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Socio-Ecological Factors of Physical Activity in Children and Adolescents With Down Syndrome: A Mixed-Methods Systematic Review

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ABSTRACT

Background: Children and adolescents with Down syndrome (DS) are at risk of obesity due to a reduced level of physical activity (PA). PA in children and adolescents with DS is a multifaceted phenomenon that includes personal, relationship, community and other social factors. Hence, a comprehensive understanding of how each component interacts with the others is required. Based on the socio-ecological model, this study aimed to identify the socio-ecological factors associated with PA in children and adolescents with DS through a mixed-methods systematic review.

Methods: A systematic search was performed using PubMed, CINAHL, PsycINFO and Web of Science databases in February 2024. Inclusion criteria were studies investigating factors related to PA, involving at least one of the target populations (children and adolescents with DS or their parents), published in peer-reviewed journals and written in English. Studies assessing the effectiveness of PA, systematic reviews and unpublished studies were excluded. The methodology was appraised using the Mixed Methods Appraisal Tool. Data were extracted and synthesised in a sequential explanatory design using the socio-ecological model.

Results: Databases were searched for 1022 articles; 17 studies (10 quantitative and 7 qualitative) were included and synthesised in this review. Based on the socio-ecological model, the following factors were identified at four levels: (1) physical and cognitive ability to participate in PA at the personal level, (2) competing family responsibilities and overprotective parenting at the family level, (3) peer support and community programmes at the community level and (4) absence of state support for PA programmes at the policy level.

Conclusions: To support the PA of children and adolescents with DS, factors affecting PA at the personal, family, community and policy levels must be considered. Community stakeholders and policymakers can use the findings of this review to develop and improve programmes for children and adolescents with DS.

Seung Hyeon Yang and Gayeong Kim contributed equally to this work.

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1 | Introduction

Down syndrome (DS), a condition in which a person is born with an extra chromosome 21, has a worldwide prevalence of 1/800 and is typically associated with physiological manifestations such as hypotonia, delayed gross motor development and cardiac abnormalities (Bull 2020; Malak et al. 2015). These DS-specific characteristics may limit an individual's physical activity (PA) range and increase their risk of obesity (Martínez-Espinosa et al. 2020). Nearly 25% of children and 50% of adults with DS experience obesity, which complicates many DS conditions, including obstructive sleep apnoea, diabetes and cardiopulmonary conditions (Bull 2020). Robust evidence is available that obesity and its complications can be prevented by increasing the moderate- to vigorous-intensity physical activity (MVPA) levels (Farooq et al. 2021; Sénéchal et al. 2021; Wyszyńska et al. 2020). The World Health Organization (WHO) recommends that children and adolescents aged 5–17 need 60 min or more of MVPA each day (Chaput et al. 2020). However, a systematic review showed that children with DS participate in less PA than their peers who are developing typically and do not meet guidelines for PA levels across age groups (Fox et al. 2019). To prevent obesity and promote PA levels in children with DS, it is necessary to understand the DS-specific facilitators and barriers to PA.

PA among children and adolescents with DS is a multifaceted phenomenon influenced by DS-specific individual factors, as well as environmental factors such as family and community (Souto et al. 2024). Previous studies primarily focused on the factors at the individual level such as children's motivation to participate in PA, self-efficacy or physical health status. However, a growing body of research applies social determinants of health perspectives and demonstrates that engaging in PA is not only a matter of an individual's responsibility but also influenced by diverse social factors from multiple levels (Albujulaya and Stevinson 2023; Rhodes et al. 2024). Hence, this review employed the social-ecological model (SEM) as the guiding framework. As SEM demonstrates the interplay of each level including intrapersonal, interpersonal, community and policy levels (Kilanowski 2017), it is a useful tool to comprehensively understand how children with DS relate to their environment and what factors influence their daily lives. Unlike single-level models, the SEM allows for examination of broader social, environmental and policy factors (Schölmerich and Kawachi 2016). This approach is particularly important for populations with complex needs such as DS, where multiple systems simultaneously affect their daily activities. Furthermore, prior research has successfully applied the SEM in analogous populations, such as children with autism spectrum disorder (Kim and Kwon 2022), supporting the relevance and applicability of the SEM framework for this review.

From the SEM perspective, previous qualitative studies have explored the experiences of children with DS related to their PA (Alesi 2017; Alesi and Pepi 2017; Alghamdi et al. 2021; Alwhaibi and Aldugahishem 2019; Barr and Shields 2011; Downs et al. 2013; Sayers Menear 2007), identifying themes that align with various SEM levels. In addition, quantitative studies have examined barriers and/or facilitators to PA in this population (Alwhaibi et al. 2022; Esposito et al. 2012; Izquierdo-Gomez et al. 2021; Izquierdo-Gomez and Marques 2017; Jain et al. 2022; Makhov and Medvedev 2019, 2020; Shields et al. 2017), with many of these factors also classified according to the SEM framework. However, there remains a notable lack of systematic reviews that synthesise both quantitative and qualitative evidence to fully capture the complexity of these multilevel factors influencing PA in this population. Therefore, this study applies the SEM framework and adopts a mixed-methods systematic review approach to comprehensively identify the factors influencing PA in children and adolescents with DS. It addresses the following research question: What are the socio-ecological factors related to PA in children and adolescents with DS at the personal, family, community and policy levels?

2 | Methods

This review particularly used a sequential exploratory mixed-methods design wherein a qualitative phase is followed by a quantitative phase (Pluye and Hong 2014). Results of qualitative studies were first analysed to develop a conceptual framework of factors related to PA among children and adolescents with DS using the SEM, which were then synthesised with the quantitative results for each identified factor.

