

## Systematic Review

# Family-systems interventions for families of people with an intellectual disability or who are autistic: a systematic review

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## Abstract

**Background** Family-systems interventions have been proposed as one way of supporting families of people with an intellectual disability (ID) or who are autistic. This systematic review aimed to summarise what family-systems interventions have been studied with this population, what evidence there is for their effectiveness and families' experiences of the interventions.

**Methods** The review was preregistered on PROSPERO (CRD42022297516). We searched five electronic databases, identified 6908 records and screened 72 full texts. Study quality was evaluated using the Mixed Methods Appraisal Tool, and a narrative synthesis was used.

**Results** We identified 13 eligible articles with 292 participating families. Most studies reported positive effects of the interventions on wellbeing and family relationships, and families reported positive experiences. However, research quality was poor and there are no any sufficiently powered randomised

controlled trials demonstrating family-systems interventions' effectiveness for this population.

**Conclusions** There is a need for higher-quality research to establish whether family-systems interventions are beneficial for families of people who have an ID or who are autistic.

**Keywords** autism, effectiveness, family-systems, intellectual disability, systematic review, systemic therapy

## Introduction

Whilst family members of people with an intellectual disability (ID) or who are autistic report positive experiences, such as personal growth and viewing their family member as a source of happiness and fulfilment (Hastings 2016; Beighton & Wills 2019), many also experience psychological and family difficulties. For example, compared with typically developing children and their parents, children with an ID or who are autistic and their parents report more symptoms of mental health problems (Buckley *et al.* 2020; Bougeard *et al.* 2021; Rydzewska *et al.* 2021) and poorer family functioning (Jackson *et al.* 2022; Desquenne Godfrey *et al.* 2023).

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Furthermore, siblings of a child with an ID may have more frequent emotional and behavioural problems themselves (Hayden *et al.* 2019). These findings suggest that having a family member with an ID or who is autistic can affect the whole family, and therefore, all members of the family may benefit from additional support.

Family-systems interventions have been proposed as one way of providing this support (Baum & Lynggaard 2006; Cridland *et al.* 2014; Simon *et al.* 2020). The central idea of family-systems interventions draws on family-systems theory: families are complex, interconnected systems in which family members influence one another (Cox & Paley 1997; Wampler & Patterson 2020). Families are also conceptualised as hierarchically structured, consisting of internal subsystems such as a spousal/couple subsystem, parental subsystem and sibling subsystem (Cox & Paley 1997). Systemic interventions aim to improve the functioning of family systems, and the difficulties of individuals within them, through targeting the interactions between family members and the beliefs that they hold (Dallos & Draper 2015).

There are two main reasons why family-systems interventions may have some utility for families of people with an ID or who are autistic. First, there is evidence for the effectiveness of family-systems interventions for a wide range of clinical needs (Carr 2020), suggesting that they may also be beneficial for these populations. Second, concepts from family-systems interventions have clear applications to families of people with an ID or who are autistic (Cridland *et al.* 2014). For example, families may experience difficulties in adapting aspects of their relationships, roles and interactions to successfully accommodate the needs of their relative with a developmental disability (Benderix & Sivberg 2007; Cridland *et al.* 2016; Seligman & Darling 2017). Because family-systems interventions are not a single approach, but a broad category encompassing diverse interventions, they may support families of people with these potential challenges in a range of ways. Carr (2012) suggests that family-systems interventions may be divided into three categories. First, there are interventions that emphasise the importance of challenging problematic behaviour patterns such as strategic family therapy (Haley 1963), which focuses on disrupting

interactions that maintain difficulties, and structural family therapy (Minuchin 1974), which promotes adaptive family organisational structures. Second, other interventions focus on maladaptive belief systems, such as Milan systemic therapy (Selvini Palazzoli *et al.* 1978), in which the therapist and family co-construct belief systems that facilitate adaptive family interaction, and narrative therapy (White & Epston 1990), which involves developing richer and less problem-saturated narratives of families' difficulties. Finally, there are interventions that aim to modify predisposing factors to problems such as multisystemic therapy (MST) (Henggeler & Schaeffer 2016), which involves targeting multiple systems surrounding a family such as school, peers and the community, and attachment-based systemic therapies (e.g. Diamond 2005; Hughes 2007), which are interested in how early attachment experiences influence later relationships. Furthermore, systemic interventions have incorporated ideas from other therapeutic modalities, leading to the development of integrative interventions such as cognitive-behavioural family therapy (Epstein & Dattilio 2020).

Despite the potential applicability of family-systems interventions for families of people with an ID or who are autistic, they are an under-researched population in family-systems intervention research (Carr 2020). For example, a Cochrane review of randomised controlled trials of the effectiveness of family therapy for autistic people and their families identified no eligible studies (Spain *et al.* 2017). This review did not include research on family-systems interventions using alternative study designs or research with families of people with IDs who are not autistic. A broader systematic review would appraise the current evidence for family-systems interventions for families of people with an ID or who are autistic and inform future research.

As well as evaluating the effectiveness of family-systems interventions, it is also important to investigate families' qualitative experiences of interventions. This includes obtaining data on families' own subjective perceptions of what is beneficial, unhelpful and their satisfaction with provided interventions. All may influence engagement with interventions in future. Process evaluation is crucial in the development of evidence for the effectiveness of complex interventions and

involves investigating the mechanisms of change and typically includes analysis of qualitative data (on the experiences of receiving an intervention) and quantitative data (e.g. mediators and moderators of outcomes) (Moore *et al.* 2014) as well as the synthesis of these different data types. Whilst there is good evidence that family-systems interventions are effective with many populations, there is a poorer understanding of *how* they achieve positive outcomes (Johnson *et al.* 2020). Data on families' subjective experiences of interventions are vital for developing a richer understanding of the processes driving positive outcomes and the barriers to successful intervention (Hardy *et al.* 2020). Through understanding these experiences, future intervention research might adapt family-systems interventions to best meet the needs of families of people with ID or who are autistic.

The aims of the current systematic review were, therefore, threefold:

- 1 To summarise what family-systems interventions for families of people with an ID or who are autistic have been reported in the literature.
- 2 To summarise evidence for the effectiveness of family-systems interventions to improve wellbeing and/or family relationships in families of people with an ID or who are autistic.
- 3 To summarise research on the subjective experiences of families of people with an ID or who are autistic receiving family-systems interventions.

## Materials and methods

This systematic review was registered on PROSPERO before the searches were conducted (CRD42022297516) and is reported in line with the PRISMA guidelines (Page *et al.* 2021).

### Search strategy

We searched Embase, Medline, PsycINFO, Web of Science (all databases), and Applied Social Sciences Index and Abstracts. The last search was conducted on 13/1/23, and no restrictions were placed on publication date. The searches consisted of sets of intellectual disability/autism, family and family-systems interventions search terms. Terms

within each group were separated with OR, and groups of terms were combined with AND. The full set of search terms can be found in Supporting Information S1. Additionally, we conducted forward and backward citation searches on included articles and contacted the corresponding authors of all included articles to identify other potentially eligible research.

### Eligibility criteria

#### Population

Studies were eligible if participants were family members of a person who had an ID, was autistic or had an associated genetic syndrome. Family members included biological, adoptive, foster or stepfamily members. Disabilities could be confirmed by report of a diagnosis, receipt of special education or services, or meeting diagnostic thresholds on psychometric tests. Studies were eligible if data were reported for a group in which  $\geq 75\%$  of participants met this population criterion. The person with an ID or who was autistic could be of any age. We did not include families of a person with a diagnosis of attention-deficit hyperactivity disorder or a specific learning disability (e.g. dyslexia) who did not also have an ID, autism or associated genetic syndrome.

#### Intervention

Records were eligible for inclusion if they were studies of a family-systems based intervention that explicitly targeted family relationships and focused on two or more family subsystems (e.g. not simply one subsystem such as parent-child as might be targeted in parenting interventions or couple therapy). Parenting interventions and interventions delivered to one or more family members that did not explicitly target family relationships were excluded. No restrictions were placed on the context of intervention delivery.

#### Comparator

Studies with or without a comparison treatment or group were included in the review.

### Outcome

Studies were eligible for inclusion if they reported outcomes related to the following: (1) the wellbeing of family members including related positive or negative constructs such as measures of life satisfaction, quality of life, mental ill-health or stress; (2) measures associated with family relationships such as measures of family functioning, the closeness, quality, supportiveness of relationships or emotions associated with these (e.g. anger or conflict); or (3) any quantitative or qualitative data on experiences of family-systems interventions by a person with a developmental disability or their family members.

