

**Understanding the impact of the Canadian Paediatric Society's
hyperbilirubinemia guidelines in Ontario:
A population health perspective**

by

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List of Abbreviations

CPS: Canadian Paediatric Society
SES: Socio-economic Status
TSB: Total Serum Bilirubin
RR: Relative Risk
CI: Confidence Interval
IOM: Institute of Medicine
LOS: Length of Stay
ED: Emergency Department
BORN: Better Outcomes Registry and Network
LHIN: Local Health Integration Network
TCB: Transcutaneous Bilirubin
GEE: Generalized Estimating Equations
ICES: Institute for Clinical Evaluative Sciences
DAD: Discharge Abstract Database
NACRS: National Ambulatory Care Reporting System
GP: General Practitioner
QIC: Quasilielihood under the Independence model Criterion
CIHI: Canadian Institute for Health Information
OHIP: Ontario Health Insurance Plan
MOHLTC: Ministry of Health and Long-Term Care
PCMCH: Provincial Council for Maternal Child Health
AIC: Akaike Information Criterion
SII: Slope Index of Inequality
RII: Relative Index of Inequality
SEP: Socioeconomic Position
BGV: Between Group Variance

Abstract

In 2007, the Canadian Paediatric Society (CPS) released a guideline aimed at preventing complications of neonatal jaundice through universal screening and guidelines for follow-up and treatment. This thesis investigates the impact of implementation of the CPS guideline on health services utilization at a population level in Ontario. First, we surveyed all Ontario hospitals providing maternal-newborn services to determine if and when they had implemented universal bilirubin screening, and to gather information about the organization of services to provide follow-up and treatment, and about the factors that influenced screening implementation. Then we conducted two population-based cohort studies using linked administrative health data to evaluate the association between 1) the implementation of universal bilirubin screening and phototherapy use (during and following birth hospitalization) length of stay (LOS), jaundice-related emergency department (ED) visits and readmissions; and 2) universal bilirubin screening implementation and access to recommended follow-up care by socio-economic status (SES). By 2012, the majority of Ontario hospitals had implemented universal bilirubin screening. There is heterogeneity in how hospitals organize services, but a notable trend towards hospital-based post-discharge care. Screening was associated with an increase in phototherapy during hospitalization at birth (relative risk (RR) 1.32, 95% confidence interval (CI) 1.09-1.59), and a decrease in jaundice-related ED visits (RR 0.79, 95% CI 0.64-0.96), but no statistically significant difference in phototherapy after discharge, length of stay, or jaundice-related readmissions after accounting for pre-existing temporal trends in healthcare service use and other patient socio-demographic and hospital characteristics. Implementation of the universal bilirubin screening in Ontario was associated with a modest increase in rates of early follow-up (adjusted RR 1.11, CI 1.0014-1.22, $p=0.0468$), but most babies were not seen within the recommended timeframe. Babies of lowest SES were least likely to receive recommended follow-up, and disparities in follow-up increased following universal bilirubin screening implementation.

En 2007, la Société canadienne de pédiatrie (SCP) a publié une directive visant à la prévention des complications de l'ictère néonatal par le dépistage universel et des lignes directrices pour le suivi et le traitement. Cette thèse étudie l'impact de la mise en œuvre de la directive SCP sur l'utilisation des services de santé à niveau de population de l'Ontario. Tout d'abord, nous avons

interrogé tous les hôpitaux de l'Ontario offrant des services de santé maternelle-nouveau-né afin de déterminer si et quand ils avaient mis en œuvre le dépistage universel de la bilirubine, et à recueillir des informations sur l'organisation des services pour assurer un suivi et de traitement, et sur les facteurs qui ont influencé la mise en œuvre de dépistage. Ensuite, nous avons mené deux études de cohorte basée sur la population à partir de données administratives sur la santé pour évaluer 1) l'association entre la mise en œuvre du dépistage de la bilirubine universel et la photothérapie utilisation lors de l'hospitalisation à la naissance, la photothérapie après avoir sortie de l'hôpital, la durée du séjour, le service des urgences liées à la jaunisse et des réadmissions liées à la jaunisse; et 2) l'association entre la mise en œuvre du dépistage universel et l'accès aux soins de suivi recommandés et si cela différait entre les quintiles de statut socioéconomique. En 2012, la majorité des hôpitaux de l'Ontario a mis en œuvre le dépistage universel de la bilirubine. Il existe une hétérogénéité de la façon dont les hôpitaux organisent des services, mais une tendance notable vers les soins post-décharge en milieu hospitalier. Le dépistage a été associé à une augmentation de la photothérapie pendant l'hospitalisation à la naissance (risque relatif (RR) de 1,32, intervalle de confiance 95 % (IC 95 %) de 1,09 à 1,59), et une diminution des visites à l'urgence liées à la jaunisse (RR 0,79, IC 95 % 0,64 à 0,96), mais aucune différence statistiquement significative dans la photothérapie après la sortie, la durée du séjour, ou réadmissions liées jaunisse - après comptabilisation des tendances temporelles pré-existants dans l'utilisation des services de soins de santé et d'autres caractéristiques socio-démographiques des patients et caractéristiques de l'hôpital. La mise en œuvre de le dépistage universel en Ontario a été associée à une légère augmentation des taux de suivi précoce (RR ajusté 1,11; IC de 1,0014 à 1,22; $p = 0,0468$), mais la plupart des bébés n'ont pas été vues dans les délais recommandés. Les bébés de statut socioéconomique faibles étaient moins susceptibles de recevoir de soins de suivi recommandés et les disparités dans le suivi ont augmenté suite à la mise en œuvre du dépistage universel de la bilirubine.

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Chapter 1

Introduction

1.1 Thesis Background

This thesis is an investigation of how the implementation of a clinical practice guideline developed by the Canadian Paediatric Society (CPS), has had an impact on health services utilization at a population level within the province of Ontario. The guideline of interest is the CPS's position statement titled '*Guidelines for the detection, management and prevention of hyperbilirubinemia in term and late preterm newborn infants (35 or more weeks' gestation)*'. The guideline is aimed at preventing a rare but serious complication of neonatal jaundice, chronic bilirubin encephalopathy, through a strategy that involves universal screening of newborns and guidelines for follow-up and treatment.

This chapter of the thesis begins by providing basic background information regarding neonatal hyperbilirubinemia, the root causes of severe hyperbilirubinemia, and the state of the evidence on universal bilirubin screening. I then outline the theoretical concepts that have shaped the research questions and design for this thesis, which draw on various elements of population health theory as well as the limited literature on socio-economic status (SES) and neonatal hyperbilirubinemia. Finally, I provide an overview of the structure of the main body of the thesis, including a description of the objectives, study design, rationale and hypotheses for three related projects. The three projects address how the guideline has been implemented by Ontario hospitals, the association between the implementation of universal bilirubin screening and jaundice-related health service use, and the association between the implementation of universal bilirubin screening and neonatal access to follow-up care and associated SES disparities.

1.1.1 Neonatal Hyperbilirubinemia

High levels of bilirubin (hyperbilirubinemia), which create visible jaundice, occur in approximately 60% of all newborns.(1) Following birth, normal physiological processes lead to a transient rise in bilirubin levels in all newborns.(1) These levels tend to peak on the third or fourth day of life.(1) In most cases, bilirubin levels subsequently spontaneously decrease, and the condition is benign.(1) In a small portion of these cases, additional underlying pathological conditions contribute to excessive hemolysis or impaired bilirubin excretion, which further increase bilirubin levels.(1) Prematurity can also limit the neonate's ability to adequately excrete bilirubin.(1) In these cases, therapeutic measures are used to prevent bilirubin levels from becoming excessively high. First line treatment for neonatal jaundice is phototherapy. Some babies will require less than 24 hours of phototherapy while others may be treated for several days. One Canadian study found the average cost of predischARGE phototherapy in 2008 to be roughly \$2000, and the cost of readmission to be approximately \$3200.(2) In most cases this simple and relatively inexpensive form of treatment is effective and sufficient. In situations where phototherapy does not control rising bilirubin levels, exchange transfusion is recommended.(3)

Concern about neonatal jaundice stems from the fact that very high levels of bilirubin can cause kernicterus (staining and necrosis of the neurons in the basal ganglia and brainstem nuclei) and associated neurological abnormalities.(3) The potential short term consequences of severe hyperbilirubinemia are referred to as acute bilirubin encephalopathy, which is described as a clinical syndrome that includes neurological symptoms such as lethargy and hypotonia, and can progress to seizures and coma.(3) The long term sequelae of this syndrome are referred to as chronic bilirubin encephalopathy, which can include cerebral palsy, seizures, hearing loss, oculomotor dysfunction, and developmental delay.(3) The life time medical costs for this condition have been estimated to surpass \$1.3 million, not including non-medical costs to families and society.(4)

Acute bilirubin encephalopathy does not occur unless hyperbilirubinemia is severe (≥ 340 $\mu\text{mol/L}$), and is rare below critical levels (≥ 425 $\mu\text{mol/L}$).(3) At the same time, it is possible for

infants with critical hyperbilirubinemia to escape long term sequelae.(5) A synthesis of the findings from studies in Canada, Denmark and the United Kingdom suggests that the risk of kernicterus for infants with a total serum bilirubin (TSB) $> 513 \mu\text{mol/L}$ is roughly one in seven.(6) The incidence of critical hyperbilirubinemia (TSB $\geq 425 \mu\text{mol/L}$) in Canada is estimated to be 1 in 2,480 live births(7) and Canadian surveillance data based on case reporting estimates the incidence of chronic bilirubin encephalopathy to be 1 in 43,000 births.(8)

1.1.2 Root Causes of Severe Hyperbilirubinemia

Throughout the 1990s, an increasing proportion of mothers and newborns were discharged from hospital within 48 hours of birth, prior to the natural peak in bilirubin levels. At the same time, rates of breastfeeding increased. A Canadian study published in 2000 comparing births from 1996-97 to those from 1989-90 demonstrated that decreased length of stay at birth was associated with substantial increases in the risk of readmission in the first seven days of life for jaundice (relative risk (RR)=3.06, 95% confidence interval (CI) 2.91-3.21), dehydration (RR=13.50, 95% CI 10.62-17.16), and feeding problems (RR=7.33, 95% CI 6.38-8.43).(9) Jaundice was the leading cause for readmission in the first 28 days of life, with an estimated rate of 16.4 per 1,000 in 1996-97.(9) These findings, and similar observations in the United States,(10-13) drew attention to the fact that healthy, term breastfed infants who have difficulty establishing successful breastfeeding are at increased risk of severe hyperbilirubinemia even in the absence of underlying pathological causes for hyperbilirubinemia, particularly if they do not receive timely follow-up care after discharge from hospital.

Subsequent research confirmed that early discharge from hospital after birth with adequate community care is not only cost-effective but safe,(14-16) and that home-visiting programs could actually prevent the need for rehospitalization for jaundice.(17) Despite a lack of direct evidence that breastfeeding support reduces the risk of severe hyperbilirubinemia, there is evidence that breastfeeding support can improve frequency and duration of breastfeeding,(3) and acknowledgement that frequent breastfeeding reduces the risk of severe hyperbilirubinemia because it promotes more rapid excretion of bilirubin.(18)

Discussion of the safety of early discharge coincided with an expression of growing concern regarding the resurgence of severe hyperbilirubinemia and kernicterus.(6, 7) A Canadian review of 258 cases of reported critical hyperbilirubinemia between July 2002 and June 2004 found that 72% of cases were identified after initial discharge from hospital, and in 64% of all cases no cause was identified.(7) Efforts to counter the problem have focused on identifying system-based causes of adverse outcomes. In a review of cases identified in the US Pilot Kernicterus Registry, the following root causes were identified:

- (a) loss or lack of concern by clinicians regarding the neurotoxic potential of bilirubin,
- (b) limitations on visual recognition of jaundice as an index to initiate further evaluation or estimate severity, (c) failure to recognize the severity of hyperbilirubinemia at a specific age in hours; (d) failure to ensure appropriate follow-up 1 to 2 days after early discharge (24 to 72 hours of age); (e) delay in intensive or timely interventions before discharge or at readmission.(19)

In response to these problems, solutions have focused on increased detection of infants at risk/requiring treatment, and improved follow-up after discharge.

1.1.3 Universal Bilirubin Screening

A key preventive strategy that has been championed is universal bilirubin screening prior to hospital discharge. In 1999, Bhutani et al. established a percentile-based bilirubin nomogram for healthy babies born at or beyond 35 weeks gestation.(20) This nomogram can be used to classify the level of risk of developing clinically significant jaundice based on an infant's TSB and his/her age in hours,(20) and has subsequently been developed to provide treatment thresholds for both phototherapy and exchange transfusion.(3) In Canada, the CPS released guidelines in 2007 that recommends universal bilirubin screening of all infants within the first 72 hours of life and use of a nomogram to guide follow-up and treatment, with the cited goal of preventing chronic bilirubin encephalopathy.(3) While the most prominent feature of the CPS guidelines is the recommendation for universal bilirubin screening, the guidelines also provide twenty-two other specific recommendations addressing issues that include investigations for infants with additional risk factors, timelines for follow-up, the provision of breastfeeding support, and

treatment recommendations. (A complete list of the specific recommendations is provided in Appendix 14).

Given that chronic bilirubin encephalopathy is such a rare outcome, the limited evidence that is available about the efficacy of universal bilirubin screening comes from observational studies rather than randomized controlled trials. Four studies that evaluated the impact of universal bilirubin screening on severe hyperbilirubinemia were identified in the literature, three of which were conducted in the United States,(21-23) and one in Canada.(24) All four studies found statistically significant reductions in the proportion of infants who developed severe or critical hyperbilirubinemia. Details of the research designs and findings of these studies are provided in Appendix 1.

The appropriateness of universal screening is still a matter of debate. In 2009, the U.S. Preventive Services Task Force released a statement on screening infants for hyperbilirubinemia to prevent chronic bilirubin encephalopathy.(25) Based on a review of the literature,(26) the task force concluded that there was insufficient evidence to assess the balance of harms and benefits, and therefore made no recommendation.(25) U.K. guidelines published in 2010 recommend measurement of bilirubin only in newborns with visible jaundice.(27) More recently the U.S. Department of Health and Human Service's Secretary's Advisory Committee on Heritable Disorders in Newborns and Children recommended against adding neonatal hyperbilirubinemia to its uniform newborn screening panel due to concerns regarding the validity of screening and the effectiveness of screening in preventing long term sequelae.(28)

In addition to concerns about potential harms of screening and treatment, critics of universal bilirubin screening also identify the potential for substantial costs associated with universal bilirubin screening.(25, 28) Gaps in evidence regarding clinical efficacy and implications for service utilization limit any economic evaluation of universal bilirubin screening. While one economic analysis that examined the potential impact of universal pre-discharge screening concluded that implementation of such a strategy would be likely to "increase health care costs significantly with uncertain benefits",(29) a more recent Ontario-based study suggested that the cost to prevent one case of kernicterus through universal screening would be lower than the

lifetime medical costs of caring for a baby affected by kernicterus.(2) However, the Ontario study was based on assumptions derived from service utilization patterns at a single hospital, and there is reason to believe that even within a single health system, there may be differences between facilities in health service utilization patterns (e.g., phototherapy use, readmission rates) following implementation of a standardized approach to screening.(22) There has been uptake of the CPS guidelines in Ontario but there was no previous literature describing how, when and the extent to which the guidelines have been adopted across Ontario, nor the impact that implementation has had on the use of health services.

1.2 Theoretical framework

This section describes the theoretical concepts that shaped the research questions, and provides an overview of the limited literature on SES and neonatal hyperbilirubinemia.

1.2.1 Social Determinants of Health and Socioeconomic Status

In contrast to previous research which has primarily focussed on clinical outcomes associated with universal bilirubin screening, this thesis examines the association of universal bilirubin screening with the utilization of health services. In doing so, I have drawn on theoretical concepts from the field of population health. Population health involves the study of health outcomes of groups, and pays particular attention to the distribution of health outcomes within groups and the ways in which the social determinants of health can give rise to health inequities.(30) Current understandings of the social determinants of health can be traced back to the 1974 Lalonde Report which presented a framework identifying four key determinants of health: human biology, environment, lifestyle and health care organization.(31) Since that time a variety of conceptualizations of the determinants of health have been presented.(32-39) Understanding of the social determinants of health has evolved to encompass a wide range of sociological factors including but not limited to income and social status, social support networks, education, employment, personal health practices, gender, and culture.(38) Common to the various current theoretical models describing the social determinants of health is an

understanding that SES is an upstream or fundamental cause of health status.(40) SES is generally understood to refer to an individual or a family's relative position in society and incorporates multiple social determinants of health. While there are variations in precisely how SES is conceptualized, measurement of SES typically relies on measures such as education, occupation or employment status, and income (or income relative to expenditures).(41)

The approach taken in this dissertation is most closely aligned with the work of Starfield, whose work has focussed on the relationship between health care and population health,(32, 42) and the work of Aday and Andersen, whose work addresses the use of health services.(43, 44) Starfield's conceptual model of health determinants at the population level incorporates the idea that health system characteristics, along with environmental characteristics, material resources, social resources, behavioural/cultural characteristics, and psychosocial characteristics influence both health and equity in health (See Figure 1.1), and will also influence each other.(32) In this model, utilization of health services is included as one component of health system characteristics.(32) The model also identifies the relationship between health policy and health system characteristics.(32) In their framework for the study of access to medical care, Aday and Andersen identify the relationships between characteristics of the health delivery system, characteristics of the population (or individuals within the population) and the utilization of health services.(43) Their notion of population characteristics includes three elements: predisposing characteristics, enabling factors, and need.(43) Both predisposing characteristics and enabling factors incorporate various social determinants of health: predisposing characteristics include factors such as education, occupation, culture, and social networks, and enabling factors include elements such as income, health insurance, a regular source of care (which Starfield refers to as a "medical home"(45)), and travel.(44) Aday and Andersen's work might be seen to fit within a piece of Starfield's larger conceptual model, but common to both bodies of work are the notions that policy shapes the characteristics of the health care system, that SES is an important predictor of health care use, and that health care use has an impact on health status and health equity.(32, 43, 44)

My research investigates a particular kind of policy, a clinical practice guideline, and focuses on the association between implementation of the guidelines and the utilization of health services in

order to explore the possibility that there may be important SES differences in health care use that could potentially lead to inequities in health. The CPS hyperbilirubinemia guidelines recommend a population-based approach (universal screening and follow-up) to prevent long-term health consequences of severe hyperbilirubinemia. My first project focuses on the relationship between the policy and the health care system, and examines the extent to which implementation of the policy was truly universal. The second project examines the association between implementation of universal bilirubin screening and health care use related to the detection and treatment of hyperbilirubinemia. The third project examines one of the key recommendations of the guidelines (timely follow-up) and investigates the relationship between implementation of universal bilirubin screening, recommended follow-up, and SES. None of the three projects incorporate all the elements of the models presented by Starfield, Aday and Andersen but each project is consistent with the concepts underlying these models and is based on the assumptions that policy influences the health care system, that SES predicts health care use, and that health care use is a determinant of health status and health equity.

1.2.2 Socioeconomic Status and Neonatal Hyperbilirubinemia

Consideration of SES is limited in the existing literature on neonatal hyperbilirubinemia, and no studies have examined potential health inequities in the risk of adverse sequelae. Assuming that pre-discharge care is consistent with recommendations, the greatest potential source of disparity arises from differences in follow-up care. The relationship between the timing of follow-up care and the risk of severe hyperbilirubinemia is sometimes difficult to untangle because of the use of proxy outcomes such as phototherapy use or readmission for jaundice, which do not consistently reflect the severity of hyperbilirubinemia. The work of Madden et al. suggests that increased rates of follow up on day 3 or 4 will likely increase ascertainment of jaundice, while the level of concern of care providers with respect to hyperbilirubinemia may mediate the impact this has on treatment and subsequently on overall rates of severe hyperbilirubinemia.(46) American studies have shown that babies born to mothers of higher SES are more likely to be readmitted for jaundice because they are more likely to have timely follow-up care (ascertainment bias), suggesting that in some contexts they may be at increased risk of over-treatment.(47-49) At the same time, delays in the diagnosis and treatment of severe hyperbilirubinemia are established

risk factors for long term sequelae,(19) and concern has been expressed that low-income families are overrepresented among those that do not receive timely follow-up.(50)

We found no Canadian literature examining rates of newborn follow-up within timelines aimed at ensuring appropriate detection of hyperbilirubinemia requiring treatment by day 3 or 4; however there is evidence to support the hypothesis that disparities exist. The limited existing Canadian literature on newborn readmission suggests that infants of lower SES are at increased risk of readmission,(51-53) but readmission specifically for hyperbilirubinemia has not been examined. Inadequate access to primary preventive care has been identified as a cause of both infant hospitalization and of severe hyperbilirubinemia,(54-57) and Ontario newborns residing in neighbourhoods with lower incomes are less likely to have a primary care visit with a physician within a week of discharge from hospital post-birth, even when physician supply is included in the model.(58) Other Ontario research has shown that socioeconomically disadvantaged women are more likely to be discharged from hospital within 24 hours of giving birth,(59) which may increase the likelihood of their babies not receiving recommended follow-up.

