**A Systematic Scoping Review on Recruiting and Retaining Seldom Heard Families in Longitudinal Qualitative Research**

**Final report 27th March 2024**

**acTorgerson, C., aAsbury, K., aFields, D., aFox, L., bHarden, M., aJackson, E. and aSundaram, V.**

**aDepartment of Education**

**University of York**

**YO10 5DD**

**bCentre for Reviews and Dissemination**

**University of York**

**YO10 5DD**

carole.torgerson@york.ac.uk

kathryn.asbury@york.ac.uk

diana.fields@york.ac.uk

laura.fox@york.ac.uk

melissa.harden@york.ac.uk

emma.jackson@york.ac.uk

vanita.sundaram@york.ac.uk

**Author contributions**

CT led on design of review, writing of protocol and report; contributed to search strategies; undertook screening, data extraction, quality appraisal and synthesis; provided methodological expertise.

KA contributed to design of review and writing of protocol and report; contributed to search strategies; undertook screening, data extraction and quality appraisal; substantive expertise.

DF contributed to protocol development and report writing; contributed to search strategies; undertook screening, data extraction and quality appraisal.

LF contributed to protocol development and report writing; contributed to search strategies; undertook screening, data extraction and quality appraisal.

MH led on developing search strategies; running all search strategies and managing the search results; contributed to report writing.

EJ contributed to report writing, undertook screening, data extraction and quality appraisal.

VS contributed to design of review and writing of protocol and report; contributed to search strategies; undertook screening, data extraction and quality appraisal; substantive expertise.

**All authors approved the final version of the report.**

**Correspondence: Professor Carole Torgerson** **carole.torgerson@york.ac.uk**

**Funded by the ESRC**

**Data management: Primarily by EndNote libraries (one for each sub-review) supplemented with Excel spreadsheets and Word documents.**

**Executive Summary**

***Introduction***

The establishment of a new Early Life Cohort (ELC) study recruiting people born in the UK in the 2020s is a priority for the ESRC, with the aim of improving recruitment to ensure all parts of the population are covered by the research. As members of vulnerable groups may not be included for robust quantitative analysis, the ESRC is currently planning longitudinal qualitative studies, focused on hard to reach and hard to hear families, to run in parallel with the planned ELC study.

*Aim*

The overarching aim of this scoping review, therefore, was to systematically scope the existing literature on the best practice in how to design and conduct longitudinal qualitative research that works for, and is welcomed by two seldom heard kinds of families: (a) families in which a child has a learning disability of chromosomal or genetic origin (sub-review 1); and (b) families in which a parent has experienced violence or abuse from a partner in the last year (sub-review 2).

***Design and Methods***

A systematic scoping review was undertaken to the standards of the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) recommendations for scoping reviews in two stages: a brief mapping stage of the research landscape and an in-depth review of the most relevant studies to the research questions (RQs). A protocol for the review, including its design and methods, was pre-registered on the Open Science Framework. In addition to longitudinal studies, relevant cross-sectional studies were included in the review.

*Research questions*

The RQs for the scoping review were:

RQ1 What challenges and elements of best practice can be found in the design and conduct of longitudinal and cross-sectional descriptive studies with hard to reach and hard to hear families?

RQ 2. What evidence is there of successful strategies for overcoming these challenges?

*Searching*

The following databases were searched in May 2023: MEDLINE (Ovid), PsycINFO (Ovid), CINAHL (Ebsco), ERIC (Ebsco), ASSIA (ProQuest), Social Science Citation Index and the Emerging Sources Citation Index (Web of Science, Clarivate Analytics), using a combination of terms for 1) children (0-19 years), 2) chromosomal or genetic learning disabilities or partner abuse and 3) qualitative data collection.

*Screening*

The total ‘hits’ from the results of the electronic searching were screened using pre-specified inclusion and exclusion criteria (see registered Protocol). All studies were screened at the first stage on titles and abstracts. Following this, a brief map of the landscape was drawn which stated the number of UK-based, non UK-based, and international (undertaken in multiple countries including UK) empirical studies as well as the number of relevant previously published systematic reviews (SRs). For sub-review 1, all UK-based, international empirical studies and all SRs were screened at the second stage based on full papers. The procedure was the same for sub-review 2 using broader inclusion criteria, namely the addition of non UK-based studies published or in the public domain since 2021. For sub-review 2 we widened the inclusion criteria for the in-depth review to include non-UK studies from 2021 because this was the date of the implementation of the UK government’s Tackling Violence Against Women and Girls (VAWG) strategy and it was felt that these recently published studies could have the potential to be directly relevant to the review questions.

*Data extraction and quality appraisal*

Data to be extracted included bibliographic details and key design and conduct items in relation to the two research questions for the review. Quality appraisal of all the included studies included challenges in design and conduct; elements of best practice in design and methods; and strategies for overcoming challenges.

*Synthesis*

The results from the data extraction process were synthesised using five themes which related to the research questions and the objectives of the project, taking into account the quality appraisal judgements. Conclusions were drawn and recommendations made based on the two research questions for each sub-review.

*Quality assurance*

The quality of screening was assured through the use of double screening at both first and second stages. The quality of data extraction and quality appraisal of included studies was assured through independent double data extraction and subsequent agreement.

***Results***

*Searching and screening*

The searching resulted in: a total of 1957 ‘hits’ for sub-review 1 and a total of 2204 ‘hits’ for sub-review 2, yielding 203 studies in the map (167 in sub-review 1 and 36 in sub-review 2), following screening at first stage; and 20 studies in the in-depth review (10 in sub-review 1 and 10 in sub-review 2) following screening at second stage.

*Quality appraisal*

Quality appraisal of the nineteen studies in the two reviews resulted in all studies meeting the minimum threshold for quality.

***Sub-review 1***

***Study characteristics***

Participant children recruited in the included studies experienced learning disabilities of genetic/chromosomal origin of the following forms: Williams Syndrome (Gillooly et al., 2022 & Gillooly, 2018); Down Syndrome (Kendall, 2019; Lightfoot and Bond, 2013; Deakin and Jahoda, 2020; Holm et al, 2021; Ridding and Williams, 2019; Bostrom and Broberg, 2014); Silver-Russell Syndrome (SRS) (Ballard et al., 2021); Prader-Willi Syndrome (Cox et al., 2021); Rett Syndrome (Williamson, 2019); Autism (Bostrom and Broberg, 2014); Chromosome 14 Deletion (Bostrom and Broberg, 2014); Mosaic Trisomy 14 Syndrome (Bostrom and Broberg, 2014).

The aims of all studies were to examine perspectives on a range of variables relating to family life from the point of view of either the children experiencing the learning disability, their parents or both children and parents.

***Challenges in design and conduct***

*Nature of sample and sample size*

Data on the populations from which samples were drawn were lacking in most of the included studies. In many cases the target sample size was not stated or justified; and the sampling strategy was not stated. The range of sample sizes was 2-70, with 80% of the studies undertaking research with fewer than 20 participants. Although small sample sizes are not necessarily an issue for qualitative research, five of the studies explicitly highlighted the limitations of their samples in terms of size and/or diversity. The reviewers judged all studies in the scoping review to have recruited small, heterogeneous samples, some unrepresentative of the population, with an inherent potential for bias in many studies due to the nature of the recruitment strategies (see below), and possible skew in demographic variables, thereby limiting any generalisability from the results to other samples. Whilst the aim of qualitative research may not necessarily be to obtain generalisable conclusions, both authors and reviewers considered this to be an issue worth investigating in future qualitative research with these kinds of families.

*Recruitment (including identification)*

Identification of potential participants in the included studies was via a variety of methods, including through relevant UK registered charities; through schools; through posting on a relevant online forum; and through databases of families registered with health clinics or through local support groups. Occasionally identification was through an existing national research study or through snowball sampling.

*Retention (including attrition and attrition mitigation)*

No study mentioned the issue of retention (between recruitment and data collection). No study mentioned attrition mitigation strategies, only one study offered a small financial incentive, and in one study there was an expectation that there would be last minute drop-outs due to the challenging nature of everyday lives for the participants and so they deliberately over-recruited the focus groups to mitigate this potential. Actual attrition rates (between recruitment and data collection) were reported as being low across all ten studies. This was thought to be due to strong positive relationships developed between the researchers and participants.

*Data collection and analysis*

The data collection method of choice for most studies with parents was face to face or telephone semi-structured interviews, usually audio recorded, during visits to the family home, to schools or at a university location or outpatient clinic. Most authors did not include the interview schedule, and in 6 studies it was not clear whether this was designed by the researchers or adapted from a published interview schedule. Piloting of the data collection methods was not mentioned in any of the included studies.

Where explicitly stated, data analysis was undertaken using thematic, ‘qualitative’, interpretative phenomenological (IPA) or systematic text condensation (STC) analysis techniques. Methods for collecting and analysing data could have been described in greater detail in most studies.

*Ethics*

Few authors mentioned ethical issues in terms of challenges. Three studies provided very limited information about ethical issues, including not stating whether ethical approval had been granted. In two cases it was stated that it was not necessary to seek and obtain ethical approval because the data collection for the research was a normal part of the activities of the organisation undertaking the research. Even where it was clear that ethical approval had been granted, in some cases no information was given about the ethics committee that had granted approval or the date when such approval was obtained. In most studies, it was clear that informed consent was given, although in one study it was unclear whether this applied to the children as well as to the parents. Only one study mentioned GDPR procedures, and no study mentioned a strategy for archiving the data or indeed what would happen to the data after the end of the research. Only two studies acknowledged the sensitive nature of the research (in terms of topic and vulnerable participants).

Although many of the studies included limited descriptions of some of the features and components of their designs (e.g., ethics), this could have been due to the word restrictions imposed by the journals in which the researchers were publishing.

***Strategies for overcoming challenges***

*Nature of sample and sample size*

In all studies an increase in the size and diversity of the sample would have overcome this challenge. For this participant group an increase in diversity is more important than an increase in sample size, however. Not being able to access the hardest to reach is more of a problem than any limitation due to a lack of the generalisability that could be derived from the results. A sampling strategy combining purposive sampling with maximum variability sampling (Bryman, 2016) would enable data to be collected from as diverse a population as possible to reflect a diversity of perspectives.

*Data collection and analysis*

Data collection from children in those studies that attempted it was challenging. More accessible, creative, and inclusive data collection methods could be used in future to obtain deeper understanding of children’s perspectives.

***Elements of best practice in design and conduct***

*Data collection*

In the case where the voices of the child participants were included successfully this was achieved through a novel approach to data collection (Williamson, 2019).

*Ethics*

One study discussed the necessity of enhanced confidentiality measures due to the possibility that the nature of the condition and its rarity could lead to individuals being known through support groups. A second study discussed the implications of the research being into a highly sensitive topic with parents of children with intellectual disabilities in relation to ethical issues. One study discussed the support available should any participant become distressed during the data collection.

***Sub-review 2***

***Study characteristics***

Participant children recruited in the included studies had a parent who had experienced violence or abuse from a partner in the last year. Participant women/mothers/fathers had experienced violence or abuse from a partner in the last year. The nine studies (one was unobtainable) explored domestic violence (DV) or intimate partner violence (IPV) from a variety of perspectives, including: adolescents’ thoughts and feelings; recovery from DV; and parenting implications of DV.

***Challenges in design and conduct***

*Nature of sample and sample size*

Data on the populations from which samples were drawn were lacking in all of the included studies and the population could not be inferred. In most cases the target sample size was not stated or justified; and the sampling strategy was not stated or unclear. In a few cases, the sampling strategy was the population, i.e., all those eligible to be recruited and contacted. The studies by McCarthy (2019) and Miranda et al. (2022) used purposive sampling; in the case of the study by McCarthy the sample was a subset of a larger purposive sample from previous research undertaken by the author. All studies worked with small, homogeneous samples, and in five cases the authors highlighted this as a limitation of their research. The range of sample sizes was 5-58, with 80% of the studies undertaking research with fewer than 20 participants. As mentioned above, although small samples are not necessarily a problem in qualitative research, five of the studies explicitly highlighted the limitations of their samples in terms of size and/or diversity. The reviewers judged all studies in the scoping review to have recruited small, homogeneous samples, some unrepresentative of the population, with an inherent potential for bias in many studies due to the nature of the recruitment strategies (see below), and possible skew in demographic variables, thereby limiting any generalisability from the results to other samples. In many cases the authors also highlighted this as a limitation. Although generalisability may not be a specific objective of qualitative research, and as both authors and reviewers highlighted this as an issue, it is worth exploring the implications of this finding for future research.

*Recruitment (including identification)*

Identification of potential participants in the included studies was via a variety of methods, including: referrals from hospitals and clinics; community centres; and family justice centres (Scrafford, 2022 Chester and Joyscelyne, date); wider service providers (Meyer and Stambe, 2022); protection programmes (Miranda et al., 2022); domestic abuse support services (voluntary and statutory, Morrison, 2014); professional contacts in learning disability services (McCarthy, 2019) and schools (Izaguirre and Kaalstrom,2021). The study by Katz (2015) identified and recruited through the voluntary sector organisation ‘Women’s Aid’; the study by Bunston (2021) identified and recruited through staff at a Refuge Centre. Some methods of recruitment could have introduced a potential source of bias.

*Retention (including attrition and attrition mitigation)*

No study mentioned the issue of retention between recruitment and data collection. No study mentioned attrition mitigation strategies, and only two studies offered a small financial incentive (Meyer and Stabe, 2022) and Katz (2015). Actual attrition rates were reported as being nil across all nine studies. This was thought to be due to strong positive relationships developed between the researchers and participants.

*Data collection and analysis*

The data collection method of choice for most studies involving mothers was face to face interview, usually audio recorded (Bunston, 2023; Katz, 2015; McCarthy, 2019; Meyer and Strombe, 2022). Chester and Joscelyne (2021) undertook interviews with children and Izaguirre and Kaalstrom (2021) used self-reported questionnaires to obtain data from children. Only one study used an innovative method of collecting data through infant observations (Bunston et al, 2021); and only one other study used creative methods for data collection to elicit data from adolescents (Morrison, 2014). Most authors did not include the interview schedule. Piloting of the data collection methods was not mentioned in any of the included studies.

Where explicitly stated, data analysis was undertaken using inductive approaches: thematic content analysis (Izaguirre and Kaalstrom, 2021), framework analysis, or interpretative phenomenological analysis (IPA). Methods for collecting and analysing data could have been described in greater detail in most studies.

There was a general brevity of content in terms of design and methods of data collection and analysis. One exception to this was in the study by Izaguirre and Kaalstrom (2021) where there was a more detailed, considered analysis.

*Ethics*

Few authors mentioned ethical issues in terms of challenges. Most of the nine studies provided limited information about ethical issues, the exception being the study undertaken by Katz (2015, pp. 156-7) which covered ethics in more detail, including involving a DV survivors’ group in the design of the study and ‘Minimising imbalances of power between researcher and participants…throughout the fieldwork, following the recommendations of Eriksson and Nasman, 2012)’. This study paid detailed attention to ethical issues. However, all nine studies stated where ethical approval for the study had been obtained and also that informed consent was sought and obtained prior to data collection. No study mentioned GDPR procedures, and no study mentioned archiving of the data or what would happen to the data after the end of the research. Few studies acknowledged the sensitive nature of the research (in terms of topic and vulnerable participants).

***Strategies for overcoming challenges***

*Nature of sample and sample size*

In all studies an increase in the size and diversity of the sample would have overcome this challenge. For this participant group an increase in diversity is more important than an increase in sample size, however. Not being able to access the hardest to reach is more of a problem than any limitation due to a lack of the generalisability that could be derived from the results. A sampling strategy combining purposive sampling with maximum variability sampling (Bryman, 2016) would enable data to be collected from as diverse a population as possible to reflect a diversity of perspectives.

*Recruitment*

Other avenues for recruitment could be considered.

*Data collection and analysis*

Data collection from children in those studies that attempted it was challenging. Bunston et al (2021) used an ‘infant-led’ approach using observation.

More accessible, creative, and inclusive data collection methods could be used in future to obtain deeper understanding of children’s perspectives.

***Elements of best practice in design and conduct***

*Data collection*

In the case where the voices of the child participants were included successfully (Morrison, 2014) this was achieved through a novel approach to data collection.

***Recommendations from sub-reviews 1 and 2: Synthesis***

In undertaking longitudinal qualitative research (to complement the new birth cohort study) with seldom heard families where a child has learning disabilities of genetic or chromosomal origins, it is recommended that the samples selected draw from the full range of genetic/chromosomal conditions, including all of the conditions explored in the research in this scoping review and any other conditions missing from this review. In undertaking longitudinal qualitative research (to complement the new birth cohort study) with seldom heard families where a parent has experienced violence or abuse from a partner in the last year it is recommended that the samples selected draw from families which are currently safe from the violence and abuse (best practice) and where the voices of all relevant parties can be heard, including mothers, fathers, and children. Where voices cannot be heard because the children are below the age of 12 months it is possible that innovative methods of infant observation could be explored (although caution is advised due to the evidence for this being based on one study only (Bunston et al., 2021).

Below, recommendations for each of the five themes explored are given.

*Nature of sample and sample size*

It is recommended that the populations from which samples are drawn to explore experiences of families in which there is a child or children with learning disabilities from genetic or chromosomal origin should, where possible, be the populations for each genetic/chromosomal condition within the UK, so that any samples could be derived from all areas of the UK and not geographically limited. It is further recommended that the population is explicitly stated. For families experiencing recent DV or IPV, the populations should, if possible, be populations within the UK and not confined to one or two geographical regions only. Sampling strategies should be developed with the aim of maximising representativeness and diversity and increasing the possibility of generalising to other samples within the populations. As above, a combination of purposive and maximum variation sampling with sample sizes as large as feasible would be the recommended approach. The target sample sizes should be explicitly stated and justified. Where there is a discrepancy between the target and achieved sample sizes this should be discussed in terms of any caveats for the interpretation of the results. Where possible, the nature of the samples should be as diverse as possible, to include samples with a variety of demographic characteristics, for example, ethnicity, socio-economic status and family domicile in rural as well as urban areas of the UK.

*Recruitment (including identification)*

It is recommended that identification of potential participants to a future longitudinal qualitative study seeking to recruit families in the seldom heard group where families have a child or children with learning disabilities of genetic or chromosomal origin, would depend on the size and nature of the sample required, the sampling strategy employed, and the level of generalisability desired from the results. Random sampling of a population would give the greatest generalisability of the results, but the scoping review (sub-review 1) provides no evidence of potential success of this strategy as it was not used in any of the included studies. Also, it may not be desirable within the context of qualitative research. The same issue applies to the other kind of seldom heard families in the scoping review.

The recruitment strategies in the studies in the scoping review were mainly successful, but the authors were generally only seeking to recruit small samples. In at least two cases the recruitment strategy led to only around a quarter or a third of the target sample size being achieved. It is recommended to recruit beyond the relevant charities’ lists of families, although this strategy was shown to be successful in the scoping review (sub-review 1). It is further recommended to recruit beyond the relevant service providers lists of families, although this strategy was shown to be successful in the scoping review (sub-review 2).

*Retention (including attrition)*

It is recommended that, because retention of recruited participants could be an issue, attrition mitigation strategies should be put into place at the planning stage of the research. There is no evidence from the scoping review that incentives would help to reduce potential attrition from recruitment to data collection, although these could be considered, along with other possible strategies, such as enhanced communication and follow-up interviews. It is further recommended that eligibility criteria are explicit and clear, again at the planning stage, so that recruited participants are not withdrawn during the data collection cycle.

*Data collection and data analysis*

It is recommended that future qualitative longitudinal research with seldom heard families plans to obtain the voices of both parents and children, including possibly including the experiences of infants, although evidence for successful use of this approach was limited to one study only in the scoping review (Bunston et al., 2021). As data collection is likely to be more challenging when the participants are young (preverbal) and/or experience communication difficulties or have limited verbal abilities, exploration of the appropriateness of a range of creative data collection strategies is recommended. For example, innovative photographic and observation methods of data collection are well suited to qualitative research with seldom heard families. For all data collection methods, these should be piloted and fine-tuned to aim to elicit relevant data from all participants.

