

Experiences of living with fetal alcohol spectrum disorders: a systematic review and synthesis of qualitative data

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ABBREVIATIONS

CERQual	Confidence in the Evidence from Reviews of Qualitative Research
FASD	Fetal alcohol spectrum disorders
GRADE	Grading of Recommendations, Assessment, Development and Evaluation

AIM To identify and assess available evidence from qualitative studies exploring experiences of individuals living with fetal alcohol spectrum disorders (FASD) or those living with a child with FASD, as well as experiences of interventions aimed at supporting individuals with FASD and their families.

METHOD A systematic literature search was conducted in six electronic databases: PubMed, Embase, Cochrane Library, CINAHL, PsycINFO, and Scopus. Included studies were analysed using manifest content analysis. Methodological limitations and confidence in the evidence were assessed using a modified version of the Critical Appraisal Skills Programme and the Grading of Recommendations, Assessment, Development and Evaluation–Confidence in the Evidence from Reviews of Qualitative Research approach respectively.

RESULTS Findings from 18 studies show that individuals with FASD experience a variation of disabilities, ranging from somatic problems, high pain tolerance, destructive behaviour, hyperactivity, and aggressiveness, to social problems with friendship, school attendance, and maintenance of steady employment. Most studies reported parents' experiences with FASD; parenting was viewed as a lifelong engagement and that the whole family is isolated and burdened because of FASD. People with FASD feel that their difficulties affect their daily life in a limiting way and make them feel different from others.

INTERPRETATION From the perspective of primarily parents, individuals with FASD and their parents face many different difficulties, for which they need societal support.

Fetal alcohol spectrum disorders (FASD) is an umbrella term describing the range of developmental deviations, such as craniofacial maldevelopment or neurodevelopmental abnormalities that can occur in an individual whose mother consumed alcohol during pregnancy.¹ The extent of developmental deviations are believed to depend on the amount, pattern, and timing of the prenatal alcohol exposure. However, other factors, including genetic, epigenetic, and social environmental factors, may also influence the development and manifestations of the disabilities.² The prevalence rates for FASD have been reported to be 2% to 5% in USA, 2% to 6% in Italy, and as high as 14% to 21% in South Africa.^{3–5}

Terms such as fetal alcohol syndrome, partial fetal alcohol syndrome, alcohol-related birth defects, and alcohol-related neurodevelopmental disorder are used to label different clusters of anomalies along the FASD spectrum.¹ The most well-known term within FASD is fetal alcohol syndrome, which is an established diagnosis ascribed to

individuals who fulfil specific criteria, such as facial dysmorphism, growth inhibition, and dysfunction of the central nervous system.⁶

Many children with FASD experience difficulties in daily life, both with their families and during the schooling period.⁶ FASD are associated with difficulties on many different fronts, concerning, for example, motor control, eyesight, hearing, attention, concentration, and impulse control. In addition, comorbidity of autism, attention-deficit–hyperactivity disorder, and intellectual disability are also seen in individuals with FASD.⁷ However, the symptoms that individuals with FASD exhibit are not always prominent enough for them to be identified by professionals in health care, social care, and the educational system, causing these children and their families to go unnoticed and leaving them without care and support.

The aim of this systematic review is to identify and assess the available evidence from qualitative studies

investigating the experiences of individuals living with FASD or those caring for a child with FASD.

METHOD

Protocol and registration

This systematic review was initiated as part of a Health Technology Assessment report written in Swedish at The Swedish Agency for Health Technology Assessment and Assessment of Social Services, which was submitted to the Swedish Ministry of Social Affairs in December 2016 (www.sbu.se/258). The Swedish Agency for Health Technology Assessment and Assessment of Social Services uses a peer-reviewed protocol for systematic reviews, available upon request. The systematic review process follows the general concepts covered by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).⁸

Inclusion criteria

Sample

Individuals with FASD or parents (biological, adoptive, or foster parents) of individuals with FASD.

Phenomenon of interest

Any experiences regarding living with FASD or experiences receiving any interventions aimed for the individuals with FASD or their parents.

Design

Peer-reviewed studies published in English, Danish, Norwegian, or Swedish reporting qualitative data, including themes and quotes, were collected using a qualitative or mixed method. Studies published before 1990 were excluded. No restrictions regarding research type or study settings were made.

Search strategy and study selection

Studies were identified by a literature search of six electronic databases performed by a health science librarian and by scanning reference lists by the reviewers. Medical subject headings and relevant terms related to FASD were used. The electronic databases PubMed, Embase, Cochrane Library, CINAHL, PsycINFO, and Scopus were searched on 29th September 2016 (see Appendix S1, online supporting information). Grey literature was not searched for.

Two review authors (HD, GB, GF, MH) independently screened the titles and abstracts for the references identified in the literature. Potentially relevant abstracts were then read in full and screened independently by two review authors. Any disagreements regarding inclusion of an article were resolved by discussions involving a third party, if necessary. For a flow chart of the review process, see Figure 1.

The methodological strengths and weaknesses of the included studies were assessed independently by two reviewers using a modified version of the Critical Appraisal Skills Programme checklist for qualitative studies.⁹ Studies were then classified as having low, moderate, or severe limitations (see Appendix S2, online supporting information).

What this paper adds

- Individuals with fetal alcohol spectrum disorders (FASD) feel their difficulties make them different from others.
- Parents think of their parenting as a lifelong engagement.
- There is a shortage of studies investigating experiences of children with FASD.
- There is a shortage of studies investigating experiences of interventions given to individuals with FASD.