Findings from two of the cohort studies investigating the efficacy of universal bilirubin screening suggest that sub-optimal follow-up for neonates may remain a problem despite implementation of universal bilirubin screening.(21, 23) Eggert et al. concluded that in twelve of thirteen cases of critical hyperbilirubinemia that occurred after the implementation of screening, “oversight or failure to ensure postdischarge follow-up of these patients, and the apparent lack of parental education regarding neonatal jaundice” may have contributed to the outcome.(21) Mah et al. found that for two of the eight post-screening cases with a TSB $\geq 513 \mu\text{mol/L}$ that they reviewed, scheduled follow-up appointments had not been kept.(23) These cases suggest that universal bilirubin screening may not be sufficient to address some of the root causes of chronic bilirubin encephalopathy, and provide further support to the hypothesis that babies of lower SES may be at greatest risk of delayed identification of severe hyperbilirubinemia.

1.2.3 Realist Review

The direction of inquiry in the first project, which examines how the CPS guideline has been implemented in Ontario, was influenced by the theoretical assumptions of realist review.(60) Realist review is a method of evaluating the impact of complex policy interventions. It acknowledges the influence of context on the impact of an intervention and aims to explain how interventions work in different circumstances.(60) The goal of realist review has been summarized as figuring out “what works for whom in what circumstances . . . and why.”(61) The implementation of the CPS hyperbilirubinemia guideline is a complex intervention. It involves establishing processes for screening, follow-up and treatment to ensure that recommended care is received both during initial hospitalization and following discharge from hospital, and requires the involvement and collaboration of multiple sectors of the health care system. The guideline itself does not prescribe exactly how services should be organized in order to meet the recommendations. In the absence of a provincially coordinated implementation of the guideline, we anticipated variation in how hospitals organized care to follow the guidelines and identified a need to gather information about the organization of care in order to understand the context for the outcomes to be examined in the second and third projects. A comprehensive realist evaluation typically involves a multi-methods, theory-driven, reflexive approach aimed at identifying the mechanisms that account for observed outcomes.(61, 62) This type of evaluation of the impact of universal bilirubin screening was beyond the scope of this dissertation, but the central premise of realist review, namely that context influences the impact of a complex intervention, shaped the content of our hospital survey. We included questions about how, where and by who services are delivered, along with questions about challenges, barriers and solutions associated with the implementation of universal bilirubin screening, in order to be able to describe the circumstances of guideline implementation in Ontario.

1.2.4 Health Services and Health Equity

Although it may not always be defined as a social determinant of health, the health care system and the services it provides are one of the determinants of the degree of health equity within a

population, with differences in access to care being an important source of health inequity related to health services.(63) While health care can reduce health inequities,(64) interventions aimed at improving health can at times result in greater inequity because the socially disadvantaged are more difficult to reach.(65) In particular, it has been noted that population-based approaches aimed at improving health (e.g., cervical screening) result in the “inequality paradox” – in which the most vulnerable populations are the least likely to benefit from the intervention.(66) Even interventions aimed at identifying and targeting vulnerable populations can sometimes “miss” a notable proportion of the most vulnerable.(67)

Consequently, the way in which health services are organized and provided can have both intended and unintended consequences with respect to health inequities. Additionally, choices regarding the allocation of health resources can have implications with respect to health equity as a result of variation in who will benefit from each of the potential options. The health equity consequences of choices regarding the organization and provision of health services are not always known in advance. These theoretical concepts support the importance of examining the relationship between implementation of the CPS hyperbilirubinemia guidelines and health service utilization. Our second project, which examines the association with length of stay, phototherapy use, and jaundice-related ED visits and readmissions, provides context specific information that is needed to understand the resource implications of guideline implementation in Ontario. Although it was beyond the scope of our project to do so, this information facilitates consideration of the opportunity costs of implementing universal bilirubin screening.

1.2.5 Access to Care

The third project was specifically influenced by the literature on access to care. Current Ontario health policy identifies both access and equity as key indicators of health care quality.(68) The conceptualization of “access” used in this project aligns with the Institute of Medicine (IOM) definition as “the timely use of personal health services to achieve the best possible outcome.”(69) Access to health services is often mistakenly equated with health insurance,(45) but it has long been acknowledged that “psychological, informational, social, organizational,

spatial, temporal” and other barriers exist in addition to financial barriers to access to health care.(70) For example, unstable housing, lack of adequate transportation and difficulty communicating with care providers have been identified as risk factors for inadequate postpartum follow-up among low-income women.(71) Similar factors could account for a socioeconomic gradient in rates of follow-up care for newborns with provincial health insurance, although more sophisticated conceptualizations of access also acknowledge that characteristics of health resources themselves can create obstacles to individuals seeking and obtaining care.(72) These concepts influenced our project in several ways. First, they provided support for our hypothesis that even within a universal health care system we might expect to observe differences in the timeliness of service use based on SES. Second, the notion that timeliness is an important element of access was incorporated into our definition of our primary outcome (recommended follow-up). Third, the IOM definition of access supports the use of actual health care utilization as a measure of access. We did not have the data necessary to examine the root causes of differences in newborn access to recommended follow-up, so our project was limited to describing the association between universal bilirubin screening and recommended follow-up and examining SES differences in this outcome.

1.3 Thesis Structure

This thesis aims to address three important gaps in knowledge related to the implementation of the CPS hyperbilirubinemia guideline through three distinct but related projects. Below is a brief overview of the objectives, study design, rationale and hypothesis of each project.

1.3.1 Project 1 – A survey of Ontario hospitals regarding implementation of the CPS hyperbilirubinemia guidelines

Objective 1 – To determine if and when hospitals implemented universal bilirubin screening

Objective 2 – To investigate organization of services to provide related follow-up and treatment

Objective 3 – To identify processes used to implement screening, and influential factors

Study Design – Internet-based survey of all Ontario hospitals providing maternal-newborn services

Rationale – Implementation of the CPS guidelines in Ontario was not provincially coordinated and occurred in an ad hoc manner. In order to subsequently measure the association between implementation and health care use, surveying hospitals directly was necessary to determine if and when individual hospitals had implemented the guidelines. Information gathered regarding the organization of services provided a context to our second and third projects which examined health care utilization outcomes. The findings of the hospital survey are presented in Chapter 2.

Hypothesis – The majority of hospitals have implemented universal bilirubin screening but there is variation across the province in terms of how services are organized.

1.3.2 Project 2 – The association between universal bilirubin screening and healthcare utilization: a cohort study

Objective – To evaluate the association between the implementation of universal bilirubin screening and five health service utilization outcomes: phototherapy use during birth hospitalization, phototherapy after hospital discharge, length of stay (LOS), jaundice-related emergency department (ED) visits, and jaundice-related readmissions.

Study Design – Population-based cohort study using administrative health data

Rationale – Phototherapy use, length of stay, jaundice-related ED visits, and jaundice-related readmissions are among the key drivers of costs associated with the prevention of severe hyperbilirubinemia. Previous studies have shown variation in the impact of universal bilirubin screening on these outcomes, and lack of information regarding resource implications is one of the gaps that limits assessment of the utility of population-based bilirubin screening. The only other Canadian evaluation of universal bilirubin screening occurred within the context of a program of universal public health home visits, which are not available in Ontario.(24) The patterns of use for these outcomes are also relevant to understanding the role that recommended follow-up care plays in preventing severe hyperbilirubinemia. This project is presented in Chapter 3.

Hypothesis –Implementation of universal bilirubin screening will be associated with increases in all of the outcomes (phototherapy use during birth hospitalization, phototherapy after hospital discharge, LOS, jaundice-related ED visits, and jaundice-related readmissions).

1.3.3 Project 3 – Guideline implementation, recommended follow-up care, and SES disparities: a cohort study

Objective 1 – To determine whether implementation of the CPS hyperbilirubinemia guidelines was associated with an increase in access to recommended follow-up care for newborns born at 35 or more weeks gestation and discharged to home from hospital within 72 hours of birth

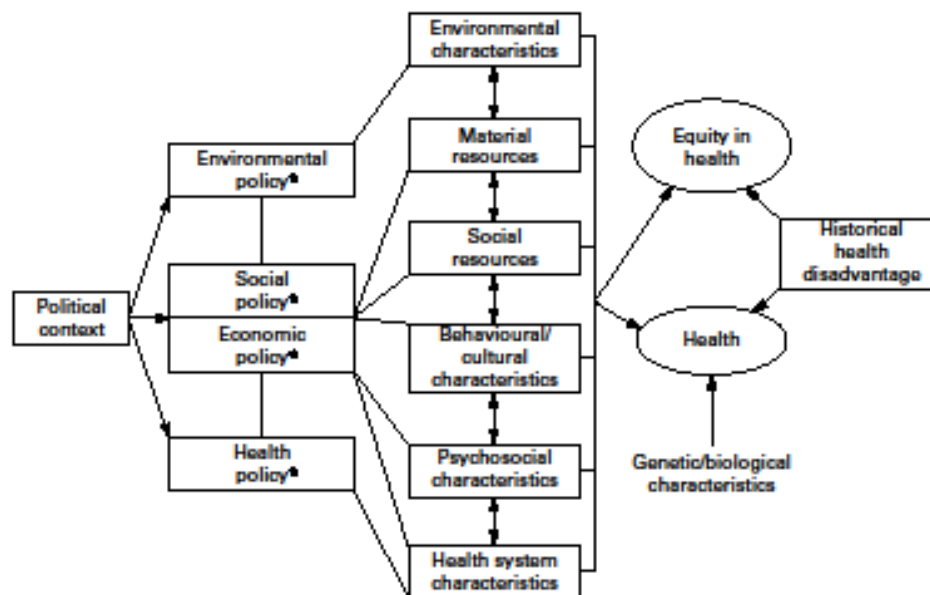
Objective 2 – If implementation of the CPS guidelines was associated with an increase in access to recommended follow-up care, to determine whether this differed between SES quintiles

Study Design – Population-based cohort study using administrative health data

Rationale – Delays in identification of the need for treatment have been identified as a root cause of kernicterus, and the CPS hyperbilirubinemia guideline addresses this concern by making specific recommendations for the timing of follow-up for newborns. The research literature supports the hypothesis that babies of lower SES are at greater risk of inadequate post-discharge follow-up, and therefore at greater risk of long-term sequelae associated with severe hyperbilirubinemia, but the impact of the CPS guideline on this potential disparity has not previously been investigated. Chapter 4 will therefore examine this issue.

Hypothesis – Following implementation of the CPS hyperbilirubinemia guideline babies will be more likely to have a follow-up visit within recommended timelines but disparities in access to this care will persist.

Figure 1.1. Starfield's conceptual framework of health determinants: population model



*Figure 1 A conceptual framework of health determinants: population model. *Policy reflects a country's approach to the distribution of power (political jurisdiction). Determinants of health at the individual level are the same but without "Equity in health outcomes."*

Reproduced from "Basic concepts in population health and health care," Journal of Epidemiology & Community Health, Barbara Starfield, 55: 452-4, 2001 with permission from BMJ Publishing Group Ltd.. (See Appendix 13)

Chapter 2

Implementation of the Canadian Paediatric Society's hyperbilirubinemia guidelines: A survey of Ontario hospitals

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Contributors Statement:

Elizabeth K. Darling: Ms. Darling conceptualized and designed the study, developed and implemented the hospital survey, carried out the data analyses, drafted the initial manuscript, and approved the final manuscript as submitted.

Astrid Guttman: Dr. Guttman contributed to the study design and the development of the hospital survey, supervised the data analyses and interpretation, reviewed and revised the manuscript, and approved the final manuscript as submitted

Ann E Sprague: Dr. Sprague contributed to the study design and the development of the hospital survey, reviewed and revised the manuscript, and approved the final manuscript as submitted.

Timothy Ramsay: Dr. Ramsay contributed to the study design, critically reviewed the manuscript, and approved the final manuscript as submitted.

Mark C. Walker: Dr. Walker contributed to the study design and the development of the hospital survey, critically reviewed the manuscript, and approved the final manuscript as submitted.

Additional Contributions: Kevin Coughlin, Sandra Dunn, Melissa Dougherty and Jennifer Medves acted as an expert reference group during the process of developing the hospital survey.

2.1 Abstract

Introduction: In 2007, the Canadian Paediatric Society (CPS) published guidelines aimed at preventing severe hyperbilirubinemia. Our objectives were to determine if hospitals had implemented these guidelines, to investigate how guideline-recommended care is organized, and to understand the factors influencing guideline implementation.

Methods: On-line survey from December 2011-May 2012 of all Ontario hospitals offering maternal-newborn services.

Results: Ninety-seven of 100 eligible hospitals responded. Seventy-seven of the 97 respondents (79%) reported having implemented universal neonatal bilirubin screening. Among these hospitals, hospital-based post-discharge follow-up was reported more frequently than community-based locations: hospital lab (n=40, 52%), mother-baby care unit (n=32, 42%), outpatient clinic (n=25, 33%), primary care-provider in community (n=19, 25%), community lab (n=8, 10%). The CPS guidelines were the most frequently reported factor influencing implementation (n=74, 96%).

Discussion: The survey provides valuable insight into the impact of a complex guideline in Canada's largest province. There is heterogeneity in how hospitals have organized services, but a notable trend towards hospital-based post-discharge care. The shift to hospital-based care runs counter to current health policy directions and highlights the lack of integration between health care sectors.

Conclusion: The majority of Ontario hospitals implemented universal bilirubin screening following release of the CPS guidelines. Further analysis is needed to determine the impact that the guidelines and the differences in implementation have had on clinical outcomes and the utilization of health services.

MeSH key words: hyperbilirubinemia; jaundice; practice guideline; guideline adherence

2.2 Introduction

Severe hyperbilirubinemia is the leading cause of neonatal readmissions in Canada.(9) Although chronic bilirubin encephalopathy is rare (affecting approximately 1 in 43,000 births), the consequences are devastating.(8) Under-recognition of severe hyperbilirubinemia places otherwise healthy infants at risk of preventable harm.(6, 7) Efforts to counter this problem target system-based causes such as the limitations of visual assessment of jaundice, failure to recognize the severity of hyperbilirubinemia based on age in hours, lack of appropriate follow-up after early discharge, and delays in treatment.(19) In 2007, the Canadian Paediatric Society (CPS) published guidelines on the detection, management and prevention of hyperbilirubinemia aimed at addressing this problem.(3) The impact of these guidelines has not been evaluated.

The CPS hyperbilirubinemia guidelines are based on universal pre-discharge bilirubin screening and use of a nomogram to guide follow-up and treatment. Evidence from three American cohort studies suggests that these measures reduce the incidence of severe hyperbilirubinemia.(21-23) The guidelines also recommend timely follow-up for all babies after hospital discharge. This element of the guidelines adds complexity to their implementation given the multiple sectors of the health care system (e.g., hospitals, community-based care providers, and public health) involved. Although clinical practice guidelines can effectively promote evidence-based practice, they are not consistently implemented in an effective and timely manner.(73-75) Systematic reviews on successful guideline implementation suggests that more complex guidelines may be less likely to be adopted and followed.(76)

As part of a larger project investigating the impact of the 2007 CPS hyperbilirubinemia guidelines in Ontario, we undertook a hospital survey to investigate hospital response to the guidelines. The survey objectives were: 1) to determine if and when hospitals had implemented universal bilirubin screening, 2) to investigate how hospitals organize services to provide related follow-up and treatment, and 3) to understand the processes used to implement screening and factors influencing these processes.

2.3 Methods

Data were collected using an on-line questionnaire administered using Survey Monkey. The target population was all Ontario hospitals offering maternal-newborn services as of March 2011 (excluding children's hospitals providing newborn services only).(77, 78) The initial questionnaire was developed by the primary investigator (LD) using research-based principles of survey design,(79-81) with input from the research team which included expertise in nursing, paediatrics, midwifery and obstetrics. Questionnaire content was based on a review of the literature on the implementation of clinical guidelines, the research objectives, and the reported experiences of a multi-disciplinary work group that developed regional hyperbilirubinemia guidelines for the Champlain Local Health Integration Network (one of 14 health regions in the province). Questions regarding the implementation process were based primarily on theoretical constructs that focus on environmental context and resources (i.e., organizational or system level factors).(82-85) Closed-ended questions were used to gather information about if and when hospitals had implemented screening and about the organization of follow-up and treatment. Multiple choice questions with the option of an open-ended response were used for questions about facilitators and challenges, and open-ended questions were used to gather information about new processes, strategies to address challenges, and perceived successes.

Face validity was assessed by a four member expert reference group with expertise in neonatology, nursing, hospital administration, and research. Based on their input the wording of four questions was revised, additional responses were added to seven multiple choice questions, and one question was deleted. The revised survey was then pilot tested at five hospitals representing small and large volume centres. Respondents to the pilot survey provided feedback about question wording and acceptability (See Appendix 3 for a list of the feedback gathered via the pilot survey). Based on their feedback no further revisions were deemed necessary. Responses from hospitals participating in the pilot were retained for inclusion in the final analysis. Each version of the questionnaire was reviewed and approved by The Ottawa Hospital Research Ethics Board prior to its use. (See Appendix 2 for copies of research ethics board approvals. A copy of the final survey is provided in Appendix 4.)

Invitations to participate in the study were administered with assistance from the Better Outcomes Registry & Network (BORN-Ontario) which maintains contact with all Ontario hospitals providing maternal-child services. BORN distributed an email to hospital contacts (primarily maternal-newborn program administrators) with a letter of information containing the link to the on-line survey and a copy of the survey questions. Participation was voluntary and participants retained the right to withdraw from the study at any time. No incentives were provided. Respondents from hospitals with multiple sites had the option of responding once for all sites, or separately for each site. The detailed consent information from the letter of information was provided to participants again on the first screen of the on-line survey. Non-responders were sent up to three reminder notices via email. Following the final reminder notice, administrators who still had not responded were contacted by telephone and offered the opportunity to conduct the questionnaire verbally over the telephone.

Survey results were described by average birth volume of eligible hospitals, calculated using data from the Discharge Abstract Database accessed at the Institute for Clinical Evaluative Sciences.

2.4 Results

The survey was piloted between December 2011 and January 2012. Remaining responses were collected between January and May 2012. Ninety-seven of one hundred hospital sites (97%) responded (92 separate responses, with 5 respondents providing a single response for two sites), see Figure 2.1. Table 2.1 presents the characteristics of the responding hospitals. The three non-responding hospitals were all from the North local health integration network (LHIN) region, provided level 1 newborn services, and had average birth volumes of <500/year. (15)

2.4.1 Universal screening

Seventy-seven of the 97 responding hospitals (79%) reported having implemented universal neonatal bilirubin screening. Nine of these hospitals (9%) reported having implemented universal screening prior to the release of the CPS guidelines. It took until May 2010 (three years following the release of the guidelines) for the next 45 hospitals to implement universal

bilirubin screening. Table 2.2 shows how many hospitals implemented screening by year. Sixty-three hospitals (82% of those conducting universal screening) reported that their approach was based on the CPS guidelines, and 12 hospitals (16%) reported that their approach was based on the guidelines with modifications.

With respect to screening, 54 of the 77 hospitals (70%) which had implemented universal screening reported using total serum bilirubin (TSB) measurements only for screening, 17 (22%) used both TSB and transcutaneous bilirubin (TcB) measurements, and 5 (6%) reported using only TcB for screening. Almost all hospitals (n=74, 96%) indicated that if a baby requires phototherapy prior to initial discharge from hospital, phototherapy treatment would normally be provided at that institution versus at another institution.

2.4.2 Follow-up testing and readmission

Follow-up for babies requiring repeat bilirubin testing after hospital discharge is organized in a variety of ways. Table 2.3 summarizes where parents are directed to take their baby if retesting is required and where babies are admitted for inpatient phototherapy following hospital discharge. Free-text comments elaborating on other follow-up included: going to the Emergency Department, referral to an outpatient newborn assessment clinic at a neighbouring hospital, and direction to see the delivering family physician if the baby does not have a physician. Another response indicated that babies under midwifery care would be provided follow-up in the community by their midwife.

2.4.3 Ensuring follow-up

Community-based care providers for newborns in Ontario include paediatricians, family physicians, midwives, and nurse practitioners. Fifty hospitals (65%) reported that bilirubin screening results are routinely communicated to the baby's primary care provider in the community, eighteen (23%) only if the results were elevated, and nine (12%) did not communicate results to community-based providers. Communication methods include: written document given to parent (n=38, 49%), direct verbal communication (n=37, 48%), fax (n=21, 27%), and email (n=1, 1%). "Other" methods described in open-ended text included discharge summary sent by mail (n=4, 5%), lab report sent by mail, and electronic access via the hospital

chart or lab result reporting system (n=2, 3%). Several respondents noted that elevated results would always be communicated directly to the provider most responsible for the baby while in hospital.

Thirty-seven hospitals (48%) reported implementing a process to verify that babies return at the appropriate time for follow-up testing. Twenty-eight hospitals (36%) reported booking follow-up outpatient appointments or keeping a list of babies who require follow-up and contacting no shows. Other approaches to verification of follow-up reported include physicians ensuring follow-up for their patients (n=4, 5%), hospitals contacting the community-based care provider if parents cannot be contacted (n=2, 3%), and verification of follow-up at health unit postpartum home visits (n=1, 1%).

Thirty-four hospitals (44%) reported using specific strategies to help ensure appropriate follow-up regarding hyperbilirubinemia for babies at high risk of experiencing barriers in access to care. Open-ended responses included referrals to public health (n=15, 19%), community-based physicians or midwives (n=7, 9%), and hospital social workers (n=4, 5%), and a number of other strategies.

