International Library of Ethics, Law, and the New Medicine 65

Eduard Verhagen
Annie Janvier *Editors*

Ethical Dilemmas for Critically Ill Babies

Springer

International Library of Ethics, Law, and the New Medicine

Volume 65

Series editor

David N. Weisstub
University of Montreal Fac. Medicine, Montreal, Québec, Canada

The book series International Library of Ethics, Law and the New Medicine comprises volumes with an international and interdisciplinary focus. The aim of the Series is to publish books on foundational issues in (bio) ethics, law, international health care and medicine. The 28 volumes that have already appeared in this series address aspects of aging, mental health, AIDS, preventive medicine, bioethics and many other current topics. This Series was conceived against the background of increasing globalization and interdependency of the world's cultures and governments, with mutual influencing occurring throughout the world in all fields, most surely in health care and its delivery. By means of this Series we aim to contribute and cooperate to meet the challenge of our time: how to aim human technology to good human ends, how to deal with changed values in the areas of religion, society, culture and the self-definition of human persons, and how to formulate a new way of thinking, a new ethic. We welcome book proposals representing the broad interest of the interdisciplinary and international focus of the series. We especially welcome proposals that address aspects of 'new medicine', meaning advances in research and clinical health care, with an emphasis on those interventions and alterations that force us to re-examine foundational issues.

More information about this series at http://www.springer.com/series/6224

Eduard Verhagen · Annie Janvier
Editors

Ethical Dilemmas for Critically Ill Babies

The Brocher Foundation mission is to encourage a research on the ethical, legal and social implications of new medical technologies. Its main activities are to host visiting researchers and to organize symposia, workshops and summer academies. More information on the Brocher Foundation program is available at www.brocher.ch.

Editors
Eduard Verhagen
Beatrix Children's Hospital, University Medical Center Groningen
University of Groningen
Groningen
The Netherlands

Annie Janvier
Department of Pediatrics and Clinical Ethics
University of Montreal
Montreal
Canada

ISSN 1567-8008 ISSN 2351-955X (electronic)
International Library of Ethics, Law, and the New Medicine
ISBN 978-94-017-7359-1 ISBN 978-94-017-7360-7 (eBook)
DOI 10.1007/978-94-017-7360-7

Library of Congress Control Number: 2015947933

Springer Dordrecht Heidelberg New York London

Printed on acid-free paper

Springer Science+Business Media B.V. Dordrecht is part of Springer Science+Business Media (www.springer.com)

Acknowledgments

We want to thank the Brocher Foundation for the support and contribution to the appearance of this book.

Contents

Chapter 1
Introduction

John Lantos

Abstract In neonatology, a key question focuses on the degree of certainty is that must be achieved in order to deem treatment morally and legally obligatory, optional or futile. With different overall patterns, trends, and categories in mind, the authors in this book examine current attitudes and practices in neonatology.

The story of neonatal intensive care is a remarkable story of medical progress but also a story of moral controversy. Every year, in NICUs around the world, many babies are saved who would have perished if they had been born 40 years ago. In the United States, roughly 250,000 preterm infants are born each year. Before 1965, many of these babies would have died. Today, most survive without long-term health problems. Neonatology has become the largest subspecialty in pediatrics.

Neonatology is not an unmitigated success. Many survivors are left with lifelong medical problems such as chronic lung disease, visual impairment, seizures or neurodevelopmental problems. These chronic health problems have led some observers to conclude that neonatology is not as successful as it sometimes seems and today in the grey zones of neonatology, parents and clinicians together decide whether life sustaining interventions for certain neonates should be withheld or withdrawn. Priest and bioethicist John Paris wrote, "There comes a point with extremely premature infants… where the risk of mortality and morbidity becomes so significant and the degree of burden and the prospects of benefit so suffused in ambiguity and uncertainty that a decision as to whether to institute or continue medical treatment properly belongs to the parents [1]". Indeed, a significant proportion of deaths in Neonatal Intensive Care Units (NICUs) follow these difficult end of life considerations. Such debates have surrounding neonatology since its inception.

Epidemiological studies of the long-term outcomes of NICU babies can also be contentious. Some such studies have concluded that the overall prevalence of cerebral palsy is increasing and that this is attributable to greater numbers of NICU

J. Lantos (✉)
Children's Mercy Bioethics Center and Department of Pediatrics,
University of Missouri-Kansas, Kansas City, MO 65211, USA
e-mail: jlantos@mch.edu

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*,
International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_1

survivors [2]. Others show that the prevalence of cerebral palsy has not changed or is actually lower than it used to be [3]. Whichever side is correct, one thing is clear. The overall prevalence of Cerebral Palsy (CP) may or may not have changed, but it now occurs in a different way. It used to just happen. Now CP occurs after a decision to save a baby's life.

This book addresses the moral, legal, economic and political questions involved in neonatology. To understand these arguments, one must first understand the sorts of medical problems that are treated in NICUs.

Neonatal mortality is defined as death before 28 days of age. Post-neonatal mortality is defined as death between 28 days and 1 year. Infant mortality is the sum of these two and is defined as death before one year of age.

Babies who weigh less than 2500 g (5.5 pounds) at birth are classified as low birth weight (LBW). Babies who weigh less than 1500 g (3.3 pounds) at birth are considered very low birthweight (VLBW). In some reports, another category, extremely low birth weight (ELBW), describes outcomes for babies who weigh less than 1000 g (2.2 pounds) at birth.

In the United States, infant mortality has dropped from 55/1000 in 1900 to 9/1000 in 2000. Trends over the century reveal much about the reasons for the improvement. In the early part of the century, the improvement was in both neonatal and post-neonatal mortality. Most epidemiologists attribute these improvements to public health measures, such as better nutrition and sanitation. In the mid-century, post-neonatal mortality rates improved faster than neonatal mortality rates. This is usually attributed to medical interventions such as antibiotics and immunizations that had greater efficacy in older babies than in neonates.

In 1960, the neonatal mortality rate in the United States was 19/1000. It steadily dropped over the next decades, to 16 in 1970, 13 in 1980, 9 in 1990 and 4 in 2000. Much of the recent drop is attributable to improvements in survival for low birthweight babies. These improvements in birthweight-specific neonatal mortality are generally attributed to improvements in neonatal intensive care and, in particular, in the care of tiny premature babies [4]. Specifically, these improvements have included prenatal steroids given to women in preterm labor, ventilators surfactant and total parenteral nutrition.

Many of the patients in NICUs are premature babies, but they are not the only group of patients admitted to NICUs. The other groups of NICU patients are both medically and ethically distinct. This can be seen by dividing the babies admitted to the NICU into three groups, recognizing that there will be some overlap between the groups.

The three groups are (1) full-term or near-term babies with acute illnesses; (2) babies with congenital anomalies; and (3) premature babies. These groups of babies raise different clinical and ethical issues. This book focuses primarily on the third group, premature babies.

Prematurity is both an acute crisis leading to many possible iatrogenic complications and a chronic condition. The acute crisis requires urgent medical interventions. At the time when these treatments are initiated, however, there is significant uncertainty about long-term prognosis. This is not true with the first two

categories of patients. With acute sepsis, for example, treatment will likely either succeed, in which case there will be complete resolution of the problem, or it will fail, in which case the baby will die. With congenital anomalies and syndromes, treatment cannot cure the underlying disease but will almost surely be successful in treating some of the associated conditions. The long-term prognosis for survivors in such cases is dependent upon the underlying condition, not the acute problems, and is usually clearly predictable with some precision.

With extremely premature babies, by contrast, there is usually a very wide range of possible outcomes. Some babies die early. Others survive for weeks or months in the NICU only to die later. Still others survive with disabilities that range from mild to severe. At the time when treatment must be initiated, doctors cannot say what the outcome for any particular baby will be.

In such situations, a key question focuses on the degree of certainty is that must be achieved in order to deem treatment morally and legally obligatory, optional or futile.

With these overall patterns, trends, and categories in mind, the authors examine current attitudes and practices in neonatology.

References

1. Paris JJ, Reardon F. Bad cases make bad law: HCA v. Miller is not a guide for resuscitation of extremely premature newborns. J Perinatol 2001:21:541–4.
2. Wichers MJ, van der Schouw YT, Moons KG, Stam HJ, van Nieuwenhui zen O. Prevalence of cerebralpalsy in The Netherlands (1977–1988). Eur J Epidemiol. 2001;17(6):7–32.
3. Winter S, Autry A, Boyle C, Yeargin-Allsopp M. Trends in the prevalence of cerebral palsy in a population-based study. Pediatrics. 2002;110(6):1220–5.
4. Lee KS, Khoshnood B, Hsieh H, Kim BI, Schreiber MD, Mittendorf R. PaediatrPerinatEpidemiol. 1995;9:420–30.

Chapter 2
How Babies Die and Why This Is Important to Clinicians, Researchers, and Parents

Eduard Verhagen and Annie Janvier

Abstract With technological developments and new knowledge, survival for many neonatal conditions has improved. Survival can be either with or without disability, and/or a chronic condition. Decision-making for neonates with uncertain futures is often influenced by considerations of length of survival, disabilities, and quality of life. In this context, deciding if an LSI is of benefit to a patient involves data about outcomes but also value judgments. These are among the hardest decisions of modern medicine. This chapter describes why discovering the evidence about the outcomes of neonates with uncertain futures in the medical literature is complex and describes research and solutions to help clinicians in this context.

In the past, when neonates died because of lack of intensive care units, ethical discussions and decision-making about life sustaining interventions (LSIs) were generally irrelevant. With technological developments and new knowledge, survival for many conditions has improved. Survival can be either with or without disability, and/or a chronic condition. Decision-making for neonates with uncertain futures is often influenced by considerations of length of survival, disabilities, and quality of life. In this context, deciding if an LSI is of benefit to a patient involves data about outcomes but also value judgments. These are among the hardest decisions of modern medicine. This article describes why discovering the evidence

E. Verhagen (✉)
Department of Pediatrics, University Medical Center Groningen, University of Groningen, 9700, RB Groningen, The Netherlands
e-mail: a.a.e.verhagen@umcg.nl

A. Janvier
Department of Pediatrics and Clinical Ethics, Neonatology and Clinical Ethics, Sainte-Justine Hospital, University of Montreal, 3175 Côte-Sainte-Catherine, Montreal, QC H3T 1C4, Canada
e-mail: anniejanvier@hotmail.com

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_2

about the outcomes of neonates with uncertain futures in the medical literature is complex and describes research and solutions to help clinicians in this context.

2.1 Values, Policies, and Facts About Survival

In industrialized countries, most neonatal deaths now occur in Neonatal Intensive Care Units (NICUs) and follow a decision to limit LSIs [1–5]. Attitudes of caregivers also influence speculation about these issues and may contribute to variations in survival and health outcomes for critically ill children [5–7]. Different values, cultures, and/or policy statements can lead to wide variations of practice [8]. For example, one of the "gray zones" in neonatology relates to intervention for extremely preterm infants. Neonates born before 22 weeks of gestational age (GA) do not survive. This is true in all publications. But if we examine the literature for survival at 24 weeks of GA, survival is not homogeneous between studies and varies widely in the literature. In some countries—including Germany, Japan, Sweden, and Norway—such a baby would almost always receive active intervention [9–12]. In Canada, the UK, and some hospitals in the U.S., prospective parents are informed about the outcomes of extreme preterm infants and must decide between intensive care and comfort care [13, 14]. In the Netherlands [15] and in some centers in the U.S. [16], intensive care for these babies is generally considered "non beneficial" and neonatologists often recommend comfort care. Not surprisingly, outcomes for babies born at 24 weeks of GA vary widely between countries. Survival for babies born at 24 weeks is 81 % in Japan, 60 % in Canada, 33 % in the UK, and is as low as zero in some centers in the Netherlands.

It is important to note that attitudes and practice regarding the care of extremely preterm babies are changing. For example, only recently in the Netherlands, obstetricians and neonatologists nationwide agreed to offer parents active resuscitation for neonates from a gestational age of 24 weeks onwards. It appears that the mortality rate among neonates with a gestational age of 24 weeks is now decreasing and is becoming more similar to the rates in other European countries actively resuscitating neonates of such a low gestational age [17]. We still need detailed data about the change in policy to understand what we can validly infer from comparison with other outcome studies.

There is an intimate relationship between values, policies, and facts. What information should parents be told: survival data for their center, survival for other centers, or the wide variation of practice between centers? What does informed consent mean in this situation? If parents are told that the chances of their baby's survival are low, they will be less likely to choose treatment. If they choose comfort care, their baby will die and in turn, will become part of the statistics. A similar variation of practice can be seen for other neonatal conditions necessitating intensive care and linked with uncertain outcomes. Some examples are hypoplastic left heart syndrome, hydrocephaly, severe renal insufficiency, and trisomy 13 and 18.

2.2 Outcome and Unit Philosophy

Finding survival statistics for fragile neonates and interpreting the data from the values may be difficult, but examining and interpreting long-term outcomes when babies survive can be even harder. Let's return to the example of a baby born at 24 weeks of GA. She develops a large unilateral grade 4 parenchymal hemorrhage at three days of life. Should the physician offer to withdraw LSI because of this finding? What can we find in the literature? What is the positive predictive value (PPV) of future cerebral palsy in preterm babies who survive with such a bleed? The outcomes after this neurological insult vary widely in the literature: from a PPV of 20–85 % [18, 19]. These differences arise in part from value judgments and not only from the NICU care babies receive. In some units, physicians will recommend withdrawing (WD) LSI in this case. In others, physicians will offer to WD LSI. And in others, parents will be informed that this parenchymal hemorrhage will increase the risk of disability for their child. Even in a single NICU with homogeneous values and so-called "neutral disclosure" to parents, different styles of disclosure may influence parents. Some babies die *with* their parenchymal hemorrhage (because of sudden bleeding and cardiovascular collapse) and others die *because* of their parenchymal hemorrhage (LSI are withdrawn for quality of life reasons). Furthermore, some units will not consider WD LSI for a small hemorrhage, but only for a large one, in a more unstable baby. The smaller the hemorrhage, the better the outcomes. Survivors in different units will have different outcomes, mainly because of the philosophy of the unit. Similar variations of practice can be seen for many other conditions where WD of LSI can occur in the NICU: hypoplastic left heart syndrome, severe NEC, severe respiratory insufficiency, hydrocephalus following IVH, seizures in very preterm infants, meningitis with shock, asphyxia with serious seizures, and others.

So how can we interpret statistics for neonates with uncertain futures and decide whether an LSI is in the best interest of our patient? How is it possible to make sense of the medical literature when outcomes are influenced not only by interventions but also by philosophies?

2.3 Description of Circumstances Around the End-of-Life

The circumstances surrounding decisions to withhold or withdraw interventions are rarely explicitly described in publications on neonatal intensive care unit (NICU) outcomes. Most studies do not make the distinction between WD/WH LSI from dying babies, and WD/WH interventions from physiologically stable children for quality-of-life reasons. This is an important distinction, as children who were stable might have lived if intervention had not been withdrawn or withheld.

NICU deaths for fragile neonates should be transparent in the medical literature. Without these distinctions, it is impossible to truly examine the literature, counsel

parents in a meaningful way, examine whether a certain intervention makes a real difference in terms of disability, or compare studies of NICU outcome (between NICUs or in one NICU at different times) in a meaningful way.

We have recently reported a useful method to classify and compare death and dying in the delivery room and NICU using strict definitions of physiology and LSI [20]. There are five ways neonatal deaths can be classified: (1) Some babies die because NICU admission is WH, (2) others die with CPR (No WH or WD), (3) or without CPR but on a respirator (no WD, WH CPR), (4) or because LSI are WD/WH in an unstable patient (baby would have died despite LSI), (5) or because LSI are WH/WD in a physiologically stable patient for quality of life reasons. In this category, many patients may have survived, had LSI been continued. Each unit can categorize their deaths in this manner. We were able to show that almost all NICU deaths in four units in three countries were accompanied by some degree of withholding/withdrawing, but that the physiologic stability of the dying infants varied considerably within and between countries. In addition, we found that dying babies with similar characteristics were treated differently with regard to initiating and increasing comfort medications. This difference in treatment between units suggests that comfort medication was not only directed at the medical condition of the baby but also at the parents and healthcare providers' comfort and outcome [21]. Uniform classification of deaths is feasible and offers an important step towards transparency about norms and values of stakeholders in decision-making and true comparison of NICU outcomes.

Studies that do describe the circumstances around the decisions about the newborn's end-of-life report that 25–45 % of NICU deaths are by withdrawing intensive care in stable newborns for quality of life reasons [22, 23]. This high proportion of deaths by 'elective' withdrawal seems even more important if we keep in mind that physicians will use outcome reports to counsel parents about decision-making regarding the fetus and/or sick neonate.

Another important step toward more transparency and comparability could be made if we use data about babies who never make it to the NICU in neonatal outcome calculations. A substantial proportion of these babies are often described as "in utero deaths." There are many ways fetuses die in utero; the most common ways are shown in Table 2.1. In addition, a group of live born babies are not admitted to the NICU for the reasons mentioned in Table 2.1.

Table 2.1 Groups of babies who never make it to the NICU: in utero deaths and DR deaths

Stillbirths (dead on arrival to the hospital)
Stillbirths (died in utero because of withholding surgical delivery)
Termination of Pregnancy (TOP) for congenital malformations
"Induction" of labor for risk of extreme prematurity
Small preterms who receive comfort care
Term babies who receive palliative care because of their predicted outcomes
Failed resuscitations

Outcomes for all of these categories of newborns can be influenced by national health policies. For example, a high rate of termination for the "most severe" hypoplastic left heart syndrome will lead to better surgical outcomes for babies with this condition. A decision to do cesarean sections only for fetuses over 25 weeks of GA will likely select bigger non-growth restricted neonates at each GA and will affect outcomes. Another striking example is what happened in Holland in 2007. The Dutch government started offering structural ultrasound at 20 weeks' gestation for all pregnant women at no extra cost. This has resulted in fewer births of babies with severe anomalies, such as spina bifida (because of termination of pregnancy), which in turn has led to fewer cases of neonatal euthanasia [24]. Another example is the policy statements for treatment of extremely premature infants. In Canada, palliative care is advised in babies born at less than 23 weeks of gestational age, and cesarean section is not recommended below 24 weeks of gestational age [14]. In the United States, gestational age is not mentioned. In the Netherlands, non-intervention is now advised below 24 weeks of gestational age, and between 24 and 25 weeks is defined as the "gray zone" where interventions are optional, depending on parental preferences [15]. The latter policy will result in fewer survivors less than 25 weeks of gestational age, fewer survivors with handicap, and also fewer survivors overall. The policy may also result in less desire to intervene below 25 weeks gestational age, thus contributing to a self-fulfilling prophecy.

For these reasons, we should consider making all fetuses and neonates who are alive at more than 450 g the denominator of all deaths, even if these fetuses are in utero. This classification is more complex than categorizing neonatal deaths and also involves another set of ethical values (and other disciplines).

The examples discussed in this paper underline that national and/or professional healthcare policies can change medical practice and shape patient outcomes. Conversely, outcomes could also influence national health policies and local/personal values. If the prevalence of individuals with disability from prematurity or congenital malformation in our communities decreases, society might adapt. It would be interesting to learn what that might do to health care infrastructure and the availability of support and acceptance of children with disabilities among the public. What effect might this situation have on policy makers and on physicians' perinatal and counseling and decision-making?

Knowledge about how babies die is important because decisions about WH/WD in the delivery room and in the NICU are in part based on personal, local and/or national values. Those values shape the patient mix, outcome in NICUs, and the context of clinical and ethical decisions. Reflection on those values and on the differences in values between units, as well as proper comparison of outcome data, can take place only if we start including in our calculations data about how babies die and who never makes it to the NICU. Only if we can recognize our biases and make more transparent the true reasoning behind our decision-making processes can we be empowered to respond appropriately and consistently to the needs of sick children and their families.

Today, decisions on when to start WH or WD life supportive interventions in critically ill children are among the most difficult decisions in pediatric practice.

These decisions directly affect the health of our societies. It is difficult to remain neutral when we make these decisions. We expect that full transparency of the circumstances around these decisions will allow us to compare outcomes better, reflect on similarities and differences in end-of-life care, and ultimately make the necessary improvements in the interest of the child.

References

1. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980–1998. Pediatrics. 2002;109(4):656–60.
2. Ryan CA, Byrne P, Kuhn S, Tyebkhan J. No resuscitation and withdrawal of therapy in a neonatal and a pediatric intensive care unit in Canada. J Pediatr. 1993;123(4):534–8.
3. Sands R, Manning JC, Vyas H, Rashid A. Characteristics of deaths in paediatric intensive care: a 10-year study. NursCrit Care. 2009;14(5):235–40.
4. Lago PM, Piva J, Garcia PC, Troster E, Bousso A, Sarno MO, et al. End-of-life practices in seven Brazilian pediatric intensive care units. Pediatr Crit Care Med. 2008;9(1):26–31.
5. Verhagen AA, Janvier A, Leuthner SR, Andrews B, Lagatta J, Bos AF, et al. Categorizing neonatal deaths: a cross-cultural study in the United States, Canada, and The Netherlands. J Pediatr. 2010;156(1):33–7.
6. Janvier A, Lantos J. Variations of practice in the care of extremely preterm infants. In: Diekema D, Mercurio M, Adam M, editors. Clinical ethics in pediatrics, a case-based textbook. Cambridge: Cambridge University Press; 2011. p. 94–100.
7. Gallagher K, Martin J, Keller M, Marlow N. European variation in decision-making and parental involvement during preterm birth. Arch Dis Child. 2014;99(3):F245–9.
8. Janvier A, Lantos J. Variations of practice in the care of extremely preterm infants. In: Diekema D, Mercurio M, Adam M, editors. Clinical ethics in pediatrics, a case-based textbook. Cambridge University Press, Oxford University Press; 2011. pp. 94–100.
9. Itabashi K, Horiuchi T, Kusuda S, Kabe K, Itani Y, Nakamura T, et al. Mortality rates for extremely low birth weight infants born in Japan in 2005. Pediatrics. 2009;123(2):445–50.
10. Sugiura T, Kouwaki M, Togawa Y, Sugimoto M, Togawa T, Koyama N. Neurodevelopmental outcomes at 18 months' corrected age of infants born at 22 weeks of gestation. Neonatology. 2011;100(3):228–32.
11. Markestad T, Kaaresen PI, Ronnestad A, Reigstad H, Lossius K, Medbo S, et al. Early death, morbidity, and need of treatment among extremely premature infants. Pediatrics. 2005;115 (5):1289–98.
12. Serenius F, Sjors G, Blennow M, Fellman V, Holmstrom G, Marsal K, et al. EXPRESS study shows significant regional differences in 1-year outcome of extremely preterm infants in Sweden. Acta Paediatr. 2014;103(1):27–37.
13. Holtrop P, Swails T, Riggs T, De Witte D, Klarr J, Pryce C. Resuscitation of infants born at 22 weeks gestation: a 20-year retrospective. J Perinatol. 2013;33(3):222–5.
14. Jefferies AL, Kirpalani HM, Canadian Paediatric Society F, Newborn C. Counselling and management for anticipated extremely preterm birth. Paediatrics Child Health. 2012;17 (8):443–446.
15. de Laat MW, Wiegerinck MM, Walther FJ, Boluyt N, Mol BW, van der Post JA, et al. Practice guideline 'Perinatal management of extremely preterm delivery'. Ned Tijdschr Geneeskd. 2010;154:A2701.
16. Kaempf JW, Tomlinson M, Arduza C, Anderson S, Campbell B, Ferguson LA, et al. Medical staff guidelines for periviability pregnancy counseling and medical treatment of extremely premature infants. Pediatrics. 2006;117(1):22–9.

17. de Kluiver E, Offringa M, Walther FJ, Duvekot JJ, de Laat MW. Perinatal policy in cases of extreme prematurity; an investigation into the implementation of the guidelines. Ned Tijdschr Geneeskd. 2013;157(38):A6362.
18. Hack M, Taylor HG, Drotar D, Schluchter M, Cartar L, Wilson-Costello D, et al. Poor predictive validity of the Bayley Scales of Infant Development for cognitive function of extremely low birth weight children at school age. Pediatrics. 2005;116(2):333–41.
19. Schmidt B, Anderson PJ, Doyle LW, Dewey D, Grunau RE, Asztalos EV, et al. Survival without disability to age 5 years after neonatal caffeine therapy for apnea of prematurity. JAMA. 2012;307(3):275–82.
20. Verhagen AA, Janvier A, Leuthner SR, Andrews B, Lagatta J, Bos AF, et al. Categorizing neonatal deaths: a cross-cultural study in the United States, Canada, and the Netherlands. J Pediatr. 2010;156(1):33–7.
21. Janvier A, Meadow W, Leuthner SR, Andrews B, Lagatta J, Bos A, et al. Whom are we comforting? An analysis of comfort medications delivered to dying neonates. J Pediatr. 2011;159(2):206–210.
22. Verhagen AA, Dorscheidt JH, Engels B, Hubben JH, Sauer PJ. End-of-life decisions in Dutch neonatal intensive care units. Arch Pediatr Adolesc Med. 2009;163(10):895–901.
23. Berger TM, Hofer A. Causes and circumstances of neonatal deaths in 108 consecutive cases over a 10-year period at the Children's Hospital of Lucerne. Switz Neonatology. 2009;95 (2):157–63.
24. Verhagen AA. The Groningen protocol for newborn euthanasia; which way did the slippery slope tilt? J Med Ethics. 2013;39(5):293–5.

Chapter 3
When Do We Become a Person and Why Should It Matter to Pediatricians?

Amélie Dupont-Thibodeau and Annie Janvier

Abstract After decades of public debate and controversy, the question about when we become a person remains largely unresolved. Many definitions of personhood have been proposed. For some, a fetus becomes a person at a specific time: either after conception, when pain can be experienced, after viability, at the emergence of sentience or consciousness, or after birth. Other conceptions of personhood define a person in terms of human relationships. Some believe a mother (or a family) will decide when her child becomes a person; others, that a person exists only when there are specific interactions with others or the environment. Various interpretations of who is a person will lead to different political and clinical decisions. For most contemporary scholars, the issue of personhood must be resolved in order for us to resolve issues such as abortion. The present chapter will review seven definitions of personhood, their effect on society and on the outcomes of neonates.