Data extraction and qualitative synthesis

Two reviewers extracted data from each study, and one to three additional reviewers checked the extraction for each study. The review authors for this systematic review possess experience in conducting systematic reviews and working in fields related to preclinical and clinical medical science, medical ethics, social medicine, and social work.

Qualitative content analysis focusing on the manifest content of the experiences was used regardless of analysis methods applied in the original studies.^{10,11} The first-level themes were identified, interpreted, specified, and verified according to the data provided in the articles. The second-level themes were similarly developed based on the first-level themes. Finally, the third-level themes were used to organize the second-level themes into broader categories.

When consensus was reached for all themes, an assessment was made of confidence in each review finding (second-level themes) using the Grading of Recommendations, Assessment, Development and Evaluation (GRADE)-Confidence in the Evidence from Reviews of Qualitative Research (CERQual) approach.¹² The data supporting the first-level themes and details regarding the CERQual assessments are described in detail in Appendix S3 (online supporting information).

CERQual assessment of confidence

CERQual is developed within GRADE to evaluate the confidence of the weighted result of qualitative data.¹² The confidence is affected by factors such as methodological limitations, relevance, coherence, and adequacy of data.

Definitions of levels of confidence in a review finding:

- High confidence (⊕⊕⊕⊕): it is highly likely that the review finding is a reasonable representation of the phenomenon of interest.
- Moderate confidence (⊕⊕⊕○): it is likely that the review finding is a reasonable representation of the phenomenon of interest.
- Low confidence (⊕⊕○○): it is possible that the review finding is a reasonable representation of the phenomenon of interest.
- Very low confidence (⊕○○○): it is not clear whether the review finding is a reasonable representation of the phenomenon of interest.

RESULTS

The literature search resulted in 3789 records. Of these, 18 studies fulfilled the inclusion criteria. Fourteen studies were assessed as having moderate methodological limitations, three as having minor methodological limitations,

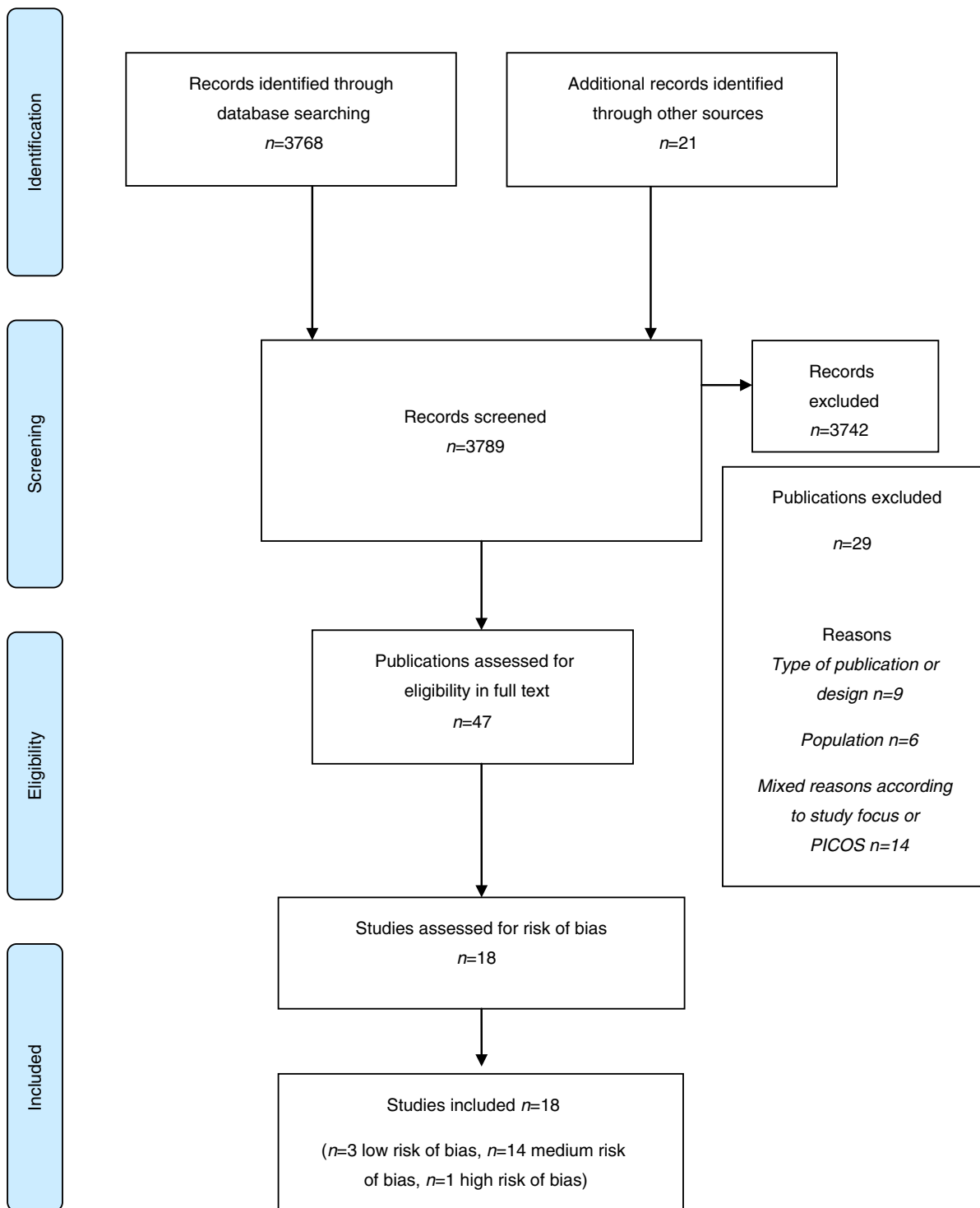


Figure 1: The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart. PICOS, population, intervention, comparison, outcome, study design. [Colour figure can be viewed at wileyonlinelibrary.com].

and one was assessed as having serious methodological limitations (see Appendix S2).

