

Université de Montréal

**Parental projections of developmental outcome, quality of life and coping in
children who require neonatal intensive care**

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Résumé

Aux soins intensifs néonataux, les professionnels et les parents évaluent le pronostic du développement et de la qualité de vie (QdV). Le but de cette thèse est de comprendre comment les parents prédisent la QdV future de leurs enfants.

Cette étude qualitative basée sur la théorisation ancrée comprend dix entrevues avec des parents. Les résultats indiquent que le pronostic développemental influence les prédictions parentales de QdV, mais il n'est pas suffisant, car la QdV est multidimensionnelle. Les parents utilisent des mécanismes d'adaptation pour gérer la maladie et l'hospitalisation de leur enfant. Ceux qui pensent qu'ils, et leur enfant, seront capables de s'adapter à un mauvais état développemental, prévoient une QdV réévaluée.

Le pronostic neuro-développemental et la QdV future ne sont pas facilement estimés et les professionnels doivent en être conscients. Aider les parents à identifier des mécanismes d'adaptation peut les amener à estimer un meilleur pronostic de la QdV.

Mots clé

Qualité de vie, néonatalogie, prématurité, pronostic, incertitude, développement, adaptation, résilience, recherché qualitative, théorisation ancrée

Summary

In the neonatal intensive care unit, professionals and parents prognosticate about developmental and quality of life (QOL) outcomes. The purpose of this thesis is to understand how parents predict future QOL for their babies during the neonatal period.

In a qualitative study using grounded theory, ten interviews with parents were conducted. The main findings indicate that *developmental prognosis* influences parental predictions of *QOL* but it is not sufficient in defining it, since *QOL* is a multidimensional construct. Parents use a variety of *coping mechanisms* in dealing with the adversity arising from the illness and hospitalization. Parents who believed that they and their child would be able to adapt to an eventual poor *developmental outcome* predicted a *re-framed QOL*.

Neurodevelopmental prognosis and future QOL are not easily estimated and health professionals should be acutely aware of this. Helping parents identify coping mechanisms might lead to more positive prognosis of future QOL.

Key words

Quality of life, neonatology, prognosis, uncertainty, development, qualitative, grounded theory, adaptation, resilience, coping

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List of Abbreviations

ADHD	Attention Deficit and Hyperactivity Disorder
CP	Cerebral Palsy
ELBW	Extremely Low Birth Weight
GRR	Generalized Resistance Resources
HRQOL	Health Related Quality of Life
HIV	Human Immunodeficiency Virus
IQ	Intelligence Quotient
MUHC	McGill University Health Centre
NICHD	National Institute of Child Health and Human Development
NICU	Neonatal Intensive Care Unit
PI	Principal Investigator
QdV	Qualité de vie
QOL	Quality of Life
R-QOL	Reframed Quality of Life
SD	Standard Deviations
SOC	Sense of Coherence
TAMS	Text Analysis Markup System
US	United States
VLBW	Very low birth weight

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Foreword

Clinical work as a paediatrician in the neonatal follow-up department allows for care of babies who start their life in the neonatal intensive care unit (NICU) and for follow-up over the years, as they grow and develop. Some mature into typically functioning children, while others have mild, moderate or severe developmental impairments. Following these children and their families overtime, one can witness how they react and adjust to their difficulties, and observe their unique experiences of quality of life.

When one keeps this long-term perspective in mind, an interest develops in learning more about the mental and emotional processes parents use when they make difficult treatment decisions in the NICU. There are significant ethical implications in the discussions clinicians have with parents, at the moment babies are hospitalized. There is a need to deepen knowledge about what meaning parents attach to terms like “future developmental outcome,” or “future quality of life” and learn more about how they see the future of their baby. A better understanding of parental perspectives of the future QOL of their children will serve to improve our counselling skills and the overall decision-making process in this field of health care.

The first chapter of this thesis briefly introduces the key concepts of developmental outcomes, quality of life, uncertainty and resilience, briefly discusses how they are

related each other and raises some of the challenges they pose to prognostication and joint decision-making.

The second chapter summarises the literature most relevant to the main concepts intended for study in this project. The studies on the premature population are prioritized, since most of the parents who participated had premature babies. The chapter is divided into the following sections: outcomes of prematurity, a common risk factor for neurodevelopmental impairment; quality of life in former preterm children and other populations; studies involving resilience (or its synonyms, adaptation and coping); and the literature relevant to the shared decision-making process.

The third chapter explains the methodology used to complete the qualitative research project that has been at the core of this thesis. The research frame and main research question are introduced. Then, a detailed presentation of the methods includes sections describing the contact with participants, sampling, data collection and management, as well as data analysis. Subsequently, the quality criteria used for the qualitative exploration are explained. The chapter concludes with some considerations of the ethical challenges of conducting this study.

The fourth chapter exposes the majority of the results, presented in the form of a journal article intended for Social Science and Medicine (Soc Sci Med). The article is still in preparation and has not yet been submitted for publication. It contains a concise introduction and methodology overview, and a large part of the results pertaining to the

future predictions that parents make for their children during the hospitalization in the NICU.

The results that could not be included in the article intended for Social Science and Medicine are presented in chapter five. They include a section on the impact of illness, involving themes of grief and attachment and a section relating to the shared decision-making process, as experienced by the participants. To facilitate understanding of the results, Table VI (*in Annex I, p. xi*) summarises the structure of the main themes and categories.

Chapter six closes with a general discussion of the results, analyzing the main concepts in further depth. A particular emphasis is placed on the practical solutions and strategies that health care professionals and families can use in order to improve care and prognostication in the NICU. A practical tool for difficult decisions in neonatology is created using knowledge acquired during the completion of the project. Lastly, the thesis is concluded with closing remarks summarising the principal ideas and recommendations as well as future research avenues.

CHAPTER 1: INTRODUCTION

With advancements in medical care, new technologies and modern surgical methods, the survival of babies requiring neonatal intensive care has increased significantly in the last four decades [1-3]. In premature babies, the most frequent patient population in the NICU, the use of surfactant, antenatal corticosteroids and screening for group B streptococcus have had a major impact on survival [4]. In many centers, babies as young as 23 weeks gestation are resuscitated at birth and given a chance to survive [5-7]. Other patient populations have also benefitted from advances in technology. For example, total body cooling programs have been shown to improve survival in asphyxiated babies [8, 9]. Newborns with cardiac or other congenital malformations now survive more frequently owing to improvement in surgical corrections and intensive care [10].

Despite this marked increase in survival, improvements in developmental outcomes have lagged behind and a significant proportion of children born prematurely continue to experience significant impairments and disabilities [2, 11-15]. In populations with birth asphyxia, therapeutic cooling techniques have given hope of improved developmental outcomes, however many of them still grow up with important health and developmental sequelae [16]. These disabilities can have an important impact on the children's ability to function and perhaps their future quality of life.

The prenatal period, the delivery and the neonatal period are critical times when difficult decisions are made [17]. Since the young age of the patients does not permit them to make autonomous decisions, the surrounding competent adults must determine what is in the best interest of the child. To do so, medical teams and parents, working together,

attempt to predict future neurodevelopmental outcomes in order to establish which course of management most appropriate for the patient. Unfortunately, the range of possible outcomes is often very wide, making these predictions extremely difficult [2]. Given the plasticity of the young brain, this is particularly true when predicting neurodevelopmental outcomes. The literature clearly documents the long-term developmental and functional outcomes of premature, asphyxiated and other populations [8, 9, 15, 18-21]. However, even though they give clinicians an overview of statistics and likely outcomes based on specific risk factors, these studies do not help predict the future of individual patients. Each child is unique and his or her future can rarely be predicted with certainty.

In addition to developmental and functional states, quality of life is increasingly recognized as a major factor in determining whether to pursue or interrupt treatment [22, 23]. A poor future quality of life is not considered to be in the best interest of the patient and is often given as a reason for discontinuing active treatment in the neonatal period. This is particularly true when the baby is very sick and requires increased amounts of painful tests and procedures. A wish to avoid harm for very little benefit in the present or future is usually behind the motivation of limiting treatments. However, making such decisions based on quality of life projections is highly problematic.

Increasingly, the literature indicates that quality of life is extremely difficult to estimate and to predict by healthy individuals. Long-term quality of life studies have provided important information regarding the outcomes of older children and adults who have required neonatal intensive care [24-30]. These reports have especially focussed on

formerly premature individuals, a frequently studied population. The majority of these studies indicate that the objective measures of health and developmental status of patients do not help to estimate how they rate their quality of life. Patients with significant physical limitations consistently rate their quality of life as better than expected by professionals working in health care and by other healthy people [31]. This is partly because quality of life is a highly subjective product of a complex interaction of physical, psychological and social factors, and the physical health of a person reflects only a limited aspect of this multifaceted construct [32, 33]. Additionally, individuals with birth or later injuries often experience an adaptation to their impairment, and this allows them to see their reality in better terms than expected by non-impaired individuals. This process of adaptation or coping with disease and adversity has been documented in the psychological literature [34-37].

Consideration of all these aspects renders the shared decision-making process between parents and medical teams very complex. Determining what is in the best interest of a non-autonomous patient becomes very challenging. In addition to extreme difficulty in predicting developmental outcomes, the future quality of life of the patient likely will not correlate well with these predicted outcomes. Being concerned primarily with physical complications and lacking experience with the long-term follow-up of similar patients, NICU workers may focus on physical health and ignore the complexity of quality of life and the human capacity for adaptation. This may lead to an underestimation of the future well being these children. Since doctors have tremendous influence on how parents interpret medical information and how they view the best interest of the child,

decision-making may not be adequately shared [38, 39]. Professionals must ensure that families receive the necessary information and support in order to make informed decisions for their baby. Parents have their personal experiences and values and they make their own projections of developmental outcomes and quality of life for their children. These projections may be similar, but sometimes they differ widely from the views and attitudes of the neonatology team. For this reason, it is extremely important for clinicians to understand parental perspectives. An in-depth comprehension of how caregivers make predictions for the future of their baby and how they share decision-making will help medical teams provide better family-centered care during an extremely stressful time.

Multiple ethical dilemmas presenting during common patient interactions have led to an intimate relationship between clinical ethics and excellent medical practice [40]. Since this project intended to improve the quality of patient-centered care, it was completed as part of a pediatric clinical ethics master's. A grounded theory framework was considered most appropriate to study the process of quality of life prognostication, because the existing literature on this topic is scarce and new theoretical insights are needed.

CHAPTER 2: LITERATURE REVIEW

This chapter will present a focused overview of the literature that is most relevant to the main themes of our study. The first section will discuss studies documenting outcomes of survival and neurodevelopment in premature patients, from early childhood to adulthood. The focus is placed on extreme prematurity because these patients form the largest and most studied population in the literature, and because most of the participants in our study had premature babies. Emphasis was put on selected articles introducing the reader to the notion of outcomes and impairment, rather than on performing an in-depth systematic review of this literature. The following section introduces the concept of QOL and its relationship with illness and developmental impairment. Since predicting future outcomes and QOL entails a great deal of uncertainty in the neonatal period, this concept is reviewed in the next section. Then, key studies involving resilience are considered, since adaptation to adversity is an important aspect of maintaining a good QOL through the illness experience. Finally, the last section introduces the process of shared decision-making, an important way to deal with the dilemmas raised by the uncertainty of outcomes and future QOL.

Outcomes of prematurity

Babies who survive after their hospitalisation in the NICU have a multitude of diagnoses and the outcomes can vary widely [12, 41]. Premature babies constitute the majority of patients admitted to the NICU and many studies have documented their outcomes [42, 43]. Therefore, this literature review of outcomes concentrates mainly on this population.

Before reviewing outcomes, a few definitions commonly used to define this patient population are introduced. Prematurity is defined by a birth before 37 weeks of gestation. An extremely premature baby is born before 28 weeks, while a very premature one is delivered between 28 to 32 weeks. The 32 to 37 week gestational age corresponds to a moderate to late prematurity [44]. Some studies recruit patients based on their weight at birth in order to eliminate uncertainty about gestational dating. A newborn weighing less than 1000 grams has an extremely low birth weight (ELBW). A very low birth weight (VLBW) falls between 1000 grams and 2500 grams [44]. For practical reasons, cohorts recording outcomes for extreme prematurity and those including extremely low birth weight babies tend to follow similar populations.

In short term studies, usually documented at 18 or 24 months of age, the focus has often been on survival and on serious impairments such as cerebral palsy, deafness, blindness and severe cognitive impairments [11, 12, 18].

Practices differ between individual centers, cities and countries and survival at the threshold of viability can vary widely. In a review of international data, survival rates at 23 weeks gestation ranged from 2 to 35%, at 24 weeks gestation they were 17 to 62% and at 25 weeks gestation they varied 35 to 72% [11]. As expected, with increased gestational age at delivery, there is improvement in survival as well. The NICHD Neonatal Research Network reports survival rates of 6%, 26%, 55% and 72% at 22, 23, 24 and 25 weeks gestation respectively [12]. The Canadian Neonatal Network records

survival rates in extremely preterm babies ranging from 18% at less than 23 weeks gestation to 81% at 25 weeks gestation [44].

Those who survive extreme prematurity are at increased risk of neurodevelopmental impairments compared to term counterparts. In a recent analysis of the Canadian Neonatal Follow-Up Network, outcomes of extreme prematurity were reviewed [45]. Out of 2528 infants recruited for follow-up, 2109 were evaluated at 18 months corrected gestational age. The results showed that 6.8% of patients were diagnosed with cerebral palsy; in an additional 3.5%, this diagnosis was suspected but not yet confirmed. Results of the Bayley III cognitive, language and motor scales showed that 14.8%, 34.9% and 22.4% respectively, scored below 85. This indicated that they performed lower than the average expected for their age. Short-term studies such as this focus mostly on tests and findings that are easier to administer and record at young ages but who do not necessarily indicate more subtle difficulties seen in older children.

Longer-term studies included patients at mid-childhood age, and used more specific testing to detect these subtle difficulties in the apparently non-impaired children. In a large meta-analysis, Bhutta et al. have concluded that prematurity can have an impact on cognition and behaviour in school-aged children [20]. They reported that prematurely born children had more externalizing and internalizing behaviours than controls, in 81% of the studies reviewed. In addition, the prematurely born children showed lower cognitive scores and more than twice the relative risk for ADHD when compared to controls [20].

At adolescence, most studies show that very low birth weight and extremely low birth weight children perform less well on academic tests and show mean IQ scores between 8 and 13 points lower than controls [2]. Interestingly, at adulthood, these earlier differences do not seem to impair their ability to integrate in society and become functional individuals [46]. Saigal et al. found no differences between adults born ELBW and normal birth weight in the rates of living independently, marriage/cohabitation or parenthood [47]. Other factors such as family socio-demographic status can influence adult outcomes, therefore, birth weight or gestational age become less powerful predictors of outcome in the long term [48]. This is particularly important, since many decisions in the neonatal period are taken in accordance with results from short-term studies. In addition, the descriptions “severe” or “significant neurodevelopmental impairment” are primarily used to simplify research terminology. As will be discussed in the next section, “moderate” or “severe” delay may have different meanings to individual people, and the effect of the delay on their quality of life can vary widely.

Quality of life

The World Health Organization describes quality of life as an **individual’s perception** of his/her place in life, in the context of the culture and value system and in relation to his/her goals, expectations, standards and concerns [32]. This broad definition illustrates the **subjectivity** and the **complexity** of the notion of quality of life. Multiple aspects of a person’s existence come together in an intricate interaction and none can be

isolated and considered separate from the others. Other researchers have described this holistic concept as having the “ability of being” (physical health, psychological health and spiritual values), as “connecting with one’s environment” (physical, psychological and community belonging) and as “achieving personal goals” (practical, leisure and personal growth) [49].

With the purpose of simplification and primarily to facilitate research, the concept of health-related quality of life (HRQOL) has been used by many studies [50-53]. HRQOL is also a subjective judgment, but it refers to the aspects of quality of life that are related to and influenced by one’s health. Testa and Simonson have defined it as the physical, psychological and social domains of health, which can be influenced by the individual’s experiences and perception [33]. Studies have attempted to isolate HRQOL, to study and quantify it through different questionnaires. Some questionnaires are disease specific (used, for example, in asthma, cancer, epilepsy), some are generic [54-57]. These tools measure in different ways the physical status, the social functioning, and the mental health of the participants living with disease. One limitation of these tools is their reductive characteristic that cannot capture the full picture of the complex phenomenon that is quality of life. In addition, overemphasising function and objective measures of health status may limit their ability to truly focus on the subjective experience. Saigal interestingly calls attention to this challenge by contrasting the concepts of measuring “ill being” instead of “well being” [58]. Despite their limitations, the use of questionnaires in studies of former premature individuals has challenged previously unquestioned assumptions. For example, when comparing ex-premature individuals with the norm,

Saigal et al [29] revealed that even though adults born ELBW reported more functional limitations in cognition, sensation, mobility and self care than controls, there was no difference in the self-reported HRQOL in comparison with controls. Similarly, a review of HRQOL of preterm children concluded that they indeed have poorer health than normal birth weight children, but that despite this, they do not perceive their quality of life as different than their healthier counterparts [59]. In the world of neonatology, these studies confronted the widely believed notions that people with illness are not as happy as healthy individuals and started the debate around the meaning of QOL.

The subjectivity of QOL added further challenges to teams and parents making decisions for a young or non-verbal child. Researchers found that the ability of healthy people to accurately estimate QOL for patients is very limited [60, 61]. In one study that included neonatologists, nurses and pairs of formerly premature adolescents and their parents, investigators asked each group to rate their preferences for a variety of hypothetical health states, ranging from mildly to severely impaired [62]. The health-care professionals (doctors and nurses combined) gave ratings, to the severely impaired hypothetical health states that were significantly lower than the adolescents or their parents. Another study recorded measurements and estimations of mood performed by a group of hemodialysis patients and matched healthy controls [63]. The healthy subjects, when they imagined having kidney disease requiring hemodialysis, rated their hypothetical moods significantly lower than the moods actually rated by the patients receiving the treatment. Remarkably, even the patients themselves tended to underestimate what their moods had been during dialysis, once the treatment was over.

The group of dialysis patients also overestimated how happy they would be if they had never developed kidney disease and imagined a mood significantly higher than the moods healthy controls actually recorded. In a very different population, Middleton examined the attitudes of 87 deaf adults toward genetic testing for hereditary deafness, using a questionnaire tool [64]. This study found that the large majority of respondents (60%) would not want to have a pregnancy test to detect deafness. Intriguingly, 74% of participants stated that they had no particular preference for a hearing or deaf child and 15% actually preferred having a hearing impaired child. Only 6% preferred a hearing child. Despite the limitations of this study (a relatively small number of participants, as well as the more educated status of the respondents), these results promote stimulating reflections on the meaning of QOL and on general assumptions of the relationship between impairment and well-being. One noteworthy explanation comes from Ubel's discussion of "focusing illusions," in his book "You're Stronger than You Think" [65]. According to this concept, when people imagine having an impairment such as paraplegia, they tend to focus on what abilities would be lost, like being confined to a wheelchair or losing the ability to participate in a favourite physical hobby. In the same way, when imagining dialysis, they focus on being stuck by needles or on the restrictions arising from being attached to the machine for several hours. However, there are many aspects of their life that would not be affected, and a defocusing exercise might change perspective. For example, aspects of their work, family life or other types of hobbies might not be affected at all, or only minimally.

As these studies show, QOL evaluations seem to correlate poorly with the physical or developmental status of a patient, making estimations and predictions of QOL very difficult. In the neonatal period, the developmental outcomes are already difficult to predict and consideration of future QOL adds another degree of complexity. This leads to a great deal of uncertainty for the parents of babies hospitalized in the NICU.

Uncertainty

Uncertainty about the future is often a major component of the parental NICU experience. According to Neville, uncertainty arises when patients are unable to sufficiently understand events such as diagnosis, symptoms, treatments and prognosis, because they are not sufficiently distinct, available, specific and familiar [66]. Studies have shown that living with uncertainty can be a heavy burden [67-69]. Sometimes, uncertainty represents a barrier to coping with the diagnosis. One study found that the suicide rate in patients with incomplete spinal cord injury was almost twice as high as in patients with complete tetraplegia, even though the prognosis of the first group was much better [70]. Similarly, another study analyzed depression scores on subjects undergoing HIV testing, before and after (at 1 week and 2 months follow-up) the result. The study revealed a high degree of pre-test suicidal ideation, in both the eventually seropositive and seronegative groups (28.6% vs 30%). At the two month follow-up, suicidal ideation had decreased to 16.3% in the seropositive group and 15.9% in the seronegative patients, an almost identical rate [71]. This points to the idea that

uncertainty is sometimes more difficult to cope with than a diagnosis of a severe illness. Similarly, parents of babies hospitalised in the NICU may wish for a more precise prognosis of development and QOL out of a need to cope with the diagnosis.

