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REVIEW

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Effectiveness of interventions for school-aged-children and adolescents with fetal alcohol spectrum disorder: a systematic review and meta-analysis

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ABSTRACT

Purpose: To describe allied health and educational interventions and their effectiveness for children and adolescents with fetal alcohol spectrum disorder (FASD). To appraise the quality and strength of studies.

Methods: Electronic databases were searched between 2005 and March 2022, identifying non-pharmacological studies supporting function, activity, or participation for FASD participants aged 5–18 years using any quantitative research design. Outcomes were coded using International Classification of Functioning, Disability and Health, family of Participation Related Constructs and behaviour categories. Multi-level random-effects meta-analysis examined intervention effects. Study methodological quality was evaluated using Cochrane risk of bias tools, RoBiNT, AMSTAR 2 and NHMRC Hierarchy levels of evidence. Certainty of findings were synthesised using GRADE approach.

Results: The systematic review included 25 studies with 735 participants, 10 of which were analysed by meta-analysis. Body function and structure, activity, behaviour, and sense of self outcomes were pooled. A small, positive effect favouring interventions was found ($g = 0.29$, 95% CI = 0.15–0.43), however the GRADE certainty was rated as low. No participation outcomes were identified.

Conclusions: Some interventions targeting body function and structure, activity and behaviour outcomes were effective. Evidence of interventions that support children's and adolescent's participation as an outcome is lacking.

IMPLICATIONS FOR REHABILITATION

- To address participation outcomes for children and adolescents with fetal alcohol spectrum disorder (FASD), it is important to understand their participation needs and directly measure these.
- Interventions identified targeted body function and structure, activity, and behaviour outcomes.
- Participation outcomes of children's/adolescent's attendance, involvement and preferences were not identified.
- A combination of individual- and context-focused interventions is recommended to maximise rehabilitation outcomes for children and adolescents with FASD.

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Introduction

Fetal Alcohol Spectrum Disorders (FASD) describe a spectrum of brain injuries, birth defects and developmental disabilities caused by prenatal alcohol exposure (PAE) [1,2]. The spectrum ranges from Fetal Alcohol Syndrome (FAS) at the most severe end, to partial Fetal Alcohol Syndrome (pFAS), Alcohol Related Neurodevelopmental Disorder (ARND), and Alcohol-Related Birth Defects, the least severe [3]. Global prevalence of FASD is estimated to be 7.7 per 1000 population of children and youth [4] based on a 2017 systematic review and meta-analysis of 24 studies in eight countries including 1416 children and youth.

People with FASD experience lifelong, mild- to- severe impairments in neurocognitive, behavioural, social, academic, language and motor functioning [1,3,5–8]. Secondary effects such as difficulties accessing education services, substance abuse, mental ill-health, difficulties living independently, problems obtaining and maintaining employment and early contact with the justice system [1,6,7,9,10] contribute to negative lifelong outcomes. Children with FASD encounter participation restrictions, difficulty adapting to adverse life situations and negotiating major life transitions, [7,11,12] caused by a range of neurological impairments impacting daily living [7,11–13].

The International Classification of Functioning, Disability and Health (ICF) framework [14] conceptualises health and functioning,

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of people with disabilities, within broader environmental and social contexts. Functioning and disability, within this framework, are further divided into (i) body functions and structures (BFS), and (ii) activities and participation [14]. The ICF framework describes how personal, social, and environmental factors impact the individual's disability and functioning, [15] which can be applied to describe the experiences of people with FASD. It was the first model to conceptualise participation and articulate the goal of healthcare intervention to enable people with disabilities full inclusion and participation in society [15]. The International Classification of Functioning for Children and Youth (ICF-CY) [16] further extends the ICF [14] to document important factors that support the growth, health, and development of children and youth with disabilities. In the ICF framework, activity and participation are grouped together. Activity is defined as "the execution of a task or action by an individual" [16, p.9] and is qualified as "performance" and "capacity" [16, p.23] to execute the task or action. It is measured through observation of a person performing the task and their capacity to perform the task. Participation is defined broadly as "involvement in life situations" [16, p.9] in the ICF framework. Measurement of participation and its separation from activity is not clearly outlined in the ICF.

Since the introduction of the ICF-CY, there has been a more concerted effort to conceptualise and operationalise the construct of participation [17]. Imms and colleagues have further defined and measured participation as a separate construct from activity [18]. The construct of participation is considered an evolving, multidimensional construct [18, p.16] defined beyond the ICF that includes the personal meanings, values and experiences of participants [18,19]. It includes "complex, socially embedded and personally meaningful life roles and activities that children undertake" [19, p.553]. Participation is important for learning, development, health, and well-being. It is advocated as an essential intervention outcome for children and youth impacted by disabilities, families, and a research priority in paediatric rehabilitation [17,18].

The Family of Participation Related Constructs (fPRC) is a conceptual framework that distinguishes activity from participation, and positions participation as both an entry point and primary outcome of intervention, extending the ICF-CY [16] definition of participation [18]. In this framework, participation has two essential components: *attendance*, defined as "being there" [18, p.18], and *involvement*, defined as "the experience of participation while attending" [18, p.18]. Attendance is measured objectively as the number and type of activities, frequency or time spent doing something [18]. Involvement is best measured subjectively by the person involved who can relate their personal experiences [18, 20]. For example, the play experiences and desires of children with disabilities. Other elements of the fPRC framework that influence, and are influenced, by participation are intrinsic person-related constructs of activity competence, such as sense of self and preferences, as well as the external elements of environment and context [18]. Recently, others have advocated using the fPRC to conceptualise and measure participation intervention outcomes and ensure critical factors like the environment are integrated into the design of interventions and outcome measurement [21].

Motor impairments experienced by children with FASD have been extensively reported [2,22–33]. They include visual motor, fine motor, balance, co-ordination, ball skills, and delayed walking development. Difficulties with these abilities impedes (i) participation in the classroom (such as writing and other tasks requiring manipulation), (ii) play with other children and (iii) participating in sports and leisure activities. At home, children with FASD and motor coordination difficulties may experience activity and participation challenges performing daily activities such as getting dressed, or having a shower or bath. This can be related to completing tasks more slowly, clumsily and requiring caregiver support [13,34].

Recent literature on the participation of children and adolescents with FASD support the above findings and demonstrate that children and adolescents with FASD have difficulty participating in everyday activities across home, school, and community environments due to the range of neurodevelopmental impairments they experience [5,12,13,35–38]. Behavioural challenges including internalising and externalising conditions, emotional control and regulation, and adaptive functioning are commonly experienced and are major factors impacting their participation and inclusion across environments. In a systematic review and meta-analysis by Tsang et al. [39] it was shown that school-aged children with PAE and/or FASD had significantly higher scores on both internalising and externalising problems than children without FASD [39] and required clinical supports. In a study conducted by Gardiner et al. [40], caregivers of 87 children and adolescents with confirmed PAE and a subset with FASD reported significantly poorer adaptive functioning skills in PAE compared with normative samples. Gardiner et al. [40] also found that poorer behavioural regulation was associated with all aspects of adaptive functioning. Children and adolescents with FASD encounter challenges with school attendance, meeting classroom expectations, difficulty adapting to adverse life situations and negotiating major life transitions [7,9,12,13,37,38,41]. Conversely, studies report children and adolescents with FASD benefit from successful participation in art, games, sports, and school to foster strengths and support wellbeing [9,38,42,43]. To optimise rehabilitation and support healthy outcomes, it is critical that evidence-based interventions supporting participation are provided to children and adolescents identified with FASD [9, 44–46].

Research of interventions to assist those with a FASD diagnosis has evolved over the past 18 years with several published reviews [6,8,9,12,45, 47–57]. These reviews demonstrate a growth in the evidence-base of interventions particularly for school-aged children with FASD [9,52,54], targeting neurocognitive functioning, specific skill development and parent and teacher training [8,9,12,47,52,54,55,57]. Very few intervention studies have addressed motor skills and these motor intervention studies have not used activity and participation outcomes [58–60]. Intervention trials have compared an intervention to either no intervention or standard care such as usual classroom, caregiver psychoeducation, referrals to allied health (speech pathology, occupational therapy, physiotherapy), or information only [9,50,52,61]. Previous systematic reviews reported inconclusive findings and have limited generalisability due to the poor methodological quality and variable designs of included studies [50–52]. While two recent systematic review protocols propose to strengthen the evidence-base on interventions for children with FASD (to improve executive functioning, cognitive, psychological, and behavioural symptoms) [44,62] no systematic review and meta-analysis has yet been undertaken to investigate outcomes classified by the ICF-CY [16] levels, the fPRC [18] and behaviour influencing children's and adolescent's participation across home, school, and community environments.

This systematic review and meta-analysis addressed the following research questions:

1. RQ 1: What types of interventions are described for school-aged children and adolescents (5–18 years) with FASD have been evaluated on outcomes at ICF-CY [16] levels (i) BFS and (ii) activity and participation, the fPRC [18] and behaviour in the home, school, and the community?

2. RQ 2: What outcome measures, as classified by the ICF-CY [16] the fPRC [18], and behaviour, have been used to evaluate intervention outcomes for school-aged children and adolescents (5–18 years) with FASD?
3. RQ 3: What is the quality of the evidence for interventions for school-aged children and adolescents (5-18 years) with FASD?
4. RQ4: How effective are these interventions compared with no intervention or standard care for school-aged children and adolescents (5-18 years) with FASD?

Methods

Design and search strategy

A systematic review and meta-analysis were conducted. The design and reporting of this followed the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA 2020) [63] and A Measurement Tool to Assess systematic Reviews version 2 (AMSTR2) [64]. It was registered on the PROSPERO registry (https://www.crd.york.ac.uk/prosperso/ CRD42020186370). Electronic databases (CINAHL, ERIC, AMED, PsychINFO, Medline EBSCO, Medline Ovid, Embase, Web of Science, Scopus, and Cochrane) were searched for eligible papers with a publication date between 2005 to June and August 2020, and again in March 2022 by CH. Trial registries were not searched as only completed peer-reviewed published studies were included. A health sciences librarian was consulted in the selection and development of the search strategy using the PICOS framework (Population, Intervention, Comparison, Outcome, and Study design) including identifying keywords and thesaurus equivalents in each database. Search terms related to FASD, allied health and education interventions, and any intervention outcomes were combined to identify relevant studies (Table S1, online supporting information). Secondary searches of reference lists from retrieved papers were hand searched to identify any additional eligible studies. Data from the search were collated by the first author (CH) using EndNote 20.0 software [65], and grouped by the database. Data were exported to Covidence [66] for duplicate removal and title and abstract screening. All abstracts were screened independently by two researchers (CH and EF) for eligibility using predetermined criteria (Table S2-online-only). Full-text articles identified as "maybe" were also retrieved and uploaded into Covidence software [66] for eligibility assessment. PW provided a third review when required, and the consensus was reached after discussion.

Eligibility criteria

Studies were eligible for inclusion if they investigated non-pharmacological, intervention studies that support BFS, activity, or participation for children and adolescents aged 5–18 years with FASD. Studies were included if they evaluated the effect of a specific intervention on any quantitative measure of BFS, activity, or participation, which enabled us to compare these effects across intervention types and studies. Studies were included if they were published between 2005 to 2022, and peer-reviewed in any language. Randomised control trials (RCTs), non-randomised trials of intervention (NRSIs), single-case experimental designs (SCEDs), and systematic reviews of intervention studies were included to best capture the available evidence. Variations from the PROSPERO registration included searching all languages and clarifying the wording of review title and questions to enhance reporting this review.

Data extraction and management

CH extracted data from eligible studies systematically, using a standardised form in Excel^{TM}. Data extracted from the included studies encompassed: study design, level of evidence, number of participants who completed the intervention and in control groups, diagnostic information, participant demographics, a description of the experimental and comparator intervention, outcome(s) of interest, and a summary of results (Table 1). Outcome data were extracted for all quantitative measures, whether questionnaire or performance-based. When available, mean, standard deviation and number of participants for control and experimental groups were extracted at pre and post intervention. When standard errors were reported, these were converted to standard deviations where necessary. When sufficient data were not available to calculate effect sizes, authors [61,67,68] were contacted to request this information; none, however, provided sufficient data. Intervention intensity was quantified by calculating the mean total duration of intervention in minutes, a total number of intervention sessions, and frequency (number of sessions per week), reported as per Smits-Engelsman and colleagues [69]. This quantification method enabled comparison and collation of data across studies. Data extraction and coding were cross-checked by RS, TM, and PW.

