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Comparing the fundamental movement skill proficiency of children with intellectual disabilities and typically developing children: a systematic review and meta-analysis

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Abstract

Background Children around the world, particularly those with intellectual disabilities (ID), are exhibiting poor motor skill proficiency. Compared with typically developing children (TDC), children with intellectual disabilities (CwID) are 65% more likely to exhibit low levels of motor competence. The purpose of this meta-analysis was to compare the motor skill proficiency levels, in terms of fundamental movement skills (FMS) of CwID to TDC. FMS are the building blocks required for lifelong participation in sport and physical activity.

Method The meta-analysis was conducted according to PRISMA statement guidelines. 6 electronic databases were searched and 16, 679 studies were found. A total of 26 studies (total participants n = 3,525) met the inclusion criteria. A multivariate maximum likelihood multivariate random effects model was fitted to the data using the metafor package in R. *Results* The study showed that the standardised mean difference (Hedges' g) in FMS between TDC and CwID is large (g = 1.24; CI 95% [.87, 1.62]). Specifically, significant differences between the two groups emerged in all five outcomes: (I) total locomotor score, (2) total object manipulation score, (3) balance, (4) run skill and (5) throw skill. *Conclusions* Further investigation into effective intervention strategies is required in order to reduce the magnitude of difference in motor skill proficiency between the two groups. In addition to developing, implementing and evaluating these interventions, researchers need to work hand in hand with national governing bodies (NGB) of sport and policy makers to ensure that teachers and coaches are being provided with opportunities to upskill in the area of FMS.

Keywords balance, children with intellectual disabilities, fundamental movement skills, locomotor skills, object manipulation skills

Introduction

The global definition of 'Motor Skill Proficiency' is reflective of a range of terminologies (e.g. motor skill performance, motor ability, motor coordination, fundamental movement skills, motor competence) that

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portray goal-directed human movement (Robinson et al. 2015; Logan et al. 2018; Laukkanen et al. 2020). Motor skill proficiency can be defined as the quality of a person's movement coordination and the level of their performance outcome when performing different movement skills (Robinson et al. 2015; Kavanagh et al. 2019). Fundamental movement skills (FMS) are one aspect of motor skill proficiency that has been commonly investigated in the literature (Logan et al. 2017). FMS are the 'building blocks' required for taking part in exercise and physical activity (Behan et al. 2019). Clark (2005) defined FMS as 'gross motor skills that involve the large force producing muscles of the trunk, arms and legs' (p. 245). They do not develop naturally but are rather learned or practised (Barnett et al. 2016). It is important that children are given opportunities to practise, learn and reinforce these skills (Goodway & Branta 2003; Valentini & Rudisill 2004; Clark 2005). FMS are the gateway to more advanced movement skills that are required for games, sports and physical activity (Logan et al. 2018). Having a strong foundation in FMS is an important contributing factor to physical activity participation and, in turn, to receiving exercise-induced health benefits (Lubans et al. 2010; Holfelder & Schott 2014; Hulteen et al. 2018), in addition to facilitating children's cognitive development (Piek et al. 2008). Piek et al. (2008) found that working memory and information processing speed could be predicted by infants' early gross motor development. Previous research identified a relationship between motor skill ability, working memory and information processing speed in school-going children (Piek 2004a,b).

It is recognised that typically developing children (TDC) have the potential to master the FMS between 6 and 10 years of age (Gallahue & Ozmun 2006; Hardy *et al.* 2010). Unfortunately, increases in sedentary time and lack of physical activity participation directly impact the development of FMS with evidence showing, in many countries around the world, that TDC are failing to perform age appropriate FMS proficiency: Singapore (Mukherjee *et al.* 2017), UK (Lawson *et al.* 2021), Canada (LeGear *et al.* 2012) and Ireland (Behan *et al.* 2019). The role of the specific subcategories of FMS are important to consider to further untangle the relationship between cognitive function and motor skill proficiency.

There are three categories of FMS: (1) object manipulation skills, which involve the manipulation

and projection of objects (e.g. underhand throw, overhand throw, catching, kicking, dribbling); (2) locomotor skills, which require the body to move through space (e.g. running, walking, skipping, jumping, hopping); and (3) balance skills (e.g. single leg balance, twisting, bending) (Barnett et al. 2016; Logan et al. 2018). Over the last decade, there have been numerous studies (Simons et al. 2008; Gkotzia et al. 2017; Maïano et al. 2019b) carried out comparing the FMS proficiency levels of children with intellectual disabilities (ID) with TDC. Compared with TDC, children with ID are 65% more vulnerable to having lower competence in FMS (Gkotzia et al. 2017). The results display that children with ID present with lower motor skill proficiency and reduced ability to perform FMS when compared with TDC of the same age (Simons et al. 2008; Gkotzia et al. 2017; Maïano et al. 2019b).

Children with ID exhibit delayed achievement in motor milestones (Jeoung 2018), with impairments in sensorimotor function, motor development delays, movement control deficiencies, motor sequencing deficits, low concentration levels and poor comprehension (Uyanik et al. 2003; Piek et al. 2012; Jeoung 2018). This affects their locomotor (Hartman et al. 2010; Westendorp et al. 2011a), object manipulation (Simons et al. 2008; Rintala & Loovis 2013) and balance skills (Gallahue et al. 2012), which influences their activities of daily living, sports, physical activity and recreational activities. Poor motor skill proficiency exhibited by children with ID is also accredited to the impairment of their intellectual functioning seen by Jeoung (2018) who found that compared with children with moderate ID (IQ = 38.9), children with mild ID (IQ = 58.7) or borderline ID (IQ = 68.8) scored significantly higher in their total motor composite score. Those results indicate a relationship between the level of ID and motor skill proficiency. In addition, low levels of motor skill proficiency hinders children with ID in their social, psychological and physical development, resulting in negative health outcomes (Frey & Chow 2006; Westendorp et al. 2011b).

