Three Decades after Baby Doe: How Neonatologists and Bioethicists Conceptualize The Best Interests Standard

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Abstract

Objective—Determine how neonatologists and bioethicists conceptualize and apply the Best Interests Standard (BIS).

Study Design—Members of the ASBH and the AAP Section on Neonatal-Perinatal Medicine were surveyed to determine how they conceptualized the BIS and ranked the appropriateness of forgoing life sustaining therapy.

Result—Neonatologists' median response supported an infant-specific BIS conceptualization that linked the infant's and family's interests. They did not support allowing limitations on the family's obligations. Ethicists' supported a conceptualization that linked the infant's and family's interests and limitations on the family's obligations, a less infant-specific conceptualization. Ethicists were less or equally likely to agree with forgoing LST in 7 of 8 cases.

Conclusion—Ethicists endorsed a conceptualization of the BIS that includes the effects on the family and rejected an infant-specific one. Neonatologists split between these two and rejected limiting the family's obligations. Critical appraisal of the BIS is needed in neonatal ethics.

Introduction

In the wake of the 1982 Baby Doe case and subsequent government interventions, Kopelman and colleagues reported the results of a survey of members of the American Academy of Pediatrics (AAP) Section on Neonatal-Perinatal Medicine on their views of the Baby Doe regulations and how the regulations affected their practice. They found that most
neonatologists thought that the regulations encouraged or required overtreatment of infants, and that they overwhelmingly wanted to consider the parent's wishes when making decisions regarding life sustaining treatment but that the regulations prevented them from doing so. Kopelman et al. concluded that the majority of neonatologists felt that the Baby Doe regulations forced them to practice in a manner contrary to infants' best interests.

While the Best Interests Standard (BIS) has remained the guiding principle of neonatal ethics during the intervening three decades since Baby Doe, competing interpretations have emerged.

Proponents of the vitalist tradition in ethics, whose views are reflected in the Baby Doe amendment to the Child Abuse Prevention and Treatment Act (CAPTA), support a narrow, infant-centric interpretation that views the BIS as being solely concerned with prolonging the infant's life. Meanwhile, the AAP has been inconsistent in its interpretation of the BIS. Ethicists, including the President's Commission on Ethics, endorsed a broader view, one in which the burden placed on caregivers could be considered when determining an infant's best interests.

Despite its widespread acceptance, there is no consensus on how the BIS should be conceptualized. One important legacy of Baby Doe is that physicians and ethicists are routinely involved in clinical ethics consultations, as well as in developing and implementing practice guidelines and organizational policies. Physicians and ethicists may share the nomenclature in neonatal ethics, but not a common concept of the BIS. Lack of conceptual consensus may result in inconsistent approaches to counseling families making life-altering and potentially irreversible decisions on whether to forego life-sustaining therapy (LST) on high-risk infants. This paper reports the results of the first national study to describe how neonatologists and bioethicists conceptualize the BIS and make judgments about setting limits on neonatal critical care. Our hypothesis was that the two groups do not share a common conceptualization of the BIS.

**Methods**

A survey was designed based on eight clinical scenarios common in the NICU, asking participants to use a 7-point Likert-type scale (from “Strongly Disagree” corresponding to “-3” to “Strongly Agree” corresponding to “3”, to rank the ethical appropriateness of foregoing LST at the parents' request. The scenarios ranged from a condition where death is imminent and inevitable, to one where the prospects for a meaningful and productive life are very good. Participants were then asked how they conceptualized the BIS in relation to the effects on the family. We modified the survey in response to feedback after piloting it with approximately 10 physicians and ported it online. Table 1 includes the case scenarios for each question. The study design and survey were approved by the Baylor College of Medicine Institutional Review Board.

An email with a link to the online survey was sent to members of the Section on Neonatal-Perinatal Medicine of the AAP (approximately 3200 members) and to the American Society of Bioethics and Humanities list serve (approximately 2000 members). After two and four weeks, a follow up email was sent to each group. The email contained informed consent...
language, and subjects were provided the opportunity to request a written copy of the consent form.

