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Communication about sudden unexpected death in epilepsy: Understanding the caregiver perspective

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Abstract

Objective: We aimed to characterize (1) the caregiver experience of learning about sudden unexpected death in epilepsy (SUDEP), and (2) caregiver preferences for SUDEP risk disclosure. **Methods:** We distributed a 24-question survey to caregivers of children with epilepsy. Free text questions were analyzed using a rapid qualitative analysis approach. **Results:** Two hundred and twelve caregivers of people with epilepsy completed the survey, including 12 bereaved caregivers. Caregivers' children had a high seizure burden, with a median seizure frequency of 24 seizures per year (range: 1 to ≥100). Most participants were aware of SUDEP at the time of the survey (193/212; 91%) though only a minority had learned about SUDEP from a healthcare provider (91/193; 47.2%). Caregivers typically learned about SUDEP from a nonprofit or online source (91/161; 56.5%). Almost all caregivers wanted to discuss SUDEP with their child's healthcare provider (209/212; 98.6%), and preferred disclosure from epileptologists (193/212; 91%), neurologists (191/212; 90.1%), and/or primary care providers (98/212; 46.2%). In open-ended responses, caregivers highlighted the value of learning about SUDEP from a healthcare provider, the importance of pairing SUDEP risk disclosure with a discussion of how to mitigate risk, and the need for educational resources and peer support. **Interpretation:** Caregivers of people with epilepsy appreciate when healthcare providers disclose information about SUDEP, yet typically hear about SUDEP elsewhere. These findings underscore the importance of interventions to improve and support SUDEP risk disclosure. Future work should evaluate strategies to disclose SUDEP risk and the impact of early SUDEP risk disclosure.

Introduction

Sudden unexpected death in epilepsy (SUDEP) is one of the most common causes of premature death in children and adults with epilepsy.[1] Current estimates of SUDEP incidence are approximately 1 in 1000 people with epilepsy annually in pediatric and adult populations, with a higher incidence in individuals of low socioeconomic status.[1–5] SUDEP incidence rates are likely underestimates due to misattributed causes of death, especially in underserved populations.[6–10] Despite SUDEP's relatively low incidence compared to death by other neurologic causes, it is second only to stroke in the number of life years lost[1] because SUDEP affects people across the age spectrum unlike other neurologic diseases such as Alzheimer's, Parkinson's, and amyotrophic lateral sclerosis, which typically affect individuals toward the end of life.[11]

The greatest risk factor for SUDEP is generalized tonic-clonic seizures, especially those occurring at night.[11] Although nocturnal tonic-clonic seizures are relatively common among people with epilepsy, a high percentage of patients and family members have not been educated on this risk of SUDEP.[11] The 2017 American Academy of Neurology/American Epilepsy Society (AAN/AES) guideline recommends that doctors inform their patients about the risk of SUDEP at or soon after epilepsy diagnosis.[1,2] Most patients and families expect their neurologists to share information about SUDEP risk.[12]

However, this counseling may not occur regularly. In one study of bereaved family members, respondents described the need for physician counseling around SUDEP risk factors, but less than 20% of family members recalled having a conversation about SUDEP with their child's clinician prior to their loved one's death.[13] A survey of Canadian and American neurologists revealed that only 6.8% had discussed SUDEP with nearly all of their patients with epilepsy.[11] Clinicians cited barriers including a perceived risk of worsening patient anxiety,[3,12] lack of personal knowledge regarding the risk,[3,14] lack of conceptualized risk due to its low incidence, and time limitations.[3,13] Additional barriers included a lack of clear guidance or training on how to engage in a conversation about SUDEP risk.[3,15] These barriers may be amplified in pediatric settings, in which clinicians must navigate both caregivers and patients at varying developmental levels.

To better facilitate conversations about SUDEP with caregivers of people with epilepsy, we must first understand caregiver preferences regarding when, how, and from whom they prefer to learn about SUDEP risk. In this study, we aimed to characterize caregiver preferences for SUDEP risk disclosure.