This review was conducted and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines for quantitative studies and the Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) guidance for qualitative studies (Moher et al. 2009; Tong et al. 2012) (see Appendices S1 and S2). The review protocol was registered with PROSPERO (Registration ID: CRD42020202321).

2.1 | Search Methods

A search strategy was developed with the assistance of a librarian using keywords in three groups: (1) DS; (2) children and adolescents (an age group); and (3) PA, defined as any bodily movement by skeletal muscles that requires energy expenditure (WHO 2021). The search was not restricted by publication year but was limited to literature in English. The databases used for the study included PubMed, CINAHL, PsycINFO and Web of Science. The specific search strategies for each database are presented in Table S1.

Inclusion criteria were studies that (1) investigated the factors related to PA in children and adolescents with DS (aged 2–21 years); (2) sampled at least one of the populations of children and adolescents with DS and their parents; (3) were published in peer-reviewed journals; and (4) were written in English. The age groups under consideration were decided based on the American Academy of Pediatrics (2017) definitions of 'children' (2–12 years) and 'adolescents' (12–21 years). This review included studies that sampled children and adolescents with DS and their parents. This decision was based on the rationale that DS is associated with intellectual disability. Individuals with intellectual disabilities face challenges in communication, which can make it difficult to gain

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a comprehensive understanding of their experiences (García et al. 2020; Smith et al. 2020). Therefore, family members were included in the study to adequately reflect the experiences of those with DS. Exclusion criteria were studies assessing the effectiveness of PA, systematic reviews, opinion papers or editorials.

2.2 | Search Outcome

The PRISMA framework was used as a guide to screen the searched articles (Figure 1). Articles identified from the search strategy were imported to Covidence, a web-based screening tool. Covidence automatically identified and removed duplicates. After removing duplicates, the initial search yielded 1022 articles. Two authors (G.K. and K.K.) independently screened the titles and abstracts that included keywords related to PA (e.g., 'physical activity', 'sports' and 'athletic') and DS, with a low threshold for consideration for further review. As a second round of screening, the full text of the 60 eligible studies was reviewed to determine eligibility for this review. In the second round, 43 out of 60 studies did not meet the inclusion criteria for one or more of the following reasons: (1) not determining factors affecting PA, (2) not including children and/or adolescents, (3) not original/published articles and (4) being systematic reviews. We included qualitative studies that sampled parents if the age range of their children was within 2–31 years, as parents' responses reflected their past experiences related to children's PA. Throughout the process, any disagreements between reviewers were resolved by consensus or by the decision of a third independent reviewer (E.K.C.). A total of 17 studies were included in the final review: 7 qualitative and 10 quantitative studies. All screening processes were performed in Covidence.

2.3 | Quality Appraisal

Quality appraisal of the studies was undertaken using the Mixed Methods Appraisal Tool (MMAT) (Hong et al. 2018). We selected a single tool capable of ensuring consistent evaluation of study quality across various designs, including qualitative, quantitative non-randomised and quantitative descriptive, as these types of studies were included in our review (Hong et al. 2018). The MMAT includes two screening questions for all study types to identify whether further methodological quality assessment is feasible: (1) Are there clear research questions? (2) Do the collected data allow us to address the research questions? These questions were asked about each included study; no studies were excluded from the review. Five core quality criteria were considered for each type of study design, including appropriateness of study design and sampling strategy, adherence to data collection methods, integration integrity and integration of findings. Each specific criterion was assessed using three response options: 'yes', 'no' and 'cannot tell'. The total score for each study, calculated as the total number of 'yes' responses, ranged from 1 to 5, with a higher score indicating better quality. Two authors (G.K. and K.K.) independently appraised the studies, and consensus was reached through discussion. Any disagreement between the