Coding

Primary outcomes of change in child performance scores were extracted and classified according to the ICF-CY [16] levels: "BFS," "activity," and "participation." We chose to code outcomes separately as either "activity" (execution of a task or skill) or "participation" (involvement in life situations) with most outcomes found to measure the execution of a task or skill [16]. Where appropriate, outcome measures were also classified by the type of person-centred construct, using the Family of Participation Related Constructs (fPRC) framework [18] (Table S3-online-only). We chose to code contextual and environmental factors according to the updated fPRC framework [18] rather than the ICF-CY [16] because the two are clearly differentiated in the former: context refers to people, place, activity, objects, and time in which participation is set and is considered from the person participating [18], whereas the environment is external, referring to the "broader, objective social and physical structure in which we live" [18, p.20]. As some items on assessments could not be coded into ICF or fPRC levels, behavioural outcomes were categorised as (i) internalising, (ii) externalising, (iii) emotional control and regulation, or (iv) adaptive functioning (caregiver reported) or (v) adaptive functioning (teacher reported) (Table S4-online-only). These categories were created by the authors informed by other FASD and learning disorders literature, including the differentiation of internalising and externalising behaviours [70], emotional control and regulation described [71], and adaptive functioning defined [72]. As the criteria for selecting studies was measurement of BFS, activity or participation outcomes for children and adolescents, it was possible to code these outcomes against ICF-CY, fPRC and/or behavioural codes; hence, there were no missing data.

Risk of bias and methodological quality assessment

CH and RS independently rated the methodological quality of included reports using appropriate assessment tools for each of the main types of study design. The methodological quality and risk of bias in the included reports were evaluated using A MeaSurment Tool to Assess systematic Reviews version 2 (AMSTAR 2) [64] for systematic reviews, the Cochrane Collaboration Risk of Bias (RoB 2.0) tool [73] for RCTs and pseudo-RCTs and the Risk of Bias In Non-randomised Studies of Interventions (ROBINS-I) tool [74] for NRSIs. The Risk of Bias assessment in N of 1 trials (RoBiNT) [75] was used to assess the risk of bias in SCEDs. The RoBiNT [75] was included after the review was registered with PROSEPRO when eligible SCEDs were identified. All included reports of studies were evaluated individually for risk of bias. Consensus was reached through discussion.

The AMSTAR 2 [64] appraises systematic reviews with both randomised and non-randomised studies. It consists of 16 items, of which six were determined as critical for determining risk of bias in this review (item 2: register protocol; item 4: comprehensiveness in literature search; item 7: justification of excluded studies; item 9: assessment of risk of bias in individual studies; item 13: account for review of risk of bias in individual studies when interpreting/discussion of results; and item 14: explanation for and discussion of heterogeneity). When six or more non-critical weaknesses were found, the overall appraisal was moved from moderate to low confidence [64]. The systematic review was rated as "high," "moderate," "low," and "critically low" according to the quality rating confidence levels (Table S5-online-only).

The RoB 2.0 [73] assesses risk of bias in five domains: bias arising from the randomisation process; bias due to deviations from intended interventions; bias due to missing outcome data; bias in the measurement of the outcome; bias in the selection of the reported result. Each potential source of bias was assessed as either "low risk," "some concerns," or "high risk." The overall study risk of bias assessment is also rated as "low," "some concerns," or "high" risk. A visual plot of findings was generated using Robvis software [76].

The ROBINS-I [74] includes the same five domains at the RoB 2.0 [73] and extends to two additional domains: risk of bias due to confounding and bias in selection of participants into the study. Each potential source of bias was assessed as either "low," "moderate," "serious," "critical," or "no information." Overall study risk of bias was also rated as "low," "moderate," "serious," "critical," or "no information." A visual plot of findings was generated using Robvis software [76].

The RoBiNT [75] consists of 15 items to evaluate internal and external validity of SCEDs. The internal validity subscale consists of 7 items: design, randomisation, behaviour sampling, participant, interventionist and assessor blinding and treatment adherence. The internal validity subscale indicates a risk of bias [77,78]. An algorithm evaluates methodological rigour and risk of bias of internal validity subscale items [78]. Methodological rigour has six categories: "very high" (corresponding to very low risk of bias), "high" (low risk of bias), "moderate" (some risk of bias), "fair" (substantial risk of bias), "low" (high risk of bias) and "very low" (very high risk of bias). The external validity subscale of the RoBiNT consists of eight items: baseline characteristics, setting, dependent variable, independent variable, raw data record, data analysis, replication, and generalisation. It assesses reporting quality and applicability of the SCED [77]. The RoBiNT is scored out of a total of 30 however authors have not provided cut-off scores to determine risk of bias and recommend using the algorithm instead [78].

The level of evidence (LOE) of each study was rated against the National Health and Research Council Hierarchy of Levels of Evidence Framework [79] (Table S6-online-only) to provide an overall view of the range of intervention evidence. This rating was completed by CH and reviewed by RS.

Data synthesis

Qualitative synthesis

Narrative synthesis was used to describe the types of interventions, effectiveness, and outcome measures that support BFS, activity, and participation for school-aged children and adolescents with FASD in the home, school, and the community. All reports meeting eligibility criteria were included in the narrative synthesis.

Quantitative synthesis

All meta-analyses were conducted using R version 4.0.5 [80] and RStudio [81]. The magnitude of effect sizes (or differences between treatment groups) was quantified using Hedge's g, which were calculated on pre-post change scores using the 'esc' package [82]. Pre-post change standard deviations were imputed using accepted methods [83], with a correlation coefficient of 0.5 used in the absence of this data being reported. Effect sizes were calculated such that positive values would always indicate greater improvement for the experimental group. Multi-level random-effects meta-analyses were performed using the "metafor" package [84]. This form of analysis better accounts for non-independence of effect sizes when multiple values are extracted from individual studies, both randomised and non-randomised, or when multiple measures of a construct are derived from different scales [85–89]. Given there were few studies included in the multi-level meta-analysis, the Hartung-Knapp-Sidik-Jonkman method was applied to estimate the variance of pooled effects [90, 91]. Interpretation of heterogeneity at both the effect size level (level 2) and the study level (level 3) was done with I^2 statistics [83, 87, 92]. We included forest plots for subgroups and overall analysis to offer further visual insight into effects.

Effects of study design and outcome

First, to investigate if there was a difference in outcomes according to study design, an overall model was run on all effect sizes with study design (RCT or NRSI) entered as a moderator variable. Next, an overall model was run on all effect sizes with outcome category entered as a moderator variable. Separate subgroup models were run for outcomes coded under the ICF-CY [16] categories of BFS, activity and for behavioural outcomes. No participation outcomes were identified to include. There was only one outcome category under fPRC Sense of Self, so subgroup analysis could not be conducted. Significant combined effect sizes were indicated by 95% confidence intervals that did not cross zero. The magnitude of mean effect size estimates (g) was interpreted according to the conventions of Cohen [92] 0.3 (small), 0.5 (moderate), 0.8 (large), >1.0 (very large). Notable heterogeneity was indicated when I^2 exceeded 50% [83].

Ratings of study certainty of evidence used in meta-analysis

The Grading of Recommendations Assessment Development and Evaluation (GRADE) approach [93] was used to rate the overall certainty of evidence of the outcomes used in the meta-analysis. It rated the quality of evidence as "high," "moderate," "low," or "very low" in the areas of risk of bias, inconsistency, indirectness, imprecision, and publication bias. Each area was downgraded by one or two points based on judgement criteria. Risk of bias were downgraded if the highest-weighted studies (i.e. those with the largest number of participants) were assessed as serious or high risk of bias according to guidelines provided by Schunemann [94] for NRSIs and RCTs. Inconsistency was downgraded if heterogeneity was considered important (>50%) or large variation in effect size among studies pooled. Indirectness was downgraded if outcomes measured did not directly measure activity or participation. Imprecision was downgraded for studies with <300 participants for each outcome. Publication bias was assessed by aggregating all effect sizes per study and visually inspecting trim-and-fill funnel plots [95] for all (pooled) outcomes, and separately for activity, behaviour and BFS outcomes. CH and BL assessed GRADE ratings. A summary of the findings table was generated using GRADEpro software [96].

Results

Flow of studies through the review

A total of 2,801 studies were identified. After duplicates were removed, the titles and abstracts of 1,367 were screened. The full texts of 70 potentially eligible studies were retrieved and screened. Of these, 25 studies met the inclusion criteria [8,9,50–52,58,67,68,97–115] - including 20 studies and 5 systematic reviews (Figure 1) - while 42 were excluded (Table S7-online-only). The 20 intervention studies were described in 23 reports [58,61,67,68,97–115]. For meta-analysis, a total of 13 reports across 10 eligible studies were included [58, 97–100, 103, 104, 108–110, 113–115] (Figure 1).

Study characteristics of included studies

Study characteristics and key findings of all eligible studies are reported in Table 1. We identified five systematic reviews [8,9,50–52], five RCTs LOE II [67,97,99,100,103,114,115], three pseudo-RCTs LOE III-1 [61,98,113,], six case-control trials (CCTs) LOE III-2 [58,68,104,108–110,112], one interrupted time series without a control LOE III-3 [106], and five LOE IV designs including three SCEDs (using an ABA design) [101,102,111], one experimental pre-post study without control [105], and one case study [107] (Table 1).

Study participants

Across the included 20 studies [58,61,67,68,97–115] there were 735 participants recruited with FASD and 674 participants who completed the intervention (92% retention). Recruited sample sizes varied considerably, with a mean of 38.6, and ranged from one participant [101,102] to 100 [104,109] (Table 1). Participant's ages varied from 5 years to 15.8 years. Mean age was 9.3 years and percentage of male participants was 60%, calculated on data from 19 studies [58,61,67,68,97-99,101,102,105-115]. Adolescents (aged 13-18 years) participated in six out of the 20 studies (30%) [58,68,105–107,114]. Mean FASD diagnosis of participants was calculated on data from 14 studies [58,68,97,98,101,102,107,109,110,113–115]. The most common FASD diagnosis was ARND (44%), then FAS (26%), and lastly pFAS (19%). A category for "other" (11%) included deferred FASD diagnosis and Neuro-developmental Disorder Alcohol Exposed (ND-AE). A variety of FASD diagnostic criteria were used to define the type of FASD of participants including the Institute of Medicine [97,99,100,103,107,116] the 4-Digit Diagnostic Code [58,61,104,109,112,117], the Gestalt Diagnostic Guidelines [98,118], and the Hoyme Criteria [114,119] The percentage of participants in guardianship care, identified in 10 studies [58,61,68,98–101,103,104,107,109–111,115], ranged from 69% [98] to 100% [101,107,111,115]. Participant's ethnicity varied across studies and was representative of diverse populations (Table 1).

Types of interventions to support outcomes on ICF [16] levels, fPRC [18] and behaviour (RQ1)

There were a diverse range of interventions supporting various outcomes of school-aged children and adolescents 5-18 years with FASD (Table 1). Interventions were classified according to their (i) aim; (ii) prime target: children, caregivers, or teachers; and (iii) the mode of intervention delivery, namely direct skills training, group or individual intervention, and technology-assisted training. Furthermore, the intensity of interventions (duration, number of sessions, and frequency) and location of interventions is also described.

Target of intervention

Intervention aims. Across the studies, intervention aims varied widely. These were categorised into those aiming to improve (i) BFS, (ii) activities and participation and (iii) behaviour. Interventions improving BFS targeted attention [67,99], executive functioning [68,108,115], and balance [58]. Interventions aimed to improve activity and participation, addressed academic skills [97,98,107], social skills [104,108–110,114], task completion [101,102], learning to use metacognitive strategies [106], and fire safety skills [111]. Behavioural interventions aimed to improve child behaviour, often by reducing problem or disruptive behaviours [61,99,100,102,103,113,115].

Prime target of intervention. In 18 of 20 studies (90%), children and adolescents with FASD received direct intervention through individual or group therapy (Table 1) [58,67,68,97,99–115]. In nine of 20 studies (45%), teachers or caregivers and children received the intervention (Table 1) [61,98–104,108–110,115]. Context-focused approaches (involving adapting the task or environment to support children/adolescents' activity and participation, such as teacher and caregiver training [12]), were used in two studies to build teacher and caregiver capacity [61,98]. In one of these, teachers received professional development training and support, and changes in child performance were measured [98]. In the other, caregiver consultation and community referrals to modify parenting attitudes and responses towards children's behaviours were used in the Families Moving Forward (FMF) study [61]. Caregiver training was mostly provided through individual consultation [61,99,100,103,104,108–110] and within a group setting in one study [115].

