

The experiences of caregivers looking after individuals with fetal alcohol spectrum disorder

A rapid synthesis of qualitative studies

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1 Introduction

Diagnostic criteria for fetal alcohol syndrome (FAS), the most severe end of a spectrum characterised by physical, neurodevelopmental and behavioural impairments, were first defined in 1973 to account for biological anomalies associated with consumption of alcohol during pregnancy.¹ Over time, other definitions have been introduced to attempt to provide better descriptions of a range of outcomes manifesting in clinically diverse presentations. Such terms have included fetal alcohol effects,² alcohol-related biological defects, alcohol-related neurodevelopment disorder and partial fetal alcohol syndrome.³ This wider constellation of effects, along with FAS, constitutes the continuum of structural anomalies and neurocognitive and behavioural disabilities associated with prenatal exposure to alcohol which is termed fetal alcohol spectrum disorder (FASD).

There are significant challenges for caregivers of individuals with FASD due to the wide range of ways the condition can affect individuals. Research has noted that parents and carers may struggle to cope with the significant demands of supporting children, particularly in the absence of an accurate diagnosis to explain the child's behaviour.⁴⁻⁶ The consistent requirement for supervision and support can have a deleterious effect on carers' well-being and family functioning.⁷

Studies have suggested that type and volume of stress associated with raising a child with FASD may be different from that of other developmental disabilities.^{8,9} Parents of children with FASD, for example, report more difficult behaviours and expressed more concerns regarding their children's future abilities to become self sufficient compared with parents of children with autism spectrum disorder.⁹

It has been suggested that both family and child characteristics contribute to the experience of stress for families of children with FASD.^{10,11} Beyond economic resources, researchers have found that having fewer personal resources to advocate for and support a child with FASD predicted higher stress in mothers from low socioeconomic status families.¹¹

Studies have suggested that a number of factors play an important part in determining the success of raising children with FASD. These include involving the extended family, access to relief for caregivers, collaborating with the school to establish support and consistency for their child, receiving respect and help from professionals, being generally informed about FASD and what the condition entails, and developing a personal network of support with other parents of children with FASD.^{12,13}

There is uncertainty about whether the underlying cause of developmental impairments is the prenatal alcohol exposure or the impact of a detrimental social environment. In many cases, these increase the likelihood that such children will be identified at risk by social services and placed into care. There is evidence that up to 75% of children in the foster care system have a family history of mental illness or substance abuse¹⁴ with estimates of the prevalence of FASD among children in care in the region of 78-80%.^{3,15,16}

To mitigate such risks, the careful and comprehensive assessment and description of a child's impairments at time of entry into the foster system will increase the likelihood of appropriate placements and allow foster or adoptive carers the opportunity to prepare for the specific challenges associated with each child's care. Within this context, the value of early assessment and diagnosis is particularly important.¹⁷

Despite the range of challenges faced by caregivers of children with FASD, foster parents have reported the impact of positive experiences on their motivation for maintaining and improving care, for example observing the child overcome problems and sharing their personal achievements, and

the desire to improve the child's life and future prospects.^{13,18} Other studies have emphasised the potential fulfilment gained by carers when raising children affected by FASD and have highlighted the role played by children in teaching parents important life lessons and enjoying the unique gifts of each individual child.^{10,19}

This qualitative synthesis explores caregivers' experiences of supporting individuals with FASD. It was conducted using a rapid review methodology in order to allow publication alongside national guidance on screening, identification and diagnosis of children and young people exposed prenatally to alcohol, which has also been developed by Healthcare Improvement Scotland (HIS) www.sign.ac.uk/sign-156-children-and-young-people-exposed-prenatally-to-alcohol.html.

The synthesis aims to describe the impact of the condition for caregiving from the experiences across a range of caregiver and family contexts and how this helps to inform the provision of quality of care and services.

2 Methods

This report synthesises primary qualitative studies which have used methods, including interviews, focus groups and participatory action research (PAR), to ask carers (biological or adoptive parents, foster parents, kinship carers or others) about their experiences of supporting an individual with FASD.

2.1 Literature search

A number of techniques can be used to identify qualitative literature, these include the use of qualitative filters within conventional databases, 'snowballing' or citation chaining, browsing of core journals, and the knowledge of colleagues or field experts as sources of information.^{20,21}

A qualitative literature search was carried out by an information specialist in December 2017, using a pragmatic approach to identify as much relevant English-language literature as possible within a short time span. Three techniques were used: formal literature searching, snowballing, and experts from the HIS guidance development group were invited to provide examples of further relevant literature in early 2017. Papers identified in the experts' list were used to generate citation chaining in Google Scholar to augment formal database searching and also checked against the literature search results.

The search methodology combined both the use of a published qualitative search filter with general key search terms for FASD (*FASD, fetal alcohol spectrum disorder, prenatal alcohol exposure*) to identify studies relating to the impact of FASD on carers. No date limits were applied. The following databases from the Scottish Knowledge Network were used:

- ASSIA
- CINAHL
- Embase
- Education Resources Information Center (ERIC)
- MEDLINE
- Midwives Information and Resource Service (MIDIRS)
- Proquest public health
- Proquest social abstracts
- PsycINFO
- Psychology and Behavioural Sciences
- Web of Science

The search filter used to identify potential qualitative studies was applied to each database.²²

Alongside the terms for FASD to help identify studies, the snowballing method included the following terms: *parent, carer, family, adopt, foster, kinship, relation, impact, qualitative and experience*. Results were scanned by the information specialist and all potentially relevant items were added to the Google Scholar results.

The reference lists of included studies were not reviewed due to the rapid nature of the review.

2.2 Selection of the literature

2.2.1 Inclusion criteria

Primary studies were included in the synthesis if they:

- included carers (biological or adoptive parents, foster parents, kinship carers or others) of individuals with FASD
- explored the impact on carers of supporting an individual with FASD
- used a qualitative study design (for example, ethnographic research, phenomenology, studies utilising focus group, and/or interview methods). Studies using mixed methods were only included if the qualitative element was reported separately to the quantitative findings
- were published in a peer-reviewed journal.

2.2.2 Exclusion criteria

Primary studies were excluded from the synthesis if they:

- used quantitative methods to gather qualitative data, for example qualitative data derived from a largely quantitative questionnaire. Such data lack the conceptual depth to contribute to a qualitative synthesis²³
- were published in a language other than English
- referred to the design or evaluation of an intervention or a service for individuals with FASD
- were not published in a peer-reviewed journal.

2.3 Selection of studies

One author reviewed all identified records by title and abstract to evaluate their eligibility. This initial sift was undertaken to exclude papers that were clearly unrelated to the aim of the synthesis. The full texts of all potentially relevant papers were obtained and assessed for eligibility by one author. A full quality appraisal of the identified papers was conducted independently by two authors. Disagreements between authors were resolved by discussion.

