

Establishing Goals of Care in Serious and Complex Pediatric Illness



Carrie M. Henderson, MD^a, Renee D. Boss, MD, MHS^{b,*}

KEYWORDS

• Goals of care • Pediatric complex care • Chronic critical illness • Palliative care

KEY POINTS

- Creating cohesive goals of care in complex pediatric illness may be threatened by a misaligned understanding of a family's values and desires.
- Cohesive goals of care require attention to team unity regarding prognosis, in-depth exploration of family context, and consistent communication.
- Blended goals of care may help families and clinicians align different viewpoints about the best course of action.

INTRODUCTION

An increasing number of children are living for months and years with enduring medical conditions that have substantial impact on their functioning and longevity. Several definitions are used to describe this population. The cohort of children with “medical complexity” are those with chronic health problems that result in functional limitations and intensive use of health services and resources.^{1,2} Children with “chronic critical illness” have repeated and prolonged hospitalizations and multiple chronic medical technologies.³ Children with “serious illness” have a high risk of death in childhood.⁴ Taken together, these definitions delineate patients with varying levels of long-term prognostic uncertainty, intensive interactions with medical systems, functional limitations, and often home medical technologies that shape the child's and family's quality of life. This article will use the shorthand “children with serious/complex illness” to denote this group of patients.

^a Department of Pediatrics, Center for Bioethics and Medical Humanities, University of Mississippi Medical Center, 2500 North State Street, Jackson, MS 39216, USA; ^b Department of Pediatrics, Johns Hopkins School of Medicine, Johns Hopkins Berman Institute of Bioethics, 200 North Wolfe Street, Suite 2019, Baltimore, MD 21287, USA

* Corresponding author. Department of Pediatrics, Johns Hopkins School of Medicine, Johns Hopkins Berman Institute of Bioethics, 200 North Wolfe Street, Suite 2019, Baltimore, MD 21287.

E-mail addresses: rboss1@jhmi.edu; Rboss1@jh.edu

Pediatr Clin N Am 71 (2024) 71–82
<https://doi.org/10.1016/j.pcl.2023.08.008>

pediatric.theclinics.com

0031-3955/24/© 2023 Elsevier Inc. All rights reserved.

Data suggest that such children account for at least one-third to one-half of patients in US general pediatric wards⁵ and neonatal (NICU)⁶ and pediatric intensive care units (PICU).⁷ They are also the primary utilizers of multidisciplinary pediatric outpatient services, including subspecialty providers, home nursing, occupational and physical therapies, and medical equipment providers.⁸ Some of these interactions with health care providers reflect decision points: *Should a new medication be added for a distressing symptom? Is a risky surgery warranted, given declining quality of life? Should home respiratory support be intensified? Or stopped? What are the options when a family's daily care needs outstrip their community supports? How do we transition a child to adult services? When is it the right time to consider advance directives?* For children with serious/complex illness, the answers to these decisions rarely derive from a strong evidence base and often depend heavily on the family's goals of care (and by "family" we include those with central roles in a child's life and decision-making, eg, parents, extended relatives, guardians, etc.).

In their systematic review, Secunda and colleagues⁹ offer an operational definition of goals of care that includes (1) what is hoped to be achieved overall for a child's care/condition, (2) grounding in both the immediate and big picture clinical course, (3) what is most important to the patient/family, and (4) a focus on how the decision at hand is impacted by these reflections on overall prognosis, clinical logistics, and values. The authors note that the essential benefit of setting intentional goals of care is to support families, to ensure that they have a role in decision-making, and so that patients do not receive less or more care than is desired.

Despite the importance of setting, communicating, and revisiting goals of care for children with serious/complex illness, multiple studies suggest that this process breaks down. Using a sample case for illustrative purposes, this article will explore common challenges to cohesive goals of care in complex pediatric illness: prognostic uncertainty, diffusion of medical responsibility, individual family context, and blended goals of care.

PROGNOSTIC UNCERTAINTY

Case: "Soraya Greene" had an uncomplicated infancy. At 18 months, she stopped gaining weight, coughed with feeds, said no words, and stopped walking. Subspecialty evaluation was delayed by transportation barriers and insurance gaps, so at 22 months Soraya was admitted for multidisciplinary work-up. During a 6 week hospitalization, her parents received the diagnosis: Soraya has a degenerative neurologic condition without cure. Among patients with this condition, the prognosis is variable: death in early childhood, slow progression with survival to young adulthood, and rare long-term survival are all possible. Progressive difficulty with feeding and breathing are likely.

