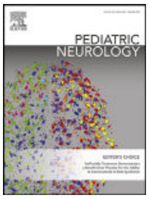
Pediatric Neurology 163 (2025) 50–57

Contents lists available at ScienceDirect

Pediatric Neurology

journal homepage: www.elsevier.com/locate/pnu



Research Paper

Communication About Sudden Unexpected Death in Epilepsy: An Adaptation of the SPIKES Protocol

Kayli Maney, MD [a], Isabella K. Pallotto, MPH [b], Simran Bansal, BS [a], Shital Patel, MD [a],
Renée A. Shellhaas, MD, MS [c], Zachary M. Grinspan, MD, MS [d],
Jeffrey Buchhalter, MD, PhD [e], Elizabeth J. Donner, MD, MSc [f],
Gardiner Lapham, MPH, RN [g], Thomas Stanton, MPP [h], Monica E. Lemmon, MD [a, *]

[a] *Department of Pediatrics, Duke University School of Medicine, Durham, North Carolina*
[b] *Department of Psychology, University of Cincinnati College of Arts & Sciences, Cincinnati, Ohio*
[c] *Department of Neurology, Washington University School of Medicine, St. Louis, Missouri*
[d] *Department of Pediatrics, Weill Cornell Medicine, New York, New York*
[e] *Department of Pediatrics, Alberta Children's Hospital, Calgary, Alberta, Canada*
[f] *Department of Neurology, The Hospital for Sick Children, Toronto, Ontario, Canada*
[g] *BAND Foundation, Washington, District of Columbia*
[h] *Danny Did Foundation, Evanston, Illinois*

ARTICLE INFO

Article history:
Received 18 April 2024
Accepted 4 October 2024
Available online 21 October 2024

Keywords:
Sudden unexpected death in epilepsy (SUDEP)
Communication
Prevention
Risk factors

ABSTRACT

Background: Sudden unexpected death in epilepsy (SUDEP) is a common cause of premature death in children and adults with epilepsy. People with epilepsy and their caregivers prefer the risk of SUDEP to be disclosed by their clinicians; however, few tools exist to support these conversations.
Methods: We aimed to (1) characterize SUDEP communication preferences of clinicians and caregivers of children with epilepsy, and (2) leverage these preferences to develop a conversation guide to support the discussion of SUDEP risk. We prospectively enrolled caregivers of children with epilepsy, child neurology clinicians, and child neurology trainees to participate in virtual focus groups and/or 1:1 interviews.
Results: Content was analyzed and collated into the following four domains: (1) *who should participate*: all participants described that the treating neurologist and/or epileptologist should lead conversations with patients and families. Caregivers preferred that clinicians disclose information about SUDEP to children only after asking permission. (2) *When and where conversations should occur*: most participants felt that the conversation should be discussed at the time of diagnosis and in the outpatient setting, (3) *content to include about SUDEP risk*: participants characterized the need to define SUDEP, what is known and unknown, as well as describe risk factors to reduce an individual child's risk, and (4) *barriers and facilitators to disclosure*: barriers included lack of time, limited evidence-based information regarding risk mitigation, lack of modifiable risk factors, and apprehension of causing undue stress on families. Participants shared that written material would help facilitate the discussion.
Conclusions: Caregivers and clinicians emphasized the value of SUDEP risk counseling in the pediatric clinical setting. Based on these findings, neurologists caring for children with epilepsy should educate caregivers about SUDEP soon after the initial epilepsy diagnosis in an empathetic manner, revisiting the conversation longitudinally over time and highlighting actionable steps to mitigate risk. Caregiver and clinician preferences informed our novel adaptation of the SPIKES protocol for use by pediatric neurologists in the context of SUDEP risk disclosure.

Introduction

Despite advances in epilepsy diagnosis and treatment, people living with epilepsy are at risk of early death and significant morbidity.[1] The incidence of sudden death among people with epilepsy is 24 times higher than that among age-matched peers

---

This work was supported by the BAND Foundation.

* Communications should be addressed to: Dr. Lemmon; Department of Pediatrics; Duke University School of Medicine; DUMC 3936; Durham, NC 27710.
E-mail address: monica.lemmon@duke.edu (M.E. Lemmon).

https://doi.org/10.1016/j.pediatrneurol.2024.10.004
0887-8994/

without epilepsy.[2] An important direct cause of mortality is sudden unexpected death in epilepsy (SUDEP).[1] The main risk factors are related to frequency of generalized tonic-clonic seizures (more frequent = higher risk), setting (sleep = higher risk), seizure type (generalized tonic-clonic = higher risk), and age of epilepsy onset (younger age = higher risk). Nocturnal supervision (i.e., having a bed partner or nighttime monitoring device) is associated with lower risk. The absolute risk of SUDEP is 1.2 cases per year for every 1000 people with epilepsy, making it second only to stroke in the number of potential years of life lost due to a neurological condition.[1,3,4] Furthermore, rates of SUDEP may be underestimated due to poor recognition, incomplete death records, and misattributed causes of mortality.[2,5]

Caregivers of children with epilepsy strongly prefer that clinicians initiate upfront and longitudinal discussions about SUDEP, which is also recommended by the American Academy of Neurology and American Epilepsy Society.[6-10] Educating caregivers and patients about SUDEP in the context of risk reduction may promote medication adherence, reduce medical misinformation, strengthen the rapport between clinicians and families, and ultimately lead to better health outcomes for people with epilepsy.[6-8,11] Despite the potential benefits of SUDEP counseling, neurologists' current practices diverge from official guidelines and family preferences, with some studies reporting SUDEP counseling incidence as low as 1% in both adult and pediatric settings.[9-12] Providers commonly cite barriers that discourage them from initiating SUDEP counseling, including a lack of high-quality evidence regarding prevention, time constraints, fear of exacerbating anxiety in patients and caregivers, and knowledge and training deficits.[6,12-14] These barriers may be magnified in the pediatric setting, where clinicians concurrently attend to the differing needs and developmental levels of children with epilepsy and their caregivers.[6,15]