### Other eligibility criteria

Records were also excluded if they were reviews, conference abstracts, not available in English or duplicates. Non-peer-reviewed articles such as theses were eligible for inclusion.

### Selection process

After completing the electronic database searches, all records were exported to EndNote and the first author conducted electronic deduplication. The first author then screened the titles and abstracts of the remaining records, and a second researcher independently screened the titles and abstracts of a randomly selected 30% of records. This initially resulted in an unsatisfactory agreement rate of 91.71% (but kappa = 0.359) indicating potentially unclear guidance about eligibility criteria. The two researchers discussed their disagreements and supplemented the eligibility criteria with additional guidance to aid screening. The first author then re-screened 100% of titles and abstracts, whilst the second author screened a randomly selected 10% of the total records. This yielded a good agreement rate of 98.88% (kappa = 0.701). All articles identified for full-text screening were independently examined for inclusion by two researchers. This resulted in a good agreement rate of 97.73% (kappa = 0.910). Forward and backward citation searches were conducted on all articles retained following full-text screening to identify other potentially relevant articles. These then underwent abstract screening and, where applicable, full-text screening by the first author. To identify eligible, but not yet published research, the

corresponding authors of all included articles were contacted and asked whether they had any other potentially eligible research in press or that were otherwise not included. Any additional studies identified also underwent full-text screening by the first author.

### Data extraction

Data were extracted from included articles using a customised data extraction form. If data could not be obtained from the text, the corresponding author was contacted to request the missing information. All results relating to eligible outcomes were sought. A full list of variables for which data were extracted is in Supporting Information S2. A second reviewer independently extracted data from 100% of articles, and differences were discussed and an agreement was reached.

### Quality assessment

Given that a wide range of study designs were eligible for inclusion, we assessed methodological quality using the Mixed Methods Appraisal Tool (MMAT; Hong *et al.* 2018). The MMAT consists of screening questions for all study designs and separate questions to be used for qualitative, quantitative randomised controlled trials, quantitative non-randomised controlled trials, quantitative descriptive research and mixed-methods research. For each screening question, assessors must answer 'yes', 'no' or 'can't tell'. A second reviewer independently appraised 100% of included studies, resulting in an item-level agreement rate of 86.32% (kappa = 0.747).

### Data synthesis

Given the heterogeneity of the study design and outcome measurement of included studies, meta-analysis and meta-synthesis were not suitable. Narrative syntheses were conducted with all the included studies.

To address review question 1 regarding what interventions have been reported, all studies were also evaluated using a TIDieR checklist format (Hoffmann *et al.* 2014) to examine whether the reported interventions were comprehensively described. The TIDieR checklist consists of information that should be included to ensure

adequate description of interventions, such as their rationale, procedures and context. To address review question 2 regarding the effectiveness of these interventions, we then summarised narratively the effectiveness evidence reported in included studies, placing more emphasis on the strongest studies in terms of quality appraisal and overall research design. Finally, we addressed review question 3 about families' experiences of interventions by describing the themes identified in included studies.

## Results

The database searches identified 6908 records. Electronic deduplication removed 2067 records, and a further 4768 records were excluded during title and abstract screening. Of the remaining 73 records that were sought for retrieval, one full text could not be obtained (Tarantino 2003) and eight were not in English and were excluded. Eight records were eligible for inclusion following full-text screening.

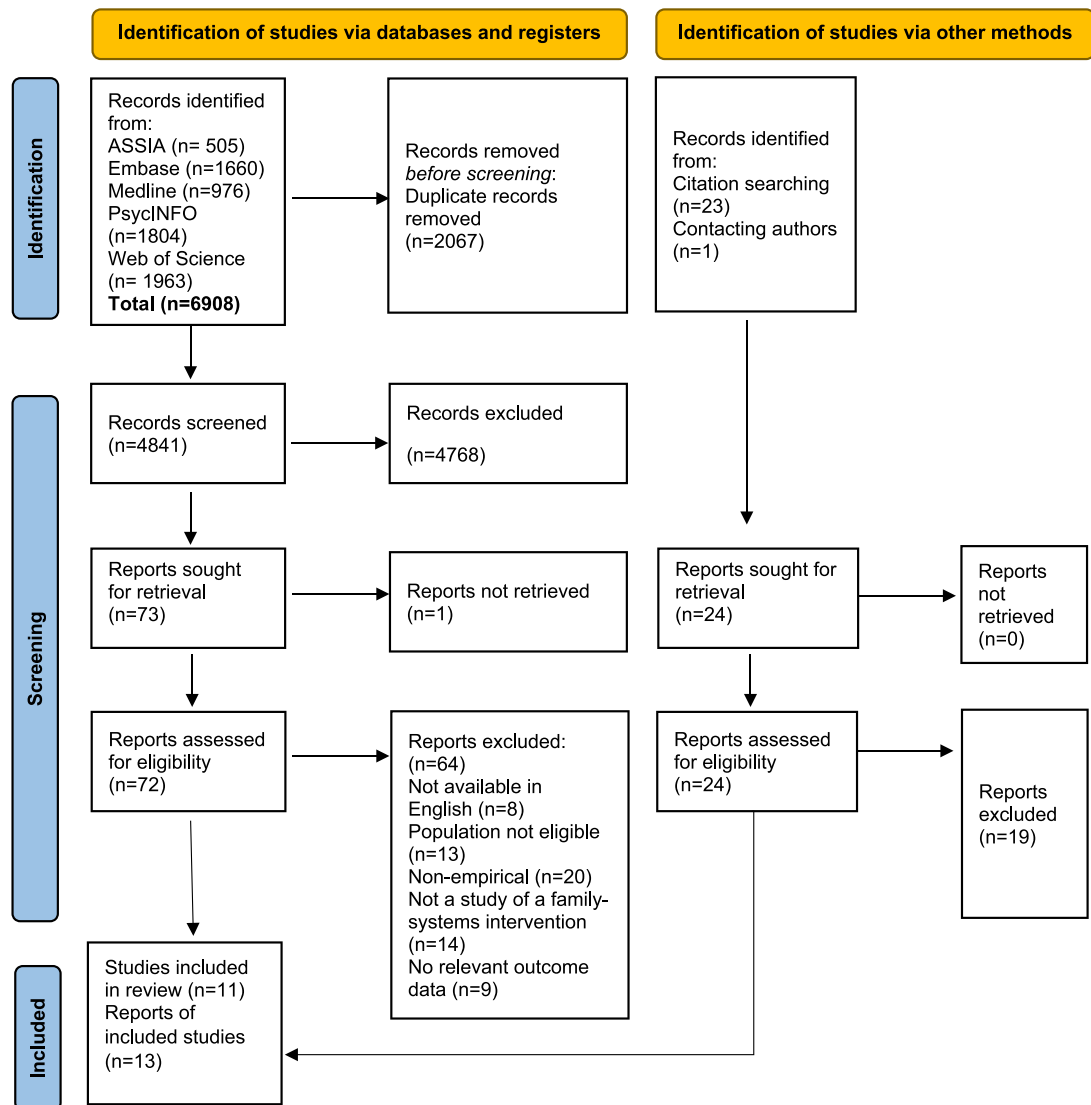


Figure 1. PRISMA flow diagram (Page *et al.* 2021) illustrating the search strategy.

Forward and backward searches identified four additional eligible studies, three of which directly cited included articles and one of which was cited in a meta-analysis that also cited an included study. One further record was identified by contacting the authors of included studies. Therefore, 13 reports from 11 studies were included in the review overall. The study selection process is illustrated in the PRISMA flow diagram (Page *et al.* 2021) (Fig. 1).

Overall, these studies included 292 families. There was a range of study designs, including two feasibility randomised controlled trials (Wagner *et al.* 2019; McKenzie *et al.* 2020), a qualitative evaluation of one of these trials (McKenzie *et al.* 2022), one non-randomised controlled trial (Blanckstein *et al.* 2019) and a separate report of follow-up data from this trial (Blanckstein *et al.* 2020), three qualitative studies (Villaescusa *et al.* 2021; Lo *et al.* 2022; Lo & Ma 2022), two pre-post studies (Parker *et al.* 1987; Baum 2006) and three case studies (Marshall & Ferris 2012; Ma *et al.* 2020a,b). Studies were conducted in the UK ( $n = 4$ ) (Baum 2006; Marshall & Ferris 2012; McKenzie *et al.* 2020, 2022), Hong Kong ( $n = 4$ ) (Ma *et al.* 2020a,b; Lo *et al.* 2022; Lo & Ma 2022), the Netherlands ( $n = 2$ ) (Blanckstein *et al.* 2019, 2020), the USA ( $n = 2$ ) (Parker *et al.* 1987; Wagner *et al.* 2019) and Spain ( $n = 1$ ) (Villaescusa *et al.* 2021).