See Appendix 2 for a summary of eligible studies (included and excluded).

2.4 Quality assessment

The inclusion criterion of studies needing to use appropriate qualitative data collection and analysis methods was used as an initial quality assessment attribute for the included studies. In addition, further quality appraisal of each eligible study was carried out. The selection of methods for quality appraisal was informed by the guidance laid out by the Cochrane Qualitative Research Methods Group.²⁴

Appraisal was performed using the Critical Appraisal Skills Programme (CASP)²⁵ quality assessment tool for qualitative studies. Two authors independently applied the CASP tool to each eligible study. A quality level of low, moderate or high (based on the Swedish Council on Health Technology Assessment's handbook for qualitative evaluation and synthesis)²⁶ was jointly applied to each study guided by the independent CASP ratings. Disagreements were resolved by discussion. See Appendix 3 for details on the quality assessment of the included and excluded studies.

There is currently no consensus among qualitative researchers on how quality criteria should be applied and how they should be used in qualitative syntheses.²⁷ It has been noted that studies considered 'low quality' due to methodological flaws may still provide valuable insights arising from the data and, conversely, methodologically strong studies may suffer from poor or limited interpretation of their data, leading to limited insights into the phenomena under investigation.^{24,28} It has further been acknowledged that poor reporting of methods does not equate with poorly-conducted research.^{24,29} Despite lacking a clear description of the analysis, three studies³⁰⁻³² were included because they still made a valuable contribution to the synthesis (*see Appendix 3 for further details*). Two low-quality studies^{33,34} were excluded from this synthesis because they did not have a clear statement of findings or were not adding any substantial findings beyond the research findings of the other included studies (*see Appendix 3*).

2.5 Data extraction

A data extraction table was populated. Information extracted included author and year of publication, study sample characteristics, number and age of participants, method of data collection, analysis approach and study setting (*see Appendix 2*).

The authors' interpretations of the included studies represent the data analysed in this synthesis.²⁷ All text labelled as discussion or results was thematically analysed and synthesised in themes (*see section 4*). The approach of analysing both the discussion and result section of the studies is thought to increase the transparency and 'auditability' of the synthesis.³⁵

QSR NVivo® 10 software for qualitative analysis was used to manage and analyse the data thematically. The results of all included studies were entered verbatim into NVivo® for analysis.

2.6 Data synthesis

Framework analysis³⁶ was used to allow the identification of common and variable patterns of themes within and across different studies. Framework analysis is a matrix-based method which involves the construction of thematic categories into which data from primary qualitative studies can be coded.³⁶ Framework synthesis has been widely utilised in the synthesis of qualitative health data and is one of the approaches to qualitative synthesis recommended by the Cochrane Qualitative Review Methods Group.²⁴ This approach involves several overlapping stages: familiarisation, identifying a thematic framework, indexing, charting and interpretation. However, not all stages were carried out in the current synthesis due to a limited timescale for conducting the synthesis and reduced research capacity.

- **Familiarisation** – Two authors read all of the included studies, with reference to the review questions.
- **Identifying a thematic framework** – In this stage, one author examined all the findings from these papers to identify the initial themes for the thematic framework. The four overarching themes which were identified arose consistently across the findings of multiple studies or were identified by one or more study as a key theme (*see section 3.2*).
- **Indexing** – One author systematically applied the thematic framework to the results and discussion section of all studies. The included papers were imported into NVivo® and the results and discussion sections were coded. The stages were iterative and additional themes which emerged at this stage further informed the thematic framework.
- **Charting** – This stages includes the creation of a matrix of findings summaries for each theme by study. Given the rapid approach of the synthesis this stage was not conducted.
- **Interpretation** – Refinement of the subthemes and search for patterns and connection between themes was performed by two authors.

2.7 Review and consultation

The discussion section of the synthesis report was presented at a Kinship Carers group meeting in Midlothian, Scotland to gather carers' feedback based on their own experiences of looking after an individual with FASD in the Scottish context. The carers' responses are summarised in Appendix 4. The final synthesis report was reviewed by the Lead Health Services Researcher and underwent editorial review by senior management from Healthcare Improvement Scotland.

3 Results

6,145 studies were identified through electronic database searching. Of these studies, 30 were selected for full text screening and 12 were subsequently included in the synthesis. See Appendix 1 for a figure illustrating the screening process.

3.1 Description of studies

3.1.1 Design

All studies used qualitative methods of data collection and analysis. Four of the included studies conducted semi-structured interviews as a method of data collection,³⁷⁻⁴⁰ six conducted unstructured interviews,^{19,30,32,41-43} one used participatory action research³¹ and one conducted focus groups.⁴⁴ The methods of data analysis used by the studies were: interpretative phenomenological analysis,^{32,37-41} phenomenological-hermeneutic approach,¹⁹ thematic analysis,^{19,44} narrative analysis,⁴³ and constant comparison.⁴² Two studies did not provide information about their approach to data analysis.^{30,31}

3.1.2 Sample sizes

Study sample sizes ranged from 4³² to 84 participants.³⁷

3.1.3 Setting and dates of studies

The studies were published between 2001 and 2016 and were conducted in four countries: Canada,^{19,30,37,39,40} USA,^{41,43} UK^{32,44} and New Zealand.⁴²

3.1.4 Participants

Four studies^{19,30,32,44} had a mixed sample of biological, adoptive and/or foster parents. In three studies^{37,39,40} participants included biological parents, foster parents, adoptive parents, step-parents, and custodial grandparents. Three studies^{38,41,43} included only foster and/or adoptive parents and two studies^{31,42} included only biological parents.

The age range of participants and their children varied across studies.

3.1.5 Study quality

Five of the included studies were rated as high quality,^{37,38,40,41,43} four as moderate quality,^{19,39,42,44} and three studies were difficult to appraise due to reporting issues.³⁰⁻³² See Appendix 3 for more details about the quality assessment of the included studies.

3.2 Results of the framework synthesis

Analysis of the twelve included studies identified four themes, three of which included subthemes (see Table 1).

Table 1. Themes and subthemes

Theme	Subtheme
Theme 1: Living and coping with FASD	1.1 Impact on carers' well-being as a result of caring for a child with FASD
	1.2 Positive caregiving experiences
	1.3 Concerns about and hope for the future
	1.4 Parenting strategies
Theme 2: Stigma and blame	
Theme 3: Diagnosis	3.1 Difficulties in obtaining diagnosis
	3.2 Formal diagnosis and awareness can help
Theme 4: Support	4.1 Lack of professionals' knowledge and appropriate support
	4.2 Formal and informal support can help

4 Synthesised themes

Studies identified various different caregiving experiences associated with looking after an individual with FASD. Although individual studies reported that parenting a child with FASD is a unique experience due to the various developmental difficulties that manifest during the child's life,^{19,40,44} caregivers shared similar experiences across studies which were synthesised in the four themes described in sections 4.1 to 4.4 (see also Table 1).