There are well-established communication tools to help clinicians disseminate difficult news to patients and families in a supportive and empathetic manner.[16,17] One example, the SPIKES protocol, is a six-step framework that improves patient-provider communication in multiple clinical settings.[17-22] Each letter represents a step: S stands for Setting up the interview, P for assessing the patient's Perception, I for obtaining the patient's Invitation, K for providing Knowledge, E for Empathetic responses, and S for Strategy/Summary.[19] Clinicians have used SPIKES to deliver information when poor prognostic outcomes are likely.[16] SUDEP counseling is different, as the conversation centers around a high-stakes but uncommon event with modifiable risk factors.[3,4] For this reason, the SPIKES protocol needs to be adapted to effectively address the communication challenges faced by neurologists when they disclose the risk of SUDEP to caregivers and/or patients. In this study, we aimed to (1) characterize the SUDEP communication preferences of clinicians and caregivers of children with epilepsy and (2) leverage these preferences to develop a conversation guide tailored to the discussion of SUDEP risk.

Methods

Participants and recruitment

We prospectively enrolled caregivers of children with epilepsy, trainee clinicians, and attending physicians who treat people with epilepsy to participate in focus groups or 1:1 interviews to discuss best practices in SUDEP risk disclosure. The Duke University Institutional Review Board approved this study. All participants were offered compensation of $50 for interview or focus group completion.

Caregiver inclusion criteria were (1) current or former primary caregiver of a child diagnosed with epilepsy and (2) currently living in the United States. Caregivers were primarily recruited from an existing cohort of caregivers who participated in a survey study of SUDEP risk disclosure preferences.[23] In this previous study, participants could opt in to further contact. We invited all opt-in participants to participate in the present study. Study team members contacted interested and eligible caregivers with a living child with epilepsy and provided information about the focus group and a link to an online form (Qualtrics; Provo, UT, USA) to sign up for a 90-minute focus group. Two additional caregivers were recruited for interviews through the Epilepsy Foundation (Landover, MD, USA).

We considered clinicians for inclusion if they were a current or former child neurology clinician. Clinicians were recruited through the Child Neurology Society platform (childneurologysociety.org) and invited to participate in interviews. Trainees were considered for inclusion if they were participating in a child neurology residency training program. Trainees were recruited from the participants of the 2021 Pellock Resident Seminar on Epilepsy of the Child Neurology Society conference. Trainees were e-mailed information about the study and invited to participate in a focus group.

Data collection

In-person and virtual focus groups were recorded by two encrypted audio recorders. Virtual focus groups were held on Zoom (San Jose, CA, USA). Audio recordings were transcribed and deidentified. All focus groups were led by a trained facilitator (M.E.L.) and followed a semistructured focus group guide. Interviews were semistructured and completed by a trained study team member (I.K.P. and M.E.L.).

Data analysis

The study team consisted of physicians, clinical research staff, members of nonprofit organizations, and parent advocates, and includes individuals with expert knowledge in qualitative study design, qualitative analysis, epilepsy, and neurology. Rapid-cycle qualitative analysis[24] was used to analyze the data after professional transcription. Domains were prespecified in a data collection guide, transcripts were summarized, and the summaries were collated into a matrix to assess key findings (Table 1). Independent team members (I.K.P., K.M., S.B.) summarized each question within each focus group using the same structured template. Structured summaries for each question were put into a matrix, and matrix analysis was used to compare across coders. The study team used an inductive approach to extract themes from the data and discussed the themes until we came to consensus. All differences were resolved and adjudicated by an independent third study team member (M.E.L.).

Application of the SPIKES framework

Findings from the focus groups and interviews were used to illustrate how the SPIKES framework can be adapted to guide SUDEP counseling. Specifically, we integrated all the responses we received and synthesized them into one of the six subcategories (S-P-I-K-E-S) of the framework.

Results

Participant characteristics

Eighteen caregivers, eight trainees, and 10 clinicians participated in focus groups and 1:1 interviews (Tables 2 and 3). Caregivers were predominantly white ($n = 15$) and female ($n = 17$). The

TABLE 1.
Focus Group Guide and Template for Structured Summaries

Domain	Questions and Probes
Who	Who should discuss SUDEP with parents? Who should discuss SUDEP with kids?
What	What should be included in conversations about SUDEP risk?
Where	What is the appropriate setting for conversations about SUDEP risk?
When	When should clinicians discuss SUDEP? How often should conversations about SUDEP be initiated? When is it inappropriate to discuss SUDEP risk?
How	How should clinicians approach conversations about SUDEP? How should clinicians engage children in conversations?
Facilitators	What tools could you use to discuss SUDEP with your child's clinician?
Barriers	What do you think the barriers are that prevent clinicians from disclosing SUDEP to caregivers?
Training (trainee and clinician groups only)	What training did you receive in communicating about SUDEP?

Abbreviation:
SUDEP = Sudden unexpected death in epilepsy

children of the caregivers had a mean age of 16 years (range: 0 to 35 years), and most had a high seizure burden, with a mean of 39 seizures per year (range: 1 to >100 seizures). Most children had a history of nocturnal seizures ($n = 14/16$, 88%), and over half had a history of generalized tonic-clonic seizures ($n = 11/16$, 69%).

Trainees were all in their final year of residency and had a mean age of 32 years (range: 30 to 37 years). Clinicians had a mean age of 50 years (range: 37 to 77 years), were predominantly white ($n = 7/10$, 70%) and female ($n = 6/10$, 60%), and all currently worked in an academic setting. Clinicians were in practice for a mean of 17 years (range: 3 to 46 years), and most were epileptologists ($n = 8/10$, 80%).