Who is a person? Rarely has a question been so ethically charged and morally divisive. After decades of public debate and controversy, it remains largely unresolved. Many definitions of personhood have been proposed, falling into the following seven categories. For some, a fetus becomes a person at a specific time: either after conception [1], when pain can be experienced [2], after viability, at the emergence of sentience or consciousness [3], or after birth [4]. Other conceptions of personhood define a person in terms of human relationships. Some believe a mother (or a family) will decide when her child becomes a person [5]; others, that a person exists only when there are specific interactions with others or the environment [6].

A. Dupont-Thibodeau (✉) · A. Janvier
Neonatology and Clinical Ethics, Sainte-Justine Hospital, 3175 Chemin de la Côte-Sainte-Catherine, H3T 1C4 Montreal, Canada
e-mail: amelie.du.pont-thibodeau@umontreal.ca

A. Janvier
e-mail: anniejanvier@hotmail.com

A. Dupont-Thibodeau · A. Janvier
Department of Pediatrics and Ethics Bureau, University of Montreal, 2900 boulevard Edouard-Montpetit, H3T 1J4 Montreal, Canada

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_3

The spectrum of divergent and conflicting opinions remains as wide as ever and continues to directly affect laws, politics, public policy, and medical decision-making. Areas affected include inheritance eligibility, and abortion laws, as well as end of life care, reproductive technology, and euthanasia.

Also, with advancing new technologies, the boundaries of what was once thought as possible are now being pushed, forcing the redefinition of fundamental concepts, such as what constitutes medical viability. With the emergence of new controversies, such as human embryo and stem cell research, we are faced with questions about what should society allow or accept as definitions of a person, morally, legally, or both.

The definition of personhood has vigorously been debated over the years as it is considered by many to be the main notion that might help us navigate these rough, murky waters and separate what is moral from what is not. Various interpretations of who is a person will lead to different political and clinical decisions. For Warren, a philosopher, "It deserves emphasis that our difficulties in saying when a person begins are primarily the result of our inability to say what a person is […]. The question which we must answer […] is this: how are we to define the moral community, the set of beings with full and equal moral rights, such that we can decide whether a human fetus is a member of this community or not?" [2]. For most contemporary scholars, the issue of personhood must be resolved in order for us to resolve issues such as abortion. The present chapter will review the seven various definitions of personhood, their effect on society and on the outcomes of neonates.

3.1 Conception

For many, personhood begins at conception. This view has been defended for centuries by various institutions, including the Roman Catholic Church, which considers all life sacred from conception until death. According to the Congregation for the Doctrine of the Faith, "Thus the fruit of human generation, from the first moment of its existence, that is to say from the moment the zygote has formed, demands the unconditional respect that is morally due to the human being in his bodily and spiritual totality. The human being is to be respected and treated as a person from the moment of conception; and therefore from that same moment his rights as a person must be recognized, among which in the first place is the inviolable right of every innocent human being to life" [7]. According to the major tradition of the Hindu faith, ensoulment of the fetus occurs at conception [8]. The fetus or embryo therefore has personhood and deserves protection. In Judaism, however, "according to the Talmud, within the first 40 days after conception the zygote is simply water" [9]. The fetus seemingly only attains full person status at birth. Yet, abortion "is not viewed as a morally neutral matter of individual desire or an acceptable form of post facto birth control" [9]. For Jehovah's witnesses, life

begins at conception, making abortion even if the life of the mother is in danger impermissible [10]. Finally, in Islam, ensoulment of the fetus is thought to occur between 40 and 120 days of gestation [11]. More recently, the concept of personhood beginning at conception has been supported by a wide range of philosophers. For scholars such as Larmer, "the potential for human consciousness is a sufficient condition of personhood and, since the fetus possesses this potential, is a person." [3]. Many others also suggest that it is simply the potential to become conscious which obligates us to protect the life of a human organism [3].

This view of a fetus as a person has been largely influential in today's political, ethical, and legal beliefs. It has shaped how abortion is viewed by many, how societies legislate in this matter, and how courts have set precedents. The notion of personhood at conception is also fundamental in the debate regarding the permissibility of in vitro fertilisation (IVF). It extends to how many embryos are created and used. During IVF, more than one embryo is frequently available to implant in the woman's body. A physician can either implant all the generated embryos, which could be more than three or four, or implant one or two and freeze the remaining embryos. Implanting more than one embryo increases the chances of a successful pregnancy, but also increases the chances of twins and triplets or higher order multiple births. Multiple births increase the risks to pregnant women and their children, mainly because of the increased risk of prematurity. It has been demonstrated that implanting a maximum of one or two embryos is safer for women and future children. But in countries where there is a tradition of considering that a person exists from the time of conception, freezing remaining embryos is deemed either morally problematic or unacceptable. In these countries, this concept of personhood often determines regulations, banning freezing of embryos, even if implanting all of the harvested embryos represents significant health risks to both the mother and the fetuses. In some countries, during IVF, in cases of a multiple pregnancy with three or more embryos, selective abortions are often performed to "reduce" the pregnancy to a safer twin pregnancy. "Selective reductions" are not acceptable when personhood is defined as occurring at conception.

This definition of personhood has led many jurisdictions to limit or prohibit embryo and stem cell research, and to introduce restrictions on IVF. For example, in Germany, a law was enacted in 1992 prohibiting all research on human embryos that is not carried out exclusively to preserve embryos or facilitate uterine transfer [4]. In addition, only a maximum of three embryos can be transferred, none of them can be frozen after a certain stage, and all of the embryos produced have to be implanted. In Austria, as of 2008, preimplantation genetic diagnosis (PIGD) remains forbidden on the basis that embryos cannot be used for anything but pregnancy. In Italy, a religious Catholic country, only three eggs can be fertilized during IVF, even if more are generally produced after the burdensome process of ovulation induction. In addition, all embryos must be implanted. PIGD, genetic screening, and pregnancy reduction are not permitted. In Switzerland, PIGD is explicitly forbidden [4]. In Finland, PIGD diagnosis as well as the number of embryos transferred are not legally determined [4]. In Quebec (not in the whole of

Canada), single embryo transfer is the norm. All remaining embryos are either destroyed, donated to science or to another couple, or frozen.

Variations also exist regarding the acceptability of embryonic research. In many countries where embryos can be frozen or donated, embryonic research—which can destroy the embryo—is permissible. It is performed on embryos that were not implanted during IVF. Regulations and ethical and scientific policies define a time during which embryonic research is acceptable, generally the first 14 days after fertilization [12]. In the USA, the Ethics Advisory Board (EAB) was appointed in 1978 with a mandate to review both IVF research and research using embryos resulting from IVF. The EAB did not support research on embryos older than 14 days but specified that embryos were different from persons: "The human embryo is entitled to profound respect; but this respect does not necessarily encompass the full legal and moral rights attributed to persons" [13]. Therefore, large variations exist in legislation, treatment of infertile couples, research, and care of pregnant women, affecting reproductive care and the outcome of children depending on the way personhood is defined.

3.2 Pain

A second answer to the question "When does the fetus become a person?" has been when the fetus begins to feel pain. As explained by Penner and his colleagues, "the claim is that the sensory input stored by the brain (along with other types of input later in life) is the essential basis upon which our experiential individuality as persons rests" [5], and "identifying the initiation of the coordinated functioning of these three aspects of the central nervous system of the fetus is the step that would convert this approach from a philosophical argument into a practical, pragmatic tool for promoting agreement among secularists regarding the time during gestation beyond which it is ethically impermissible to abort a fetus except under special circumstances." [5]. The central nervous system appears during the third week post conception. The basic divisions of the brain start developing around a gestational age of six weeks. The first fetal movements have been detected at eight weeks of gestational age, but these are believed to be automatic, not voluntary, and are under the control of the brainstem [6]. This potential pain perception has influenced certain laws and regulations in relation to embryonic research.

However, pain is felt by all vertebrate animals. If pain is to define a person, then the cortical response to pain is what should be examined. Rudimentary cortical evoked responses to somatosensory stimulation can be recorded as early as 23–24 weeks of post-menstrual age (PMA), reflecting the beginning ingrowth of thalamocortical axons in the somatosensory cortex [6]. Functional thalamocortical connections are required for more advanced conscious perception of pain, and these appear around 29–30 weeks of gestation.

Already, these findings have had repercussions on the way current medicine is practiced. For example, analgesia is now given to fetuses before third trimester

abortions in many centers. Most protocols include fentanyl administration before the feticide in order to make sure the fetus does not suffer from the termination of pregnancy. In the United States, legislation requiring physicians to inform pregnant women consulting for an abortion about fetal pain and fetal anesthesia is becoming more and more popular. States such as Nebraska, Idaho, Oklahoma, Kansas, Indiana, and Alabama have already passed laws to this effect [14].

3.3 Viability

Viability, too, has been a frequent answer to the question of when a fetus becomes a person. This argument has been crucial most notably in key legal cases such as Roe versus Wade in 1973, a famous American lawsuit. Jane Roe filed a suit to challenge the constitutionality of Texas abortion laws, which at that time made abortion a crime, except if it was performed to save the mother's life. The U.S. Supreme Court chose to sidestep the issue of when the fetus becomes a person because of the sheer complexity of the religious, ethical, and philosophical implications. Instead, the majority of the Court decided to recognize the woman's fundamental right to privacy and to control her own body, including making the decision to terminate a pregnancy. The majority of the Court acknowledged that although maternal health was important, protecting the potential of human life was also an important factor [15]. The state's interest in fetal life becomes compelling when the fetus is declared viable, referring to the fetus' ability to be "capable of a meaningful life outside the mother's womb" [15]. After viability, the protection of the fetus becomes at least as important as the protection of the mother's health. The Supreme Court's decision has allowed the legal prohibition of third trimester abortions in many American states, except when medically necessary for the mother, and has greatly influenced all cases pertaining to unborn children, for example on statutes of wrongful death and homicide [15].

So far, the threshold for viability has been set around the beginning of the third trimester. But viability cannot be exactly defined. There is a great deal of variation in outcomes for extreme preterm infants born "at the threshold of viability." A fetus can survive as early as 22 weeks of gestation outside the womb; however, it does not survive without medical help. To survive, it needs a full neonatal intensive care unit with incubators, ventilator support, and parenteral nutrition. Thus, early third trimester viability is extremely technology-dependant. Viability in rich countries versus poor countries is therefore not equivalent. These differences raise questions as to whether a fetus in an American womb is more of a person than a fetus with the same gestational age and characteristics in an African womb. Should the socio-economic context have such a radical influence on moral rights and protection?

Survival becomes a possibility in high resource countries at around 22 weeks of gestation, even though the chances of survival are poor. Other fetal characteristics will increase potential survival: girls survive more than boys, singletons more than

twins, etc. Should personhood be different for girls and boys, or for twins? Also, in rich countries, there are large variations of practice for neonates at the threshold of viability. Survival and consequently viability will be influenced by the attitudes of physicians and professional guidelines. Some countries, such as Japan, are more aggressive with treatment and offer life sustaining interventions at 22 weeks of gestational age. Others, such as the Netherlands, generally do not intervene before 25 weeks of GA. Should public policy, or professional guidelines determine when one becomes a person? Should viability be defined by the youngest baby ever saved? If, in the future, maintaining life outside the womb becomes possible before viability, this new reality may also affect our idea of what is morally or ethically permissible. Viability therefore constitutes a complex—and messy—threshold for personhood.

3.4 Sentience and Consciousness

Some scholars define personhood as the emergence of sentience and consciousness. This definition may comprise wakefulness, somato-sensory awareness, hearing and seeing, awareness of smell, self-awareness, purposeful behavior, memory, internal perspective, and language. There is some evidence that neonates can display all these components and signs of consciousness [16]. Neonates remember rhythmic sounds and vowels to which they have been exposed during fetal life. At the end of the 23rd post menstrual week, thalamocortical afferents accumulate in the superficial part of the subplate zone: it has been documented in the visual, frontal, auditory, and somatosensory cortex [17, 18]. Sounds can be heard from around 23 weeks of gestational age. A histological study of prestriate visual projections in human fetuses, each at a different developmental age, concluded that thalamic projections reach the visual cortex at 21–25 weeks of post conceptional age [17–19]. Based on the above findings, at around six months of age, the fetus is able to receive stimuli and inputs from the environment around him. The fetus becoming self-aware and able to experience perceptions and reactions is a phenomenon known as fetal sentience.

For some people, consciousness defines human viability and for others, personhood. This theory of personhood is appealing for many reasons. It is demonstrable and offers some conceptual stability. Contrary to the viability argument already discussed, fetal development is universally similar. There is no reason to believe that significant variability exists between individuals of different socio-economic backgrounds, or from different countries. Also, while a viability threshold is unequal and constantly redefined by advancing technologies, our accepted understandings of brain development appear more stable and less likely to change with time. Finally, supporters of the idea of fetal sentience as the criterion for personhood believe that this more scientific approach can be more easily accepted by all and truly provide a unified answer to the question of when one becomes a person.

3.5 Birth

In a book called The Moral Relevance of Birth, Warren argues that "birth remains the most appropriate place to mark the existence of a new legal person" [20]. But for many philosophers, birth is irrelevant to moral rights. For some, those rights are present at conception; whether the individual is in the womb or outside it is not truly relevant. For others, these rights are acquired at viability or when the fetus becomes sentient, events that are much anterior to birth in most cases. Those who believe that birth makes no difference to personhood point out the conflict between currently occurring late term abortions and prohibited neonatal infanticides. For those who accept third trimester abortions, there has to be significance to birth or how else does one differentiate between abortion and infanticide? Some scholars, however, have argued that both are acceptable to a certain degree [21].

In many countries, a fetus becomes a person, legally, at the moment of birth. At that moment, the baby becomes a full-fledged citizen, endowed with legal rights that should be no different from those of any other citizen. In the medical context, these rights are said to be identical to those of any other vulnerable, incompetent patient who lacks decision-making capacity. This instantaneous transition from intrauterine to extrauterine life has important legal implications. For example, in Canada, maternal autonomy is paramount [22]: the fetus' only rights are to inherit, if born alive, or to sue for avoidable insults that may have been experienced during the pregnancy [23]. On the other hand, the fetus has no right to life. Indeed, abortion at any gestational age is not illegal in Canada [24] and, moreover, a father cannot legally prevent the abortion of the fetus by the woman carrying his baby [25]. If a fetus dies in utero from professional negligence [26], or because of battery to the woman, the person causing the injury can be prosecuted for the injury to the woman, but cannot be charged with murder or homicide, because the fetus is not considered to be a person [27]. In Canada, as the highest judicial Court has repeatedly ruled, despite varying beliefs about when the fetus becomes a person, a fetus acquires legal rights only if born and born alive. The same appears true in many other countries but is not universal. A growing number of legal cases throughout the USA show a trend toward forced treatment of pregnant women: for example, forced cesarean sections, mandatory diet restrictions, and incarceration for failing to follow medical advice. In countries where a fetus becomes a person at birth, certain paradoxes are observed. A fetus close to term can die in utero because his mother refuses a fetal transfusion, a somewhat simple technique that causes minimal harm to the woman, but if that same baby were born, transfusion would be legally enforced to avoid death or significant harm to the child. Moral opinion on the forced treatment of pregnant women is sharply divided. All parties agree that every person has a fundamental right to freedom of choice and control over their own body. But those who support forced treatment on pregnant women over and against their will—for example in cases of drug abuse or anti-HIV medication—claim that society has a duty to protect the future child. Is the future child a person

that has the same rights as his mother? The answer to this question will change the ethical analysis of forced treatment of pregnant women.

3.6 Human Relationships

Feminists over time have influenced the personhood debate, especially in connection to abortion. While some feminists [2, 20, 28] defend views comparable to non-feminist philosophers, others, such as Susan Sherwin, a philosopher, have defended the "relational" conception of personhood [29]. This view rests on the relationship between the mother and the fetus, recognizing that it is the only direct relationship the fetus can have: "Their very existence is relationally defined, reflecting their development within particular women's bodies: that relationship gives those women reason to be concerned about them." [30]. Feminists consider the actual concerns that women face in their decision-making, such as their feelings about the fetus, relationships with partners and other children, and their other obligations [30]. For many feminists, the fetus exists only in relationship to its mother, on whom they are entirely dependent for survival. Basically, the feminist position is that the woman carrying a fetus decides when it becomes a person: "Hence the specific status of a fetus will vary according to the value ascribed to it by the woman in whose womb it is developing." [29]. As Sherwin adds, "the fact that fetal lives can be neither sustained nor destroyed without affecting the women who support them implies that whatever value others may attach to fetuses generally or to specific fetuses individually should not be allowed to outweigh the ranking that is assigned to them by the pregnant women themselves" [30]. In practice, we can see many examples of this relationship. Some women who become pregnant after a rape never consider their fetus to be a person and request abortion. Conversely, some women who have recurrent miscarriages may say "I had four children and they all died". For feminists, personhood becomes a matter of social relationship.

These relationships will be influenced by culture and the fragility of the life of fetuses or neonates. Some women give the child a name only after 12 weeks of pregnancy, or after the first ultrasound, or after the genetic screening tests results are normal. They consider their fetus to be a person when their life is unlikely to be threatened. In some countries, babies who don't breathe immediately after birth are left to die; many are full term babies who need only a few assisted breaths to survive intact. Five to ten percent of babies are born this way and need a little help to start breathing on their own. In many countries, a neonate receives the same urgent care as would an older child only after he has "declared himself" and breathed at birth. In some cultures, children get named after their first weeks of life, when their survival is more likely. In all these cases, neonates seem to be

considered morally different from older children. Neonates seem to become persons —who have the same rights as any other citizen—once it seems likely they will survive and will develop relationships with others.

3.7 Unique Human Characteristics

For some, a human being needs to demonstrate certain unique characteristics to be considered a person. For Peter Singer, one of the most influential contemporary philosophers, a neonate becomes a person when he can start interacting with his environment. Peter Singer is not against abortion because, in his view, being alive and being human do not automatically give an embryo or fetus a right to life. For him, an embryo, a fetus, and even a neonate are not yet persons because they lack essential characteristics of personhood, mainly, rationality, autonomy, and self-consciousness [31].

Other philosophers, such as Tooley [32] and MacMahan [33] also believe that personhood is gradual, that it occurs during infancy rather than at birth. For them, a neonate becomes a person at a certain time in development, for example at the first smile or with the emergence of language. This view has widely been criticized by other academics who claim that even humans lacking these characteristics retain the same moral significance. For them, merely being a member of a species that typically exhibits rationality is enough. Singer disagrees that all human beings have superior value, labeling this attitude Speciesism. On the other hand, he believes that persons (by his definition) are equal, but that non-human animals also have value. For example, he is against eating meat from animals that were made to lead a miserable life so that their flesh can be consumed by humans at the lowest possible cost.

Although this extreme view of personhood has widely been criticized, there is evidence that some human beings are not treated like others. The philosopher Savulescu [34] has pointed out that many think that humans who lack the most important mental characteristics possessed by typical adult humans also lack some of their rights, claims, and interests. For example, it is widely accepted that brain-dead humans in intensive-care units have significantly weaker claims to life-sustaining treatment than ordinary living persons. When the brain dies, everything that matters in the life of that person also seems to die. We harvest organs from brain dead humans and give them to other human beings. Also, even permanently unconscious persons who do not meet the criteria for brain death are often thought to have lost many of their interests and may have life sustaining interventions withheld or withdrawn.

Are neonates considered persons? Legally, they are. But are they “full persons”? Historically, they have not always benefited from the same protection that older or adult patients have. In many societies, infanticide was tolerated if not permitted, whereas homicide was banned. Considering a category of human beings as being

lesser persons than others leads to many injustices. In the past, some groups heavily discriminated against were granted a second-tier citizen status, while others were not considered persons at all: women, homosexuals, the disabled, and African-Americans, for example. Much has improved over the last few decades, although some discrimination remains.

Despite vivid criticisms of Singer's opinions, we have observed over the years in our practice and empirically demonstrated in our research that neonates are treated differently compared to older persons. We will examine this discrepancy in the fourth chapter of this book. This discrepancy may be observed in unequal pain treatment, variations of practice for neonates, and end-of-life decisions. Also, economic analysis of neonatal intensive care has often emphasized how expensive it is, when in fact it is a much better value in terms of resources spent than is adult intensive care [35]. Neonatal mortality in low-resource countries is staggeringly higher than in wealthier nations. However, until recently there were no worldwide attempts to improve this situation. Most of the resources were put into saving older children or "productive" adults.

Factors that have contributed to the perceived difference in moral status between neonates and older persons are many and probably include religious rites and previously high infant mortality rates. Until the late twentieth century, most parents experienced the death of at least one newborn or infant. Is it possible that to survive the common reality of infant death, some protective cultural and emotional mechanisms in the form of moral (philosophic) differentiation of the newborn from older people might have been selected? Waring, a philosopher, remarks that the way we value a person is indicated by how we react to their death. Indeed, feelings of tragedy, loss, and sharp regret are often more appropriate responses to the deaths of younger people than to those of very old persons [36]. One might view the deaths of older people as tolerable. Indeed, it is not rare to hear that it is "better this way, nature took its course," or that "he/she lived long enough" for an older individual. Similar statements such as "it is better this way, nature took its course" and "at least he/she did not suffer too long" are also said for the death of neonates. Maybe a newborn infant has not yet lived long enough to justify feelings of tragedy and regret. Do we have feelings of detachment that desensitize us to the loss of newborns? Our society has evolved such that individuals with disabilities and psychiatric illness are now integrated into society, although at times imperfectly. Wheelchair ramps are common, and seeing-eye dogs are admitted in all public institutions. Persons who live with deafness or with other limitations are often able to work. Will our society evolve to find the death of premature infants less acceptable, the way we now find that the death of a three-year-old a tragedy, something that "is not supposed to happen"?

In conclusion, the debate on personhood and everything that it influences is far from resolved. All of the tentative definitions of personhood described in this chapter have potential impacts on reproductive research, on fundamental research which uses embryos or embryonic stem cells, on abortion laws, on reproductive technologies and their regulation, on the number of implanted embryos during IVF, and on procedures and programs for prenatal diagnosis, to name only a few. All of

these areas of modern life affect individual people and families, with considerable impact on health outcomes. In addition, mounting evidence shows that, even when we move past the fetus and the abortion debate, differential treatment still seems to be occurring between neonates and older patients, in society, and among health care professionals. While likely failing to provide us with a complete answer, the detailed analysis of the personhood concept and how it is applied in various cultures will most likely allow further analysis and a better understanding why neonates and older children are treated differently and why neonates might be perceived as being a different kind of person. By having a better understanding of the moral status of neonates, its origin, legitimacy, and moral acceptability, we hope to enable easier navigation through these controversial issues. Issues such as abortion, a pregnant woman's right to refuse treatment, and her responsibilities and liabilities during pregnancy may be illuminated, even as new issues arise from future technological and societal advances. It may be that our evolutionary "wiring" has made us inherently tolerant of neonatal deaths, and the relative devaluation of the newborn could be more nature than nurture. Considerations of personhood will continue to be important for decision-making for fetuses, pregnant women, and newborns. Will our society's understanding of morality change with regard to the relative devaluation of neonates? Only time will tell.

References

1. Kerckhove LF, Waller S. Fetal personhood and the sorites paradox. J Value Inq. 1998;32:175–89.
2. Warren MA. On the moral and legal status of abortion. The Monist. 1973;57:43–61.
3. Larmer R. Abortion, personhood and the potential for consciousness. J Appl Philos. 1995;12:241–52.
4. Beauchamp S, Lance JM. Fecondation in vitro: portrait comparatif de pays d'Europe et d'Océanie. (In vitre fertilization: comparative profile European and Oceania countries. Montréal, Agence d'évaluation des technologies et des modes d'intervention en santé (AETMIS). 2009.
5. Penner PS, Hull RT. The beginning of individual human personhood. J Med Philos. 2008;33:174–82.
6. Gatti MG, Becucci E, Fargnoli F, Fagioli M, Aden U, Buonocore G. Functional maturation of neocortex: a base of viability. J Matern Fetal Neonat Med Off J Eur Assoc Perinat Med Fed Asia Oceania Perinat Soc Int Soc Perinat Obstet. 2012;25(Suppl 1):101–3.
7. Ratzinger CJ, Bovone A. Instruction on respect for human life in its origin and on the dignity of procreation. Congregation Doctrine Faith Feast Chair St. Peter Apostle Rome February. 1987;22.
8. Coward HG. Hindu ethics: purity. Abortion, Euthanasia: State University of New York Press; 1989.
9. Dorff EN. The Jewish tradition: religious beliefs and healthcare decisions. In: Park ridge center for the study of health, faith and ethics.
10. Edwin R. Dubose MJP. The Jehovah's witness tradition: religious beliefs and healthcare decisions. In: Park ridge center for the study of health, faith and ethics.
11. Waugh EH. The Islamic tradition: religious beliefs and healthcare decisions. In: the park ridge center for the study of health, faith, and ethics.

12. Moro T, et al. Neonatal end-of-life care: a review of the research literature. J Perinat Neonatal Nurs. 2006;20(3):262–73.
13. HEW support of research involving human in vitro fertilization and embryo transfer. In: U.S. Department of Health E, and Welfare, editors. Washington, DC: U.S. Government Printing Office; 1979.
14. Fausset. Abortion wars: "Fetal pain" bill advances in Georgia. LA times 2012.
15. Peterfy A. Fetal viability as a threshold to personhood. A legal analysis. J Leg Med. 1995;16:607–36.
16. Lagerkrantz H. The birth of consciousness. Early Hum Dev. 2009;85:S57–8.
17. Kostovic I, Jovanov-Milosevic N. The development of cerebral connections during the first 20–45 weeks' gestation. Seminars Fetal Neonatal Med. 2006;11:415–22.
18. Kostovic I, Rakic P. Development of prestriate visual projections in the monkey and human fetal cerebrum revealed by transient cholinesterase staining. J Neurosci Off J Soc Neurosci. 1984;4:25–42.
19. Lee SJ, Ralston HJ, Drey EA, Partridge JC, Rosen MA. Fetal pain: a systematic multidisciplinary review of the evidence. JAMA. 2005;294:947–54.
20. Warren MA. The moral significance of birth. Hypatia. 1989;4:46–65.
21. Tooley M. Abortion and infanticide. Philos Public Aff. 1972;2:37–65.
22. Weiler KM, Catton K. The unborn child in Canadian law. Osgoode Hall Law J 1976;14:643–59.
23. Klasing MS. Death of an unborn child: jurisprudential inconsistencies in wrongful death, criminal homicide, and abortion cases. Pepperdine Law Rev. 1995;22:933–79.
24. Morgentaler Rv. 3 S.C.R. 463 Supreme Court of Canada; 1993.
25. Tremblay Dv. 2 S.C.R. 530 Supreme Court of Canada; 1989.
26. Sullivan Rv. 1 S.C.R. 489 Supreme Court of Canada; 1991.
27. McConnell ML. Sui generis: the legal nature of the fetus in Canada. Can Bar Rev. 1991;70:548–56.
28. Thomson JJ. A defense of abortion. Philos Public Aff. 1971;1:47–66.
29. Gibson S. The problem of abortion: essentially contested concepts and moral autonomy. Bioethics. 2004;18:221–33.
30. Sherwin S. No longer patient: feminist ethics and health care. Philadelphia: Temple University Press; 1992.
31. Singer P. Practical ethics, 2nd ed. Cambridge University Press; 1993.
32. Tooley. Abortion and infanticide, philosophy and public affairs; 1972. pp. 37–65.
33. McMahan. The ethics of killing: Problems at the margins of life. Oxford University Press; 2002.
34. Savulescu. Brain damage and the moral significance of consciousness. J Med Philos 2009.
35. Meadow W, Lagatta J, Andrews B, Lantos J. The mathematics of morality for neonatal resuscitation. Clin Perinatol 2012;39:941–56.
36. Waring DR. Fair innings and need over lifetime. In: Springer, editors. Medical Benefit and the human lottery: an egalitarian approach to patient selection. Toronto, Ontario, Canada: York University; 2004:89–97.