*Ethics*

The reviewers recommend as a minimum any future research with these populations (as with any research population) should state explicitly that best practice ethical guidelines (for example BERA guidelines) have been implemented at every stage of the research, including: describing in detail the process of obtaining ethical approval for the research and all data protection (GDPR) measures; and providing protocols for dealing with potential distress caused by the data collection process. In addition, details of data archiving should be planned and recorded.

**Main Report**

**Introduction and Background**

The UK has a rich tradition of undertaking longitudinal cohort studies of children (Pearson, 2016). The contribution such studies have made to science and society is immense. However, a weakness they share is a systematic absence of hard to reach and seldom asked voices within the studies. This sometimes occurs because of barriers to recruitment, retention, and data collection but, even when data are available, some participants are routinely excluded from quantitative analyses due to qualitative differences between them and the general population (e.g., children with genetic disabilities are qualitatively different from children in the general population).

The establishment of a new Early Life Cohort (ELC) study recruiting people born in the UK in the 2020s is a priority for the UK Economic and Social Research Council (ESRC), with the aim of improving recruitment to ensure all parts of the population are covered by the research. As members of vulnerable groups may not be included for robust quantitative analysis, the ESRC is currently planning longitudinal qualitative studies, focused on hard to reach and hard to hear families, to run in parallel with the planned ELC. This matters from an equality, diversity and inclusion perspective. It is also important because it will maximise the opportunity for ESRC research to benefit seldom heard groups, and enable learning about social processes within such families, including how relevant social policies and practices play out in individual lives.

***Aim***

The overarching aim of this scoping review was to systematically scope the literature and synthesise the evidence on the best practice in how to design and conduct longitudinal qualitative research that works for, and is welcomed by the two seldom heard kinds of families on which we are focusing: (a) families in which a child has a learning disability of chromosomal or genetic origin (i.e., a disability with a known genetic origin that could be tested for in infancy); and (b) families in which a parent has experienced violence or abuse from a partner in the last year.

***Policy background: Learning disability of chromosomal or genetic origin***

Disabilities, including learning disabilities of chromosomal or genetic origin, are enshrined as protected characteristics in The Equality Act (2010). Disabled people experience a substantial and long-term negative effect on their capacity to engage in everyday activities. The Equality Act confers rights that protect disabled people from discrimination in Great Britain, including in employment, education and dealing with the police. In 2022 a specific Down Syndrome Act [[Down Syndrome Act 2022 (legislation.gov.uk)](https://www.legislation.gov.uk/ukpga/2022/18/enacted)] was passed but this has proved controversial, partly because it is perceived in some quarters as pursuing the agenda of the religious right and partly because it is seen as creating unnecessary division and hierarchy among the learning disabled community.

***Policy background: Intimate partner abuse in families***

The UK government launched its most recent Tackling Violence Against Women and Girls (VAWG) strategy in 2021. The VAWG strategy acknowledges that a consequence of domestic violence is harm to children living in affected families. The Domestic Abuse Act (2021) also specifies - for the first time - a statutory definition of domestic abuse that specifies different types of relationships, including ex-partners and family members, and that captures a range of different abusive behaviours (including financial, emotional, physical). The Act also acknowledges the impact of domestic abuse on children and treats children as victims of domestic abuse where they are under parental responsibility of the abuser or the victim.

Relationships, Sex and Health Education was made a statutory part of the school curriculum (in England in 2020) and is noted as a crucial part of government strategy for tackling gender-based violence.

Specifically, the main aim of the review was to, firstly, to scope - identify, locate and briefly map - previous studies in the two topic areas, and, secondly, data extract, quality appraise and synthesise in an in-depth review, the UK-based and international longitudinal studies (undertaken anywhere in the world) and UK-based cross-sectional studies which address the practical, ethical and methodological challenges involved in recruiting families from the two seldom heard groups, and minimising attrition over time (longitudinal studies). Therefore, the mapping section is international in scope, including all longitudinal and cross-sectional studies undertaken in English-speaking countries since 2013 which met the inclusion criteria (see below). The focus of the in-depth review was on UK-based studies and any international studies undertaken in multiple countries including the UK. Longitudinal studies undertaken anywhere in the world were included in the in-depth review, however. Best practice in design features, such as sample size and sampling strategies; recruitment and retention strategies; attrition mitigation strategies; and data collection and analysis methods were explored. The evidence on particular challenges was also synthesised.

The main rationale for limiting the review to 2013 to the present was that the Data Protection Act 2018 (DPA 2018), and the UK General Data Protection Regulation (UK GDPR 2016) were established and implemented in the last 8 years. Therefore, limiting the review to the last ten years enabled us to focus on the most relevant research in terms of handling sensitive data and archiving challenges (should the evidence exist in the relevant studies).

The rationale for focusing on the UK-based literature for the in-depth review was because the evidence synthesis is intended to inform a future UK-based longitudinal qualitative cohort study and because the ethical, logistical and data protection issues germane to the review are not directly transferable across countries.

Although the scoping review intended to inform recruitment to future qualitative longitudinal studies, we included cross-sectional studies which recruited from these two groups in the last 10 years. This was because the empirical longitudinal literature was hypothesised to not be extensive, and useful methodological and practical lessons could also be drawn from cross-sectional studies which recruited participants from the two seldom heard groups.

Previous high impact longitudinal research studies, typically surveys or cohort studies, for example the UK Twins’ Early Development Study (Rimfeld et al., 2019) have tended to exclude, directly or indirectly, seldom heard families such as those with the two characteristics described above. This may be due to a variety of reasons, for example: because such families have not traditionally welcomed an approach to participate in such research; because they have signed up to participate but then faced too many barriers to continue; because they have participated but their data are excluded from analyses because they are considered qualitatively different (e.g., genetically different) from the main sample; because funders of such research deem it too challenging to attempt to recruit a sample from these groups; or because of challenges related to communication or a perception that these groups are ‘hard to reach’ (Neale, 2016). There is a smaller sub-population of families with these characteristics from which to sample and recruit, than the population of families as a whole, eligible to be included in longitudinal qualitative research cohorts or samples.

The rationales for the scoping review included: an equality and diversity imperative that families like these are studied alongside main longitudinal research cohorts; the need to establish a strong evidence base on how to recruit and retain seldom heard families within the context of future longitudinal research; and the aim of impacting on research funding priorities for future longitudinal research specifically in terms of families with certain characteristics which mean they have previously been directly or indirectly marginalised or excluded from such research. The scoping review has yielded findings relevant to other groups who are vulnerable or marginalised in similar or overlapping ways.

**Design and Methods**

The scoping review was designed, conducted and reported to PRISMA guidelines for scoping reviews [PRISMA (prisma-statement.org](http://prisma-statement.org/Extensions/ScopingReviews)) in order to minimise bias and ensure rigour. In addition, all methods and procedures for undertaking the review explicitly pre-specified in the protocol (and pre-registered with OSF: [OSF | A Systematic Scoping Review on Recruiting and Retaining Seldom Heard Families in Longitudinal Qualitative Research Review Protocol](https://osf.io/jvekr/)) included methods of quality assuring each stage of the review. This ensures transparency and replicability, and ultimately high quality.

We systematically searched for, quality appraised and synthesised empirical research using descriptive designs (both longitudinal and cross-sectional) undertaken with samples of the two kinds of families. There were two stages to the scoping review. At the brief *mapping stage* of the review all studies whether UK-based or international were included. These were mapped in terms of numbers in each category only. At the *in-depth stage* of the review for sub-review 1 UK-based studies, longitudinal studies undertaken anywhere in the world and studies undertaken in multiple countries including the UK were included; for sub-review 2 not UK-based studies published after 2021 were also included (see below).

The two kinds of families offer very different methodological challenges in including them in longitudinal research (Neale, 2021). Key issues are similar for both groups in terms of: theoretical frameworks underpinning the research; research design; design features and components, such as sampling; logistical issues with quality implications, such as recruitment, retention and attrition; research relationships and ethics; participant incentives; approaches to data collection and data management and analysis. These offer a helpful starting point when considering issues specific to longitudinal descriptive research with seldom-heard groups, but the evidence base in the literature was hypothesised to be likely to be divergent because different strategies may be required for groups with different characteristics. Therefore, the scoping review was divided into two distinct sub-reviews:

Sub-review 1: families in which a child has a chromosomal or genetic learning disability; and

Sub-review 2: families in which a parent has experienced violence or abuse from a partner in the last year.

The two sub-reviews ran concurrently, but independently. However, where there was overlap, this is summarised in the conclusions and, by doing so, the aim was to provide maximum generalisability to these and any other marginalised or vulnerable groups of families that should be included in population-based longitudinal research.

The scoping review addressed the following research questions (RQs), two for each sub-review:

***Sub-review 1***

RQ 1. What challenges and elements of best practice can be found in the design and conduct of longitudinal and cross-sectional descriptive studies with families in which a child has a learning disability of chromosomal or genetic origin?

RQ 2. What evidence is there of successful strategies for overcoming these challenges?

***Sub-review 2***

RQ 1. What challenges and elements of best practice can be found in the design and conduct of longitudinal and cross-sectional descriptive studies with families in which an adult has experienced violence or abuse from a partner in the previous year?

RQ 2. What evidence is there of successful strategies for overcoming these challenges?

Promising and/or perceived effective approaches to the following key design and conduct issues were identified for each review: recruitment; sampling strategy, target and achieved sample size; minimising attrition; participant incentives; data collection and data storage; data analysis; safeguarding and archiving of sensitive data; ethical issues. Challenges were narratively synthesised thematically from the body of literature using five themes which related to the research questions and the objectives of the project, considering the quality appraisal judgements. Any lessons learned which could be applied to future research have been highlighted.

***Sub-review 1***

All international cross-sectional and longitudinal empirical research and all systematic reviews that met the criteria were included in a brief map of the research landscape. An in-depth focus on the UK-based studies and the international longitudinal studies narrowed the scope of the review for data extraction, quality appraisal and synthesis.

*Inclusion criteria (mapping)*

1. Language and country: For a study to be included in the map it must be written in the English language and have been undertaken in an English-speaking country. To be included in the in-depth review a study must be UK-based or based in multiple countries including the UK
2. Date: For a study to be included it must be published or reported between the years 2013 and 2023 (last 10 years)
3. Publication status: For a study to be included it must be published (e.g., journal articles) or unpublished but in the public domain (e.g., reports to funders)
4. Topic and focus: For a study to be included it must be about chromosomal or genetic learning disability and must explore the family life, educational and health and wellbeing experiences and perceptions of the families and/or children
5. Sample: For a study to be included it must include either: (a) a sample of families with a child or children experiencing chromosomal or genetic learning difficulty or (b) a sample of children experiencing chromosomal or genetic learning difficulty
6. Age: For a study to be included it must include a sample of children or young people experiencing a chromosomal or genetic learning disability between the ages of 0 and 19 [in the UK young people experiencing a learning disability are entitled to an additional year of formal education]
7. Design: For a study to be included it must use a cross-sectional or longitudinal descriptive design or a systematic review design
8. Methods of data collection: For a cross-sectional or longitudinal study to be included it must use one or more of the following methods to collect the data: surveys (with qualitative outcomes); interviews (structured or semi-structured or unstructured); focus groups; participatory approaches; creative approaches; any other qualitative approaches of data collection

*Exclusion criteria (mapping)*

1. Language and country: Studies not written in the English language or not undertaken in English-speaking countries will be excluded
2. Date: Studies published or reported in or before 2012
3. Publication status: Studies not in the public domain
4. Topic: Not about chromosomal or genetic learning disability and perceptions and experiences of families and children [clinical studies in these areas will be excluded]
5. Sample: Sample does not include either: (a) families with a child or children experiencing chromosomal or genetic learning disability or (b) a sample of children experiencing chromosomal or genetic learning disability
6. Age: Studies with samples of participants over the age of 19 [if data are analysed separately for < 19 and > 19 these studies will be included]
7. Design: Studies not using a cross-sectional or longitudinal descriptive design (e.g., medical case reports or histories, intervention studies) or a systematic review design
8. Methods of data collection: Studies not using one or more of the following methods to collect the data: surveys; interviews; focus groups; participatory approaches; creative approaches; any other qualitative approaches of data collection

*Inclusion criteria (in-depth review)*

1. Must be a UK-based study or a study undertaken in multiple countries including the UK or
2. Must be a longitudinal study undertaken anywhere in the world

*Exclusion criteria (in-depth review)*

1. Study not UK-based or not based in multiple countries including the UK or
2. International study not longitudinal in design

It was not necessary to further refine the inclusion criteria for the in-depth review.

***Methods for identifying studies: Searching and screening***

***Searching***

The aim of the search was to identify published qualitative studies of families with children aged 0-19 years with chromosomal or genetic learning disabilities. The search was undertaken through the electronic searching of seven databases covering health, education and social science literature.

An information specialist (MH) developed a search strategy in Ovid MEDLINE, with guidance on relevant terminology for chromosomal or genetic learning disabilities provided by the research team. Several articles that were known to meet the inclusion criteria were examined to help inform which text word search terms and subject headings to include in the strategy, in conjunction with examination of relevant terms in database thesauri. The strategy comprised search terms for: 1) children (0-19 years), 2) chromosomal or genetic learning disabilities, and 3) qualitative data collection. Each set of terms was combined with the Boolean operator AND. The search terms for qualitative data collection were based on a sensitive search filter designed to restrict retrieval to qualitative studies (Qualitative search filters for Ovid MEDLINE and Ovid PsycINFO. Library and Graduate Communication Centre, School of Public Health, University of Texas. https://libguides.sph.uth.tmc.edu/search\_filters [accessed: 3 May 2023]).

Further limits were applied to restrict retrieval to English language studies published from 2013 onwards.

The MEDLINE strategy was translated across to the other databases, ensuring that correct search syntax and subject headings were used in each database. The following databases were searched in May 2023: MEDLINE (Ovid), PsycINFO (Ovid), CINAHL (Ebsco), ERIC (Ebsco), ASSIA (ProQuest), Social Science Citation Index and the Emerging Sources Citation Index (Web of Science, Clarivate Analytics).

After searching all databases 3542 records were retrieved in total. The records were imported into EndNote 20 (Clarivate Analytics) and deduplicated. After duplicate records were removed 1957 records remained for screening.

The full search strategy for MEDLINE can be found in Appendix A. Full search strategies for each database can be found in the Supplementary material.

***Screening for inclusion***

Screening for inclusion at the mapping (first) stage of screening was undertaken using the pre-specified inclusion and exclusion criteria. At this stage the titles and abstracts were independently double screened for inclusion or exclusion by three pairs of reviewers (CT and KA; CT and DF; CT and LF). CT screened the entire database to ensure concordance in study identification with the other reviewers. Where two reviewers were unable to agree on a decision to include or exclude a third reviewer was called on to arbitrate. A brief map of the landscape was drawn, giving numbers of included systematic reviews, UK-based cross-sectional studies, non UK-based cross-sectional studies, international studies (multiple countries, including the UK and longitudinal studies undertaken anywhere in the world.

Full papers/reports of all UK-based studies, all longitudinal studies and all studies undertaken in multiple countries including the UK (which were included on the basis of titles and abstracts) and all systematic reviews were sought and obtained. Studies were coded using the following codes: SR (systematic review); UK (UK-based study); NON (non UK-based study); INT (study undertaken in multiple countries including the UK). An additional code - LONG - was applied to any empirical study using longitudinal design. At the in-depth review (second) stage of screening these full papers were independently double screened for inclusion or exclusion by the same three pairs of reviewers. Where the two reviewers were unable to agree on a decision to include or exclude a third reviewer was called on to arbitrate. This process assured the quality of the decisions to include or exclude at each stage. At the second stage of screening the reason for excluding any study was recorded.

***Data extraction***

Data extraction was undertaken by three pairs of two reviewers (CT and KA; CT and DF; CT and LF) onto a specially developed template. Data to be extracted included bibliographic details and all the key design and conduct items in relation to the two research questions for the review: participants and identification; incentives; design; recruitment and retention; attrition; data collection methods; and ethical issues (see Appendix C for the blank data extraction template).

***Quality appraisal***

Quality appraisal of all the included studies was undertaken by three pairs of two reviewers (CT and KA; CT and DF; CT and LF) onto a specially developed template, developed from Hong et al.’s (2018) Mixed Methods Appraisal Tool (MMAT) (generic and qualitative questions only): research questions; qualitative design and methods; qualitative data collection method; findings; interpretation; coherence; challenges in design and conduct; elements of best practice in design and methods; strategies for overcoming challenges (all as judged by reviewers). A summary statement indicated whether the individual studies met the threshold for quality (see Appendix D for the blank quality appraisal template).

***Narrative synthesis***

A narrative synthesis was undertaken based on the objectives for the research (research questions) and the data extraction and quality appraisal themes. Template analysis (King, 2012) was used to thematically describe the qualitative data extracted from the studies. The data extraction template items used for the synthesis were derived from the research questions by the team members: participants and identification; population, sampling and sample; incentives; recruitment; retention and attrition; data collection and analysis methods; ethics. Challenges and elements of best practice were noted as were any strategies for overcoming the challenges. Strengths and limitations according to both authors of the papers and reviewers were noted as were the summary judgements for all studies based on the quality appraisal template derived from Hong et al. (2018).

***Report***

The report was written up using the PRISMA guidelines and checklist for scoping reviews: [PRISMA (prisma-statement.org)](http://prisma-statement.org/?AspxAutoDetectCookieSupport=1)

***Sub-review 2***

*Inclusion criteria (mapping)*

1. Language and country: For a study to be included in the map it must be written in the English language and have been undertaken in an English-speaking country. To be included in the in-depth review a study must be UK-based or based in multiple countries including the UK
2. Date: For a study to be included in must be published or reported between the years 2013 and 2023 (last 10 years)
3. Publication status: For a study to be included it must be published (e.g., journal articles) or unpublished but in the public domain (e.g., reports to funders)
4. Topic: For a study to be included it must be about domestic violence or abuse and must explore the family life, educational and health and well being experiences and perceptions of the families and/or children
5. Sample: For a study to be included it must include a sample of families which include a child or children AND in which an adult has experienced violence or abuse from a partner
6. Age: For a study to be included the it must include a sample of families in which there is a child or children between the ages of 0 and 18 and in which the abuse occurred within the last year
7. Design: For a study to be included it must use a cross-sectional or longitudinal qualitative or a systematic review design
8. Methods of data collection: For a study to be included it must use one or more of the following methods to collect the data: surveys (with qualitative outcomes); interviews (structured or semi-structured or unstructured); focus groups; participatory approaches; creative approaches; any other qualitative approaches of data collection

*Exclusion criteria (mapping)*

1. Language: Studies not written in the English language or not undertaken in English-speaking countries
2. Date: Studies published or reported in or before 2012
3. Publication status: Studies not in the public domain
4. Topic: Studies not about domestic violence or abuse and perceptions and experiences of families and children [clinical studies will be excluded]
5. Sample: Studies where sample does not include families with a child or children AND an adult who has experienced violence or abuse from a partner
6. Age: Studies in which all participant children were over the age of 18 at the time of the research or in which the abuse occurred earlier than a year previously
7. Design: Studies not using a cross-sectional or longitudinal qualitative or a systematic review design (e.g., clinical case histories, intervention studies)
8. Methods of data collection: Studies not using one or more of the following methods to collect the data: surveys (with qualitative outcomes); interviews (structured or semi-structured or unstructured); focus groups; participatory approaches; creative approaches; any other qualitative approaches of data collection

For sub-review 2 we widened the inclusion criteria for in-depth review to include non-UK studies from 2021 (the date of the UK government’s Tackling Violence Against Women and Girls (VAWG) strategy) due to these recently published studies potentially being directly relevant to the review questions.