2.4.4 Challenges, solutions, successes

Table 2.4 summarizes challenges encountered in implementing universal bilirubin screening. Table 2.5 summarizes the more common new processes or services that respondents described in open-ended text. Forty-two hospitals (55%) reported that they developed new processes or services in order to implement the CPS guidelines – in many cases these changes were introduced to address challenges identified in Table 2.4. Forty-one respondents (53%) described in open-ended text the successes they felt had been achieved in implementing universal bilirubin screening. Perceived successes included: improved early identification of infants at risk or in need of treatment (n=11, 14%), better follow-up (n=8, 10%), decreased length of stay (n=5, 6%), high satisfaction with approach (n=3, 4%), streamlining of care (n=3, 4%), consistency in care (n=2, 3%), reassurance for care providers that cases less likely to be missed (n=2, 3%), and reduction in painful procedures (blood draws) for newborns (n=2, 3%).

2.4.5 Factors influencing implementation

Factors that influenced the decision to implement universal bilirubin screening are summarized in Table 2.6. Seventeen hospitals (22%) reported that there had been regional coordination of guideline implementation, and 38 hospitals (49%) reported regional engagement about a broader set of issues around the optimal provision of newborn health services following hospital discharge. Hospitals also reported engagement with local community-based primary care providers (n=32, 42%), regional perinatal partnerships (n=21, 27%), and local public health (n=19, 25%).

Hospitals that reported not having implemented universal bilirubin screening (n=20) were asked to identify barriers to implementation. These responses are summarized in Table 2.7. “Other” barriers described in open-ended text were: universal screening perceived as unnecessary because other processes in place (e.g., risk-based screening) (15%), physician preference (5%) and lack of knowledge (5%).

2.5 DISCUSSION

The findings provide valuable insight into the impact of a complex pediatric guideline in the largest Canadian province. Uptake of the CPS hyperbilirubinemia guidelines was gradual over the first five years since their release, but as of May 2012, approximately 88% of births in Ontario occurred at a hospital that had implemented universal bilirubin screening. There is substantial heterogeneity in how hospitals have organized services in order to implement the guidelines. This likely reflects differences in availability of services (including laboratory services), the mix of providers, the degree of involvement of community-based physicians in hospital care, work flow and administrative processes, birth volumes, patient demographics and the size of the geographic area served. One notable trend is that the majority of hospitals in the province have extended their responsibility for babies at higher risk of severe hyperbilirubinemia to include care during the first few days following discharge from hospital. The shift to hospital-based neonatal follow-up has been influenced by limitations in the availability of timely bilirubin

testing in the community as well as limitations in access to community-based physicians within the recommended time-frames.

Hospitals experienced a variety of challenges in implementing the guidelines, and the solutions they developed to address these challenges often involved creating new processes or re-organizing existing services. Several of the more frequently identified challenges that hospitals encountered have notable implications for health care resources. While a small number of respondents indicated that they had been able to reduce lengths of stay with universal bilirubin screening, a greater number of respondents perceived that screening had led to delays in hospital discharge. These delays were attributed to both the time required to screen babies and interpret the results, and to decisions to keep babies in hospital for longer when they require follow-up testing but not treatment. Some respondents also noted that challenges in interpreting the guidelines sometimes contributed to over-testing and over-treatment. Also, hospitals have not been allocated extra resources to fund the provision of post-discharge follow-up services. Another common theme in reported challenges was difficulty arising from a lack of integration between different sectors of the health care system (e.g., hospitals and community-based physicians).

The results of the survey suggest that there might be efficiencies to be gained from provincial coordination of guideline implementation. While some diversity in service delivery models is to be expected given the contextual variations across the province, a provincially coordinated approach to guideline implementation might better support hospitals to benefit from the experiences of others by sharing solutions to challenges, and might also facilitate timelier implementation in low volume hospitals with limited human resources for guideline implementation. Provincially-led implementation might also ensure that changes arising from the implementation of a guideline develop in directions that are consistent with current health policy. For example, increased use of hospital-based neonatal follow-up care runs counter to current Ontario health policy directions which aim to deliver services in the community when that option is safe, effective, and less expensive. (23) Current Ontario initiatives such as the Health Links program, (24) which facilitates good coordination of care between different health care sectors and providers, may provide a model for the development of a coordinated system of

community-based post-discharge maternal-newborn care in the province which could reduce the need for hospital-based follow-up for neonatal hyperbilirubinemia.

A major strength of our study is that we achieved a 97% response rate to the survey. One limitation of the survey is that responses may have been subject to errors of recall, or limited by the extent of experience of the respondent with respect to the questions being asked. We attempted to minimize this problem by providing a soft copy of the survey in advance to allow time to gather responses and encouraging respondents to gather input from others as needed. Our findings are not universally generalizable outside of Ontario, but the Ontario experience may be of relevance in settings where universal bilirubin screening has been adopted in an ad hoc manner by hospitals or in settings where there is not a coordinated system of universal community-based post-discharge maternal-newborn care.

Additional research is needed to determine the impact that the guidelines have had on clinical outcomes and the utilization of health services. Recent surveillance data collected by the Canadian Paediatric Surveillance Program should provide basic information related to the impact of the guidelines on the incidence of severe neonatal hyperbilirubinemia across Canada. Data collected in our study on the month and year of screening implementation at each hospital will allow us to examine the impact of the guidelines while taking into account the lag between guideline release and implementation. Further analyses are planned which will determine the impact on health service utilization and recommended newborn follow-up.

Figure 2.1. Study flow through invitation to participate, assessment of eligibility, and survey response

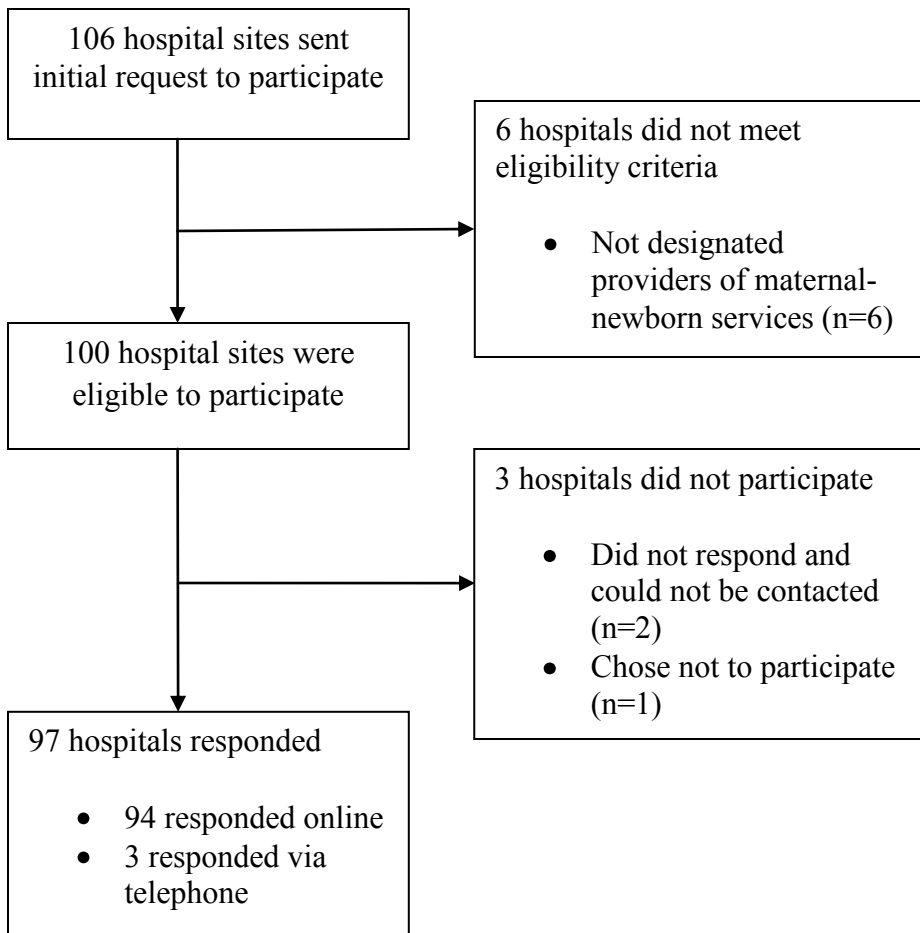


Table 2.1. Characteristics of responding hospitals

		Hospitals that implemented screening		Hospitals that did not implement screening	
		n	%	n	%
Neonatal Level of Care *					
	Level 1	31	67	15	33
	Level 2	39	89	5	11
	Level 3	7	100	0	0
Local Health Integration Network Region					
	South West (LHINS 1 & 2)	15	100	0	0
	Central West (LHINS 3 & 4)	13	87	2	13
	Greater Toronto Area (LHINS 5-9)	24	83	5	17
	South East (LHINS 10-11)	10	71	4	29
	North (LHINS 12-14)	15	63	9	37
Annual Birth Volume †					
	< 240 newborns per year	16	64	9	36
	240-500 newborns per year	9	64	5	36
	501-1000 newborns per year	14	93	1	7
	> 1000 newborns per year	38	88	5	12

* Standardized criteria are used to define the level of neonatal services provided by Ontario hospitals, with Level 1 being the most basic level of service and Level 3 being the most specialized level of service. (14)

† Annual birth volumes were calculated using data from the Discharge Abstract Database held at the Institute for Clinical Evaluative Sciences.

Table 2.2. Year of implementation of universal bilirubin screening

Hospitals that implemented universal bilirubin screening			
Year	n	% *	Cumulative %
1993	1	1	1
2003	2	2	3
2005	3	3	6
2006	1	1	7
2007	12	12	20
2008	13	13	33
2009	18	19	53
2010	12	12	65
2011	9	9	74
2012	2	2	76
Missing	4	4	80

* Of 97 hospitals who responded to the survey

Table 2.3. Location of follow-up for hyperbilirubinemia

Location	Hospitals	
	n	%
<i>Location for Follow-up Testing</i>		
Hospital Lab	40	52
Mother-baby Care Unit	32	42
Outpatient Clinic	25	33
Primary Care-Provider in Community	19	25
Community Lab	8	10
Neonatal Nursery	6	8
Community-based Family Medicine Clinic	3	4
Community-based Paediatric Clinic	1	1
Community Health Centre	1	1
Another Hospital in the Region	1	1
<i>Location for Readmission for Treatment</i>		
Paediatric Unit	48	62
Normal Newborn/Postpartum Unit	23	30
Neonatal Nursery	17	22
Nearest Children's Hospital	7	9
Another Hospital in the Region	5	6

Table 2.4. Challenges experienced with implementation of universal bilirubin screening

Challenges of implementation	Hospitals	
	n	%
Arranging post-discharge follow-up on weekends and holidays	34	44
Delays in newborn discharge from hospital	33	43
Arranging post-discharge follow-up for babies who live far from hospital	30	39
Ensuring appropriate communication of results	22	29
Arranging access to TSB or TcB testing in the community	20	26
Cost	13	17
Over-testing	11	14
Resistance from care providers to screen all babies	7	9

Table 2.5. New processes and services accompanying implementation

New process/services	Hospitals	
	n	%
Creation of new paediatric or neonatal outpatient clinic	11	14
Changes in staff roles	11	14
Arrangements for outpatient laboratory access	11	14
Implementation of TcB testing	8	10
Arrangements for outpatient follow-up via existing locations (paediatric department, mother-baby unit, breastfeeding clinics)	6	8
New readmission procedures or location	4	5

Table 2.6. Factors contributing to implementation of universal bilirubin screening

Factor contributing to implementation	Hospitals	
	n	%
Release of Canadian Paediatric Society guidelines	74	96
Leadership from physicians providing paediatric care	55	71
Leadership from Maternal-Newborn program leaders	55	71
Standard practices of other hospitals in region	41	53
Risk-management considerations	40	52
Leadership from nurse educator/clinical specialist	37	48
Activities of regional perinatal program	29	38
Leadership from an interdisciplinary committee	28	36
Medico-legal considerations	25	32
Case(s) of severe hyperbilirubinemia in hospital/community	17	22

Table 2.7. Barriers to implementation in hospitals without universal screening (n=20)

Barrier to implementation	Hospitals	
	n	%
Lack of human resources to coordinate implementation of clinical protocols	7	35
Resistance from care-providers to screen all babies	6	30
Cost	5	25
Difficulties arranging post-discharge follow-up on weekends and holidays	3	15
Difficulties arranging access to TSB or TcB testing in the community	3	15
Patient population living at a significant distance from hospital	3	15

Chapter 3

Universal bilirubin screening and healthcare utilization

This chapter has been removed because of copyright restrictions.

The chapter reported the findings of our second project. The objective of this study was to evaluate the impact of the implementation of universal bilirubin screening on neonatal healthcare use in a Canadian province with universal health insurance. The study design was a population-based retrospective cohort study. The study population was all newborns discharged following birth between April, 2003 and February, 2011 from 42 hospitals that implemented universal bilirubin screening between July, 2007 and June, 2010 in Ontario, Canada. We determine their screening implementation date using findings of the survey reported in Chapter 2. We used multiple linked administrative health datasets to measure phototherapy use, length of stay, jaundice-related emergency department (ED) visits and jaundice-related readmissions. We modelled the relationship between universal bilirubin screening and outcomes using generalized estimating equations (GEE) to account for clustering by hospital, underlying temporal trends, and important covariates. Universal bilirubin screening was associated with an increase in phototherapy during hospitalization at birth (relative risk (RR) 1.32, 95% confidence interval (CI) 1.09-1.59), and a decrease in jaundice-related ED visits (RR 0.79, 95% CI 0.64-0.96). We found no statistically significant difference in phototherapy after discharge, length of stay, or jaundice-related readmissions. Our findings facilitate determining the resource implications of universal bilirubin screening within Ontario, but highlight the limitations in the generalizability of previous research estimating the impact on healthcare utilization and underline the importance of context-specific local evaluation of guideline implementation.

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Contributors Statement:

Elizabeth K. Darling: Ms. Darling conceptualized and designed the study, carried out the data analyses, drafted the initial manuscript, and approved the final manuscript as submitted.

Timothy Ramsay: Dr. Ramsay contributed to the study design, provided advice regarding statistical analyses and interpretation, critically reviewed the manuscript, and approved the final manuscript as submitted.

Ann E. Sprague, and Mark C. Walker: Drs. Sprague and Walker contributed to the study design and interpretation of data, critically reviewed the manuscript, and approved the final manuscript as submitted.

Astrid Guttman: Dr. Guttman contributed to the study design, supervised the data analyses and interpretation, reviewed and revised the manuscript, and approved the final manuscript as submitted.

Supporting Materials:

A detailed rationale for the modelling strategy that was used in this study is provided in Appendix 5.

Appendix 6 presents a table summarizing the clinical and socio-economic characteristics of included vs. excluded babies.

Chapter 4

Association of hyperbilirubinemia guidelines with newborn follow-up care and socioeconomic disparities in follow-up

Contributors Statement:

Elizabeth K. Darling: Ms. Darling conceptualized and designed the study, carried out the data analyses, drafted the initial manuscript and incorporated the contributions of co-authors, and approved the final manuscript as submitted.

Timothy Ramsay: Dr. Ramsay contributed to the study design, provided advice regarding statistical analyses and interpretation, critically reviewed the manuscript, and approved the final manuscript as submitted.

Doug Manuel, Ann E. Sprague, and Mark C. Walker: Drs. Manuel, Sprague, and Walker contributed to the study design and interpretation of data, critically reviewed the manuscript, and approved the final manuscript as submitted.

Astrid Guttman: Dr. Guttman contributed to the study design, supervised the data analyses and interpretation, reviewed and revised the manuscript, and approved the final manuscript as submitted.

4.1 Abstract

Objectives: To determine whether implementation of universal bilirubin screening was associated with 1) an increase in use of recommended follow-up care, and 2) a differential effect between material deprivation quintiles.

Design: Retrospective population-based cohort study.

Setting: 97 of 100 hospitals providing maternity care in Ontario, Canada.

Population: 733,990 newborns born at 35 or more weeks gestation, with a valid health card number and a hospital record linked to a single maternal record, and discharged to home from hospital within 72 hours of birth between April 1, 2003 and February 28, 2011. (68,328 additional babies were excluded for the following reasons: transfer to another facility, died on the day of discharge, higher order multiples, randomly selected twin from twin sibling pairs, or mother received midwifery care.)

Intervention: Implementation of universal bilirubin screening.

Main outcome measures: Recommended follow-up care, defined as a physician visit within one calendar day after discharge for babies discharged ≤ 24 hours after birth, or a physician visit one or two calendar days after discharge for babies discharged between 24-72 hours after birth.

Results: Implementation of the guidelines was associated with a modest increase in receiving recommended follow-up (adjusted RR: 1.11, CI: 1.0014 to 1.22, $p=0.047$), but the majority of babies still do not receive follow-up within the timeframe recommended by the guidelines.

There was a striking gradient in the crude percentage increase in recommended follow-up associated with guideline implementation (ranging from 0.3% in the lowest quintile to 29.0% in the highest quintile), and we observed a significant interaction between guideline implementation and material deprivation status. Disparity in recommended follow-up increased following guideline implementation, with 40% of the crude increase attributable to the highest SES quintile and none of it to the lowest SES quintile.

Conclusions: Implementation of universal bilirubin screening has had limited impact in ensuring timely follow-up for all newborns in Ontario. Lack of timely follow-up represents an ongoing weakness in efforts to prevent severe hyperbilirubinemia. The observed widening of the SES disparity in access to recommended follow-up associated with implementation of universal

bilirubin screening illustrates that universal programs which fail to address root causes of disparities may lead to overall improvements in population outcomes but increased inequity.

4.2 Introduction

Concern regarding the rare but serious long term sequelae associated with severe neonatal hyperbilirubinemia has led several organizations to develop clinical practice guidelines providing screening and treatment recommendations.(3, 27, 86) Newborn bilirubin levels typically peak between day three and five of life, after most babies born in hospital have been discharged. Lack of timely follow-up can result in delays in the diagnosis and treatment of severe hyperbilirubinemia, which are established risk factors for long term sequelae.(19) It has been recommended that newborns receive follow-up within 24-48 hours of hospital discharge, particularly if they are discharged within 72 hours of birth,(7, 87) and in fact follow-up recommendations are a key element in current North American hyperbilirubinemia guidelines.(3, 86) In Canada, the Canadian Paediatric Society's (CPS) 2007 hyperbilirubinemia guidelines tightened a previous recommendation that newborns be assessed by a health professional within 48 hours of early discharge(88) to specify that infants discharged in the first 24 hours of life should be assessed within 24 hours.(3) While some Canadian jurisdictions have well-established systems of timely community-based follow-up,(24) ensuring timely follow-up remains a challenge in settings where there is less coordination between health care sectors (i.e., hospitals, public health, and community-based primary care providers).(89) In a survey of Ontario hospitals, we found that efforts to improve post-discharge follow-up focussed on ensuring access to repeat bilirubin testing.(89)

Cohort studies examining the effectiveness of universal bilirubin screening identified that failure to keep scheduled follow-up appointments was a contributing factor in cases of severe hyperbilirubinemia following discharge.(21, 23) Research on neonatal hyperbilirubinemia has not examined the role of socio-economic status (SES) with respect to missed follow-up, but there is evidence that even within a system of universal health insurance there is socio-economic disparity in health care use by newborns.(58) To explore potential disparities in the degree of benefit derived from guideline implementation, we examined follow-up outcomes associated with the implementation of universal bilirubin screening in response to the CPS's 2007 hyperbilirubinemia guidelines in Ontario (Canada's largest province: population 13 million). The objective of our research was twofold: to determine whether implementation of universal bilirubin screening was associated with 1) an increase in use of recommended follow-up care for newborns born at 35 or more weeks gestation and discharged to home from hospital within 72

hours of birth, and 2) a differential effect between material deprivation quintiles. We expected to find disparities in access to recommended follow-up care based on SES, and hypothesized that following implementation of screening, babies would be more likely to have a follow-up visit within recommended timelines but that disparities in follow-up would persist.

4.3 Methods

4.3.1 Study Design

This was a population-based retrospective cohort study of all newborns discharged to home from an Ontario hospital within 72 hours following birth between April 1, 2003 and February 28, 2011. We conducted a survey of all 100 Ontario hospitals providing maternal-newborn services as of March 2011 to determine if and when hospitals had implemented universal bilirubin screening, and included births from the 97 hospitals that responded to the survey. Details of the survey methods and findings have been reported previously.⁽⁸⁹⁾ Survey data were linked with administrative health datasets held at the Institute for Clinical Evaluative Sciences to evaluate the association between the implementation of screening and post-discharge follow-up visits. Scrambled health insurance numbers were used to link datasets. The study cohort was created using linked maternal and newborn records from the Canadian Institute for Health Information's (CIHI) hospital Discharge Abstract Database (DAD). Additional linked data sources were the Ontario Health Insurance Plan (OHIP) and CIHI's National Ambulatory Care Reporting System (NACRS). Research ethics approval was obtained from the Ottawa Hospital Research Ethics Board.