4.1 Living and coping with FASD

4.1.1 Impact on carers' well-being as a result of caring for a child with FASD

"It's probably like Post Traumatic Stress Syndrome, to be honest because you dread the police coming to the door, you dread the phone ringing, you absolutely dread it, well, nobody rings us up anymore because they know that we didn't used to answer the phone and it's like it's left us with that and I remember the police coming to my door one day and I'm thinking, and I looked at them and I said is he still alive? What's happened?... but it was like that all the time, there was always this dread and fear inside of what is going to happen next (biological mother)." [Whitehurst 2011, p.191]

Studies revealed that caring for an individual with FASD can have an impact on carers' well-being. Stress was identified as a common experience of caregivers^{19,30-32,39,42,44} particularly when parents had to cope with children's externalising behaviour problems, such as hyperactivity, aggression or violence.^{19,42,43} Studies highlighted that caregivers' stress could be caused by constantly fighting with the child about accepting support because of the child's inability to accept their own limitations.^{31,32} The need for constant supervision for their children was seen by some adoptive and foster parents as emotionally demanding.⁴³ Several parents explained that a further cause for concern for them is having to cope with multiple disabilities in their children in addition to FASD.^{19,41} Factors explored under other themes, such as poor or incorrect support from school and health professionals,^{19,42,44} or missing knowledge and lack of training about FASD diagnosis,³¹ can further contribute to caregivers' stress. In one study, biological, adoptive and foster parents referred to their experience as a 'continual battle'.³²

Several studies identified that caring for an individual with FASD can have a negative effect on the overall family functioning.^{19,37,44} In one study biological families indicated that they were living in constant crisis, struggling to move from adjustment to adaptation.⁴² For some families, having a child with FASD can have a significant impact on family members' mental health and relationships.^{19,37,44} The challenging behaviour that children with FASD demonstrate can also create a chaotic home environment where *"it's very hard to make a family seem like a family"* [Sanders 2010, p. 314]. Some parents spoke about their difficulties balancing their parenting responsibilities with other family demands, such as caring for an ageing family members or a sick spouse.³⁷ The challenges in looking after an individual with FASD, which can be increased by the lack of support, led, in some cases, to marital breakdown.^{43,44}

4.1.2 Positive caregiving experiences

"I think I've been blessed that I have these two girls in my life; they are very special in their own way. I don't know what I would do without them if they weren't in my life. It was a long journey! But it was worth it... I could be helping them the rest of their lives... They probably remember all the things I helped them on and what I did for them. I think they will appreciate all things I did. I give myself a pat on the back!" [Sanders & Buck, 2010, p. 321]

Multiple studies described that caregivers' experiences can change over time when they start appreciating their children's differences and uniqueness.^{19,31,37,40,41,44} Three studies mentioned biological parents' positive outlook of raising a child with FASD,^{31,37,44} with one mother sharing the experience of staying sober in order to be able to look after her child.³⁷ Two studies indicated that adoptive parents increasingly appreciate their children's accomplishments, gifts and skills over time.^{19,41} Another two studies highlighted biological and non-biological caregivers' experiences of personal growth and a 'changed worldview' as a result of looking after a child with FASD.^{19,37} One study highlighted that both biological and non-biological parents try to focus on the positive contributions that their children bring to their lives and often described their lives as an 'adventure'.⁴²

4.1.3 Concerns about and hope for the future

"I'm just kind of afraid of the future... I try not to think about the future." [Watson 2013, p.81]

A number of studies described caregivers' concerns about their children's future.^{19,31,39,41} One study identified that for some carers letting go of their hopes for the child's future was described as an experience of grief.³⁹ Several studies discussed caregivers' concerns in relation to their children's ability to live independently as an adult.^{12,20,22} The risk of teenage pregnancy,³⁹ as well as the lack of support services when children with FASD transition into adulthood^{12,31,39,41} were identified by both biological and non-biological caregivers as possible barriers for independent living. Anticipated difficulties varied depending on whether the child is transitioning into primary or secondary school, or into adulthood.⁴⁴ One study showed that concerns for the child's future were also influenced by the lack of information and diagnosis that caregivers receive from professionals.⁴¹

Caregivers were also able to describe hope for the future of their children.^{19,31,39} One study described the hope of an adoptive mother who stated: *"I think she'll be pretty independent. I think she'll be... I think, like I said, once she's got it she's got it. And I think she'll, she's going to be okay. I think she'll be okay"* [Watson 2013, p. 85]. Several studies identified that parents had hopes for their children achieving independence.^{19,31,39} The experience of being hopeful for the future for some caregivers was related to external factors such as the child finding the right partner or receiving adequate support.^{31,39}

4.1.4 Parenting strategies

"It's taken us a while to adapt to having these kids...so now that we're on a routine and understand that kids need that routine, it's easier for us now as well... If you have everything as a routine, it's very simple." [Coons et al. 2016, p. 156]

Studies described caregivers using a range of parenting strategies for their children with FASD. Both biological and adoptive carers found parenting a child with FASD to be a very different experience from parenting other children.^{31,43} Two studies suggested that for some biological and non-biological carers it is important to set realistic expectations for their children in light of their deficits.^{19,39} One study found that many carers are adapting to specific challenges on a day-to-day basis and that this adaptation is influenced by various different factors (for example, age of the child, family type, etc).³⁷ The study also indicated that both biological and non-biological carers preferred to 'pick their battles' with their children which, in some cases, led to improved relationships. For example, arguing about daily issues was not considered as important as dealing with serious behaviour problems, such as aggression or depression.^{37,38} Parents, furthermore, considered the importance of maintaining a routine and consistency to aid the challenges of everyday life.³⁷ Maintaining a routine in the home, as well as using visual prompts and signs was seen by parents as a way to co-ordinate their children's day and to prevent children's challenging behaviour. Two studies indicated that some parents find altering the home environment to be a more effective strategy in preventing feelings of overstimulation than prescribing consequences for behaviour or trying to change behaviour.^{31,37} Foster and adoptive

parents, as well as one biological mother, elaborated on their need to protect and even ‘control’ their children’s environments in order to keep them away from harm.^{19,43} Biological mothers in another study, however, identified that providing supervision which is balanced with an unconditional acceptance is a necessary condition for children to stay home.³¹

