Neuroethics (2023) 16:17
https://doi.org/10.1007/s12152-023-09524-3

ORIGINAL PAPER

Hope and Optimism in Pediatric Deep Brain Stimulation: Key Stakeholder Perspectives

Natalie Dorfman · Lilly Snellman · Ynez Kerley · Kristin Kostick-Quenet · Gabriel Lazaro-Munoz · Eric A. Storch · Jennifer Blumenthal-Barby

Received: 22 November 2022 / Accepted: 29 June 2023 / Published online: 1 August 2023

Abstract

Introduction Deep brain stimulation (DBS) is utilized to treat pediatric refractory dystonia and its use in pediatric patients is expected to grow. One important question concerns the impact of hope and unrealistic optimism on decision-making, especially in "last resort" intervention scenarios such as DBS for refractory conditions.

Objective This study examined stakeholder experiences and perspectives on hope and unrealistic optimism in the context of decision-making about DBS for childhood dystonia and provides insights for clinicians seeking to implement effective communication strategies.

Materials and Methods Semi-structured interviews with clinicians ($n = 29$) and caregivers ($n = 44$) were conducted, transcribed, and coded.

Results Using thematic content analysis, four major themes from clinician interviews and five major themes from caregiver interviews related to hopes and expectations were identified. Clinicians expressed concerns about caregiver false hopes (86%, 25/29) and desperation (68.9%, 20/29) in light of DBS being a last resort. As a result, 68.9% of clinicians (20/29) expressed that they intentionally tried to lower caregiver expectations about DBS outcomes. Clinicians also expressed concern that, on the flip side, unrealistic pessimism drives away some patients who might otherwise benefit from DBS (34.5%, 10/29). Caregivers viewed DBS as the last option that they had to try (61.3%, 27/44), and 73% of caregivers (32/44) viewed themselves as having high hopes but reasonable expectations. Fewer than half (43%, 19/44) expressed that they struggled setting outcome expectations due to the uncertainty of DBS, and 50% of post-DBS caregivers (14/28) expressed some negative feelings post treatment due to unmet expectations. 43% of caregivers (19/44) had experiences with clinicians who tried to set low expectations about the potential benefits of DBS.

N. Dorfman (✉) · K. Kostick-Quenet · E. A. Storch · J. Blumenthal-Barby
Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, TX, USA
e-mail: natalie.dorfman@bcm.edu; njkd@uw.edu

K. Kostick-Quenet
e-mail: Kristin.kostick@bcm.edu

E. A. Storch
e-mail: eric.storch@bcm.edu

J. Blumenthal-Barby
e-mail: Jennifer.blumenthal-barby@bcm.edu

L. Snellman
Baylor College of Medicine, Houston, TX, USA
e-mail: lilly.snellman@bcm.edu

Y. Kerley
Rive University, Houston, TX, USA
e-mail: yhk1@rice.edu

G. Lazaro-Munoz
Harvard Medical School, Boston, MA, USA
e-mail: glazaro@hms.harvard.edu

Conclusion Thoughtful clinician-stakeholder discussion is needed to ensure realistic outcome expectations.

Keywords Dystonia · Deep brain stimulation (DBS) · Pediatric · Hope · Unrealistic optimism · Expectations

Introduction

Deep brain stimulation (DBS) is utilized to treat movement disorders, including pediatric refractory dystonia under an FDA Humanitarian Device Exemption [1, 2]. The growing use of DBS in children raises several important ethical questions and concerns [3, 4]. The role that hopes and expectations play in decision-making is a key ethical issue. Do caregiver decision makers have realistic expectations? Does hope inspire or cloud good decision-making in this context? Do clinicians' and caregivers' views on these matters differ? How ought clinicians respond to high hopes in the clinical setting without quelling them or permitting "false hopes" that might compromise informed decision making? A better understanding of these issues is vital for understanding informed decision-making process about deep brain stimulation for children.

When a patient hopes for a certain outcome, they desire it, believe the desired outcome is possible (probability greater than zero), and tend to engage in certain behaviors such as praying for the outcome, thinking about it or fantasizing about it—even planning for it [5]. Unrealistic optimism, on the other hand, occurs when a patient has a desire for a certain outcome and *overestimates the probability* of the desired outcome [5–9]. Similarly, denial occurs when a patient has a desire for an outcome and fails to think about or "face" the high probability of the undesired outcome [5, 7, 10, 11]. Self-deception occurs when a patient has a desire for a certain outcome and actively lies to herself or tells herself a narrative about the probability which is in tension with actual evidence but in line with the desired outcome [5, 10, 12]. There is also the phenomenon of "false" hope, which occurs when a patient has a desire for a certain outcome and believes it is possible when in fact it is not possible. Related concepts include "unrealistic hope" and "self-deceived hope" [5]. Although these latter terms include the word "hope," their defining essence is unrealistic beliefs. One important thing to note about these definitions is an implicit distinction between cases of hope vs. unrealistic optimism/denial/self-deception. In the case of hope, although a person may respond to unfavorable probabilities in very positive ways, she still possesses an accurate understanding of them [5, 10]. In cases of unrealistic optimism/denial/self-deception, on the other hand, there is something awry with the person's beliefs about the probabilities [5, 13].

Understanding the existence and impact of hope and unrealistic optimism in caregivers is integral to analyzing clinical interactions and enabling shared decision-making. Understanding the ranges of unrealistic hopes as well as their impacts on clinical decision-making can better inform clinician-patient interactions.

Methods

Participants and Procedures

Semi-structured interviews were conducted with clinicians (n = 29) working in DBS programs treating children and adolescents with dystonia as well as caregivers (n = 44) to understand stakeholder perspectives on informed consent and decision making about pediatric DBS. An interview guide was developed based on issues raised in the clinical and ethics literature and through discussions with DBS and movement disorders experts. A draft of the guide was reviewed by an advisory team and piloted with a neurologist specializing in movement disorders, resulting in minor changes for improved clarity. The study was approved by the Institutional Review Board at Baylor College of Medicine. Interviews were conducted via phone or Zoom, lasting an average of 50 min.

