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Which outcomes are key to the pre-intervention assessment profile of a child with developmental coordination disorder? A systematic review and meta-analysis

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ABSTRACT

Background: Purpose of this study was to determine what key aspects of function should be incorporated to make up a pre-intervention assessment profile of a child with Developmental Coordination Disorder (DCD); more specifically, what aspects of functioning are implicated in DCD and what is their relative impact?
Methods: A systematic review and meta-analysis were conducted, for which Pubmed, Web of Science, Scopus and Proquest were searched (last update: April 2023, PROSPERO: CRD42023461619). Case-control studies were included to determine point estimates for performances on field-based tests in different domains of functioning. The risk of bias was assessed, and the level of evidence was estimated. Random-effect meta-analyses were performed to calculate the pooled standardized mean differences for domains of functioning and subgrouping was done for clinically relevant subdomains. Heterogeneity was determined with I^2.
Results: 121 papers were included for analyses. Data of 5923 children with DCD were included (59.8% boys) and 23 619 Typically Developing (TD) children (45.8% boys). The mean (SD) age of the DCD group was 10.3y (1.2) and 9.3y (1.3) for the TD children. Moderate evidence was found for motor performance, executive functions, sensory processing and perceptions, cognitive functions and sports and leisure activities to be affected in children with DCD.
Conclusion: Differences between the two groups varied per domain of functioning. This emphasizes the diversity present within children with DCD and provides a rationale for explaining the heterogeneity in this patient group. Yet, results highlight the potential involvement of all these domains and call for clinicians to be alert not only to examine motor skill difficulties but also other aspects of function. Results indicate the need to develop an individualized pre-intervention multi-dimensional assessment profile for each child with DCD. It also supports the important role that clinicians play in an interdisciplinary team to tackle the difficulties encountered by children with DCD.

1. Introduction

Developmental Coordination Disorder (DCD) is a chronic and pervasive neurodevelopmental disorder that is defined by an impaired ability to acquire age-appropriate motor skill levels [1]. With a prevalence of 5–6%, DCD is one of the most common disorders of child development [2]. Individuals with DCD present with awkward, inefficient, and often slow performance of fine- and/or gross motor

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movements, affecting their everyday activities, and participation in leisure activities or organized sports [3]. Although the disorder is primarily a motor disorder, DCD tends to impact different aspects of functioning, including, but not limited to academic achievements, social acceptance, quality of life and the ability to develop a healthy lifestyle [4]. Because of the complexity and the extensiveness of the disorder, many original studies have been published exploring the difficulties in children with DCD. These investigations are usually focused on one aspect of the disorder, depending on the background of the research team, which has caused unclarity about the key features that are vital to be assessed in children with DCD to get a profile of the child covering domains implicated and to plan treatment. Although this disorder can have a major impact on children and their families, DCD is still largely underrecognized by health care professionals, despite the fact that options for diagnosis and intervention are available [5,6].

The primary domain of impact, which forms the basis for diagnosing DCD, is the motor domain. In recent decades, extensive research has revealed deficiencies in various motor areas [4,7,8]. Deficits in both fine- and gross motor tasks, along with difficulties in balance-related activities, are frequently reported in children with DCD. Fine motor problems may include challenges with activities such as handwriting [9], grasping [10], or using utensils [11], while gross motor difficulties can affect activities such as running [12], jumping [13], or catching a ball [14], and balance problems [8]. Balance problems can manifest as difficulties maintaining stability during static activities such as standing or sitting, as well as dynamic activities such as walking, running, or playing sports [8]. It should be noted that even motor planning itself can be altered in children with DCD [15,16]. Motor planning involves the ability to mentally organize and sequence the steps required for a specific motor task. Children with DCD experience difficulties in planning and coordinating the sequence of actions needed for various activities, such as tying shoelaces, manipulating various objects, or performing complex movements.

As a result of their poor motor coordination, children with DCD experience lower perceived motor competence. In addition, peers, parents, and teachers may react negatively to the clumsy motor behavior of children with DCD. Teasing, exclusion, or even bullying by peers occurs regularly in children with DCD [11], which may lower the perception of social acceptance in DCD children compared to typically developed (TD) peers [17,18]. Both lower perceived motor competence and lower social acceptance are associated with internalizing symptoms in children with DCD [19].

Importantly, the inability to acquire the appropriate motor skills also greatly limits the development of a healthy level of physical fitness [20–22]. Moreover, the low levels of physical fitness in children with DCD are still present later in life, indicating that many children and adolescents do not participate regularly in physical activity [23]. The combination of lower physical activity levels and more sedentary behavior may lead to overweight/obesity and related secondary health problems [24]. Early help for children to cope with their limitations has been shown to be effective in improving motor competence and fitness related to motor skills in the short term [25].

