Parent perceptions and decision-making about treatments for epilepsy: A qualitative evidence synthesis

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TITLE:

Parent perceptions and decision-making about treatments for epilepsy: A qualitative evidence synthesis

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Abstract

Objectives: A qualitative evidence synthesis on parents' perspectives and experiences of treatments for childhood epilepsy.

Methods: Searched Embase, MEDLINE, PubMed, the Cochrane Library, CINAHL, PsycINFO, SocINDEX, and Web of Science from January 1, 1999, to August 19, 2021. We selected qualitative studies about caregiver's perspectives on anti-epileptic drugs, diet, or surgery treatments for childhood epilepsy. We used the Theoretical Domains Framework (TDF) to guide the extraction and synthesis of the qualitative findings as per a best-fit framework approach. One reviewer extracted study data and methodological characteristics, and two reviewers extracted qualitative findings. The team verified all extractions.

Results: We identified themes within TDF domains and synthesized summary statements of these themes. We assessed our confidence in our summary statements using GRADE-CERQual. We identified 5 studies (in 6 reports) of good methodological quality focused on caregiver perceptions of neurosurgery; we found limited indirect evidence on parents' perceptions of medications or diet. We identified themes within six of the 14 TDF domains relevant to treatment decisions: knowledge, emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources.

Conclusions: Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery. This process involves gathering evidence, discussing it with peers and professionals, working through emotions and overcoming barriers in the healthcare system. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed to fully understand the diversity of experiences of caregivers across various points in the decision-making pathway and among different healthcare contexts.

Strengths and Limitations

- This synthesis followed contemporary standards for the conduct of qualitative synthesis, which includes using a best-fit framework approach to categorize and synthesize findings based on the Theoretical Domains Framework (TDF).
- Risks to rigor of included studies were assessed using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies.
- Confidence in the conclusions drawn from this synthesis was rated using the GRADE CerQual.
- As with all qualitative synthesis, selecting which data to extract and how to code it is ultimately a subjective process.
- Limited to studies that examined parents' perceptions, experiences, and decision-making about pursuing surgery to treat their child's epilepsy.

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Epilepsy is a common neurologic disorder in children, affecting about 1% of children in the United States.[1] While there are different types of childhood epilepsy, each type involves recurring seizures caused by abnormal electrical activity in the brain. Epilepsy is categorized by seizure type (e.g., focal, generalized, or unknown), epilepsy type (e.g., focal, generalized, unknown), and syndrome type (e.g., childhood absence epilepsy, Dravet syndrome).[2, 3] No matter the type, epilepsy is a chaotic and unpredictable condition for both the affected children and their caregivers.[4]

Treatment and ongoing management approaches typically depend on the type of the epilepsy and prior treatment response. Treatment options for childhood epilepsy include anti-epileptic drugs, ketogenic diets, or surgery. Although many children with new-onset epilepsy achieve seizure freedom with anti-epileptic drugs,[5] these drugs are associated with numerous adverse effects (e.g., tiredness, nausea, headache, difficulty concentrating, depression, and suicidal ideation).[6] Furthermore, about 20% of children continue to experience seizures despite drug treatment.[7] The effectiveness of ketogenic diets (e.g., Atkins diet) is supported by low quality evidence.[8] However, these diets are also associated with adverse effects (e.g., gastrointestinal symptoms, dyslipidemia, decreased growth, and kidney stones), and require considerable caregiver effort to maintain. Surgical interventions may isolate and remove the underlying neurological cause of seizures, but carry risks of bleeding, infection, hydrocephalus, new neurologic deficits, and death.

Epilepsy treatment decision-making is thus complex and needs to consider each child's unique form of epilepsy, evidence of each treatment's potential benefits and harms, previous or ongoing experiences with treatment(s), and family's values and preferences. Although studies of treatment effectiveness rarely explore how families navigate these complex considerations or how these considerations may evolve over time, qualitative studies can offer context on caregiver experiences and decision-making. Syntheses of these qualitative studies can help shape caregiver and healthcare provider interactions and inform shared decision-making tools and processes. The purpose of this review is to summarize the qualitative research regarding caregivers' perspectives and experiences of treatments for childhood epilepsy.

Methods

We conducted a qualitative evidence synthesis in accordance with standards for qualitative evidence syntheses.[9-11]

Data Sources and Searches

We searched Embase, MEDLINE (in process), PubMed, the Cochrane Library, CINAHL, PsycINFO, SocINDEX, and Web of Science for studies published from January 1, 1999, to August 19, 2021 using terms related to epilepsy, treatments, and caregiver (or parent) perspectives (Appendix A). The search was limited to English-language. We also ran forward and backward citation searches (i.e., snowball searching) on included studies to ensure relevant studies were not missed.

Study Selection

We included qualitative studies that sought to understand caregiver's perspectives on anti-epileptic drugs, diet, or surgical treatments for childhood epilepsy. Studies needed to use qualitative methods for both data collection (e.g., focus group, individual interviews, or open-ended survey questions) and data analysis (e.g., thematic analysis). We excluded studies that focused only on non-caregiver perceptions (e.g., patient or healthcare provider) or that focused only on caregiver stress or caregiver expectations of treatment. We did not restrict by study country or care setting.

One reviewer screened titles and abstracts of citations retrieved from searches for eligibility. Two independent reviewers screened the full text of potentially relevant citations in PICO Portal© (https://picoportal.org/). Disagreements were resolved through discussion of the full team.

Data Extraction and Quality Assessment

For each study, we extracted details on the study design and methodological features, population characteristics, and qualitative analysis findings.

One reviewer assessed risks to rigor using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies, which appraises research aims, congruence between research aims and methodological approach, quality of sampling and data collection, appropriateness of application of methods, richness and conceptual depth of findings, appropriateness of interrogation of findings, and researcher reflexivity.[12] All team members reviewed the CASP assessments to ensure consistency of ratings across studies.

Two team members independently extracted and coded the qualitative findings of the included studies in MaxQDA© 2020 (Berlin, Germany), an online platform designed to support qualitative data management, extraction, and analysis. The extracted data included direct quotes from the participants (first order statements) and summary statements written by the study authors (second order statements). Extracted data were imported into spreadsheets to facilitate data cleaning, confirmation of themes, and synthesis.

To categorize the extracted data, we used the Theoretical Domains Framework (TDF),[13, 14] which was developed to assist in identifying the cognitive, affective, social, and environmental factors that may influence an individuals' performance of a health behavior. The health behavior of interest for this review was the decision to pursue an epileptic treatment. The 14 domains include: knowledge; skills; social/professional role and identity; belief about capabilities; optimism; beliefs about consequences; reinforcement; intention; goals; memory, attention, and decision processes; environmental context and resources; social influences; emotion and behavioral regulation (Appendix B). A third reviewer confirmed TDF domain codes and the team discussed the coding to ensure accuracy and consistency both within and across TDF domains. One reviewer did a final confirmation of extracted text and coding to ensure no data were missed and that there was consistency across domains.

Data synthesis and analysis

We adopted a best-fit framework approach to guide our qualitative synthesis. In this approach, data are coded according to TDF domains. One reviewer then summarized key themes within each TDF domain. Themes were discussed and debated among the team until consensus was achieved. We used the finalized themes to develop summary statements and assessed our confidence in these statements using GRADE-CERQual.[15-17] We rated our confidence in the summary statements as either high, moderate, low, or very low based on our assessment of the four GRADE-CERQual domains: methodological limitations, relevance, coherence, and adequacy of the data (Appendix C for definition of domains). "High confidence" refers to a finding that is highly likely to be a reasonable representation of the phenomenon of interest, whereas "very low confidence" refers to a review finding in which it was unclear if the finding was a reasonable representation.

Patient and public involvement: Patients or the public were not involved in the design, or conduct, or reporting, or dissemination plans of our research.

Results

The literature search yielded 1,447 citations from searches (see PRISMA flow diagram, Figure 1). We found 66 citations to retrieve for full-text screening of which five studies (in six publications) were included in the final sample.[18-23] Table 1 presents characteristics of included studies.

All studies examined parents' perceptions, experiences, and decision-making leading to surgery. All studies were conducted after the children had undergone surgery, with one study also surveying parents "just prior" to surgery.[23] Three studies reported data on parents' perceptions and experiences with medications and diet, but only in the context of selecting surgery (e.g., parents considering surgery due to the undesirable side effects or uncontrolled epilepsy with prior treatments).[18, 20, 23] In total, the views of 186 parents are represented in the five studies (the majority of whom were mothers, n=115). The time from children's surgery to data collection ranged from 6 months to 10 years among the three studies reporting on timing. Four studies collected parent perspectives through semi-structured interviews conducted in-person or over the telephone (length ranging from 10 to 75 minutes),[18-22] and one study captured parent perceptions through open-ended survey questions.[23]

All studies were assessed to have minor risk to rigor (Appendix D) due to the retrospective nature of recruitment and the lack of consideration (or reporting) of the relationship between researchers and participants. Otherwise, studies were considered appropriate in their use of qualitative design, methods of data collection, and analysis. The retrospective nature of the included studies raises concerns about recall bias, as surgical outcomes may have affected retrospective perceptions, and selection bias since studies only included parents of children who were: a) referred to surgery and b) proceeded with surgery (and possibly experienced some level of success with surgery though this was not reported explicitly). All but one study[22] did not report whether interviewers were part of the child's care team, which may have influenced responses. Ozanne et al. reported that the researchers that interviewed parents and analyzed the data were not part of the epilepsy surgery team. [22]

We identified and coded data for six of the 14 TDF domains: knowledge; emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources (Figure 2). We did not find evidence from extracted qualitative data for the remaining eight TDF domains. Appendix E provides the extracted text from studies linked to their synthesized themes.

The GRADE-CERQual table (Table 2) summarizes findings and conclusions for each TDF domain. We had no or minor concerns with the coherence of the findings (i.e., the synthesized findings reflects the complexity and variation of the data) or their relevance (i.e., the extent to which synthesized findings are applicable to the context specified in the review question). We had minor or moderate concerns with the adequacy (i.e., the degree of richness and quantity of the data supporting the synthesized finding) of the findings related to knowledge and environmental context and resources, respectively. Below, we report the summary statement (and associated GRADE-CERQual level of confidence) under each identified TDF domain, and summarize the key themes that contributed to the statements.

TDF Domain 1: Knowledge

Summary statement: *Evidence from four studies indicated that caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes contradictory information, caregivers value learning this new language so they can become better advocates for their child (Moderate confidence)* [18-22]

Once parents recognized "something [was] wrong" with the health of their child, they sought information from multiple sources "to understand seizures and epilepsy and become better advocates for their child."[18] Parents wanted information about medications,[18] surgery,[18, 20] and what types of doctors were needed to care for their child.[19] They also noted needing to learn about how to navigate the health system including multiple specialties, hospitals, and insurance procedures.[18]

Parents often perceived the information they received from professionals as inadequate to make informed decisions.[20] They described seeking lay language information from multiple sources, including additional professionals, the internet, books, family, friends, and other parents.[18, 22] Parents expressed discomfort in the "lack of understandable information" and "information imbalance" between themselves and their child's providers and sought to become "epilepsy experts in order to be effective advocates for their children."[19] After increasing their knowledge, parents felt more empowered to ask questions and participate more actively in the decision-making.[18, 20] Parents thus described active learning to ensure productive information exchanges with providers (including using clinical terms) to support a positive shared-decision making process.[19]

TDF Domain 2: Social role and identity

Summary statement: E*vidence from one study indicated that caregivers feel a sense of duty and need to do the right thing in selecting a treatment for their child (Moderate confidence).*[20]

Parents reported a "sense of duty" to be "strong" and "brave." They also reported feeling pressure to "'do the right thing' by their child; that is, choosing the treatment option that would give their child the best chance of "reaching their full potential."[20]

TDF Domain 3: Emotion

Summary statement: *Five studies provided evidence to suggest that caregivers experience the journey of navigating their child's epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation, fear to relief, and hope (Moderate confidence).*[18-22]

Parents expressed feeling drained and stressed from always being "on call,"[18] worrying about a seizure,[18, 21] or mood and behavioral difficulties.[21] Parents recalled feeling frustrated[18, 19] and desperate to "find a treatment option that would work."[19] For example, one mother was frustrated that her child's provider continued to perform medication trials to manage seizures and not discuss surgery as an option,[18] while another mother was desperate to get her son to another doctor but needed to wait for referrals because of insurance.

When considering surgery as a potential treatment option, parents reported the decision process as "difficult," "frightening," and "stressful."[19, 20] Parents feared surgery would lead to worse health outcomes for their child, [20, 23] change their child's personality, or cause a loss of function (e.g., partial or complete loss in speech or movement).[23] Parents also feared the possibility of surgery making seizures worse (e.g., increased frequency, duration). Thus, making the decision to have surgery "in vain."[23]

However, for many parents, the decision to pursue surgery brought relief and feelings of hope. Parents in one study expressed gratitude once they finally decided to pursue surgery as they felt it was "demanding to not know if surgery would be possible."[22] Parents also reported feeling hope for improvements in their child's health and wellbeing after they decided to choose surgery. Finally, after making the decision to pursue surgery (or learning it was a viable option), parents reported that they hoped for candidacy,[18, 20] and experienced relief when a date was set.[20]

TDF Domain 4: Social Influence

Summary statement: *Three studies provided evidence to suggest that surgery is a family decision that requires outside support from other caregivers "experiencing the same thing" and from a healthcare professional acting as "champions." (Moderate confidence)* [19-21]

Parents reported the value of connecting with other parents who were on a similar treatment journey (e.g., had a child with epilepsy that was unresponsive to medication) and had experiences and expertise that they could draw on.[19, 20] Peer connections helped parents understand the surgical procedural from a "parental perspective" and provided "emotional support."[20] Parents also reported the value of having a good team of epilepsy providers and a "champion" (e.g., doctor, nurse, or social worker) to advocate for them and help them navigate purposefully through their journey.[19] One mother described one such champion, a nurse coordinator, as a "life saver," who helped in getting referrals and pushed her to seek better care for her child."[19] Another mother described her child's pediatrician as her champion because "he encouraged me and gave me…confidence."[19]

Parents described seeing treatment decision-making as a family choice and indicated the importance of involving partners, siblings, grandparents, and the child (where developmentally appropriate). Parents found engaging the whole family in the decision-making process to be generally helpful, as the experience of the child's epilepsy and potential consequences of treatment impacted the whole family.[20] However, for some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary parent, she "witnessed" the true extent of their child's seizures.[20]

TDF Domain 5: Beliefs about Consequences

Summary statement*: Five studies provided evidence to suggest that parents undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.* [18-23]

Parents of children who went on to have surgery reported going through a transformation in thinking of surgery as a "last resort" to a "necessary and hopeful option."[18] This transformation in thinking evolved as parents acquired greater understanding about their child's illness and prognosis, experience in treating their child with other treatments, acquired knowledge about the potential risks and benefits of surgery, and processed the diverse emotions associated with surgery.[18, 19]

An important part of parents' moving toward a decision about surgery was weighing what they perceived to be the benefits and harms of surgery.[20, 23] In terms of benefits, parents hoped surgery would lead to improved outcomes such as seizure reduction and the opportunity for a "normal life" without the side effects of medications.[20, 23] Some parents were skeptical about the efficacy of surgery due to their experiences with medications and the ketogenic diet.

According to parents, these treatments were associated with side effects, negatively affected their child's quality of life, and provided only minimal improvement in seizure control.[20] In terms of adverse effects, parents were concerned that surgery would lead to surgical complications or various post-surgical impairments, including a change in personality, loss of speech or motor function, and behavioral problems.[23] A small number of parents reported having no concerns before surgery.[23]

Beyond rational consideration of the benefits and risks of surgery, parents often reported coming to the decision to select surgery only after exhausting all other treatment options. [20-22] Parents expressed feeling like "it was the only choice [they] could make"[20] and their "only option"[20] in improving their child's outcomes or preventing their condition from getting worse. Parents understood there were potential complications associated with surgery, but "preferred to take a risk [in proceeding with surgery] rather than live in constant fear"[22] or having a child that "was unconscious all the time."[21]

TDF Domain 6: Environmental Context and Resources

Summary statement*: Three studies provided evidence that parents face challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process (Low confidence)*[18-20, 22]

Parents from one study in the U.S.[18, 19] and another in Sweden.[22] reported experiencing significant barriers with navigating the healthcare system. In the U.S.,[18, 19] parents expressed frustration with the extensive time it took to find the right doctor after navigating various doctors from different specialties across multiple institutions. Prior to selecting surgery for their child, parents reported difficulties in finding the "right doctor" with knowledge to "effectively identify the problem, and then make a clear plan of action." Parents attributed these difficulties to the lack of pediatric neurologists in their local area, inconsistencies in treatment recommendations, and rigid adherence to center-specific treatment protocols.[18, 19] Once parents made a decision to pursue surgery, they reported battles with insurance companies to pay for surgery.[18, 19] Parents in the Sweden reported similar frustrations with "the bureaucracy" as they felt it took "a long time to get a correct diagnosis" and "adequate support." They "thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary.[22]

Parents listed several provider-specific interactions they found to be either enablers or barriers to their experience of identifying and selecting appropriate treatment for their child. With respect to enablers, parents valued when providers validated (and shared) their concerns, gave their time and fostered trust, and engaged parents in the treatment decision-making process. Barriers noted by parents included having their concerns doubted or ignored,[18, 22] receiving inadequate information or support (especially before the epilepsy was recognized to be drug-resistant),[22] and feeling excluded from discussions about their child's surgical candidacy.[20] One study reported that parents perceived physician variability in knowledge about epilepsy and their lack of understanding about the pre-surgical referral process and appropriateness of surgery as barriers.[18] Parents in one study reported how they felt more reassured when their child received care from a comprehensive team of professionals with diverse expertise.[19] Parents from one study suggested providers give an "earlier and softer introduction of surgery as a possible treatment option."[20]

[Box 1 goes here]

Discussion

Understanding parent perspectives and experiences about treatments for childhood epilepsy is important for helping ensure that parents are appropriately supported during their decision-making process. Our qualitative evidence synthesis of five studies, which had minimal risks to rigor, identified key findings across six domains: Knowledge, Emotions, Social/Professional Role and Identify, Social Influences, Beliefs about Consequences, Environmental Context and Resources. However, the evidence was limited to considerations regarding surgical treatment. Overall, these findings suggest that the decision to select surgery for childhood epilepsy involves parents going through a complex journey of acquiring extensive knowledge, working through intense emotions and perceived parental responsibilities, needing family and peer support, transforming beliefs about epilepsy and potential treatment options, and navigating various barriers and facilitators of the healthcare system.

The findings provide important evidence of domains amenable to action, which may support parents' decision-making process. For example, parents often feel that they do not have enough information to make decisions about surgery. Parents also reported experiencing intense psychological distress and exhaustion during their decision-making process and felt these emotions acutely through their role as parents responsible for making a potentially life-altering decision for their child. They also reported valuing connections with peers who were going through (or had gone through) the same experience. Healthcare systems may therefore consider providing parents with resource to connect with other parents. Further work is needed to address parents' perceived lack of shared-decision making with providers and healthcare system barriers.. Patient navigators may help as a first step to helping parent's navigate the healthcare system and gain access to additional information and resources.

The findings also identified gaps in the evidence base regarding parents' perceptions and decision-making processes. For instance, we did not identify evidence mapping to certain TDF domains that we expected to find evidence for, such as Goals. Studies included in this review were vague and inconsistent in reporting parent's goals or their desired outcomes of treatment. Some studies noted that parents wanted what 'was best' for their child or they would be happy with a reduction in seizure frequency. Survey data collected from parents considering surgery found the primary goal was seizure freedom (98%), followed closely by reduced medication (90%), and improved cognition (82%).[24] We also did not identify evidence for the domain of Memory, Attention, and Decision process. Thus, we could not determine if parents become more skilled and confident in their decision-making over time through experience and acquired knowledge.

Similarly, we only found limited evidence for the domain of Environmental context and Resources. Further evidence is needed to understand how culture and equity play a role in parents' perceptions about treatment and their capacity to access care for their child. For example, many parents' reported important barriers in knowledge, access to professionals to diagnose and treat their child, and challenges with accessing and paying for surgery. The extent to which these factors would be the same or increase among families from different socio-economic backgrounds and cultures with varying economic, educational, and social resources needs further exploration. One may hypothesize that parents' perceptions of treatments for epilepsy vary depending on cultural perspectives of the condition and availability and access to care.

Strengths and limitations

This review followed contemporary standards for the conduct of qualitative synthesis, which includes the use of a best-fit framework approach, a theory-informed framework to guide our synthesis and extraction, and use of the CASP and GRADE CerQual tool to assess the rigor and confidence of our findings. The use of the TDF is a particular strength of this review as it lends itself to both actionable interventions (e.g., mapping intervention strategies to key domains identified) and future research (e.g., further examination of domains not identified in the evidence such as 'memory, attention, and decision processes'). However, as with all qualitative research, selecting which data to extract and how to code it is ultimately a subjective process. We attempted to limit subjectivity within our group by coding in duplicate and having regular meetings to ensure consistency across and within domains.

One primary limitation of the evidence included in this review is that the data in all studies were collected retrospectively. Parents were asked for their perceptions *after* their child had undergone surgery. This may have resulted in selection bias, as studies recruited parents of children referred to and proceeded with surgery. Thus, findings may not be fully reflective of the wider population of parents who are making decisions regarding surgery. The perceptions of parents who declined surgery were not captured. The retrospective nature of the data collection may have also resulted in recall bias. The time from children's surgery to data collection ranged from 6 months to 10 years. Parents may have forgotten important aspects of their journey to surgery or filled in gaps of memory due to experiences with the child's outcome. As described by one study, parent responses were "memories processed through emotions and colored from further experiences, which were then developed into opinions and personal views."[22]

Conclusion

Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery involving gathering evidence, discussing it with peers and providers, working through emotions and overcoming barriers in the healthcare system. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed to fully understand the diversity of experiences of parents across various points in the decision-making pathway and among different healthcare contexts. In particular, qualitative studies are needed that address parents' perceptions and experiences with selecting alternative epilepsy treatment options such as diet and medications.

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Footnotes

Contributors: SU served as project lead to coordinate and participate as a team member in identifying studies for inclusion, extracting and coding data, synthesizing findings, developing and grading summary statements, and drafting of initial report and manuscript; KK participated as team member in all aspects of data extraction, coding, synthesis, developing and grading summary statements, validating all findings, and revising initial drafts of manuscript; RW participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; GA coordinated and revised search strategies and participated in all aspects of data extraction, coding, synthesis, and review of manuscript; KR participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; MV participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript.

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Patient consent: Not required.

Ethics approval: This study does not involve human or animal participants

Data sharing statement: No additional data available.

Box 1. Example excerpts for TDF domains

Box 1. Example excerpts for TDF domains

Knowledge

- [Parent]: “I googled it until I couldn’t google anymore, and I think that gave me a better understanding of what was going on.”[20].
- A mother who stated, “I just think knowledge is power and it also brings some comfort to making a good decision,” exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy[18]

Social/Professional Role and Identity

- Parents discussed feeling pressure from a need to “do the right thing” by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential.[20]

Emotion

- Participant descriptions highlighted the significant impact that epilepsy had upon on the child’s and family’s lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: “you’re tired yourself before you even go out the door”[21]
- [Parent]: “At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric”[19]

Social Influences

- Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. “Gosh, we didn’t tell anybody. We were ashamed.”[21]
- [E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information” [19].

Beliefs about Consequences

- This transformation occurred as parents became increasingly aware of the severity of their child’s epilepsy and its impact on their child’s and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment [18]
- They wanted that their child could do everything their friends could do like “play football, cycle, and swim independently”. Some parents mentioned that they wanted their child to be able to go back to school and have a “proper social life” while some parents referenced the

future hoping that the child would be better placed to get a job and driving license as a result of surgery[23]

- Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.[22]

Environmental Context and Resources

- Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.[22]
- Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, "'I don't believe that she is a surgical candidate.' As a parent, I listened and I wanted to think otherwise.... In hindsight, I'm glad I didn't listen to him."[18]

Table 1. Characteristics of included studies

Study, year, country	Population	Number of parents	Time from surgery to data Collection	Study aim	Data collection methods	Data analysis methods
Baca[1], 2015; Pieters, 2016, USA	Parents of children who had previously undergone resective epilepsy surgery	37 (individual parents) 31 mothers; 6 other	NR	Baca, 2015: To identify the nature and range of parent-perceived barriers to timely receipt of pediatric epilepsy surgery. Pieters, 2016: A) To describe thoroughly the parental experiences and perceptions of this slow and arduous period prior to the presurgical referral and evaluation to a comprehensive pediatric epilepsy center. B) To delineate the range of parent identified factors, or facilitators, that helped move families forward along their journey to surgery	Semi-structured interviews lasting on average 29 minutes (range 10 to 60 mins)	Thematic analysis
Heath, 2016, UK	Parents of children who had undergone pediatric epilepsy surgery Also interviewed healthcare professionals caring for children with epilepsy	9 (individual parents) 8 mothers; 1 father 10 healthcare professionals	NR	To explore how parents and health professionals make decisions regarding pediatric epilepsy surgery to identify: A) factors that influence the process of decision-making regarding pediatric epilepsy surgery from the perspective of parents and professionals and B) the support needs of those considering surgery as a treatment option for a child with medically intractable epilepsy	Semi-structured interviews lasting about 1 hour, and non-participant observations	Thematic analysis
O'Brien, 2020, UK	Parents and their children who had gone through resective epilepsy surgery	9 parents (1 couple; 7 individual parents) 8 mothers; 1 father	6 months to 3 years	To explore children's and parents' perspectives on the journey prior to and following surgical treatment, with a focus on the emotional experiences of children and their parents throughout the surgery journey	Semi-structured interviews lasting 25 to 75 mins	Thematic analysis
Ozanne, 2016, Sweden	Parents of children who had previously undergone resective epilepsy surgery	24 parents 13 mothers; 11 fathers	5 to 10 years	To explore parental experiences before and after hemispherotomy as reported at a long-term follow-up and the parents' views on received information and support	Interviews (length of time not reported)	Content analysis
Sylven, 2020, Sweden	Parents of children who were undergoing resective epilepsy surgery (and 2 years post-surgery)	107 (parental responses to both surveys)	2 years	A) To understand parental hopes and worries before their child underwent epilepsy surgery and B) To understand parental satisfaction two years after their child had undergone epilepsy surgery	Surveys (open-ended questions)	Thematic analysis

[1] Baca & Pieters included the same patient population but reported on different aspects of parent perspectives.

Table 2. GRADE-CERQual summary of findings statements and ratings

Summary of Review Findings	Studies contributing	Methodological limitations (CASP ratings)	Coherence	Adequacy[2]	Relevance	CERQual assessment	Explanation of CERQual assessment
Knowledge: Caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes contradictory information, caregivers value learning this new language so they can become better advocates for their child	Baca/Pieters Heath O'Brien Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concerns*: Included studies limited to surgery as treatment decision. We did not identify studies meeting inclusion criteria that addressed other treatments, such as diet or medications. Each study addressed knowledge acquisition and reported on it in some depth.	*No or very minor concerns*	*Moderate confidence*	Four studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Social/professional role and identity, parents feel a sense of duty and need to do the right thing in selecting a treatment for their child.	Heath	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concern:* Included studies limited to surgery as treatment decision.	*No or very minor concerns*	*Moderate confidence*	One study; minor concerns related to methods and limited treatment scope (surgery).
Emotion, parents experience the journey of navigating their child's epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation and fear to relief and hope.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*: All 4 studies contributed to the rich depth of the varied emotions from fear of the illness to fear of the surgery to the mental toll that took on families and hope	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).