Codes and themes were organized into four domains: (1) who should participate in conversations, (2) when and where should conversations occur, (3) content to include in a conversation, and (4) barriers and facilitators to risk disclosure. Representative quotations are in Table 4.

Who should lead and participate in conversations about SUDEP risk

Caregivers, trainees, and clinicians uniformly agreed that the treating neurologist and/or epileptologist should be the primary source of SUDEP risk communication for patients and families. In caregiver focus groups, some caregivers noted that with prerequisite knowledge about SUDEP, other provider types such as social/case workers, advanced practice providers, and primary care providers may be appropriate to support or lead conversations. Some clinicians, however, expressed concern that primary care providers, as well as support staff, may not be trained to facilitate these conversations.

Viewpoints varied on whether children ought to be included in conversations about SUDEP risk. Several clinicians and caregivers felt that older children with the emotional and cognitive maturity necessary to understand the conversation could benefit from hearing information about SUDEP risk directly. Caregivers expressed that even without learning about SUDEP in the clinical setting, these older children often already have a sense that their epilepsy comes with risks: “*You have to assume that kids are… already checking things out themselves. Especially now, on the internet…. I think [my daughter] was already asking those questions herself, like ‘Am I going to die?’*” However, participants had reservations about including younger children and those with cognitive impairment in SUDEP risk conversations, explaining that they may

TABLE 2.
Caregiver Demographic Characteristics

Mean	*M* (*S.D.*)	Min-Max	*n* (%)
Caregivers ($N = 18$)			
Age (years)	48 (10.81)	31-68	
Gender			
Woman			17 (93.75)
Man			1 (6.25)
Race			
White			15 (93.75)
Black or African American			3 (6.25)
Ethnicity			
Non-Hispanic			12 (75.0)
Hispanic			2 (12.5)
Not reported			2 (12.5)
Region of residence in the United States			
South			9 (56.3)
Midwest			3 (18.8)
Northeast			3 (18.8)
West			1 (6.3)

Abbreviations:
Max = Maximum
Min = Minimum

TABLE 3.
Trainee and Clinician Demographic Characteristics

Mean	*M* (*S.D.*)	Min-Max	*n* (%)
Trainees ($N = 8$)			
Age (years)	32.38 (2.67)	30-37	
Gender			
Woman			3 (37.5)
Man			5 (62.5)
Race			
White			4 (50)
Asian			3 (37.5)
Prefer not to answer			1 (12.5)
Ethnicity			
Non-Hispanic			7 (87.5)
Hispanic			1 (12.5)
Year in training			
PGY5			8 (100)
Clinicians ($N = 10$)			
Age (years)	50.1 (13.88)	37-77	
Gender			
Woman			6 (60)
Man			4 (40)
Race			
White			7 (70)
Asian			2 (20)
Black or African American			1 (10)
Ethnicity			
Non-Hispanic			10 (100)
Hispanic			0
Years in practice	17.2 (14.6)	3-46	
Specialty			
Neurologist			2 (20)
Epileptologist			8 (80)

Abbreviations:
Max = Maximum
Min = Minimum
PGY = Postgraduate year

TABLE 4.
Themes and Representative Quotations

- Who should lead and participate in conversations about SUDEP risk
  - *"I think it should be the diagnosing neurologist or epileptologist. They're the ones that are really giving you that this is epilepsy diagnosis."* —Caregiver
  - *"I think the neurologist is the most logical. Hopefully, they know the most about it, but I think the primary care physician should also, because they might see that person more often."* —Caregiver
  - *"I think it has to be done on a case by case basis because my son has intellectual and developmental disabilities, so sharing the information with him just wouldn't make sense."* —Caregiver
  - *"With different family dynamics, who they stay with, if they have multiple caretakers then I think the conversation probably needs to be had with various caretakers the child is having."* —Trainee
- When and where conversations about SUDEP risk should occur
  - *"It's not just what needs to be included but when. What I find in a new diagnosis of epilepsy or recurrent seizures most people shut down pretty fast and don't hear very much and there's also a phenomenal amount of anxiety so it's not something I'd like to entertain that first visit unless they bring it up."* —Clinician
  - *"Maybe not after the first seizure because everybody can have a seizure if the conditions are just right, but after a child is diagnosed with epilepsy and is taking medication, I do think this difficult conversation needs to happen."* —Caregiver
  - *"You have to hear it several times in several different ways before it sinks in."* —Caregiver
  - *"It depends on the epilepsy and the patient, so if it, for most benign epilepsies I think it should be talked about as early as possible, but I don't know that you need to keep reiterating it. As the patient evolves, if they evolve to intractable epilepsy or having nocturnal seizures or have medication compliance issues, I think those are times where it should be revisited."* —Trainee
- Content to include in a conversation about SUDEP risk
  - *"Define SUDEP. Give risks associated with the diagnosis. Communicate ways to minimize SUDEP that are in caregivers control and then give local supports for more information. For example, Epilepsy Foundation, the medical team."* —Caregiver
  - *"You know, the first thing I'm going to want to know are what are my odds? You know, what are the chances that we're going to be in this percentage, what are we doing, what are his risk factors, and what can I do to minimize those risks? You know, how do you have a normal life with that constant anxiety of the things that you can't control with just 100% epilepsy anyway, but how do you sleep at night?"* —Caregiver
  - *"I think naming it is a big ingredient in the conversation and then also going through what we know to be the risk factors about it and that's to the agency piece. It's not just the information. It's also the agency and how do I make it actionable for myself, for my kid, for my patient, all of these things and that's where people feel like they have a sense of control, understanding that this feels like something that feels out of control."* -Clinician
  - *"Let [the family] guide [the conversation]. I think that's a good framework to start. I think the family will open up and ask you any number of questions that you might not have anticipated, so I think, just feeling comfortable with the material is useful. And also meeting them where they're at and going from there."* -Trainee
- Barriers and facilitators to SUDEP risk disclosure
  - *"Even the low-risk conversation and certainly the high-risk conversation risks unearth all kinds of family issues with anxiety and maladaptive coping, and I don't have access to an army of social workers or psychologists to help those families."* —Clinician
  - *"I think that's probably what stops me from talking about it, is, when I deliver this potentially terrible news that's probably not going to happen, and then there's this whole uncertainty about it, I think it makes it harder to talk about it, especially in a small time frame."* —Trainee
  - *"Not to take anything away from doctors, cause they're amazing, but I feel like the best information I've gotten has been from parents and other people that have gone through a somewhat similar situation or had a child with a health condition. I feel like that's where I've gotten most of my good information and most of my support. Maybe any kind of support material would include you know, some kind of real-life parent experience or some kind of real-life parent contact information, because I don't, those were things I had to hunt for and were not necessarily just given to me."* —Caregiver
  - *"Even if it's a pamphlet, if [doctors] don't feel comfortable having that conversation themselves, have a pamphlet that you can give the families that maybe has some website links in there. Here is some more information on this. Here are some websites that you can go to that might explain it better than I can."* -Caregiver