Methods

In this prospective cross-sectional study, we disseminated a 24-question survey (20 close-ended, 4 free texts; Supporting Information: Appendix) to caregivers of people with epilepsy from October to November 2021. This survey was developed by the study team and informed by existing literature around SUDEP risk factors and communication preferences.[3,16] A stakeholder advisory committee, inclusive of epileptologists, advocates, and caregivers, advised on study design and survey content. Caregivers completed the survey through REDCap, a secure web application for managing online surveys.[17] The survey assessed caregivers' knowledge of SUDEP (how they learned about SUDEP), communication preferences about SUDEP, and demographic information. Skip logic was presented throughout the survey to ensure questions were relevant to each individual. For example, caregivers who indicated the death of a loved one due to SUDEP were not presented with questions related to current child demographic variables, in an effort to mitigate distress.

Community partners and advocacy organizations posted a link to the survey in their membership groups. Participating organizations included the Child Neurology Foundation, Danny Did Foundation, Hope for hypoxic ischemic encephalopathy (HIE), and Epilepsy Foundation.

Close-ended questions were analyzed using SPSS Statistics for Windows, Version 24.0 (IBM). For close-ended questions with a free text option (e.g., How did you first learn of SUDEP? Please specify "Other" source), we derived categories from the responses. Free text questions were coded using rapid-cycle qualitative analysis.[18] While a variety of rapid assessment approaches have been described, we adopted the approach outlined by Hamilton.[19] In rapid-cycle analysis, the text is summarized and organized using a matrix. Two independent coders (I. K. P., J. K. D.) evaluated responses to each free text question prompt and used a structured summary document to aggregate responses. Themes were inductively derived within and across questions in consensus with the senior author (M. E. L.).

The protocol was approved by the Duke University Institutional Review Board. The first page of the survey included study information and consent to participate in the study.

Results

Participant characteristics

A total of 318 participants opened the survey; of these, 212 participants completed the survey in full and were the basis for further analysis. Participants who did not identify as parents or caregivers of a child (e.g., caregiver of spouse) with epilepsy were excluded ($n = 2$). The majority of participants identified as White ($n = 194/212$, 94.2%), with a mean age of 42 years of age (range: 18–72 years). Some participants reported the death of a child due to SUDEP ($n = 12/212$, 5.7%). The majority of participants lived in the United States ($n = 174/212$, 84.5%), and other common countries represented included the United Kingdom ($n = 19$) and Canada ($n = 5$) (Table 1).

Most participants had children with a high seizure burden, with a median of 24 seizures per year. Over half of the participants ($n = 106/181$, 58.6%) reported a history of generalized tonic-clonic seizures, and over one-third reported nocturnal generalized tonic-clonic seizures ($n = 73/181$, 40.3%) (Table 2).

The majority of caregivers ($n = 193/212$, 91.0%) had at least some knowledge of SUDEP before taking the survey. A minority of caregivers ($n = 19/212$, 9.0%), however,

Table 1. Demographic characteristics of caregivers.

	M (SD) *N* = 205	Min–max *N* = 205	*n*/*N* (%) *N* = 212[a]
Age (years)	42.37 (9.88)	18–72	
Not reported			7 (3.3)
Race			
White			194 (94.2)
Mixed			4 (1.9)
Asian			2 (1.0)
Black or African American			1 (0.5)
Prefer not to answer			5 (2.4)
Not reported			6 (2.8)
Ethnicity			
Non-Hispanic			186 (93.5)
Hispanic			13 (6.5)
Not reported			13 (6.1)
Bereaved			12 (5.7)
US resident			174 (84.5)
Not reported			5 (2.4)
Region of residence in United States			
Midwest			50/174 (28.7)
South			50/174 (28.7)
West			37/174 (21.3)
Northeast			25/174 (14.4)
Not reported			12/174 (6.9)

[a] Unless otherwise reported.

Table 2. Demographic and epilepsy characteristics of caregivers' children.