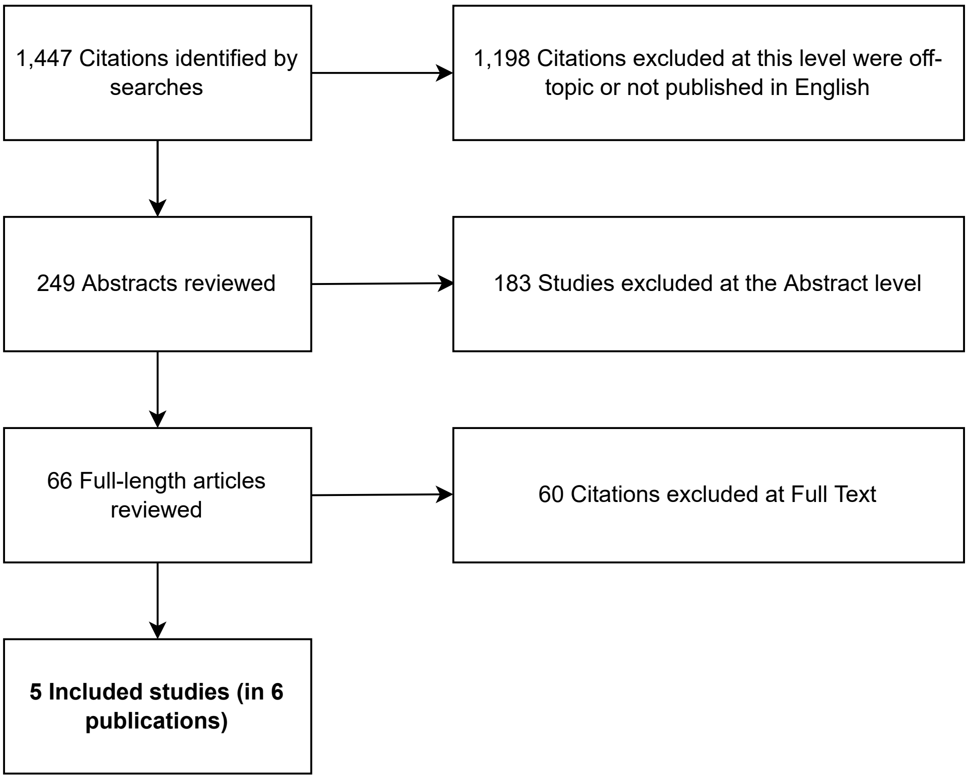
Summary of Review Findings	Studies contributing	Methodological limitations (CASP ratings)	Coherence	Adequacy[2]	Relevance	CERQual assessment	Explanation of CERQual assessment
				and relief that surgery may offer			
Social influences, surgery is a family decision that requires outside support from other parents "experiencing the same thing" and from a healthcare professional acting as "champions."	Pieters Heath O'Brien	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Three studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Beliefs about consequences, caregivers undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Environmental context and resources, parents expressed challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process.	Baca/Pieters Heath Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*Moderate concerns*: The three studies are limited to surgery and to US, Sweden and the UK. Healthcare barriers seemed particularly salient to U.S. respondents (including insurance barriers)	*No or very minor concerns*	*Low confidence*	Three studies with similar findings; moderate concerns related to methods, limited scope (surgery), and context-specific healthcare experiences of participants

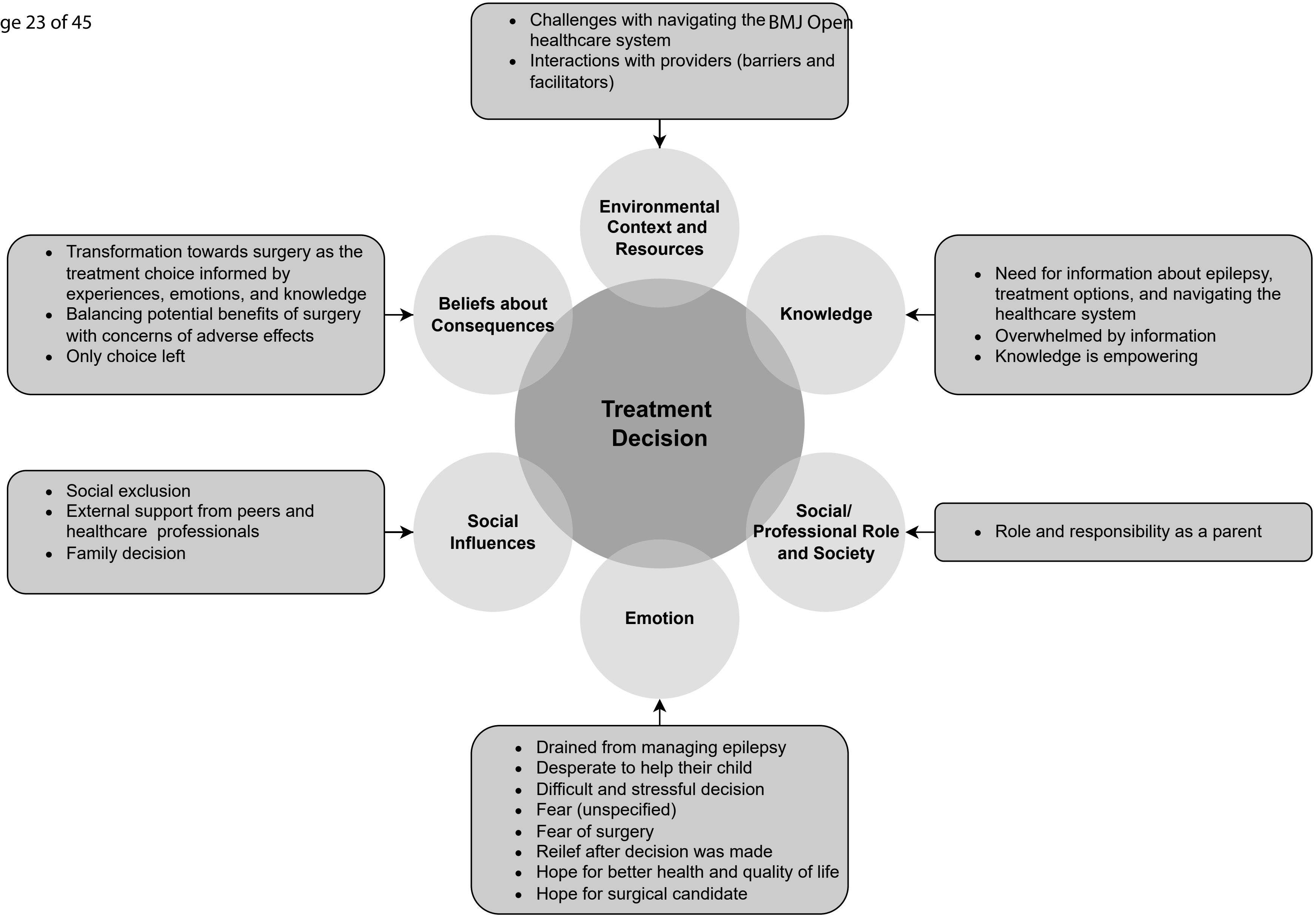
[1] Retrospective recruitment raises concerns about selection bias.

[2] We only downgraded for adequacy due to studies being limited to surgery when the summarized theme was based on parent's general feelings about epilepsy and could thus potentially apply to parent decisions regarding other treatment choices.

Figure 1 Legend: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram of studies identified

Figure 2 Legend: Figure 2 presents the six theoretical domains identified in this review that impacted treatment decision-making and presents the main themes representing those domains


1,447 Citations identified by searches
1,198 Citations excluded at this level were off-topic or not published in English
249 Abstracts reviewed
183 Studies excluded at the Abstract level
66 Full-length articles reviewed
60 Citations excluded at Full Text
5 Included studies (in 6 publications)


Challenges with navigating the healthcare system
Interactions with providers (barriers and facilitators)
Environmental Context and Resources
Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Balancing potential benefits of surgery with concerns of adverse effects
Only choice left
Beliefs about Consequences
Knowledge
Need for information about epilepsy, treatment options, and navigating the healthcare system
Overwhelmed by information
Knowledge is empowering
Treatment Decision
Social exclusion
External support from peers and healthcare professionals
Family decision
Social Influences
Social/ Professional Role and Society
Role and responsibility as a parent
Emotion
Drained from managing epilepsy
Desperate to help their child
Difficult and stressful decision
Fear (unspecified)
Fear of surgery
Reilief after decision was made
Hope for better health and quality of life
Hope for surgical candidate

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Title page
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Background, pg 3
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Pg 3
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Pg 3-4
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Pg 3, Appendix A
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Appendix A
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Pg 4-5
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Pg 4-5
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Pg 4-5
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	NA
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Pg 4-5, Table 1
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Pg 4-5
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Pg 4-5
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Pg 4-5
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	Pg 4-5
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	NA
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Pg 4-5< Appendix D

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Pg. 4-5, Table 2
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Pg 5
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Pg 5
Study characteristics	17	Cite each included study and present its characteristics.	Pg 5, Table 1
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Appendix D
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Table 2
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Pg 5
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Pg 6-8
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	Pg 6-8
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	NA
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	NA
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	Pg 6-8, Table 2
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Pg 9
	23b	Discuss any limitations of the evidence included in the review.	Pg 10
	23c	Discuss any limitations of the review processes used.	Pg 10
	23d	Discuss implications of the results for practice, policy, and future research.	Pg 9-10
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Not registered
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	NA
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	NA
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Pg 11
Competing interests	26	Declare any competing interests of review authors.	Pg 11
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Pg 11

PRISMA 2020 Checklist

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

For more information, visit: http://www.prisma-statement.org/

APPENDIX A: Search Strategy

EMBASE (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Infantile/ Pediatric Epilepsies	'benign childhood epilepsy'/exp OR 'childhood absence epilepsy'/exp OR 'severe myoclonic epilepsy in infancy'/exp OR (dravet* NEXT/1 (disease OR syndrome))
2	0-3 Age Group and Epilepsy	[infant]/lim OR [newborn]/lim OR newborn/exp OR [preschool]/lim OR 'preschool child'/exp OR toddler/exp OR (babies OR baby OR child*:ti OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR 'very young'):ab,ti,kw OR ('younger than' OR under OR below) NEAR/3 (3 OR three) OR (3 OR three) NEAR/3 ('or below' OR 'or under' OR 'or younger') AND ('epilepsy'/exp OR 'epileptic patient'/exp OR epilep*:ti)
3	Infantile Spasms/ Neonatal Seizures (not requiring epilepsy)	'infantile spasm'/exp OR (((infan* OR neonat* OR newborn*) NEAR/2 (convuls* OR seizure* OR spasm*)):ab,ti,kw)
4	Other Seizures (not requiring Epilepsy)	([infant]/lim OR [newborn]/lim OR 'newborn'/exp OR babies:ab,ti,kw OR baby:ab,ti,kw OR infan*:ab,ti,kw OR neonat*:ab,ti,kw OR newborn*:ab,ti,kw OR nicu:ab,ti,kw) AND ('febrile convulsion'/exp OR 'seizure'/exp OR convuls*:ab,ti,kw OR spasm*:ab,ti,kw OR seizure*:ab,ti,kw)
5	Pharmacologic/ Vitamin Treatment	acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide
6	Diet Therapy	'ketogenic diet'/de OR (keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic'):ab,ti,kw
7	Surgical Procedures	craniotomy/de OR hemispherectomy/de OR 'laser surgery'/de OR lobectomy/de OR 'corpus callosotomy' OR craniotom* OR (disconnect* NEAR/3 (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* NEAR/3 (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar NEAR/3 disconnect*) OR (palliat* NEAR/3 operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection'
8	Brain Stimulation	'brain depth stimulation'/de OR 'brain responsive neurostimulator'/de OR 'deep brain stimulator'/de OR 'nerve stimulation'/de OR 'nerve stimulator'/de OR 'vagus nerve stimulation'/de OR ('brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') NEAR/2 (electrostim* OR stimulat*)) OR neurostim*

9	Harms	anhidrosis/de OR 'adverse event'/de OR 'adverse drug reaction'/de OR 'behavior disorder'/de OR 'cognitive defect'/de OR 'developmental delay'/de OR 'developmental disorder'/de OR dystonia/de OR 'liver injury'/de OR 'loss of appetite'/de OR 'motor dysfunction'/de OR 'organ damage'/de OR 'patient harm'/de OR 'sleep disorder'/de OR sweating/de OR (advers* OR harm* OR 'side effect'):ab,ti,kw OR anhidrosis OR (appetite NEAR/3 (lose OR losing OR loss)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) NEAR/3 (effect* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR neurodevelopment*) NEAR/3 (delay* OR disorder* OR regress*)) OR dystonia OR hypohidrosis OR hypohydrosis OR (liver NEAR/3 (damag* OR injur*)) OR (miss* NEAR/3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) NEAR/3 (disorder* OR inability OR unable))
10	Parental Preferences	parent/de OR (parent* OR mother* OR father*):ab,ti,kw
11	Untreated Disease	'treatment refusal'/de OR ('not treated' OR 'no treatment' OR untreat*):ab,ti,kw OR (declin* OR forgo* OR 'not' OR no OR refus* OR withheld OR withhold*) NEXT/3 (treated OR treatment*)
12	Study Designs/ Publication Types	[english]/lim AND [1999-2020]/py NOT ([animals]/lim NOT [humans]/lim OR abstract:nc OR annual:nc OR 'book'/de OR ((case NEXT/1 (report* OR stud*)):ti) OR 'case report'/de OR 'case study'/de OR conference:nc OR 'conference abstract':it OR 'conference paper'/de OR 'conference paper':it OR 'conference proceeding':pt OR 'conference review':it OR congress:nc OR diagnos*:ti OR 'diagnosis'/mj OR 'diagnostic accuracy'/mj OR 'diagnostic procedures'/mj OR 'diagnostic test'/mj OR 'diagnostic test accuracy study'/mj OR 'differential diagnosis'/mj OR 'editorial'/de OR editorial:it OR 'erratum'/de OR guideline*:ti OR letter:it OR 'note'/de OR note:it OR meeting:nc OR 'practice guideline'/de OR 'review'/exp OR sessions:nc OR 'short survey'/de OR symposium:nc OR animal*:ti OR experimental:ti OR (vitro:ti NOT vivo:ti) OR canine:ti OR dog:ti OR dogs:ti OR mouse:ti OR mice:ti OR rabbit*:ti OR rat:ti OR rats:ti OR rodent*:ti OR sheep:ti OR swine:ti)
13	Combine sets	#1 OR #2 OR #3 OR #4
14	Combine sets (KQ1 Pharmacology)	#13 AND #12 AND #5
15	Combine sets (KQ2 Diet, Surgery, Brain Stimulation)	#13 AND #12 AND (#6 OR #7 OR #8)
16	Combine sets (KQ3 Treatment Harms)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND 9
17	Combine sets (CQ1 Parental Preferences)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND #10
18	Combine sets (CQ2 Untreated/ Uncontrolled Epilepsy)	#11 AND #12 AND #13

19	Combine Sets All KQs	#14 OR #15 OR #16
20	Combine Sets All CQs	#17 OR #18

SocINDEX and Web of Science databases (Searched January 1, 1999, to August 19, 2021)

(epilep* OR convuls* OR seizure* OR spasm*)

AND

(acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide OR keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic' OR 'corpus callosotomy' OR craniotom* OR (disconnect* AND (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* AND (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar AND disconnect*) OR (palliat* AND operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection' OR 'brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') AND (electrostim* OR stimulat*)) OR neurostim*)

AND

(parent* OR mother* OR father*)

AND

(perception OR factor* OR decision)

CINAHL (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR dravet*
2	Epilepsy/ 0-3 Age Group	(babies OR baby OR child* OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR "very young" OR "younger than three" OR "younger than 3" OR "under three" OR "under 3" OR "below three" OR "below 3" OR "3 or below" OR "3 or younger" OR "three or below" OR "three or younger") AND epilep*
3	Infantile Spasm/ Neonatal Seizure	"infantile spasm*" OR "neonatal seizure*" OR ((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) AND (convuls* OR seizure* OR spasm*))
4	Parental Preferences	father* OR matern* OR mother* OR parent* OR patern*
5	Harms	"adverse drug reaction" OR "adverse effect*" OR "adverse event*" OR anhidrosis OR appetite* OR behavior* OR behaviour* OR cognitiv* OR defect* OR delay* OR development* OR disorder* OR dysfunction* OR dyston* OR harm* OR impair* OR injur* OR liver OR motor OR "organ damage*" OR "sleep disorder*" OR sweat* OR "side effect*"
6	Untreated Disease	"treatment refusal" OR "not treated" OR "no treatment" OR untreat* OR "decline treatment" OR "declined treatment" OR "forgo* treatment" OR "refuse treatment" OR "refused treatment" OR "refusing treatment" OR "withheld treatment" OR "withhold treatment" OR "withholding treatment"
7	Combine Concepts	(S1 OR S2 OR S3) AND S4
8	Apply Limits	S7 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)
9	Combine concepts	(S1 OR S2 OR S3) AND (S5 AND S6)
10	Apply Limits	S9 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)

PsycINFO (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR (dravet* adj1 (disease OR syndrome)) OR ((child* OR infan* OR paediatr* OR pediatr*) adj1 epilep*)
2	Epilepsy/ 0-3 Age Group	(baby OR babies OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool OR toddler* OR "very young" OR (("younger than" OR under OR below) adj3 ("3" OR three)) OR (("3" OR three) adj3 ("or below" OR "or under" OR "or younger")) AND (epilepsy/ OR epileptic seizures/ OR epileps* OR epileptic*))
3	Infantile Spasm/ Neonatal Seizure	((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) adj3 (convuls* OR seizure* OR spasm*))
4	Parental Preferences	parents/ OR (patern* OR parent* OR matern* OR mother* OR father*)
5	Harms	behavior disorders/ OR cognitive impairment/ OR delayed development/ OR developmental disabilities/ OR liver disorders/ OR motor development/ OR "side effects (drug)"/ OR "side effects (treatment)"/ OR sleep wake disorders/ OR sweating/ OR (advers* OR anhidrosis OR dystoni* OR harm* OR harm* OR hypohidrosis OR hypohydrosis OR "organ damage") OR (appetite adj3 (lose* OR losing OR loss OR

		lost)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) adj3 (effect* OR deficit* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR motor* OR neurodevelopment*) adj3 (delay* OR disorder* OR regress*)) OR ((liver OR organ*) adj3 (damag* OR injur*)) OR (miss* adj3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) adj3 (disorder* OR inability OR unable))
6	Untreated Disease	treatment refusal/ OR ("not treated" OR "no treatment" OR untreat*) OR ((declin* OR forgo* OR "not" OR no OR refus* OR withheld OR withhold*) adj3 (treated OR treatment*))
7	Combine Sets	(1 OR 2 OR 3) AND 4
8	Apply Limits	limit 7 to yr="1999 - 2021"
9	Apply Limits	limit 8 to up=19000101-20210819
10	Apply Limits	9 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
11	CQ1	limit 10 to english language
12	Combine Sets	(1 OR 2 OR 3) AND (5 AND 6)
13	Apply Limits Apply Limits	Limit 12 to yr="1999 - 2021"
14	Apply Limits	limit 13 to up=19000101-20210819
15	Apply Limits	14 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
16	CQ2	limit 5 to english language

APPENDIX B: The Theoretical Domains Framework*

Domain	Definition	Constructs
Knowledge	An awareness of the existence of something	Knowledge (including knowledge of condition/scientific rationale) Procedural knowledge Knowledge of task environment
Skills	An ability or proficiency acquired through practice	Skills Skills development Competence Ability Interpersonal skills Practice Skill assessment
Social/professional role and identity	A coherent set of behaviors and displayed personal qualities of an individual in a social or work setting	Professional identity Professional role Social identity Identity Professional boundaries Professional confidence Group identity Leadership Organizational commitment
Beliefs about capabilities	Acceptance of the truth, reality or validity about an ability, talent or facility that a person can put to constructive use)	Self-confidence Perceived competence Self-efficacy Perceived behavioral control Beliefs Self-esteem Empowerment Professional confidence
Optimism	The confidence that things will happen for the best or that desired goals will be attained	Optimism Pessimism Unrealistic optimism Identity
Beliefs about Consequences	Acceptance of the truth, reality, or validity about outcomes of a behaviour in a given situation	Beliefs Outcome expectancies Characteristics of outcome expectancies Anticipated regret Consequents
Reinforcement:	Increasing the probability of a response by arranging a dependent relationship, or contingency, between the response and a given stimulus	Rewards (proximal/distal, valued/not valued, probable/improbable) Incentives Punishment Consequents Reinforcement Contingencies Sanctions

Domain	Definition	Constructs
Intention	A conscious decision to perform a behaviour or a resolve to act in a certain way	Stability of intentions Stages of change model Transtheoretical model and stages of change
Goals	Mental representations of outcomes or end states that an individual wants to achieve	Goals (distal/proximal) Goal priority Goal/target setting Goals (autonomous/controlled) Action planning Implementation intention
Memory, attention and decision processes	The ability to retain information, focus selectively on aspects of the environment and choose between two or more alternatives	Memory Attention Attention control Decision-making Cognitive overload/tiredness
Environmental context and resources	Any circumstance of a person's situation or environment that discourages or encourages the development of skills and abilities, independence, social competence and adaptive behaviour	Environmental stressors Resources/material resources Organizational culture/climate Salient events/critical incidents Person environment interaction Barriers and facilitators
Social influences	Those interpersonal processes that can cause individuals to change their thoughts, feelings, or behaviours	Social pressure Social norms Group conformity Social comparisons Group norms Social support Power Intergroup conflict Alienation Group identity Modelling
Emotion	A complex reaction pattern, involving experiential, behavioural, and physiological elements, by which the individual attempts to deal with a personally significant matter or event	Fear Anxiety Affect Stress Depression Positive/negative affect Burn-out
Behavioral regulation	Anything aimed at managing or changing objectively observed or measured actions	Self-monitoring Breaking habit Action planning

*British spelling of definitions is kept from original publication

Atkins L, Francis J, Islam R, O'Connor D, Patey A, Ivers N, Foy R, Duncan EM, Colquhoun H, Grimshaw JM, Lawton R, Michie S. A guide to using the Theoretical Domains Framework of behaviour change to investigate implementation problems. Implement Sci. 2017 Jun 21;12(1):77. doi: 10.1186/s13012-017-0605-9. PMID: 28637486; PMCID: PMC5480145.

APPENDIX C. CERQual assessment components

CERQual Components	Definition
Methodological limitations	The extent to which there are problems in the design or conduct of the primary studies supporting a review finding. We used our CASP assessment of each study to guide our assessment of this component.
Coherence	An assessment of how clear and cogent the fit is between the data from the primary studies and the review finding.
Relevance	The extent to which the body of evidence from the primary studies supporting a review finding is applicable to the context specified in the review question.
Adequacy	The degree of richness and quantity of data supporting a review finding.

Noyes J, Booth A, Lewin S, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 6: how to assess relevance of the data. Implement Sci. 2018 Jan 25;13(Suppl 1):4. doi: 10.1186/s13012-017-0693-6. PMID: 29384080.

APPENDIX D. Methodological appraisal ratings using the CASP tool

	Study				
CASP question	**O'Brien, 2020**	**Sylven, 2020**	**Heath et al. 2016**	**Ozanne, 2016**	**Pieters, 2016; Baca, 2015**
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the qualitative methodology appropriate?	Yes	Yes	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the recruitment strategy appropriate to the aims of the research?	No[1]	No[1]	No[1]	No[1]	No[1]
Was data collected in a way that addressed the research issue	Yes	Yes	Yes	Yes	Yes
Was the relationship between researcher and participants adequately considered?	Can't tell	Can't tell	Can't tell	Yes[2]	Can't tell
Were ethical issues taken into consideration?	Yes[3]	Yes[3]	Yes[3]	Yes[3]	Can't tell
Was there sufficient rigor?	Yes	Yes	Yes	Yes	Yes
Is there a clear statement of findings?	Yes	Yes	Yes	Yes	Yes
How valuable is the research?[4]	Yes	Yes	Yes	Yes	Yes
Overall risk to rigor	Low	Low	Low	Low	Low

Abbreviations: CASP: Critical Appraisal Skills Programme

[1] Participants in these studies were recruited after their child underwent surgery, raising concerns about selection bias due to selecting parents who were a) referred to surgery, b) proceeded with surgery, and c) experience some level of success with surgery. Excludes parents of a child with drug-resistant surgery who were not referred to surgery, who choose not to proceed with surgery, or whose child did not achieve desired outcomes.

[2] Interviewers and researchers not part of care/surgery team

[3] Approved by university ethics committee

[4] "Yes" rating given when study: a) discussed contribution to existing knowledge or understanding, b) identified new areas where research is necessary, and c) discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

APPENDIX E. Coding extracts for treatment decisions for childhood epilepsy into the Theoretical Domains Framework

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Knowledge	Baca, 2015, 25894906	With the recognition that "something is wrong," parents sought information from multiple sources in an attempt to understand seizures and epilepsy and become better advocates for their child.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Parents reported difficulties as the they sought information about seizures, epilepsy, medications, and surgery….Parents sought information from a variety of disparate sources including physicians, the Internet, books, and family/friends.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Through this journey, parents learned to navigate a complicated medical system filled with doctors of different specialties, multiple hospitals, insurance approvals, and numerous treatment options	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Pieters, 2016, 27521720	Parents also had to learn about the intricacies of the healthcare system including the types of doctors that were needed to care for their children	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Parents perceived the information they received from professionals as inadequate to make an informed decision. They therefore sought out additional information regarding the surgical procedure and outcomes. Aiming to comprehend what surgery would entail, parents requested information in lay language and searched the internet for information and videos.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	"I googled it until I couldn't google anymore and I think that gave me a better understanding of what was going on."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Finally, parents emphasized the importance of receiving adequate information regarding epilepsy surgery, including information presented in a variety of formats (e.g., booklets, photographs, videos). They also suggested that a 'frequently asked questions' information sheet would have been beneficial. "I know I got a little booklet about epilepsy surgery services, but it would have been nice for us to have some extra information… just some written up case studies of kids that have gone through, had the same, you know, operation."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	These included …additional information provision	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	O'Brien, 2020, 33120326	[I]t was felt that a greater awareness of potential changes besides seizure control prior to surgery would have been helpful for children and their families in developing realistic expectations and preparing for post-surgical adjustment. They just tell you medical stuff about it, the recovery, not that it'll be different…They just tell you about having no seizures […] They don't tell you what it's actually going to be like.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Sometimes, parents received more information from other parents, than from the physician. They were disappointed if they felt that information was withheld	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Parents thought that it was important to receive the information that surgery was aimed to treat the epilepsy and not other disabilities, such as behavioral problems, in order to have realistic expectations. They emphasized the importance of being informed about both positive and negative outcomes and risks. Even if surgery was a medical success, the family's life situation was	Need for information about epilepsy, treatment options, and navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text^A	Theme
		sometimes characterized by sorrow and problems. The deterioration, expected or unexpected, of e.g., motor function, speech, visual field, or attention made life more difficult and burdensome.	
Knowledge	Baca, 2015, 25894906	Parents also reported little initial familiarity with epilepsy and that they felt overwhelmed trying to learn a new language of epilepsy because of the "avalanche of information coming at us."	Overwhelmed by information
Knowledge	Pieters, 2016, 27521720	She described the hard and time-consuming struggle of trying to synthesize information on her own "because there really wasn't a lot out there and when you're hearing different things from different specialists it's very difficult for parents to make heads or tails of it". She spent "six months combing through the literature reading everybody's publications from places all over the world … because I wasn't getting a straight answer … my husband was saying to me you know are you going to go and get your degree in neuroscience after this … but you know it's your child, you want to do the best by them and you know you want them to be able to achieve their potential".	Overwhelmed by information
Knowledge	Baca, 2015, 25894906	As they acquired more epilepsy knowledge, however, many parents felt more empowered and able to ask questions and participate more effectively in the care of their child	Knowledge is empowering
Knowledge	Baca, 2015, 25894906	A mother who stated, "I just think knowledge is power and it also brings some comfort to making a good decision," exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy	Knowledge is empowering
Knowledge	Heath, 2016, 27371883	Increasing their own knowledge about medically intractable epilepsy and the available treatment options empowered parents to participate more fully in the decision-making process. "I said to him 'don't tell me in a doctor's term, tell it me in a parent's term. I want to know the ins and outs; I want to know exactly what the risks are but in terms that I understand'"	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	[P]arents took on the responsibility for learning as exemplified by the parent who naturally 'corrected' her language to imply her understanding of the different terms that clinicians use, "(the seizures) were not controlled … or intractable as you guys call them."	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	Being open to learn and learning itself occurred while parents became epilepsy experts in order to be effective advocates for their children	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	In order to have productive information exchanges with clinicians and engage in shared decision-making, parents realized that they had to learn about a very unfamiliar and unforeseen situation	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	The lack of understandable information and the experience of information imbalance between the parent and the physician were overwhelming as experienced by a father who described him and his wife as "pretty pragmatic." He said, "The beginning was terrible, because you're of course dealing with … specialists in things and that you've pretty much never even heard of …. It was tough in the beginning, really tough … (the information that we found) was written by doctors for doctors. It's kinda tough when you don't know what you're talking about to get through that.	Knowledge is empowering
Social role and identity	Heath, 2016, 27371883	However, they also reported a sense of duty to be "strong" (P1) and "brave" (P3, 4) for their children, wishing to protect them from their feelings of anxiety.	Role and responsibility as a parent
Social role and identity	Heath, 2016, 27371883	Parents discussed feeling pressure from a need to 'do the right thing' by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential	Role and responsibility as a parent

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Emotion	Baca, 2015, 25894906	Parents reported personal and family stress throughout the journey. Caring for children with a chronic, severe, and paroxysmal neurologic condition meant being "on call" all the time; they had to be prepared to respond to an emergency at all times	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Participant descriptions highlighted the significant impact that epilepsy had upon on the child's and family's lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: "you're tired yourself before you even go out the door" (Charlie – mother). In some cases, experiences of epilepsy impacted on their lives to such a degree that parents referred to children as "not having a life" (Charlie - mother)	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Child mood and behavioural difficulties-stress, secrecy and shame. Before surgery children's mood and behavioura difficulties were a common, causing fear and stress which negatively impacted upon family relationships: "If there was something there, she would get it, she would want to attack you with it". (Jessica – mother) Many described how increased supervision led to a change in their parenting role, negatively impacting on the relationship they or their other children had with their child: "I was just his carer really and didn't have a relationship with him" (Jack - mother)	Drained from managing epilepsy
Emotion	Baca, 2015, 25894906	Parents frequently described that medication trials continued often with little to no mention of surgery. One mother told of her frustrations with multiple medications, "It was, 'Let's work on all the different medical aspects or medicines before going to surgery or even talking about surgery."	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Parents felt desperate not only to find a treatment option that would work, but also to persevere and actively do something that was productive and necessary to help their child	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Wanted to play an active role	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[W]anted to use time constructively	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Another mother remembered that she felt "desperate" to take her adolescent son to another doctor, but she needed to wait for referrals because of insurance	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[P]arents felt frustrated and desperate to get help for their child.	Desperate to help their child
Emotion	Heath, 2016, 27371883	Following indication of surgery as a potential treatment, parents and families faced what they described as a "difficult decision" (P5) which was experienced as "daunting", "frightening", "scary", and "stressful"	Difficult and stressful decision
Emotion	Pieters, 2016, 27521720	"It was not only in terms of time, it was mentally draining as well. It took a lot of time and it took a lot of concentrated thought to figure this out."	Difficult and stressful decision
Emotion	Ozanne, 2016, 27176878	Parents were afraid and worried	Fear (unspecified)