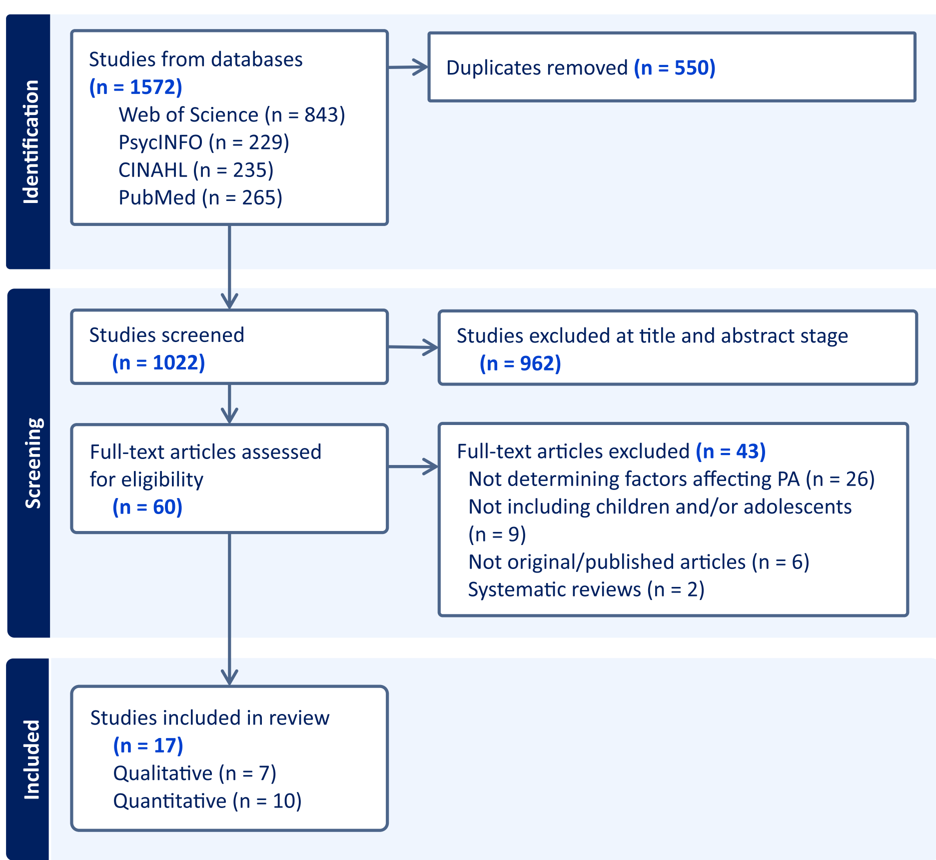

FIGURE 1 | Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram.

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two reviewers was discussed with a third reviewer (E.K.C.). The specific quality criteria for each category and the appraisal results are shown in Appendices S3–S5.

2.4 | Data Extraction

Two authors (G.K. and K.K.) extracted data from 17 full-text articles. The authors developed and pre-piloted a data extraction tool on three studies before full data extraction. Both quantitative and qualitative data were extracted and managed using the Microsoft Excel spreadsheets for coding and analysis. The articles were coded based on study aims, methods, population, data collection methods/measurements and reported factors. The findings related to the influencing factors were specifically extracted and coded within the four levels of SEM: personal, family, community and policy. Mutual agreement on coding during data extraction was assessed by randomly selecting three articles. The two authors (G.K. and K.K.) double-checked the extracted data and collectively reviewed it repeatedly. Any conflicts in the extraction process were resolved through discussion with a third member (E.K.C.) of the research team.

2.5 | Data Synthesis

In the first phase, thematic synthesis was used to analyse the data from the included qualitative studies. Two researchers (G.K. and K.K.) conducted free line-by-line open coding of the qualitative findings in primary studies and constructed the descriptive themes based on the SEM. Through an iterative process, researchers inductively identified the factors affecting PA by repeatedly reading and grouping codes at each socio-ecological level. Coding units were summarised in a matrix. The included studies were presented in columns, and the factors affecting PA were presented in rows by personal, family, community and policy levels.

In Phase 2, the results of quantitative studies were pooled. The qualitative findings in Phase 1 were used to guide and interpret the quantitative results in this phase. The factors, including affecting factors of PA and their coefficients with the probability level, were extracted and synthesised by supporting the qualitative data from the first phase or adding new evidence.

In Phase 3, the results from Phases 1 and 2 were integrated and compared to identify a common factor across the studies, and qualitatively indicated and/or quantitatively measured factors were identified. Therefore, this synthesis clarifies the socio-ecological factors related to PA in children and adolescents with DS and reveals knowledge gaps.

3 | Results

3.1 | Study Characteristics

The 17 studies included in this review were published between 2007 and 2024, eight of which were published within the last 5 years (Table 1). The studies were conducted in various countries, including the United States ($n=3$), Australia ($n=3$), Italy ($n=2$), Saudi Arabia ($n=3$), Russia ($n=2$) and India, Spain, Brazil and the United Kingdom ($n=1$ each). Of the 17 studies included, seven were qualitative studies (Alesi 2017; Alesi and Pepi 2017; Alghamdi et al. 2021; Alwhaibi and Aldugahishem 2019; Barr and Shields 2011; Downs et al. 2013; Sayers Menear 2007), one was a quantitative non-randomised control trial (Adamo et al. 2015), one was a quantitative randomised pilot study (Hassan et al. 2021) and eight were quantitative descriptive studies (Alwhaibi et al. 2022; Esposito et al. 2012; Izquierdo-Gomez et al. 2021; Izquierdo-Gomez and Marques 2017; Jain et al. 2022; Makhov and Medvedev 2019, 2020; Shields et al. 2017). All reported factors affecting PA of children and adolescents with DS.

Qualitative studies in this review used different designs, such as a phenomenological approach (Barr and Shields 2011) or a descriptive design (Alesi 2017; Alesi and Pepi 2017; Alghamdi et al. 2021; Downs et al. 2013; Sayers Menear 2007). Qualitative data were collected by in-depth interviews with individuals or focus group interviews. Among the seven qualitative studies, one involved interviews with eight children with DS aged 6–21 years (Downs et al. 2013), whereas the other six involved interviews with parents of individuals, exploring their perceptions of the factors that impact the engagement of their children with DS aged 2–31 years in PA.

Quantitative studies in this review used a descriptive design (Alwhaibi and Aldugahishem 2019; Alwhaibi et al. 2022; Esposito et al. 2012; Izquierdo-Gomez et al. 2021; Izquierdo-Gomez and Marques 2017; Jain et al. 2022; Makhov and Medvedev 2019, 2020), a prospective cohort design (Shields et al. 2017), a non-randomised trial (Adamo et al. 2015) or a randomised pilot study (Hassan et al. 2021). Among the 10 quantitative studies, seven collected data from 3 to 1249 children and adolescents with DS aged 3–20 years (Adamo et al. 2015; Esposito et al. 2012; Hassan et al. 2021; Izquierdo-Gomez et al. 2021; Izquierdo-Gomez and Marques 2017; Jain et al. 2022; Shields et al. 2017), whereas the other three included families or only mothers (Alwhaibi et al. 2022; Makhov and Medvedev 2019, 2020). Two studies used an accelerometer attached to a participant's hip with a belt to measure PA and monitored their movements during waking hours for 7–8 consecutive days (Esposito et al. 2012; Shields et al. 2017). In addition, two studies measured the capability and limitations of physical fitness of individuals using the curl-up test, medicine ball throw, standing long jump, agility square test and shuttle run speed test (Izquierdo-Gomez et al. 2021; Izquierdo-Gomez and Marques 2017). To identify the relationships between PA and the participant's physical conditions, three studies measured anthropometric variables such as weight, body fat percentage, skin fold thickness and waist circumference (Esposito et al. 2012; Jain et al. 2022; Shields et al. 2017). In the non-randomised trial study, an intervention was implemented to assess whether video modelling and reinforcements such as prompts or praise could increase moderate-to-vigorous PA levels (Adamo et al. 2015). In the randomised pilot study, the efficacy of custom-fitted footwear on PA levels of children with DS was examined in intervention and control groups (Hassan et al. 2021).