The parents initially doubted the diagnosis and only intermittently attended subspecialty outpatient visits. When Soraya reached 3 years, the neurologist called the pediatrician with worries about disease progression. "I cannot be sure what her course will be," the neurologist said, "but if the family wants to prolong her life with medical interventions, we need to prepare for those decisions." The pediatrician meets with the parents to discuss goals of care and, if they are amenable, an advance directive. Soraya's parents say they want whatever it takes to give Soraya a "good quality of life" but don't want her to have "unnecessary suffering."

Clinical Questions

- How is "good quality of life" determined?
- How is "suffering" determined?

- What is intersection between pediatric goals of care and advance directives?

With advances in diagnostic and therapeutic tools, clinicians are increasingly able to put a name to a child's medical condition and to treat or at least ameliorate that condition. Yet prognostic uncertainty may only increase: *Will a novel treatment make the child's daily life better or worse? Will interventions prolong life? Will a plan of care allow a child to go home, or will they become dependent on inpatient care?* Some parents find prognostic uncertainty liberating: it allows hope that the child will exceed expectations. Families of infants recently diagnosed with serious/complex illness, for example, are more likely to opt for tracheostomy/home ventilation than are families of older children who have been living with similar conditions, because families of infants are more optimistic about clinical improvement.¹⁰ Other families find prognostic uncertainty extremely burdensome, as demonstrated by Aite and colleagues¹¹ who showed that lower-mortality fetal conditions with widely variable outcomes were more anxiety-provoking to pregnant women than higher-mortality fetal conditions with a predictable outcome. Clinicians also have different reactions to prognostic uncertainty, with risk of being overly pessimistic¹² or overly optimistic.¹³ In Soraya's case, the prognosis for her serious/complex illness is uncertain, but a shortened lifespan is likely. Her parents are already thinking about her quality of life and potential suffering.

"Quality of life" is a term that first showed up in the medical literature in the 1960s.¹⁴ A proliferation of quality-of-life definitions and measures have been proposed since then, typically including the domains of physical, mental, and social functioning. Quality of life is generally in the eye of the beholder, meaning individuals with similar states of functioning may rate their quality of life differently. An individual's perception of a "good" or "acceptable" quality of life may also expand as the person adjusts to evolving functional status.

Assessing quality of life in children of varying developmental stages and abilities is especially complicated. Children with serious/complex illness may not be able to directly report their experiences of comfort, contentment, pain, relationships, etc. Often we rely on family report, which may conflate child and family experience. For instance, a mother and father of a child with serious/complex illnesses may rate the child's quality of life differently based on the parent's own role in the child's daily care. Adding to the intricacy of gauging a child's quality of life are the multiple studies showing that pediatric health care providers tend to rate a child's quality of life lower than families do,^{15,16} especially for patients with neurologic impairment.¹⁷ Given these factors, clinicians should generally defer to family assessment of an acceptable quality of life for a child who cannot contribute to the discussion. For Soraya's family, the next questions the pediatrician could ask might be, "*What does a good quality of life look like for your daughter?*" and "*What would an unacceptable quality of life look like?*"

Like quality of life, "suffering" is a concept that is often central to decision-making for children with serious/complex illness, yet equally vexing to define and assess. Core elements of suffering are generally physical and/or mental pain and distress. As with quality of life, the assessment of a child's suffering can be hard to disentangle from the experience of family. A parent may feel intense distress when their child has seizures, for example, but it is hard to know what the child is experiencing. The concept of "unnecessary suffering" that Soraya's parents raise with their pediatrician suggests they may find some degree of suffering acceptable to attain a goal. Some families may accept their child's discomfort with a tracheostomy, for example, if it means the child can be home with and a part of the family. There is no clear-cut threshold at which a child's suffering is so severe that irrevocable acts, like withholding

or withdrawing life-sustaining therapies, should be considered. The risk for health care provider bias is real, when provider perceptions of a child's suffering might actually reflect personal values that the child's life is not worth living.¹⁸ There are notable cases where clinician assessment of a child's suffering led them to seek to override the parent's assessment,¹⁹ and ethical and/or legal consultation can be helpful in extreme cases. Absent such concerns, clinicians should defer to the family assessment of suffering when a child cannot contribute to the discussion. For Soraya's family, the clinician could ask "*What do you think would be some signs that would tell us that Soraya is suffering?*"