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Emotion	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options. This perception was influenced by parental fear regarding the irreversibility of surgery, as well as by professionals who had informed them that surgery was the only remaining treatment for controlling their child's seizures	Fear of surgery
Emotion	Pieters, 2016, 27521720	For the majority of parents, brain surgery was a frightening possibility as described by the mother who said, “At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric.” Another mother's statement reflected upon her initial feelings of fear and avoidance when she said that she and her husband, “just couldn’t deal with (having our daughter go through brain surgery)	Fear of surgery
Emotion	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience “a change in personality” as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Fear of surgery
Emotion	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Fear of surgery
Emotion	Sylven, 2020, 32480305	Many parents were worried that their child's epilepsy would be “worse” after the surgery, and as a result, the operation would have been “in vain”. This included a risk that seizure frequency might increase, that seizures might be longer or be of a different character, or that seizure frequency might not improve.	Fear of surgery
Emotion	Ozanne, 2016, 27176878	It was demanding not to know if surgery would be possible, and the parents were thankful when the decision was finally made	Relief after decision was made
Emotion	Sylven, 2020, 32480305	Many parents described a general hope for improved behavior and emotional functioning for their child. Behavior encompassed a wide variety of outcomes including that the child would be “calmer” with fewer behavioral outbursts, have better attention and concentration, and “feel better”	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents hoped that the surgery would lead to better health and quality of life for their child	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents often expressed a desire that surgery would result in their child being “more alert”, less “tired”, and “have more energy”. They also mentioned that they hoped that surgery would result in better sleep quality	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including “tiredness” would lead to a “normal life” for their child	Hope for better health and quality of life
Emotion	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they “hoped” for candidacy	Hope for surgical candidacy
Emotion	Heath, 2016, 27371883	When a decision to accept surgery had been made, parents hoped for the procedure to be carried out as soon as possible. Waiting for a date for surgery was described as an anxious time for parents, who experienced relief when a date was set.	Hope for surgical candidacy

TDF domain	Author, Year, PMID	Extracted text^{A}	Theme
Social influence	O'Brien, 2020, 33120326	This sense of exclusion impacted on the family as a whole, with siblings being unable to attend clubs or invite friends to their house, and parents feeling that they "couldn't join in with what other families were doing" (Lucy – mother). A common theme was parental concern that siblings had less time or attention: as "the focus was on him [child with epilepsy] a lot of the time" (Andrew – mother).	Social exclusion
Social influence	O'Brien, 2020, 33120326	Difficulties appeared to be exacerbated by a perceived a lack of understanding from others, which appeared to be tied to a notion of epilepsy being an invisible condition, as seizures commonly occurred behind closed doors: "At the beginning, it was just generally at home, they didn't understand, they didn't get it. (Jessica – mother). Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. "Gosh, we didn't tell anybody. We were ashamed". (Jack – mother)	Social exclusion
Social influence	Heath, 2016, 27371883	In addition, parents emphasized the importance of accessing the experience and expertise of other parents who had followed a similar treatment pathway. Such peer support was suggested to facilitate understanding of the surgical procedure from a 'parental perspective', as well as provide emotional support. "If people could freely put their experiences on either a forum or even if you get a group, you know, parents talking to other parents who have gone through the experience to get a real idea of what's happening."	External support from peers and healthcare professionals
Social influences	Heath, 2016, 27371883	These included … peer support	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Those parents who were able to connect with other parents and families of children with epilepsy found it to be very helpful to be able to communicate with peers with seizures unresponsive to medication who were on the same journey.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	[E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Three key facilitators were identified as follows: (a) having a champion, (b) finding a good team of epilepsy providers, and (c) connecting with others who shared the experience of epilepsy. (Fig. 1; Table 2). Such facilitators played a key role during the times when parents especially needed support to act purposefully and move out of vulnerable periods.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	However, using time constructively happened after the parents described finding a champion that advocated for them and helped them navigate their journey. It was a champion, whether a doctor, nurse, or social worker, that facilitated steady and purposeful progress over more manageable portions of the journey	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	One mother favorably described one such champion, a nurse coordinator, "she's like wonderful for us. She's been our lifesaver." These champions helped parents get referrals and seek different treatment options or were the persons that believed them and pushed them to seek better or additional care. For one mother, her daughter's pediatrician acted as her champion. She said, "He encouraged me and gave me the confidence that if I wasn't happy with what five other pediatric neurologists were saying that it was okay to go and you know continue to seek out other opinions."	External support from peers and healthcare professionals

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Social influence	Heath, 2016, 27371883	Family decisions. Parents described decision-making as a family choice involving partners, siblings, grandparents, and the child (where developmentally appropriate). Discussing treatment options with family members was generally considered to be helpful. "I remember my mother-in-law was in Pakistan and I called her up and I was very teary…I felt like I had been punched in the stomach to be honest 'cause, you know, brain surgery is not something you take lightly and she said 'well if it's for his betterment and to improve his life then we will do whatever it takes'."	Family decision
Social influence	Heath, 2016, 27371883	For some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary carer, she 'witnessed' the true extent of their child's seizures. In cases where the child's epilepsy was considered to impact on the whole family (e.g., where a parent could not return to work because of a need to look after the child), then, the decision was understood as a responsibility of the whole family	Family decision
Belief about consequences	Baca, 2015, 25894906	On their journey, many parents transformed their view of surgery from a scary, last resort treatment to a necessary and hopeful option. One mother explained, "By the time she got it—it got so serious I was looking toward that as being an answer to our prayers rather than just something that I couldn't even think about." This transformation occurred as parents became increasingly aware of the severity of their child's epilepsy and its impact on their child's and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	Many parents initially perceived epilepsy brain surgery to be "pretty horrific" and a treatment of "last resort."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	After an arduous journey, parents reached the right doctor and center; epilepsy surgery, although previously seen as a last ditch option, became a viable option and a source of hope.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	After persevering through hard times with unsuccessful medications and interventions, they eventually decided to proceed with surgery, the option that they perceived to be the best treatment for the epilepsy.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	Because surgery was viewed as a treatment of last resort for many parents, time was required to process the initial fear, make sense of the emotional information, and consider the factual risks and benefits in order to decide on surgery as the best option for their child. Reflecting on why it took longer than six years to get to surgery for her 9-year-old daughter, a mother said, "Of course you need to take a lot of time to think about it. It's very invasive…. I did as much research as I could about it, and talked to the doctor a lot about it, and I decided that that was her best chance of having a normal life in the future."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge

TDF domain	Author, Year, PMID	Extracted text^{A}	Theme
Beliefs about consequences	Pieters, 2016, 27521720	Through internal processing, parents worked through the diverse emotions and large amount of information that they were receiving and gathering to move forward and thereby develop a greater understanding of their child's illness and prognosis in order to ultimately reach the point where surgery became the treatment of choice	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Parents were clear that putting their child forward for epilepsy surgery was the best and right decision for their child, demonstrating a high level of satisfaction with the treatment outcome. However, initial perceptions of surgery were often negative, particularly if it was not a treatment option that had previously been considered or that professionals had indicated. Many parents described attending their child's clinic appointment expecting to talk about more conventional treatments (e.g., medication or diet), leading to them feeling surprised when surgery was raised for discussion. "We went originally to talk about the ketogenic diet and then the doctor said 'oh I think surgery's your best option' which came as a bit of a shock really because no one had ever mentioned the surgery before." (P5) "It actually came as a bit of a surprise to us that option, in that it was mentioned in a review visit, that maybe the time had come to look at the option of surgery but we weren't really aware of that option at that point so it came as quite a shock to us." (P6) While some parents took comfort from the possibility of an additional treatment pathway, others felt excluded from the decision to assess their child for surgical candidacy, leading to their feeling unable to participate in an informed discussion	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Some parents were unconvinced about the efficacy of surgery for reducing their child's seizures. Skepticism stemmed from their experience of other pharmacological and dietary treatments which had resulted in little improvement in seizure control. In weighing up treatment options, parents also discussed the side effects of alternative treatments. For example, medications were perceived to have a range of adverse side effects, whereas ketogenic diets were viewed as negatively affecting the child's ability to live a 'normal' life. "We thought about the effects of the medication if we chose to continue on the path of finding a different medication… but then how long would that have taken us? Like how long would it work for? Then it would develop, then how long 'til we have to find something else, with all the side-effects that can happen? I didn't want to put her through that as well." (P9)	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	Balancing potential costs and benefits. When deliberating surgery, parents balanced their perceived necessity of the treatment in terms of how epilepsy currently affected and would, in the future, affect their child's quality of life, against concerns regarding the possibility of negative outcomes. In other words, parents weighed up the potential benefits of surgery such as freedom from seizures against what they understood as the risks, including cognitive impairments. "There was no quality of life for him; we had to do what was best. We were told of the risks of the surgery and we were told that actually this would be the best thing for him… we don't regret it, but it was, it's not an easy decision to take because you're worried about the risks of things."	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned that while they would like seizure freedom, any reduction in seizures would be "welcome" and a clear improvement for their child and for the family in general.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including "tiredness" would lead to a "normal life" for their child.	Balancing potential benefits of surgery with concerns of adverse effects

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Sylven, 2020, 32480305	Many parents mentioned a desire for seizure freedom for their child after surgery. They wanted their child to be free from the "trouble" of seizures and everything that follows from seizures	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	They wanted that their child could do everything their friends could do like "play football, cycle, and swim independently". Some parents mentioned that they wanted their child to be able to go back to school and have a "proper social life" while some parents referenced the future hoping that the child would be better placed to get a job and driving license as a result of surgery.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience "a change in personality" as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The majority of parents mentioned concerns about possible complications that could occur during or soon after the surgical procedure	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	A small number of parents said that they had no concerns before the surgery. One parent said that they were not worried as "no concerns arose in the discussions with professionals" before the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The results of the current study suggest that parents frequently worry about that the child would lose skills or function, or complications would arise during surgery that could result in injury.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	For all parents in this study, the perceived need of surgery outweighed the possible risks and side effects. "We thought this is going to be no life for him… He was on this diet which meant he couldn't go out with friends; he couldn't stay at friends' houses, all the things that other kids do, he couldn't do… and then he got these fits virtually every night and we thought… you know what, there is no decision to make, we gotta do it… We knew there were risks, we talked about them with [child] and he was frightened but he said no, we gotta do it." (P3)	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options…Some parents felt that they had little alternative but to accept surgical intervention in order to provide their child with the best possible chance of improving their quality of life. "We felt it was our only option really in her having a more independent future."	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	"My husband and I walked out of the appointment saying we feel like we don't have a choice… we felt like it was the only choice we could make."	Only choice left

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Heath, 2016, 27371883	For one family with an adolescent child, it was considered necessary to accept surgery while the child received care from the children's hospital as they believed that the treatment would become unavailable once transitioned to adult services.	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Parents described a process of weighing up the pros and cons based on the information provided by the medical team, which led to the decision to pursue surgery. Many perceived that surgery was the only hope "a simple decision" (Alex – mother), they were "in no doubt" (Lucy – mother) and that they "had no choice" (Jessica mother) as the health and wellbeing of their child would deteriorate or become more serious if surgery was not pursued. There was also a sense of 'not knowing' outcomes if surgery was not pursued. We felt we've got to do this because where is she going to end up if we don't?	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Decision making – is there a choice? When asked what informed the decision-making process when considering the option of surgery as a treatment for epilepsy, parents discussed how the perceived risks to their child's safety, as well as the safety of others, were key factors. Even with all the risks, it wasn't going to be much worse than having a little girl that was unconscious all the time anyway.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Information about possible complications related to surgery did not prevent the parents from wishing to proceed with the operation. They preferred to take a risk rather than live in constant fear	Only choice left
Environmental context and resources	Baca, 2015, 25894906	The journey to finding the right doctor was further complicated by parental perceptions that doctors sometimes lacked knowledge of or familiarity with epilepsy. Many parents went to numerous doctors searching for the "right" doctor, or one who had a strong epilepsy knowledge base, could effectively identify the problem, and then make a clear plan of action	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents expressed that their children were seen, evaluated, and cared for by a variety of doctors of different specialties throughout their journey. Parents frequently described difficulty finding the "right" doctor(s). Difficulty was associated with a lack of pediatric neurologists in their local area and with different doctors having different recommendations.	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents depicted a pathway filled with multiple doctors and treatments, insurance battles, and work and life obstacles as they sought a cure for their child's seizures, becoming ever more knowledgeable about epilepsy	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	Further, finding the best clinicians for the needs of their child required an understanding of the relationship between different medical (sub)-specialties and thereby represented dual and concurrent use of processing and navigating mechanisms	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	The second mechanism, navigating, was when the parents kept and directed their way along their journey often through uncharted territory as they steered through various aspects of the health-care system and learned about epilepsy itself and its various treatments.	Challenges with navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text^{A}	Theme
Environmental context and resources	Pieters, 2016, 27521720	A mother described her disappointment with being denied by insurance despite doing all the work that was required	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	"We'd already been fighting her seizures for two and a half, approaching three years, and because we were new there and per protocol, they wanted to try these other treatments for another year."	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	The Swedish Social Insurance Agency, technical aids center, school, and municipality were criticized by the parents. Based on their experience, they felt that the bureaucracy had been problematic and had taken a great deal of time. They had to fight for their rights, despite physicians' certificates. Without specific diagnoses of comorbidities, it was even more difficult to get adequate support. They thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	Parents were disappointed if it took a long time to get a correct diagnosis	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Several parents experienced feeling doubted by a doctor; some were told that nothing was wrong with their child initially	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they "hoped" for candidacy. This hoping required persistence and sometimes disregarding doctors who initially felt surgery was not an option. Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, "'I don't believe that she is a surgical candidate.' As a parent, I listened and I wanted to think otherwise.... In hindsight, I'm glad I didn't listen to him."	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	Parents also reported notable physician-based barriers including variability in perceived knowledge of epilepsy, criteria for presurgical referral and appropriateness of epilepsy surgery	Interactions with providers (barriers)
Environmental context and resources	Pieters, 2016, 27521720	[C]rossing such an information divide was an uphill battle as stated by a mother who described feeling dismissed by doctors until she found someone who was approachable and was "gracious enough to talk to a mere mortal mother on the phone … because a lot of docs won't, they've got lots of protective armor around them shielding off potential patients or worse yet parents."	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	Parents perceived a lack of control over the decision-making process through their limited involvement in initial discussions regarding surgical candidacy	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	These included earlier and softer introduction of surgery as a possible treatment option	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	During the long hospitalizations, some parents felt that nobody had time for them. Arrogance of some professionals made them very disappointed	Interactions with providers (barriers)

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Environmental context and resources	Ozanne, 2016, 27176878	If the pediatric neurologists revealed their own worries and insecurities regarding epilepsy surgery, this could have negative effects	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	Parents described that support and information were inadequate before the epilepsy was recognized to be drug-resistant, especially from the local hospital. Their concerns were not taken seriously	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	The support and information went from poor to strong when specialists at the university hospitals were involved	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	The pediatric neurologist's worry and sympathy for the family could also strengthen them since they felt invited and could be insecure together	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	Trust in the information that they received was greater when their child was in the hands of a team with different expertise, as exemplified by the mother who said, "They get all the doctors together to look at all the information … that felt a lot more helpful because if we were at a different hospital and you only had the one doctor, then they would have misdiagnosed him."	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	When parents finally reached a comprehensive center to seek evaluation for potential pediatric epilepsy surgery, they recognized the importance of having a medical team of experts	Interactions with providers (facilitators)

[A] Study participant (first order statement) or study author (second order statement). We kept original spelling from primary study.

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Parent perceptions and decision-making about treatments for epilepsy: A qualitative evidence synthesis

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Abstract

Objectives Epilepsy treatment decision-making is complex, and understanding what informs caregiver decision making about treatment for childhood epilepsy is crucial to better support caregivers and their children. We synthesized evidence on caregivers' perspectives and experiences of treatments for childhood epilepsy.

Design Systematic review of qualitative studies using a best-fit framework and Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual) approach**.**

Data sources Searched Embase, PubMed, CINAHL, PsycINFO, SocINDEX, and Web of Science from January 1, 1999, to August 19, 2021.

Eligibility criteria We included qualitative studies examining caregiver's perspectives on anti-seizure medication, diet, or surgical treatments for childhood epilepsy. We excluded studies not reported in English.

Data extraction and synthesis We extracted qualitative evidence into one of 14 domains defined by the Theoretical Domains Framework (TDF). One reviewer extracted study data and methodological characteristics, and two reviewers extracted qualitative findings. The team verified all extractions. We identified themes within TDF domains and synthesized summary statements of these themes. We assessed our confidence in our summary statements using GRADE-CERQual.

Results We identified 5 studies (in 6 reports) of good methodological quality focused on parent perceptions of neurosurgery; we found limited indirect evidence on parents' perceptions of medications or diet. We identified themes within six of the 14 TDF domains relevant to treatment decisions: knowledge, emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources.

Conclusions Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed on non-surgical treatments for epilepsy and among caregivers from different cultural and socio-economic backgrounds to fully understand the diversity of perspectives that informs treatment decision-making.

Strengths and limitations of this study

- This synthesis followed contemporary standards for the conduct of qualitative synthesis, which includes using a best-fit framework approach to categorize and synthesize findings based on the Theoretical Domains Framework (TDF).
- Risks to rigor of included studies were assessed using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies.
- Confidence in the conclusions drawn from this synthesis was rated using the Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE CERQual).
- As with all qualitative synthesis, selecting which data to extract and how to code it is ultimately a subjective process.
- Limited to studies that examined parents' perceptions, experiences, and decision-making about pursuing surgery to treat their child's epilepsy.

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Introduction

Epilepsy is a common neurologic disorder in children, affecting about 1% of children in the United States.[1] While there are different types of childhood epilepsy, each type involves recurring seizures caused by abnormal electrical activity in the brain. Epilepsy is categorized by seizure type (e.g., focal, generalized, or unknown), epilepsy type (e.g., focal, generalized, unknown), and syndrome type (e.g., childhood absence epilepsy, Dravet syndrome).[2-4] No matter the type, epilepsy is a chaotic and unpredictable condition for both the affected children and their caregivers.[5]

Treatment and ongoing management approaches depend on the type of the epilepsy and prior treatment response. Treatment options for childhood epilepsy include anti-seizure medications (ASMs), ketogenic diets, or surgery. Although many children with new-onset epilepsy achieve seizure freedom with ASMs,[6] these drugs are associated with numerous adverse effects (e.g., tiredness, nausea, headache, difficulty concentrating, depression, and suicidal ideation).[7] Furthermore, about 20% of children continue to experience seizures despite drug treatment.[8] The effectiveness of ketogenic diets (e.g., Atkins diet) is supported by low quality evidence.[9] However, these diets are also associated with adverse effects (e.g., gastrointestinal symptoms, dyslipidemia, decreased growth, and kidney stones), and require considerable caregiver effort to maintain. Surgical interventions may isolate and remove the underlying neurological cause of seizures, but carry risks of bleeding, infection, hydrocephalus, and new neurologic deficits.

Epilepsy treatment decision-making is thus complex and needs to consider each child's unique form of epilepsy, evidence of each treatment's potential benefits and harms, previous or ongoing experiences with treatment(s), and family's values and preferences. Although studies of treatment effectiveness rarely explore how families navigate these complex considerations or how these considerations may evolve over time, qualitative studies can offer context on caregiver experiences and decision-making. Syntheses of these qualitative studies can help shape caregiver and healthcare provider interactions and inform shared decision-making tools and processes. The purpose of this review is to summarize the qualitative research regarding caregivers' perspectives and experiences of treatments for childhood epilepsy.

Methods

We conducted a qualitative evidence synthesis in accordance with standards for qualitative evidence syntheses.[10-12] Patients or the public were not involved in the design, conduct, reporting, or dissemination of our synthesis.

Data Sources and Searches

We searched EMBASE, PubMed (in process), CINAHL, PsycINFO, SocINDEX, and Web of Science for studies published from January 1, 1999, to August 19, 2021 using terms related to epilepsy, treatments, and caregiver perspectives (Appendix A). The search was limited to English-language. We also ran forward and backward citation searches (i.e., snowball searching) on included studies to ensure relevant studies were not missed.

Study Selection

We included qualitative studies that sought to understand caregiver's perspectives on ASMs, diet, or surgical treatments for childhood epilepsy. Studies needed to use qualitative methods for both data collection (e.g., focus group, individual interviews, or open-ended survey questions) and data analysis (e.g., thematic analysis). We excluded studies that focused only on non-caregiver perceptions (e.g., patient or healthcare provider) or that focused only on caregiver stress or caregiver expectations of treatment. We did not restrict by study country or care setting.

One reviewer screened titles and abstracts of citations retrieved from searches for eligibility. Two independent reviewers screened the full text of potentially relevant citations in PICO Portal© (https://picoportal.org/). Disagreements were resolved through discussion of the full team.

Data Extraction and Quality Assessment

For each study, we extracted details on the study design and methodological features, population characteristics, and qualitative analysis findings.

One reviewer assessed risks to rigor using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies, which appraises research aims, congruence between research aims and methodological approach, quality of sampling and data collection, appropriateness of application of methods, richness and conceptual depth of findings, appropriateness of interrogation of findings, and researcher reflexivity.[13] All team members reviewed the CASP assessments to ensure consistency of ratings across studies.

Two team members independently extracted and coded the qualitative findings of the included studies in MaxQDA© 2020 (Berlin, Germany), an online platform designed to support qualitative data management, extraction, and analysis. The extracted data included direct quotes from the participants (first order statements) and summary statements written by the study authors (second order statements). Extracted data were imported into spreadsheets to facilitate data cleaning, confirmation of themes, and synthesis.

To categorize the extracted data, we used the Theoretical Domains Framework (TDF),[14, 15] which was developed to assist in identifying the cognitive, affective, social, and environmental factors that may influence an individuals' performance of a health behavior. The health behavior of interest for this review was the decision to pursue an epileptic treatment. The 14 domains include: knowledge; skills; social/professional role and identity; belief about capabilities; optimism; beliefs about consequences; reinforcement; intention; goals; memory, attention, and decision processes; environmental context and resources; social influences; emotion and behavioral regulation (Appendix B). A third reviewer confirmed TDF domain codes and the team discussed the coding to ensure accuracy and consistency both within and across TDF domains. One reviewer did a final confirmation of extracted text and coding to ensure no data were missed and that there was consistency across domains.

Data synthesis and analysis

We adopted a best-fit framework approach to guide our qualitative synthesis. In this approach, data are coded according to TDF domains. One reviewer then summarized key themes within each TDF domain. Themes were discussed and debated among the team until consensus was achieved. We used the finalized themes to develop summary statements and assessed our confidence in these statements using Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual).[16-18] We rated our confidence in the summary statements as either high, moderate, low, or very low based on our assessment of the four GRADE-CERQual domains: methodological limitations, relevance, coherence, and adequacy of the data (Appendix C for definition of domains). "High confidence" refers to a finding that is highly likely to be a reasonable representation of the phenomenon of interest, whereas "very low confidence" refers to a review finding in which it was unclear if the finding was a reasonable representation.

Patient and public involvement: Patients or the public were not involved in the design, or conduct, or reporting, or dissemination plans of our research.

Results

The literature search yielded 1,447 citations from searches (see PRISMA flow diagram, Figure 1). We found 66 citations to retrieve for full-text screening of which five studies (in six publications) were included in the final sample.[19-24] Table 1 presents characteristics of included studies.

All studies examined parents' perceptions, experiences, and decision-making leading to surgery. All studies were conducted after the children had undergone surgery, with one study also surveying parents "just prior" to surgery.[24] Three studies reported data on parents' perceptions and experiences with medications and diet, but only in the context of selecting surgery (e.g., parents considering surgery due to the undesirable side effects or uncontrolled epilepsy with prior treatments).[19, 21, 24] In total, the views of 186 parents are represented in the five studies (the majority of whom were mothers, n=115). The time from children's surgery to data collection ranged from 6 months to 10 years among the three studies reporting on timing. Four studies collected parent perspectives through semi-structured interviews conducted in-person or over the telephone (length ranging from 10 to 75 minutes),[19-23] and one study captured parent perceptions through open-ended survey questions.[24]

All studies were assessed to have minor risk to rigor (Appendix D) due to the retrospective nature of recruitment and the lack of consideration (or reporting) of the relationship between researchers and participants. Otherwise, studies were considered appropriate in their use of qualitative design, methods of data collection, and analysis. The retrospective nature of the included studies raises concerns about recall bias, as surgical outcomes may have affected retrospective perceptions, and selection bias since studies only included parents of children who were referred to surgery and proceeded with surgery (and possibly experienced some level of success with surgery). All but one study[23] did not report whether interviewers were part of the child's care team, which may have influenced responses. Ozanne et al. reported that the researchers that interviewed parents and analyzed the data were not part of the epilepsy surgery team.[23]

We identified and coded data for six of the 14 TDF domains: knowledge; emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources (Figure 2). We did not find evidence from extracted qualitative data for the remaining eight TDF domains. Appendix E provides the extracted text from studies linked to their synthesized themes.

The GRADE-CERQual table (Table 2) summarizes findings and conclusions for each TDF domain. We had no or minor concerns with the coherence of the findings (i.e., the synthesized findings reflects the complexity and variation of the data) or their relevance (i.e., the extent to which synthesized findings are applicable to the context specified in the review question). We had minor or moderate concerns with the adequacy (i.e., the degree of richness and quantity of the data supporting the synthesized finding) of the findings related to knowledge and environmental context and resources, respectively. Below, we report the summary statement (and associated GRADE-CERQual level of confidence) under each identified TDF domain, and summarize the key themes that contributed to the statements.

TDF Domain 1: Knowledge

Summary statement: *Evidence from four studies indicated that caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes contradictory information, caregivers value learning this new language so they can become better advocates for their child (Moderate confidence)* [19-23]

Once parents recognized “something [was] wrong” with the health of their child, they sought information from multiple sources “to understand seizures and epilepsy and become better advocates for their child.”[19] Parents wanted information about medications,[19] surgery,[19, 21] and what types of doctors were needed to care for their child.[20] They also noted needing to learn about how to navigate the health system including multiple specialties, hospitals, and insurance procedures.[19]

Parents often perceived the information they received from professionals as inadequate to make informed decisions.[21] They described seeking lay language information from multiple sources, including additional professionals, the internet, books, family, friends, and other parents.[19, 23] Parents expressed discomfort in the “lack of understandable information” and “information imbalance” between themselves and their child’s providers and sought to become “epilepsy experts in order to be effective advocates for their children.”[20] After increasing their knowledge, parents felt more empowered to ask questions and participate more actively in the decision-making.[19, 21] Parents thus described active learning to ensure productive information exchanges with providers (including using clinical terms) to support a positive shared-decision making process.[20]

TDF Domain 2: Social role and identity

Summary statement: E*vidence from one study indicated that caregivers feel a sense of duty and need to do the right thing in selecting a treatment for their child (Moderate confidence).*[21]

Parents reported a “sense of duty” to be “strong” and “brave.” They also reported feeling pressure to “’do the right thing’ by their child; that is, choosing the treatment option that would give their child the best chance of “reaching their full potential.”[21]

TDF Domain 3: Emotion

Summary statement: *Five studies provided evidence to suggest that caregivers experience the journey of navigating their child’s epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation, fear to relief, and hope (Moderate confidence).*[19-23]

Parents expressed feeling drained and stressed from always being “on call,”[19] worrying about a seizure,[19, 22] or mood and behavioral difficulties.[22] Parents recalled feeling frustrated[19, 20] and desperate to “find a treatment option that would work.”[20] For example, one mother was frustrated that her child’s provider continued to perform medication trials to manage seizures and not discuss surgery as an option,[19] while another mother was desperate to get her son to another doctor but needed to wait for referrals because of insurance.