Research carried out by Hartman *et al.* (2010), Westendorp *et al.* (2011a) and Zikl *et al.* (2013) compared the FMS of children with borderline and mild ID to TDC of the same age using the Test of Gross Motor Development (TGMD-2). The TGMD-2 is an assessment tool that is considered to be the gold

standard for measuring FMS, ensuring a true representation and accurate reflection of the locomotor and object control components of the FMS. All three studies found that the locomotor and object control skills of children with ID were significantly lower than their typically developing peers, with large effect sizes for children with a mild ID and moderate to large effect sizes for children with a borderline ID (Westendorp et al. 2011a). For both ID groups, the object control skill results were relatively lower than the locomotor skills tested, showing that even with borderline ID, object control performance is affected (Westendorp et al. 2011a). A closer look at the literature shows a shortcoming in the methodology of the above studies. The authors failed to measure the FMS of balance as part of their FMS assessment battery, despite the literature indicating balance is an area of weakness for children with ID (Palisano et al. 2001; Capio & Rotor 2010). They display lower motor skill proficiency compared with TDC, particularly in the area of balance; as a result of the FMS deficits, they experience as a population (Capio et al. 2017).

Maïano et al. (2019b) recently published a systematic review with a broad research question, examining the FMS of children and adolescents with ID. More specifically, these authors focused on mastery, deficits and developmental delays in FMS, as well as the correlates of FMS among this population. Although their review provided some valuable insights, there are still some limitations that need to be addressed. Firstly, only a section of the review, composed of six studies, was dedicated to comparing the motor skill proficiency levels of children with ID to TDC. As a result, the review only shows a snapshot of the breadth of research available in this area. Thus, the findings of all studies comparing motor skill proficiency between these groups have not previously been synthesised and summarised using quantitative methods to give a precise estimate of the magnitude of difference. This limitation presents difficulties for the readers in terms of interpreting results and deriving firm conclusions. The precise estimate of the magnitude of difference increases the accuracy of results and conclusions by increasing participant numbers, provides readers with an objective appraisal of the evidence and guides intervention development.

Secondly, the majority of studies included in the systematic review by Maïano *et al.* (2019b) exclusively focused on specific FMS assessed within the TGMD

(Ulrich 2000) to include locomotor and object manipulation skills. As a result of this, the subcategory of balance was not comprehensively examined. Thus, it is possible that the researchers overlooked studies focusing on all subcategories of FMS and those involving alternative motor and movement batteries. As a result, the performance of children with ID compared with TDC in the FMS dimension of balance was not sufficiently investigated.

Based on the aforementioned limitations, which demonstrate the need for further and more advanced analysis comparing motor skill proficiency between children with ID and TDC on a macroscopic level, the main objective of this meta-analysis is to establish a statistical significance across studies in the field of ID research in order to ensure generalisability of results for researchers and practitioners. Generalisability of results would not have been previously possible due to the inconsistencies in FMS assessment tools used across studies in the field and the smaller participant sample sizes. In addition, we want to quantify the direction, the magnitude and its precision (confidence intervals) of the effect between children with ID and TDC, as well as find out if there are study and participant-related variables that might moderate the size of the standardised mean difference. The findings will provide sports coaches, physical education (PE) teachers and policymakers with more reliable and valid evidence. Such findings will in turn influence the development and modification of current practices and policies in this domain. Similarly, the moderators between FMS proficiency and ID level have not been previously synthesised, which potentially has important implications for the design of FMS interventions for children with ID. Therefore, the aim of this study was to synthesise and meta-analyse evidence from crosssectional, experimental and longitudinal studies comparing the motor skill proficiency level of children with ID and TDC aged 4-12 years.

Method

Literature search

The meta-analysis was conducted according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analyses) statement guidelines (Moher *et al.* 2015). This meta-analysis was pre-registered on Open Science Framework. The

articles used in this meta-analysis were conducted before September 2021. Six electronic databases were searched: MEDLINE, Sport Discus, ProQuest, Scopus, PubMed and Google Scholar. The authors of the study gathered the articles from these databases using different combinations of keywords, such as intellectual disability, developmental disability, intellectual impairment AND fundamental movement skill, motorskill proficiency, movement skills and gross motor skills. The complete Boolean search string is listed in Table S4.

Firstly, 16 679 articles including published, unpublished, Doctoral dissertations and conference presentations found were uploaded in RIS reference format to the software Covidence (Covidence systematic review software). At this point, all duplicate publications were removed. After this, each article found was scanned independently by the title and abstract by two of the study authors. Each article that was carried forward was finally fully reviewed by the same authors.

In addition to the database search, the authors reviewed the reference lists of all studies included in the quantitative analysis as well as every relevant review article found. Lastly, we tried to identify unpublished research and missing data by contacting individual authors of the articles that were included in the qualitative analysis via email or via the website ResearchGate. The authors were contacted a maximum of three times with a 2-week time between the contact efforts.

