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Discussing sudden unexpected death in epilepsy with children and young people with epilepsy and their parents/carers: a mixed methods systematic review.

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Title Page

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Discussing sudden unexpected death in epilepsy with children and young people with epilepsy and their parents/carers: a mixed methods systematic review

Author names & affiliations

Kay Cooper[1,2], Pamela Kirkpatrick[1,3], Celia Brand[4], Alix Rolfe[5], Sarah Florida-James[6]

1. The Scottish Centre for Evidence-based, Multi-professional Practice: A Joanna Briggs Institute Centre of Excellence, Robert Gordon University, Garthdee Road, Aberdeen, UK
2. School of Health Sciences, Robert Gordon University, Garthdee Road, Aberdeen, UK k.cooper@rgu.ac.uk (corresponding author)
3. School of Nursing & Midwifery, Robert Gordon University, Garthdee Road, Aberdeen, UK p.kirkpatrick@rgu.ac.uk
4. NHS Lothian, Royal Hospital for Sick Children, Edinburgh UK Celia.Brand@nhslothian.scot.nhs.uk
5. University of Edinburgh, west Richmond Street, Edinburgh, UK Alix.Rolfe@ed.ac.uk
6. Scottish Intercollegiate Guidleines Network, Gyle Square, Edinburgh, UK sarah.florida-james@nhs.net

Corresponding Author

Kay Cooper
School of Health Sciences, Robert Gordon University, Garthdee Road, Aberdeen, AB10 7QG k.cooper@rgu.ac.uk

Abstract

Purpose: To synthesise the quantitative and qualitative evidence on the views and experiences of children and young people with epilepsy (CYPwE), their family members/caregivers and healthcare professionals on conversations between healthcare professionals and CYPwE/caregivers about the possibility of sudden unexplained death in epilepsy (SUDEP).

Methods: Mixed methods systematic review in accordance with Joanna Briggs Institute methodology, PRISMA guidelines and guided by an a-priori protocol.

Results: 656 potentially relevant studies were identified, 11 of which fulfilled the inclusion criteria for the review: 6 quantitative studies, 4 qualitative studies and 1 opinion/text article. Data synthesis resulted in the following 2 integrated findings: (i) Caregivers, and where appropriate CYPwE, should be provided with information on SUDEP and how it relates to them; (ii) Information on SUDEP should be delivered face-to-face, with supporting written information, by a suitably knowledgeable healthcare professional whom the caregiver/CYPwE feels comfortable with, at an appropriate time at or close to diagnosis.

Conclusion: This review confirms that healthcare professionals should discus SUDEP with CYPwE and/or their caregivers at or around the time of diagnosis and that the discussion should include prevalence of SUDEP, risk factors and risk reduction methods relative to the individual concerned. Apart from delivering SUDEP information face-to-face, with written or online information provided to reinforce messages, there is a lack of evidence on "how" to impart this sensitive information. Further research exploring the most acceptable and effective methods of discussing SUDEP with CYPwE and their caregivers is therefore indicated.

Keywords

Epilepsy, Sudden Death, SUDEP, Systematic Review

Introduction

Sudden unexplained death in epilepsy (SUDEP) is commonly defined as "sudden, unexpected, witnessed or unwitnessed, nontraumatic and nondrowning death in patients with epilepsy, with or without evidence for a seizure and excluding documented status epilepticus, in which post-mortem examination does not reveal a toxicologic or anatomic cause for death" [1]. Due to difficulties with definitive diagnosis it is increasingly classified as definite, probable or possible [1]. Epilepsy is a common neurological disease in children, and whilst SUDEP is considered a relatively rare event, it has a devastating effect on families and carers and it therefore remains an important topic for research [2]. The incidence of SUDEP in children (under 16 years) was previously considered to be around five times less than the incidence in adults, with reported incidence rates of between 1.1 and 3.4 per 10,000 person-years [3,4]. However, recent research conducted in Sweden [5] and Canada [6] has found similar rates of SUDEP in children and adults, reporting an incidence of 1.11 and 1.17 per 1,000 person-years respectively, suggesting that the risk of SUDEP in children may have previously been underestimated.

SUDEP risk is difficult to predict for individual children with epilepsy, but a number of risk factors have been reported that can support discussion of SUDEP with children and young people with epilepsy (CYPwE) and their caregivers to help them recognize, and where possible, reduce their risk [2]. These risk factors are categorised as aetiology-related, environmental, seizure-related, treatment-related, and other (change in physical health status), with uncontrolled seizures being the prime risk factor for SUDEP in children [2].

Discussing SUDEP risk is an emotive subject. It has been reported that patients and families want to be told what SUDEP is, and the risk factors and risk-reduction measures that can be taken [7-8]. However, there does not appear to be consensus from healthcare professionals on whether all patients, or selected patients (e.g. those with uncontrolled seizures only), should be informed about the risk of SUDEP [9-10], despite practice guidelines recommending that discussions should take place [11]. There is a growing body of research on the discussion of SUDEP with CYPwE. However, a search of Medline, CINAHL, the Cochrane Database of Systematic Reviews, the Joanna Briggs Institute (JBI) Database of Systematic Reviews and Implementation Reports, and PROSPERO failed to find a review (completed or in progress) that has synthesised the research to date.

It would be beneficial to practitioners and researchers for a systematic review to be conducted in order to make recommendations for practice, and to identify remaining gaps in the evidence-base. Previous systematic reviews have focussed on the incidence, causes/risk factors and public health burden of SUDEP (e.g. 12-14]. We identified one review in progress on the development of an "information-based patient information" to communicate risks about SUDEP [15] which, when completed, will complement the review reported here. We conducted a mixed methods systematic review to address the following question: What are CYPwE, family members/carers' and healthcare professionals' views and experiences of conversations between healthcare professionals and patients'/family members' about the possibility of SUDEP? The specific review questions were: 1. ***When*** should conversations about SUDEP in CYPwE take place? 2. ***Where*** should conversations about SUDEP in CYPwE take place? 3. ***Who*** should be involved in conversations about SUDEP in CYPwE? 4. ***What*** should be included in a conversation about SUDEP in CYPwE? The review was conducted according to Joanna Briggs Institute (JBI) methodology for mixed methods systematic reviews [16] and an a-priori protocol [17] and is reported according to PRISMA guidelines [18].