Mode of intervention delivery

Direct skills training. Most interventions in this review aimed to enhance the participant's activity competence [18] and execution of a task or skill [16] using skills-based training approaches (15 of 20 studies, 75%). These training approaches targeted activity competence required for participation across home, school, and the community. Skills targeted included: mathematics [107], literacy [97], social skills [104,109,110,114], self-regulation/attention [105,108,112,113,115], persistence [67], metacognitive strategy learning [67,99,100,103,105–107], fire safety [111], task completion [101,102] and problem solving [109].

Individual versus group training. Training was provided to children and adolescents individually in 15 of 18 (83%) of the studies [58,67,68,99–103,105–108,111–114]. Two studies did not involve children [61, 98]. In six of 18 studies (33%), training was delivered in groups, mainly addressing social skills

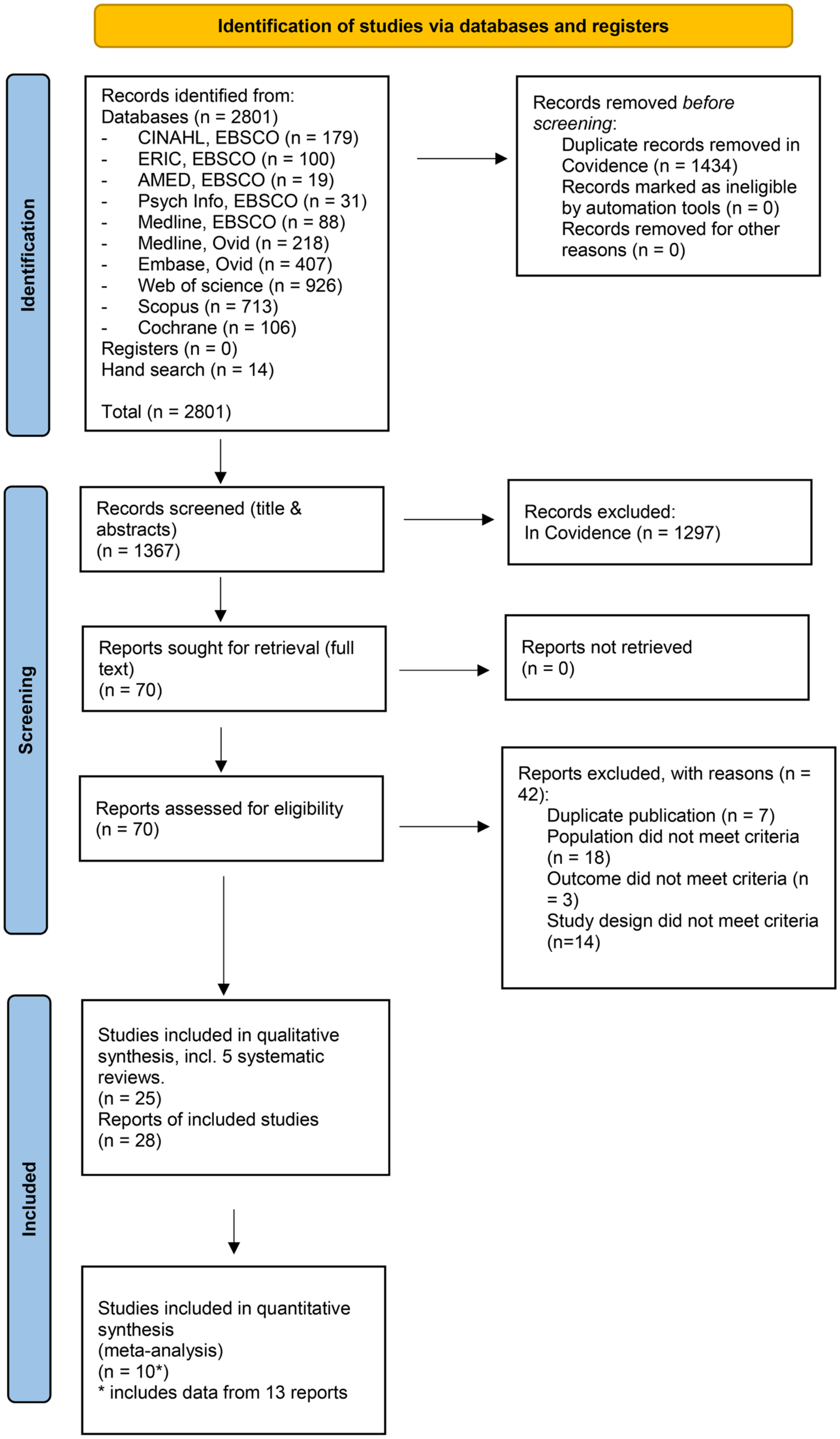

Figure 1. PRISMA 2020 flow diagram of included studies in the review and meta-analysis.

[68,97,104,109,110,114]. Two studies engaged participants in both individual and group training [68,114]. In the FAST Club physical activity study [68], participants received individual coaching and physical activity training in a group context. In a study by Vidal et al. [114] dog assisted training was used to support self-regulation and social skills via individual and group sessions [114] (Table 1).

Technology assisted training. Computers and other technology delivered individual training, targeting specific skills, were found in seven studies with positive intervention effects [58,67,99,100,103,105,106,111,112]. Technology included computer games to teach meta-cognitive strategy skill learning [67,99,100,103,106], fire safety [111], a computer program to improve attention skills [105], a virtual reality balance training system [58], and a mindfulness app [112].