Inclusion/exclusion criteria

The inclusion criteria for this meta-analysis were as follows: (1) Study design: cross-sectional, longitudinal and experimental studies. (2) Participant characteristics: children with ID including those with borderline ID, mild ID, moderate ID, developmental disability and Down syndrome and those with a dual diagnosis of ID and/or autism spectrum disorder, attention deficit hyperactivity disorder (case) and TDC (comparison). All participants had a mean age between 4 and 12 years. (3) Outcome measurements: motor skill assessment tests used to measure FMS and/or at least one of the following FMS categories of (a) object control skills, ball skills or manipulative skills (e.g. catching, kicking, throwing), (b) locomotor skills (e.g. running, hopping, jumping) and (c) balance or stability skills (e.g. single leg balance). (4) Publications written in English.

The exclusion criteria were as follows: (1) Study design: qualitative and review papers. (2) Participant characteristics: youth aged 13 years plus and adults. Participants with an ID in addition to presenting with additional diagnostic criteria such as cerebral palsy, ADHD, dyspraxia, physical disabilities and visual impairments were excluded on the basis that these additional cognitive, behavioural and/or physical conditions could influence participants motor performance. (3) Outcome measurements: motor skill assessment tests used to measure physical fitness (e.g. endurance, flexibility, strength) or fine motor skills (e.g. manual dexterity, bilateral co-ordination).

Study selection

Together, all the different search strategies identified 23 766 studies, of which the first and the second authors initially fully reviewed 16 679 papers, after 7087 duplicates were removed. Out of these papers, 230 were included for further screening. One hundred and seventy-nine papers were deemed eligible for full-text review. All the conflicts were resolved by discussion. In the case of two studies with incomplete reporting of standard deviations or errors, we calculated the standard deviations that were retrieved. In total, 26 articles were carried to the quantitative analysis. Data extraction was completed in dependently by two of the study authors. The complete flowchart representing the study selection process is provided in Fig. 1.

Effect size calculation

We computed the standardised mean difference with heteroscedastic variances between the TDC and children with ID by subtracting the ID group mean values from the TDC group means and by pooling the standard deviations of the two groups by taking the square root of their mean variance (Bonett 2009). The positive bias in the standardised mean difference was corrected, resulting in Hedges' g effect sizes (Hedges 1981). In the studies that had several measurement points, the statistics of the first time point was used. TDC performed better than children with ID, resulting in a positive effect size. Cohen's (1988) criteria of small (.2), medium (.5), and large (.8) were used to estimate the magnitude of the effects.

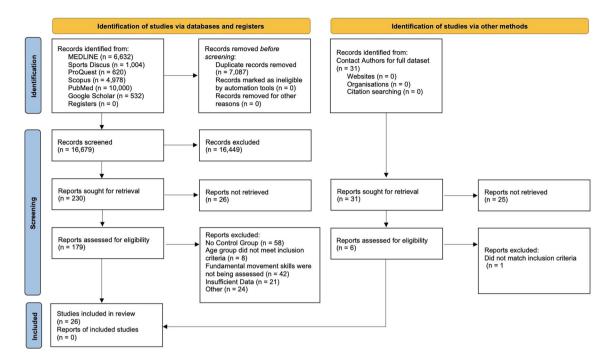


Figure 1. PRISMA flow diagram for the identification, screening, eligibility and inclusion of studies.

The reliability of the extracted statistics was assessed with an unweighted Cohen's kappa.

The initial agreement on the data that was used to compute the effect sizes (means, standard deviations and participants numbers) was high (Cohen's kappa, [2, 1328] = .87, z = 325, P < .001, 95% confidence interval (CI) = [.85, .89]; rough percentage agreement = 86.8%). Before the analyses, a full agreement was achieved by locating and resolving dissimilarities, which were mainly due to typos and differences in interpreting the participant numbers.

Selection and coding of the moderators

A moderator selection based on reason and relevant literature was conducted to explain the expected variation in the effect sizes. A total number of three moderators were categorised as the ID categories (Down syndrome, mild ID, ID and mixed group, meaning those who had an ID and another condition, e.g. ID and autism spectrum disorder), age, IQ and study quality measured by a modified JBI checklist (Moola *et al.* 2020) for correlational studies (see **Supporting Information** for additional tables). The agreement of moderator coding between the raters was initially high (Cohen's kappa, [2, 17 198 = .93], z = 76.6, P < .001, 95% CI = [.89, .96]; rough percentage agreement = 92.9%). The differences in coding were reconciled via a discussion before the analyses.

Statistical analysis

A multivariate maximum likelihood multivariate random effects model (Berkey *et al.* 1998) was fitted to the data using the metafor package (Viechtbauer 2010) in R (version 4.1.0) (R Core Team 2018). The selected modelling approach considers possible non-independence of the effect sizes by including a random effect for each effect size within a study and using a variance–covariance matrix in the model. In this study, this dependence resulted from several comparisons within studies and multiple effects sizes coming from a same study.

The between outcome correlations required for the variance–covariance matrix were derived from the study by Behan *et al.* (2019) with almost 2000 participants.

However, as the precise amount of dependence of the effects was unknown, a robust variance estimator from the club Sandwich package was applied to improve the accuracy of the estimates (Pustojevsky & Tipton 2022). We modelled the standardised mean

differences together for balance, locomotion and object skills and for run and throw skills as the run and throw scores were part of the locomotion and object total scores in some studies.

The standard parameters tau^2 and I^2 were computed to examine the between-study heterogeneity of the true standardised mean differences (Higgins *et al.* 2003; Jackson *et al.* 2012). Additionally, a likelihood ratio test examining the effect of tau^2 on all the outcomes was used as an indicator of significant between study heterogeneity.