Methods

Inclusion criteria

The **Population** was CYPwE (any type/severity) aged up to 25 years, family members/carers of CYPwE, and health professionals caring for CYPwE in any setting. We did not limit inclusion to under 16's as people with epilepsy can stay in paediatric services beyond age 16 in many healthcare systems. Some studies included mixed populations; they were included in the review if the data relating to CYPwE or their parents/carers or healthcare professionals could be extracted separately or if they constituted a substantial (at least 30%) proportion of the sample. The **Phenomena of Interest** was conversations between healthcare professionals and patients/family members/carers about the possibility of SUDEP in CYPwE. The **Context** was anywhere that conversations about SUDEP might take place, including primary and secondary care and home settings. The **Types of Studies** considered for inclusion were: (i) Quantitative descriptive, cross-sectional (e.g. surveys) and observational studies, (ii) Qualitative studies using designs such as phenomenology, grounded theory, ethnography and action research, and (iii) narrative, opinion and text articles where they contained data relevant to the review questions.

Search strategy

The search strategy aimed to find both published and unpublished studies. A 3-step search strategy was employed. First, an initial limited search of MEDLINE and CINAHL was undertaken followed by analysis of the text words contained in the title and abstract and the index terms used to describe the articles. The search strategy, including all identified keywords and index terms was adapted for each included information source and a second search was undertaken The full search strategies are provided in Appendix 1. Finally, the reference lists of all studies selected for critical appraisal were screened for additional studies. Studies published in the English language from 2007 to December 2018 were included, and the following databases were searched: MEDLINE, CINAHL, EmBASE, AMED, PsycINFO, Psycharticles. The search for unpublished studies included Google Scholar, openSIGLE, Mednar, and the New York Academy of Medicine Library Gray Literature Report.

Study selection

All identified citations were collated and uploaded to Refworks and duplicates removed. Titles and abstracts were screened by two independent reviewers for assessment against the inclusion criteria for the review. Studies that met the inclusion criteria were retrieved in full and assessed in detail against the inclusion criteria by two independent reviewers. Full text studies that did not meet the inclusion criteria were excluded and reasons for exclusion are provided in Supplementary File 1. Any disagreements that arose between reviewers were resolved through discussion.

Assessment of methodological quality

Eligible studies were critically appraised by two independent reviewers for methodological quality using JBI critical appraisal tools for analytical cross-sectional, case-series and qualitative studies, and text & opinion[19]. See Supplementary File 2 for details of the items contained in each critical appraisal tool. Disagreements between the reviewers were resolved through discussion. In order to be comprehensive, no studies were excluded on the basis of methodological quality.

Data extraction

Quantitative and qualitative data were extracted from included studies by two independent reviewers using standardized JBI data extraction tools. The data extracted included specific details about the population, study methods, the phenomenon of interest, context and outcomes of relevance to the review questions. Specifically, quantitative data comprised of data-based outcomes of descriptive statistical tests which were then converted into ‘qualitized’ data [20]. Qualitative data comprised of themes or subthemes with corresponding illustrations, which were assigned a level of credibility [21]. Any disagreements that arose between the reviewers were resolved through discussion.

Data synthesis

The convergent integrated approach to synthesis according to the JBI methodology for mixed methods systematic reviews, based on previous work by Sandelowski et al [20] and Hong et al [22], was used in this review. This involved assembling the ‘qualitized’ data with the qualitative data. Assembled data were categorized based on similarity in meaning to produce a comprehensive set of synthesized findings in the form of a set of recommendations or conclusions.

Results

Study inclusion

The search identified 656 potentially relevant articles after removal of duplicates (Figure 1). After screening titles/abstracts and full-text articles, 6 quantitative studies, 4 qualitative studies and 1 opinion/text article were included in the review.

Methodological quality

Full details of methodological assessment can be found in Supplementary File 2. One study [9] was low quality, 5 were moderate [23-27] and 5 high quality [10, 28-31] according to cut-off scores determined by the review team a-priori.

Characteristics of included studies

The 11 studies originated from Europe [10,30], the United Kingdom [9,24,25,27], North America[23,26,29,31] and Malaysia[28] (Table 1). The quantitative studies employed surveys [9,23,24,28,10] or structured interviews [29] and the qualitative studies focus groups and interviews [25-27]with one study reporting data from an open survey question [30]. The narrative article reported the views of two bereaved fathers [31]. Studies explored the views of healthcare professionals' [10,23-24,30], CYPwE [25,27], family members of CYPwE [28], or bereaved families' [26,29,31], with one study [9] including both parents and healthcare professionals. Sample sizes ranged from 2 [31] to 161[10], with the qualitative studies generally having smaller samples. A total of 732 participants are represented in the review findings.

Findings of the review

Table 2 presents the review findings, which are discussed below. The qualitized data are assembled with the qualitative data (column 1). Categorisation based on similarity of meaning can be seen in column 2, and the contribution of each category to the final integrated findings (column 3). For the qualitative data, only findings rated as unequivocal or credible according to JBI methodology [21] were included in the synthesis; any studies rated as "not supported" were omitted.

Integrated finding 1: *Caregivers, and where appropriate CYPwE, should be provided with information on SUDEP and how it relates to them: The information on SUDEP should include prevalence, risk factors and risk reduction measures that can be taken.* Thirteen findings from 9 studies [9-10,24-27,29-31] formed the 2 categories that comprised integrated finding 1. This finding revealed that SUDEP information should be provided to caregivers, and where appropriate to CYPwE themselves, and that it should provide information on the prevalence and risk factors associated with SUDEP as well as personalised information on risk-reduction measures.

Category 1: Some discordance between carers'/family members' and healthcare professionals on discussing the risk of SUDEP in CYPwE

The findings relating to family members and CYPwE themselves [9,25-27,30-31] all highlighted the importance to them of being informed about SUDEP. The participants in these studies felt they had a right to know and that discussing SUDEP enhanced the trust between them and their healthcare providers [25,27,31]. Although SUDEP is difficult and emotive to discuss, family members described the burden of guilt they would feel if their child experienced SUDEP and they had not had the opportunity to implement risk-reduction measures [26]. There was however some discordance between these findings and those from the studies on healthcare professionals. Rates of discussing SUDEP with all or most patients were 8.7% [10], 20% [9], and 29% [24], with intractable seizures being the most common reason for choosing to discuss SUDEP [9,30]. Age 12 or 13 onwards was reported as the age at which CYPwE themselves were commonly included in discussions about SUDEP [9,33].