Soraya's parents convey that their goals of care include a "good quality of life" and avoiding "unnecessary suffering." The clinicians want to help them consider how these goals map onto decisions about medical care, including life-sustaining therapies. Nonurgent discussions of these topics, when a child is not in a medical crisis, may feel uncomfortable for clinicians who worry about frightening or distressing patients and families.²⁰ Multiple studies show low rates of pediatric advance directives, even among children with the most serious/chronic illnesses.²¹ A formal advance directive may be less critical for pediatric versus adult patients, since parents are usually the legal decision-makers and are usually present to make medical decisions. It is important to note the multiple studies showing that many parents are open to discussions about life-sustaining therapies, perhaps particularly when they are worried that the treating medical team won't really know their child.²⁰⁻²² For children with serious/complex illness who have multiple hospitalizations and dozens of involved clinicians, there is a real risk that the clinicians managing a medical crisis will not be fully aware of the child's complex history. Importantly for the pediatrician, families prefer having conversations about advance directives in the context of a longitudinal relationship.²³

In sum, intentional and timely conversations with families are the first step in establishing goals of care in situations with prognostic uncertainty. These conversations can build trust with families, even when the topics are emotionally difficult. Clinicians can guard against inserting their own values into these discussions by helping the family articulate their definitions of an acceptable and unacceptable quality of life.

DIFFUSION OF RESPONSIBILITY

Case, continued: Soraya presents to the emergency department 6 months later with an aspiration pneumonia and is intubated for acute respiratory failure. Her labs and imaging show evidence of chronic respiratory insufficiency and neurologic progression of her disease rendering her airway protection mechanisms dysfunctional. She has difficulty separating from the ventilator. During a long PICU stay, this new group of physicians directing her care recommend surgical interventions to mitigate ongoing risk of respiratory failure and acute illness: a gastric feeding tube (G tube), a tracheostomy (trach), and a ventilator for home. Pulmonary physicians are involved as well as the neurology team; prognostic uncertainty persists. The family has not shared with this new team the details of the recent goals of care discussion had with the primary pediatrician; the family is confused by the recommendations of the new team.

Clinical Questions

- How do acute care hospital processes impact the goals of care for children with serious/complex illness?
- What strategies exist for closing gaps in inpatient/outpatient care coordination?

Diffusion of responsibility in clinical care is a long-recognized phenomenon wherein organ- or disease-based teams operate in individual silos with little interteam

communication about a child's overall prognosis. Rooted in the psychological phenomenon of the bystander effect,²⁴ multiple stakeholders focus on their own disease or organ system, believing someone else is taking on the mantle of responsibility, ultimately leaving no one in charge.²⁵ In ICU systems, rotating critical care physicians may act as the "team leader" to coordinate with various subspecialists and team members, but this has its difficulties as well. ICU physicians are trained to react to acute problems, not chronic care coordination, and ICU training programs focus more on technical skills than communication. The moving parts of a busy ICU dictate frequent attending turnover with handoffs of information targeted to urgent problems. Large interdisciplinary teams with multiple learner levels try to coordinate the flow the information between all of the subspecialty teams and the family.²⁶

The limitations of these processes likely have the greatest impact on the growing group of children with serious/complex illness who often spend time in an ICU.²⁷ These patients require extensive care coordination, broadly defined as a family-centered, assessment-driven, and team-based activity that addresses the comprehensive needs of families.²⁸ In the context of large and siloed medical teams, care coordination between them is key to ensuring that interventions are both medically beneficial and in line with the family's goals. Without an intentional approach to team care coordination, families are left to filter and interpret conflicting information and to bridge communication gaps between care teams. In many institutions, this has led to the rise of inpatient/outpatient pediatric hospitalist and complex care teams to serve as the "quarterbacks" for children with serious/complex illness, filling the growing deficits between the pediatric health care system, and the historic surge in childhood morbidity and chronic disease.²⁹ Across the country, the growth of complex care teams has worked to promote care coordination and alleviate fragmentation and gaps, especially as the child goes from outpatient to inpatient and back home again.