Studies included families of a person with an ID ( $n = 4$ ) (Parker *et al.* 1987; Baum 2006; Blanckstein *et al.* 2019, 2020), autistic people without an ID ( $n = 4$ ) (McKenzie *et al.* 2020, 2022; Ma *et al.* 2020a, b), people with an ID some of whom were also autistic ( $n = 2$ ) (Lo *et al.* 2022; Lo & Ma 2022), autistic people some of whom also had an ID ( $n = 1$ ) (Wagner *et al.* 2019) and autistic people with an ID ( $n = 1$ ) (Marshall & Ferris 2012), and one study referred to participants as having 'intellectual/developmental disabilities' (Villaescusa *et al.* 2021).

### What family-systems interventions for families of people with an intellectual disability or who are autistic have been reported in the literature?

The 13 included reports described 11 family-systems interventions, which are summarised in Table 1 according to the TIDieR checklist (Hoffmann *et al.* 2014). The comprehensiveness with which interventions were reported was variable, with the

number of items from the TIDieR checklist where at least some relevant information was reported ranging from 4/12 to 11/12, and in some studies, little information was given about the content of the interventions. In particular, most studies did not report whether the intervention was delivered as planned (e.g. treatment fidelity) or whether the intervention was modified in any way. The interventions were generally more comprehensively described in articles reporting on controlled trials (all scoring  $\geq 10$  on the TIDieR checklist), whilst qualitative studies, case studies and pre-studies-post-studies typically received lower scores. This could reflect the controlled trials all involving manualised interventions where details such as materials, tailoring and adherence/fidelity monitoring are more likely to be standardised and reported.

Three articles (Blanckstein *et al.* 2019, 2020; Wagner *et al.* 2019) were about adaptations of MST (Henggeler & Schaeffer 2016). MST is a manualised intervention for adolescents displaying antisocial behaviour, which involves targeting multiple surrounding systems such as family, peers, school and the community. Two articles studied an adapted form of MST for adolescents with an ID (MST-ID) (Blanckstein *et al.* 2019, 2020), which included training MST therapists about ID and its effect on the family, using more accessible materials and a greater focus on mobilising social support networks. One study evaluated an adaptation for autistic adolescents [MST for youths with autism spectrum disorder (ASD)] (Wagner *et al.* 2019), which involved a focus on the role of interactions between autistic traits and surrounding systems in maintaining disruptive behaviour. These MST adaptations were generally well described, and Blanckstein *et al.* (2019, 2020) were the only included studies to report a measure of therapist's adherence to the intervention. However, precisely how some of the adaptations were implemented in practice was unclear.

Two studies (McKenzie *et al.* 2020, 2022) evaluated a manualised intervention for families of autistic children called Systemic Autism-related Family Enabling (SAFE). SAFE integrated techniques from attachment-based systemic therapies, solution-focused therapy and narrative therapy. The first and final sessions involved several families meeting together, whilst the other three sessions were attended by individual families and

D. Sutherland *et al.* • Family-systems interventions systematic review**Table 1** A TIDieR checklist (Hoffmann *et al.* 2014) summary of family-systems interventions for families of people with an intellectual disability/who are autistic

| Study                                | Intervention name | Why   | What (materials)                          | What (procedures)  | Who provided   | How   | Where  | When and how much   |
|--------------------------------------|-------------------|---|---|--|--|---|--|---|
| Baum 2006                            | Family therapy    | Many difficulties best understood from systemic perspective                           | None                                      | Structural family therapy focusing on strengthening subsystems and boundaries and increasing communication and also with Milan systemic and narrative therapy techniques   | Clinical psychologists, lead therapist and reflecting team | In person. All family members present for first session 'where possible'        | Three families at home and six at clinic     | 90–120 min with the family; 1–9 sessions over 1–17 months   |
| Blanckstein <i>et al.</i> 2019, 2020 | MST-ID            | MST reduces externalising behaviour. MST-ID is adapted for families of people with ID | MST manual (Henggeler <i>et al.</i> 2009) | MST targets multiple systems (e.g. family, school and peers) including using family therapy skills. MST-ID involves training therapists about ID, using accessible resources and promoting support from the social network | MST therapists with 5-day MST training                     | In person. MST typically involves at least the adolescent and primary caregiver | Family home                                  | Mean treatment duration = 5.1 months (range 2–8). Number and frequency of sessions not reported but MST usually 2–15 h a week |
| Lo & Ma 2022                         | SFT               | Manage family boundaries and improve family interactions                              | None reported                             | SFT decentralising the person with ID as the source of the problem, exploring interaction patterns, the influence of past experiences and solutions  | Family therapists  | Total of 24 face-to-face sessions and 6 online sessions                         | Face-to face sessions on a university campus | Sessions: mean = 7.5, range = 4–10<br>Duration: mean = 12.25 months, range = 8–15   |

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Table 1. (Continued)

| Study                  | Intervention name               | Why   | What (materials)                        | What (procedures)  | Who provided   | How  | Where  | When and how much  |
|------------------------|---------------------------------|---|---|--|--|--|--|--|
| Lo <i>et al.</i> 2022  | Multi-family group intervention | Reduce parenting stress and improve relationships and social support    | None reported                           | Psychoeducational talk, intrafamilial activities promoting understanding within families and interfamilial activities sharing support between families                           | Therapist (unspecified)  | Group of six families. One hybrid session, four online sessions and five face-to-face sessions | Not reported   | 30 h over 10 sessions  |
| Ma <i>et al.</i> 2020a | SFT and MFT                     | Repair the mother–daughter relationship and improve family interactions | None reported                           | SFT focused on family communication. MFT involved psychoeducation, group activity programmes and an overnight camp   | SFT: not reported<br>MFT: a family therapist, clinical psychologist and two social workers | Face-to-face. SFT with mother, father and daughter. MFT with a group of five families          | SFT: not reported<br>MFT: university campus and a campsite | SFT: six sessions of unspecified duration<br>MFT: 42 h over 3 months |
| Ma <i>et al.</i> 2020b | SFT                             | Manage communication difficulties and support in parenting              | None reported                           | SFT involved shifting blame of difficulties from the wife being autistic to examining broader family interactions  | Family therapist and clinical psychologist   | Sessions with husband, autistic wife and autistic daughter                                     | Not reported   | Eight sessions over 12 months  |
| Marshall & Ferris 2012 | BFT                             | Thought it would be more effective to work with surrounding systems     | BFT manual (Falloon <i>et al.</i> 1993) | Provided information about ID, schizophrenia and autism and helped the family and team to problem-solve, improve communication and make a crisis prevention and maintenance plan | A CBT therapist and a clinical psychologist  | Sessions with participant's mother and two support workers. Not specified whether face-to-face | Not reported   | 11 sessions of unspecified duration                                  |



Table 1. (Continued)

| Study                             | Intervention name                          | Why  | What (materials)                     | What (procedures)   | Who provided   | How  | Where   | When and how much   |
|-----------------------------------|--|--|--------------------------------------|---|--|--|---|---|
| McKenzie <i>et al.</i> 2020, 2022 | SAFE                                       | SAFE targets important autism-related family needs   | SAFE manual and homework activities  | SAFE has an attachment focus and also uses solution-focused and narrative therapy techniques. First and last sessions are based on multiple family therapy. The remaining sessions are single family sessions | Accredited systemic therapist with >30 years' experience and a support therapist | Sessions 1 and 5 involve only parents. Sessions 2–4 also involve children. Not reported whether face-to-face | Sessions 1 and 5 in a community setting. Sessions 2–4 in community venue or family home | Five 3-h sessions and one group follow-up session at 24 weeks post-allocation |
| Parker <i>et al.</i> 1987         | Multiple family therapy                    | To help transition into adulthood                    | None reported                        | Not reported  | Not reported   | Delivered to group of three families. Not reported whether face-to-face                                      | Not reported  | 90-min sessions every other week for 9 months                                 |
| Villaescusa <i>et al.</i> 2021    | The Family Quality of Life Support Program | The child's wellbeing is inseparable from the family | Manual (Martínez <i>et al.</i> 2016) | Sessions to develop a family improvement plan based upon identifying and addressing difficulties and building upon families' strengths  | A 'specialist' 'with extensive experience in working with families'              | Aim for the whole family to take part (Martínez <i>et al.</i> 2016)  | Not reported  | Duration of 1 year. Number of meetings is adjustable, with a minimum of five  |