With an interesting model, Mishel, proposed a framework for understanding uncertainty in illness [72]. She has done extensive work in explaining how people cope when their illness has an uncertain prognosis. In this model, she shows how the patient interprets uncertainty either as a danger or as an opportunity, and mobilizes different coping strategies in each case. When uncertainty is perceived as a danger, as in the case of a possible diagnosis with poor prognosis, some individuals will attempt to decrease uncertainty by trying to gain control over the situation and gather as much information about the illness as possible. On the other hand, other people interpret uncertainty as an opportunity. For instance, in the case of a poor prognosis that patients believe could still be improved by stimulation or treatments, delaying the certainty of a severe diagnosis might be desirable. These individuals tend to focus on finding new treatments that could still positively influence the outcome.

As Mishel later argues in a revision of her original model [73], medicine has been significantly influenced by the mechanistic view of life of the industrialized society, where a particular cause is believed to lead to a certain effect. In this view, predictability of illness course, accuracy of diagnosis and control over outcome are highly valued, while uncertainty is seen as undesirable. This creates the expectation that doctors should be able to identify the reason for illness and prescribe a treatment that will lead to the

desired outcome of a return to health [73]. This scenario can be often expected in common illnesses that are easily treatable, over a short period of time. However, predicting developmental outcomes early in a child's life is a very different scenario. These outcomes evolve over long periods of time and can be influenced by many environmental factors along the way. As Mishel describes, when illness and the resulting uncertainty invade important aspects of a person's life, the impact moves the patient past a threshold, away from a balanced state, and into a chaotic system. Eventually, people re-organize themselves in a new equilibrium state, through a process of adaptation [73]. Further details and links with this concept are provided in the general discussion section (*p. 93*).

As the previous sections have shown, a hospitalisation in the NICU comes with a great deal of uncertainty about future survival and developmental outcomes. Also, QOL seems to be very difficult to predict, given its complexity and subjectivity. In addition, the adaptation process people tend to experience when challenged with adversity can have a significant impact on how they value their QOL. The concept of adaptation, or resilience, is discussed next.

Resilience

The realization that certain people seem to have a good quality of life despite physical impairment has led researchers to study this phenomenon. The literature on adaptation

to illness is extensive. Many studies of resilience have been conducted in different contexts and populations [36, 37, 74-78]; the most relevant to our study are introduced below.

Albrecht and Devlieger have described the “*disability paradox*” in their qualitative study, in which 153 people with disabilities were interviewed [34]. They found that more than half of the individuals with moderate to serious disabilities experienced a good or excellent QOL. This was surprising to the authors, who also performed individual interviews with each participant. The results subsequently showed that these patients maintained a high QOL by establishing a sense of balance between the illness and their social context, or environment. They were able to understand their condition, take control of their lives, seek knowledge, educate others and form strong social relationships and networks [34].

Adding to the resilience framework, Antonovsky has introduced the concept of “sense of coherence” (SOC) in which “generalized resistance resources” (GRR) facilitate successful coping with the inherent stressors of human existence [79, 80]. According to Antonovsky, when confronted with a stressor, a person with a high sense of coherence is motivated to cope (meaningfulness), understands the challenge (comprehensibility) and believes that resources to cope are available (manageability). This concept led to the construction of a questionnaire directed at measuring the sense of coherence of the subjects studied [79]. In a systematic review, Eriksson identified several studies that used the SOC to predict quality of life [81]. These studies involved mostly adult patients

with psychiatric illness, coronary heart disease, ischemia and hip fractures. Nonetheless, the review found that family SOC was strongly and positively related to quality of life in families who had at least one member with a serious illness. In addition, the results showed that the family SOC was the largest predictor of family quality of life.

Other models of adaptation include the “response shift,” [82] which refers to a change in the meaning people attach to QOL. This shift results from changes in internal standards, values and the conceptualization of QOL, as coping with illness progresses. The “response shift” model builds on the antecedents (or characteristics) of an individual and the mechanisms (or coping strategies) that he or she uses in response to a catalyst (or an adverse health event). As a result, a “response shift” occurs that changes the internal standards people use in order to interpret QOL. Other work has concentrated on cognitive adaptation to threatening events. Taylor [83] describes a theory explaining how individuals, when challenged with personal tragedies, respond with cognitively adaptive efforts in order to return to or exceed previous psychological levels. These efforts include a search for meaning, efforts to achieve mastery and attempts to enhance the self.

On the other hand, not all individuals with impairments experience a good QOL. Adaptation is a long process, with many highs and lows. Chronic pain or other uncomfortable sensations, such as difficulty breathing or extreme itching can impact a person’s QOL [65, 84]. When living with pain for a long time, sensitization of the skin can occur and even a usually benign stimulus, such as simple touch, can become a painful

experience [85]. Social isolation, as well as an individual's personality, can have a significant impact of the experience of illness and the ability to adapt [65].

In addition to the concepts introduced by these resilience studies, which deal primarily with previously healthy individuals who suffered illness, it is important to remember that neonates suffer their injuries at, or shortly after birth. Growing up, they do not have the occasion to experience life without their impairments. Therefore, they evolve and “adapt” to the only life they know, just like every other person has lived with their own set of circumstances and challenges.

Shared decision-making

In the NICU, decisions about pursuing or discontinuing active treatment must be made for the sickest neonates. In such situations, questions arise about the likely prognosis and the meaning of the predicted scenarios for the future QOL of the child and the family. All parties involved usually desire a decision that respects the best interest of the child. However, there are considerable challenges to determining what is in the best interest of the child and which persons are best placed to establish it. Parents and professionals struggle together to make such decisions and an adequate doctor-patient (parent) relationship is crucial to the success of the process.

Adequate shared decision-making is based on establishing a good relationship between the medical team and the patient or the family [39]. Through their encounters, doctors and patients share information and engage in a mutual discussion to establish the best course of management [39].

The therapeutic relationship between doctors (and other health professionals) and patients, can take various forms, depending on the background, experience and comfort level of each of the two parties involved. Over the course of the history of medicine, the “ideal” doctor-patient relationship has changed, under the influence of culture, technology and population education. In today’s western world, the typical patient tends to be more involved and to access medical information more easily [86]. Despite this overall trend, patients may vary widely in their preferences and expectations of their relationship with the medical team [87, 88]. To better understand the doctor-patient relationship, four models are described next [89]. The paternalistic model refers to the type of interaction in which doctors assume the responsibility of making sure that the interventions and management plan are in, what the physician believes, the best interest of the patient. The physician might present only selected information that will encourage the patient to give consent. In this model, the importance of patient well being, as judged by the physician, surpasses that of autonomy. In the informative, or consumer model, the physician provides the patient with the known information about diagnosis, investigation and treatments, with their advantages and risks. It is the patient’s role to decide which management to choose, based on his or her own values. This model promotes a very high level of patient autonomy. These two first models represent the

extremes of patient autonomy and responsibility of the physician. According to a third, interpretive model, the physician provides the patient with information about the medical condition and the available interventions, and helps the patient clarify his or her values, in order to choose the best-suited management. The doctor assumes the role of an advisor counsellor, while the patient autonomy is fully preserved. Finally, in the deliberative model, patient and doctor engage in an active dialogue to exchange information and discuss what health-related values the patient could/should follow. In this model, the patient's autonomy is achieved through self-development and the physician refrains from coercion. These illustrations of doctor-patient relationship are rather theoretical, but help establish a useful framework to understand the different challenges arising in counselling. In reality, patients vary widely in their preferences of interactions with doctors, and even these preferences may fluctuate based on the type of decision that needs to be made. In addition, these models are mostly applicable to adult patients, who have the capacity to achieve informed consent. In paediatrics, the situation is very different. Young children cannot consent to care, thus autonomy refers rather to the freedom that parents have to make decisions for their children and give informed consent, as proxies. This autonomy however, is limited by the extent to which a particular decision is made in the best interest of the child [90]. Establishing best interest for non-verbal children can sometimes be very difficult. Both medical teams and families act as proxies in decision-making and conflicting values or different perceptions of responsibilities can lead to conflicts. How health care professionals interpret best interest is not always consistent across patient populations. A thought-provoking study of physicians and students used an anonymous questionnaire to describe 8 incompetent

patients with potential mortality and neurologic sequelae [91]. Among the scenarios, a very premature baby, a term infant and a 2-month-old patient had identical projected survival and morbidity outcomes. Participants were asked if resuscitation was in the best interest of the patient. The respondents believed that resuscitation was more in the best interest of the 2-month-old baby (97% of respondents agreed) than the term infant (87% agreement). Only 69% of participants thought it was in the best interest of the very preterm baby to be reanimated. This illustrates, that outcome alone is not always sufficient for medical professionals to recommend the same management deemed appropriate for another patient and that value judgments still exist about certain populations, in particular, preterm babies.

The current Canadian guidelines on decision making in extremely preterm babies were published by the Canadian Pediatric Society [92]. In these guidelines, a great deal of emphasis is put on the importance of gestational age and antenatal therapies in establishing a prognosis. Outcomes are described mainly in terms of survival and neurodevelopmental status recorded in short-term studies. Studies reporting long-term outcomes and HRQOL are discussed only briefly.

In a commentary letter, several authors have indicated that current guidelines are insufficient and that the difficult task of decision-making in the NICU needs to be supported with broader and clearer recommendations [93]. They argue that a future position statement should include several additional recommendations: personalization of decision-making, promoting family centered care, encouraging professionals to

discuss QOL, clarifying the definitions of minor and severe disability while avoiding value judgments, and including the role of adaptation and coping.

This chapter outlined the concepts and studies that are most relevant to the main themes arising from this project. The literature on neurodevelopment outcomes of prematurity and quality of life will be pertinent in understanding the way parents make future prognoses for their babies. The uncertainty and joint decision-making sections are important in comprehending the challenges that parents and health professionals face when attempting to prognosticate at a young age and the way they share the responsibility of caring for the ill child. The literature on resilience facilitates understanding of the processes parents use to deal with difficult challenges such as the hospitalization of their baby in the NICU. With this overview in mind, the next chapter outlines the practical methodological details of this study: research question and frame, details on participants and interviews, information about data collection and analysis, quality criteria and some of the ethical considerations arising from interviewing parents during a stressful time.

CHAPTER 3: METHODS

As described in the literature review, parents and health care professionals have the difficult task of making decisions for babies who are ill in the neonatal period. Making such decisions is extremely difficult, due to the uncertainty of the outcomes and the challenge of estimating QOL for a non-verbal child. In addition, considering the role of adaptation to illness further complicates the prognosticating process. When making such future projections early in the life of a baby, professionals and parents may be in agreement with each other, or may hold differing views. The way parents make these predictions for their baby is an important understudied area in the medical literature. This study intends to enrich the knowledge about the parental prognostication process, in the hope of improving the shared decision-making process for parents and team alike.

Research question and frame

Research Question

The main purpose of this study was to investigate the parental perspective of the predicted quality of life of babies admitted to the NICU.

Research Frame

Ethical theory and philosophical foundations are invaluable to understanding the dynamic between different ethical dilemmas presenting in neonatal prognostication.

However, the area of parental predictions of QOL with the purpose of making treatment decisions for their baby has so far been obscure in the medical literature. Little is known of the process parents use to make these predictions. The basis of clinical ethics involves exploring complex questions as they arise from the clinical setting. Therefore, this research question requires more than the application of known theories to a practical situation and needs to be studied empirically, with the help of the subjects who are living through the experience. To understand the full picture of subjective and personal concepts such as QOL and shared decision-making, it is very important that parents are recruited to participate in this study and provide their perspectives. The insights learned from such empirical research can then be used to formulate practical recommendations for clinicians in order to improve practices in the NICU.

Given the exploratory nature of the research question, it was judged that qualitative methods were best suited for this study. Therefore, the project was based on the well-established and widely utilized grounded theory method [94] that helps researchers create new theory in order to explain a particular process to be studied. As an explorative tool, this method uses comparative analysis to compare differences between participants, identify patterns in the data (e.g. behaviours, processes, etc.) and incorporate them into general concepts, which can then be unified into broader theories. Data used in qualitative research can be provided by a variety of different sources (individual interviews, focus groups, videos, photographs, etc.). In our study, it was essentially provided by semi-structured individual interviews.

In data analysis based on grounded theory, codes are assigned to each basic idea. Then, codes are grouped into more general categories. As participant encounters progress, comparisons are made to identify similarities and differences between the ideas and categories identified in each interview. As patterns of categories emerge, these are grouped as themes. At this point, further data can be gathered to test, broaden and adjust them. This is often used as part of quality control in qualitative studies to strengthen the value of the analysis. Finally, a theory unifying the identified themes is generated. Initially, a substantive theory is formulated, then, the analysis is taken further to allow formal theory to emerge from the substantive theory.

Methods:

Timing of the interviews:

Semi-structured interviews were conducted to explore the way the parents project the future quality of life of their children, while they are still in the NICU. Typically, premature newborns are hospitalized during many weeks or months, generally up to the expected term delivery date. During this period, the future quality of life is still uncertain and many treatment decisions are being made. We chose this period for its likeliness to lead to rich information about future projections of QOL.

Setting:

The study was conducted with parents of babies hospitalized in two NICUs in Montreal, Quebec. One of the NICUs is part of the McGill University Health Centre (MUHC) and currently does not have an on-site birthing centre. This unit typically receives patients transferred from secondary care birthing centres in the province or from another MUHC NICU that has a birthing centre and tertiary care neonatal support but lacks the availability of consulting specialists. The other NICU is affiliated with Université de Montréal and has a birthing centre, neonatology support and consulting specialists. Since health care professionals practicing in different hospitals may have unique parent counselling approaches about newborns with uncertain prognosis, selecting parents from different units was particularly interesting for the purpose of this study. The study was approved by the Research Ethics Committees of both universities.

Contact with participants:

Eligible participants were first identified by the neonatologist/ fellow/resident on service responsible for the NICU. Eligibility criteria and theoretical sampling (*Table I, p. 33*) were used to identify patients. Subsequently, the responsible member of the treating team approached eligible parents to inform them of the existence of the study. Those parents who expressed interest met with the principal investigator (PI) who provided written information and full study details. Most who were approached agreed to

participate and informed consent was obtained (*Annex III¹, p. xii*). A copy of the consent form was offered to the participants. Two families refused participation due to a lack of availability. The interviews were conducted privately in the available clinic or office rooms, at a time convenient for the participants.

Sampling:

Theoretical sampling [94] was used to select the study population. This method of sampling allowed us to select patients who were most susceptible to provide useful information and evolved throughout the analysis process. The entire sample was not selected from the beginning. Instead, it used a continuously dynamic process of data gathering and analysis, while preparing where to direct the study next. At the start, the first subjects were selected using the inclusion and exclusion criteria (*Table I*). Given the general characteristic of QOL, the inclusion criteria were voluntarily not restrictive to any particular diagnosis or prognosis. Subsequently, our recruitment focused on parents of patients with uncertain prognosis. Each interview was analyzed before the next and the data obtained guided the choice of the subsequent subjects. In this way, participants were selected according to their relevance to the further development of the emerging theory. The sample size was dependent on saturation of the main emerging themes required for building theory. When no additional data further developed the core conceptual categories, the data collection process stopped.

¹ *Consent form refers to Phase 1 and 2 of a longer project, planned over several years; this thesis was only based on Phase 1*

Table I: Inclusion and exclusion criteria for participants

<i>Inclusion criteria</i>	<i>Exclusion criteria</i>
Parents of children hospitalized in the NICU Fluent in English or French Resident of Quebec, living in the greater Montreal area	Less than 18 years old The PI is already involved in the care of the child

Data collection:

Individual interviews lasted between 45 and 60 minutes. Most of the interviews were conducted with only one parent. In one case, both parents were interested in participating and they were interviewed together. A discussion guide containing open-ended questions was used to lead the encounter (*Annex III, p. xx*). Questions were structured around the experience in the NICU and the predicted future quality of life. Since the interviews were exploratory, the PI provided a minimal amount of guidance. Questions were used primarily to clarify and deepen the discussion. The interview did not provide medical information to the participants and the PI refrained from assuming a counselling role. As part of routine NICU care, services of a psychologist or a social worker were available for parents in need of support. Several participants were actively followed.

Interviews were audio-recorded and transcribed, word for word. Notes were taken immediately post-interview recording non-verbal cues and interviewer's impressions.

All the transcriptions were read by at least two of the co-authors.

All the electronic data were password protected and stored securely. All printed documents containing data were kept in a locked filing cabinet. Only the researchers had access to the data. After transcription, written material was carefully examined and all identifiable information was removed/coded (names, pseudonym, place, names of hospitals, age, rare disease diagnosis). Analysis was performed with anonymous data. After completion of the project and publication of the results, all audio material and full text written material will be securely kept for five years as required and then destroyed.

Data analysis

Analysis was performed with the help of the TAMS Analyzer software [95]. Open codes were assigned to each basic idea and then grouped in general categories. Similarities and differences between identified ideas and categories were noted. As patterns emerged, categories were grouped into themes. With interview progression, as analysis advanced, the focus of the subsequent interviews changed to explore new categories. Grounded theory was constructed as described in the research frame section.

Frequent interactions and discussions between the PI and the main supervisor took place in order to exchange insights, ideas and ensure the quality of analysis. The PI and the main supervisor coded selected samples separately, in order to ensure adequate

coding methodology and coding reliability. The thesis director frequently reviewed the code list and grouping into categories and themes.

Quality criteria

Quality of the analysis was ensured following criteria for qualitative methods [96-98].

Triangulation consisted in interviewing parents from different hospitals, using audio and non-verbal information and involving two of the co-authors in the data analysis.

Throughout the analysis of qualitative data, particular attention to “deviant” cases was paid. Elements that contradicted the emerging explanation of the phenomena being studied were identified and further explored when needed. This continued until all or most of the cases observed could be included in the theory proposed. Constant discussion and feedback between the co-authors added to a significant reflexive effort made throughout the data analysis.

Ethical considerations

This project considers a question primarily in the clinical ethics domain. The difficult decisions made in the neonatal period involving very sick babies and predictions of quality of life have an ethical nature and are part of the daily experience of parents and clinicians. There is significant value in studying these questions and further elucidation

of this understudied area will improve the quality of parental counselling. The methods chosen are well established in the scientific qualitative research literature.

Informed consent was obtained for all participants (interviewees). Each had the right to refuse participation or withdraw at his/her own discretion, without any consequences on their care. Their privacy was protected through careful confidentiality measures.

Beneficence guided us in our intention to further the scientific knowledge and benefit society. The participants might also have benefited from the encounters by finding comfort in being listened to during such difficult times. There was initial concern that talking about such emotionally charged subjects might cause them distress. Participants were fully informed of this risk in the consent forms and they had the opportunity to withdraw at their discretion. Several parents cried during the interviews, although none expressed the desire to stop the interview or to withdraw from the study. It is estimated that this emotional risk was not higher than the risk they experience in their routine counselling encounters with the medical team.

In conclusion, this methodology chapter introduced the context in which the research frame and questions were chosen for this empirical ethics research project. It also provided details about participant choice and interview conduction, as well as methods of data collection and analysis. The next chapter, details the main results of the analysis, presented in the form of an article intended for publication in Social Science and Medicine.

CHAPTER 4: RESULTS

**JOURNAL ARTICLE INTENDED FOR SOCIAL SCIENCE AND
MEDICINE (*SOC SCI MED*)**

The contribution of each author to the completion of the research project and manuscript/article writing is outlined below:

Andreea Gorgos (principal investigator): performed the literature review; wrote the initial study proposal for science and ethics board approval; met with participants to explain study; conducted all the interviews; performed the data analysis; composed the manuscript and article for publication.

Shuvo Ghosh: provided guidance and insights about research methodology proposal submission and approval process; edited different versions of the written proposal, manuscript and article.

Patricia Riley: provided insights in research methodology in the neonatal field; helped define the study structure; made recommendations for the written proposal, manuscript and article.

Antoine Payot (principal master's thesis supervisor): provided insights and very close guidance for all the steps involved in this project: from choosing the research question, to the final version of the manuscript and article.

Parental projections of developmental outcome, quality of life and coping in children who require neonatal intensive care and their implications for decision-making

Gorgos A, Ghosh, S, Riley P, Payot, A

INTRODUCTION AND BACKGROUND

(Abbreviations are identical to those used in manuscript).

Many difficult treatment decisions are made for infants with severe illnesses in the neonatal intensive care unit (NICU). Despite the increasing survival rates resulting from improved medical and surgical care [1-3], an important proportion of these children continue to experience long-term neurodevelopmental impairments [2, 4, 5]. When babies are ill, health care professionals prognosticate about future outcomes and quality of life (QOL), in order to help decide what management might in the best interest of the patient. In particular, prediction of QOL is the major criterion used in decisions to withdraw or withhold treatments in neonatology [6].

Using predictions of development and QOL in the decision-making process can be problematic because of the inherent limitations of prognosis at very young ages.

The plasticity of the developing brain and the role of environmental factors allow for a wide range of possible neurodevelopmental outcomes[2]. Population based studies,

while giving clinicians an overview of statistics and likely outcomes based on specific risk factors, do not predict the future of each unique individual. Furthermore, even if future developmental outcomes were easier to predict, they do not correlate well with future QOL. Many studies show that patients with significant physical limitations rate their QOL as better than expected by health care professionals [7] or other healthy individuals [8]. This discrepancy is partly explained by the fact that QOL is a product of a complex interaction of physical, psychological and social factors, and physical health is only one of them [9, 10]. In addition, many patients eventually cope with illness and adapt to their new reality. Both the process of adaptation to disease and resilience are described in the literature [11, 12].