	Median	Min–max	*n* (%) *N* = 181[a]
Age (years)	10.50	0–45	
Not reported			5 (2.8)
Number of seizures			
Per month	3.00	1–100	
Not reported			8 (4.4)
Per year	24.00	1–100	
Not reported			12 (6.6)
Presence of >1 seizure per month			96 (53.0)
Not reported			8 (4.4)
Presence of tonic-clonic seizures			106 (58.6)
Not reported			6 (3.3)
Number per year	4.00	1–100	
Not reported			4/106 (3.8)
Presence of nocturnal seizures			123 (68.0)
Not reported			5 (2.8)
Tonic-clonic			73 (40.3)
Not reported			6 (3.3)
Number of prescribed seizure medications	2.00	0–6	
Not reported			8 (4.4)

Of the 212 caregivers, 31 caregivers were not shown child demographic questions due to skip logic. Of the 181 caregivers shown the child demographic questions, 5 caregivers chose not to answer any child demographic questions. Percentages may not add to 100 due to skip logic.
[a] Unless otherwise reported.

learned about SUDEP from the survey itself. Of those caregivers who had some baseline knowledge of SUDEP, a minority (*n* = 31/193, 16.1%) first learned about SUDEP from a healthcare provider. More commonly, caregivers learned about SUDEP from a general online source, such as Google or Facebook (*n* = 49/161, 30.4%), nonprofits (*n* = 41/161, 25.5%), other epilepsy caregivers (*n* = 12/161, 7.5%), or the news (*n* = 10/161, 6.2%) (Table 3). Caregivers learned about SUDEP from nonprofits in a variety of ways, including accessing the nonprofit online at their webpage or a moderated social media group. In open-ended responses, seven of the twelve bereaved caregivers reported that they had not heard of SUDEP until their child's death.

Caregiver preferences for SUDEP risk disclosure

Nearly all participants (*n* = 209/212, 98.6%) shared the desire to have a conversation about SUDEP with their healthcare provider (Table 4). The majority of caregivers reported that SUDEP risk discussions should be led by epileptologists (*n* = 193/212, 91.0%) and/or neurologists (*n* = 191/212, 90.1%). Nearly half of participants (*n* = 98/212, 46.2%) reported wanting to hear about and discuss SUDEP with primary care physicians; this preference was more common for bereaved caregivers (*n* = 10/12, 83.3%) than nonbereaved caregivers (*n* = 88/200, 44.0%). Over one-third of participants reported that nurses and/or nurse practitioners and other caregivers should be engaged to disclose SUDEP risk.

Most participants desired this conversation to occur at the first epilepsy visit (*n* = 140/212, 66%) or with changes in their child's level of risk (*n* = 127/212, 59.9%). Many participants wanted to discuss SUDEP both at the first epilepsy visit and with changes in the child's level of risk (*n* = 90/212, 42.5%). Some caregivers (*n* = 35/212, 16.5%) preferred discussions of SUDEP to occur at each visit. In open-ended responses, some caregivers described the need for conversations to occur away from the child or for the conversation to occur after first asking permission to discuss SUDEP in the child's presence. As shared by one caregiver:

> It is a very uncomfortable topic. It would best be discussed without a young child in the room.

In open-ended responses, caregivers elaborated on the key information to include in conversations about SUDEP. Many caregivers emphasized the importance of describing key risk factors in order to know their child's individual

Table 3. Epilepsy caregivers' knowledge of SUDEP.

	n/N (%)		
	Nonbereaved	Bereaved	Total
SUDEP knowledge before survey			
Some knowledge	181/200 (90.5)	12/12 (100.0)	193/212 (91.0)
No knowledge	19/200 (9.5)	0/12 (0.0)	19/212 (9.0)
Source where initially learned about SUDEP[a]			
Healthcare provider	30/181 (16.7)	1/12 (8.3)	31/193 (16.1)
Neurologist	15/30 (50.0)	-	15/31 (48.4)
Epilepsy specialist	10/30 (33.3)	1/1 (100.0)	11/31 (35.5)
Other provider	3/30 (10.0)	-	3/31 (9.7)
Primary care provider	2/30 (6.7)	-	2/31 (6.5)
Somewhere else	150/181 (83.3)	11/12 (91.7)	161/193 (83.4)
General online source	47/150 (31.3)	2/11 (18.2)	49/161 (30.4)
Nonprofit	39/150 (26.0)	2/11 (18.2)	41/161 (25.5)
Did not report	20/150 (13.3)	-	20/161 (12.4)
Unknown	13/150 (8.7)	-	13/161 (8.1)
Epilepsy caregiver	12/150 (8.0)	-	12/161 (7.5)
News	10/150 (6.7)	-	10/161 (6.2)
Personal experience	3/150 (2.0)	6/11 (54.5)	9/161 (5.6)
Other	6/150 (4.0)	1/11 (9.1)	7/161 (4.3)
Did not report source	1/181 (0.55)	-	1/193 (0.52)

Percentages may not add to 100 due to skip logic.