Abbreviation:
SUDEP = Sudden unexpected death in epilepsy

not have the necessary cognitive ability to process the information. Some caregivers warned that sharing information about SUDEP in the presence of a young child, who may understand words like "die" and "seizure," but not nuanced information about risk, could result in significant fear: "*….I think if [my son] was in the conversation, he would hear certain key words…but not have the mental ability to understand that and would walk away with, 'Every time I have a seizure, I'm going to die.'*" Regardless of a child's age or cognitive abilities, caregivers shared that families should decide whether information about SUDEP should be disclosed to their children.

When and where to have conversations about SUDEP risk

In caregiver focus groups, most participants felt that the conversation surrounding SUDEP risk should occur at the time of diagnosis: "*As parents of a child with seizures, that should be the very first thing that they talk about…. The doctors need to be forthcoming with information. Education is power and education will save lives.*" Although clinicians and trainees agreed that SUDEP disclosure should occur early, there was some disagreement regarding whether it should be brought up at the time of diagnosis or at a subsequent visit, after rapport had been established. Many clinicians recommended that SUDEP should instead be disclosed at the second or third visit out of concern that families might be too overwhelmed at the time of diagnosis to have the discussion.

Caregiver focus groups felt that SUDEP should be revisited often, allowing families the chance to ask questions as they arise and engage in ongoing discussion. In contrast, many clinicians and trainees feared that repeated conversations about SUDEP might lead families to experience undue stress and urges to be overprotective. Instead, they recommended discussing SUDEP at future visits only with changes in risk such as worsening seizures and medication noncompliance.

Although all participants felt that SUDEP could be discussed in the inpatient or outpatient setting, most favored the outpatient setting. As shared by a caregiver, *"I don't think you can wait until you have the perfect setting to give the information…. Ideally it would be part of your regular office visit, but I wouldn't wait. You never know when you're not going to get another office visit."* Clinicians had mixed perspectives on the role of telehealth as a medium to discuss SUDEP. Some felt that it would be appropriate to discuss via a telephone call, whereas others felt that this approach would be too impersonal.

Content to include in conversations about SUDEP risk

Caregivers, trainees, and clinicians agreed about the content to include in a conversation about SUDEP risk. Counseling should include the definition of SUDEP, known causes, risk factors, and actionable ways to mitigate individuals' risk. Risk reduction strategies include medication adherence, regular communication with clinicians, and safe sleep practices. Caregivers additionally emphasized the need for clinicians to be transparent about uncertainties associated with SUDEP and to offer information on seizure monitoring devices.

Importantly, caregivers recommended that clinicians complement the disclosure of SUDEP risk with acknowledgement of the potential emotional distress that may arise, actively helping families process and incorporate information about risk into their daily lives: "*It's not just what can we do to minimize risks. Of course, that's important. It's also how do we live with this information? What does it mean for our daily life?*" To help manage expectations, caregivers emphasized the need for clinicians to direct families to informational resources and peer support, citing the value of organizations like the Epilepsy Foundation (www.epilepsy.com; Landover, MD, USA), Child Neurology Foundation (childneurologyfoundation.org), and Danny Did (dannydid.org; Evanston, IL, USA).

Although caregivers and clinicians described topics that should be integrated into SUDEP risk disclosures, they also acknowledged the importance of tailoring these conversations to the individual caregiver and child. One caregiver expressed the need to consider the unique values and experiences of families that may impact communication and understanding, including religious and cultural beliefs: *"[It] is very important that you may want to consider the cultural backgrounds of the people that you're working with, [be]cause sometimes there is culture that kind of dictates how people may respond to a conversation like SUDEP."* When asked how clinicians might probe for this type of information, the caregiver suggested open-ended questions like "*What are your thoughts about your child's epilepsy diagnosis?"* or *"Tell me about the way you understand your child's epilepsy diagnosis*."

Barriers and facilitators to SUDEP risk disclosure

All participants described common barriers to SUDEP risk disclosure. The fear of imposing undue stress and anxiety on families and patients was a central theme that emerged during focus groups and interviews. Clinicians and trainees described the discomfort associated with leading these important, yet emotionally charged conversations: *"I hate making families cry, I feel like I do it all day…. I know that families are going to be thankful that we've had this conversation, but that still doesn't mean that it's not an existentially uncomfortable conversation."* They noted how time constraints further exacerbate their reluctance, as it takes time not only to disclose the information but also to allow patients and families room to digest it.