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TABLE 1 | Summary of included studies ($N = 17$).

No.	First author (Year) Country	Study design	Aims	Participants and settings	Methods/measurements
1	Adamo (2015) USA	Quantitative non-randomised	To identify the use of a packaged intervention including peer video modelling on iPad, prompting and behaviour-specific praise from an adult to increase moderate-to-vigorous PA in preschoolers with DS	3 preschool children with DS (aged 3–5) and 6 children without disabilities From a university-affiliated inclusive early childhood programme	Intervention implemented showing a video of the activity, providing prompts or praises as reinforcements Observational System for Recording Physical Activity in Children-Preschool (OSRAC-P)
2	Alesi (2017a) Italy	Qualitative	To compare the beliefs on facilitators and barriers to participation in PA among parents of children with DS and parents of typically developing children	35 parents into two groups: 19 parents with children with DS and adolescents (aged 7–31) and 16 with typically developing children (aged 5–10) From gyms or not-for-profit associations for people with DS and their families	Semi-structured interview Thematic analysis
3	Alesi (2017b) Italy	Qualitative	To explore parental beliefs concerning involvement, facilitators/barriers and benefits of PA in children with DS	13 parents of children with DS (children aged 7–27) From gyms or not-for-profit associations for people with DS and their families	Semi-structured interview Thematic analysis
4	Alwhaibi (2019) Saudi Arabia	Qualitative	To explore factors affecting participation in PA in children with DS from their mothers' perspectives	36 mothers of children with DS (children aged 6–11) From a DS centre in Saudi Arabia	Face-to-face interview Thematic analysis
5	Alghamdi (2021) Saudi Arabia	Qualitative	To explore maternal perceptions regarding physical activity of children with DS	17 mothers of children with DS (children aged 3–17) From four DS centres in the western region of Saudi Arabia and other regions in Saudi Arabia	Semi-structured interview Thematic analysis
6	Alwhaibi (2022) Saudi Arabia	Quantitative descriptive	To investigate the correlation between the use of technology and the level of physical activity among children with DS	49 mothers of children with DS (children aged 6–12) From three DS centres in Saudi Arabia	Children's Physical Activity Questionnaire (CPAQ)
7	Barr (2011) Australia	Qualitative	To explore the barriers and facilitators to PA for children with DS	20 parents of children with DS (children aged 2–17) In a metropolitan and a regional area	Phenomenological approach In-depth interviews Thematic analysis

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TABLE 1 | (Continued)

No.	First author (Year) Country	Study design	Aims	Participants and settings	Methods/measurements
8	Downs (2013) UK	Qualitative	To explore PA among children with DS	8 children with DS (aged 6–21) From a DS organisation in Liverpool	Semi-structured interview Thematic analysis
9	Esposito (2012) USA	Quantitative descriptive	To examine the PA patterns of children with DS	104 children with DS (aged 8–16) From DS parents' support groups and organisations throughout the state of Michigan	PA: accelerometer (using Mini Mitter/Respironics, Co Inc., Bend, OR, USA) Anthropometric measures: weight, the percentage of body fat, skinfolds
10	Hassan (2019) Australia	Quantitative randomised (pilot study)	To determine the feasibility of conducting a definitive randomised trial to determine the efficacy of custom-fitted footwear to increase physical activity in children with DS	33 children with DS: 17 children with DS in the intervention group (mean age 9.7 ± 3.6) and 16 children with DS in the control group (mean age 9.6 ± 4.0) From a community, member-based disability organisation for people with DS	The intervention group was provided with two pairs of custom-fitted footwear Physical activity level at baseline, 6 and 12 weeks using an ActiGraph wGT3x-BT accelerometer (ActiGraph, Pensacola, FL, USA)
11	Izquierdo-Gomez (2018) Brazil	Quantitative descriptive	To examine the association of potential socio-economic indicators with obesity and physical fitness components in children with DS	1249 children with DS (aged 10–20) From the Association of Parents and Friends of People with Disabilities, APAE	Socio-economic indicators: family income, parental education level Body mass index (BMI) Physical fitness components: curl-up test (trunk strength), medicine ball throw (upper body strength), standing long jump test (lower body strength), agility square test (speed agility), shuttle run speed test (running speed)
12	Izquierdo-Gomez (2020) Spain	Quantitative descriptive	To assess the bidirectional longitudinal associations of several markers of fatness and physical fitness in adolescents with DS	111 adolescents with DS (aged 11–20) From special education schools, associations and foundations for people with intellectual disabilities from the regions of Madrid and Toledo, Spain	Fatness: weight, height, BMI, waist-to-height ratio (%), body fat percentage (%) Physical fitness: muscular strength (handgrip strength test), motor fitness (4 × 10-m shuttle run test), cardiorespiratory fitness (20-m shuttle run test)