Of the 18 included studies, two described experiences of children and adults with FASD ($n=36$), whereas 15 studies provided information on parents' experiences of raising a

child with FASD, and one study described parental experiences of the intervention respite care (at least 237 caregivers). Included studies were conducted in Canada, New Zealand, USA, UK, and Australia. The studies are described in Table I. The third-level themes, the second-

Table I: Studies meeting the inclusion criteria

Study and country	Populations	Method	Risk of bias
Chamberlain et al., ¹⁷ Australia	10 caregivers (4 foster parents, 1 adoptive parent, 5 legal guardians)	Phenomenological approach and thematic analysis (NVivo 10)	Moderate
Coons et al., ²⁴ Canada	84 parents from 59 families (16 birth parents, 5 step-parents, 55 adoptive parents, 8 foster parents)	Semi-structured interview; interpretative phenomenological analysis	Moderate
Doig et al., ²² Canada	10 parents (7 birth, foster, or adoptive mothers, 2 birth fathers, 1 grandmother)	Constructivist grounded theory approach	Low
Gardner, ²⁵ USA	8 foster mothers	Content analysis	Moderate
Granitsas, ²⁶ USA	5 adoptive parents	Qualitative phenomenological approach: 'What has it been like for you to parent a child with FAS?'	Moderate
Johnston and Boyle, ²⁷ Canada	8 birth mothers	Three semiformal interviews, participant observations; ethnographic analysis	Low
Michaud and Temple, ²⁸ Canada	5 mothers (adoptive, foster, or birth mothers)	Participatory action research model (bringing together people to facilitate collaboration, advocacy, and social change)	Moderate
Mukherjee et al., ¹⁸ UK	30 parents, mainly foster and adoptive parents and maybe a few biological parents	Focus groups from advertising, randomly max. 10 parents/group; mixed method, semi-structured analysis method; parental stress index and knowledge questionnaire NVivo	Moderate
Poth et al., ²⁹ Canada	6 caregivers (2 grandmothers, 3 foster mothers, 1 adoptive mother)	Qualitative analysis	Moderate
Ryan and Ferguson, ²¹ USA	Biological, adoptive, and foster parents and other extended family members	'Peer debriefing' of interviews	Moderate
Salmon and Buetow, ¹³ New Zealand	14 individuals with FASD (7 with FAS, 7 with FAE); 9 males and 5 females; aged 14–37y where 6 were ≤18y	Unstructured interviews; mixed method (transcendental phenomenology phase 1 and grounded theory in phase 2)	Moderate
Salmon, ¹⁶ New Zealand	8 birth mothers	Unstructured open-ended questions. Analysis constant comparative method. Validity: triangulation, participant checking, and closeness of fit	Moderate
Sanders and Buck, ¹⁵ Canada	11 parents (7 adoptive, 3 birth, and 1 foster parent)	Interview using 'unstructured format' to find out parent's experiences and thoughts. After the interview, the parents summarize the interview in two phrases. A phenomenological–hermeneutic approach and thematic analysis were used to analyse and organize the data into themes. QSR NVivo7 qualitative data analysis was used to help identify, categorize, and compare the themes	Moderate
Stade et al., ¹⁴ Canada	22 individuals with FASD (11 with ARND, 6 with pFAS, 5 with FAS); 10 females, 12 males, 6–18y	In-depth interview (phenomenological)	Moderate
Swart et al., ³⁰ Canada	17 caregivers (9 birth parents, 1 step parent, 5 adoptive parents, 2 foster parents)	Grounded theory	Moderate
Watson et al., ¹⁹ Canada	31 parents	Interpretative phenomenological analysis	Low
Watson et al., ²⁰ Canada	31 family members (birth parents, foster parents, adoptive parents, step-parents, custodial grandparents)	Qualitative interviews	Moderate
Whitehurst, ³¹ UK	4 mothers (1 birth, 3 adoptive)	Interpretative phenomenological approach	High

FAE, fetal alcohol exposure; FASD, fetal alcohol spectrum disorders; ARND, alcohol-related neurodevelopmental disorder; FAS, fetal alcohol syndrome; pFAS, partial fetal alcohol syndrome.

level themes, and the supporting first-level themes are summarized in Appendix S3 (online supporting information).

The CERQual assessments of the second-level themes are summarized in Tables II–VII and described in more detail in Appendix S3. The themes for which we found a moderate confidence are also described in the text below.

A summary of the second- and third-level themes is given in Table VIII.

Experiences of individuals with FASD

In two studies, individuals were interviewed regarding their experiences of living with FASD. They included 14 individuals aged 14 to 37 years and 22 individuals aged 6 to

18 years.^{13,14} From these two studies, six second-level themes were organized into two third-level themes: 'Difficulties experienced by persons with FASD' and 'Experiences by persons with FASD of how their difficulties are met by others' (Tables II and III).