Table 1. (Continued)

| Study                                 | Intervention name  | Why   | What (materials)  | What (procedures)  | Who provided   | How  | Where   | When and how much  |
|---------------------------------------|--|---|---|--|--|--|---|--|
| Wagner <i>et al.</i> 2019             | MST for youths with ASD  | Good evidence for MST. It has been adapted for other groups | MST manual (Henggeler <i>et al.</i> 2009)   | MST targets multiple systems including using skills from family therapy. MST for youths with ASD focuses on interactions between autistic traits and surrounding systems | MST therapists in doctoral courses with >6 months working with children or adolescents but no MST experience | Face-to-face. MST involves at least the adolescent and primary caregiver. Other family member's involvement not reported | The family's natural environment (home, school and neighbourhood) | Sessions as convenient. Frequency depended on needs, with more (e.g. 2–3) in initial weeks and less later. Lasted 5–7 months |
| Study                                 | Tailoring  | Modifications   | How well, planned (e.g. fidelity maintenance and assessment)  | How well, actual (e.g. fidelity outcome)   | TIDieR items reported  |  |   |  |
| Baum 2006                             | Families identified own goals to work on. Interpreter used with one family | Not reported  | A family therapist supervised the team for 3 h per month  | No fidelity measures. Three families attended $\leq 2$ sessions  | 10/12  |  |   |  |
| Blankenstein <i>et al.</i> 2019, 2020 | Tailored to capabilities of youth and caregivers                           | Not reported  | Therapists completed MST training, weekly supervision and quarterly booster sessions. Adherence monitored using the TAM-R | Mean TAM-R score = 4.3 (SD = 0.56); 1 = 'not at all' to 5 = 'very much'  | 11/12  |  |   |  |
| Lo & Ma 2022                          | Not reported   | Some sessions conducted online due to COVID-19              | Not reported  | Not reported   | 8/12   |  |   |  |
| Lo <i>et al.</i> 2022                 | Not reported   | Adapted to partial online delivery due to COVID-19          | Not reported  | Not reported   | 7/12   |  |   |  |
| Ma <i>et al.</i> 2020a                | MFT 'is process-oriented, and thus program changes often occur'            | Not reported  | Weekly staff meetings held for planning and evaluation of the MFT activities  | Not reported   | 9/12   |  |   |  |

Table 1. (Continued)

| Study                             | Tailoring   | Modifications   | How well, planned (e.g. fidelity maintenance and assessment)  | How well, actual (e.g. fidelity outcome)  | TIDieR items reported |
|-----------------------------------|---|---|---|---|-----------------------|
| Ma <i>et al.</i> 2020b            | Not reported  | Not reported  | Not reported  | Not reported  | 6/12                  |
| Marshall & Ferris 2012            | BFT 'is manualized ... but remains flexible in its application ...' | Minor adjustments to the BFT assessment to ensure relevance | Not reported  | Not reported  | 9/12                  |
| McKenzie <i>et al.</i> 2020, 2022 | SAFE tools can be 'employed flexibly' depending on families' needs  | Not reported  | Therapists received 4-day training and ongoing supervision. Therapists completed TCQ to monitor adherence   | 82% of lead and 63% of support therapists felt confident. One mother missed one session | 11/12                 |
| Parker <i>et al.</i> 1987         | Not reported  | Not reported  | Not reported  | Not reported  | 4/12                  |
| Villaescusa <i>et al.</i> 2021    | Number of meetings adjusted to families' needs                      | Not reported  | Not reported  | Not reported  | 6/12                  |
| Wagner <i>et al.</i> 2019         | Intervention is tailored to the strengths and needs of families     | Not reported  | Therapists received MST training and weekly supervision. Other therapies discontinued at start of treatment | Not reported  | 10/12                 |

ASD, autism spectrum disorder; BFT, behavioural family therapy; CBT, cognitive-behavioural therapy; ID, intellectual disability; MFT, multiple family therapy; MST, multisystemic therapy; MST-ID, multisystemic therapy – intellectual disability; SAFE, Systemic Autism-related Family Enabling; SD, standard deviation; SFT, structural family therapy; TAM-R, Therapist Adherence Measures – Revised (Henggeler *et al.* 2006); TCQ, Training Checklist and Questionnaire.

address their specific needs. SAFE was relatively comprehensively described, with the only item with no detail being how well SAFE was delivered.

Three studies investigated multi-family systemic interventions (Parker *et al.* 1987; Ma *et al.* 2020a; Lo *et al.* 2022). Lo *et al.* (2022) described a group programme with six families of an adolescent with an ID, which involved intrafamilial activities to promote relationships between family members and interfamilial activities to build social support between families. However, no information was provided about the therapist who delivered the intervention or how well it was delivered. Ma *et al.* (2020a) described using the combination of a multi-family therapy programme and structural family therapy (Minuchin 1974) with the family of an adult autistic woman without an ID. Both interventions focused on improving communication between the daughter and her mother. Multiple family therapy was reasonably well described, but very little information was given about the structural family therapy. Last, Parker *et al.* (1987) described a study of multiple family therapy with three families of a person with an ID. However, very little information was given about the intervention beyond its format and frequency.

Two studies investigated manualised interventions other than MST or SAFE (Marshall & Ferris 2012; Villaescusa *et al.* 2021). Villaescusa *et al.* (2021) studied an intervention called the Family Quality of Life Support Program for families of people with an intellectual/developmental disability. This involved the family developing a 'family improvement plan' by addressing family members' concerns and building upon strengths, although many details about precisely what this involved were unclear. Marshall & Ferris (2012) reported a case study of using manualised behavioural family therapy with the family and support workers of an autistic man with a mild ID and schizophrenia. This focused on preventing placement breakdown by helping the family and support workers to communicate and problem-solve effectively during crises. The intervention was largely well described, but details were missing about where sessions took place and whether they were delivered as planned.

Finally, three studies reported using non-manualised family therapy (Baum 2006; Ma *et al.* 2020b; Lo & Ma 2022). Baum (2006) evaluated family therapy based upon structural

(Minuchin 1974), Milan systemic (Selvini Palazzoli *et al.* 1978) and narrative therapy (White & Epston 1990) techniques in an ID service. Lo & Ma (2022) and Ma *et al.* (2020b) both used structural family therapy with families of adolescents with an ID and a family consisting of a father, autistic mother and autistic daughter, respectively. These interventions were generally well described.

The inconsistent reporting of session frequency and duration make overall synthesis challenging. However, many of the interventions appeared to be of relatively high intensity. The most intensive interventions were the MST adaptations. MST for youths with ASD began with roughly two to three sessions each week before tapering down over 5–7 months (Wagner *et al.* 2019), and whilst Blanckstein *et al.* (2019, 2020) did not report session frequency or duration, MST typically involves 60–100 h of contact and adaptations for other populations often involve more (Henggeler & Schaeffer 2016). The multi-family interventions described by Lo *et al.* (2022), Ma *et al.* (2020a) and Parker *et al.* (1987) involved 42 h, 30 h and 90 min every 2 weeks for 6 months, respectively. SAFE consisted of 18 h over six sessions (McKenzie *et al.* 2020, 2022), and behavioural family therapy involved 11 sessions of unspecified duration (Marshall & Ferris 2012). The least intensive interventions were structural family therapy, which consisted of on average 5.85 sessions (range = 1 to 10) across the four studies (Baum 2006; Ma *et al.* 2020a,b; Lo & Ma 2022) and the Family Quality of Life Support Program, which had a minimum of five sessions (Villaescusa *et al.* 2021).

Interventions were delivered by a range of professionals including clinical psychologists ( $n = 4$ ) (Baum 2006; Marshall & Ferris 2012; Ma *et al.* 2020a, b), systemic therapists ( $n = 4$ ) (McKenzie *et al.* 2020, 2022; Ma *et al.* 2020a,b; Lo & Ma 2022), MST therapists ( $n = 2$ ) (Blanckstein *et al.* 2019, 2020; Wagner *et al.* 2019), non-systemic therapists ( $n = 2$ ) (Marshall & Ferris 2012; McKenzie *et al.* 2020, 2022), unspecified therapists ( $n = 1$ ) (Lo *et al.* 2022) and social workers ( $n = 1$ ) (Ma *et al.* 2020a), but two studies did not report who delivered the interventions.