Data Analysis

Interviews were audio-recorded, transcribed verbatim, and analyzed using MAXQDA 2018 qualitative data analysis software. Team members (led by KK-Q, a qualitative methods expert and medical anthropologist) developed a codebook to identify thematic patterns in clinicians' and patients' responses to questions about hope and unrealistic

Table 1 Clinician characteristics

Clinical Specialty ($n = 29$)	% (n)
Pediatric Neurologist	31 (9)
Neurosurgeon	24 (7)
Neurologist	21 (6)
Pediatric Neurosurgeon	7 (2)
Pediatric Nurse Practitioner / DBS Programmer	7 (2)
Nurse Practitioner / DBS Programmer	3 (1)
Physician Assistant	3 (1)
Child Life Specialist	3 (1)
Years specialized in pediatric movement disorders ($n = 23$)	
Less than 5 years	4 (1)
5–10 years	48 (11)
10–15 years	17 (4)
15–20 years	17 (4)
20–25 years	13 (3)
Country ($n = 29$)	
United States	86 (25)
United Kingdom, Canada, France	17 (4)

Table 2 Caregiver Characteristics

Prospective caregivers ($n = 16$)	36.36%
Post-DBS caregivers ($n = 28$)	63.63%

optimism [14]. Team members (KK-Q, JBB, LS, and YK) all coded 5 transcripts together to ensure inter-coder reliability in application of the codebook and then LS and YK independently coded each interview transcript. With oversight from KK-Q and JBB, LS and YK then iteratively abstracted the coded information and identified thematic patterns across interviews utilizing thematic content analysis. ND calculated final theme and sub-theme frequencies and was responsible for primary data analysis.

Results

A total of 56 clinicians working with pediatric patients with dystonia specializing in pediatrics or having substantial experience with pediatric patients were contacted, and 29 participated (response rate = 52%) (Table 1). A total of 44 caregivers of children receiving DBS for dystonia were contacted (Table 2). We first present the thematic findings from clinician interviews followed by those from caregiver interviews.

Clinician Views on Hope and Unrealistic Optimism in Pediatric Deep Brain Stimulation

Clinicians said that because DBS for pediatric dystonia is a relatively new, experimental treatment with variable impacts on dystonia symptoms in pediatric patients, they worry about the potential for unrealistic hopes to impact caregiver decision making, as described below.

Concerns about Unrealistic Expectations and False Hopes

The most common unrealistic hope that clinicians reported seeing in caregivers was that DBS will be a *"cure,"* with a third of all clinicians interviewed explicitly mentioning that many caregivers hold this unrealistic hope. One clinician explained that some caregivers believe that DBS will completely heal their child and enable them to do things that they were unable to previously. Some parents go so far as to say that they believe their child will be "reborn" through DBS (see Tables 3 and 4).

A majority (86%, 25/29) of clinicians expressed concern that caregivers may have false hopes that may lead caregivers to overestimate the potential benefits and therefore *inadequately assess the risk/benefit ratio*. One clinician explained that realistic expectations are important for meaningful engagement in the informed consent process. However, some (21%, 6/29) clinicians noted that the rise in access to videos and testimonials on social media websites depicting significant improvements for other dystonia pediatric patients has increased false hopes among some caregivers.[1] One clinician explained that caregivers see videos that depict patients with significant improvements, which leads them to believe that DBS will

---

[1] Our findings on social media impacting levels of hope are similar to those in other studies, which have found that DBS can increase levels of unrealistic hope among patients and caregivers[15, 16] but patients feel that utilizing social media can allow them to more fully participate in healthcare conversations. [17.]

Table 3 Clinician views on hope and unrealistic optimism in DBS

Unrealistic expectations and false hopes		
Physician	004	People have the feeling that it's going to be this dramatic, quote unquote miraculous cure, like, they're going to get, it has got to… make everything better and perfectly back to normal. They're never going to have to take medicine again. I know, they're going to walk away from all their medical problems
Physician	007	I think that they think that it's going to get better right away as soon as you turn it on. That they're going to be cured. That everything is going to be better as soon as you turn on the DBS. That they're not going to take any medications afterwards. That … I mean, those are the major ones
Physician	011	[The caregivers] see these stories of people on the internet, they see these miraculous videos, they see this awesome stuff that's out there and they say, ah, here's an answer for my child and this is going to solve everything. And so having realistic expectations specifically in the pediatric deep brain stimulation for dystonia population, both patients and family
Physician	014	Maybe the biggest misconception is that it can be a total cure, because people have seen the DY21 videos
Physician	017	So, I think some patients believe that it's a cure, that you do this and then you're free of the disease forever. So, we try to establish realistic expectations that it can improve dystonia, but never eliminate it. So, it's not a cure for the disease, it's a management of the symptoms
Physician	019	So the misconception that deep brain stimulation is a cure for the dystonia, for example… there's sometimes a misconception that DBS is like other operations where you come in, you have an operation, and you go home and it's over
Physician	022	Far and away, the kid is going to get out of the wheelchair and there's been… they're rebirthed, they're reborn. I would call it the reborn expectation. They're going to do this and they're going to be reborn
Physician	024	A lot of patients will go on YouTube and search DBS. And what they often end up seeing is adults with Parkinson's disease where they're shaking and then they turn it on, and then moments later they stop shaking. And so, then they assume DBS is going to do the exact same thing for the dystonia
Physician	026	And obviously these families are desperate to get any help. I mean they hear about DBS, and read about DBS, they think DBS cures their kids dystonia, and obviously they pressurize us. But we have to make sure that we select these patients very, very carefully because we know DBS is unlikely or less likely to be very effective in children with secondary dystonia. So that is the problem
Last resort options and desperation		
Physician	002	I had a mom begging for a DBS on a kid who was violent and knocking people around… And we were thinking of it as rage behavior or something like that, I was thinking of DBS for it. Well, she latched on that as the hope. And that was one of the dumbest things I've done is to entertain that openly to her. Because that's a desperate human being. And she's not thinking… And I can't have a rational conversation anymore.
Physician	002	I had a mom begging for a DBS on a kid who was violent and knocking people around… And we were thinking of it as rage behavior or something like that, I was thinking of DBS for it. Well, she latched on that as the hope. And that was one of the dumbest things I've done is to entertain that openly to her. Because that's a desperate human being. And she's not thinking… And I can't have a rational conversation anymore
Physician	008	We've just touched on which is that these families are desperate, they've got a very sick child, the child's been tried on numerous medications. The DBS in most instances is sort of a Hail Mary if the child's not DYT1. So no matter what you say in the office in the back of their mind is this is going to be the miracle. This is going to be the Hollywood movie. So, it's not so much as a misconception it's just human nature that you believe in the miracle and that this is going to be the thing and it's going to completely change the trajectory of your child's life