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TABLE 1 | (Continued)

No.	First author (Year) Country	Study design	Aims	Participants and settings	Methods/measurements
13	Jain (2022) India	Quantitative cohort	To assess the relationship between trunk muscle strength with reaching ability and balance among children with DS	14 children with DS (mean age 10.71 ± 3.00 years) and 14 typically developing children (mean age 10.71 ± 3.00 years) From the Neuro-sensory Developmental Unit for Physiotherapy, Mangalore, India	Anthropometric measures: weight, height Muscle strength of trunk flexors, extensors, lateral flexors and rotators: using a Baseline Hydraulic Push-Pull handheld dynamometer
14	Makhova (2019) Russia	Quantitative descriptive	To show the impact of athletic activity in children with DS and to determine modern problems of its wide application	300 families having children with DS	6-item questionnaire asking the perceived basic problems of sports' development
15	Makhova (2020) Russia	Quantitative descriptive	To determine the importance degree of various parental motivations in PA participation of their children with DS	137 families having children with DS 22 regions of Russia	23-item questionnaire asking the level of the importance of motivational statements
16	Sayers Menear (2007) USA	Qualitative	To investigate parents' perceptions of the health and PA needs of their children with DS	21 parents of children with DS (children aged 3–22) In a metropolitan area of the southeastern United States	Focus group interviews Thematic analysis
17	Shields (2017) Australia	Quantitative prospective cohort	To investigate the association of foot posture, deformity and footwear fit with PA in children with DS	50 children with DS (aged 5–18) In an outpatient podiatry clinic based at the university campus in Australia	Foot structure: foot posture, foot deformity Footwear fit PA: accelerometer (using RT3 triaxial accelerometer, Stayhealthy, Inc., Monrovia, CA, USA) Anthropometric measures: weight, height, waist circumference

Abbreviations: DS: Down syndrome, PA: physical activity.

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3.2 | Quality Appraisal

The quality of included studies was appraised using the MMAT. All qualitative studies met the five criteria, and most authors specifically described the rationale for sample size that achieves data saturation (Appendix S3). The quality of the quantitative descriptive studies varied, but most met more than four out of five criteria (Appendix S4). However, one quantitative non-randomised study was assessed as low quality owing to its small sample size ($N = 3$) and lack of representativeness of the target population, as well as the absence of reported statistics, including p-values (Adamo et al. 2015) (Appendix S5). In addition, one quantitative randomised study was assessed as good quality by meeting all quality criteria (Appendix S6).

3.3 | Findings From the Qualitative Studies (Table S2)

3.3.1 | Theme 1: Lack of Motivation to Engage in PA Among Individuals With DS

Motivation is a critical factor in encouraging individuals with DS to engage in PA, according to parents of individuals with DS (Alghamdi et al. 2021; Sayers Menear 2007). In Sayers Menear's study (2007), a parent mentioned, 'I can see that as they get older, you would like for them to have the motivation to stay physically fit. And my concern is finding a niche that would fit their abilities, give them self-esteem, and that they would enjoy doing without us having to make them do it.' The parent perceived that the gap in PA between their child and peers without DS widened with age. This perceived gap was considered one of the factors resulting in the lack of motivation for PA among children with DS. The possible reasons for this lack of motivation include the challenging nature of PA (Alesi 2017), cognitive ability to understand PA rules (Barr and Shields 2011), physical capability to participate in PA (Barr and Shields 2011) and increased dependence on electronic devices (Alwhaibi and Aldugahishem 2019). In the Alghamdi et al. study (2021), mothers of children with DS mentioned that their children lose interest and motivation in PA easily and feel bored when participating in PA. According to the mothers in the study, their children spend more time using technology, which reduces the time spent in PA (Alghamdi et al. 2021).

3.3.2 | Theme 2: DS-Specific Condition

DS-specific physical problems may hinder participation in PA. These include limitations in physical abilities and cognitive difficulties in understanding and following the rules of certain sports (Alesi 2017; Alwhaibi and Aldugahishem 2019; Barr and Shields 2011; Downs et al. 2013). For example, one parent in Barr and Shields' study (2011) stated, 'All the kids can ride their bicycles and he can't … he is getting frustrated that he can't keep up with the other kids and some of them are even younger than him.' Physical characteristics of DS, as mentioned in studies, that may affect participation in PA included gross motor impairment, hypotonicity (Alghamdi et al. 2021; Barr and Shields 2011), heart impairments (Alesi and Pepi 2017; Barr and Shields 2011), hearing impairments (Downs et al. 2013) and lack of coordination skills (Barr and Shields 2011). Owing to DS-specific conditions, some parents found that individual sports or activities were more suitable for their children to develop motor skills and understand the activities at their pace.

3.3.3 | Theme 3: Protectiveness of Parents

Some parents described using protective mechanisms over their children with DS in PA if any PA setting seemed too difficult for their children's capabilities. One parent said, 'I don't want him to get disappointed or hurt … that is probably my weakness; I am protecting him too much' (Barr and Shields 2011). If parents perceived their child as vulnerable in any PA setting, they were less likely to encourage engagement. In another study, a parent said, 'I know that I'm one of the barriers to [physical activity for] my daughter, but I cannot help it' (Alwhaibi and Aldugahishem 2019). The parents commonly perceived their overprotectiveness as a barrier for their children to participate in PA.