Chapter 4
Neonates Are Devalued Compared to Older Patients

Annie Janvier, Carlo Bellieni and Keith Barrington

Abstract According to bioethical principles, babies and older patients should be treated according to the same standards. In practice, newborn infants are treated differently in ways which show that they are valued less than older individuals. We provide six specific examples of this differential treatment and analyse the possible consequences of this devaluation.

The Neonatal Resuscitation Programme textbook, which is the standard neonatal resuscitation text used in North America and many other countries states: "The ethical principles regarding resuscitation of newborns should be no different from those followed in resuscitating an older child or adult [1]. The Nuffield report, created following a multidisciplinary consultation process involving physicians, parents, ethicists and lawyers in the UK [2] specifies that "independent of gestational age, we consider, for example, a child of six days, months or years to be worthy of equal consideration". According to such principles, babies and older patients should be treated according to the same standards. In practice, newborn infants are treated differently in ways which show that they are valued less than older individuals. We provide six specific examples of this differential treatment and analyse the possible consequences of this devaluation.

A. Janvier (✉)
Department of Pediatrics and Clinical Ethics, University of Montreal, Neonatology and Clinical Ethics, Sainte-Justine Hospital, 3175 Côte-Sainte-Catherine, QC H3T 1C4 Montréal, Canada

C. Bellieni
Department of Pediatrics, Obstetrics and Reproduction Medicine, University of Siena, Via Banchi Di Sotto 55, 53100 Siena, Italy

K. Barrington
Department of Pediatrics, University of Montreal, 2900 Boulevard Edouard-Montpetit, QC H3T 1J4 Montréal, Canada
e-mail: keith.barrington@umontreal.ca

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_4

4.1 Pain Treatment

Pain is not good for patients of any age, but the consequences of pain for neonatal patients are perhaps more important than for older patients. Pain has serious short and long term consequences in the newborn. In the short term, it provokes an acute increase of intracranial pressure and blood pressure, oxygen desaturation, and a release of free radicals [3]. Subsequent metabolic and endocrine responses lead to hyperglycemia, and cardiovascular and respiratory instability. Long term effects of untreated pain include altered responses to pain in older life, extending into adolescence [4]. And yet, despite these well-known consequences, analgesia is grossly underused for newborn infants [5–7]. As an example, even though caregivers recognize that endotracheal intubation is painful, as many as 87 % of units do not premedicate before an endotracheal intubation in non-emergency conditions. Analgesia is rarely given to newborns for lumbar punctures and even sometimes chest tubes are inserted without local or systemic analgesia [5, 7]. This would be inconceivable in older patients, despite there being much less evidence of long term adverse consequences, perhaps because they can physically protest during painful procedures. Although there are concerns regarding the overutilization of opioids, in neonatology underutilization of analgesia is more of a concern than overutilization.

For more minor procedures, oral administration of a sucrose solution is known to be effective, but even this inexpensive simple intervention is underutilized [8]. Rigorous studies are performed to find out how to better alleviate or eliminate this pain. Unfortunately, during many of these clinical trials, babies in the control groups receive no analgesia [9, 10]. Placebo groups in such studies raise serious ethical concerns, but have unfortunately been included in 89 % of neonatal analgesia studies [11]. Some of these studies examine intramuscular injections or heelstick punctures, which might be considered to be relatively minor, but even very invasive procedures such as circumcision have undergone clinical trials where no analgesia was provided to the control-babies [12, 13]. The declaration of Helsinki [14] requires that in clinical trials any new treatment or drug should be compared to standard of care, if any exists, and not to a placebo. Ironically, prolonged pain and suffering are often used as reasons for withholding active intensive care in preterm infants. Intolerable, persistent and untreatable pain is an ethically appropriate reason for limiting interventions. Failure to prevent and treat pain is not.

4.2 Nursery Environment

Noises in an incubator are far higher than those allowed for other patients in the hospital. The incubator fans produce noises of 45 dB or even higher than this level in some cases [2, 15] while in hospital the background noise should not exceed 35 dB [16]. It is hard to understand why the maximal noise threshold tolerated in nurseries is higher than 35 dB.

4.3 Variations of Practice for Neonates

Variations of practices exist everywhere in medicine. Usually, these variations are due to random factors, economic incentives, availability of resources or a different combination of therapies. Variations of outcomes for preterm infants are frequently due explicit policies or philosophies. Interestingly, different industrialized countries, which all have access to the same information from the medical literature, come to very different conclusions. Survival for babies born at 24 weeks is 81 % in Japan, 60 % in Canada, 33 % in the United Kingdom, and is as low as zero in some centers in the Netherlands [17]. Those who are more aggressive obviously think that intervention is ethically appropriate. Those who defend less aggressive treatment think that over-treatment is more problematic than under-treatment. Both argue that they are doing what is in the best interest of neonates and their families. Both invoke local or national policies, based on local or national outcome data, to justify their approach. In the end, values become facts, which reinforce the values and policies. Policy statements for preterm infants often state survival and handicap as justification for optional intervention. The fact that such outcome statistics would not be used to justify non-intervention approaches at later ages suggests that some other powerful factors are at work [18, 19].

4.4 Resuscitation and End of Life Decisions

In empiric research, physicians and students were more frequently willing to withhold resuscitation and intensive care from sick neonates than from older children or adults with similar or even much worse prognoses [20–25]. Many healthcare providers thought that resuscitation was in the best interests of a preterm infant born at 24 weeks gestation. Interestingly, even more thought that resuscitation was in the best interests of a 2 month-old or a multiply disabled 7 year-old who were described as having similar or worse outcomes [20–25]. More than 50 % of those who thought resuscitation to be in the best interest of the 24-weaker also said that they would accept a parental decision to provide comfort care [23]. On the other hand, a minority would do so for the 2 month old and the disabled 7 year-old. These findings were found in multiple countries with different healthcare and cultures [20–25]. Such responses suggest that decisions for preterm newborns are made using different values than those for older children. Many clinical guidelines propose withholding interventions for neonates as a result of poor survival at mortality rates that have never been used to propose withholding resuscitation later in life [26, 27].

These observations extend to resuscitation decisions in real life. For example, survival to hospital discharge of adults after an out of hospital cardiac arrest is about 7 % with a high risk of subsequent disability [28]. This outcome is much worse than

survival/disability outcomes of babies born at 23 weeks of gestation. Yet, the former routinely have resuscitation instituted, whereas among the latter in many countries, resuscitation is actively discouraged, and in others it is considered optional. For example, among 12,390 adults with cardiac arrest recently studied, 159 were lucky enough to have a witnessed cardiac arrest in public close to an automatic defibrillating device and had it applied by a bystander, which increased their survival to about 34 % [28]. This survival rate is considered sufficiently positive to drive the widespread purchase, installation and maintenance of automated external defibrillators throughout North America. In stark contrast, many clinical guidelines propose avoiding the resuscitation of 23 week infants, who have a similar survival rate and better long term outcomes if active care is instituted [19].

For older children, near certain death or profound disability seem necessary before withholding or withdrawing LSIs are considered by HCP [29]. For the newborn, end of life decisions are generally taken considering not only babies' best interest, but also the parents' interests [30]. In a questionnaire study [31], medical respondents were more likely to wish to resuscitate a "precious IVF" baby of an older mother than a baby of an young single mother. Are physicians who care for older incompetent patients also influenced by older maternal age for life and death decisions of their patients?

4.5 Economic Analysis of NICUs Compared to Other ICUs

NICUs cost-effectiveness has been highly scrutinized and seems to be held to higher standard than ICU for older patients. Some policy statements regarding counseling of women at risk of delivering preterm have used the just distribution of resources as a reason for being cautious in resuscitating fragile preterm infants [26]. While it is true that NICU care for neonates is expensive, so is intensive care for older patients. Surprisingly, in spite of these high costs, every study of the cost-effectiveness of NICUs shows them to be far more cost effective than many widely accepted treatments [32]. Twenty-nine percent of adult ICU bed days are used by patients who die, compared to 8 % in the NICU [33]. NICUs are cost effective because most of the money is spent on babies who survive. Even when the long term costs of survivors who are disabled are included, the advantage of neonatal units still holds. A standard measure of cost-effectiveness that combines survival and quality of life is dollars per quality-adjusted life-year (QALY). Most NICU survivors live a long time without serious impairments and economically productive lives. Thus, the high initial costs are amortized over a lifetime and lead to relatively low figures on dollars/QALY. The quality adjusted costs for life of an infant born at 24 weeks has been calculated at around 6000$ US/QALY [34]. Most critical interventions for adults cost more than 70,000$/QALY.

4.6 Neonatal Mortality in Poor Countries

The former discussions have mainly focussed on neonates in industrialized countries. Unfortunately, most babies in the world do not have access to an NICU. Each year, 10 million children die, of these, 7 million are less than a year old and 4 million are neonates. 10,000 neonates die everyday. The majority are term neonates or neonates older than 35 weeks of GA who would not need intensive care to survive [35]. One of the UN millenium development goals established in 2000 was to decrease child mortality and maternal health. Improving neonatal mortality/health was not described as a goal. Child and post-neonatal mortality have decreased, but the goals that were set were not achieved [36]. This is mainly because neonatal mortality has not changed at all since 2000. The majority of neonatal deaths in the world are due to lack of access to basic medical care. About 3 million of those deaths could be prevented with simple inexpensive interventions: breast feeding, basic temperature management, early treatment of infections and more importantly, education. Five percent of term babies need a little help to start breathing on their own. In many countries, babies who don't breathe immediately after birth are left to die. Most of these deaths are in the first month of life. Saving one life for a neonate in Zambia costs about 200$ and 5$ per disability-adjusted life-year averted [37]. Saving lives for older children is more expensive, technologically more difficult and costly, yet it seems to be favored over saving the lives of newborn infants.

Neonates are favoured in every equation of the QALY analysis, because of their young age. But are they too young to be worthy of as much resources as older children? Of interest, physicians and parents in these poor countries have been interviewed. Many see these neonatal deaths are considered to be "natural": "We try not to make waste for the family. It is better for them to go for a new baby" [38, 39]. The Disease Control Priorities Project is an ongoing effort to produce evidence-based analysis and resource materials to inform health policymaking in developing countries and ultimately lead to improving the health of people in developing countries. In one of their publications, authors discuss fetal and neonatal mortality. When calculating how to compare deaths at different ages, they suggest a calculus of ALP (Acquisition of Life Potential), where the death of a 20 year old is worse than the death of a neonate: "An individual life acquires value only as it acquired self-awareness […] an individual life acquired value as it develop bonds with others" [40]. This analysis was based on their own appreciation of the moral differences between neonates and older children, but also on other sources. An Institute of Medicine (1985) review of vaccine development priorities judged that the loss from a death at age 20 should be about two times that from an infant death [41]. However, some studies in lay individuals suggest a value closer to three or four times [42, 43].

4.7 Why Are These Differential Considerations Tolerated for Neonates?

Neonates are not like older patients. In a matter of minutes, just after birth, they acquire legal status. Sick babies in the NICU have never been home, never experienced a normal family life, they have no self-awareness. For some philosophers, babies are not full persons, and therefore they have a lower moral status than older babies [44, 45]. This attitude seems to be present even in global health. Is it easier to see a newborn baby die compared to a child who is 3 years old or an adult who is 20 years old? This systematic devaluation of the newborn could be due to some deep-rooted anthropological, cultural, social and evolutionary factors. Until the late twentieth century, most parents experienced the death of at least one newborn or infant. Perhaps the commonness of infant death led to protective cultural and emotional mechanisms. Waring, a philosopher, remarks that the way we value a person is indicated by how we react to their death, "feelings of tragedy, evil, loss and sharp regret are supposedly more appropriate responses to the deaths of younger people" [46]. One might view the deaths of older people as tolerable. Similar statements of "it is better this way, nature took its course" and "at least he/she did not suffer" are held for the elderly and preterms alike. Maybe the neonate has not yet lived long enough to justify feelings of tragedy and regret secondary to instilled feelings of detachment that desensitize society from the loss of newborns? [47]. If we are to apply principle of justice to patients in our society, we should treat persons who have similar outcomes similarly. The differential treatment of newborns can only be justified if neonates are considered to be morally different to older persons.

4.8 Conclusion

Decisions to treat newborns differently suggest that we value their lives differently. There are three possible responses to this: we should intervene at the same predicted outcomes as we currently do for older patients, we should continue to treat them differently, or we should intervene for older patients at the same predicted outcomes as we do for newborn. We favor the first position. The devaluation of newborns has led to unacceptable rates of pain control, policy statements that produce large variations of practices and unacceptable stagnation of neonatal mortality in poor countries. Fragile lives should not have fragile rights, even if they are seen as being "not like other lives yet". Often, practitioners are unaware of their own implicit valuations. In many cases, careful analysis of such implicit valuations will lead us away from treating newborns differently. Such analysis may often place us in conflict with prevailing social and professional norms.

References

1. American Academy of Pediatrics. Neonatal Resuscitation Program (NRP) Textbook—6th edition. 2010.
2. The Nuffield Council on Bioethics. ccd web version 22 June 2007. 2007 [cited; Available from: http://www.nuffieldbioethics.org/sites/default/files/CCD web version 22 June 07 (updated).pdf.
3. Bellieni CV, Iantorno L, Perrone S, Rodriguez A, Longini M, Capitani S, et al. Even routine painful procedures can be harmful for the newborn. Pain. 2009;147(1–3):128–31.
4. Peters JW, Schouw R, Anand KJ, van Dijk M, Duivenvoorden HJ, Tibboel D. Does neonatal surgery lead to increased pain sensitivity in later childhood? Pain. 2005;114(3):444–54.
5. Carbajal R, Rousset A, Danan C, Coquery S, Nolent P, Ducrocq S, et al. Epidemiology and treatment of painful procedures in neonates in intensive care units. JAMA. 2008;300 (1):60–70.
6. Losacco V, Cuttini M, Greisen G, Haumont D, Pallas-Alonso CR, Pierrat V, et al. Heel blood sampling in European neonatal intensive care units: compliance with pain management guidelines. Arch Dis Child Fetal Neonatal Ed. 2011;96(1):F65–8.
7. Johnston C, Barrington KJ, Taddio A, Carbajal R, Filion F. Pain in Canadian NICUs: have we improved over the past 12 years? Clin J Pain. 2011;27(3):225–32.
8. Simons SH, van Dijk M, Anand KS, Roofthooft D, van Lingen RA, Tibboel D. Do we still hurt newborn babies? A prospective study of procedural pain and analgesia in neonates. Arch PediatrAdolesc Med. 2003;157(11):1058–64.
9. Yilmaz F, Arikan D. The effects of various interventions to newborns on pain and duration of crying. J ClinNurs. 2011;20(7–8):1008–17.
10. Cong X, Ludington-Hoe SM, Walsh S. Randomized crossover trial of kangaroo care to reduce biobehavioral pain responses in preterm infants: a pilot study. Biological research for nursing. 2011; 13(2):204–16.
11. Harrison D, Bueno M, Yamada J, Adams-Webber T, Stevens B. Analgesic effects of sweet-tasting solutions for infants: current state of equipoise. Pediatrics. 2010;126(5): 894–902.
12. Brady-Fryer B, Wiebe N, Lander JA. Pain relief for neonatal circumcision. Cochrane Database Syst Rev. 2004; (4):CD004217.
13. Schiavenato M, Butler-O'Hara M, Scovanner P. Exploring the association between pain intensity and facial display in term newborns. Pain research &management: the journal of the Canadian Pain Society = journal de la societecanadienne pour le traitement de la douleur. 2011; 16(1):10–2.
14. World Medical Association. WMA Declaration of Helsinki—Ethical Principles for Medical Research Involving Human Subjects. 2008 [cited; Available from: http://www.wma.net/en/30publications/10policies/b3/index.html. 2008.
15. World Health Organization. WHO Guidelines for Community Noise. 2010 [cited; Available from: http://www.who.int/docstore/peh/noise/ComnoiseExec.htm. 2010.
16. Philbin MK, Robertson A, Hall JW 3rd. Recommended permissible noise criteria for occupied, newly constructed or renovated hospital nurseries. Adv Neonatal Care. 2008;8(5): S11–5.
17. Janvier A, Lantos J. Variations of practice in the care of extremely preterm infants. In: Diekema D, Mercurio M, Adam M, editors. Clinical Ethics in Pediatrics, a Case-Based Textbook: Cambridge University Press; 2011. p. 94–100.
18. Janvier A, Barrington KJ, Aziz K, Lantos J. Ethics ain't easy: do we need simple rules for complicated ethical decisions? Acta Paediatr. 2008;97(4):402–6.
19. Dupont-Thibodeau A, Barrington KJ, Farlow B, Janvier A. End-of-life decisions for extremely low-gestational-age infants: why simple rules for complicated decisions should be avoided. SeminPerinatol. 2014;38(1):31–7.

20. Laventhal N, Spelke MB, Andrews B, Larkin LK, Meadow W, Janvier A. Ethics of resuscitation at different stages of life: a survey of perinatal physicians. Pediatrics. 2011;127 (5):e1221–9.
21. Armstrong K, Ryan CA, Hawkes CP, Janvier A, Dempsey EM. Life and death decisions for incompetent patients: determining best interests–the Irish perspective. Acta Paediatr. 2011;100 (4):519–23.
22. Janvier A, Leblanc I, Barrington KJ. Nobody likes premies: the relative value of patients' lives. J Perinatol. 2008;28(12):821–6.
23. Janvier A, Leblanc I, Barrington KJ. The best-interest standard is not applied for neonatal resuscitation decisions. Pediatrics. 2008;121(5):963–9.
24. Hansen TW, Janvier A, Aasland O, Forde R. Ethics, choices, and decisions in acute medicine: a national survey of Norwegian physicians' attitudes. PediatrCrit Care Med. 2013;14(2): e63–9.
25. Hagen EM, Therkelsen OB, Førde R, Aasland O, Janvier A, Hansen TW. Challenges in reconciling best interest and parental exercise of autonomy in pediatric life-or-death situations. J Pediatr. 2012;161(1):146–51.
26. Jefferies AL, Kirpalani HM. Counselling and management for anticipated extremely preterm birth. Paediatrics Child Health. 2012; 17(8):443–6.
27. Kaempf JW, Tomlinson M, Arduza C, Anderson S, Campbell B, Ferguson LA, et al. Medical staff guidelines for periviability pregnancy counseling and medical treatment of extremely premature infants. Pediatrics. 2006;117(1):22–9.
28. Weisfeldt ML, Everson-Stewart S, Sitlani C, Rea T, Aufderheide TP, Atkins DL, et al. Ventricular tachyarrhythmias after cardiac arrest in public versus at home. N Engl J Med. 2011;364(4):313–21.
29. Fontana MS, Farrell C, Gauvin F, Lacroix J, Janvier A. Modes of death in pediatrics: differences in the ethical approach in neonatal and pediatric patients. J Pediatr. 2013;162 (6):1107–11.
30. Rebagliato M, Cuttini M, Broggin L, Berbik I, de Vonderweid U, Hansen G, et al. Neonatal end-of-life decision making: Physicians' attitudes and relationship with self-reported practices in 10 European countries. JAMA. 2000;284(19):2451–9.
31. Marcello KR, Stefano JL, Lampron K, Barrington KJ, Mackley AB, Janvier A. The influence of family characteristics on perinatal decision making. Pediatrics. 2011;127(4):e934–9.
32. Doyle LW. Neonatal intensive care at borderline viability—is it worth it? Early Hum Dev. 2004;80(2):103–13.
33. Lantos JD, Mokalla M, Meadow W. Resource allocation in neonatal and medical ICUs. Epidemiology and rationing at the extremes of life. Am JRespir Crit Care Med. 1997; 156 (1):185–9.
34. Doyle LW, The Victorian Infant Collaborative Study Group. Evaluation of neonatal intensive care for extremely low birth weight infants in Victoria over two decades: II. Efficiency. Pediatrics. 2004;113(3 Pt 1):510–4.
35. Lawn JE, Cousens SN, Darmstadt GL, Bhutta ZA, Martines J, Paul V, et al. 1 year after the lancet neonatal survival series–was the call for action heard? Lancet. 2006;367(9521):1541–7.
36. Lozano R, Wang H, Foreman KJ, Rajaratnam JK, Naghavi M, Marcus JR, et al. Progress towards Millennium Development Goals 4 and 5 on maternal and child mortality: an updated systematic analysis. Lancet. 2011;378(9797):1139–65.
37. Manasyan A, Chomba E, McClure EM, Wright LL, Krzywanski S, Carlo WA. Cost-effectiveness of essential newborn care training in urban first-level facilities. Pediatrics. 2011;127(5):e1176–81.
38. Miljeteig I, Johansson KA, Sayeed SA, Norheim OF. End-of-life decisions as bedside rationing. An ethical analysis of life support restrictions in an Indian neonatal unit. J Med Ethics. 2010;36(8):473–8.
39. Miljeteig I, Sayeed SA, Jesani A, Johansson KA, Norheim OF. Impact of ethics and economics on end-of-life decisions in an Indian neonatal unit. Pediatrics. 2009;124(2):e322–8.

40. Jamison DT, Shahid-Salles SA, Jamison J, Lawn JE, Zupan J. Incorporating Deaths Near the Time of Birth into Estimates of the Global Burden of Disease. In: Lopez AD, Mathers CD, Ezzati M, Jamison DT, Murray CJL, editors. Global Burden of Disease and Risk Factors. Washington (DC); 2006.
41. Committee on Issues PfNVD, Division of Health Promotion, Disease Prevention IoM. New Vaccine Development: Establishing Priorities: Diseases of Importance in the United States: The National Academies Press; 1985.
42. Johannesson M, Johansson PO. Is the valuation of a QALY gained independent of age? Some empirical evidence. J Health Econ. 1997;16(5):589–99.
43. Cropper ML, Aydede SK, Portney PR. Preferences for life saving programs: how the public discounts time and age. Journal of Risk and Uncertainty. 1994;8(3):243–65.
44. McMahan J. The ethics of killing: problems at the margins of life. Oxford: Oxford University Press; 2002.
45. Singer P. Practical ethics. Cambridge: Cambridge University Press; 2011.
46. Waring DWR. Medical benefit and the human lottery: an egalitarian approach to patient selection. Berlin: Springer; 2004.
47. Janvier A, Bauer KL, Lantos JD. Are newborns morally different from older children? TheorMedBioeth. 2007;28(5):413–25.

Chapter 5
Who Makes It to the NICU? The Association Between Prenatal Decisions and Neonatal Outcomes

Amélie Dupont-Thibodeau and Annie Janvier

Abstract Statistics are often used in various contexts in neonatal and perinatal medicine: in determining the efficacy of new treatments and innovations, in quantifying mortality and morbidity outcomes, in determining the success of a unit, or even in defending NICU budgets. In order to use statistics well, one has to know how to interpret them. But while they can be mathematically and conceptually complex, statistics are not purely objective: they are influenced by values and beliefs. This influence becomes particularly important when outcome data is used to make critical decisions, such as life and death decisions. In this chapter, we will discuss how perinatal mortality and health care professionals' values and beliefs intersect when examining neonatal outcomes. We will suggest a classification method for perinatal deaths that enables outcome data to be ethically transparent and accurate.

Mrs. Smith is 23 weeks pregnant. She and her husband have named their unborn child Christine. They have just discovered today that Christine has a serious heart condition called hypoplastic left heart syndrome (HLHS). They were told that the left side of the heart, the side that pumps the blood to important organs, is underdeveloped, and that this is a very serious condition. They were presented with different options. A series of surgeries exists for this condition, but they are considered palliative: they will help the heart function better, but they will not repair it. These surgeries have a survival rate of about 70 %. Survivors do not have normal cardiac function and they also have other co-morbidities. Christine could be listed for a cardiac transplantation, but a small donor is very hard to find. Because of the seriousness of the condition, another option is palliative care at birth. Christine would die in her first days of life in the neonatal intensive care unit (NICU), in her

A. Dupont-Thibodeau (✉) · A. Janvier
Neonatology and Clinical Ethics, Sainte-Justine Hospital, 3175 Chemin de la Côte-Sainte-Catherine, Montreal H3T 1C4, Canada
e-mail: amelie.du.pont-thibodeau@umontreal.ca

A. Janvier
e-mail: anniejanvier@hotmail.com

A. Dupont-Thibodeau · A. Janvier
Department of Pediatrics and Ethics Bureau, University of Montreal, 2900 Boulevard Edouard-Montpetit, Montreal H3T 1J4, Canada

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_5

mother's room on the maternity ward, or at home. Termination of pregnancy is also an option. At this point, hearing all this, the parents are overwhelmed. They have heard many statistics and are considering the options for themselves and their child.

Statistics have always played an important role in neonatal and perinatal medicine. They are used in various contexts: in determining the efficacy of new treatments and innovations, in quantifying mortality and morbidity outcomes, in determining the success of a unit, or even in defending NICU budgets. In order to use statistics well, one has to know how to interpret them. But while they can be mathematically and conceptually complex, statistics in the case above are not purely objective: they are influenced by values and beliefs. This influence becomes particularly important when outcome data is used to make critical decisions, such as life and death decisions.