We computed the mean and median responses, as well as the interquartile ranges (IQR), standard (SD) deviations, and standard errors of the mean (SEM) for each of the clinical questions and conceptualization questions. The response distributions were analyzed via Mann-Whitney U test. To identify potentially confounding factors, we performed a chi-square analysis on the demographic variables. We did not evaluate differences in degree earned, work setting, or academic appointment, as we believe that these categories represent innate differences between the two groups, rather than confounding variables. For those variables that differed significantly between the two groups, we performed a Mann-Whitney U test or Kruskal-Wallis test as appropriate to determine if there was a significant difference in how the demographic groups in question answered the questions. The variable groups needed to differ on at least 2 of 11 questions to be considered confounding variables. The confounding variables were then included in a linear regression analysis. The AAP provided basic demographic data on the members of the Section on Neonatal-Perinatal Medicine from 2011, and this was compared to the neonatologist group via a chi-square analysis.

**Results**

We had a total of 666 responses from the neonatologist group and 242 from the ethicists. Table 2, which details the demographic information we collected and the data provided by the AAP, is available online as Supplementary Material. There were statistically significant differences between the groups in regards to age group, gender, marital status, having children, and religion (p ≤ 0.05 for all). There were no significant differences in mean age (p = 0.88), years in practice (p = 0.07), years of experience (p = 0.15), or religiosity (p = 0.79). In regards to gender and having children, the two groups only differed in their response to 1 of 11 questions (p ≤ 0.05), and so did not undergo further analysis. Analysis of age group, marital status and religion revealed differing response distributions on 3, 4, and 8 of 11 questions (p ≤ 0.05 for all), respectively. These confounding variables were controlled for in a linear regression analysis and had no significant effect on our results. The neonatologist group did not differ from the demographic data provided by the AAP with regards to gender (p = 0.09) but did so with regards to age distribution (p = 0.045). As there was no demographic data available from the ASBH, further analysis on whether the age of our respondents differed from the sample group was not done.

Figure 1 illustrates the responses to how the subjects conceptualized the BIS. Neonatologists agreed (median 1; IQR -0.5 to 2, ethicists median 1; IQR -2 to 2) that the BIS is exclusive of all effects or interests except those directly affecting the infant. Both groups agreed (neonatologists median 1; IQR -0.5 to 2, ethicists median 2; IQR 0 to 2) that the interests of the infant and family are inherently and inextricably linked, though the ethicists agreed more strongly and this conceptualization was their top choice. The neonatologists disagreed (median -1; IQR -2 to 1), while the ethicists were neutral (median 0; IQR -2 to 2) on whether the effects on the family or their interests may place limits on their obligations towards the infant.
In response to the questions on the appropriateness of forgoing LST, the neonatologists were more likely to agree with forgoing LST (median 1 to 3) than the ethicists (median 0 to 3) in 6 of 8 clinical scenarios (p ≤0.05), while the ethicists were more likely to agree with foregoing LST than the neonatologists only in the least severe case, question 8 (p ≤0.05; median -2; IQR -3 to -0.75 and -2; IQR -3 to -2 respectively), and finally for question 6, there was no difference in the median response between neonatologists and ethicists (-1; IQR -2 to 0 for each)(Figure 2).

Discussion

Our hypothesis was confirmed: in their conceptualization of the BIS, neonatologists and ethicists differ in small, but clinically and ethically significant ways. On average, the neonatologists split evenly between viewing the BIS as exclusive of all interests except those of the infant, and one in which the effects on the family must be considered. However, neonatologists rejected the idea that familial burdens may place limits on the family’s obligations to the infant. In contrast, the ethicists agreed most strongly with the view that a determination of an infant's Best Interests must account for familial effects. They were neutral on allowing familial burden to place a limit to their obligations to the infant, and rejected an interpretation of the BIS that focuses exclusively on the effects on the infant.

These results, three decades after Baby Doe, raise a number of significant ethical concerns. First, even though they share a common nomenclature, neonatologists and ethicists do not share a common conceptualization of the BIS.