Table 3 (continued)

Physician	017	At that point, they are medically refractory and we know there's no further medication that's going to help them, including things like Botox or Baclofen or other anti-spasmodic agents. At that point, we think the next line for many of these patients, assuming they have the overall general medical health to undergo it, would be something like deep brain stimulation
Physician	019	Think about the monumental experience that this represents for a kid to go through brain surgery and then another operation to put in a battery and then to have all this stuff implanted in their body and then with high expectations end up like not that much better. That can I think be real psychological blow that could do more harm than good
The practice of setting low expectations to combat unrealistic optimism		
Physician	003	it's really just mitigating that conversation about, "I really can't promise the extent of benefits." I mean it is a risky procedure, there's a lot of implications to the procedure and so how to balance the risk benefit conversation. That to me seems to be the biggest struggle in my mind
Physician	008	So, I explain even to the other DYT1s and say, "Look these are the results we've been able to get. I can't promise you this is what's going to happen."
Physician	008	Managing expectations is very important and being honest with people and then when you got into it with the families with the children with secondary dystonia you got to be very, very clear with them that this is a completely different entity and that the results are very, very mixed and might be just modest
Physician	011	But here's a case where I cannot, with any certainty, say what we can expect the benefit to be. So, we have to be very open to saying that there's a possibility here that there will be no benefit after we've gone through this whole process, and they have to be very comfortable with that odds
Physician	014	I usually underestimate the likelihood of success and overestimate the rate of complications just because I think people don't interpret statistics in a consistent way. And, so, when you tell a person a number like 85% chance of success, they hear 100% because 85 is closer to 100. So, I'll usually use a number like 50% because I just want people to make a decision… I want people to be comfortable with the possibility that it really wouldn't work
Physician	019	But the predicted benefit is the side of the equation that I worry more about in these kids and the potential psychiatric impact, psychological impact of what I would consider unsuccessful DBS. Think about the monumental experience that this represents for a kid to go through brain surgery and then another operation to put in a battery and then to have all this stuff implanted in their body and then with high expectations end up like not that much better. That can I think be real psychological blow that could do more harm than good
Physician	027	So, we try to downplay the outcomes as best we can because we don't really even know how well the patients will do, what sort of benefit they'll achieve after the surgery
Physician	027	The second is understanding that we really don't understand a lot about this disease process. And so, our expectations are not necessarily born in the final results. So, we try to downplay the outcomes as best we can because we don't really even know how well the patients will do, what sort of benefit they'll achieve after the surgery
Physician	029	I say, "You need to be okay with, in your mind, if this never works, okay? I want you to have that mindset going into this; that if this doesn't work, yes, you'll be disappointed but you're going to be okay with having made that decision because that is a possibility. And I set expectations very low, and I say, 'This is what we're hoping to improve. Everything else is gravy. So, this is our goal'
Physician	029	I think they need to be aware that there's a possibility that it's not going to work. That's the biggest thing for most cases. They need to be prepared that there could be complications and if it gets infected, which is the most common complication, then the device is going to have to come out. Then, yes, you can put it back in, but just be mentally prepared for that possibility. They need to be mindful that this is going to indicate multiple surgeries over their lifetime

Table 3 (continued)

Unrealistic pessimism		
Physician	004	I think before they're fully informed about kind of like what it actually is, they're nervous about big brain surgery, and that's sort of complications they imagine like the bleeding and stroke that could go with big brain surgery, but those are actually very low risk in general
Physician	013	I've seen some families have negative misconceptions about DBS where they think it is very invasive, it's very dangerous, it's very risky, which is of course not the case
Physician	016	I think the biggest misconception is that they're subjecting their child to a highly risky procedure that doesn't have the upside that they appreciate
Physician	016	What I worry about is that if they have a history of depression, or maybe even suicidal ideation or something like that, you worry if you don't meet their expectation, that it could be worsened. Because they're putting themselves through a big risk to get the surgery. And if their expectations aren't met, a lot of times their psychological profile could put them at risk, for example
Physician	018	You know there's potential for that if the kids or the parents have the misconception that the device makes them very delicate, and they have to be very careful of the device. So, I always make sure to tell them that, while pieces of the hardware and wires can break, it's almost never from some trauma or something that they did
Physician	018	It was very, very difficult and painful episode to have parents really misunderstand us as people who were providing bad care to their daughter, when in fact the truth was that their daughter had a very bad condition, and many clinicians were really trying to help her. And the fact that the parents could not perceive that, and they just thought actually she had a simple condition and all of you incompetent physicians are just not able to handle it
Physician	019	If anybody tells them anything that I can fix your kid, they'll do it, right. And so, they're desperate…The psychological blow of the disappointment of it not solving all their problems like they expect
Physician	029	In some cases where you got a slam-dunk case like a DYT1, and the family is reluctant or doesn't… and you just want to shake them and be like, "Yes, I understand brain surgery is scary. You have to do this for your child," and having to watch them suffer, sometimes for years before the family comes around and agrees to it
Physician	016	What I worry about is that if they have a history of depression, or maybe even suicidal ideation or something like that, you worry if you don't meet their expectation, that it could be worsened. Because they're putting themselves through a big risk to get the surgery. And if their expectations aren't met, a lot of times their psychological profile could put them at risk, for example

solve all the patient's problems (see Tables 3 and 4). Another clinician said caregivers can also be swayed by results from DBS for adults with Parkinson's, explaining that the caregivers who view these videos assume that DBS will have the same outcomes for dystonia patients.

Another unrealistic expectation that clinicians cited is that caregivers believed DBS will be *a simple procedure* rather than acknowledging the complexity of the treatment process. Multiple clinicians expressed that many caregivers believe that DBS is a single operation procedure, and neglect to factor in the fact that DBS will require multiple programming appointments. If caregivers are under the impression that their child will only need one surgery or be miraculously cured by DBS, they may overlook the potential complications and number of surgeries their child will require. Further, one clinician explained that caregivers "need to be prepared that there can be complications and if it gets infected, which is the most common complication, then the device is going to come out… they need to be mindful that this is going to indicate multiple surgeries over their lifetime" (C_Dy_029).