When their child is at risk of death or disability, parents often have to make critical decisions with healthcare professionals. These decisions are varied in their type and complexity, ranging from a surgical intervention to withdrawal of a respirator and other end-of-life decisions. In most cases, parents have little knowledge of neonatal intensive care and very little experience in a NICU. In order to help them make decisions, it is one of our responsibilities as the team of healthcare providers to inform them and present them with accurate data. The same is true during pregnancy. Some parents, like Mr. and Mrs. Smith, are faced with difficult decisions during their pregnancy. Should they terminate the pregnancy? If they choose to continue the pregnancy, should they favor intensive care or comfort care at birth? When do they have to decide? For these difficult conversations, we usually enlighten our discussions with our knowledge and professional experience. Generally, we will inform parents of outcomes for children in similar cases. We will tell them about similar cases we have experienced, or cases we know about. We will also generally examine published and local statistics for the condition we are discussing with the parents. But these published statistics are not only objective numbers. They also reflect values, values that are not made transparent in the current literature.

In the following chapter, we will discuss how perinatal mortality and health care professionals' values and beliefs intersect when examining neonatal outcomes. We will suggest a classification method for perinatal deaths that enables outcome data to be ethically transparent and accurate.

5.1 How Babies Die Outside of the NICU

Thankfully, not every conceived baby goes to the neonatal intensive care unit (NICU). The NICU will admit around 10–15 % of babies born alive. While far from admitting the majority of babies born alive, NICU beds represent a large part of pediatric inpatient beds. When thinking of the babies who are not admitted to the NICU, one will most likely think of healthy term neonates in no distress, requiring

Table 5.1 Classification of fetal and neonatal deaths after viability occurring outside of the NICU

	Cause
Late termination of pregnancy	1. Maternal risks 2. Psychosocial 3. Fetal anomaly 4. High probability of very premature birth
Stillbirths	1. Unanticipated fetal demise 2. Anticipated fetal demise following medical withholding of intervention 3. Fetal demise following maternal-fetal intervention
Unsuccessful neonatal resuscitation	1. Unanticipated resuscitation 2. Anticipated resuscitation
Palliative care at birth	1. Prematurity 2. Serious pre-existing pathology

routine care. If these babies are born in the hospital, they usually stay with their mothers and are discharged soon after birth. But there are other neonates who are neither healthy nor admitted to the NICU. Some neonates will die without ever being admitted to the NICU. These patients are rarely part of neonatal outcome statistics. Who are the babies who never make it to the NICU? We have classified perinatal deaths and early neonatal deaths into four large categories (Table 5.1).

5.1.1 Late Terminations of Pregnancy

Late terminations of pregnancy are rare and account for only a small percentage of the total number of terminations of pregnancy, which generally occur in the first trimester. In Canada, for example, in 2012, 70.4 % of terminations occurred during the first 12 weeks of pregnancy, and only 2.2 % after 21 weeks [1]. This category is not homogeneous. The reasons for the terminations are varied, and so are the methods used for terminating the pregnancy.

(a) Reasons for termination
Late terminations of pregnancy can happen for a number of reasons. For some women, certain social and personal situations are so difficult that bringing the child to term is not an option. In other cases, it is because the mother's life is endangered by the continuation of the pregnancy and termination occurs to save the mother. For others, the pregnancy is terminated because of a fetal condition. These conditions can arise from an adverse complication of pregnancy, such a severe intra-uterine growth retardation (IUGR), a fear of prematurity and its consequences in women at high risk of delivering early, or most often the presence of one or more congenital anomalies, similar to the case described at the beginning of this chapter. The occurrence of these terminations is greatly influenced by politics, laws, and values in different

countries or institutions. In some countries such as Ireland, abortion is illegal at any gestational age, unless it is necessary to save the mother's life or unless continuing the pregnancy poses a serious risk to her health [2]. In Canada, though there is no law establishing a threshold after which termination becomes illegal, abortions are not offered in every province. In the United States, restrictions vary state-by-state, but late terminations are frequently not permitted and remain highly controversial [3].

(b) Method of termination
Not only are the reasons for termination diverse, the method of termination can also vary. Late terminations of pregnancy can be performed with or without feticide. A feticide consists of injecting potassium chloride in the heart of the fetus to cause a cardiac arrest. For example, Mr. and Mrs. Smith could choose to have a feticide before inducing labor. In this case, Christine would be born without a heartbeat. But labor could also be induced prior to fetal viability, or at a viable but early gestational age with palliative care after birth. In this case, Christine would be born alive but would most likely die quickly of both her prematurity and her heart condition.

These approaches to termination are delicate and sensitive issues, both to parents and to healthcare teams and need to be well explained and transparent. But the approaches also influence outcome statistics differently. In the case of the feticide, Christine will be counted as a stillbirth, and in the other, as a neonatal death. For parents, beyond the emotional and psychological impact, practical considerations will also matter. For example, work compensations for parents, such as sick leave, are often shorter for an in utero death than for a neonatal death.

5.1.2 Stillbirths

The second category is comprised of stillbirths. Even with our increasing technological prowess, some fetuses die in utero. With an increased ability to monitor pregnancies, fetal mortality is less common than before in industrialized countries. In the United States, for example, fetal mortality was estimated at 7.83 per 1000 live births in 1985, compared to 6.05 in 2006 [4]. Further categorization of stillbirths is important. They can be categorized as follows: (a) unanticipated fetal demise, (b) anticipated fetal demise following medical withholding of intervention (delivery or operative delivery), and (c) fetal demise following maternal-fetal interventions.

Some fetal deaths remain unanticipated and are often inevitable, but others are avoided by inducing the birth of an at-risk fetus. For example, in some cases of placental insufficiency, the fetus no longer grows and fetal demise is thought to be a significant risk. In these situations, medical induction of birth can prevent fetal death. In other cases, however, even if the medical team and the parents are aware that the fetus is at significant risk of in utero demise, induction of birth is not performed, because the team and parents choose to let "nature follow its course." The reasons for

this choice vary: some fetuses show signs of distress at a gestational age that is too early to be compatible with extra utero life, some demonstrate a level of fetal maturity that is compatible with life but with a significant risk of future mortality or sequelae, and others present a known fetal condition that will affect their future quality of life. Also, compromised fetuses often do not tolerate labor well and may develop fetal distress with contractions. Although it may be indicated to ensure survival, a cesarean section at the limits of viability increases maternal risks and future reproductive risks more than one later in pregnancy. In summary, the estimation of risks and benefits of a cesarean section sometimes points towards withholding surgical intervention when the compromised fetus has a poor predicted outcome.

A recent category of stillbirths is comprised of fetal deaths following maternal-fetal interventions. In utero interventions have evolved in the last decades. Fetal centers have developed in many academic centers. Interventions may prevent fetal demise and/or improve future quality of life. For example, in utero transfusions in cases of severe anemia can prevent fetal death. In utero repair of a meningomyelocele has been demonstrated to decrease the risk of future motor disability [5]. Even in the context of increasing success, each of these interventions still carries a risk of maternal complications and fetal death.

5.1.3 Unsuccessful Resuscitations

A third category is comprised of neonates who are born in significant distress but for whom resuscitation is unsuccessful. Although rare, these cases still occur even with significant improvement in antenatal care and neonatal resuscitation techniques.

5.1.4 Neonates Who Receive Palliative Care at Birth

A fourth and final category is comprised of babies who receive palliative care at birth. Some babies are born with a known condition or malformation, and prior to delivery the team and family choose palliative care. For others, a short resuscitation is attempted, and an inadequate response to resuscitation is followed by reorientation of care. In some centers, when comfort care is administered in the NICU, these deaths will count in the NICU mortality statistics. In other centers, comfort care can be administered in the delivery room, in the mother's room on the maternity ward, on pediatric units, or at home.

The number of babies who die outside of the NICU and way these babies die influence neonatal statistics and should be considered when studying NICU outcomes. They directly influence the denominator used to study these outcomes. For example, in units where cesarean sections for compromised extremely preterm fetuses are frequent, the babies admitted to the NICU are likely to be sicker than

those in other units where operative deliveries before 26 weeks are rare. This discrepancy may translate to a higher proportion of total NICU survivors for preterm infants admitted in the unit where earlier cesarean sections are less frequent. This proportion would probably be different if the denominator of this calculation were not the number of NICU admissions, but rather the number of viable fetuses [6].

5.2 Telling the Values from the Facts

In neonatology, most measures of survival and outcome have improved over the last thirty years. This change is largely due to significant improvement in obstetrical, neonatal, surgical, and specialized care. For example, several decades ago, Patrick Bouvier Kennedy, the son of President John F. Kennedy, died of hyaline membrane disease at 35 weeks of gestational age, an outcome that is unthinkable today [7].

In parallel, antenatal diagnosis has also improved: we can detect the majority of fetal anomalies. We are detecting them more often than previously. Now, more and more tests are being proposed to pregnant women and families during pregnancy to help diagnose conditions that would otherwise be detectable only after birth. Nuchal translucency tests, quadruple tests, fetal ultrasounds, fetal heart ultrasounds, fetal MRIs, and fetal DNA analysis are just a few examples of what is now available. Detecting fetal anomalies prior to delivery can be helpful to parents in two ways. First, parents can prepare for a neonate with a specific condition. Sometimes, early detection can ensure delivery at a tertiary care hospital. For neonates with certain cardiac anomalies, detecting the anomalies before delivery has been shown to improve outcomes [8]. Secondly, sometimes, it will lead to a termination of pregnancy or a decision to provide comfort care at birth.

During discussions with parents in the antenatal period, neonatal outcomes data need to be transparent. As clinicians, we need to know the denominator of the outcome statistics we use in order to make the most informed decision. We often do not know who is included and who is excluded from the outcome data we give parents. If the most severe cases of a particular pathology are detected and pregnancies in these cases are frequently terminated, survival and outcome statistics will improve. These numbers depend not only on clinical skills and competency, but also on the general culture, values, beliefs, and consequent choices of the environment in which these pregnancies are followed and these babies born. Parents should be made aware of these factors when receiving antenatal counseling. They need to know if the statistics they are being told are for babies like their baby, and if not, how their baby is different. A second reason for increased transparency in perinatal and early neonatal deaths is for adequate quality control. In neonatology, it is imperative to compare our neonatal outcomes from one year to the next, as well as to compare ourselves with those with better outcomes in order to learn from each other and optimize neonatal care. If the culture of a particular center is to offer frequent terminations for severe pathologies, then the data generated by the center

in question might not in fact be comparable with data from a different epoch or from a center where termination is not possible. A center's culture surrounding palliative care can also directly affect these outcomes in a similar fashion.

5.3 Conclusion

In this chapter, we have classified antenatal deaths that occur after viability and explained why these deaths matter when examining neonatal statistics. In our opinion, neonatal statistics should have as a denominator all fetuses that are alive at the threshold of viability. If neonatal outcomes were presented using this denominator, one could more easily tell facts from values when reviewing the neonatal literature. In the analysis of outcome statistics, this classification of antenatal deaths should be coupled with a classification of deaths in the NICU, as described by Verhagen and Janvier in the second chapter of this book. This transparency would enable clinicians to better distinguish values from facts when examining neonatal outcomes and speaking with prospective parents. For example, in the case of baby Christine, we would know what a survival of 70 % for HLHS means by accounting for the severity of the cardiopathy for the neonates who were not admitted to the NICU.

There is increasing literature being published regarding how babies die in the NICU. But very little is published regarding how babies and fetuses die when they are not admitted to the NICU. Yet this information, while sensitive and sometimes controversial, is important not only for clinicians, but also for parents. Due to societal influences and changes in values, different centers have developed different approaches regarding end of life care and termination of pregnancy. These approaches vary tremendously and therefore have a real impact on how we interpret neonatal outcomes. When we look at data from our institution, it is important to know what our institutional culture is in order to better interpret our own data and better counsel parents. When parents are making difficult decisions regarding the life or death of their child, they deserve this transparency in order to make the most informed decision. Knowing how babies die when they are not admitted to the NICU is part of this information, and research regarding this should therefore be encouraged, supported, and shared.

References

1. Canadian Institute for Health Information, Induced Abortions Reported in Canada in 2012, https://www.cihi.ca/sites/default/files/document/ta_11_alldatatables20140221_en.pdf.
2. Connolly M-L. Northern Ireland abortion law: public consultation on proposed changes. BBC News; 2014.
3. Abortion Restrictions in States. The New York Times; 2013.

4. Macdorman MF, Kirmeyer SE, Wilson EC. Fetal and perinatal mortality, United States, 2006. Natl Vital Stat Rep. 2012;60(8):1–22.
5. Adzick NS, Thom EA, Spong CY, Brock JW 3rd, Burrows PK, Johnson MP, et al. A randomized trial of prenatal versus postnatal repair of myelomeningocele. New Engl J Med. 2011;364(11):993–1004.
6. Lorenz JM, Paneth N, Jetton JR, den Ouden L, Tyson JE. Comparison of management strategies for extreme prematurity in New Jersey and the Netherlands: outcomes and resource expenditure. Pediatrics. 2001;108(6):1269–74.
7. Altman LK. A Kennedy baby's life and death. The New York Times; 2013.
8. Hunter LE, Simpson JM. Prenatal screening for structural congenital heart disease. Nat Rev Cardiol. 2014;11(6):323–34.

Chapter 6
Termination of Pregnancy and Perinatal Palliative Care in the Case of Fetal Anomaly: Why Is There so Much Incoherence?

Antoine Payot

Abstract The question of the fetus as a patient is sensitive and we cannot ignore it. To deny the fetus any kind of status is troubling, especially when all prenatal screening and diagnostic tests are directed toward evaluating the fetus's health and development. On the other hand, one can understand the risks of allowing the fetus to have its own juridical status, leading to the possible slippery slope where a pregnant woman could be neglected as a person for the benefit of her fetus. Can we find middle ground between these extremes to allow the fetus some kind of a social importance, without neglecting women's right to be in charge of their own bodies and responsible for their fetuses? While medicine has been giving a face to fetuses through technology, shouldn't we recognize its responsibility towards defining the status of the fetus?

6.1 The Fetus as a Patient

Physicians typically consider the pregnant woman as one patient, while establishing dual goals of a good outcome for the woman and her fetus. However, advances in perinatal medicine have led to the concept of two separate patients by many clinicians and authors. It is now possible to intervene on the fetus and these fetal interventions are often referred to as fetal surgery. But when one intervenes on the fetus as a patient, they need to also operate on the pregnant woman [1].

The fetus does not need to be seen as a second patient to create a moral duty for the obstetrician. When a pregnant woman presents herself to an obstetrician, she

A. Payot (✉)
Pediatrics and Clinical Ethics, University of Montreal, Sainte-Justine Hospital, 3175 Chemin de la Côte-Sainte-Catherine, Montréal QC H3T 1C4, Canada
e-mail: antoine.payot@umontreal.ca

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_6

implicitly establishes a link between the fetus and the child-to-be by wanting to continue the pregnancy. The pregnant woman confers the status of patient to her fetus [2]. Although this argument allows this status to be withheld in the case of abortion, it also requires persons on both sides of the abortion debate to acknowledge that, once a woman confers patient status on her fetus, the obstetrician and the mother-to-be have a moral duty to provide care for the fetus as a future child. This duty extends to the pediatrician, who can be involved in helping the future parent make good decisions for the not-yet-born child. In some circumstances, one of these decisions might be to provide palliative care when a neonate will be born with a serious condition [3].

Even if the question of the fetus as a patient is sensitive, we cannot ignore it. To deny the fetus any kind of status is troubling, especially when all prenatal screening and diagnostic tests are directed toward evaluating the fetus's health and development. On the other hand, one can understand the risks of allowing the fetus to have its own juridical status, leading to the possible slippery slope where a pregnant woman could be neglected as a person for the benefit of her fetus.

There must be middle ground between these extremes to allow the fetus some kind of a social importance, without neglecting women's right to be in charge of their own bodies and responsible for their fetuses. While medicine has been giving a face to fetuses through technology, it must now recognize its responsibility towards defining the status of the fetus.

So wherever the ethical delimitation of the fetus stands, it will inevitably and continually raise important ethical questions.

6.2 Abortion

In many societies, the right to terminate a pregnancy represents a victory for women's freedom and autonomy. In many countries, it is legal for women to be able to choose to pursue or not a pregnancy or to have a child. However, can this reproductive choice really compare to the choice of interrupting a pregnancy because of a fetal anomaly? In fact, the medicalization of pregnancy and materialization of the fetus through imaging and other diagnostic techniques imply that medicine has a responsibility in such a decision-making process. Several states and countries have addressed this difference by delimiting a period of time during the pregnancy where termination of pregnancy is acceptable, for example, when the fetus reaches viability.

Thus, for several societies, abortion can essentially be categorized as follows: voluntary abortion (reproductive) during the first trimester of pregnancy, abortion for fetal anomaly during the second trimester, or demarcated by the limit of viability being between 22 and 24 weeks of pregnancy. Late termination of pregnancy remains legal in many countries, at any gestation until birth and in exceptional conditions, such as when the fetus has substantial risk of severe disability and/or when the pregnancy represents a threat to the woman's health.

6.3 The Challenge of Coherence Between Pre and Postnatal Periods

The discovery of a fetal anomaly generally occurs during the second trimester of a pregnancy. A fetal ultrasound, which is routinely done during pregnancy, makes it possible to visualize fetal morphology at a relatively early stage, typically between 17 and 20 weeks. A radiologist specialized in this field or an obstetrician usually carries out this highly specialized ultrasound.

From an obstetrical point of view, the interests of the mother are primary. If she wishes to stop her pregnancy following the discovery of an anomaly and the information she has received, the obstetrician will probably agree to the request.

In Canada, in contrast with many other countries, there is no law regulating late terminations of pregnancy. Although the viability of the fetus to extra-uterine life seems to be a criterion adopted by many clinicians, any delimitation is at the discretion of the physician who will carry out the procedure. The fetus does not have any rights of its own and is in fact considered as an integral part of its mother. Legal rights, as any other person, occur at birth. As termination of pregnancy is outlined in the standards of care for women, it is often perceived as a right. And since the healthcare system covers abortions, many obstetricians often feel they are obligated to offer it and/or comply with abortion requests at later stages of pregnancy.

This dichotomy between the absence of rights of a fetus inside its mother's womb and full citizen's rights after the birth has led to a particular practice of termination of pregnancy after viability. Indeed, in many countries, to avoid the birth of a living child with full legal rights, it is not uncommon to carry out late abortions, or a feticide (lethal injection in the uterine cavity, the umbilical cord, or the heart of the fetus) that ensures the death of the fetus.

This practice raises particularly complex ethical questions. On the one hand, aside from considerations of rights, are there ethical differences between a later fetal euthanasia, after viability and a postnatal euthanasia? Does the "geographical" position of the fetus allow this ethical distinction? Beyond this thorny question, one foresees that without some boundaries, criteria that make it possible to decide on a late termination of pregnancy cannot be the same ones used to decide whether to systematically offer palliative care after the birth of the child. In fact, it would be medically unethical to offer palliative care to a child who only presents with a mild anomaly.

6.4 In Practice

Nearly 5 % of pregnancies are diagnosed with a form of fetal anomaly, and 15 % of these anomalies are potentially life-threatening. The discovery of an anomaly during second trimester ultrasound will often lead to other testing techniques to improve diagnostic accuracy, and sometimes elaborate a prognosis.

After a diagnosis of fetal anomaly, there may be limited time to make decisions before the fetus is considered viable. However, parents can be offered many possible courses of action. They can decide to induce delivery and give birth to the child immediately. The child will not be viable and will likely die quickly after birth. They can alternatively decide to go further with diagnostic procedures (amniocentesis, cardiac echography, magnetic resonance imaging, etc.). This choice will allow for specifying the diagnosis, but may also bring the pregnancy beyond the viability of the fetus, which may mean that abortion is no longer an option in some institutions and consequently, the birth of a living child may become inevitable. Parents could finally decide to continue the pregnancy.

6.5 Different Practice—Different Views

One of the major difficulties in this perinatal decision-making process lies in counseling. Parents experiencing the news of having a fetus with an anomaly are often in a state of shock after the diagnosis. In some cases, this psychological state of mind can cause them to detach from the pregnancy [4]. How can physicians and healthcare teams best inform and counsel them?

From a recent study we carried out in Quebec, we discovered very different opinions of the acceptability of late termination of pregnancy for fetal anomaly between physicians involved in prenatal diagnosis and pediatric specialists who are somehow involved in the care of such infants later in life [5].

Physicians involved in prenatal diagnosis were significantly and systematically more willing to accept later termination of pregnancy for specific diseases compared to pediatricians who were involved in postnatal care, such as for cystic fibrosis (43.5 % vs. 17.8 %), cardiopathy with a curable prognosis such as tetralogy of Fallot (78.3 % vs. 23.6 %), cardiopathy with a palliative prognosis such as hypoplastic left heart syndrome (95.7 % vs. 63 %), Down syndrome without cardiopathy (77.3 % vs. 37.5 %), and Down syndrome with cardiopathy (100 % vs. 74 %). When physicians were asked how much they believed they influenced decisions of parents, 76 % believed they influenced the parental decision to terminate the pregnancy. This finding is especially important in regard to the decision-making process. One can wonder how informed consent and medical autonomy of parents, (often in a vulnerable position) are possible in such circumstances. These findings are consistent with a recent review showing that maternal-fetal medicine specialists from the United States were more disposed to talk about pregnancy termination than were fetal care specialists [6].

One can presume that such counseling is given because of the poor quality of life and suffering anticipated for the coming child and its family. However, it has been demonstrated extensively that health care professionals underestimate the quality of life of their patients and, furthermore, are poor at predicting future quality of life [7–9]. It could also be argued that a decision to terminate a pregnancy could protect parents from grief or the difficulty of living with a child who would require palliative care.

6.6 Parental Outcomes

Spontaneous and induced pregnancy losses are common. Evidence shows that potential parents undergo a grief reaction and require support and counseling in the long term. Some studies of women after termination of pregnancy for fetal anomaly show that they often experience pathological posttraumatic stress disorders symptoms [7, 10]. Pathological posttraumatic stress symptoms are high the year after termination of a pregnancy (almost 46 % of women). These symptoms decrease afterwards but remain present after 16 months in 20.5 % of women. Most interestingly, for those women, these symptoms don't seem to resolve and will stay at the same intensity level even seven years after a termination. Pathological grief and psychological malfunctioning follow the same pattern, although they are less frequently reported and almost completely resolved after 16 months.

The impact of a feticide on parents is less known. In a qualitative study done in the UK, parents' decisions about accepting or declining feticide seemed to reflect differences in perceptions of suffering in the birth process and dying after birth. In that study, many parents believed that the child's suffering after birth would be worse than feticide [8]. However, for most parents, feticide came together with the pregnancy termination process. Some parents were so focused on the feticide decision and procedure that they didn't seem to realize that there would be a delivery after the technique.

Finally, one can wonder if the severity of the diagnosis has an impact on parents' experience of termination of pregnancy. Literature shows that when pathologies leading to termination of pregnancy are categorized, the survival potential of the baby after birth is associated with a higher level of psychological morbidity on the part of the parents. These studies also show that pathological grief or posttraumatic stress disorder symptoms could be predicted by educational level of the mothers, gestational age at the time of the termination of pregnancy (greater age meaning more symptoms), and lethality of the fetal disease.

The management of such situations has an enduring effect on the psychological and emotional wellbeing of parents and the wider family. Family-centered care has become a crucial part of care of neonates. Should it also be the case for prenatal care?

6.7 Could Perinatal Palliative Care Be an Alternative to a Late Interruption of Pregnancy for Fetal Anomaly?

Perinatal palliative care is the holistic provision of supportive care and end of life care [9]. For a long time, pediatric palliative care has often been equated to end of life care during the dying process. But pediatric palliative care encompasses more than the dying process: a child can benefit from palliative care when he suffers from

any kind of incurable disease or has a limited life expectancy [11]. Nevertheless, any child having a potentially life-limiting condition, even if it is potentially curable, could benefit from palliative care during periods of uncertainty or when the curative treatments prove to be ineffective. Thus, the concept of uncertainty becomes the heart of the palliative paradigm and does not exclude a concomitant palliative approach with curative care.

What primarily differentiates the concept of perinatal palliative care from pregnancy termination is its intentionality. Whereas termination of pregnancy intends death for the fetus, perinatal palliative care is focused on the future of the child and its family: what one can do to make the life of this child and its family the most harmonious and joyful, looking at quality rather than quantity of life.

Palliative care should be considered and discussed with parents in the prenatal or early neonatal period if a baby suffers from a life-limiting illness [12]. Many situations in which it may be appropriate to withhold or withdraw life-sustaining interventions in babies have been described.

A recent report of a working group of the British Association of Perinatal Medicine concerning palliative care proposes that antenatal planning of palliative care starts with the identification and precise definition of the pathology, followed by an agreement within the multidisciplinary team about the diagnosis and prognosis, which should be shared and discussed with the family. The care is centered on the family and includes psychological, spiritual, and social support. All the studies indicate that communication among hospital personnel is of primary importance. The antenatal maternal evaluation should be disclosed in a precise and significant way. Plans of care should be openly discussed, including all options. Decisions should be written and communicated to all the caregivers implicated in the pregnancy, and should include the planned management of the childbirth. In many cases, this plan will imply awaiting the spontaneous beginning of labor and avoiding any useless intervention [9, 13].

Nevertheless, a cesarean could sometimes be justified to respond to maternal indications or to increase the possibility for a baby of being born alive, even if its life is likely to be short. The meaning that the parents give to the life of this baby in their family belongs to them, even if caregivers sometimes do not agree with their values.

6.8 Why Is Postnatal Palliative Care so Far from Termination of Pregnancy?

Many factors can explain why palliative care cannot simply be understood as another option for families facing a diagnosis of fetal anomaly. These are mostly related to the difference between abortion for reproductive autonomy and termination of pregnancy for fetal anomaly. In the first case, the pregnancy is not an acceptable option. In the latter, the child is generally desired, but not a child with a

particular anomaly. Nevertheless, due to the common acceptance of reproductive autonomy, many see later terminations of pregnancy as a right, and obstetricians may feel obligated to respond to the demand for abortion or even to systematically offer it. The absence of legal status of the fetus, even if designed to protect the rights of women, gives even more strength to this difference and sometimes seems to overshadow the ethical differences between these two kinds of intentions to terminate a pregnancy.

Furthermore, for palliative care to be an alternative to termination of pregnancy, it should be restricted to the same category of patients. As proposed by Leuthner and colleagues [3], these would be fetuses with diagnostic and prognostic certainty. However, termination of pregnancy could also be offered for those whose prognosis or even diagnosis is predominantly uncertain. On the other hand, while late terminations of pregnancy may be provided for some conditions, such as Down syndrome, palliative care at similar gestations would not be morally acceptable.