Biological and non-biological caregivers highlighted the importance of obtaining information and knowledge about FASD in order to better understand their child’s behaviour and to adopt appropriate parenting strategies.^{19,37,38,43} Behavioural strategies such as following up bad behaviour with consequences were seen as ineffective because of challenges in learning and memory associated with FASD.^{19,32} Two studies identified that parenting strategies and coping improved as parents learned about the impact of FASD on the behaviour and development of children.^{19,38} Acquiring new skills or support was recognised by parents as important in coping and adapting to their children’s behaviour.³⁷ The ability to effectively look after children with FASD was considered by adoptive and foster parents as something that develops over time and with experience.⁴³

In addition to having to educate themselves about FASD, parents also identified having to take on a role of advocate for their children.^{19,37,40} Parents described this advocating role as a ‘fight’, especially when having to argue in favour of support services or adequate support at school.^{19,37}

4.2 Stigma and blame

“It’s very hard to forgive yourself when you’ve actually damaged someone’s life, irrevocably, you know, you can’t turn the clock back... I’ve said sorry to him so many times and said I really, really wouldn’t have wanted this life for you and I really am so sorry... but all I can do is be there for you to help you get through it, you know.” [Whitehurst 2011, p.191]

Guilt for drinking during pregnancy was identified as common experience for biological mothers.^{19,31,32,40,42} Mothers also felt guilty for assuming that their children were purposefully misbehaving and felt that they were ‘mishandling’ situations by not using appropriate parenting strategies.¹⁹ In one study the experience of guilt was related to the inability to meet the complex needs of children with FASD.³¹ Some biological mothers also shared the experience of having to ask their children to leave home when their behaviour became unmanageable.³¹ The mothers expressed a sense of failure for not being able to continue caring for their children.³¹ One study reported a feeling of guilt from an adoptive mother who shared: *“So when they say that biological moms who are raising these kids feel guilty, the adopted moms feel just as guilty because you want to do so much”* [Morrissette 2001, p.11].

Several studies indicated that carers of individuals with FASD feel isolated and judged by the public and professionals.^{32,37,42,44} Biological and non-biological carers often felt criticised by community members and professionals for their ‘poor parenting skills’ because of the assumption that child’s behaviour is caused by poor parenting.^{37,44} Some non-biological carers expressed feelings of anger or judgement toward biological parents for their drinking during pregnancy.^{19,38}

In one study adoptive parents reported feelings of uneasiness and embarrassment to have to explain themselves when people make assumptions and judge them for drinking during pregnancy or when taking their child with FASD to public places.³² Some families expressed a concern that the difficulties which are experienced with their children are belittled or distorted.⁴⁴ In one study parents shared that community members and professionals perceived the child to be deliberately acting poorly³⁷ although noted that these judgements are a result of that the lack of knowledge about FASD, as well as the fact that FASD is not always physically recognisable.³⁷ Parents expressed the view that educating the public and raising awareness about FASD and its accompanying behaviours is needed for parents to feel a greater amount of community support.³⁷

4.3 Diagnosis

4.3.1 Difficulties in obtaining a diagnosis

“It’s [the diagnostic process] like hitting a brick wall.” [Watson et al. 2013, p.108]

More than half of the studies highlighted that families experienced challenges in regards to obtaining a diagnosis of FASD for their children.^{19,30,32,37,38,40-42,44} Carers expressed that healthcare professionals (HCPs) may fail to make a timely diagnosis because of lack of awareness, understanding or training about functional diagnosis of FASD.^{19,30,32,38,40,42} One study indicated that sleeping problems in people with FASD are also underdiagnosed as a result of ‘inadequate explanatory models’.³⁰ Studies also mentioned that children with FASD could receive an incorrect diagnosis, such as attention deficit hyperactivity disorder (ADHD) or conduct disorder.^{30,37,42} Furthermore, both biological and non-biological carers felt that HCPs do not provide consistent messages regarding alcohol consumption during pregnancy which can further impact the diagnosis of FASD.^{38,40} One study, for example, reported several mothers who were encouraged by HCPs to consume alcohol during pregnancy.³⁷

Alongside professionals’ lack of knowledge and awareness (*see section 4.4.1*), studies identified further factors such as the lack of confirmed maternal drinking and the variability in children’s characteristics which can make the diagnosis of FASD more challenging.^{19,40} Because FASD is a spectrum disorder, the lack of physical impairments can sometimes lead to unrealistic assumptions and expectations about children’s development.¹⁹ Studies highlighted that the lack of, or incorrect, diagnosis led to caregivers being unaware of the impact of alcohol consumption during pregnancy on the child’s development.^{19,37} This can have further implications on family adaptation. Several studies, for example, identified that adoptive parents were not given a formal FASD diagnosis or enough information about the condition prior to adoption, which left them feeling that they were not able to make an informed decision or that they had a false sense that the child will develop normally.^{32,38,41,44} In two studies, however, parents started investigating the possibility of child’s prenatal alcohol exposure and initiated the process of diagnosis as they became aware that ‘something was not right’.^{19,32}

4.3.2 Formal diagnosis and awareness can help

“Without the name there’s no place, there’s no place to begin and there’s no way to try and get other people to understand.” [Watson et al. 2013, p.109]

Several studies highlighted that formal diagnosis of FASD can help caregivers to understand children’s unique needs and challenges, and be able to access appropriate support services for their children.^{19,37,40} Two studies identified that a formal diagnosis can provide relief for both biological and non-biological caregivers and alleviate family tension as they gain more understanding about the cause of their children’s behaviour and adapt their parenting strategies.^{19,42} One study, however, highlighted that parents were struggling to obtain support services for their children even after they were diagnosed with fetal alcohol effects⁴⁵ which is considered as a less severe diagnosis than fetal alcohol syndrome within the FASD spectrum.⁴⁰

4.4 Support

4.4.1 Lack of professionals’ knowledge and appropriate support

“I would want someone to say you’re not going to have to fight social services, you’re not going to have to fight education to get a placement, you’re not going to have to argue with health over resources or whatever, and then I would have felt more confident about carrying on.” [Mukherjee & Wray 2013, p.50]

Lack of knowledge and awareness of FASD among HCPs was one of the most common themes in the studies.^{19,30,32,37-40,43,44} Some caregivers talked about lack of trust in professionals because of their lack of understanding of the behavioural challenges of children with FASD.^{19,32,37} Caregivers often referred to their experience of obtaining HCP support as a ‘struggle’ and a ‘fight’.^{19,32,44} Professionals’ lack of knowledge and understanding could lead to lack of support to meet the needs of caregivers.^{32,38,44} In one study both biological and non-biological carers expressed their concerns that the lack of support can add to their children experiencing difficulties in establishing independence.³⁹

Carers sometimes felt that they were ‘not listened to’ and that children’s needs were neglected or forgotten by professionals.^{39,42} One study pointed out the lack of recognition of the challenge of sleep problems of children with FASD and the failure to integrate information about these problems into the clinical assessment of FASD which can have a negative effect on the whole family.³⁰

