reason that I request this exemption for the online version is primarily because this is a pilot project being conducted as master's thesis research, and therefore has significant time and cost constraints. These have been compounded by the technical challenges faced in producing the online engagement exercise, which integrates video animation and carefully timed audio recording as well as the survey questions. The video component incorporates specific terminology and phrasing.

The goal of this small pilot project is to inform a future definitive bilingual version, which would be professionally developed.

If you have any questions, please contact me or Dr. Stuart Nicholls.

Thank you for your consideration of this request.

Sincerely

Dr Brenda J Wilson

November 20, 2013

Re: 2009275-01H “CIHR Emerging Team in Genomics in Screening: Public Engagement Study” – submission of online content

Dear Dr Saginur

On May 23, 2013 our amendment was approved to conduct in-person and online versions of health professional engagement workshops. At the time of approval the online version was incomplete, as we had prioritized the in-person version. A condition of the amendment approval was that we submit the online version once ready, and before being put into use.

We now submit the online version, administered using FluidSurveys, which incorporates a survey component and embedded video presentations. The target audience and survey content is identical to that of the in-person workshop (already approved). The video content is similar to that of the in-person workshop but has been condensed. These videos can be viewed from within FluidSurveys by accessing the following link:

We also attach scripts of the four videos.

If you have any questions, please contact me or Dr. Stuart Nicholls.

Sincerely

Dr Brenda J Wilson



Ethics Office
Suite 200, Eastern Trust Building
95 Bonaventure Avenue
St. John's, NL
A1B 2X5

November 2, 2012



Dear Dr. Etchegary:



**RE: Pilot study of professional engagement approaches on personalized medicine:
Newfoundland and Labrador component**

At the meeting held on **November 1, 2012**, the health Research Ethics Board has reviewed your application and **granted full board** approval as submitted.

Full board approval of this research study is granted for one year effective **November 1, 2012**.

This approval will lapse on **October 31, 2013**. It is your responsibility to ensure that the Ethics Renewal form is forwarded to the HREB office prior to the renewal date. *The information provided in this form must be **current to the time of submission** and submitted to the HREB **not less than 30 nor more than 45 days** of the anniversary of your approval date.* The Ethics Renewal form can be downloaded from the HIC website <http://www.hrea.ca>

The health Research Ethics Board advises THAT IF YOU DO NOT return the completed Ethics Renewal form prior to date of renewal:

- *Your ethics approval will lapse*
- *You will be required to stop research activity immediately*
- *You may not be permitted to restart the study until you reapply for and receive approval to undertake the study again*

Lapse in ethics approval may result in interruption or termination of funding

email: info@hrea.ca

Phone: 777-8949

FAX: 777-8776

It is **your responsibility to seek the necessary approval from the Regional Health Authority or other organization as appropriate.**

Modifications of the protocol/consent are not permitted without prior approval from the Health Research Ethics Boards. Implementing changes in the protocol/consent without HREB approval may result in the approval of your research study being revoked, necessitating cessation of all related research activity. Request for modification to the protocol/consent must be outlined on an amendment form (available on the HREA website) and submitted to the HREB for review.

This research ethics board (the HREB) has reviewed and approved the research protocol and documentation as noted above for the study which is to be conducted by you as the qualified investigator named above at the specified site. This approval and the views of this Research Ethics Board have been documented in writing. In addition, please be advised that the Health Research Ethics Board currently operates according to *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans*; *ICH Guidance E6: Good Clinical Practice* and applicable laws and regulations. The membership of this research ethics board is constituted in compliance with the membership requirements for research ethics boards as defined by *Health Canada Food and Drug Regulations Division 5; Part C*.

Notwithstanding the approval of the HREB, the primary responsibility for the ethical conduct of the investigation remains with you.

We wish you every success with your study.