These issues raise many ethical questions about the discrepancy between termination of pregnancy and neonatal palliative care. Why is there such a difference of perspectives among physicians? Is it due to a fear of uncertainty, fear of the gray zone and of the uncertain outcome of the child and its family? Could it also be related to feelings of inadequacy: responsibility or guilt for leading future parents to make a decision that may reduce their quality of life? The multiple and highly publicized "wrongful life" cases have certainly contributed to such anxieties. However, the abundant literature about quality of life and resilience of families and individuals living with a physical or mental disability should make us think differently.

One could then ask if the fear of having a baby born alive and seeing it die could be alleviated by receiving good palliative care. Unfortunately, we don't know much about the experience of parents who have experienced palliative care compared with those who have had late pregnancy termination. Future research in this domain would be of great interest to see differences in outcomes of family quality of life and of posttraumatic stress symptoms.

Perinatal palliative care offers a new way of thinking about prenatal diagnosis and late termination of pregnancy. It involves a humanistic approach towards parents who are in a state of shock, whose families are shaken by the discovery of an unexpected anomaly in their expected child. As caregivers, we should ask ourselves the importance we ought to give to this unique family project.

References

1. Harrison MR, Adzick NS. The fetus as a patient. Surgical considerations. Ann Surg. 1991;213 (4):279–91; discussion 277–8.
2. Chervenak FA, McCullough LB. The fetus as a patient: an essential concept for the ethics of perinatal medicine. Am J Perinatol. 2003;20(8):399–404.
3. Leuthner SR. Fetal palliative care. Clin Perinatol. 2004;31(3):649–65.
4. Ahman A, Runestam K, Sarkadi A. Did I really want to know this? Pregnant women's reaction to detection of a soft marker during ultrasound screening. Patient Educ Couns. 2010;81(1):87–93.

5. Payot A, Dupont-Thibodeau A, Diadori P, Gorincour G. Grey zone or Black zone: disagreements between pediatricians and prenatal diagnosis physicians about critical prenatal decisions. Pediatr. Acad. Soc. 2012.
6. Brown SD, et al. Prenatally diagnosed fetal conditions in the age of fetal care: does who counsels matter? Am J Obstet Gynecol. 2012;206(5):409.e1–409.e11.
7. Korenromp MJ, et al. Long-term psychological consequences of pregnancy termination for fetal abnormality: a cross-sectional study. Prenat Diagn. 2005;25(3):253–60.
8. Graham RH, et al. The role of feticide in the context of late termination of pregnancy: a qualitative study of health professionals' and parents' views. Prenat Diagn. 2009;29(9):875–81.
9. Kilby MD, Pretlove SJ, Bedford Russell AR. Multidisciplinary palliative care in unborn and newborn babies. BMJ (Clinical research ed), 2011;342(Apr11 1):d1808–d1808.
10. Korenromp MJ et al. Adjustment to termination of pregnancy for fetal anomaly: a longitudinal study in women at 4, 8, and 16 months. Am J Obstet Gynecol. 2009;201(2):160.e1–7.
11. Santé et Services Sociaux, Québec. Normes en matière de soins palliatifs pédiatriques. 2006; pp. 1–88. http://publications.msss.gouv.qc.ca/acrobat/f/documentation/2006/06-902-05.pdf
12. Bhatia J. Palliative care in the fetus and newborn. J Perinatol. 2006;26:S24–6.
13. British Association of Perinatal Medicine. Working group report, Palliative care (supportive and end of life care). A framework for clinical practice in perinatal medicine. 2010.

Chapter 7
Predicting Outcomes in the Very Preterm Infant

Keith Barrington

Abstract Extremely preterm infants have a high mortality and increased long term morbidity compared to babies born at term or later in gestation. They may have delays in motor, cognitive or developmental domains or persistent impairments. Frequently, in the NICU, investigations or evaluations are performed with the goal of trying to predict the long term outcomes for many different purposes. The characteristics of a test required for such predictions differ according to the different purposes. Individual prediction of profoundly abnormal outcome, based on any currently available test, is severely limited, and the use of any test in order to limit or redirect intensive care is difficult to justify, particularly because survivors of neonatal intensive care have almost universally good quality of life.

Extremely preterm infants have a high mortality and increased long term morbidity compared to babies born at term or later in gestation. They may have delays in motor, cognitive or developmental domains or persistent impairments. Frequently, in the NICU, investigations or evaluations are performed with the goal of trying to predict whether an individual infant is likely to be impaired.

According to the widely followed WHO definitions 'An impairment is any loss or abnormality of psychological, physiological or anatomical structure or function; whereas a disability is any restriction or lack (resulting from an impairment) of ability to perform an activity in the manner or within the range considered normal for a human being; and a handicap is a disadvantage for a given individual, resulting from an impairment or a disability, that prevents the fulfilment of a role that is considered normal (depending on age, sex and social and cultural factors) for that individual'.

Although many publications speak blithely of handicap, we should preferably refer to attempts to predict impairment, as whether or not an impairment leads to significant disability starts to imply societal values, such as, what are our expectations? Two children with the same impairment, may have different disabilities.

K. Barrington (✉)
Department of Pediatrics, University of Montreal, 2900 Boulevard Edouard-Montpetit, Montreal, QC H3T 1J4, Canada
e-mail: keith.barrington@umontreal.ca

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_7

Handicap, using this definition, can often be minimized depending on how we define the roles of individuals with impairments, and on whether society seeks to integrate or to marginalize those with disabilities.

7.1 What Do We Want to Predict?

If we then suppose that predicting impairment may be possible, we need to ask which impairments are important to predict, I suggest that minor impairments with little effect on function, and which rarely lead to disability, are unlikely to be predictable, and may not be worthwhile to try and predict. We should focus on impairments which lead to important effects on clinical functioning, or which affect quality of life, which is where I focus in this chapter.

The majority of studies examining neonatal outcomes, and associating them with evaluations in the neonatal period have examined developmental assessments at 2 years of age or earlier. This has been done for a number of reasons: follow up programs become more expensive, and follow-up rates fall, the later the follow up is performed. So in order to achieve reasonable proportions of babies followed up, which is essential in order to have accurate descriptions of the outcomes of the whole group, a compromise has to be reached; a compromise between how predictive the tests really are for long term function on the one hand, and loss to follow up on the other. As a result developmental screening tests at between 18 and 24 months of age have been largely used, most commonly the Bayley Scales of Infant Development (BSID), as well as the Griffiths and other tests. Although valuable as a screening test for developmental delay, there are good data that demonstrate how poorly 2 year BSID scores predict longer term intellectual outcomes, and even more poorly functional outcomes. Maureen Hack, for example, showed that only 1/3 of preterm infants who had a low BSID score at 20 months had an IQ that was below normal at 8 years of age [1]. The 5 year CAP study outcomes (with evaluation by IQ testing) have also been compared with the 2 year BSID scores [2]. There was overall a substantial improvement in mean scores between the 2 time periods, and a reduction in the proportion of infants with scores more than 2 SD below the mean. In addition those infants with the lowest scores on the BSID tended to have the greatest improvements. We have shown that the biggest influence on whether a child would have an improvement in scores was in fact their socio-economic status. Some children with BSID scores more than 3 SD below the mean at 2 years were within the normal range at 5 years of age. Such data make the reliance on 2 year BSID and other developmental delay screening tools as indicators of neurodevelopmental impairment highly suspect.

The outcomes that we should be trying to predict, and the kind of reliability that we require in our predictions, should really depend on the purpose for which the predictions are being made. For some purposes, a statistically significant correlation between a particular finding and a poor Bayley score at 2 years might be important.

For others a very high positive predictive value for profound disability may be required.

To re-emphasize, a low BSID score is not a disability. It is not an impairment, it is just a screening test trigger that should lead to further evaluation and surveillance. As Colombo and Carlson put it [3], 'The BSID is, to be charitable, only modestly related to school-age cognitive development (i.e., the outcome that is most meaningful to investigators in this field). The BSID is a global measure of developmental status in infancy that assesses and aggregates the timely attainment of relatively crude milestones in infancy and early childhood.'

So the ability of a neonatal finding to predict a low BSID score is of limited interest, and is of extremely questionable value when we are discussing major changes in medical care based on predictions from prior literature. Long term outcomes which have impacts on function and quality of life are the outcomes most important to families in terms of their impact on the family, and their significance for their daily lives.

I would say that if the purpose of attempting to predict outcomes is in order to select patients for follow up, then a high sensitivity is required, to ensure that few patients with delays or impairments are missed, and prediction of a low 2-year Bayley score (which itself has a fairly high sensitivity (and low specificity) for long term functional problems) would be reasonable. Enrolling identified children in a follow up program is not generally a harm, so enrolling more children than the proportion that will truly eventually have significant impairments would be acceptable.

If the reasoning is to initiate a targeted early intervention, then again high sensitivity is required, so that all infants who will potentially benefit will be enrolled; if the intervention carries potential risks, or is costly, then a high specificity is also required.

If we wish, by our predictions, to prepare parents for their future, we need a high positive predictive value (PPV) for outcomes that are going to impact on their lives, and a high negative predictive value (NPV) to ensure that we do not inappropriately reassure them. In this case a high PPV for a low BSID score is of questionable benefit, as there is little evidence that a BSID <70 affects the function of families.

If we are trying to understand the causes of disability among preterm infants, then statistically significant associations, even if PPV is low, may help to direct our attention to findings which require further study.

In our efforts to perform research to reduce disability, or the impacts of disability, we really need predictive methods that have a high PPV for these outcomes (for example to enroll infants in prevention trials without enrolling infants at low risk) and a high sensitivity, so that a high proportion of affected children will be enrolled. In this case, minor or moderately severe impairments might be worthwhile predicting.

In contrast some articles explicitly state that the purpose of performing a particular test is that, with the results, we can redirect intensive care to comfort care,

and prevent the survival of disabled children. I propose that for such a purpose, if it is considered morally acceptable, we should demand findings that have extremely high PPVs, and only for profoundly abnormal outcomes. In such a situation a prediction that a 2 year BSID will be 2 SD below the mean is entirely inadequate.

7.2 Predicting Outcomes Before Birth, Lack of Prediction by Gestational Age

When deciding on immediate neonatal intervention for an infant about to deliver in the extremely preterm range, we have limited data to use. Decisions are usually based on estimated gestational age (EGA) and often follow recommendations from professional societies. It is clear from multiple data sources that gestational age among the extremely preterm infant is strongly correlated with survival, even though birth weight is a better predictor of survival. In contrast, a recent systematic review [4] shows that when infants are examined at a sufficiently advanced age (over 4 years at least) there is no distinction in intellectual impairment between infants born at 22, 23, 24, 25 or 26 weeks. Although they differ in their survival, among survivors there is little evidence of different frequencies of significant impairments [5].

Therefore gestational age cannot be used to predict long term outcomes, however sex can be, as there are consistent and substantial differences between boys and girls [6]. This would suggest that if predictions of long term intellectual impairments are going to be made to decide on active intervention, it makes no sense to alter intervention based on gestational age, it would be more rational to resuscitate girls, and not boys. The moral acceptability of this is worthy of discussion.

7.3 Predicting Outcomes in the Delivery Room, Lack of Prediction by Condition at Birth

A frequent decision in previous years was to suggest that immediately after birth we could evaluate the infant, and institute intensive care if the condition was favorable. It has become clear that this is inappropriate, infants evaluated as being in poor condition, or even requiring cardiac massage often survive, and, generally speaking do not have worse long term outcomes than infants in ‘good’ condition [7]. Even infants who at 5 min of age are still bradycardic or asystolic (Apgar less than 2) have a significant percentage of survival (about 30 %) although they do appear to have 2 year BSID scores which are substantially lower than extremely low birth weight infants who have better Apgar scores [8].

These data make it clear that decisions to intervene actively to support transition of an extremely preterm infant should not generally be based on the infant's condition in the delivery room. It would support the contention of Dr W Meadow that a 'trial of therapy' is most commonly the appropriate approach to take [9].

7.4 Predicting Outcomes by Early Ultrasound Findings

After admission to the NICU, very preterm babies will often have early, and then repeated, head ultrasounds. These are performed in order to follow professional guidelines, and because of known significant associations of some findings with impairments. But are such findings adequate indicators for long term serious impairment? A recent systematic review [10] examined the positive predictive value for neuromotor impairment (cerebral palsy) of various findings on head ultrasound. Of those findings which are often visible in the first ultrasound the PPV of a grade 3 hemorrhage for cerebral palsy was 26 % and of a grade 4 hemorrhage was 53 % (ranging from 29 to 76 % between studies). No such proportions for developmental delay or intellectual impairment were calculated. I suggest that a PPV of around 50 %, for an outcome which is often only moderately severe in the former extremely preterm infant, is inadequate for making life or death decisions.

The recent large prospective multicenter cohort known as the ELGAN study provided further information, both about the poor inter-observer reliability of head ultrasound interpretation [11], and the predictive value of various findings. They note, for example, that peri-ventricular hemorrhagic infarction was associated with an increase in the frequency of a BSID MDI (mental developmental index) of less than 70 from the background rate of 28 up to 44 % [12], that is, a likelihood ratio of 1.6. Lesions which usually are apparent later, such as the presence of ventriculomegaly or the appearance of echolucent lesions, each had a PPV for developmental delay, as detected by the BSID, of only 45 %.

Despite extensive literature review I have been unable to find any abnormality, routinely visible on an early head ultrasound before 1 week of age, which has a very high PPV (over 90 %) for profoundly abnormal outcomes (profound reductions in intellectual ability, or severely disabling cerebral palsy).

The common practice of describing head ultrasound by the Papile classification is also problematic, a small localized intracerebral hemorrhage is given the same grade [4] as the hemorrhagic destruction of an entire hemisphere, or both hemispheres. Studies which have examined the impact of the extent of hemorrhage, which would logically seem likely to be associated with the impacts of the hemorrhage, are few. One study which compared the effects of unilateral or bilateral hemorrhages, showed no apparent effect of a unilateral grade 4 PVH, but a significant impact of bilateral PVH [13] on the association with low BSID at 2 years. For all grades of hemorrhage, the effects on outcome were modified by the presence

of postnatal infection, postnatal steroid use, or (additively) both. Infants with grade 1 hemorrhages who had both adverse factors had substantially worse outcomes than infants with bilateral grade 4 hemorrhages who had neither of them.

7.5 Predicting Outcome by Clinical Course

Complications during the hospital stay of the extremely preterm infant do, in contrast have substantial effects on developmental delay, and on cerebral palsy (CP). The data concerning these impacts are also limited by the same reliance on early developmental screening tools such as the BSID, and on the detection of cerebral palsy of all grades. One large database study from the USA demonstrated that when postnatal complications were included in the prediction model, there were no findings on early head ultrasound that contributed to prediction of the combined outcome of low BSID scores or all grades of CP [14]. The only findings on later ultrasound that were predictive were cystic periventricular leukomalacia (PVL) or hydrocephalus requiring a shunt.

An episode of necrotising enterocolitis or sepsis increases the risk of developmental delay and of all grades of CP by about 2. For an infant who has both, the increase in risk is about 4 fold. Receiving postnatal dexamethasone increases risk by at least double, and again can be multiplied by the other factors. Any neonatal surgery increases risks, as does a diagnosis of bronchopulmonary dysplasia (BPD).

7.6 Predicting Outcomes by Other Modalities of Investigation

Other modalities of investigation, such as early and repeated amplitude integrated EEG, routine EEG to search of rolandic sharp waves, early neurologic evaluation, and NIRS have all been investigated in recent years. None of them have high PPV for profoundly abnormal long term outcomes. Although aEEG and NIRS may be statistically related to poorer outcomes, and therefore may be useful as research tools for evaluation of the mechanisms of injury, or even clinical tools as a research target for improving outcomes, in the future.

7.7 Predicting Outcomes Near to Discharge

In contrast to the rationale for prediction of outcome early in the hospital course, prediction of outcomes near to discharge, or at term equivalent age (TEA) is not often being performed for decision making regarding on-going active intervention or not. The other reasons for attempting prediction are all still important, however.

Many studies have recently been performed which demonstrate that much more detail is visible on TEA MRI scans than on head ultrasound. Several studies [15–17] have demonstrated that a series of different findings are statistically correlated with worse outcomes (2 year BSID scores!) These are most commonly markers of white matter injury, although reduction in grey matter volume also appears to be significantly correlated with outcomes.

However when the data are presented as the PPV for a reduced BSID score at 2 years of age or for CP (of all grades) it becomes clear that in an individual case the PPV is very low, and very often below 50 %. If we add the unreliability of BSID scores for important long term outcomes, it becomes very clear that the TEA MRI is of little value for individual patients, and should be considered a research tool, for which its value is probably very high. I think it highly likely that in the future, the detailed differences in brain development and function between the former preterm infant and controls, revealed by MRI, will clarify why preterm babies have a high load of impairments, and allow research directed at reducing that burden.

Currently however, there is little evidence that TEA MRI is a better predictor of clinically important long term disability than a TEA ultrasound. Indeed 4 studies comparing the modalities have found the opposite, that, despite the increased fidelity of the images they are equivalent for the, perhaps dubious, purpose of predicting low BSID scores or all grades of CP.

7.8 Which Test Should Be Used as a Screening Test?

Despite the recommendations of the CPS, the American academy of Neurology and other learned societies, it is clear from this review that none of the additional tests, head ultrasound, TEA MRIs, aEEG, NIRS, or routine EEG to detect rolandic sharp waves, fulfill widely accepted criteria for a screening test. That is, to be introduced for screening the test should be highly sensitive and very specific, it should identify a treatable condition, the distribution of test values in the target population should be known and a suitable cut-off level defined and agreed, there should be an agreed policy on the further diagnostic investigation of individuals with a positive test result and on the choices available to those individuals, there should be an effective treatment or intervention for patients identified through early detection, with evidence of early treatment leading to better outcomes than late treatment, there should be evidence from high quality Randomised Controlled Trials that the screening programme is effective in reducing mortality or morbidity. Where screening is aimed solely at providing information to allow the person being screened to make an "informed choice" (e.g. Down's syndrome, cystic fibrosis carrier screening), there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened.

None of the tests mentioned qualify by these criteria.

7.9 Can We Predict Adverse Long Term Outcomes, Should We, and Which Ones?

It is clear from this review that our ability to predict changes in the proportions of groups of survivors that will have early developmental or motor disability is limited. Even our ability to predict survival, either before or after birth is poor. Our ability to predict profoundly adverse outcomes in individuals, with a great degree of confidence, is almost non-existent.

In this case is there any value for early and repeated head ultrasound, routinely performed around the world? I think moderately early ultrasound is reasonable as a means to screen for treatable disorders, specifically post-hemorrhage ventricular dilatation. To detect this condition, although the indications for treatment are not crystal clear, an ultrasound towards the end of the first week of life will detect those infants who are at risk, or who already have early changes. A head ultrasound which is normal at this time is very unlikely to change, at least in an infant who is clinically reasonably stable, and therefore does not need to be repeated. Imaging around the time of discharge adds little for the infant who had an initially normal head ultrasound, but, for those with initial abnormalities, or with very complicated courses, a head ultrasound near to discharge has more predictive ability for 2 year BSID scores or all grades of CP, but still with a low specificity and low PPV. Extensive bilateral cystic periventricular leukomalacia (grade 3 PVL) is one exception to this discussion, affected infants frequently have severe disabling CP [18], and detection of this condition may assist in early re-adaptation and intervention. Although commonly associated with serious complications, including perinatal and late-onset sepsis, occasional babies with grade 3 PVL are unexpected, a head ultrasound specifically directed to detecting this condition could be considered between 36 and 40 weeks post-menstrual age.

7.10 Predicting Quality of Life

Every year there are many publications describing outcomes of groups of extremely preterm infants. Although it is clear that the incidence of impairments is much higher among such infants than among control term-delivered babies, it must be emphasized that the large majority of extremely preterm infants function very well, with no or modest impacts of their life history on their lives or their families. Perhaps the most important question of all then should be: what is the impact of extreme prematurity on the quality of life of survivors, and how can we predict those infants with a poor quality of life?

Many ethicists have proposed that in order to consider re-direction of care to comfort care, rather than curative care, the major consideration should be: does this patient have a potential for a good quality of life, or not?

There is one test which we can indeed use to predict future quality of life in the extremely preterm infant at discharge from the hospital. The test is simple: 'is the baby alive?' If the answer is yes, the positive predictive value for an acceptable to excellent quality of life is over 95 %.

Every study of quality of life expectations of former extremely preterm infants is consistent. The quality of life, when rated either by themselves, or their families is excellent, and usually indistinguishable from controls [19]. Even when restricted to those infants with impairments, even severe impairments, the results are the same [20].

Although impairments are difficult or impossible to predict in the early neonatal period, and although the quality of life of impaired individuals is excellent, this is not to imply that we should ignore the high burden of impairment in the extremely preterm infant. Reducing that burden and finding ways to minimize the impacts of impairment are vitally important issues for families experiencing the birth of an extremely preterm infant. Improving functional outcomes, reducing brain injury, improving re-adaptation techniques and services are all vital to reduce the impacts of prematurity, and ease the lives of our patients.

Further studies simply describing the adverse outcomes of groups of preterm infants will undoubtedly be required, to determine if there are changes over time. On the other hand research to find ways to reduce those adverse outcomes and reduce their impacts should be the priority for the future.

7.11 Conclusion

Predicting the survival and the long term outcomes of preterm infants may be considered for many different purposes. The characteristics of a test required for such predictions differ according to the different purposes. Individual prediction of profoundly abnormal outcome, based on any currently available test, is severely limited, and the use of any test in order to limit or redirect intensive care is difficult to justify, particularly because survivors of neonatal intensive care have almost universally good quality of life.

References

1. Hack M, Taylor HG, Drotar D, Schluchter M, Cartar L, Wilson-Costello D, et al. Poor predictive validity of the Bayley scales of infant development for cognitive function of extremely low birth weight children at school age. Pediatrics. 2005;116(2):333–41.
2. Schmidt B, Anderson PJ, Doyle LW, Dewey D, Grunau RE, Asztalos EV, et al. Survival without disability to age 5 years after Neonatal Caffeine therapy for Apnea of prematurity. JAMA J Am Med Assoc. 2012;307(3):275–82.
3. Colombo J, Carlson SE. Is the measure the message: the bsid and nutritional interventions. Pediatrics. 2012.

4. Moore GP, Lemyre B, Barrowman N, Daboval T. Neurodevelopmental outcomes at 4 to 8 years of children born at 22 to 25 weeks' gestational age: a meta-analysis. JAMA Pediatr. 2013.
5. Andrews B, Lagatta J, Chu A, Plesha-Troyke S, Schreiber M, Lantos J, et al. The nonimpact of gestational age on neurodevelopmental outcome for ventilated survivors born at 23–28 weeks of gestation. Acta Paediatr. 2012;101(6):574–8.
6. Moore T, Hennessy EM, Myles J, Johnson SJ, Draper ES, Costeloe KL, et al. Neurological and developmental outcome in extremely preterm children born in England in 1995 and 2006: the EPICure studies. BMJ. 2012;345:e7961.
7. Finer NN, Tarin T, Vaucher YE, Barrington K, Bejar R. Intact survival in extremely low birth weight infants after delivery room resuscitation. Pediatrics. 1999;104(4):e40.
8. Wyckoff MH, Salhab WA, Heyne RJ, Kendrick DE, Stoll BJ, LaptookAR. Outcome of extremely low birth weight infants who received delivery room cardiopulmonary resuscitation. J Pediatr. 2012;160(2):239–44.e2.
9. Meadow W, Lagatta J, Andrews B, Lantos J. The mathematics of morality for neonatal resuscitation. Clin Perinatol. 2012;39(4):941–56.
10. Nongena P, Ederies A, Azzopardi DV, Edwards AD. Confidence in the prediction of neurodevelopmental outcome by cranial ultrasound and MRI in preterm infants. Arch Dis Child Fetal Neonatal Ed. 2010;95(6):F388–F90.
11. Kuban K, Adler I, Allred E, Batton D, Bezinque S, Betz B, et al. Observer variability assessing US scans of the preterm brain: the ELGAN study. Pediatr Radiol. 2007;37(12):1201–8.
12. O'Shea TM, Kuban KC, Allred EN, Paneth N, Pagano M, Dammann O, et al. Neonatal cranial ultrasound lesions and developmental delays at 2 years of age among extremely low gestational age children. Pediatrics. 2008;122(3):e662–9.
13. Merhar SL, Tabangin ME, Meinzen-Derr J, Schibler KR. Grade and laterality of intraventricular haemorrhage to predict 18–22 month neurodevelopmental outcomes in extremely low birthweight infants. Acta Paediatr. 2012;101(4):414–8.
14. Broitman E, Ambalavanan N, Higgins RD, Vohr BR, Das A, Bhaskar B, et al. Clinical data predict neurodevelopmental outcome better than head ultrasound in extremely low birth weight infants. J Pediatr. 2007;151(5):500–5.e2.
15. Woodward LJ, Anderson PJ, Austin NC, Howard K, Inder TE. Neonatal MRI to predict neurodevelopmental outcomes in preterm infants. N Eng J Med. 2006;355(7):685–94.
16. Miller SP, Ferriero DM, Leonard C, Piecuch R, Glidden DV, Partridge JC, et al. Early brain injury in premature newborns detected with magnetic resonance imaging is associated with adverse early neurodevelopmental outcome. J Pediatr. 2005;147(5):609–16.
17. Mirmiran M, Barnes PD, Keller K, Constantinou JC, Fleisher BE, Hintz SR, et al. Neonatal brain magnetic resonance imaging before discharge is better than serial cranial ultrasound in predicting cerebral palsy in very low birth weight preterm infants. Pediatrics. 2004;114 (4):992–8.
18. Pierrat V, Duquennoy C, van Haastert IC, Ernst M, Guilley N, de Vries LS. Ultrasound diagnosis and neurodevelopmental outcome of localised and extensive cystic periventricular leucomalacia. Arch Dis Child Fetal Neonatal Ed. 2001;84(3):F151–6.
19. Saigal S, Tyson J. Measurement of quality of life of survivors of neonatal intensive care: critique and implications. Semin Perinatol. 2008;32(1):59–66.
20. Payot A, Barrington KJ. The quality of life of young children and infants with chronic medical problems: review of the literature. Curr Probl Pediatr Adolesc Health Care. 2011;41(4):91–101.

Chapter 8
Predicting the Future of Preterm Infants: Should We Use Quality of Life and Social Determinants Criteria?

Antoine Payot

Abstract In neonatology, many choices to withhold or withdraw treatments are made based on long-term projections of the child's quality of life. These projections are essentially made considering medical facts of possible physical or intellectual disabilities. Physicians and parents generally talk about quality of life, whether it is to decide not to resuscitate a very young premature baby or to withdraw treatments from a neonate. These issues raise numerous ethical issues, two of those are discussed in this paper: clinicians are mostly wrong in their predictions of quality of life for their patients, and medical facts are only part of the evaluation and prediction of quality of life; socio-economic factors have a great influence on outcome of premature babies. Why are physicians so reluctant to use them in their assessments?

In neonatology, many choices to withhold or withdraw treatments are made based on long-term projections of the child's quality of life. These projections are essentially made considering medical facts of possible physical or intellectual disabilities. Physicians and parents generally talk about quality of life, whether it is to decide not to resuscitate a very young premature baby or to withdraw treatments from a neonate [1]. These issues raise numerous ethical issues. I will discuss two of them in this paper: clinicians are mostly wrong in their predictions of quality of life for their patients, and medical facts are only part of the evaluation and prediction of quality of life.

8.1 Predicting Quality of Life

Many tools have been developed to assess health related quality of life (HRQoL). The interest in these tools relates to their ability to measure the evolving perception of HRQoL throughout the growth of the child as it evolves in its family. For ex premature babies, the wide body of literature tells us that HRQoL can be relatively

A. Payot (✉)
Pediatrics and Clinical Ethics, University of Montreal, Sainte-Justine Hospital, 3175 Chemin de la Côte-Sainte-Catherine, Montreal, QC H3T 1C4, Canada
e-mail: antoine.payot@umontreal.ca

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_8

low in early life. This is especially true for parents submerged by the burden of care, numerous hospital appointments, and possibly low maternal mood [2]. However, their HRQoL improves gradually during infancy and even more during adolescence and young adulthood. Ultimately, there seems to remain very little difference in HRQoL between ex-VLBW and control groups at a later age [3]. It seems obvious that most physical disabilities have been dealt with, allowing more emphasis on the social and psychological aspects in the self-assessment of Quality of Life [4]. From most of these studies, it is clear is that having a biological impairment does not automatically translate into a poor self-assessed quality of life.

One of the main problems in assessing babies' quality of life is that parents and health professionals are the key reporters of the health status of children, not the children themselves. Studies show that proxy respondents tend to report higher morbidity and lower quality of life than the individuals whose perceived health status and quality of life is being judged. In perinatology, one of the first studies in this area was conducted by Saigal et al. [5], who compared perceptions of health practitioners and parents of very low birth weight children who were adolescents at that time. Clinicians systematically rated four vignettes about disabled adolescents lower than parents did, especially when the vignettes presented a severe mental or physical disability. One case was even perceived by clinicians as being worse than death. However, there was much more concordance in perceptions of the four vignettes between adolescents and their parents. In general, agreement between parent and child may vary depending on the domains that are being measured. For example, there is good agreement between parent and child about observable areas, such as physical activity and functioning, somatic distress, and chronic illness. Agreement is poorer for domains such as social and emotional functioning, pain, and cognitive functioning. Ironically, it is not possible to obtain the personal perspectives where these are most needed: from those who are too young, sick, disabled, or cognitively impaired to respond themselves. Under these circumstances, we have no other choice than to rely on proxy evaluations.

Another important limitation in the evaluation of the quality of life of babies in the neonatal intensive care unit lies in the fact that we are not really measuring it. In fact, we are predicting future quality of life, sometimes to make life and death decisions. Most of the time, we are not even completely certain that the child will present with a physical or intellectual disability, or of the degree of such impairment. Furthermore, such a prediction doesn't take into account many factors, such as the child's resilience and the parents capacity to adapt to potential challenges. Resilience is an individual's ability to cope with difficulties, in this case, to overcome physical disabilities and develop social and relational skills. For small children who will be dependent on their parents for a long time, one might also talk about family resilience, although this is rarely mentioned in the medical literature. Adaptation to disability related to perinatal problems has been well demonstrated. In particular, social and psychological factors are considered to be more important than physical impairment in the self-assessed quality of life of teenagers and young adults.

Resilience over time has been studied in the Kauai Longitudinal Study, which followed 700 children from birth until age 40 [6]. It clearly demonstrated that

individuals are not programmed to fail in life. One third of the children who had experienced high risk factors in infancy (poverty, perinatal complications, parental psychopathology, low education, etc.) were able to overcome these problems and become highly competent adults. Many factors explained how children worked to override anticipated problems. Most resilient children showed individual protective factors. At age one, their mothers tended to characterize them as active, affectionate, cuddly, good-natured, and easy to deal with; at age two, independent observers described the resilient toddlers as agreeable, cheerful, friendly, responsive, and sociable. Family factors also played a role in their resilience. Children who succeeded against the odds had the opportunity to establish, early on, a close bond with at least one competent, emotionally stable person who was sensitive to their needs. Much of this nurturing came from substitute caregivers, such as grandparents, older siblings, aunts, and uncles. Resilient children seemed to be especially adept at "recruiting" such surrogate parents. Finally, community was also a factor in allowing some of these children to overcome difficulties. Resilient youngsters tended to rely on elders and peers in their community for emotional support and sought them out for counsel in times of crisis. Those elders and peers could be, for instance, teachers, neighbors, youth leaders, and ministers.

Factors such as resilience have not been studied in children born premature to be able to predict their effect on outcome in life. However, it is interesting to see that in many recent studies, socio-economic status seems to be predictive of outcome in premature children. Could socio-economic status be related to a better potential for resilience in these families?

What about quality of life of families who have a very low birth weight child (VLBW)? Studies show that quality of life for these families is affected, but mostly during the first two years of the child's life [7, 8]. Authors studying families of VLBW children showed greater stress, but greater family cohesion and fewer conflicts than typical families. They also found no significant difference in parental divorce between these families and the control population. However, these families are subject to more stress as the child's emotional function increases. In studies, the most affected are often parents of children with low measured IQ, or severe disability, or parents of low socio-economic status.

8.2 Outcome and Socio-Economic Factors

Individuals with a low socio-economic status have long been described as being more affected by chronic diseases and having a higher mortality. This has been demonstrated in many adult studies. In perinatology, low-socio-economic status has been shown to be a risk factor for both chronic disease and high mortality [9, 10]. More recent epidemiological studies have shown that parents' socio-economic status was the main predictor of mild and severe cognitive deficiency in their children, with an increased risk of up to more than threefold [11].

The cause of this negative impact is debatable. Is it directly linked to the poor socio-economic status of the parents or is it a consequence of low parental education and insufficient stimulation during the first years of development? One could argue that these families have limited resources to cope with the birth and care of a very low birth weight child, and that it may interfere considerably with aspects of their daily life. The level of impact on a family over time is also largely influenced by social support, as demonstrated in a recent study [7]. Having a very low birth weight child greatly increases family stress. This stress tends to decrease over time until there is no significant difference from those families who had a healthy, term baby. However, many families with little social support don't experience such a reduction of stress, even after many years [7]. In other words, they seem to have less capacity to cope with adversity, or additional adversity.

These findings, which seem to be confirmed across time and for many different countries, are troubling. It seems intuitively easy, for example, to decide which babies should receive neonatal resuscitation based on medical facts: birth weight, gestational age, periventricular leukomalacia, etc. However, are we ready to make this determination on social grounds: because of the parents' low socio-economic background, low education, or marital status? To do so would probably be perceived as negative discrimination, even if epidemiologically it appears to be worse for a VLBW baby to be born of parents of low socio-economic status than to have a severe intraventricular hemorrhage [12].

As healthcare providers, we are focused on the outcomes of babies in our care. We want to base our medical decisions on biological and objective facts for two main reasons. The first comes from our education, from our having been trained to take care of biological dysfunctions. The other reason is that those are the factors we can identify and try to change. Social and psychological aspects can seem much more nebulous and difficult to grasp. We don't know how we could have a positive impact on such factors. The mixing of biological and socio-economic influences makes these factors even more difficult to deal with.

However, as research in this domain evolves, health care providers should no longer be able to claim ignorance. As social science continues taking its place in medicine, we will face difficult questions as we decide how to use this information. For example, will our care differ for extremely premature babies of a low socio-economic status because they show lower than average outcomes in the long term? Or hopefully will we get involved in reducing socio-economic disparities and improving education. Public health programs could be of benefit for these fragile patients. However, adverse outcomes could be caused not only by deprivation, but also by an adverse lower socio-economic status, as sociologists have described. To be able to respond to such questions and improve our knowledge as well as the potential impact of such programs, one will need to develop broader interdisciplinary research and collaboration between medicine and social science. In the future, we may be able to screen the potential resilience of families and adapt our follow-up services where they are most needed.

References

1. 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):1487–500 Epub 2007/01/02.
2. Saigal S, Tyson J. Measurement of quality of life of survivors of neonatal intensive care: critique and implications. Semin Perinatol. 2008;32(1):59–66 Epub 2008/02/06.
3. Saigal S, Stoskopf B, Streiner D, Boyle M, Pinelli J, Paneth N, et al. Transition of extremely low-birth-weight infants from adolescence to young adulthood: comparison with normal birth-weight controls. JAMA. 2006;295(6):667–75.
4. Payot A, Barrington KJ. The quality of life of young children and infants with chronic medical problems: review of the literature. Curr Probl Pediatr Adolesc Health Care. 2011;41(4):91–101 Epub 2011/03/29.
5. Saigal S, Feeny D, Furlong W, Rosenbaum P, Burrows E, Torrance G. Comparison of the health-related quality of life of extremely low birth weight children and a reference group of children at age eight years. J Pediatr. 1994;125(3):418–25 Epub 1994/09/01.
6. Werner E. Resilience and recovery: findings from the Kauai longitudinal study. Res Policy Pract Child Ment Health. 2005;19(1):11–4.
7. Singer LT, Fulton S, Kirchner HL, Eisengart S, Lewis B, Short E, et al. Longitudinal predictors of maternal stress and coping after very low-birth-weight birth. Arch Pediatr Adolesc Med. 2010;164(6):518–24.
8. Singer LT, Fulton S, Kirchner HL, Eisengart S, Lewis B, Short E, et al. Parenting very low birth weight children at school age: maternal stress and coping. J Pediatr. 2007;151(5):463–9.
9. Taylor HG, Klein N, Schatschneider C, Hack M. Predictors of early school age outcomes in very low birth weight children. J Dev Behav Pediatr. 1998;19(4):235–43 Epub 1998/08/26.
10. Hack M, Fanaroff AA. Outcomes of children of extremely low birthweight and gestational age in the 1990s. Semin Neonatol. 2000;5(2):89–106 Epub 2000/06/22.
11. Beaino G, Khoshnood B, Kaminski M, Marret S, Pierrat V, Vieux R, et al. Predictors of the risk of cognitive deficiency in very preterm infants: the EPIPAGE prospective cohort. Acta Paediatr. 2011;100(3):370–8 Epub 2011/01/19.
12. Voss W, Jungmann T, Wachtendorf M, Neubauer AP. Long-term cognitive outcomes of extremely low-birth-weight infants: the influence of the maternal educational background. Acta Paediatrica. 2012;101(6):569–573. doi:10.1111/j.1651-2227.2012.02601.x.

Chapter 9
End-of-Life Decisions in Neonatology from a Children's Rights Perspective: Dutch Developments Examined

Jozef Dorscheidt

Abstract End-of-life decisions regarding severely suffering newborn infants remain one of the most difficult issues in health care. In the last two decades the Netherlands have witnessed several developments regarding this issue. New research results and policy developments on legal and medical aspects of neonatal end-of-life decisions in the Netherlands have vitalized the Dutch debate once more and provide good cause for further reflection on specific issues. This chapter offers a reflection on several issues in current neonatal end-of-life practice in the Netherlands and discusses how these issues relate to the United Nations Convention on the Rights of the Child.

9.1 Introduction

End-of-life decisions regarding severely suffering newborn infants remain one of the most difficult issues in health care. In the last two decades the Netherlands have witnessed several developments regarding this issue. Developments such as the introduction of a statutory reporting procedure, the acquittal of two physicians charged with murder of a hopeless and unbearably suffering neonate, and the establishment of a national multidisciplinary expert committee serving as an advisory board to the Public Prosecutor, have shaped current Dutch understanding of the matter. New research results and policy developments on legal and medical aspects of neonatal end-of-life decisions in the Netherlands have vitalized the Dutch debate once more and provide good cause for further reflection on specific issues.

This chapter offers a reflection on several issues in current neonatal end-of-life practice in the Netherlands and discusses how these issues relate to the United Nations Convention on the Rights of the Child (CRC). First, the chapter explains how neonatal end-of-life decisions have developed as a children's rights issue.

J. Dorscheidt (✉)
Section of Health Law, University of Groningen/University Medical Center Groningen, Oude Kijk in 't Jatstraat 26, Groningen 9766PB Netherlands
e-mail: j.h.h.m.dorscheidt@rug.nl; j.h.h.m.dorscheidt@umcg.nl

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_9

Second, after a short overview of the applicable Dutch legal framework, it discusses some results of medical-empirical research on the practice end-of-life decisions in Dutch Neonatal Intensive Care Units (NICUs). Various questions raised by this research are subsequently examined against the background of the newborn infant's inherent right to life and the norm of a child's best interest. To other Contracting States to the CRC, this approach may offer a point of reference in defining their own position on the presented issues.

9.2 End-of-Life Decisions in Neonatolgy: A Children's Rights Issue

On January 31st 2004 the United Nations Committee on the Rights of the Child in Geneva (35th session) addressed end-of-life decisions in neonatology in its Concluding Observations on the Second Periodic Report of the Kingdom of the Netherlands. Remarkably, the Dutch government did not specifically provide any information on this topic to the CRC-Committee. Obviously, the CRC-Committee relied on its own informational sources in this regard, for instance by intensifying its contacts with the United Nations Committee on Civil and Political Rights. In its response to the Third Periodic Report of the Kingdom of the Netherlands under the International Covenant on Civil and Political Rights (ICCPR) [1], this Committee had already expressed concerns about the enforcement qualities of the Dutch Euthanasia Act, particularly since this Act addresses end-of-life requests by competent minors as well. In paragraph 6 of its Concluding Observations, the ICCPR-Committee also referred to the issue of end-of-life decisions in neonatology and asked the Dutch government to provide explanatory information on current practice in this area. By readdressing the issue the CRC-Committee has acknowledged the topic as a genuine children's rights matter.

With regard to Article 6 CRC, the CRC-Committee also expressed concerns regarding the legal position of minors in the Dutch legislation on euthanasia and physician-assisted suicide (PAS). Furthermore, the CRC-Committee was particularly concerned about information that Dutch physicians had ended the life of newborn infants with severe abnormalities. As a result, the Committee asked the Dutch government to provide detailed information with regard to these issues and stressed that regulations and procedures concerning end-of-life decisions in neonatology must be in conformity with the child's inherent right to life under Article 6 of the Convention.

However, in its Third Periodic Report of March 2007, the Dutch government neglected to provide the requested information, even though sufficient data on this issue were available. The Periodic Report merely recalled the establishment of the earlier mentioned national multidisciplinary expert committee and referred to the intention to induce doctors to be more open about the frequency and medical characteristics of neonatal end-of-life cases [2]. In consequence, the CRC-Committee reiterated its concerns in January 2009 (in its 50th Session) and urged the Netherlands

to investigate the application of criminal law to the termination of life of neonatal children. The Committee subsequently repeated—in Recommendation 31(a)—its previous recommendation to frequently evaluate, and if necessary revise, the regulations and procedures on euthanasia and PAS, in order to ensure the special protection of children, including newborn infants with severe abnormalities, under Article 6 of the Convention and additionally pointed to the need to prevent non-reporting.

In November 2013 the Dutch government delivered its Fourth Periodic Report to the CRC-Committee. In this Report [3] the government confirms, in response to Recommendation 31(a), that regulations and procedures concerning neonatal end-life decisions require careful evaluation and, if necessary, revision. Furthermore, the government states that an evaluation of the procedure for reporting and reviewing cases in which the life of a newborn infant has been terminated, was commissioned in 2010. No other information is presented to the CRC-Committee.

It is unclear what drives this governmental reluctance to provide relevant data on medical neonaticide. Yet, it is a fact that the Netherlands are unwilling to share available information on neonatal end-of-life practice and legal research on the conformity of this practice with Article 6 CRC with the CRC-Committee, and consequently the children's rights community as a whole. One might ask whether this suggests that country reports to the CRC-Committee are at risk of being political statements rather than reflections of a country's truthful intentions to respect the rights of children. Still, to complement the CRC-Committee's views the next paragraphs discuss opinions and considerations resulting from available Dutch research data on the matter. These data may show that some of the medical-empirical findings and related legal analyses give rise to criticize current Dutch neonatal end-of-life practice from a children's rights perspective.

9.3 Dutch Legal Framework

In order to fully understand the Dutch juridical perspective on neonatal end-of-life decisions and how it relates to Article 6 CRC, for instance, a short explanation of the applicable Dutch legal framework is presented below [4].

According to Dutch legal understanding the wording 'end-of-life decisions in neonatal practice' refers to several types of professional decisions in neonatal health care, leading to the death of a newborn patient. This wording is a collective noun that refers to decisions to withdraw or withhold medical treatment, to administer (or increase) sedative medication or—in extreme cases—to deliberately end a neonate's life. In general, the neonatologist in charge is responsible for these end-of-life decisions.

A non-treatment decision remains without legal consequences provided such a decision is based on admissible professional grounds, such as inability to realize the treatment's goal or the disproportionality between the ends and means of the treatment. If the grounds for a non-treatment decision are legally unsound or even

negligent, the physician in charge can be held accountable. In appropriate circumstances this scrutiny may lead to disciplinary or even criminal charges against the physician. To deliberately end the life of a hopeless and unbearably suffering newborn child (=medical neonaticide) counts as homicide or murder under Dutch criminal law.

While earlier Dutch burial law required physicians to report cases of medical neonaticide directly to the judicial authorities [5], as of March 2007 [6] these cases must be examined by a national multidisciplinary expert committee [7]. This committee—composed of lawyer (=chair), an ethicist and three neonatologists—reviews such cases against special requirements of due care and provides a recommendation to the Public Prosecutor, who decides whether or not the physician in charge will be prosecuted. In cases where the physician has met these due care requirements and a successful appeal to a ground for impunity known as 'defense of necessity' (as in Article 40 of the Dutch Penal Code) is expected, no prosecution will be initiated.

In 2009 the expert committee received its first reported case [8]. The committee's recommendation in this case declared the responsible physician's performance to be careful and the Board of Procurators-General concluded that this physician had acted in accordance with the due care requirements. In consequence, the Public Prosecutor did not begin criminal proceedings against the physician.

In 2005 the so-called "Groningen Protocol" [9] was accepted by the Dutch Paediatrics Association as a guideline for physicians confronted with situations in which medical neonaticide might become inevitable. The document contains a list of special requirements of careful decision making. It originates from the content of the statutory reporting procedure, from a governmental report published in 1997 and considerations articulated by the district and appellate courts in the criminal cases of Prins and Kadijk in 1995 and 1996.

In the summer of 2013 the Royal Dutch Medical Association (RDMA) issued a Position Statement on the matter. Its content will be discussed in paragraph 6.

9.4 Dutch Research

In recent years, the issue of neonatal end-of-life decision has been researched extensively in the Netherlands, from a legal as well as a medical-empirical perspective. An important part of this research was conducted by scholars at the University Medical Center in Groningen.

At Groningen, it is generally understood that in order to deal effectively with issues about end-of-life decisions in neonatology in general and its permissibility in particular, legal and medical scholars need to collaborate instead of opposing one another. This belief in the need for collaboration resulted in joint research.

In my contribution to this research, I studied the meaning of the child's inherent right to life (Article 6 CRC) and its right to health care (Article 24 CRC) in the

context of neonatal end-of-life decisions. Additionally, I focused on whether the legal prohibition of disability discrimination mentioned in Article 2 (1) CRC needs special consideration in the decision-making process. In medical practices in the USA, the UK, Germany and also the Netherlands disabled neonates who suffer from severe abnormalities are at risk of being discriminated against on the basis of their disability and could be wrongfully limited in the enjoyment of their fundamental rights when end-of-life decisions—particularly non-treatment decisions—arise.

I concluded that proper notice of the legal prohibition of disability discrimination during the decision making process can ensure the equal protection of the right to life and to health care of the severely suffering neonate when end-of-life decisions occur. In those situations, application of a special scheme of reference derived from the concept of "objective justification" is discrimination jurisprudence, can help to guard against such decisions being unlawfully based on an infant's disability (or disabilities) [10].

Subsequently, the departments of health law and of paediatrics at the University Medical Center Groningen studied current end-of-life practice in all 10 Dutch Neonatal Intensive Care Units (NICUs). Specific issues in this practice were defined and its characteristics were analyzed. The findings were then reviewed with respect to current Dutch legal understanding and considered in relation to norms of international human rights law.

In the next paragraph some of the significant findings and reflections on three issues are presented: (1) the interpretation of "hopeless and unbearable suffering", (2) the position of parents in deciding what's best for their newborn infant, and (3) the use of particular medication regimens.

9.5 End-of-Life Practice in Dutch NICU's

Between October 2005 and September 2006 Verhagen c.s. analyzed the medical files of 359 neonates who died in Dutch NICUs [11]. In 340 of these 359 cases a decision to withhold or withdraw treatment preceded death. The decision to withdraw life-sustaining treatment (mostly withdrawal from mechanical respiration) was made in 294 cases. Death resulting from withholding other treatment occurred in 46 cases, while deliberate ending of life officially took place in one case.

All cases in which end-of-life decisions occurred were classified into three groups. Group I involved infants (208/359 = 58 %) suffering from disorders that were expected to offer no real chance of survival. Group II included infants (150/359 = 42 %) with no terminal disorder, but a rather poor prognosis with regard to the future quality of life. Group III consisted of only one child, who was stable and not dependent on NICU-care, but known to have a poor prognosis and to be subjected to severe and manifest suffering.

The performed analyses were particularly focused on the 147 [12] group II-cases, as the end-of-life decisions in this group were believed to provide the most food for thought. Some 80 physicians in charge of infants within this group were interviewed by a medical scholar (EV), as these physicians were likely to provide further details on the background of an end-of-life decision in a particular case. During several of these interviews, a legal scholar (JD) was present as well.

9.5.1 Hopeless and Unbearable Suffering

The analysis of the data shows that in 135 (92 %) of the (147) cases in group II, a non-treatment decision—mainly termination of mechanical respiration—was based on the infant's expected quality of life. In 112 (76 %) of these cases, this decision concerned future suffering or expected inability to be engaged in verbal or non-verbal communication with others. In 71 (48 %) of these cases, considerations regarding the infant's present quality of life were used.

These findings raise the question of the type of "hopeless and unbearable suffering" that can justify an end-of-life decision: *present* pain and suffering or *future* pain and suffering as well? But what about the mere expectancy of a child's pain and suffering? [13].

In November 2005 the Dutch government stressed that the presence of "hopeless and unbearable suffering" is the only admissible ground for decisions to deliberately end neonatal life [14]. Even though understandable, on the basis of the Groningen research this governmental point of view obviously needs further clarification. For instance, the government's statement refers only to decisions regarding deliberate ending of life and does not address non-treatment decisions or decisions regarding administering pain relief medication, although these decisions may also include considerations concerning a child's future quality of life, particularly when discussing the futility of a specific medical treatment.

Whether or not it is legally sound to use expected, but not yet manifest pain and suffering as a ground for an end-of-life decision, obviously depends on the specific circumstances of a case. To allow the use of such reasons as the sole ground for such decisions is highly problematic, since the basis of these decisions would require certainty that the predicted pain and suffering will actually occur. Generally, such guarantees are hardly possible. In addition, there is limited data as to the survival rates and level of quality of life of infants with specific abnormalities, in particular those who have received every possible treatment in the past. Moreover, there is hardly any data as to the levels of suffering of such infants who do survive.

In any case, the Dutch Health Council declared it to be necessary to learn more about the requirement of *present* suffering [15], while the Dutch Paediatrics Association encourages the use of professional guidelines as an aid in establishing *expected* hopeless and unbearable suffering [16]. At present, the latter Association is preparing a guideline, which—among other things—will elaborate on the use of various representations of suffering in this regard.

9.5.2 Parental Requests to End Neonatal Life

The medical-empirical data also reveal that in seven cases (six in group II and the one in group III) the parents explicitly informed the attending physician that they did not want their child to suffer any further. These cases give rise to the question of whether parents have a legal right to request their infant's death. And they raise questions about the legal responsibility of the physician who complies with such a request.

Authoritative literature confirms that a parental competency to decide about life and death of a child is considered incompatible with the child's inherent right to life, protected under international human rights law. The basic reason for this view lies in a particular quality attributed to the right to life: its inalienable character. This inalienability reflects that the exercise of this right is not transferable to others, that respect for this right cannot be suspended, annulled or made void and that the bearer of this right cannot waive it.

However, the meaning of the inalienable character of the right to life's as well as its inherent quality is being debated. It is difficult to establish whether withdrawal or withholding of life-sustaining treatment, a particular medicinal regimen or even medical neonaticide constitutes a violation of the right to life. There is simply no univocal standard for legal interpretation of the inherency and inalienability concepts [17]. Moreover, the Groningen research has revealed that parents—at least in Dutch NICUs—sometimes act contrary to the above mentioned, presupposed characteristics of their infants' right to life. Physicians who are confronted with a parental request to end their child's suffering, that is life, are sometimes willing to satisfy such a request, by adapting the infant's medication in order to hasten death. This practice indicates a gap between what the theoretical nature of the right to life would allow and actual parental and medical conduct in the neonatal end-of-life decision-making process. The question remains: is this conduct unreservedly unacceptable, that is, illegitimate, or do end-of-life decisions in neonatology approach the limits of what the application of a human being's legal right to life can resolve? [18].

9.5.3 Use of Medication

Another issue revealed by the Groningen research is that the use of analgesics, sedatives and neuromuscular blockers (NMBs) is an important part of end-of-life care in Dutch NICU's [19]. This finding confronts legal scholars with the challenge to classify this use of medication in legal terms, that is to say, to determine whether or not a certain medicinal regimen is legally permissible.