[a] General online source includes: social media, search engines, unspecified web pages, web pages unaffiliated with nonprofits (e.g., WebMD), and unspecified online groups. Nonprofit includes: any communication from nonprofits, nonprofit websites, and social media pages or online groups moderated by nonprofits. Unknown includes: cannot remember. Other includes: unspecified pamphlet, academic training, word of mouth, and radio commercial.

SUDEP, sudden unexpected death in epilepsy.

Table 4. Guidance for healthcare providers on approaching the SUDEP conversation.

	n/N (%)		
	Nonbereaved *N* = 200	Bereaved *N* = 12	Total *N* = 212
Caregivers who want to have discussion with healthcare provider	197 (98.5)	12 (100)	209 (98.6)
Time of discussion			
First epilepsy visit	131 (65.5)	9 (75.0)	140 (66.0)
Risk level changes	120 (60.0)	7 (58.3)	127 (59.9)
Annually	73 (36.5)	1 (8.3)	74 (34.9)
Every visit	31 (15.5)	4 (33.3)	35 (16.5)
Other	12 (6.0)	3 (25.0)	15 (7.1)
Providers who should discuss			
Epilepsy specialists	181 (90.5)	12 (100)	193 (91.0)
Neurologists	180 (90.0)	11 (91.7)	191 (90.1)
Primary care doctors	88 (44.0)	10 (83.3)	98 (46.2)
Nurses/nurse practitioners	70 (35.0)	8 (66.7)	78 (36.8)
Psychologists	41 (20.5)	8 (66.7)	49 (23.1)
Other caregivers	19 (9.5)	4 (33.3)	23 (10.8)
Other	12 (6.0)	1 (8.3)	13 (6.1)

Percentages may not add to 100 due to the ability to select multiple response options.

SUDEP, sudden unexpected death in epilepsy.

risk for SUDEP, particularly in the context of different diagnoses or seizure types. In the open-ended question "What questions would you recommend other patients and families ask their healthcare providers about SUDEP?," survey participants often asked their own questions about SUDEP such as *what* the risk factors for SUDEP are, indicating a knowledge gap. Caregivers valued when clinicians defined and discussed risk factors for SUDEP alongside counseling around how caregivers could reduce their child's risk. Caregivers appreciated hearing information about SUDEP "warning signs," the ways to lower their child's risk, the importance of medication adherence, and counseling regarding whether monitoring devices could benefit their child.

Caregivers described the need to balance information about SUDEP risk with support from other epilepsy caregivers and the care team. Some parents spoke to the anxiety and fear that can accompany living with SUDEP risk and encouraged clinicians to validate those emotions:

> Do not downplay the fear parents have regarding SUDEP. We are scared, and it is reasonable to feel that way. There is very little we can do to prevent, but awareness is a crucial step in the right direction.

Caregivers characterized how clinicians could provide support by connecting families with resources for additional information and peer support. Caregivers advocated for the provision of written resources; some caregivers specifically asked for resources that were free of jargon, data-driven, and included infographics to aid accessibility. Some caregivers appreciated information on monitoring devices and new research findings. Regarding peer support, caregivers appreciated referrals to parent support groups and local chapters of epilepsy nonprofits.

The value of learning about SUDEP from a healthcare provider

In open-ended responses, caregivers underscored the value of learning about SUDEP from a healthcare provider and that hearing it from a trusted source was less scary and more accurate than the internet. Some caregivers described that they "deserved to know" their child's risk of SUDEP and that understanding information about SUDEP risk was key to understanding their child's care. As shared by one caregiver:

> It's a risk. Just like they discuss side effects of medications. This is a 'side effect' of epilepsy, and everyone affected deserves to know the risks.

Other caregivers directly addressed concerns that discussing SUDEP could exacerbate anxiety or fear as misplaced:

> "Protecting" the caregivers or "not wanting to scare them" does a disservice.

Some caregivers described SUDEP risk disclosure as an element of a trusting therapeutic relationship. As shared by one caregiver:

> Finding out about SUDEP on you[r] own is frightening. It decreases trust in the medical community. If they are unwilling to discuss something life-threatening, why should you trust any recommendations they have?