A second concern is whether the Baby Doe regulations have had an effect on how the neonatologists interpret the BIS. The neonatologists in Kopelman et al.’s survey overwhelmingly agreed that the parents’ wishes would influence their decision (77% in one of the three cases presented and 87% in a second case) and disagreed that the parents’ wishes would not enter into their decision-making (86%), but 66% felt that the Baby Doe regulations required them to treat contrary to what the family believed was in the infant’s best interest.¹ Though our study is not designed to follow up on the Kopelman et al. study, some inferences can be made. Neonatologists in the Baby Doe era strongly believed that the parents’ wishes should be considered and that the Baby Doe regulations prevented them from doing so. In our study, over half the neonatologists rejected the idea that the obligation to their infant could be limited by the effects on the family and on their interests, while over half endorsed a BIS that excludes all effects or interests other than those directly affecting the infant. This suggests that, in the decades since the promulgation of the Baby Doe regulations, neonatologists now place a lesser importance on the role of the family. Has it come to pass that, in trying to comply with the Baby Doe regulations, many neonatologists have altered their understanding of the BIS, perhaps to avoid the cognitive dissonance of acting contrary to the best interests of the infant? Or did the regulations have their desired effect, despite having never been actively enforced, by decreasing the bias against infants with potential cognitive and/or physical disabilities?

We were surprised to find that ethicists were less likely than the neonatologists to agree with the ethical appropriateness of forgoing LST. At first glance, this may seem contradictory to
the results of the conceptualization question, a possibility that cannot be altogether ruled out. A closer examination of the questions however suggests another interpretation. Case scenarios 1 and 2 describe essentially medically futile cases (fulminant necrotizing enterocolitis and severe asphyxiating thoracic syndrome, respectively). Though their median responses are similar, the ethicists had lower mean responses in each case and a significant difference in the distribution of responses (more than 10% of ethicists disagreed with forgoing LST in scenario 2). While these conditions are clearly futile to physicians, we cannot know if this is obvious to ethicists. Perhaps the discrepancy may represent a disconnect between a conceptualization of the BIS and its application, or that clinical application of the BIS may be influenced by other moral concerns not measured here.

These data underscore the importance of organizational policies and practices that call for clarifying the range of views on the BIS in the counseling of parents regarding forgoing life sustaining treatment. Policies that are the intellectual progeny of solely one discipline are necessarily heirs to that discipline’s distinct viewpoint, which may not be clinically or ethically adequate. In light of the fact that neonatologists and ethicists conceptualize the BIS differently, a multidisciplinary approach should be used, based on explicitly incorporating input from both disciplines and possibly others such as social work, chaplaincy, or family advocates. The goal should be policies and practices that better reflect the varied challenges and obligations that parents who are faced with these terrible decisions must juggle. Such policies and practices should prevent the “whipsawing” that parents experience when presented with competing “best” choices by different members of the medical team.6

There are a number of important limitations of our data. Likert-type scales can introduce biases, including a social desirability bias or a central tendency bias in which responses cluster in the neutral range. We saw a different phenomenon altogether. The responses to the “futile” cases, questions 1 and 2, demonstrated a “clustering” effect along answer “3” or “Strongly Agree.” This made it difficult to interpret subtle differences between the two groups and to describe basic statistical results such as the percentile groups. Likert-type responses though ordinal, do not have a regular interval, and so should best be described using terms such as “median” and “percentile.” Therefore, while we chose a 7-point scale in an attempt to find subtle differences, nonetheless the median responses between the two groups were often similar, even though the distribution of responses was significantly different.

The potential for overlap between the two subject groups exists as well. Nearly one-quarter of the ethicists had a medical degree. This may have diminished differences between the two subject groups, especially if any of the physician-ethicists were neonatologists.

For each scenario we chose to include a disease process that in our opinion matched the description we provided. In piloting our survey, the respondents may have demonstrated preexisting biases against certain diseases, such as Trisomy 18 or spinal muscular atrophy, leading them to respond differently than if they had only the clinical description available. In response, we chose other conditions that, we believed, were less likely to elicit biased responses. However, we cannot know for certain if the disease examples we chose might
have elicited biased responses of their own, for example, cases 2 (asphyxiating thoracic syndrome) and 8 (Down Syndrome).

This subject area merits continued investigation. Our study did not address potential conceptual variation about the BIS on the part of parents. Information on how parents and other family members conceptualize the BIS, as well as other ethical principles, and about how they understand relevant medical terminology would provide valuable insight as to how families make end-of-life decisions. Furthermore, determining what other factors, if any, families use to make their decisions, and where they obtain their information would have profound implications for how neonatologists and ethicists should counsel parents.