Another barrier noted by clinicians and trainees was the lack of formal training, related both to the specific context of SUDEP and more broadly to the provision of emotional support during serious conversations. As shared by one trainee, *"I haven't received any formal training in SUDEP communication. I don't think it's come up in our weekly lectures or in cases. I similarly can't think of a time that I've seen it modeled either."* Even with training, inherent constraints persist due to the gaps in existing knowledge of SUDEP risk mitigation. As explained by one caregiver, this uncertainty may discourage certain clinicians from initiating discussions about SUDEP: *"One of the things I think is a real barrier for doctors, in talking about SUDEP, is the unexplained. In my experience, doctors don't like to talk about things they don't understand."*

Participants also identified facilitators of SUDEP risk disclosure alongside barriers. There was consensus among participants that written material in the form of handouts could promote effective discussions about SUDEP. A few caregivers further specified that having QR codes on the handouts directing them to reputable web sites would allow for easier access to useful information after visits. Some clinicians stated that having caregivers and/or patients fill out previsit surveys assessing their baseline understanding and interest in learning about SUDEP could provide the context to lead collaborative, individualized conversations.

Discussion

We characterized participant experiences and preferences for conversations about SUDEP risk. Most parents of children with epilepsy hope to learn about SUDEP from neurologists, as recommended by the American Academy of Neurology and American Epilepsy Society.[6,10,11,15,25] Yet, barriers such as lack of training, time constraints, clinician knowledge deficits, and the fear of exacerbating distress often prevent these important conversations from occurring.[11,12,14,26] Our findings similarly suggest that clinicians and caregivers simultaneously find value in neurologists engaging caregivers in early, transparent conversations about SUDEP risk while acknowledging common challenges that arise in the clinical setting. Nearly all participants emphasized the importance of educating families about known risk factors, actionable steps to mitigate risk, and treatment adherence. Although interventions involving triggers in electronic health records have shown promising results in improving the frequency of SUDEP risk disclosures, few tools exist to improve the quality of communication.[9,27,28] The SPIKES protocol is a communication framework widely adapted to help clinicians facilitate difficult conversations in various settings.[16,18,19] Here, we outline key findings to support the utility of adapting the patient-centered, empathy-driven SPIKES protocol to the SUDEP context (Table 5).[18,29]

Before the start of SUDEP risk disclosure, it is important to consider the *Setting*, thinking about who should be present, as well as when and where the conversation should occur.[25] Our data suggest that the ideal place for the conversation is in the outpatient setting, when possible, or a telemedicine video visit. The conversation should ideally be led by the neurology clinician who has an established relationship with the patient. Importantly, both trainees and experienced clinicians in this cohort highlighted little to no formal training in how to communicate SUDEP risk. The importance of training is underscored by recent data suggesting that physicians with less training are more likely to leave patients and caregivers dissatisfied after SUDEP disclosure.[12] Taken together, our findings and the existing literature suggest an opportunity for structured communication tools and skills training, both for trainees and experienced clinicians, that include reaching primary care clinicians who may serve as the treating clinician in many settings. Participants also described the benefits of leveraging the diverse skills of interdisciplinary members of the health care team in these conversations. Given the emotionally-charged nature surrounding SUDEP, psychologists and/or social workers can offer families additional social support and offer resources such as caregiver support groups.[15,18,30,31] The timing of the conversation based on our data supported discussion of SUDEP at the time of diagnosis (or by the end of the second visit/interaction) and the need to revisit iteratively with changes in risk and/or lack of attention to modifiable risk factors.

Understanding the caregiver and/or patient's baseline knowledge about SUDEP is the goal of the *Perception* stage. To do so, questions should be asked in an open-ended manner to assess understanding and associated worries surrounding the topic. Some questions can include: "*What have you heard about the risks that can come with seizures*?" and "*Have you ever heard of something called SUDEP? [If yes], Tell me about what you've heard*." By assessing the understanding of families, clinicians can also gain valuable information about any misconceptions that caregivers or patients may hold about SUDEP, as well as information about their health literacy.[32,33] Clinicians can then guide the conversation informed by the caregiver and/or patient's baseline knowledge.

After exploring the perception of families, clinicians can then extend an *Invitation* to further conversation about SUDEP risk. Our data support encouraging clinicians to explicitly invite caregivers to