Difficulties experienced by persons with FASD

Persons with FASD experience that their cognitive and mental health, as well as social difficulties regarding friends, school, and work, limit their daily life and make them feel different from others. In this second-level theme, children and young adults with FASD describe their difficulties regarding concentration, motor control, and social interactions.^{13,14} When comparing themselves to other children, children with FASD notice that other children do not possess these problems. One 8-year-old male said: 'I think differently from everyone. Everybody makes fun of me'; and one 18-year old individual argues 'They [teachers, employers] expected me to do things I couldn't. I have a hard time doing some things. Others [his age] do things easier, get through work ...'

In a study by Salmon and Buetow,¹³ young adults and adults describe their memory and emotional and social problems, as well as the hurdles they meet in school and work life. A 23-year-old male describes his memory problems as such: 'My memory blocks itself out so that everything to me is new. Every day is a new day, everything that I do is a new thing and even though I might have done it a thousand times before, it's still new. My long-term memory is the one that reminds me of the things that I've done before'. A 24-year-old individual says: 'Holding down a stable job is difficult; my attention goes very quickly. If I'm interested in something, I can hold my attention'.

Experiences among parents raising children with FASD

Sixteen studies investigating experiences among parents raising children with FASD were included. The number of participants in the included studies varied between four and 84. One study included only biological mothers, whereas the majority of studies included foster or adoptive parents. From these 16 studies, 17 second-level themes were organized into four third-level themes: 'Parents' experiences regarding their child's disabilities (Table IV)', 'Parents' experiences of parenthood in relation to FASD' (Table V), 'Parents' experiences regarding society in their role as parents to a child with FASD' (Table VI), and 'Parents experiences from respite care intervention (Table VII)'.

Parents' experiences regarding their child's disabilities

Parents of children with FASD experience their children showing various combinations of cognitive and physical disabilities. The parents reported that their children showed diminished memory and comprehension, various somatic problems, high pain tolerance, destructive behaviour, hyperactivity, and aggressiveness. In addition, parents also

described their child's lack of fear and judgement, as well as understanding of the consequences of their actions. In one study by Sanders and Buck,¹⁵ a mother explains: 'It's frustrating because you get mad telling your kid the same thing a million and one times but you know he doesn't remember'.

Parents experience various social disadvantages tied to their child's FASD

Parents reported that their children exhibit behavioural problems and difficulties with maintaining friendships, school attendance, and maintaining employments. Furthermore, the parents described their children as naïve, taken advantage of, and tending to end up in the wrong social environment. One biological mother in the study by Salmon describes it like this¹⁶: 'He's been involved in gangs, we know that. But because he doesn't understand things the way other people do, they will give him drugs to sell, and he'll sell the drugs and then spend the money himself. So we've had several instances where there have been gang hits on him, so it's either we pay the debt and free him up, or we risk having him killed'.

Parents' experiences of parenthood in relation to FASD

Parents experience a number of feelings when their child is diagnosed with FASD. Parents explained that receiving the diagnosis of FASD for their child was like a double-edged sword, whereas biological mothers felt grief and guilt for having consumed alcohol during their pregnancy. In the study by Sanders and Buck,¹⁵ one mother stated: 'I had a lot of guilt for many years ... very much depressed, knowing that my drinking caused this damage. Like I said, that just threw me over the edge. I cried a lot. I felt bad that I did this damage to Dylan. I thought what kind of a parent am I to do that to my own kid'.

In addition, when connecting their child's behaviour to exposure to alcohol during the foster period, both biological and non-biological parents expressed regrets. Parents brought up that having a diagnosis also means that the child can be labelled by the society in a judgemental way. However, receiving a diagnosis for their child also meant a great relief for the parents because it gave them an explanation for their child's behaviour.

Parents describe the importance of getting a formal diagnosis in order to get tailored help from society. Parents explained that a diagnosis is needed to receive support and to be able to address proper interventions for their child. In the study by Chamberlain et al.,¹⁷ a parent explained: 'Well they currently see a psychologist now ... she said she can tailor anything that we need to see her for. Now she's got those diagnoses she can tailor things to a more individual program for them'.

Parents express positive feelings such as joy, pride, and devotion towards their children (in spite of all difficulties). Parents feel devoted to and proud of their children, who bring them joy. Parents described that their children are gifted and talented in many ways. In the study by Chamberlain et al.,¹⁷

Table II: Difficulties experienced by individuals with fetal alcohol spectrum disorders (FASD): Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of confidence in the evidence of second-level themes within the third-level theme

Study	Review finding (second-level themes)	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Salmon and Buetow, ¹³ Stade et al. ¹⁴	Persons with FASD felt that their cognitive, mental health, as well as social difficulties regarding friends, school, and work limit their daily life and make them feel different from others	Moderate confidence	Two studies contributed to this finding, one from Canada and one from New Zealand. The reasons for rating down are minor to moderate methodological concerns and minor concerns about adequacy
Stade et al. ¹⁴	Children with FASD experience unfairness relating to the aetiology of FASD	Very low confidence	One study from Canada contributed to this finding. The reasons for rating down are moderate concerns regarding methodological and adequacy and minor concerns regarding relevance
Stade et al. ¹⁴	Children with FASD try hard to overcome the limitations of the disabilities in daily life activities	Very low confidence	One study from Canada contributed to this finding. The reasons for rating down are moderate concerns regarding methodological limitations and adequacy and minor concerns regarding relevance
Salmon and Buetow ¹³	Individuals with FASD exhibit destructive behaviour such as criminal activities and use of alcohol and drugs, as a way of coping with mental and social difficulties	Very low confidence	One study from New Zealand contributed to this finding. The reasons for rating down are moderate concerns regarding methodological limitations, serious concerns regarding adequacy, and minor concerns regarding relevance