Table 1. Study characteristics and key findings.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
1	Adnams [97] Improve academic skills Country: South Africa Funding: NR	II 3 arm RCT Some risk of bias concerns	$n = 65$ Male: 53% Guardian care: NR Mean age: 9 years Age range: NR FASD criteria: Revised IOM FASD diagnosis: FAS 67.5% pFAS 20% deferred 7.5% Ethnicity: NR	**Language and literacy training (LLT)** School-based group SLP $n = 20$ Delivered by SLP	FASD-C (no intervention) $n = 20$ Non-FASD – C (no intervention) $n = 25$	• 30′ session • 2x/week • 9 months	• Academic skills (UCT Reading test and Spelling tests; Ballard addition and subtraction tests); direct measure. • Language skills (PAELT); direct measure.ICF level: A fPRC: AC	• No statistically sig. gains of the LLT group compared to control group in scholastic tests. • Statistically sig. gains in LLT group in some sub-tests of PAELT compared to control group ranging from an overall effect $d = 0.31$ to 1.2. • Most gains for LLT group were educationally significant >0.25. • All groups showed improvements on scholastic and language scores.
2	Bertrand [61] Reduce child problem behaviours Country: USA Funding: CDC Federal funding.	III-1 RCT-BR High risk of bias	$n = 52$ Male: 52% Guardian care: 87% Mean age: 8 years Age range: 5-11 years FASD criteria: 4 digit code FASD diagnosis: FAS 7.7% Ethnicity: 50% Caucasian / Hispanic 25% African American 25% NR	**Families Moving Forward** Home-based individual caregiver training & consultation with teachers & others Based on positive behaviour support techniques Provided by psychologists or occupational therapists or both $n = 26$ Delivered by mental health providers	Standard care $n = 26$	• 16 × 90′ sessions • 1 x/fortnight • 9-11 months	• Child behaviour (ECBI Problem score); caregiver ratedBehaviour: externalising	• FMF group showed statistically sig. decrease in number of challenging behaviours $p < 0.05$.
3	Clark [98] Improve classroom behavior Country: Canada Funding: Victoria Foundation FASD Action Fund.	III-1 Mixed methods incl. pseudo-RCT High risk of bias	$n = 22$ Male: 77% Guardian care: 69% Mean age: 7 years 9 months. Age range: 6-12 years FASD criteria: Gestalt diagnostic guidelines FASD diagnosis: FAS 100% Ethnicity: 62% Aboriginal 38% Caucasian	**Teacher PD program** School-based group and individual teacher training $n = 7$ Delivered by interprofessional team (psychology, OT, SLP)	Not described $n = 6$	• Ran over a school year • 2 x full day and 4 x half day workshops • Weekly mentoring	• Academic skills (CBM); teacher rated • Classroom behaviour (BASC-2TRS & SOS); teacher and clinician ratedICF levels: A fPRC: AC Behaviour: externalising, internalising, adaptive functioning	• No statistically sig. change in academic achievement. • Statistically sig. differences in adaptive skills $p = 0.02$ and school problems $p = 0.04$ reported by teachers in experimental group. • No statistical sig. changes observed in experimental group behaviour.
4a	Coles [99] Improve attention, behaviour, and adaptive functioning Country: USA Funding: National Institute of Health/ National Institute of Alcohol Abuse & Alcoholism grant	II 3 arm RCT High risk of bias	$n = 30$ Male: 60% Guardian care: 96% Mean age: 7 years 1 month Age range: 5-10 years FASD criteria: IOM FASD diagnosis: FAS n NR pFAS n NR Ethnicity: 47% Caucasian, 30% African American23% mixed/other	**GoFAR** Computer-based MC training with individual coaching & individual caregiver training $n = 10$ **Faceland** Computer-based emotion recognition training with individual coaching & individual caregiver training $n = 10$ Dyad Behavioural analogy therapy (BAT) to reinforce FAR technique (Focus and plan, Act, Reflect) Both GoFAR and Faceland received BAT Clinic-based with home practice for BAT Delivered by clinical psychologists	No intervention $n = 10$	• 1 hour session (concurrent child computer training & caregiver training) • 1 x week • 5 weeks • 1 h/ BAT session • 1x/week • 5 x weeks	• Neurocognition (TOVA, NESPY); direct measure. • Child behviour (CBQ, CBCL, BRIEF); caregiver reported. • Adaptive functioning (VABS-2); caregiver reported.ICF levels: BFS & A fPRC: AC Behaviour: adaptive functioning, emotional control & regulation, externalising, internalising	• Sig. improvements in attention (TOVA API score) between GoFAR, Faceland and control groups $p < 0.05$. • GoFAR group showed sig. reduction in fear score (CBQ) compared to Faceland and control groups $p < 0.01$. • Both GoFAR and Faceland groups showed sig. improvement on daily living skills (VABS-2) compared to control group $p < 0.02$.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
4b	Coles [100] Improve behaviour Country: USA Funding: National Institute of Health/ National Institute of Alcohol Abuse & Alcoholism grant	II 3 arm RCT Some risk of bias concerns	$n=30$ Male: 60% Guardianship: NR Mean age: 7 years 1 month Age range: 5-10 years FASD criteria: IOM FASD diagnosis: FAS n NR pFAS n NR Ethnicity: 47% Caucasian, 30% African American23% mixed/other	**GoFAR** (see Coles et al. 2018) $n=10$ **Faceland** (see Coles et al. 2018) $n=10$ Dyad Behavioural analogy therapy (BAT) to reinforce FAR technique (Focus and plan, Act, Reflect) Both GoFAR and Faceland received BAT Clinic-based with home practice for BAT Delivered by clinical psychologists	No intervention $n=10$	• See Coles et al. [100]	• Child's game play • Ave session time (mins); computer rated • Attention to task; clinician rated • Enthusiasm; clinician rated • No. prompts to continue play; clinician rated • Enjoyment; child rated • Disruptive behaviour; caregiver reported.fPRC: C, At, I Behaviour: externalising	• A sig. session effect $p<0.044$ and session by group $p<0.047$ were found for total game time. • No overall time point effect, but a sig. effect on the disruptive behaviour composite, $p<0.035$. • Session effects were found on attention to task $p<0.005$, enthusiasm $p<0.044$ and prompts to continue to play $p<0.016$ • No sig. differences between Go FAR and Faceland on child's enjoyment rating. • Both GoFAR and Faceland groups showed reduced disruptive behaviour.
4c	Kable [103] Improve self-regulation Country: USA Funding: National Institute of Health/ National Institute of Alcohol Abuse & Alcoholism grant	II 3 arm RCT High risk of bias	$n=30$ Male: 64% Guardianship: 96% Mean age: 7 years Age range: 5-10 years FASD criteria: IOM FASD diagnosis: FAS n NR pFAS n NR Ethnicity: 47% Caucasian, 30% African American23% mixed/other	**GoFAR** Computer-based MC training with individual coaching Individual caregiver training $n=10$ **Faceland** Computer-based emotion recognition training with individual coaching Individual caregiver training $n=10$ Delivered by clinical psychologists *NB: No BAT in this study*	No intervention $n=10$	• 1 h session (concurrent child computer trianing & caregiver training) • 1 x week • 5 weeks	• Disruptive child behaviour; caregiver reported.Behaviour: externalising, emotional control & regulation	• No sig. differences between groups in disruptive behaviour $p=0.134$.
5	Copeland [101] Independent homework and bedroom cleaning task completion and improve behaviour Country: USA Funding: NR	IV Single case study 12-month follow-up High risk of bias	$n=1$ Male: 100% Guardianship: Adopted Age: 9 years FASD criteria: NR FASD diagnosis: ARND 100% Ethnicity: Indigenous American	**Individual self-monitoring** Home-based individual child and caregiver training sessions. Profession delivered sessions Certified behaviour analyst (special education)		• 14 x sessions • Mean training 26' • Mean intervention 38'(Session frequency NR)	• Child task completion; observation/ checklists • Intensity of challenging; clinician rated behaviour (ECBI); caregiver ratedICF levels: A fPRC – AC Behaviour: externalising	• Effect size $\tau=0.79$ indicated large magnitude of change on task completion. • Reduction in intensity of problem behaviours however slight increase in number of problem behaviours seen.
6	Griffin & Copeland [102] Independent homework and bedroom cleaning task completion and improve behaviour Country: USA Funding: NR	IV Single case study High risk of bias	$n=1$ Male: 100% Guardianship: NR Age: 11 years FASD criteria: NR FASD diagnosis: ARND 100% Ethnicity: Hispanic	**Individual self-monitoring** Home-based individual child and caregiver training sessions. Profession delivered sessions NR		• 20 x sessions(Frequency and length NR)	• Child task completion; observation/ checklists • Reduction in argumentative statements; clinician rated • Challenging behaviour (ECBI); caregiver ratedICF levels: A fPRC – AC Behaviour: externalising	• Improved behaviour during training/interventions. • Counter therapeutic effect at baseline and reversal. • Slight reduction in number and intensity of problem behaviours. • Increased task completion that was maintained.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
7	Jirikowic [58] Improve balance Country: USA Funding: NR	III-2 CCT 3 arm pilot One month follow-up Serious risk of bias	$n = 29$ Male: 53% Guardian care: 96% Mean age: 10 years 8 months Age range: 8-15 years and 8 months FASD criteria: 4-digit code FASD diagnosis: FAS 7.5 % SE-AE 48.3% ND-AE 44.2% Ethnicity: NR	**STABLE Clinic** Individual task-specific balance practice delivered via VR game using a pliable surface. Delivered in clinic. $n = 6$ Delivered by physiotherapist **STABLE Home** As above yet delivered at home. $n = 15$	No intervention $n = 8$	• 30-35′ sessions • 1 to 2 x /week • 1 x/month	• Balance (MABC2 total movement score and balance sub-score; P-CTSIB-2 total score and 3 sensory systems); direct measure • Gait (DGI); direct measureICF Level: BFS	• Sig. differences on MABC2 balance scores by session $p = 0.02$ and group $p = 0.04$. • Sig. treatment effects for MABC2 total movement scores ($p = 0.05$) • Sig. total sensory score in home STABE group compared to control group $p = 0.02$. • No sig. effects found on gait scores for any group pre- and post- intervention. • No sig. difference in scores one-month post- intervention for either STABLE group $p = 0.11$.
8	Kerns [105] Improve attention Country: Canada Funding: Victoria Foundation and Ministry of Children and Family Development	IV Pre-post single arm Serious risk of bias	$n = 10$ Male: 60% Guardian care: NR Mean age: 12 years 3 months Age range: 8-15 years FASD criteria: NR FASD diagnosis: NR Ethnicity: NR	**Computerised Progressive Attention Training (CPAT)** School-based individual computer attention training with coaching using MC strategies. $n = 10$ Delivered by computer & supervised by psychologists and learning assistant integration support teacher.		• 30′ sessions • 4 x/week • Total 16 h	• Working memory (WISC-III/NI, CSOT); direct measure • Attention (ANT-C, KiTAP, TEA-Ch) • Literacy (WJ-III test of maths and reading fluency); direct measure-ICF levels: BFS & A fPRC: AC	• Fewer commission error effects and increase in reaction time effects. • Large attention effects: neutral attention conditions $d = 1.01$, incongruent attention conditions $d = 1.05$, auditory sustained attention $d = 1.26$. • Largest effect for reading fluency effect $d = 1.40$ $p < .01$.
9	Makela [106] Learn to use metacognitive strategies Country: NR Funding NR	III-3 Single arm ITS Moderate risk of bias	$n = 7$ Male: 71% Guardian care: NR Mean age:12 years Age range: 8-16 years FASD criteria: NR FASD diagnosis: NR Ethnicity: NR	**Cognitive Carnival** School-based computer MC training with individual coaching for strategy use. $n = 7$ Delivered by computer & supervised by research assistants profession NR.		• 60-90′ sessions • 2-3x/week • 12 x/weeks • 720′ total	• No. metacognitive strategies used via prompting or spontaneous; clinician rated.ICF levels: A fPRC: AC & C	• Sig. increase in spontaneous use of strategies over time *p=.037* • Sig. reduction in prompting over time $p = 0.014$.
10	Millians and Coles [107] Improve learning and academic skills Country: USA Funding: Gift from the Spray Foundation.	IV Case studies Serious risk of bias	$n = 5$ Male: 40% Guardian care: 100% Mean age: 12 years Age range: 10-13 years FASD criteria: IOM FASD diagnosis: FAS 40% pFAS 20% no alcohol related Dx 20% deferred 20 % Ethnicity: 20% Caucasian, 80% African American	**Saturday Cognitive Habilitation Program** Individual literacy tutoring with MC coaching. Clinic-based with home practice. $n = 5$ Delivered by a special education teacher.		• 1 h/ session incl. 50′ direct instruction & 10′ caregiver feedback • 20 x sessions	• Academic skills (Varied literacy standardised tests for each child); direct measure.ICF Level: A fPRC: AC	• 4/5 participants made clinically sig. gains in reading comprehension, maths reasoning and non-verbal reasoning. • No gains in word reading or recall of mathematical facts.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
11	Nash [108] Improve executive function, behaviour, and social skills Country: Canada Funding: NR	III-2 CCT 6 months follow-up Serious risk of bias	$n = 25$ Male: 52% Guardian care: NR Mean age: 10 years Age range: 8-12 years FASD criteria: Canadian Diagnostic Guidelines & 4-digit code FASD diagnosis: FAS 0% pFAS n NR ARND n NR Ethnicity: NR	**Alert Program** Individual, manualised Alert Program with caregiver support. Clinic-based $n = 12$ Delivered by post-graduate doctoral students profession NR.	Delayed treatment $n = 13$	• 1-hour sessions • 1 x/week • 12 x weeks over 14 weeks	• Cognitive executive function (NEPSYII, TEA-Ch, CANTAB); direct measure. • Socio-affective executive functioning (NEPSYII, TSC); direct measure. • Emotional/behavioural functioning and social skills (BRIEF, CBCL, SSIS-P); caregiver rated.ICF levels: BFS & A fPRC: AC Behaviour: emotional control & regulation	• Sig. treatment effect for improvements in inhibition-naming $p = 0.001$. • Sig. treatment effect for affect recognition $p = 0.05$. • Sig. treatment effect for improved caregiver reported emotional control $p = 0.03$ and reductions in externalising behaviours $p = 0.08$. • No change in caregiver-reported social skills. • Improvements maintained over 6 months.
12a	O'Connor [109] Improve social skills Country: USA Funding: CDC Federal funding grant.	III-2 CCT 3 months follow-up Serious risk of bias	$n = 100$ Male: 51% Guardian care: 79% Mean age:8 years Age range: 6-12 years FASD criteria: 4-digit diagnostic code FASD diagnosis: FAS 11% pFAS 43% ARND 46% ARBD 0% Ethnicity: Caucasian 54%, African American 17%, Hispanic 17%, Asian 2%, Mixed 10%	**Children's Friendship Training (CFT).** Group social skills manualised training. Caregiver training provided. Clinic-based with home practice. $n = 51$ Delivered by clinical psychologists	Delayed treatment $n = 49$	• 90' sessions • 1 x/week • 12 x weeks	• Child social skill knowledge (TSSK); self-report • Social skills and problem behaviours (PSSRS-SS; PSSRS-PB; TSSRS-SS; TSSRS-PB); caregiver & teacher rated.ICF levels: A fPRC: AC Behaviour: externalising	• Improvement social skill knowledge $p < 0.0001$ and large treatment effect d 1.28. • Improved social skill reported by caregivers compared to control $p < 0.03$ and large treatment effect, $d = 0.52$. • Reduced problem behaviours reported by caregivers compared to control $p < 0.05$ and moderate treatment effect $d = 0.40$. • No teacher rated improvements in social skills. • Improvements in social skill knowledge maintained over 3 months compared to control $p < 0.0001$, with large treatment effect d 1.73. • Improved social skill reported by caregivers maintained over 3 months compared to control $p < 0.0001$ with large treatment effect $d = 1.15$.
12b	Keil [104] Improve social skills. Country: USA Funding: CDC Federal funding grant.	III-2 CCT 3 months follow-up Moderate risk of bias	See O'Connor et al. 2006	See O'Connor et al. 2006	See O'Connor et al. 2006	See O'Connor et al. 2006	• Hostile attributions (HAT provocation and group entry measures); direct measure.Behaviour: externalising	• Sig. reduction in hostile attributions in peer group entry compared to control $p < 0.05$ and moderate treatment effect $d = 0.42$. • No sig. effect for reduction in provocation $p = 0.88$. • Decline in hostile attributions was maintained over 3 months compared to control $p = 0.65$.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
13	O'Connor [110] Improve social skills Country: USA Funding: CDC Federal funding grant.	III-2 CCT Moderate risk of bias	$n = 85$ Male: 78% Guardian care: NR Mean age: 8 years Age range: 6-12 years FASD criteria: 4-digit code FASD diagnosis: FAS 0% pFAS 31.3% ARND 38.8% Ethnicity: Caucasian 24.7%	**CFT** Group social skills manualised training & caregiver training. Community centre-based with home practice. $n = 41$ Delivered by mental health professionals profession NR.	Standard care social skills program without caregiver training. $n = 44$	• 90′ sessions • 1 x/week • 12 x weeks	• Child social skill knowledge (TSSK); direct measure. • Child self-concepts (Piers Harris-2); child rated. • Social skills (PSSRS-SS); caregiver rated.ICF level: BFS & A fPRC: AC & SS	• Improved child social skill knowledge compared to control $p < 0.0001$. • Improved overall self-concept compared to control $p < 0.05$. • No improvement in overall caregiver rated social skills compared to control group $p = 0.12$. • Improvement on assertion $p < 0.05$ and responsibility $p < 0.04$ compared to control.
14	Padgett [111] Improve fire safety skills Country: USA Funding: National Institute of Health, National Institute on Alcohol Abuse & Alcoholism Small Business Innovation research grant	IV Multiple baseline, multiple probe pre-post case series High risk of bias	$n = 5$ Male: 80% Guardian care: 100% Mean age: 6 years Age range: 5 -7 years FASD criteria: NR FASD diagnosis: FAS 60% pFAS 40% Ethnicity: Caucasian 80%, African American 20%	**VR Fire Safety Training** Individual clinic-based with computer program simulation of steps. $n = 5$ Delivered by computer. Profession of examiner NR.		• 2 x visits (duration NR) • Second visit 1/week after first visit	• Correct ordering a set of 3 pictures representing fire safety steps. • Correct response to simulated fire in building of 3 steps.ICF level: A fPRC: AC	• After intervention 5/5 completed all 3 steps in virtual world. • Immediately after intervention 4/5 correctly sequenced pictures and responded to steps 1 and 2 of simulation. 5/5 responded to simulation step 3. • 1-week post-intervention 3/5 correctly sequenced pictures and 5/5 responded to simulation of all 3 steps.
15	Pei [67] Improve underlying impaired cognitive processes and activate neural attentional processes. Country: Canada Funding NR	II Pilot RCT Efficacy RCT High risk of bias	$n = 18$ pilot RCT $n = 21$ RCT Male: NR Guardian care: NR Mean age: NR Age range: 6-12 years FASD criteria: NR FASD diagnosis: NR Ethnicity: NR	**Cognitive Carnival** School-based computer MC training with individual coaching for strategy use Pilot RCT $n = 9$ Efficacy RCT n = NR Delivered by computer & interventionist. Profession of interventionist NR.	Alternate educational computer game with coaching Pilot RCT $n = 9$ Efficacy RCT Delayed treatment control n = NR	• 30′ sessions • 24 x sessions	• Changes in cognitive skills; direct measure. • Persistence; computer rated. • Total no. strategies used via prompting or spontaneous; clinician rated. • Diffusion tensor imaging (DTI).ICF level: BFS fPRC: AC & C	• Both groups improved on several cognitive measures in memory, attention, and quantitative tasks. • Participants used up to 25 strategies. • Less prompting for strategies used by end of game play. • Differences between groups on DTI. • Sig. attention and working memory improvements maintained 3 months post-intervention.
16	Pritchard Orr [68] Improve executive function Country: Canada Funding: Victoria Foundation grant	III-2 CCT 2 months follow-up Moderate risk of bias	$n = 30$ Male: 47% Guardian care: 87% Mean age: 10 years Age range: 7-14 years FASD criteria: Canadian Diagnostic Guidelines & 4-digit code FASD diagnosis: FAS 10% pFAS 33.3% ARND 50% PAE without FASD 6.7% Ethnicity: NR	**FAST Club physical activity (PA) program** Individual PA program within a group setting Based at a school gymnasium. $n = 15$ Delivered by student instructors profession NR.	Delayed treatment $n = 15$	• 90′ sessions • 2 x sessions/ week • 8 weeks	• Executive function (CCTT1 and CCTT2); direct measure.ICF level: BFS	• No differences seen in CCTT1 T scores $p = 0.173$. • Sig. difference seen in CCTT2 T scores $p = 0.014$ with medium treatment effect $d = 0.46$.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
17	Reid [112] Reduce respiratory sinus arrhythmia Country: Australia Funding: Griffith University PhD scholarship, Australian Government Research Training Program Scholarship.	III-2 CCT Serious risk of bias	$n = 34$ Male: 64% Guardian care: NR Mean age: 8 years, 6 months Age range: 6-10 years FASD criteria: 4 digit code FASD diagnosis: pFAS 13% ND-AE 40% SE-AE 40% Ethnicity: Caucasian 35.7%, racial background 64.3%	**Mindfulness** Body scan task using Smiling Mind App Clinic-based $n = 14$ Delivered by mindfulness app	Mindfulness Typical developing children $n = 20$	• 5′40 session • 1 x session • 5′ of neutral video prior to mindfulness	• Respiratory sinus arrhythmia (RSA); heart rate activity across band of frequencies of spontaneous breathing; direct measure. • Child mindfulness compliance; observed behaviours.ICF Levels: BFS & A fPRC: AC	• Both groups demonstrated sig. increase in RSA during intervention $p < 0.0001$. • No sig. between group effect on RSA was found $p = 0.70$. • Both groups of participants complied with mindfulness instructions.
18	Soh [113] Improve emotional control and inhibition; and change neuroanatomy underlying self-regulation Country: Canada Funding: Canadian Institute of Health Research, trainee scholarship & student scholarship	III-1 3-arm RCT Some risk of bias concerns	$n = 65$ Male: 45% Guardian care: NR Mean age: 9 years Age range: 8-12 years FASD criteria: Canadian Diagnostic Guidelines & 4-digit code FASD diagnosis: FAS 3.4% pFAS 13.8% ARND 75.9% Unspecified 6.9% Ethnicity: NR	**Alert Program** Individual, manualised Alert Program Clinic-based $n = 20$ Delivered by clinical psychologists	Typical developing children (TDC) No intervention $n = 27$ Delayed treatment group $n = 13$	• 90′ sessions • 1 x/week • 12 x weeks	• Emotion regulation (BRIEF); caregiver rated. • Inhibition (NEPSY-II); direct measure. • Brain structure and function (MRI); direct measure.ICF Level: BFS Behaviour: emotional control & regulation	• Alert group showed sig. improvements in caregiver reported emotional regulation compared to control $p < 0.0001$. • Experimental group showed sig. improvements in caregiver reported inhibition compared to control $p < 0.001$. • Alert group showed marginally sig. larger grey matter volume than control in the left superior frontal gyrus, $p < 0.005$.
19	Vidal [114] Improve social skills and reduce severity symptoms Country: Spain Funding: Grant from Fundacion Probitas.	II RCT High risk of bias	$n = 33$ Male: 67% Guardian care: NR Mean age: 12 years Age range: 6-18 years FASD criteria: Hoyme criteria FASD diagnosis: FAS 39.4% pFAS 33.3% ARND 27.3% Ethnicity: NR	**Dog Assisted Therapy (DAT)** Individual and group manualised DAT program to attain individualised goals. Clinic-based. Groups involved 3-4 participants. Psychiatrist monitored medication adherence. $n = 17$ Delivered by DAT professionals, certified therapy dogs and a psychologist.	Treatment as usual (TAU), no DAT Psychiatrist monitored medication adherence. $n = 16$	• 45′ sessions • 1 x/week • 12 weeks, incl. 6 x individual and 6 x group sessions	• Behaviour (CBCL); caregiver rated. • Social skills (SSIS-P); caregiver rated. • Symptom severity (CGI-S); clinician rated.ICF Levels: A fPRC: AC Behaviour: externalising, internalising	• Large treatment effect on social skills $d = 0.8$. • Moderate treatment effect on reduction of externalising symptoms $d = 0.56$ • No treatment effect on internalising symptoms. • Medium treatment effect on severity of symptoms $d = 0.5$.
20	Wells [115] Improve executive functioning, emotional and problem-solving skills Country: USA Funding: CDC Federal funding grant.	II RCT High risk of bias	$n = 78$ Male: 68% Guardian care: 100% Mean age: 8 years Age range: 6-11 years and 11 months FASD criteria: NR FASD diagnosis FAS 26.9% pFAS 12.8% ARND 60.3% Ethnicity: Caucasian 37%, African American 5%, Indigenous American 1%, mixed 15%	**Neurohabilitation Program** and extensive psychology feedback and report Group-based manaualised integrated Alert Program with other TBI strategies Concurrent caregiver education Session dyads to practice learning. Clinic-based $n = 40$ Delivered by OT and psychologist.	No program Extensive psychology feedback and report $n = 38$	• 75′ therapy/education session including 15-30′ dyad practice. • 1 x/week • 12 weeks	• Executive functioning (BRIEF); caregiver report. • Emotional and social problem solving (RATC); direct measure.ICF levels: BFS Behaviour: adaptive functioning, emotional control & regulation	• Sig. treatment effects executive function $p = 0.006$, emotional and social problem-solving abilities $p < 0.01$.