Category 2: SUDEP information should include prevalence, risk factors, and risk reduction

CYPwE and family members in the included studies felt that "basic information" [27] on SUDEP should be provided and should include prevalence, risk factors and preventive strategies, including the importance of drug adherence [27,29]. Some highlighted the importance of having an explanation of how the risk of SUDEP related to them personally [25], and some highlighted the need to balance the discussion of SUDEP risk with the information that it is a relatively rare event [26].

Integrated finding 2: *Information on SUDEP should be delivered face-to-face, with supporting written information, by a suitably knowledgeable healthcare professional whom the caregiver/CYPwE feels comfortable with, at an appropriate time at or close to diagnosis.* Twenty-five findings from 9 studies [9,23-28,30-31] formed the 3 categories (categories 3-5, Table 2) that comprised integrated finding 2. This finding identified that information on SUDEP should ideally be delivered face-to-face, with appropriate written information to reinforce the discussion, and that the discussion should happen at or close to the time of a diagnosis of epilepsy being made. It also identified that the healthcare professional providing the information on SUDEP should be knowledgeable and have good rapport with the caregiver and/or CYPwE.

Category 3: Consideration of the timing and who should be present during SUDEP discussions is important.

CYPwE reported not wanting to be "in the dark" about their risk of SUDEP, whilst recognising that the time needed to be chosen carefully [27]. CYPwE also reported that their healthcare professional should be able to determine whether they were ready for such a discussion to take place, and that if necessary the information could be delivered "in chunks" [25]. Parents and guardians [28] wanted themselves and their CYPwE to be told about SUDEP at the time of diagnosis. Bereaved parents agreed with this view and generally felt that the information should be provided routinely at the time of a diagnosis of epilepsy being made [9,26]. However, in surveys of UK and Malaysian parents of children with epilepsy 16% [9] and 18% [28] respectively reported that SUDEP should be discussed when seizures became difficult to control, and 3% did not want to know at all [9]. There was also recognition by medical doctors that any meeting to discuss SUDEP should be given sufficient time and not be rushed [30].

Regarding who should be present, the findings revealed that an epilepsy specialist should provide information on SUDEP. Definition of specialist varied across the studies with pediatric neurologist [26], "doctor" [31], and nurse [25] being named in some studies, while others just referred to "specialists" [23-24]. There was recognition in in one US survey [24] that primary care providers lacked the relevant knowledge for discussing SUDEP, with 79% citing lack of knowledge as the reason they did not discuss SUDEP, stating that they expected the neurologist to conduct this role. Some of the findings related to the inclusion of CYPwE themselves in discussions about SUDEP. Parents generally felt it was their role to decide whether their children should be present or when to inform them about SUDEP [9,26,28]. However, 31% of parents in one study [9] felt the healthcare professional should inform their child.

Category 4: SUDEP discussions should be face-to-face wherever possible and followed-up with written information.

Face-to-face discussion of SUDEP information was identified as the most suitable method [9,24-28]. In one study [24], 91% of UK clinical nurse specialists sampled reported that home visits or telephone were suitable methods. However, as these methods were reported together it is not known how many felt that SUDEP information should be provided over the telephone. Canadian parents who had lost children to SUDEP [26] were clear that such information should not be provided over the telephone. Written information was seen as important to reinforce face-to-face counselling [9,25-26] but not to replace it [25], and could be provided by an educational software programme or leaflet [28].

Category 5: The person discussing SUDEP should be knowledgeable and have a good rapport with family members/carers/CYPwE, taking their emotions into account.

Two of the included qualitative studies' findings contributed to this category. "Medical doctors" [30] described the role that emotions played when discussing SUDEP, explaining that they can be useful or can impede the discussion. CYPwE discussed the importance of discussing SUDEP with someone whom they felt knew them and had time for them [27], in this case suggesting that the epilepsy nurse was more appropriate than the medical doctor (UK context).

Discussion

This mixed methods systematic review aimed to address the "when, where, who and what" regarding discussion of SUDEP with CYPwE and their caregivers. The review findings can be summarised as follows. SUDEP information should be provided: (i) at or close to the time of a diagnosis of epilepsy being made; (ii) face-to-face in the clinical or home setting; (iii) by a specialist known to the caregiver/CYPwE, with or without the CYPwE being present, and (iv) in the form of prevalence, risk factors and risk reduction measures.

Despite the integrated finding recommending that SUDEP information should be provided at or close to the time of diagnosis, there was some discordance between the caregivers and CYPwE who wanted information, and the healthcare providers who did not routinely provide SUDEP information to all caregivers or CYPwE. The routine provision of SUDEP information, in adults as well as CYPwE, has been the topic of some debate in the literature for a number of years. A fatal accident inquiry in Scotland ruled in 2011 that "the vast majority of patients with epilepsy, or their parents or carers where appropriate, should be advised of the risk of SUDEP on first diagnosis…"[32]. The same recommendation was subsequently made in practice guidelines [11]. However, several studies have since reported that healthcare professionals can be reluctant to discuss SUDEP routinely [29,30,33]. Now that risk factors and prevalence of SUDEP in CYPwE are better understood [2,5-6], it has been suggested that there is a need now more than ever to raise awareness of SUDEP in CYPwE, not just among healthcare professionals and their patients, but across policy-makers and the wider public health community [12].

The provision of SUDEP information face-to-face, with supporting written or online material, by a specialist known to the caregiver or CYPwE is in keeping with person-centred care [34] and contemporary practice for disclosing any information of an emotive nature. Whether or not to include the CYPwE in the discussion is perhaps less clear; only two of the studies in the review mentioned an age of 12+ and 13.5 years old respectively [9-10]. More commonly, parents felt they should choose whether and when to involve CYPwE in the discussion. We also found a lack of evidence on the age at which CYPwE should be included in the discussion about SUDEP, and on how to modify information provision across different age groups, which would logically be expected to be appropriate.

The findings recommend that SUDEP information includes prevalence, risk factors, and risk reduction methods. Although not specific to CYPwE, the SUDEP and seizure safety checklist developed by Shankar et al has been found to encourage healthcare professionals to discuss SUDEP with their patients [35]. It takes approximately 10-minutes to complete [36] and allows a risk assessment to be conducted, providing an individualised SUDEP risk rating which can be used to begin discussions about SUDEP and to provide a tailored risk reduction plan involving lifestyle modifications such as medication compliance and night-time surveillance [37]. The checklist developers recently highlighted the need for SUDEP risk assessment, discussion and reduction to be an ongoing process throughout the course of the individual’s epilepsy [36]. This is again in keeping with person-centred care. Specific risk-reduction methods are out with the scope of this review. However, due to the lack of evidence on the aetiology of paediatric SUDEP, optimising the management of epilepsy is currently the best practice available for its prevention, with further large-scale studies required to fully explore pathology, risk factors and possible preventive measures [12].