*Inclusion criteria (in-depth review)*

1. Must be a UK-based study or a study undertaken in multiple countries including the UK or
2. Must be a longitudinal study undertaken anywhere in the world or
3. Must be a not UK-based study undertaken after 2021

*Exclusion criteria (in-depth review)*

1. Study not UK-based or not based in multiple countries including the UK or
2. International study not longitudinal in design
3. Non-UK study published in 2020 or earlier

***Methods for identifying studies: Searching and screening***

***Searching***

The aim of the search was to identify published qualitative studies of families living with DV, i.e., where parents or caregivers experienced violence or abuse from a partner. The search was undertaken through the electronic searching of seven databases covering health, education and social science literature.

An information specialist (MH) developed a search strategy in Ovid MEDLINE, with guidance on relevant terminology for domestic violence provided by the research team. Several articles that were known to meet the inclusion criteria were examined to help inform which text word search terms and subject headings to include in the strategy, in conjunction with examination of relevant terms in database thesauri. The strategy comprised search terms for: 1) children (0-18 years), 2) domestic violence, and 3) qualitative data collection. Each set of terms was combined with the Boolean operator AND. The search terms for qualitative data collection were based on a sensitive search filter designed to restrict retrieval to qualitative studies (Qualitative search filters for Ovid MEDLINE and Ovid PsycINFO. Library and Graduate Communication Centre, School of Public Health, University of Texas. https://libguides.sph.uth.tmc.edu/search\_filters [accessed: 3 May 2023]). Further limits were applied to restrict retrieval to English language studies published from 2013 onwards. Where possible, dissertations, theses, comments, editorials and letters were removed from the search to focus retrieval on published research studies.

The MEDLINE strategy was translated across to the other databases, ensuring that correct search syntax and subject headings were used in each database. The following databases were searched in June 2023: MEDLINE (Ovid), PsycINFO (Ovid), CINAHL (Ebsco), ERIC (Ebsco), ASSIA (ProQuest), Social Science Citation Index and the Emerging Sources Citation Index (Web of Science, Clarivate Analytics).

After searching all databases 4737 records were retrieved in total [MH1] . The records were imported into EndNote 20 (Clarivate Analytics) and deduplicated. After duplicate records were removed 2204 records remained for screening.

The full search strategy for MEDLINE can be found in Appendix B.

Full search strategies for each database can be found in the supplementary materials.

***Screening for inclusion***

Screening for inclusion at the mapping stage at the first stage of screening was undertaken using the pre-specified inclusion and exclusion criteria. At this stage of screening the titles and abstracts were independently double screened for inclusion or exclusion by three pairs of reviewers (CT and VS; CT and EJ; CT and DF). CT screened the entire database to ensure concordance in study identification with the other reviewers. Where two reviewers were unable to agree on a decision to include or exclude a third reviewer was called on to arbitrate. Studies were coded using the following codes: SR (systematic review); UK (UK-based study); NON (not UK-based study); INT (study undertaken in multiple countries including the UK). An additional code - LONG - was applied to any empirical study using longitudinal design. A brief map of the landscape was drawn. Full papers/reports of all UK-based studies, all longitudinal studies and all studies undertaken in multiple countries including the UK (which were included on the basis of titles and abstracts) were sought and obtained. At the second stage of screening these full papers were independently double screened for inclusion or exclusion by the same three pairs of reviewers. Where the two reviewers were unable to agree on a decision to include or exclude a third reviewer was called on to arbitrate. This process assured the quality of the decisions to include or exclude at each stage. At the second stage of screening the reason for excluding any study was recorded.

***Data extraction***

Data extraction was undertaken by three pairs of two reviewers (CT and VS; CT and DF; CT and EJ) onto a specially developed template. Data to be extracted included bibliographic details and all the key design and conduct items in relation to the two research questions for the review (see Appendix C).

***Quality appraisal***

Quality appraisal of all the included studies was undertaken by pairs of two reviewers (CT and VS; CT and EJ; CT and DF) onto a specially developed template, developed from Hong et al.’s (2018) Mixed Methods Appraisal Tool (MMAT) (see Appendix D).

***Narrative synthesis***

A narrative synthesis was undertaken based on the objectives for the research (research questions) and the data extraction and quality appraisal themes. Template analysis was used to thematically describe the qualitative data extracted from the studies. The data extraction template items used for the synthesis were derived from the research questions by the team members: participants and identification; population, sampling and sample; incentives; recruitment; retention and attrition; data collection and analysis methods; ethics. Challenges and elements of best practice were noted as were any strategies for overcoming the challenges. Strengths and limitations according to both authors of the papers and reviewers were noted as were the summary judgements for all studies based on the quality appraisal template derived from Hong et al. (2018).

***Report***

The report was written up using the PRISMA guidelines and checklist for scoping reviews: [PRISMA (prisma-statement.org)](http://prisma-statement.org/?AspxAutoDetectCookieSupport=1)

**Results**

***Searching***

The searching and screening at first and second stages resulted in a total of 1957 ‘hits’ for sub-review 1 and a total of 2204 ‘hits’ for sub-review 2 yielding 203 studies in the map (167 in sub-review 1 and 35 studies in sub-review 2) and 20 studies in the in-depth reviews (10 in sub-review 1 and 10 in sub-review 2). One study was unobtainable in the in-depth review for sub-review 2 and could not be included for data extraction (Moe et al., 2022). This left 9 studies remaining for data extraction and quality appraisal for sub-review 2. (See below the PRISMA flow diagrams for each sub-review: Figures 1 and 2)

**Figure 1: Flow of studies for Sub-review 1 (Chromosomal and Genetic Learning Disabilities)**





























**Figure 2: Flow of studies for Sub-review 2 (Partner Abuse)**





























***Sub-review 1***

***Map and in-depth review***

Table 1 shows the number of studies included at first and second stages of screening and included in the map and the in-depth review.

**Table 1: Map and in-depth review**

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Screening and inclusion** | **SR** | **UK** | **NON** | **NON LONG** | **INT** | **Total** |
| Number of studies included at first stage of screening (titles and abstracts) | 3 | 20 | 135 | 4 | 5 | 167 |
| Number of studies included at second stage of screening for inclusion in the in-depth review: 10 empirical studies data extracted and quality appraised | 0 | 8 | N/A | 1 | 1 | **10** |

Three systematic reviews were included in the map. Twenty empirical studies undertaken in the UK and 135 studies undertaken in non-UK countries were included as were 4 non-UK studies with a longitudinal design and 5 studies undertaken internationally across more than one country.

***Screening for in-depth review***

After applying the criteria for the in-depth review to the 135 studies in the map, ten studies (in eleven publications) were included in the in-depth review for full data extraction and quality appraisal (Ballard et al, 2021; Bostrom and Broberg, 2014; Cox et al, 2021; Deakin and Jahoda, 2020; Gillooly 2018 & Gillooly et al, 2022; Kendall, 2019; Lightfoot and Bond, 2013; Holm et al, 2021; Ridding and Williams, 2019; Williamson, 2019). Table 2 gives the study codes, authors and publication dates of the ten included studies.

**Table 2: In-depth review**

|  |  |  |
| --- | --- | --- |
| **Study code** | **Number of studies** | **Author(s), date** |
| UK:UK-basedCross-sectional design | 8 | Ballard et al, 2021; Cox et al, 2021; Deakin and Jahoda, 2020; Gillooly 2018 & Gilooly et al, 2022; Kendall, 2019; Lightfoot and Bond, 2013; Ridding and Williams, 2019; Williamson, 2019 |
| NON LONG:Non UK-based Longitudinal design | 1 | Bostrom and Broberg, 2014 |
| INT:International studyCross-sectional study but part of a longitudinal study | 1 | Holm et al, 2021 |

***Data extraction and quality appraisal of studies***

The ten studies were independently double data extracted and quality appraised using the templates specifically designed for this review (see Appendices C and D). The quality appraisal process involved the reviewers making summary judgements about the quality of the studies relevant to three aspects of the two research questions (challenges; strategies to overcome challenges and best practice). A final completed and agreed data extraction and quality appraisal table for each study is included in Appendix E. Nine studies used a cross-sectional design; one used a longitudinal design (Bostrom and Broberg, 2014); and one used a cross-sectional design, but was part of a larger longitudinal study (Holm et al, 2021). Eight studies were undertaken in the UK (Ballard et al, 2021; Cox et al, 2021; Deakin and Jahoda, 2020; Gillooly 2018 & Gillooly et al, 2022; Kendall, 2019; Lightfoot and Bond, 2013; Ridding and Williams, 2019; Williamson, 2019); one study was undertaken in Sweden (Bostrom and Broberg, 2014); and one study was undertaken in Denmark, Italy, Poland and Spain (Holm et al, 2021).

Figure 1 shows the full searching and screening process for the map and in-depth review using the PRISMA flow-diagram.

***Study characteristics***

Participant children recruited in the included studies experienced learning disabilities of genetic/chromosomal origin of the following forms: Williams Syndrome (Gillooly et al 2022 & Gillooly, 2018); Down Syndrome (Kendall, 2019; Lightfoot and Bond, 2013; Deakin and Jahoda, 2020; Holm et al, 2021; Ridding and Williams, 2019; Bostrom and Broberg, 2014); Silver-Russell Syndrome (SRS) (Ballard et al, 2021); Prader-Willi Syndrome (Cox et al, 2021); Rett Syndrome (Williamson, 2019); autism (Bostrom and Broberg, 2014); Chromosome 14 Deletion (Bostrom and Broberg, 2014); Mosaic Trisomy 14 Syndrome (Bostrom and Broberg, 2014).

The aims of all studies were to examine perspectives on a range of variables relating to family life from the point of view of either the children experiencing the learning disability, their parents or both children and parents. The number of studies recruiting child only, both parent and child, and parent only participants for data collection was one, two and seven participants respectively, with most studies only including the parental voice. Individual studies focused either on one specific variable, for example, friendships (as in Gillooly et al, 2022) or inclusion in mainstream education (as in Kendall, 2019; Lightfoot and Bond, 2013) or more generally on a range of variables, for example, the ‘lived experiences of adolescents with SRS’ (as in Ballard et al, 2021) fathers’ experiences of parenthood (as in Bostrom and Broberg, 2014; Ridding and Williams, 2019) and mothers’ views and experiences of family life with a child experiencing Down syndrome (as in Deakin and Jahoda, 2010).

The inclusion of ten studies (published in the last ten years) for in-depth review shows that cross-sectional research collecting and analysing qualitative data with a sample of participants from the seldom heard group of families with a child or children with learning disabilities stemming from a genetic or chromosomal origin is possible. Very little longitudinal qualitative research with this population was retrieved through the scoping review, the only exception being the study by Bostrom and Broberg (2014). However, lessons can be learned from the scoping review results to inform future planned longitudinal qualitative research with this population, as studies using a cross-sectional design to undertake research with this population still need to: recruit participants meeting the eligibility criteria; conduct the research in a sensitive and ethical manner; and collect and analyse data appropriately, considering the nature of the population.

To address the two research questions, data extraction and quality appraisal judgements were synthesised thematically, using template analysis with codes from the data extraction templates. Five themes were explored in respect of: challenges in design and conduct; strategies for overcoming these challenges and best practice. The five themes were: the nature of sample and sample size; recruitment (including identification; and incentives); retention (including attrition); data collection and analysis; and ethics.

*Quality appraisal*

Quality appraisal of the ten studies resulted in all ten studies meeting the minimum threshold for quality. Apart from explicitly stating their research questions, all studies were judged to be adequate in all domains. In those cases where the research questions were not explicitly stated, they could be inferred from the aim(s) of the study. All ten studies used an appropriate qualitative design and appropriate data collection and data analysis methods. Findings were adequately derived from the data in all cases and the interpretation of results was sufficiently substantiated by the data. In most cases, where there was a limited sample, this was acknowledged and there was coherence between the data collection, analysis, and interpretation. Overgeneralization beyond the small samples was generally avoided in most cases. In summary, although there was variation in the quality of the included studies, all studies were of sufficient quality for the data around the themes to be included.

***Challenges in design and conduct***

*Nature of sample and sample size*

Data on the populations from which samples were drawn were lacking in most of the included studies. In a few cases the population was stated (for example, Cox et al, 2021; Gillooly et al, 2022); in a few studies the population could be inferred from the information on identification (for example, Kendall, 2019). In many cases the target sample size was not stated or justified; and the sampling strategy was not stated. In a few cases, the sampling strategy was the population, i.e., all those eligible to be recruited and contacted. The studies by Kendall (2019) and Williamson (2019) used purposive sampling. Ridding and Williams (2019) used theoretical sampling as outlined by Strauss and Corbin (1998) to obtain ‘...as much variation as possible in the first participants, followed by sampling based on concepts and themes emerging from the data in subsequent phases’ (p.680). Gillooly et al (2022) justified their sample size by stating that it was in line with previous research of this kind. In this study the target sample size was stated (79) but only 27% of the target sample participants were recruited (21). All studies worked with small sample sizes, and in most cases the authors highlighted this as a limitation of their research. The range of sample sizes was 2-70, with 80% of the studies undertaking research with fewer than 20 participants. The largest sample size of 70 was achieved in the international study undertaken in four countries longitudinally (Holm et al, 2021), although the authors originally planned a higher sample size which was not achievable due to logistical reasons associated with the COVID-19 pandemic. Although small sample sizes are not necessarily an issue for qualitative research, five of the studies explicitly highlighted the limitations of their samples in terms of size and/or diversity. The reviewers judged all studies in the scoping review to have recruited small, heterogeneous samples, some unrepresentative of the population, with an inherent potential for bias in many studies due to the nature of the recruitment strategies (see below), and possible skew in demographic variables, thereby limiting any generalisability from the results to other samples. Whilst the aim of qualitative research may not necessarily be to obtain generalisable conclusions, both authors and reviewers considered this to be an issue worth investigating in future qualitative research with these kinds of families.

*Recruitment (including identification)*

Identification of potential participants in the included studies was via a variety of methods, including: through a relevant UK registered charity, for example through the Williams Syndrome Foundation (Gillooly et al, 2021), Down Syndrome Foundation (Kendall, 2019), Rett UK (Williamson, 2019); through schools; through posting on the relevant online forum, for example the Down Syndrome Association forum (Kendal et al, 2019); and through databases of families registered with health clinics or through local support groups. Occasionally identification was through an existing national research study (Ballard et al, 2021) or through snowball sampling. In at least one study the participants were previously known to the researchers who were therefore not completely independent (Cox et al, 2021).

*Retention (including attrition and attrition mitigation)*

No study mentioned the issue of retention. No study mentioned attrition mitigation strategies, only one study offered a small financial incentive (Bostrom and Broberg, 2014), and in one study there was an expectation that there would be last minute drop-outs due to the challenging nature of everyday lives for the participants and so they over-recruited to the focus groups (Holm et al, 2021). Actual attrition rates from recruitment to data collection were reported as being low across all ten studies. This was thought to be due to strong positive relationships developed between the researchers and participants. In one study, one child was unable to take part in the interview due to communication difficulties (Gillooly et al, 2022). In another study, one participant was excluded from the study after 3 years due to a formal diagnosis of learning disabilities being ruled out (no further explanation given) (Bostrom and Broberg, 2014). In the study by Holm et al (2021) there was attrition between recruitment and data collection from the focus groups to the extent that two of the focus groups had to be undertaken with a sub-optimal number of participants. This was a specific issue in relation to the problems of data collection during the time of the COVID-19 pandemic.

*Data collection and analysis*

The data collection method of choice for most studies for data collection from parents was face to face or telephone semi-structured interview, usually audio recorded, during visits to the family home, to schools or at a university location or outpatient clinic. Most authors did not include the interview or focus group schedules and in 6 studies it was not clear whether this was researcher-designed or adapted from a published interview schedule. The exceptions to this were the studies by Bostrom and Broberg (2014), Gillooly et al (2021), Holm et al (2021) and Lightfoot and Bond (2013), where the authors explicitly stated a published interview schedule was adapted for use in their research. Focus groups were also used in one study.

The study by Gillooly et al (2021) adapted a published interview schedule to collect data from children, but this was challenging, as many of the children had limited verbal skills and struggled to retain attention. The data collected from the children could not be analysed in the planned way due to its limited nature. Piloting of the data collection methods was not mentioned in any of the included studies.

Where explicitly stated, data analysis was undertaken using thematic, ‘qualitative’, interpretative phenomenological (IPA) or systematic text condensation (STC) analysis techniques. Methods for collecting and analysing data could have been described in greater detail in most studies. Holm et al (2021), however, described their use of STC analysis in detail.

*Ethics*

Few authors mentioned ethical issues in terms of challenges. Three studies provided very limited information about ethical issues, including not stating whether ethical approval had been granted. In two cases it was stated that it was not necessary to seek and obtain ethical approval because the data collection for the research was a normal part of the activities of the organisation undertaking the research. Even where it was clear that ethical approval had been granted, in some cases no information was given about the ethics committee that had granted approval or the date when such approval was obtained. In most studies, it was clear that informed consent was given, although in one study it was unclear whether this applied to the children as well as to the parents. Only one study mentioned GDPR procedures, and no study mentioned a strategy for archiving the data or indeed what would happen to the data after the end of the research. Only two studies (see below in best practice) acknowledged the sensitive nature of the research (in terms of topic and vulnerable participants).

Although many of the studies included limited descriptions of some of the features and components of their designs (e.g., ethics), this could have been due to the word restrictions imposed by the journals in which the researchers were publishing.

***Strategies for overcoming challenges***

*Nature of sample and sample size*

In all studies an increase in the size and diversity of the sample would have overcome this challenge.

*Data collection and analysis*

Data collection from children in those studies that attempted it was challenging. Gillooly et al (2021) suggested the future use of ‘accessible participatory methods’ to collect data from children, to capture their voices. Lightfoot and Bond (2013) undertook ‘voice of the child interviews’ using school-based images and happy or sad emojis for the children’s responses. Williamson (2019) used photographic images taken by the parents prior to the interview to use as prompts for the discussion.

More accessible, creative, and inclusive data collection methods could be used in future to obtain deeper understanding of children’s perspectives.

***Elements of best practice in design and conduct***

*Data collection*

In the case where the voices of the child participants were included successfully (Lightfoot and Bond, 2013) this was achieved through a novel approach to data collection, whereby children were asked to use emojis to indicate their preferences, in response to school based images, and to ‘peg’ those preferences on a ‘washing line’ to indicate how happy or sad they made them feel.

*Ethics*

One study discussed the necessity of enhanced confidentiality measures due to the possibility that the nature of the condition (SRS) and its rarity could lead to individuals being known through support groups (Ballard et al, 2021). A second study discussed the implications of the research being into a highly sensitive topic with parents of children with intellectual disabilities in relation to ethical issues (Deakin and Jahoda, 2020). One study discussed the support available should any participant become distressed during the data collection.

The study by Deakin and Jahoda (2020) used IPA and the steps used to quality assure this process are outlined in detail in their study write-up.

***Sub-review 2***

***Map and in-depth review***

Table 3 shows the number of studies included at first and second stages of screening and included in the map and the in-depth review.

**Table 3: Map and in-depth review**

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Screening and inclusion** | **SR** | **UK** | **NON** | **NON****LONG** | **INT** | **Total** |
| Number of studies included at first stage of screening (titles and abstracts) | 8 | 5 | 22 | 0 | 1 | 36 |
| Number of studies included at second stage of screening for inclusion in the in-depth review: 7 SRs summarised; \*9 empirical studies data extracted and quality appraised | 7 | 4 | 4 (\*5) | 0 | 1 | **(16) \*17** |

\* 1 unobtainable

8 systematic reviews were included in the map. Five empirical studies undertaken in the UK and 22 studies undertaken in non-UK countries were included, as was 1 study undertaken internationally across more than one country.