Sincerely,


Dr. Fern Brunger
Chair, Non-Clinical Trials
Health Research Ethics Board

C C VP Research c/o Office of Research, MUN
VP Research c/o Patient Research Centre, Eastern Health

email: info@hrea.ca

Phone: 777-8949

FAX: 777-8776

Using Genomics in Health Care: Health Professionals [Online] Workshop

- 1) Pre-workshop survey
- 2) Writing booklet
- 3) Post-workshop survey

All your answers will be kept confidential
Your name is not on the questionnaire

**Please wait for further instructions
before opening the booklet**

Pre [online] workshop survey

This is what genomic medicine means to me. (Check all that apply):

- ☐ Taking a family history
 - ☐ Screening
 - ☐ Stratifying risk of disease in a healthy patient
 - ☐ Making a treatment decision
 - ☐ Making a prognostic prediction
 - ☐ Something else - please describe
-

In my opinion, genomic medicine will be useful in my clinical practice.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

Irrespective of the nature of the test, being able to more accurately quantify a patient's risk of colorectal cancer would make a difference to my clinical decision-making.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

Irrespective of the nature of the test, being able to identify infants at higher than average risk of type 1 diabetes would make a difference to my clinical decision-making.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

On the whole, I find family history information to be a useful tool in my everyday practice.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

Irrespective of the nature of the test, being able to more accurately predict a patient's response to a drug would make a difference to my clinical decision-making.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

Writing booklet

As we go through the workshop, please use these pages to capture your thoughts, as they occur, on any aspects of genomic medicine.

You may write in prose, bullet points, or any unstructured format.

We can supply more pages if you run out of space!

Writing booklet

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We can supply more pages if you run out of space!

[illegible]

Post [online] workshop survey

COLORECTAL CANCER

Irrespective of the nature of the test, being able to more accurately quantify a patient's risk of colorectal cancer would make a difference to my clinical decision-making.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

If a properly validated genomic profiling test for colorectal cancer became available,

I think it would generally

Help patients **1** **2** **3** **4** **5** **Harm patients**

I think most of my patients would expect me to recommend it

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

I think family physicians should be capable of ordering it

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

I would recommend it to patients

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

If you **agree** or **strongly agree** that you would recommend this test to patients, are there any groups of patients for whom you would not recommend it? Check all that apply.

- ☐ Patients not yet eligible for standard colorectal cancer screening (e.g., under 50)
- ☐ Patients without a family history of colorectal cancer
- ☐ Patients who don't actively request this kind of test
- ☐ Patients who are not good at managing their health risks
- ☐ Patients who are anxious
- ☐ None (I would recommend this test for everyone)
- ☐ Other, please specify:

DIABETES

Irrespective of the nature of the test, being able to identify infants at higher than average risk of type 1 diabetes would make a difference to my clinical decision-making.

Strongly agree 1 2 3 4 5 Strongly disagree

If a properly validated genomic profiling test for type 1 diabetes risk in infants became available,

I think knowing the test result would generally

Help children 1 2 3 4 5 Harm children

I think knowing the test result would generally be beneficial to parents

Strongly agree 1 2 3 4 5 Strongly disagree

I think most parents would expect me to recommend this test

Strongly agree 1 2 3 4 5 Strongly disagree

I think family physicians should be capable of ordering this test

Strongly agree 1 2 3 4 5 Strongly disagree

I would recommend this test to parents

Strongly agree 1 2 3 4 5 Strongly disagree

If you **agree** or **strongly agree** that you would recommend this test, are there any groups of patients for whom you would not recommend it? Check all that apply.

- ☐ Infants of parents who don't actively request the test for their child
 - ☐ Infants where there is no family history of type 1 diabetes
 - ☐ Infants of parents who are not good at managing their own health risks
 - ☐ Infants of parents who are anxious
 - ☐ None (I would recommend this test for all newborns)
 - ☐ Other, please specify
-

FAMILY HISTORY

On the whole, I find family history information to be a useful tool in my everyday practice.

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

In what kinds of situations do you actively ask a patient about his or her family history?