When considering surgery as a potential treatment option, parents reported the decision process as “difficult,” “frightening,” and “stressful.”[20, 21] Parents feared surgery would lead to worse health outcomes for their child, [21, 24] change their child’s personality, or cause a loss of function (e.g., partial or complete loss in speech or movement).[24] Parents also feared the possibility of surgery making seizures worse (e.g., increased frequency, duration). Thus, making the decision to have surgery “in vain.”[24]

However, for many parents, the decision to pursue surgery brought relief and feelings of hope. Parents in one study expressed gratitude once they finally decided to pursue surgery as they felt it was "demanding to not know if surgery would be possible."[23] Parents also reported feeling hope for improvements in their child's health and wellbeing after they decided to choose surgery. Finally, after making the decision to pursue surgery (or learning it was a viable option), parents reported that they hoped for candidacy,[19, 21] and experienced relief when a date was set.[21]

TDF Domain 4: Social Influence

Summary statement: *Three studies provided evidence to suggest that surgery is a family decision that requires outside support from other caregivers "experiencing the same thing" and from a healthcare professional acting as "champions." (Moderate confidence)* [20-22]

Parents reported the value of connecting with other parents who were on a similar treatment journey (e.g., had a child with epilepsy that was unresponsive to medication) and had experiences and expertise that they could draw on.[20, 21] Peer connections helped parents understand the surgical procedural from a "parental perspective" and provided "emotional support."[21] Parents also reported the value of having a good team of epilepsy providers and a "champion" (e.g., doctor, nurse, or social worker) to advocate for them and help them navigate purposefully through their journey.[20] One mother described one such champion, a nurse coordinator, as a "life saver," who helped in getting referrals and pushed her to seek better care for her child."[20] Another mother described her child's pediatrician as her champion because "he encouraged me and gave me…confidence."[20]

Parents described seeing treatment decision-making as a family choice and indicated the importance of involving partners, siblings, grandparents, and the child (where developmentally appropriate). Parents found engaging the whole family in the decision-making process to be generally helpful, as the experience of the child's epilepsy and potential consequences of treatment impacted the whole family.[21] However, for some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary parent, she "witnessed" the true extent of their child's seizures.[21]

TDF Domain 5: Beliefs about Consequences

Summary statement*: Five studies provided evidence to suggest that parents undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.* [19-24]

Parents of children who went on to have surgery reported going through a transformation in thinking of surgery as a "last resort" to a "necessary and hopeful option."[19] This transformation in thinking evolved as parents acquired greater understanding about their child's illness and prognosis, experience in treating their child with other treatments, acquired knowledge about the potential risks and benefits of surgery, and processed the diverse emotions associated with surgery.[19, 20]

An important part of parents' moving toward a decision about surgery was weighing what they perceived to be the benefits and harms of surgery.[21, 24] In terms of benefits, parents hoped surgery would lead to improved outcomes such as seizure reduction and the opportunity for a "normal life" without the side effects of medications.[21, 24] Some parents were skeptical about the efficacy of surgery due to their experiences with medications and the ketogenic diet.

According to parents, these treatments were associated with side effects, negatively affected their child's quality of life, and provided only minimal improvement in seizure control.[21] In terms of adverse effects, parents were concerned that surgery would lead to surgical complications or various post-surgical impairments, including a change in personality, loss of speech or motor function, and behavioral problems.[24] A small number of parents reported having no concerns before surgery.[24]

Beyond rational consideration of the benefits and risks of surgery, parents often reported coming to the decision to select surgery only after exhausting all other treatment options. [21-23] Parents expressed feeling like "it was the only choice [they] could make"[21] and their "only option"[21] in improving their child's outcomes or preventing their condition from getting worse. Parents understood there were potential complications associated with surgery, but "preferred to take a risk [in proceeding with surgery] rather than live in constant fear"[23] or having a child that "was unconscious all the time."[22]

TDF Domain 6: Environmental Context and Resources

Summary statement: *Three studies provided evidence that parents face challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process (Low confidence)*[19-21, 23]

Parents from one study in the U.S.[19, 20] and another in Sweden.[23] reported experiencing significant barriers with navigating the healthcare system. In the U.S.,[19, 20] parents expressed frustration with the extensive time it took to find the right doctor after navigating various doctors from different specialties across multiple institutions. Prior to selecting surgery for their child, parents reported difficulties in finding the "right doctor" with knowledge to "effectively identify the problem, and then make a clear plan of action." Parents attributed these difficulties to the lack of pediatric neurologists in their local area, inconsistencies in treatment recommendations, and rigid adherence to center-specific treatment protocols.[19, 20] Once parents made a decision to pursue surgery, they reported battles with insurance companies to pay for surgery.[19, 20] Parents in the Sweden reported similar frustrations with "the bureaucracy" as they felt it took "a long time to get a correct diagnosis" and "adequate support." They "thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary.[23]

Parents listed several provider-specific interactions they found to be either enablers or barriers to their experience of identifying and selecting appropriate treatment for their child. With respect to enablers, parents valued when providers validated (and shared) their concerns, gave their time and fostered trust, and engaged parents in the treatment decision-making process. Barriers noted by parents included having their concerns doubted or ignored,[19, 23] receiving inadequate information or support (especially before the epilepsy was recognized to be drug-resistant),[23] and feeling excluded from discussions about their child's surgical candidacy.[21] One study reported that parents perceived physician variability in knowledge about epilepsy and their lack of understanding about the pre-surgical referral process and appropriateness of surgery as barriers.[19] Parents in one study reported how they felt more reassured when their child received care from a comprehensive team of professionals with diverse expertise.[20] Parents from one study suggested providers give an "earlier and softer introduction of surgery as a possible treatment option."[21]

[Box 1 goes here]

Discussion

Understanding caregiver perspectives about treatments for childhood epilepsy is important to ensure that parents are appropriately supported during their decision-making process. Our qualitative evidence synthesis of five studies, which had minimal risks to rigor, identified key findings across six domains: Knowledge, Emotions, Social/Professional Role and Identify, Social Influences, Beliefs about Consequences, Environmental Context and Resources. However, the evidence was limited surgical treatment. Overall, these findings suggest that the decision to select surgery for childhood epilepsy involves parents going through a complex journey of acquiring extensive knowledge, working through intense emotions and perceived parental responsibilities, needing family and peer support, transforming beliefs about epilepsy and potential treatment options, and navigating various barriers and facilitators of the healthcare system.

These findings are supported by a similar review conducted by Samanta et al. (2021) that sought to understand caregiver decision-making around epilepsy surgery for children with drug-resistant epilepsy. [25] In their synthesis of a similar body of evidence, these reviewers identified the following as key determinants: knowledge and information, communication and care coordination, caregivers' emotional state, and socioeconomic factors. While the findings of our review are similar to Samanta, our scope and methodology differ. We sought to explore caregiver decision-making around all treatments for epilepsy, not just surgery. We also used rigorous methodological tools (e.g., TDF and GRADE-CerQual). Use of these tools strengthens our findings by placing them within a framework that identifies facilitators and barriers, and by providing criteria to establish confidence in the certainty of these findings.

Through the TDF framework, we identified decision domains that presented potential barriers to care that are amenable to action. For example, parents often felt that they did not have enough information to make decisions about surgery. They reported experiencing intense psychological distress and exhaustion during their decision-making process and felt these emotions acutely through their role as parents responsible for making a potentially life-altering decision for their child. Parents also reported valuing connections with peers who were going through (or had gone through) the same experience. Healthcare systems and providers may therefore consider providing parents with resources, such as patient navigators to help guide them through the healthcare system and better understand the care pathway. They can also help parents to access peer support and advocates[26, 27]

The findings also identified gaps in the evidence base regarding parents' perceptions and decision-making processes. Foremost, we did not identify any studies exploring parent decision-making around non-surgical treatment options. Qualitative studies that prospectively explore caregiver decision-making about these treatments are needed to determine if perspectives differ from those about surgery. We also did not identify evidence mapping to certain TDF domains that we expected to find evidence for, such as Goals. Studies included in this review were vague and inconsistent in reporting parent's goals or their desired outcomes of treatment. Some studies noted that parents wanted what 'was best' for their child or they would be happy with a reduction in seizure frequency. Survey data collected from parents considering surgery found the primary goal was seizure freedom (98%), followed by reduced medication (90%), and improved cognition (82%).[28] We also did not identify evidence for the domain of Memory, Attention, and Decision process. Thus, we could not determine if parents become more skilled and confident in their decision-making over time through experience and acquired knowledge.

Similarly, we found limited evidence for the domain of Environmental context and Resources. Further evidence is needed to understand how culture and equity play a role in parents' perceptions about treatment and their capacity to access care for their child. For example, many parents' reported important barriers in knowledge, access to professionals to diagnose and treat their child, and challenges with accessing and paying for surgery. The extent to which these factors would be the same or increase among families from different socio-economic backgrounds and cultures with varying economic, educational, and social resources needs further exploration. One may hypothesize that parents' perceptions of treatments for epilepsy vary depending on cultural perspectives of the condition and availability and access to care.

Strengths and limitations

This review followed contemporary standards for the conduct of qualitative synthesis, which includes the use of a best-fit framework approach, a theory-informed framework to guide our synthesis and extraction, and use of the CASP and GRADE-CerQual tool to assess the rigor and confidence of our findings. The use of the TDF is a particular strength of this review as it lends itself to both actionable interventions (e.g., mapping intervention strategies to key domains identified) and future research (e.g., further examination of domains not identified in the evidence such as 'memory, attention, and decision processes'). However, as with all qualitative research, selecting which data to extract and how to code it is ultimately a subjective process. We attempted to limit subjectivity within our group by coding in duplicate and having regular meetings to ensure consistency across and within domains.

One primary limitation is that the data in all studies included in this were collected retrospectively. Parents were asked for their perceptions *after* their child had undergone surgery. This may have resulted in selection bias, as studies recruited parents of children referred to and proceeded with surgery. Thus, findings may not be fully reflective of the wider population of parents who are making decisions regarding surgery. The perceptions of parents who declined surgery were not captured. The retrospective nature of the data collection may have also resulted in recall bias. The time from children's surgery to data collection ranged from 6 months to 10 years. Parents may have forgotten important aspects of their journey to surgery or filled in gaps of memory due to experiences with the child's outcome. As described by one study, parent responses were "memories processed through emotions and colored from further experiences, which were then developed into opinions and personal views."[23]

Conclusion

Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed to fully understand the diversity of experiences of parents across various points in the decision-making pathway and among different healthcare contexts. In particular, qualitative studies are needed that address parents' perceptions and experiences with selecting non-surgical epilepsy treatment options such as diet and medications.

Footnotes

Contributors: SU served as project lead and participated as a team member in identifying studies for inclusion, extracting and coding data, synthesizing findings, developing and grading summary statements, and drafting of initial report and manuscript; KK participated as team member in all aspects of data extraction, coding, synthesis, developing and grading summary statements, validating all findings, and revising initial drafts of manuscript; RW participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; GA coordinated and revised search strategies and participated in all aspects of data extraction, coding, synthesis, and review of manuscript; KR participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; MV participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript.

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Box 1. Example excerpts for TDF domains

Box 1. Example excerpts for TDF domains

Knowledge

- [Parent]: “I googled it until I couldn’t google anymore, and I think that gave me a better understanding of what was going on.”[21].
- A mother who stated, “I just think knowledge is power and it also brings some comfort to making a good decision,” exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy[19]

Social/Professional Role and Identity

- Parents discussed feeling pressure from a need to “do the right thing” by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential.[21]

Emotion

- Participant descriptions highlighted the significant impact that epilepsy had upon on the child’s and family’s lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: “you’re tired yourself before you even go out the door”[22]
- [Parent]: “At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric”[20]

Social Influences

- Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. “Gosh, we didn’t tell anybody. We were ashamed.”[22]
- [E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information” [20].

Beliefs about Consequences

- This transformation occurred as parents became increasingly aware of the severity of their child’s epilepsy and its impact on their child’s and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment [19]
- They wanted that their child could do everything their friends could do like “play football, cycle, and swim independently”. Some parents mentioned that they wanted their child to be able to go back to school and have a “proper social life” while some parents referenced the future hoping that the child would be better placed to get a job and driving license as a result of surgery[24]
- Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.[23]

Environmental Context and Resources

- Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.[23]
- Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, “‘I don’t believe that she is a surgical candidate.’ As a parent, I listened and I wanted to think otherwise.... In hindsight, I’m glad I didn’t listen to him.”[19]

Table 1. Characteristics of included studies

Study, year, country	Population	Number of parents	Time from surgery to data Collection	Study aim	Data collection methods	Data analysis methods
Baca[1], 2015; Pieters, 2016, USA	Parents of children who had previously undergone resective epilepsy surgery	37 (individual parents) 31 mothers; 6 other	NR	Baca, 2015: To identify the nature and range of parent-perceived barriers to timely receipt of pediatric epilepsy surgery. Pieters, 2016: A) To describe thoroughly the parental experiences and perceptions of this slow and arduous period prior to the presurgical referral and evaluation to a comprehensive pediatric epilepsy center. B) To delineate the range of parent identified factors, or facilitators, that helped move families forward along their journey to surgery	Semi-structured interviews lasting on average 29 minutes (range 10 to 60 mins)	Thematic analysis
Heath, 2016, UK	Parents of children who had undergone pediatric epilepsy surgery Also interviewed healthcare professionals caring for children with epilepsy	9 (individual parents) 8 mothers; 1 father 10 healthcare professionals	NR	To explore how parents and health professionals make decisions regarding pediatric epilepsy surgery to identify: A) factors that influence the process of decision-making regarding pediatric epilepsy surgery from the perspective of parents and professionals and B) the support needs of those considering surgery as a treatment option for a child with medically intractable epilepsy	Semi-structured interviews lasting about 1 hour, and non-participant observations	Thematic analysis
O'Brien, 2020, UK	Parents and their children who had gone through resective epilepsy surgery	9 parents (1 couple; 7 individual parents) 8 mothers; 1 father	6 months to 3 years	To explore children's and parents' perspectives on the journey prior to and following surgical treatment, with a focus on the emotional experiences of children and their parents throughout the surgery journey	Semi-structured interviews lasting 25 to 75 mins	Thematic analysis
Ozanne, 2016, Sweden	Parents of children who had previously undergone resective epilepsy surgery	24 parents 13 mothers; 11 fathers	5 to 10 years	To explore parental experiences before and after hemispherotomy as reported at a long-term follow-up and the parents' views on received information and support	Interviews (length of time not reported)	Content analysis
Sylven, 2020, Sweden	Parents of children who were undergoing resective epilepsy surgery (and 2 years post-surgery)	107 (parental responses to both surveys)	2 years	A) To understand parental hopes and worries before their child underwent epilepsy surgery and B) To understand parental satisfaction two years after their child had undergone epilepsy surgery	Surveys (open-ended questions)	Thematic analysis

[1] Baca & Pieters included the same patient population but reported on different aspects of parent perspectives.

Table 2. GRADE-CERQual summary of findings statements and ratings

Summary of Review Findings	Studies contributing	Methodological limitations (CASP ratings)	Coherence	Adequacy[2]	Relevance	CERQual assessment	Explanation of CERQual assessment
Knowledge: Caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes contradictory information, caregivers value learning this new language so they can become better advocates for their child	Baca/Pieters Heath O'Brien Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concerns*: Included studies limited to surgery as treatment decision. We did not identify studies meeting inclusion criteria that addressed other treatments, such as diet or medications. Each study addressed knowledge acquisition and reported on it in some depth.	*No or very minor concerns*	*Moderate confidence*	Four studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Social/professional role and identity, parents feel a sense of duty and need to do the right thing in selecting a treatment for their child.	Heath	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concern:* Included studies limited to surgery as treatment decision.	*No or very minor concerns*	*Moderate confidence*	One study; minor concerns related to methods and limited treatment scope (surgery).
Emotion, parents experience the journey of navigating their child's epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation and fear to relief and hope.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*: All 4 studies contributed to the rich depth of the varied emotions from fear of the illness to fear of the surgery to the mental toll that took on families and hope	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).

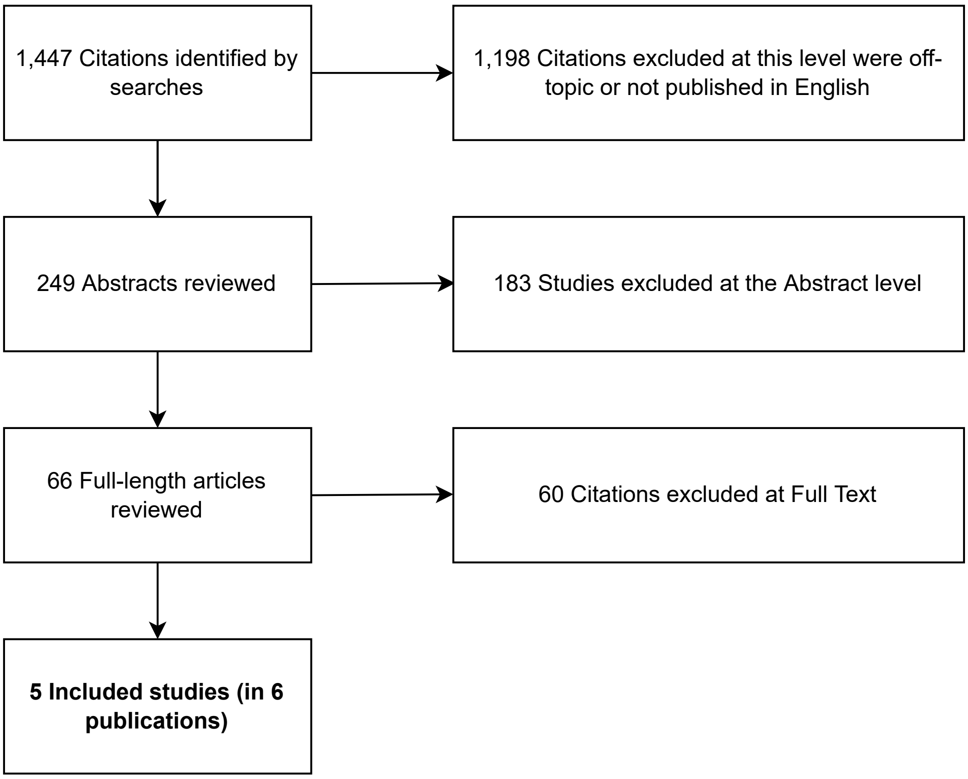
Summary of Review Findings	Studies contributing	Methodological limitations (CASP ratings)	Coherence	Adequacy[2]	Relevance	CERQual assessment	Explanation of CERQual assessment
				and relief that surgery may offer			
Social influences, surgery is a family decision that requires outside support from other parents "experiencing the same thing" and from a healthcare professional acting as "champions."	Pieters Heath O'Brien	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Three studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Beliefs about consequences, caregivers undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Environmental context and resources, parents expressed challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process.	Baca/Pieters Heath Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*Moderate concerns*: The three studies are limited to surgery and to US, Sweden and the UK. Healthcare barriers seemed particularly salient to U.S. respondents (including insurance barriers)	*No or very minor concerns*	*Low confidence*	Three studies with similar findings; moderate concerns related to methods, limited scope (surgery), and context-specific healthcare experiences of participants

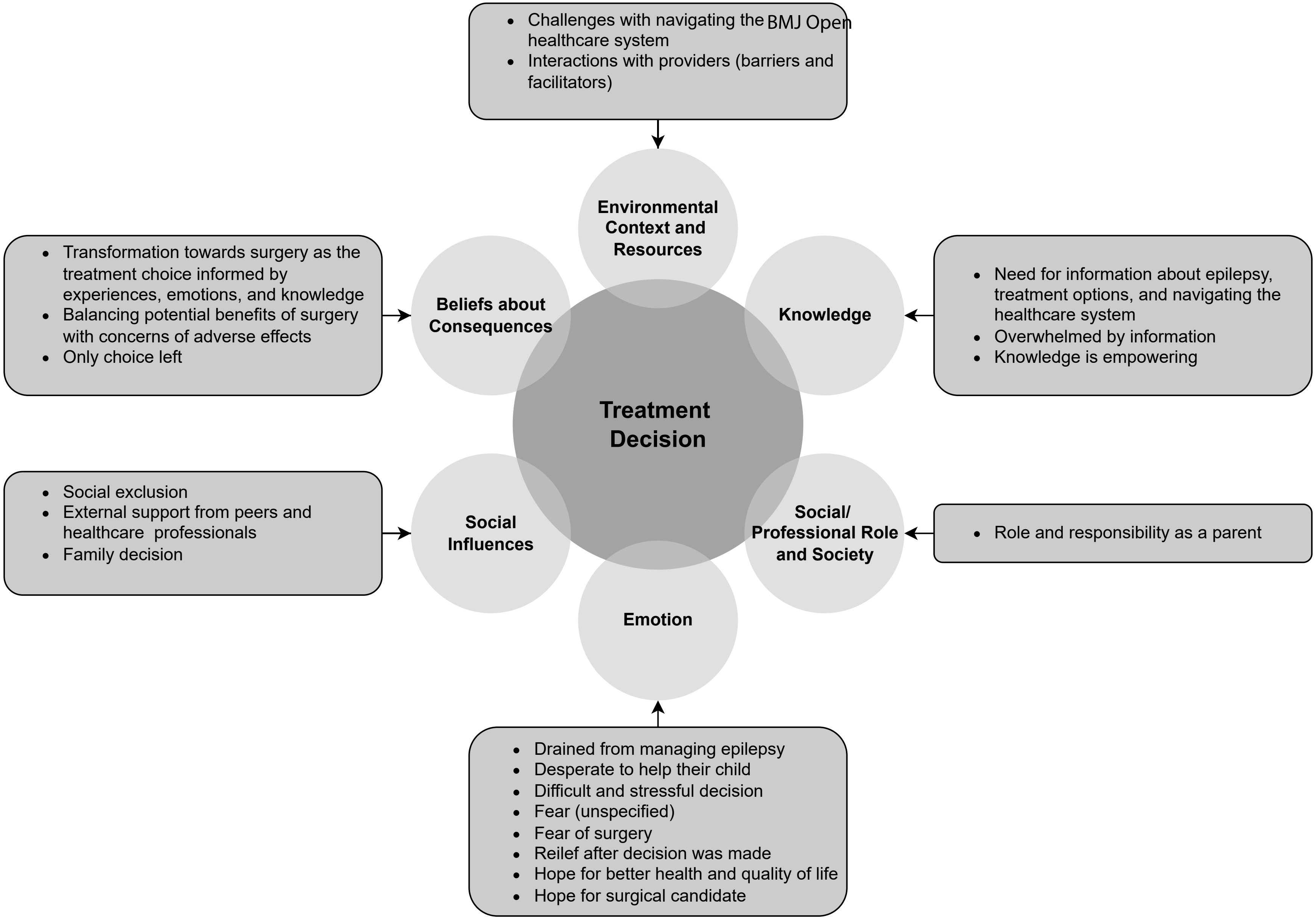
[1] Retrospective recruitment raises concerns about selection bias.

[2] We only downgraded for adequacy due to studies being limited to surgery when the summarized theme was based on parent's general feelings about epilepsy and could thus potentially apply to parent decisions regarding other treatment choices.

Figure 1 Legend: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram of studies identified

Figure 2 Legend: Figure 2 presents the six theoretical domains identified in this review that impacted treatment decision-making and presents the main themes representing those domains


1,447 Citations identified by searches
1,198 Citations excluded at this level were off-topic or not published in English
249 Abstracts reviewed
183 Studies excluded at the Abstract level
66 Full-length articles reviewed
60 Citations excluded at Full Text
5 Included studies (in 6 publications)


Challenges with navigating the healthcare system
Interactions with providers (barriers and facilitators)
Environmental Context and Resources
Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Balancing potential benefits of surgery with concerns of adverse effects
Only choice left
Beliefs about Consequences
Knowledge
Need for information about epilepsy, treatment options, and navigating the healthcare system
Overwhelmed by information
Knowledge is empowering
Treatment Decision
Social exclusion
External support from peers and healthcare professionals
Family decision
Social Influences
Social/ Professional Role and Society
Role and responsibility as a parent
Emotion
Drained from managing epilepsy
Desperate to help their child
Difficult and stressful decision
Fear (unspecified)
Fear of surgery
Reilef after decision was made
Hope for better health and quality of life
Hope for surgical candidate

APPENDIX A: Search Strategy

EMBASE (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Infantile/ Pediatric Epilepsies	'benign childhood epilepsy'/exp OR 'childhood absence epilepsy'/exp OR 'severe myoclonic epilepsy in infancy'/exp OR (dravet* NEXT/1 (disease OR syndrome))
2	0-3 Age Group and Epilepsy	[infant]/lim OR [newborn]/lim OR newborn/exp OR [preschool]/lim OR 'preschool child'/exp OR toddler/exp OR (babies OR baby OR child*:ti OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR 'very young'):ab,ti,kw OR ('younger than' OR under OR below) NEAR/3 (3 OR three) OR (3 OR three) NEAR/3 ('or below' OR 'or under' OR 'or younger') AND ('epilepsy'/exp OR 'epileptic patient'/exp OR epilep*:ti)
3	Infantile Spasms/ Neonatal Seizures (not requiring epilepsy)	'infantile spasm'/exp OR (((infan* OR neonat* OR newborn*) NEAR/2 (convuls* OR seizure* OR spasm*)):ab,ti,kw)
4	Other Seizures (not requiring Epilepsy)	([infant]/lim OR [newborn]/lim OR 'newborn'/exp OR babies:ab,ti,kw OR baby:ab,ti,kw OR infan*:ab,ti,kw OR neonat*:ab,ti,kw OR newborn*:ab,ti,kw OR nicu:ab,ti,kw) AND ('febrile convulsion'/exp OR 'seizure'/exp OR convuls*:ab,ti,kw OR spasm*:ab,ti,kw OR seizure*:ab,ti,kw)
5	Pharmacologic/ Vitamin Treatment	acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide
6	Diet Therapy	'ketogenic diet'/de OR (keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic'):ab,ti,kw
7	Surgical Procedures	craniotomy/de OR hemispherectomy/de OR 'laser surgery'/de OR lobectomy/de OR 'corpus callosotomy' OR craniotom* OR (disconnect* NEAR/3 (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* NEAR/3 (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar NEAR/3 disconnect*) OR (palliat* NEAR/3 operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection'
8	Brain Stimulation	'brain depth stimulation'/de OR 'brain responsive neurostimulator'/de OR 'deep brain stimulator'/de OR 'nerve stimulation'/de OR 'nerve stimulator'/de OR 'vagus nerve stimulation'/de OR ('brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') NEAR/2 (electrostim* OR stimulat*)) OR neurostim*

9	Harms	anhidrosis/de OR 'adverse event'/de OR 'adverse drug reaction'/de OR 'behavior disorder'/de OR 'cognitive defect'/de OR 'developmental delay'/de OR 'developmental disorder'/de OR dystonia/de OR 'liver injury'/de OR 'loss of appetite'/de OR 'motor dysfunction'/de OR 'organ damage'/de OR 'patient harm'/de OR 'sleep disorder'/de OR sweating/de OR (advers* OR harm* OR 'side effect'):ab,ti,kw OR anhidrosis OR (appetite NEAR/3 (lose OR losing OR loss)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) NEAR/3 (effect* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR neurodevelopment*) NEAR/3 (delay* OR disorder* OR regress*)) OR dystonia OR hypohidrosis OR hypohydrosis OR (liver NEAR/3 (damag* OR injur*)) OR (miss* NEAR/3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) NEAR/3 (disorder* OR inability OR unable))
10	Parental Preferences	parent/de OR (parent* OR mother* OR father*):ab,ti,kw
11	Untreated Disease	'treatment refusal'/de OR ('not treated' OR 'no treatment' OR untreat*):ab,ti,kw OR (declin* OR forgo* OR 'not' OR no OR refus* OR withheld OR withhold*) NEXT/3 (treated OR treatment*)
12	Study Designs/ Publication Types	[english]/lim AND [1999-2020]/py NOT ([animals]/lim NOT [humans]/lim OR abstract:nc OR annual:nc OR 'book'/de OR ((case NEXT/1 (report* OR stud*)):ti) OR 'case report'/de OR 'case study'/de OR conference:nc OR 'conference abstract':it OR 'conference paper'/de OR 'conference paper':it OR 'conference proceeding':pt OR 'conference review':it OR congress:nc OR diagnos*:ti OR 'diagnosis'/mj OR 'diagnostic accuracy'/mj OR 'diagnostic procedures'/mj OR 'diagnostic test'/mj OR 'diagnostic test accuracy study'/mj OR 'differential diagnosis'/mj OR 'editorial'/de OR editorial:it OR 'erratum'/de OR guideline*:ti OR letter:it OR 'note'/de OR note:it OR meeting:nc OR 'practice guideline'/de OR 'review'/exp OR sessions:nc OR 'short survey'/de OR symposium:nc OR animal*:ti OR experimental:ti OR (vitro:ti NOT vivo:ti) OR canine:ti OR dog:ti OR dogs:ti OR mouse:ti OR mice:ti OR rabbit*:ti OR rat:ti OR rats:ti OR rodent*:ti OR sheep:ti OR swine:ti)
13	Combine sets	#1 OR #2 OR #3 OR #4
14	Combine sets (KQ1 Pharmacology)	#13 AND #12 AND #5
15	Combine sets (KQ2 Diet, Surgery, Brain Stimulation)	#13 AND #12 AND (#6 OR #7 OR #8)
16	Combine sets (KQ3 Treatment Harms)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND 9
17	Combine sets (CQ1 Parental Preferences)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND #10
18	Combine sets (CQ2 Untreated/ Uncontrolled Epilepsy)	#11 AND #12 AND #13