***Screening for in-depth review***

After applying the criteria for the in-depth review to the 36 studies in the map, ten studies were included in the in-depth review for full data extraction and quality appraisal (Chester & Joscelyne, 2021; Katz, 2015; Morrison, 2015; McCarthy, 2019; Izaguirre, 2021; Meyer & Stambe, 2022; Miranda et al, 2023; Scrafford, 2022; Moe et al, 2022; Bunston et al, 2021) One study was unobtainable (Moe et al, 2022), leaving nine studies for full data extraction and quality appraisal. Table 4 gives the authors and publication dates of the ten studies included in the in-depth review.

**Table 4: In-depth review**

|  |  |  |
| --- | --- | --- |
| **Study code** | **Number of studies** | **Author(s), date** |
| SR:Systematic review | 7 (in 8 publications) | Arai et al, 2021; Houghton, 2021; Hui & Maddern, 2021; Noble-Carr et al, 2020 & Noble-Carr et al, 2021; Rogers & Berger, 2022; Shory & Baladram, 2023; Sousa et al, 2022 |
| UK:UK-based research | 4 | Chester & Joscelyne, 2021; Katz, 2015; Morrison, 2015; McCarthy, 2019  |
| NON-UK:Non-UK-based research | 4 (\*5) | Izaguirre, 2021; Meyer & Stambe, 2022; Miranda et al, 2023; Scrafford, 2022; \*Moe et al, 2022 |
| INT:International research | 1 | Bunston et al, 2021 |

***Data extraction and quality appraisal of studies***

The nine studies were independently double data extracted and quality appraised using the templates specifically designed for this review (see Appendices C and D). The quality appraisal process involved the reviewer making summary judgements about the quality of the studies and judgements relevant to three aspects of the two research questions. A final completed and agreed data extraction and quality appraisal table for each study is included in Appendix F. All nine studies used a cross-sectional design; four studies were undertaken in the UK (Chester and Joyscelyne, 2021; Morrison, 2014; McCarthy, 2019; Katz, 2015); one study was undertaken in each of the following countries: Spain (Izaguirre and Kaalstrom, 2021); Australia (Meyer and Stambe, 2022); Chile (Miranda et al, 2022) and the United States of America (Scrafford, 2021). One study was undertaken in two countries: Australia and the UK (Bunston et al, 2021).

Figure 2 shows the full searching and screening process using the PRISMA flow-diagram.

***Study characteristics***

Participant children recruited in the included studies had a parent who had experienced violence or abuse from a partner in the last year. Participant women/mothers/fathers had experienced violence or abuse from a partner in the last year. The nine studies explored domestic violence (DV) or intimate partner violence (IPV) from a variety of perspectives, including: adolescents’ thoughts and feelings; recovery from DV; and parenting implications of DV.

The number of studies recruiting child only, both parent and child, and parent only participants for data collection was 3, 3 and 3 participants respectively. The sample size of included participants in the nine studies ranged from 5 to 55, with seven of the nine studies including fewer than 20 participants.

It is clear from the inclusion of nine recently published studies for in-depth review that cross-sectional research collecting and analysing qualitative data is possible with a sample of participants from the seldom heard group of families with a child or children who had a parent who had experienced intimate partner violence in the past year. No longitudinal qualitative research with this population was retrieved through the scoping review. However, many lessons can be learned from the scoping review results to inform future planned longitudinal qualitative research with this population as studies using a cross-sectional design to undertake research with this population still need to recruit and retain participants meeting eligibility criteria; conduct the research in a sensitive and ethical manner and collect and analyse data appropriately, considering the nature of the population.

To address the two research questions, data extracted from the nine studies and quality appraisal judgements about the nine studies were synthesised thematically, using codes from the data extraction templates. Five themes were explored in respect of: challenges in design and conduct; strategies for overcoming these challenges and best practice. These were: the nature of sample and sample size; recruitment (including identification; and incentives); retention (including attrition); and ethics.

*Quality appraisal*

Quality appraisal of the nine studies resulted in all nine studies meeting the minimum threshold for quality. Apart from the question about research questions, all studies were judged to be adequate in all domains. In those cases where the research questions were not explicitly stated, they could be inferred from the aim(s) of the study. All nine studies used an appropriate qualitative design and appropriate data collection and data analysis methods. Findings were adequately derived from the data in all cases and the interpretation of results was sufficiently substantiated by the data. In most cases, where there was a limited sample, this was acknowledged and there was coherence between the data collection, analysis and interpretation. Overgeneralisation beyond the small samples was generally avoided in most cases.

***Challenges in design and conduct***

*Nature of sample and sample size*

Data on the populations from which samples were drawn were lacking in all of the included studies and the population could not be inferred. In most cases the target sample size was not stated or justified; and the sampling strategy was not stated or unclear. In a few cases, the sampling strategy was the population, i.e., all those eligible to be recruited and contacted. The studies by McCarthy (2019) and Miranda et al (2022) used purposive sampling; in the case of the study by McCarthy the sample was a subset of a larger purposive sample from previous research undertaken by the author. All studies worked with small, homogeneous samples, and in five cases the authors highlighted this as a limitation of their research. The range of sample sizes was 5-58, with 80% of the studies undertaking research with fewer than 20 participants. Five of the studies explicitly highlighted the limitations of their samples in terms of size and/or diversity. The reviewers judged all studies in the scoping review to have recruited small, homogeneous samples, some unrepresentative of the population, with an inherent potential for bias in many studies due to the nature of the recruitment strategies (see below), and possible skew in demographic variables, thereby limiting any generalisability from the results to other samples.

*Recruitment (including identification)*

Identification of potential participants in the included studies was via a variety of methods, including: referrals from hospitals and clinics; community centres; and family justice centres (Scrafford, 2022 Chester and Joyscelyne, 2021); wider service providers (Meyer and Stambe, 2022); protection programmes (Miranda et al, 2022); domestic abuse support services (voluntary and statutory, Morrison, 2014); professional contacts in learning disability services (McCarthy, 2019) and schools (Izaguirre and Kaalstrom, 2021). The study by Katz (2015) identified and recruited through the voluntary sector organisation ‘Women’s Aid’; the study by Bunston (2021) identified and recruited through staff at a Refuge Centre. Some methods of recruitment could have introduced a potential source of bias.

*Retention (including attrition and attrition mitigation)*

No study mentioned the issue of retention. No study mentioned attrition mitigation strategies, and only two studies offered a small financial incentive, $25 in the case of Meyer and Stabe (2022) and £10 in the case of Katz (2015). Actual attrition rates were reported as being nil across all nine studies. This was thought to be due to strong positive relationships developed between the researchers and participants.

*Data collection and analysis*

The data collection method of choice for most studies for data collection from mothers was face to face interview, usually audio recorded (Bunston, 2023; Katz, 2015; McCarthy, 2019; Meyer and Strombe, 2022). Chester and Joscelyne (2021) undertook interviews with children and Izaguirre and Kaalstrom (2021) used self-reported questionnaires to obtain data from children. Only one study used an innovative method of collecting data through infant observations (Bunston et al, 2021); and only one other study used participatory methods for data collection to elicit data from adolescents (Morrison, 2015). Most authors did not include the interview schedule. Piloting of the data collection methods was not mentioned in any of the included studies.

Where explicitly stated, data analysis was undertaken using inductive approaches: thematic content analysis (Izaguirre and Kaalstrom, 2021), framework analysis, or interpretative phenomenological analysis (IPA) (e.g., McCarthy, 2019. Methods for collecting and analysing data could have been described in greater detail in most studies.

There was a general brevity of content in terms of design and methods of data collection and analysis. One exception to this was in the study by Izaguirre and Kaalstrom (2021) where there was a more detailed, considered analysis.

*Ethics*

Few authors mentioned ethical issues in terms of challenges. Most of the nine studies provided limited information about ethical issues, the exception being the study undertaken by Katz (2015) which covered ethics in more detail, including involving a DV survivors’ group in the design of the study and ‘Minimising imbalances of power between researcher and participants…throughout the fieldwork, following the recommendations of Eriksson and Nasman, 2012)’. This study paid detailed attention to ethical issues. However, all nine studies stated where ethical approval for the study had been obtained and that informed consent was sought and obtained prior to data collection. No study mentioned GDPR procedures, and no study mentioned archiving of the data or what would happen to the data after the end of the research. Few studies acknowledged the sensitive nature of the research (in terms of topic and vulnerable participants), although this could have been due to the nature of the journals in which the studies were published and any restrictions on word length etc.

***Strategies for overcoming challenges***

*Nature of sample and sample size*

In all studies an increase in the size and diversity of the sample would have overcome this challenge. For this participant group an increase in diversity is more important than an increase in sample size, however. Not being able to access the hardest to reach is more of a problem than any limitation due to a lack of the generalisability that could be derived from the results. A sampling strategy combining purposive sampling with maximum variability sampling (Bryman, 2016) would enable data to be collected from as diverse a population as possible to reflect a diversity of perspectives.

*Recruitment*

Other avenues for recruitment could be considered, although the avenues used in the studies in the review did work quite successfully.

*Data collection and analysis*

Data collection from children in those studies that attempted it was challenging due mainly to challenges in regard to verbal communication. Bunston et al (2021) used an ‘infant-led’ approach using observation.

More accessible, creative and inclusive data collection methods could be used in future to obtain deeper understanding of children’s perspectives.

***Elements of best practice in design and conduct***

*Data collection*

In the case where the voices of the child participants were included successfully (Bunston et al., 2021) this was achieved through a novel approach to data collection (infant-led observation, see above).

***Recommendations from sub-reviews 1 and 2: Synthesis***

In undertaking longitudinal qualitative research (to complement the new birth cohort study) with seldom heard families where a child experiences learning disabilities of genetic or chromosomal origins, it is recommended that the samples selected draw from the full range of genetic/chromosomal conditions, including all of the conditions explored in the research in this scoping review and any other conditions missing from this review. In undertaking longitudinal qualitative research (to complement the new birth cohort study) with seldom heard families where a parent has experienced violence or abuse from a partner in the last year it is recommended that in selecting samples, issues arising in the scoping review are considered, including drawing from families which are currently safe from the violence and abuse and including the voices of all relevant parties, including mothers, fathers and children.

Below, recommendations for each of the five themes explored are given.

*Nature of sample and sample size*

It is recommended that the populations from which samples are drawn to explore experiences of families in which there is a child or children with learning disabilities from genetic or chromosomal origin should, where possible, be populations for each genetic/chromosomal condition within the UK, so that any samples are from all areas of the UK and not geographically limited. This could be achieved using a combination of a purposive, maximum sampling variation approach and place-based strategy. It is further recommended that the population is explicitly stated. For families experiencing recent DV or IPV, the populations should, if possible, be populations within the UK and not confined to one or two geographical regions only. Sampling strategies should be developed with the aim of maximising generalisability to other samples within the populations where possible. The target sample sizes should be explicitly stated and justified. Where there is a discrepancy between the target and achieved sample sizes this should be discussed in terms of any caveats for the interpretation of the results. Where possible, the nature of the samples should be as diverse as possible, to include samples with a variety of demographic characteristics, for example, ethnicity, socio-economic status and family domicile in rural as well as urban areas of the UK.

*Recruitment (including identification)*

It is recommended that identification of potential participants to a future longitudinal qualitative study seeking to recruit families in the seldom heard group where families have a child or children with learning disabilities of genetic or chromosomal origin, would depend on the size and nature of the sample required, the sampling strategy employed, and the level of generalisability desired from the results. Random sampling of a population would give the greatest generalisability of the results, but the scoping review (sub-review 1) provides no evidence of potential success of this strategy as it was not used in any of the included studies. The same issue applies to the other seldom heard families in the scoping review.

The recruitment strategies in the studies in the scoping review were mainly successful, but the authors were generally only seeking to recruit small samples. In at least two cases the recruitment strategy led to only around a quarter or a third of the target sample size being achieved. It is recommended to recruit beyond the relevant charities’ lists of families, although this strategy was shown to be successful in the scoping review (sub-review 1). It is further recommended to recruit beyond the relevant service providers lists of families, although this strategy was shown to be successful in the scoping review (sub-review 2). The least asked families (most marginalised) are unlikely to be on these lists.

*Retention (including attrition)*

It is recommended that, because retention of recruited participants could be an issue, attrition mitigation strategies should be put into place at the planning stage of the research. There is no evidence from the scoping review that financial incentives would help to reduce potential attrition from recruitment to data collection, although these could be considered, along with other possible strategies, such as enhanced communication and follow-up interviews. It is further recommended that eligibility criteria are explicit and clear, again at the planning stage, so that recruited participants are not withdrawn during the data collection cycle.

*Data collection and data analysis*

It is recommended that future qualitative longitudinal research with seldom heard families plans to obtain the voices of both parents and children. As data collection is likely to be more challenging when the participants are young (preverbal) and/or experience communication difficulties or have limited verbal abilities (because they experience learning disabilities, for example), exploration of the appropriateness of a range of participatory data collection strategies is recommended. For example, innovative photographic and observation methods of data collection were well suited to one study in the review researching seldom heard families. For all data collection methods, these should be piloted and fine-tuned to elicit relevant data from all participants.

*Ethics*

The reviewers recommend as a minimum any future research with these populations (as with any research population) should state explicitly that best practice ethical guidelines (for example BERA guidelines) have been implemented at every stage of the research, including: describing in detail the process of obtaining ethical approval for the research and all data protection (GDPR) measures; and providing protocols for dealing with potential distress caused by the data collection process. In addition, details of data archiving should be planned and recorded.

**References**

**Background**

Bryman, A. (2018). *Social Research Methods.* Oxford University Press.

Hong QN, Pluye P, Fabregues P, Gagnon M-P, Griffiths F, Nicolau B, O’Cathain A, Rousseau M-C, Vedel I. Mixed Methods Appraisal Tool (MMAT), version 2018. Registration of Copyright (#1148552), Canadian Intellectual Property Office, Industry Canada

Neale, B. (2016). Introduction: Young fatherhood: Lived experiences and policy challenges. *Social Policy and Society*, *15*(1), 75-83

Neale, B. (2021). The craft of qualitative longitudinal research: the craft of researching lives through time. *The Craft of Qualitative Longitudinal Research*, 1-388

Pearson, H. (2016). *The life project: The extraordinary story of 70,000 ordinary lives*. Catapult

Rimfeld, K., Malanchini, M., Spargo, T., Spickernell, G., Selzam, S., McMillan, A., ... & Plomin, R. (2019). Twins early development study: A genetically sensitive investigation into behavioral and cognitive development from infancy to emerging adulthood. *Twin Research and Human Genetics*, *22*(6), 508-513

Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647

**Sub-review 1: Studies included in in-depth review**

Ballard LM, Jenkinson E, Byrne CD, Child JC, Inskip H, Lokulo-Sodipe U, Mackay DJG, Wakeling EL, Davies JH, Temple IK and Fenwick K Experiences of adolescents living with Silver-Russell syndrome, *Archives of Disease in Childhood* 2021;106:1195–1201

Bostrom PK and Broberg M (2014) Openness and avoidance – a longitudinal study of fathers of children with intellectual disability *Journal of Intellectual Disability Research* 58(9) pp. 810-21

Cox JS, Semple C, Augustus R, Wenn M, Easter S, Broadbent R, Giri D and Hinton EC Qualitative parental perceptions of a paediatric multidisciplinary team clinic for Prader-Willi syndrome *Journal of Clinical Research in Pediatric Endocrinology* 2021;13(4):439-445

Deakin K and Jahoda A, A supporting role: Mothers’ perceptions of their child’s developing awareness of Down syndrome *Journal of Applied Research in Intellectual Disabilities* 2020 33:1380-1389

Gillooly AE, Riby DM, Durkin K and Rhodes SM (2022) *Journal of Autism and Developmental Disorders*

Gillooly,A Atypicalities of Social Functioning in Children with Williams Syndrome University of Strathclyde School of Psychological Sciences and Health PhD thesis 2018

Holm KG, Neville AJ, Peirini A, Bielenska AL, Jamry-Dziuria, Cavero-Carbonell C, Garne E and Clemenson J The voice of parents of children with a congenital anomaly – A EUROlinkCAT study *Frontiers in Pediatrics* Volume 9 Article 654883 November 2021.

Kendall L (2019) Supporting children with Down syndrome within mainstream education settings: parental reflections *Education 3-13*, 47:2, 135-147.

King, N. (2012). Doing template analysis. *Qualitative organizational research: Core methods and current challenges*, *426*, 426-450.

Lightfoot L & Bond C (2013) An exploration of primary to secondary school transition planning for children with Down’s syndrome, *Educational Psychology in Practice*, 29:2, 163-179

Ridding A and Williams J Being a dad to a child with Down’s syndrome: Overcoming the challenges to adjustment *Journal of Applied Research in Intellectual Disability* 2019:32;678-690

Williamson I. (2019) *‘I am everything but myself’*: exploring visual voice accounts of single mothers caring for a daughter with Rett syndrome *Qualitative Research in Psychology* 16:4, 566-590

**Sub-review 2: Studies included in in-depth review**

Bunston W, Frederico M and Whiteside M (2021) The experience of the infant entering refuge (shelter) setting after fleeing family violence, Journal of Family Violence, 36 pp. 953-965

Chester J and Joscelyne T “I Thought It Was Normal”: Adolescents’ Attempts to Make Sense of Their Experiences of Domestic Violence in Their Families *Journal of Interpersonal Violence*, Vol 36 Issue 11-12

Izaguirre A and Kaalstrom A (2021) Differences in the reactions of adolescents to family violence *Child and Family Social Work* 26;425-433

Katz E (2015) Recovery-Promoters: Ways in which Children and Mothers Support One Another's Recoveries from Domestic Violence, *The British Journal of Social Work*, Volume 45, Issue suppl\_1, December 2015

McCarthy M (2019) ‘All I wanted was a happy life’: the struggles of women with learning disabilities to raise their children whilst also experiencing domestic violence *Journal of Gender Based Violence* 3 (1) pp.101-118

Meyer S & Stambe R-M (2022). Mothering in the Context of Violence: Indigenous and Non-Indigenous Mothers’ Experiences in Regional Settings in Australia. *Journal of Interpersonal Violence*, 37(9-10)

Miranda JK, Olivares N and Crockett MA (2022) Growing up with intimate partner violence at home: Adolescents’ narratives on their coping strategies *Journal of Family Violence* 38 pp. 105-116

\*Moe AM & Kothari C L (2023). Perinatal Intimate Partner Violence: A Qualitative Analysis of Mothers Perceptions of Early Childhood Exposure. *Violence and Gender*, 10(2), 85-90

Morrison F (2014) ‘All Over Now?’ The Ongoing Relational Consequences of Domestic Abuse through Children’s Contact Arrangements *Child Abuse Review* 24: 274-284

Scrafford, K. E., Miller-Graff, L. E., Umunyana, A. G., Schwartz, L. E., & Howell, K. H. (2022). “I Did It to Save My Children”: Parenting Strengths and Fears of Women Exposed to Intimate Partner Violence. *Journal of Interpersonal Violence*, 37(9-10)

\*unobtainable

**Appendices**

**Appendix A**

*Full search strategy for MEDLINE for Sub-review 1*

**Database: Ovid MEDLINE(R) ALL <1946 to May 01, 2023>
Search Strategy:**

1 Intellectual Disability/ (59547)

2 (intellectual$ adj2 (disabilit$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or incapacit$ or difficult$ or handicap$)).ti,ab. (27304)

3 (cogniti$ adj2 (disabilit$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or incapacit$ or difficult$ or handicap$)).ti,ab. (142481)

4 (intelligen$ adj2 (disabilit$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or incapacit$ or difficult$ or handicap$)).ti,ab. (613)

5 ((low or reduced or decreased or limited or subnormal$ or sub-normal$) adj2 (intellect$ or cognitive or cognition$ or intelligen$)).ti,ab. (6682)

6 mental$ retard$.ti,ab. (33733)

7 1 or 2 or 3 or 4 or 5 or 6 (235626)

8 exp Learning Disabilities/ (23317)

9 (learning adj2 (disabilit$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or difficult$)).ti,ab. (25541)

10 (dyslexia$ or alexia$ or dyscalculia$ or acalculia$ or dysgraphia$ or agraphia$).ti,ab. (7992)