In DCD, as well as in other neurodevelopmental disorders, co-occurring disorders are the rule rather than the exception. Specifically, learning disabilities, Attention Deficit Hyperactivity Disorder (ADHD) [26,27], with an overlap of about 50%, and deficits in executive function (inhibitory control, working memory, and executive attention) have been consistently reported [4]. One deficit suggested by multiple studies is an increased prevalence of sensory integration or processing issues [28–31]. Within the realm of sensory integration, visual processing, and visual perception are recognized as pivotal components, playing a vital role not only in the overall sensory-motor experience but also exerting a significant influence on the academic performance of children [32–34].

Both the primary problems of children with DCD and the co-occurring problems may have an impact on Health-Related Quality of Life (HRQoL), which can be defined as the perceived consequences of a disorder on a child's well-being and functioning in daily life [35]. Lower HRQoL compared to TD children is reported in the physical domain, but also in the psychological and social domains [36–38]. These findings again highlight that the impact of DCD extends beyond the motor domain. Research investigating which factors moderate or mediate HRQoL in children with DCD is paramount to improve HRQoL.

It has been established that DCD is a heterogeneous disorder, which concerns the nature and severity of primary motor problems as well as the presence of non-motor-related signs and symptoms and co-occurring disorders. However, previously published systematic reviews and meta-analyses focused on only one aspect of functioning [7–9,35], neglecting the multi-dimensional character of DCD. The focus of this study is to document the full range of key aspects of functioning that may be implicated in the individual child with DCD during their daily activities. These insights are important for clinicians as well as scientists to create awareness of the multi-faceted character of the disorder. In order to help or study these children, we first need to find them, using questionnaires and discriminative test items, i.e. field-based tests, that are easily applicable in the clinical context. Once diagnosed, treatment based on the child's multi-dimensional assessment profile should aim to alleviate the burden of the disorder on the acquisition of skills by the child and improve participation in age-appropriate activities, and quality of life and health. Therefore, the main purpose of this study is to determine which key aspects should be incorporated into the assessment to form a profile of a child with DCD that can serve as a basis for a multi- or interdisciplinary treatment approach. More specifically, which domains are implicated, and how large is the impact of that problem, deficit, or limitation? There is consensus that DCD defines a heterogeneous group of children with marked impairments in motor skills, but no consensus exists on key symptoms and domains to include in the assessment. Thus, gathering the scientific information that is necessary to identify those key aspects that should be part of the assessment requires a data-driven meta-analytic approach. This will the evidence of the choice of easily accessible pre-intervention assessment tools that help standardize domains for the assessment of children with DCD around the world. It will also facilitate the management of children with DCD and, through that, enable prediction and improvement of intervention efficiency.

2. Methods

2.1. Protocol and registration

This review is written in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [39]. The protocol was registered in the PROSPERO database (CRD42023461619).

2.2. Information sources and search strategy

The electronic databases of Medline (Pubmed interface), Web of Science, Scopus, and Proquest were searched systematically (last search: April 11th, 2023) using controlled terminology and free text terms. The search string was developed in Pubmed and transformed to meet the requirements of the other databases [Appendix 1]. No filters or restrictions were applied.

2.3. Selection process

Relevant studies were identified using predefined selection criteria according to the Population Intervention Comparison Outcome Study Design (PICOS) method:

- **Population**: Children with (probable) DCD with(out) comorbidities were included if: 1) A primary diagnosis of DCD was established by a pediatrician or multidisciplinary team, 2) the ICD-10, DSM-IV-TR or DSM-5-TR criteria were fulfilled in the (probable) DCD children.

Children were excluded if they did not have a diagnosis of DCD in one of the following situations: 1) Mixed study groups including DCD (different diagnoses, not all with DCD as the primary diagnosis), 2) Children with another diagnosis such as Cerebral palsy, Autism Spectrum Disorder, Attention Deficit Hyperactivity Disorder, 3) a diagnosis of DCD based on the DCD-Q only.

- **Intervention**: Functional or field-based assessment tools were of interest: performance measures (e.g. fitness test) or questionnaires (e.g. quality of life questionnaire). Laboratory tests and experimental studies were excluded because they were not easily accessible. To determine profiles in children with DCD, sensitive measures to distinguish children with DCD from TD controls are required. Such measures must be related to functional activities of daily life and, therefore, should be related to the assessment profile under consideration.
- **Comparison**: Only typically developing (TD) peers were included.
- **Outcome**: Performances on functional or field-based assessment tools had to be reported separately for children with DCD and controls and expressed with numeric values: mean (SD or 95% CI), and median (IQR). Graphical expression of results; statistics without any raw data were excluded.
- Original studies written in English, Dutch, French, Spanish, Portuguese or German, with a case-control design were considered relevant as well as studies in which norm-referenced (based on healthy normative data) assessment tools were applied. Conference proceedings/-reports, editorials, letters, case studies/-series, abstract only, (systematic) reviews and meta-analyses were excluded.