Concerns about Last Resort Options and Desperation

Many (68.9%, 20/29) clinicians pointed out that most pediatric candidates for DBS have refractory conditions and their caregivers and patients are considering DBS as a last resort. Clinicians note that most patients who enter the DBS program have exhausted all other known clinical options. As one clinician explained, dystonia patients who are eligible for DBS are very sick, so caregivers view DBS as a "Hail Mary" (see Tables 3 and 4). The clinician then explained that this can lead to unrealistic optimism, explaining that it is difficult to dissuade caregivers from holding this view.

Patients and families may also arrive frustrated with the lack of progress in their clinical care to date. One clinician explained that the frustration that caregivers feel can lead to desperation and limit adaptive decision-making on the part of the caregivers. Desperate parents can beg clinicians for DBS and can get angry if the clinician believes that DBS would not be helpful (see Tables 3 and 4). In these scenarios, clinicians feel that caregivers are irrational because their evaluations of DBS are colored by emotion over reason.

In deciding treatment course, clinicians try to evaluate how desperate caregivers are (see Tables 3 and 4). Clinicians said that in some cases this desperation can lead to a deterioration of clinician-parent trust and communication. One clinician recounted a situation wherein the caregivers overestimated the potential benefits of DBS and began to believe that the physicians were not providing good care for their child.

Another concern about this being a last resort option is that caregivers and patients may be *disappointed* if their unrealistic hopes remain unmet. One clinician explained that some caregivers are so desperate that there is a significant psychological and emotional impact to poor DBS outcomes. Another clinician stated that this psychological impact may be even worse for patients than for caregivers (see Tables 3 and 4).

The Practice of Setting Low Expectations to Combat Unrealistic Optimism

To combat unrealistic optimism and decrease unrealistic expectations, 68.9% (20/29) of clinicians expressed that they intentionally try to lower parent expectations about DBS outcomes (the practice of "laying crepe" as we expand on in the discussion). Clinicians emphasized the importance of managing expectations. One clinician stated that clinicians have to be very clear that DBS results are mixed and uncertain (see Tables 3 and 4).

In order to help clinicians set good expectations, one clinician expressed that they would significantly underestimate the chances of success so that clinicians would engage in good decision-making (see Tables 3 and 4). Another clinician agreed that most DBS clinicians are harsh in their estimation of potential outcomes. The clinician explained that when the family is engaged in the decision-making process, the clinician sets low goals for treatment outcomes (see Tables 3 and 4). Clinicians also expressed that downplaying potential outcomes was important because there is little clinical information about how patients will do post-DBS. One clinician explained that because there is a lack of information about potential treatment outcomes, caregivers should not be setting high or low expectations.

Some Caregivers are Too Pessimistic, Overweigh Risks, and do not Have Enough Hope

A smaller percentage of clinicians (34.5%, 10/29) expressed the opposite concern that some *caregivers* may be too pessimistic about DBS and over-estimate potential risks, which may dissuade patients and families from proceeding. Clinicians expressed that caregivers have misconceptions about DBS and perceive it to be overly dangerous and risky without significant benefits (see Tables 3 and 4).

Many clinicians noted that there were significant consequences of undue pessimism—it could prevent patients from accessing treatment. One clinician gave the example of a case wherein a patient was a perfect candidate for DBS but never got DBS as the caregivers perceived surgery to be too dangerous. Another clinician expressed frustration, stating that there are some cases wherein the child is a perfect candidate for DBS, but the caregivers are too scared to consent to brain surgery for a young patient (see Tables 3 and 4). The clinician elaborated that the patient will sometimes suffer for years before caregivers will consent to surgery. These families are so focused on risks, and neglect to consider the potential benefits of DBS.

Caregivers' Views on Hope and Unrealistic Optimism in Pediatric Deep Brain Stimulation

Realistic Hopes and Expectations for DBS

A majority of caregivers (73%, 32/44) reported that *contrary to clinician concerns* they had realistic hopes about DBS. The most common hopes expressed by caregivers included a better quality of life for the child (45%n 20/44), for the child to have [some of] their "life back" (32%, 14/44), for the child

Table 4 Caregiver views on hope and unrealistic optimism in DBS

Realistic hopes and expectations for DBS		
Post-DBS Caregiver	014	Yeah. I mean, our hopes were high, but really realistic expectations were pretty low
Post-DBS Caregiver	014	I mean, our expectations were pretty low, because when you're looking at a kid with his significant challenges, even the smallest amount of change, that one little thing can make a big difference. So we weren't going for the big stuff. We were really hoping for just really small changes. And like I said, I mean, we were super happy he was sleeping. That alone was amazing
Post-DBS Caregiver	014	I mean, we actually, our expectations were fairly low. I wasn't like, "Oh, he's going to be walking." We were just hoping, "Wow, if he could even get control over one hand enough to drive a chair. If he could get enough head control, maybe he can sit up." I mean, our expectations were pretty low, because when you're looking at a kid with his significant challenges, even the smallest amount of change, that one little thing can make a big difference
Post-DBS Caregiver	015	When you have DBS, you set goals to what you want to achieve yet after the operation… So we didn't set very high goals… Because we wanted to build her confidence and we knew, because there was a chance of zero success, we didn't want to be too built-up
Post-DBS Caregiver	024	So, I truly didn't have expectations. I mean, I dream, I dream big, but also know that a lot of my dreams aren't realistic. Like I said, even just seeing how when he turned her on and off, and the difference it made in her tone was worth it to me
Post-DBS Caregiver	026	And so, I don't think anybody was setting specific goals for exactly the kind of outcome we were hoping for. I think after discussing with them my expectations, my sort of good case scenario was it helped to take the edge off his dystonic movements and give him more control. That seemed like a really realistic thing to hope for. And of course, hope for lots more, but that's sort of the realistic place to be. They seemed like that was a realistic hope. Again, not a guarantee
Last resort options and desperation		
Post-DBS Caregiver	003	It was torture. She was falling all the time. She'd walk, she'd fall. I was getting emergency calls all the time. She couldn't function. She really wanted it. It wasn't like I was contemplating this for days and everything
Post-DBS Caregiver	003	By the time it rolled around where [the clinician] suggested so one day I said, "I don't what to do because she can't write, she can't go to school. What are we going to do?" And then he said, "Have the surgery." I will never forget that moment that he said, "Have the surgery." He said, "If you have the surgery, then you'll never need to see me again."
Post-DBS Caregiver	004	I still think this is the last thing you go. I think you get to the last thing fast. It was very obvious to us that there was no other solution. Nothing was working. He was deteriorating faster and faster and faster. We could see it happening. I think that's really important
Post-DBS Caregiver	008	I mean, so we were kind of desperate to give him as much independence as we could, so it was, like, whatever we could do at that point
Post-DBS Caregiver	010	As a dad, I'm thinking, "Will she ever walk again?" Right? She was in a wheelchair, she couldn't walk anymore. She couldn't use her right hand anymore. So, I was looking for something that might give her the quality of life back. And when we finally started talking to doctors about the DBS that was the only option to give her a hope of quality of life?
Post-DBS Caregiver	012	To be fair, I think at that point, the progression was so fast and scary and awful that whatever they needed to do, I was sort of like, "Just do it." I think we thought it was going to be a miracle and it was on so many levels, like so many levels. It was miracle, but it wasn't like the miracle. And I think that I should have probably asked more questions about, "What if it doesn't work as well as you're saying it will." Maybe I should have asked about that