11 8 or 9 or 10 (43302)

12 Persons with Mental Disabilities/ (3694)

13 (mental$ adj2 (disabilit$ or disable$ or handicap$ or impair$ or deficit$ or deficien$ or incapacit$ or subnormal$ or sub-normal$)).ti,ab. (12731)

14 12 or 13 (15880)

15 Developmental Disabilities/ (22248)

16 (developmental$ adj (disabilit$ or delay$ or deviat$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or incapacit$ or difficult$ or handicap$)).ti,ab. (36586)

17 (delay$ adj4 cogniti$ adj4 develop$).ti,ab. (756)

18 (severe neurological impairment$ or SNI).ti,ab. (1659)

19 15 or 16 or 17 or 18 (51843)

20 7 or 11 or 14 or 19 (318666)

21 exp Chromosome Aberrations/ (162484)

22 Chromosome Disorders/ (20370)

23 (chromosom$ or gene or genes or genetic$).ti,ab. (3319911)

24 21 or 22 or 23 (3367359)

25 20 and 24 (54671)

26 ((rare or unique) adj3 chromosom$ adj3 (disabilit$ or disable$ or impair$ or disorder$ or dysfunction$ or deficit$ or deficien$ or incapacit$ or difficult$ or handicap$)).ti,ab. (183)

27 25 or 26 (54776)

28 cri-du-chat syndrome/ or de lange syndrome/ or down syndrome/ or exp mental retardation, x-linked/ or prader-willi syndrome/ or rubinstein-taybi syndrome/ or trisomy 13 syndrome/ or wagr syndrome/ or williams syndrome/ (48741)

29 jacobsen distal 11q deletion syndrome/ or klinefelter syndrome/ or turner syndrome/ or trisomy 18 syndrome/ (12068)

30 sex chromosome disorders/ or "sex chromosome disorders of sex development"/ (902)

31 (down$ syndrome or trisomy 21 or trisomy21).ti,ab. (28342)

32 (edward$ syndrome or trisomy 18 or trisomy18).ti,ab. (2758)

33 (isodicentric chromosome 15 syndrome or Idic15 or Idic 15 or 15q duplication syndrome or dup15q or marker chromosome 15 syndrome or inverted duplication 15 or inv dup 15 or invdup 15 or invdup15 or inv dup15).ti,ab. (218)

34 (turner$ syndrome or bonnevie-ullrich syndrome or gonadal dysgenesis or monosomy X or XO syndrome).ti,ab. (9480)

35 (patau$ syndrome or trisomy 13 or trisomy13 or 13 trisomy or 13trisomy or ullrich-feichtiger$ syndrome or bartholin-patau syndrome).ti,ab. (1931)

36 (jacobsen$ syndrome or 11q deletion or 11q terminal deletion or 11q monosomy or 11qmonosomy).ti,ab. (374)

37 (klinefelter$ syndrome or XXY syndrome or XXY trisomy).ti,ab. (3430)

38 (XXX or triple X syndrome or trisomy X).ti,ab. (2110)

39 (sex chromosome adj (aneuploid$ or disorder$ or abnormalit$ or aberration$)).ti,ab. (1309)

40 ((cri-du-chat or cat cry or crying cat or 5p deletion or 5p minus or 5p) adj syndrome$).ti,ab. (539)

41 (de lange$ syndrome$ or brachmann syndrome$ or typus degenerativus amstelodamensis or CdLS$).ti,ab. (1138)

42 X linked mental retardation.ti,ab. (999)

43 (adrenoleukodystrophy or X ALD).ti,ab. (2265)

44 coffin lowry syndrome.ti,ab. (230)

45 ((fragile X or marker X or Martin Bell or FRAX$) adj syndrome$).ti,ab. (5461)

46 (glycogen storage disease adj2 (type IIb or IIb or type 2B)).ti,ab. (1)

47 (GSD-IIb or GSD2 or Danon disease or Antopol disease).ti,ab. (268)

48 (lesch-nyhan adj (disease or syndrome)).ti,ab. (993)

49 ((menkes or kinky hair or steely hair) adj (syndrome or disease)).ti,ab. (1127)

50 (mucopolysaccharidosis II or hunter$ syndrome).ti,ab. (939)

51 ((pyruvate dehydrogenase complex or (PDH or PDHC)) adj deficiency).ti,ab. (231)

52 (rett$ adj (syndrome or disorder)).ti,ab. (3885)

53 ((prader willi or prader labhart willi or royer$) adj syndrome).ti,ab. (3587)

54 ((rubinstein-taybi or rubinstein) adj syndrome).ti,ab. (4)

55 ((WAGR adj (syndrome or complex)) or 11p deletion syndrome).ti,ab. (188)

56 ((williams or williams beuren) adj syndrome).ti,ab. (1864)

57 DiGeorge Syndrome/ or Angelman Syndrome/ or Holoprosencephaly/ or Smith-Magenis Syndrome/ or Sotos Syndrome/ or Wolf-Hirschhorn Syndrome/ (5747)

58 (di george or digeorge).ti,ab. (1903)

59 angelman syndrome.ti,ab. (1645)

60 (holoprosencephal$ or arhinencephal$).ti,ab. (2169)

61 (smith-magenis syndrome or (17p adj2 (monosomy or syndrome))).ti,ab. (466)

62 ((sotos adj (syndrome or sequence)) or cerebral gigantism).ti,ab. (584)

63 (wolf hirschhorn syndrome or (4p adj2 (syndrome or monosomy))).ti,ab. (643)

64 or/28-63 (92427)

65 27 or 64 (136639)

66 exp Child/ (2140276)

67 Adolescent/ (2209789)

68 exp Infant/ (1246560)

69 Young Adult/ (1008398)

70 (child$ or infant$ or infancy or pediat$ or paediat$ or preschool$ or pre school$ or schoolchild$ or school age$ or schoolage$ or schoolboy$ or schoolgirl$ or student$).ti,ab. (2453160)

71 (girl or girls or boy or boys or kid or kids).ti,ab. (271389)

72 (adolescen$ or puberty or prepuberty or pubescen$ or prepubescen$ or teen$ or youth$ or preteen$ or juvenil$).ti,ab. (533400)

73 (young adj (person or persons or people or adult or adults)).ti,ab. (147333)

74 66 or 67 or 68 or 69 or 70 or 71 or 72 or 73 (5332164)

75 65 and 74 (71948)

76 exp Qualitative Research/ (81113)

77 Interview/ (30621)

78 Focus Groups/ (35574)

79 (interview\* or focus group\* or qualitative or ethnograph\* or fieldwork or field work or key informant).ti,ab. (673771)

80 ((semi-structured or semistructured or unstructured or informal or in-depth or indepth or face-to-face or structured or guide$) adj3 (discussion\* or question\*)).ti,ab. (44189)

81 Interviews as Topic/ (66817)

82 Narration/ (10059)

83 ((theme$ or thematic$) adj2 analys$).ti,ab. (51473)

84 or/76-83 (747574)

85 75 and 84 (1515)

86 ((parent$ or family or families or mother$ or father$ or mum$ or dad$ or caregiver$ or care-giver$ or grandparent$ or sibling$ or brother$ or sister$) adj6 experience$).ti,ab. (42719)

87 ((child$ or adolescen$ or young person$ or young people or young adult$) adj6 experience$).ti,ab. (60332)

88 86 or 87 (92298)

89 75 and 88 (800)

90 85 or 89 (2129)

91 limit 90 to (english language and yr="2013 -Current") (1034)

 **Appendix B**

*Full search strategy for MEDLINE for sub-review 2*

**Database: Ovid MEDLINE(R) ALL <1946 to June 19, 2023>
Search Strategy:**

1 Domestic Violence/ (7638)

2 Intimate Partner Violence/ (5949)

3 Spouse Abuse/ (7574)

4 1 or 2 or 3 (19763)

5 Family/ (84597)

6 exp Parents/ (141589)

7 Parenting/ (21146)

8 5 or 6 or 7 (230852)

9 4 and 8 (1662)

10 ((domestic$ or famil$) adj6 (abus$ or violen$) adj6 (parent$ or mother$ or father$ or mum$ or dad$ or stepmother$ or stepfather$ or stepmum$ or stepdad$ or caregiver$ or care-giver$)).ti,ab. (1278)

11 ((domestic$ or famil$) adj6 (abus$ or violen$) adj6 (wife or wives or husband or husbands or partner or partners or spous$ or marital or marriage$)).ti,ab. (1183)

12 (intimate partner$ adj6 (abus$ or violen$) adj6 (family or families or parent$ or mother$ or father$ or mum$ or dad$ or stepmother$ or stepfather$ or stepmum$ or stepdad$ or caregiver$ or care-giver$)).ti,ab. (813)

13 (IPV adj6 (family or families or parent$ or mother$ or father$ or mum$ or dad$ or stepmother$ or stepfather$ or stepmum$ or stepdad$ or caregiver$ or care-giver$)).ti,ab. (834)

14 (intimate partner$ adj6 (abus$ or violen$) adj6 (wife or wives or husband or husbands or spous$ or marital or marriage$)).ti,ab. (179)

15 (IPV adj6 (wife or wives or husband or husbands or spous$ or marital or marriage$)).ti,ab. (228)

16 ((interparent$ or inter-parent$) adj3 (abus$ or violen$)).ti,ab. (195)

17 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 (4759)

18 exp Child/ (2148979)

19 Adolescent/ (2214498)

20 exp Infant/ (1250208)

21 (child$ or infant$ or infancy or pediat$ or paediat$ or preschool$ or pre school$ or schoolchild$ or school age$ or schoolage$ or schoolboy$ or schoolgirl$ or student$).ti,ab. (2470365)

22 (girl or girls or boy or boys or kid or kids).ti,ab. (272877)

23 (adolescen$ or puberty or prepuberty or pubescen$ or prepubescen$ or teen$ or youth$ or preteen$ or juvenil$).ti,ab. (538024)

24 18 or 19 or 20 or 21 or 22 or 23 (4908061)

25 17 and 24 (3747)

26 Exposure to Violence/ (1126)

27 4 and 24 and 26 (209)

28 ((child$ or girl or girls or boy or boys or kid or kids or preschool$ or pre school$ or adolescen$ or teen$ or youth$ or juvenil$) adj6 (expos$ or witness$)).ti,ab. (48711)

29 ((abus$ or violen$) adj3 (domestic$ or famil$ or intimate partner$ or spous$ or parent$ or interparent$)).ti,ab. (24741)

30 IPV.ti,ab. (8621)

31 29 or 30 (26797)

32 28 and 31 (1928)

33 ((child$ or girl or girls or boy or boys or kid or kids or preschool$ or pre school$ or adolescen$ or teen$ or youth$ or juvenil$) adj3 (abus$ or violen$) adj3 (home$ or household$ or living)).ti,ab. (411)

34 25 or 27 or 32 or 33 (5271)

35 exp Qualitative Research/ (82039)

36 Interview/ (30670)

37 Focus Groups/ (35760)

38 (interview\* or focus group\* or qualitative or ethnograph\* or fieldwork or field work or key informant\*).ti,ab. (681525)

39 ((semi-structured or semistructured or unstructured or informal or in-depth or indepth or face-to-face or structured or guide\*) adj3 (discussion\* or question\*)).ti,ab. (44800)

40 Interviews as Topic/ (66816)

41 Narration/ (10126)

42 vignett$.ti,ab. (13988)

43 ((theme$ or thematic$) adj2 analys$).ti,ab. (52709)

44 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 (768035)

45 34 and 44 (1240)

46 limit 45 to (english language and yr="2013 - 2023") (721)

47 (comment or editorial or letter).pt. (2167488)

48 46 not 47 (721)

49 exp animals/ not humans/ (5133116)

50 48 not 49 (721)

 **Appendix C**

*Data extraction template for sub-review 1 & 2*

|  |  |
| --- | --- |
| DATA EXTRACTION |  |
| Accession number (EndNote library) |  |
| Author, date, country, setting |  |
| Topic |  |
| Participants and identification: population; target and achieved sample size; and sampling strategy(ies) |  |
| Participant incentives |  |
| Design (longitudinal or cross-sectional) |  |
| Recruitment and retention |  |
| Attrition: attrition mitigation; actual attrition |  |
| Data collection methods and measurement |  |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) |  |
| Comments |  |

**Appendix D**

*Quality appraisal template (from Hong et al, 2018) for Sub-review 1 and Sub-review 2*

|  |  |  |
| --- | --- | --- |
| QUALITY APPRAISAL |  | \*Yes; No; Unclear |
| Accession number (EndNote library) |  |  |
| Author, date, country, setting |  |  |
| \*Research questions  | Are there clear RQs? |  |
| \*Qualitative design and methods | Are the design and methods adopted appropriate? |  |
| \*Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? |  |
| \*Findings | Are the findings adequately derived from the data? |  |
| \*Interpretation | Is the interpretation of results sufficiently substantiated by data? |  |
| \*Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? |  |
| **Summary comments** |  |  |
| **Challenges** in design and conduct (as judged by **reviewers**) |  |  |
| Elements of **best practice** in design and conduct (as judged by **reviewers**)  |  |  |
| Strategies for **overcoming challenges** (as judged by **reviewers**) |  |  |