Relevant studies were identified in two screening phases (title/abstract and full text) using predefined selection criteria. Each study eligibility was assessed by a pair of independent researchers (BS-E & JL; LV & EV; DC & MD). Discrepancies were discussed and in case of remaining disagreement, the opinion of a third reviewer was decisive. Rayyan software was used during this selection process [40].

2.4. Methodological quality

The modified version of the Critical Appraisal Skills Program (CASP) [4] was used by four pairs of independent reviewers (BS-E & JL; LV & EV; DC & MD; MS & MD) to rate methodological issues in case-control studies. The CASP contains 10 items, which are each rated as low risk of bias (1 point), some concerns (0.5 points) and high risk of bias (0 points), providing a total score between 0 and 10. Due to the aim of this study, finding discriminative items between DCD and TD children, five items were specifically important [Appendix 3]. We used the same interpretation rules as for the entire scale, but now converted to a total of 5 instead of 10: $\geq$4/5 indicates high quality/low risk of bias, 2.5–3.5/5 indicates moderate quality, $<$2.5/5 indicates low quality/high risk of bias. Covidence software was used to assess and compare the methodological rating of individual studies within the pairs.

2.5. Data extraction

Data were extracted and cross-checked by independent pairs (BS-E and JL/LV and EV/DC and MD/MM and BS-E). Data were extracted using a standardized extraction spreadsheet in Microsoft Excel, including information about: a) DCD population (source, DSM-IV-TR or 5-TR/ICD-10 criteria, number of participants, sex distribution, mean age (SD) and age range), b) control group (number of participants, sex distribution, mean age (SD) and age range), c) task domain/category (i.e. motor skills, physical activity, sports and leisure, sensory processing and perception, cognitive functions, executive functions, self-perceived competence, behavior, quality of life) expressed by mean/median and SD/IQR or the recovery percentage. To include similar statistical measures, the medians and IQR were converted to means and SDs; the median was considered equal to the mean and the IQR was multiplied by 0.75 [41]. The data extracted from each study was coded and grouped according to the task domain/category.

2.6. Data analysis

Statistical Package for Social Sciences (SPSS) version 28.0 software was used to perform random-effects meta-analyses to estimate pooled standardized mean differences (SMD) between children with DCD and the control group for the outcomes. Random-effects meta-analyses were chosen to incorporate the expected random variation in the effect of the difference in performance across the studies into the pooled differences [42]. Total scores of an instrument were removed from the analyses when item scores were available to reduce dependence among the variables. Outcomes used to identify children with DCD (diagnostic criteria) were removed from the analyses. Outcomes indicating better performances with lower scores were multiplied with -1 to have all differences pointing in the same direction. Duplicate data and outliers were removed from the analyses. The meta-analyses were performed for each task domain/category separately.

Clinical diversity (variability among participants, types of outcome measures), methodological quality, and statistical diversity were considered to assess heterogeneity. Statistical heterogeneity was assessed using a standard Chi^2 test to check the strength of evidence of heterogeneity and the I^2 test to assess the impact of heterogeneity on the meta-analysis. I^2 values of 75%–100% indicate considerable heterogeneity [42]. The studies variance component (Tau^2) was obtained through a random-effects meta-analysis model. When heterogeneity was too large, subgroup analysis was considered, if at least five or more studies were available, accounting for clinical diversity with the types of outcome measures. For each subgroup analysis, expert duos in that specific domain defined the clinical subdomains. The pooled effects SMD, standard error (SE), z-score, p-value and confidence interval were calculated and reported per domain and subdomain.

For each subgroup analysis the most sensitive measures were selected based on the magnitude of the SMD (minimum -2.0, which leads to 1.2 number to treat; SMD of 1 approx. 2 children). Clinical decision-making is facilitated by consideration magnitude of the SMD thus indicating the clinical relevance of an item.

2.7. Level of evidence

The quality of the evidence was evaluated with the Grading of Recommendation, Assessment, Development and Evaluation (GRADE)-method [43]. Per task domain/category, the certainty of evidence that children with DCD and typically developing children were different was estimated by combining the risk of bias, precision, consistency, directness, and publication bias [explained in appendix 3 Risk of Bias]. Publication bias was automatically corrected for as point estimates had to be reported in the individual studies to be included and was therefore not rated separately. Each aspect (risk of bias, precision, consistency and directness) was rated resulting in an overall judgement: 1) High quality: further research is very unlikely to change our confidence in the estimate of effect; 2) Moderate quality: future research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate, 3) Low quality: future research is very likely to have an important impact on our confidence in the estimate of affect and is likely to change the estimate, 4) Very low quality: we are very uncertain about the estimate.

3. Results

3.1. Study selection

The search string revealed 5838 unique records, 846 of which remained for full-text screening. A total of 121 articles were eligible for methodological quality assessment and data extraction [PRISMAflowchart in Appendix 2].