A between study heterogeneity of the mean estimates was specified if the likelihood ratio test ($\chi 2$) reached a significance level of P < .05, and the sampling error contributed to the observed total variance less than 75% (Hedges & Olkins 1985; Lipsey & Wilson 2001). The precision of effect sizes was indicated by 95% CIs.

The selected moderators were used in a linear regression analysis as univariate categorical or continuous independent variables in order to explain the heterogeneity of the mean differences. To detect publication bias, we used a modification of the Egger's test (Egger *et al.* 1997) using the standard error of the observed outcomes as a predictor in a multivariate model. Furthermore, we visually examined the normal and contour enhanced funnel plots to detect asymmetry and potential publication bias. Lastly, the influential studies and effects were located using Cook's distances and the existence of outlier studies and effects were determined by analysing the distribution of studentised residuals (Viechtbauer & Cheung 2010).

Results

Study characteristics

In total, 91 effects from 26 studies were analysed for six outcomes: total FMS (k = 11), locomotor skills (k = 23), object manipulation skills (k = 22), balance (k = 17), run skill (k = 9) and throw skill (k = 9). The total number of participants in the experimental group (children with ID) was 1232 (sample size ranged from 7 to 156) and 2293 in the control group (TDC) (sample size ranged from 14 to 977). The average percentage of male participants in the ID group was 65%, whereas in the TD group it was 64%, with four studies not reporting gender distribution. In the 25 studies with the detailed ages of the participants, the weighted mean age for TDC was 8.21 years, and for children with ID was 8.15 years. Eighty per cent of the studies were cross-sectional by design. The most frequently used FMS tools to measure locomotor and object manipulation skills were the Test of Gross Motor Development (TGMD) (n = 15) and the MABC (n = 2), whereas for balance it was the BOT (n = 4) or a variation of a single leg stand (n = 4). Thirteen of the studies were conducted in Europe, eleven in the USA and two in Asia. The full details of the independent studies are listed in Table 1.

Standardised mean differences in motor skill proficiency

The observed mean estimates of 17 effects for balance ranged from -.16 to 3.76, with 94% of the effects favouring the TD group (see Fig. 2). Based on the multivariate model, the standardised mean difference between the TD and ID groups for balance was 1.26 (CI 95% [.52, 1.99], t = 3.79, P = .004). The difference was strongly heterogeneous

 $(\chi_2(\mathbf{I}) = 67.24, P < 0.001, tau^2 = 0.99, I^2 = 92.76\%).$

For locomotion skill, the mean outcome estimates of 23 effects ranged from .09 to 4.87 with 100% of the effects favouring the TDC. The standardised mean difference based on the multivariate model was 1.14 (CI 95% [.71, 1.57], t = 5.68, P = <.001). The mean estimate was strongly heterogeneous ($\chi 2(1) = 50.56$, P < 0.001, $tau^2 = 0.48$, $I^2 = 93.50\%$).

For object manipulation, the outcomes of 22 effects ranged from .09 to 4.87 with 100% of the effects having a positive estimate. The standardised mean difference based on the multivariate model was 1.26 (CI 95% [.81, 1.70], t = 6.09, P < .001). The estimated standardised mean difference was strongly heterogeneous

 $(\chi_2(1) = 29.33, P < 0.001, tau^2 = 0.50, I^2 = 92.50\%).$

For the run skill, the estimates of nine effects ranged from .07 to 3.89 with all the effects favouring the TD group. The multivariate model indicated that the standardised mean difference between the groups was .86 (CI 95% [.33, 1.39], t = 3.99, P = .008). The standardised mean difference was strongly heterogeneous ($\chi_2(1) = 50.88$, P < 0.001, $tau^2 = 0.21$, $I^2 = 89.49\%$).

The observed standardised mean differences for throw skill of nine studies ranged from .11 to 3.11 with all outcomes measures being positive. According to the multivariate model, the standardised mean

Study	Country	Design	Gross motor assessment tool	FMS assessed	Sample size ID	% Males ID	Age ID	ID Type	Sample Size TDC	% Males TDC	Age TDC
Alesi et al. (2018)	Italy	C	TGMD	Locomotor and	18	50%	9.32 ± 6.1	di nim	8	50%	9.28 ± 8.1
				object manipulation Locomotor and	18	72%	8.22 ± 2.82	DS			
Bruininks & Bruininks (1977)	NSA	S	BOT	ooject manipulation Balance	55	%69	9.25	Mild ID	55	%69	9.25
Capio	Hong Kong	C	TGMD-2	Locomotor and	20	%09	7.10 ± 2.90	DS	20	80%	7.25 ± 2.5
et al. (2018) Craig et al. (2018)	Italy	S	MABC-2	object manipulation Balance Balance	42 46	76.20% 82.60%	4.8 ± 1.3 4.6 ± 1.1	ID Mixed	43	62.80%	4.6 ± 1.5
Folsom-Meek	NSA	S	BOT	Balance	20	%00 I	8–11	(UI + USA) ID	20	%00 I	8-11
(1787) Golubović	Serbia	S	Flamingo Balance Test	Balance	42	Not	8.6 ± 1.89	di nim	45	Not	8.6 ± 1.89
er al. (2012) Hartman	Netherlands	CS	TGMD2	Locomotor and	67	stated 58%	9.76 ± 1.50	di nim	67	58%	9.76 ± 1.50
et al. (2010) Howe (1959)	NSA	S	Balancing on one foot	object manipulation Balance	43	72%	6-12	₽	43	72%	6-12
Jung et al. (2017)	NSA	S	TGMD2	Locomotor and	32	59.40%	4.25 ± 0.66	Mixed (DD)	28	57.10%	4.25 ± 0.66
le Blanc	NSA	S	Cratty SCGMT	object manipulation Balance	25	Not	12.32 ± 2.07	DS	25	Not	I2.33 ± I.99
Lejčarová (2009)	Czech Republic	S	One Leg Standing Endurance Test	Balance	139	56.30%	10.62 ± 0.56	di Miid	4	85.70%	10.62 ± 0.56
Magistro et al. (2018)	Italy	Ŋ	TGMD3	Locomotor and object manipulation	98	72%	8.28 ± 1.98	Mixed (ID, ASD, ADHD)	977	50%	8.68 ± 1.84
Mehrman (1983)	NSA	CS	Southern California Perceptual-Motor Tests (SCDMT)	Balance	17	Not stated	7	CI PIIM	20	Not stated	7
Pitchford & Webster (2021)	NSA	S	TGMD3	Locomotor and object manipulation	22	55%	6.41 ± 2.01	Q	22	55%	6.41 ± 2.01