19	Combine Sets All KQs	#14 OR #15 OR #16
20	Combine Sets All CQs	#17 OR #18

SocINDEX and Web of Science databases (Searched January 1, 1999, to August 19, 2021)

(epilep* OR convuls* OR seizure* OR spasm*)

AND

(acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide OR keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic' OR 'corpus callosotomy' OR craniotom* OR (disconnect* AND (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* AND (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar AND disconnect*) OR (palliat* AND operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection' OR 'brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') AND (electrostim* OR stimulat*)) OR neurostim*)

AND

(parent* OR mother* OR father*)

AND

(perception OR factor* OR decision)

CINAHL (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR dravet*
2	Epilepsy/ 0-3 Age Group	(babies OR baby OR child* OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR "very young" OR "younger than three" OR "younger than 3" OR "under three" OR "under 3" OR "below three" OR "below 3" OR "3 or below" OR "3 or younger" OR "three or below" OR "three or younger") AND epilep*
3	Infantile Spasm/ Neonatal Seizure	"infantile spasm*" OR "neonatal seizure*" OR ((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) AND (convuls* OR seizure* OR spasm*))
4	Parental Preferences	father* OR matern* OR mother* OR parent* OR patern*
5	Harms	"adverse drug reaction" OR "adverse effect*" OR "adverse event*" OR anhidrosis OR appetite* OR behavior* OR behaviour* OR cognitiv* OR defect* OR delay* OR development* OR disorder* OR dysfunction* OR dyston* OR harm* OR impair* OR injur* OR liver OR motor OR "organ damage*" OR "sleep disorder*" OR sweat* OR "side effect*"
6	Untreated Disease	"treatment refusal" OR "not treated" OR "no treatment" OR untreat* OR "decline treatment" OR "declined treatment" OR "forgo* treatment" OR "refuse treatment" OR "refused treatment" OR "refusing treatment" OR "withheld treatment" OR "withhold treatment" OR "withholding treatment"
7	Combine Concepts	(S1 OR S2 OR S3) AND S4
8	Apply Limits	S7 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)
9	Combine concepts	(S1 OR S2 OR S3) AND (S5 AND S6)
10	Apply Limits	S9 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)

PsycINFO (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR (dravet* adj1 (disease OR syndrome)) OR ((child* OR infan* OR paediatr* OR pediatr*) adj1 epilep*)
2	Epilepsy/ 0-3 Age Group	(baby OR babies OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool OR toddler* OR "very young" OR (("younger than" OR under OR below) adj3 ("3" OR three)) OR (("3" OR three) adj3 ("or below" OR "or under" OR "or younger")) AND (epilepsy/ OR epileptic seizures/ OR epileps* OR epileptic*))
3	Infantile Spasm/ Neonatal Seizure	((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) adj3 (convuls* OR seizure* OR spasm*))
4	Parental Preferences	parents/ OR (patern* OR parent* OR matern* OR mother* OR father*)
5	Harms	behavior disorders/ OR cognitive impairment/ OR delayed development/ OR developmental disabilities/ OR liver disorders/ OR motor development/ OR "side effects (drug)"/ OR "side effects (treatment)"/ OR sleep wake disorders/ OR sweating/ OR (advers* OR anhidrosis OR dystoni* OR harm* OR harm* OR hypohidrosis OR hypohydrosis OR "organ damage") OR (appetite adj3 (lose* OR losing OR loss OR

		lost)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) adj3 (effect* OR deficit* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR motor* OR neurodevelopment*) adj3 (delay* OR disorder* OR regress*)) OR ((liver OR organ*) adj3 (damag* OR injur*)) OR (miss* adj3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) adj3 (disorder* OR inability OR unable))
6	Untreated Disease	treatment refusal/ OR ("not treated" OR "no treatment" OR untreat*) OR ((declin* OR forgo* OR "not" OR no OR refus* OR withheld OR withhold*) adj3 (treated OR treatment*))
7	Combine Sets	(1 OR 2 OR 3) AND 4
8	Apply Limits	limit 7 to yr="1999 - 2021"
9	Apply Limits	limit 8 to up=19000101-20210819
10	Apply Limits	9 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
11	CQ1	limit 10 to english language
12	Combine Sets	(1 OR 2 OR 3) AND (5 AND 6)
13	Apply Limits Apply Limits	Limit 12 to yr="1999 - 2021"
14	Apply Limits	limit 13 to up=19000101-20210819
15	Apply Limits	14 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
16	CQ2	limit 5 to english language

APPENDIX B: The Theoretical Domains Framework*

Domain	Definition	Constructs
Knowledge	An awareness of the existence of something	Knowledge (including knowledge of condition/scientific rationale) Procedural knowledge Knowledge of task environment
Skills	An ability or proficiency acquired through practice	Skills Skills development Competence Ability Interpersonal skills Practice Skill assessment
Social/professional role and identity	A coherent set of behaviors and displayed personal qualities of an individual in a social or work setting	Professional identity Professional role Social identity Identity Professional boundaries Professional confidence Group identity Leadership Organizational commitment
Beliefs about capabilities	Acceptance of the truth, reality or validity about an ability, talent or facility that a person can put to constructive use)	Self-confidence Perceived competence Self-efficacy Perceived behavioral control Beliefs Self-esteem Empowerment Professional confidence
Optimism	The confidence that things will happen for the best or that desired goals will be attained	Optimism Pessimism Unrealistic optimism Identity
Beliefs about Consequences	Acceptance of the truth, reality, or validity about outcomes of a behaviour in a given situation	Beliefs Outcome expectancies Characteristics of outcome expectancies Anticipated regret Consequents
Reinforcement:	Increasing the probability of a response by arranging a dependent relationship, or contingency, between the response and a given stimulus	Rewards (proximal/distal, valued/not valued, probable/improbable) Incentives Punishment Consequents Reinforcement Contingencies Sanctions

Domain	Definition	Constructs
Intention	A conscious decision to perform a behaviour or a resolve to act in a certain way	Stability of intentions Stages of change model Transtheoretical model and stages of change
Goals	Mental representations of outcomes or end states that an individual wants to achieve	Goals (distal/proximal) Goal priority Goal/target setting Goals (autonomous/controlled) Action planning Implementation intention
Memory, attention and decision processes	The ability to retain information, focus selectively on aspects of the environment and choose between two or more alternatives	Memory Attention Attention control Decision-making Cognitive overload/tiredness
Environmental context and resources	Any circumstance of a person's situation or environment that discourages or encourages the development of skills and abilities, independence, social competence and adaptive behaviour	Environmental stressors Resources/material resources Organizational culture/climate Salient events/critical incidents Person environment interaction Barriers and facilitators
Social influences	Those interpersonal processes that can cause individuals to change their thoughts, feelings, or behaviours	Social pressure Social norms Group conformity Social comparisons Group norms Social support Power Intergroup conflict Alienation Group identity Modelling
Emotion	A complex reaction pattern, involving experiential, behavioural, and physiological elements, by which the individual attempts to deal with a personally significant matter or event	Fear Anxiety Affect Stress Depression Positive/negative affect Burn-out
Behavioral regulation	Anything aimed at managing or changing objectively observed or measured actions	Self-monitoring Breaking habit Action planning

*British spelling of definitions is kept from original publication

Atkins L, Francis J, Islam R, O'Connor D, Patey A, Ivers N, Foy R, Duncan EM, Colquhoun H, Grimshaw JM, Lawton R, Michie S. A guide to using the Theoretical Domains Framework of behaviour change to investigate implementation problems. Implement Sci. 2017 Jun 21;12(1):77. doi: 10.1186/s13012-017-0605-9. PMID: 28637486; PMCID: PMC5480145.

APPENDIX C. CERQual assessment components

CERQual Components	Definition
Methodological limitations	The extent to which there are problems in the design or conduct of the primary studies supporting a review finding. We used our CASP assessment of each study to guide our assessment of this component.
Coherence	An assessment of how clear and cogent the fit is between the data from the primary studies and the review finding.
Relevance	The extent to which the body of evidence from the primary studies supporting a review finding is applicable to the context specified in the review question.
Adequacy	The degree of richness and quantity of data supporting a review finding.

Noyes J, Booth A, Lewin S, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 6: how to assess relevance of the data. Implement Sci. 2018 Jan 25;13(Suppl 1):4. doi: 10.1186/s13012-017-0693-6. PMID: 29384080.

APPENDIX D. Methodological appraisal ratings using the CASP tool

CASP question	Study O'Brien, 2020	Sylven, 2020	Heath et al. 2016	Ozanne, 2016	Pieters, 2016; Baca, 2015
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the qualitative methodology appropriate?	Yes	Yes	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the recruitment strategy appropriate to the aims of the research?	No[1]	No[1]	No[1]	No[1]	No[1]
Was data collected in a way that addressed the research issue	Yes	Yes	Yes	Yes	Yes
Was the relationship between researcher and participants adequately considered?	Can't tell	Can't tell	Can't tell	Yes[2]	Can't tell
Were ethical issues taken into consideration?	Yes[3]	Yes[3]	Yes[3]	Yes[3]	Can't tell
Was there sufficient rigor?	Yes	Yes	Yes	Yes	Yes
Is there a clear statement of findings?	Yes	Yes	Yes	Yes	Yes
How valuable is the research?[4]	Yes	Yes	Yes	Yes	Yes
Overall risk to rigor	Low	Low	Low	Low	Low

Abbreviations: CASP: Critical Appraisal Skills Programme

[1] Participants in these studies were recruited after their child underwent surgery, raising concerns about selection bias due to selecting parents who were a) referred to surgery, b) proceeded with surgery, and c) experience some level of success with surgery. Excludes parents of a child with drug-resistant surgery who were not referred to surgery, who choose not to proceed with surgery, or whose child did not achieve desired outcomes.

[2] Interviewers and researchers not part of care/surgery team

[3] Approved by university ethics committee

[4] "Yes" rating given when study: a) discussed contribution to existing knowledge or understanding, b) identified new areas where research is necessary, and c) discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

APPENDIX E. Coding extracts for treatment decisions for childhood epilepsy into the Theoretical Domains Framework

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Knowledge	Baca, 2015, 25894906	With the recognition that "something is wrong," parents sought information from multiple sources in an attempt to understand seizures and epilepsy and become better advocates for their child.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Parents reported difficulties as the they sought information about seizures, epilepsy, medications, and surgery….Parents sought information from a variety of disparate sources including physicians, the Internet, books, and family/friends.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Through this journey, parents learned to navigate a complicated medical system filled with doctors of different specialties, multiple hospitals, insurance approvals, and numerous treatment options	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Pieters, 2016, 27521720	Parents also had to learn about the intricacies of the healthcare system including the types of doctors that were needed to care for their children	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Parents perceived the information they received from professionals as inadequate to make an informed decision. They therefore sought out additional information regarding the surgical procedure and outcomes. Aiming to comprehend what surgery would entail, parents requested information in lay language and searched the internet for information and videos.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	"I googled it until I couldn't google anymore and I think that gave me a better understanding of what was going on."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Finally, parents emphasized the importance of receiving adequate information regarding epilepsy surgery, including information presented in a variety of formats (e.g., booklets, photographs, videos). They also suggested that a 'frequently asked questions' information sheet would have been beneficial. "I know I got a little booklet about epilepsy surgery services, but it would have been nice for us to have some extra information… just some written up case studies of kids that have gone through, had the same, you know, operation."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	These included …additional information provision	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	O'Brien, 2020, 33120326	[I]t was felt that a greater awareness of potential changes besides seizure control prior to surgery would have been helpful for children and their families in developing realistic expectations and preparing for post-surgical adjustment. They just tell you medical stuff about it, the recovery, not that it'll be different…They just tell you about having no seizures […] They don't tell you what it's actually going to be like.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Sometimes, parents received more information from other parents, than from the physician. They were disappointed if they felt that information was withheld	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Parents thought that it was important to receive the information that surgery was aimed to treat the epilepsy and not other disabilities, such as behavioral problems, in order to have realistic expectations. They emphasized the importance of being informed about both positive and negative outcomes and risks. Even if surgery was a medical success, the family's life situation was	Need for information about epilepsy, treatment options, and navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
		sometimes characterized by sorrow and problems. The deterioration, expected or unexpected, of e.g., motor function, speech, visual field, or attention made life more difficult and burdensome.	
Knowledge	Baca, 2015, 25894906	Parents also reported little initial familiarity with epilepsy and that they felt overwhelmed trying to learn a new language of epilepsy because of the "avalanche of information coming at us."	Overwhelmed by information
Knowledge	Pieters, 2016, 27521720	She described the hard and time-consuming struggle of trying to synthesize information on her own "because there really wasn't a lot out there and when you're hearing different things from different specialists it's very difficult for parents to make heads or tails of it". She spent "six months combing through the literature reading everybody's publications from places all over the world … because I wasn't getting a straight answer … my husband was saying to me you know are you going to go and get your degree in neuroscience after this … but you know it's your child, you want to do the best by them and you know you want them to be able to achieve their potential".	Overwhelmed by information
Knowledge	Baca, 2015, 25894906	As they acquired more epilepsy knowledge, however, many parents felt more empowered and able to ask questions and participate more effectively in the care of their child	Knowledge is empowering
Knowledge	Baca, 2015, 25894906	A mother who stated, "I just think knowledge is power and it also brings some comfort to making a good decision," exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy	Knowledge is empowering
Knowledge	Heath, 2016, 27371883	Increasing their own knowledge about medically intractable epilepsy and the available treatment options empowered parents to participate more fully in the decision-making process. "I said to him 'don't tell me in a doctor's term, tell it me in a parent's term. I want to know the ins and outs; I want to know exactly what the risks are but in terms that I understand'"	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	[P]arents took on the responsibility for learning as exemplified by the parent who naturally 'corrected' her language to imply her understanding of the different terms that clinicians use, "(the seizures) were not controlled … or intractable as you guys call them."	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	Being open to learn and learning itself occurred while parents became epilepsy experts in order to be effective advocates for their children	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	In order to have productive information exchanges with clinicians and engage in shared decision-making, parents realized that they had to learn about a very unfamiliar and unforeseen situation	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	The lack of understandable information and the experience of information imbalance between the parent and the physician were overwhelming as experienced by a father who described him and his wife as "pretty pragmatic." He said, "The beginning was terrible, because you're of course dealing with … specialists in things and that you've pretty much never even heard of …. It was tough in the beginning, really tough … (the information that we found) was written by doctors for doctors. It's kinda tough when you don't know what you're talking about to get through that.	Knowledge is empowering
Social role and identity	Heath, 2016, 27371883	However, they also reported a sense of duty to be "strong" (P1) and "brave" (P3, 4) for their children, wishing to protect them from their feelings of anxiety.	Role and responsibility as a parent
Social role and identity	Heath, 2016, 27371883	Parents discussed feeling pressure from a need to 'do the right thing' by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential	Role and responsibility as a parent

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Emotion	Baca, 2015, 25894906	Parents reported personal and family stress throughout the journey. Caring for children with a chronic, severe, and paroxysmal neurologic condition meant being "on call" all the time; they had to be prepared to respond to an emergency at all times	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Participant descriptions highlighted the significant impact that epilepsy had upon on the child's and family's lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: "you're tired yourself before you even go out the door" (Charlie – mother). In some cases, experiences of epilepsy impacted on their lives to such a degree that parents referred to children as "not having a life" (Charlie - mother)	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Child mood and behavioural difficulties-stress, secrecy and shame. Before surgery children's mood and behavioura difficulties were a common, causing fear and stress which negatively impacted upon family relationships: "If there was something there, she would get it, she would want to attack you with it". (Jessica – mother) Many described how increased supervision led to a change in their parenting role, negatively impacting on the relationship they or their other children had with their child: "I was just his carer really and didn't have a relationship with him" (Jack - mother)	Drained from managing epilepsy
Emotion	Baca, 2015, 25894906	Parents frequently described that medication trials continued often with little to no mention of surgery. One mother told of her frustrations with multiple medications, "It was, 'Let's work on all the different medical aspects or medicines before going to surgery or even talking about surgery."	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Parents felt desperate not only to find a treatment option that would work, but also to persevere and actively do something that was productive and necessary to help their child	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Wanted to play an active role	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[W]anted to use time constructively	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Another mother remembered that she felt "desperate" to take her adolescent son to another doctor, but she needed to wait for referrals because of insurance	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[P]arents felt frustrated and desperate to get help for their child.	Desperate to help their child
Emotion	Heath, 2016, 27371883	Following indication of surgery as a potential treatment, parents and families faced what they described as a "difficult decision" (P5) which was experienced as "daunting", "frightening", "scary", and "stressful"	Difficult and stressful decision
Emotion	Pieters, 2016, 27521720	"It was not only in terms of time, it was mentally draining as well. It took a lot of time and it took a lot of concentrated thought to figure this out."	Difficult and stressful decision
Emotion	Ozanne, 2016, 27176878	Parents were afraid and worried	Fear (unspecified)

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Emotion	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options. This perception was influenced by parental fear regarding the irreversibility of surgery, as well as by professionals who had informed them that surgery was the only remaining treatment for controlling their child's seizures	Fear of surgery
Emotion	Pieters, 2016, 27521720	For the majority of parents, brain surgery was a frightening possibility as described by the mother who said, "At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric." Another mother's statement reflected upon her initial feelings of fear and avoidance when she said that she and her husband, "just couldn't deal with (having our daughter go through brain surgery)	Fear of surgery
Emotion	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience "a change in personality" as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Fear of surgery
Emotion	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Fear of surgery
Emotion	Sylven, 2020, 32480305	Many parents were worried that their child's epilepsy would be "worse" after the surgery, and as a result, the operation would have been "in vain". This included a risk that seizure frequency might increase, that seizures might be longer or be of a different character, or that seizure frequency might not improve.	Fear of surgery
Emotion	Ozanne, 2016, 27176878	It was demanding not to know if surgery would be possible, and the parents were thankful when the decision was finally made	Relief after decision was made
Emotion	Sylven, 2020, 32480305	Many parents described a general hope for improved behavior and emotional functioning for their child. Behavior encompassed a wide variety of outcomes including that the child would be "calmer" with fewer behavioral outbursts, have better attention and concentration, and "feel better"	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents hoped that the surgery would lead to better health and quality of life for their child	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents often expressed a desire that surgery would result in their child being "more alert", less "tired", and "have more energy". They also mentioned that they hoped that surgery would result in better sleep quality	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including "tiredness" would lead to a "normal life" for their child	Hope for better health and quality of life
Emotion	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they "hoped" for candidacy	Hope for surgical candidacy
Emotion	Heath, 2016, 27371883	When a decision to accept surgery had been made, parents hoped for the procedure to be carried out as soon as possible. Waiting for a date for surgery was described as an anxious time for parents, who experienced relief when a date was set.	Hope for surgical candidacy

TDF domain	**Author, Year, PMID**	**Extracted text^A**	**Theme**
Social influence	O'Brien, 2020, 33120326	This sense of exclusion impacted on the family as a whole, with siblings being unable to attend clubs or invite friends to their house, and parents feeling that they "couldn't join in with what other families were doing" (Lucy – mother). A common theme was parental concern that siblings had less time or attention: as "the focus was on him [child with epilepsy] a lot of the time" (Andrew – mother).	Social exclusion
Social influence	O'Brien, 2020, 33120326	Difficulties appeared to be exacerbated by a perceived a lack of understanding from others, which appeared to be tied to a notion of epilepsy being an invisible condition, as seizures commonly occurred behind closed doors: "At the beginning, it was just generally at home, they didn't understand, they didn't get it. (Jessica – mother). Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. "Gosh, we didn't tell anybody. We were ashamed". (Jack – mother)	Social exclusion
Social influence	Heath, 2016, 27371883	In addition, parents emphasized the importance of accessing the experience and expertise of other parents who had followed a similar treatment pathway. Such peer support was suggested to facilitate understanding of the surgical procedure from a 'parental perspective', as well as provide emotional support. "If people could freely put their experiences on either a forum or even if you get a group, you know, parents talking to other parents who have gone through the experience to get a real idea of what's happening."	External support from peers and healthcare professionals
Social influences	Heath, 2016, 27371883	These included … peer support	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Those parents who were able to connect with other parents and families of children with epilepsy found it to be very helpful to be able to communicate with peers with seizures unresponsive to medication who were on the same journey.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	[E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Three key facilitators were identified as follows: (a) having a champion, (b) finding a good team of epilepsy providers, and (c) connecting with others who shared the experience of epilepsy. (Fig. 1; Table 2). Such facilitators played a key role during the times when parents especially needed support to act purposefully and move out of vulnerable periods.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	However, using time constructively happened after the parents described finding a champion that advocated for them and helped them navigate their journey. It was a champion, whether a doctor, nurse, or social worker, that facilitated steady and purposeful progress over more manageable portions of the journey	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	One mother favorably described one such champion, a nurse coordinator, "she's like wonderful for us. She's been our lifesaver." These champions helped parents get referrals and seek different treatment options or were the persons that believed them and pushed them to seek better or additional care. For one mother, her daughter's pediatrician acted as her champion. She said, "He encouraged me and gave me the confidence that if I wasn't happy with what five other pediatric neurologists were saying that it was okay to go and you know continue to seek out other opinions."	External support from peers and healthcare professionals

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Social influence	Heath, 2016, 27371883	Family decisions. Parents described decision-making as a family choice involving partners, siblings, grandparents, and the child (where developmentally appropriate). Discussing treatment options with family members was generally considered to be helpful. "I remember my mother-in-law was in Pakistan and I called her up and I was very teary…I felt like I had been punched in the stomach to be honest 'cause, you know, brain surgery is not something you take lightly and she said 'well if it's for his betterment and to improve his life then we will do whatever it takes'."	Family decision
Social influence	Heath, 2016, 27371883	For some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary carer, she 'witnessed' the true extent of their child's seizures. In cases where the child's epilepsy was considered to impact on the whole family (e.g., where a parent could not return to work because of a need to look after the child), then, the decision was understood as a responsibility of the whole family	Family decision
Belief about consequences	Baca, 2015, 25894906	On their journey, many parents transformed their view of surgery from a scary, last resort treatment to a necessary and hopeful option. One mother explained, "By the time she got it—it got so serious I was looking toward that as being an answer to our prayers rather than just something that I couldn't even think about." This transformation occurred as parents became increasingly aware of the severity of their child's epilepsy and its impact on their child's and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	Many parents initially perceived epilepsy brain surgery to be "pretty horrific" and a treatment of "last resort."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	After an arduous journey, parents reached the right doctor and center; epilepsy surgery, although previously seen as a last ditch option, became a viable option and a source of hope.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	After persevering through hard times with unsuccessful medications and interventions, they eventually decided to proceed with surgery, the option that they perceived to be the best treatment for the epilepsy.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	Because surgery was viewed as a treatment of last resort for many parents, time was required to process the initial fear, make sense of the emotional information, and consider the factual risks and benefits in order to decide on surgery as the best option for their child. Reflecting on why it took longer than six years to get to surgery for her 9-year-old daughter, a mother said, "Of course you need to take a lot of time to think about it. It's very invasive.… I did as much research as I could about it, and talked to the doctor a lot about it, and I decided that that was her best chance of having a normal life in the future."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Pieters, 2016, 27521720	Through internal processing, parents worked through the diverse emotions and large amount of information that they were receiving and gathering to move forward and thereby develop a greater understanding of their child's illness and prognosis in order to ultimately reach the point where surgery became the treatment of choice	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Parents were clear that putting their child forward for epilepsy surgery was the best and right decision for their child, demonstrating a high level of satisfaction with the treatment outcome. However, initial perceptions of surgery were often negative, particularly if it was not a treatment option that had previously been considered or that professionals had indicated. Many parents described attending their child's clinic appointment expecting to talk about more conventional treatments (e.g., medication or diet), leading to them feeling surprised when surgery was raised for discussion. “We went originally to talk about the ketogenic diet and then the doctor said ‘oh I think surgery's your best option’ which came as a bit of a shock really because no one had ever mentioned the surgery before.” (P5) “It actually came as a bit of a surprise to us that option, in that it was mentioned in a review visit, that maybe the time had come to look at the option of surgery but we weren't really aware of that option at that point so it came as quite a shock to us.” (P6) While some parents took comfort from the possibility of an additional treatment pathway, others felt excluded from the decision to assess their child for surgical candidacy, leading to their feeling unable to participate in an informed discussion	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Some parents were unconvinced about the efficacy of surgery for reducing their child's seizures. Skepticism stemmed from their experience of other pharmacological and dietary treatments which had resulted in little improvement in seizure control. In weighing up treatment options, parents also discussed the side effects of alternative treatments. For example, medications were perceived to have a range of adverse side effects, whereas ketogenic diets were viewed as negatively affecting the child's ability to live a ‘normal’ life. “We thought about the effects of the medication if we chose to continue on the path of finding a different medication… but then how long would that have taken us? Like how long would it work for? Then it would develop, then how long 'til we have to find something else, with all the side-effects that can happen? I didn't want to put her through that as well.” (P9)	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	Balancing potential costs and benefits. When deliberating surgery, parents balanced their perceived necessity of the treatment in terms of how epilepsy currently affected and would, in the future, affect their child's quality of life, against concerns regarding the possibility of negative outcomes. In other words, parents weighed up the potential benefits of surgery such as freedom from seizures against what they understood as the risks, including cognitive impairments. “There was no quality of life for him; we had to do what was best. We were told of the risks of the surgery and we were told that actually this would be the best thing for him… we don't regret it, but it was, it's not an easy decision to take because you're worried about the risks of things.”	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned that while they would like seizure freedom, any reduction in seizures would be “welcome” and a clear improvement for their child and for the family in general.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including “tiredness” would lead to a “normal life” for their child.	Balancing potential benefits of surgery with concerns of adverse effects

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Sylven, 2020, 32480305	Many parents mentioned a desire for seizure freedom for their child after surgery. They wanted their child to be free from the "trouble" of seizures and everything that follows from seizures	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	They wanted that their child could do everything their friends could do like "play football, cycle, and swim independently". Some parents mentioned that they wanted their child to be able to go back to school and have a "proper social life" while some parents referenced the future hoping that the child would be better placed to get a job and driving license as a result of surgery.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience "a change in personality" as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The majority of parents mentioned concerns about possible complications that could occur during or soon after the surgical procedure	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	A small number of parents said that they had no concerns before the surgery. One parent said that they were not worried as "no concerns arose in the discussions with professionals" before the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The results of the current study suggest that parents frequently worry about that the child would lose skills or function, or complications would arise during surgery that could result in injury.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	For all parents in this study, the perceived need of surgery outweighed the possible risks and side effects. "We thought this is going to be no life for him… He was on this diet which meant he couldn't go out with friends; he couldn't stay at friends' houses, all the things that other kids do, he couldn't do… and then he got these fits virtually every night and we thought… you know what, there is no decision to make, we gotta do it… We knew there were risks, we talked about them with [child] and he was frightened but he said no, we gotta do it." (P3)	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options…Some parents felt that they had little alternative but to accept surgical intervention in order to provide their child with the best possible chance of improving their quality of life. "We felt it was our only option really in her having a more independent future."	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	"My husband and I walked out of the appointment saying we feel like we don't have a choice… we felt like it was the only choice we could make."	Only choice left

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Heath, 2016, 27371883	For one family with an adolescent child, it was considered necessary to accept surgery while the child received care from the children's hospital as they believed that the treatment would become unavailable once transitioned to adult services.	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Parents described a process of weighing up the pros and cons based on the information provided by the medical team, which led to the decision to pursue surgery. Many perceived that surgery was the only hope "a simple decision" (Alex – mother), they were "in no doubt" (Lucy – mother) and that they "had no choice" (Jessica mother) as the health and wellbeing of their child would deteriorate or become more serious if surgery was not pursued. There was also a sense of 'not knowing' outcomes if surgery was not pursued. We felt we've got to do this because where is she going to end up if we don't?	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Decision making – is there a choice? When asked what informed the decision-making process when considering the option of surgery as a treatment for epilepsy, parents discussed how the perceived risks to their child's safety, as well as the safety of others, were key factors. Even with all the risks, it wasn't going to be much worse than having a little girl that was unconscious all the time anyway.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Information about possible complications related to surgery did not prevent the parents from wishing to proceed with the operation. They preferred to take a risk rather than live in constant fear	Only choice left
Environmental context and resources	Baca, 2015, 25894906	The journey to finding the right doctor was further complicated by parental perceptions that doctors sometimes lacked knowledge of or familiarity with epilepsy. Many parents went to numerous doctors searching for the "right" doctor, or one who had a strong epilepsy knowledge base, could effectively identify the problem, and then make a clear plan of action	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents expressed that their children were seen, evaluated, and cared for by a variety of doctors of different specialties throughout their journey. Parents frequently described difficulty finding the "right" doctor(s). Difficulty was associated with a lack of pediatric neurologists in their local area and with different doctors having different recommendations.	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents depicted a pathway filled with multiple doctors and treatments, insurance battles, and work and life obstacles as they sought a cure for their child's seizures, becoming ever more knowledgeable about epilepsy	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	Further, finding the best clinicians for the needs of their child required an understanding of the relationship between different medical (sub)-specialties and thereby represented dual and concurrent use of processing and navigating mechanisms	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	The second mechanism, navigating, was when the parents kept and directed their way along their journey often through uncharted territory as they steered through various aspects of the health-care system and learned about epilepsy itself and its various treatments.	Challenges with navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Environmental context and resources	Pieters, 2016, 27521720	A mother described her disappointment with being denied by insurance despite doing all the work that was required	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	"We'd already been fighting her seizures for two and a half, approaching three years, and because we were new there and per protocol, they wanted to try these other treatments for another year."	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	The Swedish Social Insurance Agency, technical aids center, school, and municipality were criticized by the parents. Based on their experience, they felt that the bureaucracy had been problematic and had taken a great deal of time. They had to fight for their rights, despite physicians' certificates. Without specific diagnoses of comorbidities, it was even more difficult to get adequate support. They thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	Parents were disappointed if it took a long time to get a correct diagnosis	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Several parents experienced feeling doubted by a doctor; some were told that nothing was wrong with their child initially	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they "hoped" for candidacy. This hoping required persistence and sometimes disregarding doctors who initially felt surgery was not an option. Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, "'I don't believe that she is a surgical candidate.' As a parent, I listened and I wanted to think otherwise.... In hindsight, I'm glad I didn't listen to him."	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	Parents also reported notable physician-based barriers including variability in perceived knowledge of epilepsy, criteria for presurgical referral and appropriateness of epilepsy surgery	Interactions with providers (barriers)
Environmental context and resources	Pieters, 2016, 27521720	[C]rossing such an information divide was an uphill battle as stated by a mother who described feeling dismissed by doctors until she found someone who was approachable and was "gracious enough to talk to a mere mortal mother on the phone … because a lot of docs won't, they've got lots of protective armor around them shielding off potential patients or worse yet parents."	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	Parents perceived a lack of control over the decision-making process through their limited involvement in initial discussions regarding surgical candidacy	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	These included earlier and softer introduction of surgery as a possible treatment option	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	During the long hospitalizations, some parents felt that nobody had time for them. Arrogance of some professionals made them very disappointed	Interactions with providers (barriers)

TDF domain	**Author, Year, PMID**	**Extracted text**[A]	**Theme**
Environmental context and resources	Ozanne, 2016, 27176878	If the pediatric neurologists revealed their own worries and insecurities regarding epilepsy surgery, this could have negative effects	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	Parents described that support and information were inadequate before the epilepsy was recognized to be drug-resistant, especially from the local hospital. Their concerns were not taken seriously	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	The support and information went from poor to strong when specialists at the university hospitals were involved	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	The pediatric neurologist's worry and sympathy for the family could also strengthen them since they felt invited and could be insecure together	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	Trust in the information that they received was greater when their child was in the hands of a team with different expertise, as exemplified by the mother who said, "They get all the doctors together to look at all the information … that felt a lot more helpful because if we were at a different hospital and you only had the one doctor, then they would have misdiagnosed him."	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	When parents finally reached a comprehensive center to seek evaluation for potential pediatric epilepsy surgery, they recognized the importance of having a medical team of experts	Interactions with providers (facilitators)

[A] Study participant (first order statement) or study author (second order statement). We kept original spelling from primary study.