TABLE 5.
SPIKES Framework for Discussing SUDEP With Families

Step	Goal	Key Points and Example Language
Setting	Set the stage for a collaborative discussion by preparing a comfortable, private space and ensuring that all parties whom the patient wishes to involve are present.	- Introduce SUDEP in the outpatient setting when possible, either in person or via a face-to-face telemedicine visit - Ensure that a neurology and/or primary care clinician with whom the caregiver and patient have an established, trusting relationship with leads the conversation when possible. - Turn off pagers to vibrate and hold nonemergent calls - Discuss SUDEP at the time of diagnosis and revisit iteratively with changes in risk - Integrate SUDEP risk disclosure into the broader discussion of epilepsy risks - Attend to the developmental age and stage of child - Demonstrate active listening skills, centering your attention on the needs of the patient
Perception	Assess the caregiver and/or patient's baseline knowledge about SUDEP using open-ended inquiry.	- *“What worries* do you *have about your child's epilepsy diagnosis?”* - *“What have* you heard *about the risks that can come along with seizures?”* - *“Have you ever heard of something called SUDEP? [If yes], Tell me about what you've heard.”*
Invitation	Ask caregivers and/or patients directly about how much and what kind of information will be helpful to them. If a child is present, ask permission before proceeding with the conversation.	- *“Some parents prefer to have conversations about serious risks of seizures without their child present*; *others prefer to include their child in these conversations. Which do you prefer?”* - *“Would you like to learn more about your child's risk of SUDEP?”* - *“Is it ok if we* discuss *SUDEP today?”*
Knowledge	Share your knowledge about SUDEP in a clear, direct, and comprehensive manner.	- Provide information about SUDEP in direct terms - Deliver information in small segments, allowing caregivers and/or patients time to process - Use simple, nonmedical language matching the caregiver's and/or patient's education level(s) - Conversations about SUDEP should include the following information: ○ Definition: “*SUDEP is the sudden unexpected death of a person or child with epilepsy. We do not yet know the cause of SUDEP, but we do know that SUDEP happens most often at night. SUDEP in children is rare.*” ○ Risk factors: “*There are a few things that can increase a child's risk of SUDEP. Those risk factors include having seizures that are not well controlled, having generalized tonic-clonic seizures where the whole body stiffens and shakes, and having seizures at night.*” ○ Patient's risk level: “*Based on everything we know about [name]'s seizures, his risk of SUDEP is [insert risk level].*” ○ Risk mitigation strategies: “*The best way to prevent SUDEP is to have as few seizures as possible. Things we can do to reduce your child's risk of SUDEP include making sure that he continues taking his medication every day, keeping a seizure diary, learning your child's seizure triggers, and creating a Seizure Action Plan. Let's talk more about [insert risk factor] ….*” ○ Resources to find additional information: “*There are many good resources where you can learn more about SUDEP. Would you like to go over some of those today?*” - Acknowledge any uncertainty that may exist. - Check the understanding of caregivers and/or patients and ask if they need additional clarification: “*This was a lot of information. What questions do you have?”*
Empathy	Acknowledge and respond to caregiver and patient emotions.	- Name emotions when present: “*It sounds like you are worried about…”* - Assess and explore the emotions in the room: *Tell me more about your concerns….* - Normalize the emotions that caregivers may be experiencing: *Some parents describe feeling overwhelmed when learning about SUDEP. How do you feel?* - Validate the efforts of caregivers: *You are such a strong advocate for [name].* - Share resources for further support: *Our team will be here to support you. Would you like to hear about organizations and resources that can help you learn more about SUDEP?*
Summarize	Summarize the information that has been presented using lay language and present a plan for next steps, including referrals to further resources.	- Ask questions to verify that the caregiver and/or patient understands the key information and proposed plan: “*We've reviewed a lot of information today*; *can you tell me what you understand about SUDEP and your child's risk?”* - *“To summarize*, *all children with epilepsy have a risk of SUDEP and your child's risk of SUDEP is [insert risk level]. You can reduce your child's risk by….*”

Abbreviation:
SUDEP = Sudden unexpected death in epilepsy

share their communication preferences, including whether SUDEP should be discussed in the presence of their child. An example of this invitation can include a statement such as *"Some parents prefer to have conversations about serious risks of seizures without their child present*; *others prefer to include their child in these conversations*. *Which do you prefer?"* If possible, personnel should be made available to supervise young children whose caregivers opt to have SUDEP conversations without them present, although we acknowledge that some clinical settings may lack access to additional personnel.[15]

The goal of the *Knowledge* stage is to share information about SUDEP in a direct and comprehensive manner. Key topics to integrate into SUDEP risk counseling include a definition of SUDEP, associated risk factors, the individual patient's risk level, risk mitigation strategies, and referrals to informational and support resources. Despite knowing that uncertainty exists, participants expressed the importance of naming the uncertainty and the limitations of current knowledge on the mechanisms behind SUDEP. A universal concern among clinicians was the fear of imposing undue anxiety on caregivers and patients by bringing up an unlikely event. However, recent findings suggest that SUDEP risk disclosures do not yield adverse long-term effects on well-being or quality of life and instead may lead to better health outcomes for patients with epilepsy by improving treatment adherence.[6-8] Critically, both the literature and anecdotes from our caregiver focus groups suggest that families are likely to stumble across the term SUDEP through outside sources.[15,25,32] If clinicians proactively initiate discussions about SUDEP, caregivers may be comforted in knowing that they can trust in the accuracy of the information they are receiving.[11,25,34]

It is well-established that patients and families feel more supported and comforted when clinicians take the time to speak to them with *Empathy* and validate their emotional responses when delivering difficult news.[31,35,36] It is important to name the emotions that are present, assess and explore those emotions, and help validate their concerns. Incentivizing clinicians who interact with patients with epilepsy to participate in evidence-based, empathy-building trainings such as VitalTalk can complement efforts to increase the quality of communication about SUDEP via use of the SPIKES protocol.[37-39] Furthermore, to mitigate the burden of time as a barrier to SUDEP counseling, clinicians can introduce SUDEP alongside other risks of premature mortality and epilepsy and offer families resources such as handouts, web sites, and peer support referrals for access to information and support to access after the visit.[34,40,41]

In the *Summarize* stage, the information relayed to the patient and/or caregiver is recapitulated using lay language and a plan is presented for next steps, including referrals to further resources, as some caregivers noted a lack of age-appropriate SUDEP resources, particularly for young children. An important and reflective question can include "*We've reviewed a lot of information today*; *can you tell me what you understand about SUDEP and your child's risk?"*

This study's findings should be considered in the context of its limitations. The cohort of study participants was small, and caregivers who participated in this study are not representative of the diverse population of people who care for children with epilepsy. Participant demographics including primary language and education level were not collected and may have influenced participant communication preferences. Furthermore, the seizure burden of the children of caregiver participants was relatively high, suggesting a higher risk of SUDEP compared with the general population of patients with epilepsy. Caregivers in our study may have received SUDEP counseling to a greater extent than the average caregiver, and their expressed interest in learning about SUDEP from clinicians may be higher than that in caregivers whose children have lower seizure burdens. Further investigation is needed to characterize how patient risk level impacts SUDEP communication preferences, particularly with regard in unique epilepsy syndromes. Similarly, recruiting through conferences, academic institutions, and professional societies may have biased our results to reflect the views of clinicians with a higher baseline knowledge about SUDEP and engagement in the epilepsy community than the general population of child neurology clinicians. Data regarding differences among practice setting, including time allotted for clinic visits, interpreter services, and the availability of written epilepsy educational materials were also not collected but may impact communication experiences and preferences.