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Study	Country	Design	Gross motor assessment tool	FMS assessed	Sample size ID	% Males ID	Age ID	ID Type	sample Size TDC	% Males TDC	Age TDC
Rider	USA	S	Stork Stand	Balance	31	65%	7.63 ± 1.02	₽	31	52%	7.41 ± 1.27
et al. (1983) Rintala &	Finland	C	TGMD2	Locomotor and	20	65%	9.5	₽	20	%09	9.5
Loovis (2013) Schott	Greece	C	TGMD2 and MABC-2	object manipulation Locomotor and	8	61%	9.06 ± 0.96	DS	8	%19	8.99 ± 0.93
et al. (2014) Smith (1989)	NSA	ш	TGMD, Jump and	object manipulation Locomotor and	7	26%	9.4 ± 1.30	₽	8	62%	7.5 ± 0.63
Sretenović	Serbia	S	l hrow BOT2	object manipulation Balance	15	%001	7.8	۵	23	%00 I	7.8
et al. (2019) Staples	NSA	ш	TGMD3	Locomotor and	13	54%	5.09 ± 0.68	DS	35	%09	5.63 ± 0.33
et al. (2021) Wang &	China	ш	BOT	object manipulation Balance	20	55%	4.6 ± 1.0	DS	30	%09	4.3 ± 0.9
Ju (2002) Westendorp	Netherlands	Ľ	TMGD2	Locomotor and	35	Not	9.5 ± 1.2	di di Mim	253	49%	9.5 ± 1.2
et al. (2014) Westendorp	Netherlands	S	TMGD2	object manipulation Locomotor and	104	stated 66%	10.1 ± 1.4	ai di Mim	104	59%	10.1 ± 1.4
et al. (2011a) Westendorp	Netherlands	S	TMGD2	object manipulation Locomotor and	156	%99	9.55 ± 1.45	di nim	255	54%	9.7 ± 1.3
et al. (2011b) Woodard &	NSA	S	TGMD	object manipulation Locomotor and	22	55%	6-8	di Mim	22	55%	68
Surburg (2001) Zikl et al. (2013)	Czech Republic	CS	TGMD	object manipulation Locomotor and object manipulation	57	%09	11.05	di nim	57	61%	10.46

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Table I. (Continued)

Locomotion

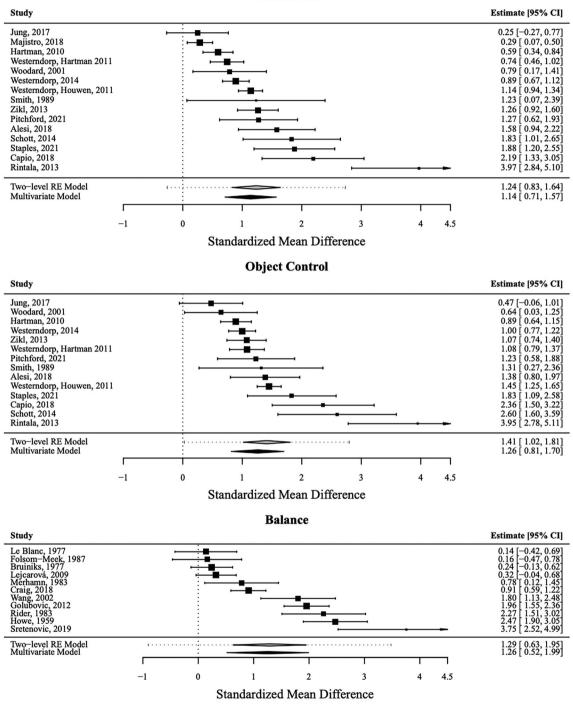


Figure 2. Forest plots with an aggregated two-level RE model and a multivariate model displayed.

difference between the TD and ID children was .89 (CI 95% [.02, 1.75], t = 2.51, P = .046). The estimate of the standardised mean difference was 15 heterogeneous ($\chi 2(1) = 24.92$, P = 0.0195, $tau^2 = 0.68$, $I^2 = 96.28\%$).