Bereaved caregivers described stakes associated with not learning about SUDEP while their child was alive. Some caregivers spoke to the potential for decreased guilt or increased meaning-making during their child's life. As shared by one caregiver:

> Perhaps if I was aware SUDEP was a possibility, I would have embraced the time I had with her more fully.

Caregivers with living children also recognized the guilt that could result from their child's death from SUDEP had they never learned of the possibility. As shared by one caregiver:

> It's better to know all the information, and that's the only way we can know that we did the best for our kids to prevent SUDEP. … This way, we know that we did everything we could for our child, even if there is a negative outcome. The worst is feeling like you could have done better for your child if you had only known what to do.

Discussion

In this study, caregivers offer actionable advice to inform SUDEP risk disclosure. Our findings affirm that the overwhelming majority of caregivers of children with epilepsy want to talk about SUDEP with their child's healthcare team. This study builds on existing literature to suggest by whom, when, and how this information should be delivered. Most caregivers described a preference to learn about SUDEP early in their child's course and longitudinally from pediatric neurology and epilepsy clinicians. Despite these preferences, most respondents did not hear about SUDEP from a healthcare professional and, instead, accessed online resources and support. Our findings highlight the need to improve SUDEP risk disclosure counseling in the healthcare setting, as well as an opportunity to leverage online resources and support communities.

The overwhelming majority of caregivers desired to hear about SUDEP from their child's healthcare provider. This affirms existing literature that patients and caregivers appreciate counseling about SUDEP risk.[2,13,20–24] Current barriers to SUDEP risk disclosure include clinician knowledge deficits, perceived risk of exacerbating parent distress, time, and lack of communication skills training.[12–15,25,26] Multiple reports suggest most physicians do not regularly discuss SUDEP with families.[2,3,21,25–27] Many physicians report only discussing SUDEP when a patient has risk factors for SUDEP.[20,25,26] Generally, physicians who have more years in practice, completed an epilepsy fellowship, work in an academic setting, and treat adults with epilepsy are more likely to discuss SUDEP with their patients.[25,26] An analysis of medical records of physicians treating patients with epilepsy at six hospitals in New York found that SUDEP counseling was only documented at two of six studied hospitals, and by only 1% of physicians.[26] Much of these data were collected before the publication of the 2017 AAN/AES practice guidelines that assert that all clinicians should disclose SUDEP to epilepsy patients and their families.[2] These guidelines did increase the frequency with which clinicians discussed SUDEP with epilepsy patients and caregivers; however, our data, in conjunction with data evaluating practice change in response to the 2017 guideline,[3] suggest an ongoing opportunity for improvement in SUDEP risk disclosure.

To reduce barriers to SUDEP risk disclosure, there are ongoing efforts to educate physicians on the SUDEP

Clinical Practice Guideline and interventions using electronic health records (EHR) to promote the disclosure of SUDEP risk. One quantitative pre-post trial of a clinical decision support system used patient-report data collected in the waiting room to provide clinical advice to the clinician using the EHR.[28] This system increased the frequency of SUDEP discussions initiated by primary care providers, suggesting that EHR triggers could be useful in prompting clinicians of many specialties working with epilepsy patients to disclose SUDEP risk.[28] A commonly cited reason for avoiding SUDEP risk counseling is to avoid causing the caregiver undue anxiety for a rare event.[25,27] Acute anxiety is a normative side effect of a new epilepsy diagnosis[29]; however, this concern must be balanced with the ethical imperative to disclose important health information. In one study of adult epilepsy patients, 30% experienced increased fear of epilepsy after learning about SUDEP; however, all participants felt it was their right to be informed about SUDEP.[21] A qualitative study of young adults with epilepsy in Scotland found that any anxiety felt after the disclosure of SUDEP was not long-lasting.[24] Importantly, while clinicians may worry that disclosing information about SUDEP worsens caregiver anxiety, our findings suggest that caregivers are likely to encounter information about SUDEP regardless of clinician counseling; ensuring that information about SUDEP risk comes first from a trusted healthcare provider may mitigate parent distress.[22] Additionally, caregivers emphasized that SUDEP risk discussions fostered trust in the doctor–patient relationship.