3.3.4 | Theme 4: Family's Time and Material Resources to Support PA

Participants reported that the availability and willingness of family members to participate in PA activities with their child or adolescent with DS were often influenced by other competing responsibilities in their families or other jobs (Alghamdi et al. 2021; Alwhaibi and Aldugahishem 2019; Barr and Shields 2011; Sayers Menear 2007). For example, a mother said that 'If we are going to do a physical activity, then I am going to have to supervise it, and I can't abandon everyone else and their needs to supervise his physical activity for three hours everyday. It's not just possible. So, I give him a book or put him at the computer or the television where I know he will stay busy and safe while I am doing other things' (Sayers Menear 2007). Participants reported that their child's PA was sometimes not a priority due to competing family responsibilities, time constraints and financial issues within their family. In addition, many parents reported that they were their child's only means of transport to and from PA activities (Downs et al. 2013). Parents also mentioned that their child's ability to use public transport could potentially increase their child's PA levels by improving access to PA participation.

3.3.5 | Theme 5: Siblings as Role Models

Family support was highlighted as a facilitator, particularly the presence of a positive role model (Alesi and Pepi 2017; Alwhaibi and Aldugahishem 2019; Barr and Shields 2011). Commonly, parents mentioned that the siblings served as role models for children with DS, encouraging them to imitate and engage in PA. In Barr and Shields' study (2011), a parent explained, 'He enjoys doing what they [siblings] are doing … if they are on their bikes, he will jump on his bike.' Similar accounts were found in other studies, which reflect that the presence of siblings may create an encouraging environment and generate further interest in PA among children with DS.

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3.3.6 | Theme 6: Lack of Social Acceptance

Parents were concerned with the lack of social acceptance for DS. In the Alghamdi et al. study (2021), mothers were worried about their children being around children without disabilities while participating in PA because they were uncertain regarding social acceptance of their children with DS. A mother mentioned 'a fear of crowding, lack of acceptance from the others, and people … looking at my child like he is an abnormal and a strange child' (Alghamdi et al. 2021). In addition, parents of children with DS reported that some of the other parents prevented their own children from playing with children with DS: 'It is really offensive to see the look on others' faces when my children plays with other kids; it is as if he is carrying a contagious disease' (Alwhaibi and Aldugahishem 2019). The participation rules of PA facilities were identified as another barrier. In Alwhaibi and Aldugahishem's study (2019), some mothers cited that the facilities did not allow their children's participation in PA without the close supervision of family.

3.3.7 | Theme 7: Peer Support

Several studies have highlighted the importance of peer interaction in promoting PA among individuals with DS (Barr and Shields 2011; Sayers Menear 2007). Parents commonly described how peer groups offer children with DS opportunities for social interactions and a purpose of engaging in PA. A parent mentioned, 'He will sit down with kids that are in basketball or footy … he will sit down and talk, and they talk back to him … that is a big thing' (Barr and Shields 2011). Parents identified that social interaction with peers during PA could give children with DS a purpose to participate in PA, as it could become more enjoyable and meaningful. Furthermore, peer support was key to promoting ongoing participation in PA.

3.3.8 | Theme 8: Lack of Structured Community-Based PA Programmes With Qualified Coaches

Parents in several studies reported a lack of structured community-based PA programmes for their children with DS (Alesi 2017; Alghamdi et al. 2021; Barr and Shields 2011; Downs et al. 2013; Sayers Menear 2007). For example, many community-based PA programmes operate only during specific seasons, limiting children from fully participating in PA. In addition, PA programmes in communities had few experts who could provide coaching in PA for children with DS. Children with DS need coaches trained specifically to address their PA needs. Due to the scarcity of experts and coaches for children with DS, parents had to educate instructors: 'I spend a lot of time up skilling teachers and instructors … so they understand her and can get the best out of her' (Barr and Shields 2011). Parents reported that educating instructors and seeking appropriate opportunities for PA were necessary to provide an adequate range of PA for their child.

TABLE 2 | Socio-ecological factors to participate in PA in children with DS.

Socio-ecological levels	Contents

Personal level	1. DS-specific characteristics
	• Physical
	  • Hypertonicity, weak trunk muscle strength
	  • Reduced gross and fine motor skills
	  • Foot deformity and inappropriate footwear
	  • Obesity or fatness
	• Cognitive ability
	  • Ability to understand rules
	  • Good verbal skills
	  • Limited cognitive ability
	2. Motivations for participating in PA
	• Wanting to be with friends and to be part of the sports team
	• Enthusiasm/enjoyment for PA vs. frustration on PA
	3. Older age of children with DS
	• Decreasing PA participation as they grow older
	• Increased dependency on electronic devices

Family level	1. Parental characteristics • Higher income of parents • Higher education level of parent • Protective parenting style 2. Parental support • Parental support with transportation, costs for PA participation • Parental active participation in PA with their children with DS • Family's motivation and awareness of PA participation of their children with DS 3. Sibling support • Sibling as a positive role model for PA participation
Community level	1. Social stigma 2. Structured community-based programmes: acceptability, availability 3. The presence of expert PA instructors and coaches 4. Peer support 5. Availability of PA facilities
Policy level	1. The absence of national support for PA programmes

Abbreviations: DS: Down syndrome, PA: physical activity.

