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Psychosocial impact on siblings of patients with developmental and epileptic encephalopathies

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ABSTRACT

Objective: Caring for children with developmental and epileptic encephalopathies (DEEs) places substantial demands on the entire family unit, including siblings. The Sibling Voices Survey assesses parental and sibling responses to questions designed to assess how children adapt to growing up with siblings with DEE.
Methods: Participants responded to 1 of 4 online, age- and role-specific surveys (9–12, 13–17, and ≥18-year-old [adult] siblings; parents responded with perceptions of their unaffected child's/children's feelings). Survey questions used visual analog scales, categorical responses, and free-form responses.
Results: Survey submissions (n = 248) included 128 parents and 120 siblings (9- to 12-year-olds, n = 24; 13- to 17-year-olds, n = 17; adults, n = 79). All groups identified home life as the most substantially affected area of their lives (71%–84%), compared with interactions at school (21%–32%) or with friends (28%–42%). The most difficult aspect across all sibling groups was "feeling worried/scared when their sibling has seizures" (58%–70%). Feeling "overly responsible" for the sibling was reported by most adult siblings (63%), 41% of 13- to 17-year-old siblings, and 34% of parents. Siblings reported more symptoms of depressed mood (e.g., "down/unhappy," 47%–62%) than their parents perceived them feeling (25%). Most sibling groups (29%–49%) reported more symptoms of anxious mood (e.g., "nightmares/bad dreams") than parents perceived (15%). Identification of potential helpful coping mechanisms varied by age group. Most respondents (68%–76%) reported positive aspects, including greater maturity and compassion.
Significance: The Sibling Voices Survey provided important insights into how DEE impacts siblings psychologically and socially. This study highlights the need for increased awareness among parents and healthcare providers to monitor siblings for potential signs of depressed or anxious mood, to provide proper support, and to decrease potential for negative long-term consequences.

1. Introduction

Severe developmental and epileptic encephalopathies (DEEs; e.g., Dravet syndrome [DS], Lennox–Gastaut syndrome [LGS]) are associated with severe, frequent, and refractory seizures, as well as developmental delay, neurocognitive impairment, and motor deficiencies, with elevated risk for sudden unexpected death in epilepsy (SUDEP) [1–5]. Patients often require long-term care [6,7]. Comprehensive care of patients with DEE places substantial humanistic and economic strain on caregivers and their families [8–10]. Patient care often involves all family members, including siblings. Siblings may attend medical visits, therapy sessions, and community events geared toward helping the affected individual function in everyday life. The impact on siblings is becoming increasingly apparent to parents/caregivers, patient advocacy groups (PAGs), and the broader healthcare community [10,11]. Siblings are attuned to their parents' worry, fear, and stress over their sibling's condition, often without access to emotional support [10,12]. Unequal attention given to the child with DEE, even if unintentional by parents, may also lead to feelings of isolation and alienation in siblings [10,12,13]. Growing up with a sibling having a chronic illness may adversely affect the transition from childhood to adulthood in unaffected siblings, potentially resulting in behavioral and emotional problems in later adolescence and young adulthood, including social functioning

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later on, such as being unable to feel trusting and comfortable in social relationships [14]. Conversely, a majority of parents report positive outcomes in their unaffected children, including increased altruism and compassion for others [15].

In a recent survey of caregivers conducted by the Dravet Syndrome Foundation regarding the stresses and concerns of having a child with DS, 74% (114/154) of caregivers reported concerns about the emotional impact on siblings. In open-response sections, impact on siblings ranked second among most common caregiver concerns ($n = 42$; the top-ranking concern was speech/communication issues of the affected child [$n = 43$]) [11]. Furthermore, previous research reflects parental perceptions of impact on their unaffected children rather than direct responses from the siblings. However, parent proxy reports may not accurately reflect sibling experience [16,17]. Although the impact on siblings of children with other chronic conditions (e.g., cancer) has been extensively reported, families with a patient with DEE experience added burdens, including unpredictability of seizures [10,18], higher risk of injury and sudden death [2], and the social stigma associated with epilepsy [12,19]. The purpose of the Sibling Voices Survey was to directly assess perspectives of siblings with DEE. Our study evaluated parental and sibling responses to questions designed to assess the unique emotional and social impact of growing up with a sibling with a DEE.