3.2. Methodological quality

The consensus scores for the methodological evaluation at a study level are presented in [Appendix 3]. Most studies were of high quality (68%). Most studies included enough children (81.8%), described how they verified typical development in the control group (79.3%), and performed adequate statistical analyses (82.6%). Only 57.9% of the studies controlled for confounders, and 45.4% of the studies clearly defined the DCD group using all diagnostic criteria.

3.3. Study population

In total 5923 children with DCD were included (3541 boys, 59.8%) and 23 619 TD children (10 815 boys, 45.8%). The overall mean age (SD) of the children with DCD was 10.3 (1.2) years, and of the TD children, 9.3 (1.3) years. The selection of the samples and the DCD criteria are described in [Table 1].

3.4. DCD profile

[Table 2] describes the number of studies, the number of comparisons, the percentage of negative results, the pooled mean SMD, the SE, significance, 95% CI, and the heterogeneity per domain. The forest plots depicting the single study comparisons are available in [Appendix 4-13].

Table 1
Description of the groups with regard to selection and DSM criteria for Developmental Coordination Disorder (DCD).

		% of studies
Selection of the DCD samples	Convenience sampling	64.2
	Clinical referral	27.6
	Parental referral	5.7
	Not specified	2.5
Criterion A – assessment tool	MABC	87.6
	BOT	5.8
	MAND	2.5
	Other	2.5
	Not specified	1.7
Criterion – cut-off value	P15/P16	57.0
	P10	6.6
	P5	28.1
	other	2.5
	Not specified	5.8
Criterion B – assessment tool	MABC checklist	13.2
	DCD-Q	17.4
	Other	19.8
	Not specified	49.6
Criterion B – source	Parents	18.2
	Teacher	5.0
	Child	7.4
	Other	9.9
	Not specified	59.5
Criterion C – assessment tool	Anamnesis	10.7
	DSM-IV or earlier	19.8
	Other	11.6
	Not specified	57.9
Criterion D	Assessment by physician	19.8
	Other	38.8
	Not specified	41.3
Motor performance cut-off for the Typically Developing group	P20	57.0
	Other	17.4
	Not specified	25.6

Abbreviations: MABC: Movement Assessment Battery for Children; BOT: Bruininks-Oseretsky Test of motor proficiency; MAND: McCarron Assessment of Neuromuscular Development; P: percentile rank; DCD-Q: DCD Questionnaire; DSM-IV: Diagnostic and statistical manual of mental disorders, 4th edition.

The most sensitive items per (sub)domain are listed in [Tables 3–7].

3.5. Level of evidence

The level of evidence for the difference between children with and without DCD is depicted in [Table 8]. Moderate evidence (low risk of bias, precise results but some inconsistencies and indirectness) was found for motor performance, sports and leisure, sensory processing and perception, and cognitive and executive functions [Fig. 1]. Hence, future research may still change the current findings. Very little evidence (moderate risk of bias, some imprecision, inconsistency, and indirectness) was found for self-perceived competence, quality of life, behavior, physical fitness, and physical activity. We are, therefore, uncertain about the current findings.

4. Discussion

The main purpose of this study was to determine which key aspects should be incorporated in the assessment to make up a pre-intervention assessment profile of a child with DCD. More specifically, we wanted to identify those domains implicated and how large the impact of that problem, deficit or limitation was. Based on an extensive literature search, the different domains [Table 2] and the most sensitive items to distinguish children with DCD from typically developing children per domain have been extracted [Table 3, Fig. 1].

With respect to the level of discrimination between DCD and TD groups, we found moderate evidence for measures of motor performance (other than motor scales used to diagnose DCD), executive functions, sensory processing and perceptions, cognitive functions, and sports and leisure activities [Fig. 1]; whereas very poor evidence was found for self-perceived competence, QOL, behavior, physical fitness and physical activity. Although the majority of the studies were of high quality (68%), there is scope to better control for confounders and better identification of DCD groups using all diagnostic criteria. Despite the moderate level of discrimination across domains, evidence for all domains was downgraded based on *indirectness* (i.e., how and what DCD criteria and outcome measures were applied), highlighting the need for future research using clear measurements and questionnaires/structured interviews that are sensitive and specific to the full diagnostic criteria for DCD. Only this way more homogeneous results can be reached, clarifying conclusions, particularly for domains where true differences are masked, which is likely for physical fitness, quality of life, self-perceived competence and behavior. Furthermore, inconsistent results (shown by non-overlapping 95% CIs across studies and high heterogeneity among them) also downgraded confidence in all domains, except for sports and leisure. Although the composition of the groups investigated may have also caused the inconsistency, other factors like the chosen statistics are also influential (and should be better justified). To afford parametric testing, the distribution of the data should be checked, as even the commonly used standardized motor tests are known to give skewed results [159].