Conclusion

Caregivers and clinicians emphasized the value of SUDEP risk counseling in the pediatric clinical setting. Based on these findings, neurologists and/or epileptologists caring for children with epilepsy should educate caregivers about SUDEP soon after the initial epilepsy diagnosis in an empathetic manner, revisiting the conversation longitudinally over time and highlighting actionable steps to mitigate risk. SUDEP risk disclosure should be standardized as part of routine care for people with epilepsy and tailored to match the unique risk profiles, emotional and informational needs, and baseline understanding of patients and families. Despite a well-established need, few communication tools exist to facilitate effective caregiver-clinician conversations about SUDEP. Here, caregiver and clinician preferences informed our novel adaptation of the SPIKES protocol for use by pediatric neurologists in the context of SUDEP risk disclosure. Although the SUDEP-specific SPIKES tool is structured with key phases of patient-centered communication, the framework acknowledges that each step is iterative and may need to be returned to over the course of the conversation. Future work will assess the feasibility and acceptability of this protocol as a mechanism to enhance communication quality and understanding about SUDEP; this could be done by assessing patient and provider satisfaction as a follow-up to assess the validity of the results.

CRediT authorship contribution statement

Kayli Maney: Writing – review & editing, Writing – original draft, Investigation, Formal analysis, Data curation. **Isabella K. Pallotto:** Writing – review & editing, Writing – original draft, Data curation. **Simran Bansal:** Writing – review & editing, Writing – original draft, Data curation. **Shital Patel:** Writing – review & editing, Writing – original draft. **Renée A. Shellhaas:** Writing – review & editing, Writing – original draft, Conceptualization. **Zachary M. Grinspan:** Writing – review & editing, Writing – original draft, Conceptualization. **Jeffrey Buchhalter:** Writing – review & editing, Writing – original draft, Conceptualization. **Elizabeth J. Donner:** Writing – review & editing, Writing – original draft, Conceptualization. **Gardiner Lapham:** Writing – review & editing, Writing – original draft, Conceptualization. **Thomas Stanton:** Writing – review & editing, Writing – original draft, Conceptualization. **Monica E. Lemmon:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

Declaration of Competing Interest

The following authors declare the following financial or non-financial interests which may be considered as potential conflicts of interest: Dr. Shellhaas serves as a consultant for the Epilepsy Study Consortium and receives royalties from UpToDate for authorship of topics related to neonatal seizures and a stipend from the Pediatric Epilepsy Research Foundation for service as president-elect. Her research is funded by NIH. Dr. Lemmon receives salary support from the NIH. Gardiner Lapham is a trustee of the BAND foundation. Apart from above, the authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this article.