Table III: Individuals with fetal alcohol spectrum disorders (FASD) experiences of how their difficulties are met by others. Shown with Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of the second-level theme within the third-level theme

Study	Review finding (second-level themes)	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Stade et al. ¹⁴	Children with FASD report that they get a substantial amount of support from their parents	Very low confidence	One study from Canada contributed to this finding. The reasons for rating down are moderate concerns regarding methodological limitations and adequacy, and minor concerns regarding relevance
Salmon and Buetow ¹³	Adults with FASD experience lack of support from professionals in the social services and the health care system	Very low confidence	One study from New Zealand contributed to this finding. The reasons for rating down are methodological limitations, serious concerns regarding adequacy, and minor concerns regarding relevance

an adoptive mother argues: ‘Without the behaviours, he was a very persevering, clever little boy. I would say “One day you are going to be an actor”, because he just has this canny, canny way with him. He just makes people laugh’.

Parents experience burdens such as stress and social isolation. Raising a child with FASD puts frustration and emotional stress on parents, who burn out and feel isolated. In the study by Mukherjee et al.,¹⁸ one parent stated the following: ‘We’re not doing any of the things that we thought we would be doing now when we adopted. She doesn’t get invited anywhere herself and we don’t go with her anywhere, do we?’

Parents experience that living with a child with FASD burdens the whole family. Parents report that living with a child exhibiting FASD puts stress on the whole family, including siblings and other family members, causing the family to

be socially isolated. One parent in the study by Mukherjee et al. described it this way¹⁸: ‘We divorced five years ago. It was just . . . the family just exploded, it was just, it broke down. Totally broke down’.

Parents report that they have to adapt their parenting strategies to their child’s specific needs. Parents of children with FASD report that they have to adapt their strategies according to their child’s specific needs, such as constantly having to repeat things for their child. In the study by Watson et al.,¹⁹ one parent described the importance of routines: ‘We stick to the same schedule like get up, you get dressed, you come and eat breakfast, you brush your teeth. We get your lunch in the bag, you get your jacket and that on for school, and catch the bus. Off days, by 11 o’clock it’s horrible’.

Table IV: Parents' experiences regarding their child's disabilities. Shown with Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of confidence in the evidence of the second-level theme within the third-level theme

Studies contributing to the review finding	Review finding (second-level themes)	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Sanders and Buck, ¹⁵ Salmon, ¹⁶ Chamberlain et al., ¹⁷ Watson et al., ¹⁹ Gardner, ²⁵ Granitsas, ²⁶ Whitehurst ³¹	Parents experience that their children show various combinations of cognitive and physical disabilities	Moderate confidence	Seven studies with moderately rich data contributed to this finding (2 from USA, 1 from UK, 1 from New Zealand, 1 from Australia, 2 from Canada). The reasons for rating down are moderate concerns with regard to methodological limitations and minor concerns regarding adequacy of data
Sanders and Buck, ¹⁵ Salmon, ¹⁶ Watson et al., ¹⁹ Granitsas, ²⁶ Johnston and Boyle, ²⁷ Michaud and Temple, ²⁸ Swart et al. ³⁰	Parents experience various social disadvantages tied to their child's FASD	Moderate confidence	Eight studies with rich data contributed to this finding (5 from Canada, 2 from USA, 1 from New Zealand). Reasons for rating down are minor-to-moderate concerns with regard to methodological limitations, as well as relevance

FASD, fetal alcohol spectrum disorders.

Parents express concern for their child's future and their own ability for successful lifelong parenting. Parents foresee that their children are likely to need a lifelong dependence and support, and that in the absence of proper support they might encounter difficulties with education, work, and in worst case tragic outcomes such as homelessness, drug abuse, or imprisonment. In the study by Sanders and Buck,¹⁵ a mother describes the lifelong engagement for a parent to a child with FASD like this: 'There's no saying you're 18 now they can go, or they can be home and you don't have to worry about them. You always have to worry about them and it's hard because you never know what they're going to do. They could end up with a gang, they could get hurt easily drinking and being in a crowd. They' so easily led astray... Yeah, it's a lifetime of worrying about them'.

Parents report that they have to advocate for their children in multiple ways. Parents experience that they have to speak for their children and constantly monitor their rights to services and support towards the school system and social care. One adoptive mum in the study by Chamberlain et al. explained¹⁷: 'It was my lack of qualifications. Yeah I am an annoying parent I guess because I would advocate. Even before we started at the school I was giving them the information...but it's kind of like [school saying] "this is how we've done it for the last however many years at the school so this is how we will continue to do it"'

Parents' experiences regarding society in their role as parents to a child with FASD

Parents striving for a diagnosis experience this as a struggle. Parents whose children have been diagnosed with FASD experienced that they had to struggle to receive this diagnosis. The process of obtaining a diagnosis was

frustrating and stressful. The parents also described the importance of acquiring a diagnosis in order to be able to receive any support from the society. In the study by Watson et al.,²⁰ a foster mother of two sons with FASD states: 'It was a lot of work. It shouldn't be that hard. We need a better diagnostic process ... if you don't have the [facial] features, good luck'.