**Appendix E**

*Data Extraction Tables Sub-review 1*

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and LF 04/09/’23** |
| Accession number (EndNote library) | 2902 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Ballard LM, Jenkinson E, Byrne CD, Child JC, Inskip H, Lokulo-Sodipe U, Mackay DJG, Wakeling EL, Davies JH, Temple IK and Fenwick K Experiences of adolescents living with Silver-Russell syndrome, Arch Dis Child 2021;106:1195–1201. doi:10.1136/archdischild-2020-321376.UKNot stated (reviewer assumes home). |
| Topic(s)Aim(s)Research question(s) | Silver-Russell syndrome (SRS); psychosocial impact of SRS; lived experiences; healthcare and support.To explore the lived experience of adolescents with SRS.Not explicitly stated but can be inferred from the aims. |
| Participants and identificationPopulationSample: target sample stated with justification? Achieved sample; sampling strategy(ies) | ‘Eight participants (five girls) aged 13–18 years were interviewed…Six participants were recruited from a broader study (of nine adolescents, six agreed to participate and three were uncontactable) (2) and two from a patient support group (4). The number of participants declined is not known as adolescents were asked to approach the researcher if they were interested in taking part.’ (p.1197). Population not known.Target sample size not stated. Achieved sample: 8Sampling strategy: unclear |
| Participant incentives | Not stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Recruitment via: ‘(1) study information disseminated by the Child Growth Foundation, UK; (2) prior recruitment to an existing national study ‘Imprinting Disorders: finding out why’ with expressed interest in further research; (3) review of positive molecular genetics tests at the Wessex Regional Genetics Laboratory; and (4) contact with regional genetic and paediatric endocrine centres at existing genetics research sites.’ (p.1196).For (1) and (2) ‘…participants were sent information by post and invited to contact the study team or were contacted by telephone if they had previously given permission.’ (p.1196). For (3) and (4) the patient’s overseeing clinician was contacted and asked to post study information to their patient’. (p.1196-7).Not stated |
| Attrition mitigationActual attrition | Not statedNone  |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Interviews undertaken by ‘a trained female health psychologist (LMB) experienced in discussing sensitive health issues with patients’ (p.1197). Interviews ranging between 30-60 minutes. No interview schedule provided. Not stated if in person or online. Parents potentially in gatekeepers’ position, but none declined on their child’s behalf. Two adolescents interviewed with parents present.LimitationsSmall sample, lacking in diversity (p.1197), no recommendations for improving data collection |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical approval for the study was granted by the NHS Research Ethics Committee South Central – Hampshire B (REC reference: 13/SC/0630). ‘Research and development approval was secured from University Hospital Southampton National Health Service Foundation Trust (study sponsor) and, for the 23 UK Genetics Centres, via the National Institute for Health Research UK Rare Genetic Disease Research Consortium Agreement.’ (p.1196). ‘As SRS is a rare condition and individuals may be known through support groups, robust confidentiality measures were instituted, including numerical participant identification and the use of age ranges.’ (p.1197)‘All participants were provided with an information sheet and the opportunity to discuss the research and ask questions. Written consent/assent was gained from adolescents and their parents. Participants were reassured that they could discontinue the interview at any time and that it would be audio recorded, but this could be stopped at their request.’ (p.1197) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No but can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes, small sample is acknowledged and data not over-generalised. |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample. Recruitment was challenging. Design was appropriate but methods details are lacking. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**)  | Issues of sensitivity addressed. Included a member of a charity who had personal experience of a growth condition at all stages of the project.  |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Provide clearer information on recruitment and interview design. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and LF 04/09/’23** |
| Accession number (EndNote library) | 3477 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Bostrom PK and Broberg M (2014) Openness and avoidance – a longitudinal study of fathers of children with intellectual disability *Journal of Intellectual Disability Research* 58(9) pp. 810-21.SwedenUniversity premises; children’s clinic; fathers’ homes. |
| Topic(s)Aim(s)Research question(s) | Autism, Down Syndrome, Chromosome 14 Deletion, Mosaic Trisomy 14 Syndrome; fathers’ personal experiences.To explore fathers’ experiences of parenthood in relation to a child with ID or DD from the initial diagnosis of the disability to 5 years later.Not stated but can be inferred from aim. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 7 fathers of children with ID or DD (aged 5 months to 5 years).Children diagnosed with autism (3), Down Syndrome (2), Chromosome 14 Deletion (1) and Mosaic Trisomy 14 Syndrome (1).Unclear, reviewer assumes all those eligible who were contacted (number not stated).Target sample size not stated; nature of sample justified (shared parental/household responsibilities); sampling strategy inferred – all those eligible who consented. Actual sample n=7. |
| Participant incentives | Ten Euros gift voucher |
| Design (longitudinal or cross-sectional) | Longitudinal (2005-10)Data collection at three time points: T1, T2 and T3 |
| Recruitment Retention | Recruitment through community-based clinics providing services for children with disabilities (part of a mailed-out questionnaire for a larger study). After written consent was received an interview was undertaken.Not stated |
| Attrition mitigationActual attrition | Not stated, but small incentive offered.No attrition, but one father excluded after 3 years when a diagnosis of ID was ruled out. |
| Data collection methods and measurementStrengths and Limitations; future recommendations for data collection | Semi-structured interviews – total of three at 3 time points – 40-90 minutes, using a guide – positive and negative aspects of present life situation; changes in perceptions of themselves and their life situation in relation to the child and the father-child relationship. Four questions from Working Model of the Child Interview (WMCI). (p.813). Fathers could pick the location of the interview and the same interviewer was used for all timepoints. Interviews audio recorded.LimitationsHeterogeneous sample – experiences may have been affected by diversity of child characteristics such as age and diagnosis. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Written consent.No other details. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No, but can be inferred from aim. |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small, heterogeneous sampleRecruitment strategy Few ethical details – no discussion of ethical approval or data sharing or sensitivity of topics.No information on the analysis from a longitudinal perspective.Used multiple coders for IPA with no discussion of if this might be aproblem and no positionality statement. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) |  |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Expand recruitment strategy.Clearer explanation for how time points were linked needed. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF 24/08/’23** |
| Accession number (EndNote library) | 254 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Cox JS, Semple C, Augustus R, Wenn M, Easter S, Broadbent R, Giri D and Hinton EC Qualitative parental perceptions of a paediatric multidisciplinary team clinic for Prader-Willi syndrome *Journal of Clinical Research in Pediatric Endocrinology* 2021;13(4):439-445.UKOutpatient ward of large community hospital; private appointment room, adjacent to clinical team. |
| Topic(s)Aim(s)Research question(s) | Prader-Willi syndrome (PWS); supporting families with children with PWSTo undertake a qualitative review of a pilot clinic to support families of children with PWS.Not explicitly stated but can be inferred from objectives.Interviews sought to explore parents’ experience of the MDT clinic compared with their previous care and understand the areas of greatest need for families. They sought to engage parents in the design of both the structure and the content of the clinic, and feedback was requested to facilitate co-design. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Families selected from the regional database of children with PWS n=6 in the South-West of England. Families included typically attended a large number of appointments with a range of health professionals.Population: Regional databaseAchieved sample: 6 families who confirmed their willingness to attend a pilot clinic. Target sample not stated. Strategy not stated. |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Unclear. Families selected from database.Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection | Researchers who conducted the interviews were external to the clinical team and were accompanied by a social worker. All families interviewed after their clinic appointment; two families interviewed before and after their appointment (n=8 interviews in total). Interviews audio recorded using a Dictaphone.StrengthsResearchers were external although division was not absolute; researchers' in-depth understanding may have influenced responses.LimitationsFive of the families were previously engaged with treatment. Further work with disengaged families is recommended. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | No ethical approval was required. The Patient Experience and Involvement Team at University Hospitals Bristol approved the research and all interviewees provided informed consent. Participant anonymity preserved through the use of pseudonyms.Parents informed about the clinic and were consented prior to interviews. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No but can be inferred from objectives |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes (limitations of the sample are clear) |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains except RQs which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Very small sample size; participants previously known to researchers who were not completely independent. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Interviews transcribed verbatim by externally approved servicesand anonymised. Transcripts independently coded by two researchers, both researchers met to refine coding (iterative analysis process). |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Larger sample size; greater independence in data collection and analysis. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF 24/08/’23** |
| Accession number (EndNote library) | 351 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Deakin K and Jahoda A, A supporting role: Mothers’ perceptions of their child’s developing awareness of Down syndrome *Journal of Applied Research in Intellectual Disabilities* 2020 33:1380-1389.UKHome setting |
| Topic(s)Aim(s)Research question(s) | Down’s Syndrome; child’s awareness of disability.To explore mothers of Down’s Syndrome children’s views and experiences of helping their children to understand their Down’s Syndrome (p.1381); family life; children’s relationships with others (siblings and school peers); stigma family may have experienced; discriminatory attitudes; mothers’ hopes for children’s futures.Not stated but implicit in aims |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Nine mothers of children with Down syndrome (5 daughters, 4 sons aged 9 – 16). Population not clearTarget sample size 9 (not justified); achieved sample size 9; sampling strategy – first 9 mothers contacted who agreed to take part. |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | ‘Families were contacted through schools in the West of Scotland and Down Syndrome Scotland, a charitable organization…’ (pp.1381-2).Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection | Semi-structured interviews (45-90 minutes in length) sought mothers’ views using (but not limited to) an interview topic guide.IPA analysis of interview dataLimitationsSmall sample‘…this study neglected the perspectives of fathers on their child's developing sense of self. Future research would do well to include both parents and perhaps interviewing couples would enable additional insights. A future study that purposefully sampled parents of children with Down Syndrome in mainstream and special school might also provide important insight into how wider social factors help to shape their views about their offspring with Down Syndrome's developing sense of self. Future research should also perhaps consider how knowledge of negative social attitudes is negotiated between families and their offspring, rather than assuming that children lack insight and families are simply protective. A longitudinal ethnographic approach may provide insight into how children's sense of self emerges from their experiences both inside and outside of the family’ (p.1388). |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical permission obtained from the ethics committee of the University of Glasgow. Participants gave permission to be interviewed at home; permission was obtained for interviews to be recorded. Demographic information collected after interview and audio recorder switched off. Interviews guide semi structured so that the interviewer could remain alert to other topics or concerns raised by the participants and explored these when they arose. Researcher ensured the development of rapport with each participant before asking about their child’s relationships with others. Pseudonyms for the mothers and their children were used throughout process. Interview data transcribed verbatim. Interpretative phenomenological analysis (IPA) justified due to having been successfully used in previous research concerning highly sensitive topics with parents of people with intellectual disabilities. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Not stated; can be inferred by reviewers from aims |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** |  Meets threshold for quality (‘Yes’ or implicit in all domains). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample; target sample size not justified.The social barriers children may face were rarely mentioned and the stigma associated with Down’s Syndrome did not feature at all.Fathers were not purposively sampled. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Method of data analysis – interpretative phenomenological analysis (IPA).The researcher took the following steps to ensure the analyses were carried out rigorously. (a) a reflective journal was used to chart the process of drawing out and integrating themes across the transcripts, (b) a summary of each interview was produced to help ensure the themes reflected the complexity and detail of the participants’ narratives as a whole. (c) the interviewer recorded reflections after each interview to allow the emotional reactions of the interviewees to inform the interpretation of the transcripts, (d) an experienced IPA researcher oversaw and audited the process of analysis, this included double coding initial transcripts anddiscussing all the emerging themes from across the sample of participants, and (e) finally, care was taken to return to the originaltranscripts to check the themes were grounded in examples from the interviews. |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Larger sample size, well justified in respect of obtaining greater representativeness and generalisability. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT KA DF LF 07/08/’23** |
| Accession number (EndNote library) | 20 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Gillooly AE, Riby DM, Durkin K and Rhodes SM (2022) *Journal of Autism and Developmental Disorders* &Gillooly,A Atypicalities of Social Functioning in Children with Williams Syndrome University of Strathclyde School of Psychological Sciences and Health PhD thesis 2018UK (Scotland, North England, North Wales)Home visits |
| Topic(s)AimRQ(s) | Williams Syndrome (WS); friendships and friendship difficultiesTo examine ‘…the perspectives of children with WS and their parents using semi-structured interviews to obtain rich qualitative insights into the characteristics of friendships in children with WS’.Journal: Not explicitly stated (can be inferred by reviewer)PhD thesis: Stated: ‘What are the experiences of peer relationships in children with Williams syndrome?’ |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | n = 21 parents and n = 21/20 children (7-16 years; mean = 11.83 years; 12 F, 9M)Identification through the Williams Syndrome Foundation UK charityAll families who had a child with WS aged between 6 and 16 and lived in Scotland, North England and North Wales (n = 79) were invited to participate.Sample: 21 parents and 21 children, 27% of 79 invited to participate (78% north/central England, 24% Scotland, 5% Wales).Justification: Sample size in line with similar previous research in this topic area.Sampling strategy – population, i.e., all those eligible. |
| Participant incentives | None stated |
| Design  | Cross-sectional |
| Recruitment Retention | 21 children and their parentsFamilies were members of the Williams Syndrome Foundation UK charity and had previously provided permission to be contacted regarding participation in research studies. Families were contacted by letter and invited to participate. Information was provided in the participant information sheets (parent and child).Not stated |
| Attrition mitigationActual attrition | Not statedNone, although one child unable to take part in interview due to communication difficulties. |
| Data collection methods and measurementLimitations and future recommendations for data collection (authors) | Semi-structured interviews audio-recorded during home visits.Two bespoke questionnaires were developed (Appendix 1 and Appendix 2 in Supplementary Materials): (adapted interview schedule Cuckle, P., & Wilson, J. (2002). Social relationships and friendships among young people with Down’s syndrome in secondary schools. *British Journal of Special Education*, 29(2), 66–71.Parent schedule = 14 items (quality of friendships, appropriateness, level of contact, social inclusion participation extra-curricular clubs). Child schedule = 13 items (measuring perceptions of quality of friendships, extra-curricular engagements, understanding of term ‘friendship’). Interviews took place with 11 mothers, 2 fathers and 8 couples, and 20 children.Data from children was limited due to data collection method; data could not be analysed in the planned way. During child interviews many of the children had limited verbal skills and struggled to pay attention. One child unable to be interviewed due to communication difficulties. ‘It was not possible to conduct an in-depth qualitative analysis on this data due to the shorter nature of the responses. Future research should explore this further by using a range of accessible participatory methods to further capture the voices of the children themselves and better understand what is important to children with WS within a friendship. Future research is also needed to identify optimal ways to support children with WS in their social interactions, targeting the identified areas of difficulty, while considering the evident individual variability.’ |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical approval from University Ethics Committee.Obtained consent to participate and to record. Choice about whether parent present.Participant Information Sheets: Parent and Child explained pseudonymity and anonymity, and GDPR using standard wording and terminology. |
| **QUALITY APPRAISAL** |  | **Yes, No, Unclear; Comments** |
| Research questions  | Are there clear RQs? | Journal article: No: RQ implicit in aim of studyPhD thesis: Yes: p.220 ‘What are the experiences of peer relationships in children with Williams Syndrome?’ |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Mainly, yes – but see below on inconsistency. Limitations of sample clear |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ or ‘mainly yes’ in all domains) |
| **Challenges** in design and conduct **(as judged by reviewers)** | Small sample size (n =21), so not generalisable; potential for bias in sample due to nature of recruitment strategy (contact through WS Foundation UK) as sample may not represent typical or full range of seldom heard families with WS child.Child data limited – see above in limitations section.27% response rate geographically differential: 78% north/central England, 24% Scotland, 5% Wales.One instance of inconsistency in reporting results: All 20 children reported having at least one best friend (PhD thesis, p.260) and ‘most’ of the children did *not* have a best friend (p.261). This may have been due to giving greater weight to the parental views. |
| Elements of **best practice** in design and conduct **(as judged by reviewers)** | Recruitment through WS Foundation UK (although see above re.: potential bias in sample). Multiple informants recruited (mothers, fathers, couples, children). |
| Strategies for **overcoming challenges** **(as judged by reviewers)** | More accessible, creative, and inclusive data collection methods for the children to obtain deeper understanding of their perspectives.Report consistently and avoid giving give adult voices more weight than children’s voices.Recruit beyond the charity’s list of families interested in research, in addition to using this resource. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF 24/08/’23** |
| Accession number (EndNote library) | 117 |
| Bibliographic details (author(s), date, publication)Country(ies)Research setting | Holm KG, Neville AJ, Peirini A, Bielenska AL, Jamry-Dziuria, Cavero-Carbonell C, Garne E and Clemenson J The voice of parents of children with a congenital anomaly – A EUROlinkCAT study *Frontiers in Pediatrics* Volume 9 Article 654883 November 2021.Denmark, Italy, Poland, SpainInterviews: Region of Southern Denmark (setting not stated)Focus groups: Spain: Barcelona meeting room; Poland: Poznan University and SWPS University of social Sciences Katowice and The Specialist Children’s Hospital in Olsztyn; Italy: Trisomy 21 Onlus Association Florence; St Anna Hospital in Cona Ferrara; Hotel Parma and Congressi, Parma; Heart Hospital ‘Gaetano Pasquinucci’, Massa. |
| Topic(s)Aim(s)Research question(s) | Congenital anomalies: Down syndrome, spina bifida, cleft lip, severe congenital heart defects requiring surgery; parental experiencesTo investigate parental experiences of having a child with a heart defect requiring surgery, cleft lip, spina bifida or Down’s Syndrome and to identify their research priorities. The EUROlinkCAT project aimed to close the gap between research and clinical treatment, and to ensure evidence-based practice where parent’s knowledge and perspectives were included.‘What is important for [parents] having a child born with a congenital anomaly?’‘What should the researcher focus on in upcoming projects?’ |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Parents of children aged between 1 and 10 with one of four pre-defined congenital anomalies. Identification from overarching study: EUROlinkCAT study, parental congenital anomaly groupsNot statedTarget sample not statedAchieved sample: n= 12 parents (interviews); n=58 parents and n=2 caregivers (focus groups). |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional but part of a longitudinal study |
| Recruitment Retention | Via 5 EUROCAT registries in Denmark, Spain, Poland and Italy. Participants were consecutively recruited through an invitation from the collaborating hospital physicians or through patient associations for the included anomalies (p. 2). The rationale for choosing Denmark, Spain, Poland and Italy was to have as broad a perspective as possible in relation to culture, organisation of health system and social circumstances as possible. Inclusion criteria were being biological parent or grandparent of a child aged between one and 10 years old with one of the selected congenital anomalies. Participants had to be able to speak the local language of their country or English. All parents participating in the interviews and focus groups had been regularly in contact with a hospital due to their child’s diagnosis.Not stated |
| Attrition mitigationActual attrition | Not stated but an expectation that last minute dropouts (asparents in the study lead challenging everyday lives) included aminimum of ten parents for each focus group.Attrition from focus groups (number not stated) and therefore two focus groups undertaken with sub-optimal number of participants. |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (according to authors) | Semi-structured interviews: interview guide initially developed in Denmark based on the literature and according to the study aim; themes included daily life.Focus groups: follow-up questions added to interview guide. Interviews and focus groups were audio recorded and transcribed and translated into English.Objective field notes from focus groups were used using transcripts to add to parental statements.StrengthsAll participating parents expressed satisfaction with the experience and energy and insights were obtained as a result. The focus groups were facilitated by strong working relationships between the EUROCAT Registry leaders, clinicians and patient associations.LimitationsData collected in 5 regions in 4 European countries. It was planned to undertake focus groups with parents of all 4 anomaly groups and more focus groups were planned in other countries (Malta, Portugal, England). However, the COVID-19 pandemic halted the continuation of the focus groups. This study looks at the opinions of parents of live parents whose views may be different from parents who decide to terminate pregnancy due to foetal anomaly. Other limitations include cultural, social and health systems heterogeneity; anomaly heterogeneity |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical approval: Danish Data Protection Agency and hospital management approval; normal part of registries’ activities so specific approval not required; no obligation for ethical approval in Poland and Spain. Parents in the four regions received written and oral information about the study. Parents gave written informed consent to participate prior to interviews and could withdraw at any time. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Yes |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality: Yes in all domains |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample sizeA common platform for training of the researchers for conducting focus groups, an interview guide and a template of how to transcribe data were developed to minimise the effect of individual beliefs and assumptions.It was challenging for parents to answer questions about research priorities so follow-up questions were added to the interview guidefor the interviewers to use during the focus groups to support talking and discussion. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Data collection: The focus groups were facilitated by the strong working relationships between EUROCAT Registry leaders, clinicians and patient associations with which they have long-standing relationships. The research group was interdisciplinary consisting of physicians, nurses, and epidemiologists from different countries. This provided the group with different perspectives and pre-understanding. Interviews carried out to test and qualify the interview guide for focus groups. Interview transcripts were shared with parents for feedback.Data analysis: systematic text condensation (STC), a method for descriptive analysis of phenomena for the development of new descriptions and concepts. STC analysis consists of decontextualization and recontextualization; procedure in 4 steps: (I) read transcripts repeatedly to identify themes; (II) identify and code units of meaning; (III) identify sub-groups of codes from step II and develop.Attrition was low, possibly due to the strong positive relationships developed between researchers and participants.Participants were recruited through an invitation either from thecollaborating hospital physicians or through patient associations for the included anomalies; this purposive sampling ensured that the target groups were reached. |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Increase sample size |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and KA 05/09/’23** |
| Accession number (EndNote library) | 1198 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Kendall, L (2019) Supporting children with Down syndrome within mainstream education settings: parental reflections *Education 3-13*, 47:2, 135-147.UK (England)Researcher’s university setting and by telephone  |
| Topic(s)Aim(s)Research question(s) | Down Syndrome; parental perspectives; inclusion in mainstreamNot explicitly stated as aims but can be inferred: to explore ‘parental perspectives, effective practice and barriers, around the inclusion of children with DS in mainstream education, across all Key Stages (early years, primary and secondary school).’ (p.135)Not stated but can be inferred. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | n=5 parents of children with DS aged between 3 and a half to 12 years old (2 in nursery setting, two in primary schools and one in secondary school).Population not explicitly stated but inferred: Children with DS educated in mainstream settings in NW England (who were also members of the NW England branch of the DSA).Target sample size not statedAchieved sample n=5Sampling strategy: purposive sampling |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | ‘An initial approach was made to the Chair-person of the Down syndrome Association (DSA) in the North West of England requesting access to that particular organisation.’ (p.138). Access granted.Research outline posted on the DSA forum.Not clear how the n=5 were selectedNot stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Face-to-face semi-structured interviews (n=4) and one telephone interview. Interviews lasted between 40 minutes and 1 hour 30 minutes. All interviews were audio recorded and transcribed verbatim. Participants sent a copy of their interview to check for accuracy.Thematic data analysis approach.StrengthsTelephone interview – an effective way of gathering data from a ‘hard to reach’ parent.LimitationsSample size is a limitation, as is lack of fathers’ perspectives. It is acknowledged that education experiences of the participants’ children will not be representative of all children with DS. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Research followed 2011 BERA ethical guidelines. No mention of ethics approval.All participants (parents) completed consent forms.Pseudonyms used to protect anonymity of children. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No but can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Reasonable – some over-generalisation |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ or ‘Reasonable’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Sample size is low.Ethical approval not stated; few details around ethical issues e.g., consent etc.The analysis is very descriptive and lacking in depth.  |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) |  |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample sizeObtain ethical approval and/or explicitly state this has been done if this is the case.Add in more details about ethical issues and methods of data collection. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and KA 05/09/’23** |
| Accession number (EndNote library) | 1562 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Lightfoot L and Bond C (2013) An exploration of primary to secondary school transition planning for children with Down’s syndrome, *Educational Psychology in Practice*, 29:2, 163-179.UK (England)Not explicitly stated but the reviewers assume at the two participating schools.  |
| Topic(s)Aim(s)Research question(s) | Down Syndrome; transition from primary to secondary school; inclusion; ‘voice of the child’1. To examine the factors influencing the transition from primary to secondary school for two children with Down’s Syndrome.
2. To consider current inclusive practice in a mainstream primary school and how this might inform successful inclusion at secondary level.