Moderator analysis

Study

Due to high significant likelihood ratio tests and high I^2 values for all the outcomes, we pursued to explain

the between-study variability using two categorical and four continuous moderators in multivariate meta-regression analyses. Based on the analyses, the standardised mean differences in locomotion skill were larger between children with Down syndrome and TDC compared to the difference between TDC and children categorised as mixed (t = -4.298, P = .35). Furthermore, for run skill, the difference between children with Down syndrome and TDC

Estimate [95% CI]



Run

Majistro, 2018		-	-			0.07 [-0.14, 0.28]
Zikl, 2013			⊢ ∎i			0.52 [0.21, 0.82]
Schott, 2014						1.13 [0.42, 1.84]
Pitchford, 2021			·			1.17 [0.53, 1.81]
Westerndorp, Houwe	n, 2011		₩	-		1.23 [1.04, 1.43]
Alesi, 2018		1	·=			1.24 [0.63, 1.84]
Rintala, 2013				•		1.40 [0.67, 2.13]
Two-level RE Mode	l	1				0.91 [0.51, 1.31]
Multivariate Model		÷				0.86 [0.33, 1.39]
		1	1	1		
	-0.5	0	1	2	3	

Standardized Mean Difference



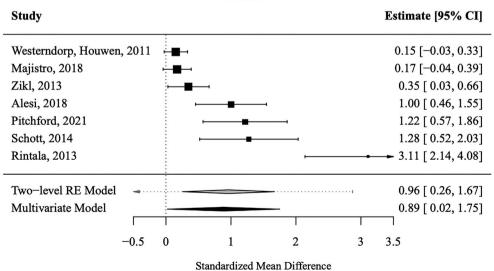


Figure 3. Forest plots with an aggregated two-level RE model and a Multivariate model displayed.

was larger compared with the difference between TDC and children with mild ID (z = -4.44, P < .001), ID (z = 1.98, P = .0475) and children categorised as mix (z = -3.72, P < .001). For throw skill, the difference in skill between children with Down syndrome and TDC was larger compared with the difference between TDC and children with mild ID (z = -6.05, P < .001) and mix (z = -5.65, P < .001). Similarly, the difference between TDC and children with ID in throw skill was larger than the difference between TDC and children with mild ID (z = -5.71, P < .001) and mix (z = 5.47, P < .001)

(see Fig. 3).

Regarding the continuous moderators, the IQ of the children with ID moderated the effect between the TDC and children with ID with higher IQ reducing the difference between the groups ($\beta = -0.017$. t = -4.65, P = .015). Lastly, for the difference in run skill, the age difference in the two groups moderated the effect size with studies having older typically developing than children with ID, yielding larger standardised mean differences ($\beta = 2.142$. t = 4.25, P = .025). The full results of the moderator analyses are displayed in Table S2 for categorical moderators and in Table S3 for the continuous moderators.

Discussion

Summary of evidence

The aim of this meta-analysis was to assess the quality and certainty of the evidence from cross-sectional, experimental and longitudinal studies comparing the motor skill proficiency level, in terms of FMS, of children with ID and TDC aged 4-12 years, whereas the secondary aim was to explore the moderators causing the variability in the outcomes (SMDs). This meta-analysis provides convincing quantitative evidence to support the hypothesis that children with ID have significantly lower motor skill proficiency levels than TDC with overall FMS score showing a very large effect size in the standardised mean difference (g = 1.24; CI 95% [.87, 1.62]) between the two groups. This means that whereas TDC are underperforming in FMS, children with ID are displaying even lower proficiencies than their typically developing peers.

These findings have two meaningful implications. Firstly, they demonstrate the vital importance of developing tailored interventions that meet the needs of children with ID to improve their FMS proficiency, knowing that there is a direct relationship with other constructs such as physical activity, health and overall quality of life. Secondly, the results demonstrate the need for a deeper understanding of FMS proficiency on an individual skill level basis with specific consideration of the skill complexity in relation to children ID levels.

The lack of motor skill proficiency among children with ID (Gkotzia *et al.* 2017; Maïano *et al.* 2019a) further emphasises the importance of developing, implementing and evaluating FMS interventions for this cohort.

In theory, PE classes are an optimal environment for children to learn, practise and reinforce their FMS (Lander et al. 2015). Globally, at the core of the primary school PE curriculum, the aim is to provide children with a baseline proficiency in FMS, in addition to providing knowledge and skills to engage in lifelong physical activity (Irish Primary School Physical Education Curriculum 1999; Australian Curriculum Assessment Reporting Authority 2014; European Education and Culture Executive Agency, Eurydice 2015). However, as demonstrated by the low FMS proficiency levels in this meta-analysis among both CwID and TDC, the impact of primary school PE on FMS proficiency is not sufficient. Research has highlighted some of the barriers faced for delivering quality PE in primary schools, including lack of teacher knowledge and confidence, importance level of PE as determined by the school/teacher, sports provision models that are not child-centred, lack of funding and facilities, lack of specialist PE teachers and lack of support from school management (Decorby et al. 2005; Morgan & Hansen 2008). A commonality among all of this research that is echoed by primary school teachers themselves is the lack of training and education provided to teachers on how to deliver PE, resulting in lack of confidence and knowledge (Decorby et al. 2005; Ma et al. 2021). This will provide children with activities where they would maximise learning opportunities and support children's skill acquisition process. This point is even more salient for those who work with children with ID considering that they may need additional support and assistance to develop their FMS. For example, Ma et al. (2021) found that teachers in primary and secondary schools have limited capacity to advance

children's FMS due to lack of education in their initial training and gaps in professional development opportunities. This is relevant for both children with and without ID as all teachers undergo similar training and professional development. As a result, many children miss out on the opportunity to be taught FMS through targeted, developmentally appropriate motor skill programmes (Goodway & Branta 2003; Valentini & Rudisill 2004; Robinson & Goodway 2009) and leave primary school without having mastered the FMS (Okely *et al.* 2001; Lander *et al.* 2015).