This review is not without limitations. We limited the literature search to the past 12-years. Whilst we hope this makes the review findings relevant to contemporary practice it is possible that we have excluded relevant literature. Likewise, we excluded conference abstracts due to the lack of data we could extract from them. We also limited the search to studies published in English, and all but one of the resulting studies were from Western countries. The findings may therefore not be generalizable to other healthcare contexts. We did not exclude any studies on methodological quality and therefore the included studies ranged from low to high quality. However, only one study was assessed as low quality, and its results were confirmed by other studies in the review; therefore we are confident that this has not had a major impact on the review findings.

Conclusions and Recommendations

When, where, how, and with whom to have discussions about the risk of SUDEP in CYPwE is an important topic. This review confirms that healthcare professionals should discus SUDEP with CYPwE and/or their caregivers at or around the time of diagnosis and that the discussion should include prevalence of SUDEP, risk factors and risk reduction methods relative to the individual concerned. Apart from delivering SUDEP information face-to-face, with written or online information provided to reinforce messages, there is a lack of evidence on "how" to impart this sensitive information across the age-range of children and young people receiving a diagnosis of epilepsy. Further research exploring the most acceptable and effective methods of discussing SUDEP with CYPwE and their caregivers is therefore indicated.

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Conflicts of Interest

Sarah Florida-James is a SIGN Programme Manager and led the development of the SIGN Guideline "Diagnosis and management of epilepsies in children". Kay Cooper, Pamela Kirkpatrick, Alix Rolfe and Celia Brand were members of the SIGN Guideline Development Group for the same guideline.

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Appendix 1: Search strategies