Besides the professionals, parents make predictions of their children's future development and QOL. These may be influenced by the medical prognosis or may be a separate vision of the future, according to personal values, familial or cultural background. In a true joint decision-making process, it is crucial that health care professionals explore and understand the perspective of the parents [13]. How parents predict future QOL is an important, but understudied area in the neonatal literature. Examining this process could improve the quality of prognostication made early in the life of these children, help parents and medical teams communicate more effectively, and ultimately inform the decision-making process. The purpose of this study was to explore the parental perspective of the predicted QOL of babies hospitalized in the NICU.

SETTING

The study was conducted with parents of babies hospitalized in two tertiary NICUs. Since health care professionals practicing in different hospitals may have unique parent counselling approaches, selecting parents from different units was particularly interesting for the purpose of this study. The protocol was approved by the Research Ethics Boards of both participating universities.

METHODS

This study used qualitative methods of patient selection and data analysis based on the grounded theory method [14]. A member of the medical team approached eligible parents. Participants who expressed interest met with the principal investigator (PI) who provided written information and full study details. Most parents consented to participate. Two families refused participation due to limited availability.

Participants:

Ten semi-structured interviews were conducted between July 2012 and February 2014. A convenience sample was chosen based on eligibility criteria: parents of children hospitalized in the NICU, fluent in English or French, Quebec residents, living in Montreal or nearby. Excluded from the study were parents of less than 18 years of age and parents

of patients receiving care from the PI. After the initial convenience sample was selected, theoretical sampling [14] was used to further identify eligible patients.

Interviews:

The PI conducted all the interviews in a private room near the NICU. Interviews were exploratory and used open-ended questions around the NICU experience and predictions of future QOL. As themes began arising from analysing the initial interviews, they were further explored in subsequent parent encounters.

As part of routine NICU care, services of a psychologist or a social worker were available for parents in need of support. Several participants were actively followed and none expressed increased emotional distress as a consequence of being interviewed.

Data analysis:

The interviews were audio-recorded, transcribed and all identifying information was removed/modified. Notes were taken immediately post-interview, recording non-verbal cues and interviewer's impressions. All the transcriptions were read by at least two of the co-authors. Analysis was performed with the help of the TAMS Analyzer software. Open codes were assigned to each basic idea; these codes then were grouped in general categories. Similarities and differences between identified ideas and categories were noted. As patterns emerged, categories were grouped into themes. Each interview was

analyzed before the next and the data obtained guided the choice of the subsequent subjects and the themes to be further defined. Subsequently gathered data was used to test, broaden and adjust the themes. When the core conceptual categories were sufficiently well defined, data collection was stopped. For publication purposes, quotations in French were translated into English.

Quality criteria:

Quality criteria for the analysis included triangulation (interviewing parents from different hospitals, using audio and non-verbal information, involving two of the co-authors in the data analysis), attention to “deviant” cases and reflexivity (frequent discussions between the co-authors took place in order to exchange insights, explore biases and ensure analysis quality).

RESULTS

Participants and infants

Participants

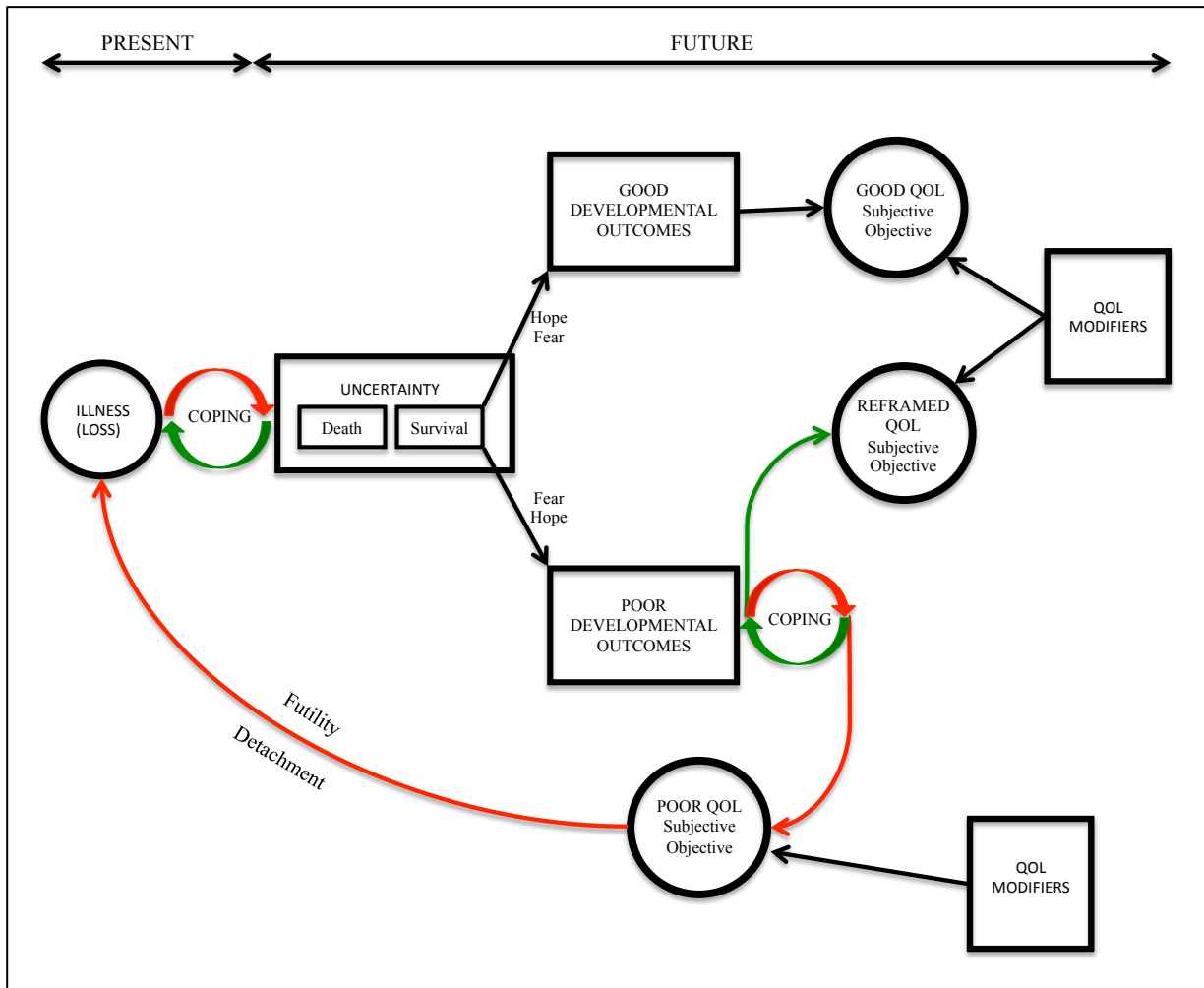
Nine interviews were performed with a single biological parent (8 mothers and 1 father). In a tenth interview, both biological parents were present. Two of the parents were health-care professionals.

Infants

One of the parents had a set of twins. The rest were singletons. Each was hospitalized for different reasons: eight of the babies were premature; their main complications were either severe intra-ventricular haemorrhage (2), broncho-pulmonary dysplasia (2) or esophageal atresia (1). One baby was hospitalized for a brain malformation and one was treated with total body cooling for moderate neonatal asphyxia.

Data analysis revealed the following themes: *uncertainty and best interest, outcomes of development and quality of life, as well as coping mechanisms*. The grounded theory emerging from these themes is illustrated in a conceptual model in (*Figure 1, p. 45*), which can be used as a visual reference while the main themes are introduced and discussed. This model also explains the relationship between the themes and proposes the concept of a re-framed QOL through predictions of coping.

Figure 1: Conceptual model of theme relationships



Uncertainty and Best interest

Uncertainty

When talking about the future, uncertainty was ubiquitous. Parents were very aware of the inherent difficulty to predict outcomes, both regarding the short-term survival, as well as the long-term developmental outcomes.

Most parents were distressed by the “rollercoaster” nature of the NICU hospitalization and were very aware of the fragility and instability of their baby’s health. They understood, and some even expected, that from one day to the next, their baby’s state might go from well to critically ill, or from imminent death to recovery. Even parents, who now believed their baby to be out of immediate danger, described in detail how difficult it had been for them when survival had been unsure. Several stated that the uncertainty of the short-term survival was an important barrier to being able to imagine their child in the long-term: *“You cannot think about the future anymore, because you don’t know if they will be alive after” (Interview #10)* and *“we are in survival mode and ... If you say <<survival>>, it means that you project (...) with difficulty, two, three years later” (Interview #5).*

Best interest

Due to the high level of uncertainty, many parents expressed great ambivalence about important decisions that might affect survival or long-term outcome of the baby. One

parent recalls reflecting, *“who am I to decide that this life will stop? But again, who am I to decide that I am going to make a handicapped child live? Because it’s not just the impact on her, it will handicap the life of those around us. It will decrease the quality of life for everybody”* (Interview #9). Another mother mentioned, *“since he was born, Michael, we have tried to ask the question a few times, to figure out if we were embarking on futile treatment, and we have a hard time drawing it, where is the line of futile treatment? We want him to live, we want him to be with us, we love him already, but we are afraid of embarking on this futile treatment at all price, of life at any price”* (Interview #2).

Parents described the values they cherished and, invariably, the best interest of the baby was a priority. However, parents differed widely in how they thought best interest should be achieved. Some held strong opinions about the sanctity of life and their wish for the medical team to do *“all they can,”* while others talked about avoiding suffering even if it meant withdrawing active treatment.

Predictions of outcomes

Developmental outcomes

Aside from uncertainty about short-term survival, described above, most outcome predictions focused on future development (*Table II, p.49*). Good developmental outcome was defined by abilities and function, such as walking, talking, eating, seeing, hearing, etc. On the other hand, an inability to perform these functions independently

was considered a poor outcome. Having a certain degree of intelligence and an ability to learn in school was important to most parents. They accepted that their child might show certain delays in their development, especially at a young age, and thought a diagnosis of attention deficit and hyperactivity disorder or some school problems were mild difficulties. A poor developmental outcome was also characterized by severe cognitive impairment, autism or lack of awareness (self or environment). Importantly, not all parents automatically feared a diagnosis of cerebral palsy (CP). Many were aware of the wide spectrum of CP and accepted a mild degree of impairment that still allowed the child to be functional. One parent said, *“for us, (...) an attention deficit, or let’s say a learning difficulty as such, you know, (...) it’s not a sequela, you understand. It’s certain that for us a sequela, (is) the fact that she would have difficulty walking when she walks with a walker”* (Interview #9). Another participant mentioned, *“that my children might be hyperactive, it doesn’t bother me. (...) I want them to be normal. Being dyslexic, needing a little computer, it’s as if it doesn’t scare me; but to be mocked because they are different. Being physically... in a wheelchair, things like that ...”* (Interview #10). Overall, parents were very motivated to stimulate development and to seek professional help if delays were present. Most of them understood that they could potentially play an important role in maximizing the future development of their child.

Table II: Summary of Developmental outcome categories

Good developmental outcome	Poor developmental outcome
Ability to walk, communicate, eat, hear or see Intelligence, learning in school Acceptable problems: mild delays in development, mild cerebral palsy, attention deficit, hyperactivity, mild school difficulties	Inability to walk, communicate, eat, hear or see Severe cerebral palsy Severe cognitive impairment Unawareness of self and environment Autism

Quality of life outcomes

The data analysis of this study revealed that parents understand QOL as a complex construct, with a subjective dimension, as well as an objective one (*Table III, p.52*). The subjective aspect was exposed through the belief of most parents that the level of quality of life was closely related to the ability of the child to be happy and to enjoy life.

“Everybody has a bit of their own definition of what is good for them, and what makes them happy, it’s a bit like that, my perception, quality of life is a bit what makes you happy”

(Interview #2). Consequently, several parents were aware that QOL estimation is very difficult. They believed QOL is personal and the child’s perceptions can be different from their parents’, just like societal and individual perceptions of QOL can be distinct. One mother mentioned, *“I think it’s something that is personal and it’s hard to say <<their quality of life is this>>. It’s something that is really abstract. (...) I think that what we perceive and what they perceive is totally different. That is completely different. And what the society thinks of the quality of life of a certain person just by looking, I think it’s totally*

different” (Interview #7). Another parent reflected, “it’s very possible that my child might be happy and I would think that he’s not happy. It’s very easy to think like that. (...) It’s our perception. (...) We are not in his head. Because we compare ourselves. We compare our own quality (of life), and the reflection that, let’s say, a three year old child can have, is totally different from the one of a 35 year old person”(Interview #9).

Other parents, who hadn’t previously considered the subjectivity of QOL, had interesting reflections during the interview. One mother who had previously stated that she believed poor developmental outcomes led to poor QOL spontaneously opened to the possibility of her child being happy even with a developmental impairment, because it would be the only life he would know. She realized that it was she who would be unhappy about her child’s state: *“He will live with his illness, no matter what he experiences. Let’s say, he has some problem, and he has medical appointments, he will only know that. So, finally, it will be part of his routine ... But what would be sad for him, it would be if his Mom doesn’t accept all this, and that I would feel bad to make him feel this ...this... my discomfort about it. And then finally, that he would be unhappy. Not because he comes here – because for him, it would be part of his life, to come here. But, that he would be unhappy because Mom is unhappy (...). I know I have a lot of work to do”(Interview #5).*

Furthermore, several parents raised the issue of their child’s ability to be aware of him/herself and of their surroundings. Even though a child’s lacking contact with the environment and being unaware of his/her own developmental status were seen by parents as a poor outcome, they believed that such a state did not automatically lead to a

poor QOL. If the child doesn't realize he/she is different, is well protected and the basic needs are fulfilled, parents believed that a good QOL was possible. On the other hand, a child with an impairment who is aware of his or her own difference or who is being teased and bullied at school can have a significantly lower QOL. One mother said, *"even if she is disabled, God forbid or something is wrong with her... In her own world, she'll be happy. Like, she won't realize that she's different (...) I mean, when they are small, because for them, that's their way of thinking. (...) Yeah, if she's a bit slow, obviously at school and, kids do, say things, so, yeah... And obviously she'll be hurt, we'll be hurt"* (Interview #3). Another parent reflected: *"You know, if he is the least bit conscious of what the others say, (...) for sure that... You know, I think you can be happy, but when you see that others are judging you and laugh at you, it bothers you a little more"* (Interview #8).

The objective dimension of the construct was revealed through the examples parents used to illustrate what they believed to be a good or bad QOL (Table III, p. 52). They focussed on the ability of the child to fulfill age appropriate roles. They associated good QOL with independence and autonomy, as well as fulfillment of social roles such as having friends, a family and children, getting an education and being employed. *"For me, quality of life is this. It's basic care, it's the needs you have without having to rely on the others, it's being able to be autonomous. I think that autonomy really helps a lot"* (Interview #9); *"It means that we have a child who is able to become fulfilled, able to play, who can make friends, who will be able to learn... (...) that he would be able to have an occupation that he likes, that he could get married, have children, if he wants. Despite differences, that he would be able to evolve roughly like a child ... for the reason we have children ... to lead*

him to a certain autonomy, to not always be dependent on his parents”(Interview #2).

Although not a universal theme, the notion of “normality” and being like others was important for some parents: “quality of life, is this, it’s being like the others” (Interview #10); “it’s being able to offer ... being able to offer him the same things his sisters had” (Interview #5).

Besides not achieving these roles, parents also associated physical pain or its subjective interpretation, suffering, with a poor QOL: “not having too much pain, too. This is something... I think pain, chronic pain and stuff can play on your quality of life. So I guess this is another thing. Pain related to surgeries or pain related to having reflux, and stuff like that” (Interview #7).

Table III: Summary of QOL categories

	Good QOL	Poor QOL	Modifiers
Subjective	Happiness Life enjoyment Personal and difficult to estimate by proxies Different from but linked with family QOL	Unhappiness Absence of life enjoyment Personal and difficult to estimate by proxies Different from but linked with family QOL	Developmental status Ability to function Personality Family’s support and love Education
Objective	Independence Autonomy Having friends Having a family Becoming educated Employment Being “normal” or like others Free from pain and suffering	Dependence Absence of autonomy Lack of friends Lack of family Being uneducated Unemployment Being “abnormal” or unlike others Experiencing pain and suffering	Placement in long term facility Friends Society Support groups Adapted schools Technological progress

QOL modifiers

Aside from the developmental status and the ability to function and fulfill age appropriate roles, a number of factors were mentioned as important in increasing or decreasing QOL (*Table III, p. 52*). As will be explained later, there was significant overlap between these QOL modifiers and the coping strategies parents used in dealing with adversity.

Among the **QOL modifiers**, the child's own personality and reaction to illness were mentioned. Parents recognized that some children might be inherently more optimistic or more pessimistic when making sense of their own illness. This reinforced the concept that QOL is a subjective experience. Participants believed parents had an important role in shaping the child's personality and helping them deal with adversity. All the parents thought the family's presence, support, love, protection and education would increase QOL: *"he will still have people around him, he will still have people who will love him and I think that, often, it's the people around who will suffer more than the person, per se"* (*Interview #8*); *"I don't know if the diagnosis really impacts the quality of life or if it's just the way we deal with it. Well, it's like if we don't make a big deal of it and we just continue just to push through it and to try to give him as a normal life as possible, I feel like he's going to be a happy boy and he's going to have a good quality of life. If he's limited or I limit him, I don't know, I can't do... I feel like he would lose a little bit of his quality of life"* (*Interview #7*). Several parents feared being forced to "abandon" their children through placement in a long-term facility, if caring for them at home became too difficult, and

believed this might negatively impact their QOL. In many interviews, participants had difficulty with separating their own or their family's QOL from the QOL of their child. There was a general awareness that the two were closely connected and had the ability to influence one another.

The extended family, friends and society at large were additional factors influencing QOL. The presence of support groups and resources, adapted schools for children with delays, medical progress and technology were positive factors. On the other hand, parents believed the stereotypes which people use to compare children with impairments to the "norm" could make their child feel inferior and decrease his or her QOL. Parents talked about the external pressures of normality and performance and the risk for their child to be bullied and rejected if different. *"I believe that, in fact, the ultra performing society makes it so (...) You know... you have to go to daycare, you have to already start to be stimulated, you have to play hockey, you have to bla, bla, bla. You know, already, there is a pressure, you have an external pressure (...). However, we have a society who could help take care (of the child), but on the other hand you have a performing society, who leaves the weak ones on the side" (Interview #10).*

Data analysis showed that many of the themes relating to the factors that could potentially increase or decrease QOL were overlapping significantly with the coping mechanisms and resilience elements that parents used to deal with their child's illness. Such themes included parental or child personality, family and friends, the role of

society, etc. Nonetheless, the analysis of coping mechanisms revealed some new and unique elements as well (*Table IV, p. 58*).

Coping mechanisms

Personal resources:

Staying positive and maintaining **hope** for good outcomes such as survival and good development was a very frequent coping mechanism. Parents focussed on positive thoughts, projected normality in the future and concentrated on the baby's resilience and ability to fight. Some parents mentioned "*finding a new strength*" they didn't know they had. Another talked about using humor to stay positive. A few discussed their efforts to reduce their perfectionist tendencies and accept their children as they are. A mother of a baby with a very uncertain neurologic diagnosis and prognosis criticized the medical team for being too negative. She emphasized that she needed some positive elements in the prognosis, in a situation where nothing could be stated with certainty. When thinking about the prognosis received from the medical team, another mother focussed on the fact that statistics don't necessarily predict the future of one child. She said, "*they're statistics. (...) they don't take people as such; they take a mass. (...) My daughter is somebody, my daughter is a personality, my daughter, she has her own strength. She is not like the others, and you know, I don't want them to compare her to anybody, I want her to show them exactly of what she is capable*" (*Interview #9*). Parents also remembered instances where the doctors had erred in prognosis and maintained hope that they would err again.

When articulating feelings of hope, fear was almost always present as well. Parents feared death and worried about the possibility of a bad developmental outcome. Many talked about fearing the unknown and future uncertainty. One parent who had been considering palliative care for the baby was worried about making the wrong decision and regretting it later. Figure 1 (p. 45) illustrates the close relationship between hope and fear in the context of uncertainty: when parents hoped for a good outcome, they were also afraid it will not come true; when they feared a poor outcome, they responded by hoping that it will be prevented. The two feelings existed concurrently when discussing the future and its uncertainty. Interestingly, when parents envisioned their child growing up with a severe developmental impairment, they often went beyond simply hoping that this will be prevented. Instead, they talked about hoping for future resilience for both parent and child. Hopeful parents were able to predict that they, and their child, would be able to cope with the future adversity arising from a poor developmental outcome. As a result, the QOL they were projecting for their child was very different from the one predicted by the parents who did not envision future resilience. This concept is further discussed in the next section, *predictions of coping in the future and QOL re-framed* (p. 59).