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
21	Flannigan [9] To identify interventions to improve mental health and substance use outcomes for individuals with PAE and FASD Country: Canada Funding: NR	Unable to rate LOE due to use of mixed study designs Systematic Review Critically low	$n = 932$ Age range: 1-50 years	Searched qualitative, quantitative and mixed methods published studies with outcomes related to mental health and/or substance abuse. Searched from 2000 onwards. Published in English. 4 databases searched. Qualitative synthesis.	33 studies included: 12 RCTs, 8 CCTs, 1 file review, 1 implementation study, 1 exploratory study. Interventions targeted: family attachment and wellness (n = 9), self-regulation (n = 5), social skills (n = 6), behavioural ($n = 5$), mental health literacy ($n = 1$), responding to risk and reducing harm ($n = 5$), substance use ($n = 4$), justice ($n = 1$). 12 studies had weak methodological rigour, 10 had moderate and 10 had strong. 1 qualitative study had adequate validity, results, and research value. Most common age group of participants was middle childhood (6–12 years). Very few studies focused on mental health or substance use as a primary target of interventions. Interventions targeting self-regulation and social skills had the strongest evidence.	• Much current intervention focuses on improving functioning and underlying skills that indirectly influence mental health. • Found "favourable and emerging evidence for interventions to support attachment and family wellness, build skills and strategies and respond to risk and reduce harm." [9](p.23) • Did not find any "evidence on effective mental health treatment approaches or best practices for individuals with FASD" [9](p. 24) and a "stark absence of traditional psychotherapeutic interventions to improve mental health or substance outcomes for individuals with PAE/FASD." [9](p. 25)		

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Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
22	Ordenewitz [8] To identify evidence-based interventions for children and adolescents with FASD developed over the last two decades Country: Germany Funding: The Bavarian State Ministry of Health and Care, the German Federal Ministry of Health, the private foundation of Robert-Vogel-Stiftung, and Soziale Bertungs-und Betresuungdienste (SBB) Bayer gGmbH.	I Systematic Review Critically low	$n = 968$ Age range: 2-18 years	Searched intervention RCTs published in English, German, or French since 1.1.2000. Search conducted September 2019. 7 databases searched. Qualitative synthesis. Methodological quality was not assessed.	• 25 RCTs included, 12 were published since 2015. • Interventions targeted: language/speech, learning/memory skills, executive functions, arithmetic skills, attention, social skills and behaviour, family capacity. • Found evidence for intervention effects of scholastic skills, attention, executive functioning, behaviour. • Questionable impact of choline supplement on the effects on memory. • No RCTs were found investigating fine motor or visual spatial skills or medication effects of attention. • Only 1 study targeted adolescents.	• Few intervention RCTs exist for children and adolescents with FASD. • Identified some positive benefits from training of functional deficits particularly combination of caregivers' and children's sessions. • Interventions targeting self-regulation and social skills appear to have a long-term, and attention and behaviour effects.		
23	Peadon [50] To identify and evaluate evidence for pharmacological and non-pharmacological interventions for children with FASD Country: Australia Funding: Drug & Alcohol Services Australia NHMRC Program Grant, fellowship & post-graduate scholarship.	Unable to rate LOE due to use of mixed study designs Systematic Review Critically low	$n = 347$ Age range: 3-16 years	Searched intervention RCTs, quasi RCTS, non-RCTs and cohort studies with pre and post controlled studies evaluating pharmacological and non-pharmacological interventions. Searched between 1950-2009. No language restrictions. Grey literature searched. 6 databases searched. Qualitative synthesis.	• 12 studies included: 6 RCTs, 1 quasi RCT, 1 CCT. • Interventions included pharmacological (n = 2), education and learning strategies (n = 7), social skills & communication (n = 2), behavioural (n = 1). • Some evidence for language and literacy, maths, attention process training, stimulant medication, virtual reality, cognitive control therapy and social skills training. • Different FASD criteria used and short-term follow-up. • Identified 7 studies in progress.	• Identified an "urgent need for more high-quality intervention research." [50](p. 7) • Limited evidence for specific interventions for children with FASD. • Significant methodological problems with included studies randomisation, concealment, blinding, very small sample sizes limiting power to detect a true treatment effect.		

(*Continued*)

Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			
24	Premji [51] To identify intervention studies to improve outcomes for children and youth with a FASD and their families. Country: Canada Funding: The Alberta Centre for Child and Family Community Research.	Unable to rate LOE due to use of mixed study designs Systematic Review Critically low	*n* = 36 Age range: 0-18 years	Searched RCTs, quasi-RCTS and case-studies. Searched between 1973- July 2004. No language or study design restrictions. Grey literature searched. 40 databases searched. Qualitative synthesis.	• 2 RCTs & 1 quasi RCT included. • Intervention involved cognitive control therapy and medication. • Outcome measures were IQ, teacher caregiver rated behaviour.	• No conclusions drawn regarding intervention effectiveness. • Identified studies lacked scientific rigour. • Identified a " dire need to conduct rigorous intervention research in this area." [51](p.394)		
25	Reid [52] To analyse the available literature on interventions for people with FASD across the lifespan. Funding: NR Country: Australia	Unable to rate LOE due to use of mixed study designs Systematic review Critically low	*n* = 32 Age range: across the lifespan	Searched RCTs, quasi RCTs and case-studies in December 2014. No language or document type or study design restrictions. Grey literature searched. 7 databases searched, 40 FASD organisations and webpages searched. Qualitative synthesis.	• 32 studies included: • Interventions targeted: improve infant outcomes (n = 2), underlying self-regulation deficits or attentional control (*n* = 6), social skills (*n* = 6), improve children's behaviour and reduce parental stress (*n* = 3), caregiver education and advocacy, (*n* = 4), support parents affected by PAE (*n* = 2). • 0 studies were rated as strong for selection bias or blinding; 19 studies were rated as strong for study design; 29 studies used measures that were valid and reliable; 17 studies rated strong for withdrawal/ dropouts. • Found mixed evidence for effectiveness in early infancy. • Studies focusing on improving self-regulation and attentional control in early to middle childhood provided strong evidence for gains, however these had limited follow-up. • The evidence for changes in specific skills was variable. • The studies improving social skills showed consistently strong results.	• Despite small sample sizes and limited follow-up, the literature showed possibility to improve functioning. • Identified paucity of long-term follow-up.		