In summary, the interventions drew upon a wide range of family-systems approaches and were varied in their content, intensity and the professionals that

delivered them. The most studied approach was MST, which generally has a larger body of effectiveness research than many other systemic interventions (Littell *et al.* 2021). However, there were also multiple studies on SAFE, structural family therapy (Minuchin 1974) and multi-family group interventions. The interventions shared many features in common – all were delivered by professionals and 11 out of 13 were delivered entirely through face-to-face sessions. The remaining two interventions began face-to-face before adapting to online delivery during the COVID-19 pandemic (Lo *et al.* 2022; Lo & Ma 2022). Additionally, all of the interventions except the behavioural family therapy described by Marshall & Ferris (2012) also involved the person with an ID or who was autistic directly participating. Where studies reported adaptations to facilitate the engagement of the family member with an ID or who was autistic, these included adaptations in language and materials (Blanckstein *et al.* 2019; Wagner *et al.* 2019), a slower pace of therapy (Lo *et al.* 2022; Lo & Ma 2022), and play and activity-based content (McKenzie *et al.* 2022).

**What evidence is there for the effectiveness of family-systems interventions to improve wellbeing and/or family relationships in families of people with an intellectual disability or who are autistic?**

Table 2 summarises the findings of the seven studies that reported outcome data on the effect of the interventions on wellbeing or family relationships, grouped based on study design. The quality of these studies was evaluated using the MMAT and is summarised in Table 3.

Two studies were feasibility randomised controlled trials (Wagner *et al.* 2019; McKenzie *et al.* 2022). Wagner *et al.* (2019) found that MST for youths with ASD ( $n = 8$ ) was significantly more effective than usual community services ( $n = 7$ ) at improving family adaptability – families' ability to adapt effectively to stress ( $\eta^2 = 0.804$ ,  $P = 0.01$ ). However, there were no significant differences for other outcomes such as adolescent conduct or internalising problems. McKenzie *et al.* (2022) found that families who received SAFE plus usual support ( $n = 22$ ) showed some non-significant and very modest improvements in family functioning compared with those who received usual support only ( $n = 12$ ). Families in the

SAFE group showed an average decrease (indicating improved functioning) of 0.5 points on a measure of family functioning with a possible range of 15–75 compared with an average increase (indicating poorer functioning) of 1.1 in the usual support group. Both trials were underpowered and, because outcomes were self-reported, unblinded. The trial of MST for youths with ASD also suffered 43% attrition in the usual community services arm, and the groups differed on several variables at baseline. Therefore, neither of these trials provide good evidence for family-systems interventions' effectiveness.

Blanckstein *et al.* (2019) conducted a non-randomised controlled trial of MST-ID ( $n = 55$ ) versus standard MST ( $n = 73$ ) for families of adolescents with an ID, and Blanckstein *et al.* (2020) reported 18-month follow-up data for the MST-ID group. There was a significant decrease in rule-breaking behaviour in the MST-ID group ( $d = -0.44$ ,  $P = 0.001$ ), which was maintained at 18-month follow-up ( $d = -0.29$ ,  $P = 0.034$ ). Adolescents in the MST-ID group were also more likely than those receiving standard MST to be living at home (100% vs. 76.5%) and have improved family relationships (100% vs. 75.8%) at the end of treatment. However, there were no differences between MST-ID and MST in parenting stress or adolescent externalising problems and rule-breaking. These studies had large amounts of missing follow-up data and no control comparison for the 12- and 18-month follow-up. It is also important to note that in the Netherlands where the studies were conducted, individuals with an IQ < 85 may access ID services. The mean IQ in the MST-ID group was 73.90 and in the MST group was 75.10. Some participants might, therefore, not be considered to have an ID in other countries.

The remaining three articles had pre–post-designs (Parker *et al.* 1987; Baum 2006; Marshall & Ferris 2012). In an evaluation of a family therapy service for families of people with an ID, therapists reported that goals were achieved in four families and not achieved in two (Baum 2006). Of the four families whose goals were achieved, three showed first-order change (symptom reduction such as reduction in behaviour that challenges) and three showed second-order change (changes in family relationships). However, the unclear validity of therapists' subjective evaluation of outcomes and the

Table 2 A summary of studies evaluating the effectiveness of family-systems interventions in families of a person with an intellectual disability/who are autistic

| Study                   | Country         | Study design                                   | Participants  | Intervention               | Outcomes  | IMMAT total score                |
|-------------------------|-----------------|--|---|----------------------------|---|----------------------------------|
| Baum 2006               | UK              | Pre-design-post-design                         | Nine families of people with ID   | Family therapy             | Therapists reported goals 'achieved' = 4, 'not achieved' = 2 and not reported = 3<br><b>Wellbeing:</b> symptom reduction: yes = 3 and not reported = 6<br><b>Family relationships:</b> therapist reported change in family relationships: yes = 3, no = 1 and not reported = 5<br>Controlled for allocation bias with propensity score analysis<br><b>Wellbeing:</b> CBCL rule-breaking in MST-ID group: $F_{1, 33} = 13.59$ , $P < 0.01$<br>6-month follow-up MST-ID vs. MST:<br>CBCL rule-breaking: $B = -0.496$ , non-sig change; CBCL externalising: $B = -3.991$ , non-sig change; OBVL parenting stress: $B = -0.274$ , non-sig change  | Quantitative descriptive: 2/5    |
| Blanckstein et al. 2019 | The Netherlands | Non-randomised controlled trial                | Families of adolescents with ID. MST: $n = 55$ , MST-ID: $n = 73$               | MST-ID vs. standard MST    | <b>Family relationships:</b> mother's CSQ scores <sup>†</sup> (high score = high caregiver strain, range = 0–84): pre-BFT: 73 and post-BFT: 27<br><b>Family relationships:</b> mother's FFQ scores <sup>†</sup> (high score = better functioning, range = 0–72): pre-BFT: 28 and post-BFT: 52<br><b>Family relationships:</b> SCORE-15 overall score: mean (SD). Lower score = improved family functioning, range = 15–75. P-values not reported<br>Child with autism: SAFE: baseline $M = 36.4$ (10.1), 24 weeks $M = 38.8$ (11.3), $\Delta = 2.4$ . SUE: baseline $M = 36.8$ (11.9), 24 weeks $M = 38.1$ (12.0), $\Delta = 1.3$ ; 95% CI difference between SAFE and SUE at 24 weeks = -9.68 to 11.22 | Quantitative non-randomised: 3/5 |
| Blanckstein et al. 2020 | The Netherlands | Pre-design-post-design with 18-month follow-up | 55 families of adolescents with an ID   | MST-ID                     | <b>Family relationships:</b> improved parenting skills: $RR = 1.232$ , $P < 0.05$ ;<br>Therapist report: improved family relations: $RR = 1.280$ , $P < 0.05$<br>Adolescent living at home: MST-ID = 100%, MST = 76.5%, $P < 0.05$<br><b>Wellbeing:</b> CBCL rule-breaking: start-end: $t = -3.296$ , $P = 0.001$ , $d = -0.44$ ; start - 18 months: $t = -2.119$ , $P = 0.034$ , $d = -0.29$   | Quantitative non-randomised: 2/5 |
| Marshall & Ferris 2012  | UK              | Pre-post single-case design                    | Family of autistic adult with mild ID and schizophrenia                         | Behavioural family therapy |   | Quantitative descriptive: 4/5    |
| McKenzie et al. 2020    | UK              | Feasibility RCT                                | Families of autistic child. SAFE: $n = 22$ . Support usually employed: $n = 12$ | SAFE vs. SUE               |   | Quantitative RCTs: 3/5           |