Acceptance of the current illness and of the possibility of a future with more adversity helped parents find some inner peace, especially when the grieving process was very difficult. One mentioned, “*we won’t have any choice but to accept it. Therefore you know, regardless, life gives you things, and you have to accept them*” (Interview #9). Parents also

drew on learned lessons from their **past experiences** in order to deal with the present. One mother remembered her own experience with illness in her youth to conclude that her child will likely have a good QOL even with a physical impairment, just like she did when she was young. Parents sometimes **compared** their baby **to other, sicker patients**, and found reassurance that their baby was doing better: *“I know we’re not allowed to look at the other babies in the other beds, but when you compare, you console yourself” (Interview #9).*

Faith was an important theme for many parents. Although some relied on traditional models of religion, others described themselves as non-practicing but needing belief nonetheless. Interestingly, one mother said she believed in her baby.

Living in the present was a frequent coping mechanism, used especially to deal with the uncertainty of the future. Many parents talked about *“taking one step at the time,”* making decisions for now and focusing on short-term goals such as the discharge home. This kept their thoughts away from the frightening possibilities of the future. To help them focus on the present, parents did their best to maintain a routine, be as present at the bedside as possible and get involved in the daily medical and developmental care of their baby: *“Having a routine helped. A routine (...) of getting involved a lot, a lot, a lot. (...) of taking part in the treatments, of being involved in medical rounds. (...) Because Dr. Jones told me: it’s with a lot of stimulation that you’ll help them. Well, I told myself: ok, this is it, we start right away; we’re going to stimulate them, we’ll do our best, and... So this helped me a lot, to take part, in a way, in the treatment (...), to feel involved” (Interview #10)*

External resources

In addition to using their own resources, parents relied significantly on surrounding help. The strength of the relationship with their life partner, as well as the surrounding family and friends helped them to become more resilient. Many found support through organizations supporting parents of babies with similar problems. Educating themselves about the baby's illness through medical literature and reading parental blogs was often helpful. However, reading on the internet sometimes had the opposite effect, worrying them about complications that might occur.

Table IV: Summary of Coping mechanisms categories

	Coping mechanisms
Personal resources	Personality Positivity and hope Acceptance Past experiences Comparison to others Faith Living in the present
External resources	Relationship with partner Family and friends Support organizations Medical literature and parental blogs Society

Predictions of future coping and QOL reframed

Most of the parents interviewed had been told by the medical team that their baby was at risk for a certain degree of developmental impairment in the future and this often ranged from mild to severe. When imagining a poor developmental outcome, most parents assumed that it might negatively impact QOL, but they did not believe that QOL was uniquely determined by developmental status. They talked about the possibility that their child may have a good QOL despite being severely impaired. Parents predicted that they, and their child, would be able to use QOL modifiers and coping mechanisms to deal with future illness and developmental problems in order to improve the QOL that might otherwise be associated with a poor developmental status. Numerous examples illustrate these predictions of coping: *“Even if he has some kind of paralysis, there are sports he can do anyway. Or maybe, he will like studying more. I don’t know. It will be his choice. But both parents will really have to be present. (...) I will really have to, at each stage of his life, follow well the development that he needs to have. So now, currently, we concentrate on the fact that he must grow, gain weight and his lungs must get better. This is the stage presently, with also, you know, exercises, (...) Then, when ... when he’ll be at home, there will be the adaptation to home as well. You know, it’s...it will really be, I think, it will be, stages to follow, little by little. (...) Everybody will have to adapt around James. We have developed so much, something beautiful together, that no matter what happens, he’s my child”* (Interview #6). Another mother said, *“I think he will learn to live with his own situation, and then he’ll succeed in finding happiness in that. I think that you adapt only to your situation and then, if you’re like that, you’re like that. He will only know his own situation,*

like me, I only knew mine. (...) The child, you give him a wheelchair and he will live with it (...), he won't have the choice anyway. He will have to accept it. It's not like somebody who has always walked, and who becomes handicapped. If the person never walks, in a way, he never knew this. For him, his normality, is the wheelchair" (Interview #8); "for sure we will not let (him) down, anyway, (...). It's accompanying him and giving him the care needed ... the best quality of life possible, in his situation... we will not hesitate, (...) to accompany him in this. (...) It will be an adaptation" (Interview #2). These predictions of coping led to the construction of a theme of Reframed QOL, or R-QOL (Figure 1, p. 45). Predictions of R-QOL illustrate a state superior to the poor QOL that could have been associated with impairment, in the absence of coping mechanisms. Interestingly, parental predictions of R-QOL did not go as far as to equate it to the QOL typically associated with a good developmental outcome. It is rather an intermediary state, more desirable than poor QOL, but not quite as good as the QOL parents believed "normal" children had. This idea was constructed from data obtained from most interviews. On the other hand, two of the parents diverged from the group in their discourse about the future and did not spontaneously envision a possible R-QOL. In fact, they did not predict resilience for their baby or themselves, which supports the notion that coping predictions are needed to envision R-QOL. Their belief was that illness and a poor developmental outcome were tightly linked with a bad QOL. When further probed during the interview, these mothers allowed for the theoretical possibility of other factors influencing QOL, such as the support from family or friends. Despite this, their belief was that development and QOL were closely linked and they had great difficulty envisioning a good future for their baby if the developmental state did not evolve positively. One said, *"for me, being ill, it's not*

having a good quality of life” (Interview #5); the other mentioned, “I couldn’t conceive that our children might not be able to follow us on the bike, to have a normal life” (Interview #10). These mothers seemed to still be actively grieving and showed few of the coping mechanisms described above. They were not able to accept their baby’s illness and the possibility of poor outcome in the future. They had a high degree of fear and little hope for the future and questioned the medical team whether the current management was in the best interest of the baby and of the family. They were worried about pursuing treatments that might prolong their baby’s suffering unnecessarily and mentioned that they had questioned the medical team about palliative care. One of them recalls, “then, (...) he extubated himself. He had to be urgently re-intubated. Then, I went to see the doctor and I told him: <<It’s becoming too difficult. At a certain point, what... you know, where are we going with him>>? And then, I told him... I said <<I find that it’s futile treatment>>” (Interview #5). In a similar scenario, the other mother describes, “we were finding that maybe it was futile treatment, while we, basically, we didn’t want these children anymore, because we were so scared” (Interview #10).

DISCUSSION

Summary of findings

The main findings of this study indicate that prognosis of developmental outcome influences parental predictions of QOL; however, this is not sufficient to fully determine

it. QOL is a multidimensional construct, of which developmental outcome is only one element. QOL, as estimated by the participants in this study, has an objective facet in addition to the subjective. Uncertainty is a central theme dominating parental discourse when making future predictions. Parents used a variety of coping mechanisms in dealing with the adversity caused by the current illness and its uncertain outcomes. Parents who believed that they, and their child, would be able to adapt to an eventual poor developmental outcome were able to predict a reframed QOL. This R-QOL was described as superior to the poor QOL parents typically associated with a poor developmental outcome, but not quite as good as the QOL believed to accompany good developmental outcomes.

Quality of life

The results reinforce the QOL definitions found in the literature and add further insight into the difficulties of QOL estimations and predictions. The World Health Organization describes quality of life as an **individual's perception** of his/her place in life, in the context of the culture and value system and in relation to his/her goals, expectations, standards and concerns [9]. With the purpose of simplification and facilitation of research, the concept of health-related quality of life (HRQOL) has also been used [10]. HRQOL refers to the aspects of quality of life that are related to and influenced by one's health. Through these definitions, and others found in the literature [15], QOL is portrayed as a personal and multifaceted construct. This subjectivity and complexity is reflected in the results of our study. Parents describe QOL as being quite personal and

difficult to estimate for another person. The complexity of the construct is seen in the parental description of the different factors that have an influence on QOL, in addition to the developmental status. These include: awareness of self and of environment, presence or absence of pain, personality, family presence, support groups and the society. Of note, results show that parents also saw an objective dimension in QOL. They associated QOL with the ability to fulfill age appropriate roles, be autonomous and independent. They indicated that good development would be more likely to lead to a good QOL while developmental impairment might influence QOL in a negative way. It is intriguing that no parents mentioned the possibility of having a poor QOL, despite a good developmental outcome, when, in reality, many people experience this situation on a regular basis. This close association of developmental state and QOL is not very surprising since it is common even in the medical field [16]. The parental discourse may have reflected personal beliefs or have been influenced by discussions with medical professionals. The literature however, does not support this association of physical health and QOL. Repeatedly, studies of QOL asking individuals with physical impairments to rate their own QOL fail to show differences compared to controls. Saigal *et al* revealed that even though adults born ELBW reported more functional limitations in cognition, sensation, mobility and self care than controls, there was no difference in the reported HRQOL in comparison with controls [17]. Similarly, a review of HRQOL of preterm children concluded that they have poorer health than normal birth weight children, but despite this, they do not perceive their quality of life as different from their healthier counterparts [18]. Albrecht and Devlieger have described the “*disability paradox*” in their qualitative study in which 153 people with disabilities were interviewed [11]. Their

study showed that despite their physical impairments, many participants experienced a high QOL. This discrepancy between our study and the literature is in part explained by different methodologies used in other studies and the fact that the objective component of QOL is probably more related to health status measures than to true QOL, or even HRQOL. Also, it reflects the difficulty parents had in estimating and predicting QOL for their children, because familial and individual QOL are so closely linked. In paediatrics, this creates challenges since young children cannot give their subjective impression of QOL, and we rely on parental estimations. These are likely to be based on objective observations or be more indicative of the parental or familial QOL. The literature shows that the correlations between parental proxies and children are imperfect [19]. In a systematic review, Eiser and Morse concluded that parents are better at estimating the physical domains of HRQOL than the social or emotional domains [20]. Riis shows that healthy controls significantly underestimate the well-being of hemodialysis patients, when asked to imagine having kidney disease [8]. Despite the limitations of proxy estimation of QOL, our study was designed to reflect the reality as much as possible. In the NICU, important decisions are made by parents and medical professionals, and are based largely on predictions of future QOL. For this reason, it is important that health care professionals remain aware of these limitations when prognosticating and fully explore parental visions of how developmental outcomes would impact future QOL

Reframed quality of life and coping

The concept of R-QOL resulted from the parents' ability to predict future coping strategies for themselves and for their baby. In the context of neonatal prognostication,

this novel concept has not been described in the literature before and its practical implications are extensive. Many health practitioners assume that predicting neurodevelopment status allows estimating future QOL [21]. However, as the results of this study indicate, assessing the presence and strength of coping mechanisms might be a more accurate way of predicting QOL. This premise would undoubtedly benefit from further investigation with larger, longitudinal and hypothesis testing studies.

The literature on coping and adaptation is vast, but few studies are specific to the neonatology environment [22-24]. In the adult literature, Antonovsky has described the concept of “*sense of coherence (SOC)*” based on “*generalized resistance resources,*” which facilitate successful coping with the inherent stressors of human existence [25, 26]. When confronted with a stressor, a person with a high sense of coherence is motivated to cope (meaningfulness), understands the challenge (comprehensibility) and believes that resources to cope are available (manageability). In a systematic review, Eriksson identified several studies that used Antonovsky’s SOC to predict quality of life [27]. These studies involved mostly adult patients with psychiatric illness, coronary heart disease, ischemia and hip fractures. The review found that family SOC was strongly and positively related to QOL in families who had at least one member with a serious illness. In addition, the family SOC was the largest predictor of family QOL. Many of the themes that resulted from our analysis are similar to those found in Antonovsky’s SOC. The baby’s illness and hospitalisation, as well as the uncertainty of survival and long-term prognosis are identified as stressors. The motivation to cope with these stressors comes from the strength of the attachment bond parents have formed with their baby. When projecting

into the future, they also find meaningfulness in the developmental prognosis by interpreting it in the context of the resulting QOL. Parents demonstrated comprehensibility of the illness through their extensive knowledge of diagnoses, complications and treatments. They understood their baby's condition and the possible prognosis by communicating with the medical team or by researching the medical literature and Internet blogs. Finally, the "*generalized resistance resources*" used to deal with the stressors correspond to the large variety of coping mechanisms described in the *internal and external resources* sections of this study. The extent to which Antonovsky's concepts are reflected in the results of this study is very compelling and suggests that the questionnaire derived from SOC studies could potentially be used with parents of babies hospitalised in the NICU [26, 28]. A limitation of this questionnaire would be, again, the difficulty of estimating SOC for another person.

Uncertainty

When parents made predictions of QOL, the inherent uncertainty of the prognosis was also revealed as a central theme. Mishel has done extensive work in explaining how people cope when their illness has an uncertain prognosis [29, 30]. Her model shows how the patient interprets uncertainty either as a danger or as an opportunity, mobilizing different coping strategies in each case [29]. This is reflected in our study, where some parents felt great anxiety about the possibility of a poor outcome and they expressed a strong wish to know with more certainty what the future had in store. Important decisions of active treatment pursuit or withdrawal were likely to follow if the

prognosis were more certain. Others were more prone to think prognosis could still be modified. They assessed evolution one day at the time and focussed on treatment and stimulation in order to maximize the baby's developmental potential. These later parents had a more positive outlook of the future and held the belief that they had an important role to play in shaping the future of their baby. Exploring how parents interpret uncertainty could help health care professionals counsel parents more effectively and support families in finding useful coping mechanisms.

CONCLUSIONS AND IMPLICATIONS FOR PRACTICE

In the NICU, professionals and parents strive to engage in a joint decision-making process in order to establish a management path that is in the best interest of the baby. Many issues make this process very challenging for families who are already undergoing intense stress caused by the baby's illness. Prognostication of future developmental outcomes is very difficult to establish with certainty at such young ages due to the immaturity of the neonatal brain and the important influence that environment has on maximizing function. Predicting future QOL further complicates prognostication with its complexity and subjectivity. Parents interpret QOL in the context of their own background and values, and no two people are identical. Health care professionals and other proxies often interpret QOL differently from parents. Also, current projections might differ from how parents and their grown children will actually experience QOL in the future. Humans have a great capacity to cope and to adapt to adversity, so they can

perceive their new reality in better terms than previously predicted, as well as more positively than estimated by others. Therefore, in addition to prognosticating about future outcomes, it is the professionals' role to help parents explore what these might mean in the context of their own lives. Functioning in an interdisciplinary fashion involves all the important members of the team and allows for adequate assessment of these aspects, even in an intensive care unit where time is often lacking. This allows medical teams to understand which of the available management options will be acceptable to the family. Parents should be encouraged to find mechanisms to cope with the current difficulties and to consider what resilience tools they can use to overcome future challenges if the poor prognosis turns out to be accurate. This exercise might change the way parents predict QOL for their baby and their family and it might ultimately influence what decisions they make in the present.

The uncertainty inherent in prognostication at such a young age should be openly discussed. In situations where treatment withdrawal is not imminently considered and uncertainty is high, focussing uniquely on the possibility of a negative prognosis might affect parental attachment and bonding with the child and reduce their ability to cope with the current illness. Reframing uncertainty as an opportunity for the parent to become involved in developmental stimulation and maximize long term potential can foster empowerment and a more secure attachment.

Finally, the health care professional's responsibility goes far beyond that of simple outcome prognostication, and instead, includes listening, exploring, understanding and

supporting parents through the challenging experiences of hospitalization of their child in the NICU and subsequent decision-making.

This study, as well as the review of literature, has allowed for the creation of a practical tool for NICU clinicians. It includes key elements to remember when establishing a therapeutic relationship with the family of an ill newborn with the purpose of achieving optimal shared decision-making. They are outlined below.

1. When difficult discussions are conducted, ensure: a private environment; a limited number of key professionals; important support people for the family.
2. First establish a partnership with the parents, fostering mutual trust and respect; refer to the baby by name when possible and avoid medical jargon; tolerate silence and allow adequate time for discussion and questions.
3. Enquire about parental preferences for information (amount, format) and provide personalized and balanced information about the baby's anticipated medical challenges; acknowledge and respond to the parents' main concerns and expectations, frequently ask for feedback and verify that the information provided has been adequately understood.
4. When prognosticating about survival and outcomes, openly discuss the inherent uncertainty involved; explain the limitations of using statistics in

predicting outcomes for an individual baby and counsel parents about the role of the environment and its influence on future development.

5. When discussing possible outcomes, and impact on quality of life, be aware of the potential for framing information according to personal opinions, attitudes or biases; give parents the opportunity to explore the effect of these possible outcomes in the context of their personal values and familial/cultural environment; encourage them to consider what role coping mechanisms might have on their ability to manage possible outcomes and on the future quality of life.
6. Explore parental emotions, validate them and help parents navigate the decision-making process without ignoring their feelings.
7. If parents indicate that they have “changed their minds”, explore reasons thoroughly (time permitting); clearly document any change to the treatment plan and its rationale; promptly update the other members of the treating team (neonatologist on call, obstetrician, etc.).
8. When appropriate, introduce palliative care early, as complementary to medical care and focused on symptom control, rather than simply as “nothing else can be done.”

9. Explain that the benefits and burdens of particular therapies will probably need to be revisited after birth and during their stay in the NICU, based on the clinical trajectory of their child and in light of their demonstrated response to therapy.

10. After a death, team members who had been involved in the decision-making process should offer parents a meeting to review the events surrounding the death, answer questions and help with the bereavement process.

REFERENCES

1. Lorenz JM, Wooliever DE, Jetton JR, Paneth N: **A quantitative review of mortality and developmental disability in extremely premature newborns.** *Archives of Pediatrics & Adolescent Medicine* 1998, **152**(5):425-435.
2. Watts JL, Saigal S: **Outcome of extreme prematurity: as information increases so do the dilemmas.** *Archives of Disease in Childhood Fetal & Neonatal Edition* 2006, **91**(3):F221-225.
3. Lantos JD, Meadow WL: **Neonatal Bioethics: The moral challenges of medical innovation.** Baltimore: The Johns Hopkins University Press; 2006.
4. Hack M, Fanaroff AA: **Outcomes of children of extremely low birthweight and gestational age in the 1990s.** *Seminars in Neonatology* 2000, **5**(2):89-106.
5. Stoll BJ, I HN, Bell EF, Shankaran S, Laptook AR, Walsh MC, Hale EC, Newman NS, Schibler K, Carlo WA *et al*: **Neonatal Outcomes of Extremely Preterm Infants From the NICHD Neonatal Research Network.** *Pediatrics* 2010, **126**:443-458.
6. Fontana M, Farrel C, Gauvin F, Lacroix J, Janvier A: **Modes of Death in Pediatrics: Differences in the Ethical Approach in Neonatal and Pediatric Patients** *The Journal of Pediatrics* 2013, **162**:1107-1111.
7. Saigal S: **Perception of health status and quality of life of extremely low-birth weight survivors. The consumer, the provider, and the child.** *Clinics in Perinatology* 2000, **27**(2):403-419.
8. Riis J, Baron J, Loewenstein G, Jepson C, Fagerlin A, Ubel PA: **Ignorance of Hedonic Adaptation to Hemodialysis: A Study Using Ecological Momentary Assessment.** *Journal of Experimental Psychology* 2005, **134**(1):3-9.
9. **Programme on mental health: WHOQOL user manual** [http://www.who.int/mental_health/resources/evidence_research/en/index.html]. Last accessed 2015-02-10
10. Testa MA, Simonson DC: **Assessment of Quality-of-Life Outcomes.** *N Engl J Med* 1996, **334**(13):835-840.
11. Albrecht G, Devlieger P: **The disability paradox: high quality of life against all odds.** *Soc Sci Med* 1999, **48**:977-988.

12. Jensen MP, Moore MR, Bockow TB, Ehde DM, Engel JM: **Psychosocial factors and adjustment to chronic pain in persons with physical disabilities: a systematic review.** *Archives of Physical Medicine & Rehabilitation* 2011, **92**(1):146-160.
13. Elwyn G, Frosch D, Thomson R, Joseph-Williams N, Lloyd A, Kimmersley P, Cording E, Tomson D, Dodd C, Rollnick S *et al*: **Shared Decision Making: A Model for Clinical Practice.** *J Gen Intern Med* 2012, **27**(10):1361-1367.
14. Glaser BG, Strauss AL: **The Discovery of Grounded Theory: Strategies for Qualitative Research.** Chicago: Aldine Publishing Company; 1967.
15. Saigal S, Rosenbaum P: **What matters in the long term: reflections on the context of adult outcomes versus detailed measures in childhood.** *Seminars In Fetal & Neonatal Medicine* 2007, **12**(5):415-422.
16. Knowles R, Griebisch I, Dezateux C: **Quality of life and congenital heart defects: comparing parent and professional values.** *Arch Dis Child* 2007, **92**(5):388-393.
17. Saigal S, Stoskopf B, Pinelli J, Streiner D, Hoult L, Paneth N, Goddeeris J: **Self-perceived health-related quality of life of former extremely low birth weight infants at young adulthood.** *Pediatrics* 2006, **118**(3):1140-1148.
18. Donohue PK: **Health-related quality of life of preterm children and their caregivers.** *Mental Retardation & Developmental Disabilities Research Reviews* 2002, **8**(4):293-297.
19. Robitail S, Simeoni M-C, Ravens-Sieberer U, Bruil J, Auquier P, for the KG: **Children proxies' quality-of-life agreement depended on the country using the European KIDSCREEN-52 questionnaire.** *Journal of Clinical Epidemiology* 2007, **60**(5):469-478.
20. Eiser C, Morse R: **Can parents rate their child's health-related quality of life? Results of a systematic review.** *Qual Life Res* 2001, **10**(4):347-357.
21. Saigal S, Stoskopf BL, Feeny D, Furlong W, Burrows E, Rosenbaum PL, Hoult L: **Differences in preferences for neonatal outcomes among health care professionals, parents, and adolescents.** *JAMA* 1999, **281**(21):1991-1997.
22. Affleck G, Tennen H: **Appraisal and Coping Predictors of Mother and Child Outcomes after Newborn Intensive Care.** *Journal of Social and Clinical Psychology* 1991, **10**(4):424-447.