Our results suggest that interindividual differences are present both within and between groups of children with DCD. Despite the fact that we are looking for test items that are particularly sensitive to DCD, not all children share the same difficulties. This is in line with previous studies that have attempted to cluster children based on test performance in different domains (e.g. executive functioning, motor skills, physical fitness) [44,160–162]; in general, the identified profiles vary according to which test items were included. Yet, we can be confident based on these results that a large number of children with DCD are prone to have difficulties with motor skills other than those included in the diagnostic assessments and with executive functions. By comparison, given the smaller SMDs, a smaller proportion of children will also struggle with sensory processing and perception, cognitive functions and sports and leisure. Precision of measurement (magnitude of the difference and optimal information size), on the other hand, was low in

Table 2
Number of studies, number of comparisons, percentage of negative results, when the studies were published, Standardized Mean Differences (SMD), standard error, z-score, p-value, 95% confidence interval of the SMD and heterogeneity in performances for each domain and subdomain between children with DCD and their TD peers.

Domain	Studies (n)	Comparisons (n)	% negative results	Time frame	SMD	SE	z-score	p-value	95% CI Lower	95% CI Upper	I^2
Motor performance [Appendix 4]	38	224	32.1	1988–2023	−0.941	0.0518	−18.178	<0.001	−1.042	−0.840	90%
Fine motor [44,45,46–48,49,50–60]	17	74	44.6	1988–2023	−0.922	0.1043	−8.838	<0.001	−1.126	−0.717	89%
Gross motor [45,61,62,54,57,59,63–70]	14	60	21.7	1992–2023	−0.837	0.0701	−11.943	<0.001	−0.975	−0.700	83%
Praxis/motor planning [28,44,71,72,73–77]	9	67	29.9	1997–2022	−0.881	0.0847	−10.400	<0.001	−1.046	−0.715	89%
Questionnaires [78,49,59,79,80]	5	23	26.1	2009–2019	−1.438	0.2165	−6.642	<0.001	−1.863	−1.014	91%
Physical fitness, physical activity and sports & leisure	43	276	38.6	2006–2023	−0.644	0.0358	−18.007	<0.001	−0.714	−0.574	88%
Physical fitness [Appendix 5]	24	132	28.8	2007–2023	−0.706	0.0516	−13.689	<0.001	−0.853	−0.465	87%
Aerobic fitness [20,22,81,82,83,84,85–96]	18	27	40.7		−0.659	0.0989	−6.665	<0.001	−0.770	−0.455	79%
Anaerobic fitness [20,21,97,81,84,67,85,88,91,92,98]	11	42	14.3		−0.872	0.0804	−10.842	<0.001	−1.030	−0.714	84%
Strength [20–22,97,81,82,62,84,67,87,88,91–94,99]	16	63	33.3		−0.612	0.0815	−7.500	<0.001	−0.771	−0.452	90%
Flexibility [22,81,100]	3	10									
Body composition [88]	1	4									
Physical activity [Appendix 6]	17	65	58.5	2005–2019	−0.550	0.0935	−5.887	<0.001	−0.734	−0.367	96%
Objective measures [87,101–105]	6	27	66.7		−0.383	0.1540	−2.490	0.013	−0.685	−0.082	98%
Questionnaires [106,107,62,80,86,108–113]	11	38	52.6		−0.673	0.1127	−5.967	<0.001	−0.893	−0.452	91%
Sports & leisure [48,59,79,80,90,102,105,110,111,114–116] [Appendix 7]	12	81	28.8	2007–2014	−0.629	0.0433	−14.518	<0.001	−0.714	−0.544	52%
Sensory integration and perception [Appendix 8]	17	96	38.5	1997–2022	−0.686	0.0540	−12.701	<0.001	−0.792	−0.580	80%
Pure sensory functions [28,33,44,61,117,118,119,120,51,121–123]	12	39	23.1	2001–2022	−0.760	0.0888	−8.555	<0.001	−0.934	−0.586	88%
Sensory tasks with a motor component [28,33,61,117,118,50,51,58,77,115,121–124]	14	57	49.1	1997–2021	−0.634	0.0686	−9.234	<0.001	−0.768	−0.499	71%
Cognitive and executive functions	31	228	37.3	1998–2023	0.762	0.0521	−14.621	<0.001	−0.864	−0.660	92%
Cognitive functions [Appendix 9]	23	111	43.9	1998–2023	−0.666	0.0625	−10.650	<0.001	−0.788	−0.543	91%
Academic skills [48,53–55,125–130]	10	45	20.0		−1.013	0.1326	−7.640	<0.001	−1.273	−0.753	96%
Mental Abilities [28,44,125,126,131,53,127,132–136]	12	31	61.3		−0.375	0.0678	−5.538	<0.001	−0.508	−0.242	76%
Mental imagery [72,74,137,138]	4	28									
Comorbidities [72]	1	4									
Emotion recognition [139]	1	6									
Executive functioning [Appendix 10]	14	115	30.7	2011–2022	−0.861	0.0835	−10.317	<0.001	−1.025	−0.698	92%
Inhibition [28,44,140,127,141–144]	8	37	45.9		−0.581	0.0704	−8.242	<0.001	−0.719	−0.443	68%
Working memory ([44,140,121,134,141,142,145,146])	8	27	23.1		−0.735	0.0837	−8.788	<0.001	−0.899	−0.571	57%
Flexibility/shifting [44,127,134,141,142,147]	6	24	29.2		−1.449	0.3485	−4.157	<0.001	−2.132	−0.766	98%
Planning [28,44,142,143]	4	7									
Dual tasking [140,54,57]	3	9									
Fluency [28,140,142]	3	8									
Attention [44,121]	2	3									
Self-perceived competence [Appendix 11]	9	31	48.4	2002–2023	−0.839	0.2452	−3.422	<0.001	−1.320	−0.359	98%
Physical abilities [148,62,58,149–151]	6	13	46.2	2002–2023	−1.308	0.4853	−2.695	0.007	−2.259	−0.357	99%
Academic skills [62,149–151]	4	7									
Self-esteem [106,90,149]	3	5									
Social competence [148,149]	2	4									
Physical appearance [148,149]	2	3									
Behavior (Appendix 12)	16	106	31.1	2007–2021	−1.909	0.3219	−5.931	<0.001	−2.540	−1.278	100%
Attention problems and hyperactivity [26,48,152,131,153,127,154]	7	13	38.5	2007–2020	−2.629	1.3229	−1.987	0.047	−5.222	−0.036	100%