3.4 | Results From the Quantitative Studies

The results of the quantitative studies provided statistical evidence supporting the findings from the qualitative studies within the SEM framework. Most quantitative studies presented personal-level factors, including children's age, reinforcement (prompts and praise), footwear fit and BMI (Adamo et al. 2015; Esposito et al. 2012; Izquierdo-Gomez et al. 2021;

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Izquierdo-Gomez and Marques 2017; Shields et al. 2017). For example, in the Esposito et al. (2012) study, the PA levels of children with DS decreased with their age in light activity ($r = -0.31$, $p < 0.01$) and in moderate-to-vigorous PA ($r = -0.32$, $p < 0.01$). Furthermore, a randomised pilot study showed that custom-fitted footwear statistically significantly increased the duration of the PA time per day in the intervention group compared with the control group at 6 weeks (adjusted mean difference = 20 min, 95% confidence interval [CI] −9 to 50, $p = 0.170$) and at 12 weeks (adjusted mean difference = 8 min, 95% CI −21 to 37, $p = 0.554$) (Hassan et al. 2021). Only two studies reported family-level factors as facilitators, such as higher family income, parental education and motivation (Izquierdo-Gomez and Marques 2017; Makhov and Medvedev 2020). In Izquierdo-Gomez and Marques' study (2017), male participants with either a higher paternal education level or a higher family income showed significantly better performance in the agility square and shuttle run speed tests ($p < 0.05$), after controlling for age and BMI, whereas female participants did not show any significant differences. Additionally, one study identified factors at the community level, which included specially prepared trainers and structured and available information about PA as facilitators (Makhov and Medvedev 2019). Finally, only one study identified policy-level factors, such as national support for sports programmes, as facilitators (Makhov and Medvedev 2019) (see Table S3).

3.5 | Synthesis of the Qualitative and Quantitative Findings

The factors affecting PA of children and adolescents with DS were synthesised based on the SEM framework (see Table 2). The authors synthesised the findings from qualitative studies at the personal, family and community levels. Further, results from quantitative studies offered statistical evidence for each level, including policy, as classified by the authors.

Children with DS possess DS-specific physical characteristics that may limit their ability to exercise, such as hypotonicity, reduced gross/fine motor skills and cardiac abnormalities. Additionally, limited cognitive abilities, which vary by case, may hamper their range of PA. According to quantitative data, as children with DS get older, PA participation decreases, and dependency on electronic devices increases. Some factors may function as either a facilitator or a barrier in certain cases, which include motivation level for PA, appropriateness of footwear fit and PA types.

At the family level, a family's competing responsibilities, such as time constraints, were identified as a barrier. The protectiveness of parents can also be a barrier to the child's engagement in PA. Some factors, such as parental support (financial/emotional support) and siblings' support, were found to be both facilitators and barriers depending on their degree. At the community level, social stigma towards individuals with DS was identified as a barrier. Structured community-based programmes with the expertise of PA instructors and coaches and peer support were identified as facilitators. Finally, at the policy level, national support for PA programmes was identified.

4 | Discussion

In this mixed-methods systematic review, we explored the multilevel factors affecting PA among children and adolescents with DS, using the SEM framework. Key findings from this review were associated with each level of the SEM and included the following: personal-level factors such as DS-specific physical characteristics (e.g., hypotonicity and foot deformity), motivation to engage in PA and older age; family-level factors such as parental or sibling support for PA; community-level factors such as social stigma and community-based PA programmes or facilities; and a policy-level factor such as the absence of national support for PA programmes. This mixed-methods systematic review enabled the analysis of multiple factors affecting PA across various levels among children with DS.

At the personal level of the SEM, DS-specific physical conditions such as congenital heart disease, hypotonia and foot deformity were identified as physical barriers in both qualitative and quantitative studies. When selecting appropriate types and levels of PA, these physical characteristics should be considered. In particular, foot deformities, including pes planus (flatfoot) and hallux valgus (prevalent in 60%–75% and 26% of children with DS, respectively), are DS-specific conditions (Concolino et al. 2006; Pau et al. 2012). These foot deformities can result in foot pain, and their severity increases among those with higher BMI levels (Perotti et al. 2018). Furthermore, an increase in the difference between the length of the foot and that of the footwear should not be overlooked as wearing appropriate footwear can alleviate foot deformities and increase PA (Shields et al. 2017). Conventional footwear does not accommodate the foot shape of individuals with DS; hence, it is necessary to regularly evaluate the fit of shoes and provide shoe education to facilitate PA among children and adolescents with DS and their parents.

Motivation to engage in PA was found to be a facilitator at the personal level, supported by both qualitative and quantitative studies. Motivation is related to the cognitive ability of children and adolescents with DS (Barr and Shields 2011), and diverse approaches are needed to enhance their motivation levels for PA. For instance, video-based PA materials would effectively motivate children to participate in PA and reduce the use of electronics because many children and adolescents with DS are visual learners (Yussof and Paris 2012). Adamo et al. (2015) used video modelling with an iPad to prompt and praise children with DS, increasing moderate-to-vigorous PA. This shows that when developing PA interventions, it is essential to consider distinctive DS characteristics and to encourage children and adolescents with DS to participate in PA by increasing their motivation.

Supports from parents and siblings were identified as significant factors in all qualitative studies included in this review. From this result, researchers are recommended to consider involving family members in PA programmes, such as a family-led or a family-centred PA programme. However, systematic review papers on PA interventions among children with DS reported that the majority of intervention studies targeted school-based settings (Hardee and Fetters 2017; Li et al. 2013). Several studies in this review indicated that parents or siblings' active participation

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in PA encouraged children with DS to be more actively engaged in PA. Thus, future intervention studies may consider not only targeting children with DS but also guiding family members to learn how to incorporate family-based PA into their children's daily lives.