Not stated but can be inferred from aims. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Two children with Down Syndrome and their mothers and learning support staff at the schools. Commissioned by a secondary school who had offered a place to a boy with DS. They (or the Educational Psychologist?) suggested including another child (from another primary school) as a comparison (convenience sample? – the strategy isn’t justified). The second child was in Year 5 and her parents were undecided about her secondary education. Not explicit but the aim seems to have been to help the mainstream secondary school to prepare for the arrival of a Year 7 child with Down Syndrome.Population not explicitly statedTarget sample size not stated.Achieved sample n=2; sampling strategy unclear – but see above in ‘identification. |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Recruitment was via the participating schools, see above.Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations and future recommendations for data collection (as reported by authors). | ‘Semi-structured interviews were conducted using open-ended questions to elicit information from the mothers of both children and the learning support assistant working with each child. These were informed by Appreciative Enquiry’s 4-D cycle and positively framed questioning (Preskill & Catsambas, 2006). Interviews were audio recorded and transcribed. The data from all of the interviews was transcribed verbatim and analysed thematically following the six stages outlined by Braun and Clarke (2006).’Voice of the child interviews: Novel approach to eliciting the voice of the children. Children were asked to use emojis to indicate their preferences, in response to school based images, and to peg those preferences on a washing line to indicate how happy or sad they made them feel. Good rationale provided for this approach, which was tailored to each child. A pre-meeting took place to help the children and researcher build a rapport and Learning Support Assistants were present and joined a general discussion at the end.The researcher led the interviews, but school staff were able to contribute to check understanding. ‘Colourful images relating to differing aspects of school were selected for discussion with the children. The images were then presented one by one and discussion was generated around them to check understanding. Once it was felt that the child had a clear understanding of the topic, the child was asked whether it made him/her feel happy, sad or ok.’LimitationsSocial desirability bias – one Learning Support Assistant wanted toknow what was going to happen to the data and was unsure howhonest she could be. Not clear how the researcher responded.Limitations with child data collection – only able to elicit their viewsto a limited extent. Potential for researcher bias in analysis.No recommendations given for overcoming challenges. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘Consent and agreed collaboration were obtained from key people at the primary and secondary schools and informed consent was obtained from the participants, in keeping with Professional Practice Guidelines (BPS, 2000)’Although it states that participants gave informed consent it is unclear whether this applied to the children unclear whether ethical approval was in place - limited information. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No, but can be inferred from the aims |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes, the interpretation is limited to the sample |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Very small, unrepresentative sample.There is a lack of methodological information and the account lacks precision and detail. No interview schedule for the parents and LSAs.  |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Voice of the child included in the research: Focus on VoC very good with good justification of the approach. The personalisation to individual children was also very good |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Increase and justify the sample size.More tightly focused research questions.More detailed methodological information. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF 24/08/’23** |
| Accession number (EndNote library) | 495 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Ridding A and Williams J Being a dad to a child with Down’s syndrome: Overcoming the challenges to adjustment *Journal of Applied Research in Intellectual Disability* 2019:32;678-690UKHome settings |
| Topic(s)Aim(s)Research question(s) | Down Syndrome (DS); fathers’ experiences‘…to develop a model to account for fathers’ experiences of parenting a child with DS; their contributions; influences on family functioning; and inclusion in their children’s support provision.’ (p. 680).Stated explicitly:‘1. How do fathers adjust to living with a child with DS and what parenting roles do they play?2. What specific factors contribute to their adjustment and how?’ (p.680) |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Fifteen fathers of children with DS (aged under 10 years – 8 months to 8 years).Population not stated.Target sample not stated; achieved sample size n=15Sampling strategy – theoretical sampling (Strauss and Corbin, 1998) to obtain ‘…as much variation as possible in the first few participants, followed by sampling based on concepts and themes emerging from the data in subsequent phases.’ (p.680). |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Support groups within NW England.As above, theoretical sampling to obtain maximum variation in variables such as employment, birth order, age of child: ‘…participants with different demographics….were selected in order to add variation to the data, and to allow the exploration of emerging analytical themes.’ (p.680)Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection | Semi-structured interviews were ‘guided by an initial schedule which provided a broad, flexible approach to exploring fathers’ experiences.’ (681). Interview schedule was piloted with a father of a child with DS who did not meet the eligibility criterion of geographical region.Each participant chose the location of the interview, usually at their home. Average length of interviews was one hour (shortest was 40 minutes). Simultaneous data collection and analysis allowed the constant comparison of themes both within and between participants.The use of Grounded Theory (GT) was considered most congruent with the study aims and allowed the researchers to examine experiences in depth, while contributing to a theoretical understanding and development of a model. Analysis – NVivo for identification and collation of codes. Analysis was undertaken in three stages: open coding; axial coding; selective coding; the coding system was developed for this research. (p.682).StrengthsFirst study to offer understanding of fathers’ adjustment to parenting a child with DS; grounded theory approach to sampling and analysis.LimitationsTheoretical sampling may have limited the generalisability to other populations; sample self-selected (mainly white British fathers in financially stable jobs); possibility of social desirability in the fathers’ responses.In futureExplore fathers’ experiences across varying diagnostic categories (not just grouping all fathers under the umbrella term ID) as specific implications could be lost; aim to reach fathers who may be struggling, fathers not currently residing with partner and child; recruit from other geographical areas and other cultures; explore mothers’ experiences of parenting a child with DS and explore the complexities of parental relationships from their perspectives; undertake replication studies (p. 688). |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical approval granted (University of Liverpool’s Doctorate in Clinical Psychology Research Committee) (p.680; p.681).Fathers were given copies of participant information sheet and signed a consent form at the time of the interview.Confidentiality was maintained throughout; interviews were digitally recorded and transcribed verbatim. Participant names were omitted during recording to preserve anonymity; other names were replaced with pseudonyms post transcription.If participants became distressed during the interviews, they were signposted to appropriate support services. Participants were made aware of their right to withdraw at any time. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Yes |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample and possibly skewed in several demographic variables.Population referred to but not stated. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Theoretical sampling (but see above); grounded theory analysis. |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Different sampling strategy; larger sample size. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and KA 05/09/’23** |
| Accession number (EndNote library) | 1249 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Williamson, I. (2019) *‘I am everything but myself’*: exploring visual voice accounts of single mothers caring for a daughter with Rett syndrome *Qualitative Research in Psychology* 16:4, 566-590UK (East England)Home settings |
| Topic(s)Aim(s)Research question(s) | Rett syndrome; experiences of lone parenting of a Rett syndrome daughter.To offer an insight into lives of women raising a daughter with Rett syndrome, with a view to inform specific interventions for parental caregivers.To make a contribution to discussions about how to use methods that blend photography and phenomenology in disability studies and qualitative research more generally.Not stated but can be inferred from aims. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Biological mothers raising a daughter with Rett syndrome alone n=5 Population not statedTarget sample size not statedAchieved sample n=5. Small sample justified in IPA.Sampling strategy: purposive sampling; participants recruited as ‘experiential experts’ |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Participants recruited in 3 ways: via an online support group hosted on a popular social media site; via a national support agency for families affected by Rett syndrome (Rett, UK); through a poster at a large centre for adults with learning disabilities in a city in the east of England.All participants were based in the east of England - geographical selection not explicitly explained. Selection process of 5 mothers not stated.Part of a larger study which included both single and non-single mothers; link to other study unclear – unknown how many participants there were in the main study, and whether they too were all based in the east of England.Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection | Two modes of data collection:1. Participants took a series of photographic images across a five-day period leading up to the subsequent interview.
2. Face-to-face interviews.

Data were collected by a trainee health psychologist who had prior experience of working with families with Rett syndrome and who received training and supervision in undertaking photo-interviews. Participants were asked to take photos that captured meaningful components of their ‘quotidian caregiving experience’ but not identifiable photos of their daughters or other people. They were asked to only take as many photos as they could comfortably manage and were asked to caption individual images and to choose five for discussion during the interview that took place after 5 days of being asked to take photographs.All participants used cameras in their mobile devices in the five-day period leading up to the interview. Interviews took place in participants’ homes and lasted between one and two hours. Interviews incorporated a discussion of the photos and the use of a loosely structured schedule (not supplied). Interviews were transcribed in full.Interpretative phenomenological analysis (IPA).Participants raised some reservations about their photos, dismissing their efforts as “silly” and saying that the task was ‘hard’ or ‘too abstract’ or that they didn’t do their experiences justice.Participants may feel more sensitive/exposed with this kind of data.Participants were drawn from support groups and may differ from those who don’t access support. They were all white and ‘not in abject poverty’. Larger-scale qualitative research with a more diverse group is needed. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Ethical approval from ‘the relevant committee at the host university’ (p.570) (no details). An information sheet was provided, and a consent form was signed and a short demographic questionnaire was completed before the participants were given a five-day period to take photographs of their caring experiences. Participants gave explicit permission for their anonymised photographs and data extracts to be used.‘A fuller discussion of managing ethical, methodological and epistemological elements of using photos in the content of phenomenological research is available’ (p. 571) but unavailable as labelled ‘Author’ in published paper. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No, but can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains except for RQs which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample size.Unclear from recruitment strategy why all participants were based in the east of England.Unclear how this study fits with the larger study mentioned, or where the additional ethical and methods information can be found.Photography instructions given to participants and interview schedule not included.  |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Innovative photographic methods of data collection are well described and well justified. Method ‘well suited to qualitative research with participants in difficult and/or disadvantaged circumstances’ (p.569). |
| Strategies for **overcoming challenges** (as judged by **reviewers**) | Recruit a larger, more diverse sample.Provide more information on: the links with larger study; geographical recruitment focus; interview schedule; photography instructions etc.When pointing to supplementary information in other papers include full bibliographic details in order that they can be obtained. |