As illustrated in the Results section, one of the key findings of this meta-analysis indicates that an area of weaknesses in motor skill proficiency for children with ID is object manipulation skills, with a large effect size seen in the standardised mean difference (g = 1.21)between children with ID and TDC. The results found in this study support the findings of previous literature by Zikl et al. (2013), Jung et al. (2017), Pitchford & Webster (2021) and ErginSitki & Özbek (2021), suggesting that this subcategory of FMS for this population is weaker in comparison with their locomotor skills. The results of the locomotor skills also indicated a large effect size (g = 1.14)between the two groups; however, as expected and in line with other studies, the standardised mean difference was not as large as with object manipulation skills. Research by Westendorp et al. (2011b) further compliments the results as they compared FMS scores between children with borderline and mild ID, which demonstrated that the children from the borderline group had a higher proficiency in locomotor skills; however, the children's proficiency in object manipulation skills was of a similar level. Therefore, it can be argued that even with lower severity of ID and higher cognitive function, proficiency in object manipulation skills is still significantly impacted.

Majority of studies included in this meta-analysis measured FMS proficiency as a whole or only observed one construct. Studies rarely focus on individual specific skills like throwing, catching, running and skipping. Therefore, a question still remains whether children with ID have poor proficiency in all FMS or only in the skills considered to be more complex.

To investigate this question within our results, further examination of individual differences in object manipulation and locomotor skills revealed that the skills of throw (g = .88) and run (g = 0.85) had large effect sizes in their standardised mean differences. Contrary to this, locomotor skills, described as 'simple skills', are less dependent on cognitive functioning and are more automatised (Bernstein et al. 1996). These results once again demonstrate that object manipulation skills are weaker compared with locomotor skills for children with ID, with their overall FMS proficiency being significantly poorer than their TD peers. This result corresponds with research by Jeoung (2013) who investigated object manipulation skill and performance level in students with varying degrees of ID and found that the skill of throwing (3.21 ± 0.15) showed the weakest proficiency of all of the object manipulation skills. A possible reason for the poor proficiency in object manipulation skills demonstrated by children with ID is because these motor skills are complex and can be described as 'open skills', which rely heavily on environmental factors including external objects and other players (Wall 2004; Westendorp et al. 2011b). Object manipulation skills are mastered during sports and play that demand rapid adaptations to changing environmental situations (Houwen et al. 2007) and involve more cognitive functions and processes (Planinsec 2002; Planinsec & Pisot 2006; ErginSıtkı & Özbek 2021). As previously discussed, children with ID experience deficits in cognitive functioning, thus indicating that due to the higher complexity of object manipulation skills and the extent to which cognitive processes are required to successfully execute and master these skills, this can account for the poor performance observed in this population (Planinsec 2002; Planinsec & Pisot 2006; Westendorp et al. 2011b; ErginSitki & Özbek 2021).

It can be argued that balance is another important construct of FMS, demonstrating low-performance outcomes as the results indicated that the largest effect size difference found in the standardised mean difference between children with ID and TDC was for balance skills (g = 1.26; CI 95% [.52, 1.99]). Gallahue *et al.* (2012) described balance skills as 'the most basic skills within the FMS family'. They are considered as the ability of the body to adapt appropriately to changes in the movement of body parts that alter a person's stability with compensating movements (Gallahue *et al.* 2012; Rudd *et al.* 2015). Few studies comparing the FMS proficiency of

children with ID and TDC include balance, despite it being one of the three pillars of FMS; this is the key reason why we included studies focusing on balance in this meta-analysis. Our findings further strengthen the existing literature in which balance has also been highlighted as an area of FMS weakness for children with ID (Palisano et al. 2001; Capio & Rotor 2010). Sretenović et al. (2019) measured balance using the BOT assessment tool and found that TDC had a total balance score (dynamic and static) of 31.07 compared with children with ID who scored 11.34. Nikolić & Ilić-Stošović (2009) identified that 28.7% of TDC present with a balance disorder compared with 86.8% of Sretenović et al. (2019) sample of children with ID who presented with balance difficulties. Further emphasis needs to be placed on the importance of assessing balance in children with ID alongside locomotor and object manipulation skill scores. From the evidence found, there is a vital need to include balance skills for this population in future FMS interventions.

As discussed in the above methodology and results sections, large variation in the outcomes (or SMDs) was found within and between studies. Potential causes of the variations include relatively small number of studies included, gender distribution and age of participants of studies, methods used to assess FMS and description of ID. Despite these variations in studies, all results favoured the TDC with the overall conclusion being that children with ID have poorer motor skill proficiency levels than TDC, up to as much as 4 standard deviations (Rintala & Loovis 2013; 3.97 [2.84, 5.10]). To further investigate the findings and variations found, a univariate categorical moderator analysis was run on all outcomes to identify if any effects could be explained by specific moderator variables. From previous literature, the level of IQ is a variable of particular interest. We wanted to see if the level of IQ impacted FMS proficiency levels of children. Some individual papers included in this meta-analysis show differences in FMS proficiency depending on the level of IQ of the participants. On the contrary, once the robust variance estimators were applied to all the data available for this meta-analysis, the effects observed on FMS proficiency in individual papers were not observed for all outcomes. It was found that the level of IQ of participants was a statistically significant moderator only for locomotor skill proficiency

(-0.017, t = -4.65, P = 0.015). This result indicates that as IQ level increases in the ID group, the difference in locomotor skill proficiency between the children with ID and TDC decreases. No significant difference for object manipulation or balance skills related to level of ID were found. A potential explanation for this is because not many of the included studies measured IQ and those that did use different IQ measurement tools. However, with this result and trends shown from previous literature, it can be gathered that there is potentially a positive association between level of ID and overall FMS proficiency and that IQ level does impact FMS proficiency of children with ID. These findings suggest a complex relationship exists between IQ and motor development. Further research is required to investigate this complex relationship and its impact on motor skill intervention planning and development.