(*Continued*)

Table 1. Continued.

#	Reference, aim, country & source of funding	LOE, design & overall risk of bias	Sample characteristics*: % male, % guardian care, Mean age (SD) & age range, FASD criteria, % type of FASD diagnosis, ethnicity	Intervention		Intervention format	Outcome of interest ICF, fPRC and behaviour classification**	Summary of results
				Experimental	Control			

A :activity; AC: activity competence; ANT-C: attentional network test adapted for children; ARND: alcohol related neuro-developmental disorder; ARBD: alcohol related birth defects; At: attendance; BASC 2-TRS: Behavior Assessment System for Children 2nd edition Teacher Rating Scale; BFS: body functions & structures; BRIEF: behior rating scale of executive function; C – context; CANTAB – Cambridge neuropsychological test automated battery; CBCL: child behavior checklist; CBM: curriculum based measure; CBQ: child behavior Questionnaire; CCT: case control trial; CCTT1: children's color trail test 1; CCTT2: children's color trail test 2; CGIS: clinical global impression scale; CSOT: children's size ordering task; DGI: dynamic gait index; ECBI : eyeberg child behavior inventory; FAS: fetal alcohol syndrome; FASD :C fetal alcohol spectrum disorders control; fPRC:family of participation related constructs; HAT: hostile attribution tendency score; I: involvement; ICF: international classification and functioning; IOM: institute of medicine guidelines; ITS: interrupted time series; KiTAP: test of attentional performance for children; LLT: language literacy training; LOE: level of evidence according to NHMRC 2009; MABC2: movement assessment battery for children 2nd edition; MC: metacognitive; MRI: magnetic resonance imaging; ND-AE: neurobehavioral disorder alcohol exposed; NEPSY: A Developmental Neuropsychological Assessment; NEPSY II: A developmental neuropsychological assessment 2nd edition; Non-FASD-C: non FASD control; NR: not reported; PAE: prenatal alcohol exposure; PAELT: phonological awareness and early literacy test; P-CTSIB-2: pediatric clinical test of sensory integration for balance 2nd edition; pFAS: partial fetal alcohol syndrome; PSSRS-SS: parent social skills rating system social skills; PSSRS-PB: parent social skills rating system problem behaviors; RATC Robert's appreciation test for children; RCT: randomised control trial; RCT-BR – randomised control trial block randomisation; SE-AE- static encephalopathy alcohol exposed; SLP: speech language pathology; SS: sense of self; SSIS-P: social skills improvement system parent rated; SOS: student observation system; TBI: traumatic brain injury; TEA-Ch: test of everyday attention for children; TSC: test of social cognition; TSSK: test of social skills knowledge; TSSRS-PB: teacher social skills rating system problem behaviors; TSSRS-SS - Teacher Social Skills rating system social skills; TOVA: test of variable attention; UCT: University of Cape Town; VABS2: vineland adaptive behavior Scale 2nd edition; VR: virtual reality; WISC III/NI: Wechsler intelligence scale for children 3rd edition: Neuropsychological Investigation; WJ III: Woodcock-Johnson 3rd edition.

In most of these studies, computer-based learning was facilitated with individual coaching from research assistants [58,67,99,100,103]. Only two studies relied on technology alone to enable participant learning and skill development [111,112].

Intensity of intervention

The mean culminated duration of intervention, calculated on 15 of 20 studies, was 976 min or 15 h and 27 min (median 960 min, range 5 – 2160 min) [61,67,68,97,99,100,103–115]. Mean frequency was 1.4 sessions weekly calculated on data from 15 of the 20 studies [58,61,67,68,97,99,100,103–106,108–110,112–115]. The average number of intervention sessions, calculated from 19 reports was 13.8 [67,68,97,99,100,103–115]. The highest number of sessions per week was four occurring in a school (Table 1) [105]. The least frequent intervention occurred fortnightly and involved home visits (Table 1) [61]. Around half of the studies scheduled interventions weekly (Table 1) [99,100,103–105,108–111,113–115]. The longest period over which intervention occurred was 39 weeks of school-based interventions (Table 1) [98]. The shortest duration was one mindfulness session of 5 min, 40 s duration (Table 1) [112].

Location of intervention

Interventions occurred in clinics, participants' homes, and schools. Most interventions were delivered in a clinic setting (11 of 20 studies or 55%) [58,99,100,103–105,107–115]. One of these clinic-based interventions, the Children's Friendship Training (CFT) [104,109,110], involved practice in the home of social skills learned by children in the clinic and caregivers practising coaching skills to support their child's social interaction during the social play at home. School-based interventions were completed in six of the 20 studies (30%) and interventions in participants' homes occurred in four (20%). School-based interventions [67,68,97,105,106] involved withdrawing participants from the classroom. One study focused on supporting teachers [98] however, details about when and where teacher training occurred were not provided.

Outcome measures, as classified by ICF-CY [16] levels, fPRC [18] and behaviour, used to evaluate intervention outcomes (RQ2)

A wide variety of outcome measures were used that covered aspects of BFS, activity, behaviour, or sense of self (Table 1), but not participation. Outcome measures were reported from 20 studies in 23 reports (Table 1). When coded against the ICF-CY levels, [16] 10 were at BFS level, 14 at activity and none at participation. When coded using the fPRC [18], measures included those of attendance (1 outcome), involvement (1), activity competence (15), sense of self (1), and context (3). No study measured child preferences or other environment factors. There were 13 reports that used one or more behavioural outcomes: externalising behaviours (10 studies), internalising (3), emotional control and regulation (5), and adaptive functioning (3).

Both objective and subjective outcome measurement tools were identified. With respect to objective tools, 16 of the 23 reports (Table 1) assessed child performance directly using standardised tools (70%) [58,67,97,99,100,104,105,107–110,112,113,115]. Subjective measures involved teacher and caregiver rating of participant performance. Teacher ratings of participant performance were reported in two reports (9%) [98,109]. Caregiver ratings of participant performance were used in 12 reports (52%) [61,99–103,108–110,113–115]. Participant's experiences were measured twice (9% of reports) [100,110]. As a measure of the fPRC construct involvement [18], participants were asked to rate their enjoyment of the GoFAR computer game intervention [100]. Sense of self construct [18] was measured used once (5% of reports) [110] using the Pier-Harris Children's Self-Concept Scale second edition [120]. Clinicians rated participant observed performance in nine reports (39%) using empirical tools and other checklists created for the research [67,98,100–102,106,111,112,114].

Quality of evidence of interventions (RQ3)

We identified studies across all six LOE of the NHMRC evidence hierarchy [79] (Table 1). This included four systematic reviews that could not be rated because they included mixed study designs with various LOE [9,50–52] and one rated as LOE 1 [8]. The quality of the five systematic reviews were rated as "critically low" using the AMSTAR 2 [64] (Table S8-online-only). Risk of bias were present in all studies. Of the 10 RCTs and pseudoRCTs reports, seven had a "high" risk of bias and three had "some" risk of bias [61,67,97–100,103,113–115] (see Figure S9-online-only). Only one study used randomisation [114]. Blinding of participants, interventionists and/or assessors was difficult to achieve and inconsistently reported. The 10 NRSI reports were considered "moderate" to "serious risk" of bias using the ROBINS-I [74] (see Figure S10-online-only) [58,68,104–110,112]. Confounding factors, subjective outcome measures and insufficient information reported all contributed to risk of bias ratings.

Total internal validity of the RoBiNT scale across the three SCEDs [101,102,111] ranged from three to six out of a possible score of 14 (see Table S11-online-only). Two of the three studies met the criteria for experimental design [101,102] and two met treatment adherence criteria [102,111]. All three studies met minimum standards for interrater agreement. Only one study had "high" methodological rigour and "low" risk of bias [102] as it used an ABAB design with four phases, had high treatment adherence, reported at least three data points in every phase and a high interrater agreement in all phases, except baseline. Two studies [101,111] were rated as "low" methodological rigour and had "high" risk of bias due to less than three data points reported in each phase. Blinding of participants, interventionists or assessors was not reported in any SCED report [101,102,111]. External validity scores across all three SCEDs ranged from 6 to 11 out of a possible score of 16. Only two of the three SCEDs [101,102] described the intervention setting and presented raw data in sufficient detail. All three SCEDs operationally defined the target behaviour effectively [101,102,111]. Only one study [101] reported visual and statistical data analysis. Total RoBiNT scores ranged from 9 to 17 out of 30.

Effectiveness and strength of interventions compared with no intervention or usual care (RQ4)

Intervention effectiveness identified through qualitative synthesis

Authors of all 20 studies reported improvements in targeted outcomes at either BFS, activity and/or behaviour (Table 1). No study measured participation outcomes in home, school, or community environments. Activity competence when completing homework and tidying bedroom in the home was measured in two SCEDs [101,102]. These studies demonstrated the positive effects of behavioural, self-monitoring interventions that were also maintained and reported as acceptable by participants and caregivers. The CFT study [104,109] measured activity competence in the child's home and school environments. The authors [104,109] reported positive effects on child's social skills following intervention rated by caregivers, but no effect on social skills rated by teachers (Table 1). Most studies measured the immediate effects of intervention. Follow-up data were present in five studies [58,68,101,104,108,109] and the follow-up time intervals varied from 1 month [58] to 12 months [101].

Intervention effectiveness identified through quantitative synthesis

Multi-level meta-analysis was conducted with study design entered as a moderator. This analysis showed no differences in the pooled effects between RCTs and NRSIs ($F_{1,\,132} = 0.61$, $p = 0.44$). Therefore, meta-analyses are reported with results from both study designs combined. Our multi-level meta-analysis included 13 reports from 10 studies [58,97–100,103,104,108–110,113–115], 131 effect sizes and a total of 405 participants. A total of 15 moderators (i.e. aspects of BFS, activity and behaviour) were included in the overall model, which demonstrated a small positive intervention effect across all outcomes $g = 0.29$ (95% CI 0.15–0.43 (Figure 2). However, the GRADE [93] certainty of the evidence was rated as low, downgraded due to inconsistency and high risk of bias amongst included studies (Table S12-online-only). There was no evidence of publication bias, indicated by a symmetrical funnel plot (Figure S13-online-only). Low overall heterogeneity of effect sizes (39.6%) was found.

Subgroup analysis indicated a small treatment effect for "activity outcomes," overall $g = 0.36$ (95% CI 0.17–0.54), while the specific categories of academic, language and social skills outcomes were small-to-moderate but generally not significant (Figure 2). A symmetrical funnel plot indicated publication bias was not present (Figure S14-online-only). The GRADE [93] certainty of the evidence was rated as low, downgraded by high risk of bias and serious imprecision due to small optimal information size (i.e. $n < 300$) (Table S12-online-only). There was a small, non-significant treatment effect for "BFS outcomes" $g = 0.33$ (95% CI −0.14, 0.79). The specific (positive) effect on motor outcomes was significant ($g = 0.89$, 95% CI 0.26, 1.53). Social cognition showed a small, positive effect ($g = 0.30$, 95% CI 0.01-0.59). Attention, inhibition, working memory, and verbal fluency sub-categories showed non-significant effects. A symmetrical funnel plot indicated publication bias was not present for BFS outcomes (FIgure S15-online-only). Overall heterogeneity for BFS outcomes was high (70.9%) (Level-2 = 21.3%, Level-3 = 49.6%). The certainty of evidence, using the GRADE approach [93], was rated as very low, downgraded by high risk of bias, very serious inconsistency, serious indirectness, and serious imprecision (Table S12-online-only). There was a small, significant treatment effect for "behavioural outcomes" $g = 0.21$ (95% CI $0.10 - 0.33$); the same magnitude of effect was also seen across sub-categories, but statistically significant for emotional control and regulation and externalising outcomes only. An asymmetrical funnel plot detected publication bias (Figure S16-online-only). The GRADE [93] certainty of evidence was rated as very low, downgraded by high risk of bias, serious indirectness, serious imprecision, and detected publication bias (Table S12-online-only). To inform future research, *post hoc* power analysis on the overall model was conducted using *metapoweR* [121], indicating that the study is likely underpowered (69%) (see Table S17).