Search Strategy

MEDLINE & CINAHL Via EBSCOhost

S1 MH epilepsy/ OR epilepsy

S2 MH death, sudden/ OR sudden unexplained death in epilepsy OR SUDEP

S3 MH paediatric/s OR pediatric* OR paediatric*

S4 MH child/ OR child*

S5 MH young adult/ OR young

S6 S3 OR S4 OR S5

S7 S1 AND S2 AND S6

Limiters 01/01/2007 – 31/12/2018

PsycARTICLES Via EBSCOhost

MH epilepsy OR epilepsy

Limiters 01/01/2007 – 31/12/2018

AMED Via EBSCOhost

S1 epilepsy

S2 SUDEP OR sudden unexplained death in epilepsy

Limiters 01/01/2007 – 31/12/2018

EMBASE Via Ovid

S1 MH epilepsy/

S2 epilepsy

S3 S1 OR S2

S4 MH sudden death/

S5 sudden unexplained death in epilepsy

S6 SUDEP

S7 S4 OR S5 OR S6

S8 MH pediatrics/

S9 pediatric*

S10 paediatric*

S11 S8 OR S9 OR S10

S12 child*

S13 MH young adult/

S14 young

S15 S11 OR S12 OR S13 OR S14

S16 S3 AND S7 AND S15

Limiters 01/01/2007 – 31/12/2018

PsychINFO Via Ovid

S1 MA epilepsy OR epilepsy

S2 MA sudden death OR SUDEP OR sudden unexplained death in epilepsy

S3 MA paediatrics OR pediatric* OR paediatric*

S4 S1 AND S2 AND S3

Grey literature

Epilepsy and SUDEP

Epilepsy and sudden unexplained death

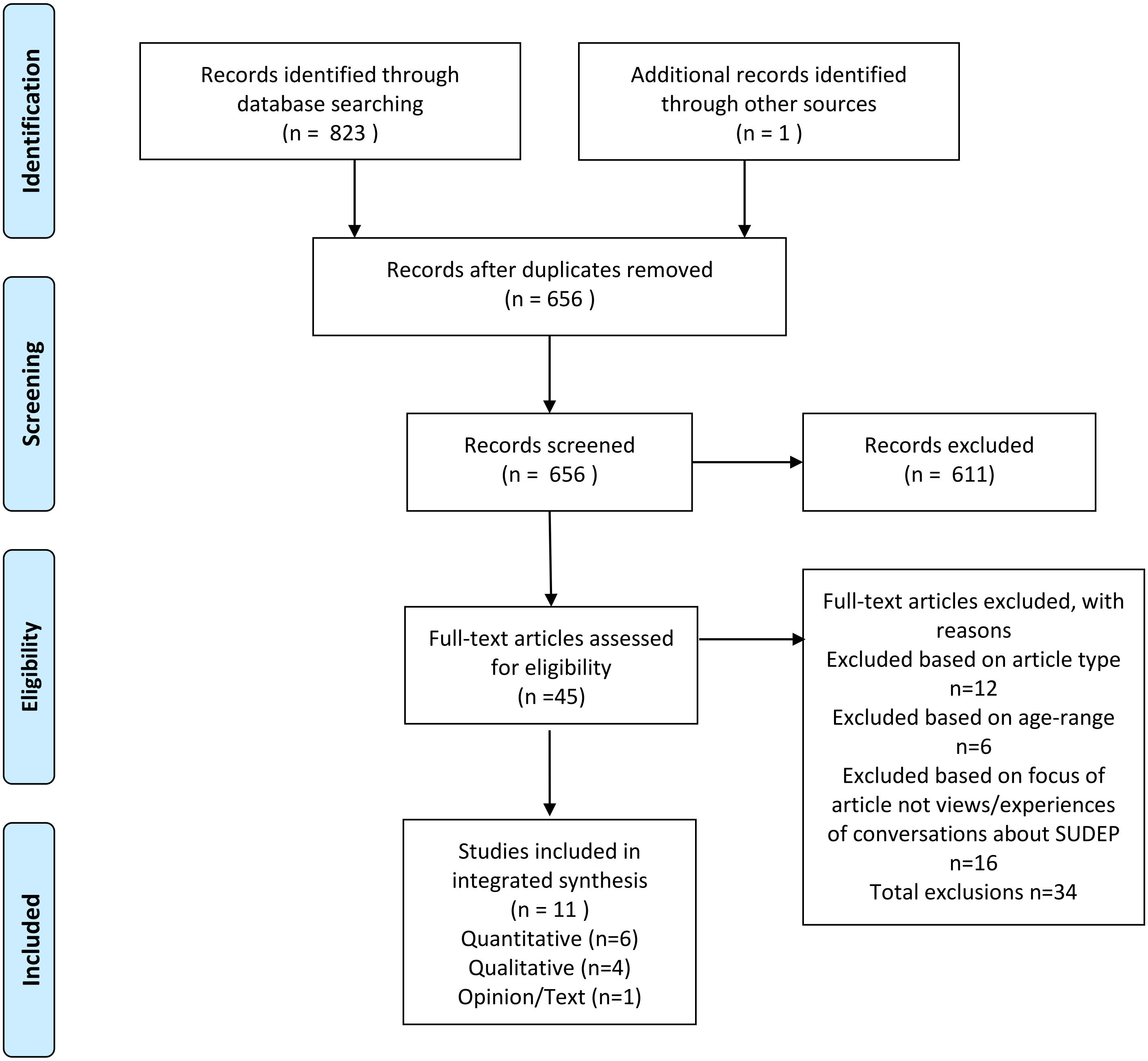

Figure 1: PRISMA Flow Diagram

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

Table 1: Characteristics of included studies

Author	Year	Location	Population	Methods	Phenomena of Interest	Main Findings
Berl et al [23]	2017	USA	Paediatric Primary Care Providers (n=149)	Survey	Knowledge of SUDEP & how it relates to care of patients with epilepsy	Only 3.4% discussed SUDEP with patients/caregivers 79% expected neurologist to discuss SUDEP
Fong et al [28]	2017	Malaysia	Parents/guardians of CYPwE aged up to 18-years (n=27) Mean age 9.7 years (±4.7)	Survey	Beliefs regarding provision of SUDEP information	SUDEP should be discussed (90.6%) SUDEP should be discussed at diagnosis (72.2%) Internet/computer programme/leaflet and discussion with neurologist best way to discuss with parents (59.1%) and CYPwE (53.9%) Should be discussed with CYPwE (70.1%)
Gayatri et al [9]	2010	UK	Parents of children attending epilepsy clinic (n=67 round 1 & n=47 round 2)	Questionnaires: immediately after receiving SUDEP information &	Beliefs regarding provision & content of SUDEP information	Parents: Most want to know about SUDEP at time of diagnosis and think they or healthcare

			Mean age 10.6 (±3.6) Paediatric neurologists (n=46)	3-month follow-up 1 Questionnaire		professional should discuss with CYPwE Neurologists: Most provide SUDEP information to selected patients via personal communication
Lewis et al [24]	2008	UK	Clinical Nurse Specialists (n=27 paediatric)	Postal survey	What, when and how issues related to SUDEP are raised and discussed	SUDEP information should be provided during home visits or by telephone, by a specialist Lack of agreement on whether all patients/carers should be informed
Louik et al [29]	2017	USA	Family members bereaved by SUDEP (n=43) Mixed population but median age at death 24; 31% aged 18 or younger	Semi-structured telephone interview	Attitudes towards SUDEP discussion	72% wished SUDEP had been discussed Wanted to discuss: general information; incidence; risk factors; prevention; importance of drug adherence
Strzelczyck et al [10]	2016	Austria Germany Switzerland	Neurologists & neuropaediatricians (n=161 paediatricians)	Online/paper survey	Attitudes towards counselling about SUDEP	SUDEP not widely discussed and generally when disease course refractory

Galli et al [30]	2017	Italy	Medical doctors caring for paediatric patients with epilepsy (n=114)	Electronic survey (data from open question extracted)	To explore disclosure of SUDEP risk	Timing important to control Handling emotions important
Harden et al [25]	2015	UK	Young adults known to have received SUDEP information (n=27) Age 18-19; mean 22 years	Semi-structured interviews	Views on SUDEP information-giving	SUDEP information should be provided face-to-face by a specialist. Lack of consensus on timing
RamachandranNair et al [26]	2013	Canada	Parents bereaved by epilepsy (n=42) Age of CYPwE not reported	Focus groups & 1-1 interviews	Views on discussing SUDEP	Should be routine, face-to-face, counselling by the neurologist, reinforced by written information. Parents should decide whether child should be present or not
Tonberg et al [27]	2015	UK	Young adults known to have received SUDEP Information (n=27) Mean age 22.1 years (±3.5)	Qualitative interviews	Views on SUDEP disclosure	CYPwE have a right to know about SUDEP Timing important Face-to-face by someone knowledgeable who CYPwE has rapport with
Stevenson et al [31]	2014	USA	Bereaved fathers (n=2)	N/A Opinion article	Views on SUDEP disclosure	Parents want to discuss SUDEP with a Doctor

	Age of CYPwE not reported

CYPwE=children & young people with epilepsy

Table 2: Integrated Findings, Categories and Qualitied or Qualitative Data

Findings from Qualitized (QZ) or Qualitative (QT) data	Categories	Integrated Findings
QZ Gayatri et al [9]: Most Paediatric Neurologists provide SUDEP information to selected patients (e.g. intractable seizures, aged 12+) and/or parents/guardians	**Category 1** Some discordance between carers'/family members' and healthcare professionals' on discussing the risk of SUDEP in CYPwE	**Integrated Finding 1** Caregivers, and where appropriate CYPwE, should be provided with information on SUDEP and how it relates to them: The information on SUDEP should include prevalence, risk factors and risk reduction measures that can be taken.
QZ Lewis et al [24]: Lack of agreement among Clinical Nurse Specialists on whether all patients & carers should be informed about SUDEP		
QZ Strzelczyck et al [10]: Most Neurologists & Neuropaediatricians discuss SUDEP with CYPwE from age 13.5; some only discuss with caregivers		
QT Harden et al [25]: **SUDEP information should be given to those with epilepsy** ("I think, everyone should have access to information and not be, like, have things withheld from them 'cos I think it kind of causes a bit of mistrust…being told what you should and shouldn't know isn't good" (pp234) Unequivocal		
QT Tonberg et al [27]: **The right to know** ("I feel like if I didn't have all the information, I would feel a bit cheated") pp 100 Unequivocal		
QT Stevenson et al [31]: **Parents want to discuss SUDEP** ("When a Dr provides that information in an open and honest way, it builds trust and respect, and offers parents the opportunity to make informed decisions") pp1496 Credible [1]		
QT RamachandranNair et al [26]: **Knowing better than not knowing** ("Not knowing about the possibility of SUDEP would be worse…they would feel angry if they were not informed, and if their child subsequently experienced SUDEP, they would feel burdened by guilt that there might have been more they could have done") (pp562) Credible		
QT Galli et al [30]: **Explaining SUDEP** ("On the one hand communication regarding SUDEP seemed to become possible when the circumstances were severe even if the related risk of death was too distant to be sudden and unexpected…on the other hand, communication about SUDEP was endorsed if the patient/parent(s) asked about SUDEP or if there was a need to improve compliance" (pp36) Credible		
QZ Gayatri et al [9]: SUDEP information more likely to be given when seizures difficult to control than at diagnosis		
QT RamachandranNair et al [26]: **Balanced counselling** ("…some parents expressed that it is important for neurologists to stress that SUDEP is rare and to balance the message with one of hope") pp562 Credible		