- ☐ New patient intake
- ☐ Periodic health examination/assessment
- ☐ When a patient presents with a new complaint
- ☐ Other, please specify:

What kind of approach do you use to capture family history information? (Check all that apply)

- ☐ I ask unstructured questions
- ☐ I ask a standard set of questions
- ☐ I use a family history tool designed for health professionals
- ☐ I ask patients to complete a family history tool
- ☐ I refer to a nurse or nurse practitioner to gather the information
- ☐ Other, please specify:

If the province promoted a standardized approach to family history taking,

I think it would generally:

Help patients **1** **2** **3** **4** **5** **Harm patients**

I would use the system instead of my current approach

Strongly agree **1** **2** **3** **4** **5** **Strongly disagree**

I think most of my patients would expect me to use such a system

Strongly agree 1 2 3 4 5 Strongly disagree

GENOMIC MEDICINE AND ME

This is what genomic medicine means to me. (Check all that apply):

- ☐ Taking a family history
 - ☐ Screening
 - ☐ Stratifying risk of disease in a healthy patient
 - ☐ Making a treatment decision
 - ☐ Making a prognostic prediction
 - ☐ Something else - please describe
-

In my opinion genomic medicine will be useful in my clinical practice.

Strongly agree 1 2 3 4 5 Strongly disagree

If properly validated genomic tests for chronic disease risk or to guide prescribing became available, which of the following would help you work out how to use them in your own practice? (Check all that apply)

- ☐ I would not wish to integrate them into my own practice
 - ☐ Specific point of care tools to use within consultation (e.g., guidelines, mobile apps)
 - ☐ Targeted CME/CPD sessions on specific tests
 - ☐ Easy access to a genetic counselor
 - ☐ A colleague in my clinic prepared to become an expert
 - ☐ Web-based support for answering questions and helping me assess patients' risk
 - ☐ Other, please specify:
-

If a standardized approach to family history taking was recommended, which of the following would help you integrate it into your own practice? (Check all that apply)

- ☐ I would not wish to change my approach to family history taking

- ☐ A family history tool that my patients could fill in (paper, web-based, mobile app)
 - ☐ A structured family history tool that could be used to gather information in telephone interviews
 - ☐ A family history tool that I could use myself (paper, web-based, mobile app)
 - ☐ A family history tool that linked data directly to an electronic medical record
 - ☐ Other, please specify:
-

THE [ONLINE] WORKSHOP

This [online] workshop was an effective way to make me think about genomic medicine in my own clinical practice

Strongly agree 1 2 3 4 5 Strongly disagree

This [online] workshop was an effective way for me to express my views on genomic medicine as it might relate to my own clinical practice

Strongly agree 1 2 3 4 5 Strongly disagree

I would participate in a similar [online] workshop in the future

Strongly agree 1 2 3 4 5 Strongly disagree

The [online] workshop raised ideas I had not previously considered

Strongly agree 1 2 3 4 5 Strongly disagree

I learned new information during the [online] workshop

Strongly agree 1 2 3 4 5 Strongly disagree

The [online] workshop presented information in an unbiased way

Strongly agree 1 2 3 4 5 Strongly disagree

I had enough time to think about the issues during the [online] workshop

Strongly agree 1 2 3 4 5 Strongly disagree

PROFESSIONAL DEMOGRAPHIC INFORMATION

What is your age? _____

What is your sex?

- ☐ Male
☐ Female

Are you a

- ☐ Primary Care Physician
☐ Specialist Physician
☐ Resident
☐ Medical Student
☐ Pharmacist
☐ Other, please specify:
-

How many years have you been practicing (not including internship but including residency)?

- ☐ 0-4
☐ 5-9
☐ 10-14
☐ 15-19
☐ 20 or more
☐ I am not in clinical practice

With respect to the patient care setting where you see most of your patients (if appropriate), which population primarily does it serve? (Please check only one)

- ☐ Urban
☐ Small urban (10,000-25,000)
☐ Rural (<10,000)
☐ I am not involved in patient care

Do you have any training in genetics?

- ☐ No formal education in genetics
☐ Yes. Please specify:
-

Do you have a special professional interest in cancer screening or treatment?

- ☐ No
☐ Yes. Please specify:

Do you have a special professional interest in paediatrics?

- ☐ No
☐ Yes. Please specify:

Do you have a special professional interest in diabetes or endocrinology?

- ☐ No
☐ Yes. Please specify:
