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Decision-making for extremely preterm infants with severe hemorrhages on head ultrasound: Science, values, and communication skills

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ABSTRACT

Severe intracranial hemorrhages are not rare in extremely preterm infants. They occur early, generally when babies require life-sustaining interventions. This may lead to ethical discussions and decision-making about levels of care. Prognosis is variable and depends on the extent, location, and laterality of the lesions, and, importantly also on the subsequent occurrence of other clinical complications or progressive ventricular dilatation. Decision-making should depend on prognosis and parental values. This article will review prognosis and the uncertainty of outcomes for different lesions and provide an outline of ways to conduct an ethically appropriate discussion on the decision of whether to continue life sustaining therapy. It is possible to communicate in a compassionate and honest way with parents and engage in decision-making, focussing on personalized information and decisions, and on function, as opposed to diagnosis.

Practice points

- Severe intracranial hemorrhage often lead to ethical discussions.
- When attempting to establish a prognosis for severe intracranial hemorrhages, clinicians should consider factors other than Papile grades, such as extent, laterality and other clinical factors. Prognosis will always be uncertain.
- Specific points regarding communication with patients in this context are provided, such as personalizing information and decisions.
- Accurate information is not sufficient to make decisions about withdrawing life sustaining intervention. Function and adaptation are essential to discuss with parents, more than diagnosis.
- Parental goals and values should be assessed for such a meeting/decisions

Case

We will use Mina's story for this article, published in the American Journal of Bioethics in 2022 [1].

"Helen came to the hospital with her husband Peter, presenting symptoms of threatened preterm labor at gestational age (GA) 23 weeks + 4 days. She had become pregnant while on contraceptives, and had 3 children, aged 3,5 and 9 years. Three hours later, Mina was born. Due to a rapid delivery, there was no possibility for proper prenatal counselling. Mina appeared vital at birth and was stabilized on non-invasive ventilation, and surfactant administration through a tracheal catheter. Her skin appeared immature, and she had transitory electrolyte disturbances during the first days of life. After 2 days, she was intubated due to apneas, and bilateral grade 2 intraventricular hemorrhage (IVH) was found, which progressed to grade 3

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Abbreviations

CA	corrected age
CP	cerebral palsy
GA	gestational age
ICH	Intracranial hemorrhage
IPH	intraparenchymal hemorrhage
IQ	intelligence quotient
LSI	Life sustaining interventions
NDI	neurodevelopmental impairment
NICU	neonatal intensive care unit
PDI	psychomotor developmental index
PHVD	post-hemorrhagic ventricular dilatation
US:	ultrasound
WLSI	withdraw of life sustaining interventions

on one side. The parents stayed in the Neonatal Intensive Care Unit (NICU) around the clock and participated in the care."

When faced with an infant who has an early brain injury who requires ongoing life-sustaining interventions (LSI), ethical decision making should be based on an evaluation of the likely outcomes of such an infant, which outcomes are relevant to the ongoing provision of LSI, and the goals of care. We will consider which outcomes of very preterm infants are relevant to parents, review the prognostic accuracy of early head imaging studies for those outcomes, and discuss how to approach LSI decisions in situations of uncertainty.

1. Introduction

Intracranial hemorrhages (ICH) are common in very preterm infants, and are usually graded using the Papile classification [2]. Grade 3 ICH, bleeding into the lateral cerebral ventricle(s) with dilatation, and grade 4 ICH, also referred to as intraparenchymal hemorrhage (IPH), are collectively referred to as severe ICH and usually combined in outcome studies. There is unfortunately some ambiguity in the classification, with grade 2 ICH (bleeding into the ventricles without dilatation) followed by early post-hemorrhagic ventricular dilatation (PHVD) sometimes being re-classified as grade 3 ICH. This complicates the interpretation of prognostic data. Additionally, IPH are very variable in location, size, and whether they are unilateral or bilateral, but outcome studies usually combine all IPH into a single group.

Severe ICH generally occur in the first week of life, with a relatively stable incidence [3], despite increasing survival of the extremely preterm [4]. Clinicians are confronted with preterm infants dependant on life-sustaining interventions (LSI) when they develop severe ICH. Ethical questions about whether LSI should be withdrawn or withheld may then arise.

1.1. What outcomes are important to parents of extremely preterm infants?

Long-term follow-up of preterm infants has four main goals: 1) to enhance developmental monitoring and early intervention for higher risk infants, 2) to improve perinatal care through audits and quality improvement, 3) to better inform parents through anticipatory guidance, 4) to measure endpoints in research studies. Physicians and researchers have chosen the outcomes systematically measured. It is only recently that parents of former preterm infants and individuals born preterm themselves have been questioned regarding outcomes that they find important [1,5–8].

Questions about the nature of outcomes that are appropriate for withholding/withdrawing life sustaining intervention (WLSI) have very rarely been investigated. Two relevant studies suggest that parents of children undergoing intensive care think that an inability of the child to communicate in the future would be a reasonable basis for WLSI [9,10]. Is it possible to predict such outcomes from early cranial imaging in the newborn?

Profound impacts on cognitive outcomes leading to an inability to communicate are uncommon in former preterm infants. The majority of prognostic data after ICH has evaluated associations with "neurodevelopmental impairment" (NDI), or severe NDI. NDI is usually defined by a low score on a developmental screening test at 18–24 months, or the diagnosis of cerebral palsy (CP). A few babies also qualify because of hearing or visual deficits. The definitions of NDI, and severe NDI, have been somewhat variable between studies. Profound intellectual disability, such as that which is generally considered appropriate by parents for WLSI, has very rarely been reported.

1.1.1. The prognosis of ICH

Table 1 is an empirical summary of the factors (imaging and clinical) to be considered during an ethical discussion regarding infants with severe intracranial hemorrhage.

1.1.1.1. Grade 3 hemorrhage vs IPH, and PVHD. There are few reliable data regarding the outcomes of grade 3 ICH in isolation, as most studies have combined them with IPH. Some studies suggest that outcomes are similar to grade 4 ICH [11], others that they are similar to grade 2 ICH [12]. A recent systematic review was unable to analyze outcomes of grade 3 hemorrhages alone, as, in 17 of 23 studies, grade 3 and 4 hemorrhages were combined [13].

In contrast, many studies describe the prognosis of infants with ventricular dilatation following intraventricular hemorrhages [14], which varies depending on severity, and also on the type and timing of intervention, which differs between centres [15] and countries [16].

1.1.1.2. Heterogeneity of IPH. Some IPH are localized and unilateral, others extensive and/or bilateral. In most cohort studies, all IPH are combined as a single group. It appears inherently likely that more extensive abnormalities will have greater short and long-term impacts [17], however, there are comparatively few data regarding laterality [18,19], or severity, and outcomes. Two scoring systems for severity of IPH have been evaluated. The Bassan score applies only to IPH and gives 1 point for each of being bilateral, affecting more than 2 brain regions, or having mid-line shift, therefore ranging from 0 to 3 [20]. The Al-Abdi score is much more complex, but can be applied to all grades of ICH, adding points for complicating features and ranging from 1 to 35; scores for IPH are between 16 and 35.

1.1.1.3. Outcome of severe ICH

1.1.1.3.1. Death. The majority of severe ICH are asymptomatic and discovered on routine screening ultrasound, (US). Life-threatening decompensations following ICH are unusual, and most reported "mortality

Table 1
Empiric data to consider before when considering palliative care for infants with intraparenchymal and/or grade III ICH.

IMPORTANT ECHOGRAPHIC AND CEREBRAL IMAGING	• Localisation of the hemorrhage • Extension of the hemorrhage • Bilaterality • Multilobar extension • Ventricular dilatation • Results of electroencephalogram
CLINICAL FACTORS ASSOCIATED WITH WORSE OUTCOMES	• Low gestationnal age, gender (chances or survival) • Postnatal sepsis • Postnatal steroids • Periventricular leukomalacia • Nectorizing enterocolitis/surgery • Failure to thrive

from ICH" is probably due to decisions to WLSI [21,22]. Death is less frequently associated with grade 3 ICH (between 10 and 30%) than IPH [14,15], presumably due to fewer decisions of WSLI. The proportion of death after diagnosis of IPH is as high as 76% in some countries [2,16], because WLSI differs according to culture and the ethical values of caregivers and parents [22–24].

When death is reported after ICH, whether WLSI was performed should be detailed, otherwise the studies are uninterpretable. A way to categorize mode of death has been described [25] which can help examine biases and ethical decision-making associated with WLSI decisions in units with different cultures, viewpoints or practices.

1.1.1.3.2. Cerebral palsy. We report in Table 2, the results of different cohorts which have studied associations with severe ICH and cerebral palsy (CP). A diagnosis of CP is based on a neurological examination, and the degree of functional impact is now usually described with the Gross Motor Functional Classification System (GMFCS). In industrialized countries, 2/3 of individuals with CP, of all grades combined, walk, 3/4 talk, and ½ have an IQ > 70 [26]. The impact of CP with GMFCS 1 or 2 on quality of life is questionable; infants are predicted to be ambulant, even though they may require therapy and assistance. A GMCFS score of 3 or more is often referred to as "disabling CP", defined as the need for assistive devices for mobility. However, most individuals with CP report a good quality of life [27], despite their disability. Indeed, many with CP or other disabling conditions articulate a social model of disability with the absence of adequate accommodations (such as access ramps) perpetuating challenges.

Grade 3 vs grade 4

Prognostication of CP based on the Papile grade is unclear but seems more frequent after grade 4 than 3. In the study of Brouwer et al., CP was less frequent after grade 3 IVH (7%) compared to IPH (50%) [28]. A French cohort study showed that 4% of preterm infants without ICH developed CP, compared to 19% of the survivors of grade 3 ICH and 45% of the small number of IPH survivors (n = 13) [29].

Location of IPH

Does the exact location of IPH predict cerebral palsy? [24] The "ELGAN" multicenter cohort suggested that anterior hyperdensity was more strongly associated with motor dysfunction than posterior lesions. In contrast, Rademaker et al. suggested that posterior lesions were more likely to lead to CP [30]. Roze found no association between location of IPH and the incidence of CP [31]. One study found that the involvement of the trigone in the IPH increased the frequency of CP among infants with IPH from 14 to 41% [27]. Dudink described that some locations (such as the area drained by complete terminal vein) had a higher frequency of contralateral hemiplegia, but the numbers were very small (n = 3 for some lesions) [32].

In summary, even though it might be considered obvious that IPH affecting motor tracts would lead to CP, the available literature does not confirm this, and there are no robust data that any specific location is higher risk.

Extent of IPH

A small number of studies have compared unilateral to bilateral IPH for their impact on CP. The ELGAN study showed bilateral IPH was much more likely to lead to CP than unilateral [33]. Maître et al. showed that 8/17 infants with bilateral IPH had CP with a GMFCS of 4 or 5, compared to 19/52 infants with unilateral lesions [19].

In one cohort >90% of survivors at 30 months corrected age (CA)

Table 2
Related incidence of death and cerebral palsy according to different studies in children with intracranial hemorrhage.

Study	N	Study type	Population	Cerebral palsy definition	Comparison/outcome studied	Outcome or death OR [IC95%]/%/(n[a])
Doyle, 2000 (Australia)	424	Cohort	VLBW 500–1499 g, born on two periods of 18 months each 1980 and 1992,	Loss motor function, abnormal muscle tone, and positive Babinski responses in affected legs	% In surviving children at 5 years with CP	IVH 3 = 31.2% (n = 5/16) IPH = 20% (n = 1/5)
Scherlock, 2005 (Australia)	298	Cohort	Preterm <1000 g, <28 weeks of GA born between 1991 and 1992	Loss motor function with abnormal muscle tone or reflexes CP moderate and severe ≥ walking with considerable difficulty	% In surviving children at 5 years with mild CP % Moderate or severe CP in surviving children at 5 years	IVH3 = 16.7% (2/12) IPH = 100% (n = 6/6) IVH3 = 8.3% (1/12) IPH = 83% (n = 5/6)
Brouwer, 2007 (Netherlands)	144	Retrospective cohort	Preterm ≤34 weeks of GA with IVH 3 and IPH	GMFCS≥1	Death and CP at 24 months of corrected age	IVH3 = 7% (n = 5/68) Mortality = 28% (n = 26/64) IPH = 49% (n = 37/76), Mortality = 37% (n = 44/120)
Roze, 2008 (Netherlands)	54	Retrospective cohort	Preterm <37 weeks of GA with IPH	GMFCS≥1	Death and CP	IPH = 66% (n = 25/38) Mortality = 30% (16/54)
Beaino, 2010 (France)	1812	Cohort (Epipage 1)	Preterm 22–32 weeks ofGA born in 1997	Involontary movement, loss of coordination or ≥ ½ abnormal posture or movement, increased tone or hyperreflexia	No IVH vs IPH/CP 18–24 month of corrected age	OR 29.66 [16.71–52.62]
Klebermass-Schrehof, 2013 (Austria)	471	Cohort	Preterm<32 weeks of GA, born between 1994 and 2005	Abnormal muscle tone in at least one extremity and abnormal control of movement or posture	% In surviving with 5 years CP	IVH3 = 63.6%[b] IPH = 90.9%[b]
Radic, 2015 (Novel Scottia)	1200	Retrospective cohort	20–30 weeks of GA, with IVH status available	CP moderate and severe ≥ walking with considerable difficulty	2–3 years moderate or severe CP	IPH = 12% (9/76)
Cizmeci, 2020 (Netherlands)	160	Retrospective cohort	Preterm with IPH ≤32 weeks of GA	GMCSF≥1	Death and CP at 24 months of corrected age	IPH = 42% (31/160), mortality 40% (64/160)

IPH, intraparenchymal hemorrhage; IVH3, grade 3 intraventricular hemorrhage.
[a] Number of infants with cerebral palsy or death/number of infants with a severe intracranial hemorrhage.
[b] Data about exact number of infants not precised.

who had a Bassan score >2 demonstrated gross motor disabilities [20]. Tsai et al. found that all infants who had an IPH involving 3 lobes had at least mild CP ("which slightly interferes with, but does not delay, motor milestones") diagnosed at 18–22 months [34]. Cizmeci et al. showed an association with higher Bassan scores and lower gross motor z-scores, but not with CP [21]. Studies using the Al-Abdi system have not specifically addressed CP [35,36].

Additional risk of post hemorrhagic ventricular dilatation

The ELGAN data showed that ventricular dilatation [33] further increased CP risk; of 42 children with IPH and dilatation, 26% had CP with a GMFCS of 2 or more. Adams-Chapman and others showed that CP followed 23% of grade 3 ICH without VP shunting, but 57% after grade 3 followed by VP shunt; IPH without a shunt was followed by a 37% incidence of CP, compared to 80% of those with IPH and shunt requirement.[14]This was confirmed in other studies [34].

1.1.1.3.3. Developmental, cognitive and sensory outcomes in early childhood. Most studies have relied on developmental testing at 18–24 months CA as the primary outcome for evaluating cognitive outcomes. Such testing has limited ability to predict longer term outcomes of importance, such as school readiness, or intelligence quotient (IQ) [37,38]. Bilateral grade 3 or 4 ICH in one cohort was associated with an increase in the proportion of survivors with impairment (either developmental delay or CP), but in both instances, the majority of survivors (>65%) were unimpaired or had mild NDI at 18–22 months CA [39]. One cohort [40] of infants with IPH showed that language and developmental outcomes (measured between 12 and 66 months CA) were not associated with severity, determined by the Bassan score. Tsai et al. [34] showed that 54% of infants with IPH had a developmental quotient in the lowest quartile (at 18–22 months) but showed no relationship with Bassan scores.

Two studies using the Al-Abdi score found that it was more predictive of long-term outcomes than the simple Papile system, both studies using "NDI" as their definition of adverse outcome [35,36]. The difficulties in prognostication from an US image are illustrated by the fact that one infant with the highest possible score of 35 (bilateral IPH affecting multiple areas in both hemispheres with midline shift) had only mild NDI.

Few studies have described feeding or ophthalmologic problems. Broitman reported in his prospective study 23% of infants were not feeding independently at 18–22 months CA [38]. Infants with IPH have almost 10% of ophthalmologic sequelae (visual acuity<6/60, smaller average visual field, and visual fixation difficulty). There is no clear association between visual impairment and occipital IPH [34].

1.1.1.3.4. Cognitive and intellectual outcomes at school age. The systematic review of Mukerji [37] found 12 studies evaluating the impact of severe ICH on longer term outcomes. The proportion of infants with IPH who had adverse outcomes varied according to the cohort and the tests used. For example, 50% had a K-ABC <70 [41], 100% (6/6) had an IQ less than 85 (average approximately 70), and 3/17 required special education.

Overall, the more severe the ICH (Grade 3 and 4 vs grade 1 and 2), the more pronounced the cognitive impacts at school age [42,43], but there is no clear difference between grade 3 and 4 ICH. Van den Bohr et al. showed that 25% of 14-year-old adolescents with all grades of ICH required specialized education compared with 10% in the general population [44] and that major disability (that "interfered seriously with everyday life and imposed a severe burden on the child, the caretakers, and society") was present at 5 years of age in 4/17 of infants with grade 3 or 4 ICH [44]. Luu et al. showed that 12 year olds with grade 3 or 4 ICH, ventricular dilatation, or PVL, had an IQ lower by about 20 points compared to full-term peers (they were not compared to preterm infants without adverse neurological outcomes) and that 32% of the former preterm infants with these lesions required special education [38]. Another study showed that 75% of 8 year olds with severe ICH had delayed numerical skills [45].

1.1.1.3.5. Clinical factors associated with worse outcome. In addition to the location and extent of the hemorrhage, other clinical factors such as sepsis and post-natal corticosteroids are very important in determining outcomes. The combination of both factors has more negative impact [18]. In several cohort studies, outcomes were much more closely related to clinical factors, and ICH, even severe ICH, added little or nothing to the prediction [46]. Most of such cohorts, however, have used a simplistic grading system, and have not evaluated the extent or laterality of the lesions.

1.1.1.4. The role of other neurological investigations in clarifying prognosis

1.1.1.4.1. Cranial ultrasound and magnetic resonance imaging. Cranial US (cUS) is a non-invasive, widely available tool, which is sensitive for centrally located brain injury, including ICH: for some lesions, such intraventricular bleeding, there is relatively good interobserver variability, however diagnosis of periventricular echodensities is much more variable [43]. Serial cUS can readily be used at the bedside for detection of ICH and the progression of ventricular dilatation [44]. Although Magnetic Resonance Imaging (MRI) gives more detailed images, new diagnoses of lesions not seen on cUS are usually minor findings of uncertain long term significance [47]. Currently there is no clear place for MRI in the acute phase of ICH to help clinicians refine prognosis to counsel parents.

1.1.1.4.2. Electrophysiology. Although most IPH are clinically silent, they are associated with an increased incidence of clinical seizures varying between 5 and 40% [48]. There is no large cohort study with routine electroencephalogram (EEG) findings on all infants having an IPH. In one small cohort study, 5 of the 11 infants with IPH had electrographic seizures [49]. Although the American Clinical Neurophysiology Society recommended long-term recording of EEG in all preterm infants with severe ICH [50], it is unclear if electrographic seizures affect their prognosis.

In summary, grade 3 ICH, even when bilateral, leads to an incidence of CP of less than 20%, most of which is GMFCS 1 or 2, and there is little clear data about cognitive outcomes. Unilateral IPH affecting 1 or 2 lobes has little effect on motor outcomes but is associated with lower developmental scores at 18–24 months corrected age. More extensive IPH leads to a higher incidence of CP, and when bilateral many survivors have severe CP, from 50 to 90%. Overall, IPH shifts developmental scores downwards by about 1½ SD; bilateral or more extensive IPH have similar cognitive impact. Later cognitive impacts of IPH, after 3 years of age, are much less well described, but show that most children are able to go to normal school, although many will need extra scholastic assistance. If severe ICH is followed by ventricular dilatation, CP is much more common, and schooling difficulties become more frequent especially if neurosurgical intervention is required [12]. The additional insult of late-onset sepsis, and bronchopulmonary dysplasia treated with steroids, increase the risks of both motor and cognitive problems.

The simplistic labelling of an ICH as "grade 4" or "IPH" is inadequate for life and death decisions; the extent and laterality of the hemorrhage should be noted prior to evaluation of prognosis. Even with knowledge of these factors, prediction of cognitive outcomes is very limited.

2. Ethical decision-making after a severe ICH

2.1. Variations of practice for end-of-life decisions after ICH

Throughout the world, there are wide variations of practice for end-of-life decisions in NICUs. Faced with an infant with an IPH the approach differs between countries [24]. In an ethnographic study comparing French and American units, Orfali highlighted that American clinicians reported that for them the worse risk was to cause a baby's death, when a meaningful life could have been lived [23]. In contrast, the worst risk French clinicians reported was "*to permit the survival of a severely*

impaired child" [23]. Clinicians should recognize these variations of practices, their biases. In recent decades, shared decision-making has been promoted. What does shared decision-making mean for end-of-life decisions after ICH? Does this approach require palliative care discussions regarding WLSI for all ICHs? More than 90% of units in Australia, NZ and Switzerland will offer redirection of care for infants with bilateral IPH, compared to less than 10% of units in Japan, Israel, and Tuscany [50]. The variation in survival for patients with IPH directly reflect difference in practices concerning WLSI. Some units may offer, while others may recommend WLSI. Facts will create values, which will feed facts and empirical data that is used to inform families. The culture and also the legislation of the country may govern these decisions.

Physicians are notoriously poor at prognostic evaluation. Brecht et al. showed that of 25 infants whose caregivers discussed limiting LSI following a unilateral IPH, 19 died (18 before discharge and 1 after). Of the 6 survivors: three (42%) were considered as functionally normal, one mildly disabled but independent, one significantly disabled [51]. A more recent study among extremely preterm infants noted that 58/529 survived after withdrawal of life-sustaining treatment with comfort care [52]. On these 58 infants, 9 died before discharge and the half of the 39 evaluated survivors had no or mild NDI at 24 months CA [52]. It is essential that neonatologists recognize that early individual prognostication, based on cranial ultrasound findings findings, is always uncertain. Communicating that uncertainty to parents is important, yet complex.

2.2. Parental values related to outcomes after severe ICH

2.2.1. Neurodevelopmental outcomes and parental perspectives

Epidemiologic studies evaluating the long-term outcome of preterm infants have usually investigated NDI, which is largely determined by scores (mild-moderate-severe) on developmental tests as well as a neuromotor examination. Developmental tests are relatively easy to administer, are well standardized, and are relatively objective, yet, they lack predictive ability [53]. The definitions of NDI (which are variable in the literature) have been the produc of clinicians' decisions. The pitfalls in using these definitions to inform families are two-fold: 1) the use of global statistics at the individual level and (2) the lack of integration of other important factors such as daily functioning of the child and the family, and the resilience of a family. In every study examining the perspective of clinicians in preterm infants' quality of life, clinicians were always more pessimistic than families or individuals born preterm. There are also major differences in views on disability between medical caregivers and families [54].

It will be important in the future to evaluate the parental perspective in follow-up studies, both in terms of the quality of life of their children, as well as coping strategies and resilience. These factors are interdependant. The parent's view of the quality of their children's lives is also driven by parental resilience.[55-57]Studies concerning the quality of life, or patient important outcomes of pre-schooled or schooled children born prematurely have emerged in recent years [55–58].

For Mina, what matters for the clinicians? In the American Journal of Bioethics, the story of Mina continues:

> "Through several conversations during the first week, within a shared decision-making approach, Helen and Peter were given support and information about the moral dilemmas and Mina's uncertain prognosis. They expressed ambiguity: they hoped for survival and a good life for her but were concerned about future disability. The doctors and nurses caring for her felt that further life support was dependent on parental wishes; all agreed that both continuing and withdrawing life support were justifiable decisions and strived to communicate that to the parents."

2.3. Ethical decision-making for Mina: when is it reasonable to offer palliative care?

Our review of the prognostic information for Mina, who is one week of age, who suffered a unilateral grade three ICH, leads us to conclude that the clinicians did not have enough elements to offer to withdrawal of LSI. From a strictly scientific point of view, Mina was born at 23 weeks and, as a survivor at 7 days, she had a 70% chance of survival [59]. She seems not to have other comorbidities (severe lung disease, NEC, infection), although these could occur later. The unilateral grade 3 IVH probably places Mina at a somewhat higher risk of NDI, most of which is due to an increase in developmental delay, and to an increased risk of CP (most likely GMFCS 1 or 2). The future is truly uncertain, but at this stage, in our opinion, in this context a decision to WLSI would be inappropriate. The duty of the physicians, in our view, is to inform the parents that although risks of "NDI" are increased, and that the future is uncertain, it is unlikely that Mina will have profound intellectual disability, and the ultrasound findings do not change that small risk substantially. On the other hand, Mina is young, and unfortunately many adverse events can happen. This information could change rapidly if ventricular dilatation occurs and/or Mina develops necrotizing enterocolitis and needs surgery. One also needs to realize that "the window of opportunity" is considered by some to be important [58]. Mina, being a preterm at 7 days, is fragile and needs considerable LSI to stay alive. The stronger she gets, the less support she needs, which make it harder to redirect care to comfort care. Although this phenomenon exists, deciding earlier, without adequate data, is ethically problematic.

We wonder, as published in several of our previous articles, why the preterm infant is treated so differently than the older child or adult? Is it because an extremely preterm baby on a respirator, at seven days of age, is still considered to have a different personhood, is not considered "a real person yet"? [60,61] Is it because we feel, as neonatal clinicians we "create" disability whereas, in other disciplines, they feel they save a sick patient? [62] Extreme preterm infants seem to be viewed as morally different than older sick patients [61]. In no other field of medicine do fragile patients, legally incompetent, with outcomes as good as those of Mina (70% survival with 50% of survivors having no disability, 15% severe disability) have suggestions that LSI be withheld or withdrawn. We doubt that for other patients with such outcomes, routine withdrawal of intensive care after a week of intensive care would ever be considered, and certainly not promoted in the pages of a bioethics journal. Other patients with worse outcomes are also not offered redirection of care in the NICU. For example, a baby born with trisomy 21 who (if slightly preterm at 36 weeks) needs continuous positive airway pressure (CPAP) for respiratory distress syndrome would not be considered for WLSI in a "window of opportunity." We consider this baby, with good reason, to have a good future quality of life, despite a predicted mean IQ of 40.2 in adolescence, and a high likelihood of not being able to live independently; such an outcome is clearly less optimal than Mina's likely outcomes.

2.4. When is it reasonable to offer palliative care for severe ICH?

While a unilateral grade 3 ICH in a stable preterm infant may not be grounds for redirection of care, some combinations of clinical factors and severe ICH will. Extensive bilateral ICH may occur in immature babies who become very unstable in their first days of life. For example, it is not rare for severe ICH to follow catastrophic pulmonary hemorrhages. In these cases, the babies are physiologically unstable, at times requiring maximal respiratory support, inotropic support, and steroids for respiratory failure. The extent of the ICH and additional clinical factors that predict both a high burden of care, mortality and poor outcomes may be grounds for offering and sometimes recommending withdrawing LSI. In all the cases, when babies are stable, clinicians and parents should know that decisions do not need to be made in an urgent fashion. A fragile child with an IPH affecting more than 2 lobes of the

brain, or bilateral, could often be observed for their evolution over, for example, 2 weeks. Some infants will remain stable and improve, others will develop dilatation that requires shunting, and perhaps in addition sepsis, which may prompt the team to discuss or recommend palliative care with the parents. Some children develop surgical NEC and, in these circumstances, it may be decided to withhold surgical interventions and redirect care.

3. Communication with parents

Historically, disability has been viewed from a biomedical lens, with a focus on what is not working or abnormal. In 2001, the World Health Organization shifted to the current International Classification of Functioning, Disability, and Health (ICF model), examining disability from the lens of function and participation. Expanding on this concept, Rosenbaum et al. described the F words as the focus for any child's development. In the context of communication with parents about disability and/or for decision-making regarding end-of-life decisions, we will discuss two frameworks: 1) The F word perspective to speak about disabilities and inform parents, aimed at speaking about function, not diagnoses [59] and 2) the SOBPIE (Situation, Opinions, Basic human interactions, Parents, Information, Emotions, see below) approach for WLSI decisions with families [63].

3.1. Function over diagnosis: the importance of F words

Speaking about disability to families, especially when so variable from one baby to another, is complex. Traditionally, the medical model identifies a diagnosis or condition and then offers a solution or cure (antibiotics, chemotherapy, surgery). For conditions that cannot be 'fixed', such as disability, this traditional medical model unravels, with only the identification of a 'severe neurological problem' or CP, and the associated limitations identified. What matters to parents, however, is how a child will function, independent of a child's diagnosis and prognosis: will a child be happy, go to school, have friends, be independent? The F words as described by Rosenbaum et al. refocuses the discussion of disability on those unifying, ubiquitous goals of parents for children: friends, family, fitness, future, fun, functioning (not the how, but the what people do) [59]. When recalibrated to these core goals, disability can be presented in the context of what parents' value. Clinicians who follow children long term (neonatal follow-up, rehabilitation medicine, physiotherapists, etc) could assist with these conversations with families. We should ensure that neonatal clinicians are as equipped to discuss function as they are diagnoses.

3.2. The SOBPIE approach

In the context of significant grade ICH, a SOBPIE approach could be used to guide the communication. The SOBPIE approach has been extensively described and will only be summarized here [63–65], practical recommendations are available in detail in the articles, for babies who are physiologically unstable, as well as those who are stable.

SOBPIE stand for the following: what is the **Situation?** Is the baby imminently dying? Should WH/WD LSI be considered? 2. **Opinions and options**: personal biases of clinicians and potential alternatives for patients 3. **Basic** human interactions 4. **Parents**: Their story, their concerns, their needs, their goals 5. **Information**: meeting parental informational needs, providing balanced information 6. **Emotions**: relational aspects of decision-making: emotions, social supports, coping with uncertainty, adaptation and resilience [63].

The first two letters do not involve communicating with parents but are prerequisites for ethical decision-making: the reflection of clinicians. These should always precede conversations with families. It is essential to remain humble and curious, to question those decisions, to think/speak about and analyze our biases, within units in an interdisciplinary fashion, as well as between units. Only then can we learn from each other and optimize our care.

3.2.1. Basic human interaction

Most clinicians believe that they interact with parents in an empathetic manner, but sometimes, this is not obvious to the families. Often, it is not *what* the doctor says, but *how* it is said [66]. This simple list of suggestions can lead to basic human interactions.

- The nurse taking care of the baby should be aware that a difficult conversation will occur with the parents and should be present.
- Do the parents want a significant support person present? Wait for that person if time permits.
- Introduce yourself to the parents.
- Limit the number of clinicians and trainees attending difficult conversations. For example, if discussing shunt placement in a sick baby, we would recommend that the baby's nurse be present and that each staff of interest (neonatology, neurosurgery, for example) chose one person to attend.
- Have difficult conversations in a place that is suitable for the parents.
- Make sure you don't get interrupted: ask a colleague to cover the urgent calls.
- Sit down for difficult conversations.
- Explain your role in the team caring for their baby and why you are there.
- Know and use the baby's name. A baby is not a "23-weeker" or "a difficult case of ICH".
- Tolerate silence.

These may seem infantilizing recommendations, but they are often not all followed [65].

3.2.2. Parents, information and emotions

We adcovate for personalized decision-making. A "one-size-fits-all" communication model focused on standardizing information does not lead to partnerships. It is possible to standardize personalized approaches and adapt to families [64].

Parents have different perspectives and informational needs. Clinicians have to be sensitive to the needs of each set of parents and not just transfer information that they think is important, in the format that they think is the most appealing. Parents whose preterm child suffers a severe ICH have generally been in the unit for several days. Ideally, in non-emergent end of life scenarios, principles of palliative care and relationships begin early in the trajectory of critical illness before acute deterioration. Clinicians caring for babies, even if they are two days old, should have a basic knowledge of the family structure.

Some parents want basic information, while others may prefer detailed statistics and medical data.

The following questions can help personalize communication:

"*What is your current understanding of the situation*?"

To understand how much information to provide, clinicians can use the "*some parents, other parents*" approach: "*Some parents want to know all the numbers, statistics and percentages while some want the big picture. What kind of parent are you*?" [19].

One of the most valued principles in decision-making is autonomy, or self-ruling. Parents in these situations may not feel autonomous. They may be in shock, feel many emotions, such as guilt. They may not want to decide alone for their child. The author Elizabeth Stone writes "*Making the decision to have a child – it is momentous. It is to decide forever to have your heart go walking around outside your body*". Many ethicists writing from a feminist viewpoint have pointed out that the autonomy principle is flawed [67]. Parents have relationships, other children, spouses, communities. Important decisions affect these relationships, and are thus rarely taken in a purely individualistic, autonomous fashion. We advocate for personalized decision-making as opposed to shared decision-making in these situations. Many parents do not want to

share these decisions and decide the date of death of their child. Some think God or nature will decide. Others may want to decide on their own. None of these situations represent shared decisions [63–65,68,69]. These sentences can help in these circumstances.

- Using the "some parents … other parents" approach can help uncover preferences with statements such as "*Some parents do not want to be the ones to make life-and-death decisions for their baby … Some want to decide with the medical team, and others want to be the ones to make the decision. How would you feel most comfortable approaching these decisions*?" and/or
- "*Some parents know in their gut what decision feels best, some want to use data to make decisions, and some do a little bit of both. Which approach seems best for you*?".

When parents want recommendations and ask, "*What would you do*?" or "*What would you do for your child*?", we should not offer choices neutrally and make them feel abandoned [70]. We can tell them what other parents decide, and why. Units should develop structured processes to offer recommendations, such as interdisciplinary meetings. These cases should routinely be discussed in a group fashion.

Other sentences can be useful, such as "*What concerns you the most*?" or "*What are your hopes for your child*?"

Answers to these questions will vary and may surprise us.

- "*I do not want her to die*";
- "*I don't think I can be the mom of a disabled child*"
- "*I do not want to have any regrets, to feel I have abandoned my child. I already abandoned her and could not stay pregnant, I want to be able to live with myself*";
- "*I hope she will have a good quality of life*";
- "*What will happen to my couple, my other children, my family*?"

Sometimes, one parent biggest concern will be "*that she will be handicapped*" while the other will answer "*that she will die*" simultaneously. These answers may help couples understand each other's perspectives. Some parents cannot imagine being the parents of a dead child; others, to be the parents of a disabled child. These serious concerns need to be addressed, as well as decreasing guilt of parents of preterm children. These discussions may be complex: what happens when children die, what happens to disabled children, how parents cope with adversity and the impact of resilience. When parents' most serious fears are addressed, it is less hard for them to engage in decision-making.

4. Conclusion

Long term outcomes after severe ICH are very variable and depend on multiple factors. The Papile grading system is very limited for prognosis; IPH which are more extensive and bilateral are more important for future motor function, especially if there is post-hemorrhagic ventricular dilatation. Cognitive functioning is even more difficult to predict, and although there are impacts, they are variable and often minor. Discussions regarding WLSI, however, often follow the diagnosis of a severe ICH. Diagnosis should be followed by an overall evaluation of the infant's status, an analysis of the number of brain regions affected and laterality, whether sepsis or other complications have occurred, and surveillance for PHVI. Following this assessment, the neonatologist should remain humble, recognizing the uncertainty of prognosis, and explore outcomes that are important to the parents. To facilitate communication with parents, the clinicians should speak about function rather than diagnosis, as well as what can be done to optimize function. The SOBPIE framework can also provide guidelines that can facilitate processes.

Parents and families will live with these experiences and decisions for the rest of their lives. How they remember the communication process and care their infants received depends on their perceptions of the relationships built with clinicians and their ability to rewrite their stories within the context of their values. Personalized communication is a process which gives as much attention to medical outcomes as to the process by which those outcomes are presented and ultimately supported. In the care of babies with a severe ICH, and at risk of death or disability, these processes are essential. Only by speaking about these decisions, while remaining humble and curious, will we improve our care to the most fragile children.

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