2. Materials and methods

2.1. Survey development and recruitment

The Sibling Voices Survey was designed to be exploratory and was not intended to clinically assess the psychological health of participants. Survey questions were developed and beta-tested in consultation with several patient advocacy organizations (e.g., Dravet Syndrome Foundation, the LGS Foundation), communities of parents/caregivers, siblings, and expert healthcare professionals who specialize in patients with DEE and their families. A review of surveys reported in the prior medical literature confirmed that no existing validated survey was designed to evaluate siblings of people with DEE over the age range assessed in this study. Validated instruments that are suitable for children assess either psychological health of the respondent in general (e.g., Pediatric Quality of Life [PedsQL] [20]) or the impact of generalized epilepsy on adult family members (Impact of Pediatric Epilepsy Scale [IPES] [21]). The Sibling Voices Survey was customized to be applied to siblings of people with DEE. Where possible, questions from the PedsQL and the IPES were adapted for age-appropriateness and applicability to siblings of people with DEE. The development of the Sibling Voices Survey was also informed by customized surveys developed for siblings of people with generalized and intractable epilepsy conditions [12,14,22,23]. The research team led focus groups of parents/caregivers and siblings to review the survey questions for clarity, age-appropriateness, and content during the drafting process. The Western Institutional Review Board (IRB; Puyallup, WA) approved the protocol. Participants were recruited through social media, patient advocacy websites, and patient community events throughout the US. Sibling respondents ≥18 years old (y/o) and caregiver/parent respondents provided informed consent. Parent or guardian permission was required for participants <18 y/o. Anonymous survey responses were submitted via internet to a central database from July 19 to December 31, 2017.

2.2. Inclusion criteria

Participants were eligible if their family had ≥2 children, with ≥1 child with a diagnosis of DS, LGS, or another DEE. Eligible participants were siblings ≥9 y/o or parents/household members with a child with DEE.

2.3. Survey design and analysis

As an initial foray into the area of direct assessment of siblings' perceptions, a set of 4 online, cohort-specific surveys were developed: (1) parents responding with their perceptions of their unaffected children, (2) siblings 9–12 y/o, (3) siblings 13–17 y/o, and (4) adult siblings ≥18 y/o (Supplementary Material). Adult siblings were asked to answer from their current perspective and retrospectively while growing up. Quality-of-life metrics used visual analog scale scores based on 0 to 10 or 0 to 3 response options. Survey questions focused on a range of psychological and emotional symptoms, including symptoms of anxious or depressed mood, fear, and worry. Questions also assessed siblings' support circles, coping mechanisms, and knowledge of their sibling's DEE diagnosis. Respondents could also provide free-form responses.

2.4. Statistical analysis

Survey responses were presented as descriptive statistics, including means, medians, and proportions, as appropriate. Categorical data were summarized by frequency, proportions, and percentages. Continuous variables were summarized as means and standard deviations or as medians and interquartile ranges, as appropriate.

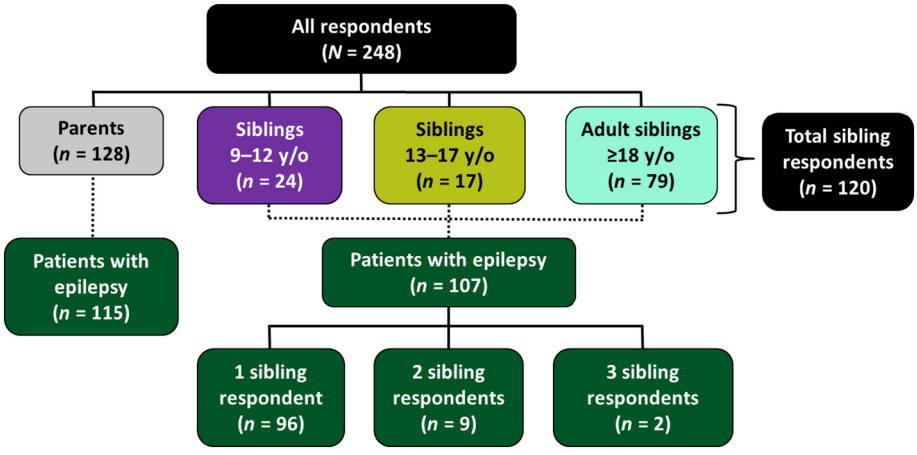

Fig. 1. Disposition of survey respondents. Survey respondents were subdivided by role (parent or sibling) and corresponding numbers of patients with epilepsy in the respondent families. Sibling respondents were further subdivided by age category. y/o, years old.

Table 1
Demographic characteristics of Sibling Voices Survey respondents.