4.3.2 Inclusion/Exclusion Criteria

Inclusion criteria included gestational age ≥ 35 weeks (the target population of the CPS guidelines), a valid health insurance number (needed to link to follow-up data), linkage to a single maternal health record (needed to create covariates), and discharge to home within 72 hours of hospital birth. We chose to examine babies leaving hospital within 72 hours of birth because lack of follow-up within one to two days for these babies has been identified as a root cause of kernicterus.⁽¹⁹⁾ 802,318 babies met the inclusion criteria. We excluded babies who

were transferred to another facility, babies who died on the day of discharge, and higher order multiples. We randomly excluded one twin from twin sibling pairs because we anticipated that their outcomes would be correlated. We also excluded babies born to mothers who received midwifery care because the standard of care is follow-up within one day of discharge, and our objective was to examine the impact of the guidelines on babies cared for by other health care providers. Many recipients of midwifery care do not receive any prenatal or follow-up visits from a physician, and they are more likely than the rest of the cohort to be discharged on day 1. Visits by midwives are not included in OHIP, which captures physicians' billings. We did not have linked access to the provincial database that captured data on all mothers and babies who received midwifery care during the study period. Therefore, in order to identify mothers who were recipients of midwifery care, we examined maternal OHIP records during a nine-month look-back window preceding the birth to identify codes for midwife-requested consultations, and examined service provider codes on the mother's delivery record in DAD. After these exclusions, 733,990 babies remained in the analytical cohort. Figure 1 provides details regarding cohort creation and exclusions.

4.3.3 Variable Definition

The exposure of interest was discharge from a hospital that had implemented universal bilirubin screening. Our hospital survey collected the year and month that screening was implemented. When month was missing we imputed the month to be July (halfway through the year). Babies were classified according to the date they were discharged from hospital and the date screening was implemented at the hospital where they were born. The CPS guidelines were published in May-June 2007. In cases where hospitals reported implementing universal bilirubin prior to July 2007, we only classified babies as exposed if they were born in July 2007 onwards because the recommendations regarding short time frames for post-discharge follow-up are an element of the guideline that may not have accompanied pre-guideline implementation of universal bilirubin screening.

Our primary outcome was recommended follow-up care. The CPS guidelines include an explicit recommendation that babies discharged within 24 hours of birth should receive follow-up within 24 hours, but other recommendations in the guidelines regarding follow-up are based on screening results rather than age at discharge. We considered previous CPS guidelines

recommending follow-up within 48 hours(88) and discussion within the hyperbilirubinemia guidelines themselves regarding the importance of follow-up as a safety net to catch occasional case where bilirubin levels rise suddenly after the first two or three days, and chose to operationalized our primary outcome as follows: for babies discharged ≤ 24 hours after birth, visits occurring one calendar day after discharge; for babies discharged between 24-72 hours after birth, visits that occurred one or two calendars days after discharge. Follow-up visits were identified using records from OHIP, and NACRS, which captures visits to emergency departments and other ambulatory clinics. In some small rural hospitals, scheduled primary care visits occasionally occur in emergency departments and are captured in NACRS. We used fee codes (OHIP) and diagnostic codes (NACRS) for primary care visits to construct the follow-up outcome. (Details of the fee codes and diagnostic codes used are provided in Appendix 7.) Our sensitivity analysis outcome was follow-up within seven days of discharge. We used the same fee codes and diagnostic codes as were used for the primary outcome and defined the outcome as occurring if a visit occurred one to seven days following the date of discharge.

We controlled for important clinical, health service, and socio-demographic variables. We established the following list of covariates a priori: gestational age category (35-38 weeks versus 39+ weeks), birth mode (spontaneous vaginal, assisted vaginal, caesarean), maternal parity (multiparous vs. primiparous), maternal residence (rural vs. urban), timing of recommended follow-up (i.e., if day fell on a weekend or statutory holiday), maternal age at first delivery (<19 vs. 19+), and maternal prenatal care provider (general practitioner (GP) vs. obstetrician or other providers vs. no prenatal care). Maternal prenatal care provider was included as a marker of access to a primary care physician who would (in most cases) also provide newborn care. We classified women as having prenatal care from a GP if they had at least one prenatal visit billed by a GP or family physician. (Details regarding the data sources used to create the covariates are provided in Appendix 5.)

We used material deprivation quintile to account for SES. This variable was derived from maternal postal code from the mother's hospital record using Pampalon's Deprivation Index for Canada.(90) The index is based on census data at the level of dissemination area (the smallest geographic census unit in Canada: population 400-700). Material deprivation in this index is most influenced by neighbourhood income, education and employment. We selected

this measure to account for SES because both income and education are associated with access to care and we did not have individual level data on these variables.

4.3.4 Statistical analysis

We graphed the trends over time in rates of recommended follow-up and of follow-up within seven days, and repeated both of these graphs stratifying the cohort by material deprivation quintile. We used log binomial generalized estimating equations to model the relationship between implementation of the guidelines and recommended follow-up at the level of the individual. The model fit a change in level but not a change in slope. We included all of the pre-selected covariates and accounted for clustering by hospital. We also adjusted for an underlying linear time trend by including a quarter variable in the model – the quarter variable was comprised of consecutively numbered three month blocks based on the date of the baby’s hospital discharge. To conduct our sensitivity analysis, we ran the same analysis using follow-up within seven days as the outcome. To examine SES disparities, we added a product (a.k.a. interaction) term between guideline implementation and material deprivation quintile to the original model in order to compare the association between guideline implementation and follow-up across SES quintiles. Cases with missing values for any covariate were excluded from the models. All analyses were conducted using SAS 9.3 (SAS Institute, Inc, Cary, NC).

4.4 Results

Characteristics of the study cohort are presented in Table 1. There are small differences in hospital characteristics, birth mode, day of discharge, and SES when babies born prior to implementation of screening are compared to those born after screening implementation. Given our very large sample, we did not conduct statistical testing as it would invariably lead to statistical significance even for very small absolute differences.

The basic trends over time in rates of recommended follow-up and follow-up within seven days are shown in Figures 2-A and 2-B. Both graphs show a vertical line at the second quarter of 2007 to indicate the release of the CPS hyperbilirubinemia guidelines. While the rate of recommended follow-up rose during the study period, the rate of overall follow-up within seven days remained stable over the same time period. There was an underlying temporal trend

of an increase in rates of recommended follow-up of 0.6% relative to the preceding quarter. Figures 3-A and 3-B illustrate the same outcomes stratified by material deprivation quintile. The rate of recommended follow-up rose in all quintiles but most sharply in the highest SES quintile, while the rates of follow-up within seven days remained stable across all quintiles. (See Figure A7.1 in Appendix 8 for a graph showing the proportion of babies within each fiscal year that had their first visit on each of the first seven days following discharge.)

Table 2 shows the results of the models for both recommended follow-up and follow-up within seven days. After babies with missing values for any covariate were excluded, 711,242 babies were included in all the models. After adjusting for covariates, implementation of screening was associated with a modest increase in receiving recommended follow-up (adjusted RR: 1.11, CI: 1.0014 to 1.22, $p=0.047$), but no difference in having a follow-up visit within seven days of birth. This effect size is approximately the equivalent of the increase that was observed over a five year period due to the underlying temporal trend. Both models demonstrated a significant gradient in outcomes based on material deprivation, with lower SES being associated with a lower probability of having a follow-up visit. Timing of recommended follow-up was a strong predictor of follow-up, with the adjusted RR of recommended follow-up being 0.34 (CI: 0.26 to 0.43, $p<0.0001$) if the day of recommended follow-up fell on a weekend or holiday.

Including a product term with screening implementation and material deprivation quintile in the model of recommended follow-up demonstrated a significant interaction between these two variables. Table 3 shows the crude rates, crude percentage increase in rates, crude RRs, adjusted RRs with confidence intervals and p-values for each material deprivation quintile for births before and after screening implementation. There was a striking SES gradient in the crude percentage increase in the rate of recommended follow-up associated with screening implementation with observed increases ranging from 0.3% in the lowest quintile to 29.0% in the highest quintile. After adjusting for confounding variables, the RRs of recommended follow-up reflect this same gradient. When rates of follow-up in each of the lowest four quintiles are compared to that of the highest quintile, the adjusted RRs illustrate a shift towards increased disparity following implementation of screening. Based on the crude rates of recommended follow-up in each quintile, the unadjusted attributable contribution to the increase in the overall rate of recommended follow-up after implementation of screening was 40% for Q1 (Highest

SES), 31% for Q2, 23% for Q3, 6 % for Q4, and 0% for Q5 (Lowest SES). (See Appendix 9 for details of calculations.)

We conducted a post-hoc descriptive analysis which showed that babies in the highest SES quintile were more likely to have their first visit with a paediatrician as opposed to a family physician (36.2% in Q1, versus 27.2%, 26.0%, 29.2% and 31.0% in Q2-5 respectively), and although overall only 29.7% of babies had their first visit with a paediatrician, on days one to three, 33.8%, 33.1%, and 34.6% of babies having their first visit on each of these respective days were seen by a paediatrician. (See Appendix 10 for details.)

4.5 Discussion

Implementation of universal bilirubin screening in Ontario in response to the CPS hyperbilirubinemia guidelines was associated with a modest increase in rates of early follow-up. The effect size that we observed was within the range of previously reported effect sizes associated with guideline dissemination.(91, 92) However, even following the implementation of screening, the majority of babies were not seen within recommended timeframes. Furthermore, despite a slight shift towards earlier visits, the proportion of babies who do not receive any visit during the first week following discharge remained stable throughout the study period at 20%. There was a significant gradient in the likelihood of receiving recommended follow-up across material deprivation quintiles, and screening implementation was associated with the greatest increase in rate of recommended follow-up for babies of the highest SES. Disparities in recommended follow-up not only persisted, but increased following the implementation of screening.

A major strength of our study is that it was population-based, and included complete follow-up of outcomes for the entire cohort. We had a 97% response rate to the survey that we used to determine if and when hospitals had implemented the CPS guidelines. Our study is the first to examine the impact of universal bilirubin screening on newborn follow-up. Previous research had shown a gradient in Ontario newborn's access to follow-up within one week of hospital discharge based on neighbourhood income,(58) but our study provides new information regarding rates of follow-up within a shorter timeframe (aimed at catching rare cases where a significant unanticipated increase in bilirubin levels occurs after initial bilirubin screening). Our findings confirm that babies of the lowest SES are at the greatest risk of inadequate follow-up,

and demonstrate that implementation of universal bilirubin screening in response to the CPS hyperbilirubinemia guidelines has actually been associated with an increase in disparity in follow-up. While some have expressed the opinion that adherence to evidence-based guidelines has the potential to reduce disparities in health care,(93) experimental research has found that use of a guideline may have variable impact on disparities in treatment.(94) It has been suggested that patient, provider and organizational factors all play a role in variations observed in the application of clinical guidelines.(95) A survey of Ontario health care providers assessing awareness of the CPS hyperbilirubinemia guidelines provides insight into provider factors that may contribute to the observed disparity. The survey found paediatricians were most aware of the guidelines, and that 89% of paediatricians reported completing newborn follow-up within 72 hours after discharge versus 60% of family physicians.(96) Post-hoc analysis of our cohort showed that babies in the highest SES quintile were most likely to have their first physician visit with a paediatrician, suggesting that a combination of provider awareness of the guidelines and parental choice of provider and/or access to provider group may account for some of the disparity observed. Of further note, the survey asked about follow-up within 72 hours, which exceeds the timeframe recommended in the CPS guideline and may reflect a prevailing acceptance that physicians will not be available for routine follow-up on weekends.

As an observational study using historical controls, our study is subject to the limitations introduced by the possibility of unobserved differences between the exposed and unexposed groups. We endeavoured to minimize bias by modelling outcomes at the individual level and adjusting for confounding variables and underlying temporal trends. The use of administrative health data also has some inherent limitations. One major concern with administrative health data is the fidelity of diagnostic information. Data re-abstraction studies examining the quality of data captured in the DAD in Ontario have found very high levels of agreement (100% for most variables) for non-medical data such as birth date, date of discharge, etc.(97) Levels of agreement for interventions have been found to be slightly lower (97%) and are even lower for diagnoses (95% of records match conditions, though some may be coded with more or less specificity).(97) In order to reduce the risk of changes in coding over time, we selected a study period that began one year after the ICD-10 coding system was implemented in Ontario.

As was the case in our study, another challenge with using administrative health data is that the available administrative data sources sometimes do not capture all of the information that

would ideally be used to answer the research question. We defined our primary and secondary outcomes using public health insurance (OHIP) billings which do not capture follow-up visits provided by public health nurses or hospital-based nurses, visits provided by a small number of physicians who work under alternate payment plans (e.g., physicians who work at community health centres, which serve 0.9% of the population (98)), or visits provided by a small number of salaried nurse-practitioners who work with physicians who do not bill for the services the nurse-practitioners provide. As of 2006 it was estimated that just over 5% of Ontario physicians worked primarily in a non-fee-for-service model, but three-quarters of them still submitted some billings to OHIP.(99) The use of OHIP billings to measure follow-up visits likely under-estimates the proportion of babies who received follow-up from any health care provider within the recommended timeframe. Given that there is not a system of universal nursing follow-up for newborns in Ontario, and given that public health nurse visits typically occur six to seven days after birth(59) we did not anticipate that this limitation would grossly over-estimate the proportion of newborns who do not received recommended follow-up. Based on comparison with Ontario's provincial midwifery database,(100) it is possible that a small portion of babies in the cohort (approximately 6800, or <1%) received postpartum care from a midwife. Given the midwifery model in Ontario, our estimates for the effect of being discharged on day 1 and the effect of having no prenatal care should be interpreted with caution. Another limitation is that the date of guideline implementation was determined retrospectively and by survey data which may have decreased accuracy. Our decision to impute the month of implementation when missing may have resulted in the misclassification of exposure for <1.7% of the cohort – we chose the mid-point in the year as the cut point for imputed values in order to minimize bias in the direction of any misclassification. Finally, we did not have individual-level SES data, so we used a postal-code derived neighbourhood-level variable based on census data. The use of neighbourhood-level variables when individual-level SES data is not available has been established as a reasonable approach based on evidence that the associations between health outcomes and neighbourhood-level SES tend to be similar to the associations between health outcomes and individual-level SES.(101, 102) Previous Ontario studies using this approach have demonstrated associations between health care use and neighbourhood-level SES in the pediatric population.(58, 103) However, it has been noted that neighbourhood-level variables measure contextual factors (as opposed to simply being proxies for individual-level data) and may have

differing effect sizes than individual-level variables, and are therefore most appropriately conceived as neighbourhood-level variables rather than substitutes for absent individual-level data.(101, 104)

At a specific level, our findings have implications for the issue of post-discharge follow-up of newborns. Implementation of the CPS guidelines has had a limited impact in ensuring timely follow-up for all newborns in Ontario and lack of timely follow-up represents an ongoing weakness in efforts to prevent severe hyperbilirubinemia. The majority of babies are still not seen within the recommended time frame, and babies of lowest socio-economic status are at greatest risk of falling through the cracks. Our finding of the significant association between timing of discharge and recommended follow-up highlights that access to care on weekends and holidays is a significant barrier to recommended follow-up. Efforts to improve rates of follow-up should take a multi-pronged approach that involves both increasing awareness of the recommendations among clinicians and parents, and developing solutions to overcome existing barriers to access. While the onus is on “the health care system” to ensure that care is accessible, the lack of integration between different sectors of the system remains a challenge, and the collaborative efforts of prenatal care providers, hospitals, and providers of primary care to newborns are needed to successfully improve newborn follow-up. In our survey of Ontario hospitals, respondents reported developing new hospital-based clinics staffed by either nurses and/or physicians to ensure follow-up seven days a week for babies identified as being at high risk for severe hyperbilirubinemia.(89) This trend runs counter to existing Ontario health policy which favours community-based service delivery when that option is safe, effective, and less expensive.(105) However, policy efforts in Ontario to improve access to primary care services through changes to family physician remuneration has not had an impact on many measures of access such as Emergency Department use,(98) highlighting the challenges of improving access to primary care services. Evaluation of community-based TcB screening within a program of universal follow-up by public health nurses in another Canadian jurisdiction found significant improvements in both safety and resource utilization.(24) It is possible that home-based follow-up may help to minimize barriers underlying disparities in follow-up.(106) Additional research probing the family perspective of the barriers would be helpful in developing solutions. Although there is no consensus in the research literature with respect to the best approach to ensure appropriate neonatal follow-up care, Canadian research suggests that effective

coordination between hospitals and community-based perinatal services can result in improved health outcomes while making efficient use of resources,(24, 107) and our findings suggest there remains room for improvement at a system level in the transition from hospital to home for Ontario newborns.

Finally, our findings also have more general implications related to the impact of clinical guidelines on health disparities. Implementation of the CPS hyperbilirubinemia guidelines appears to have had the unintended consequence of increasing disparity in rates of recommended follow-up for newborns. This finding is consistent with previous research in Ontario which has shown that even within the context of universal health care, efforts to improve access to care can result in increased disparity in health service utilization.(108) While we hypothesize that our findings may be partially explained by differences between SES groups in provider types, further research examining patient, provider, and organizational factors is needed to fully understand the observed increase in disparity. Our findings suggest that when disparities exist, implementing guidelines which fail to take into account and address the root of these disparities may lead to improvements in outcomes at a population level but increased inequity within that population. As a corollary, our findings suggest that in order for clinical practice guidelines to reduce health disparities, attention to reducing disparities should be incorporated from the beginning of the guideline development process through to the knowledge translation and implementation stages.

Figure 4.1. Cohort Creation Flow Diagram

802,318 records of live births that met the following inclusion criteria were extracted:

- Baby was discharged between April 1, 2003 and February 28, 2011
- Baby's health record is linked to a single maternal health record
- Baby's health record has a valid OHIP number
- Maternal postal code is known and indicates Ontario residence
- Baby's gestational age at birth is known and is at least 35 weeks
- Baby was discharged directly to home within 72 hours of birth

68,328 (8.52%) of the babies were excluded, for the following reasons:

Reason for Exclusion	n (%)
• Baby died before discharge or on the day of discharge	353 (0.04)
• Were triplets or quadruplets, or randomly selected twin from twin sibling pairs	4294 (0.54)
• Were born at a hospital that did not provide maternity services, or that closed maternity services prior to March 2011	2491 (0.31)
• Mother had prenatal care from a midwife	57,273 (7.14)
• Were missing information about length of stay (LOS) or had an LOS less than 0 hours	207 (0.03)
• Were born at a hospital (n=3) that did not respond to the survey	3710 (0.46)

733,990 babies were included in the analytic cohort

Table 4.1. Cohort Characteristics by Screening Implementation Status

	Total		Screening implementation status			
	N	%age	Screening Not Implemented		Screening Implemented	
	N	%age	N	%age	N	%age
Gestational age at birth						
35-38 weeks	203207	27.7	154238	27.3	48969	28.8
39+ weeks	530783	72.3	410004	72.7	120779	71.2
Mode of birth						
Missing	3387	0.5	3384	0.6	3	0
Spontaneous Vaginal	517970	70.6	400228	70.9	117742	69.4
Assisted Vaginal	88346	12	68696	12.2	19650	11.6
Cesarean Section	124287	16.9	91934	16.3	32353	19.1
Maternal Parity						
Missing	15	0	13	0	2	0
Multiparous	429204	58.5	330362	58.5	98842	58.2
Primiparous	304771	41.5	233867	41.4	70904	41.8
Day of Newborn Discharge						
Day 1 (≤ 24 h)	23108	3.2	20051	3.6	3057	1.8
Day 2 (>24-48 h)	459153	62.6	352438	62.5	106715	62.9
Day 3 (>48-72 h)	251729	34.3	191753	34.0	59976	35.33
Timing of recommended Follow-Up						
Not weekend or holiday	488852	66.6	375398	66.5	113454	66.8
Weekend or holiday	245138	33.4	188844	33.5	56294	33.2
Maternal Prenatal Care Provider						
No Prenatal Care	7162	1	5691	1	1471	0.9
Prenatal Care – Any GP Care	437360	59.6	335765	59.5	101595	59.9
Prenatal Care – OB & other	289468	39.4	222786	39.5	66682	39.3

Table 4.1. Cohort Characteristics by Screening Implementation Status (cont'd.)