Table 2. (Continued)

| Study                     | Country | Study design    | Participants   | Intervention            | Outcomes   | IMMAT total score                |
|---------------------------|---------|-----------------|--|-------------------------|--|----------------------------------|
| Parker <i>et al.</i> 1987 | USA     | Pre-post-design | Three families of person with ID   | Multiple family therapy | <p>Primary caregiver: SAFE: baseline <math>M = 34.3</math> (9.7), 24 weeks <math>M = 32.5</math> (7.2), <math>\Delta = -1.8</math>; SUE: baseline <math>M = 31.1</math> (7.6), 24 weeks <math>M = 33.0</math> (7.9), <math>\Delta = 1.9</math>; 95% CI difference between SAFE and SUE at 24 weeks = <math>-3.49</math> to <math>7.35</math></p> <p>Whole family average: SAFE: baseline <math>M = 35.7</math> (9.7), 24 weeks SAFE <math>M = 35.2</math> (8.6), <math>\Delta = -0.5</math>; SUE: baseline <math>M = 34.1</math> (9.5), 24 weeks <math>M = 35.2</math> (9.8), <math>\Delta = 1.1</math>; 95% CI difference between SAFE and SUE at 24 weeks = <math>-2.09</math> to <math>5.26</math></p> <p><b>Wellbeing:</b> pre-change–post-change in mean FRI self-esteem scores Person with intellectual disability: <math>+1.66</math>. Brothers: <math>+1</math>. Sisters: <math>-6</math>. Mothers: <math>-2</math>. Fathers: <math>-2.33</math></p> <p><b>Family relationships:</b> Pre-change–post-change in mean total FRI scores Person with an intellectual disability: <math>+1</math>. Brothers: <math>+1</math>. Sisters: <math>-7.5</math>. Mothers: <math>+5</math>. Fathers: <math>+1.66</math></p> | Quantitative non-randomised: 1/5 |
| Wagner <i>et al.</i> 2019 | USA     | Feasibility RCT | 15 families of autistic adolescent MST: $n = 8$ . Usual support: $n = 7$ | MST for youths with ASD | <p>ANCOVAs analysed condition (MST vs. usual support) <math>\times</math> time (baseline and 6 and 12 months) effects controlling for PSI total, FACES adaptability and BASC conduct problems scores</p> <p><b>Family relationships</b></p> <p>FACES-II cohesion: <math>F = 4.14</math>, <math>\eta^2 = 0.433</math>, non-sig change; adaptability: <math>F = 10.36</math>, <math>\eta^2 = 0.804</math>, <math>P = 0.01</math></p> <p><b>Wellbeing</b></p> <p>BASC-2 conduct problems: <math>F = 2.92</math>, <math>\eta^2 = 0.325</math>, non-sig change; internalising problems: <math>F = 0.032</math>, <math>\eta^2 = 0.079</math>, non-sig change</p> <p>PSI-SF difficult child: <math>F = 2.00</math>, <math>\eta^2 = 0.238</math>, non-sig change; caregiver distress: <math>F = 1.03</math>, <math>\eta^2 = 0.146</math>, non-sig change</p> <p>Caregiver BSI Global Severity Index: <math>F = 0.077</math>, <math>\eta^2 = 0.057</math>, non-sig change</p>   | Quantitative RCTs: 1/5           |

<sup>a</sup>Values were read from a graph and are approximations.

ANCOVAs, analyses of covariance; ASD, autism spectrum disorder; BASC-2, Behavior Assessment System for Children – Second Edition (Reynolds & Kamphaus 2004); BSI, Brief Symptom Inventory (Derogatis 1993); CBCL, Child Behaviour Checklist (Achenbach & Rescorla 2001); CI, confidence interval; CSQ, Caregiver Strain Questionnaire (Brannan *et al.* 1997); FACES-II, Family Adaptability and Cohesion Evaluation Scales-II (Olson *et al.* 1982); FFCQ, Family Functioning Questionnaire (Roncone *et al.* 2007); FRI, Family Relationships Inventory (Michaelson & Bascom 1978); ID, intellectual disability; IMMAT, Mixed Methods Appraisal Tool; MST, multisystemic therapy; MST-ID, multisystemic therapy – intellectual disability; OBYL, Opvoedingsbelastingvragenlijst Burden of Parenting Questionnaire (Vermulst *et al.* 2012); PSI, Parenting Stress Index; PSI-SF, Parenting Stress Index – Short Form (Abidin 1995); RCT, randomised controlled trial; RR, risk ratio; SAFE, Systemic Autism-related Family Enabling; SCORE-15, Systemic Core-15 (Stratton *et al.* 2010); SD, standard deviation; SUE, support usually employed.

Data in bold indicate domains of outcome (i.e. wellbeing or family relationships). Underlined data indicate which family member the data relate to.

**Table 3** Quality appraisal of studies evaluating the effectiveness of family-systems interventions using the Mixed Methods Appraisal Tool (MMAT)

| Screening questions                           |                               | Method-specific quality assessment questions |                                     |                                    |   |                               | MMAT total score |
|---|-------------------------------|--|-------------------------------------|------------------------------------|---|-------------------------------|------------------|
| S2. Data allow to address research questions? |                               | 2. Quantitative randomised controlled trials |                                     |                                    |   |                               |                  |
| Study   | S1. Clear research questions? | 2.1. Randomisation appropriately performed?  | 2.2. Groups comparable at baseline? | 2.3. Complete outcome data?        | 2.4. Outcome assessors blinded?             | 2.5. Adhered to intervention? | MMAT total score |
| McKenzie <i>et al.</i> 2020                   | Yes                           | Yes  | Yes                                 | No                                 | No  | Yes                           |                  |
| Wagner <i>et al.</i> 2019                     | Yes                           | Yes  | No                                  | No                                 | No  | Can't tell                    | 1/5              |
| 3. Quantitative non-randomised                |                               |  |                                     |                                    |   |                               |                  |
| S2. Data allow to address research questions? |                               | 3.2 Measurements appropriate?                |                                     |                                    | 3.5. Intervention administered as intended? |                               | MMAT total score |
| Study   | S1. Clear research questions? | 3.1 Participants representative?             | 3.3 Complete outcome data?          | 3.4. Confounders accounted for?    | 3.5. Intervention administered as intended? |                               |                  |
| Blanckstein <i>et al.</i> 2019                | Yes                           | No   | No                                  | Yes                                | Yes   |                               | 3/5              |
| Blanckstein <i>et al.</i> 2020                | Yes                           | No   | No                                  | No                                 | Yes   |                               | 2/5              |
| Parker <i>et al.</i> 1987                     | Yes                           | No   | Yes                                 | No                                 | Can't tell                                  |                               | 1/5              |
| 4. Quantitative descriptive                   |                               |  |                                     |                                    |   |                               |                  |
| S2. Data allow to address research questions? |                               | 4.2. Sample representative?                  |                                     | 4.4. Risk of nonresponse bias low? |   | 4.5. Analysis appropriate?    | MMAT total score |
| Study   | S1. Clear research questions? | 4.1 Relevant sampling strategy?              | 4.3. Measurements appropriate?      | 4.4. Risk of nonresponse bias low? |   | 4.5. Analysis appropriate?    |                  |
| Baum 2006                                     | Yes                           | Yes  | No                                  | No                                 | No  | No                            | 2/5              |
| Marshall & Ferris 2012                        | Yes                           | Yes  | No                                  | Yes                                | Yes   | Yes                           | 4/5              |



incomplete outcome data mean that the effectiveness of family therapy in this study is unclear. A case study found that behavioural family therapy reduced caregiver strain and improved family functioning in the family of an adult autistic man with a mild ID and schizophrenia (Marshall & Ferris 2012). Finally, a study with three families of an adolescent/young adult with an ID found that multiple family therapy led to improvements in some family members' self-esteem and perception of one another but decreases among others (Parker *et al.* 1987). Overall, the small samples and lack of control comparisons in these studies mean that they provide weak evidence for family-systems interventions' effectiveness.

Scores on the MMAT ranged from 1 (Parker *et al.* 1987; Wagner *et al.* 2019) to 4 (Marshall & Ferris 2012). However, because the MMAT uses different quality criteria for different study designs, comparisons across study designs should be interpreted cautiously. A quantitative case study with an MMAT score of 4 (Marshall & Ferris 2012) does not constitute stronger evidence than a randomised controlled trial with a score of 3 (McKenzie *et al.* 2020). The randomised controlled trials received scores of 3 (McKenzie *et al.* 2020), indicating a reasonably well-conducted trial with some weaknesses, and 1 (Wagner *et al.* 2019), indicating a severely limited trial. Non-randomised studies' scores ranged from 3 (Blankenstein *et al.* 2019), constituting informative evaluations that should still be interpreted with caution given the lack of randomisation, to 1 (Parker *et al.* 1987), which provide little information about intervention effectiveness. Finally, quantitative descriptive studies scored 2 (Baum 2006) and 4 (Marshall & Ferris 2012), but, given their study designs, neither represent robust intervention evaluations. In summary, all of the studies described tentative positive effects of the interventions on family relationships and/or wellbeing, except for Parker *et al.* (1987) whose findings were mixed. However, given the methodological flaws in the studies, none provide strong evidence supporting family-systems interventions' effectiveness. Currently, the only intervention with early evidence indicating possible clinically meaningful and sustained effectiveness appears to be MST-ID for adolescents displaying antisocial behaviour. SAFE may have very modest effectiveness at improving family functioning in families of autistic children, but a larger trial is

required to ascertain whether these effects are clinically meaningful. Small samples, lack of control groups and methodological weaknesses mean that there is currently inadequate evidence to determine whether other systemic interventions are effective with families of people with an ID or who are autistic.