Sibling respondents				
Characteristic	Sibling age group			All siblings
	9–12 y/o	13–17 y/o	Adult siblings	
N	24	17	79	120
Sex, *n* (%)				
Male	12 (50)	11 (65)	23 (29)	46 (38)
Female	12 (50)	6 (35)	56 (71)	74 (62)
Age, years, mean (range)	10 (9–12)	14 (13–15)	24 (18–63)	21 (9–63)
School grade, mean (range)	5 (3–7)	9 (8–11)	NA	NA
Family size, mean (range)	5 (4–8)	5 (4–10)	4 (1–30)	4 (1–30)
No. of children/family, mean (range)	4 (2–6)	4 (2–8)	3 (0–10)	3 (0–13)
Twin sibling, *n* (%)	2 (8)[a]	0 (0)	1 (1)	3 (2)

Parent respondents	
Characteristic	Parents
N	128
Siblings, *n*	198
Sibling sex,[b] *n* (%)	
Male	106 (54)
Female	92 (46)
Sibling age, years, median (range)[b]	12 (0–39)
Relationship, n (%)	
Mother	116 (91)
Father	10 (8)
Other	2 (<2)

NA, not applicable; y/o, years old.
[a] Excludes a set of 3 triplet siblings who selected "no" for "twin sibling."
[b] Some parents did not fill out surveys for all siblings in their family.

3. Results

3.1. Sibling voices survey: respondents and demographics

During the time that the survey was posted and open for response online, there were 4614 total page views and 248 nonduplicate responses. A total of 120 siblings and 128 parents submitted survey responses (Fig. 1; Table 1), representing 107 and 115 patients with DEE, respectively (Table 2).

3.2. Most difficult aspects of living with siblings with DEEs

All groups were comparable in their reports on the aspects of life that are harder/more difficult. Home life was reported as most disrupted across all groups (71%–84%; Table 3). Relationships with friends were reported to be affected by 28%–42% of respondents. Most (63%) parents reported that they believed the sibling had difficulties having friends over to the house, with 7% stating that the sibling did not want friends over at all. Relationships at school were reported to be affected by 21%–32% of respondents.

The 2 most difficult aspects reported by the 9–12 y/o group were feeling "worried/scared" (58%) and receiving "less [parental] attention" (50%); for the 13–17 y/o and adult sibling groups, the most difficult aspects were being "worried/scared" (59%; 70%) and "parents being stressed/unhappy" (59%; 77%) (Table 3). Parents reported that the 2 most difficult aspects for their unaffected child were receiving "less [parental] attention" (56%) and "having activities disrupted" (53%). Most siblings reported feeling worried or afraid when their sibling has a seizure (58%–70%). Responses to questions on whether and how these difficult aspects changed over time show that parental attention over time was reported as "improved" by 66% of adult siblings, 0% of 13–17 y/o siblings, and 38% of parents. Most (67%) 13–17 y/o siblings reported worsening over time.

Table 2
Demographic characteristics of patients with DEE.

Characteristic	Patients with DEE corresponding to:	
	Parent respondents (*n* = 128)	Sibling respondents (*n* = 120)
Patients, *n*	115	107
Sex, *n* (%)		
Male	53 (46)	44 (41)
Female	62 (54)	63 (59)
Age, years, mean (range)[a]	10 (1–50)	16 (1–61)
Patient diagnosis, *n* (%)		
Dravet syndrome	47 (41)	41 (38)
Lennox–Gastaut syndrome	13 (11)	13 (12)
Other DEE	55 (48)	53 (50)

DEE, developmental and epileptic encephalopathy.
[a] Excludes 2 deceased patients with DEE (both cohorts).

3.3. Burden of responsibility

Feelings of having "lost their childhood" were reported by 18% of 13–17 y/o siblings and 29% of adult siblings (see Fig. 3). Feeling overly responsible for their sibling with DEE was reported by 63% of adult siblings, 41% of 13–17 y/o siblings, and 34% of parents. Burden of responsibility was reflected in free-form comments, including "started working a lot around 18–19 to help pay the bills," "ended up being more like a second parent," and "taking more responsibility for his care; it occupies my daily life."

Of the adult siblings, 61% reported having discussed a plan for transitioning care of the affected sibling from the parent. Of these, 52% of adult siblings planned to assume caregiving. Top concerns in transitioning care were (1) "fear of something bad happening to the sibling while in my care" (70%), "fear of what will happen to my sibling if something happens to me" (67%), providing "a fulfilling and happy life for my sibling [with DEE]" (67%), and "the psychological/emotional toll" of caregiving (65%) (Table 3).

Table 3
Challenges faced by siblings of patients with DEE.