23. Doering LV, Moser DK, Dracup K: **Correlates of anxiety, hostility, depression, and psychosocial adjustment in parents of NICU infants.** *Neonatal Network - Journal of Neonatal Nursing* 2000, **19**(5):15-23.
24. Lau R, Morse CA: **Parent's coping in the neonatal intensive care unit: a theoretical framework.** *Journal of Psychosomatic Obstetrics & Gynecology* 2001, **22**(1):41-47.
25. Antonovsky A: **The salutogenic model as a theory to guide health promotion.** *Health Promotion International* 1996, **11**(1):11-18.
26. Antonovsky A: **The Structure and Properties of the Sense of Coherence Scale.** *Soc Sci Med* 1993, **36**(6):725-733.
27. Eriksson M, Lindstrom B: **Antonovsky's sense of coherence scale and its relation with quality of life: a systematic review.** *J Epidemiol Community Health* 2007, **61**:938-944.
28. Thomson G, Moran VH, Axelin A, Dykes F, Flacking R: **Integrating a sense of coherence into the neonatal environment.** *BMC Pediatrics* 2013, **13**(84):1-8.
29. Mishel M: **Uncertainty in illness.** *Journal of Nursing Scholarship* 1988, **4**:225-232.
30. Mishel M: **Reconceptualization of the Uncertainty in Illness Theory.** *Journal of Nursing Scholarship* 1990, **22**(4):256-262.

**CHAPTER 5: RESULTS NOT INCLUDED IN THE JOURNAL
ARTICLE INTENDED FOR SOCIAL SCIENCE AND MEDICINE (*SOC
SCI MED*)**

This chapter presents the results derived from the study that were not included in the journal article intended for publication in the Social Science and Medicine (Soc Sci Med). These results are more related to the parental experience of the current hospitalization. They include themes of *illness impact* (comprising themes of *Grief* and *Attachment*) and a parental perspective of the *shared decision-making process*.

Impact of illness

Given the exploratory purpose of the interviews, a great deal of discourse freedom was allowed at the beginning of the encounter. The large majority of parents spontaneously provided a detailed narrative of the events leading up to the current hospitalization. They demonstrated great familiarity with their infant's diagnoses, the ensuing complications and the different tests and treatments required. Many used medical terms and acronyms, despite no previous knowledge of medical jargon. This indicated a high level of involvement in the care of their baby and acquired familiarity over time in a highly technical environment.

The parents described the effects of illness and hospitalization on their baby and themselves. Description of impact on the child focussed mostly on pain and physical discomfort resulting from procedures and incubator confinement. The main factors impacting parents were fatigue; decreased personal, couple and family time; isolation and loneliness; and financial difficulties. In families with older children, the siblings

missed their parent(s); when the latter spent long periods in hospital, the siblings were jealous or misbehaved to gain the parent's attention. In addition to these findings, much was revealed about the emotional impact of the illness on the parents.

Grief

Participants described the evolution of their emotional reactions to the losses suffered throughout the pregnancy, delivery, and NICU hospitalization. The losses were multiple: loss of healthy pregnancy and well newborn, loss of future healthy child, modified family projects, impact on personal life, etc. The emotional reactions were diverse. Parents described shock when receiving bad news, denial to themselves and to others, fear and anxiety, as well as deep sadness. One mother recalls, *"I was like <<I don't want to hear anything 'cause she's not getting delivered right now and don't tell me anything>>, 'cause I was really (...) scared (...). Maybe I was trying to block it off, like, this is not happening to my baby or something"* (Interview #3). Another parent describes her initial reactions: *"for us, having a premature baby was out of the question. (...) We were dreaming that (the pregnancy) was going to continue...yes, so, a lot of denial"* (Interview #10). Some reported feeling shame about having a sick child. Others felt guilty for somehow causing the illness: *"I have the guilt (...) that still haunts me often, because (...) I wasn't able to carry the (pregnancy) long enough (...). I envy the people I see (...) pregnant"* (Interview #9). Some experienced anger or felt they were objects of injustice. The powerlessness and discomfort with the lack of control were prevalent: *"you know, we feel destitute. We feel, (...) from the beginning, that we don't have control"* (Interview #9). Mixed with these

feelings, positive reactions were also described. Parents reported feeling love and affection for their newborn, empathy with the baby's suffering, as well as pride and optimism when progress occurred.

During data analysis, a general theme of **grief** in response to various losses best explained this mixture of feelings. Figure 2 (*Annex IV, p. xxi*) includes grief in the previously presented diagram of theme relationships. As the interviews captured only snapshots in time of what is typically a long and complex process, each parent seemed to be at a different point in the course of their grieving response. Some expressed predominant feelings of shock, sadness and anger, while others had achieved, at least partially, a certain acceptance.

The grieving process was complex. Because the babies were still hospitalized, parents did not have a clear, well-defined loss, which they could accept and from which they could move on. Multiple or successive losses happened throughout the course of the hospitalization and others were still in sight: *"each time, there was mourning (...). Again, again, again a difference; again (...) different babies. Then, for two months, we always had big blows; you know, when it wasn't one, it was the other, when it wasn't both. We had no good news. (...) So at a certain point, you say: I don't want anymore, I don't want anymore"* (Interview #10). The uncertainty of short-term survival and long term outcomes prevented some parents from even knowing what to grieve: *"yes, we were sad because we knew that probably he was going to have something. But we don't know what, so (...) we can't say: <<oh, we have to mourn this, or mourn that>>. We don't know. So I can't say:*

<<oh, he will never walk>>. Maybe he'll walk. <<Oh, maybe he'll never go to school>>. We don't know, maybe he will. (...) So it's difficult to say <<well, ok, I have mourning to do. It's this. He'll never do this his whole life. Ok. It makes me sad>>. (...) We don't know what to mourn. We don't even know **if** we need to mourn... it's difficult" (Interview #8).

Attachment

Most parents formed a strong attachment with their child, but talked at length about the difficulties they encountered. Many found exclusion from routine care like feeding and bathing to be difficult and often felt like observers, especially when the child was unstable. One mother mentioned feeling that her baby belonged to the NICU more than to her: "For me, he's a NICU baby. For now, he's not.... Yes, he's my son, but I... he doesn't really belong to me. He's not my baby. He's ...(...) the nurses' baby" (Interview #5).

The inability to be close to their baby at all times, the fear of infections, the incubator with its alarms, and the lack of privacy were among the identified barriers to forming a strong attachment bond. One participant recalls " (I was) more restrained a little, because in fact, we had the barrier all the time. I could touch him, but I couldn't hold him, I couldn't feed him. (...) But as soon as (...) they took a few of the wires away, (...) they let us take him freely, then for sure it helped a lot" (Interview # 8).

One mother, who had been extremely involved in the care of her baby and who frequently helped with procedures, mentioned sometimes feeling like a nurse and

becoming habituated to seeing painful procedures. She felt attached to her baby, but was afraid of eventually becoming less sensitive to her child's suffering: *"In the beginning, the nurses were performing techniques on my child and I wasn't really used to it. (...) Often, when it was intense, you know, often I had a hard time keeping myself from crying. (...) It was weird because with time, you know, I was watching these techniques, and it was as if it had become normal (...). It wasn't as intense as at the beginning because I had gotten used to it (...) ... as if...not that it didn't bother me, but it bothered me less than at the beginning. I didn't like this (Interview #6).*

For other mothers, witnessing their children's severe illness, believing they were suffering and being afraid they would soon die led them to feel detached. They believed a disabled child would have a poor QOL and the possibility of poor long-term prognosis prevented them from feeling strongly attached to their babies: *"For me, now, I'm starting to.... like, detach myself from him because... for me, he's suffering and it's not what I want for my child" (Interview #5).* One mother revealed being present in the NICU out of duty, and not because she felt attachment. She said *"I was very, very, very ambivalent, because the attachment was very long, very, very, very, very long. But it's as if I was going every day, out of duty. (...) I was making sure to come to the medical rounds, I was trying to follow the evolution. And it's as if I was always telling myself: <<ah, in the worst case, we can unplug him>>. Then, when (the doctor) said: <<well, we can't unplug him anymore>> – because he was extubated, then, it meant, I felt really trapped" (Interview # 10).* In this mother's case,

feeling “trapped” seemed to be related to the improvement of her baby’s illness before she had been ready to accept having a child with potential long-term neurodevelopmental impairments. From her discourse, it did not appear that discussions about withdrawing artificial nutrition had been considered appropriate by the medical team.

Shared decision-making process

Parents shared their experience of the decision-making process in a variety of situations. These ranged from important moments, such as deciding whether to proceed with a potentially risky eye surgery, to less stressful ones like agreeing to routine care. The decision-making process included the following sub-themes: *health care professional and parental roles; communication; best interest*; this last sub-theme was discussed in the context of uncertainty, and was already described in Chapter 4, *Uncertainty and Best interest (p.46)*.

Health care professional roles

Parents described health care professionals as specialists in the medical issues of their baby. Certain levels of competence and knowledge, as well as the ability to give an “objective” opinion were expected. Since all the infants in the NICU were ill in varying degrees, parents trusted that the medical team had the experience to identify the

difference between “typically ill,” such as a premature baby who needs ventilator support for his/her immature lungs and the “critical” level of illness of a child at risk of imminent death. Parents also presumed that experience allows medical teams to make predictions of usual illness evolution and typical health and developmental outcomes. They expected professionals to know what usually happens to children with similar diagnoses in the long-term and to help estimate future QOL based on the well-being of older children who grew up with the developmental outcomes predicted for their baby. One parent found it useful that her doctor put in perspective the different degrees of cerebral palsy, ranging from barely noticeable to severe impairment, and stressed the importance of the environment and the developmental stimulation provided by parents. *“You know, for me, <<cerebral palsy>> was really big. It meant almost, handicapped (...) (The resident) said <<with cerebral palsy, you could have people in front of you and you might not even realize they have cerebral palsy>>” (Interview #6).*

Parental roles

Many of the parents saw themselves as the main advocates for their baby. They were frequently present at the bedside and were concerned with only one baby (or two, in the case of the twins). They often provided continuity in a unit where teams of doctors and nurses changed frequently. Over time, they acquired a large amount of medical knowledge and were able to actively participate in treatment decisions. In addition, parents stressed their role in predicting what values will be important in the long term and how the baby’s condition and future impairments might integrate in the family: *“Of*

course, I don't have a nursing diploma, but I know my son and I know what is good for him and what is not." The participant added, "I am here so much of the time, that I am able to advise a doctor, a nurse, in a way that what I say makes sense" (Interview #6).

Some parents talked about their duty to bear the long-term consequences of the current decisions and felt this gave them the ultimate responsibility of agreeing with the management proposed by the medical team. One mother recalls telling a nurse, *"you treat him now, but (...) after, I will live with him my whole life."* She added, *"I question all the time... what we do to him now, what will be the impact for him, later. (...) Giving morphine, yes, ... the nurse will have an easier time, maybe, changing his diaper or performing his routine care (...) because he will be calmer. But giving morphine now, what will be the impact (...) on his brain, on his development (...). The dexamethasone, it's similar. We give it to him now, to extubate, to take away the inflammation. But what will be the impact for him later? In fact, that has always been my questioning" (Interview #5).*

Communication

Communication between the medical team and the parents was an essential theme emerging from the interview process. Parents appreciated being promptly informed of test results and regularly updated about their baby's state. Establishing a trusting and respectful relationship seemed dependent on the particular team consulting and staying open to parental opinion. Parents wanted the team to communicate honestly, with compassion and empathy. One parent recalls the initial meeting with the medical team:

“The doctor we saw at that moment, she made me... made me enthusiastic despite everything, despite the situation, in the way she presented things, that it had gone well and that she was breathing” (Interview #9).

In certain cases, counselling on outcomes and management options before delivery of a pre-term baby was completely absent, despite the time availability: *“All that day I spent here with monitors, (...) hours, only seeing a nurse that came to check on things ... Now, I regret that nobody came to talk to me about this. What is a premature baby of 26 weeks (...) and what were the options I had, when (...) they hadn't yet done my Caesarian. (...) because, sincerely, (...) had they explained all this to me, I ... I would have said <<No. We let it be natural>> (Interview #5).* The lack of antenatal consultation and the feeling of loss of control triggered regret, resentment and seemed to damage this mother's trust in the medical team. Other parents reported meeting with the neonatologist, but only after birth or only to discuss the short-term issues pertaining to the neonatal period, leaving out long-term concerns. Exclusion from team discussions at rounds with parents present was also an intermittent problem. Parents expressed frustration when lack of continuity led to conflicting messages from different team members. In one case, this was particularly difficult since doctors had diverging visions of what was in the best interest of the child. This led to some recommending continued active care, and others advising palliative care, creating confusion and anguish for the parents. Lastly, although they understood the teaching mission of the hospital, some parents reported instances when trainees were clumsy in their communication, worrying parents unnecessarily or focusing on unimportant details.

The additional results presented in this chapter contribute to better illustrate the relationship between illness, predicted QOL and coping mechanisms. The parent encounters were performed during a very difficult time, when their child is still gravely ill and there is tremendous uncertainty about the future. The grieving process, an important theme in the parental discourse, seemed to be the catalyst that uncovered the multitude of coping mechanisms used in this stressful situation. Reflecting on the ways they dealt with loss and uncertainty allowed parents to identify the various coping mechanisms they were using in the present. Then, when asked to make projections for the future, the parents who had strong coping mechanisms in the present predicted that they, and their child will be able to cope in the future as well. This led to the projection of a re-evaluated QOL by these parents. For other parents, the extreme and complex grief seemed to impede them from feeling attached to their baby and from forecasting a positive future. Those mothers who were still actively grieving the illness of their child had the most negative views of the future QOL of their child and were contemplating the option of withdrawing active treatment. This insight has tremendous implications for practice, suggesting that helping parents with the grieving process may impact their vision of their child's future. The parental perspectives of shared decision-making helped better define the influence medical teams have on how families view the QOL of their child. Most parents looked to health professionals for guidance, counseling and support while they desired an active role in decision-making. They felt responsible to ensure their child's best interest is maximized and had great difficulty when they felt they lacked control or were not empowered. In fact, one of the mothers who were mostly afraid of

the future expressed that a trusting relationship between her and the team had not been established. This illustrates how forming solid alliances with caregivers can have positive impacts on many aspects of the hospitalization, prognostication and decision-making.

This chapter has described themes that are key to the understanding of the dynamic relationship between the illness of a child hospitalized in the NICU, the prognostic of QOL and the adaptation process that results. Grieving their baby's illness, forming attachment and navigating the medical system, while bearing the responsibility of important treatment decisions, are tremendous challenges for the NICU parents and their babies. The next chapter aims to further examine the results of this study in the context of the existing literature and to outline practical recommendations for clinicians operating in the NICU.

**CHAPTER 6: GENERAL DISCUSSION AND PRACTICAL
RECOMMENDATIONS FOR HEALTH CARE PROFESSIONALS**

Summary of findings

The purpose of this thesis was to qualitatively examine the parental perspective on the future QOL of their child, during the hospitalisation in the NICU. In order to achieve this goal, several steps were necessary. First, an initial literature review of neurodevelopment and QOL led to further examining other related topics, such as uncertainty, adaptation and shared decision-making. Throughout this exercise, there was realization that the literature on the complex feature of prognosticating QOL for a child, whose parents are yet unable to know his or her preferences and values, was very limited. This led to the motivation of designing a study to investigate how parents project the future QOL of their ill neonate. The qualitative method of grounded theory using parental interviews was considered the most appropriate approach for studying the research question and for fully capturing the cognitive and emotional aspects of the process of parental prognostication.

The main findings of our research indicate that the prognosis of developmental outcome influences parental predictions of QOL; however, this is not sufficient to fully determine it. QOL is a multidimensional construct, of which developmental outcome is only one part. Uncertainty is a central theme dominating parental discourse when making future predictions. Parents used a variety of coping mechanisms in dealing with the adversity caused by the current illness and its uncertain outcomes. Parents who believed that they, and their child, would be able to adapt to an eventual poor developmental outcome were able to predict a reframed QOL. This reframed QOL was described as superior to the QOL

parents typically associated with a poor developmental outcome, but not quite as good as the QOL believed to accompany good developmental outcomes.

Secondary findings revealed the grief process ensuing in response to the losses experienced by parents during pregnancy, delivery and NICU hospitalization. This grief process is complicated by uncertainty and multiple losses happening over time. The NICU environment and the outcome uncertainty are important barriers to parental-child attachment. Furthermore, insight is gained on the parental perspectives of the shared decision-making process.

This next section will elaborate a general discussion of the main themes of neurodevelopmental outcomes, uncertainty, QOL and adaptation and examine their tight, interconnected relationship. In abbreviated form, these notions have been discussed in the article intended for Social Science and Medicine. In this section, the discussion will build on previous arguments, interpret the results in further depth and make additional links with the literature. Since they were not included in the previous article, the themes of grief and attachment will be examined more closely and discussed in this section. Finally, the process of shared decision-making will be examined, and practical solutions for clinicians faced with the challenges arising from prognostication will be presented.

Neurodevelopmental Outcomes

When projecting future outcomes in this study, parents tended to dichotomize development into two categories: good outcomes, characterized by an ability to function in a variety of domains; and poor outcomes, indicating various incapacities. Although parents were aware of the uncertainty surrounding how good or how poor the outcomes would become, the reality of the wide spectrum of development was not very evident. Given that life has many colors and shades, developmental ability is a continuous scale even among typically functioning people. Strengths and weaknesses vary widely between individuals, with unique traits, who together form a diverse collective. The participants' focus on the extremes of the developmental spectrum informs us of the process of projection that parents use, possibly influenced by medical professionals. When describing a spectrum, it is sometimes easier for professionals to define the limits of "best and worst case scenario" and then allow parents to make projections within these limits [3]. Although defining these boundaries places prognosis within a specific context, emphasis still needs to be placed on the wide variety of outcomes within that range, which are collectively much more likely to occur than the absolute extremes.

When faced with the difficult task of prognosticating about an individual child, professionals often use risk factors; these may include the degree of prematurity, birth weight and additional complications with potential impact on neurologic function, such as brain haemorrhage, retinopathy of prematurity or hearing loss, etc. [103]. These risk factors help establish a prognosis by comparing the given child with evidence from the

literature regarding outcomes of other children with similar risk factors. To quote one example, research may show 100 out of 1000 (10%) premature children develop cerebral palsy [104]. There are two major pitfalls that health care professionals and parents must beware of when considering this number. One is the difficulty to identify, early in life, which 100 children will eventually go on to develop this complication. As previously mentioned, statistics do not predict individual outcomes, and for each unique child, a particular outcome may or may not materialize. Professionals must take great care in quoting statistics when counselling parents, since it has been shown that many people have great difficulty interpreting risk and percentages when dealing with health issues [105]. The second pitfall in using statistics for prognosis is the manner in which outcomes are defined in research. Studies may report cerebral palsy as a severe outcome, without differentiating its degree of significance or the level of function that the child achieved. In fact, many children with cerebral palsy function very well, with no or minimal aid, and are very well integrated in their environment. When counselling parents early in the life of their newborn, it is important to discuss these aspects. Many parents do not have the experience with illness that professionals have and need guidance in interpreting statistics and prognosis. This is notion is important due the common link often made between developmental outcome and future QOL. As discussed before, the two concepts are not equivalent. Even if development can influence QOL, prognosticating is so difficult that professionals should beware projecting outcomes with too much assurance and should discuss the inherent uncertainty of prognostication. Practical tools on how to counsel parents will be discussed further in the shared decision-making section. The large spectrum of developmental possibilities and the

difficulties of interpreting risk and statistics lead to a high level of uncertainty early in the life of a sick neonate.

Uncertainty

Even though it was not the initial focus of the study, the uncertainty of the future was a prevalent theme, tightly intertwined with prognostication. Understanding how parents dealt with the uncertainty of the outcomes and analyzing this process in the context of the literature was a fascinating exercise. As discussed in the literature review, studies demonstrate that living with uncertainty is very difficult for most. Suicide rates in patients with incomplete spinal cord injury are higher than in patients with complete tetraplegia [70]. Similarly, depression scores before undergoing HIV testing are higher than the scores recorded after the results are known, regardless of the diagnosis [71]. This indicates that uncertainty is sometimes more difficult to manage than the diagnosis of a severe illness. In our study, parents often expressed how challenging it was for them to live in a state of uncertainty. They also revealed that the uncertainty of short-term survival was a barrier to their ability to project into the future. This is not a surprising finding, as parents often used the “living in the present” coping mechanism to deal with uncertainty.