Parents report lack of understanding and knowledge about FASD among professionals and feel blamed for bad parenting. Parents feel that doctors, social workers, and educators lack knowledge about FASD. Parents also experience that there is little or no knowledge of FASD among the police. Instead, they feel blamed for their children's behaviour on inappropriate parenting. In the study by Ryan and Ferguson,²¹ a parent described the following: 'We are on our own. No one understands our situation. In fact, we feel like they blame us for Alan's behaviour. We call [the social service agency] and they say that there are no services for kids with fetal alcohol syndrome'.

Parents stress the importance of support to children with FASD and their parents from professionals within health care, social care, and school. Parents experienced lack of support from professionals and appreciate the times when support is provided. In the study by Sanders and Buck,¹⁵ a mother described her daughter's situation at school: 'She had great support through school, terrific support ... I felt kind of guilty because sometimes you see kids and they are struggling and they're not getting that support. Her siblings never got it either because nobody fought for them ...'

Experiences of interventions for parents raising children with FASD

Only one of the included studies investigated the experiences of an intervention aimed for parents raising children

Table V: Parents' experiences of parenthood in relation to fetal alcohol spectrum disorders (FASD). Shown with Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of the second-level theme within the third-level theme

Studies contributing to the review finding	Review finding (second-level themes)	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Sanders and Buck, ¹⁵ Salmon, ¹⁶ Whitehurst ³¹	Parents experience a number of feelings when their child is diagnosed with FASD	Moderate confidence	Three studies contributed to this finding (1 from Canada, 1 from New Zealand, 1 from UK). The reasons for rating down are moderate concerns regarding methodological limitations and adequacy of data
Salmon, ¹⁶ Chamberlain et al., ¹⁷ Watson et al., ²⁰ Johnston and Boyle ²⁷	Parents describe the importance of getting a formal diagnosis in order to get tailored help from society	Moderate confidence	Four studies contributed to this finding (2 from Canada, 1 from New Zealand, 1 from Australia). The reasons for rating down are moderate concerns regarding methodological limitations and minor concerns regarding adequacy of data
Sanders and Buck, ¹⁵ Chamberlain et al., ¹⁷ Ryan and Ferguson, ²¹ Granitsas, ²⁶ Michaud and Temple ²⁸	Parents express positive feelings such as joy, pride, and devotion to their children (in spite of all difficulties)	Moderate confidence	Five studies contributed to this finding (2 from Canada, 2 from USA, 1 from Australia). The reasons for rating down are moderate concerns with regard to methodological limitations and adequacy of data
Mukherjee et al., ¹⁸ Michaud and Temple, ²⁸ Swart et al., ³⁰ Whitehurst ³¹	Parents experience burdens such as stress and social isolation	Moderate confidence	Four studies contributed to this finding (2 from UK, 2 from Canada). The reasons for rating down are minor to moderate concerns with regard to methodological limitations and adequacy of data
Sanders and Buck, ¹⁵ Mukherjee et al., ¹⁸ Ryan and Ferguson, ²¹ Whitehurst ³¹	Parents experience that living with a FASD child burdens the whole family	Moderate confidence	Four studies contributed to this finding (2 from UK, 1 from USA, 1 from Canada). The reasons for rating down are moderate concerns with regard to methodological limitations and relevance and minor concerns regarding adequacy
Sanders and Buck, ¹⁵ Mukherjee et al., ¹⁸ Watson et al., ²⁰ Ryan and Ferguson, ²¹ Coons et al., ²⁴ Gardner, ²⁵ Michaud and Temple ²⁸	Parents experience that they have to adapt their parenting strategies to their child's specific needs	Moderate confidence	Seven studies contributed to this finding (4 from Canada, 1 from UK, 2 from USA). The reasons for rating down are moderate concerns with regard to methodological limitations
Sanders and Buck, ¹⁵ Chamberlain et al., ¹⁷ Mukherjee et al., ¹⁸ Watson et al., ¹⁹ Gardner, ²⁵ Granitsas, ²⁶ Michaud and Temple ²⁸	Parents express concern for their child's future and their own ability for a successful lifelong parenting	Moderate confidence	Seven studies contributed to this finding (3 from Canada, 2 from USA, 1 from Australia, 1 from UK). The reasons for rating down are moderate concerns with regard to methodological limitations and minor concerns regarding relevance
Chamberlain et al., ¹⁷ Mukherjee et al., ¹⁸ Watson et al., ²⁰ Coons et al. ²⁴	Parents experience that they have to advocate for their children in multiple ways	Moderate confidence	Four studies contributed to this finding (2 from Canada, 1 from UK, 1 from Australia). The reasons for rating down are moderate concerns with regard to methodological limitations and minor concerns regarding relevance and adequacy

with FASD (Table VII and Appendix S3). This study, conducted in Canada, investigated respite care and the theme is therefore 'Parents' experiences from respite care intervention'.²²

DISCUSSION

This systematic review synthesizes qualitative data regarding the experiences of individuals living with FASD or their parents. It should be noted that our aim was to stay close to the descriptive study data and not to build

hypotheses, to describe possible causation, or convey to theory development.