Caregivers also spoke about school professionals’ lack of awareness and knowledge about FASD and the associated child’s behaviour^{19,32,37,38,40,42,43} which made some parents feel that they need to educate professionals or advocate for their children’s schooling.^{37,38} Parents also stated that the lack of knowledge can lead to inappropriate schooling support and failure to meet children’s needs.^{32,38,40,42} Parents described experiences of frustration, resentment or lack of trust towards school professionals.^{19,38,42,43}

4.4.2 Formal and informal support can help

“It’s nice to know you are not alone.” [Coons et al. 2016, p.158]

In more than half of the studies caregivers shared experiences of the benefits of receiving support from professionals in managing their child’s well-being.^{19,31,32,37,38,40,42,43} Professional support was perceived by caregivers as necessary in promoting successful family adaptation and functioning.^{19,38} Professional support, both for children and their caregivers, was seen as crucial particularly in transition periods, such as from childhood into teenage years, or from teenage into adulthood.⁴³ Studies acknowledged that by providing support to the children and understanding not only their challenges but also their strengths, professionals were able to provide better assistance to parents.^{19,37} Three studies highlighted that caregivers value the support that is provided by paediatricians and other clinicians as a point of reference in accessing services or for prescribing medications.^{32,37,38} In one study biological and adoptive parents reported that the support received from counsellors helped them to alleviate stress.³⁷ Furthermore, three studies identified formal support groups as valuable in providing caregivers with an opportunity to build relationships, support each other and exchange information about parenting strategies.^{31,37,42} One study, however, highlighted that support can be beneficial only if it is relevant and meaningful to carers’ needs, with one mother reporting that she had to stop the support she was receiving in the mornings as this created ‘more of a hassle’.⁴⁰

Studies also identified the importance of informal support in the upbringing of individuals with FASD. For example, caregivers placed importance on the mutual support between spouses in the upbringing of their children,⁴³ as well as the support they receive from extended family or friends.³⁷ A biological great-grandmother shared: *“It takes a community to raise a child [with FASD]. And I believe it, I really believe it now. There’s nobody. I don’t care where you come from or where they go, there’s nobody that can do it on their own”* [Coons et al. 2016, p.157].

5 Discussion

This report synthesised qualitative studies looking at the experiences of caregivers of individuals with FASD. The aim of the synthesis was to integrate the findings from twelve primary qualitative studies by staying close to the descriptive study data. The synthesis identified four main themes: living and coping with FASD, stigma and blame, diagnosis and support.

The most prevalent theme in the synthesis related to caregivers' experience of obtaining a diagnosis of FASD. The theme of diagnosis identified two subthemes: 'difficulties in obtaining a diagnosis' and 'formal diagnosis and awareness can help'. The findings from the synthesis indicated that obtaining a formal diagnosis of FASD can be beneficial for both the individuals with the condition and their caregivers in bringing understanding into individuals' unique needs and challenges, helping caregivers adopt appropriate parenting strategies and access support services. The potential advantages of having a diagnosis in providing access to different resources and appropriate support services was also identified by a review of studies investigating the ethical aspects and consequences of diagnostics, interventions, and family support in relation to FASD.⁴⁶ The synthesis, however, identified caregivers' experiences of healthcare professionals who undermined or avoided the diagnosis of FASD because it was perceived by them as a 'label'. Some disadvantages related to FASD diagnosis were also identified by the review which found that diagnosis of FASD can have negative consequences for families related to the experience of stigma and blame, as well as feelings of guilt.⁴⁶ In contrast, the synthesis reported that some caregivers considered the experience of judgement by community members and professionals to be a result of the lack of knowledge and awareness about FASD.

The second most common theme in the synthesis captured the attitudes of parents towards receiving appropriate support to assist them in caring for an individual with FASD. Two subthemes were identified under this theme: 'lack of professionals' knowledge and appropriate support', and 'formal and informal support can help'. The synthesis found that appropriate support promotes family adaptation and functioning, helps children transition into adulthood and maintain their independence. This reflects the findings from a recent systematic review of FASD intervention studies which found that involving caregivers as 'change agents in outcome of children with FASD', by supporting them in improving their parenting skills, understanding their child's behaviour and connecting them with appropriate resources, can impact the developmental and behavioural outcome of their children.⁴⁷ Furthermore, studies in the systematic review showed that increasing parents' knowledge about the condition and the associated children's behaviour alters caregivers' attitudes and behaviours towards the child.

The synthesis identified that caring for an individual with FASD can have a deleterious impact on caregivers' individual well-being, as well as on the overall family functioning. This finding is consistent with previous research which showed that children's behaviour problems are associated with parental wellbeing.⁴⁸ Studies in the synthesis showed that contributing factors to caregivers' experiences of stress could be caused by children's externalising behaviour, fighting with the child about accepting support, having to cope with multiple disabilities in addition to FASD or poor or incorrect support from school and health professionals or missing knowledge and lack of training about FASD diagnosis.

The synthesis identified a range of strategies that caregivers used to look after their with children FASD, such as setting realistic expectations for their children, maintaining a routine and consistency in the home, picking their battles, and adapting on a day-to-day basis. The synthesis suggested that caregivers can experience positive change and improve their parenting strategies when they receive more information about the condition and the associated child's behaviour and start appreciating their children's differences and uniqueness.

Limitations

More than half of the studies included in the synthesis were conducted in Canada. Findings from qualitative research are often dependent on the context in which they were produced⁴⁹ and, therefore, transferability to other sociocultural environments needs to be considered carefully. The nature of the spectrum disorder and the complexity of the behaviours and symptoms associated with it adds a further challenge for findings to be representative of all potential caregivers' experiences of looking after individuals with FASD.

6 Conclusion

The findings from this synthesis suggest that caregivers and their families are impacted in numerous ways as they navigate the difficulties in recognising and understanding the symptoms of FASD and obtaining a diagnosis, and the consequences and management of FASD throughout the life course. A lack of knowledge and understanding of FASD among healthcare professionals is experienced as a key barrier to formal diagnosis and receiving meaningful support, which suggests the need for professional interventions, such as improved training. While there can also be negative consequences of FASD diagnosis including stigma, the synthesis suggests that an accurate diagnosis is important for receiving timely support that can help caregivers make positive changes towards managing the effects of FASD and improving the outcomes for their families. Furthermore, support that is meaningful and that meets the need of individuals with FASD and their caregivers also depends on the availability of appropriate support interventions.

6.1 Implications for practice

Caregivers believed that healthcare professionals often had poor knowledge and awareness of FASD which may limit opportunities for diagnosis, prevention and early intervention.