In instances when parents were able to make future projections, the emotions of hope and fear were almost always intertwined (*Figure 2, Annex IV, p. xxi*). Parents hoped for

good outcomes but feared they would not materialize; they dreaded the extremes of poor prognosis but hoped they would be prevented. This combination of hope and fear has been noted by other investigators [106]. It also reflects Mishel's model of uncertainty interpretation as danger or opportunity, previously described in the literature review. This way of experiencing uncertainty was reflected in our findings. When talking about the possibility of a poor outcome, some parents expressed a great deal of distress. They coped with this possibility by trying to reduce the unknown as much as possible. In doing so, they acquired as much information as they could through discussions with the health care team, or through personal research in books or on the internet. For these parents, additional knowledge decreased the uncomfortable feeling of uncertainty. It is important that professionals realize that uncertainty happens at many other levels, in addition to the diagnosis and the prognosis [107]. For example, the unfamiliar environment of the NICU can cause parents significant anxiety and helping them acclimatise to the setting can be comforting. Promptly updating families about recent developments, meaning of medical terms, planned investigations and current treatments are ways to help parents navigate the hospitalization process and ease the unfamiliarity [108]. On the other hand, some parents in our study had a different attitude about the unknown prognosis. They interpreted uncertainty as an opportunity because they believed outcomes could still be influenced. These parents talked about taking things one day at the time and desired to get fully involved in developmental stimulation, in order to maximize the baby's potential. They had a more positive outlook of the future, and took very seriously their role in shaping the future of their baby.

Health care professionals must be aware of the effect that uncertainty has on parents. Reducing it, as much as possible, can help decrease stress and anxiety. When uncertainty cannot be further diminished, helping parents and professionals allow its presence, understand it and regard it as an opportunity to act on the child's development may lead to feelings of empowerment and improved coping mechanisms. The ability to deal with illness and uncertainty can have an important role in improving QOL.

Quality of life

This study's findings on QOL are also very illuminating. The literature's description of QOL as a complex and subjective construct is reflected in our results. As discussed previously (*p. 64*), the objective aspect of QOL seems to be more indicative of the functional or health status typically measured on HRQOL tools [100]. Additionally, some of the parental association of "good" development with "good" QOL and "poor" outcomes with "poor" QOL, is not what the literature supports. In fact, as detailed in the literature review (*p. 13*), many patients who experience physical or intellectual impairments have a good QOL, and non-impaired proxies typically underestimate their well-being. This discrepancy with the literature can be explained by the mental processes that healthy individuals (in this case parents), use to imagine what life would be like for their children, if they grew up with a physical or mental impairment. One such mental process is provided by Ubel's "focussing illusions," introduced in the literature review (*p. 16*). Solomon offers another noteworthy perspective of these mental processes in his

extensive study of human identity “Far from the tree” [109]. According to Solomon, many people interpret parenting primarily as a vertical transmission of identity from one generation to the next. Attributes and values such as ethnicity, skin pigmentation, language, nationality, etc. are vertically transmitted and form the basis of an identity fostering a sense of acceptance and belonging in a family or a cultural group. Deaf children born to deaf parents also experience such a transmission of identity through a common physical trait, even though society considers this trait to be an impairment. When children with physical or mental impairments are born to able-bodied parents, they acquire an identity that is horizontal and unfamiliar to their parents. Another example is that of gay and gender fluid individuals; since most have heteronormative parents, sexual orientation and gender identity often generate a horizontal identity. In his work, Solomon interviewed hundreds of parents of children with horizontal identities and he described their challenges, as well as the growth resulting from their experiences. He found that most parents struggle with the idea of having a different child, but with time, many find positive aspects that enrich their life and that of their families. Solomon argues that acceptance of a child with such different characteristics from their parents requires understanding and adaptation over time, through building a strong attachment between parent and child. When done adequately, this in turn builds the child’s self-acceptance and resilience [109]. Based on the literature and the results of our study, it is interesting to note that the subjective part of QOL is in fact, an interpretation regarding life circumstances, rather than a direct consequence tightly linked to the conditions of health at and around birth.

When considering the QOL modifiers and future coping mechanisms, most parents in our study were able to envision a reframed QOL, even if poor developmental outcomes occurred. In fact, they were projecting resilience for themselves and their child. In her work, Mishel creates fascinating links between the fields of psychology and physics using the chaos theory [73]. She builds on the work of Prigogine and Stengers, who demonstrated that most systems include subsystems that are in constant flux and that large fluctuations can shatter the organization of the system, sending it past a certain threshold, into chaos [110]. Subsequent to such an event, a pattern of self re-organization into a new equilibrium occurs, with eventual stabilization in a state that may be very different from the original. Mishel makes parallels between this model and the way humans respond to severe adversity. When illness and uncertainty invade multiple aspects of a person's life with a disruptive impact, then the individual is sent into a state far from equilibrium. Eventually (especially if the illness and the uncertainty continue), a new view of life emerges, where lessons are learned, priorities are re-evaluated, and growth occurs [73]. Many of the parents in our study, being aware of an adaptive process, or having learned about their strengths from past experiences, were able to maintain hope for the future and especially, to hope for future resilience. As former Czech president Vaclav Havel is known to have said, "hope is not the conviction that something will turn out well, but the certainty that something makes sense regardless of how it turns out" [111]. In addition to representing a feeling or an expectation, hope is also a mobilizing force. It is a process-oriented complex of many thoughts, feelings and actions. It is often a response to a threat that results in planning and using internal and external resources to achieve a desired goal. This aspect of

mobilizing people into action contributes to resilience and empowerment [112, 113].

Further discussion on these themes follows below.

Adaptation

The adaptation themes in our study are also seen in other studies [37, 114-116] and in Antonovsky's concept of Sense of Coherence. As previously mentioned in the Discussion section of Chapter 4 (*p. 65*), the illness, the hospitalisation and the uncertainty of the future are important stressors for parents and they trigger the need for coping mechanisms through comprehensibility, meaningfulness and manageability. Thomson et al. used the principles of SOC to propose an approach destined to empower parents in the NICU [102]. The authors argue that in order to maximize comprehensibility, professionals must help parents understand the stressors by conveying clear information presented in an organized fashion. As we found in our study as well, ongoing and consistent communication that involves both listening to parent's concerns and keeping them informed is necessary. Meaningfulness is interpreted as the extent to which the demands are worthy of investment. Promoting parent-child bonding and fostering a strong and secure attachment will provide the illness experience with meaning and help parents feel more invested in the care of their child. Manageability refers to the degree to which generalised resistance resources help the parent deal with the tension created by the stressors. In our study, these GRRs are reflected in the internal and external resources that parents used to cope. Internal resources included positivity

and hope, acceptance, faith, and living in the present. Externally, family and friends, support organisations and parental groups were particularly helpful.

Furthermore, an appropriate balance between underload and overload is necessary [102]. Inappropriate underload reflects situations where parents are reduced to simple visitors and do not feel sufficiently involved in the care of their child. On the other hand, examples of overload include instances when parents do not trust the medical team to administer adequate care and feel the need to be constantly present in order to “supervise” the team. Several examples of these situations were found in our study, with some mothers feeling like visitors, while others felt they needed to verify everything. Adequate balance of parental “load” can be achieved by involving parents as partners in a joint decision-making process, as discussed in a later section. As grief and attachment were important themes that influenced how parents viewed the future of their baby, they are discussed below.

Grief and Attachment

Although not part of the primary objective, acute grief was revealed in our study as a complex reaction to a variety of losses, mainly relating to health and personal (or familial) projects.

Grief is a complex process that is unique to each individual and can take various periods of time to complete. It exhibits a range of physical, emotional, intellectual, social, spiritual and occupational manifestations [117, 118]. In the later stages of grief, an adjustment occurs when parents find ways to reorganize their life and adapt to the changes that triggered the process [117]. Most of the parents in our study were still in various stages of acute grief. Parents described many difficult emotions, including shock, sadness, anger, as well as positive feelings of love, affection and empathy. Some of these emotions had been experienced in the recent past, some were being felt during the interview, and others were anticipated, in response to future losses. These early reactions are part of the initial steps of adaptation and they represent how people deal with intense feelings. They are part of the daily coping process necessary for eventual adjustment [117]. In our study, the parents who talked more about adjustment and acceptance seemed to have greater ease focussing on the future. They were likely moving towards the later stages of grief, where they had begun to adapt to the changes they had experienced. According to Dyer, a new equilibrium, a re-organization of life and an integration of the loss eventually occurs in people who complete the mourning process [117]. This view of grief also reinforces the earlier discussion on adaptation and reframed QOL (*p. 64*).

Grief and attachment are closely linked. A longitudinal study explored the association between mothers' unresolved grief and infant-mother attachment security in 74 preterm infants [119]. The study found a relative risk of 1.59 (95% CI: 1.03-2.44) for insecure attachment development, when mothers had feelings of unresolved grief relating to the preterm birth experience. The results also indicated that maternal grief resolution and

interaction quality independently predicted mother-child attachment security. In our study, two of the mothers who seemed to be actively grieving, and who had the most fearful view of the future, also described not feeling very securely attached to their babies.

In another relevant qualitative study of 25 mothers, Bialoskurski analysed the nature of attachment in the NICU [120]. The results showed that the attachment process was not automatic and mothers were often delayed in their attachment to their premature infants. Obstacles included a small and fragile appearance of the baby, immature behaviour of the infant, as well as physical barriers that the child or maternal health introduced. The mothers also used the delay in attachment as a coping mechanism to protect themselves from the extreme emotions that might occur in the case of the baby's death. Our study also confirmed these barriers to attachment. Particularly telling was the discourse of the mothers who thought their babies were really ill and suffering, and who questioned the appropriateness of treatment pursuit. They felt less attached to their children and one even mentioned feeling "trapped" when extubation was no longer possible. The medical team should be aware of these barriers, explore them with each individual parent, and support the establishment of a strong attachment between parent and child. In order to do so, strong and trusting relationships between parents and health care professionals must be established and maintained through a real partnership. This has often been presented as a shared decision-making process.

Shared decision-making and practical solutions

The literature on joint decision-making advocates for the establishment of a partnership between clinicians and parents, with the purpose of selecting a management course that is founded on both evidence-based clinical information, as well as the family's values and priorities [121]. In this study, parental perspectives of the shared decision-making process confirmed aspects already found in the literature [122]. Parents saw doctors as the specialists in their baby's diagnoses and treatment options, as well as their risks and benefits. Participants wanted their doctor to be knowledgeable and competent and have the ability to guide them through the various management choices. At the same time, participants felt the parents knew their baby best and could judge how the treatment options would fit with their family and life circumstances, in the long term. They wanted guidance from the team, but they believed parents were the most competent advocates for the best interest of their child. Moreover, since they will bear the long-term consequences of the decisions taken at the moment, some parents felt that the ultimate responsibility of agreeing with a treatment course fell to them.

To adequately share the decision-making process, parents and health care professionals must be able to share information, to recognise one another's expertise and to share responsibility for joint decisions [121]. When working in a field with a level of mortality and morbidity as high as neonatology, professionals must have solid communication skills and an adequate knowledge of human psychology and behaviour in times of stress.

Firstly, adequate communication fosters a relationship based on trust, where parents feel respected and empowered [123]. In this study, the antenatal consultation was revealed to be an important way of informing and preparing parents to the short and long term effects of a complicated delivery. When an antenatal consultation hadn't taken place, it created a significant amount of regret and resentment and it negatively influenced the parent's relationship with the medical team. In practice, for adequate communication to occur, the setting must be suitable [124]. A private environment free of interruptions should be arranged. The physician should invite a limited number of key professionals (bed side nurse, social worker, etc.) and the role of each should be clearly explained. If the family has identified important support people, they can be included in the discussions as well. Professionals should refer to the baby by name when possible and avoid medical jargon. They should tolerate silence and allow adequate time for discussion and questions. When opening the discussion, physicians can first enquire about the parents' preferences for information and provide personalized and balanced information about the baby's anticipated medical challenges. Professionals should ascertain and respond to the parents' questions, frequently ask for feedback and verify that the information provided has been adequately understood [122].

Secondly, health care professionals should explore the parents' main concerns, expectations, past experiences, values and family and life context [124]. Involving all the important members of the team in an interdisciplinary fashion allows for satisfactory

assessment of these personal aspects, even in an intensive care unit where time is precious. This approach helps medical teams fully explore and understand which management options are acceptable to the family and which are not. At the same time, it gives parents the opportunity to explore what the projected outcome means for them and their family, in the long term. When taking into account their own values and life situation, parents may vary widely in the way they judge the future QOL of their baby or might have a different view from the health care team. In this case, professionals should be aware of their personal values and biases and ensure that these don't inappropriately influence patients during counselling. Physicians sometimes "frame", often without realizing, the information based on their own preferences for a particular outcome, which may not necessarily coincide with the patient's choice [125]. Health professionals can aim to help parents identify and use coping mechanisms to deal with the illness of their baby. In addition, they can encourage parents to consider how these coping tools will lead to adaptation and resilience in the future and how this will help them deal with an undesirable outcome. This exercise might change the way parents predict QOL for their baby and their family and it might ultimately influence what decisions they make in the present.

Furthermore, the uncertainty inherent in prognostication at such a young age should also be openly discussed. Understanding how parents interpret uncertainty, as a threat or as an opportunity, can guide medical teams in their counselling. In situations where treatment withdrawal is not imminently considered and uncertainty is high, focussing uniquely on the possibility of a negative prognosis might affect parental attachment and

bonding with the child and reduce their ability to cope with the current illness.

Reframing uncertainty as an opportunity is an essential way for the parent to become involved in developmental stimulation and maximize long term potential. This can empower parents and allow them to regain some feelings of control over an overwhelming situation. Involving parents in the developmental care of their baby can be achieved by practicing adequate patient and family centered care [126].

Parents' emotions may play a critical role in their interpretation of their child's best-interests [124]. During counselling, physicians should make efforts to explore parental emotions, validate them and help parents navigate the decision-making process while using both their intellect and their feelings.

Palliative care specialists often play an important role in counselling and supporting families of very ill babies. However, when palliation discussions are introduced, sometimes families feel the medical team is giving up on treating their child. To prevent this, palliative care discussions should occur early in the course of a deteriorating patient and be introduced as complementary to medical care and focussed on symptom control, rather than simply as "nothing else can be done" [127]. Families experiencing particularly challenging situations, whose infants are offered an approach based on comfort care, should be offered the opportunity to meet with a palliative care team [128]. After a death, team members who had been involved in the decision-making process should offer parents a meeting to review the events

surrounding the death, answer questions and help with the bereavement process [129].

Finally, the joint decision process is a continuum that allows respect for the parents' desired level of participation and accountability in making decisions for their baby. Health care professionals must respect the level of autonomy parents desire and adapt to a wide range of preferences [121]. In other words, the doctor-patient relationship, as described in the literature review, must be adequately tailored to each case. Some parents might want to be very involved in making decisions, while other prefer to leave more of the responsibility to the medical team [130]. In the latter case, professionals should ensure that parents truly agree with the management and understand its implications instead of merely passively acquiescing to suggestions [131]. The medical team must support families in their decision-making process; this may include making a treatment recommendation that is in keeping with the family's expressed values and sharing in the moral accountability for that decision. A practical tool for difficult decisions in neonatology has been developed with the help of this research project (Table V). The tool summarizes the key messages that professionals should remember when counselling parents of premature or significantly ill babies. These recommendations are largely based on the results stemmed from this study, although the existing literature served to further reinforce and polish them into a practical tool.

Table V: Practical tool for difficult decisions in neonatology

Environment	<p>When difficult discussions are conducted, a private environment free of interruptions should be arranged. The physician should invite a limited number of key professionals (bed side nurse, social worker, etc.) and the role of each should be clearly explained. If the family has identified important support people, they can be included in the discussions.</p>
Relationship	<p>Physicians should first aim to establish a partnership with the parents, fostering development of mutual trust and respect. They should refer to the baby by name when possible and avoid medical jargon. They should tolerate silence and allow adequate time for discussion and questions.</p>
Parental concerns	<p>Physicians should enquire about the parents' preferences for information (amount, format) and provide personalized and balanced information about the baby's anticipated medical challenges. They should ascertain and respond to the parents' main concerns and expectations, frequently ask for feedback and verify that the information provided has been adequately understood.</p>

<p>Uncertainty</p>	<p>When prognosticating about survival and outcomes, physicians should openly discuss the inherent uncertainty involved. They should explain the limitations of using statistics in predicting outcomes for an individual baby and counsel parents about the opportunity offered by the influence of environment on future development.</p>
<p>Quality of Life and Coping</p>	<p>Physicians should be cognizant of the potential for <i>framing</i> information about possible outcomes and their impact on quality of life according to their personal opinions, attitudes or biases, and do their best to avoid it.</p> <p>When discussing quality of life, physicians should give the parents the opportunity to explore the effect of these possible outcomes in the context of their own personal values, as well as their familial and cultural environment. Parents should be encouraged to consider what role coping mechanisms might have on their ability to manage possible outcomes and on the future quality of life.</p>
<p>Emotions</p>	<p>Parents' feelings may play a critical role in their interpretation of their child's best interests. During counselling, physicians should make efforts to explore parental emotions, validate them and help parents navigate the decision-making process while using both their intellect and their feelings.</p>

<p>Care plan changes</p>	<p>If parents indicate that they have “changed their minds”, the reasons behind such changes should be explored thoroughly (time permitting) not simply unquestioningly accepted. When there is a change to the treatment plan, these changes and the rationale for them must be clearly documented and the other members of the treating team (neonatologist on call, obstetrician, etc.) promptly updated.</p>
<p>Palliative care</p>	<p>Palliative care discussions should occur early and be introduced as complementary to medical care and focused on symptom control, rather than simply as “nothing else can be done.” Families experiencing particularly challenging situations, whose infants are offered a palliative care approach, should be offered the opportunity to meet with a palliative care team.</p>
<p>Ongoing assessment</p>	<p>The counselling process should be ongoing and parents should be explained that the benefits and burdens of particular therapies will probably need to be revisited after birth and during their stay in the NICU, based on the clinical trajectory of their child and in light of their demonstrated response to therapy.</p>

Bereavement	After a death, team members who had been involved in the decision-making process should offer parents a meeting to review the events surrounding the death, answer questions and help with the bereavement process.
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CONCLUSIONS

This thesis aimed to study the parental perspective of the future QOL of ill babies, while they are hospitalized in the NICU. An initial review of literature of the most relevant concepts identified the difficult challenges that parents and professional faced when projecting into the future of their baby. Medical teams try to prognosticate about future developmental outcomes, but these are very difficult to establish with certainty. The neonatal brain is still immature and has great potential for improvement, when developing in a stimulating environment. Each child is unique, and statistics do not help predict which patient will develop impairments, nor what their degree might be. Trying to associate the possible developmental impairments with future QOL further complicates prognostication, because QOL is personal and is influenced by many factors that are unrelated to physical health. The literature also showed that the role of adaptation to illness is very important in individuals who experienced impairments. Humans have a great capacity to cope and to adapt to adversity, so they can perceive their new reality in better terms than previously predicted, as well as more positively than estimated by others. This makes QOL very difficult to predict and to estimate for another person. Health care professionals and other proxies can interpret QOL differently from parents. Also, current projections might differ from how parents and their grown children will actually experience QOL in the future. Therefore, trying to determine the course of management in the best interest of the baby is very challenging for parents and health professionals.

To better understand how parents see the future QOL of their baby, a research project was conducted, using parental interviews in order to gain insight into their perspectives.

Grounded theory was used to analyse the data and construct a coherent model integrating the various themes elicited by the research.

This project showed that uncertainty of short-term survival and long-term outcomes was an important theme for the parents of the NICU. The roller coastal nature of the illness and not knowing whether their child will survive made it difficult for parents to project too far into the future. When able to think about the long-term outcomes, some parents felt a great deal of anxiety, while others interpreted uncertainty as an opportunity to stimulate their child and to maximize future potential. This high level of uncertainty exposed a spectrum of parental beliefs concerning the best interest of the baby. Some parents thought it was best for all possible treatments to be attempted, others wanted to avoid too much suffering, while some were torn between the different choices. Grieving multiple losses related to the baby's illness was an important theme for parents. The uncertainty of the survival and outcomes complicated the process and prevented parents from "moving-on." Most parents were able to form strong attachment relationships with their baby. Caregivers who expressed the least amount of coping mechanisms had the weakest feelings of attachment.