Survey responses of "Sometimes" or "A lot" on questions related to challenges faced by siblings of patients with DEE, *n* (%)				
	9–12 y/o siblings ($n = 24$)	13–17 y/o siblings ($n = 17$)	Adult siblings ($n = 79$)	Parents ($n = 128$)
How is life different or harder?				
Home	17 (71)	14 (82)	66 (84)	93 (73)
School	5 (21)	5 (29)	19 (24)	41 (32)
Friends	10 (42)	6 (35)	22 (28)	36 (28)
Most difficult aspects				
Worried/scared when sibling has a seizure	14 (58)	10 (59)	55 (70)	52 (41)
Parents stressed/unhappy	6 (25)	10 (59)	61 (77)	–
More responsibilities at home	8 (33)	9 (53)	49 (62)	38 (30)
Embarrassed by how they look	–	2 (12)	34 (43)	45 (35)
Having activities disrupted	11 (46)	7 (41)	33 (42)	68 (53)
Less attention from mom and dad	12 (50)	5 (29)	26 (33)	72 (56)
Coping with odd behavior or lack of control	7 (29)	9 (53)	41 (52)	61 (48)
Feeling as if you lost your childhood	–	3 (18)	23 (29)	–
Feeling overly responsible for sibling with DEE	–	7 (41)	50 (63)	44 (34)
Greatest concerns in transitioning care from parent to adult sibling (adult siblings were asked to check all that apply)				
Fear of something bad happening to my sibling while in my care	–	–	55 (70)	–
Fear of what will happen to my sibling if something happens to me	–	–	53 (67)	–
Being able to provide a fulfilling and happy life for my sibling	–	–	53 (67)	–
Psychological or emotional toll on me to care for my sibling	–	–	51 (65)	–
Having less control over my life (having my options limited)	–	–	44 (56)	–
Financial burden	–	–	44 (56)	–
Significant limitations on my freedom and independence	–	–	43 (54)	–
Fear that my sibling might die in my care	–	–	43 (54)	–
Physical demands required to take care of my sibling	–	–	37 (47)	–

3.4. Impact of DEE on sibling emotional well-being

All age groups, especially the adult group, reported impact on their emotional well-being.

3.4.1. Potential symptoms of anxious mood

In questions capturing potential symptoms of anxiety, being easily startled was reported by 58% of 9–12 y/o siblings, 53%–54% of adult siblings, 38% of parents, and 24% of 13–17 y/o siblings (Fig. 2A). "Having bad dreams/nightmares" was reported by 29%–49% of siblings compared with 14% of parents responding about their own perceptions of their unaffected children. Most respondents reported being afraid that their sibling might die (Table 4). Free-form responses reported worry, stress, and anxiety as chief concerns (Table 4). Adult siblings reported the highest levels of feeling stressed, worried, and afraid.

3.4.2. Potential symptoms of depressed mood

In questions related to depressed mood symptoms, feeling "down/unhappy" was reported by 47%–62% of siblings compared with 25% of parents who shared perceptions of their unaffected children (Fig. 2B). Levels of sadness (>8 on 0–10 scales) were reported by up to 42% of siblings compared with 14% of parents who shared perceptions of their unaffected children (Fig. 2C). Greater than 50% of siblings in most groups reported feelings of unhappiness or irritability; 35% of adult siblings reported a history of treatment for clinical depression. Free-form responses underscored feelings of sadness among siblings (e.g., "my younger brother has developed severe depression in part, I believe, due to growing up with my sister"; "negative energy constantly in our family").

3.5. Associations between mood scores, knowledge about DEE, and social support

Most siblings had knowledge of their sibling's condition (≥67%) and ways to help during seizures (≥83%), although 69% of 13–17 y/o and adult siblings reported feeling uncomfortable being alone with their brother/sister with DEE (Fig. 3). More than twice as many 13–17 y/o (88%) as 9–12 y/o siblings (42%) felt comfortable talking with others about their sibling with DEE, compared with 68% of adult siblings. Knowledge about DEE corresponded to being comfortable talking with others in more 13–17 y/o (86%) and adult siblings (62%) than 9–12 y/o siblings (38%). Better mood scores corresponded to having more DEE knowledge and/or being informed of ways to help, especially in younger groups (67% of siblings 9–12 y/o, 76% of siblings 13–17 y/o, and 52% of adult siblings).