	Total		Screening implementation status			
	N	%age	Screening Not Implemented		Screening Implemented	
	N	%age	N	%age	N	%age
Mother's age at first birth						
<19	47790	6.5	37400	6.6	10390	6.1
19+	686200	93.5	526842	93.4	159358	93.9
Maternal Residence						
Missing	582	0.1	467	0.1	115	0.1
Rural	73363	10	57615	10.2	15748	9.3
Urban	660045	89.9	506160	89.7	153885	90.7
Material Deprivation Quintile						
Unknown	18881	2.6	14462	2.6	4419	2.6
Q1 (High SES)	126801	17.3	92149	16.3	34652	20.4
Q2	146887	20	108315	19.2	38572	22.7
Q3	150367	20.5	115291	20.4	35076	20.7
Q4	156944	21.4	124527	22.1	32417	19.1
Q5 (Low SES)	134110	18.3	109498	19.4	24612	14.5
Hospital Type						
Non-teaching large	511318	69.7	399478	70.8	111840	65.9
Non-teaching small	52257	7.1	41544	7.4	10713	6.3
Teaching	170415	23.2	123220	21.8	47195	27.8
Average Annual Birth Volume						
< 240	18369	2.5	14963	2.7	3406	2
240-500	32400	4.4	26901	4.8	5499	3.2
501-1000	63627	8.7	46871	8.3	16756	9.9
> 1000	619594	84.4	475507	84.3	144087	84.9
Level of Newborn Services						
Level 1 (lowest)	82926	11.3	69976	12.4	12950	7.6
Level 2	539421	73.5	410028	72.7	129393	76.2
Level 3 (highest)	111643	15.2	84238	14.9	27405	16.1

Figure 4.2. Frequency of Recommended Follow-up by Quarter

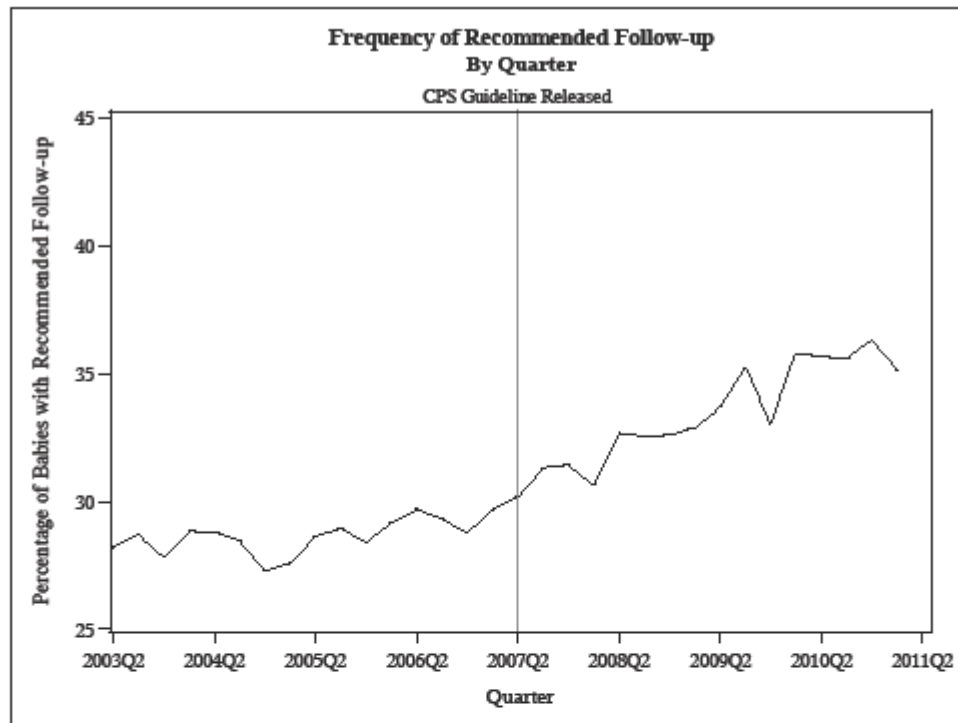


Figure 4.3. Frequency of Follow-up Within Seven Days by Quarter

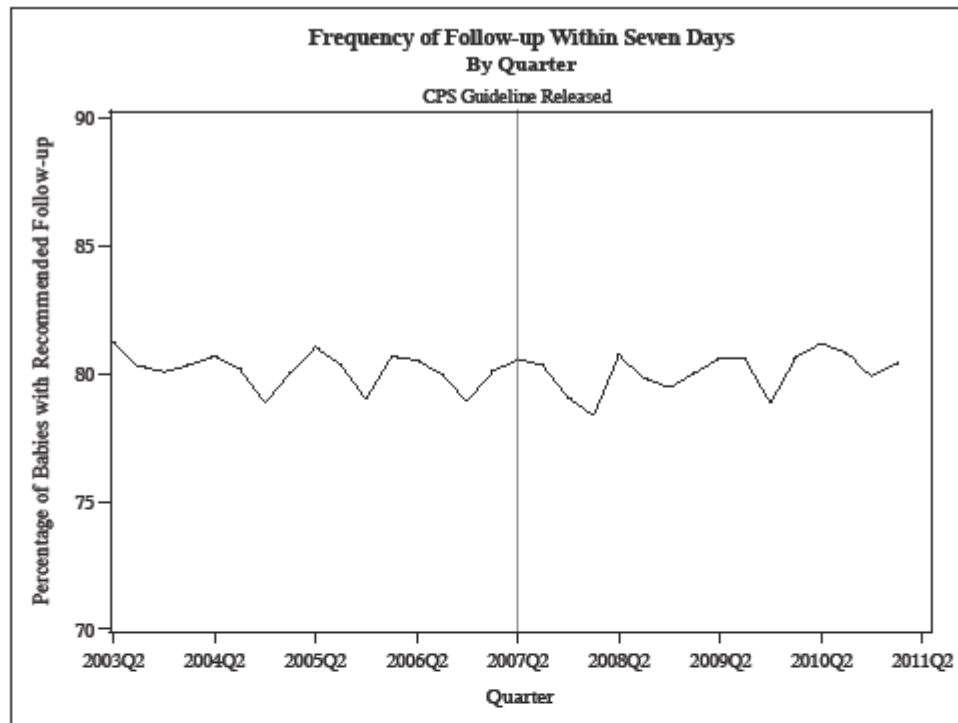


Figure 4.4. Frequency of Recommended Follow-up by Material Deprivation Index

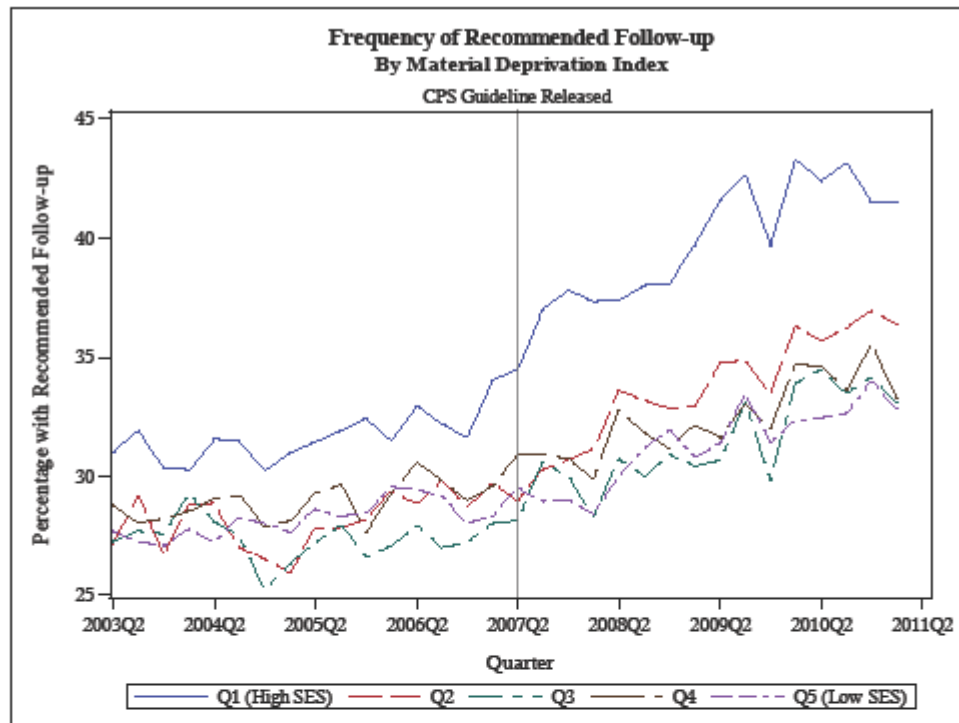


Figure 4.5. Frequency of Follow-up within Seven Days by Material Deprivation Index

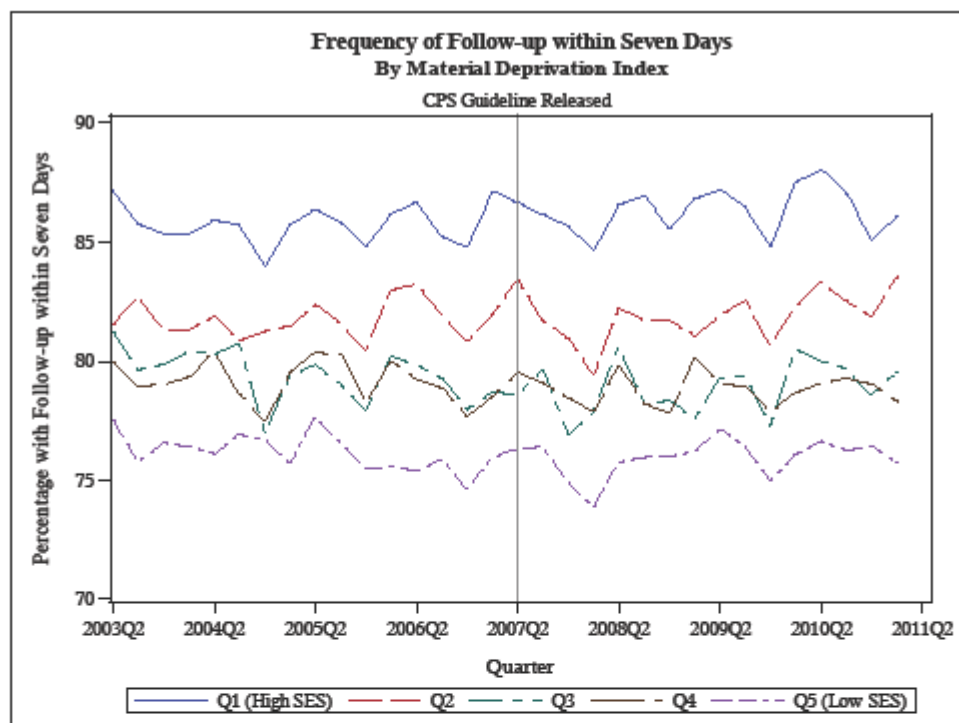


Table 4.2. Models of Recommended Follow-Up and Follow-Up within 7 Days

Variable	Crude Rate of Recommend ed Follow- Up	Unadjusted RR of Recommended Follow-up	Adjusted RR of Recommended Follow-Up (95% CI)	p	Crude Rate of Follow-Up within 7 days	Unadjusted RR of Follow- Up within 7 days	Adjusted RR of Follow-Up within 7 days (95% CI)	p
Overall	31.2				80.3			
Intercept	.		0.44 (0.34,0.56)	<.0001	.		0.75 (0.71,0.78)	<0.0001
Quarter (temporal trend)	.		1.01 (1.00,1.01)	0.0031	.		1.00 (1.00,1.00)	0.2516
Screening Implementation								
Screening Implemented	35.0	1.17	1.11(1.00,1.22)	0.0468	79.2	0.99	1.01 (0.99,1.03)	0.1836
Screening Not Implemented	29.9	1.00	1.00	.	80.4	1.00	1.00	.
Gestational Age								
35-38 weeks	32.4	1.06	1.03 (1.02,1.05)	<.0001	81.0	1.02	1.01 (1.01,1.02)	<.0001
39+ weeks	30.6	1.00	1.00	.	79.8	1.00	1.00	.
Parity								
Multiparous	28.6	0.83	0.88 (0.85,0.90)	<.0001	78.7	0.96	0.96 (0.95,0.97)	<.0001
Primiparous	34.6	1.00	1.00	.	82.2	1.00	1.00	.
Mode of Birth								
Caesarean	29.4	0.95	0.86 (0.82,0.89)	<.0001	79.9	1.00	0.98(0.97,0.98)	<.0001
Assisted Vaginal	34.3	1.11	1.02 (1.00,1.03)	0.0569	82.4	1.03	1.01 (1.00,1.01)	0.0002
Spontaneous Vaginal	30.9	1.00	1.00	.	79.8	1.00		.
Day of Discharge								
Day 1 (≤ 24 h)	7.7	0.24	0.34 (0.26,0.45)	<.0001	72.9	0.91	0.93 (0.91,0.95)	<.0001
Day 2 (>24-48 h)	31.5	0.97	0.93 (0.90,0.96)	<.0001	80.3	1.00	0.98 (0.98,0.99)	<.0001
Day 3 (>48-72 h)	32.5	1.00	1.00	.	80.5	1.00	1.00	.

Table 4.2. Models of Recommended Follow-Up and Follow-Up within 7 Days (cont'd.)

Timing of recommended

follow-up day								
Weekend or holiday	13.6	0.34	0.34 (0.26,0.43)	<.0001	80.4	1.01	1.02 (1.01,1.03)	0.0012
Not weekend or holiday	39.8	1.00	1.00	.	80.0	1.00	1.00	.
Prenatal Care								
No Prenatal Care	12.2	0.40	0.51 (0.40,0.64)	<.0001	40.0	0.49	0.46 (0.34,0.61)	<.0001
Prenatal Care - Ob or other	32.7	1.08	0.95 (0.91,0.99)	0.0174	79.6	0.98	0.92 (0.89,0.94)	<.0001
Prenatal Care – Any GP Care	30.3	1.00	1.00	.	81.2	1.00	1.00	.
Mother's age at first birth								
<19 at first birth	19.7	0.62	0.85 (0.82,0.88)	<.0001	65.0	0.80	0.89 (0.88,0.91)	<.0001
19+ at first birth	31.9	1.00	1.00	.	81.2	1.00	1.00	.
Maternal Residence								
Rural	14.2	0.43	0.90 (0.85,0.95)	0.0004	61.0	0.74	0.94 (0.91,0.97)	<0.0001
Urban	32.9	1.00	1.00	.	82.3	1.00	1.00	.
Material Deprivation Quintile								
Q5 (Low SES)	29.7	0.83	0.86 (0.82,0.89)	<.0001	76.0	0.88	0.92 (0.90,0.93)	<.0001
Q4	30.6	0.86	0.91 (0.88,0.93)	<.0001	79.0	0.92	0.95 (0.94,0.96)	<.0001
Q3	29.4	0.82	0.93 (0.91,0.95)	<.0001	79.2	0.92	0.97 (0.96,0.98)	<.0001
Q2	31.0	0.87	0.96 (0.95,0.98)	<.0001	81.8	0.95	0.99 (0.98,0.99)	0.0003
Q1 (High SES)	35.7	1.00	1.00	.	86.1	1.00	1.00	.

Table 4.3. Estimates of RR of Recommended Follow-Up by Material Deprivation Quintile (Before and After Screening Implementation)

Quintile	Crude Rate of Recommended Follow-Up		Crude % Increase in Recommended Follow-Up	Crude RR of Recommended Follow-Up		Adjusted RR of Recommended Follow-Up (95% CI)		p-value for adjusted RR	
Overall	29.9	35.0	17.1	1.17		1.11		0.0468	
	Before	After		Before	After	Before	After	Before	After
Q5 (Low SES)	29.7	29.8	0.3	0.90	0.70	0.90 (0.86,0.94)	0.87 (0.79,0.95)	<0.0001	0.003
Q4	30.3	31.9	5.3	0.92	0.75	0.95 (0.91,0.98)	0.88 (0.81,0.95)	0.002	0.002
Q3	28.1	33.5	19.2	0.85	0.78	0.96 (0.93,0.99)	0.90 (0.84,0.97)	0.01	0.004
Q2	29.2	36.0	23.3	0.88	0.84	0.99 (0.97,1.02)	0.92 (0.87,0.97)	0.5	0.002
Q1 (High SES)	33.1	42.7	29.0	1.00	1.00	1.00	1.00	.	.

Other variables in the model: hospital, quarter (temporal trend), gestational age, parity, mode of birth, day of discharge, timing of recommended follow-up (weekend or holiday), prenatal care, mother's age at first birth, maternal residence (rural/urban)

Chapter 5

Discussion

This final chapter draws together the findings from the three projects to present an overall summary of the contributions of this dissertation. First we review the objectives for each project and evaluate whether the objectives were met. Next we provide a synthesis of the key findings and messages related first to the impact of the CPS hyperbilirubinemia guidelines in Ontario and second to role of SES disparities in the prevention of severe hyperbilirubinemia. The following section outlines the knowledge translation plan for our research. Finally a brief summary of the overall work is provided.

5.1 Review of Thesis Objectives

5.1.1 Project 1

The objectives of this project were to determine if and when hospitals had implemented universal bilirubin screening, to investigate the organization of services to provide follow-up and treatment, and to identify factors and processes influencing screening implementation.

We achieved a very high response rate to our survey with 97 of 100 eligible hospital sites responding. The survey collected information on the CPS guideline implementation dates which allowed us to describe uptake of the guideline and to conduct our subsequent projects which relied on this data. Respondents described a variety of new processes and services developed to support implementation of the guidelines including a notable shift towards hospital-based post-discharge follow-up for newborns identified as being at high risk for severe hyperbilirubinemia. The survey also gathered information on the factors that influenced whether or not the guidelines were implemented. Overall the objectives of this project were met, and our findings supported our original hypothesis that the majority of hospitals had implemented universal bilirubin screening but with variation in how services were organized.

5.1.2. Project 2

The objective of the second project was to evaluate the association between the implementation of universal bilirubin screening and five health service utilization outcomes.

Contrary to our original hypothesis that we would find the implementation of universal bilirubin screening to be associated with increases in all of the outcomes, after adjusting for pre-existing temporal trends and other important covariates, screening was only associated with an increase in phototherapy during hospitalization at birth (relative risk (RR) 1.32, 95% confidence interval (CI) 1.09-1.59). We found universal screening to be associated with a decrease in jaundice-related ED visits (RR 0.79, 95% CI 0.64-0.96), but no statistically significant difference in phototherapy after discharge, length of stay, or jaundice-related readmissions. The objectives of this project were met.

5.1.3 Project 3

The objectives of the third project were to determine whether implementation of the CPS hyperbilirubinemia guidelines was associated with an increase in access to recommended follow-up care, and if so, whether this differed between SES quintiles.

In keeping with our hypothesis, implementation of the CPS hyperbilirubinemia guideline was associated with a modest increase in receiving recommended follow-up (adjusted RR: 1.11, CI: 1.0014 to 1.2239, $p=0.0468$), but there was a strong SES gradient in this outcome. The crude percentage increase in the rate of recommended follow-up associated with guideline implementation ranged from 0.3% in the lowest quintile to 29.0% in the highest quintile. After adjusting for confounding variables, the adjusted RRs comparing rates of follow-up in each of the lowest four quintiles to that of the highest quintile illustrate a shift towards increased disparity following implementation of the guidelines. The objectives of this project were also met.

5.2 KEY FINDINGS AND MESSAGES

5.2.1 Impact of the CPS hyperbilirubinemia guidelines in Ontario

This dissertation has provided new evidence on the impact of the CPS hyperbilirubinemia guidelines in Ontario with respect to guideline adoption by hospitals, healthcare utilization, and the timing of newborn follow-up. Below are highlights of these findings:

- Uptake of the CPS hyperbilirubinemia guidelines by Ontario hospitals occurred gradually, but by 5 years following their release the majority of hospitals had adopted the guidelines and nearly 90% hospital births occur at a hospital that has implemented universal bilirubin screening.
- Hospitals adopted the guidelines in an ad hoc process which allowed them to develop tailor-made approaches to service organization. However, provincial coordination of guideline implementation might have offered some efficiency, such as allowing hospitals to benefit from the experiences of others by sharing solutions to challenges, and facilitating timelier implementation in low-volume hospitals with limited human resources for guideline implementation.
- Implementation of the guidelines has been associated with increased use of hospital-based neonatal follow-up care. This runs counter to current Ontario health policy directions which aim to deliver services in the community when that option is safe, effective, and less expensive.
- After accounting for pre-existing trends, implementation of universal screening was associated with an increase in phototherapy before discharge, a decrease in jaundice-related ED visits and no differences in length of stay, phototherapy after initial discharge, and jaundice-related readmission. These findings are specific to the Ontario context, including a growing awareness of neonatal hyperbilirubinemia which preceded the release of the guidelines. The improved organization of follow-up care for at-risk babies reported by hospitals likely explains the reduction in jaundice-related ED visits.
- Data on breastfeeding would be helpful in further evaluating the impact of the guidelines.
- The impact of universal screening on health care use in Ontario differed from the findings of other studies and highlights the limitations in the generalizability of previous research and the importance of context-specific local evaluation of guideline implementation.
- Our findings also highlight the limitations of previous economic analyses, including a cost-effectiveness analysis specific to Ontario, and provide a foundation for a more accurate assessment of the cost implications of universal bilirubin screening in Ontario.

- Implementation of the CPS guidelines has had a limited impact in ensuring timely follow-up for all newborns in Ontario. After excluding babies in midwifery care, less than 40% of babies who leave the hospital within 72 hours of birth have a follow-up visit within the recommended time-frame. Approximately 20% of these babies do not have any follow-up visit in the first seven days of life and this has not changed following guideline implementation.
- Access to care on weekends and holidays is a significant barrier to recommended follow-up.
- Lack of timely follow-up represents an ongoing weakness in efforts to prevent severe hyperbilirubinemia. There is room for improvement at a system level in the transition from hospital to home for Ontario newborns.