Table 4 (continued)

Post-DBS Caregiver	018	I mean, for [PATIENT], it wasn't worth living a life like that. And it may be the same for a lot of people with depression or OCD if it is that bad in their life that they've got to give something a last shot. And that's where I was. I said, "Our backs are up against the wall. We've got to try something."
Post-DBS Caregiver	020	They [the clinicians] were strongly saying this is the last option that we got and we weren't really thinking about side effects
Post-DBS Caregiver	020	My dad was a big part of it and [the clinicians] explained everything to him. They were saying that, "We cannot guarantee what's going to happen. This is our last option, you have to try that out." And that's how we decided to do it
Post-DBS Caregiver	023	I honestly didn't know if it would work, nothing else had worked. They had given him medicine. He was doing Botox injections in his neck, and he was in so much that after a month he'd be begging for more Botox injections. So, it was just very hard to watch your child in a state like that
Expectation setting can be difficult		
Prospective DBS Caregiver	003	I worry a little bit about, and maybe this is back to the medical piece, but putting her through something like that and then it having zero effect. That would bum me out. I like having the knowledge that it might not work, but I want to think that it will help a little bit, otherwise, why would I be putting my kid who can't talk or can't make these big decisions for herself through something so major?
Post-DBS Caregiver	007	I think that's just so hard to tell because every kid is different, and there's millions of settings
Post-DBS Caregiver	009	I wish they would have done the genetic paneling. Because knowing that DBS does a little better with other types of dystonias. I would have liked to rather have known, okay, do we have that kind of dystonia, or is this going to… Are we just shooting in the dark?
Post-DBS Caregiver	011	There's a child in the Boston area that had DBS prior to the diagnosis, and he was very young, but he was so severe they felt that the needed to do it. It didn't appear that it helped at all, so I mean, I guess that is the thing, is some of these more rare conditions, they really don't know if they're going to help. To put your child through that amount of surgery and have it not help is always a risk
Post-DBS Caregiver	012	I also think just being careful about miracle stories, because you do hear them and they are out there and I am so jealous of all of the miracles, but there are a lot of like… This is a really serious illness. It's a really serious disability and although DBS is great, I wish I would've known that it could not be a miracle
Post-DBS Caregiver	018	I met a lot of caregivers in hospital that they were at the beginning of their journey in even understanding their child had a disability and so if they were offered this magic surgery, I think that their expectations could be also set too high
Post-DBS Caregiver	020	It wasn't guaranteed either. They were testing it out to see if it worked. They couldn't 100% say, it would work or not. But we wanted to give it a try because there was nothing else to do at that time
Unmet expectations and disappointment		
Post-DBS Caregiver	008	I always tried to explain that it probably wouldn't. That we were hopeful, but I think he thought it was going to work no matter what. And it has. Like I said, he's seen more than we have, but I think he was expecting more than what he's gotten so far
Post-DBS Caregiver	015	Because back that feelings of, "Yes we had it done, we tried it, it's been safe, it's not been as successful as we hoped, but we wouldn't regret having it done." That type of thing
Post-DBS Caregiver	016	But [family member] expectations were like he would start walking. Or has he started running. It's quite a challenge to explain them, but I think in their hearts, they know. But their minds refuse to accept the reality. But they're being supportive. We say that he's done something at school. It's nice that they support that ways, but I think they're not completely 100%

Table 4 (continued)

Post-DBS Caregiver	021	I think you need to hope that the patient understands what's going on and it doesn't make his or her worry or anxiety or depression worse if it doesn't work. Are they more despondent because now they tried this potentially miraculous thing and it didn't work and now they're feeling worse?
Post-DBS Caregiver	024	So, I mean, just putting a lot of our faith in him and him talking us through how much further she could go with that than Botox, and how it would affect her whole body and not just her legs. So, if I was to say that I wasn't disappointed every time we left a programming appointment, I would be lying, because I really just wanted them to turn it on and have her be like, "Hey, mom. How are you?" And that didn't happen
Post-DBS Caregiver	026	There were no behavioral issues per se, but that was surprising to me and a little bit concerning initially, not knowing if that was just cognitive fatigue from all this new input. And actually, that was even before he was turned on, just post-surgical, and lastly, lingered a lot longer than I expected. That was surprising
Benefits of hope		
Post-DBS Caregiver	012	There's a couple, the lady who's sent me your link, she has a teenager and they were one of the first people I talked to. We actually use the same doctor and her daughter now runs marathons and stuff. So that was my first, [NAME 2]was my first toe in the water with DBS and I actually talked to her and her husband on the phone when [PATIENT] was first sick and I thought, "Okay, well this is perfect. Sold." Right? [NAME 2]was, I mean, bless them… It was the hope I needed to move forward, but, and she didn't know that you can go that way either. But you can see where it gets a little bit hairy
Post-DBS Caregiver	019	For me, as I said before, what really helped was seeing visually other people who'd had similar dystonia to [patient]'s and being able to see that, to see what it was like before, to see what it was like after, because it did… It gave hope. It was like actually, we could imagine [patient] walking again, because he was in a wheelchair
Experiences with clinicians setting low expectations		
Prospective DBS Caregiver	003	I think it's important to know that there aren't guarantees because I don't think I would want to go into this being told, "This is the thing you do to fix GNA01," or "When your kid wakes up and we turn this on, it's going to be better." As shitty as it is to hear that it might not work, I would rather go into something with full knowledge of potential outcomes
Post-DBS Caregiver	003	When we went to the neurosurgeon and the neurosurgeon said to us, he said, "I make no guarantees. Sometimes it works, sometimes it's different."
Post-DBS Caregiver	013	Well, [the clinician] started with, "This is not a hundred percent sure that it's going to work. It might help a little bit. It's worth trying because of her health." But I was like, "Yes, let's do it. If that's the only thing we can try, let's struck by it."
Post-DBS Caregiver	020	My dad was a big part of it and [the clinicians] explained everything to him. They were saying that, "We cannot guarantee what's going to happen. This is our last option, you have to try that out." And that's how we decided to do it
Post-DBS Caregiver	022	But the way they explained it to us, her dad at the time, the way they explained it to us, it was just scary to me. And to think that her brain was going to be open and they were going to stick stuff in there. And the risks, and then it might not even work. And so we said, "No, let's hold off until that's the last resort." So we went through more medications
Post-DBS Caregiver	024	I think I didn't necessarily know what to expect. I knew they were telling me there's a chance that this will not work at all. They said, "There is a chance that this won't work at all." We'll go through all of this, and that won't work. That was my biggest fear, we're going to do all of this and it's not going to work
Post-DBS Caregiver	026	And so, [the clinicians] were really upfront and transparent that, "We can make no promises. I wouldn't do it if I didn't think he was a good candidate, but he's not a great candidate."