In essence, this permissibility depends on the particular use of a pharmaceutical substance. Relevant in this regard are the nature and potential of a drug, the indication for administering, its dosage, the moment of administering, and not in the least, its foreseeable effect in relation to previously provided medication.

The medical-empirical data show, among other things, that in group I as well as in group II medication was increased after terminating the mechanical respiration. Furthermore, although the used dosages were sufficiently documented in the infants' medical files, the medical indications for the increase were not. To be specific, in 51 of the 94 group I-cases in which medication was increased after the decision to withdraw treatment, no reasons for the increase appeared to be documented. A similar lack of documentation was established in 100 of the 110 group II-cases in which medication was increased after treatment was withdrawn.

A decisive element in judging the use of medication is knowledge of the medication's nature. Generally, the use of analgesics and sedatives is regarded as part of a medicinal regimen that has no particular role in the death of a severely ill infant. The use of NMBs, however, is suspect, as it is common knowledge among physicians that paralyses result from administering NMBs and can cause the death of an infant. Because of this danger, the use of NMBs is believed to reveal a physician's obvious intention to let a child die and to have part in that process.

It is controversial that in 7 out of 22 group I-cases in which NMBs were administered after the non-treatment decision was effectuated no reasons for this administering were found in the infant's medical files. It is worrisome that in 19 of the 26 infants in group II who received NMBs after a non-treatment decision was made, no explanation for administering these NMBs was documented in the files. The physician's intentions to speed up the child's dying process became known only through the interviews. It goes without saying that without proper documentation, adequate external review of a case is hardly possible.

Remarkably, the traced use of analgesics and sedatives raised questions as well. The Groningen research shows that even the use of analgesics and sedatives sometimes occurs beyond intentions to prevent and treat symptoms (primarily restlessness), discomfort and gasping. In up to 10 % of all cases—primarily in gasping and moribund newborns—an increase of analgesics and sedatives also occurred with the intention to hasten death, a practice which was confirmed by some of the interviewed physicians in relation to 11 cases in group II. In essence, the question of how medication relates to an infant's death involves more than the mere use of NMBs.

Regarding the use of NMBs, the medical-empirical data show that these muscle relaxants were administered to a total of 55 infants (16 %) after termination of the mechanical respiration. In view of the lethal effect of NMBs, one would expect physicians to examine more than one case to identify medical neonaticide. However, none of the infant death cases which involved the use of NMBs has been reported to the judicial authorities [20].

Neither the physicians in charge of the 11 cases in group II qualified their conduct as medical neonaticide and they too failed to inform the judicial authorities. In their comments, the physicians claimed they were not convinced that this increase of medication actually caused the children to die. I believe this claim points to two problems: *First*, to actually prove that an increase of certain medication causes the

death of a very ill infant can sometimes be rather difficult. *Second*, physicians and lawyers evidently hold different views concerning the concept of "causation" [21].

9.6 Recent Developments

In recent years, new developments have fuelled the Dutch debate on how to deal with neonatal end-of-life decisions. Arguably, these developments suggest that (a part of) the Dutch neonatological community remains unsatisfied with the current legal-procedural settlement of medical neonaticide. The results of the official evaluation of the national multidisciplinary expert committee show that the current reporting procedure, which involves the scrutiny of this committee, does not produce the aimed results. Physicians believe the procedure offers no legal certainty and they have no confidence in it as well. Furthermore, they consider the defined due care requirements to be unclear and feel insecure about the consequences of their neonatal end-of-life performances and their reporting about it. Quite some physicians even seem unfamiliar with the reporting procedure as a whole. In line with these impressions the evaluation shows that since 2007 at least 11 cases of medical neonaticide occurred, of which a single case was merely reported [22]. In response to this evaluation the Dutch Ministers of Heath Care and of Justice have condemned this on-going non-reporting practice, but also realized that a procedure that does not accomplish its initial goal, is not effective. In result, the Ministers have decided to draw up a new settlement procedure, which shall include a more clear framework of review [23].

Some months prior to the publication of the expert committee's evaluation, the RDMA issued a Position Statement on neonatal end-of-life-decisions [24]. The purpose of this document is to provide an inventory of possible problems in end-of-life decisions concerning severely ill neonates and to offer recommendations on solutions for the signalized problems. While aiming to support neonatologists in their respective actions as well as the review of these actions, the inventory includes a clarification of the criteria for permissible end-of-decisions concerning neonates from a medical-professional perspective. The Position Statement's starting point is that parents are always to be involved in the decision making and that palliative care, including palliative sedation, may offer appropriate relief for a neonate's severe suffering. Yet, in cases in which the latter is not possible or achievable, the RDMA holds explicit views. Crucial in all this is the (internationally disputed) opinion that gasping may impose suffering to the neonate. In consequence, the administering of muscle relaxants in gasping children, whose pain cannot be alleviated by available pain relief medication and whose mechanical respiration is discontinued, is considered to be justified. The same goes for situations where the child's dying process becomes long-lasting, so as to cause severe suffering to the infant's parents. In such a case, the use of muscle relaxants is considered permissible too. In both situations, according to the RDMA, the case must be reported to the national multidisciplinary expert committee. Yet, in case muscle relaxants were

already part of a child's regular treatment, while awaiting this medication to be exhausted is not appropriate, the administering of muscle relaxants is considered normal palliative care. A neonate's death resulting from this is to constitute a natural death and should not be reported, because the use of the muscle relaxants occurs within the context of continuing medical treatment.

Several conclusions of the expert committee's evaluation and the RDMA's Positions Statement may be questioned, at least from a legal point of view. For instance, what has caused the Dutch neonatologists' negative impression of the expert committee?

In the past the neonatologists' community repeatedly insisted on a multidisciplinary panel of review experts, so as to be assured of more than a legal examination of cases. Besides, this medical community showed a clear willingness to assume and also share responsibility for its neonathanatic actions. Since the expert committee's review perception must relate to legal considerations too, the non-reporting of apparent medical neonaticide cases suggests that this committee, or any similar body, would be considered effective only if it shares the medical-ethical viewpoints prescribed by the neonatological community. However much understandable this may seem from a medically standpoint, such an approach can hardly be considered a prudent one. A medical-professional approach which does not take due account of well-considered legal views, other than the ones obviously used to demonstrate but one's formal attention to legal affairs, is at risk of gabling away one's necessary societal support. Since the actions of the multidisciplinary expert committee are being disqualified although no reviews of reported cases have proved the committee's inability to deal with the issues involved, the present non-reporting of apparent medical neonaticide cases might raise questions about the Dutch neonatalogical community's credibility all the same. For, while it pledged willingness to contribute to an acceptable societal settlement of medical neonaticide, it does not substantiate this willingness by following the self-requested procedural settlement, notably instigated by the Dutch government.

Specific parts of the RDMA's Position Statement can be criticized as well. For instance, the professional statement that parental suffering can be a justified cause for ending a neonate's life, lacks substantial argumentation against the view that this viewpoint is incompatible with a neonates inherent right to life. At present, the decisive justification for any neonatal end-of-life decision, apart from fulfilling due care requirements, is an newborn infant's present and manifest hopeless and unbearable suffering. To relativize this norm in terms of weighing or giving decisive meaning to the suffering of others as well, means to abandon the interest of the child as the core element of the whole issue.

A similar comment can be made against the RDMA-statement that neonatal death resulting from the use of muscle relaxants in a neonatological treatment context constitutes a natural death, which does not need to be reported. Let alone the evident legal objections against such an opinion, previous protests in this matter

by the Dutch Medical Forensic Association did not cause the RDMA to reconsider this viewpoint [25]. The experts committee's disapproving response may not have come as a surprise [26].

9.7 The Best Interest of the Child

Obviously, parents and physicians are expected to respect the fundamental rights of the child. At the same time, it is clear that parents generally aim to do only what is best for their child. This disposition even serves a legal interest of the infant, as to do what is in a child's best interest is obligatory under Article 3 (1) CRC. This provision generally articulates this key imperative of the Convention and states that in all actions concerning children, whether undertaken by public or private institutions, the best interest of the child shall be a primary consideration.

The key question in our context is of course which conduct actually serves the sick and severely suffering infant's best interest. To my knowledge, no serious attempt has yet been made in children's rights literature to investigate how this quality of a specific conduct concerning a hopeless and unbearable suffering infant can be determined. Certain criteria for an adequate viewpoint on this matter must be considered important, if not decisive. In consequence, I believe that to substantiate a strong opinion on such a viewpoint one must:

1. identify the alleged best interest of the child, which means clarifying this interest in a specified, case-related and legally essential wording;
2. demonstrate why complying with this interest is *best* for the infant, which means showing that meeting this interest is the decisive need of the child in comparison to other needs;
3. show that all other relevant interests of the child have been identified and their relative weight has been assessed on explicit grounds;
4. show that the conduct necessary to meet the child's decisive need is adequate and sufficient to supply the child's particular need;
5. certify to what extent this conduct is a measure regularly used by medical professionals in order to serve the selected interest.

To comply with these "best-interest criteria" requires serious thought. Although these criteria do not simplify the decision-making process, they can be used as a guide to support an accountable interpretation of "the best interest of the child" in the context of end-of-life decisions in neonatology. However, certain challenges will remain, such as how to separate an infant's needs from the needs of the parents or from the needs attributed to the child by the physician due to his personal rather than medical-professional beliefs. Separating and identifying these needs makes it possible to estimate their true nature and makes it more clear as to whether they are rightly weighed. This line of thought may prevent one from making decisions unaware of their paramount incentives.

9.8 Conclusion

In this chapter some of the findings and thoughts on recent Dutch research and policy developments on end-of-life decisions in neonatology have been highlighted. Several more could have been addressed and analyzed.

To show that the issue of end-of-life decisions in neonatology is a children's rights issue, is one thing. To come to a more profound interpretation of a neonate's fundamental right to life in this particular context, is quite another. The Groningen research has shown that insights into actual medical practice and the particular difficulties neonatologists are confronted with, can improve the understanding of the purpose as well as the limits of the fundamental rights of the newborn child in the context of end-of-life decision-making. Such insights may enable us to ask keen and perhaps even more difficult question, and at the same time open possibilities to find other, more differentiated answers. It is one of the aims of the Groningen Center for Children's Rights in Health Care [27] to keep articulating such questions, to provide corresponding responses and to share them with all concerned.

References

1. Concluding observations of the Human Right Committee: Netherlands. Seventy-second session, 20/07/01, CCPR/CO/72/NET.
2. The studies of Van der Maas and Van der Wal revealed that physicians are still reluctant to report such cases to the judicial authorities. See van der Heide A, van der Maas PJ, van der Wal et al G. Medical end-of-life decisions made for neonates and infants in the Netherlands. Lancet 1997;350(9073):251–255; Vrakking AM, van der Heide A, Arts et al WF. Medical end-of-life decisions for children in the Netherlands. Archives Pediatrics Adolescent Med 2005;159(9):802–809. This conclusion was confirmed in van de Vathorst S c.s. Evaluatie Regeling deskundigen commissie late zwangerschapsafbreking in een categorie-2 geval en levensbeëindiging bij pasgeborenen. Den Haag: ZonMw, 2013, pp. 102–4.
3. See Fourth Periodic Report of the Kingdom of the Netherlands concerning the Implementation of the International Convention on the Rights of the Child (October 2006–December 2012), 2013, Annex blg-266402 of a Letter on 28th November 2013 (kst-26150–134) to the Second Chamber of Dutch Parliament. On the draft of this Report, see Annex of Parliamentary Papers II 2011–2012, 26150, nr. 123.
4. An international legal perspective on the matter is presented in Dorscheidt JHHM. Legal aspects of end-of-life decisions in neonatology. In: Roy GB, editor. Legal and Forensic medicine. Berlin, Heidelberg: Springer, 2013, pp. 1175–200.
5. Decree of 29 April 1994, Bulletin of Acts and Decrees (Staatsblad) 1994, 321.
6. Aanwijzing vervolgingsbeslissing levensbeëindiging niet op verzoek en late zwangerschapsafbreking,(GovernmentGazette (Staatscourant) 6 Mar 2007, nr. 46, p. 10.
7. A legally substantiated plea for such a committee can be found in Dorscheidt JHHM. Assessment procedures regarding end-of-life decisions in neonatology in the Netherlands. Med Law 2005;24:803–829.
8. Combined Annual Report of the Committee on late termination of pregnancy and ending of neonatal life 2009–2010, The Hague 2011, p. 6 and pp. 21–3. Up to this date (May 2015) no other cases were reported. See the Committee's Combined Annual Report 2011–2012, The Hague, 2013, p. 6 as well as Parliamentary Papers II 2013–2014, 33750 XVI, nr. 110, p. 5.

9. The Protocol is discussed by Verhagen AAE, Lantos JD. The groningen protocol. In: Diekema DS, Mercurio MR, Adam MB, editors. Clinical ethics in pediatrics. a case-based textbook. Cambridge: Cambridge University Press, 2011, pp. 131–6. A medical update is presented by Verhagen AAE. Neonatal Euthanasia: lessons from the groningen protocol. Seminars Fetal Neonat Med 2014;19:296–9.
10. Dorscheidt JHHM. Levensbeëindiging bij gehandicapte pasgeborenen. Strijdig met het non-discriminatiebeginsel? The Hague: SduUitgeverij, 2006. See also Dorscheidt JHHM. End-of-life decisions in neonatology and the right to life of the disabled newborn child: impressions from the Netherlands. In: Clements L, Read J, editors. Disabled people and the right to life, the protection and violation of disabled people's most basic human right. Oxford: Routledge, 2008, pp. 176–4.
11. Verhagen AAE, Dorscheidt JHHM, Engels B, Hubben JH, Sauer PJJ. End-of-life decisions in severely ill newborns in the Dutch NICU. Arch Pediatr Adolesc Med. 2009;163(10):895–901.
12. In 3 of the 150 cases in group II, data regarding the death of the particular child could not be traced due to a lack of information on who was the physician in charge.
13. Verhagen AAE, De Vos M, Dorscheidt JHHM, Engels B, Hubben JH, Sauer PJJ. Conflicts about end-of-life decisions in NICUs in the Netherlands. Pediatrics 2009;124:e-112–e-119.
14. Parliamentary Papers II 2005–2006, 30300 XVI, nr. 90, p. 4.
15. Dutch Health Council. Overwegingen bij het beëindigen van het leven van pasgeborenen, Signalering ethiek en gezondheid 2007/1. The Hague, pp. 35–6.
16. Verhagen AAE, van der Hoeven MA, Goudoever JB, de Vries MC, Van Schouten-Van Meete-ren AY, Alberts MJ. Uitzichtloos en ondraaglijk lijden en actieve levensbeëindiging bij pasgeborenen. Dutch J Med. 2007;151(26):1474–7.
17. Dorscheidt JHHM, for an in-depth discussion of this question. In: Clements L, Read J, editors., Open circuits pp. 182–186.
18. For a further reflection on this question as well as others in relation to the parental perspective, see Dorscheidt JHHM, Verhagen AAE, Sauer PJJ, Hubben JH. Parental involvement in end-of-life decisions in neonatology: legal considerations with regard to Dutch medical practice. Med Law Int 2011;11(1):1–22.
19. Verhagen AAE, Dorscheidt JHHM, Engels B, Hubben JH, Sauer PJJ. Analgesics, sedatives and neuromuscular blockers as part of end-of-life decisions in Dutch NICUs. Arch Dis Childhood Fetal Neonat Ed. 2009;94(6):F434–8.
20. According to the national multidisciplinary expert committee the use of muscle relaxants in the terminal phase equals active ending of life. For the purpose of pain relief other medication should be administered. See Combined Annual Report 2009–2010, Open Circuit, p. 17.
21. Dorscheidt JHHM, Verhagen AAE, Sauer PJJ, Hubben JH. Medication regimes in the context of end-of-life decisions in neonatology: legal considerations with regard to Dutch NICU-practice. Med Law 31.
22. van de Vathorst S c.s. Evaluatie Regeling deskundigencommissie late zwangerschap safbreking in een categorie-2 geval en levensbeëindiging bij pasgeborenen, Den Haag: ZonMw, 2013, pp. 99–104.
23. Parliamentary Papers II 2013–2014, 33750 XVI, nr. 110, pp. 8–12.
24. Royal Dutch MedicalAssociationPosition Statement (KNMG-Standpunt): Medische beslissingen rond het levenseinde bij pasgeborenen met zeer ernstige afwijkingen, Utrecht, May 2013.
25. See http://medischcontact.artsennet.nl/Actueel/Nieuws/Nieuwsbericht/133294/Arts-mag-leven-pasgeborene-beeindigen.htm. Viewed—once again—in Dec 2014.
26. See the experts committee's letter to the Dutch Minister of Health Care, 16 Sept 2013. http://www.lzalp.nl/Images/Standpunt%20commissie%20LZALP%20inzake%20KNMG%20rapport%20pasgeborenen%202013_tcm14-37363.pdf.
27. http://www.rug.nl/research/groningen-centre-for-law-and%20governance/organisatie/gckg/.

Chapter 10
The Lure of Technology: Considerations in Newborns with Technology-Dependence

Brian Carter and Laura Miller-Smith

Abstract For a minority of children managed in the NICU, there is a need for more complex technologic assistance in order to sustain life, mitigate a more chronic debilitation from a pervasive life-limiting condition, or provide a bridge from life-sustaining therapy to a more semi-permanent treatment such as organ transplantation. This chapter will address two major types of technology assistance for infants and children—tracheostomy and assisted home ventilation, and dialysis—and the myriad complications and considerations that they raise. Some attention to why clinicians may be so inclined to impose technology as a solution to life-limiting conditions will be noted, as well as why some parents may seem to insist on pursuing technology.

> There is a technological possession and impetus, and a mandate and momentum of technology in health care [1]

Newborns with prenatal or postnatal diagnoses of life-limiting or life threatening conditions, regardless of gestational age, are commonly managed in the neonatal ICU (NICU). They may have an urgent need to receive life-supportive technology and still cling precariously to life as their physiology and potential for continued life unfolds over the initial days following birth. In North America, many of these children are born in subspecialty hospital settings with pediatric expertise. Their diagnoses include those in Table 10.1, among others. Some may have been evaluated and their mothers cared for prenatally in what have become known as Fetal Care Centers [2]. Others may be diagnosed after birth—perhaps even after experiencing some physiologic decompensation during transition to extra-uterine life. These infants are then referred and transported to specialty pediatric centers and

B. Carter (✉)
Department of Pediatrics, University of Missouri-Kansas City School of Medicine, 2411 Holmes Street, Kansas City, MO 64108, USA
e-mail: bscarter@cmh.edu

L. Miller-Smith
University of Missouri-Kansas City School of Medicine, Children's Mercy Hospital, Division of Critical Care Pediatrics, 2401 Gillham Road, Kansas City, MO 64108, USA
e-mail: lmillersmith@cmh.edu

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_10

Table 10.1 Diagnostic conditions in technology-dependent newborns

CNS	
– Apnea	Cardiorespiratory monitor
– Central Hypoventilation Syndrome	Tracheostomy and ventilation; P/ox
– Ohtahara Syndrome	Ketogenic diet; medications; gastrostomy tube
– Spinal Muscular Atrophy	Gastrostomy tube; tracheostomy +/− ventilation
– Myotonic Dystrophy	Pulse-oximeter; Percussive vest/cough-assist
Lung	
– Tracheal occlusion/stenosis	Tracheostomy +/− ventilation
– CLD /BPD	Oxygen; tracheostomy/ventilation, pulse-oximeter, inhalational agents
– Tracheo-bronchomalacia	Tracheostomy + PEEP/ventilation; P/ox
– Pulmonary hypertension	Oxygen; tracheostomy + PEEP +/− ventilation; inhalational agents; pulse-oximeter; medications; gastrostomy tube
– Hemi-diaphragmatic paralysis	Diaphragmatic pacer
Heart	
– Congenital heart block	Pacemaker
– Cyanotic congenital heart disease	Medications; surgery; pulse-oximeter; gastrostomy tube; transplantation
Kidney	
– Chronic renal failure (multi-cystic Dysplastic kidneys, obstructive uropathy, ARPKD)	Peritoneal dialysis; gastrostomy tube
Gastrointestinal	
– Short-gut	Ostomy supplies; gastrostomy tube; possible home IV nutrition (per central line)

continue to receive care in the NICU. They generally require life-supportive technologies that are commonly applied for a relatively short duration—with an expectation of physiologic stabilization, then improvement and a future that generally does not rely heavily on medical technology. Discharge care plans and coordination of home care services may include the temporary provision of oxygen, medications, cardiopulmonary monitoring, tube feedings, or urinary catheterization. On occasion, a child may require continued intravenous antibiotics (per central venous access), serial casting (e.g. for club feet), wound care following a serious infection, or surgical dressing changes. These post-hospitalization care requirements have become fairly normative in North America over the past 25 years, affecting increasing numbers of children [3].

For a minority of children managed in the NICU, there is a need for more complex technologic assistance in order to sustain life, mitigate a more chronic debilitation from a pervasive life-limiting condition, or provide a bridge from life-sustaining therapy to a more semi-permanent treatment such as organ

transplantation. This chapter will address two major types of technology assistance for infants and children—tracheostomy and assisted home ventilation, and dialysis—and the myriad complications and considerations that they raise. Some attention to why clinicians may be so inclined to impose technology as a solution to life-limiting conditions will be noted, as well as why some parents may seem to insist on pursuing technology.

10.1 Tracheostomy

Tracheostomy is an increasingly common procedure in chronically ill and ventilator dependent infants in the NICU. Tracheostomy can be performed to overcome anatomical airway abnormalities—such as congenital tracheal stenosis o postnatal subglottic stenosis—and may not require ongoing mechanical ventilation. In other instances, the presence of tracheomalacia may prompt the placement of a tracheostomy to facilitate long-term assisted ventilation and positive-end-expiratory pressure (PEEP) as a therapeutic intervention for months to years as the airway continues to grow and become more rigid. Tracheostomy also can be used for managing chronic neonatal lung disease (chronic lung disease, CLD; or bronchopulmonary dysplasia, BPD) and be accompanied by chronic ventilator use. In some neurologically impaired newborns and young infants, tracheostomy may be performed as a measure to protect their airway that is considered to be at risk for collapse, or aspiration, and/or assisting with ventilation when there are disorders of central respiratory drive. Regardless of the indication, tracheostomy can be a life-saving procedure. However, it can be accompanied by serious medical complications, in addition to social and ethical burdens that may impact the patient and family's long-term outcome. As the use of tracheotomy has been increasing in North America, limited data is available about the outcomes of these children, and how to best advise families on pursuing this treatment [4].

In a recent study conducted in 2006, it was noted that there were approximately 7800 patients discharged from US hospitals on long term mechanical ventilation following tracheostomy. This was a 55 % increase since 2000, with hospital charges increasing 70 % over that same period [5]. In addition to significant in-hospital time and expense, these patients also required significant in-home health care resources, with home nursing and medical supplies. While not the focus of the study, there were in this same era children who received tracheostomy without associated ventilator dependence. Such children would also require escalated in-home support.

Regardless of the need for assisted ventilation following tracheostomy, infants receiving a tracheostomy are at risk for related medical complications. A study from Alberta Children's Hospital in Canada reported a 90 % incidence of infection, 56 % incidence of tracheal granulation, 10 % incidence of mucous plugging resulting in cardiopulmonary arrest, and a 10 % risk of accidental decannulation [6]. Tracheostomy placement can be seen as a long-term medical commitment. Evidence from Children's Hospital of Los Angeles, looking at patients with

tracheostomy with long-term mechanical ventilation between 1977 and 2009, revealed that 61 % of patients remained on outpatient mechanical ventilation, 18 % were off of ventilator support, and the remainders were deceased [7].

Following tracheostomy, there are considerable delays in discharging patients home from the hospital. These delays have been associated with a myriad of issues, including insurance coverage, recruiting local home health nursing and respiratory care, limitations of support in certain geographic locations, constructing the optimal home environment to safely provide needed technology, and overcoming various social issues regarding family relationships, substance abuse, language barriers, education level and employment [8, 9]. Educating parents on necessary tracheotomy care and close patient monitoring can be time consuming, and can also account for discharge delays beyond a patient's clinical readiness. In addition, tracheostomy can be associated with more frequent outpatient clinic visits, ED visits, and inpatient hospitalizations.

There is limited data on how parents perceive both their own and their child's quality of life following tracheostomy. One study from London found that tracheostomy on a child had a varying impact on the families' quality of life. Caregivers reported an impact on their personal health, ability to sleep, and have strong relationships within and outside of the family. They also reported more difficulty maintaining paid employment—and thereby had reduced annual income. Caregivers also report difficulty receiving the amount of nursing hours they feel is needed to support their child [10]. A survey from Duquesne University in the United States, using a validated model to determine family distress, found that parental caregivers of infants and toddlers with a tracheostomy at home were in moderate distress and had a quality of life lower than that of the average age-matched healthy adult [11].

Because of these factors, decisions regarding the placement of a tracheostomy and embarking on long-term home respiratory care can create moral distress among health care workers and parents alike. When surveyed, >60 % of physicians report that they would definitely not want life sustaining treatment should they require being maintained on a ventilator [12]. However, as stated, tracheostomy is being performed at an increasing rate, with many such children requiring long-term ventilation. This creates an ethical tension in the decision to pursue tracheostomy, as clinicians may be recommending a procedure and associated therapy that they would not want for themselves. This issue is further compounded when a child has other complex medical problems, perhaps birth anomalies requiring other surgeries, and/or profound neurological disabilities. Ethical questions may arise about whether pursuing tracheostomy with home ventilation is in the best interest of the child patient, and whether it is worth the costs and consequences accrued [13]. Yet, there is little published data to describe how often this tension translates into medical practice, and how often involvement of an ethics committee or palliative care service may be pursued to investigate these concerns.

In the authors' center, there are more than 50 tracheotomies performed each year, with the number continuing to climb. The patients come either from the neonatal or pediatric intensive care units. The process for identifying which patients are optimal

candidates for the procedure is, at times, inconsistent and a better way to predict potential outcomes or complications is currently being sought. Many of the long-term outpatient outcomes are unknown to the inpatient physician that conduct counseling for families and advise them about tracheotomy. Neonatologists and critical care clinicians that do not participate in long-term follow-up clinics are in a sense blind to the net risks and benefits over time—being more familiar with tracheostomy as a means to getting the patient out of the ICU. In such a setting, the adequacy of procedural informed consent may not be optimal for the family. Patients that will ultimately be followed by a home ventilator team, pulmonologist or otolaryngologist as an outpatient, should likely receive a pre-tracheostomy consultation by this future managing team in order to gain the greatest understanding of their potential needs and outcome. Hopefully, in the future having data on the outcomes and complications for these patients will optimize informed consent for such procedures.