QT Tonberg et al [27]: **Basic information is sufficient** (“…relating to prevalence, causation, and preventive strategies”) pp101 Credible	**Category 2** SUDEP information should include prevalence, risk factors, and risk reduction	
QZ Louik et al [29]:Family members want information on SUDEP, including general information, incidence, risk factors, preventive measures and importance of drug adherence		
QT Harden et al [25]: **Magnitude of SUDEP risk within context of own epilepsy most important** (“I liked being told how it related to me…just having someone to put it into perspective” (pp235) Unequivocal		
QZ Gayatri et al [9]: Majority of parents want to know about SUDEP at time of diagnosis, not just when seizures difficult to control	**Category 3** Consideration of the timing and who should be present during SUDEP discussions is important	**Integrated Finding 2** Information on SUDEP should be delivered face-to-face, with supporting written information, by a suitably knowledgeable healthcare professional whom the caregiver/CYPwE feels comfortable with, at an appropriate time at or close to diagnosis
QT: Galli et a[30]: **Controlling the timing of communication** (“It is important to schedule the meeting and not be in a hurry” (pp35)) Unequivocal		
QT Harden et al [25]: **Disagreement regarding the timing of SUDEP information-giving** (“I suppose that’s a judgement for the doctors and consultants to make if they think you can handle then you may as well have it all as soon as possible, but if you’re an anxious person, or it’s more complicated…maybe do it in chunks” (pp235) Unequivocal		
QT Fong et al [28]: Majority of parents/guardians want to know about SUDEP at time of diagnosis		
QT Fong et al [28]: Parents/guardians felt SUDEP should be discussed with CYPwE either at diagnosis or when epilepsy becomes poorly controlled		
QT Fong et al [28]: Some wanted to tell their child themselves		
QT RamachandranNair et al [26]: **Who should receive counselling** (“…it should be the parents’ decision as to whether or not the child should be present at the meeting or when to inform the child about the risk of SUDEP”) pp563 Credible		
QT Tonberg et al [27]: **Importance of timing (“**You don’t wanna be in the dark about something that’s part of you…but I think that the time needs to be chosen carefully”) pp101 Unequivocal		
QT Timing of SUDEP counselling RamachandranNair et al [26]: (“Parents generally expressed a preference for receiving routine SUDEP counselling at the time of the diagnosis of epilepsy”) pp 562 Credible		
QZ Berl et al [24]: Primary Care Providers lack SUDEP knowledge and expect specialists to discuss with patients/caregivers		
QZ Lewis et al [25]: When provided, specialists should provide information about SUDEP		
QT RamachandranNair et al [26]: **The pediatric neurologist’s responsibility** (“…while it is the responsibility of the pediatric neurologist to conduct the SUDEP counselling, parents expressed a		

preference for meeting with a clinical nurse who could "translate what the doctors say" or a clinical social worker "to soften the blow"" pp563 Credible		
QT Stevenson et al [31]: **A doctor should provide information on SUDEP** ("Information about SUDEP should optimally come from a Doctor") pp1499 Credible		
QT Harden et al [25]: **Information should be provided by a specialist** ("It was more the nurse that you'd speak to and it's the nursing that you can phone at any time and speak to them…they probably have a much better of view how the patients see their epilepsy (pp235) Unequivocal		
QZ Gayatri et al [9]:Parents think that parents or healthcare professionals supported by written information should discuss SUDEP with CYPwE	**Category 4** SUDEP discussions should be face-to-face wherever possible and supported by written information	
QZ Gayatri et al [9]: Personal communication preferred method of providing SUDEP information for most Paediatric Neurologists		
QZ Lewis et al [24]: SUDEP information should be provided during home visits or by telephone		
QT Harden et al [25]: **Importance of face-to-face delivery** ("The majority of participants said that it was important that this information was presented face-to-face, within the context of a consultation rather than via an information leaflet" (pp235. Credible)		
QT RamachandranNair et al [26]: **Importance of face-to-face counselling** ("Across all groups of parents, it was identified that SUDEP counselling should occur in a face-to-face interaction between the neurologist and the parents and not be provided in a phone consultation") pp562 Credible		
QT Fong et al [28]: Combined internet/computer programme/leaflet *and* discussion with neurologist preferred method of receiving SUDEP information for most parents/guardians		
QT Fong et al [28]: Parents/guardians felt internet/computer programme/leaflet and discussion with neurologist preferred method of discussing SUDEP with CYPwE but also support for healthcare professionals alone		
QT: RamachandranNair et al [26]:: **Written information to reinforce counselling** ("…an important strategy in supporting parents to manage the extensive amount of information received during their appointments") pp562 Credible		
QT: Tonberg et al [27]: **Importance of face-to-face information provision** ("There was strong consensus among participants that information about SUDEP should be delivered face-to-face") pp101 Unequivocal		
QT Galli et al [30]: **Handling emotions** ("The respondents described a varied and often polarized spectrum of emotions. In some cases the presence of emotions seemed to be useful for		

disclosure…On the other hand, emotions may have impeded or even denied the possibility of disclosure for other physicians” (pp35) Credible	**Category 5** The person discussing SUDEP should be knowledgeable and have a good rapport with family members/carers/CYPwE, taking their emotions into account	
QT Tonberg et al [27]: **Importance of knowledge and rapport** (“The epilepsy nurse I would say…I feel they’ve got a bit more time for you I would say, and they probably become…attached is not the right word but they, they know you more, I would say, than the doctors”) pp101 Unequivocal		

Studies excluded at full-text screening, with reasons (n=34)

ABDALLA, I.G., SCORZA, C.A., FIORINI, A.C., CAVALHEIRO, E.A. and SCORZA, F.A., 2014. Sudden unexpected death in children with epilepsy: Hearing from parents. *Epilepsy & Behavior,* **31**, pp. 48-49.