Finally, the goal should be to identify clinically and ethically sound conceptualizations in order to perform a critical appraisal of the BIS. Achieving this goal is necessary if the BIS is to remain the guiding ethical principle of neonatal ethics. A similar critical appraisal of the BIS may also be needed for other, related areas of medical practice in which the interests of patients are inextricably bound up with the ability and willingness of family members to provide long-term care and support. For example, there are geriatric patients with advance forms of dementia whose preferences for clinical management are unknown. In such cases, the best interests standard of surrogate decision making guides family members.

Conclusions

Our study found that neonatologists and ethicists conceptualize the BIS in significantly different ways. Neonatologists split on whether the determination of a child's best interests should focus exclusively on the effect on the infant or whether those interests are inseparable from those of the family. Neonatologists reject the idea that the family's interests can act as an external restraint on their obligation towards the child. Ethicists most strongly support an interpretation that links the child's and family's interests; are neutral on whether the family's interests can act as a constraint on their obligation towards the child, and reject the idea that a determination of a child's best interests can exclude the effects on the family. However, in most clinical scenarios, the neonatologists were more likely to agree with the family's request to discontinue life sustaining treatment than the ethicists. These data raise serious concerns as to how ethicists and neonatologists make decisions regarding life-sustaining treatment, how they create and implement hospital and organizational policies, and ultimately how they counsel parents in regards to end-of-life decision making. This study raises implications for organizational policies and practices. This study also has supports the need to study parental views as an unknown variable in end of life decision making, because parents are presumed to be in authority over the end of life decisions for their infants and knowing how they conceptualize the BIS is critical to understanding how they make these decisions.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.
Acknowledgments
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Figure 1. Median Response and Interquartile Ranges to Conceptualization Question
Shown are the median and mean responses and interquartile ranges to the conceptualization question, “How do you conceptualize the Best Interests standard as it applies to end-of-life decision-making for infants?” The median response is represented by the black bar and the mean response by the white diamond. The colored bars represent the 50% interquartile range, while the error bars represent the 10% and 90%iles. The distribution of responses were analyzed via the Mann-Whitney U test. An asterisk (*) represents a p value of ≤0.05.
Figure 2. Median Response and Interquartile Ranges to Questions on Permissibility of Forgoing Life-Sustaining Therapy

Show are the median responses and interquartile ranges to the 8 clinical questions on the ethical permissibility of forgoing life-sustaining therapy at the family’s request. The median response is represented by a blue square for the neonatologists and a red triangle for the ethicists. The error bars represent the 25% and 75%iles. The distribution of responses were analyzed via the Mann-Whitney U test. An asterisk represents a p value ≤0.05.
Table 1

| Question | Case Description |
|----------|------------------|
| 1        | Where death is imminent and no longer preventable. Example: a premature infant with fulminant necrotizing enterocolitis who cannot be adequately oxygenated/ventilated despite maximum cardiorespiratory support. |
| 2        | Where death can be predicted in the near future (e.g. six months) with near certainty. Example: an infant with an asphyxiating thoracic syndrome on 100% FiO2. |
| 3        | Where death is likely, but not certain in the near future. Example: an infant with a severe variant of hypoplastic left-heart syndrome that is not surgically reparable. |
| 4        | Where overall mortality is high, but death is unlikely in the near future, and there is irreversible loss or absence of cognitive developmental capacity. Example: an infant with hydranencephaly. |
| 5        | Where overall mortality is high, but death is unlikely in the near future, and the infant has limited cognitive developmental capacity and faces a future of multiple medical procedures. Example: an infant with severe hypoxic-ischemic encephalopathy who is ventilator dependent, has a tracheostomy, a history of seizures, and is fed via gastrostomy tube. |
| 6        | Where death is unlikely in the near future and cognitive developmental capacity is intact, but the infant faces a future of multiple medical procedures. Example: an infant with prune belly syndrome who will require dialysis. |
| 7        | Where mortality is unknown, but some degree of neurodevelopmental impairment is certain, and the infant faces multiple medical procedures. If the infant reaches adulthood he/she will never be capable of basic activities of daily living. Example: an infant with semi-lober holoprosencephaly and severe gastroschisis. |
| 8        | Where mortality is unknown but some degree of neurodevelopmental impairment is certain and the infant faces multiple medical procedures. If the infant reaches adulthood, he or she will most likely be capable of basic activities of daily living. Example: an infant with Trisomy 21 and a correctable cyanotic heart lesion. |