Relationships with peer groups are especially important during adolescence; time spent with peers increases, and family influence decreases compared to earlier developmental stages (Lam et al. 2014). In this review, peer group support was identified as influencing PA in children and adolescents with DS at the community level (Barr and Shields 2011; Sayers Menear 2007). Because children with DS interact relatively better with their peers than those with severe intellectual developmental disabilities, group interventions could effectively stimulate their participation in PA (Mitchell et al. 2015). However, before facilitating group PA interventions commonly found in the qualitative studies of this review, the characteristics of children and adolescents with DS, who possess both positive and negative aspects of peer-related abilities, must be thoroughly understood and carefully implemented. In addition, as children with DS grow older, the discrepancy between their physical and cognitive abilities and those of their peer groups increases, resulting in fewer PAs that can be enjoyed together (Downs et al. 2013). Based on the socio-ecological perspective, to solve these problems, it is necessary to understand the phenomenon by expanding it beyond the personal level to the social level. In other words, for adolescents with DS to continue PA, social support and policy are needed to form peer groups with physical abilities similar to theirs and to encourage positive peer interaction.

Community-based PA programmes and facilities were commonly identified in several studies included in this review. The community-based approach can promote more inclusive interventions by engaging children and adolescents with DS, families and the wider community (Berra et al. 2017). However, the reviewed studies found that current community-based programmes have limitations such as temporary or seasonal operating times (Sayers Menear 2007) and lack DS-specific PA experts and/or instructors (Alesi 2017; Alesi and Pepi 2017). Moreover, programme limitations vary across communities, which may lead to health inequities. To develop a well-designed community-based PA programme for children and adolescents with DS, stakeholders and healthcare providers should identify DS-specific physical and psychosocial needs for PA. Such programmes require specially trained PA instructors who are knowledgeable about the unique PA needs of individuals with DS, which can directly enhance the long-term participation in PA of children and adolescents with DS.

At the policy level, when establishing policies to promote PA for people with disabilities, it is necessary to systematically understand the characteristics of each disability and adjust the approach accordingly. For example, although the levels of cognitive and physical disabilities differ for DS, cerebral palsy and spina bifida, the disabilities are classified as a single group (Florian et al. 2006). As a result, the social infrastructure for conditions involving only physical disabilities, without cognitive impairment, tends to be relatively well established compared to those with cognitive impairment (Mackenzie and Watts 2011). This may be attributed to the fact that it is difficult for people with cognitive disabilities to actively advocate for the establishment of infrastructure and policies to meet their needs. However, given the limited number of studies addressing policy-level factors in this review, these interpretations should be drawn with caution. Further research is needed to better understand and inform policy approaches that adequately address the distinct needs of children and adolescents with DS.

This review has several strengths, including the use of the mixed-methods systematic review approach and the SEM framework. First, the mixed-methods systematic review methodology facilitated a better understanding of the results obtained in qualitative studies from quantitative studies. Furthermore, we identified personal-, family- and community-level factors from qualitative studies, whereas policy-level factors were generally not captured in the literature. Most quantitative studies provided statistically measured evidence that confirmed the qualitative results, although one quantitative study reported a policy-level factor. Thus, reviewing both qualitative and quantitative studies enabled the identification of factors across all levels of the SEM. Second, the SEM framework provided valuable insights into understanding the multiple factors influencing PA and their interactions across the various levels of the SEM.

There were limitations to this review. First, most of the included studies were conducted in Western countries, which may limit the generalisability of the findings to other cultures and regions. Researchers should conduct further studies in other continents, such as Asia, to gain a more comprehensive understanding of PA in children and adolescents with DS, considering cultural differences. Second, we included children and adolescents with DS but did not classify them by developmental stage. Whereas older age was identified as an influencing factor at the personal level, developmental characteristics were not fully reflected. Future research should consider the different developmental stages of children and adolescents with DS. Third, a few low-quality studies were included in the synthesis due to the limited number of studies on this topic. For example, there were no randomised controlled trials in the quantitative research, but descriptive, cohort and non-randomised design studies were included. We utilised the results from low-quality studies to support other studies to provide additional support to other findings. Lastly, some included studies relied on retrospective parental reporting of PA for individuals extending beyond the adolescent age range, which may introduce recall bias. This potential limitation was carefully considered during quality appraisal and data synthesis, and findings from these studies were interpreted with caution to minimise the impact of recall-related inaccuracies on the overall conclusions.

5 | Conclusions

This mixed-methods systematic review focused on identifying the socio-ecological factors of PA participation in children and adolescents with DS. PA is a multifaceted and complex phenomenon. Our review identified factors at different levels, such as personal, family, community and policy levels, to be considered when developing PA intervention programmes for children and adolescents with DS. To prevent obesity and improve health

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status in individuals with DS, it is important to address both individual and socio-ecological factors surrounding them to effectively promote their participation in PA.

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Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

Data are available from the corresponding author upon reasonable request.

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Supporting Information

Additional supporting information can be found online in the Supporting Information section. **Appendix S1:** Preferred Reporting Items for Systematic Reviews and Meta-Analyses checklist (PRISMA). **Appendix S2:** The enhancing transparency in reporting the synthesis of qualitative research guidance (ENTREQ). **Appendix S3:** Mixed Method Appraisal Tool (MMAT) in selected qualitative studies ($N=7$). **Appendix S4:** Mixed Method Appraisal Tool (MMAT) in selected quantitative descriptive studies ($N=8$). **Appendix S5:** Mixed Method Appraisal Tool (MMAT) in selected quantitative non-randomised study ($N=1$). **Appendix S6:** Mixed Method Appraisal Tool (MMAT) in selected quantitative randomised controlled trial ($N=1$). **Table S1:** Literature search strategy. **Table S2:** Summary of Socio-ecological factors from qualitative results. **Table S3:** Summary of Socio-ecological factors from quantitative results.

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