Reason for exclusion: Commentary on another article

BAXTER, P., 2011. Explaining risks and benefits. *Developmental Medicine & Child Neurology,* **53**(10), pp. 873.

Reason for exclusion: Editorial; not focussed on views & experiences

BELLON, M., PANELLI, R.J. and RILLOTTA, F., 2015. Epilepsy-related deaths: An Australian survey of the experiences and needs of people bereaved by epilepsy. *Seizure,* **29**, pp. 162-168.

Reason for exclusion: Mixed age-range

BRODIE, M.J. and HOLMES, G.L., 2008. Should all patients be told about sudden unexpected death in epilepsy (SUDEP)? Pros and Cons. *Epilepsia,* **49**(SUPPL. 9), pp. 99-101.

Reason for exclusion: Not focussed on views & experiences

DEVINSKY, O., 2011. Sudden, unexpected death in epilepsy. *New England Journal of Medicine,* **365**(19), pp. 1801-1811.

Reason for exclusion: Not focussed on views & experiences

DONNER, E. and BUCHHALTER, J., 2014. Commentary: It's time to talk about SUDEP. *Epilepsia,* **55**(10), pp. 1501-1503.

Reason for exclusion: Commentary; not focussed on views & experiences

DONNER, E.J., 2011. Explaining the unexplained; expecting the unexpected: where are we with sudden unexpected death in epilepsy? *Epilepsy Currents,* **11**(2), pp. 45-49.

Reason for exclusion: Not focussed on views & experiences

DONNER, E.J., CAMFIELD, P., BROOKS, L., BUCHHALTER, J., CAMFIELD, C., LODDENKEMPER, T. and WIRRELL, E., 2017. Understanding Death in Children With Epilepsy. *Pediatric neurology,* **70**, pp. 7-15.

Reason for exclusion: Review

DONNER, E.J., WADDELL, B., OSLAND, K., LEACH, J.P., DUNCAN, S., NASHEF, L. and PICOT, M.C., 2016. After sudden unexpected death in epilepsy: Lessons learned and the road forward. *Epilepsia,* **57**(Supplement 1) (pp 46-53), pp. ate of Pubaton: 01 Jan 2016.

Reason for exclusion: Not focussed on views & experiences

DOUMLELE, K., BUCHHALTER, J., CRANDALL, L., DEVINSKY, O., DONNER, E., DIXONSALAZAR, T., FOWLER, D., HESDORFFER, D., WRIGHT, C. and FRIEDMAN, D., 2016. Experiences with premorbid SUDEP discussion among participants in the North American SUDEP registry (NASR). *Neurology,* **Conference**, pp. 68th.

Reason for exclusion: Mixed age-range

DUNCAN, S., TONBERG, A., HARDEN, J., MCLELLAN, A. and CHIN, R., 2014. Sudep disclosure in young adults with epilepsy: Patients' reaction, perception of risk, views on timing and behavioural change. *Epilepsy Currents.Conference: 2013 Annual Meeting of the American Epilepsy Society, AES 2013.Washington, DC United States.Conference Publication: (var.pagings),* **14**(SUPPL. 1), pp. 164.

Reason for Exclusion: Conference abstract only

FISHER, P.G., 2017. Do you know what SUDEP is? *Journal of Pediatrics,* **188**, pp. 2.

Reason for exclusion: Commentary on another article

FONG, C.Y., LIM, W.K., KONG, A.N., LUA, P.L. and ONG, L.C., 2017. Provision of sudden unexpected death in epilepsy (SUDEP) information among Malaysian parents of children with epilepsy. *Epilepsy & Behavior,* **75**, pp. 6-12.

Reason for Exclusion: Focus on epilepsy education software programme

FRIEDMAN, D., DONNER, E.J., STEPHENS, D., WRIGHT, C. and DEVINSKY, O., 2014. Sudden unexpected death in epilepsy: Knowledge and experience among US and Canadian neurologists. *Epilepsy & Behavior,* **35**, pp. 13-18.

Reason for Exclusion: Mixed population

HENNING, O., NAKKEN, K.O. and LOSSIUS, M.I., 2018. People with epilepsy and their relatives want more information about risks of injuries and premature death. *Epilepsy and Behavior,* **82**, pp. 6-10.

Reason for Exclusion: Nor about conversations & not CYPwE

JONES, L. and TE WATER NAUDE, J., 2013. Sudden unexpected death in epilepsy information provision to parents of children with epilepsy - A service evaluation. *Journal of Neurology, Neurosurgery and Psychiatry,* **Conference**

Reason for Exclusion: Not about conversations

KHAN, A., BAHEERATHAN, A. and HUSSAIN, N., 2014. SUDEP--patients' 'right to know' or 'right not to know'. *Epilepsy & behavior : E&B,* **41**, pp. 78.

Reason for Exclusion: Letter to Editor

KRUJA, J. and VYSHKA, G., 2012. Avoiding parental distress when discussing about SUDEP: the Albanian experience. *Medicinski arhiv,* **66**(3), pp. 201-203.

Reason for Exclusion: Not focussed on views & experiences

LAPHAM, G., 2017. Increasing Awareness of Sudden Death in Pediatric Epilepsy Together. *Pediatrics,* **139**(2), pp. 10-13.

Reason for Exclusion: Not about conversations

MASSEY, S., FRIEDMAN, D., BERBERI, N., DEVINSKY, O. and KOTHARE, S., 2015. Sudep awareness among pediatric practitioners: A descriptive survey of united states physicians. *Epilepsy Currents.Conference: 68th Annual Meeting of the American Epilepsy Society, AES 2014.Seattle, WA United States.Conference Publication: (var.pagings),* **15**(SUPPL. 1), pp. 262.

Reason for Exclusion: Conference abstract; not focussed on conversations

MEHTA, S., 2015. Getting it right for young people with epilepsy. *British Journal of Neuroscience Nursing,* **11**(5), pp. 238.

Reason for Exclusion: Not focussed on conversations

MILLER, W.R., YOUNG, N., FRIEDMAN, D., BUELOW, J.M. and DEVINSKY, O., 2014. Discussing sudden unexpected death in epilepsy (SUDEP) with patients: Practices of health-care providers. *Epilepsy & Behavior,* **32**, pp. 38-41.