Complex care teams may or may not serve as an outpatient medical home; when they do not, as in Soraya's case, the outpatient pediatrician may still be excluded from contributing to goals of care discussions in the hospital. Because inpatient medical care for children with serious/complex illness is often centralized to academic and/or large hospitals,⁷ families like Soraya's may rely heavily on local pediatricians. For Soraya, who is experiencing an acute-on-chronic decline, her pediatrician holds unique perspective about her life up to now, her family's wishes and home life context, and a more comprehensive view of her gradual decline. Families of children who require frequent hospitalization clearly value their primary pediatrician's input when they are hospitalized, stating it improves interdisciplinary coordination and aids in decision-making.³⁰ Invitations to planned care conferences or family meetings, weekly phone calls for updates or check-ins, and web-based face-to-face participation in rounds or meetings are ways to include a primary care provider in creating goals of care.

It should be noted that, even when a reasonable degree of care coordination exists for children with serious/complex illness, clinicians may still struggle with a team-based approach to medical decision-making. A survey of NICU and PICU providers about professional responsibility for consensus decision-making noted that most providers did not feel a responsibility to achieve consensus, although most stated they would try for consensus around a high-stakes intervention for a child.³¹ Most also agreed that conflicting recommendations about an intervention should be disclosed to a family. The literature reveals descriptions of a few team decision-making processes for children with serious/complex illness,³² but this is an area that must clearly expand to meet the needs of this growing population.

In sum, the majority of children with serious/complex illness are cared for by large numbers of clinicians in the inpatient and outpatient setting. When these clinicians act in silos rather than as a coordinated team, families can become confused by disparate information and recommendations. The goals of care for a child will become fragmented without intentional processes for team coordination.

INDIVIDUAL FAMILY CONTEXT

Case, continued: During her PICU admission, Soraya's parents consider how their social circumstances relate to the home medical technologies (g-tube, tracheostomy, home ventilator) the doctors are recommending. The parents have limited transportation and are stressed about living several hours away from any pediatric hospital. They worry about the impact of bringing medical technology and home nursing into their home, and how it might affect their other children. In their interactions with the clinical team, they want to appear to be agreeable to "do what Soraya needs" but privately they wonder if it would be wrong to not pursue home medical technology and allow Soraya to die naturally.

Clinical Questions

- How does individual family context impact medical decisions for children with serious/complex illness?
- What strategies exist for preventing bias when considering family context?

Family context relevant to a child's well-being includes many elements of home and community: housing adequacy, transportation, financial vulnerability, parent physical and mental health, sibling needs, degree of extended family supports, access to community resources, childcare options, school resources, local respite, etc. For children with serious/complex illness, medical needs often place growing stressors on family context. A stressed family context, in turn, will have impact on a child's medical care and medical outcomes.³³

Family life can be profoundly impacted by pediatric home medical technology.^{34,35} Home ventilation via a tracheostomy, along with feeding per surgical gastric tube, requires some of the most complex home care because 24/7 assessment, monitoring, and intervention is needed. Historically, mechanical ventilation was only provided in an ICU setting but advances in equipment development and tracheostomy care enable children to receive this technology in a home setting. Parents become responsible for a level of care otherwise delivered by highly trained clinicians in a hospital; the rate of adverse events with home ventilation is significant.^{36,37} Despite the training that families get to manage medical equipment, recent data suggest families feel unprepared for the enormous effect of home ventilation on their day-to-day lives. They report significant stress on their relationships, employment, financial security, and personal well-being—most families report these factors were not emphasized during the decision-making process about home medical technology.^{34,38} The impact of home ventilation also shapes the child's life in a variety of ways: limitations to travel and mobility, hindering some aspects of development, continued infections or complications, and isolation due to medical fragility.³⁹ Most families who choose home ventilation report that they would make the same decision again, but also admit to significant challenges and burdens that were not discussed or anticipated.^{34,40}

Establishing goals of care regarding treatments or interventions with broad impact on home life should intentionally explore social realities and expectations.³² This might include discussing that one parent may need to stop working, or the family might need

to buy a different vehicle, or may have limited home nursing, or may even want to relocate to a community with more resources. All of these factors could impact family stability, which impacts home care, which impacts child health outcomes.