### What does research suggest about the experiences of families of people with an intellectual disability or who are autistic receiving family-systems interventions?

Table 4 summarises the design and findings of the seven included studies reporting data on families' experiences of family-systems interventions. The quality of these studies was evaluated using the MMAT and is summarised in Table 5. Most studies were of good quality and were well described. However, Ma *et al.* (2020a,b) presented very little information about the research questions or data collection, and details about the analysis methods in several studies were unclear (Ma *et al.* 2020a,b; Villaescusa *et al.* 2021).

Three studies reported quantitative data on families' experiences (Villaescusa *et al.* 2021; Lo *et al.* 2022; McKenzie *et al.* 2022). Participants gave high ratings of the helpfulness of SAFE and the multi-family group intervention (Lo *et al.* 2022; McKenzie *et al.* 2022). High levels of satisfaction were also reported for both the multi-family group intervention and the Family Quality of Life Support Program (Villaescusa *et al.* 2021). However, participants in Villaescusa *et al.* (2021) completed the questionnaires in the form of an interview with the 'specialist' who delivered the programme, which may have biased their responses.

Seven articles reported qualitative data on families' experiences of family-systems interventions from focus groups ( $n = 5$ ) (McKenzie *et al.* 2020, 2022; Villaescusa *et al.* 2021; Lo *et al.* 2022; Lo & Ma 2022), questionnaires ( $n = 3$ ) (Villaescusa *et al.* 2021; Lo *et al.* 2022; McKenzie *et al.* 2022) and interviews ( $n = 3$ ) (Ma *et al.* 2020a,b; Lo & Ma 2022). However, the McKenzie *et al.* (2020, 2022) studies both reported data from the same study. These studies described generally positive experiences and processes. In all studies, participants described experiencing changes in family relationships and interactions such as reduced conflict or improved

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Table 4 A summary of the studies on families' experiences of family-systems interventions

| Study                  | Study design                            | Participants  | Intervention                    | Data collection   | Analysis method   | Quantitative satisfaction data   | Qualitative themes/summary of qualitative data  | MMAT scores   |
|------------------------|---|---|---------------------------------|---|-------------------|--|---|---|
| Lo & Ma 2022           | Qualitative focus groups/interviews     | Four families of adolescent with ID; nine participants in total | SFT                             | Focus groups or individual interviews                               | Thematic analysis |  | Themes: (1) sharing care responsibilities among family members, (2) using more open communication styles in the family, (3) mediating family problems, (4) actively involving adolescents with intellectual disabilities in family interactions and (5) guiding the future development of individuals with intellectual disabilities and their families | Qualitative: 5/5  |
| Lo <i>et al.</i> 2022  | Qualitative focus groups/questionnaires | Six families of adolescent with ID; 17 participants in total    | Multi-family group intervention | Focus groups and Client Satisfaction Questionnaire – Parent Version | Thematic analysis | Client satisfaction questionnaire data: 1 (strongly disagree/strongly agree/strongly satisfied) to 5 (strongly disagree/strongly satisfied). Mean satisfaction and helpfulness of sessions >4 for face-to-face and >4.50 for online sessions | Themes: improved family relationships, mutual helpful influences occurring among the families and a new understanding of the family members with intellectual disabilities  | Qualitative: 5/5<br>Quantitative descriptive: 3/5<br>Mixed-methods: 0/5 |
| Ma <i>et al.</i> 2020a | Qualitative interview                   | One family (mother, father and adult autistic daughter)         | SFT and MFT                     | Interview with mother and daughter                                  | Not reported      |  | Daughter described family therapy as helping her understand what had been going wrong in their family. Parents viewed their daughter more like an adult   | NA: no clear research questions   |

Table 4. (Continued)

| Study  | Study design                           | Participants  | Intervention                               | Data collection  | Analysis method   | Quantitative satisfaction data  | Qualitative themes/summary of qualitative data  | MMAT scores   |
|--|--|---|--|--|---|---|---|---|
| Ma <i>et al.</i> 2020b                         | Qualitative interview                  | One family (father, autistic mother and daughter)   | SFT  | Interviews with mother and father  | Not reported  |   | Therapy provided space to voice and resolve concerns. Understood themselves better. Related to extended family in more adaptive way                       | NA: no clear research questions   |
| McKenzie <i>et al.</i> 2020, 2022 <sup>†</sup> | Qualitative focus group/questionnaires | 22 families completed HAT; 10 adults and 6 children took part in focus groups             | SAFE                                       | Completed HAT after each SAFE session. Separate adult and child focus groups     | Thematic analysis (Braun & Clarke 2006)   | HAT Likert scale data: adults (1 = not helpful; 7 = very helpful): M = 5.83, SD = 0.87; children (1 = not helpful; 5 = very helpful): M = 4.07, SD = 0.79 | Themes: therapist as helping reflection, increased understanding, feeling closer, more confident to reflect and problem-solve, and improved communication | Qualitative: 5/5<br>Quantitative descriptive: 3/5<br>Mixed-methods: 1/5 |
| Villaescusa <i>et al.</i> 2021                 | Qualitative focus group/questionnaires | 13 families; 28 relatives and 15 individuals with intellectual/developmental disabilities | The Family Quality of Life Support Program | Questionnaire with Likert scales and open questions; n = 5 completed focus group | Two researchers identified general and specific categories, but method is unclear | 100% families pleased to participate; 58.3% satisfied and 41.6% very satisfied  | Themes: programme design, professional practice, and impact and satisfaction with the programme   | Qualitative: 3/5<br>Quantitative descriptive: 3/5<br>Mixed-methods: 2/5 |

McKenzie *et al.* (2020, 2022) both report qualitative data from the same study and so are reported together.  
 HAT, Helpful Aspects of Therapy Questionnaire (Llewelyn *et al.* 1988); ID, intellectual disability; MFT, multiple family therapy; MMAT, Mixed Methods Appraisal Tool; SAFE, Systemic Autism-Related Family Enabling; SD, standard deviation; SFT, structural family therapy.

**Table 5** Quality appraisal of studies investigating experiences of family-systems interventions using the Mixed Methods Appraisal Tool (MMAT)

| Screening questions             |                               | Method-specific quality assessment questions  |                                   |                                       |  |                                       | MMAT total score   |     |
|---------------------------------|-------------------------------|---|-----------------------------------|---------------------------------------|--|---------------------------------------|--|-----|
|                                 |                               | I. Qualitative                                |                                   |                                       |  |                                       |  |     |
| Study                           | S1. Clear research questions? | S2. Data allow to address research questions? | 1.1                               | 1.2                                   | 1.3                                    | 1.4                                   | 1.5  |     |
|                                 |                               |   | Appropriate qualitative approach? | Methods adequate to address question? | Findings adequately derived from data? | Interpretation substantiated by data? | Coherence between data sources, collection, analysis and interpretation? |     |
| Lo & Ma 2022                    | Yes                           | Yes   | Yes                               | Yes                                   | Yes                                    | Yes                                   | Yes  | 5/5 |
| Lo <i>et al.</i> 2022           | Yes                           | Yes   | Yes                               | Yes                                   | Yes                                    | Yes                                   | Yes  | 5/5 |
| Ma <i>et al.</i> 2020a          | No                            | No  | N/A                               | N/A                                   | N/A                                    | N/A                                   | N/A  | N/A |
| Ma <i>et al.</i> 2020b          | No                            | No  | N/A                               | N/A                                   | N/A                                    | N/A                                   | N/A  | N/A |
| McKenzie <i>et al.</i> 2020†    | Yes                           | Yes   | Yes                               | Yes                                   | Yes                                    | Yes                                   | Yes  | 5/5 |
| McKenzie <i>et al.</i> 2022†    | Yes                           | Yes   | Yes                               | Yes                                   | Yes                                    | Yes                                   | Yes  | 5/5 |
| Villaescusa <i>et al.</i> 2021† | Yes                           | Yes   | Yes                               | Yes                                   | No                                     | Yes                                   | Yes  | 4/5 |

| Screening questions             |                               | 4. Quantitative descriptive                   |                             |                        |                           |                               | MMAT total score      |     |
|---------------------------------|-------------------------------|---|-----------------------------|------------------------|---------------------------|-------------------------------|-----------------------|-----|
|                                 |                               |   |                             |                        |                           |                               |                       |     |
| Study                           | S1. Clear research questions? | S2. Data allow to address research questions? | 4.1                         | 4.2                    | 4.3                       | 4.4                           | 4.5                   |     |
|                                 |                               |   | Relevant sampling strategy? | Sample representative? | Measurements appropriate? | Risk of nonresponse bias low? | Analysis appropriate? |     |
| Lo <i>et al.</i> 2022           | Yes                           | Yes   | Yes                         | Yes                    | No                        | Yes                           | Yes                   | 3/5 |
| McKenzie <i>et al.</i> 2022†    | Yes                           | Yes   | Yes                         | No                     | Yes                       | No                            | Yes                   | 3/5 |
| Villaescusa <i>et al.</i> 2021† | Yes                           | Yes   | Can't tell                  | Yes                    | No                        | Yes                           | Yes                   | 3/5 |