Reason for Exclusion: Mixed age-range

PYSDEN, K., FERRIE, C. and GAYATRI, N., 2007. A survey into the practice of paediatric neurologists in the United Kingdom and Ireland when providing information about sudden unexpected death in epilepsy...British Paediatric Neurology Association Annual Meeting 2007, 17th-19th January. *Developmental Medicine & Child Neurology,* **49**, pp. 18.

Reason for Exclusion: Conference abstract only

RAMACHANDRAN NAIR, R., JACK, S.M. and STROHM, S., 2016. SUDEP: To discuss or not? Recommendations from bereaved relatives. *Epilepsy & Behavior: E&B,* **56**, pp. 20-25.

Reason for Exclusion: Mixed age-range

RAMACHANDRANNAIR, R., JACK, S.M., MEANEY, B.F. and RONEN, G.M., 2012. SUDEP: Is there an optimal way to inform parents?. *Canadian Journal of Neurological Sciences.Conference: 47th Annual Congress of the Canadian Neurological Sciences Federation.Ottawa, ON Canada.Conference Publication: (var.pagings),* **39**(3 SUPPL. 3), pp. S13.

Reason for Exclusion: Conference abstract only

REESE JR., J., FRIEDMAN, D. and GAILLARD, W., 2015. Sudep experience and practice in a large group of child neurologists. *Epilepsy Currents.Conference: 68th Annual Meeting of the American Epilepsy Society, AES 2014.Seattle, WA United States.Conference Publication: (var.pagings),* **15**(SUPPL. 1), pp. 248-249.

Reason for Exclusion: Conference abstract only

RONEN, G.M., 2017. Applying ethical principles in discussing SUDEP with patients and families. *Epilepsy & Behavior,* **70**, pp. 269-270.

Reason for Exclusion: Not focussed on views & experiences

SCORZA, F.A., 2015a. Breaking bad news on the possible occurrence of sudden death in children with epilepsy sleeping on sofas. *Epilepsy & Behavior: E&B,* **50**, pp. 88-89.

Reason for Exclusion: Focus on sleeping on sofas

SHAFER, P.O. and BUCHHALTER, J., 2016. Patient Education: Identifying Risks and Self-Management Approaches for Adherence and Sudden Unexpected Death in Epilepsy. *Neurologic clinics,* **34**(2), pp. 443.

Reason for Exclusion: Not focussed on conversations

SHANKAR, R., DONNER, E.J., MCLEAN, B., NASHEF, L. and TOMSON, T., 2017. Sudden unexpected death in epilepsy (SUDEP): what every neurologist should know. *Epileptic Disorders: International Epilepsy Journal With Videotape,* **19**(1), pp. 1-9.

Reason for Exclusion: Review

TOMSON, T., SURGES, R., DELAMONT, R., HAYWOOD, S. and HESDORFFER, D.C., 2016. Who to target in sudden unexpected death in epilepsy prevention and how? Risk factors, biomarkers, and intervention study designs. *Epilepsia,* **57 Suppl 1**, pp. 4-16.

Reason for Exclusion: Review

VERMA, A. and KUMAR, A., 2015a. Sudden unexpected death in epilepsy: some approaches for its prevention and medico-legal consideration. *Acta Neurologica Belgica,* **115**(3), pp. 207-212.

Reason for exclusion: Not focussed on conversations

XU, Z., AYYAPPAN, S. and SENEVIRATNE, U., 2015. Sudden unexpected death in epilepsy (SUDEP): What do patients think? *Epilepsy & Behavior,* **42**, pp. 29-34.

Reason for Exclusion: Adult patients

YOUNG, C., SHANKAR, R., HENLEY, W., ROSE, A., CHEATLE, K. and SANDER, J.W., 2018. SUDEP and seizure safety communication: Assessing if people hear and act. *Epilepsy & Behavior: E&B,* **86**, pp. 200-203.

Reason for Exclusion Not focussed on conversations

Methodological Quality Assessment

Analytical cross-sectional studies

	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	
Berl et al 2017	Y	Y	NA	NA	N	N	Y	Y	Moderate
Fong et al 2017	Y	Y	NA	NA	Y	N/A	Y	Y	High
Gayatri et al 2010	Y	N	NA	NA	N	N	Y	Y	Low
Lewis et al 2008	Y	Y	NA	NA	N	N	Y	Y	Moderate
Louik et al 2017	Y	Y	NA	NA	Y	Y	Y	Y	High
Strzelczyck et al 2016	Y	Y	NA	NA	Y	Y	Y	Y	High

Key: Q1 Were the criteria for inclusion in the sample clearly defined? Q2 Were the study subjects and the setting described in detail? Q3 Was the exposure measured in a valid and reliable way? Q4 Were objective, standard criteria used for measurement of the condition? Q5 Were confounding factors identified? Q6 Were strategies to deal withj confounding factors stated? Q7 Were the outcomes reported in a valid and reliable way? Q8 Was appropriate statistical analysis used? NA Not applicable

Qualitative studies

	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	
Galli et al 2017	Y	Y	Y	Y	Y	NA	NA	Y	Y	Y	Moderate
Harden et al 2015	N	Y	Y	Y	Y	N	N	Y	Y	Y	Moderate
RamachandranNair et al 2013	Y	Y	Y	Y	Y	N	N	N	Y	Y	Moderate
Tonberg et al 2015	N	Y	Y	Y	Y	N	N	Y	Y	Y	Moderate

Key: Q1 Is there congruity between the stated philosophical perspective and the research methodology? Q2 Is there congruity between the research methodology and the research question or objectives? Q3 Is there congruity between the research methodology and the methods used to collect data? Q4 Is there congruity between the research methodology and the representation and analysis of the data? Q5 Is there congruity between the research methodology and the interpretation of results? Q6 Is there a statement locating the researcher culturally or theoretically? Q7 Is the influence of the researcher on the research, and vice-versa, addressed? Q8 Are participants, and their voices, adequately represented? Q9 Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body? Q10 Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data? NA Not applicable

Text & Opinion

	Q1	Q2	Q3	Q4	Q5	Q6	
Stevenson et al 2014	Y	Y	Y	Y	Y	NA	Moderate

Key: Q1 Is the source of opinion clearly identified? Q2 Does the source of opinion have standing in the field? Q3 Are the interests of the relevant population the central focus of the opinion? Q4 Is the stated position the result of an analytical process, and is there logic in the opinion expressed? Q5 Is there reference to the extant literature? Q6 Is any incongruence with the literature/sources logically defended? NA Not applicable