Caregivers offered advice about the logistics of SUDEP risk disclosure, including preferences to hear about SUDEP at epilepsy diagnosis and longitudinally with changes in risk. These preferences underscore findings from existing literature.[20,22,23] Systems that prompt clinicians to evaluate and discuss SUDEP risk may aid counseling.[28] Not surprisingly, parents wished for neurology clinicians to share this information; however, approximately half desired to hear information directly from their primary care physician. This finding suggests that interventions to improve SUDEP risk disclosure should target primary care physicians in addition to neurology providers, especially since access to epileptologists is variable among settings. Primary care providers often serve as front-line clinicians for children with epilepsy, since access can limit their ability to refer to a specialist.[30] However, primary care providers may not have adequate training in SUDEP risk or may assume that a neurologist or epileptologist treating the patient's epilepsy will discuss SUDEP with caregivers upon referral.[31] Taken together, our findings and existing data highlight the need for interventions about SUDEP risk disclosure to include primary care providers.

Caregivers additionally offered advice on what information to include in SUDEP risk counseling, including a thorough discussion of risk factors, how caregivers could mitigate risk, and associated resources and support. Important risk factors for SUDEP include generalized tonic-clonic seizures, nocturnal seizures, and uncontrolled seizures.[1,23] Clinicians can emphasize the importance of medication adherence and avoidance of seizure triggers in risk reduction. Some studies indicate that patients who receive SUDEP counseling are more adherent to medication regimens.[21,32] Many caregivers in our sample discussed the value of monitoring devices, a desire that aligns with the concept of early intervention, based on the belief that cases of witnessed SUDEP are rare.[33] While data on the value of these devices mixed and evolving, clinicians should engage parents directly in a conversation about the potential harms and benefits of monitoring.[34–36] This finding acknowledges a gap in the literature around if, when, and how to utilize monitoring devices as a strategy to reduce SUDEP risk. Some caregivers acknowledged the need for clinicians to validate caregiver emotions. Naming and validating emotions is a well-documented strategy for effectively communicating challenging information.[37] Since SUDEP is only one cause of premature mortality in epilepsy, counseling around SUDEP risk need not be isolated. Clinicians commonly cite limited time for appointments as a barrier to SUDEP risk disclosure.[3] Leveraging ways to incorporate SUDEP counseling into well-established parts of the visit may decrease the potential time burden and improve clinician disclosure. As a caregiver in this cohort offered, SUDEP counseling may most naturally fall amidst discussion of epilepsy risks more broadly. Future work should study how the incorporation of SUDEP risk disclosure with the discussion of other risks of premature mortality in epilepsy impacts the frequency of SUDEP risk disclosure by clinicians, time spent counseling the patient and family, and patient and caregiver satisfaction.

Limitations of this study include the racial/ethnic homogeneity of the sample, highlighting that our social media-driven recruitment strategy did not yield a representative sample of caregivers of children with epilepsy. Alternatively, our sample's racial and ethnic homogeneity may reflect less engagement in advocacy groups by individuals with minoritized identities.[38] Data suggest that systemic barriers contribute to reduced rates of seizure remission and medication adherence in racial and ethnic minoritized children; it is unknown whether SUDEP risk disclosure occurs less often in these populations as well.[39–41] It is important for future work to target these underrepresented voices, especially in the context of the high burden of disease and likely underreporting of SUDEP in underserved populations.[9,10] Some people living with epilepsy reside in congregant settings; their experiences may be underrepresented here. Worth noting, our survey had a relatively equal geographic distribution of responses, which may indicate that perspectives from parents from both well-resourced and under-resourced regions were included. Child seizure burden was limited to self-report and was high in comparison to the general population of children with epilepsy. Our sample is likely at higher risk for SUDEP than the general population of children with epilepsy; opinions expressed by caregivers may reflect this. The high yield and recruitment strategy through parent groups of our survey have the potential to bias our results toward parents who are highly engaged in the epilepsy community and, therefore, wish for a high level of information from and communication with clinicians. Our methodology risks recall bias. While our design and analytic strategy offer a broad view of caregiver preferences, caregiver interviews and/or focus groups may yield more nuanced data.