Fairness and justice are important considerations: only addressing family context for some families of children with serious/complex illness risks bias. A standardized approach to including family context in serious decision-making is one way to protect against that bias. Palliative care engagement, with their philosophy and orientation toward family-centered decision-making, can be helpful. Palliative care providers also bring skills in considering alternatives to life-sustaining treatments and interventions, as Soraya's family wonders what it might look like if they decline the physicians' recommendation for medical technology. Ethics consultants may also be helpful in weighing risks and benefits of pediatric interventions with respect to a family's unique context. Families should be encouraged to reach out to other experienced parents, perhaps via online groups, to learn more about what life might be like if they choose, or decline, a particular medical intervention for their child.^{41,42}

In sum, family context is an important consideration in medical decisions with broad impact on home life. When family context is not explored, we risk leaving families unprepared, undermining family stability and child medical outcomes. Systematic, skilled evaluations can reduce bias in determining how family context maps onto medical decisions.

BLENDING GOALS OF CARE

Case, continued: Soraya's parents feel she is still fighting and are not ready to fully shift to end-of-life care. However, they have decided that home medical technology is not right for Soraya or their family. The family wants to take Soraya home on her medicines and nasal cannula, and to continue feeding her by mouth, understanding this approach will leave Soraya susceptible to aspiration and lung infections. The inpatient provider teams worry Soraya will have poor nutrition, increased infections, and hypoxia. Her outpatient pediatrician has concerns about accepting professional liability for Soraya's well-being once she is home, given unclear parameters for growth and vital signs. The family is offered home hospice but is not ready to enroll. They would like to avoid, though have not ruled out, further hospitalizations as Soraya's health declines.

Clinical Questions

- What are blended goals of care?
- How can clinicians support families who have blended goals of care?

Pediatric medical decision-making is often based in the best interest standard wherein clinicians and families come together to create plans of care that best serve the child's health interests. Clinicians typically establish the scientific guardrails defining which treatment options are entirely beneficial to the child and therefore "obligatory" (eg, the parents cannot be allowed to refuse), and which treatment options are entirely without benefit and therefore "unacceptable" (eg, the parents cannot be allowed to elect them). These scientific guardrails delineating benefit and harm are generally based on clinical evidence regarding pediatric populations without serious/complex illness. Substantial ambiguity characterizes which treatment options might help or harm a child with a progressive illness and limited lifespan. If Soraya goes home with a nasal cannula and oral feeding, she may die sooner, but the time she has can be focused on comfort and family. If she receives a tracheostomy and g-tube, she might live longer (though not necessarily), but the extra time she has might

be lived in the hospital, away from her family. Neither option is without potential benefit or harm, so it is difficult to consider either option “obligatory.”

Some ethicists advocate for the “zone of parental discretion,” suggesting that parents have ultimate decisional authority about the plan of care for their child even when clinicians feel the parents’ plan is suboptimal.^{43,44} Many families of children with serious/complex illness reject singular goals of care, opting for some blend of life prolongation and life enrichment. “Blended” goals of care often translate into a trial of, or limited use of, therapies with potential to reverse or slow a child’s decline, with a priority for withholding/withdrawing those therapies that reduce the child’s quality of life.⁴⁵ The process of forming blended goals of care varies with individual circumstances. Blended goals may be the result of a compromise when families seek one treatment path but clinicians recommend another. Blended goals may be derived for one specific treatment scenario, for example, a child’s acute pneumonia, or they may describe the global approach to that child’s care. Blended goals may limit one very burdensome intervention even as they escalate multiple, less burdensome interventions. Finally, blended goals may evolve by default, for example, simply because some treatments are tried and others are not, or they may be intentional goals set to direct a child’s care. Blended goals are often a progressive series of compromises that aim to match family’s values regarding their child’s care. Blended goals avoid the “do everything versus do nothing” dichotomy and respect parents’ wisdom regarding their child’s experiences of their medical care.⁴⁶