- It is therefore important that efforts are made to provide training and guidance, and increase awareness of prenatal alcohol exposure and FASD among healthcare professionals, as well as other relevant professional stakeholders, such as social work, criminal justice and education.
- Training on the prevention, diagnosis and management of FASD should be integrated into undergraduate and postgraduate curricula and continued professional development.

Caregivers reported that families experienced challenges in obtaining a diagnosis of FASD for their children, which presented a barrier to fully understanding the children's unique needs and challenges, and the ability to access appropriate support services. The British Medical Association Board of Science has published a report⁵⁰ calling for the development of national guidance on diagnosis and referral of FASD, based on the Canadian diagnostic guidelines as a model of best-practice.

- Guidance on the diagnosis of the full range of fetal alcohol spectrum disorders should be developed and made available to all healthcare professionals. Guidance on diagnosis should emphasise the need for testing to exclude other disorders.

6.2 Implications for policy

There are no specific services funded to oversee the diagnosis or management of individuals exposed prenatally to alcohol in the UK. The National Clinic for Fetal Alcohol Spectrum Disorders (the only specialist FASD behavioural clinic of its type in the UK) relies on individual funding requests, whereby clinicians in England apply to their local clinical commissioning group for funding in order to refer individual patients. The lack of proper commissioning acts as a barrier to the development of services required to adequately address the needs of individuals affected by prenatal alcohol exposure. Scottish Government has funded a pilot to set up a novel multidisciplinary clinic in NHS Ayrshire and Arran for the assessment and support of children with suspected FASD. The aim of the team is to work with multidisciplinary teams in health boards across NHSScotland to improve access to diagnostic services and improve clinician confidence.

- The Fetal Alcohol Advisory and Support Team in NHS Ayrshire and Arran should publish the service evaluation of the pilot project, showing how the experience gained can be used to support clinicians to develop appropriate services for families and individuals affected by FASD.

6.3 Implications for further research

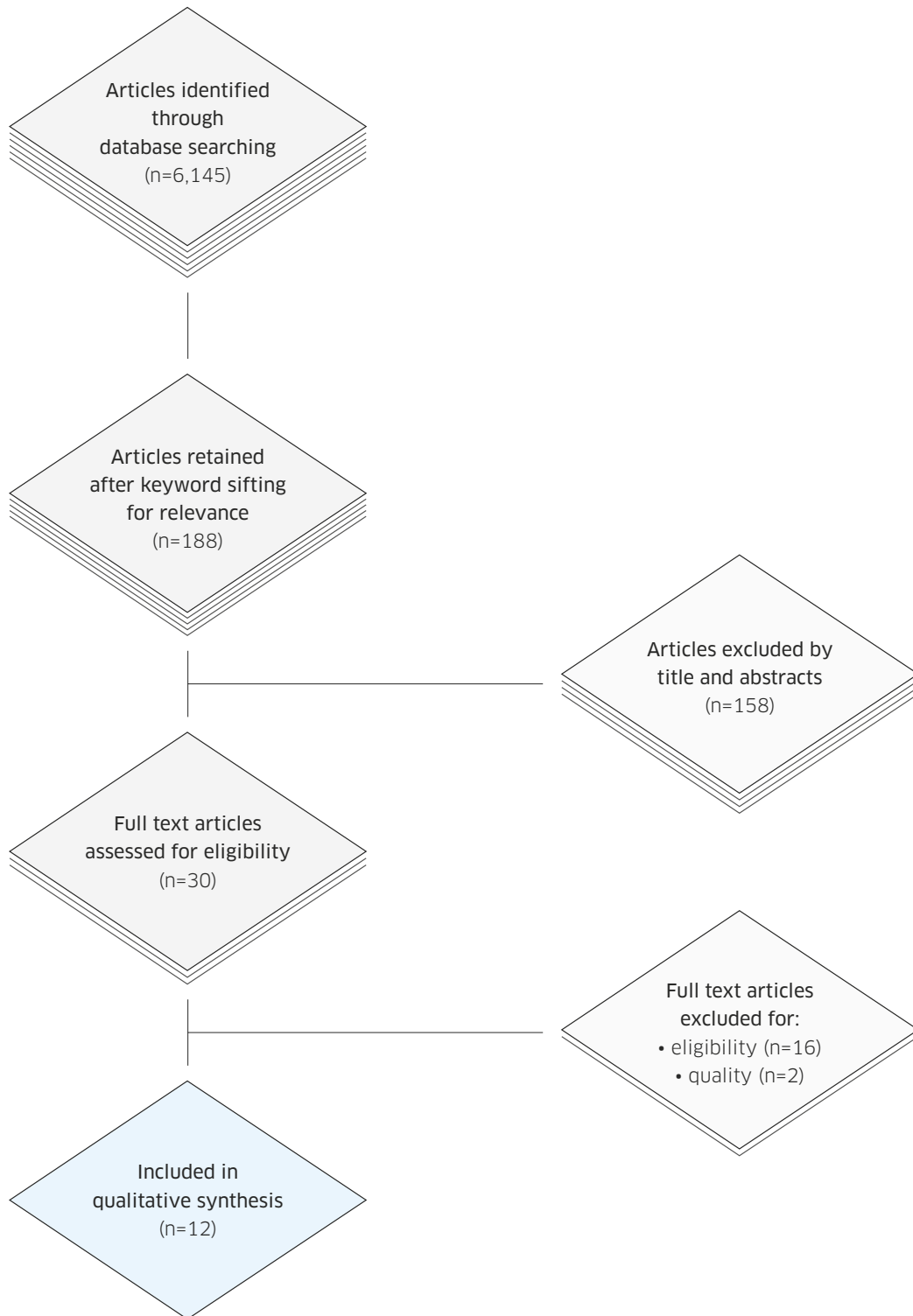
No qualitative studies were identified describing the experiences of individuals in Scotland affected by prenatal exposure to alcohol and only a single study was identified in the UK setting. Further research involving Scottish caregivers is required to inform the context of care provision within Scotland.