5.2.2 Prevention of severe hyperbilirubinemia and SES disparities

This dissertation has provided a novel investigation of role of SES disparities in newborn follow-up as potential source of inequity in the prevention of severe hyperbilirubinemia. These are key findings related to this dimension of our research:

- Previous studies suggest that sub-optimal follow-up remains a potential causal factor in cases of severe hyperbilirubinemia that occur despite universal bilirubin screening. Our research confirms that in Ontario there is a strong SES gradient in access to timely follow-up care as recommended by the CPS hyperbilirubinemia guideline, with babies in the lowest SES quintile being the least likely to receive recommended follow-up and babies in the highest SES quintile being the most likely to receive it.
- Guideline implementation was associated with a modest increase in the proportion of babies receiving recommended follow-up care but 40% of this gain was attributable to the highest SES quintile and none of it to the lowest SES quintile. In other words, the SES gradient in access to timely follow-up became more pronounced after guideline implementation.
- Further research is required to understand the factors that contribute to the gradient in newborn access to recommended follow-up care. Better understanding of the barriers in access is a prerequisite to developing solutions such as new service delivery models aimed at improving post-discharge follow-up.
- The observed widening of the SES disparity in access to recommended follow-up associated with implementation of the CPS guideline illustrates that universal programs which fail to address root causes of disparities may lead to overall improvements in population outcomes but increased inequity.

- In order for clinical practice guidelines to be universally effective, explicit strategies to reduce disparities should be incorporated from the beginning of the guideline development process through to the knowledge translation and implementation stages.

5.4 KNOWLEDGE TRANSLATION PLAN

5.4.1 Knowledge Translation Goals

The goals of our knowledge translation plan are to communicate the key findings and messages identified in the previous two sections in order to increase knowledge and awareness regarding the impact of the CPS hyperbilirubinemia guidelines, to inform policy and clinical guidelines regarding the organization of newborn health services related to the transition from hospital to home, to stimulate future research that builds on our findings, and to improve newborn access to recommended follow-up care and reduce disparity in such access.

5.4.2 Knowledge-User Audience

In order to meet these goals, our audience will include clinicians providing newborn care (e.g., paediatricians and family physicians), managers and administrators responsible for maternal newborn programs, health care policy makers (i.e., the Ontario Ministry of Health & Long Term Care (MOHLTC) via the Provincial Council for Maternal Child Health (PCMCH)), the public (and particularly expectant parents) via the media, researchers, and professional associations which develop guidelines and disseminate knowledge around their implementation (in particular the CPS).

5.4.3 Knowledge Translation Strategy

The following diffusion-based strategies will be used: publication in peer-reviewed journals, simultaneous publication of ICES studies on ICES web-site, presentations at conferences and BORN provincial rounds.

The following dissemination-based strategies will be employed: media releases summarizing results and key messages to coincide with publications, the use of social media to communicate with parents and other members of the public, and summary briefing to key stakeholders (i.e., the

Canadian Paediatric Society, and the Ontario MOHLTC through PCMCH). Findings of the first project have already presented directly to two provincial committees, the Neonatal Hyperbilirubinemia Task Force of the Maternal-Child Screening Committee (reporting to PCMCH & BORN-Ontario), and the Hyperbilirubinemia Clinical Expert Advisory Group (reporting to PCMCH) by ED, who was an invited member of both of these groups.

Our findings point to a need for provincial coordination of a knowledge translation strategy to support the implementation of maternal newborn guidelines in general, and in particular to support further improvements in the quality of care related to the prevention of severe hyperbilirubinemia. The following provincially coordinated strategies could be used to translate our research findings into direct improvements in the health care system:

- Development of a provincial guideline or directive on universal hyperbilirubinemia screening and subsequent treatment and follow-up which hospitals are mandated to adopt (PCMCH and the MOHLTC have published a clinical handbook on hyperbilirubinemia in 2013 but hospitals have not been mandated to adopt its recommendations)(109)
- Use of interactive webinars for knowledge exchange to support hospitals in the process of implementing such a directive
- Development of a performance evaluation framework for the prevention of severe hyperbilirubinemia which incorporates performance evaluation metrics that measure timeliness of newborn post-discharge follow-up
- Regular reporting to hospitals and community care providers based on such an evaluation framework (i.e., a form of audit and feedback)
- Funding for research to investigate the patient, provider and organizational barriers to timely newborn follow-up in order to develop strategies to improve newborn follow-up, and further funding to implement and evaluate such strategies.

5.4.4 Evaluation of Knowledge Translation Plan

Evaluation of our knowledge translation plan will be guided by the following questions:

- Were the results published in peer-reviewed journals?
- Were the findings presented at conferences?

- Were the findings reported in the media?
- Were the findings communicated directly to key stakeholders?
- Was a provincial strategy to improve the quality of care related to universal bilirubin screening and subsequent follow-up developed, enacted, and evaluated?

5.5 SUMMARY

By 2012, the majority of Ontario hospitals had implemented universal bilirubin screening. There is heterogeneity in how hospitals organize services, but a notable trend towards hospital-based post-discharge care. Screening was associated with an increase in phototherapy during hospitalization at birth (relative risk (RR) 1.32, 95% confidence interval (CI) 1.09-1.59), and a decrease in jaundice-related ED visits (RR 0.79, 95% CI 0.64-0.96), but no statistically significant difference in phototherapy after discharge, length of stay, or jaundice-related readmissions after accounting for pre-existing temporal trends in healthcare service use and other patient socio-demographic and hospital characteristics. Implementation of the guideline in Ontario was associated with a modest increase in rates of early follow-up (adjusted RR 1.11, CI 1.0014-1.22, $p=0.0468$), but most babies were not seen within the recommended timeframe. Babies of lowest SES were least likely to receive recommended follow-up, and disparities in follow-up increased following guideline implementation.

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Appendices

Appendix 1. Summary of studies examining the impact of universal pre-discharge bilirubin screening

Table A1.1. Summary of studies examining the impact of universal pre-discharge bilirubin screening

Study	Setting	Design	Sample Size	Outcomes
Eggert et al., 2006	18-hospital health system in Utah - Mar 1, 2001 to Dec 31, 2004	retrospective cohort study	101 272 infants 48,789 before and 52,483 after	- severe hyperbilirubinemia (≥ 20 mg/dL) decreased from 1:77 to 1:142 ($p < 0.0001$) - critical hyperbilirubinemia (≥ 25 mg/dL) decreased from 1:1522 to 1:4037 ($p < 0.005$) - hospital readmission decreased from 0.55% to 0.43% ($p < 0.005$)
Kuzniewicz et al., 2009	11 hospitals in Northern California Kaiser Permanente Medical Care Program - Jan 1, 1995 to June 30, 2007	retrospective cohort study	358 086 infants 319 904 before and 38 182 after	- lower risk of TSB levels > AAP exchange guidelines (0.17% vs. 0.45%, $p < 0.001$) - higher overall risk of being treated with phototherapy (9.1% vs. 4.2%, $p < 0.001$) - higher risk of readmission for phototherapy (2.7% vs. 1.6%, $p < 0.001$) - slightly longer length of stay at birth (50.9 vs. 48.7 hours, $p < 0.001$)
Mah et al., 2010	116 hospitals across U.S. (Hospital Corporation of America) - May 1, 2004 to Dec 31, 2008	prospective cohort study	1 028 817 infants 129 345 before and 899 472 after	- total bilirubin 25.0-29.9 mg/dL decreased from 43/100,000 (1:2325) to 27/100,000 (1:3703) ($p = .0019$) - total bilirubin ≥ 30.0 mg/dL decreased from 9/100,000 to 3/100,000 ($p = .0051$)

Table A1.1. Summary of studies examining the impact of universal pre-discharge bilirubin screening (cont'd)

Study	Setting	Design	Sample Size	Outcomes
Mah et al., 2010 (continued)				• phototherapy not reported for pre/post groups, increased from 4.4% in 2004 to 5.1% in 2008 ($p < .001$) • 94.2% of babies who received phototherapy were treated during birth admission only
Wainer et al., 2012	• Calgary Health Region (AB, Canada) • Jun 1, 2006 to May 31, 2008	• prospective cohort study with historical control group	• 28 908 infants • 14 112 before and 14 796 after	• 54.9% reduction in incidence of TSB >342 mol/L/ 20 mg/dL (odds ratio: 2.219, 95% confidence interval: 1.543-3.193; $p < 0.0001$) • Reductions in overall phototherapy rate (5.27% vs 4.3%, OR: 1.241(1.122-1.374), $p < 0.0001$), age at readmission for phototherapy (104.3 ± 52.1 vs 88.9 ± 70.5, $p < 0.005$), and duration of phototherapy readmission (24.8 ± 13.6 vs 23.2 ± 9.8, $p < 0.05$)

Appendix 2. Research Ethics Board Approvals

Figure A2.1. Original letter of approval for project 1 from Ottawa Hospital Research Ethics Board

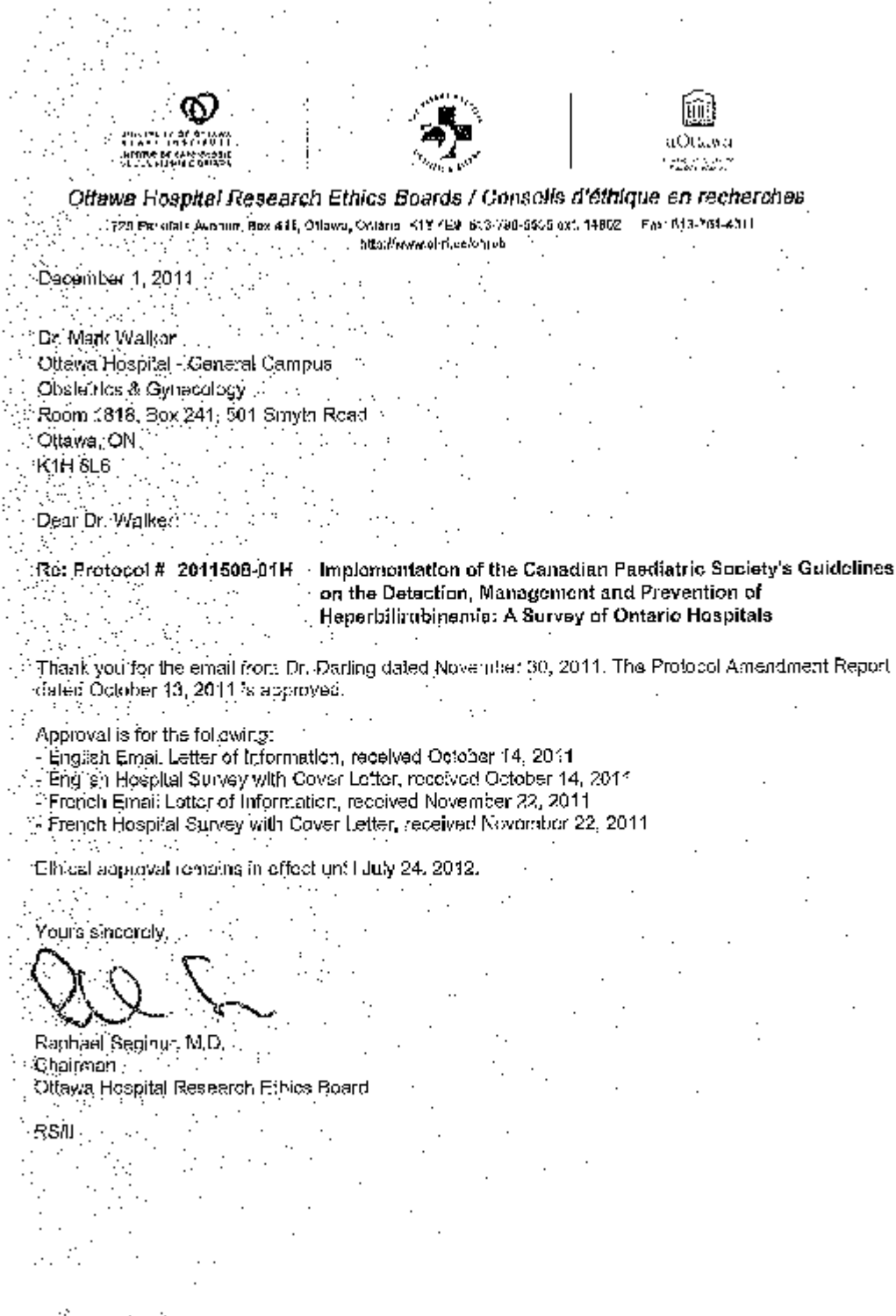


Figure A2.2. Original letter of approval for project 1 from University of Ottawa Research Ethics Board

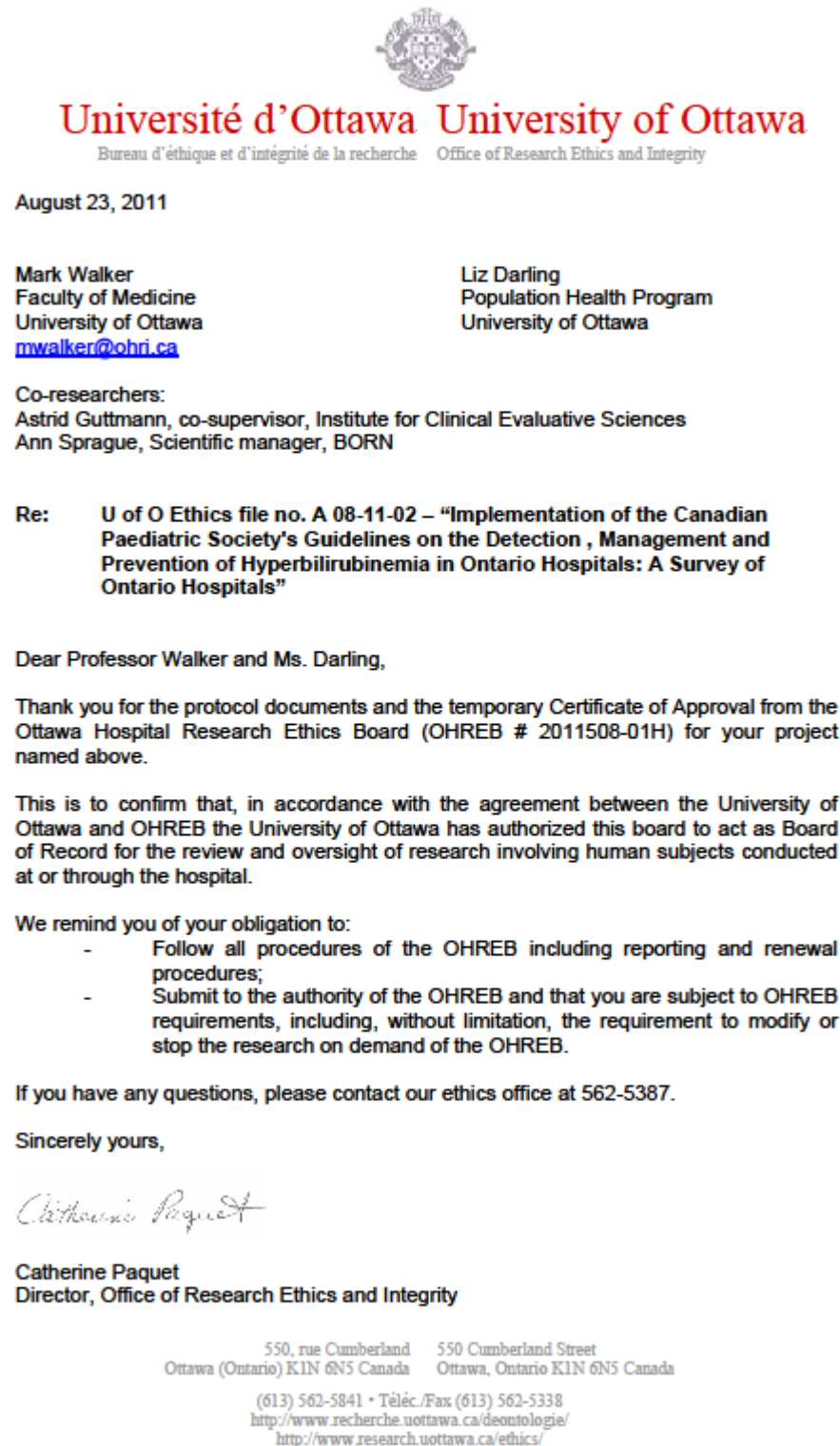


Figure A2.3. Original letter of approval for projects 2 & 3 from Ottawa Hospital Research Ethics Board

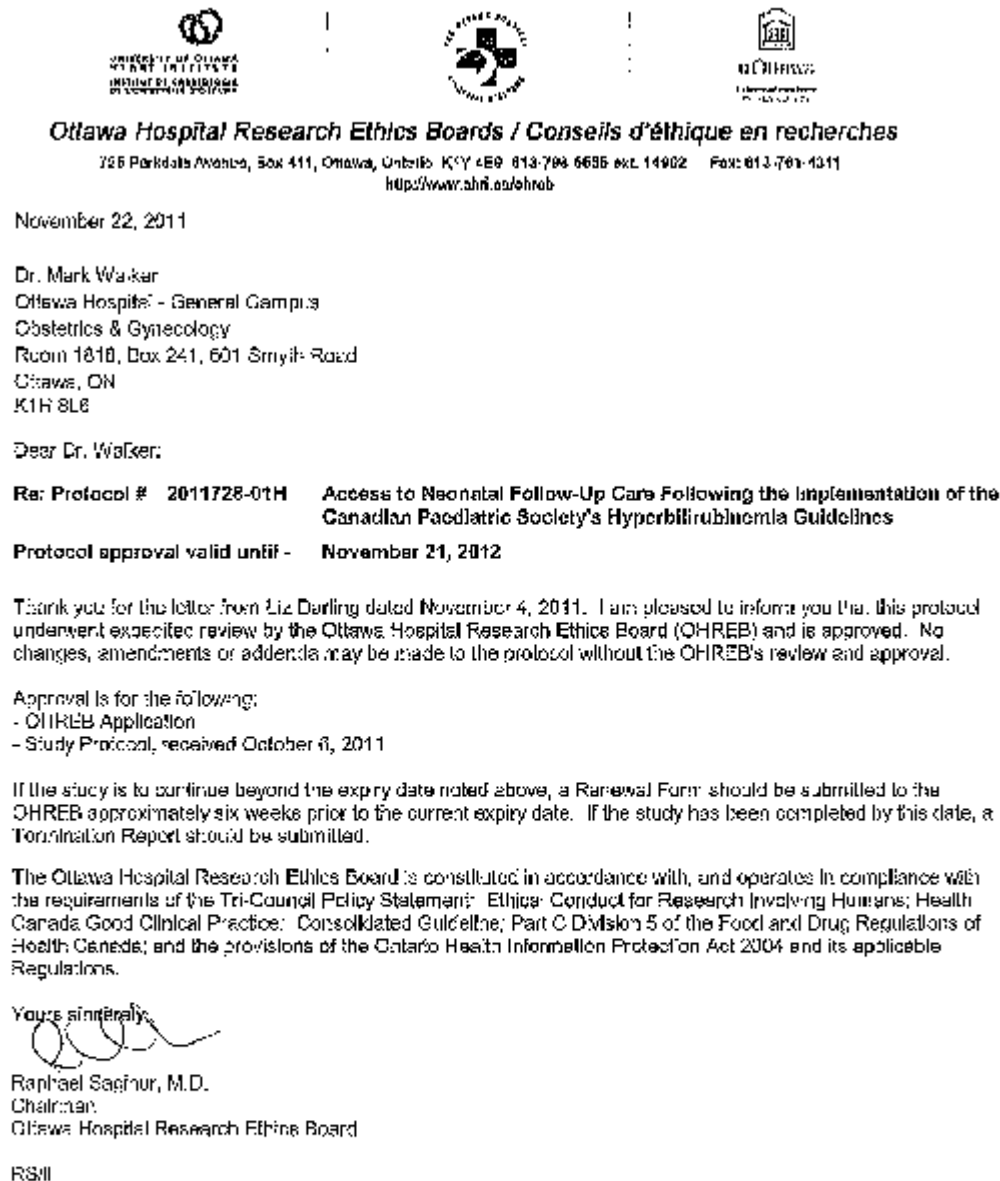
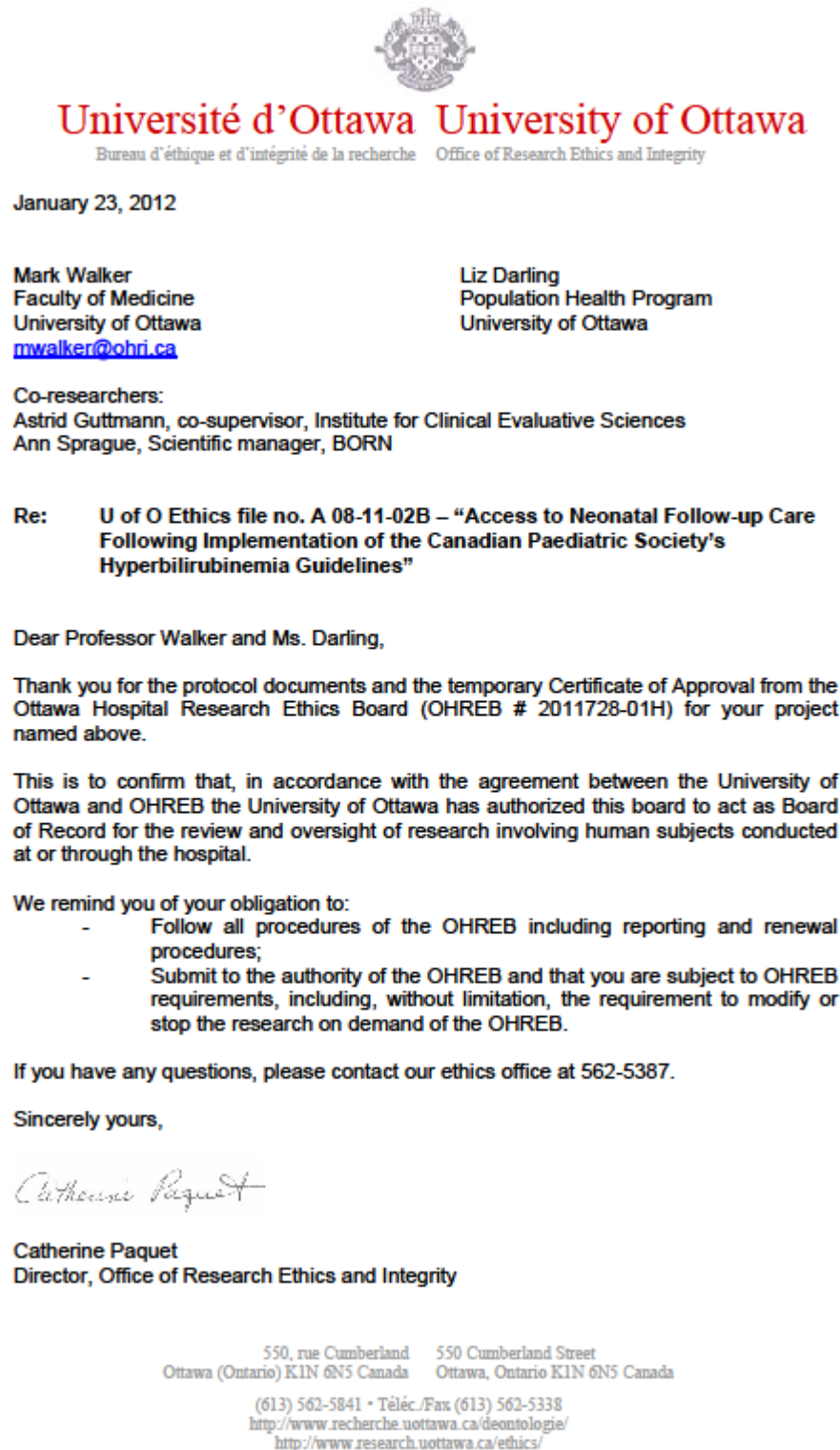


Figure A2.4. Original letter of approval for projects 2 &3 from University of Ottawa Research Ethics Board



Appendix 3. Questions Used in Pilot Survey to Gather Feedback

1. How long did it take you to complete the survey?
2. Did you want to stop answering the survey at any point?
3. Did you find any of the questions difficult to answer or inappropriately worded?
4. Would you be likely to complete this survey if you received an email asking you to complete it?