to regain independent skills (32%, 14/44), and for the child to gain the ability to walk or run (34%, 13/44).

While caregivers acknowledged that they had high hopes for the potential outcomes of DBS, they explained that their expectations were realistic. One post-DBS caregiver explained that they had set realistic expectations while maintaining high hopes (see Table 5). Many caregivers (60%, 26/44) had hopes for small improvements: these included hopes for a decrease in dystonic storms, controlling chorea, improved sleep, increased confidence, and for DBS to make feeding the child easier. Achieving minor improvements in quality of life for both patients and caregivers was the central hope for most caregivers. One caregiver explained that even small improvements to patient functioning were important (see Table 5).

Another parent explained that they simply hoped that their child would experience some decrease in pain (see Table 5). Another caregiver noted that they were hoping for a miracle cure, but also accepted that DBS would likely only help with symptom management (see Table 5). High (even "unrealistic") hopes could coexist with realistic expectations for outcomes.

Table 5 Patient demographics

Patients	$n=9$	%
Gender		
Male	5	26.3
Female	8	42.1
Prefer not to answer	0	0
No Response	6	31.6
Race/ethnicity		
Hispanic/Latinx	1	5.2
White or European American	12	63.2
Native Hawaiian or Other Pacific Islander	1	5.2
Prefer not to answer	0	0
No Response	5	26.3
Age		
12	3	15.8
13	1	5.2
16	1	5.2
19	2	10.5
21	1	5.2
22	1	5.2
24	1	5.2
25	2	10.5
Prefer not to answer	1	5.2
No Response	6	31.6

DBS Simply had to be Done—Choice (and Hope) was Irrelevant

Caregivers (77%, 34/44) expressed that DBS essentially had to be done because it was their last option. Some caregivers expressed that the patients wanted to get DBS due to the significant impact on quality of life and ability to engage in normal activities (see Table 5).

Some caregivers reported that their child's condition had progressed so far that they felt they had no other choice. One caregiver expressed that they were desperate to get DBS in order to increase patient independence. Another caregiver expressed that DBS progressed so quickly that the caregivers felt they had no other option (see Table 5). One caregiver stated that clinicians had stated that DBS was the last option for the patient, which led the caregiver to ignore potential risks. In their view, hope and choice did not play much of a role.

Expectation Setting for Self/Child can be Difficult

Caregivers (54.3%) expressed that they struggled setting outcome expectations due to the uncertainty of DBS. One caregiver explained that variability and complexity of the disease made expectation setting difficult. Caregivers also cited the lack of information about *childhood* dystonia as another issue making expectation setting difficult. The fac that DBS was an experimental treatment with uncertain outcomes complicated decision-making and made expectation-setting impossible. Because even clinicians could not provide estimates for potential outcomes with high levels of confidence, caregivers felt ill-equipped to set expectations (see Table 5).

One caregiver explained that they wished the clinic had given more information for what to expect during DBS surgery and programming in order to help caregivers set better expectations (see Table 5). Another caregiver explained that because DBS is situated as a miracle cure, it can be difficult to set realistic expectations about DBS outcomes (see Table 5). This highlights the need for realistic expectations from clinicians to help facilitate goal setting for caregivers.

Unmet Expectations can Result in Disappointment

There was a large number (43%, 19/44) of caregivers who expressed negative feelings post-DBS treatment due to unmet expectations. caregivers expressed that difficulty in setting expectations was one factor leading to disappointment. One caregiver explained that the family expected the child would begin walking and running. When those expectations were unrealized, family members were unhappy (see Table 5). Multiple caregivers expressed that they had high hopes for how DBS would impact patients, and they felt disappointed post-intervention when those expectations were unmet (see Table 5).

Caregivers also expressed that the patients experienced disappointment with outcomes as well. One caregiver stated that their child believed DBS would be a complete cure, but the patient did not internalize this and was disappointed that they had undergone a complicated procedure for imperfect results (see Table 5). While caregivers tried to set realistic expectations with patients, they were not always successful, which could lead to disappointment and other negative outcomes among patient populations.

Another factor leading to disappointment was the length of time that side effects from DBS lasted. One caregiver explained that post-DBS, the child would display delays in processing and replying to questions, which was a surprising outcome (see Table 5). Lack of expectation setting and management regarding outcomes and side effects of DBS leads caregivers to feel disappointed with outcomes that they otherwise would be happy with.

The finding that 54.3% of caregivers expressed that they struggled setting outcome expectations and 43% of caregivers expressed disappointment with outcomes post-DBS highlights the need for clinicians to set and manage expectations throughout the treatment process.