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Title page
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Background, pg 3
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Pg 3
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Pg 3-4
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Pg 3, Appendix A
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Appendix A
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Pg 4-5
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Pg 4-5
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Pg 4-5
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	NA
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Pg 4-5, Table 1
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Pg 4-5
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Pg 4-5
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Pg 4-5
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	Pg 4-5
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	NA
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Pg 4-5< Appendix D

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Pg. 4-5, Table 2
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Pg 5
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Pg 5
Study characteristics	17	Cite each included study and present its characteristics.	Pg 5, Table 1
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Appendix D
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Table 2
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Pg 5
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Pg 6-8
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	Pg 6-8
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	NA
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	NA
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	Pg 6-8, Table 2
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Pg 9
	23b	Discuss any limitations of the evidence included in the review.	Pg 10
	23c	Discuss any limitations of the review processes used.	Pg 10
	23d	Discuss implications of the results for practice, policy, and future research.	Pg 9-10
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Not registered
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	NA
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	NA
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Pg 11
Competing interests	26	Declare any competing interests of review authors.	Pg 11
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Pg 11

PRISMA 2020 Checklist

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

For more information, visit: http://www.prisma-statement.org/

BMJ Open

Parent perceptions and decision-making about treatments for epilepsy: a qualitative evidence synthesis

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Parent perceptions and decision-making about treatments for epilepsy: a qualitative evidence synthesis

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Abstract

Objectives Epilepsy treatment decision-making is complex and understanding what informs caregiver decision making about treatment for childhood epilepsy is crucial to better support caregivers and their children. We synthesized evidence on caregivers' perspectives and experiences of treatments for childhood epilepsy.

Design Systematic review of qualitative studies using a best-fit framework and Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual) approach**.**

Data sources Searched Embase, PubMed, CINAHL, PsycINFO, SocINDEX, and Web of Science from January 1, 1999, to August 19, 2021.

Eligibility criteria We included qualitative studies examining caregiver's perspectives on anti-seizure medication, diet, or surgical treatments for childhood epilepsy. We excluded studies not reported in English.

Data extraction and synthesis We extracted qualitative evidence into one of 14 domains defined by the Theoretical Domains Framework (TDF). One reviewer extracted study data and methodological characteristics, and two reviewers extracted qualitative findings. The team verified all extractions. We identified themes within TDF domains and synthesized summary statements of these themes. We assessed our confidence in our summary statements using GRADE-CERQual.

Results We identified five studies (in six reports) of good methodological quality focused on parent perceptions of neurosurgery; we found limited indirect evidence on parents' perceptions of medications or diet. We identified themes within six of the 14 TDF domains relevant to treatment decisions: knowledge, emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources.

Conclusions Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed on non-surgical treatments for epilepsy and among caregivers from different cultural and socio-economic backgrounds to fully understand the diversity of perspectives that informs treatment decision-making.

Strengths and limitations of this study

- This synthesis followed contemporary standards for the conduct of qualitative synthesis, which includes using a best-fit framework approach to categorize and synthesize findings based on the Theoretical Domains Framework (TDF).
- Risks to rigor of included studies were assessed using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies.
- Confidence in the conclusions drawn from this synthesis was rated using the Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE CERQual).
- As with all qualitative synthesis, selecting which data to extract and how to code it is ultimately a subjective process.
- This synthesis was limited to studies that examined parents' perceptions, experiences, and decision-making about pursuing surgery to treat their child's epilepsy.

Introduction

Epilepsy is a common neurologic disorder in children, affecting about 1% of children in the United States.[1] While there are different types of childhood epilepsy, each type involves recurring seizures caused by abnormal electrical activity in the brain. Epilepsy is categorized by seizure type (e.g., focal, generalized, or unknown), epilepsy type (e.g., focal, generalized, unknown), and syndrome type (e.g., childhood absence epilepsy, Dravet syndrome).[2-4] No matter the type, epilepsy is a chaotic and unpredictable condition for both the affected children and their caregivers.[5]

Treatment and ongoing management approaches depend on the type of the epilepsy and prior treatment response. Treatment options for childhood epilepsy include anti-seizure medications (ASMs), ketogenic diets, or surgery. Although many children with new-onset epilepsy achieve seizure freedom with ASMs,[6] these drugs are associated with numerous adverse effects (e.g., tiredness, nausea, headache, difficulty concentrating, depression, and suicidal ideation).[7] Furthermore, about 20% of children continue to experience seizures despite drug treatment.[8] The effectiveness of ketogenic diets (e.g., Atkins diet) is supported by low quality evidence.[9] However, these diets are also associated with adverse effects (e.g., gastrointestinal symptoms, dyslipidemia, decreased growth, and kidney stones), and require considerable caregiver effort to maintain. Surgical interventions may isolate and remove the underlying neurological cause of seizures, but carry risks of bleeding, infection, hydrocephalus, and new neurologic deficits.

Epilepsy treatment decision-making is thus complex and needs to consider each child's unique form of epilepsy, evidence of each treatment's potential benefits and harms, previous or ongoing experiences with treatment(s), and family's values and preferences. Although studies of treatment effectiveness rarely explore how families navigate these complex considerations or how these considerations may evolve over time, qualitative studies can offer context on caregiver experiences and decision-making. Syntheses of these qualitative studies can help shape caregiver and healthcare provider interactions and inform shared decision-making tools and processes. The purpose of this review is to summarize the qualitative research regarding caregivers' perspectives and experiences of treatments for childhood epilepsy.

Methods

We conducted a qualitative evidence synthesis in accordance with standards for qualitative evidence syntheses.[10-12]

Data sources and searches

We searched EMBASE, PubMed (in process), CINAHL, PsycINFO, SocINDEX, and Web of Science for studies published from January 1, 1999, to August 19, 2021, using terms related to epilepsy, treatments, and caregiver perspectives (Appendix A). The search was limited to English-language. We also ran forward and backward citation searches (i.e., snowball searching) on included studies to ensure relevant studies were not missed.

Study selection

We included qualitative studies that sought to understand caregiver's perspectives on ASMs, diet, or surgical treatments for childhood epilepsy. Studies needed to use qualitative methods for both data collection (e.g., focus group, individual interviews, or open-ended survey questions) and data analysis (e.g., thematic analysis). We excluded studies that focused only on non-caregiver perceptions (e.g., patient or healthcare provider) or that focused only on caregiver stress or caregiver expectations of treatment. We did not restrict by study country or care setting.

One reviewer screened titles and abstracts of citations retrieved from searches for eligibility. Two independent reviewers screened the full text of potentially relevant citations in PICO Portal© (https://picoportal.org/). Disagreements were resolved through discussion of the full team.

Data extraction and quality assessment

For each study, we extracted details on the study design and methodological features, population characteristics, and qualitative analysis findings.

One reviewer assessed risks to rigor using the Critical Appraisal Skills Programme (CASP) tool for qualitative studies, which appraises research aims, congruence between research aims and methodological approach, quality of sampling and data collection, appropriateness of application of methods, richness and conceptual depth of findings, appropriateness of interrogation of findings, and researcher reflexivity.[13] All team members reviewed the CASP assessments to ensure consistency of ratings across studies.

Two team members independently extracted and coded the qualitative findings of the included studies in MaxQDA© 2020 (Berlin, Germany), an online platform designed to support qualitative data management, extraction, and analysis. The extracted data included direct quotes from the participants (first order statements) and summary statements written by the study authors (second order statements). Extracted data were imported into spreadsheets to facilitate data cleaning, confirmation of themes, and synthesis.

To categorize the extracted data, we used the Theoretical Domains Framework (TDF),[14, 15] which was developed to assist in identifying the cognitive, affective, social, and environmental factors that may influence an individuals' performance of a health behavior. The health behavior of interest for this review was the decision to pursue an epileptic treatment. The 14 domains include: knowledge; skills; social/professional role and identity; belief about capabilities; optimism; beliefs about consequences; reinforcement; intention; goals; memory, attention, and decision processes; environmental context and resources; social influences; emotion and behavioral regulation (Appendix B). A third reviewer confirmed TDF domain codes, and the team discussed the coding to ensure accuracy and consistency both within and across TDF domains. One reviewer did a final confirmation of extracted text and coding to ensure no data were missed and that there was consistency across domains.

Data synthesis and analysis

We adopted a best-fit framework approach to guide our qualitative synthesis. In this approach, data are coded according to TDF domains. One reviewer then summarized key themes within each TDF domain. Themes were discussed and debated among the team until consensus was achieved. We used the finalized themes to develop summary statements and assessed our confidence in these statements using Grading of Recommendations Assessment, Development, and Evaluation Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual).[16-18] We rated our confidence in the summary statements as either high, moderate, low, or very low based on our assessment of the four GRADE-CERQual domains: methodological limitations, relevance, coherence, and adequacy of the data (Appendix C for definition of domains). "High confidence" refers to a finding that is highly likely to be a reasonable representation of the phenomenon of interest, whereas "very low confidence" refers to a review finding in which it was unclear if the finding was a reasonable representation.

Patient and public involvement

None.

Results

The literature search yielded 1,447 citations from searches (see PRISMA flow diagram, Figure 1). We found 66 citations to retrieve for full-text screening of which five studies (in six publications) were included in the final sample.[19-24] Table 1 presents characteristics of included studies.

All studies examined parents' perceptions, experiences, and decision-making leading to surgery. All studies were conducted after the children had undergone surgery, with one study also surveying parents "just prior" to surgery.[24] Three studies reported data on parents' perceptions and experiences with medications and diet, but only in the context of selecting surgery (e.g., parents considering surgery due to the undesirable side effects or uncontrolled epilepsy with prior treatments).[19, 21, 24] In total, the views of 186 parents are represented in the five studies (the majority of whom were mothers, n=115). The time from children's surgery to data collection ranged from 6 months to 10 years among the three studies reporting on timing. Four studies collected parent perspectives through semi-structured interviews conducted in-person or over the telephone (length ranging from 10 to 75 minutes),[19-23] and one study captured parent perceptions through open-ended survey questions.[24]

All studies were assessed to have minor risk to rigor (Appendix D) due to the retrospective nature of recruitment and the lack of consideration (or reporting) of the relationship between researchers and participants. Otherwise, studies were considered appropriate in their use of qualitative design, methods of data collection, and analysis. The retrospective nature of the included studies raises concerns about recall bias, as surgical outcomes may have affected retrospective perceptions, and selection bias since studies only included parents of children who were referred to surgery and proceeded with surgery (and possibly experienced some level of success with surgery). All but one study[23] did not report whether interviewers were part of the child's care team, which may have influenced responses. Ozanne et al. reported that the researchers that interviewed parents and analyzed the data were not part of the epilepsy surgery team.[23]

We identified and coded data for six of the 14 TDF domains: knowledge; emotion; social/professional role and identity; social influence; beliefs about consequences; and environmental context and resources (Figure 2). We did not find evidence from extracted qualitative data for the remaining eight TDF domains. Appendix E provides the extracted text from studies linked to their synthesized themes.

The GRADE-CERQual table (Table 2) summarizes findings and conclusions for each TDF domain. We had no or minor concerns with the coherence of the findings (i.e., the synthesized findings reflect the complexity and variation of the data) or their relevance (i.e., the extent to which synthesized findings are applicable to the context specified in the review question). We had minor or moderate concerns with the adequacy (i.e., the degree of richness and quantity of the data supporting the synthesized finding) of the findings related to knowledge and environmental context and resources, respectively. Below, we report the summary statement (and associated GRADE-CERQual level of confidence) under each identified TDF domain and summarize the key themes that contributed to the statements.

TDF Domain 1: Knowledge

Summary statement: *Evidence from four studies indicated that caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes* *contradictory information, caregivers value learning this new language so they can become better advocates for their child (Moderate confidence)* [19-23]

Once parents recognized "something [was] wrong" with the health of their child, they sought information from multiple sources "to understand seizures and epilepsy and become better advocates for their child."[19] Parents wanted information about medications,[19] surgery,[19, 21] and what types of doctors were needed to care for their child.[20] They also noted needing to learn about how to navigate the health system including multiple specialties, hospitals, and insurance procedures.[19]

Parents often perceived the information they received from professionals as inadequate to make informed decisions.[21] They described seeking lay language information from multiple sources, including additional professionals, the internet, books, family, friends, and other parents.[19, 23] Parents expressed discomfort in the "lack of understandable information" and "information imbalance" between themselves and their child's providers and sought to become "epilepsy experts in order to be effective advocates for their children."[20] After increasing their knowledge, parents felt more empowered to ask questions and participate more actively in the decision-making.[19, 21] Parents thus described active learning to ensure productive information exchanges with providers (including using clinical terms) to support a positive shared-decision making process.[20]

TDF Domain 2: Social role and identity

Summary statement: E*vidence from one study indicated that caregivers feel a sense of duty and need to do the right thing in selecting a treatment for their child (Moderate confidence).*[21]

Parents reported a "sense of duty" to be "strong" and "brave." They also reported feeling pressure to "'do the right thing' by their child; that is, choosing the treatment option that would give their child the best chance of "reaching their full potential."[21]

TDF Domain 3: Emotion

Summary statement: *Five studies provided evidence to suggest that caregivers experience the journey of navigating their child's epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation, fear to relief, and hope (Moderate confidence).*[19-23]

Parents expressed feeling drained and stressed from always being "on call,"[19] worrying about a seizure,[19, 22] or mood and behavioral difficulties.[22] Parents recalled feeling frustrated[19, 20] and desperate to "find a treatment option that would work."[20] For example, one mother was frustrated that her child's provider continued to perform medication trials to manage seizures and not discuss surgery as an option,[19] while another mother was desperate to get her son to another doctor but needed to wait for referrals because of insurance.

When considering surgery as a potential treatment option, parents reported the decision process as "difficult," "frightening," and "stressful."[20, 21] Parents feared surgery would lead to worse health outcomes for their child, [21, 24] change their child's personality, or cause a loss of function (e.g., partial or complete loss in speech or movement).[24] Parents also feared the possibility of surgery making seizures worse (e.g., increased frequency, duration). Thus, making the decision to have surgery "in vain."[24]

However, for many parents, the decision to pursue surgery brought relief and feelings of hope. Parents in one study expressed gratitude once they finally decided to pursue surgery as they felt it was "demanding to not know if surgery would be possible."[23] Parents also reported feeling hope for improvements in their child's health and wellbeing after they decided to choose surgery. Finally, after making the decision to pursue surgery (or learning it was a viable option), parents reported that they hoped for candidacy,[19, 21] and experienced relief when a date was set.[21]

TDF Domain 4: Social influence

Summary statement: *Three studies provided evidence to suggest that surgery is a family decision that requires outside support from other caregivers "experiencing the same thing" and from a healthcare professional acting as "champions." (Moderate confidence)* [20-22]

Parents reported the value of connecting with other parents who were on a similar treatment journey (e.g., had a child with epilepsy that was unresponsive to medication) and had experiences and expertise that they could draw on.[20, 21] Peer connections helped parents understand the surgical procedural from a "parental perspective" and provided "emotional support."[21] Parents also reported the value of having a good team of epilepsy providers and a "champion" (e.g., doctor, nurse, or social worker) to advocate for them and help them navigate purposefully through their journey.[20] One mother described one such champion, a nurse coordinator, as a "life saver," who helped in getting referrals and pushed her to seek better care for her child."[20] Another mother described her child's pediatrician as her champion because "he encouraged me and gave me…confidence."[20]

Parents described seeing treatment decision-making as a family choice and indicated the importance of involving partners, siblings, grandparents, and the child (where developmentally appropriate). Parents found engaging the whole family in the decision-making process to be generally helpful, as the experience of the child's epilepsy and potential consequences of treatment impacted the whole family.[21] However, for some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary parent, she "witnessed" the true extent of their child's seizures.[21]

TDF Domain 5: Beliefs about consequences

Summary statement*: Five studies provided evidence to suggest that parents undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.* [19-24]

Parents of children who went on to have surgery reported going through a transformation in thinking of surgery as a "last resort" to a "necessary and hopeful option."[19] This transformation in thinking evolved as parents acquired greater understanding about their child's illness and prognosis, experience in treating their child with other treatments, acquired knowledge about the potential risks and benefits of surgery, and processed the diverse emotions associated with surgery.[19, 20]

An important part of parents' moving toward a decision about surgery was weighing what they perceived to be the benefits and harms of surgery.[21, 24] In terms of benefits, parents hoped surgery would lead to improved outcomes such as seizure reduction and the opportunity for a "normal life" without the side effects of medications.[21, 24] Some parents were skeptical about the efficacy of surgery due to their experiences with medications and the ketogenic diet. According to parents, these treatments were associated with side effects, negatively affected their child's quality of life, and provided only minimal improvement in seizure control.[21] In terms of adverse effects, parents were concerned that surgery would lead to surgical complications or various post-surgical impairments, including a change in personality, loss of speech or motor function, and behavioral problems.[24] A small number of parents reported having no concerns before surgery.[24]

Beyond rational consideration of the benefits and risks of surgery, parents often reported coming to the decision to select surgery only after exhausting all other treatment options. [21-23] Parents expressed feeling like "it was the only choice [they] could make"[21] and their "only option"[21] in improving their child's outcomes or preventing their condition from getting worse. Parents understood there were potential complications associated with surgery, but "preferred to take a risk [in proceeding with surgery] rather than live in constant fear"[23] or having a child that "was unconscious all the time."[22]

TDF Domain 6: Environmental context and resources

Summary statement: *Three studies provided evidence that parents face challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process (Low confidence)*[19-21, 23]

Parents from one study in the U.S.[19, 20] and another in Sweden.[23] reported experiencing significant barriers with navigating the healthcare system. In the U.S.,[19, 20] parents expressed frustration with the extensive time it took to find the right doctor after navigating various doctors from different specialties across multiple institutions. Prior to selecting surgery for their child, parents reported difficulties in finding the "right doctor" with knowledge to "effectively identify the problem, and then make a clear plan of action." Parents attributed these difficulties to the lack of pediatric neurologists in their local area, inconsistencies in treatment recommendations, and rigid adherence to center-specific treatment protocols.[19, 20] Once parents made a decision to pursue surgery, they reported battles with insurance companies to pay for surgery.[19, 20] Parents in the Sweden study reported similar frustrations with "the bureaucracy" as they felt it took "a long time to get a correct diagnosis" and "adequate support." They "thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary.[23]

Parents listed several provider-specific interactions they found to be either enablers or barriers to their experience of identifying and selecting appropriate treatment for their child. With respect to enablers, parents valued when providers validated (and shared) their concerns, gave their time and fostered trust, and engaged parents in the treatment decision-making process. Barriers noted by parents included having their concerns doubted or ignored,[19, 23] receiving inadequate information or support (especially before the epilepsy was recognized to be drug-resistant),[23] and feeling excluded from discussions about their child's surgical candidacy.[21] One study reported that parents perceived physician variability in knowledge about epilepsy and their lack of understanding about the pre-surgical referral process and appropriateness of surgery as barriers.[19] Parents in one study reported how they felt more reassured when their child received care from a comprehensive team of professionals with diverse expertise.[20] Parents from one study suggested providers give an "earlier and softer introduction of surgery as a possible treatment option."[21]

See Box 1 for example excerpts for TDF domains.

Discussion

Understanding caregiver perspectives about treatments for childhood epilepsy is important to ensure that parents are appropriately supported during their decision-making process. Our qualitative evidence synthesis of five studies, which had minimal risks to rigor, identified key findings across six domains: Knowledge, Emotions, Social/Professional Role and Identify, Social Influences, Beliefs about Consequences, Environmental Context and Resources. However, the evidence was limited surgical treatment. Overall, these findings suggest that the decision to select surgery for childhood epilepsy involves parents going through a complex journey of acquiring extensive knowledge, working through intense emotions and perceived parental responsibilities, needing family and peer support, transforming beliefs about epilepsy and potential treatment options, and navigating various barriers and facilitators of the healthcare system.

These findings are supported by a similar review conducted by Samanta et al. (2021) that sought to understand caregiver decision-making around epilepsy surgery for children with drug-resistant epilepsy. [25] In their synthesis of a similar body of evidence, these reviewers identified the following as key determinants: knowledge and information, communication and care coordination, caregivers' emotional state, and socioeconomic factors. While the findings of our review are similar to Samanta, our scope and methodology differ. We sought to explore caregiver decision-making around all treatments for epilepsy, not just surgery. We also used rigorous methodological tools (e.g., TDF and GRADE-CerQual). Use of these tools strengthens our findings by placing them within a framework that identifies facilitators and barriers, and by providing criteria to establish confidence in the certainty of these findings.

Through the TDF framework, we identified decision domains that presented potential barriers to care that are amenable to action. For example, parents often felt that they did not have enough information to make decisions about surgery. They reported experiencing intense psychological distress and exhaustion during their decision-making process and felt these emotions acutely through their role as parents responsible for making a potentially life-altering decision for their child. Parents also reported valuing connections with peers who were going through (or had gone through) the same experience. Healthcare systems and providers may therefore consider providing parents with resources, such as patient navigators to help guide them through the healthcare system and better understand the care pathway. They can also help parents to access peer support and advocates[26, 27]

The findings also identified gaps in the evidence base regarding parents' perceptions and decision-making processes. Foremost, we did not identify any studies exploring parent decision-making around non-surgical treatment options. Qualitative studies that prospectively explore caregiver decision-making about these treatments are needed to determine if perspectives differ from those about surgery. We also did not identify evidence mapping to certain TDF domains that we expected to find evidence for, such as Goals. Studies included in this review were vague and inconsistent in reporting parent's goals or their desired outcomes of treatment. Some studies noted that parents wanted what 'was best' for their child or they would be happy with a reduction in seizure frequency. Survey data collected from parents considering surgery found the primary goal was seizure freedom (98%), followed by reduced medication (90%), and improved cognition (82%).[28] We also did not identify evidence for the domain of Memory, Attention, and Decision process. Thus, we could not determine if parents become more skilled and confident in their decision-making over time through experience and acquired knowledge.

Similarly, we found limited evidence for the domain of Environmental Context and Resources. Further evidence is needed to understand how culture and equity play a role in parents' perceptions about treatment and their capacity to access care for their child. For example, parents reported important barriers in knowledge, access to professionals to diagnose and treat their child, and challenges with accessing and paying for surgery. The extent to which these factors would be the similar among families from different cultural or socio-economic backgrounds or from countries with varying economic, educational, and social resources needs further exploration.

Strengths and limitations

This review followed contemporary standards for the conduct of qualitative synthesis, which includes the use of a best-fit framework approach, a theory-informed framework to guide our synthesis and extraction, and use of the CASP and GRADE-CerQual tool to assess the rigor and confidence of our findings. The use of the TDF is a particular strength of this review as it lends itself to both actionable interventions (e.g., mapping intervention strategies to key domains identified) and future research (e.g., further examination of domains not identified in the evidence such as 'memory, attention, and decision processes'). However, as with all qualitative research, selecting which data to extract and how to code it is ultimately a subjective process. We attempted to limit subjectivity within our group by coding in duplicate and having regular meetings to ensure consistency across and within domains.

One primary limitation is that the data in all studies included in this were collected retrospectively. Parents were asked for their perceptions *after* their child had undergone surgery. This may have resulted in selection bias, as studies recruited parents of children referred to and proceeded with surgery. Thus, findings may not be fully reflective of the wider population of parents who are making decisions regarding surgery. The perceptions of parents who declined surgery were not captured. The retrospective nature of the data collection may have also resulted in recall bias. The time from children's surgery to data collection ranged from 6 months to 10 years. Parents may have forgotten important aspects of their journey to surgery or filled in gaps of memory due to experiences with the child's outcome. As described by one study, parent responses were "memories processed through emotions and colored from further experiences, which were then developed into opinions and personal views."[23]

Conclusion

Parents of children with epilepsy navigate a complex process to decide whether to have their child undergo surgery. Educational resources, peer support, and patient navigators may help support parents through this process. More qualitative studies are needed to fully understand the diversity of experiences of parents across various points in the decision-making pathway and among different healthcare contexts. Qualitative studies are needed that address parents' perceptions and experiences with selecting non-surgical epilepsy treatment options such as diet and medications.

Contributors: SU served as project lead and participated as a team member in identifying studies for inclusion, extracting and coding data, synthesizing findings, developing and grading summary statements, and drafting of initial report and manuscript; KK participated as team member in all aspects of data extraction, coding, synthesis, developing and grading summary statements, validating all findings, and revising initial drafts of manuscript; RW participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; GA coordinated and revised search strategies and participated in all aspects of data extraction, coding, synthesis, and review of manuscript; KR participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript; MV participated in all aspects of data extraction, coding, synthesis, developing summary statements, and reviewing manuscript.

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Box 1. Example excerpts for TDF domains

Knowledge

- [Parent]: “I googled it until I couldn’t google anymore, and I think that gave me a better understanding of what was going on.”[21].
- A mother who stated, “I just think knowledge is power and it also brings some comfort to making a good decision,” exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy[19]

Social/Professional Role and Identity

- Parents discussed feeling pressure from a need to “do the right thing” by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential.[21]

Emotion

- Participant descriptions highlighted the significant impact that epilepsy had upon on the child’s and family’s lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: “you’re tired yourself before you even go out the door”[22]
- [Parent]: “At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric”[20]

Social Influences

- Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. “Gosh, we didn’t tell anybody. We were ashamed.”[22]
- [E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information” [20].