In a review conducted by Gkotzia et al. (2017), it was concluded that the degree of ID, otherwise known as IQ level, negatively impacted the motor skill proficiency score of children. Four studies included in the meta-analysis have divided children according to their ID level into borderline (BID) and mild ID (MID) (Lejčarová 2009, Hartman et al. 2010, Westendorp et al. 2011b) and autism spectrum disorder (ASD) with ID (Craig et al. 2018). In all of these studies, the ID groups scored significantly lower than the TDC. Participants with MID scored lower on the locomotor skill subtest $(36 \pm 0.79/34.5 \pm 0.61)$ and the object control test $(34.73 \pm 0.72/31.8 \pm 0.56)$ compared with participants with BID who scored $(38.09 \pm 0.61/36.9 \pm 0.41)$ and $(35.53 \pm 0.55/$ 33.2 \pm 0.51), respectively (Hartman *et al.* 2010, Westendorp et al. 2011b) using the TGMD assessment tool. Craig et al. (2018) used the MABC-2 to assess motor skill proficiency and found that children with ID scored significantly higher P < 0.001in their overall test score (2.7 ± 1.4) compared with children with a dual diagnosis of ASD and ID (1.87 ± 1.2) . Differences in balance scores between participants with BID and MID were investigated by Lejčarová (2009) who found a large effect size existed for the balance skill of 'Standing on one leg' for these groups; participants with MID scored significantly lower (7.55 ± 5.77) than those with BID (19.49 \pm 12.94). To summarise, from the results displayed in the literature and those found in the meta-analysis, the level of IQ or degree of ID must be

taken into consideration when designing, developing and implementing FMS interventions for children with ID.

Limitations

A limitation observed among all articles analysed in this meta-analysis is the inconsistent language and descriptions used to describe the participants with ID and their degree of ID. Many terms within the included studies such as learning disability, developmental delay, ID, mild ID, borderline ID, autism spectrum disorder and ID, mental and behavioural disorder, appeared to be used interchangeably across the studies. Although these terms can be used to describe ID without the level of IQ stated and different methods of assessing IQ used between studies, grouping participants according to degree of ID was difficult. The low number of papers and unclear reporting in some cases in the current field of research hindered the analyses as some of the moderators' analyses were underpowered to detect potential statistical differences relating to degree of ID and its impact on FMS proficiency level. This highlights the need for researchers to be as specific as possible when describing the participants included in their studies and their ability level. Future studies could benefit from defining the level of IQ of their participants or at least could be more precise in describing the ID groups. More accurate reporting of IQ is important for studies like this meta-analysis, as it allows for a deeper understanding and further discussions of the impact of IQ level on FMS proficiency.

Implications for research and practice

From the results found in this meta-analysis, it is evident that there is an immediate need for developmentally and structurally appropriate FMS interventions for children with ID in order to improve their overall FMS proficiency and thus quality of life. In addition to developing, implementing and evaluating these interventions, researchers need to work hand in hand with national governing bodies of sport and policymakers to ensure that teachers and coaches are being provided with opportunities to upskill in the area of FMS. Future research needs to assess the knowledge level of teachers and coaches working with children with ID, alongside evaluating the current continuous professional development opportunities available in the area of FMS.

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Conflict of interest

The authors have no conflict of interest to report.

Ethics statement

Dublin City University Research Ethics Committee has approved this research proposal DCUREC/2022/ 181.

Data availability statement

The data that support the findings of this study are openly available on Open Science Framework at https://osf.io/v7pja/ (doi: 10.17605/OSF.IO/V7PJA).

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Supporting Information

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

Table S1. PRISMA checklist

 Table S2. Categorical moderator analyses

Table S3. Continuous moderator analyses

Table S4. Boolean List of Search Terms Used **Figure S1.** Cook's distances and Studentized residuals for effects and studies (clustered). For Cook's distances, values exceeding the median plus six times the interquartile range are considered influential. For studentized residuals (effects), cut-off z-value is set at 3.16. For studentized residuals for studies, three chi-squared critical values are displayed (red line = studies with ten effects, blue line = studies with four effects, green line = studies with two effects and black line = studies with one effect).

Figure S2. Cook's distances and Studentized residuals for effects and clustered for studies. For Cook's distances, values exceeding the median plus six times the interquartile range are considered influential. For studentized residuals (effects), cut-off z-value is set at 2.77. For studentized residuals for studies, two chi-squared critical values are displayed (blue line = studies with four effects, green line = studies with two).

Table S5. Sensitivity analyses based on the influentialand outlier effect/study diagnostic and study quality**Table S6.** Quality Assessment of Studies

Figure S3. Contour enhanced Funnel plots for Locomotion, Object Control, Balance, Run, and Throw Skills. Effects aggregated at the study level (i.e., only one effect per study is plotted)

Figure S4. Standard Funnel plots for Locomotion, Object Control, Balance, Run, and Throw Skills. Effects aggregated at the study level (i.e., only one effect per study is plotted).