3.6. Coping mechanisms

Cohort-specific questions assessed worrisome coping behaviors. Of the 13–17 y/o siblings, 6% reported using alcohol to cope with their situation at home. Parents reported that some siblings had lower grades (20%) and demanded attention (43%). Most parents (68%) responded that the sibling felt compelled to defend his or her sibling with DEE, and nearly a third (29%) reported that the sibling engaged in verbal/physical altercations. When survey respondents were asked what would help them cope better with their sibling's DEE, from a list of possible options, the 3 most common varied by group (Table 5). In the 9–12 y/o group, these choices included going on special outings alone with parents (38%), having a fun game or App to distract when they are stressed (38%), and having more information about their sibling's DEE (38%). The 13–17 y/o group identified going on special outings alone with parents (35%), having a fun game or App to distract when they are stressed (35%), and spending more time with friends (35%). Adult siblings noted learning ways to help with their stress (54%), spending more time with siblings of kids with DEE (51%), and gathering more information about their sibling's DEE (52%) as potentially most helpful. Parents reported that the top 3 things they thought would help their unaffected children were going on special outings alone with parents (76%), spending time with other siblings of kids with DEE (73%), and learning ways to help with stress, worry, and sadness (50%).

In free-form responses, most respondents (siblings, 68%; parents who shared perceptions of their unaffected children, 76%) identified some positive aspects about their brother/sister with DEE (Table 5). The youngest group tended to cite tangible evidence of special treatment (eating new foods, having more family time); adolescents and adult siblings tended to focus on their sibling's positive aspects (sense of humor, good nature). Some adult siblings also cited greater family