Conclusions

Caregivers in this cohort overwhelmingly valued learning about SUDEP risk from a healthcare provider. Caregivers appreciated disclosure by epilepsy and/or neurology clinicians, the opportunity to revisit SUDEP risk longitudinally over time, and when clinicians discussed mitigation strategies alongside risk. Future work should evaluate strategies to disclose SUDEP risk and the impact of early SUDEP risk disclosure on the therapeutic alliance, medication adherence, and patient and caregiver well-being.

Author Contributions

Isabella K. Pallotto: Formal analysis; project administration; writing – original draft. **Renée A. Shellhaas**: Conceptualization; writing – review and editing. **Kayli Maney**: Formal analysis; writing – original draft. **Madelyn Milazzo**: Conceptualization; writing – review and editing. **Zachary Grinspan**: Conceptualization; writing – review and editing. **Jeffrey Buchhalter**: Conceptualization; writing – review and editing. **Elizabeth J. Donner**: Conceptualization; writing – review and editing. **Gardiner Lapham**: Conceptualization; writing – review and editing. **Thomas Stanton**: Conceptualization; writing – review and editing. **J. Kelly Davis**: Formal analysis; project administration; writing – review and editing. **Monica E. Lemmon**: Conceptualization; formal analysis; supervision; writing – review and editing.

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Conflicts of Interest

Dr. Shellhaas receives a stipend from the American Academy of Neurology for service as an associate editor for *Neurology*, is a consultant for the Epilepsy Study Consortium, and receives royalties from UpToDate for authorship of topics related to neonatal seizures. Further, she is a member of the Board of Directors for the Pediatric Epilepsy Research Foundation. Her research is supported by grants from NIH. Dr. Buchhalter receives compensation for serving as a consultant to the Epilepsy Foundation, Epilepsy Study Consortium, Epilog, Neurocrine Inc., and UCB. He provides consultation services without compensation to the American Academy of Neurology and Partners Against Mortality in Epilepsy. Dr. Lemmon's research is supported by grants from NIH and the Doris Duke Charitable Foundation.

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References

1. Whitney R, Donner EJ. Risk factors for sudden unexpected death in epilepsy (SUDEP) and their mitigation. *Curr Treat Options Neurol*. 2019;21(2):7.
2. 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(17):1674-1680.
3. Keller AE, Whitney R, Donner EJ. Why child neurologists talk about SUDEP: results from two cross-sectional surveys. *Epilepsia Open*. 2021;6(1):195-205.
4. Cihan E, Hesdorffer DC, Brandsoy M, et al. Socioeconomic disparities in SUDEP in the US. *Neurology*. 2020;94(24):e2555-e2566.
5. Blank LJ. Socioeconomic disparities in epilepsy care. *Curr Opin Neurol*. 2022;35(2):169-174.
6. 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(9):886-892.
7. Ryvlin P, Cascino GD. Sudden unexpected death in epilepsy patients is often misdiagnosed as sudden cardiac death. *Neurology*. 2017;89(9):878-879.