This review identified six third-level themes and 23 second-level themes. For each of the second-level themes we assessed the confidence using the GRADE-CERQual approach, where the weighted result of qualitative data was assessed taking factors such as methodological limitations, relevance, coherence, and adequacy of data into consideration. A low level of confidence does not imply that the finding is irrelevant but points out that future research

Table VI: Parents' experiences regarding society in their role as parents to a child with fetal alcohol spectrum disorders (FASD). Shown with Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of the second-level theme within the third-level theme

Study	Review finding (second-level theme)	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Sanders and Buck, ¹⁵ Mukherjee et al., ¹⁸ Ryan and Ferguson, ²¹ Granitsas, ²⁶ Whitehurst ³¹	When taking a child on board, adoptive and foster parents felt that they were not given sufficient information by social workers or health care that their child had FAS/FASD, or its potential impact	Low confidence	Five studies contributed to this finding (1 from Canada, 2 from UK, 2 from USA). This finding was graded as low confidence because of moderate concerns regarding methodological limitations and serious concerns regarding relevance
Salmon ¹⁶	Biological mothers feel uninformed, blamed, not listened to, and not taken seriously	Very low confidence	One study from New Zealand contributed to this finding. This finding was graded as very low certainty because of moderate concerns regarding methodological limitations, adequacy, and relevance
Johnston and Boyle ²⁷	Biological parents recognize that FASD is related to socio-cultural patterns	Very low confidence	One study from Canada contributed to this finding. This finding was graded as very low certainty because of moderate concerns regarding relevance and serious concerns regarding adequacy of data
Sanders and Buck, ¹⁵ Salmon, ¹⁶ Chamberlain et al., ¹⁷ Watson et al. ¹⁹	Parents striving for a diagnosis experience this as a struggle	Moderate confidence	Four studies contributed to this finding (2 from Canada, 1 from New Zealand). The reasons for rating down are moderate concerns with regard to methodological limitations and minor concerns regarding adequacy
Salmon, ¹⁶ Chamberlain et al., ¹⁷ Mukherjee et al., ¹⁸ Ryan and Ferguson, ²¹ Coons et al., ²⁴ Swart et al., ³⁰ Whitehurst ³¹	Parents experience lack of understanding and knowledge about FASD among professionals and feel blamed for bad parenting	Moderate confidence	Seven studies contributed to this finding (2 from UK, 2 from Canada, 1 from New Zealand, 1 from USA, 1 from Australia). The reasons for rating down are moderate concerns with regard to methodological limitations and no to minor concerns regarding adequacy and relevance
Sanders and Buck, ¹⁵ Salmon, ¹⁶ Ryan and Ferguson, ²¹ Coons et al., ²⁴ Poth et al., ²⁹ Whitehurst ³¹	Parents stress the importance of support for children with FASD and their parents from professionals within health care, social care, and school	Moderate confidence	Six studies contributed to this finding (3 from Canada, 1 from New Zealand, 1 from UK, 1 from USA). The reasons for rating down are moderate concerns with regard to methodological limitations and minor concerns regarding adequacy

FAS, fetal alcohol syndrome.

may or may not support and strengthen the confidence in this theme. The major reason for considering a theme to be of lower confidence was that there were not enough data to support the finding or that there were concerns regarding quality or relevance of the included studies. Relevance refers to what extent the primary studies represent parts of the phenomenon of interest in terms of, for example, population or context. A result that is based on a specific population or context may not reflect the general perspective, leading to uncertainty of the finding. For findings that were rated down for relevance, assessments were conducted in such a manner that the specific findings could be sensitive to societal structures in the countries where the studies were performed. Interestingly, similar experiences were expressed across several different contexts and there were no indications that the experiences would be culturally dependent.

The results based on themes with moderate risk of bias demonstrate that individuals with FASD show a variance

in the disabilities that they experience, ranging in the included studies from somatic and cognitive problems, high pain tolerance, destructive behaviour, hyperactivity, and aggressiveness to social problems associated with friendship, school attendance, and maintenance of employment. Furthermore, parents also describe that their children are naïve and can be taken advantage of, which can lead to the development of destructive behaviour and criminal activities. Parents describe their parenting as being a lifelong engagement in the sense that they need to form a protective environment around their child for whom they constantly need to advocate. Although the parents are proud of their children and describe that they have great talents and bring their families joy, they also feel that the whole family is isolated and burdened.

The fact that persons with FASD have been exposed to alcohol prenatally makes the disorder complex. Many parents describe that there is a lack of knowledge among professionals in health care, social care, and the school system

Table VII: Parents' experiences of respite care intervention. Shown with Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of the second-level theme within the third-level theme

Study	Review finding	CERQual assessment of confidence in the evidence	Explanation
Doig et al. ²²	Parents experience respite care as beneficial for the family, but have seen recurrent difficulties in receiving it and also a deficiency in system coordination	Very low confidence	One study with serious concerns regarding adequacy and moderate concerns regarding relevance

Table VIII: Summary of the Confidence in the Evidence from Reviews of Qualitative Research (CERQual) assessment of the second-level themes within the different third-level themes