Appendix 4. Hospital Survey

1. Please select the hospital you represent: [drop-down menu]
2. Does your hospital routinely conduct bilirubin screening for all babies born at ≥ 35 weeks gestation?
 - ☐ Yes
 - ☐ No

[If respondent answers "No" to Question 2, survey skips to Question 22]
3. When did your hospital start to conduct bilirubin screening for all babies born at ≥ 35 weeks gestation?
Month [drop-down menu] Year [drop-down menu]
4. Is your hospital's approach to bilirubin screening based on the Canadian Paediatric Society's (CPS's) Guidelines for the detection, management and prevention of hyperbilirubinemia in term and late preterm newborn infants (35 or more weeks' gestation)?
 - ☐ No
 - ☐ Yes with modifications
 - ☐ Yes
5. What methods of bilirubin measurement do you use for screening? (Select all that apply)
 - ☐ Total serum bilirubin (TSB)
 - ☐ Transcutaneous bilirubin (TcB)
6. Are bilirubin screening results routinely communicated to the baby's primary care provider in the community?
 - ☐ No
 - ☐ Only if results are elevated
 - ☐ Yes
7. How are results communicated to primary care providers? (Select all that apply)
 - ☐ Written document given to parents
 - ☐ Fax to primary care provider
 - ☐ Email to primary care provider
 - ☐ Direct verbal communication with primary care provider
 - ☐ Other (please specify [free text box below])
8. When bilirubin screening results prior to hospital discharge indicate that a baby should be retested within 24 to 48 hours, where are parents directed to take the baby for recommended follow-up? (Select all that apply)
 - ☐ Outpatient clinic at your hospital
 - ☐ Neonatal nursery at your hospital
 - ☐ Mother baby care unit at your hospital
 - ☐ Lab at your hospital
 - ☐ Lab in the community

- Primary care provider in the community
 - Local Children's Hospital
 - Another hospital in the region
 - Community based paediatric clinic
 - Community based family medicine clinic
 - Community Health Centre
 - Other (please specify [free text box below])
9. Has your hospital implemented any doublechecking or verification strategies to ensure that parents take their baby for follow-up within the recommended time frame?
- No
 - Yes (please describe [free text box below])
10. Does your hospital rely on any programs or other supports available in your community to help ensure appropriate follow-up regarding hyperbilirubinemia for babies at high risk of experiencing barriers in access to care (e.g. parents with language or transportation barriers)?
- No
 - Yes (please describe [free text box below])
11. If a baby born at your hospital requires phototherapy prior to initial discharge from hospital, would phototherapy treatment normally be provided at your hospital?
- No
 - Yes
12. If a baby born at your hospital requires inpatient phototherapy after their initial discharge, where is the baby admitted? (Select all that apply)
- Neonatal nursery at your hospital
 - Paediatric unit at your hospital
 - Normal newborn/postpartum unit at your hospital
 - Nearest children's hospital
 - Another hospital in the region
13. Is outpatient phototherapy available in your community?
- No
 - Yes (please specify who provides this service [free text box below])
14. Please identify all the factors that contributed to the implementation of universal bilirubin screening at your hospital. (Select all that apply)
- Release of the CPS guidelines
 - Standard practices of other hospitals in the region
 - Activities of regional perinatal program
 - Leadership from Maternal newborn program leaders
 - Leadership from an interdisciplinary perinatal committee
 - Leadership from nurse educator
 - Leadership from physicians providing paediatric care
 - Risk management considerations

- Medicolegal considerations
 - Case(s) of severe hyperbilirubinemia at your institution/in your community
 - None of the above
 - Other (please specify [free text box below])
15. During the process of implementing universal bilirubin screening, did your hospital engage in dialogue about the optimal provision or organization of newborn health services following hospital discharge with any of the following parties? (Select all that apply)
- Other hospitals in your region
 - Local community based primary care providers
 - Local public health
 - Regional perinatal partnerships
16. Has there been any regional coordination related to the implementation of universal bilirubin screening in your region?
- No
 - Yes
17. Has your hospital or region developed any new processes or services related to newborn follow-up in order to implement the guidelines (e.g., established outpatient access to laboratory services, setup an outpatient clinic for follow-up assessments, changed procedure for readmission for phototherapy, implemented point of care testing with bilimeters)?
- No
 - Yes (please describe [free text box below])
18. What challenges has your hospital encountered in implementing universal bilirubin screening? (Select all that apply)
- Arranging post-discharge follow-up on weekends and holidays
 - Arranging post-discharge follow-up for babies who live far from hospital
 - Arranging access to TSB or TcB testing in the community
 - Resistance from care providers to screen all babies
 - Over-testing
 - Ensuring appropriate communication of results
 - Delays in newborn discharge from hospital
 - Cost
 - Other (please describe [free text box below])
19. What strategies/solutions have been helpful in addressing these challenges? [free text box below]
20. Has your hospital had any successes related to the implementation of universal bilirubin screening that you would like to share? [free text box below]
21. Please provide any other comments you wish to make about your hospital's experience implementing universal bilirubin screening. [free text box below]
- [Survey ends here for respondents who report having implemented universal bilirubin screening]

22. If a baby born at your hospital requires phototherapy prior to initial discharge from hospital, would phototherapy treatment normally be provided at your hospital?
- ☐ Yes
 - ☐ No
23. When a baby born at your institution develops hyperbilirubinemia requiring therapy, is your policy for treatment based on the CPS's Guidelines for the detection, management and prevention of hyperbilirubinemia in term and late preterm newborn infants (35 or more weeks' gestation)?
- ☐ Yes
 - ☐ Yes with modifications
 - ☐ No
 - ☐ Hospital does not have a policy
24. If a baby born at your hospital requires inpatient phototherapy after their initial discharge, where is the baby admitted? (Select all that apply)
- ☐ Neonatal nursery at your hospital
 - ☐ Paediatric unit at your hospital
 - ☐ Normal newborn/postpartum unit at your hospital
 - ☐ Nearest children's hospital
 - ☐ Another hospital in the region
25. Is outpatient phototherapy available in your community?
- ☐ No
 - ☐ Yes (please specify who provides this service [free text box below])
26. What factors have created barriers to your hospital implementing universal bilirubin screening? (Select all that apply)
- ☐ Human resources to coordinate the implementation of clinical protocols
 - ☐ Resistance from careproviders to screen all babies
 - ☐ Difficulties arranging postdischarge followup on weekends and holidays
 - ☐ Difficulties arranging access to TSB or TcB testing in the community
 - ☐ Patient population living a significant distance from hospital
 - ☐ Perception of medicolegal risk for inhospital physicians
 - ☐ Cost
 - ☐ Other (please describe [free text box below])
27. Please provide any other comments you wish to make about your hospital's experience related to universal bilirubin screening. [free text box below]

THANK YOU FOR COMPLETING THIS SURVEY.

Appendix 5. Modelling Strategy for Cohort Studies

We used slightly different modelling strategies for the two cohort studies (projects 2 and 3). In both cases we began by developing an a priori list of variables for inclusion in the model. One advantage of a priori variable identification is that encourages consideration of the relationships between variables at a theoretical level and leads to selection of variables that have clinically plausible relationships with the outcome of interest based on previous research or the general expertise of the research team. A priori specification of variables also prevents data-driven modelling. For our third project (on recommended follow-up) we simply used a priori selection of the covariates because we were quite confident that each of the covariates should be included in our final model.

For our second project (on health service utilization) we chose to use backward selection to reduce the models for each of the outcomes to the simplest model reasonable. While each of the covariates we had specified a priori had a theoretical rationale that we believed to be plausible based on our clinical expertise and our understanding of the context of our research, some of the variables we identified had not been previously tested in the research literature. We also were cognisant of the potential for confounding between some of the covariates we had identified. Furthermore, we were primarily interested in obtaining accurate estimates for our primary outcome as opposed to creating an optimal predictive model. For these reasons we determined that the most appropriate approach to modelling would be to include all of the covariates in the model and then selectively remove one at a time (i.e., backward selection). We used a p value of >0.10 to identify variables for removal, and removed the variable with the largest p value first. At each step in the process we reviewed the impact of removing covariates from the model and found that in all cases removal of non-significant variables had negligible to no impact on the estimates for the primary outcome.

We faced one additional challenge that prevented us from using a pure backward selection approach for project 2. For all but one outcome, we hypothesized SES to be a covariate. We had three SES variables available in the data set: material deprivation quintile, social deprivation quintile, and income quintile. As more complex constructs, we anticipated that the deprivation variables would be more sensitive covariates than the income variable but had no evidence from previous analyses to guide our hypotheses with respect to which of the three SES constructs would have the strongest association with each outcome. Consequently, we determined that the best approach would be to test each SES construct and use the objective measure of QIC to determine which model had the best fit. [The QIC, a.k.a. Quasi-AIC (Akaike Information Criterion) is a statistical test used for model selection when using generalized estimating equations.] Collinearity of the three SES variables prevented us from including more than one of them in the model at a time, so we kept these variables out of the model until we had eliminated other covariates using backward selection and then tested each SES covariate one at a time. Table A4.1 presents the covariates identified a priori for each of the outcomes of interest in project 2, and indicates whether they were included or excluded from the final model.

Table A5.1 Covariates including in modelling in project 2 (health service utilization)

Outcome Covariate	LOS	Phototherapy at Birth	Phototherapy after Discharge	Jaundice- Related ED Visit	Jaundice- Related Readmission
Hospital	•	•	•	•	•
Quarter	•	•	•	•	•
Gestational age category	•	•	•	•	•
Maternal parity	•	•	•	•	•
Mode of birth	•	•	•	•	•
Hospital level	X	X	X	X	•
Midwifery care	•	•	•		•
Prenatal care from GP				X	•
Rural	X	X	X	X	
Age category at discharge			X	•	X
Phototherapy at birth					X
Material deprivation quintile	x		•	x	•
Social deprivation quintile	•		x	•	x
Income quintile	x		x	x	x

• = included in the model

X= identified a priori but excluded during backward selection

x = excluded based on QIC

The following two tables present descriptions of the covariates for the two cohort studies, including identification of the categories for each variable and the data source(s) used to create the variable.

Table A5.2 Description of covariates used in project 2 (health service utilization)

Variable	Categories	Data source
Gestational age category	35-38 weeks 39+ weeks (reference)	Baby's record in DAD (or maternal record in DAD if missing from baby's record)
Maternal parity	Primiparous (reference) Multiparous	Mother's record in DAD, calculated based on sum of previous preterm deliveries and previous term deliveries

Table A5.2 Description of covariates used in project 2 (health service utilization) cont'd.

Variable	Categories	Data source
Birth mode	Spontaneous Vaginal (reference) Vacuum or Forceps Cesarean	Mother's record in DAD, determined using intervention codes
Hospital Level	Level 1 Level 2 Level 3 (reference)	Provincial Council for Maternal Child Health designations for Criti-Call regarding the level of neonatal services provided
Midwifery care	Midwife No Midwife	Mother's OHIP record (to identify midwife-requested consultations) and Mother's record in DAD (to identify service provider codes)
Prenatal care from GP	Mother had at least one prenatal visit billed by GP Mother had no prenatal visit billed by GP	Mother's OHIP record. Determined using a nine-month look-back for prenatal fee codes and the specialty codes associated with these.
Rural	Rural Urban (reference)	Mother's record in DAD used to identify postal code, variable calculated using the Rurality Index of Ontario 2008 score, with the two available rural categories collapsed into single rural category
Age category at discharge	1 (≤ 24 h) 2 (>24 -48 h) 3 (>48 -72 h) (reference)	Baby's record in DAD, categorized based on length of time between birth datetime variable and discharge datetime variable
Phototherapy at birth	Yes No (reference)	Baby's record in DAD, identified records of hospitalization at birth with Canadian Classification of Intervention procedure code for phototherapy

Table A5.3 Description of covariates used in project 3 (recommended follow-up)

Variable	Categories	Data source
Gestational age category	35-38 weeks 39+ weeks (reference)	Baby's record in DAD (or maternal record in DAD if missing from baby's record)
Maternal parity	Primiparous (reference) Multiparous	Mother's record in DAD, calculated based on sum of previous preterm deliveries and previous term deliveries
Birth mode	Spontaneous Vaginal (reference) Vacuum or Forceps Cesarean	Mother's record in DAD, determined using intervention codes
Day of Discharge	1 (≤ 24 h) 2 (>24 -48 h) 3 (>48 -72 h) (reference)	Baby's record in DAD, categorized based on length of time between birth datetime variable and discharge datetime variable
Timing of recommended Follow-Up	Holiday or weekend Not a holiday or weekend (reference)	Baby's record in DAD, calculated based on LOS and whether the day that follow-up was recommended fell on a Saturday or Sunday, or one of the following statutory holidays: Boxing Day, Christmas Day, New Year's Day, Canada Day, Labour Day, Thanksgiving, Victoria Day, Good Friday, Civic Holiday
Maternal prenatal care provider	Prenatal Care - GP (reference) Prenatal Care – OB & other No Prenatal Care	Determined using a nine-month look-back in the OHIP database for prenatal fee codes and the specialty codes associated with these. The GP group includes women with at least one prenatal visit with a GP/family physician, the no prenatal care group includes women with no OHIP billings for prenatal visits, all other women with OHIP billings are in the OB & other group.

Table A5.3 Description of covariates used in project 3 (recommended follow-up) cont'd.

Variable	Categories	Data source
Maternal age at first birth	19+ (reference) <19	Maternal record in a dataset of linked maternal and newborn records from DAD (MOMBABY), looking back to the first delivery for each mother and categorizing based on maternal age.
Maternal residence	Rural Urban (reference)	Mother's record in DAD used to identify postal code, variable calculated using the Rurality Index of Ontario 2008 score, with the two available rural categories collapsed into single rural category

**Appendix 6. Comparison of Included vs. Excluded Babies in Analysis of
Universal Bilirubin Screening and Healthcare Utilization**

Table A6.1 Comparison of Included vs. Excluded Babies in Analysis of Universal Bilirubin Screening and Healthcare Utilization

		Total		Excluded		Included	
		N	%age	N	%age	N	%age
Overall		972718	100.0	438615	45.1	534103	54.9
Mode of birth							
	Missing	3811	0.4	765	0.2	3046	0.6
	Vaginal - Spontaneous	602117	61.9	278094	63.4	324023	60.7
	Vaginal - Vacuum/Forceps	105543	10.9	43878	10.0	61665	11.5
	Caesarean Section	261247	26.9	115878	26.4	145369	27.2
Parity							
	Unknown	24	0.0	18	0.0	6	0.0
	Multiparous	544704	56.0	250144	57.0	294560	55.2
	Primiparous	427990	44.0	188453	43.0	239537	44.8
Age at Discharge (in hours)							
	<24 Hours	53587	5.5	28507	6.5	25080	4.7
	24-72 Hours	737433	75.8	331780	75.6	405653	76.0
	>72 Hours	181698	18.7	78328	17.9	103370	19.4
Gestational Age in Weeks							
	35-38 weeks gestation	297106	30.5	132855	30.3	164251	30.8
	39+ weeks gestation	675612	69.5	305760	69.7	369852	69.2
Mother had at least one prenatal visit billed by GP							
	No	415034	42.7	180266	41.1	234768	44.0
	Yes	557684	57.3	258349	58.9	299335	56.0
Mother had prenatal care from midwife							
	No	909035	93.5	409108	93.3	499927	93.6
	Yes	63683	6.5	29507	6.7	34176	6.4

Table A6.1 Comparison of Included vs. Excluded Babies in Analysis of Universal Bilirubin Screening on Healthcare Utilization (cont'd.)

		Total		Excluded		Included	
		N	%age	N	%age	N	%age
Level of Newborn Services							
	Level 1	106935	11.0	64477	14.7	42458	7.9
	Level 2	704739	72.5	330370	75.3	374369	70.1
	Level 3	161044	16.6	43768	10.0	117276	22.0
Geographical Area (2008 RIO score)							
	Unknown	772	0.1	381	0.1	391	0.1
	Rural	98805	10.2	57690	13.2	41115	7.7
	Urban	873141	89.8	380544	86.8	492597	92.2
Material Deprivation Quintile							
	Unknown	25349	2.6	11943	2.7	13406	2.5
	1	176235	18.1	65532	14.9	110703	20.7
	2	197333	20.3	79030	18.0	118303	22.1
	3	198774	20.4	90198	20.6	108576	20.3
	4	203247	20.9	100308	22.9	102939	19.3
	5	171780	17.7	91604	20.9	80176	15.0
Social Deprivation Quintile							
	Unknown	25349	2.6	11943	2.7	13406	2.5
	1	236427	24.3	109230	24.9	127197	23.8
	2	190723	19.6	86962	19.8	103761	19.4
	3	174735	18.0	79875	18.2	94860	17.8
	4	176294	18.1	76777	17.5	99517	18.6
	5	169190	17.4	73828	16.8	95362	17.9

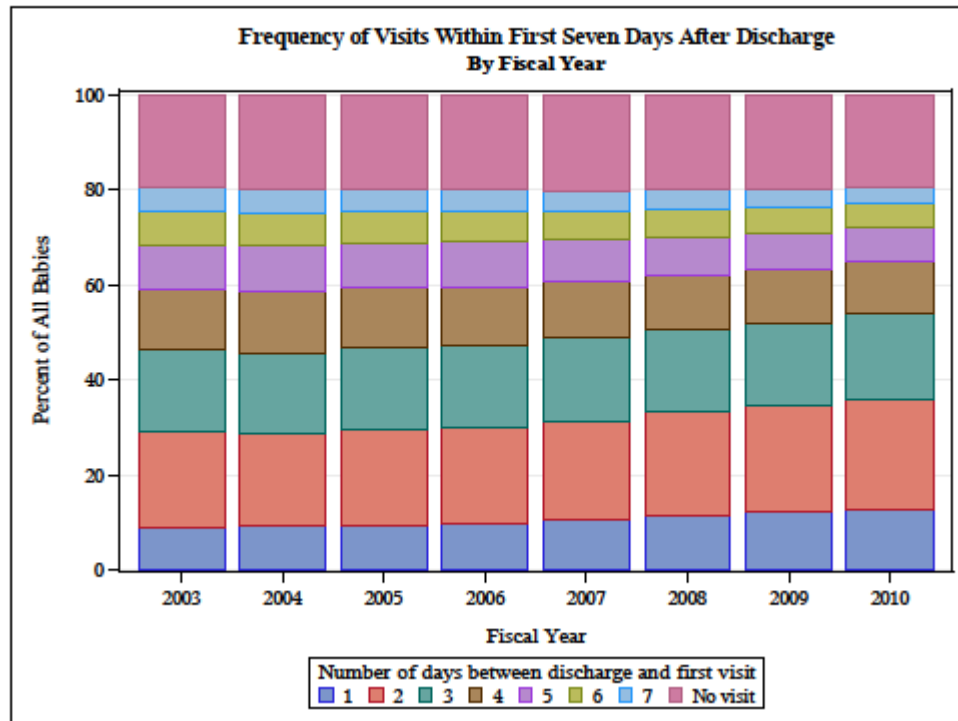
Appendix 7. Fee codes and diagnostic codes used to define follow-up visits

Table A7.1. Codes used to define follow-up visits

OHIP billings	• Excludes billings with a location code indicating inpatient services or phone-based services • Restricted to billings made by the following specialties: family medicine, community medicine, paediatrics, or nurse practitioner • Included fee codes: A001, A003, A004, A007, A225, A260, A261, A262, A263, A264, A265, A266, A565, A661, A662, A813, A815, and A901
NACRS visits	• Restricted to scheduled visits • Included diagnostic codes: P599 (jaundice), Z001 (routine child health exam), or A762 (health supervision & care of a healthy infant or child)

Appendix 8. Frequency of Visits Within First Seven Days after Discharge by Fiscal Year

Figure A8.1. Frequency of Visits Within First Seven Days after Discharge by Fiscal Year



Appendix 9. Calculation of unadjusted attributable proportion for each material deprivation quintile of the observed increase in recommended follow-up associated with guideline implementation

To estimate the proportion of the overall increase in the rate of recommended follow-up associated with implementation of the guidelines that was attributable to each material deprivation quintile, we followed the following process:

1. Determined the total number of babies within each quintile born following guideline implementation.
2. Calculated the expected number of babies to receive recommended follow-up within each quintile by multiplying the number of babies within the quintile by the quintile-specific rate of recommended follow-up prior to guideline implementation.
3. Calculated the difference between the expected number of babies to receive recommended follow-up and the observed number for each quintile.
4. Calculated the attributable portion of babies who received recommended follow-up within quintile by dividing the difference between expected and observed for each quintile by the total number of excess number of babies who received recommended follow-up (8359).