Table 5. (Continued)

| Study                                       | 5. Mixed-methods              |   |   |   |   |  |  | MMAT total score |
|---|-------------------------------|---|---|---|---|--|--|------------------|
|   | S1. Clear research questions? | S2. Data allow to address research questions? | 5.1. Adequate rationale for mixed-methods design? | 5.2. Study components effectively integrated? | 5.3. Integration of qualitative and quantitative components adequately interpreted? | 5.4. Differences between quantitative and qualitative results addressed? | 5.5. Adhere to quality criteria of each of the methods involved? |                  |
| Lo <i>et al.</i> 2022                       | Yes                           | Yes   | No  | No  | No  | No   | No   | 0/5              |
| McKenzie                                    | Yes                           | Yes   | No  | No  | No  | Yes  | No   | 1/5              |
| <i>et al.</i> 2022 <sup>†</sup>             | Yes                           | Yes   | No  | Yes   | No  | Yes  | No   | 2/5              |
| Villaescusa <i>et al.</i> 2021 <sup>†</sup> | Yes                           | Yes   | No  | Yes   | No  | Yes  | No   | 2/5              |

<sup>†</sup>McKenzie *et al.* (2020, 2022) and Villaescusa *et al.* (2021) are mixed-methods studies and so are evaluated in the qualitative and quantitative description sections as well as in the mixed-methods section of the MMAT. Although McKenzie *et al.* (2020) is a mixed-methods study, it only includes qualitative data on experiences of the intervention and so is only evaluated in the qualitative section.

communication. Some participants also reported a change in their perception of their family member with an ID or who was autistic such as a greater understanding of their behaviour (Ma *et al.* 2020a; McKenzie *et al.* 2022) or becoming able to grant them greater independence (Ma *et al.* 2020a; Lo *et al.* 2022; Lo & Ma 2022). Common factors that were perceived to be important across multiple interventions included the value of the therapeutic relationship (Villaescusa *et al.* 2021; Lo & Ma 2022; McKenzie *et al.* 2022) and having space to reflect upon difficulties (Ma *et al.* 2020b; Villaescusa *et al.* 2021; Lo *et al.* 2022; McKenzie *et al.* 2022). The generally high quality of most of these studies provides good evidence that family-systems interventions are perceived to be beneficial and acceptable by families of people with an ID or who are autistic.

## Discussion

There is currently limited research on family-systems interventions with families of people with an ID or who are autistic. The interventions that were reported were from a range of systemic approaches, largely manualised and often integrated techniques from several models. Families generally reported positive experiences, and important processes, such as changes in family interactions or changes in perceptions of family members with an ID or who are autistic, appeared to overlap between interventions. Despite this, the effectiveness of family-systems interventions for these populations is unclear. Almost all studies identified some evidence of positive effects of the interventions on family relationships and/or wellbeing, suggesting that systemic approaches may be promising. The strongest preliminary evidence was for MST-ID as an intervention for adolescents with a mild/borderline ID displaying antisocial behaviour. Whilst McKenzie *et al.* (2020) concluded that SAFE appeared to be beneficial, the changes in family functioning were small. There was insufficient/inadequate data to draw conclusions about the effectiveness of other family-systems interventions. However, the small-scale studies described here are nevertheless useful illustrations of how family-systems interventions may be used and adapted with these populations.

The review findings suggest several foci for future research. Large-scale, carefully powered, randomised

controlled trials with appropriate comparative interventions for the study control arm may be warranted for promising interventions (MST-ID and SAFE). Given that four out of seven studies evaluating effectiveness in this review had samples of 15 or fewer families, effectiveness research in this field must progress to larger-scale controlled trials. Several studies have indicated that families have positive experiences of structural family therapy and that therapists perceive positive effects, but evidence is currently limited in relation to process evaluation (including questions of intervention process and moderation/subgroup effects). Mixed-methods process evaluations are a clear priority for future research. Based upon the finding of the quality assessment, future research must also focus on recruiting more representative samples and improving retention as well as more clearly reporting aspects of the research. It is also crucial that future interventions are clearly and comprehensively described to enable replication. In particular, interventions involving people with an ID or who are autistic may require substantial adaptations and tailoring to suit people with varied support needs, and these must be described more rigorously.

The lack of well-powered clinical trials potentially reflects debates about the role of scientific research on family-systems interventions. Some systemic practitioners, in particular those influenced by social constructionist approaches, have expressed caution about the informativeness of large-scale trials for clinical practice (Sexton & Datchi 2014; Lebow 2016). However, many systemic researchers and practitioners defend the importance of efficacy and effectiveness research for establishing whether family-systems interventions are beneficial and for encouraging their more widespread adoption by services (Sexton & Datchi 2014; Lebow 2016). A sophisticated understanding of how family-systems interventions may benefit families of a person with an ID or who are autistic will depend on integrating quantitative and qualitative methods in particular through process evaluations embedded in large-scale trials.

These findings may also inform clinical practice. Given the evidence reviewed here, family-systems interventions cannot currently be considered evidence-based interventions or recommended for routine implementation in services for families of

people with an ID or who are autistic. Nevertheless, clinicians may wish to explore the use of interventions such as MST for which there is preliminary evidence for potential positive effects. Alternatively, clinicians may consider integrating systemic techniques and ideas with other methods such as those from behavioural interventions (Rhodes *et al.* 2014). Where clinicians do use family-systems approaches, other research has highlighted the importance of interventions being adapted appropriately for people with an ID or who are autistic (e.g. to reduce the inaccessibility of the language, communication or environment that may prevent active engagement) (Arkless 2004; Baum 2007). Families' priorities should also guide which support is provided because some families expressed that practical support would have been more beneficial than systemic interventions (Arkless 2004; Baum 2006; Lo *et al.* 2022).

The findings of this review must be considered in the context of several limitations. As with all systematic reviews, it is possible that a small number of relevant articles were missed. Because the review only included interventions that targeted multiple subsystems, it did not include interventions within the systemic tradition that were applied in a more individually focused way (e.g. where one individual attended sessions and without a core focus on family relationships), as is sometimes the case with narrative and solution-focused therapies. This review also did not include the considerable number of narratively described individual case studies, which reported no quantitative or qualitative outcome data (e.g. Hill-Weld 2011; Digma 2021). These may nevertheless be informative illustrations of the application of family-systems interventions with families of people with an ID or who are autistic (although they would not add strength to any conclusions about their effectiveness). It is also important to recognise that, whilst there has been some research conducted in Hong Kong on experiences of family-systems interventions, this review identified no eligible studies, which evaluated their effectiveness outside of European or North American countries. It is possible that conducting the searches in English could have led to articles from other countries being missed, but it should be a priority for future research to be carried out in a variety of cultures.

Future research must also consider two differing approaches to intervention development reflected in

this review: the adaptation of existing interventions or the creation of bespoke interventions for families of people with an ID or who are autistic. Adapting existing family-systems approaches to suit the needs of families of people with an ID or who are autistic has the advantage that these may have existing evidence for their effectiveness with other populations. Modifying these without altering their core methods may therefore have similar benefits. However, these interventions may sometimes be inadequately adapted (Arkless 2004). An alternative approach involves developing bespoke interventions such as SAFE, which are based upon family-systems principles, but designed around the priorities of people with an ID/who are autistic and their families. These may lack the pre-existing research of other approaches but could more directly target families' needs. Whilst evidence is not currently available to evaluate the relative merits of these approaches, the advantages and disadvantages of each approach should be considered.

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### Conflict of interest

No conflicts of interest have been declared.

### Data availability statement

Data sharing is not applicable to this article as no new data were created or analysed in this study.

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## Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

**Supporting Information S1.** Example search string for Web of Science. The same search terms were used in all databases.

**Supporting Information S2.** Full list of variables for which data was extracted.