Experiences of individuals with FASD or of parents of children with FASD

Second-level themes	Third-level themes
Persons with FASD report that their cognitive and mental health, as well as social difficulties regarding friends, school, and work, limit their daily lives and make them feel different from others	Difficulties experienced by persons with FASD
Children with FASD experience unfairness relating to the aetiology of FASD. The children understand the cause of FASD and that it is preventable Children with FASD try hard to overcome the limitations of the disabilities in daily life activities	
Persons with FASD exhibit destructive behaviour such as criminal activities and use of alcohol and drugs, as a way of coping with mental and social difficulties	Experiences by individuals with FASD of how their difficulties are met by others
Children with FASD report that they get a substantial amount of support from their parents	
Adults with FASD experience lack of support from professionals in the social services and the health care system	Parents' experiences regarding their child's disabilities
Parents experience that their children show various combinations of cognitive and physical disabilities	
Parents experience various social disadvantages tied to their child's FASD	Parents' experiences of parenthood in relation to FASD
Parents experience a variety of feelings when their child is diagnosed with FASD	
Parents describe the importance of getting a formal diagnosis in order to get tailored help from society	Parents' experiences regarding society in their role as parents to a child with FASD
Parents express positive feelings such as joy, pride, and devotion to their children (despite all difficulties)	
Parents experience burdens such as stress and social isolation	Parents' experiences from respite care intervention
Parents report that living with a child with FASD burdens the whole family	
Parents report that they have to adapt their parenting strategies to their child's specific needs	Parent's experiences regarding society in their role as parents to a child with FASD
Parents express concern for their child's future and their own ability for successful expected lifelong parenting	
Parents report that they have to advocate for their children in multiple ways	Parent's experiences from respite care intervention
When taking a child on board, adoptive and foster parents felt that they were not given sufficient information by social workers or health care that their child had FAS/FASD, or its potential impact	
Biological mothers feel uninformed, blamed, not listened to, and not taken seriously	Parent's experiences from respite care intervention
Biological parents recognize that FASD is related to socio-cultural patterns	
Parents striving for a diagnosis experience this as a struggle	Parent's experiences from respite care intervention
Parents experience lack of understanding and knowledge about FASD among professionals and feel blamed for bad parenting	
Parents stress the importance of support to children with FASD and their parents from professionals within health care, social care, and school	Parent's experiences from respite care intervention
Parents experience respite care as beneficial for the family but have seen recurrent difficulties in receiving it and also a deficiency in system coordination	

The confidence in the evidence for the themes is highlighted as: light grey for moderate confidence, grey for low confidence, and dark grey for very low confidence in the evidence. FASD, fetal alcohol spectrum disorders; FAS, fetal alcohol syndrome.

regarding FASD, causing the families and children to feel that it is a hurdle to receive a diagnosis and to receive support from society. These experiences may indicate that FASD are not always easily identifiable to outsiders and

therefore not recognized. In addition, receiving a diagnosis may cause guilt for the child and their families. Parents whose child has received a diagnosis recall that this provides them with a mixed set of feelings. Apart from feelings

of guilt, there is also a perceived advantage to understand the reason for their child's behaviour, whereby parents experience that a diagnosis of a FASD-related condition opens up pathways for appropriate support. Regardless of diagnosis, individuals with FASD and their parents need to be treated in a professional manner and provided with appropriate care and support from the health care and social services, as well as proper support during schooling. It is therefore of great importance that health care, social care, and the school system hold competence about FASD in order to support their professionals in their care of these individuals and their families. Parents who have received support stress the importance of obtaining support from health care, social care, and educational systems.

Although there is very low confidence in the theme 'Parents' experiences from respite care', owing to the fact that it is only reported in one study with few parents, it still indicates that respite care could be beneficial for the parents. It is, however, worth noting that there is a lack of studies investigating the experiences of interventions for individuals with FASD or for their families. The need for studies on the effect of interventions on measurable effects for individuals with FASD has been concluded in a systematic review by Reid et al.²³ The lack of studies on interventions for these individuals could be a result of the fact that these individuals do not receive any interventions.

Although more studies are needed, in particular regarding experiences of interventions for individuals with FASD or their families and regarding experiences from biological parents, it is gratifying to see that current qualitative studies with scientific rigour have been performed. The results of this qualitative evidence synthesis enhances the picture that living with FASD, independent of cultural and societal contexts, is experienced as a lifelong struggle.

Limitations

FASD is an umbrella term for several different conditions, which while manifesting some similarities, can also exhibit considerable differences, thus resulting in FASD group heterogeneity. In this review, we have looked at experiences of FASD as a group and have not been able to consider possible differences or similarities among the different subgroups, mainly owing to the lack of reporting on the different subgroups. Furthermore, there might be some additional literature, for example doctoral theses, that

have not been published in peer-reviewed journals and are therefore not considered for this review.

The themes in this systematic review were developed through an iterative process of analysis where the themes were phrased so as to have no or minor concerns regarding coherence. Although this may cause a risk of developing findings that can seem banal, the themes reflect important experiences presented in the included studies.

CONCLUSIONS/IMPLICATIONS

The findings from this review suggest that individuals affected by FASD feel there is a lack of knowledge or understanding among the professionals they encounter in health care, social care, and education services. These professionals may benefit from more information related to FASD, in order to better understand the requirements of individuals and families living with FASD, which should aid in the establishment of the necessary support that should be provided. Individuals with FASD and their parents need to be acknowledged, accepted, and supported by society.

Until now, most research has focused on parents' experiences of living with a child with FASD. Future research should also include the perspective of those individuals living with FASD. Furthermore, very little is known about experiences and expectations of supportive interventions for individuals with FASD and their families.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Search strategy.

Appendix S2: Methodological limitations of the included studies.

Appendix S3: Presentation of themes and GRADE-CERQual Evidence Profiles.

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