The Benefits of Hope

While physicians viewed caregivers' unrealistic hopes and expectations as potentially having negative consequences, caregivers described that maintaining high hopes – even if potentially unrealistic – could still be beneficial for them. For example, caregivers said that hoping for the dramatic improvements in dystonia symptoms portrayed in certain of the educational videos allowed caregivers to overcome their fear of potential risks. One parent explained that seeing a child who received DBS and who was now able to run marathons gave her "the hope I needed to move forward" with DBS treatment (see Table 5). Another caregiver expressed that viewing videos of patients who had significant improvements post-DBS was necessary in order to consent to DBS.

Experiences with Clinicians Setting Low Expectations

Many caregivers (43%, 19/44) had experiences with clinicians who seemed to intentionally try to set low expectations about the potential benefits of DBS (described earlier as "laying crepe"). Many caregivers expressed that the clinician warned that they were unable to guarantee any specific positive outcomes. One caregiver explained that the clinician emphasized DBS may only slightly improve patient symptoms (see Table 5). Multiple caregivers explained that clinicians explained that DBS had uncertain outcomes and clinicians could not guarantee significant improvements. Caregivers expressed that clinicians seemed to intentionally lower their expectations from the outset to decrease instances of disappointment.

Some caregivers felt that the clinician laying crepe helped them make a more informed decision. One caregiver explained that it was better to know potential realistic outcomes than be ignorant (see Table 5). However, laying crepe sometimes led to caregivers unnecessarily delaying DBS. One caregiver expressed that the clinician laying crepe made them nervous about deciding to consent to DBS, explaining that the over-emphasis on risks of DBS was scary and led them to put DBS off (see Table 5). The overly-negative expression of potential risks and benefits scared some caregivers from utilizing the intervention earlier in the disease course.

Discussion

There has been previous research on suggested and clinician-utilized attempts to manage caregiver hope and optimism within DBS which has found that clinicians attempt to showcase the uncertainty about potential outcomes for patients. [15, 18] Our findings build off of these data: we found that clinicians worried that caregivers developed unrealistic expectations for DBS outcomes in a way that could compromise informed consent, and thus attempted to temper caregiver hopes intentionally setting low expectations and/or emphasizing the uncertainties involved (a practice called “laying crepe”). The phrase “laying crepe” or “hanging crepe” is used in medical literature (often surgery) to refer to the practice of providing pessimistic and dismal portrayal of outcomes, often to avoid patient disappointment. The origin of the term comes from funeral history—undertaker assistants would hang black cloth (crepe) over the windows of homes of the deceased and over their casket—these assistants were called “crepe hangers” [19]. The clinicians that we interviewed reported that they believed the practice of laying crepe would help caregivers develop more realistic expectations and thereby aid in good decision-making. However, many caregivers considered themselves to indeed have high hopes for DBS for their child but counterbalanced those hopes with realistic expectations. What are we to make of this practice of clinicians laying crepe to counter potential unrealistic expectations or false hopes by families during decision making?

One issue with laying crepe is that, instead of enabling quality decision-making, it can be a barrier to caregivers engaging in an accurate risk-benefit analysis. Clinicians have an obligation to facilitate informed consent and help patients make informed medical decisions. Understanding the risk-benefit profile of medical interventions is critical for informed consent (and assent in children to the extent appropriate) as well as establishing realistic expectations of treatment response. While clinicians engaged in laying crepe with the intention of improving decision-making, this practice can undermine the process of informed consent.

On the other hand, clinicians engage in laying crepe in order to prevent disappointment. The concern that caregivers will be disappointed is warranted, as 50% (14/28) caregivers expressed disappointment with outcomes post-DBS. However, feelings of disappointment with outcome are not exclusive or incongruent with caregiver’s making good, informed decisions.

Given this information, it is clear that there are both benefits and drawbacks of clinicians laying crepe. Among caregivers that have high hopes and unrealistic expectations for DBS outcomes, laying crepe may be a necessary precaution for clinicians to take in order to ensure good decision-making on the part of caregivers. If caregivers believe that DBS will be a miraculous cure, they are unlikely to be engaging in an accurate and informative risk–benefit analysis. Clinicians could be right to engage in laying crepe in situations like this to help facilitate good decision-making and prevent potential decisional-regret post-intervention. However, it appears that among most caregivers pursuing DBS, laying crepe may be an unnecessary and even ethically dubious practice, as the practice may pose a barrier to effective decision-making. If caregiver ability to accurately evaluate the risks and benefits of DBS is disrupted by laying crepe, clinicians should change their risk communication methods accordingly. Furthermore, if clinicians engage in the practice of laying crepe among caregivers who already have unrealistically pessimistic expectations for DBS outcomes, clinicians can unintentionally disincentivize caregivers from pursuing a helpful intervention.

Importantly, laying crepe is not always a strictly negative endeavor. For example, with some forms of dystonia, DBS is significantly less effective. In these cases, clinicians have an obligation to help patients and caregivers understand that the benefits of DBS may be limited. In these cases, engaging in the practice of laying crepe can better prepare caregivers for realistic outcomes. However, given that DBS is only offered for patients with refractory dystonia and is a last-resort treatment, many caregivers already have low expectations for outcomes. This was reflected in our results showing that most caregivers self-reported realistic expectations for DBS outcomes.

Limitations

Empirically evaluating the effects of unrealistic hope and optimism in the case of DBS for pediatric refractory dystonia is complicated and multi-faceted. One limitation of our study was that it is difficult to parse whether caregiver self-reports about levels of realistic hope and optimism. While 74% of caregivers viewed themselves as having high hopes but reasonable expectations, this self-report may be inaccurate. Firstly, caregivers’ self-perceptions of accuracy of expectations is subjective. This self-perception can be influenced by biases, such as hindsight bias, making such self-reports inaccurate. Even caregivers who had overwhelmingly realistic expectations still voiced a mix of some unrealistic expectations as well. This included caregivers who stated that they had hopes for a miraculous cure while simultaneously hoping for very minor patient improvements, such as the patient sleeping better.

Another major challenge in differentiating unrealistic optimism from realistic hope is that some patients and caregivers may outwardly present themselves as overly optimistic in communication with physicians. This could be due to concerns that others may prefer or expect them to remain positive, or due to fears of 'jinxing' themselves (Blumenthal-Barby and Ubel). Indeed, this may be what was happening with our 74% of caregivers who viewed themselves as having high hopes but reasonable (even low) expectations for DBS when the clinicians (85%) worried they had false hopes. As a result, clinicians may end up laying crepe with caregivers who actually hold realistic expectations for DBS. If laying crepe negatively skews previously realistic expectations, then the practice may compromise the informed decision making of caregivers about DBS for their children.