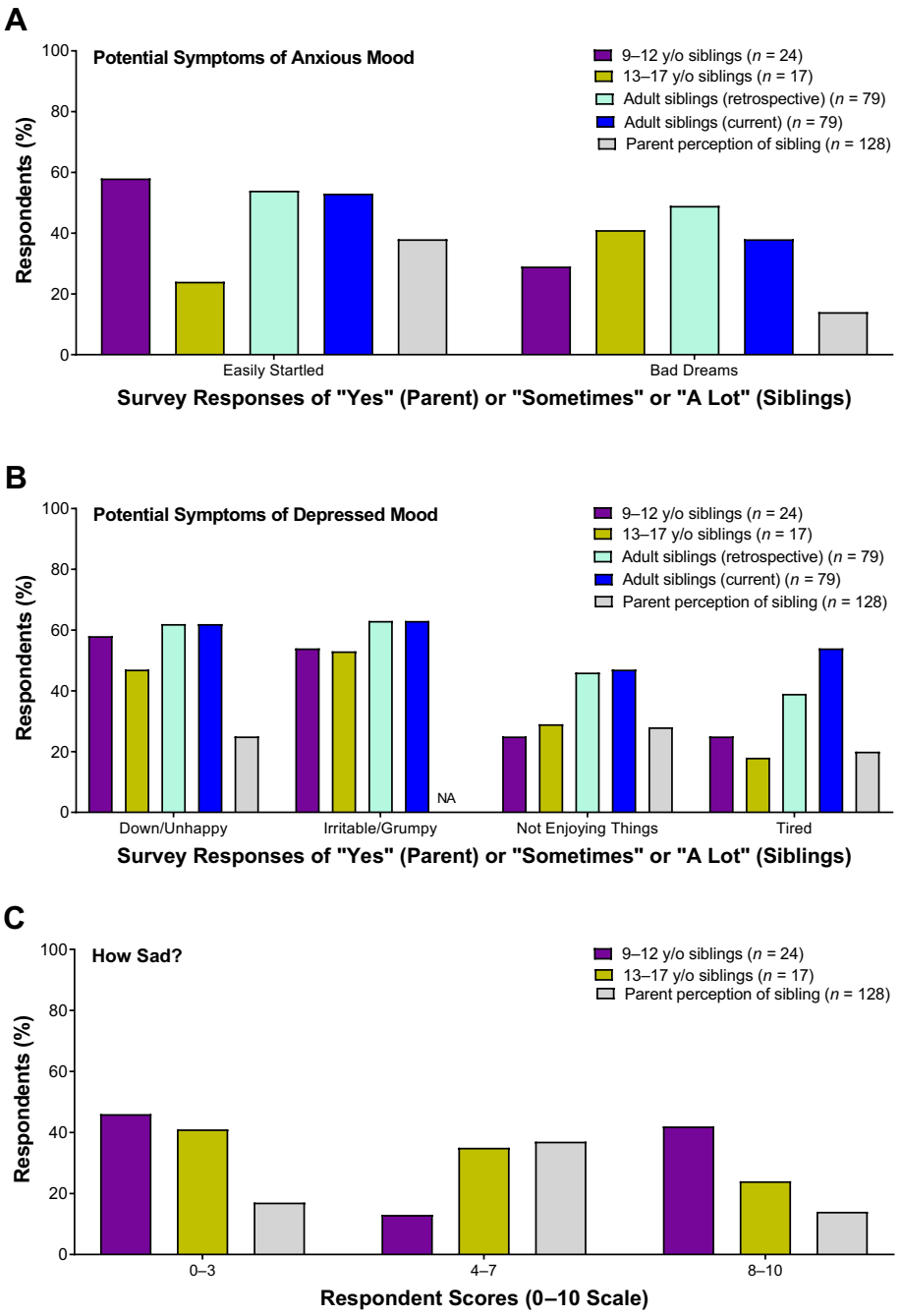

Fig. 2. Responses indicating potential symptoms of (A) anxious mood or (B) depressed mood or (C) level of sadness experienced by siblings related to their brother's or sister's DEE diagnosis.

cohesion and a more mature outlook on life. Parents emphasized positive attributes of their unaffected child's interactions with others (heightened empathy, maturity).

4. Discussion

Several recent studies describe the humanistic and economic impact of DEE on the broader family unit, including both caregivers and siblings [8–10]. Effects on siblings reported in previous studies most often have been obtained via proxy reports from parents/caregivers, which may not accurately reflect these siblings' actual feelings and experiences [16,17]. To our knowledge, the Sibling Voices Survey is the first study of its kind and seeks to directly investigate the psychosocial impact of DEEs on siblings versus being solely based on parent/caregiver proxy report. Survey responses indicate that, in general, siblings experienced depressed or anxious mood symptoms more often than parents perceived. Mechanisms for dealing with depressed or anxious feelings varied among groups. However, the youngest survey respondents (9–12 y/o) were much less comfortable talking to others about their sibling's diagnosis and their feelings and therefore may present potential risk for having unrecognized and undiagnosed symptoms of depressed mood and anxiety.

The present study highlights the potential unmet need for greater education and healthcare provider awareness of how DEE family dynamics may affect sibling mood and coping. Specialty medical visits for patients with DEE presumably focus on the affected sibling, with

Table 4
Fear that the sibling may die.

Fear that the sibling might die: Survey responses of "Sometimes" or "A lot" (siblings) or "Yes" (parents), *n* (%)[a]	
9–12 y/o siblings ($n = 24$)	19 (79)
13–17 y/o siblings ($n = 42$)	7 (42)
Adult siblings (retrospective; $n = 61$)	35 (57)
Adult siblings (current; $n = 61$)	42 (69)
Parents ($n = 16$)	15 (94)

Free-form responses identifying how fear that the sibling may die affected respondents	
Group	Response
Parents	"I think it caused a lot of anxiety that most kids never have to deal with."
	"I know he worried about it…my LGS child passed due to SUDEP."
Adult siblings	"Questioning every seizure and wondering if this one will take him."
	"It scared me and caused me a lot of stress that I did not believe I should share with others. I kept it to myself and it made me shut people out."
	"I was a child, so of course I spent countless nights up crying, wondering what would happen if I never saw [my sibling] again; it was hard."
	"I believe there was an ever looming feeling of worry, dread, and to a smaller extent, helplessness whenever a seizure occurred…by living the day-to-day stresses of living with my brother, I found that a wedge had formed between myself and my social interactions."
	"I acted out and made a lot of bad decisions, including substance abuse, because I wanted to be numb and not think about it."

[a] This question was optional for parents and adult siblings; not all participants responded.

the potential for little or no attention paid to the psychological health of caregivers/parents or other family members, particularly given the increasing demands on healthcare providers to see an increasing number of patients in their practices. Our survey results highlight the need to ensure that siblings are being adequately assessed for depressed or anxious mood symptoms. Families caring for children with DEE live in an environment permeated by stress, fear, and anxiety [8,10,24]. Although our study was not designed to identify which specific aspects contributed most to sibling mood, a majority of all groups identified disruptions in home life, fear that the sibling might die, and concerns about the burdens of responsibility and financial strain. These concerns align with stressors reported to adversely affect family psychological health when caring for children with DEEs, generalized epilepsies, and chronic illnesses in general [14,25,26]. Taken together, these stressors may lead siblings growing up in a DEE household to experience feelings of depressed mood, anxiety, and/or social isolation [10,25,27]. This may place these siblings at long-term risk for developing clinical depression or anxiety disorders. In our study, 35% of adult siblings reported that they had a history of receiving treatment for clinical depression and uncovered considerable impact on adult siblings. Healthcare providers and other clinicians are encouraged to discuss with parents the potential elevated risk for symptoms of sibling anxiety and depressed mood, especially in cases where these symptoms may be arising from challenges specific to growing up with a sibling with a DEE [11,18,28]. Furthermore, PAGs (e.g., LGS Foundation, Dravet Syndrome Foundation) appear to be aware of this risk and are increasingly offering emotional and social support for the entire family unit, including siblings—both children and adults [18,24,29]. The current study suggests that empowering siblings with knowledge of ways to help care for their affected sibling and to be informed about the clinical condition could potentially lower anxiety, reduce feelings of helplessness, and improve overall functioning, with closer bonds formed within the family unit [14,22,30]. Further research could include leveraging PAG networks as one means of communicating ways for siblings to feel more engaged with the family around providing care for the affected child.