(*continued on next page*)

Table 2 (*continued*)

Domain	Studies (n)	Comparisons (n)	% negative results	Time frame	SMD	SE	z-score	p-value	95% CI		I^2
									Lower	Upper	
Internalizing problems [26,48,152,106,153,155]	6	29	41.4	2007–2021	−1.978	0.6223	−3.179	0.001	−3.198	−0.759	100%
Social problems [26,48,152,105,135]	5	15	20.0	2007–2020	−2.515	1.1785	−2.134	0.033	−4.824	−0.205	100%
Somatic complaints [48,152,153,155]	4	5									
Externalizing problems [48,152,153]	3	11									
Sleep problems [37,153,156]	3	7									
Thought problems [48,152]	2	3									
Bullying [157,133]	2	12									
Stress [83]	1	5									
Emotional behavior [26]	1	1									
Self-esteem [133]	1	2									
Prosocial behavior [157]	1	1									
Independence [105]	1	1									
Autism features [153]	1	1									
Quality of life [Appendix 13]	6	43	34.8	2009–2021	−1.065	0.1916	−5.560	<0.001	−1.441	−0.690	99%
Social [37,84,156–158]	5	15	26.7	2009–2021	−1.108	0.3116	−3.554	<0.001	−1.718	−0.497	99%
Physical pain [37,84,157,158]	4	7									
Emotional [37,84,157,158]	4	6									
Self-esteem [37,157,84]	3	5									
Cognitive [84,157,158]	3	3									
Fatigue [156]	1	6									
Autonomy [157]	1	1									
Sleep [156]	1	1									

Abbreviations: n: number; SMD: Standardized Mean Difference; SE: standard error; 95% CI: 95% confidence interval; p-value: two-tailed; I^2: statistical measure for heterogeneity between studies.

studies showing very low levels of evidence. Larger 95% CIs indicate more variability in performance, highlighting the potential lack of sensitivity of specific tasks or tests within a domain. Therefore, we also reported the percentage of insignificant results, which varied between 14.3 and 66.7%. The varying levels of evidence and their implications for assessment and theory building warrant careful consideration at a domain level.

4.1. Motor performance

Results of this study also underline the heterogeneity of the primary motor problems. Individuals with DCD consistently exhibit motor skills below the expected level when compared to TD individuals across all domains of motor performance. Specifically, children with DCD demonstrated deficits in fine motor skills, gross motor skills, and praxis/motor planning compared with their TD peers [Table 2]. However, it is crucial to acknowledge that a substantial degree of heterogeneity exists even within the core domain of the disorder, being motor performance, with 32.1% of the 224 analyzed comparisons showing no discernible differences between children with DCD and their TD counterparts.