**Appendix F**

*Data Extraction Tables Sub-review 2*

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and EJ agreed 27/11/’23** |
| Accession number (EndNote library) | 95 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Bunston W, Frederico M and Whiteside M (2021) The Experience of the Infant Entering Refuge (Shelter) Setting with Their Mothers After Fleeing Family Violence *Journal of Family Violence* 36: 953-965Australia, Scotland and EnglandRefuges (medium to maximum security) |
| Topic(s)Aim(s)Research question(s) | Refuges; family violence; infant perspectivesTo explore how infants experience refuge in a Refuge settingStated in findings section:What was experienced as refuge for the infant?How are the infant/mother attended to in order to bring the infant into an emotionally regulated and healthy state?How does entry into Refuge impact the infant/mother relationship?How does the infant experience safety (‘refuge’) in a Refuge environment?What knowledge do both staff and mothers have in relation to the needs of infants entering Refuge? |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Ten infants (aged 3 weeks to 16 months) and ten mothers in eight Refuges. ‘Accessing a suitable number of Refuges for this research (all of which were medium to maximum security) was initially problematic and necessitated eventually following up leads across three separate countries. Getting access to the secret and appropriately, well-guarded world of the Refuge eventually came down to using existing contacts and networks the researcher had available to her. Furthermore, it was imperative to recognise the sensitivity of Refuge/Shelter settings and ensure the confidentiality of all research participants and all residents in the Refuge space’ (p.954)‘Nine of the ten infants were under 12 months with the age range 3 weeks to 16 months; also involved were 10. Eight Refuges participated, three in Melbourne and two in remote Australia, one in London, England and two in Glasgow, Scotland’ (p. 956)Population not explicitly statedTarget sample size not statedAchieved sample size 10 infants and 10 mothersSampling strategy not stated |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | ‘All participants were recruited by Refuge staff once the staff themselves deemed the researcher trustworthy.’ (p.954). ‘The mothers then had a brief meeting with the researcher to decide if they would like to participate. Only one mother declined to be involved before her scheduled interview following a disagreement with the Refuge over an unrelated issue.’Not stated |
| Attrition mitigationActual attrition | Not statedNot stated |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Data was collected through infant observation and interviews with mothers. The research involved the collection of observational data and interviews.‘The data collected consisted of 18 (1 h) ‘infant observation’ sessions involving the 10 infants. One infant had been in Refuge previously, two were born in Refuge and over half had been in Refuge for less than 7 days, two less than a month and three between 3 and 12 months (see Bunston 2016, p:105). Of the six infants who recently entered Refuge, one was observed during admission and three others within 3 days of admission. The number of infant observations were determined by the availability of the mother, and the researcher’ (p. 956). Following the infant observation mothers were interviewed (and audio recorded) using a set of semi structured questions. Analysis was undertaken using ‘constructivist grounded theory’ approach.The research was not objective and depended on the researcher’s interpretation.‘The brevity of contact with each infant, mother and Refuge offered only a snapshot rather than a sequential sense of the infants’ experience to any large degree, and the number of participants overall was small. To extrapolate extensively from this under-researched area using this small cohort of participants would be a mistake and was not the intended purpose. The lack of research regarding the infant in the setting of Refuge, even in the increasingly expansive arena of infant mental health, is indicative of the extent to which this environment in which so many infants are accommodated tends to be overlooked. This study offers a starting point for others to follow’ (p.963) |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Fully informed consent.‘University ethics approval for this research was gained, and each Refuge was asked if further amendments were required as appropriate to their setting’ (p.955) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Yes (stated in findings section) |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample size; self-selected sample.The research questions were not stated in the methods section. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | ‘Infant-led’ approach using observation. |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample sizeStating the RQs in the methods section.Giving more detail about the length of the study and reasoning behind the country choices beyond convenience. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF Agreed 24/10/’23** |
| Accession number (EndNote library) | 237 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Chester J and Joscelyne T “I Thought It Was Normal”: Adolescents’ Attempts to Make Sense of Their Experiences of Domestic Violence in Their Families *Journal of Interpersonal Violence*, Vol 36 Issue 11-12UKChild and Family Health service |
| Topic(s)Aim(s)Research question(s) | Adolescents’ thoughts and feelings about domestic violence (DV) experiencesTo describe the experience of being exposed to and living with DV directly from the perspective of children and adolescentsNot explicitly stated but can be inferred from aims of the study |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 5 adolescents (3 female and two male) who had all witnessed or been the victims of parental domestic abuse.‘To be considered for participation, the adolescents needed to have been safe from DV for at least a year prior to the interview (for safeguarding reasons). Participants were accessed via a Child and Family Mental Health service where the second author was working at the time. Possible participants were suggested by clinicians and then approached by the researchers. Five out of the 10 adolescents approached agreed to take part in the study’ (p. 5257)The adolescents’ age ranged from 14 to 18 years and they were all of White British origin, which was representative for that service and area. Four participants had siblings. None had ever resided at a shelter for victims of DV. Their age when the DV began varied from 2 to 8 years, and it ended when they were 7 to 14 years old. On average, the DV had lasted 7½ years (ranging from 4 to 12 years). With one exception in which a male and female parent were violent toward each other, the violence described was from a male parent toward a female parent. One mother had lived with several male partners who had been violent toward her (p. 5257)Population not statedTarget sample n=10Achieved sample n=5Sampling strategy not stated |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Via a Child and Family Mental Health ServicePotential participants were suggested by clinicians and approached by researchers.Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by **authors**) | ‘The participant interviews were semi structured. The main areas explored included the participants’ experience of being exposed to DV, the meaning they made of their experiences, how they felt it had affected them, how any changes had happened, and about receiving support from others. The first author (J.C.) conducted all five interviews which lasted between 60 and 90 min’ (p. 5257)StrengthsA personal research diary was kept to reflect on the interactive andinterpretive process of developing the themes in the analysis. A presentation to Child and Mental Health Service clinicians indicated that main themes “resonated” with their clinical experienceStrategies to enhance validity of analysisLimitationsNone statedRecommendations for future data collectionUse of larger sample and quantitative methods; interview at time of experiences rather than some time after experiencesUse of longitudinal design to look at change over timeUse of more diverse sample |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘A National Health Service (NHS) ethics panel gave approval for this project. All adolescents and their parents gave informed consent to their participation and for the anonymized results of the study to be published. Safeguarding was assured by the adolescents being told that there would be limits to confidentiality. All adolescents had the opportunity to talk to a known clinician after the interview for further support’ (p. 5257) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | None stated but can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes, the results from the small sample have not been over-generalised |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample; interview schedule not given (questions can be inferred from results) |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Data analysis methods: interpretative phenomenological analysis (IPA); strategies to increase data analysis validity (see above) |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample size; add details on population and sampling strategies; justify target sample size |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and EJ agreed 22/11/’23** |
| Accession number (EndNote library) | 1736 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Izaguirre A and Kaalstrom A (2021) Differences in the reactions of adolescents to family violence *Child and Family Social Work* 26;425-433Spain Secondary schools (state and private) |
| Topic(s)Aim(s)Research question(s) | Domestic violence; IPV; adolescents’ reactions‘…to qualitatively explore variations among adolescents' reactions to violence occurring at their homes from their own point of view.’ (p. 427)What is the relation between the beginning of family violence and the adolescents' reactions to such episodes from their own point of view? |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Fifty-eight adolescents (26 boys and 32 girls, 13–18 years of age) recruited in their schools.Population not explicitly stated. Reviewers assume the initial 34 state and private secondary schools.Target sample size not stated.Achieved sample size – those adolescents who answered the 7 qualitative questions on the self-reported questionnaire.Sampling strategy – self-selected. |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | ‘The initial sample of the study comprised 845 adolescents (410 boys, 398 girls, and 37 who did not specify their sex) who were between 13 and 18 years of age (M = 15.89; SD = 0.84). They were recruited from six public and private schools in San Sabastian and its surrounding areas in the Basque Country in Spain. They completed a self-report questionnaire which contained, among other different measures, qualitative open questions regarding the beginning of the violent incidents against their mother and the adolescents' reactions when the violence happened. For the purpose of this study, adolescents who did not answer these qualitative questions were excluded being the final sample of the study 58 adolescents (26 boys and 32 girls) (p. 427)Not stated |
| Attrition mitigationActual attrition | Not applicableNot applicable |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by **authors**) | ‘Seven open questions regarding child exposure to violence were added to the self-report questionnaire. These questions referred to the nature of the violent episodes that happened at home in order to characterize the beginning and the frequency of the violence (e.g., when did the violence start to occur? how long did the violence last? and when did the violence stop happening?), and the reactions of the adolescents whenever they witnessed this violence against their mother (e.g., how did you react when the violence happened?)’ (p. 427).Thematic content analysis: codes were generated and then classified.LimitationsStudy relies on only one type of data collected at one point in time. Data collected focused on physical violence. Future recommendationsStudies investigating young people's reactions to other forms of threats. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘…passive informed consent was obtained from the adolescents' parents. None of the parents disagreed with their children participating in the study.’ (p.427). Schools provided with a report of the overall findings of the study. ‘Parents and adolescents were informed that the data would be used for research purposes only and that the children's identities would remain anonymous to the researchers. Data were collected in April 2014 and participants completed the questionnaires and demographic data individually in their classes. They were also encouraged to discuss any questions and doubts individually with the researcher in charge of the classroom assessment. The Ethics Committee of the university had approved this study.’ (p.427) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Yes |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes (although limited) |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Mainly, although there is some generalisation |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (although data collection method limited and some over-generalisation). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Method of recruiting sample (those who completed the open questions) introduces selection biasThe sample was restricted to one region |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | The analysis was rigorous and considered |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | A broader study in more than one region. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF Agreed 26/10/’23****[NOTE: page numbers not available for quotations due to online publication]** |
| Accession number (EndNote library) | 1310 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Katz E, (2015) Recovery-Promoters: Ways in which Children and Mothers Support One Another's Recoveries from Domestic Violence, *The British Journal of Social Work*, Volume 45, Issue suppl\_1, December 2015UK (Midlands region)Participants’ homes |
| Topic(s)Aim(s)Research question(s) | Recovery from domestic violence (DV); gendered coercive control; mothering; empowerment; practitionersTo explore how children and mothers with past experiences of domestic violence may play key roles in effectively promoting three aspects of one another’s recoveries: (i) decreases in abuse, (ii) formal supports from social workers and/or other practitioners which assisted recovery and (iii) mothers and children promoting one another's recoveries within their everyday family livesNot explicitly stated, although alluded to (see above) and can be inferred |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 15 mothers and 15 childrenNot explicitly stated. Reviewer infers all those contacted via 12 voluntary sector organisations.Target sample not statedAchieved sample n= 15 mothers and n=15 children‘In seven families the mother and one child were interviewed, in four families the mother and two of her children participated, and in four families it was only possible to interview the mother. The four mother-only interviews were not purposefully sought out, but were ultimately included in the analysis because they tended to provide data on families in slightly different circumstances, compared to the eleven families where children participated. The children who could not be interviewed seemed to be either too much or too little recovered to wish to participate.’ |
| Participant incentives | £10 vouchers per family |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Mothers and children were current or former users of voluntary sector organisations such as Women’s Aid. Recruitment was via these organisations. ‘Approximately twelve organisations were contacted by the researcher. Some organisations agreed to help the researcher to contact mothers using their service, so that these mothers could be informed about the study and asked whether they and their children were interested in participating. Participants were ultimately recruited from six of these organisations. Three families were also recruited through ‘snowball sampling’, where families that had been interviewed put the researcher in contact with further participants. All participants were residing in the community (rather than in refuges) at the time of interview.’ Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurement; data analysisStrengths and limitations; future recommendations for data collection (as reported by authors) | Data were ‘gathered through one-to-one, semi-structured interviews. Semi-structured interviews enabled data to be gathered on specific research questions, and relevant themes to be introduced that were not present in the topic guide (Esterberg, 2002), while also giving participants ‘the opportunity to shape the flow and content of the discussion’ (Moe, 2009, p.247). One-to-one interviews were also seen as an appropriate method for researching with children aged ten and over (Baker, 2005). Open questions were asked about mother–child relationships and (i) the domestic violence, (ii) the process of separating from perpetrators/fathers, (iii) experiences of services and (iv) participants' post-separation lives.’Data analysis used framework analysis, analysed thematically –creating framework from research questions and topic guide.StrengthsRecruiting families through organisations was advantageous, enabling access to a hidden and hard-to-reach population. ‘Sampling of mothers and children residing in the community, most of whom had never accessed refuges, and its interviews with children attending UK primary and secondary schools (i.e. children aged younger and older than eleven).’Limitations‘…only thirty participants from fifteen families were interviewed, and it cannot be known whether the results are generalisable to wider populations of domestic violence survivors. Several of the families studied had multiple, complex problems during the domestic violence (e.g. the perpetrator/father abusing the mother and child/ren, mental illness, drug/alcohol misuse and children with significant behavioural issues) and some of the families had experienced social service interventions.’‘The research also under-represents certain groups, including ethnic minority families and families living in rural areas.’ ‘Further research would be helpful in establishing whether the findings of this study are applicable to other populations of domestic violence survivors. In a small number of cases, it was only possible to interview mothers, or mothers and children requested joint rather than separate interviews. When children's views were absent, we cannot assume that they would agree with their mothers' accounts; and, when joint interviews occurred, this may have constrained participants' discussions of negative aspects of their mother–child relationships.’ |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘Approval was granted by the University of Nottingham's Research Ethics Committee. A domestic violence survivors' group was consulted prior to the fieldwork to confirm the appropriateness of the research design.’‘To participate, children had to be aged ten or over, and mothers and children needed to have separated from perpetrators/fathers and to be largely living in safety. The researcher established that these criteria were met through conversations with gate-keepers and mothers prior to the signing of consent forms. Interviews were usually conducted in participants' homes and, in most cases, mothers and children were interviewed separately (though some elected to have their mother/child with them during their interview). Interviews were digitally voice-recorded, and all participants were thanked via a £10 gift voucher. Participants were given detailed, age-appropriate information about the study, and formally agreed to participate by signing a consent form. Minimising imbalances of power between researcher and participants was attempted throughout the fieldwork, following the recommendations of Eriksson and Nasman, 2012). Participants were informed prior to interview that a referral to an appropriate statutory agency would be made if concerns arose about the safety of someone under eighteen (Baker, 2005). Fortunately, this situation did not occur during the study. Participants chose pseudonyms, and care was taken to maintain confidentiality and anonymity (Mullender et al, 2002).’ |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No, but alluded to and can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes, although a little overgeneralisation from the sample |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Generally, yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Moderate sample |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Detailed attention to ethical issues (see above) throughout study.Framework analysis – created from research questions and topicGuide.Analytical strategy: use of the CHIME framework, recovery was conceptualised as involving ‘connectedness, hope and optimism about the future, identity, meaning in life, and empowerment’ (Leamy et al, 2011, p.448) |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample size; widen diversity of sample with regards to ethnicity and increase number of families living in rural areas.Increase children’s voices in future research. |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and VS agreed 29/11/’23** |
| Accession number (EndNote library) | 3843 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | McCarthy M (2019) ‘All I wanted was a happy life’: the struggles of women with learning disabilities to raise their children whilst also experiencing domestic violence *Journal of Gender Based Violence* 3 (1) pp.101-118UK (London and SE England) (p.5)Homes or venues of participants’ choice |
| Topic(s)Aim(s)Research question(s) | DV; mothers with learning disabilities‘…to give some insights into the lives of women with learning disabilities as they experience the demands of trying to raise and protect their children, during or after the extreme stresses of domestic violence.’ (p.2)Not stated but can be inferred from aim. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 6 mothers with mild learning disabilities who had experienced violence from male partners.Population not statedSubset of a larger purposive sample of 15 women with learning disabilities who took part in a qualitative study on their experience of DV with the author. Target sample size not stated. Achieved sample size n = 6 |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Via professional contacts in a variety of learning disability services in London and SE EnglandNot stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Lengthy interviews (2 hours) to elicit the women’s ‘understanding and experience of domestic violence; the impact on themselves and their children; their coping strategies; whether and how they sought help to leave the relationship and life after the abusive relationship.’ (pp. 5-6)‘The principles and practices of Interpretative Phenomenological Analysis (IPA) were used to guide the data analysis. The rationale for using this approach is that IPA is well suited to a small sample size, as it allows for in-depth exploration of interview data and is deemed particularly suitable for under-researched topics of inquiry’ (p.6)Limitations‘…the sample was relatively small and participants were all volunteers, which may have resulted in unknown biases…all the women in this study were no longer in violent relationships (ethical approval was not given to include such women)…(so) they were relying on memory…Including women who were still actively mothering through domestic violence may have yielded different results’ (p.21)Participants may not be representative of women at the ‘more severe’ end of the learning disability spectrum.Recommendations for future data collectionEarly identification of women with learning disabilities who experience DV– close working is needed with health care professionals, includingmidwivesPeer support for women who are vulnerable mothers and are separatedfrom their childrenAdvocates for parents with learning disabilities are extremely important toempower them to speak up for themselves |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘Ethical approval for the study was given by the Social Care Research Ethics Committee (SCREC) (Ref. 12/IEC08/0028). It received Research Governance approvals from all the participating Local and Health Authorities and unconditional approval from the Association of Directors of Adult Social Services (ADASS)’ (p.6)‘All women were able to give informed consent to participate and were given accessible information, time and support to come to a decision.’ (p.6) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Not stated but can be inferred |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains except RQs, which can be inferred.). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample size with inherent biasesNo interview schedule to refer to, and only participants who have been in touch with services could be included. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Use of IPA in analysisDrawing on voices of under-represented group and ensuring diversity insample |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample size; think of more avenues for recruitment to furtherwiden range of women being included in sample; fuller description ofsampling, design of study (interview schedule, alternative methods foreliciting a wider range of voices). |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and VS agreed 01/12/23** |
| Accession number (EndNote library) | 162 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Meyer, S., & Stambe, R.-M. (2022). Mothering in the Context of Violence: Indigenous and Non-Indigenous Mothers’ Experiences in Regional Settings in Australia. *Journal of Interpersonal Violence*, 37(9-10)Australia (two regional settings)Not explicitly stated; reviewer assumes community setting; service provider setting |
| Topic(s)Aim(s)Research question(s) | DFV; mothering; indigenous and non-indigenous womenTo examine the experiences of mothering of Indigenous and non-Indigenous mothers affected by DFV in regional Queensland, Australia. To ‘examine mothers’ experiences of DFV as a gendered experience at the intersection of cultural and socio-spatial contexts’ (p. 7961)Not explicitly stated but can be inferred from aims. |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 17 mothers from one of two regional settings who participated in a wider study of women’s experiences of DFV and related homelessness.Identification through a stakeholder or service provider the women were currently or had previously been in contact with. 9 Indigenous and 8 non-Indigenous, ranging in age from 18-42‘Two Indigenous and one non- Indigenous mothers had previously experienced the removal of their dependent children by child protection services and described their children as living with maternal grandparents in two cases and in foster care in the third case. The number of children in women’s care ranged from one to six, with an average number of 3.5 children across mothers. Indigenous women reported a higher average number of children to care for than non-Indigenous women (3.9 compared to 3.1).’ (p. 7966)Population not statedTarget sample size not statedAchieved sample size n = 17Sampling strategy unclear |
| Participant incentives | 25 dollar shopping voucher for each participant |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Through stakeholder or service provider (see above)Not stated |
| Attrition mitigationActual attrition | Not statedNot stated |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | After providing preliminary consent to meet with the researcher, full informed consent was sought from all interviewees who participated in a one to one interview. All interviews were guided by semi-structured, open-ended questions, enquiring about women’s experiences of DFV, help-seeking, past and current housing stability, access to informal and formal support, future outlook, and feedback to service providers (housing related). 14 mothers were audio recorded. The data from the other 3 mothers were captured through written field notes. Analysis – inductive approach; thematic analysis, using codes and generated themesStrengthsFindings address a knowledge gapLimitationsSmall sample – not generalisableRecommendations for future data collection‘Self-determined’ research that draws on the voices of underrepresentedgroups is essential to informing community-led support and interventionsfor DV survivors. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | The research was approved by The University of Queensland’s Human Research Ethics Committee (clearance number: 2012000134). Ethical clearance covered field observations and interviews, written and verbal informed consent, and data collection through fieldnotes and audio recording. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Not stated but can be inferred from aims |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes – findings not generalised beyond the sample |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs which can be inferred from aims) |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample size (acknowledged by authors)Relationship between earlier study and the current study unclear with regard to sampleNo interview schedule to refer to, only participants who have been in touch with services could be included. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Listening to voices of Indigenous womenEnsuring diversity in sample |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample sizeMore avenues for recruitment to further widen range of women being included in sample.Outline the relationship between the current study and the earlier study in relation to participants |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and EJ agreed 27/11/’23** |
| Accession number (EndNote library) | 34 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Miranda JK, Olivares N and Crockett MA (2022) Growing up with intimate partner violence at home: Adolescents’ narratives on their coping strategies *Journal of Family Violence* 38 pp. 105-116ChileSchools |
| Topic(s)Aim(s)Research question(s) | Experiences of IPV in children and adolescents; impact; coping strategies ‘…to understand adolescents’ coping strategies regarding IPV between their parents or caregivers, from their perspectives.’ (p.105)Not stated but can be inferred from aim |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | Ten adolescents between 12 and 17 years old (4 female and 6 male); identification through use of protection programmes. A risk assessment was carried out between members of the research team and the programme professionals with the aim of ensuring that children and adolescents in situations of high risk of possible harm did not participate in the study. Therapists referred any adolescents who met the research criteria to the research team.‘The inclusion criteria were: (a) aged between 12 and 17 years old, b) having lived experiences of IPV (psychological, physical, and/or sexual) during the last year, (c) children having a court referral certificate stipulating “Witness of Domestic Violence” or having a documented history of living with IPV, depending on the program that he or she is attending, (d) currently living with their mothers and having lived with her for at least 6 months during the previous year, and (e) undergoing diagnostic assessment in the program. This was in order to avoid a re-victimization of the participants (Eriksson & Näsman, 2012) and contribute to the psychotherapeutic process. To protect the well-being of the participants, the following exclusion criteria were established: (a) children and mothers with a protected name and/or address, and (b) cases with a court order indicating that children are currently living in a situation assessed as high risk.’ (p. 107)Not statedTarget sample size not statedAchieved sample size n=10 (not justified).Sampling strategy – potential participants identified through therapists. |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | The therapists identified families which met criteria, told the mothers about the research. If a mother expressed their interest in participating, a member of the team informed them about the project, asked about their desire to participate, answered their concerns and asked them to consult with their children about their wish to participate. When both (mother and adolescent) agreed to participate, the research team first asked separately for informed consent from the mother and then for informed assent from the adolescent.Not stated |
| Attrition mitigationActual attrition | Not statedNone |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Audio recorded semi-structured interviews using a schedule adapted from a previous research study undertaken by the first author. The interviews were designed to ‘explore the three thematic axes addressed by this research: experiences of IPV in children and adolescents, its impact and coping strategies’ Thematic narrative analysis. ‘Thematic narrative analysis…allows the systematic study of personal experience and meaning, i.e., how events have been constructed and ordered by people to give meaning to the events and actions in their lives . The interview transcripts were analysed by members of the research team. Then, the following steps were taken: 1) Intra-case analysis was conducted by each member of the research, coding and condensing each adolescent’s interview. Joint coding sessions were held between team members, implementing analyst triangulation and carrying out an intra-case analysis. 2) Inter-case analysis was carried out to visualize differences and similarities, discussing and organizing themes and subthemes.’Strengths‘Our study contributes to the literature on youth who have experienced IPV between their parents by exploring the coping strategies of a group of adolescents who have experienced repeated/chronic IPV at home. Our results distinguish various ways in which adolescents acknowledge and respond to violence, and how the various ways of responding to violence may change with age. More generally, they contribute to the scarce research on this type of violence in Latin America and, especially, in Chile.’Limitations‘ the homogeneity of the sample must be recognized: the adolescents of this study were characterized by having had repeated or chronic IPV experiences during their lives, were all participating in protection programs and resided in urban areas. Future studies with larger and more diverse samples continues to be a challenge for clinical research.’ |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | University Ethics Committee granted ethical approvalProgramme professionals trained by the research team in the investigation protocol; ‘Evaluation and Containment Protocol’ in case of participants’ distress or disclosure. Assent was requested from the adolescents together with the informed consent of their mothers. Names of the participants were changed to protect their anonymity. |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No but can be inferred from aim |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** |  |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample size; sample homogeneous |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Working alongside therapist and relationship building worked well with this demographic |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample size and recruit more diverse sample |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and DF agreed 23/10/’23** |
| Accession number (EndNote library) | 1328 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Morrison F (2014) ‘All Over Now?’ The Ongoing Relational Consequences of Domestic Abuse through Children’s Contact Arrangements *Child Abuse Review* 24: 274-284UK (Scotland)Not explicitly stated. Reviewer assumes domestic abuse support service |
| Topic(s)Aim(s)Research question(s) | Domestic abuse/violence; child contact; post-separation parentingTo explore children’s and mothers’ experiences of contact with non-resident fathers when there is domestic abuse. Not stated |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 18 children aged eight to 14 years and 16 mothers who had experienced domestic abuse. The age of child participants ranged from eight to 14 years: eight were boys and ten were girls. The majority of the families identified as White Scottish, one family identified as Asian and one as Chinese.Population not stated.Target sample not stated. Achieved sample 18 children and 16 mothers.Purposive sampling strategy  |
| Participant incentives | None |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| Recruitment Retention | Participants recruited from domestic abuse support services in both the voluntary and statutory sectors.Not stated |
| Attrition mitigationActual attrition | Not statedNot stated |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Separate in-depth interviews were carried out with children and their resident mothers.Interviews with children: participative research activities which provided a physical entity to focus on rather than the dialogue and interaction between the child and researcher, e.g., ‘storyboard’, a pictorial vignette to explore three key areas: parental separation; participation in decisions about contact; and experiences of contact; ‘My Story’ which encouraged children to map their own experiences of contact onto paper.‘Interviews with women were open and tended to be directed by the woman being interviewed. At the start of the interview, women were asked about the legal processes that had taken place pertaining to contact and anything else that they thought was important to their child’s contact. Although a topic guide was developed that explored these areas, women tended to provide full accounts without much prompting, with the topic guide tending to be referred to more as a ‘checklist’ during the interview.’ (p.277)Interviews recorded and later transcribed.Inductive analytic strategy adopted for analysis: Interview transcripts were coded and analysed thematically. StrengthsEthical and practical advantages of recruitment strategy (via domestic abuse support services).LimitationsSmall sample‘Participants had spoken about their experiences prior to the research and participants who matched the sample criteria for the research were more easily identified than might have otherwise. However, this could mean that particular groups or perspectives are not represented in the sample. It would be useful to carry out similar research and recruit participants from a more general population’ (p.282)Perspectives of non-resident fathers of child participants not included. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | ‘Given the vulnerability of participants and the sensitivity of the topic, the research paid particular attention to ethical issues throughout including addressing informed consent, dealing with risk and child protection, privacy and confidentiality, and distress and damage. Ethical approval for the research was sought from and given by the University of Edinburgh’s Ethics Committee.’ (p. 277) |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | No, but can be inferred from aims |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, except RQs, which can be inferred). |
| **Challenges** in design and conduct (as judged by **reviewers**) | Small sample |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Methods of data collection with children innovatory.A purposive sampling strategy was used with families recruited from domestic abuse support services in both the voluntary and statutory sectors – to recruit ‘hard to reach’ families |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase the sample size |

|  |  |
| --- | --- |
| **DATA EXTRACTION** | **CT and VS agreed 29/11/’23** |
| Accession number (EndNote library) | 163 |
| Bibliographic details (author(s), date, publication)CountryResearch setting | Scrafford, K. E., Miller-Graff, L. E., Umunyana, A. G., Schwartz, L. E., & Howell, K. H. (2022). “I Did It to Save My Children”: Parenting Strengths and Fears of Women Exposed to Intimate Partner Violence. *Journal of Interpersonal Violence*, 37(9-10)USA (mid south and mid west)Private conference room at a university-affiliated research centre or at a community organization affiliated with the study |
| Topic(s)Aim(s)Research question(s) | IPV; parenting strengths and concernsTo explore mothers’ reports of women’s parenting strengths in the context of IPVNot stated in methods but stated in results:Where do women exposed to domestic violence excel in parenting? What are some common fears and worries that women exposed to domestic violence have about parenting? |
| Participants and identificationPopulationSample: target sample stated with justification? achieved sample size? sampling strategy(ies) | 22 IPV-exposed mothers (some of whom were pregnant) were recruited via flyers and direct referrals from hospitals, community centres, OBGYN offices and family justice centres. All participants contacted study staff via phone and were screened for eligibility. ‘IPV-affected mothers’ ages ranged from 18-42 years. The composition of this participant group covered African-American, Asian American, Hispanic, and Mixed race. 18 participants had children, 10 were pregnant. Six participants were currently experiencing IPV and 10 had experienced IPV during their most recent pregnancy. In terms of marital status, nine mothers were single, seven were married/living with a partner, and six were divorced or separated.’Population not statedTarget sample size not statedAchieved sample size n=22Sampling strategy unclear |
| Participant incentives | None stated |
| Design (longitudinal or cross-sectional) | Cross-sectional |
| RecruitmentRetention | As above recruitment was via flyers and direct referrals from hospitals, community centres, OBGYN offices and family justice centresNot stated |
| Attrition mitigationActual attrition | Not statedNot stated |
| Data collection methods and measurementStrengths and limitations; future recommendations for data collection (as reported by authors) | Focus groups:‘Two graduate-level research assistants co facilitated the focus groups, with one asking a set of semi structured questions and the other taking notes. All group facilitators received training on safety and mandated reporting in the context of IPV, additional education and experiential training on focus group facilitation, and weekly supervision throughout the study…All focus groups were audio recorded for later transcription. As part of the transcription process, the data was deidentified by replacing participant names with unique numerical identifiers.’ (p. 7780)Thematic analysis:Use of ‘Braun and Clarke’s (2006) six step guide: (a) read the transcripts to familiarize ourselves with the data, (b) generated a list of annotations, (c) organized annotations into subthemes and broader themes, (d) reviewed themes, collapsing some and removing others that had insufficient support from the data, (e) named the themes and subthemes, and (f) wrote up the thematic map and results.’ (p. 7782)Strengths‘The social-ecological model of resilience (Ungar, 2012) provides a useful framework for understanding relationships between motherhood and partner violence’ (p. 7794)Limitations‘Our data are limited to the two questions posed about parenting. Had we asked questions about how motherhood intersects with decisions to leave the partner, or how IPV impacts parenting, we would have received more diverse information about these topics that may have altered our thematic map…Furthermore, mothers participating in the study represent women of low SES; mothers of higher SES may have provided different responses due to different social experiences. Our samples were also limited to two urban locations in the Mid-South and Mid-West.’ (p.7797)Recommendations for future data collectionUsing socio-ecological model of resilience may reduce power imbalances between service providers and IPV-affected mothers, and thereby help service providers to approach IPV-affected mothers as allies and be better at asking screening questions and considering safety procedures, and barriers to help seeking for this population. |
| Ethical issues (sample identification, consent, data collection, archiving, GDPR) | Institutional review board (IRB) approval at both sitesUpon arrival at the focus groups, all participants completed an informed consent form and a brief demographics form |
| **QUALITY APPRAISAL** |  | **Yes; No; Unclear****Comments** |
| Research questions  | Are there clear RQs? | Not in methods but stated in results |
| Qualitative design and methods | Are the design and methods adopted appropriate? | Yes |
| Qualitative data collection methods | Are the data collection methods adequate to address the RQ(s)? | Yes |
| Findings | Are the findings adequately derived from the data? | Yes |
| Interpretation | Is the interpretation of results sufficiently substantiated by data? | Yes, the results from the small sample have not been over-generalised |
| Coherence | Is there coherence between qualitative data sources, collection, analysis and interpretation? | Yes |
| **Summary comments** | Meets threshold for quality (‘Yes’ in all domains, although RQs not explicitly stated in methods) |
| **Challenges** in design and conduct (as judged by **reviewers**) | Limited information on components of design including recruitment, sampling, ethicsSmall sampleOnly two open ended questionsAnalysis of link between demographics (e.g. race, SES) and parenting strengths has not been conducted. |
| Elements of **best practice** in design and conduct (as judged by **reviewers**) | Data analysis methods: socio-ecological framework of resilience |
| Strategies for **overcoming challenges (**as judged by **reviewers)** | Increase sample size; pose more detailed and nuanced questions about experiences of parenting; conduct intersectional analysis. |