Discussion

We found effective interventions (conducted in the home and school) that supported BFS, activity, and behaviour outcomes for school-aged children and adolescents (5–18 years) with FASD. Although it is well understood that children and adolescents with

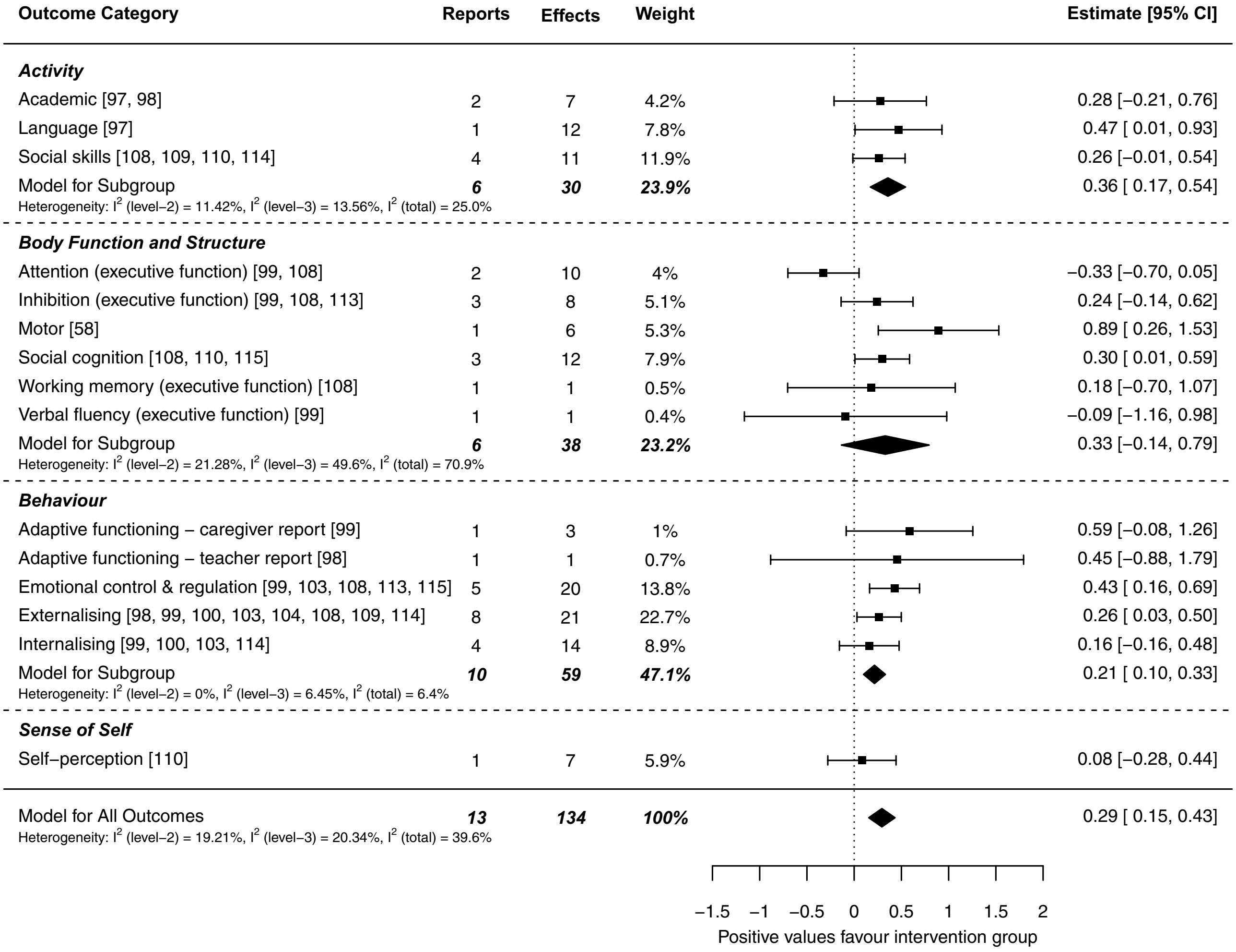

Figure 2. Forrest plot showing the meta-analysis for (a) activity outcomes, (b) BFS outcomes, (c) behaviour outcomes, (d) sense of self outcomes and all treatment effects combined.

FASD experience significant participation challenges in home, school, and community environments [5,7–9,11–13,37,38, 41], participation outcomes and community-based interventions were not investigated in any study included in this review. Our findings indicate that pooled intervention outcomes targeting BFS, activity, and behaviour and self-perception show mild effect (RQ4); however, the certainty of this evidence was rated as low using the GRADE approach [93], providing little confidence in the estimated effect. We identified context-focused interventions used in conjunction with child-focused interventions supported improved activity performance of children and adolescents with FASD in home and school environments [97,98,108–110]. In the remainder of the discussion, we discuss these effects according to the level at which outcomes were assessed.

RQ1: Types of interventions for school-aged children and adolescents with FASD evaluated at ICF [16] levels, fPRC [18] and behaviour

Diverse types of interventions (RQ1) were shown to effect some significant changes in children and adolescents with FASD at the level of BFS, activity performance and behaviour. Most participants were middle childhood aged (8-12 years), although adolescents (13-18 years) participated in nearly one-third of all studies. Modalities included direct skills training with children and adolescents; individual and group sessions; and technology-assisted training to support child and adolescent skill development. These child-focused interventions appear to ameliorate some of the key neurodevelopmental deficits caused by prenatal alcohol exposure (PAE) [1,3,5,6]. Specifically, intervention studies targeted executive functioning [68,99,105,108,113], social cognition [108,110,115], use of metacognitive strategies [67,99,100,103,106], self-regulation and emotional problem solving [108,112,113,115], social and adaptive behaviour [61, 98–104, 108–111,114], attention and working memory [105,108], academic skills [97,98,105,107], and balance outcomes [58]. Earlier systematic reviews have also identified diverse interventions for school-aged children with FASD targeting neurodevelopmental deficits caused by PAE [8,9,50,52].

Consistent with previous reviews, we found only one study [58] that addressed motor performance impairments for children with FASD [59,60] despite the extensive evidence of motor impairment caused by PAE. In this pilot study, participants practised standing balance to complete virtual reality games on a platform and were evaluated at the BFS level of balance, using the Movement Assessment Battery for Children second edition (MABC-2) [122]. Jirikowic et al. [58] reported clinically and statistically significant improvements on the MABC-2 balance sub-scores by session and group, and MABC-2 total scores. We found a positive, significant intervention effect on balance in our meta-analysis from this study, despite suggestions that the MABC-2 is not designed as a measure of change [123]. The study by Jirikowic et al. [58] scored an overall risk of bias as "serious" due to serious bias in confounding and missing data. The intervention effects of improved balance on daily activity and participation were not measured, however, improvements on the MABC-2 were demonstrated.

Contexts of interventions and their influence on activity and participation outcomes

Interventions were mostly delivered in clinic contexts and rarely delivered in the participant's lived environment, where daily participation occurs. Context-situated interventions involve directly practising and learning skills within the environment of their eventual usage. Skill learning is highly context-specific and generalisation (or far transfer) to a broader context is the ultimate goal for training or intervention [124]. Context-specific practice enhances skill learning by providing opportunity for essential elements of learning, namely practice variability, frequency and problem solving [125]. There were four studies providing home-based interventions [58,61,101,102]. School-based interventions involved removing participants from their participation context rather than directly implementing intervention to the classroom or playground [67,68,97,105,106].

Participation, as a target of intervention, in the participant's lived environment was addressed in three studies although not directly measured [101,102,109]. Two SCEDs [101,102] identified in this review used individual self-monitoring behavioural therapy to improve participant's homework and chore completion in participants' homes. Although these studies showed promise and positive effects, they could not be pooled with other SCEDs to strengthen findings of effect. The Children's Friendship Training (CFT) intervention [104,109,110] involved social skills training for children, combined with coaching strategies for caregivers to support their child's participation in a clinic setting and applying these to home practice. The social skills were not directly practised in the school environment. O'Connor and colleagues [109] found large treatment effects on the social skills of children when evaluated by caregivers, but little effect by teacher report; however, the risk of bias for this study was rated as "serious." This pattern of finding was also shown by Storbero et al. [126] who completed a systematic review and meta-analysis on 11 social skills interventions for school-aged children with attention deficit hyperactivity disorder (ADHD).

Effective context-focused approaches, aimed at building teachers and caregiver's capacity to support children's activity performance were identified [61,98–104,108–110,115]. These studies increased caregivers' and teachers' knowledge about FASD and approach to supporting children's behaviour and social skills at home and school. Skorka et al. [12] completed a narrative review exploring the experiences of children with FASD and their families and the impact on their daily function. They advocated a "paramount" [12, p.12] need to use context-focused approaches for children with FASD to support their participation across environments.

Innovative and complimentary interventions for children and adolescents with FASD

Innovative, complimentary therapeutic tools were among those used to support activity performance, body function and behavioural outcomes for children with FASD. Such tools included computer programmes [67,99,100,103,105,106,111], an iPad application [112], virtual reality balance platform [58], dog-assisted therapy [114] and physical activity [68]. Importantly, these tools are readily available in children's participation contexts and may support participation outcomes however participation outcomes were not assessed in these studies. These therapeutic tools were not described in previously published intervention systematic reviews [8,9,50–52] and demonstrate creative, promising interventions to support activity outcomes for school-aged children and adolescents with FASD.

RQ2: Outcome measures used to evaluate interventions for school-aged children and adolescents with FASD as classified by ICF-CY [16] levels, fPRC [18], and behaviour

A wide range of outcome measures were used to detect changes in participant outcomes at ICF-CY [16] levels: BFS, activity and fPRC [18] and behaviour (RQ2). No studies measured participation.

fPRC [18] outcomes measured

Although several constructs of the fPRC [18] were measured, participants' attendance and involvement in daily activities in their lived environment were not measured using dedicated participation metrics. Rather, measurement of participant's attendance and involvement was limited to specific activities like computer game play [100]. Attendance [18] was measured once by measuring participants' time attending to a computer game intervention recorded by a clinician [100]. Involvement [18], while a key element of participation, was evaluated in only one study by asking participants to rate their enjoyment in playing a computer game [100]. This highlights the paucity of self-reported participation measures of children with FASD in the literature. By comparison, activity competence [18] was the most common fPRC construct measured using standardised tools.

Sense of self, another self-reported construct "related to one's confidence, satisfaction, self-esteem, and self-determination" [18, p.19] was also measured once by O'Connor et al. [110] In this study [110], participant's self-perception after playing the computer game was measured using the Piers Harris Self-Concept Scale second edition [120].

Another fPRC construct, context [18], was identified in three studies by measuring the reduction of clinician prompting to support participant's success in playing computer games [67,100,106]. Context measures are useful to inform how others in the environment affect participation. In these studies, clinician prompting was used to support the participant's learning demonstrated through reduction of prompting.

Participation is recognised as a complex, challenging construct to measure and is broadly defined by the ICF-CY [16,21]. A "paradigm shift" towards participation-focused practice in paediatric rehabilitation has been advocated by Anaby et al. [17, p.1747] and achieved by situating the intervention in the child's natural environment, incorporating caregiver education and training elements, and using participation outcome measures. Similar to a recent systematic review by Czencz et al. [21] that explored the effects of exercise on the quality of life and participation of adolescents and adults with cerebral palsy, our review noted the absence of participation measures and community-based interventions for children and adolescents with FASD. In the CFT study [109] included in our review, social skills training outcomes were measured at school and not at the training location of the home and the clinic. Given its recency, researchers and clinicians may not be familiar with using the fPRC [18] to guide participation intervention and outcomes for children and adolescents with FASD and other disabilities. Anaby et al. [17] have recently attempted to support the adoption of participation-focused practice by creating a Participation-Knowledge Translation roadmap emphasising the importance of participation interventions.

Gaps in the measurement of participation and child/adolescent preferences and goals

The exclusion of outcomes measuring child and adolescent preferences, goals and valued life roles is a significant gap identified by this review. There is increasing support to include children's and adolescent's subjective experiences of their healthcare and participation in their daily lives to contribute to intervention goal setting [127–129]. Vroland-Nordstrand et al. [129] demonstrated that children with disabilities can identify and achieve meaningful intervention goals just like their caregivers using the Swedish version of the Perceived Efficacy and Goal-Setting System [130]. The authors [129] found that children reached their self-identified goals and prioritised different goals to their caregivers across a wider range of participation contexts (i.e. home and school). As children with disabilities can participate in goal setting, the absence of studies that evaluate goal setting in children and adolescents with FASD is a notable concern.

Behavioural outcomes of intervention for children and adolescents with FASD

Behavioural outcomes, using caregiver and teacher standardised questionnaires, were the second most common outcome measured [61,98–104,108,109,113–115]. Behavioural challenges in children with FASD who need intervention to support participation are well documented [2,5,13,39,56,131]. It is unsurprising that this review identified more than half of the intervention studies measuring behavioural outcomes.