8. Zhuo L, Zhang Y, Zielke HR, et al. Sudden unexpected death in epilepsy: evaluation of forensic autopsy cases. *Forensic Sci Int*. 2012;223(1-3):171-175.
9. Burneo JG, Jette N, Theodore W, et al. Disparities in epilepsy: report of a systematic review by the North American Commission of the International League Against Epilepsy. *Epilepsia*. 2009;50(10):2285-2295.
10. Verducci C, Hussain F, Donner E, et al. SUDEP in the North American SUDEP Registry: the full spectrum of epilepsies. *Neurology*. 2019;93(3):e227-e236.
11. Marin Collazo IV, Tatum WO. Sudden unexpected death in epilepsy (SUDEP): are all your patients informed? *Neurologist*. 2016;21(4):66-71.
12. Gayatri NA, Morrall MCHJ, 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: parental beliefs regarding SUDEP information. *Epilepsia*. 2010;51(5):777-782.
13. 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.
14. Elmali AD, Bebek N, Baykan B. Let's talk SUDEP. *Noro Psikiyatr Ars*. 2019;56(4):292-301.
15. So EL, Bainbridge J, Buchhalter JR, et al. Report of the American Epilepsy Society and the Epilepsy Foundation joint task force on sudden unexplained death in epilepsy. *Epilepsia*. 2009;50(4):917-922.
16. Donner EJ, Waddell B, Osland K, et al. After sudden unexpected death in epilepsy: lessons learned and the road forward. *Epilepsia*. 2016;57:46-53.
17. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)—a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform*. 2009;42(2):377-381.
18. Hamilton AB, Finley EP. Reprint of: qualitative methods in implementation research: an introduction. *Psychiatry Res*. 2020;283:112629.
19. Hamilton AB. Rapid qualitative methods in health services research: spotlight on women's health; 2013.
20. Cooper K, Kirkpatrick P, Brand C, Rolfe A, Florida-James S. Discussing sudden unexpected death in epilepsy with children and young people with epilepsy and their parents/carers: a mixed methods systematic review. *Seizure*. 2020;78:159-167.
21. Long L, Cotterman-Hart S, Shelby J. To reveal or conceal? Adult patient perspectives on SUDEP disclosure. *Epilepsy Behav*. 2018;86:79-84.
22. Ramachandrannair R, Jack SM, Meaney BF, Ronen GM. SUDEP: what do parents want to know? *Epilepsy Behav*. 2013;29(3):560-564.
23. Ramachandrannair R, Jack SM, Strohm S. SUDEP: to discuss or not? Recommendations from bereaved relatives. *Epilepsy Behav*. 2016;56:20-25.
24. Tonberg A, Harden J, McLellan A, Chin RFM, Duncan S. A qualitative study of the reactions of young adults with epilepsy to SUDEP disclosure, perceptions of risks, views on the timing of disclosure, and behavioural change. *Epilepsy Behav*. 2015;42:98-106.
25. 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.
26. 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.
27. Brodie MJ, Holmes GL. Should all patients be told about sudden unexpected death in epilepsy (SUDEP)? Pros and cons. *Epilepsia*. 2008;49:99-101.
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(01):90-99.
29. Jones C, Reilly C. Parental anxiety in childhood epilepsy: a systematic review. *Epilepsia*. 2016;57(4):529-537.
30. Kobau R, Zack MM, Sapkota S, Sajatovic M, Kiriakopoulos E. When and why US primary care providers do and do not refer their patients with new-onset seizures or existing epilepsy or seizure disorders to neurologists—2018 DocStyles. *Epilepsy Behav*. 2021;125:108385.
31. Berl MM, Goodkin HP, Kroner BL, Bumbut A, Lapham G, Gaillard WD. Sudden death in epilepsy: knowledge among pediatric providers. *J Pediatr*. 2017;188:291-293.
32. 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(5):417-424.
33. Pack AM. Sudden unexpected death in the epilepsy monitoring unit. *Epilepsy Curr*. 2014;14(2):78-80.
34. Brinkmann BH, Karoly PJ, Nurse ES, et al. Seizure diaries and forecasting with wearables: epilepsy monitoring outside the clinic. *Front Neurol*. 2021;12:690404.
35. Bruno E, Viana PF, Sperling MR, Richardson MP. Seizure detection at home: do devices on the market match the needs of people living with epilepsy and their caregivers? *Epilepsia*. 2020;61:S11-S24.
36. Shum J, Friedman D. Commercially available seizure detection devices: a systematic review. *J Neurol Sci*. 2021;428:117611.
37. Back AL, Arnold RM, Baile WF, Tulsky JA, Fryer-Edwards K. Approaching difficult communication tasks in oncology. *CA Cancer J Clin*. 2005;55(3):164-177.

38. Zemore SE, Gilbert PA, Pinedo M, Tsutsumi S, McGeough B, Dickerson DL. Racial/ethnic disparities in mutual help group participation for substance use problems. *Alcohol Res*. 2021;41(1):03.
39. Bautista RED, Graham C, Mukardamwala S. Health disparities in medication adherence between African-Americans and Caucasians with epilepsy. *Epilepsy Behav*. 2011;22(3):495-498.
40. Gutierrez-Colina AM, Wetter SE, Mara CA, et al. Racial disparities in medication adherence barriers: pediatric epilepsy as an exemplar. *J Pediatr Psychol*. 2022; jsac001.
41. Gregerson CHY, Bakian AV, Wilkes J, et al. Disparities in pediatric epilepsy remission are associated with race and ethnicity. *J Child Neurol*. 2019;34(14):928-936.

Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article.