Details for these calculations are shown in the table below.

Table A9.1. Calculation of unadjusted attributable portion for each material deprivation quintile of the observed increase in recommended follow-up associated with guideline implementation

Material Deprivation Quintile	A. Number born following guideline implementation	B. Rate of recommended follow-up pre-implementation	C. Expected n with recommended follow-up (AxB)	D. Observed n with recommended follow-up	E. Difference between expected and observed (D-C)	Attributable proportion of increase (E/8359)
Q5 (Lowest)	24612	29.7	7310	7344	34	0%
Q4	32417	30.3	9822	10327	505	6%
Q3	35076	28.1	9856	11742	1886	23%
Q2	38572	29.2	11263	13885	2622	31%
Q1 (Highest)	34652	33.1	11470	14782	3312	40%
Total					8359	

Appendix 10. Post-hoc analyses examining provider type, SES, and day of follow-up

Based on research literature specific to Ontario, we identified one possible factor that might account for some of the socio-economic disparity that we observed in rates of recommended follow-up. Paediatricians are more likely than family physicians both to be aware of the CPS hyperbilirubinemia guidelines and to report routinely seeing newborns within 72 hours of discharge from hospital.(96) We hypothesized that babies of lower SES might be less likely to be seen by a paediatrician, which could account for some of the increase in disparity following guideline implementation. To explore this post hoc hypothesis we examined two basic descriptive analyses shown below: the proportion of provider type for first visits by material deprivation quintile, and the proportion of provider type for first visit by day of first visit. While babies in the highest SES quintile were most likely to be seen by a paediatrician, the relationship between provider type and SES was U-shaped rather than linear, suggesting that there are probably additional factors contributing to the observed disparity. It is also important to keep in mind that these are simple bivariate analyses that do not adjust for other covariates which might account for some of the observed differences between groups.

Table A10.1. Proportion of provider type for first visits by material deprivation quintile

	Q1 (High SES)	Q2	Q3	Q4	Q5 (Low SES)
Family Physician	63.7	72.8	74.0	70.8	69.0
Paediatrician	36.2	27.2	26.0	29.2	31.0

Table A10.2. Proportion of provider type for first visit by day of first visit

	Day 1	Day2	Day 3	Day 4	Day 5	Day 6	Day 7	Total
Family Physician	66.2	66.9	65.4	73.6	77.7	79.2	79.3	70.3
Paediatrician	33.8	33.1	34.6	26.4	22.3	20.8	20.7	29.71

Appendix 11. Measuring changes in disparity

We considered several methods of examining whether there was a change in disparity over time, including the Slope Index of Inequality, the Relative Index of Inequality, Between-Group Variance, and use of a product (or interaction) term in the regression model (guideline implementation x material deprivation). This appendix provides a brief description of these alternative approaches and presents the results of an analysis of Between-Group Variance.

Slope Index of Inequality and Relative Index of Inequality

We considered using two specific regression-based measures for examining patterns of disparity: the Slope Index of Inequality (SII) and the Relative Index of Inequality (RII). The SII and RII measure disparity by calculating the slope of the relationship between a health outcome and ranked socioeconomic groups.(110) Both measures take into account the size of the social groups within the population (i.e., they are weighted), and can be interpreted as the difference in the outcome of interest between the lowest and the highest socioeconomic groups.(110) The SII is an absolute measure of this difference between lowest and highest groups, and the RII is a relative measure which presents the absolute difference as a proportion of the average rate of the outcome of interest in the population.(110) The basic equation used to derive the SII and the RII is $Y = \beta_0 + \beta_1 (\text{SEP midpoint}) + \varepsilon$, where Y = the health outcome, and the socioeconomic position (SEP) midpoint = the midpoint of the range for the cumulative percentage of the population accounted for by each group.(110) From this basic equation, the SII and RII are calculated as follows:

- $\text{SII} = -\beta_1$
- $\text{RII} = (-\beta_1) / y$, where y = average outcome for the population(110)

One limitation of this approach to measuring disparity is that it assumes a linear relationship between the outcome and the ordered socioeconomic groups.(110) We determined that it would not be appropriate to use this measure because the relationship between material deprivation and recommended follow-up did not appear to be linear.

Between-Group Variance

Another measure of disparity that we considered using is Between-Group Variance (BGV). This measure is also weighted by group size. The basic equation use to calculate BGV is:

$$\sum_{j=1}^J p_j (y_j - \mu)^2, \text{ where:}$$

p_j is the group's share of the population,

y_j is the rate of the outcome of interest in group j , and

μ is the population average(110)

Inclusion of a squared term in this equation results in increased sensitivity to deviations which are further from the population average.(110) BGV summarizes the variance with respect to a health outcome across all social groups, but does not describe the changes within specific groups.(110) This measure can be used to address basic questions pertaining to whether differences between groups have increased or decreased over time.(110) Below we show the calculations of BGV in the relationship between material deprivation and recommended follow-up for 2003 and 2010 (the first and last years in our study).

Table A11.1. Calculation of BGV (Material Deprivation and Recommended Follow-Up) for 2003

Material Deprivation Quintile	Group Size	n with Recommended Follow-Up	y_j	p_j	$(y_j - \mu)^2$	$p_j (y_j - \mu)^2$
1	15680	4845	30.90	0.181820	5.885661	1.070133
2	17055	4769	27.96	0.197764	0.260837	0.051584
3	18055	5039	27.91	0.209360	0.318130	0.066604
4	19000	5391	28.37	0.220318	0.009903	0.002182
5	16449	4511	27.42	0.190737	1.100479	0.209902

$$\mu_{2003} = 24555/86239 = 28.74$$

$$BGV_{2003} = \sum p_j (y_j - \mu)^2 = 1.40$$

Table A11.2. Calculation of BGV (Material Deprivation and Recommended Follow-Up) for 2010

Material Deprivation Quintile	Group Size	n with Recommended Follow-Up	y_j	p_j	$(y_j - \mu)^2$	$p_j (y_j - \mu)^2$
1	15038	6347	42.21	0.178017	40.982204	7.295536
2	17869	6486	36.30	0.211530	0.242874	0.051375
3	17779	6020	33.86	0.210465	3.781095	0.795787
4	18127	6227	34.35	0.214584	2.110060	0.452785
5	15662	5166	32.98	0.185404	7.954559	1.474807

$$\mu_{2010} = 30246/84475 = 35.80$$

$$BGV_{2010} = \sum p_j (y_j - \mu)^2 = 10.07$$

The calculations of BGV for the relationship between material deprivation and recommended follow-up show that BGV increased from 1.40 in 2003 to 10.07 in 2010, indicating that disparity increased over this time period.

Use of a Product (Interaction) Term

The final option that we considered, and ultimately the one that we chose to use, was the use of a product term to account for differential association between guideline implementation and recommended follow-up depending on material deprivation quintile. This approach offered several advantages over the previous measures of disparity: it allowed us to specifically examine the question of whether guideline implementation was associated with a change in disparity, it allowed us to adjust for other important covariates when examining disparity in the relationship between material deprivation and recommended follow-up, and it built on the modelling approach that we were using to examine the relationship between guideline implementation and follow-up rather than introducing a completely new measure. The methods and results of this approach are presented in Chapter 4.

Appendix 12. Comparison of SES measures and disparity in recommended follow-up

This appendix presents graphical analyses that we conducted comparing the association between recommended follow-up and material deprivation quintile with the associations between recommended follow-up and two additional SES variables (income quintile and social deprivation quintile). All three of these SES measures are neighbourhood level measures created using individual postal codes and census data at the level of the dissemination area (the smallest Canadian census geographical area). Income quintile is based on household income. The two deprivation measures are composite measures that are calculated using multiple components. The material deprivation index is intended to measure relative disadvantage in terms of access to goods and resources, whereas the social deprivation index is intended to measure relative disadvantage with respect to social relationships (combining concepts of social capital, social fragmentation and social isolation).(78) The two measures were constructed using the same six indicators, but the weighting of these indicators differed between the two.(78) Material deprivation is influenced most strongly by the proportion of people aged 15 years and older without a high school diploma, the employment/population ratio of people aged 15 years and older, and the average income of people aged 15 years and older.(78) Social deprivation is influenced more by the proportion of individuals aged 15 years and older living alone, the proportion of single-parent families, and the proportion of individuals aged 15 years and older who are separated, divorced or widowed.(78)

When planning the analysis for project 3, we selected material deprivation as our SES measure because it measures the combined effect of education, employment and income, and based on the existing literature related to access to care we felt that this measure would be most strongly associated with newborn access to recommended follow-up care (see section 1.2.5 in Chapter 1 for discussion of access to care). However we were also interested in exploring the differences between the three measures that we had available. As a supplementary analysis, we ran time trend graphs demonstrating the association between recommended follow-up and the two other measures: income quintile and social deprivation quintile (See Figures A11.1 and A11.2.; the graph for material deprivation is Figure 4.4 in Chapter 4).

While all three SES measures illustrate some form of gradient with recommended follow-up, there are interesting differences between the three measures. In direct contrast to material deprivation, where the highest SES quintile was notably higher than the four lower quintiles, the graph by income quintile shows that the lowest income quintile was notably lower than the four higher quintiles. The graph for social deprivation appears to be somewhat in between these two patterns with the middle three quintiles clustered close together and the top and bottom quintiles notably higher and lower respectively. One other difference between the three graphs is that the spread from lowest to highest quintile increases notably over time for material deprivation but does not increase obviously for income quintile or social deprivation quintile.

The differences we observed confirm that the three SES measures are in fact measuring quite different constructs. The notable relative disadvantage with respect to recommended follow-up that was observed for the lowest income quintile is consistent with research which suggests that barriers such as lack of (affordable) transport may limit access to health care, and suggests that such barriers may be much more of an issue for the lowest income quintile than for others. We hypothesize that the relative advantage with respect to recommended follow-up that was observed for the highest material deprivation quintile may reflect the education component of this measure; the highest levels of maternal education might be associated with higher levels of recommended follow-up for several reasons, including maternal awareness of the importance of follow-up, and maternal ability to navigate the health care system and access care in a timely manner. Additional research would be required to confirm the causal pathways or associations that explain the different patterns of association between recommended follow-up and each of the three SES measures. The differences that we observed between these measures suggest that further research examining the associations between various neonatal outcomes and each of the measures may help to improve our understanding of the associations between SES and health outcomes and to guide appropriate selection of SES measures for future research.

Figure A12.1. Frequency of Recommended Follow-up by Income Quintile

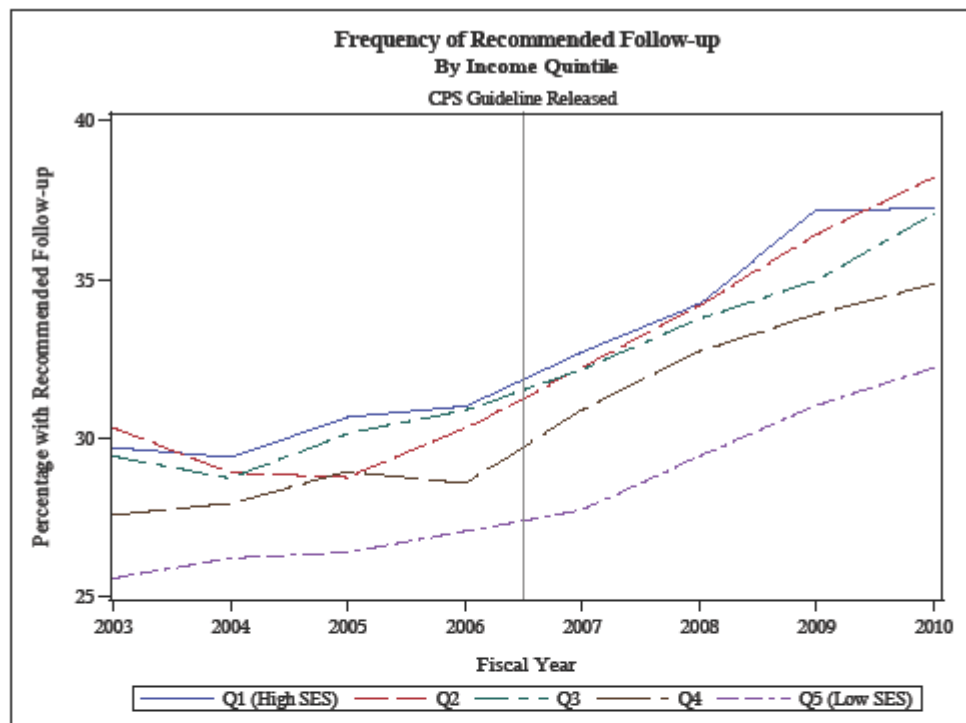
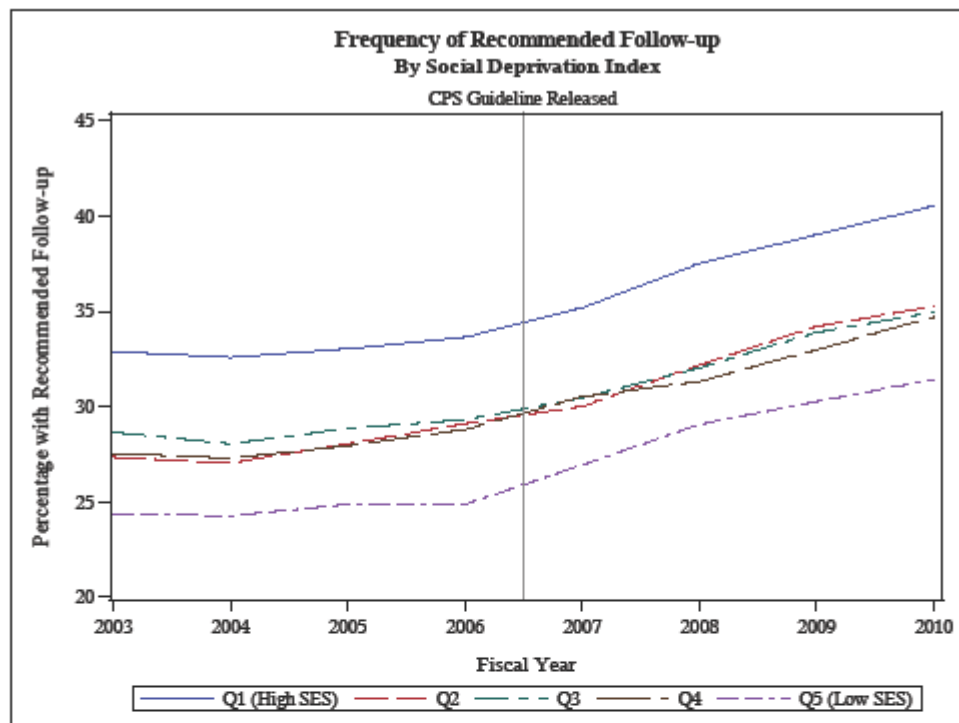


Figure A12.2. Frequency of Recommended Follow-up by Social Deprivation Quintile



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My article - Implementation of the Canadian Paediatric Society's hyperbilirubinemia guidelines: a survey of Ontario hospitals - was published in Paediatrics & Child Health earlier this year. The research reported in the article is part of my doctoral research at the University of Ottawa. I am currently in the process of finalizing my dissertation for submission and wish to include the manuscript of the article as a chapter in my dissertation. The chapter acknowledges where the article was published and provides a complete bibliographic reference. I wish to request permission to include the manuscript in the dissertation, and am required by the University of Ottawa to include a copy of the written permission from the publisher as part of the dissertation itself.

Thank-you,
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Appendix 14. Recommendations from the Canadian Paediatric Society's Guidelines for detection, management and prevention of hyperbilirubinemia in term and late preterm newborn infants (35 weeks or more gestation)

- All mothers should be tested for ABO and Rh(D) blood types and be screened for red cell antibodies during pregnancy (recommendation grade D).
- If the mother was not tested, cord blood from the infant should be sent for evaluation of the blood group and a DAT (Coombs test) (recommendation grade D).
- Blood group evaluation and a DAT should be performed in infants with early jaundice of mothers of blood group O (recommendation grade B).
- Selected at-risk infants (Mediterranean, Middle Eastern, African or Southeast Asian origin) should be screened for G6PD deficiency (recommendation grade D).
- A test for G6PD deficiency should be considered in all infants with severe hyperbilirubinemia (recommendation grade D).
- Either TSB or TcB concentration should be measured in all infants during the first 72 h of life. If not required earlier because of clinical jaundice, a TSB measurement should be obtained at the same time as the metabolic screening test; alternatively, a TcB measurement should be obtained either at discharge or, if not yet discharged, at 72 h of life (recommendation grade C).
- If the TSB concentration does not require immediate intervention, the results should be plotted on the predictive nomogram. The result of the TSB measurement, the time at which it was obtained and the zone should be recorded, and a copy should be given to the parents. Follow-up of the infant should be individualized according to the risk assessment (recommendation grade D).
- Any infant discharged before 24 h of life should be reviewed within 24 h by an individual with experience in the care of the newborn who has access to testing and treatment facilities (recommendation grade D).
- There should be a systematic approach to the risk assessment of all infants before discharge and institution of follow-up care if the infant develops jaundice (recommendation grade D).
- All newborns who are visibly jaundiced in the first 24 h of life should have their bilirubin level determined (recommendation grade D).
- Transcutaneous bilirubinometry is an acceptable method, either as a routine procedure or in infants with visible jaundice. The result should be summed with the 95% CI of the device to estimate the maximum probable TSB concentration (recommendation grade C).
- TSB concentration may be estimated on either a capillary or a venous blood sample (recommendation grade C).
- Infants with severe or prolonged hyperbilirubinemia should be further investigated, including measurement of the conjugated component of bilirubin (recommendation grade C).
- A program for breastfeeding support should be instituted in every facility where babies are delivered (recommendation grade D).

- Routine supplementation of breastfed infants with water or dextrose water is not recommended (recommendation grade B).
- Infants with a positive DAT who have predicted severe disease based on antenatal investigation or an elevated risk of progressing to exchange transfusion based on the postnatal progression of TSB concentration should receive IVIG at a dose of 1 g/kg (recommendation grade A).
- A TSB concentration consistent with increased risk (figure 1)¹ and (table 4)¹ should lead to enhanced surveillance for development of severe hyperbilirubinemia, with follow-up within 24 h to 48 h, either in hospital or in the community, and repeat estimation of TSB or TcB concentration in most circumstances (recommendation grade C).
- Intensive phototherapy should be given according to the guidelines shown in figure 2¹ (recommendation grade D).
- Conventional phototherapy is an option at TSB concentrations 35 µmol/L to 50 µmol/L lower than the guidelines in figure 2¹ (recommendation grade D).
- Breastfeeding should be continued during phototherapy (recommendation grade A).
- Supplemental fluids should be administered, orally or by intravenous infusion, in infants receiving phototherapy who are at an elevated risk of progressing to exchange transfusion (recommendation grade A).
- Infants with a TSB concentration above the thresholds shown on figure 3¹ should have immediate intensive phototherapy, and should be referred for further investigation and preparation for exchange transfusion (recommendation grade B).
- An infant with clinical signs of acute bilirubin encephalopathy should have an immediate exchange transfusion (recommendation grade D).

Original source: Canadian Paediatric Society. Guidelines for detection, management and prevention of hyperbilirubinemia in term and late preterm newborn infants (35 or more weeks' gestation). Paediatric Child Health. 2007;12(Suppl B):1B-12B. Available at: <http://www.cps.ca/english/statements/FN/FN07-2.pdf> (Accessed on September 8, 2010).

1. See original source for figures and tables providing details related to follow-up and treatment.