Abbreviations

ADHD	attention deficit hyperactivity disorder
ASD	autism spectrum disorder
CASP	Critical Appraisal Skills Programme
FAE	fetal alcohol effects
FAS	fetal alcohol syndrome
FASD	fetal alcohol spectrum disorder
HCP	healthcare professional
HIS	Healthcare Improvement Scotland
PAR	participatory action research
SIGN	Scottish Intercollegiate Guidelines Network
UK	United Kingdom
USA	United States of America

Appendices

Appendix 1: Prisma flow chart of articles



Appendix 2: Summary of eligible studies (included and excluded)

Author and year	Study sample	Participant characteristics	Children: number and age
Chamberlain et al. (2017) ³³	Foster parents (n=4), adoptive parents (n=1), legal guardians (n=5)	Married (n=5), single (n=2), widowed (n=1), divorced (n=1), domestic partnership (n=1)	n/a
Coons et al. (2016) ³⁷	Mothers (n=58): biological (n=12), step-parent (n=2), adoptive (n=39), foster (n=5). Fathers (n=26): biological (n=4), step-parent (n=3), adoptive (n=16), foster (n=3)	Mothers' average age - 50.74, married - 67.2% Father's average age - 54.35, married - 80.8%	Age range: 1–36
Coons et al. (2016) ³⁸	Adoptive mothers (n=37), adoptive fathers (n=14)	Mothers: average age - 51.73; married - 86.1% Fathers: average age - 55.45%; married 100%	Average age: 15.85 (n=53) Gender (% female): 58.49
Gardner (2000) ³⁴	Foster mothers (n=8)	Age range: 30–60	Age range - 2–16 (n=17)
Granitsas (2004) ⁴¹	Adoptive parents (n=5)	n/a	n/a
Ipsiroglu et al. (2013) ³⁰	Biological parents (n=1), adoptive parents (n=2); foster parents (n=3)	n/a	n/a
Michaud & Temple (2013) ³¹	Adoptive, foster or biological mothers (n=5)	n/a	n/a
Morrisette (2001) ⁴³	Foster and adoptive parents (n=6); 1 male/5 female	Mean number of years of experience as a foster/ adoptive parent: 10	n/a

n/a - not available

Method of data collection	Method of data analysis	Recruitment/Setting	Quality rating	Included/excluded
Semi-structured interviews conducted at two months post diagnosis	Thematic analysis	Specialist diagnostic service, Australia	Low to moderate	Excluded. Lacking depth in analysis and findings.
Semi-structured interviews	Interpretative phenomenological analysis	Ontario, Canada	High	Included
Semi-structured interviews	Interpretative phenomenological analysis	Northern Ontario, Canada	High	Included
Unstructured interviews	Content analysis	n/a	Low	Excluded. Doesn't add any substantial findings beyond body of existing literature
Unstructured interviews	Interpretative phenomenological analysis	Developmental clinic, Commonwealth of Massachusetts, USA	High	Included
Unstructured interviews	No detail given	Pediatric Sleep Assessment Clinic of British Columbia Children's Hospital, Vancouver, Canada	Difficult to appraise due to reporting	Included because of its specific focus on sleep
Participatory action research	No detail given	Sioux Lookout, Ontario, Canada	Difficult to appraise due to reporting	Included because of valuable insights provided by PAR methodology
Unstructured interviews	Narrative analysis	Missoula, Montana, USA	High	Included

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Appendix 2: Summary of eligible studies (included and excluded) continued

Author and year	Study sample	Participant characteristics	Children: number and age
Mukherjee et al. (2013) ⁴⁴	Foster and adoptive parents (n=28), biological mothers (n=2)	Age range: 30–62	n/a
Salmon (2008) ⁴²	Biological mothers (n=8)	n/a	Age range: 8.5–30 (n=9). Sample included two males and two females diagnosed with FAS, three males and one female diagnosed with fetal alcohol effects (FAE) ⁴⁵ , and one female who had partial FAE and ADHD.
Sanders (2010) ¹⁹	Biological (n=3), adoptive (n=7), and foster (n=1) parents (n=11): 2 male/9 female	Married (n=6), separated (n=1), single (n=1), divorced (n=3). Age range: 32–59	Mean age: 13.5 (n=14)
Watson et al. (2013) ⁴⁰	Biological, foster, adoptive, step-, and custodial parents (n=31)	n/a	Age range: 1–3, mean age: 16.51
Watson et al. (2013) ³⁹	Mothers (n=42): biological (n=5), step-mother (n=2), adoptive (n=32), foster (n=3) Fathers (n=15): biological (n=2), adoptive (n=11), foster (n=2)	Mothers: married – 80.95% Fathers: married – 93.33%	Mean age: 15.25
Whitehurst (2011) ³²	Adoptive mothers (n=3), biological mother (n=1)	n/a	Mean age: 10.28 (n=7) Gender: 4 female/3 male

n/a - not available

Method of data collection	Method of data analysis	Recruitment/Setting	Quality rating	Included/excluded
Focus groups	Thematic analysis	England, London and Oxfordshire, UK	Moderate	Included
Unstructured interviews	Constant comparison	Hamilton, New Zealand	Moderate	Included
Unstructured interviews	Phenomenological-hermeneutic approach and thematic analysis	Alberta, Canada	Moderate	Included
Semi-structured interviews	Interpretative phenomenological analysis	Disability support organisations, Northern Ontario, Canada	High	Included
Semi-structured interviews	Interpretative phenomenological analysis (not reported in methods section, only in results)	Disability support organisations, Northern Ontario, Canada	Moderate	Included
Unstructured interviews	Interpretative phenomenological analysis	UK	Difficult to appraise due to reporting	Included. Valuable study given its UK setting

Appendix 3: Quality assessment of included and excluded studies

Author and year	Clear statement of aims	Qualitative methodology appropriate	Research design appropriate	Recruitment strategy appropriate	Data collected appropriately	Relationship between author and participant considered
Chamberlain et al. (2017) ³³	Yes	Yes	Yes	Yes	Yes	Yes
Coons et al. (2016) ³⁷	Yes	Yes	Yes	Yes	Yes	Yes
Coons et al. (2016) ³⁸	Yes	Yes	Yes	Yes	Yes	Yes
Gardner (2000) ³⁴	Yes	Yes	Yes	Yes	Yes	No
Granitsas (2004) ⁴¹	Yes	Yes	Yes	Yes	Yes	Yes
Ipsiroglu et al. (2013) ³⁰	Yes	Yes	Yes	Can't tell	Yes	No
Michaud & Temple (2013) ³¹	Yes	Yes	Yes	Can't tell	Yes	Yes

Ethical issues taken into consideration	Data analysis sufficiently rigorous	Statement of findings clear	Value of research	Summary rating	Exclusion
Yes	No	No	The focus on diagnosis could have been more valuable if a greater depth of findings had been achieved.	Low to moderate quality. Lacking depth in analysis and findings.	Excluded
Yes	Yes	Yes	Focus on adaptation and well conducted.	High quality	Included
Yes	Yes	Yes	Detailed description of analysis given. Conclusions well supported by quotes and triangulated by other research where identified.	High quality	Included
Can't tell	Can't tell	No	Doesn't add any substantial findings beyond body of existing literature.	Low quality	Excluded
Yes	Yes	Yes	Good quotes and stayed close to lived experience. Some themes emphasised are different to other included studies.	High quality	Included
Can't tell	Can't tell	Yes	Valuable given specific focus on sleep, which is not covered in detail in any other studies included.	Reporting makes difficult to appraise probably because a part of mixed method study but a valuable study for this analysis.	Included
Can't tell	Can't tell	Yes	Findings have a different emphasis due to use of PAR methodology. Voice close to participants. May have limited transferability due to small, rural, Canadian sample but does cover common ground with other studies.	Reporting makes difficult to appraise due to PAR methodology but a valuable study for this analysis.	Included