Clinician–family disagreement about Soraya’s care likely reflects different perspectives regarding the desired outcome for Soraya. The clinicians are prioritizing best practice standards with a goal of sustained periods of health and longevity of life. The parents appear to be situated in the gray zone between palliative and curative intent. Their blended goals to orally feed Soraya and go home with nasal cannula, while accepting future hospitalizations for antibiotics and supportive care, create unease among inpatient and outpatient clinicians who wonder about their medical, legal, and ethical responsibilities to the child. Hospice engagement is one way to align clinician–family goals and create an outpatient, individualized treatment plan that is family-centric and medico-legally straightforward. This option is increasingly available and flexible with the rise in insurance coverage for concurrent care, for example, simultaneous curative and hospice services.⁴⁷

In situations where home hospice is not involved, inpatient and outpatient clinicians must decide how to accommodate blended goals of care for children with serious/complex illness. Palliative care teams may offer support, given their expertise in managing children of all ages and diagnoses who are approaching end of life. They may be able to make recommendations regarding what vital signs parameters and nutrition goals could bridge the gap between what is achievable at home and what will optimize the child’s well-being. Clear documentation about why nonstandard clinical parameters are being followed, and what those parameters are, will promote consistency between team members. Clinicians should also clearly communicate to families, and document, any potential risk of blended goals. In Soraya’s case, if clinicians accept the parents’ goal of minimal home respiratory support (nasal cannula), the parents should know that low oxygen can injure Soraya’s brain and body, making future hospital interventions less likely to work. Finally, where clinicians cannot come to agreement about blended care goals, ethics consultation is recommended.

Although most decision-making models have a clear beginning and end, decisions for children with serious/complex illness resemble an evolving process without discrete boundaries.⁴⁸ Many families of children with serious/complex illness do not have dichotomized care goals of prolonging life versus comfort care. Instead, families

often have iterative goals that are blended, that is, some medical interventions to extend life but not at the expense of quality of life. Clinicians can engage palliative care, hospice, and ethics resources to help them support families in developing blended goals of care.

SUMMARY

An increasing number of children are living for months and years with serious/complex illness that includes long-term prognostic uncertainty, intensive interactions with medical systems, functional limitations, and often chronic medical technologies that shape the child's and family's quality of life. These families often face many medical decision points and are supported by intentional and iterative discussions about goals of care. Threats to cohesive goals of care in complex pediatric illness include prognostic uncertainty, diffusion of medical responsibility, individual family context, and blended goals of care. This article offers strategies for addressing each of these challenges—in all cases success involves recognizing how standard care approaches need adaptations to meet the needs of children with serious/complex illness.

CLINICS CARE POINTS

- Creating cohesive goals of care in complex pediatric illness may be threatened by a misaligned understanding of a family's values and desires.
- Mitigating barriers to cohesive goals of care requires attention to team unity around prognosis and outcome(s) and in-depth exploration of family context, in addition to clear and consistent communication.
- Blended goals of care may be a way to meet some expectations, but not all, for families and clinicians alike who may have different viewpoints about the best course of action.

DISCLOSURE

None.

REFERENCES

1. Kuo DZ, Houtrow AJ. Council On Children With D. Recognition and Management of Medical Complexity. *Pediatrics* 2016;138(6). <https://doi.org/10.1542/peds.2016-3021>.
2. Cohen E, Kuo DZ, Agrawal R, et al. Children with medical complexity: an emerging population for clinical and research initiatives. *Pediatrics* 2011; 127(3):529–38.
3. Shapiro MC, Henderson CM, Hutton N, et al. Defining Pediatric Chronic Critical Illness for Clinical Care, Research, and Policy. *Hosp Pediatr* 2017;7(4):236–44.
4. Guttmann K, Kelley A, Weintraub A, et al. Defining Neonatal Serious Illness. *J Palliat Med* 2022;25(11):1655–60.
5. Rogozinski L, Young A, Grybauskas C, et al. Point Prevalence of Children Hospitalized With Chronic Critical Illness in the General Inpatient Units. *Hosp Pediatr* 2019;9(7):545–9.
6. Boss RD, Henderson CM, Weiss EM, et al. The Changing Landscape in Pediatric Hospitals: A Multicenter Study of How Pediatric Chronic Critical Illness Impacts NICU Throughput. *Am J Perinatol* 2020. <https://doi.org/10.1055/s-0040-1718572>.