References

1. Beghi E. The epidemiology of epilepsy. Neuroepidemiology. 2020;54:185–191.
2. Whitney R, Donner EJ. Risk factors for sudden unexpected death in epilepsy (SUDEP) and their mitigation. Curr Treat Options Neurol. 2019;21:7.
3. Thurman DJ, Hesdorffer DC, French JA. Sudden unexpected death in epilepsy: assessing the public health burden. Epilepsia. 2014;55:1479–1485.
4. Devinsky O, Spruill T, Thurman D, Friedman D. Recognizing and preventing epilepsy-related mortality: a call for action. Neurology. 2016;86:779–786.
5. Devinsky O, Friedman D, Cheng JY, Moffatt E, Kim A, Tseng ZH. Underestimation of sudden deaths among patients with seizures and epilepsy. Neurology. 2017;89:886–892.
6. Gayatri NA, Morrall MC, Jain V, Kashyape P, Pysden K, Ferrie C. Parental and physician beliefs regarding the provision and content of written sudden unexpected death in epilepsy (SUDEP) information. Epilepsia. 2010;51:777–782.
7. Collard SS, Regmi P. Qualitative insights into the feelings, knowledge, and impact of SUDEP: a narrative synthesis. Epilepsy Behav. 2019;94:20–28.
8. Radhakrishnan DM, Ramanujam B, Srivastava P, Dash D, Tripathi M. Effect of providing sudden unexpected death in epilepsy (SUDEP) information to persons with epilepsy (PWE) and their caregivers-experience from a tertiary care hospital. Acta Neurol Scand. 2018;138:417–424.
9. Barbour K, Hesdorffer DC, Tian N, et al. Automated detection of sudden unexpected death in epilepsy risk factors in electronic medical records using natural language processing. Epilepsia. 2019;60:1209–1220.
10. Harden C, Tomson T, Gloss D, et al. Practice guideline summary: sudden unexpected death in epilepsy incidence rates and risk factors: report of the guideline development, dissemination, and implementation Subcommittee of the American Academy of Neurology and the American Epilepsy Society. Neurology. 2017;88:1674–1680.
11. Louik J, Doumlele K, Hussain F, et al. Experiences with premorbid SUDEP discussion among participants in the North American SUDEP Registry (NASR). Epilepsy Behav. 2017;70:131–134.
12. Asadi-Pooya AA, Trinka E, Brigo F, et al. Counseling about sudden unexpected death in epilepsy (SUDEP): a global survey of neurologists' opinions. Epilepsy Behav. 2022;128:108570.
13. Long L, Cotterman-Hart S, Shelby J. To reveal or conceal? Adult patient perspectives on SUDEP disclosure. Epilepsy Behav. 2018;86:79–84.
14. Nisbet T, Turbull S, Mulhern S, Razvi S. 'Breaking Good news': neurologists' experiences of discussing SUDEP with patients in Scotland. Epilepsy Behav. 2017;70:72–79.
15. Ramachandrannair R, Jack SM, Meaney BF, Ronen GM. SUDEP: what do parents want to know? Epilepsy Behav. 2013;29:560–564.
16. Baile WF, Buckman R, Lenzi R, Glober G, Beale EA, Kudelka AP. SPIKES—a six-step protocol for delivering bad news: application to the patient with cancer. Oncologist. 2000;5:302–311.
17. The SPIKES Strategy. Patient Brochure. Pamphlet. Child Neurology Foundation. Available at: https://www.childneurologyfoundation.org/wp-content/uploads/2017/05/SPIKES_Pocket_Folder_Handout.pdf; 2021. Accessed September 30, 2022.
18. Milton AC, Mullan B. Views and experience of communication when receiving a serious mental health diagnosis: satisfaction levels, communication preferences, and acceptability of the SPIKES protocol. J Ment Health. 2017;26:395–404.
19. Mirza RD, Ren M, Agarwal A, Guyatt GH. Assessing patient perspectives on receiving bad news: a survey of 1337 patients with life-changing diagnoses. AJOB Empir Bioeth. 2019;10:36–43.
20. Fukui S, Ogawa K, Yamagishi A. Effectiveness of communication skills training of nurses on the quality of life and satisfaction with healthcare professionals among newly diagnosed cancer patients: a preliminary study. Psychooncology. 2011;20:1285–1291.
21. Shaw DJ, Davidson JE, Smilde RI, Sondoozi T, Agan D. Multidisciplinary team training to enhance family communication in the ICU. Crit Care Med. 2014;42:265–271.
22. Wolfe AD, Frierdich SA, Wish J, Kilgore-Carlin J, Plotkin JA, Hoover-Regan M. Sharing life-altering information: development of pediatric hospital guidelines and team training. J Palliat Med. 2014;17:1011–1018.
23. Pallotto IK, Shellhaas RA, Maney K, et al. Communication about sudden unexpected death in epilepsy: understanding the caregiver perspective. Ann Child Neurol Soc. 2023;1:66–74.
24. Hamilton AB, Finley EP. Qualitative methods in implementation research: an introduction. Psychiatry Res. 2019;280, 112516.
25. Ramachandran Nair R, Jack SM, Strohm S. SUDEP: to discuss or not? Recommendations from bereaved relatives. Epilepsy Behav. 2016;56:20–25.
26. Xu Z, Ayyappan S, Seneviratne U. Sudden unexpected death in epilepsy (SUDEP): what do patients think? Epilepsy Behav. 2015;42:29–34.
27. Barbour K, Yozawitz EG, McGoldrick PE, Wolf S, Nelson A, Grinspan ZM. Predictors of SUDEP counseling and implications for designing interventions. Epilepsy Behav. 2021;117:107828.
28. Grout RW, Buchhalter J, Patel AD, et al. Improving patient-centered communication about sudden unexpected death in epilepsy through computerized clinical decision support. Appl Clin Inform. 2021;12:90–99.
29. von Blanckenburg P, Hofmann M, Rief W, Seifart U, Seifart C. Assessing patients' preferences for breaking bad news according to the SPIKES-Protocol: the MABBAN scale. Patient Educ Counsel. 2020;103:1623–1629.
30. Lewis S, Higgins S, Goodwin M. Informing patients about sudden unexpected death in epilepsy: a survey of specialist nurses. Br J Neurosci Nurs. 2008;4:30–34.
31. McColl-Kennedy JR, Danaher TS, Gallan AS, Orsingher C, Lervik-Olsen L, Verma R. How do you feel today? Managing patient emotions during health care experiences to enhance well-being. J Bus Res. 2017;79:247–259.
32. Brigo F, Igwe SC, Ausserer H, et al. Why do people Google epilepsy?: an infodemiological study of online behavior for epilepsy-related search terms. Epilepsy Behav. 2014;31:67–70.
33. Jabbour D, Masri JE, Nawfal R, Malaeb D, Salameh P. Social media medical misinformation: impact on mental health and vaccination decision among university students. Ir J Med Sci. 2022;192:291–301.
34. Mesraoua B, Tomson T, Brodie M, Asadi-Pooya AA. Sudden unexpected death in epilepsy (SUDEP): definition, epidemiology, and significance of education. Epilepsy Behav. 2022;132, 108742.
35. Zwingmann J, Baile WF, Schmier JW, Bernhard J, Keller M. Effects of patient-centered communication on anxiety, negative affect, and trust in the physician in delivering a cancer diagnosis: a randomized, experimental study. Cancer. 2017;123:3167–3175.
36. Kasat K, Stoffels G, Ellington M. Improving communication with parents: the neonatal intensive care unit empathy workshop. J Perinatol. 2020;40:1423–1432.
37. Back AL, Arnold RM, Baile WF, et al. Efficacy of communication skills training for giving bad news and discussing transitions to palliative care. Arch Intern Med. 2007;167:453–460.
38. Bays AM, Engelberg RA, Back AL, et al. Interprofessional communication skills training for serious illness: evaluation of a small-group, simulated patient intervention. J Palliat Med. 2014;17:159–166.
39. Feudtner C. Empathy in action. Pediatrics. 2020;145:e20193116.
40. Leach JP. SUDEP discussions with patients and families. Pract Neurol. 2012;12:103.
41. Shankar R, Ashby S, McLean B, Newman C. Bridging the gap of risk communication and management using the SUDEP and seizure safety checklist. Epilepsy Behav. 2020;103:106419.