The current study underscores the need for improved communication between parents/caregivers and their unaffected children regarding their emotional well-being and impact of having a sibling with DEE. Our study supports prior reports (in generalized epilepsy conditions) of discrepancies between sibling self-reports and parent proxies [16,17,25]. This difference was especially apparent in questions related to sibling mood, suggesting that potential symptoms of anxious or depressed mood may go undetected by parents [23,28,31]. Parents burdened by the emotional and physical burden of caring for their child with DEE may inadvertently overlook their unaffected children's emotional and psychological struggles [9,12]. Indeed, both our sibling and parent groups reported reduced parental attention and worry/fear as substantial contributing factors to challenges at home [15,21]. Children and adolescents may not have access to resources to appropriately cope with the stresses unique to families of patients with DEE. Without an

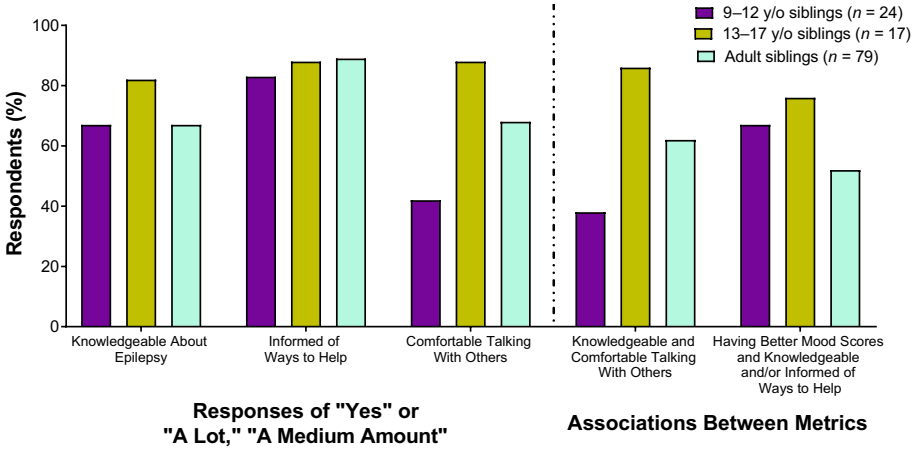

Fig. 3. Associations between knowledge, ways to help, comfort level talking to others, and mood scores in sibling survey respondents.

Table 5
Ways to cope and positive aspects.

Ways to cope with the sibling's condition				
Survey item, *n* (%) (Respondents were asked to check all that apply)	9–12 y/o siblings (*n* = 24)	13–17 y/o siblings (*n* = 17)	Adult siblings (*n* = 79)	Parents (*n* = 128)
Special outings with my parents (and just me)	9 (38)	6 (35)	30 (38)	97 (76)
A fun game, app, or video that would help me when I am feeling stressed	9 (38)	6 (35)	14 (18)	44 (34)
More information about my sibling's epilepsy	9 (38)	4 (24)	41 (52)	46 (36)
Spending more time with my friends	7 (29)	6 (35)	19 (24)	58 (45)
Spending more time with other siblings of kids with epilepsy	7 (29)	4 (24)	40 (51)	93 (73)
Learning new ways to help with my stress, worry, and sadness	5 (21)	2 (12)	43 (54)	64 (50)