Beliefs about Consequences

- This transformation occurred as parents became increasingly aware of the severity of their child’s epilepsy and its impact on their child’s and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment [19]
- They wanted that their child could do everything their friends could do like “play football, cycle, and swim independently”. Some parents mentioned that they wanted their child to be able to go back to school and have a “proper social life” while some parents referenced the future hoping that the child would be better placed to get a job and driving license as a result of surgery[24]
- Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.[23]

Environmental Context and Resources

- Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.[23]
- Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, “‘I don’t believe that she is a surgical candidate.’ As a parent, I listened and I wanted to think otherwise.... In hindsight, I’m glad I didn’t listen to him.”[19]

Table 1. Characteristics of included studies

Study, year, country	Population	Number of parents	Time from surgery to data Collection	Study aim	Data collection methods	Data analysis methods
Baca[1], 2015; Pieters, 2016, USA	Parents of children who had previously undergone resective epilepsy surgery	37 (individual parents) 31 mothers; 6 other	NR	Baca, 2015: To identify the nature and range of parent-perceived barriers to timely receipt of pediatric epilepsy surgery. Pieters, 2016: A) To describe thoroughly the parental experiences and perceptions of this slow and arduous period prior to the presurgical referral and evaluation to a comprehensive pediatric epilepsy center. B) To delineate the range of parent identified factors, or facilitators, that helped move families forward along their journey to surgery	Semi-structured interviews lasting on average 29 minutes (range 10 to 60 mins)	Thematic analysis
Heath, 2016, UK	Parents of children who had undergone pediatric epilepsy surgery Also interviewed healthcare professionals caring for children with epilepsy	9 (individual parents) 8 mothers; 1 father 10 healthcare professionals	NR	To explore how parents and health professionals make decisions regarding pediatric epilepsy surgery to identify: A) factors that influence the process of decision-making regarding pediatric epilepsy surgery from the perspective of parents and professionals and B) the support needs of those considering surgery as a treatment option for a child with medically intractable epilepsy	Semi-structured interviews lasting about 1 hour, and non-participant observations	Thematic analysis
O'Brien, 2020, UK	Parents and their children who had gone through resective epilepsy surgery	9 parents (1 couple; 7 individual parents) 8 mothers; 1 father	6 months to 3 years	To explore children's and parents' perspectives on the journey prior to and following surgical treatment, with a focus on the emotional experiences of children and their parents throughout the surgery journey	Semi-structured interviews lasting 25 to 75 mins	Thematic analysis
Ozanne, 2016, Sweden	Parents of children who had previously undergone resective epilepsy surgery	24 parents 13 mothers; 11 fathers	5 to 10 years	To explore parental experiences before and after hemispherotomy as reported at a long-term follow-up and the parents' views on received information and support	Interviews (length of time not reported)	Content analysis
Sylven, 2020, Sweden	Parents of children who were undergoing resective epilepsy surgery (and 2 years post-surgery)	107 (parental responses to both surveys)	2 years	A) To understand parental hopes and worries before their child underwent epilepsy surgery and B) To understand parental satisfaction two years after their child had undergone epilepsy surgery	Surveys (open-ended questions)	Thematic analysis

[1] Baca & Pieters included the same patient population but reported on different aspects of parent perspectives.

Table *2.* GRADE-CERQual summary of findings statements and ratings

Summary of Review Findings	**Studies contributing**	**Methodological limitations (CASP ratings)**	**Coherence**	**Adequacy[2]**	**Relevance**	**CERQual assessment**	**Explanation of CERQual assessment**
Knowledge: Caregivers value information about epilepsy, its treatment options, and navigating the healthcare system to access timely and effective treatment for their child. Despite feeling overwhelmed by the complexity and sometimes contradictory information, caregivers value learning this new language so they can become better advocates for their child	Baca/Pieters Heath O'Brien Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concerns*: Included studies limited to surgery as treatment decision. We did not identify studies meeting inclusion criteria that addressed other treatments, such as diet or medications. Each study addressed knowledge acquisition and reported on it in some depth.	*No or very minor concerns*	*Moderate confidence*	Four studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Social/professional role and identity, parents feel a sense of duty and need to do the right thing in selecting a treatment for their child.	Heath	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*Minor concern:* Included studies limited to surgery as treatment decision.	*No or very minor concerns*	*Moderate confidence*	One study; minor concerns related to methods and limited treatment scope (surgery).
Emotion, parents experience the journey of navigating their child's epilepsy and ultimately selecting surgical treatment as an emotionally fraught one with emotions ranging from exhaustion, desperation and fear to relief and hope.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*: All 4 studies contributed to the rich depth of the varied emotions from fear of the illness to fear of the surgery to the mental toll that took on families and hope	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).

Summary of Review Findings	**Studies contributing**	**Methodological limitations (CASP ratings)**	**Coherence**	**Adequacy**[2]	**Relevance**	**CERQual assessment**	**Explanation of CERQual assessment**
				and relief that surgery may offer			
Social influences, surgery is a family decision that requires outside support from other parents "experiencing the same thing" and from a healthcare professional acting as "champions."	Pieters Heath O'Brien	*Minor concerns:* Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns*: Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Three studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Beliefs about consequences, caregivers undergo a transformation from seeing surgery as a last resort to the only option for their child to have a chance at a better life. Surgery became a viable option as parents realized that their child's current treatment was not working or it had unacceptable side effects, and some saw the side effects of surgery as less daunting than the disease.	Baca/Pieters Heath O'Brien Ozanne Sylvén	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*No or very minor concerns*	*No or very minor concerns*	*Moderate confidence*	Five studies with similar findings; minor concerns related to methods and limited treatment scope (surgery).
Environmental context and resources, parents expressed challenges in navigating the healthcare system and interacting with professionals to find the "right doctor" or care team for their child. Parents value having their concerns heard and being engaged in the decision-making process.	Baca/Pieters Heath Ozanne	*Minor concerns*: Nature of relationship between researchers and participants not reported; retrospective recruitment.[1]	*No or very minor concerns:* Findings reflect the complexity and variation of data	*Moderate concerns*: The three studies are limited to surgery and to US, Sweden and the UK. Healthcare barriers seemed particularly salient to U.S. respondents (including insurance barriers)	*No or very minor concerns*	*Low confidence*	Three studies with similar findings; moderate concerns related to methods, limited scope (surgery), and context-specific healthcare experiences of participants

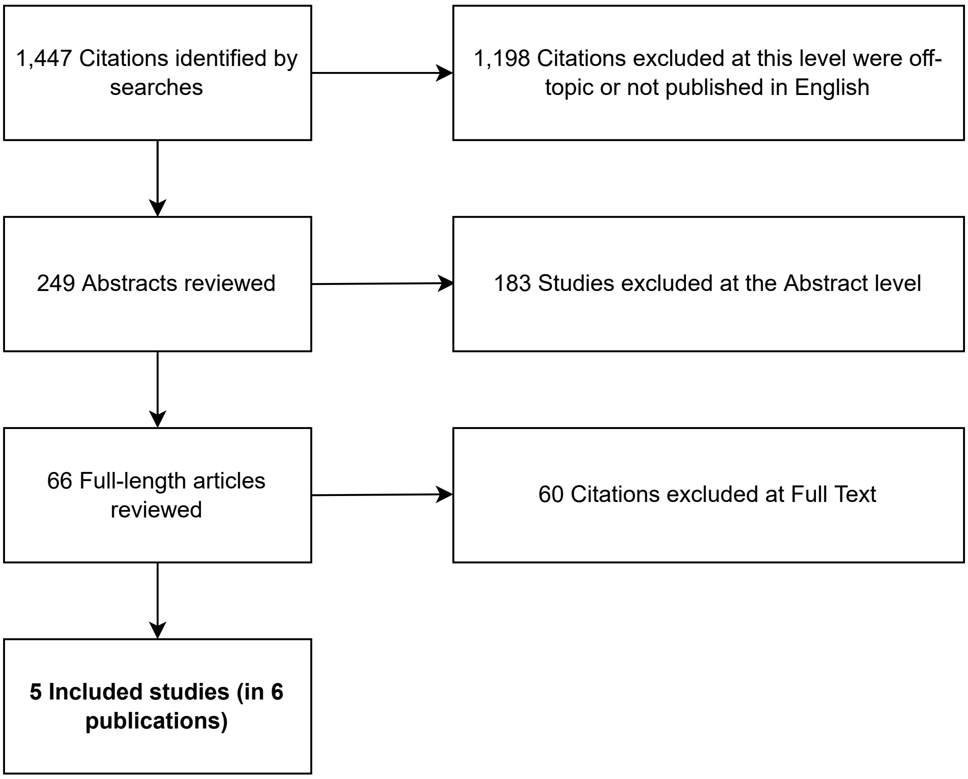
[1] Retrospective recruitment raises concerns about selection bias.

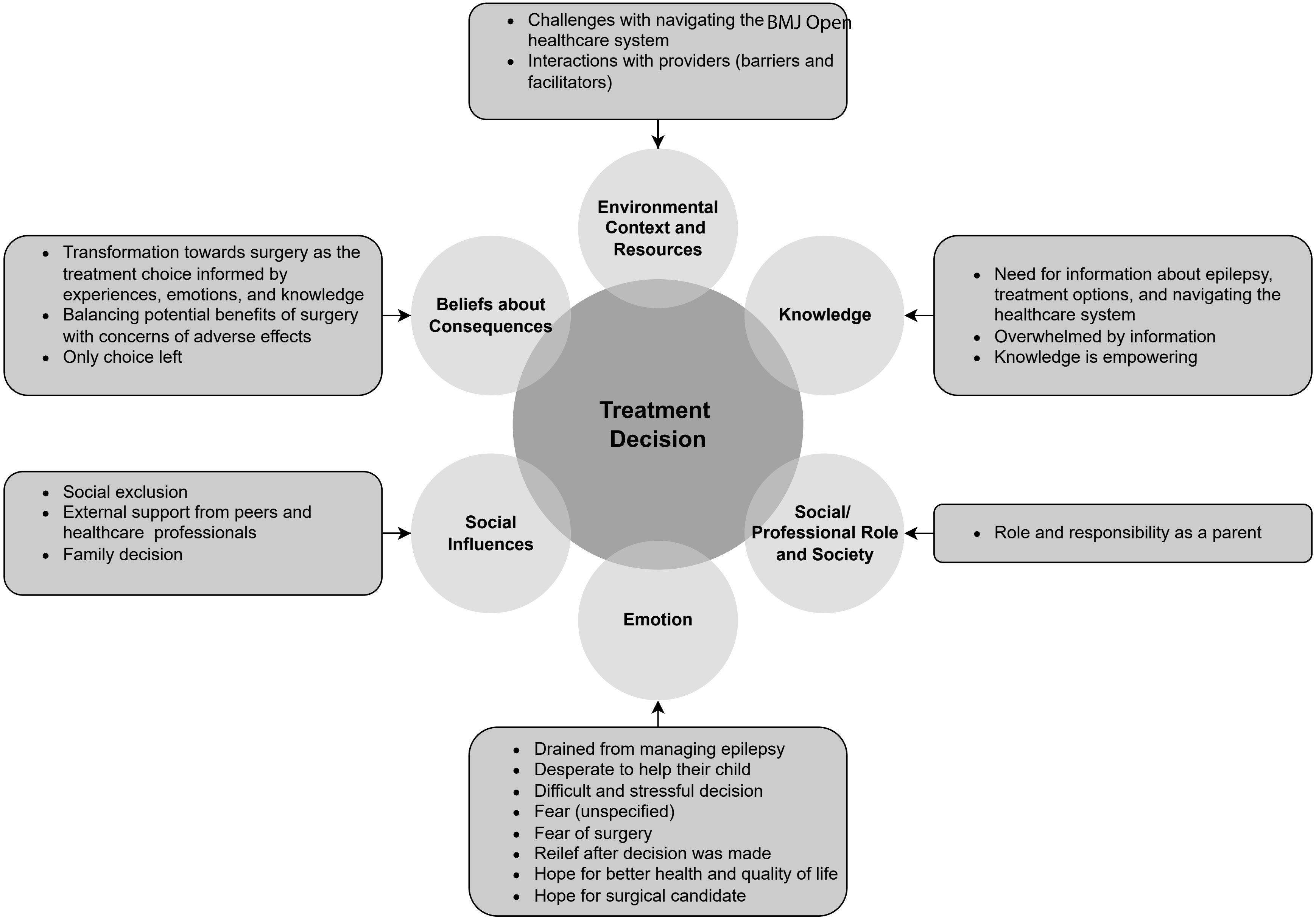
[2] We only downgraded for adequacy due to studies being limited to surgery when the summarized theme was based on parent's general feelings about epilepsy and could thus potentially apply to parent decisions regarding other treatment choices.

FIGURE TITLES

Figure 1. PRISMA flow diagram of studies identified

Figure 2. The six theoretical domains identified in this review that impacted treatment decision-making and the main themes representing those domains


1,447 Citations identified by searches
1,198 Citations excluded at this level were off-topic or not published in English
249 Abstracts reviewed
183 Studies excluded at the Abstract level
66 Full-length articles reviewed
60 Citations excluded at Full Text
5 Included studies (in 6 publications)


Challenges with navigating the healthcare system
Interactions with providers (barriers and facilitators)
Environmental Context and Resources
Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Balancing potential benefits of surgery with concerns of adverse effects
Only choice left
Beliefs about Consequences
Knowledge
Need for information about epilepsy, treatment options, and navigating the healthcare system
Overwhelmed by information
Knowledge is empowering
Treatment Decision
Social exclusion
External support from peers and healthcare professionals
Family decision
Social Influences
Social/ Professional Role and Society
Role and responsibility as a parent
Emotion
Drained from managing epilepsy
Desperate to help their child
Difficult and stressful decision
Fear (unspecified)
Fear of surgery
Reilef after decision was made
Hope for better health and quality of life
Hope for surgical candidate

APPENDIX A: Search Strategy

EMBASE (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Infantile/ Pediatric Epilepsies	'benign childhood epilepsy'/exp OR 'childhood absence epilepsy'/exp OR 'severe myoclonic epilepsy in infancy'/exp OR (dravet* NEXT/1 (disease OR syndrome))
2	0-3 Age Group and Epilepsy	[infant]/lim OR [newborn]/lim OR newborn/exp OR [preschool]/lim OR 'preschool child'/exp OR toddler/exp OR (babies OR baby OR child*:ti OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR 'very young'):ab,ti,kw OR ('younger than' OR under OR below) NEAR/3 (3 OR three) OR (3 OR three) NEAR/3 ('or below' OR 'or under' OR 'or younger') AND ('epilepsy'/exp OR 'epileptic patient'/exp OR epilep*:ti)
3	Infantile Spasms/ Neonatal Seizures (not requiring epilepsy)	'infantile spasm'/exp OR (((infan* OR neonat* OR newborn*) NEAR/2 (convuls* OR seizure* OR spasm*)):ab,ti,kw)
4	Other Seizures (not requiring Epilepsy)	([infant]/lim OR [newborn]/lim OR 'newborn'/exp OR babies:ab,ti,kw OR baby:ab,ti,kw OR infan*:ab,ti,kw OR neonat*:ab,ti,kw OR newborn*:ab,ti,kw OR nicu:ab,ti,kw) AND ('febrile convulsion'/exp OR 'seizure'/exp OR convuls*:ab,ti,kw OR spasm*:ab,ti,kw OR seizure*:ab,ti,kw)
5	Pharmacologic/ Vitamin Treatment	acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide
6	Diet Therapy	'ketogenic diet'/de OR (keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic'):ab,ti,kw
7	Surgical Procedures	craniotomy/de OR hemispherectomy/de OR 'laser surgery'/de OR lobectomy/de OR 'corpus callosotomy' OR craniotom* OR (disconnect* NEAR/3 (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* NEAR/3 (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar NEAR/3 disconnect*) OR (palliat* NEAR/3 operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection'
8	Brain Stimulation	'brain depth stimulation'/de OR 'brain responsive neurostimulator'/de OR 'deep brain stimulator'/de OR 'nerve stimulation'/de OR 'nerve stimulator'/de OR 'vagus nerve stimulation'/de OR ('brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') NEAR/2 (electrostim* OR stimulat*)) OR neurostim*

9	Harms	anhidrosis/de OR 'adverse event'/de OR 'adverse drug reaction'/de OR 'behavior disorder'/de OR 'cognitive defect'/de OR 'developmental delay'/de OR 'developmental disorder'/de OR dystonia/de OR 'liver injury'/de OR 'loss of appetite'/de OR 'motor dysfunction'/de OR 'organ damage'/de OR 'patient harm'/de OR 'sleep disorder'/de OR sweating/de OR (advers* OR harm* OR 'side effect'):ab,ti,kw OR anhidrosis OR (appetite NEAR/3 (lose OR losing OR loss)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) NEAR/3 (effect* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR neurodevelopment*) NEAR/3 (delay* OR disorder* OR regress*)) OR dystonia OR hypohidrosis OR hypohydrosis OR (liver NEAR/3 (damag* OR injur*)) OR (miss* NEAR/3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) NEAR/3 (disorder* OR inability OR unable))
10	Parental Preferences	parent/de OR (parent* OR mother* OR father*):ab,ti,kw
11	Untreated Disease	'treatment refusal'/de OR ('not treated' OR 'no treatment' OR untreat*):ab,ti,kw OR (declin* OR forgo* OR 'not' OR no OR refus* OR withheld OR withhold*) NEXT/3 (treated OR treatment*)
12	Study Designs/ Publication Types	[english]/lim AND [1999-2020]/py NOT ([animals]/lim NOT [humans]/lim OR abstract:nc OR annual:nc OR 'book'/de OR ((case NEXT/1 (report* OR stud*)):ti) OR 'case report'/de OR 'case study'/de OR conference:nc OR 'conference abstract':it OR 'conference paper'/de OR 'conference paper':it OR 'conference proceeding':pt OR 'conference review':it OR congress:nc OR diagnos*:ti OR 'diagnosis'/mj OR 'diagnostic accuracy'/mj OR 'diagnostic procedures'/mj OR 'diagnostic test'/mj OR 'diagnostic test accuracy study'/mj OR 'differential diagnosis'/mj OR 'editorial'/de OR editorial:it OR 'erratum'/de OR guideline*:ti OR letter:it OR 'note'/de OR note:it OR meeting:nc OR 'practice guideline'/de OR 'review'/exp OR sessions:nc OR 'short survey'/de OR symposium:nc OR animal*:ti OR experimental:ti OR (vitro:ti NOT vivo:ti) OR canine:ti OR dog:ti OR dogs:ti OR mouse:ti OR mice:ti OR rabbit*:ti OR rat:ti OR rats:ti OR rodent*:ti OR sheep:ti OR swine:ti)
13	Combine sets	#1 OR #2 OR #3 OR #4
14	Combine sets (KQ1 Pharmacology)	#13 AND #12 AND #5
15	Combine sets (KQ2 Diet, Surgery, Brain Stimulation)	#13 AND #12 AND (#6 OR #7 OR #8)
16	Combine sets (KQ3 Treatment Harms)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND 9
17	Combine sets (CQ1 Parental Preferences)	#13 AND #12 AND (#5 OR #6 OR #7 OR #8) AND #10
18	Combine sets (CQ2 Untreated/ Uncontrolled Epilepsy)	#11 AND #12 AND #13

19	Combine Sets All KQs	#14 OR #15 OR #16
20	Combine Sets All CQs	#17 OR #18

SocINDEX and Web of Science databases (Searched January 1, 1999, to August 19, 2021)

(epilep* OR convuls* OR seizure* OR spasm*)

AND

(acetazolamide OR acth OR 'adrenocorticotropic hormone' OR benzodiazepine* OR brivaracetam OR bromide OR cannabidiol OR carbamazepine OR clobazam OR clonazepam OR clorazepate OR corticotropin OR divalproex OR eslicarbazepine OR ethosuximide OR everolimus OR felbamate OR fenfluramine OR folate OR 'folic acid' OR frisium OR gabapentin OR lacosamide OR lamotrigine OR levetiracetam OR liposteroid OR lorazepam OR mesuximide OR methsuximide OR onfi OR oxcarbazepine OR perampanel OR phenobarbital OR phenytoin OR prednisone OR pregabalin OR primidone OR pyridoxine OR 'pyridoxal 5 phosphate' OR rufinamide OR sabril OR stiripentol OR thiopental OR thiopentone OR tiagabine OR topiramate OR valproate OR 'valproate semisodium' OR 'valproic acid' OR vigabatrin OR zonisamide OR keto* OR ketogenic OR 'low glycemic index' OR 'medium chain triglyceride' OR 'modified atkins' OR 'modified keto' OR 'modified ketogenic' OR 'corpus callosotomy' OR craniotom* OR (disconnect* AND (hemispher* OR surg* OR procedure*)) OR hemispherecotom* OR hemispherotom* OR lesionectom* OR lobectom* OR (laser* AND (ablat* OR operat* OR procedure* OR surg*)) OR (multilobar AND disconnect*) OR (palliat* AND operat* OR procedure* OR surg*) OR resect* OR resection OR transect* OR transection* OR 'sublobar resection' OR 'subpial transection' OR 'brain stimulat*' OR 'deep brain stimulat*' OR 'electric brain stimulat*' OR 'external trigeminal nerve stimulat*' OR 'responsive brain stimulat*' OR 'responsive neurostimulat*' OR 'vagus nerve stimulat*' OR stimulation OR stimulator*) OR ((brain OR 'deep brain' OR electric* OR responsive OR 'vagus nerve') AND (electrostim* OR stimulat*)) OR neurostim*)

AND

(parent* OR mother* OR father*)

AND

(perception OR factor* OR decision)

CINAHL (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR dravet*
2	Epilepsy/ 0-3 Age Group	(babies OR baby OR child* OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool* OR toddler* OR "very young" OR "younger than three" OR "younger than 3" OR "under three" OR "under 3" OR "below three" OR "below 3" OR "3 or below" OR "3 or younger" OR "three or below" OR "three or younger") AND epilep*
3	Infantile Spasm/ Neonatal Seizure	"infantile spasm*" OR "neonatal seizure*" OR ((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) AND (convuls* OR seizure* OR spasm*))
4	Parental Preferences	father* OR matern* OR mother* OR parent* OR patern*
5	Harms	"adverse drug reaction" OR "adverse effect*" OR "adverse event*" OR anhidrosis OR appetite* OR behavior* OR behaviour* OR cognitiv* OR defect* OR delay* OR development* OR disorder* OR dysfunction* OR dyston* OR harm* OR impair* OR injur* OR liver OR motor OR "organ damage*" OR "sleep disorder*" OR sweat* OR "side effect*"
6	Untreated Disease	"treatment refusal" OR "not treated" OR "no treatment" OR untreat* OR "decline treatment" OR "declined treatment" OR "forgo* treatment" OR "refuse treatment" OR "refused treatment" OR "refusing treatment" OR "withheld treatment" OR "withhold treatment" OR "withholding treatment"
7	Combine Concepts	(S1 OR S2 OR S3) AND S4
8	Apply Limits	S7 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)
9	Combine concepts	(S1 OR S2 OR S3) AND (S5 AND S6)
10	Apply Limits	S9 AND (Published Date: 19990101-20210831; English Language; Exclude MEDLINE records)

PsycINFO (Searched January 1, 1999, to August 19, 2021)

Set	Concept	Search statement
1	Childhood Epilepsy	"benign childhood epilepsy" OR "childhood absence epilepsy" OR "severe myoclonic epilepsy in infancy" OR (dravet* adj1 (disease OR syndrome)) OR ((child* OR infan* OR paediatr* OR pediatr*) adj1 epilep*)
2	Epilepsy/ 0-3 Age Group	(baby OR babies OR infan* OR neonat* OR newborn* OR nicu OR paediatric* OR pediatric* OR preschool OR toddler* OR "very young" OR (("younger than" OR under OR below) adj3 ("3" OR three)) OR (("3" OR three) adj3 ("or below" OR "or under" OR "or younger")) AND (epilepsy/ OR epileptic seizures/ OR epileps* OR epileptic*))
3	Infantile Spasm/ Neonatal Seizure	((babies OR baby OR infan* OR neonat* OR newborn* OR nicu) adj3 (convuls* OR seizure* OR spasm*))
4	Parental Preferences	parents/ OR (patern* OR parent* OR matern* OR mother* OR father*)
5	Harms	behavior disorders/ OR cognitive impairment/ OR delayed development/ OR developmental disabilities/ OR liver disorders/ OR motor development/ OR "side effects (drug)"/ OR "side effects (treatment)"/ OR sleep wake disorders/ OR sweating/ OR (advers* OR anhidrosis OR dystoni* OR harm* OR harm* OR hypohidrosis OR hypohydrosis OR "organ damage") OR (appetite adj3 (lose* OR losing OR loss OR

		lost)) OR ((cognitiv* OR behavior* OR develop* OR motor OR movement OR neurodevelop*) adj3 (effect* OR deficit* OR disorder* OR problem* OR symptom*)) OR ((cognitiv* OR develop* OR motor* OR neurodevelopment*) adj3 (delay* OR disorder* OR regress*)) OR ((liver OR organ*) adj3 (damag* OR injur*)) OR (miss* adj3 milestone*) OR ((eat* OR perspir* OR sweat* OR sleep*) adj3 (disorder* OR inability OR unable))
6	Untreated Disease	treatment refusal/ OR ("not treated" OR "no treatment" OR untreat*) OR ((declin* OR forgo* OR "not" OR no OR refus* OR withheld OR withhold*) adj3 (treated OR treatment*))
7	Combine Sets	(1 OR 2 OR 3) AND 4
8	Apply Limits	limit 7 to yr="1999 - 2021"
9	Apply Limits	limit 8 to up=19000101-20210819
10	Apply Limits	9 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
11	CQ1	limit 10 to english language
12	Combine Sets	(1 OR 2 OR 3) AND (5 AND 6)
13	Apply Limits Apply Limits	Limit 12 to yr="1999 - 2021"
14	Apply Limits	limit 13 to up=19000101-20210819
15	Apply Limits	14 NOT (1* or 2* or 3* or 4* or 5* or 6* or 7* or 8* or 9*).pm.
16	CQ2	limit 5 to english language

APPENDIX B: The Theoretical Domains Framework*

Domain	Definition	Constructs
Knowledge	An awareness of the existence of something	Knowledge (including knowledge of condition/scientific rationale) Procedural knowledge Knowledge of task environment
Skills	An ability or proficiency acquired through practice	Skills Skills development Competence Ability Interpersonal skills Practice Skill assessment
Social/professional role and identity	A coherent set of behaviors and displayed personal qualities of an individual in a social or work setting	Professional identity Professional role Social identity Identity Professional boundaries Professional confidence Group identity Leadership Organizational commitment
Beliefs about capabilities	Acceptance of the truth, reality or validity about an ability, talent or facility that a person can put to constructive use)	Self-confidence Perceived competence Self-efficacy Perceived behavioral control Beliefs Self-esteem Empowerment Professional confidence
Optimism	The confidence that things will happen for the best or that desired goals will be attained	Optimism Pessimism Unrealistic optimism Identity
Beliefs about Consequences	Acceptance of the truth, reality, or validity about outcomes of a behaviour in a given situation	Beliefs Outcome expectancies Characteristics of outcome expectancies Anticipated regret Consequents
Reinforcement:	Increasing the probability of a response by arranging a dependent relationship, or contingency, between the response and a given stimulus	Rewards (proximal/distal, valued/not valued, probable/improbable) Incentives Punishment Consequents Reinforcement Contingencies Sanctions

Domain	Definition	Constructs
Intention	A conscious decision to perform a behaviour or a resolve to act in a certain way	Stability of intentions Stages of change model Transtheoretical model and stages of change
Goals	Mental representations of outcomes or end states that an individual wants to achieve	Goals (distal/proximal) Goal priority Goal/target setting Goals (autonomous/controlled) Action planning Implementation intention
Memory, attention and decision processes	The ability to retain information, focus selectively on aspects of the environment and choose between two or more alternatives	Memory Attention Attention control Decision-making Cognitive overload/tiredness
Environmental context and resources	Any circumstance of a person's situation or environment that discourages or encourages the development of skills and abilities, independence, social competence and adaptive behaviour	Environmental stressors Resources/material resources Organizational culture/climate Salient events/critical incidents Person environment interaction Barriers and facilitators
Social influences	Those interpersonal processes that can cause individuals to change their thoughts, feelings, or behaviours	Social pressure Social norms Group conformity Social comparisons Group norms Social support Power Intergroup conflict Alienation Group identity Modelling
Emotion	A complex reaction pattern, involving experiential, behavioural, and physiological elements, by which the individual attempts to deal with a personally significant matter or event	Fear Anxiety Affect Stress Depression Positive/negative affect Burn-out
Behavioral regulation	Anything aimed at managing or changing objectively observed or measured actions	Self-monitoring Breaking habit Action planning

*British spelling of definitions is kept from original publication

Atkins L, Francis J, Islam R, O'Connor D, Patey A, Ivers N, Foy R, Duncan EM, Colquhoun H, Grimshaw JM, Lawton R, Michie S. A guide to using the Theoretical Domains Framework of behaviour change to investigate implementation problems. Implement Sci. 2017 Jun 21;12(1):77. doi: 10.1186/s13012-017-0605-9. PMID: 28637486; PMCID: PMC5480145.

APPENDIX C. CERQual assessment components

CERQual Components	Definition
Methodological limitations	The extent to which there are problems in the design or conduct of the primary studies supporting a review finding. We used our CASP assessment of each study to guide our assessment of this component.
Coherence	An assessment of how clear and cogent the fit is between the data from the primary studies and the review finding.
Relevance	The extent to which the body of evidence from the primary studies supporting a review finding is applicable to the context specified in the review question.
Adequacy	The degree of richness and quantity of data supporting a review finding.

Noyes J, Booth A, Lewin S, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 6: how to assess relevance of the data. Implement Sci. 2018 Jan 25;13(Suppl 1):4. doi: 10.1186/s13012-017-0693-6. PMID: 29384080.