When making predictions about developmental outcomes, parents focused on the extremes of the abilities spectrum. A good outcome meant having the capacity to perform activities of daily living, being intelligent and experiencing only minor problems such as mild developmental delays, mild cerebral palsy or mild school difficulties. At the

other end of the range, a bad developmental outcome was represented by inability to perform such activities and extreme severity of problems such as cerebral palsy, cognitive impairment or autism. Quality of life was revealed as a complex construct with a subjective, as well as an objective dimension. The subjective aspects were related to the parental belief that their child must be happy and enjoy life in order to have a good QOL. This subjectivity also meant that the QOL of the child is difficult to estimate by caregivers. Parents also had great difficulty in separating the QOL of the child from their own, which indicated the interdependence of the two. The objective dimension of future QOL was related to the functional status of the child, as observed by parents. This functional status was characterized by the fulfillment of age appropriate roles, such as being autonomous, having friends, going to school, being employed, etc. The developmental prognosis influenced parental predictions of QOL, however many other factors were contributory (personality, family, friends, support groups and societal norms). Parents used various coping mechanisms to deal with the current illness and the ensuing uncertainty. Some of the resources were personal, such as personality type, the ability to maintain an attitude of positivity and hope, acceptance of their child's illness, use of lessons from past experiences, comparison with sicker patients, faith, as well as focusing on the present. Other coping mechanisms included external factors like one's relationship with the partner, family, friends or support organizations. A key and innovating finding of this study was revealed by the **relationship between coping skills and future QOL**. When imagining the future adversity of a developmental impairment, parents who believed that they, and their child, would have the ability to cope, predicted a reframed QOL. This reframed QOL was greater than the QOL parents supposed children

with a poor developmental outcome had, but was not as good as the QOL assumed to accompany good developmental outcomes. The notion of a **reframed QOL**, fostered by identifying and predicting coping mechanisms is very interesting, due to its great potential for clinical application. With better understanding of the processes parents use to project into the future, clinicians can work to empower them.

Perspectives of the joint decision-making process revealed that parents expected health care professionals to be objective, knowledgeable about long-term outcomes and QOL, and have the ability to guide parents in their decisions. Parents viewed themselves as the main advocates for their baby, who bear the ultimate responsibility of the decisions made. Open, honest communication between parents and the medical team was very important and several barriers were identified.

A practical tool for difficult decisions in neonatology was constructed based on the insights gained from this study and on the literature review. Recommendations are made about how to best prepare the setting for difficult discussions and how to establish a trusting and respectful relationship with parents. The appropriate style of communication is described, as well as the most important themes to discuss: prognostication about survival and outcomes, uncertainty, interpretations of QOL and parental feelings. Recommendations about appropriate documentation, introduction of palliative care and bereavement counseling are made.

Health professionals can use this tool to improve their skills in shared decision-making. They should aim to fully understand the parental perspectives, concerns and values and enable parents to consider useful resilience tools that may help them overcome future challenges, if the poor prognosis turns out to be accurate. Aiding parents to interpret uncertainty as an opportunity to maximize developmental potential will also empower them to get involved in the routine care of their child, will foster a more secure attachment and help parents face an extremely challenging situation with a greater sense of control.

This project sets the stage for future research avenues on QOL with parents of ill neonates. Specifically, the study will continue using the acquired knowledge to create a mixed methods questionnaire for parents of babies hospitalized in the NICU. This questionnaire will aim to use qualitative and quantitative methods to characterize how parents view future QOL and to identify their coping mechanisms. Eventually, the parents interviewed in this project will be re-interviewed when their children have reached the preschool age. This longitudinal approach involving two qualitative phases of interviews with the same parents will allow for a true analysis of the adaptation process to illness over time and will test the accuracy of the predictions made in the NICU.

In sum, this thesis advocates for health professionals working in neonatology to remain aware of the limitations of prognostication, of the shortcomings of equating developmental impairments with QOL and of their responsibility to enable parents to

create, find and use coping mechanisms during their child's hospitalization. They should strive to adequately apply the principles of shared decision-making, acquire good communication skills and understand the common psychological processes associated with illness.

REFERENCES

1. Lorenz JM, Wooliever DE, Jetton JR, Paneth N: **A quantitative review of mortality and developmental disability in extremely premature newborns.** *Archives of Pediatrics & Adolescent Medicine* 1998, **152**(5):425-435.
2. Watts JL, Saigal S: **Outcome of extreme prematurity: as information increases so do the dilemmas.** *Archives of Disease in Childhood Fetal & Neonatal Edition* 2006, **91**(3):F221-225.
3. Lantos JD, Meadow WL: **Neonatal Bioethics: The moral challenges of medical innovation.** Baltimore: The Johns Hopkins University Press; 2006.
4. Caughey A, Robinson J, Norwitz E: **Contemporary diagnosis and management of preterm premature rupture of membranes.** *Reviews in Obstetrics and Gynecology* 2008, **1**(1):11-12.
5. Synnes A, Buchanan L, Ruth C, Albersheim S: **Management of the newborn delivered at the threshold of viability.** *BCMJ* 2008, **50**(9):498-508.
6. Chan K, Ohlsson A, Synnes A, Lee DS, Chien LY, Lee SK, Canadian Neonatal N: **Survival, morbidity, and resource use of infants of 25 weeks' gestational age or less.** *American Journal of Obstetrics & Gynecology* 2001, **185**(1):220-226.
7. Ho S, Saigal S: **Current Survival and Early Outcomes of Infants of Borderline Viability.** *Neoreviews* 2005, **6**:e123-e132.
8. Gluckman P, Wyatt J, Azzopardi D, Ballard R, Edwards A, Ferriero D, Polin R, Robertson C, Thoresen M, Whitelaw A *et al*: **Selective head cooling with mild systemic hypothermia after neonatal encephalopathy: multicentre randomised trial.** *Lancet* 2005, **365**:663-670.
9. Shankaran S, Laptook AR, Ehrenkranz RA, Tyson JE, McDonald SA, Donovan EF, Fanaroff AA, Poole WK, Wright LL, Higgins RD *et al*: **Whole-body hypothermia for neonates with hypoxic-ischemic encephalopathy.** *N Engl J Med* 2005, **353**(15):1574-1584.

10. van der Bom T, Zomer AC, Zwinderman AH, Meijboom FJ, Bouma BJ, Mulder BJM: **The changing epidemiology of congenital heart disease.** *Nat Rev Cardiol* 2011, **8**(1):50-60.
11. Hack M, Fanaroff AA: **Outcomes of children of extremely low birthweight and gestational age in the 1990s.** *Seminars in Neonatology* 2000, **5**(2):89-106.
12. Stoll BJ, I HN, Bell EF, Shankaran S, Laptook AR, Walsh MC, Hale EC, Newman NS, Schibler K, Carlo WA *et al*: **Neonatal Outcomes of Extremely Preterm Infants From the NICHD Neonatal Research Network.** *Pediatrics* 2010, **126**:443-458.
13. Hack M, Flannery DJ, Schluchter M, Cartar L, Borawski E, Klein N: **Outcomes in young adulthood for very-low-birth-weight infants.** *N Engl J Med* 2002, **346**(3):149-157.
14. Hack M: **Young adult outcomes of very-low-birth-weight children.** *Seminars In Fetal & Neonatal Medicine* 2006, **11**(2):127-137.
15. Hack M: **Adult outcomes of preterm children.** *Journal of Developmental & Behavioral Pediatrics* 2009, **30**(5):460-470.
16. Jacobs S, Hunt R, Tarnow-Mordi W, Inder T, Davis P: **Cooling for newborns with hypoxic ischaemic encephalopathy.** *Cochrane Database of Systematic Reviews* 2007(4).
17. Payot A, Gendron S, Lefebvre F, Doucet H: **Deciding to resuscitate extremely premature babies: how do parents and neonatologists engage in the decision?** *Soc Sci Med* 2007, **64**(7):1487-1500.
18. Vohr BR, Wright LL, Dusick AM, Mele L, Verter J, Steichen JJ, Simon NP, Wilson DC, Broyles S, Bauer CR *et al*: **Neurodevelopmental and functional outcomes of extremely low birth weight infants in the National Institute of Child Health and Human Development Neonatal Research Network, 1993-1994.** *Pediatrics* 2000, **105**(6):1216-1226.
19. Davis DW: **Cognitive outcomes in school-age children born prematurely.** *Neonatal Network - Journal of Neonatal Nursing* 2003, **22**(3):27-38.

20. Bhutta AT, Cleves MA, Casey PH, Cradock MM, Anand K: **Cognitive and behavioral outcomes of school-aged children who were born preterm: A meta-analysis.** *JAMA: Journal of the American Medical Association* 2002, **288**(6):728-737.
21. Miatton M, De Wolf D, Francois K, Thiery E, Vingerhoets G: **Neuropsychological performance in school-aged children with surgically corrected congenital heart disease.** *J Pediatr* 2007, **151**(1):73-78, 78.e71.
22. Fontana M, Farrel C, Gauvin F, Lacroix J, Janvier A: **Modes of Death in Pediatrics: Differences in the Ethical Approach in Neonatal and Pediatric Patients** *The Journal of Pediatrics* 2013, **162**:1107-1111.
23. Payot: **The Quality of Life of Young Children and Infants with Chronic Medical Problems: Review of Literature.** *Curr Probl Pediatr Adolesc Health Care* 2011, **41**:91-101.
24. Dickinson HO, Parkinson KN, Ravens-Sieberer U, Schirripa G, Thyen U, Arnaud C, Beckung E, Fauconnier J, McManus V, Michelsen SI *et al*: **Self-reported quality of life of 8-12-year-old children with cerebral palsy: a cross-sectional European study.** *Lancet* 2007, **369**(9580):2171-2178.
25. Dinesen SJ, Greisen G: **Quality of life in young adults with very low birth weight.** *Archives of Disease in Childhood Fetal & Neonatal Edition* 2001, **85**(3):F165-169.
26. Gray R, Petrou S, Hockley C, Gardner F: **Self-reported health status and health-related quality of life of teenagers who were born before 29 weeks' gestational age.** *Pediatrics* 2007, **120**(1):e86-93.
27. Jozefiak T, Larsson B, Wichstrom L, Mattejat F, Ravens-Sieberer U: **Quality of Life as reported by school children and their parents: a cross-sectional survey.** *Health & Quality of Life Outcomes* 2008, **6**:34.
28. Saigal S, Feeny D, Rosenbaum P, Furlong W, Burrows E, Stoskopf B: **Self-perceived health status and health-related quality of life of extremely low-birth-weight infants at adolescence.** *JAMA* 1996, **276**(6):453-459.

29. Saigal S, Stoskopf B, Pinelli J, Streiner D, Hoult L, Paneth N, Goddeeris J: **Self-perceived health-related quality of life of former extremely low birth weight infants at young adulthood.** *Pediatrics* 2006, **118**(3):1140-1148.
30. Verrips E, Vogels T, Saigal S, Wolke D, Meyer R, Hoult L, Verloove-Vanhorick SP: **Health-related quality of life for extremely low birth weight adolescents in Canada, Germany, and the Netherlands.** *Pediatrics* 2008, **122**(3):556-561.
31. Saigal S: **Perception of health status and quality of life of extremely low-birth weight survivors. The consumer, the provider, and the child.** *Clinics in Perinatology* 2000, **27**(2):403-419.
32. WHO: **Programme on mental health: WHOQOL user manual.** Geneva, Switzerland; 1998. Last accessed 2015-02-10
http://www.who.int/mental_health/resources/evidence_research/en/index.html
33. Testa MA, Simonson DC: **Assessment of Quality-of-Life Outcomes.** *N Engl J Med* 1996, **334**(13):835-840.
34. Albrecht G, Devlieger P: **The disability paradox: high quality of life against all odds.** *Soc Sci Med* 1999, **48**:977-988.
35. Jensen MP, Moore MR, Bockow TB, Ehde DM, Engel JM: **Psychosocial factors and adjustment to chronic pain in persons with physical disabilities: a systematic review.** *Archives of Physical Medicine & Rehabilitation* 2011, **92**(1):146-160.
36. Brasfield CD: **An exploration of the stressors, coping resources, and resiliency of rural mothers of children with special needs.** *Dissertation Abstracts International: Section B: The Sciences and Engineering* 2008, **69**(1-B):665.
37. Heiman T: **Parents of children with disabilities: Resilience, coping, and future expectations.** *Journal of Developmental and Physical Disabilities* 2002, **14**(2):159-171.

38. Lee SK, McMillan DD, Ohlsson A, Pendray M, Synnes A, Whyte R, Chien LY, Sale J: **Variations in practice and outcomes in the Canadian NICU network: 1996-1997.** *Pediatrics* 2000, **106**(5):1070-1079.
39. Morrison J: **Twin Gestation and Premature Birth.** *J Perinatol* 2005, **25**(1):1-3.
40. Reuner G, Hassenpflug A, Pietz J, Philippi H: **Long-term development of low-risk low birth weight preterm born infants: neurodevelopmental aspects from childhood to late adolescence.** *Early Human Development* 2009, **85**(7):409-413.
41. Shah P, Lee S, Yoon EW, Param V: **The Canadian Neonatal Network: Annual Report;** 2013. www.canadianneonatalnetwork.org. *Last accessed 2015-02-10*
42. Synnes A, Luu T, Moddemann D, Church P, Lee D, Vincer M, Ballantyne M, Majnemer A, Creighton D, McGuire M *et al*: **The Canadian Neonatal Follow-Up Network.** In: *Pediatric Academic Society.* Vancouver; 2014.
43. Zwicker JG, Harris SR: **Quality of Life of Formerly Preterm and Very Low Birth Weight Infants From Preschool Age to Adulthood: A Systematic Review.** *Pediatrics* 2008, **121**(2):e366-e376.
44. Saigal S, Stoskopf B, Streiner D, Boyle M, Pinelli J, Paneth N, Goddeeris J: **Transition of extremely low-birth-weight infants from adolescence to young adulthood: comparison with normal birth-weight controls.** *JAMA* 2006, **295**(6):667-675.
45. Saigal S, Rosenbaum P: **What matters in the long term: reflections on the context of adult outcomes versus detailed measures in childhood.** *Seminars In Fetal & Neonatal Medicine* 2007, **12**(5):415-422.
46. **The Quality of Life Model** [<http://www.utoronto.ca/qol/concepts.htm>]. *Last accessed: 2014-09-24*
47. Detmar SB, Bruil J, Ravens-Sieberer U, Gosch A, Bisegger C, European Kg: **The use of focus groups in the development of the KIDSCREEN HRQL questionnaire.** *Qual Life Res* 2006, **15**(8):1345-1353.

48. Erhart M, Ellert U, Kurth B-M, Ravens-Sieberer U: **Measuring adolescents' HRQoL via self reports and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument.** *Health & Quality of Life Outcomes* 2009, **7**:77.
49. Klassen AF, Lee SK, Raina P, Chan HWP, Matthew D, Brabyn D: **Health status and health-related quality of life in a population-based sample of neonatal intensive care unit graduates.** *Pediatrics* 2004, **113**(3 Pt 1):594-600.
50. Sagheri D, Ravens-Sieberer U, Braumann B, von Mackensen S: **An Evaluation of Health-Related Quality of Life (HRQoL) in a group of 4-7 year-old children with cleft lip and palate.** *Journal of Orofacial Orthopedics* 2009, **70**(4):274-284.
51. Horsman J, Furlong W, Feeny D, Torrance G: **The Health Utilities Index: concepts, measurement properties and applications.** *Health and Quality of Life Outcomes* 2003, **1**(54).
52. Rajmil L, Herdman M, Fernandez de Sanmamed M-J, Detmar S, Bruil J, Ravens-Sieberer U, Bullinger M, Simeoni M-C, Auquier P, Kidscreen G: **Generic health-related quality of life instruments in children and adolescents: a qualitative analysis of content.** *Journal of Adolescent Health* 2004, **34**(1):37-45.
53. Ravens-Sieberer U, Erhart M, Wille N, Wetzel R, Nickel J, Bullinger M: **Generic health-related quality-of-life assessment in children and adolescents: methodological considerations.** *Pharmacoeconomics* 2006, **24**(12):1199-1220.
54. Robitail S, Simeoni M-C, Erhart M, Ravens-Sieberer U, Bruil J, Auquier P, European Kidscreen G: **Validation of the European proxy KIDSCREEN-52 pilot test health-related quality of life questionnaire: first results.** *Journal of Adolescent Health* 2006, **39**(4):596.e591-510.
55. Saigal S: **Quality of life of former premature infants during adolescence and beyond.** *Early Human Development* 2013, **89**(4):209-213.
56. Donohue PK: **Health-related quality of life of preterm children and their caregivers.** *Mental Retardation & Developmental Disabilities Research Reviews* 2002, **8**(4):293-297.

57. Upton P, Lawford J, Eiser C: **Parent-child agreement across child health-related quality of life instruments: a review of the literature.** *Qual Life Res* 2008, **17**(6):895-913.
58. Robitail S, Simeoni M-C, Ravens-Sieberer U, Bruil J, Auquier P, for the KG: **Children proxies' quality-of-life agreement depended on the country using the European KIDSCREEN-52 questionnaire.** *Journal of Clinical Epidemiology* 2007, **60**(5):469-478.
59. Saigal S, Stoskopf BL, Feeny D, Furlong W, Burrows E, Rosenbaum PL, Hoult L: **Differences in preferences for neonatal outcomes among health care professionals, parents, and adolescents.** *JAMA* 1999, **281**(21):1991-1997.
60. Riis J, Baron J, Loewenstein G, Jepson C, Fagerlin A, Ubel PA: **Ignorance of Hedonic Adaptation to Hemodialysis: A Study Using Ecological Momentary Assessment.** *Journal of Experimental Psychology* 2005, **134**(1):3-9.
61. Middleton A, Hewison J, Mueller R: **Attitudes of deaf adults toward genetic testing for hereditary deafness.** *American Journal of Human Genetics* 1998, **63**(4):1175-1180.
62. Ubel PA: **You're Stronger than You Think.** New York: McGraw-Hill; 2006.
63. Neville KL: **Uncertainty in illness: an integrative review.** *Orthopedic Nursing* 2003, **22**(3):206-214.
64. McCormick KM: **A Concept Analysis of Uncertainty in Illness.** *Journal of Nursing Scholarship* 2002, **34**(2):127-131.
65. Wright L, Afari N, Zautra A: **The illness uncertainty concept: A review.** *Current Science Inc* 2009, **13**(2):133-138.
66. Santacroce SJ: **Measuring parental uncertainty during the diagnosis phase of serious illness in a child.** *Journal of Pediatric Nursing* 2001, **16**(1):3-12.
67. Hartkopp A, Bronnum-Hansen H, Seidenschnur AM, Biering-Sorensen F: **Suicide in a spinal cord injured population: its relation to functional status.** *Arch Phys Med Rehabil* 1998.

68. Perry S, Jacobsberg L, Fishman B: **Suicidal ideation and HIV testing.** *Jama* 1990, **263**(5):679-682.
69. Mishel M: **Uncertainty in illness.** *Journal of Nursing Scholarship* 1988, **4**:225-232.
70. Mishel M: **Reconceptualization of the Uncertainty in Illness Theory.** *Journal of Nursing Scholarship* 1990, **22**(4):256-262.
71. Affleck G, Tennen H: **Appraisal and Coping Predictors of Mother and Child Outcomes after Newborn Intensive Care.** *Journal of Social and Clinical Psychology* 1991, **10**(4):424-447.
72. Chehrazi A: **Healthy adaptation in parents of children with autism: Implications of personality and resilience.** *Dissertation Abstracts International: Section B: The Sciences and Engineering* 2002, **63**(2-B):1017.
73. Grotberg EH: **The International Resilience Research Project.** In: *International perspectives on human development.* edn. Lengerich, Germany: Pabst Science Publishers; Germany; 2000: 379-399.
74. Hawley DR, DeHaan L: **Toward a Definition of Family Resilience: Integrating Life-Span and Family Perspectives.** *Family Process* 1996, **35**(3):283-298.
75. Lau R, Morse CA: **Parent's coping in the neonatal intensive care unit: a theoretical framework.** *Journal of Psychosomatic Obstetrics & Gynecology* 2001, **22**(1):41-47.
76. Antonovsky A: **The Structure and Properties of the Sense of Coherence Scale.** *Soc Sci Med* 1993, **36**(6):725-733.
77. Antonovsky A: **The salutogenic model as a theory to guide health promotion.** *Health Promotion International* 1996, **11**(1):11-18.
78. Eriksson M, Lindstrom B: **Antonovsky's sense of coherence scale and its relation with quality of life: a systematic review.** *J Epidemiol Community Health* 2007, **61**:938-944.

79. Sprangers MA, Schwartz CE: **Integrating response shift into health-related quality of life research: A theoretical model.** *Soc Sci Med* 1999, **48**(11):1507-1515.
80. Taylor SE: **Adjustment to threatening events: A theory of cognitive adaptation.** *American Psychologist* 1983, **38**(11):1161-1173.
81. Elliott T, Renier C, Palcher J: **Chronic pain, depression, and quality of life: correlations and predictive value of the SF-36.** *Pain Med* 2003, **4**(4):331-339.
82. Woolf C: **Central sensitization: Implications for the diagnosis and treatment of pain.** *Pain* 2011, **152**(3 Suppl):S2-15.
83. Elwyn G, Frosch D, Thomson R, Joseph-Williams N, Lloyd A, Kimmersley P, Cording E, Tomson D, Dodd C, Rollnick S *et al*: **Shared Decision Making: A Model for Clinical Practice.** *J Gen Intern Med* 2012, **27**(10):1361-1367.
84. Gerber B: **The patient-physician relationship in the internet age: future prospects and the research agenda.** *J Med Internet Res* 2001, **3**(2):e15.
85. Orfali K, Gordon E: **Autonomy gone awry: a cross-cultural study of parents, experiences in neonatal intensive care units.** *Theor Med Bioeth* 2004, **25**(4):329-365.
86. Orfali K: **Parental role in medical decision-making: fact or fiction? A comparative study of ethical dilemmas in French and American neonatal intensive care units.** *Soc Sci Med* 2004, **58**(10):2009-2022.
87. Emanuel EJ, Emanuel LL: **Four models of the physician-patient relationship.** *Jama* 1992, **267**(16):2221-2226.
88. Baines P: **Medical ethics for children: applying the four principles to paediatrics.** *Journal of Medical Ethics* 2008, **34**(3):141-145.
89. Janvier A, Leblanc I, Barrington KJ: **The best-interest standard is not applied for neonatal resuscitation decisions.** *Pediatrics* 2008, **121**(5):963-969.