Free-form responses identifying positive aspects or coping mechanisms	
Group	Response
9–12 y/o siblings	"We get family time, we get to see the firemen, we get to try new food."
	"He is different, and that's why people love him."
13–17 y/o siblings	"You are able to look at life from another perspective."
	"He is very funny, happy, and always super cute."
	"He isn't bashful, which makes him fun to be around, and he is very sweet."
	"She is an amazing person and never gives up…she doesn't let her condition get in the way of her dreams."
Adult siblings	"I am good in stressful situations and don't panic when most would…I feel like a stronger person due to the hardships we've gone through."
	"She has taught me how to love unconditionally, how to put another's happiness above my own. That the most important things in life are people, not things."
	"Wow, where to begin…he and it [epilepsy] have taught me to appreciate, to value, to live life to the fullest and the true meaning of being grateful."
	"The career that I want to do is primarily decided through my brother. A harder upbringing than some makes you more empathetic and understanding to other people who might have problems; it has given me some skills that I otherwise may not have had."
Parents	"They truly developed a close bond and love each other deeply…The positive impact is an unbreakable bond and unconditional love."
	"He is more responsible than other kids his age. He has great compassion for the sick. He is attending nursing school and plans eventually to be a pediatric nurse practitioner."
	"I think it has taught her to appreciate life, although I'm sure she feels sorry for herself because it does so affect her, but I think she realizes how much she cares for her brother and other kids with conditions, since it now personally affects her family."
	"He has done so well with other kids who have differences in school, and he is recognized for his loving, caring, and accepting ways."

outlet to voice concerns and share feelings, these children may experience a variety of feelings including "growing up too fast" [10], having "lost their childhood," or "feeling overly responsible" for their sibling with DEE [10,23]. Some larger treatment centers in urban, resource-rich areas provide emotional and social outlets specifically for siblings of children with a variety of chronic illnesses (e.g., Sib2Sib, Cook Children's Hospital, Fort Worth, TX, https://cookchildrens.org/patients/support-groups/Pages/default.aspx; Sibshops, siblingsupport.org) [18]. Sharing positive and negative experiences in a safe, empathetic environment could help improve coping and functioning [13,30,32,33]. Expanding such resources in areas of greatest concern to DEE communities specifically could provide additional outlets for siblings of children with DEEs to voice their concerns in a supportive environment. To our knowledge, studies assessing the effectiveness of sibling programs or psychotherapy interventions (e.g., cognitive behavior therapy) on DEE family unit populations have not been conducted. Further research is needed to identify the best approaches to support these siblings.

Additionally, our findings suggest the need for more effective support mechanisms and resources focused on this vulnerable sibling population. Most survey respondents (68%–76%) identified some positive aspects of siblings' relationships with their brother or sister with DEE, including fostering maturity and a sense of resilience. Other studies have identified closer familial bonds, emotional maturity, and a strong sense of unconditional love among siblings of children with chronic illnesses, including generalized epilepsies and DEEs [14,15,22,28,33]. Further research is needed to determine how to provide siblings with the support they need to reorient to the positive to the greatest extent possible.

This survey-based study has some limitations. Survey respondents were self-selected, with the associated potential for selection bias. Our study was not validated to diagnose clinical depression or anxiety, nor designed to identify specific concerns of sibling-reported anxiety, stress, or fear. Questions in the 4 age- and role-specific survey versions were developed solely for the purposes of this study and were not previously validated, although they were vetted in focus groups composed of expert clinicians, researchers, PAGs, patient families, etc. The small sample size of siblings <18 y/o precluded statistical analyses beyond descriptive statistics. Economic data for the respondents were not captured. Finally, the anonymized format of data collection precluded the possibility of analyzing parent perceptions of their own children. The survey response rate could not be accurately calculated. The 4614 page views were not necessarily from unique and potentially eligible participants who had a family member with a DEE. The site was in the public domain, and access was not limited to PAG members. Response rates could not be calculated based on the number of eligible participants compared with the number of respondents.

Further research is warranted to better understand how to identify when siblings in DEE households are struggling with symptoms of depressed and/or anxious mood potentially arising from the strain of their families' unique challenges. Moreover, research is needed to identify optimal interventions and coping mechanisms that reduce the emotional and psychological impact of specific stressors, including better support for parent-to-sibling care transitions where applicable [10,18,34,35].

5. Conclusions

The Sibling Voices Survey presents the impact of growing up with siblings with DEE on emotional well-being directly from the sibling perspective. Focusing resources on sibling psychosocial health early in life could improve sibling well-being, functioning, and resilience.

Declaration of competing interest

This study was sponsored by Zogenix, Inc. (Emeryville, CA). Laurie Bailey, Bradley Galer, Arnold Gammaitoni, and Carla Schad are employees of Zogenix, Inc., and own stock in the company. Lauren

Schwartz is a consultant of Zogenix, Inc. Mary Anne Meskis has nothing to disclose. Tracy Dixon-Salazar is a consultant at the Lennox–Gastaut Syndrome Foundation and the Chan Zuckerberg Initiative (Redwood City, CA). We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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Appendix A. Supplementary data

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