APPENDIX D. Methodological appraisal ratings using the CASP tool

	Study				
CASP question	**O'Brien, 2020**	**Sylven, 2020**	**Heath et al. 2016**	**Ozanne, 2016**	**Pieters, 2016; Baca, 2015**
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the qualitative methodology appropriate?	Yes	Yes	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	Yes	Yes	Yes	Yes	Yes
Was the recruitment strategy appropriate to the aims of the research?	No[1]	No[1]	No[1]	No[1]	No[1]
Was data collected in a way that addressed the research issue	Yes	Yes	Yes	Yes	Yes
Was the relationship between researcher and participants adequately considered?	Can't tell	Can't tell	Can't tell	Yes[2]	Can't tell
Were ethical issues taken into consideration?	Yes[3]	Yes[3]	Yes[3]	Yes[3]	Can't tell
Was there sufficient rigor?	Yes	Yes	Yes	Yes	Yes
Is there a clear statement of findings?	Yes	Yes	Yes	Yes	Yes
How valuable is the research?[4]	Yes	Yes	Yes	Yes	Yes
Overall risk to rigor	Low	Low	Low	Low	Low

Abbreviations: CASP: Critical Appraisal Skills Programme

[1] Participants in these studies were recruited after their child underwent surgery, raising concerns about selection bias due to selecting parents who were a) referred to surgery, b) proceeded with surgery, and c) experience some level of success with surgery. Excludes parents of a child with drug-resistant surgery who were not referred to surgery, who choose not to proceed with surgery, or whose child did not achieve desired outcomes.

[2] Interviewers and researchers not part of care/surgery team

[3] Approved by university ethics committee

[4] "Yes" rating given when study: a) discussed contribution to existing knowledge or understanding, b) identified new areas where research is necessary, and c) discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

APPENDIX E. Coding extracts for treatment decisions for childhood epilepsy into the Theoretical Domains Framework

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Knowledge	Baca, 2015, 25894906	With the recognition that "something is wrong," parents sought information from multiple sources in an attempt to understand seizures and epilepsy and become better advocates for their child.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Parents reported difficulties as the they sought information about seizures, epilepsy, medications, and surgery….Parents sought information from a variety of disparate sources including physicians, the Internet, books, and family/friends.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Baca, 2015, 25894906	Through this journey, parents learned to navigate a complicated medical system filled with doctors of different specialties, multiple hospitals, insurance approvals, and numerous treatment options	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Pieters, 2016, 27521720	Parents also had to learn about the intricacies of the healthcare system including the types of doctors that were needed to care for their children	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Parents perceived the information they received from professionals as inadequate to make an informed decision. They therefore sought out additional information regarding the surgical procedure and outcomes. Aiming to comprehend what surgery would entail, parents requested information in lay language and searched the internet for information and videos.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	"I googled it until I couldn't google anymore and I think that gave me a better understanding of what was going on."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	Finally, parents emphasized the importance of receiving adequate information regarding epilepsy surgery, including information presented in a variety of formats (e.g., booklets, photographs, videos). They also suggested that a 'frequently asked questions' information sheet would have been beneficial. "I know I got a little booklet about epilepsy surgery services, but it would have been nice for us to have some extra information… just some written up case studies of kids that have gone through, had the same, you know, operation."	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Heath, 2016, 27371883	These included …additional information provision	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	O'Brien, 2020, 33120326	[I]t was felt that a greater awareness of potential changes besides seizure control prior to surgery would have been helpful for children and their families in developing realistic expectations and preparing for post-surgical adjustment. They just tell you medical stuff about it, the recovery, not that it'll be different…They just tell you about having no seizures […] They don't tell you what it's actually going to be like.	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Sometimes, parents received more information from other parents, than from the physician. They were disappointed if they felt that information was withheld	Need for information about epilepsy, treatment options, and navigating the healthcare system
Knowledge	Ozanne, 2016, 27176878	Parents thought that it was important to receive the information that surgery was aimed to treat the epilepsy and not other disabilities, such as behavioral problems, in order to have realistic expectations. They emphasized the importance of being informed about both positive and negative outcomes and risks. Even if surgery was a medical success, the family's life situation was	Need for information about epilepsy, treatment options, and navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
		sometimes characterized by sorrow and problems. The deterioration, expected or unexpected, of e.g., motor function, speech, visual field, or attention made life more difficult and burdensome.	
Knowledge	Baca, 2015, 25894906	Parents also reported little initial familiarity with epilepsy and that they felt overwhelmed trying to learn a new language of epilepsy because of the "avalanche of information coming at us."	Overwhelmed by information
Knowledge	Pieters, 2016, 27521720	She described the hard and time-consuming struggle of trying to synthesize information on her own "because there really wasn't a lot out there and when you're hearing different things from different specialists it's very difficult for parents to make heads or tails of it". She spent "six months combing through the literature reading everybody's publications from places all over the world … because I wasn't getting a straight answer … my husband was saying to me you know are you going to go and get your degree in neuroscience after this … but you know it's your child, you want to do the best by them and you know you want them to be able to achieve their potential".	Overwhelmed by information
Knowledge	Baca, 2015, 25894906	As they acquired more epilepsy knowledge, however, many parents felt more empowered and able to ask questions and participate more effectively in the care of their child	Knowledge is empowering
Knowledge	Baca, 2015, 25894906	A mother who stated, "I just think knowledge is power and it also brings some comfort to making a good decision," exemplified the sense of empowerment associated with gaining fluency in the language of epilepsy	Knowledge is empowering
Knowledge	Heath, 2016, 27371883	Increasing their own knowledge about medically intractable epilepsy and the available treatment options empowered parents to participate more fully in the decision-making process. "I said to him 'don't tell me in a doctor's term, tell it me in a parent's term. I want to know the ins and outs; I want to know exactly what the risks are but in terms that I understand'"	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	[P]arents took on the responsibility for learning as exemplified by the parent who naturally 'corrected' her language to imply her understanding of the different terms that clinicians use, "(the seizures) were not controlled … or intractable as you guys call them."	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	Being open to learn and learning itself occurred while parents became epilepsy experts in order to be effective advocates for their children	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	In order to have productive information exchanges with clinicians and engage in shared decision-making, parents realized that they had to learn about a very unfamiliar and unforeseen situation	Knowledge is empowering
Knowledge	Pieters, 2016, 27521720	The lack of understandable information and the experience of information imbalance between the parent and the physician were overwhelming as experienced by a father who described him and his wife as "pretty pragmatic." He said, "The beginning was terrible, because you're of course dealing with … specialists in things and that you've pretty much never even heard of …. It was tough in the beginning, really tough … (the information that we found) was written by doctors for doctors. It's kinda tough when you don't know what you're talking about to get through that.	Knowledge is empowering
Social role and identity	Heath, 2016, 27371883	However, they also reported a sense of duty to be "strong" (P1) and "brave" (P3, 4) for their children, wishing to protect them from their feelings of anxiety.	Role and responsibility as a parent
Social role and identity	Heath, 2016, 27371883	Parents discussed feeling pressure from a need to 'do the right thing' by their child; that is, choosing the treatment option that would give their child the best chance of reaching their full potential	Role and responsibility as a parent

TDF domain	Author, Year, PMID	Extracted text^{A}	Theme
Emotion	Baca, 2015, 25894906	Parents reported personal and family stress throughout the journey. Caring for children with a chronic, severe, and paroxysmal neurologic condition meant being "on call" all the time; they had to be prepared to respond to an emergency at all times	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Participant descriptions highlighted the significant impact that epilepsy had upon on the child's and family's lives prior to epilepsy surgery. Worries about seizures and the associated risks led to constant fatigue for some: "you're tired yourself before you even go out the door" (Charlie – mother). In some cases, experiences of epilepsy impacted on their lives to such a degree that parents referred to children as "not having a life" (Charlie - mother)	Drained from managing epilepsy
Emotion	O'Brien, 2020, 33120326	Child mood and behavioural difficulties-stress, secrecy and shame. Before surgery children's mood and behavioura difficulties were a common, causing fear and stress which negatively impacted upon family relationships: "If there was something there, she would get it, she would want to attack you with it". (Jessica – mother) Many described how increased supervision led to a change in their parenting role, negatively impacting on the relationship they or their other children had with their child: "I was just his carer really and didn't have a relationship with him" (Jack - mother)	Drained from managing epilepsy
Emotion	Baca, 2015, 25894906	Parents frequently described that medication trials continued often with little to no mention of surgery. One mother told of her frustrations with multiple medications, "It was, 'Let's work on all the different medical aspects or medicines before going to surgery or even talking about surgery."	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Parents felt desperate not only to find a treatment option that would work, but also to persevere and actively do something that was productive and necessary to help their child	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Wanted to play an active role	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[W]anted to use time constructively	Desperate to help their child
Emotion	Pieters, 2016, 27521720	Another mother remembered that she felt "desperate" to take her adolescent son to another doctor, but she needed to wait for referrals because of insurance	Desperate to help their child
Emotion	Pieters, 2016, 27521720	[P]arents felt frustrated and desperate to get help for their child.	Desperate to help their child
Emotion	Heath, 2016, 27371883	Following indication of surgery as a potential treatment, parents and families faced what they described as a "difficult decision" (P5) which was experienced as "daunting", "frightening", "scary", and "stressful"	Difficult and stressful decision
Emotion	Pieters, 2016, 27521720	"It was not only in terms of time, it was mentally draining as well. It took a lot of time and it took a lot of concentrated thought to figure this out."	Difficult and stressful decision
Emotion	Ozanne, 2016, 27176878	Parents were afraid and worried	Fear (unspecified)

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Emotion	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options. This perception was influenced by parental fear regarding the irreversibility of surgery, as well as by professionals who had informed them that surgery was the only remaining treatment for controlling their child's seizures	Fear of surgery
Emotion	Pieters, 2016, 27521720	For the majority of parents, brain surgery was a frightening possibility as described by the mother who said, "At first I was horrified at the thought of this innocent – having her brain opened and operated on. It just seemed so barbaric." Another mother's statement reflected upon her initial feelings of fear and avoidance when she said that she and her husband, "just couldn't deal with (having our daughter go through brain surgery)	Fear of surgery
Emotion	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience "a change in personality" as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Fear of surgery
Emotion	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Fear of surgery
Emotion	Sylven, 2020, 32480305	Many parents were worried that their child's epilepsy would be "worse" after the surgery, and as a result, the operation would have been "in vain". This included a risk that seizure frequency might increase, that seizures might be longer or be of a different character, or that seizure frequency might not improve.	Fear of surgery
Emotion	Ozanne, 2016, 27176878	It was demanding not to know if surgery would be possible, and the parents were thankful when the decision was finally made	Relief after decision was made
Emotion	Sylven, 2020, 32480305	Many parents described a general hope for improved behavior and emotional functioning for their child. Behavior encompassed a wide variety of outcomes including that the child would be "calmer" with fewer behavioral outbursts, have better attention and concentration, and "feel better"	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents hoped that the surgery would lead to better health and quality of life for their child	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Parents often expressed a desire that surgery would result in their child being "more alert", less "tired", and "have more energy". They also mentioned that they hoped that surgery would result in better sleep quality	Hope for better health and quality of life
Emotion	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including "tiredness" would lead to a "normal life" for their child	Hope for better health and quality of life
Emotion	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they "hoped" for candidacy	Hope for surgical candidacy
Emotion	Heath, 2016, 27371883	When a decision to accept surgery had been made, parents hoped for the procedure to be carried out as soon as possible. Waiting for a date for surgery was described as an anxious time for parents, who experienced relief when a date was set.	Hope for surgical candidacy

TDF domain	**Author, Year, PMID**	**Extracted text**^{A}	**Theme**
Social influence	O'Brien, 2020, 33120326	This sense of exclusion impacted on the family as a whole, with siblings being unable to attend clubs or invite friends to their house, and parents feeling that they "couldn't join in with what other families were doing" (Lucy – mother). A common theme was parental concern that siblings had less time or attention: as "the focus was on him [child with epilepsy] a lot of the time" (Andrew – mother).	Social exclusion
Social influence	O'Brien, 2020, 33120326	Difficulties appeared to be exacerbated by a perceived a lack of understanding from others, which appeared to be tied to a notion of epilepsy being an invisible condition, as seizures commonly occurred behind closed doors: "At the beginning, it was just generally at home, they didn't understand, they didn't get it. (Jessica – mother). Most participants described finding it difficult to talk to others about epilepsy, with several reporting that they limited the information they shared, or withholding the diagnosis altogether. "Gosh, we didn't tell anybody. We were ashamed". (Jack – mother)	Social exclusion
Social influence	Heath, 2016, 27371883	In addition, parents emphasized the importance of accessing the experience and expertise of other parents who had followed a similar treatment pathway. Such peer support was suggested to facilitate understanding of the surgical procedure from a 'parental perspective', as well as provide emotional support. "If people could freely put their experiences on either a forum or even if you get a group, you know, parents talking to other parents who have gone through the experience to get a real idea of what's happening."	External support from peers and healthcare professionals
Social influences	Heath, 2016, 27371883	These included … peer support	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Those parents who were able to connect with other parents and families of children with epilepsy found it to be very helpful to be able to communicate with peers with seizures unresponsive to medication who were on the same journey.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	[E]xchanging shared experiences with peers gave direction to decision-making because it helped with processing complex factual and emotional information	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	Three key facilitators were identified as follows: (a) having a champion, (b) finding a good team of epilepsy providers, and (c) connecting with others who shared the experience of epilepsy. (Fig. 1; Table 2). Such facilitators played a key role during the times when parents especially needed support to act purposefully and move out of vulnerable periods.	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	However, using time constructively happened after the parents described finding a champion that advocated for them and helped them navigate their journey. It was a champion, whether a doctor, nurse, or social worker, that facilitated steady and purposeful progress over more manageable portions of the journey	External support from peers and healthcare professionals
Social influence	Pieters, 2016, 27521720	One mother favorably described one such champion, a nurse coordinator, "she's like wonderful for us. She's been our lifesaver." These champions helped parents get referrals and seek different treatment options or were the persons that believed them and pushed them to seek better or additional care. For one mother, her daughter's pediatrician acted as her champion. She said, "He encouraged me and gave me the confidence that if I wasn't happy with what five other pediatric neurologists were saying that it was okay to go and you know continue to seek out other opinions."	External support from peers and healthcare professionals

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Social influence	Heath, 2016, 27371883	Family decisions. Parents described decision-making as a family choice involving partners, siblings, grandparents, and the child (where developmentally appropriate). Discussing treatment options with family members was generally considered to be helpful. "I remember my mother-in-law was in Pakistan and I called her up and I was very teary…I felt like I had been punched in the stomach to be honest 'cause, you know, brain surgery is not something you take lightly and she said 'well if it's for his betterment and to improve his life then we will do whatever it takes'."	Family decision
Social influence	Heath, 2016, 27371883	For some families, decision-making regarding surgery gave rise to conflicts. For example, one mother described herself as more agreeable to surgery than the child's father because, as the primary carer, she 'witnessed' the true extent of their child's seizures. In cases where the child's epilepsy was considered to impact on the whole family (e.g., where a parent could not return to work because of a need to look after the child), then, the decision was understood as a responsibility of the whole family	Family decision
Belief about consequences	Baca, 2015, 25894906	On their journey, many parents transformed their view of surgery from a scary, last resort treatment to a necessary and hopeful option. One mother explained, "By the time she got it—it got so serious I was looking toward that as being an answer to our prayers rather than just something that I couldn't even think about." This transformation occurred as parents became increasingly aware of the severity of their child's epilepsy and its impact on their child's and their future life. Although surgery remained scary, epilepsy was or became scarier than surgery. As some parents feared that their child would die from epilepsy, surgery was no longer perceived as an elective treatment.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	Many parents initially perceived epilepsy brain surgery to be "pretty horrific" and a treatment of "last resort."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Baca, 2015, 25894906	After an arduous journey, parents reached the right doctor and center; epilepsy surgery, although previously seen as a last ditch option, became a viable option and a source of hope.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	After persevering through hard times with unsuccessful medications and interventions, they eventually decided to proceed with surgery, the option that they perceived to be the best treatment for the epilepsy.	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Pieters, 2016, 27521720	Because surgery was viewed as a treatment of last resort for many parents, time was required to process the initial fear, make sense of the emotional information, and consider the factual risks and benefits in order to decide on surgery as the best option for their child. Reflecting on why it took longer than six years to get to surgery for her 9-year-old daughter, a mother said, "Of course you need to take a lot of time to think about it. It's very invasive…. I did as much research as I could about it, and talked to the doctor a lot about it, and I decided that that was her best chance of having a normal life in the future."	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Pieters, 2016, 27521720	Through internal processing, parents worked through the diverse emotions and large amount of information that they were receiving and gathering to move forward and thereby develop a greater understanding of their child's illness and prognosis in order to ultimately reach the point where surgery became the treatment of choice	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Parents were clear that putting their child forward for epilepsy surgery was the best and right decision for their child, demonstrating a high level of satisfaction with the treatment outcome. However, initial perceptions of surgery were often negative, particularly if it was not a treatment option that had previously been considered or that professionals had indicated. Many parents described attending their child's clinic appointment expecting to talk about more conventional treatments (e.g., medication or diet), leading to them feeling surprised when surgery was raised for discussion. "We went originally to talk about the ketogenic diet and then the doctor said 'oh I think surgery's your best option' which came as a bit of a shock really because no one had ever mentioned the surgery before." (P5) "It actually came as a bit of a surprise to us that option, in that it was mentioned in a review visit, that maybe the time had come to look at the option of surgery but we weren't really aware of that option at that point so it came as quite a shock to us." (P6) While some parents took comfort from the possibility of an additional treatment pathway, others felt excluded from the decision to assess their child for surgical candidacy, leading to their feeling unable to participate in an informed discussion	Transformation towards surgery as the treatment choice informed by experiences, emotions, and knowledge
Beliefs about consequences	Heath, 2016, 27371883	Some parents were unconvinced about the efficacy of surgery for reducing their child's seizures. Skepticism stemmed from their experience of other pharmacological and dietary treatments which had resulted in little improvement in seizure control. In weighing up treatment options, parents also discussed the side effects of alternative treatments. For example, medications were perceived to have a range of adverse side effects, whereas ketogenic diets were viewed as negatively affecting the child's ability to live a 'normal' life. "We thought about the effects of the medication if we chose to continue on the path of finding a different medication… but then how long would that have taken us? Like how long would it work for? Then it would develop, then how long 'til we have to find something else, with all the side-effects that can happen? I didn't want to put her through that as well." (P9)	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	Balancing potential costs and benefits. When deliberating surgery, parents balanced their perceived necessity of the treatment in terms of how epilepsy currently affected and would, in the future, affect their child's quality of life, against concerns regarding the possibility of negative outcomes. In other words, parents weighed up the potential benefits of surgery such as freedom from seizures against what they understood as the risks, including cognitive impairments. "There was no quality of life for him; we had to do what was best. We were told of the risks of the surgery and we were told that actually this would be the best thing for him… we don't regret it, but it was, it's not an easy decision to take because you're worried about the risks of things."	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned that while they would like seizure freedom, any reduction in seizures would be "welcome" and a clear improvement for their child and for the family in general.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents mentioned complete freedom from medicine while others mentioned a reduction in AEDs. Parents believed a reduction in AEDs and associated adverse side effects of AEDs including "tiredness" would lead to a "normal life" for their child.	Balancing potential benefits of surgery with concerns of adverse effects

TDF domain	Author, Year, PMID	Extracted text^{A}	Theme
Beliefs about consequences	Sylven, 2020, 32480305	Many parents mentioned a desire for seizure freedom for their child after surgery. They wanted their child to be free from the “trouble” of seizures and everything that follows from seizures	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	They wanted that their child could do everything their friends could do like “play football, cycle, and swim independently”. Some parents mentioned that they wanted their child to be able to go back to school and have a “proper social life” while some parents referenced the future hoping that the child would be better placed to get a job and driving license as a result of surgery.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Parents mentioned the possibility that their child would experience “a change in personality” as a result of the surgery. They did not mention particular personality characteristics but feared that the child's personality would change for the worst	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	Some parents reported that they were concerned that surgery would lead to a loss of function including that language/speech would be negatively affected. This concern included a worry that the child would develop speech difficulties or that they would lose the ability to talk completely as a result of the surgery. Parents also mentioned concerns that their child would acquire motor difficulties or even be paralyzed as a result of the surgery. Parents were also worried that any loss of function would be irreversible after the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The majority of parents mentioned concerns about possible complications that could occur during or soon after the surgical procedure	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	A small number of parents said that they had no concerns before the surgery. One parent said that they were not worried as “no concerns arose in the discussions with professionals” before the surgery	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Sylven, 2020, 32480305	The results of the current study suggest that parents frequently worry about that the child would lose skills or function, or complications would arise during surgery that could result in injury.	Balancing potential benefits of surgery with concerns of adverse effects
Beliefs about consequences	Heath, 2016, 27371883	For all parents in this study, the perceived need of surgery outweighed the possible risks and side effects. “We thought this is going to be no life for him… He was on this diet which meant he couldn't go out with friends; he couldn't stay at friends' houses, all the things that other kids do, he couldn't do… and then he got these fits virtually every night and we thought… you know what, there is no decision to make, we gotta do it… We knew there were risks, we talked about them with [child] and he was frightened but he said no, we gotta do it.” (P3)	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	Last treatment of choice. For most parents, surgery was a last-resort treatment option that would only be considered following the exhaustion of all other options…Some parents felt that they had little alternative but to accept surgical intervention in order to provide their child with the best possible chance of improving their quality of life. “We felt it was our only option really in her having a more independent future.”	Only choice left
Beliefs about consequences	Heath, 2016, 27371883	“My husband and I walked out of the appointment saying we feel like we don't have a choice… we felt like it was the only choice we could make.”	Only choice left

TDF domain	Author, Year, PMID	Extracted text[A]	Theme
Beliefs about consequences	Heath, 2016, 27371883	For one family with an adolescent child, it was considered necessary to accept surgery while the child received care from the children's hospital as they believed that the treatment would become unavailable once transitioned to adult services.	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Parents described a process of weighing up the pros and cons based on the information provided by the medical team, which led to the decision to pursue surgery. Many perceived that surgery was the only hope "a simple decision" (Alex – mother), they were "in no doubt" (Lucy – mother) and that they "had no choice" (Jessica mother) as the health and wellbeing of their child would deteriorate or become more serious if surgery was not pursued. There was also a sense of 'not knowing' outcomes if surgery was not pursued. We felt we've got to do this because where is she going to end up if we don't?	Only choice left
Beliefs about consequences	O'Brien, 2020, 33120326	Decision making – is there a choice? When asked what informed the decision-making process when considering the option of surgery as a treatment for epilepsy, parents discussed how the perceived risks to their child's safety, as well as the safety of others, were key factors. Even with all the risks, it wasn't going to be much worse than having a little girl that was unconscious all the time anyway.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Parents felt that epilepsy surgery was a question of life or death, of chaos or control. It was terrifying to see the child so ill. The epilepsy was uncontrolled, and drugs gave side effects. Parents felt that there was no alternative to surgery. However, it was a difficult decision since they did not know the outcome.	Only choice left
Beliefs about consequences	Ozanne, 2016, 27176878	Information about possible complications related to surgery did not prevent the parents from wishing to proceed with the operation. They preferred to take a risk rather than live in constant fear	Only choice left
Environmental context and resources	Baca, 2015, 25894906	The journey to finding the right doctor was further complicated by parental perceptions that doctors sometimes lacked knowledge of or familiarity with epilepsy. Many parents went to numerous doctors searching for the "right" doctor, or one who had a strong epilepsy knowledge base, could effectively identify the problem, and then make a clear plan of action	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents expressed that their children were seen, evaluated, and cared for by a variety of doctors of different specialties throughout their journey. Parents frequently described difficulty finding the "right" doctor(s). Difficulty was associated with a lack of pediatric neurologists in their local area and with different doctors having different recommendations.	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Parents depicted a pathway filled with multiple doctors and treatments, insurance battles, and work and life obstacles as they sought a cure for their child's seizures, becoming ever more knowledgeable about epilepsy	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	Further, finding the best clinicians for the needs of their child required an understanding of the relationship between different medical (sub)-specialties and thereby represented dual and concurrent use of processing and navigating mechanisms	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	The second mechanism, navigating, was when the parents kept and directed their way along their journey often through uncharted territory as they steered through various aspects of the health-care system and learned about epilepsy itself and its various treatments.	Challenges with navigating the healthcare system

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Environmental context and resources	Pieters, 2016, 27521720	A mother described her disappointment with being denied by insurance despite doing all the work that was required	Challenges with navigating the healthcare system
Environmental context and resources	Pieters, 2016, 27521720	"We'd already been fighting her seizures for two and a half, approaching three years, and because we were new there and per protocol, they wanted to try these other treatments for another year."	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	The Swedish Social Insurance Agency, technical aids center, school, and municipality were criticized by the parents. Based on their experience, they felt that the bureaucracy had been problematic and had taken a great deal of time. They had to fight for their rights, despite physicians' certificates. Without specific diagnoses of comorbidities, it was even more difficult to get adequate support. They thought that it would be helpful if the authorities understood that parents only asked for help when they had reached their limit, and then, urgent help was necessary	Challenges with navigating the healthcare system
Environmental context and resources	Ozanne, 2016, 27176878	Parents were disappointed if it took a long time to get a correct diagnosis	Challenges with navigating the healthcare system
Environmental context and resources	Baca, 2015, 25894906	Several parents experienced feeling doubted by a doctor; some were told that nothing was wrong with their child initially	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	As parents learned that surgery was a potential viable option for their child, they "hoped" for candidacy. This hoping required persistence and sometimes disregarding doctors who initially felt surgery was not an option. Looking back, one father voiced happiness that he listened to his parental instincts rather than the opinion of one doctor. He (doctor) said, "'I don't believe that she is a surgical candidate.' As a parent, I listened and I wanted to think otherwise.... In hindsight, I'm glad I didn't listen to him."	Interactions with providers (barriers)
Environmental context and resources	Baca, 2015, 25894906	Parents also reported notable physician-based barriers including variability in perceived knowledge of epilepsy, criteria for presurgical referral and appropriateness of epilepsy surgery	Interactions with providers (barriers)
Environmental context and resources	Pieters, 2016, 27521720	[C]rossing such an information divide was an uphill battle as stated by a mother who described feeling dismissed by doctors until she found someone who was approachable and was "gracious enough to talk to a mere mortal mother on the phone … because a lot of docs won't, they've got lots of protective armor around them shielding off potential patients or worse yet parents."	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	Parents perceived a lack of control over the decision-making process through their limited involvement in initial discussions regarding surgical candidacy	Interactions with providers (barriers)
Environmental context and resources	Heath, 2016, 27371883	These included earlier and softer introduction of surgery as a possible treatment option	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	During the long hospitalizations, some parents felt that nobody had time for them. Arrogance of some professionals made them very disappointed	Interactions with providers (barriers)

TDF domain	Author, Year, PMID	Extracted text^A	Theme
Environmental context and resources	Ozanne, 2016, 27176878	If the pediatric neurologists revealed their own worries and insecurities regarding epilepsy surgery, this could have negative effects	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	Parents described that support and information were inadequate before the epilepsy was recognized to be drug-resistant, especially from the local hospital. Their concerns were not taken seriously	Interactions with providers (barriers)
Environmental context and resources	Ozanne, 2016, 27176878	The support and information went from poor to strong when specialists at the university hospitals were involved	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	The pediatric neurologist's worry and sympathy for the family could also strengthen them since they felt invited and could be insecure together	Interactions with providers (facilitators)
Environmental context and resources	Ozanne, 2016, 27176878	Trust and safety characterized the relations to the pediatric neurologists and specialist nurses. It strengthened parents' trust to see that the pediatric neurologist fought for the child and that the parents' opinions counted in the decision process. Parents appreciated continuous contact.	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	Trust in the information that they received was greater when their child was in the hands of a team with different expertise, as exemplified by the mother who said, "They get all the doctors together to look at all the information … that felt a lot more helpful because if we were at a different hospital and you only had the one doctor, then they would have misdiagnosed him."	Interactions with providers (facilitators)
Environmental context and resources	Pieters, 2016, 27521720	When parents finally reached a comprehensive center to seek evaluation for potential pediatric epilepsy surgery, they recognized the importance of having a medical team of experts	Interactions with providers (facilitators)

[A] Study participant (first order statement) or study author (second order statement). We kept original spelling from primary study.

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Title page
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Background, pg 3
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Pg 3
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Pg 3-4
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Pg 3, Appendix A
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Appendix A
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Pg 4-5
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	Pg 4-5
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Pg 4-5
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 4-5
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	NA
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Pg 4-5, Table 1
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Pg 4-5
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Pg 4-5
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Pg 4-5
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	Pg 4-5
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	NA
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Pg 4-5< Appendix D

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	Pg. 4-5, Table 2
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Pg 5
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Pg 5
Study characteristics	17	Cite each included study and present its characteristics.	Pg 5, Table 1
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Appendix D
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Table 2
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Pg 5
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Pg 6-8
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	Pg 6-8
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	NA
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	NA
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	Pg 6-8, Table 2
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Pg 9
	23b	Discuss any limitations of the evidence included in the review.	Pg 10
	23c	Discuss any limitations of the review processes used.	Pg 10
	23d	Discuss implications of the results for practice, policy, and future research.	Pg 9-10
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Not registered
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	NA
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	NA
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Pg 11
Competing interests	26	Declare any competing interests of review authors.	Pg 11
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Pg 11

PRISMA 2020 Checklist

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

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