The task with the most notable discriminative power within the gross motor component appears to be the side jump [Table 3], a finding acknowledged in two separate studies [45,61]. It can be observed that the fine motor tasks displaying the highest discrimination ability are linked with activities of daily living, such as preparing the meal [46] or handwriting [47,48]. This assumption finds additional support in the results from the motor performance questionnaires, where items related to daily activities [78] and handwriting [49] also demonstrated the highest discrimination ability. Moreover, the implication of daily activities extends beyond the fine motor domain, as evidenced by the results from the praxis/motor planning domain. It is noteworthy that the items demonstrating the highest discrimination ability in this domain originate from the Gestures Test [71,72]. In this test, participants are required to perform gestures mimicking daily tasks like brushing teeth with a toothbrush or combing hair with a comb. Thus, the importance of including ecologically valid tasks is not confined to specific motor domains. Ensuring that assessment tasks mirror daily activities enhances the relevance and applicability of these tasks, thereby promoting a more authentic reflection of an individual's motor proficiency in everyday scenarios. Importantly, poor imitation in children with DCD also has repercussions for how we instruct children during testing and intervention.

4.2. Physical fitness, physical activity, and sports & leisure

The results indicate that children with DCD have lower physical fitness performance compared to their TD peers. This indicates that overall fitness levels need to be part of the performance profile of children with DCD, as deficits and limitations in all these areas were found. This supports other studies that have found similar results [20,23,81,97,163]. The low levels of fitness shown in children with DCD can increase the risk for health problems such as cardiovascular disease [164]. Also, low fitness levels will limit participation in active leisure, increasing the risk of lower levels of physical activity than their TD counterparts.

The results of the study corroborate other studies that have found differences in physical activity levels between children with DCD and TD children [165]. Although most studies confirmed that children with DCD were less active, the magnitude of the difference seems to depend on the tools used. The results indicated that children and parents reported greater differences in physical activity compared to objective measures of physical activity. This further highlights the need to include both objective and subjective measures of overall activity level in assessment to create a profile for children with DCD. Such a profile can help to develop appropriate intervention programs to improve physical fitness levels and increase participation in physical activity and leisure activities.

The study highlighted some gaps in the literature regarding physical activity levels in children with DCD. The study results indicated a smaller difference between children with DCD and TD children when using objective measures for physical activity. More studies are needed that compare subjective and objective measures and children's own reports and proxy reports (parent/teacher). In addition, the different

Table 3
Most sensitive items for motor performances (Standardized Mean Differences < −2).

Subdomain	Item	SMD	SE
Fine motor	the sensorimotor component of the "Do-Eat" test item "preparing chocolate milk" [46]	−3.99	0.46
	the sensorimotor component of the "Do-Eat" test item "tying an apron and making a sandwich" [46]	−3.76	0.44
	the sensorimotor component of the "Do-Eat" test item "filling out a certificate" [46]	−3.25	0.40
	the task performance of the "Do-Eat" test item "tying an apron and making a sandwich" [46]	−2.69	0.37
	letter formation during handwriting [47]	−2.40	0.35
	handwriting to dictation [48]	−2.30	0.34
Gross motor	Körperkoordinationstest für Kinder (KTK) side jump item [61]	−2.90	0.36
	KTK side jump item [45]	−2.36	0.31
Praxis/motor planning	the gestures test (total errors – imitation) [71]	−2.73	0.26
	the gestures test (total errors – on command) [71]	−2.15	0.54
	transitive reciprocal imitation [72]	−2.43	0.41
	transitive gestures on verbal command [72]	−2.41	0.41
	intransitive reciprocal imitation [72]	−2.24	0.24
Questionnaires	the Handwriting Proficiency Screening Questionnaire (HPSQ) time and speed of performance sum score [49]	−4.17	0.48
	the HSPQ physical and emotional well-being sum score [49]	−3.16	0.40
	the HSPQ legibility sum score [49]	−2.39	0.35
	the DCD Daily Questionnaire (DCDDaily-Q) Performance of self-care and self-maintenance activities [78]	−2.28	0.37
	the DCD Daily Questionnaire (DCDDaily-Q) Learning of self-care and self-maintenance activities [78]	−2.20	0.36
	the DCDDaily-Q Learning of gross motor activities [78]	−2.17	0.36
	the Children's Activity Scale for Parents (ChAS-P) ADL score [80]	−2.11	0.32

Abbreviations: SMD: Standardized Mean Difference; SE: Standard Error.

Table 4
Most sensitive items for physical fitness, physical activity and sports & leisure (Standardized Mean Differences < −2).

Subdomain	Item	SMD	SE
Physical fitness [Appendix 5]	strength: two-legged side hop [88]	−2.67	0.25
	strength: pushing a medicine ball [92]	−2.52	0.67
	strength: lower limb strength [62]	−2.43	0.13
	anaerobic capacity [97]	−2.14	0.21
	aerobic capacity [91]	−2.08	0.23
Physical activity [Appendix 6]	accelerometry [104]	−4.16	0.18
	the Children's Activity Scale for Parents (ChAS-P) by the parents [80]	−2.93	0.37
	the ChAS-P by the teacher [113]	−2.12	0.33

Abbreviations: SMD: Standardized Mean Difference; SE: Standard Error.

Table 5
Most sensitive items for sensory processing and perception (Standardized Mean Differences < −2).