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Appendix 3: Quality assessment of included and excluded studies continued

Author and year	Clear statement of aims	Qualitative methodology appropriate	Research design appropriate	Recruitment strategy appropriate	Data collected appropriately	Relationship between author and participant considered
Morrisette (2001) ⁴³	Yes	Yes	Yes	Yes	Yes	Yes
Mukherjee et al. (2013) ⁴⁴	Yes	Yes	Yes	Can't tell	No	No
Salmon (2008) ⁴²	Yes	Yes	Yes	Yes	Yes	No
Sanders (2010) ¹⁹	Yes	Yes	Yes	Can't tell	Yes	No
Watson et al. (2013) ⁴⁰	Yes	Yes	Yes	Yes	Yes	Yes

Ethical issues taken into consideration	Data analysis sufficiently rigorous	Statement of findings clear	Value of research	Summary rating	Exclusion
Can't tell	Yes	Yes	Has some unique contributions, focus on detail of how might support families better and discussion of family dynamics such as 'manipulation' and splitting of parents by children with FASD.	High quality with a valuable focus on how best to support families. Sample may be unrepresentative of broader group of families supporting children with FASD.	Included
Can't tell	Can't tell	Yes	Yes, does contextualise in evidence base and is a UK setting so valuable for this analysis.	Moderate quality. Valuable study in UK setting.	Included
Yes	Yes	Yes	Valuable exploration of biological mothers experiences (under-represented in most other studies). Contextualised with extensive literature review.	Moderate quality, with possible problem concerning bias of solo author.	Included
Can't tell	No	Yes	Study does make an effort to separate biological and non-biological parents experience and describes these experiences in detail, which is useful given lack of sampling of biological mothers in most other included studies.	Moderate quality	Included
Yes	Yes	Yes	Yes. Good comparison of ASD/FASD.	High quality	Included

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Appendix 3: Quality assessment of included and excluded studies continued

Author and year	Clear statement of aims	Qualitative methodology appropriate	Research design appropriate	Recruitment strategy appropriate	Data collected appropriately	Relationship between author and participant considered
Watson et al. (2013) ³⁹	Yes	Yes	Yes	Yes	Yes	Can't tell
Whitehurst (2011) ³²	Yes	Yes	Yes	Can't tell	Can't tell	Can't tell

Ethical issues taken into consideration	Data analysis sufficiently rigorous	Statement of findings clear	Value of research	Summary rating	Exclusion
Can't tell	Can't tell	Yes	One of two studies in sample that compares ASD and FASD. Interesting application of hope theory. Clear description of lower hope for future outcomes for children with FASD and degree to which this group is ignored and unsupported relative to ASD.	Moderate quality	Included
Can't tell	Yes	Yes	Yes. Valuable given UK setting and in-depth and explicit findings with good supporting quotes.	Reporting makes difficult to appraise but a valuable study for this analysis.	Included

Appendix 4: Feedback from carers

A focus group with members of the Grandparents Parenting Again and Kinship Carers (Midlothian), consisting of family members of individuals affected by prenatal alcohol exposure, was conducted in November 2018 to find out whether they identify with caregivers' experiences reported in the discussion section of the report. Carers provided feedback which was summarised according to each individual theme. This consultation aims to demonstrate what relevance each theme identified in the discussion might have for carers in Scotland.

Living with and coping with FASD

Carers identified with the experience of stress from living life through their emotions all the time and the impact that stress has on their mental and physical health as a result of caring for an individual with FASD. They further spoke about how stress about their child causes them to feel a failure and indirectly affects the relationship with their partners or other family members. Carers shared feelings of exhaustion and also shame due to unawareness about the reasons for the child's behaviour. One carer did not identify with the positive caregiving outlook described in the synthesis but shared that they would have not taken the child if they had known he/she was affected by FASD. The caregiver, however, appreciated that the child is safe and looked after.

Some carers shared concerns about their own mortality and the impact it could have on their children not being able to cope on their own. One carer expressed worry about their child's developmental delay which could have an impact on their future but noted that one-to-one support through school has helped their child.

Carers agreed with the concept described in the parenting strategies subtheme that parenting a child with FASD was a very different experience from conventional parenting. One carer made the point that what was needed was not just 'good enough parenting' but 'exceptional parenting'. Carers felt they needed to attend parenting courses such as Incredible Years and Growing with Confidence. Carers shared using different parenting strategies to cope with their child's behaviour. Carers avoided accusation but still sanctioned the behaviour of the child while reaffirming their love for them.

Stigma and Blame

Consistent with the theme synthesised from the published evidence, carers shared experiences of being criticised by community members and professionals in relation to the child's behaviours being due to poor parenting. They also shared their own criticism of their own children for their lifestyle choices. One carer showed clear disapproval towards their daughter's behaviour as it has resulted in the child being badly affected.

Diagnosis

One carer shared the experience of being told by child and adolescent mental health services that due to the lack of facial features, the child did not have FASD. Professionals suggested that the terrifying breakdown in the child's regulation and prolonged tantrums lasting hours were due to early puberty which caused the carers to feel frustrated. Sometimes carers were told that their child's behaviour could be partly due to attachment issues and partly due to autism spectrum disorder (ASD). Some carers shared the experience of their child refusing to be assessed or having to wait long periods of time for the results of assessment and subsequently being informed of no impairment. Overall, carers not only shared the experience of being frustrated because of the lack of formal diagnosis but also from being left uncertain about whether a formal diagnosis could help.

Support

One carer shared the experience of passing from one professional to another without being able to find out what was wrong with their children. Lack of knowledge and awareness of FASD among healthcare professionals was one of the most common themes identified in the studies within the literature included in the synthesis. Similarly, Scottish carers felt that both social workers and healthcare professionals should be informed about FASD. Furthermore, carers pointed out the lack of awareness and knowledge of FASD among school professionals and wanted every school and member of staff to have the pack from the National Organisation for Foetal Alcohol Syndrome-UK (NOFAS-UK) which contains a Teacher's Toolkit with checklist of things which will help the child manage on a day-to-day basis in the classroom.

Caregivers identified the significant and unique role that the Kinship Care group has in their lives because it provides them with an opportunity to support each other and exchange invaluable information around parenting, benefits, arrangements with the local authority, entitlements and much more. In regards to the importance of mutual support between spouses, one carer talked about the lack of emotional support provided by her spouse and shared the experience of feeling as being 'only one person'.

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