7. Shapiro MC, Boss RD, Donohue PK, et al. A snapshot of chronic critical illness in pediatric intensive care units. *J Pediatr Intensive Care* 2021.
8. Berry JG, Goodman DM, Coller RJ, et al. Association of Home Respiratory Equipment and Supply Use with Health Care Resource Utilization in Children. *J Pediatr* 2019;207:169–175 e2.
9. Secunda K, Wirpsa MJ, Neely KJ, et al. Use and Meaning of "Goals of Care" in the Healthcare Literature: a Systematic Review and Qualitative Discourse Analysis. *J Gen Intern Med* 2020;35(5):1559–66.
10. Boss RD, Henderson CM, Raisanen JC, et al. Family Experiences Deciding For and Against Pediatric Home Ventilation. *J Pediatr* 2021;229:223–31.
11. Aite L, Zaccara A, Trucchi A, et al. When uncertainty generates more anxiety than severity: the prenatal experience with cystic adenomatoid malformation of the lung. *J Perinat Med* 2009;37(5):539–42.
12. Bogetz JF, Munjapara V, Henderson CM, et al. Home mechanical ventilation for children with severe neurological impairment: Parents' perspectives on clinician counselling. *Dev Med Child Neurol* 2022. <https://doi.org/10.1111/dmcn.15151>.
13. Boss RD, Lemmon ME, Arnold RM, et al. Communicating prognosis with parents of critically ill infants: direct observation of clinician behaviors. *J Perinatol* 2017; 37(11):1224–9.
14. Elkinton JR. Medicine and the quality of life. *Ann Intern Med* 1966;64(3):711–4.
15. Payot A. Best interest standards do not correlate with the reality of physicians' decision making in life and death choices. *Evid Based Nurs* 2012;15(1):9.
16. Saigal S, Stoskopf BL, Feeny D, et al. Differences in preferences for neonatal outcomes among health care professionals, parents, and adolescents. *JAMA* 1999; 281(21):1991–7.
17. Bogetz JF, Boss RD, Lemmon ME. Communicating With Families of Children With Severe Neurological Impairment. *J Pain Symptom Manage* 2022;63(2):e264–6.
18. Tate T. Pediatric Suffering and the Burden of Proof. *Pediatrics* 2020;146(Suppl 1): S70–4.
19. Miller KE, Coleman RD, Eisenberg L, et al. Unilateral Withdrawal of Life-sustaining Therapy in a Severely Impaired Child. *Pediatrics* 2018;142(5). <https://doi.org/10.1542/peds.2018-0131>.
20. Boss RD, Hutton N, Griffin PL, et al. Novel legislation for pediatric advance directives: surveys and focus groups capture parent and clinician perspectives. *Palliat Med* 2015;29(4):346–53.
21. Liberman DB, Pham PK, Nager AL. Pediatric advance directives: parents' knowledge, experience, and preferences. *Pediatrics* 2014;134(2):e436–43.
22. Wharton RH, Levine KR, Buka S, et al. Advance care planning for children with special health care needs: a survey of parental attitudes. *Pediatrics* 1996;97(5): 682–7.
23. Lotz JD, Daxer M, Jox RJ, et al. "Hope for the best, prepare for the worst": A qualitative interview study on parents' needs and fears in pediatric advance care planning. *Palliat Med* 2017;31(8):764–71.
24. Fischer P, Krueger JI, Greitemeyer T, et al. The bystander-effect: a meta-analytic review on bystander intervention in dangerous and non-dangerous emergencies. *Psychol Bull* 2011;137(4):517–37.
25. Stavert RR, Lott JP. The bystander effect in medical care. *N Engl J Med* 2013; 368(1):8–9.
26. Marcus KL, Henderson CM, Boss RD. Chronic Critical Illness in Infants and Children: A Speculative Synthesis on Adapting ICU Care to Meet the Needs of Long-Stay Patients. *Pediatr Crit Care Med* 2016;17(8):743–52.