Tracheotomy can be a life-saving procedure, but it can have significant complications that impact the patient and family over an extended period of time and across care environments. In order to ensure the best possible outcome, clinicians need to understand who is receiving the procedure, their long term outcomes, and their medical, social and ethical ramifications.

10.2 Dialysis for Newborns and Young Infants

Renal failure may occur in newborns and young infants due to acute crises (asphyxia, sepsis, shock) or as a reflection of a longer-standing end-stage renal disease (ESRD). In the former, acute kidney injury (AKI) can be transient or complicated by protracted oligo-anuria, systemic hypotension requiring vasopressors drugs, multiple organ-system failure and acidemia, or the need for assisted ventilation and dialysis—all of which have been associated with increased mortality in children [14–16]. For newborns with ESRD, the common etiologies are long-standing problems of renal maldevelopment and include autosomal recessive polycystic kidney disease (ARPKD), renal dysplasia that may result in small kidneys or cystic dysplastic kidneys, and obstructive uropathies. Congenital absence of the kidneys (Potter's syndrome) is generally considered to be lethal, although technologic support can theoretically be mustered [17].

In many cases of prenatally diagnosed ESRD, questions arise about what might be accomplished to ameliorate the renal disease or the associated oligohydramnios and pulmonary hypoplasia. The previous section addressing tracheostomy and potential home ventilation may be relevant for some children with ESRD associated with pulmonary hypoplasia, pulmonary hypertension, or eventual chronic lung disease from long-term ventilator assistance to manage these entities. Should an infant with severe ESRD as a newborn survive initial management in the NICU with assisted ventilation, ECMO, CVVH or dialysis s/he may yet remain ventilator dependent in addition to dialysis dependent.

For newborns requiring early dialysis (the first postnatal week or month), many conditions might impact the success or failure of efforts to support the infant's renal function and maintain short-term fluid and electrolyte balance and longer-term growth. These include issues of prematurity—is the baby born at under 37 weeks' gestation; if so, how early, and how small? Matters of patient ethnicity (African-American children fare worst), postnatal oligo-anuria, and co-morbid conditions also become important in prognosticating successful dialysis and infant growth. Such complications might include chromosomal anomalies or syndromes, other birth defects, severe cardiopulmonary disease, sepsis, and anything that might preclude placement and reliable dependence upon a peritoneal dialysis (PD) catheter. For those receiving PD, the potential for sepsis/peritonitis, PD catheter dysfunction, and poor nutritional maintenance may complicate many patients' courses [18–20].

Little has been written about the parental realities of living with children on home-dialysis while awaiting potential kidney transplantation. However, across North America the expectation that parents accommodate to this level of care is often assumed. Locally, at Children's Mercy Hospital in Kansas City, Missouri (USA) all families having a newborn for which dialysis is being considered are seen by an ethics consultant and also by a palliative care consultant. The intent is to illuminate parental awareness to the life-changing nature of their child's condition, explore values and psychosocial, spiritual, and logistical considerations for ongoing care and to enter into PD in a state of informed awareness. As noted by the parental reports from Australia, there is a rather involved process of adaptation to accepting the need for medical technology and providing it in their homes [21]. Parents in the Australian study reported a number of steps in their home-care journey: first, learning about and struggling with the diagnosis of ESRD and its permanence. Parents even noted the trauma—commented upon by other parents whose children have different conditions—in seeing their child having to endure numerous procedures while hospitalized and even in having to be strong in advocating and negotiating care for their child. Secondly, these parents speak to the medicalization of parenting—their becoming exhausted and overwhelmed as they assumed a caregiver role in their child's daily dialysis, contending with both psychological and behavioral issues with their child as well as dialysis-related complications and the ever present fear of potential rehospitalizations—for which they would assign self-blame—and parenting in a manner they never anticipated. Thirdly, family norms became disrupted affecting spousal relations, sibling attention, and even future family planning [21, 22]. Finally, these parents, in a manner of accommodation and adjustment over time revealed how much they relied upon others—including their child's clinicians and other parents of children with ESRD—for support in coping [21].

It would appear that parents are at the same time, then, juggling the tasks of absorbing the clinical environment, adapting to the medicalization of parenting, contending with disrupted family norms—and creating the "new normal" as they develop effective coping strategies and support structures [21]. They need information that addresses nutrition, financial and psychological realities and available

support, and access to peer support groups. Behavioral health expertise may also be beneficial for parents, the family unit, and the affected child. Building these supports into any dialysis program seems essential when technology is anticipated to "come home" with the child. While these measures are taken, the quality of the child's life is at times questioned by some parents. Few existing studies address quality of life for children on dialysis, but early indications are that the health-related quality of life of the child as reported by parents differs for that reported by the affected child in the same household [23, 24].

Must all neonatal and young infant dialysis candidates, then, receive this technology? The ethical considerations are myriad, and perhaps a bit unique when compared to other innovative or even standardized treatments of chronically ill children with technology. In one attempt to answer this question, Lantos and Warady note some of the unique ethical challenges posed by infant dialysis [25]. It is a relatively "new" technology in pediatric medicine, and it is potentially applicable to an array of patients who cannot all be viewed as necessarily likely to benefit similarly. As the causes of ESRD vary, and the existence of other comorbidities must be considered, families may have a difficult time hearing that their child does not likely stand to benefit from dialysis—or may only do so briefly. In addition to those concerns voiced by parents noted above, the time commitment, hospital/clinic utilization, home-care needs and resource utilization may be viewed by some parents as excessive. Some have expressed that they see a disproportionate burden on the child and family compared to any potential benefit. As dialysis is always a short-term form of management, a bridge until kidney transplantation—the average age at which transplantation can be reasonably anticipated being between 18 and 30 months—they may see dialysis as imposing a very long-term commitment to a life encumbered by technology.

For parents of children whose lives may be dependent upon technology, there is a need to consider all of what the technological solution brings with it. Parents must endure their decisions—live with them—and be able to construct, or write their narrative in a manner that they will be able to live with. Ultimately the decision to employ technology at home for their chronically and critically ill children is theirs. One mother who refused technological attempts to ameliorate her newborn's condition stated this eloquently, "So, from day one, life becomes about 'not dying' and I don't see that is the same thing as 'living' [26]. In the end, she—as all parents—needed to be able to answer the question that often lingers, "*What will I think of myself in the years ahead when I remember the decision I make today?*" [26].

10.3 Responding to "Do Everything"

When hearing parental insistence to "*do everything*," clinicians need to pause and make some inquiry into what, precisely, is meant by this request. "*Everything*" may imply all that is imaginable, or available; what might be affordable, safe, or beneficial; anything that might be worthy of a trial; what the patients whose stories I

read on the Internet say I should expect; or more. "Do everything," might be an imploring request for clinicians to understand the predicament a parent finds themselves in while facing a life-long, life-changing, or life-limiting diagnosis in their child—at the mercy of a complicated health care system, stripped of power, control, and an ability to 'parent,' protect or nurture their child [27]. In understanding this latter possibility, the clinician may be asked by the parent(s) or others in the family, "*Doctor, what would you do?*" [28]. Such a request from parents need not be responded to by platitudes such as "*I don't know, I'm not you and haven't lived your experiences;*" or "*I could never answer that, we are different people with different values and belief systems*" [28, 29]. Rather, the clinician can take the query put forward this way as an invitation to empathy—a parents' request to "*put yourself in my shoes*," or "*try to understand what it must be like to be me right now,*" and render an empathic response that is both informed by clinical experience and wisdom, and also an identification with another member of the human community who is presently suffering [30].

But can we really "do everything?" Of course not—we are limited by the choices we make and the consequences that these choices entail. To pretend otherwise is to intentionally ignore clinical and logistical realities. Persisting in the language of "doing everything" also becomes confusing, for it is subject to the interpretations that are rendered by various individuals who see, and hear, and expect different things—"everything" being a reflection of their lived, hoped for, or feared perspectives [31].

Parents may ask that everything be done because what they have been told by generally well-meaning clinicians—or at least what they have heard—is, "*Well, we can do this [place a tracheotomy; perform dialysis; operate; cannulate a baby for ECMO,* etc.*];*" simply as a response to a crisis and a desire to "do something." Clinicians are inclined to "do"—by nature, training, and the reinforcement of practical clinical experience. When intensive care clinicians "do something" they often employ the tools, the *techne*, and the technology at their disposal. When parents hear "We can do…" they may believe that doing such is both reasonable and likely beneficial, or else the physician wouldn't bring it up. Carefully attending to not only how words are spoken, but the choice of words and phrases used—and how they may be heard or interpreted –may mitigate this misunderstanding of what can be done being what should be done. In some instances, specialist clinicians need to understand that a good clinical outcome that can possibly be obtained by doing something may not be the best goal to work toward [32].

In one sense, seeing that clinical caregivers "*do something*" can be reassuring to families—all is not lost because there is something else to do [33]. So the imploring cry to "do everything" speaks to a family's desire to not be abandoned, be minimized, or be excluded from consideration [31]. It may also be a response to parental support and address of their needs during their child's hospitalization [34]. Finally, as well described by neonatologist Art Kopelman, parental insistence on "doing everything"—even what may appear to be futile or apparently inappropriate care—may represent some deeper underlying need, misunderstanding, belief or world view [35]. Parents may believe that an infant can survive and be normal or to have

only mild learning problems as they clearly misunderstand a prognosis (perhaps due to the use of medical jargon), or their emotions affect a sense of denial. They may believe that a higher power will perform a miracle; that the great number of members of their faith community who are praying for their infant will effect such a miraculous outcome. Or perhaps they believe that if they agreed to limit or stop life support, members of their faith community would see them as lacking faith in God's ability to heal—their understanding of God to be all powerful and inclined to intervene to affect a healing. Parents may also know of similar cases, or have a history in their own infant's case, where the doctors' predictions were wrong, engendering doubt. Should they be distrusting or feel disenfranchised, they might suspect that their infant has not received the best possible care. When parents see survival of their infant as the only solution to a life crisis, perhaps relational, they may imbue the survival of their infant with unrealistic promise for holding a family together or giving meaning and purpose to their lives. The likelihood of an infant having moderate-to-severe disabilities may also not be as important to some parents as might be suggested by their clinical caregivers. Parents may believe that all life, for every child, even when complicated by profound disabilities, has great value and should be preserved. They may be prepared to commit themselves to raising their child if s/he survives, regardless of any limitations [35].

Pediatric specialty care, and neonatal intensive care in particular, has benefited from the availability of technology. New tools have advanced the field, decreased mortality and made often life-threatening or life-limiting conditions bearable, survivable and offered both parents and children new hopes. But as with all innovations, new tools, and technologies, life-support and chronic complex care technologies in healthcare have come with a price. Not only measured in dollars and finite resources, the price includes human cost—lives of families, relationships, parental roles, spiritual and existential meaning, the place and role of healthcare within individual and family life, and more broadly within society. As noted by Postman, "*…embedded in every tool is an ideological bias, a predisposition to construct the world as one thing rather than another*" [36]. Clinicians and parents need to work together and be willing to explore such biases before—and even after—the implementation of technological tools for newborns and young infants.

References

1. Hofmannin B. Is there a technological imperative in health care? Internat J Technol Assess Health Care. 2002;18(32):675–89.
2. American College of Obstetricians and Gynecologists Committee on Ethics and American Academy of Pediatrics Committee on Bioethics. Maternal-fetal intervention and fetal care centers. Committee Opinion No. 501. Obstet Gynecol. 2011;118:405–10.
3. Feudtner C, Villareale NL, Morray B, Sharp V, Hays RM, Neff JM. Technology-dependency among patients discharged from a children's hospital: a retrospective cohort study. BMC Pediatrics. 2005;5(1):8.

4. Amin R, et al. Pediatric long-term home mechanical ventilation: twenty years of follow-up from one Canadian center. Pediatr Pulmonol. 2014;49:816–24.
5. Benneyworth BD, et al. Inpatient health care utilization for children dependent on long-term mechanical ventilation. Pediatrics. 2011;127:e1533–41.
6. Al-Samri M, et al. Tracheostomy in children: a population-based experience over 17 years. Pediatric Pulmonol. 2010;45:487–93.
7. Edwards JD, et al. Outcomes and causes of death in children on home mechanical ventilation via tracheostomy: an institutional and literature review. J Pediatr. 2010;157:955–9.
8. Graf JM, et al. Children with new tracheostomies: Planning for family education and common impediments to discharge. Pediatr Pulmonol. 2008;43:788–94.
9. Rafferty A, et al. Retrospective, cross-sectional review of delayed discharge after paediatric tracheostomy. J Laryngol Otol. 2012;126:1247–53.
10. Hopkins C, et al. The impact on paediatric tracheostomy on both patient and parent. Int J Pediatr Otorhinolaryngol. 2009;73:15–20.
11. Joseph RA, et al. Parental quality of life: caring for an infant or toddler with a tracheostomy at home. Neonatal Netw. 2014;33(2):86–94.
12. Needle JS, et al. Influence of personal preferences for life-sustaining treatment on medical decision making among pediatric intensivists. Crit Care Med. 2012;40:2464–9.
13. Wilfond BS. Tracheostomies and assisted ventilation in children with profound disabilities: navigating family and professional values. Pediatrics. 2014;133(1):S44–9.
14. Bresolin N, Silva C, Halllal A, et al. Prognosis for children with acute kidney injury in the intensive care unit. Pediatr Nephrol. 2009;24:537–44.
15. DurkanAM Alexander T. Acute kidney injury post neonatal asphyxia. J Pediatr. 2011;158: e29–33.
16. Chang JW, Tsai HL, Wang HH, Yang LY. Outcome and risk factors for mortality in children with acute renal failure. Clin Nephrol. 2008;70:485–9.
17. Bienstock JL, Birsner ML, Coleman F, Hueppchen NA. Successful in utero intervention for bilateral renal agenesis. Obstet Gynecol. 2014;124 (2 Pt 2 Suppl. 1):413–5.
18. Hijazi R, Abitbol CL, Chandar J, Seeherunvong W, Freundlich M, Zilleruelo G. Twenty-five years of infant dialysis: a single center experience. J Pediatr. 2009;155:111–7.
19. Kari JA, Gonzalez C, Ledermann SE, Shaw V, Rees L. Outcome and growth of infants with severe chronic renal failure. Kidney Internat. 2000;57:1681–7.
20. Klaassen I, Neuhaus TJ, Mueller-Wiefel DE, Kemper MJ. Antenatal oligohydramnios of renal origin: long-term outcome. Nephrol Dial Transplant. 2007;22:432–9.
21. Tong A, Lowe A, Sainsbury P, Craig JC. Parental perspectives on caring for a child with chronic kidney disease: an in-depth interview study. Child: Care Health Dev. 36, 4:549–557.
22. Laakkonen H, Taskinen S, Rönnholm K, Holmberg C, Sandberg S. Parent–child and spousal relationships in families with a young child with end-stage renal disease. Pediatr Nephrol. 2014;29:289–95.
23. Abrahão SS, Ricas J, Andrade DF, et al. Difficulties experienced by children/adolescents with chronic kidney disease and by their families. J Bras Nefrol. 2010;32:16–20.
24. Park KS, Hwang YJ, Cho MH, et al. Quality of life in children with end-stage renal disease based on a PedsQL ESRD module. Pediatr Nephrol. 2012;27:2293–300.
25. Lantos JD, Warady BA. The evolving ethics of infant dialysis. Pediatr Nephrol. 2013;28:1943–47.
26. Carter BS. How can we say to neonatal intensive care unit parents amid crisis, "You are not alone"? Pediatrics. 2002;110:1245.
27. October TW, Fisher KR, Feudtner C, Hinds PS. The parent perspective: "being a good parent" when making critical decisions in the PICU. PediatrCrit Care Med. 2014;15:291–8.
28. Meyer EC, Lamiani G, Foer MR, Truog RD. "What would you do if this were your child?" Practitioners' responses during enacted conversations in the United States. PediatrCrit Care Med. 2012;13:e372.
29. Korones DN. What would you do if it were your kid? N Engl J Med. 2013;369:1291–3.
30. Halpern J. What is clinical empathy? J Gen Intern Med. 2003;18:670–4.

31. Feudtner C, Morrison W. The darkening veil of “do everything. JAMA Pediatr. 2012;166:694–5.
32. Hoffmann PB, Schneiderman LJ. Physicians should not always pursue a good “clinical” outcome. Hastings Cent Rep. 2007;inside back cover.
33. Hirni K, Carter B. Hearing others’ perspectives when we hear, “do everything!”. JAMA Pediatr. 2015;169(5):423–4.
34. Shields l, Kristensson-Hallstrom I, O’Callaghan M. An examination of the needs of parents of hospitalized children: comparing parents’ and staff’s perceptions. Scand J Caring Sci. 2003;17:176–184.
35. Kopelman A. Understanding, avoiding, and resolving end-of-life conflicts in the NICU. Mount Sinai J Med. 2006;73:580–6.
36. Postman N. Technopoly: The surrender of culture to technology. New York: Vintage Books;1993. p. 13.

Chapter 11
Making Tough Ethical Choices in a Morally Pluralistic World

John Lantos

Abstract This book uses neonatal intensive care as an example of the complex decisions that must be made as we try to harness ambiguous technology to human ends. The answers that we have come to about decisions in the NICU are not settled or final. The process of coming to moral consensus is iterative, non-linear, and ongoing. Scientific discoveries change the way we think about what it means to be human and what it means to live in community. Each story of scientific discovery and innovation is a story of the struggle to find the balance between the new emerging possibilities for human flourishing, and the risks and dangers that are held within it.

This book presents discussions of difficult dilemmas in neonatal bioethics. There are many issues about which reasonable people disagree. The process of hammering out a moral consensus within pluralistic, diverse, democratic societies is a messy one. It requires discussion and debate among scholars in fields as diverse as economics, moral philosophy, law, literature, disability studies, epidemiology, psychology, communication studies, theology, sociology, political science and anthropology. Discussions take place with families and doctors at the bedsides of critically ill babies, in the media, in film and literature, in scholarly journals, in the courts, in profess and in government advisory committees and task forces. This complicated discussion has allowed us, as a society, to clarify our understanding of and commitment to certain moral obligations to premature babies.

As the various chapters in this book show, there is still plenty of debate and discussion. Some people view today's policies and practices as inadequate or morally problematic in all sorts of ways. Some argue that parents' rights are violated by the current approach that focuses on the independent rights of baby. Some think we spend too much money on neonatal intensive care and that we should spend more on prevention. Some think that all of neonatology is a vast medical experiment being conducted without the consent of the research subjects and

J. Lantos (✉)
Children's Mercy Bioethics Center, Department of Pediatrics, University of Missouri-Kansas, Columbia, Kansas City, MO 65211, USA
e-mail: jlantos@mch.edu

E. Verhagen and A. Janvier (eds.), *Ethical Dilemmas for Critically Ill Babies*, International Library of Ethics, Law, and the New Medicine 65,
DOI 10.1007/978-94-017-7360-7_11

without appropriate research methodology. Such debates and disagreements are necessary because neonatal care, like all health care, is a collective effort.

The medical care of critically ill newborns and the research to improve that care are collectively financed. The mechanisms for governance of this multidisciplinary enterprise are not straightforward. Decisions are made and policies adopted as a result of the collective pushing and pulling of multiple interest groups, hundreds of lobbies, thousands of micro-markets, and millions of people expressing needs or desires of one sort or another.

The answers that we have come to about decisions in the NICU were not in any way obvious at the start. The fundamental question at the center of all bioethical debate is a question of value. Which efforts are worth the economic and psychological cost? We have limited financial and moral and psychological and intellectual resources. How should we allocate them?

One way to understand the ways in which we've made such allocation choices is to look at alternative choices that were considered but not adopted in our effort to meet our moral obligations to critically ill babies. As an example, an alternative approach to our current approach might have been to fund more preventive prenatal care, rather than funding neonatal intensive care. Many people argued for this approach. Some studies suggested that better prenatal care would reduce the need for intensive care. Wilson and colleagues studied the costs of prematurity and concluded that, "…had all women with inadequate prenatal care received Medicaid-covered adequate prenatal care, expenditure for this care would yield more than a two to one return in savings in NICU costs." [1]. Gorski and Colby came to similar conclusions, "For each additional $1 spent on prenatal care, $2.57 in medical care costs would be saved." [2].

By this view, neonatal intensive care was simply not the most cost-effective way to save more babies. Sinclair and colleagues noted, "The costs of neonatal intensive care are very high, and efficiency analyses comparing the costs and outcomes of intensive care programs and alternative programs will be required, if we are to continue to justify the existence and expansion of neonatal-intensive-care programs." [3]. Silverman suggested that neonatal intensive care was an "unreviewed and unlegislated social policy." He asked, "How much of its resources should the community invest in social interventions to prevent premature birth, and how much in medical rescue in neonatal intensive care?" [4]. Similarly, Kliegman argued that resources should be "redirected to…large population-based efforts to reduce the number of low-birthweight infants." [5].

Others questioned these analyses. Huntington and Connell showed that prenatal care did not really save money, even if it did lead to lower infant mortality [6]. Lu and colleagues examined "original research, systematic reviews, meta-analyses and commentaries for evidence of effectiveness of the three core components of prenatal care—risk assessment, health promotion and medical and psychosocial interventions—for preventing the two constituents of LBW: preterm birth and intrauterine growth restriction (IUGR)." After careful examination of the evidence, they conclude that "neither preterm birth nor intrauterine growth retardation can be effectively prevented by prenatal care." Similarly, Alexander and Koronbrot concluded

that "the empirical evidence supporting the association between prenatal care and reduced rates of low birth weight…has been equivocal."

These facts put to rest the idea that we had to make a choice between prevention and crisis-intervention. Instead, we had to provide both. Every country with a modern health care system has developed regionalized neonatal intensive care programs to as part of their response to the problems of infant mortality.

Another view that has been discarded is that we should not "play God" by either over-treating or forgoing treatment of critically ill babies. Early pioneers in neonatology understood that technology would change the way we thought about sick babies, about parents, about pediatric care, and about infant mortality. They recognized that this was a massive interference in nature but it was one that they thought was morally appropriate. Their vision challenged the conventional moral wisdom of the time.

Theologian Richard McCormick summarized the situation thus, "The availability of powerful new technologies that can sustain life almost indefinitely has forced us to ask: what are we doing when we intervene to stave off death? What values are we seeking to serve? How should we formulate these values in our time if we are to maintain (individually and collectively) our grasp on the basic values that define our well-being? Ought we sustain life when the individual gains nothing from such sustenance? And what does 'stand to gain nothing' mean?" [7].

These sorts of questions led to discussion about the distinction between a moral view that emphasized the sanctity of life with moral views that considered quality of life. The term "quality of life" has multiple meanings. In some contexts, it is used to refer to the subjective assessments of competent adults as to their own sense of happiness or well-being. As such, it is an important complement to objective measures of outcome. In other contexts, such as the one in which decisions must be made about treatment of critically ill newborns, the term refers to a third party's assessment of the suffering or the burdens experienced by a patient who is unable to express his or her own opinions. In that context, in particular, quality of life assessments have been particularly problematic and seemed to open the door to policies that would try to eliminate all citizens with disabilities of any sort. C. Everett Koop, a pediatric surgeon who would go on to become Surgeon General of the United States, expressed this fear,

> We are rapidly moving from the state of mind where destruction of life is advocated for children who are considered to be socially useless or have non-meaningful lives to a place where we are willing to destroy a child because he is socially disturbing. What we need is alternatives, either in the form of education or palliative measures for the individual as well as for society. [8]

As Surgeon General, Koop tried to incorporate these concerns and fears into the federal regulations governing treatment decisions in the NICU. His efforts to do so made clear the inevitability of incorporating some consideration of quality of life into decision making. The alternative, it seemed, was not heightened moral sensibility but, instead, an intolerant and intolerable vitalist ideology. If quality of life was not considered in any way, then it would be necessary to continue treatment of

all babies with every available technology all the time until the last heartbeat. That seemed more problematic than a more nuanced view of dangers of ever considering quality of life.

Once a technology becomes available, a decision must be made about its proper use. There is no moral imperative to use all available technology but there is a moral imperative to decide whether and how new technology should be used.

A proper response to this aspect of the moral imperative of technology is to make decisions as openly and deliberately as possible. One of the roles of modern bioethics is to facilitate that process of decision making.

This book uses neonatal intensive care as an example of the complex decisions that must be made as we try to harness ambiguous technology to human ends.

The answers that we have come to about decisions in the NICU are not settled or final. The process of coming to moral consensus is iterative, non-linear, and ongoing. Scientific discoveries change the way we think about what it means to be human and what it means to live in community. Things that once seemed good may come to seem problematic. We change the way we view cars, or pesticides, or nuclear fission, or life-sustaining medical technology. Usually, we don't accept or reject an innovation outright. We modify it. We regulate it. We build upon the initial discoveries in order to develop new and perhaps safer ways of deploying the new innovations. Sometimes, new possibilities for human flourishing emerge. But each new possibility is as double-stranded as a DNA molecule, holding within it both hopes and fears, risks and benefits, dangers and possibilities for the escape from danger. Each story of scientific discovery and innovation is a story of the struggle to find the balance between the two.

References

1. Wilson AL, Munson DP, Schubot DB, Leonardson G, Stevens DC. Does prenatal care decrease the incidence and cost of neonatal intensive care admissions? Am J Perinatol. 1992;9:281–4.
2. Gorsky RD, Colby JP Jr. The cost effectiveness of prenatal care in reducing low birth weight in New Hampshire. Health Serv Res. 1989;24:583–98.
3. Sinclair JC, Torrance GW, Boyle MH, Horwood SP, Saigal S, Sackett DL. Evaluation of neonatal-intensive-care programs. N Engl J Med. 1981;305:489–94.
4. Silverman WA. Neonatal pediatrics at the century mark. Per Biol Med. 1989;32:159–69.
5. Kliegman RM. Neonatal technology, perinatal survival, social consequences, and the perinatal paradox. Am J Public Health. 1995;85(7):909–13.
6. Huntington J, Connell FA. For every dollar spent—the cost-savings argument for prenatal care. N Engl J Med. 1994;331:1303–7.
7. McCormick RA. The quality of life, the sanctity of life. Hast Ctr Rep. 1978;8(1):30–6.
8. Koop CE. The slide to Auschwitz. The Pro-Life Forum. Accessed at http://www.prolifeforum.org/ethics/everett.asp on July 14 2004.