Finally, our research was focused on the perspectives of the legal decision-makers (i.e., clinicians and caregivers) on realistic versus unrealistic hope and optimism. One area for future research is to include the perspectives of pediatric patients. Given that patients have insight into their own disorder, it would be beneficial to elicit their perspectives.

Conclusion

Understanding caregiver and clinician perspectives on hope and optimism in decision-making is vital for implementing novel neurotechnologies in clinical settings [20, 21]. Many of the concerns that clinicians expressed about false hope and unrealistic optimism distorting caregivers informed decision making about DBS for their child were minimized by caregivers. While clinicians expressed concerns that caregivers would have unrealistic hopes about DBS outcomes, 74.3% of caregivers reported that they believed their hopes were realistic and that they often hoped for small changes. Clinicians expressed concerns that unrealistic hopes might be generated by social media, and conversely, caregivers reported that social media improved their support network and enabled them to make difficult healthcare decisions. Caregivers felt that high hopes paired with realistic expectations enabled them to make difficult healthcare decisions. Collectively, these data highlight the need for ongoing and thorough discussion between clinicians and stakeholders to set realistic expectations for child health outcomes.

Acknowledgements Research for this article was funded by the BRAIN Initiative-National Institutes of Health [parent grant R01MH121371] (Blumenthal-Barby, Storch, Lazaro-Munoz). The views expressed are those of the authors and do not necessarily reflect views of the NIH, Baylor College of Medicine, or Harvard Medical School.

Data Availability The participants of this study did not give written consent for their data to be shared publicly, so due to the sensitive nature of the research supporting data is not available.

Declarations

Conflicts of Interest The authors have no conflicts of interest to disclose.

References

1. Hale, A.T., M.A. Monsour, J.D. Rolston, R.P. Naftel, and D.J. Englot. 2020. Deep brain stimulation in pediatric dystonia: A systematic review. *Neurosurgical Review* 43 (3): 873–880. https://doi.org/10.1007/s10143-018-1047-9.
2. Elkaim, L.M., N.M. Alotaibi, A. Sigal, H.M. Alotaibi, N. Lipsman, S.K. Kalia, et al. 2019. Deep brain stimulation for pediatric dystonia: A meta-analysis with individual participant data. *Developmental Medicine and Child Neurology* 61 (1): 49–56. https://doi.org/10.1111/dmcn.14063.
3. Muñoz, K.A., J. Blumenthal-Barby, E.A. Storch, L. Torgerson, and G. Lázaro-Muñoz. 2020. Pediatric Deep brain stimulation for dystonia: Current state and ethical considerations. *Cambridge Quarterly of Healthcare Ethics* 29 (4): 557–573. https://doi.org/10.1017/S0963180120000316.
4. Zuk, P., and G. Lázaro-Muñoz. 2021. Treatment search fatigue and informed consent. *AJOB Neuroscience* 12 (1): 77–79. https://doi.org/10.1080/21507740.2020.1866115.
5. Blumenthal-Barby, J.S., and P.A. Ubel. 2018. In defense of 'Denial': Difficulty knowing when beliefs are unrealistic and whether unrealistic beliefs are bad. *The American Journal of Bioethics* 18 (9): 4–15. https://doi.org/10.1080/15265161.2018.1498934.
6. Weinstein, N. 1980. Unrealistic optimism about future life events. *Journal of Personality and Social Psychology* 39 (5): 806–820.
7. Kadlac, A. 2015. The virtue of hope. *Ethical Theory and Moral Practice* 18 (2): 54–337.
8. Jansen, L.A., et al. 2016. The impact of unrealistic optimism on informed consent in early-Phase Oncology trials. *IRB* 38 (5): 1–7.
9. Schwitzgebel, E. 2010. Acting contrary to our professed beliefs or the Gulf between occurrent judgment and dispositional belief. *Pacific Philosophical Quarterly* 91 (4): 531–553.
10. Mele, A.R. 2000. *Self-deception unmasked*. Princeton: Princeton University Press.
11. Lear, J. 2006. *Radical hope: Ethics in the face of cultural devastation*. Cambridge: Harvard University Press Kindle edition.

12. Martin, A. 2013. *How we hope: A moral psychology*. Princeton: Princeton University Press.
13. Bell, E.E., B. Maxwell, M.P. McAndrews, A.F. Sadikot, and E. Racine. 2010. Hope and patients' expectations in deep brain stimulation: Healthcare providers' perspectives and approaches. *The Journal of Clinical Ethics* 21 (2): 112–124.
14. Boyatzis, R. 1998. *Transforming qualitative information*. https://us.sagepub.com/en-us/nam/transforming-qualitative-information/book7714. Accessed 27 Jun 2023.
15. Gardner, J., G. Samuel, and C. Williams. 2015. Sociology of low expectations: Recalibration as innovation work in biomedicine. *Science, Technology & Human Values* 40 (6): 998–1021.
16. Benetoli, A., et al. 2018. "How Patients' use of social media impacts their interactions with healthcare professionals. *Patient Education and Counseling* 101 (3): 439–44. https://doi.org/10.1016/j.pec.2017.08.015. ScienceDirect.
17. Braun, V., and V. Clarke. 2021. One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology* 18 (3): 328–352.
18. Gardner, J., et al. 2019. Persuasive bodies: testimonies of deep brain stimulation and Parkinson's on youtube. *Social Science & Medicine* 222: 44–51. https://doi.org/10.1016/j.socscimed.2018.12.036. ScienceDirect.
19. Hundley, J. (2015). Laying crepe. *OrthopaedicLIST.Com Blog*. https://www.orthopaediclist.com/blog/laying-crepe/
20. Gardner, J., and A. Cribb. 2016. The dispositions of things: The non-human dimension of power and ethics in patient-centered medicine. *Sociology of Health & Illness* 38 (7): 1043–1057.
21. Gardner, J. 2017. Securing a future for responsible neuromodulation in children: The importance of maintaining a broad clinical gaze. *European Journal of Paediatric Neurology* 21 (5): 49–55. https://doi.org/10.1016/j.ejpn.2016.04.019.