27. Kuo DZ, Cohen E, Agrawal R, et al. A national profile of caregiver challenges among more medically complex children with special health care needs. *Arch Pediatr Adolesc Med* 2011;165(11):1020–6.
28. Kuo DZ, McAllister JW, Rossignol L, et al. Care Coordination for Children With Medical Complexity: Whose Care Is It, Anyway? *Pediatrics* 2018;141(Suppl 3):S224–32.
29. Cohen E, Berry JG, Sanders L, et al. Status Complexicus? The Emergence of Pediatric Complex Care. *Pediatrics* 2018;141(Suppl 3):S202–11.
30. Rieger EY, Kushner JNS, Sriram V, et al. Primary care physician involvement during hospitalisation: a qualitative analysis of perspectives from frequently hospitalised patients. *BMJ Open* 2021;11(12). e053784.
31. Shapiro MC, Donohue PK, Kudchadkar SR, et al. Professional Responsibility, Consensus, and Conflict: A Survey of Physician Decisions for the Chronically Critically Ill in Neonatal and Pediatric Intensive Care Units. *Pediatr Crit Care Med* 2017;18(9):e415–22.
32. Henderson CM, Wilfond BS, Boss RD. Bringing Social Context Into the Conversation About Pediatric Long-term Ventilation. *Hosp Pediatr* 2018. <https://doi.org/10.1542/hpeds.2016-0168>.
33. Stein RE, Siegel MJ, Bauman LJ. Double jeopardy: what social risk adds to biomedical risk in understanding child health and health care utilization. *Acad Pediatr* 2010;10(3):165–71.
34. Henderson CM, Raisanen JC, Shipman KJ, et al. Life with pediatric home ventilation: Expectations versus experience. *Pediatr Pulmonol* 2021;56(10):3366–73.
35. Carnevale FA, Alexander E, Davis M, et al. Daily living with distress and enrichment: the moral experience of families with ventilator-assisted children at home. *Pediatrics* 2006;117(1):e48–60.
36. Ong T, Liu CC, Elder L, et al. The Trach Safe Initiative: A Quality Improvement Initiative to Reduce Mortality among Pediatric Tracheostomy Patients. *Otolaryngol Head Neck Surg* 2020;163(2):221–31.
37. Edwards JD, Kun SS, Keens TG. Outcomes and causes of death in children on home mechanical ventilation via tracheostomy: an institutional and literature review. *J Pediatr* 2010;157(6):955–959 e2.
38. Brehaut JC, Garner RE, Miller AR, et al. Changes over time in the health of caregivers of children with health problems: growth-curve findings from a 10-year Canadian population-based study. *Am J Public Health* 2011;101(12):2308–16.
39. Wright-Sexton LA, Compretta CE, Blackshear C, et al. Isolation in Parents and Providers of Children With Chronic Critical Illness. *Pediatr Crit Care Med* 2020;21(8):e530–7.
40. Hebert LM, Watson AC, Madrigal V, et al. Discussing Benefits and Risks of Tracheostomy: What Physicians Actually Say. *Pediatr Crit Care Med* 2017;18(12):e592–7.
41. Available at: www.Family-Reflections.com. Accessed April 7, 2023.
42. Available at: www.CourageousParentsNetwork.com. Accessed April 7, 2023.
43. Caruso Brown AE, Ciurria J. "Prix Fixe" or "A La Carte"? Pediatric Decision Making When the Goals of Care Lie in the Zone of Parental Discretion. *J Clin Ethics* 2021;32(4):299–306.
44. McDougall R, Gillam L, Spriggs M, et al. The zone of parental discretion and the complexity of paediatrics: A response to Alderson. *Clin Ethics* 2018;13(4):172–4.
45. Ullrich C, Morrison RS. Pediatric palliative care research comes of age: what we stand to learn from children with life-threatening illness. *J Palliat Med* 2013;16(4):334–6.

46. McGraw SA, Truog RD, Solomon MZ, et al. "I was able to still be her mom"—parenting at end of life in the pediatric intensive care unit. *Pediatr Crit Care Med* 2012;13(6):e350–6.
47. Lindley LC, Cozad MJ, Mack JW, et al. Effectiveness of Pediatric Concurrent Hospice Care to Improve Continuity of Care. *Am J Hosp Palliat Care* 2022;39(10):1129–36.
48. Lipstein EA, Brinkman WB, Britto MT. What is known about parents' treatment decisions? A narrative review of pediatric decision making. *Med Decis Making* 2012;32(2):246–58.