90. Jefferies A, Kirpalani H: **Counselling and management for anticipated extremely preterm birth.** *Paediatr Child Health* 2012, **17**(8):443.
91. Janvier A, Barrington KJ, Aziz K, Bancalari E, Batton D, Bellieni C, Bensouda B, Blanco C, Cheung PY, Cohn F *et al*: **CPS position statement for prenatal counselling before a premature birth: Simple rules for complicated decisions.** *Paediatr Child Health* 2014, **19**(1):22-24.
92. Glaser BG, Strauss AL: **The Discovery of Grounded Theory: Strategies for Qualitative Research.** Chicago: Aldine Publishing Company; 1967.
93. <http://www.macupdate.com/app/mac/8358/tams-analyzer>. *Last accessed: 2014-10-02*
94. Mays N, Pope C: **Qualitative research in health care. Assessing quality in qualitative research.** *Bmj* 2000, **320**(7226):50-52.
95. Morse JM: **A review committee's guide for evaluating qualitative proposals.** *Qual Health Res* 2003, **13**(6):833-851.
96. Neuman WL: **Social research methods: qualitative and quantitative approaches**, 3rd edn. Wisconsin: Allyn and Bacon; 1997.
97. Dorling JS, Field DJ, Manktelow B: **Neonatal disease severity scoring systems.** *Archives of Disease in Childhood - Fetal and Neonatal Edition* 2005, **90**(1):F11-F16.
98. Vincer M, Allen A, Joseph K, Stinson D, Scott H, Wood E: **Increasing prevalence of cerebral palsy among very preterm infants: a population-based study.** *Pediatrics* 2006, **118**(6):e1621-1626. Epub 2006 Oct 1630.
99. Visschers VHM, Meertens RM, Passchier WWF, De Vries NNK: **Probability Information in Risk Communication: A Review of the Research Literature.** *Risk Analysis* 2009, **29**(2):267-287.
100. De Graves S, Aranda S: **Living With Hope and Fear-The Uncertainty of Childhood Cancer After Relapse.** *Cancer Nursing* 2008, **31**(4):292-301
210.1097/1001.NCC.0000305745.0000341582.0000305773.

101. Hansen BS, RØRtveit K, Leiknes I, Morken I, Testad I, Joa I, Severinsson E: **Patient experiences of uncertainty – a synthesis to guide nursing practice and research.** *Journal of Nursing Management* 2012, **20**(2):266-277.
102. De Rouck S, Leys M: **Information needs of parents of children admitted to a neonatal intensive care unit: A review of the literature** *Patient Education and Counseling* 2009, **76**(2):159-173.
103. Eiser C, Morse R: **Can parents rate their child's health-related quality of life? Results of a systematic review.** *Qual Life Res* 2001, **10**(4):347-357.
104. Solomon A: **Far from the tree.** London: The Random House Group Limited; 2014.
105. Prigogine I, Stengers I: **Order out of chaos: Man's new dialogue with nature.** New York: Bantam Books; 1984.
106. Halpin D: **The Nature of Hope and its Significance for Education.** *British Journal of Educational Studies* 2001, **49**(4):392-410.
107. Amendolia B: **Hope and Parents of the Critically Ill Newborn: A Concept Analysis.** *Advances in Neonatal Care* 2010, **10**(3):140-144
110.1097/ANC.1090b1013e3181dee1083b.
108. Mednick L, Cogen F, Henderson C, Rohrbeck CA, Kitessa D, Streisand R: **Hope more, worry less: Hope as a potential resilience factor in mothers of very young children with type 1 diabetes.** *Children's Health Care* 2007, **36**(4):385-396.
109. Horton TV, Wallander JL: **Hope and social support as resilience factors against psychological distress of mothers who care for children with chronic physical conditions.** *Rehabilitation Psychology* 2001, **46**(4):382-399.
110. Huang CF: **Families of children with developmental disabilities: The test of a structural model of family hardiness, social support, stress, coping, and family functioning.** *Dissertation Abstracts International: Section B: The Sciences and Engineering* 1997, **58**(1-B):0135.

111. McCubbin M, Balling K, Possin P, Frierdich S, Bryne B: **Family resiliency in childhood cancer.** *Family Relations: An Interdisciplinary Journal of Applied Family Studies* 2002, **51**(2):103-111.
112. Thomson G, Moran VH, Axelin A, Dykes F, Flacking R: **Integrating a sense of coherence into the neonatal environment.** *BMC Pediatrics* 2013, **13**(84):1-8.
113. Dyer K: **Identifying, Understanding, and Working with Grieving Parents in the NICU, Part I: Identifying and Understanding Loss and the Grief Response.** *Neonatal Network: The Journal of Neonatal Nursing* 2005, **24**(3):35-46.
114. Dyer K: **Identifying, Understanding, and Working with Grieving Parents in the NICU, Part II: Strategies.** *Neonatal Network: The Journal of Neonatal Nursing* 2005, **24**(4):27-40.
115. Shah PE, Clements M, Poehlmann J: **Maternal Resolution of Grief After Preterm Birth: Implications for Infant Attachment Security.** *Pediatrics* 2011, **127**(2):284-292.
116. Bialoskurski M, Cox CL, Hayes J: **The Nature of Attachment in a Neonatal Intensive Care Unit.** *Journal of Perinatal and Neonatal Nursing* 1999, **13**(1):66-77.
117. Godolphin W: **Shared Decision-Making.** *Healthcare Quarterly* 2009, **2009**(12):186-190.
118. Towle A, Godolphin W: **Framework for teaching and learning informed shared decision making.** *Bmj* 1999, **319**(766-71).
119. Ward F: **Parents and Professionals in the NICU: Communication within the Context of Ethical Decision Making—An Integrative Review.** *Neonatal Network: The Journal of Neonatal Nursing* 2005, **24**(3):25-33.
120. Janvier A, Barrington K, Farlow B: **Communication with parents concerning withholding or withdrawing of life-sustaining interventions in neonatology.** *Seminars in perinatology* 2014, **38**(1):38-46.

121. Haward M, Murphy R, Lorenz J: **Message Framing and Perinatal Decisions.** *Pediatrics* 2008, **122**:109-118.
122. Kuo D, Houtrow A, Neff J: **Family-Centered Care: Current applications and future directions in pediatric health care.** *Matern Child Health J* 2012, **16**(2):297-305.
123. Tsai E: **Advance care planning for paediatric patients.** *Paediatr Child Health* 2008, **13**(9):791-796.
124. Romesberg T: **Understanding Grief: A Component of Neonatal Palliative Care.** *Journal of Hospice and Palliative Nursing* 2004, **6**(3):161-170.
125. Carter BS: **Bereavement in the neonatal period: What we know and what we wish.** *Pediatric Critical Care Medicine* 2011, **12**(3):364-366.
126. Benbassat J, Pilpel D, Tidhar M: **Patients' preferences for participation in clinical decision making: a review of published surveys.** *Behav Med* 1998, **24**(2):81-88.
127. Alderson P, Hawthorne J, Killen M: **Parents' experiences of sharing neonatal information and decisions: Consent, cost and risk.** *Soc Sci Med* 2006, **62**(6):1319-1329.

ANNEX I

Table VI: Theme structure

Grounded theory	Main Themes	Main Categories	Detailed
Reframed QOL and Future coping	Future Projections	Uncertainty Best Interest Developmental outcomes QOL outcomes QOL modifiers	Chapter 4
	Coping	Personal resources External resources	
	Impact of Illness	Grief Attachment	Chapter 5
	Shared Decision Making	Professional role Parental role Communication (Best interest)	

ANNEX II
INFORMATION FORM FOR STUDY PARTICIPATION
(Phase One)



Title of study:

Parental perspectives of the quality of life in children who required neonatal intensive care: the evolution from projection to experience

Principal investigator:

[REDACTED]

Neonatal Follow-up Program
Montreal Children's Hospital, McGill University
MS Candidate
Hôpital Sainte Justine, Université de Montréal
Tel: [REDACTED]

Supervisors:

[REDACTED]

Neonatology
Mother & Child Clinical Ethics Unit
Hôpital Sainte-Justine, Université de Montréal
Tel: [REDACTED]

[REDACTED]

Neonatal Follow-up Program
Montreal Children's Hospital, McGill University
Tel: [REDACTED]

Invitation to participate in a study

Dear Madam, Sir,

I am a paediatrician working in the neonatal follow-up program at the Montreal Children's Hospital. I am also completing a Master's program in Pediatric Clinical Ethics at Université de Montréal.

I am interested in conducting a study with parents of children requiring neonatal intensive care in order to improve the quality of the care provided. I invite you to read this consent form and decide if you're interested in participating in this study.

Nature of the study

Having a child in the neonatal intensive care unit is a very difficult experience. One of the many reasons making this experience challenging is the uncertainty of the future. Throughout their discussions with you, the medical team helps you imagine the future of your baby. Among the predictions, may be the future development (for example, "walking", "talking", "seeing", "hearing", etc.) and the quality of life of your baby.

In this study, I want to explore the way parents foresee their baby's future quality of life while he/she is still in the intensive care unit. When your child is 3-5 years old, I will discuss with you again how these predictions compare with your actual lived experience. Information obtained from this study will help the medical teams improve their communication and counselling skills while discussing with parents the future of the baby.

Study process

If you agree to participate, you will be involved in two steps of the study.

The first step will take place in the next few days. We will meet here at the hospital, for a discussion of how you envision the future of your baby. The discussion will take between 30 and 60 minutes and it will be recorded. You can come alone, or with your partner. After our encounter, I will conduct a brief chart review and record information pertinent for this research project.

I will conduct several such discussions with other parents as well. After finishing all the interviews, I will create a questionnaire for parents using the information obtained. Then, I will contact you again, and ask you to read it and give me your impressions of the quality of this questionnaire.

The second step will take place in 3-5 years from now. I will re-contact you and arrange for another discussion about your child's quality of life. At that time, if you agree to continue the study, the details of the project will be re-discussed with you.

Confidentiality assurance

The interviews will be recorded, transcribed and analysed by myself (and my supervisors as needed). All personal information will be coded with numbers so that your name or your baby's name cannot be identified. The material and recordings will be kept under lock at the Montreal Children's Hospital, under my responsibility. All the information will remain confidential, unless you indicate otherwise or unless required by the law. Neither you, nor your baby will be identified in final publications or conference presentations. The data (recordings and transcriptions) will be destroyed 5 years after the completion of the study. Nevertheless, in order to verify the adequacy of the study process and your protection, a representative of the research ethics committee of CHU Ste-Justine may consult the research data and the medical file of your child.

Risks and inconveniences

There are no physical risks if you participate in this study. However, I understand that talking about such difficult subjects may cause you distress. If this occurs, support from a social worker or psychologist will be available to you. You will be free to quit the study at any time. During the discussions, you will not be required to answer questions that make you uncomfortable. This will not affect at all the quality of the care and the follow-up offered to your baby. The time spent participating in this study may be inconvenient for you. We will try our best to respect your availability.

Advantages and benefits

Your participation is valuable in improving the quality of care provided to parents and babies in the neonatal intensive care unit and during follow-up. At the end of the project, I will be happy to share with you general results of the study, if you wish so.

Researcher responsibility

If you consent to participate in this study, you do not give up any of your or your baby's rights. The researchers remain bound by their legal and professional responsibilities should any situation possibly causing harm to you or your baby occur.

Freedom to participate

Your participation to this study is entirely free and voluntary. Should you decide to decline participation, or to withdraw during the study, the care provided to you and your baby will not be affected and the data will be destroyed.

Contact persons: For any question, dissatisfaction or problem with this study you can contact

[REDACTED]

Consent to participate in the study (Phase One)

Parental perspective of the quality of life of children who required neonatal intensive care: evolution from the projection to the experience

Principal investigator: [REDACTED] MD, Montreal Children's Hospital
Supervisors: [REDACTED], Hôpital Sainte-Justine;
[REDACTED] Montreal Children's Hospital

The nature and conduct of the study have been explained to me. I have read this consent form or it has been explained to me and I have received a copy. I have had the opportunity to ask questions, which have been answered to my satisfaction. After thinking about it, I agree that my child and I participate in this study. I authorize the research team to consult my child's medical record to obtain information relevant to this project.

Child First Name, Last Name

Participant First Name, Last Name

Signature

Date

I wish to be contacted to receive information about the study results:

I have explained the research project to the participant and I have answered all his/her questions. I have informed him/her that the participation is free and voluntary. I promise to respect what has been agreed upon in the consent form.

Signature of researcher [REDACTED]

Date

**FORMULAIRE D'INFORMATION POUR PARTICIPER À UNE ÉTUDE
(Phase 1)**



Titre de l'étude:

Perspective parentale de la qualité de vie des enfants nécessitant des soins intensifs néonataux: l'évolution de la projection à l'expérience

Chercheur principal :

[REDACTED]

Programme de suivi néonatal
Hôpital de Montréal pour Enfants, Université McGill
Candidate MS
Hôpital Sainte Justine, Université de Montréal
Tél: [REDACTED]

Superviseurs :

[REDACTED]

Néonatalogie
Unité d'éthique clinique mère et enfant
Hôpital Sainte-Justine, Université de Montréal
Tél : [REDACTED]

[REDACTED]

Programme de suivi néonatal
Hôpital de Montréal pour Enfants, Université McGill
Tél: [REDACTED]

Invitation à participer à une étude.

Chère Madame, Monsieur,

Je suis une pédiatre travaillant dans le programme de suivi néonatal à l'Hôpital de Montréal pour Enfants. Je suis également en formation dans le programme de maîtrise en éthique clinique pédiatrique à l'Université de Montréal.

Je suis intéressée à faire une étude avec les parents d'enfants ayant besoin de soins intensifs néonataux, afin d'améliorer la qualité des soins. Je vous invite à lire ce formulaire de consentement et de décider si vous êtes intéressé(e) à participer à cette étude.

Description de l'étude

Avoir un enfant dans l'unité de soins intensifs néonataux est une expérience très difficile. L'une des nombreuses raisons rendant cette expérience pénible est l'incertitude de l'avenir. Tout au long de leurs discussions avec vous, les membres de l'équipe médicale vous aident à imaginer l'avenir de votre bébé. Parmi les prédictions faites, il y a le développement futur (par exemple, «marcher», «parler», «voir», «entendre», etc.) et la qualité de vie de votre bébé.

Dans cette étude, je veux explorer la façon dont les parents prévoient la future qualité de vie de leur bébé, alors qu'il/elle est encore dans l'unité de soins intensifs. Quand votre enfant aura 3-5 ans, j'explorerai comment ces prédictions se comparent aux expériences vécues réellement. Les informations obtenues dans cette étude aideront les équipes médicales à améliorer leur communication et leurs conseils aux parents lors des discussions au sujet de l'avenir du bébé.

Déroulement de l'étude

Si vous acceptez de participer, vous serez impliqué(e) dans deux étapes de l'étude.

La première étape se déroulera dans les prochains jours. Nous allons nous rencontrer ici à l'hôpital, pour une discussion sur la façon dont vous imaginez l'avenir de votre bébé. La discussion durera entre 30 et 60 minutes, et sera enregistrée. Vous pouvez venir seul(e), ou avec votre conjoint(e). Après notre rencontre, j'effectuerai une brève revue du dossier médical de votre enfant pour noter les informations pertinentes à ce projet de recherche.

Je vais effectuer plusieurs discussions avec d'autres parents également. Après avoir terminé toutes les entrevues, je vais créer un questionnaire à l'aide de l'information obtenue. Ensuite, je vais vous recontacter et vous demander de le lire et de me donner vos impressions sur la qualité de ce questionnaire.

La deuxième étape se déroulera dans 3 à 5 ans. Je vous recontacterai et organiserai une autre discussion au sujet de la qualité de vie de votre enfant. À ce moment, si vous êtes d'accord à continuer l'étude, je discuterai les détails du projet avec vous.

Assurance de confidentialité

Les entrevues seront enregistrées, transcrites et analysées par moi-même (et mes superviseurs, au besoin). Tous les renseignements personnels seront codés avec des chiffres afin que votre nom et celui de votre bébé ne soient identifiés. Les documents et enregistrements seront conservés sous clef à l'Hôpital de Montréal pour Enfants, sous ma responsabilité. Toutes les informations resteront confidentielles, sauf si vous indiquez autrement ou si requis par la loi. Ni vous, ni votre bébé serez identifiés dans les publications finales ou les présentations aux conférences. Les données (enregistrements et transcriptions) seront détruites 5 ans après la fin de l'étude. Cependant, aux fins de vérifier le bon déroulement de la recherche et d'assurer votre protection, il est possible qu'un délégué du comité d'éthique de la recherche du CHU Ste-Justine pourrait consulter les données de recherche et le dossier médical de votre enfant.

Risques et inconvénients

Il n'y a aucun risque physique associé à cette étude. Cependant, je comprends que discuter des sujets aussi difficiles peut vous causer de la détresse. Si cela se produit, le soutien d'un travailleur social ou psychologue sera à votre disposition. Vous serez libre de quitter l'étude à tout moment. Au cours des discussions, vous n'aurez pas à répondre aux questions qui vous rendent inconfortable. Cela n'affectera pas la qualité des soins et le suivi offert à votre bébé. Le temps passé à participer à cette étude peut être un inconvénient. Je ferai de mon mieux pour accommoder vos disponibilités.

Avantages et bénéfices

Votre participation est précieuse dans l'amélioration de la qualité des soins offerts aux parents et aux bébés dans l'unité des soins intensifs néonataux, ainsi qu'au cours du suivi. À la fin du projet, je serai heureuse de partager avec vous des résultats généraux de l'étude, si vous le souhaitez.

Responsabilité du chercheur

Si vous consentez à participer à cette étude, vous ne renoncez à aucun de vos droits, ni ceux de votre bébé. Les chercheurs restent liés par leurs responsabilités légales et professionnelles dans toute situation qui pourrait causer un dommage à vous ou à votre bébé.

Liberté de participer

Votre participation à cette étude reste entièrement libre et volontaire. Si vous décidez de refuser la participation, ou de vous retirer au cours de l'étude, les soins offerts à vous et à votre bébé ne seront pas affectés et les données seront détruites.

Personnes ressources: Pour toute question, insatisfaction ou problème avec cette étude vous pouvez contacter : [REDACTED] [REDACTED]

Consentement à participer à l'étude (Phase 1)

Perspective parentale de la qualité de vie des enfants nécessitant des soins intensifs néonataux: l'évolution de la projection à l'expérience

Chercheur principal: [REDACTED] MD, Hôpital de Montréal pour Enfants
Superviseurs : [REDACTED], l'Hôpital Sainte-Justine ;
[REDACTED] Hôpital de Montréal pour Enfants

On m'a expliqué la nature et le déroulement du projet de recherche. J'ai pris connaissance du formulaire de consentement et on m'en a remis un exemplaire. J'ai eu l'occasion de poser des questions auxquelles on a répondu à ma satisfaction. Après réflexion, j'accepte de participer et que mon enfant participe à ce projet de recherche. J'autorise l'équipe de recherche à consulter le dossier médical de mon enfant pour obtenir les informations pertinentes à ce projet.

Prénom, nom de famille de l'enfant

Prénom, nom de famille du participant

Signature

Date

Je souhaite être contacté(e) pour recevoir des informations sur les résultats

J'ai expliqué le projet de recherche au participant et j'ai répondu à toutes ses questions. Je l'ai informé que la participation est libre et volontaire. Je m'engage à respecter ce qui a été convenu dans le formulaire de consentement.

Signature du chercheur [REDACTED]

Date

ANNEX III

SAMPLE DISCUSSION GUIDE

How do you find your experience in the neonatal intensive care so far?

Did the team meet with you to explain the condition of your baby? How did the meeting go?

Has the team made any prediction with respect to the future development of your baby (walking, talking, seeing, hearing, etc.)?

What do you think of these predictions?

Has the team made any prediction with respect to the future quality of life of your baby?

What do you think of these predictions?

How do you envision the future quality of life of your baby?

In your opinion, what will determine the future quality of life of your baby?

Do you think treatment decisions should be based on these predictions?

In your opinion, how could we improve the predictions of future quality of life?

ANNEX IV

Figure 2: Conceptual model of theme relationships (including Grief)