RQ3: Quality of evidence for intervention studies of school-aged children and adolescents with FASD

All the studies identified in this review had methodological limitations. The RCTs were rated as "some risk" to "high risk" of bias and the ROBINS-I were rated as "moderate to serious risk" of bias. Of the three SCEDs, two were rated as "high" risk of bias and only one was rated as "low." The external validity scores within the three SCEDs were higher than the internal validity scores and there was variability in methodological rigour and reporting across studies. All systematic reviews were assessed as "critically low" (RQ3) [8,9,50–52] using the AMSTAR 2 [64] mostly by not reporting protocol registration, justifying language used in search and reason for excluded studies in review. Strict word limits in journals may explain some of these omissions. However, reporting criteria for systematic reviews have evolved since some of these reviews were published [50–52] with the update of the PRISMA 2020 Statement [63] and the AMSTAR 2 [64].

Most studies identified had low participant numbers and measured outcomes immediately post-intervention rather than long term. Several studies did not report how randomisation occurred or blinding and numbers of participants that completed intervention in each group. Although comparison data between participant groups on IQ, age, gender, and guardianship status were often reported, the authors often failed to report other interventions (e.g. medical, therapy or educational) received during the studies or other potential confounding factors. The evidence of intervention effects for school-aged children and adolescents with FASD will remain questionable until low risk of bias is achieved.

As found in other systematic reviews of intervention studies for children with FASD [8,9,50–52] this review identified a lack of high-quality intervention evidence. We acknowledge the challenges in recruiting children and adolescents with FASD into studies as finding those with similar neurodevelopmental profiles is difficult due to their varied presentation and diagnostic criteria.

RQ4: Intervention effectiveness for school-aged children and adolescents with FASD

Child-focused interventions addressing academic performance, language skills, task completion and social skills were found to be effective activity outcomes for children and adolescents with FASD at home and school (RQ4). Likewise, context-focused interventions aimed to support caregiver/teacher awareness and capacity to support children and adolescent activity outcomes were also effective at home and school [12,16].

Of the 14 studies that addressed activity performance, 10 reported significant changes in outcomes (see Table 1). Improved outcomes were evidence across language skills [97], daily living skills [99], focused attention to game play [100], task completion [101,102], reading fluency [105], ability to spontaneously use metacognitive skills in computer game play [106], social skills [109, 110, 114] and problem-solving [111]. Many studies relied on caregiver or teacher reports of behavioural change rather than directly measuring performance in context. As well, studies failed to randomly assign participants into intervention and control groups, elevating the risk of bias.

By pooling child outcomes from included RCTs and NRSIs, we identified small positive intervention effects to improve activity ($g = 0.36$, 95% CI 0.17–0.54) and behavioural ($g = 0.21$, 95% CI 0.10–0.33) outcomes for school-aged children with FASD (RQ4). These studies used language [97] and social skills group-based intervention [109,110,114] with children and adolescents and caregivers/teacher capacity-building strategies [98,109,110] to support child application of skills in context. Caution needs to be taken when interpreting these results as the certainty of this finding was rated as low using the GRADE approach [93]. Previous reviews have not pooled outcomes from both RCTs and NRSIs into a meta-analysis. Two other systematic reviews have synthesised evidence on interventions qualitatively and noted positive effects for activity (learning and social skills), behavioural and BFS (executive function and attention) outcomes [8,50]. Our review is the first to combine data from both studies designs into a meta-analysis.

The reported effects of studies addressing executive functioning, social cognition and motor skills were mixed. Collectively, the pooled effect of intervention across all these measures was large and non-significant. Sub-analysis showed a small positive effect for social cognition ($g = 0.30$, 95% CI 0.01–0.59), reflecting knowledge of social rules. The intervention that targeted social cognition mainly used group-based learning, with caregiver training to support skill transfer. The certainty of this finding was rated very low using GRADE [93] indicating little confidence in the estimated effects [132]. Motor outcomes, using standardised balance measures, showed a significant, positive effect for virtual-reality balance training ($g = 0.89$, 95% CI 0.26, 1.53) [58]. This study compared balance training completed at home and in the clinic setting to no intervention. Skill transfer to participation and movement in daily living was not evaluated, however. BFS outcomes were mostly assessed using robust, direct measures of child performance and often used in conjunction with caregiver reports to gauge transfer to daily living. Analysis of executive functioning measures of attention, inhibition, working memory and verbal fluency outcomes were non-significant. Recently, Betts and colleagues [57] undertook a small meta-analysis of RCTs and quasi-experimental designs across three studies that investigated the effect of intervention on executive function outcomes (visual attention and cognitive flexibility) using direct child performance measures. They found positive (but non-significant) intervention effects for all three different types of executive functioning measures.

Problem behaviour in children with FASD is frequently reported in the literature [39] and was also a focus of evaluation in those studies we reviewed, rated mainly by caregivers. A wide variety of tools were used to measure adaptive functioning, emotional control and regulation, externalising, and internalising behaviours. Our meta-analysis showed a small, pooled treatment effect overall for behavioural outcomes ($g = 0.21$ 95% CI 0.10, 0.33), and more specifically, significant positive effects for emotion control and regulation and externalising outcomes. Like BFS outcomes, the certainty of these outcomes was rated very low using GRADE [93]. Risk of bias were higher for caregiver ratings as they were not blind to the intervention status of their child, perhaps promoting a halo effect on outcomes. Direct observation of child behaviour and/or teacher ratings were used in two studies [98, 109] but did not corroborate the reports of caregivers, suggesting a possible response bias.

Recent studies of neurodevelopmental disorders (that have included FASD participants), have used various direct and indirect measures of BFS, behaviour, activity, and participation to determine intervention effectiveness. For example, in two studies using a computerised game (i.e. Caribbean Quest) with educational assistants (EAs) to teach metacognitive strategies have shown some positive effects on attention, executive function, metacognition, self-monitoring behaviour, daily activities in the home, and classroom learning and behaviour [133,134]. Intervention effects were evaluated in a number of ways: participant performance on academic tasks and metacognitive strategy use (activity outcomes); attention and working memory (BFS outcomes); interviews with EAs about how children transferred the skills into classroom learning behaviours (i.e. behaviour and activity outcomes), and parent and educator questionnaires about their perceptions of their child's metacognition (activity outcomes) [133,134]. These studies show promising benefits on all of the aforementioned measures. A limitation of these studies is that authors did not directly measure participation from the participant's perspective or use participation outcome tools.

Strengths and limitations of this review

This is the first systematic review and meta-analysis examining the effectiveness of interventions on outcomes at ICF-CY [16] levels, the fPRC [18] and behaviour for school-aged children and adolescents (5-18 years) with FASD. Previous systematic reviews identified few RCTs and a small meta-analysis was performed on measures of executive function which indicated no significant effects [8,50–52,57]. We used a multi-level meta-analysis technique [85–89], which allowed the pooling of all available outcome data from both RCTs and NRSIs to evaluate effects of interventions supporting activity and participation for school-aged children and adolescents (5–18 years) with FASD.

While every effort was made to limit potential sources of error, our review has some limitations that need to be considered when interpreting its findings. Firstly, although we pre-registered our review with PROSPERO, minor changes were made to the inclusion criteria and the wording of the review title and questions. For example, we included the RoBiNT to assess risk of bias in SCEDs after we identified SCED reports from our search of the databases. Our age range criteria excluded several studies including preschool-aged children and limit the summation of interventions for all school-aged children and adolescents (5-18 years) with FASD identified. Our meta-analysis was limited to 10 studies, with small sample sizes and rated with moderate to high levels of risk of bias. Power analysis for the meta-analysis was completed *post hoc* based on the finding from the overall model which indicated that the study is likely to be underpowered. We accept that the meta-analysis does have an element of unresolved error in combining effect size estimates and including studies with moderate to high levels of risk of bias. The GRADE approach [93] was used to mitigate risk of bias in reporting results from our meta-analysis by evaluating the certainty of the evidence. We were unable to examine the long-term impact of interventions as only five studies included any follow up data and follow-up time periods varied from one to 12 months [58,68,101,104,108,109].

Recommendations for future research and clinical practice

Limited high-quality evidence currently exists regarding the effectiveness of interventions for school-aged children and adolescents with FASD compared with no intervention or standard care. No study reviewed measured participation. Measuring the impact of interventions on participation is essential to optimise health and development outcomes [9,18] as children and adolescents with FASD are known to experience participation challenges. Clinicians and researchers can optimise children's and adolescents' rehabilitation outcomes by asking children, adolescents, and families about their participation goals, how they experience intervention, and by providing support to address these [127].

Picture and survey-based goal setting and participation tools have been used successfully to support goal setting of children with other disabilities. The former includes the Perceived Efficacy Goal Setting System second edition (PEGS-2) [135], Children's Assessment of Participation and Enjoyment & Preferences for Activities of Children (CAPE/PAC) [136] and Picture My Participation [137]. Recently photo-elicitation interview methods have explored the lived experiences of adolescents with FASD to understand how their challenges influence daily participation [38]. Participation needs and challenges identified by caregivers measured using the Participation and Environment Measure for Children and Youth (PEM-CY) [138] and the Child and Adolescent Scale of Participation (CASP) [139] have also been used with caregivers of children with other disabilities. Researchers and clinicians could trial these tools with caregivers/teachers of children with FASD to identify participation needs and intervention outcomes.

As recommended by others [17,21], to support participation outcomes we also suggest that clinicians and researchers shift intervention context from the clinic to community and other settings, such as school and home, where children and adolescents with FASD will benefit from using the intervention in daily living. In addition, evaluation of activity and participation outcomes are best measured in the context in which they are delivered. For example, future studies could investigate the impact of social skills taught, practiced, and coached directly in the school environment which may then support children's participation at school in classroom learning activities and playtime outside of the classroom [140–142].

There continues to be a paucity in motor interventions for children and adolescents with FASD [59,60] despite the increasing awareness of motor impairments in this group [2,22–32]. As advocated by Lucas et al. [59], we recommend more high-quality motor intervention studies that measure activity and participation outcomes for children and adolescents with FASD and motor impairments.

Given difficulties sourcing representative samples in RCTs and large sample sizes, Anderson and Carr [143] have proposed using high quality SCEDs which can be included in meta-analysis. Marked variation in individual abilities can make randomisation difficult when using RCTs, diluting the ability to detect real treatment effects. High quality intervention SCEDs also enables individualised interventions to be described and meta-analysis of pooled treatment effects. Although three SCEDs [101,102,111] were identified in this review, examining effectiveness of self-monitoring behavioural and virtual reality interventions, their methodical quality reporting varied; two of the three were rated as "high" risk of bias due to poor sampling. Addressing internal and external validity constructs through improved study design and reporting is required in future SCED studies.

Reporting practices of studies involving children and adolescents with FASD can be improved to provide more confidence in the value of interventions and reduce potential confounding bias (especially via provision of all details on participant abilities). These abilities include aspects of IQ, adaptive behaviour, social skills, language, executive functioning, academic skills, and motor skills. Specifying the type of FASD and diagnostic criteria will also enhance comparisons between studies. Describing all other interventions that participants receive during studies (including medication, education, and other support) will also reduce potential sources of confounding. Including both objective and subjective outcome measures will reduce bias and increase validity of measures and impact on everyday living.

Conclusions

This systematic review and meta-analysis demonstrated a small, positive pooled effect for interventions for children and adolescents (5-18 years) with FASD that were designed to support "BFS," "activity," and" behaviour outcomes;" however, the certainty of this estimated effect was rated low using the GRADE approach [93]. Sub-group analysis showed positive, small intervention effects on "activity" and "behaviour" outcomes. No significant effect was found for pooled BFS outcomes, however, the one motor intervention study that targeted balance showed a significant, positive effect on balance metrics, while the effect on social cognition outcomes was very small, but positive. Combining child/adolescent interventions with caregiver and teacher training appears to support activity, social cognition, and behaviour outcomes effectively. As such, interventions provided in home and school environments show promising effects. Notably, no reports directly measured children's or adolescents' participation, preferences, or goals, and few measured their involvement and self-perception. Interventions to support community participation were not identified which highlights an area of dire need for future research. In summary, we recommend that interventions address the child/adolescent's own participation goals, be embedded in everyday environments, and incorporate caregiver/teacher training to optimise participation and health outcomes for children and adolescents with FASD.

Disclosure statement

The authors have no conflicts of interest to declare.

Data availability statement

Data extracted from included studies and used for analyses can be obtained from the corresponding author.

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