Subdomain	Item	SMD	SE
Pure sensory function	depth perception [120]	−3.89	0.38
Sensory tasks with a motor component	copying items of the Developmental Test of Visual Perception (2nd edition) [118]	−2.17	0.41

Abbreviations: SMD: Standardized Mean Difference; SE: Standard Error.

Table 6
Most sensitive items for cognitive and executive functioning (Standardized Mean Differences < −2).

Subdomain	Item	SMD	SE
Cognitive functions	academic skills: basic reading skill of phrase making [48]	−4.50	0.35
	academic skills: basic reading skill of character pronunciation (MABC <5th percentile)	−3.85	0.32
	academic skills: basic reading skill of character pronunciation (MABC <15th/16th percentile)	2.98	0.29
Executive functions	the Wisconsin Card Sorting Test (WCST) when completing the first category [147]	−5.92	0.40
	the WCST, the total number of correct answers [147]	−4.95	0.34
	the WCST, the number of perseverative errors [147]	−4.62	0.33
	the WCST, the number of perseverative responses [147]	−4.29	0.31
	the WCST, the number of categories completed [147]	−2.26	0.22
Flexibility/ shifting	the Five Digit Test (Movement Assessment Battery for Children (MABC) < 15th/16th percentile) [134]	−2.28	0.28
	the Five Digit Test (MABC <5th percentile) [134]	−2.04	0.22

Abbreviations: SMD: Standardized Mean Difference; SE: Standard Error.

Table 7
Most sensitive items for self-perceived competence, behavior and quality of life (SMDs < −2).

Subdomain	Item	SMD	SE
Self-perceived competence	physical abilities: the Perceived Efficacy and Goal Setting System (PEGS) self-efficacy in leisure activities (Movement Assessment Battery for Children (MABC) <5th percentile) [151]	−5.47	0.51
	physical abilities: the PEGS self-care [151]	−4.18	0.42
	physical abilities: the PEGS self-efficacy in leisure activities (MABC <15th/16th percentile) [151]	−3.45	0.36
	academic skills: the PEGS schoolwork (MABC <5th percentile) [151]	−3.40	0.37
	academic skills: the PEGS schoolwork (MABC <15th/16th) [151]	−2.33	0.29
Behavior	the Child Behavior Checklist items [48]	[-15.38, −2.24]	[1.03; 0.25]
	the hyperactivity and attention subscale of the Strengths and Difficulties Questionnaire [127]	−2.22	0.34
Quality of life	the KINDL-R[a] school [84]	−5.79	0.46
	the KINDL-R[a] physical well-being [84]	−5.13	0.43
	the KINDL-R[a] friends [84]	−4.88	0.41
	the KINDL-R[a] emotional [84]	−2.02	0.29
	the PedsQL[b] psychosocial [158]	−2.04	0.11

Legend.
[a] KINDL-R: a questionnaire for health-related quality of life assessment in children, the revised version.
[b] PedsQL: Pediatric Quality of Life Inventory.

methods used to determine the levels of physical activity should be compared to determine the most reliable and ecological valid method for measuring physical activity.

4.3. Sensory processing and perception domain

Our findings [Table 2] revealed consistently lower scores for sensory processing and perception for the DCD group compared to TD peers, but

Table 8
Level of evidence for each subdomain.

Subdomain	Risk of bias	Precision*	Consistency**	Directness***	Level of evidence
Cognitive functions	High (n=1), moderate (n=6), low (n=16) (↑)	Precise (=)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊕⊕⊖
Executive functioning	Moderate (n=4), low (n=10) (↑)	Precise (=)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊕⊕⊖
Self-perceived competence	High (n=1), Moderate (n=3), low (n=5) (=)	Some imprecision (↓)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊖⊖⊖
Quality of life	Moderate (n=5), low (n=1) (=)	Some imprecision (↓)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊖⊖⊖
Behavior	High (n=1), moderate (n=5), low (n=10) (=)	Some imprecision (↓)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊖⊖⊖
Motor performance	Moderate (n=9), low (n=29) (↑)	Precise (=)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊕⊕⊖
Sensory processing and perception	Moderate (n=2), low (n=14) (↑)	Precise (=)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊕⊕⊖
Physical fitness	High (n=1), moderate (n=9), low (n=14) (=)	Some imprecision (↓)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊖⊖⊖
Physical activity	High (n=1), moderate (n=6), low (n=10) (=)	Some imprecision (↓)	Some inconsistency (↓)	Some indirectness (↓)	⊕⊖⊖⊖
Sports and leisure	Moderate (n=5) to low (n=7) (=)	Precise (=)	Consistent (=)	Some indirectness (↓)	⊕⊕⊕⊖

Legend: * Precision was graded through the magnitude of the 95% CI (smaller confidence intervals indicate higher precision) combined with the optimal information size (adequate sample size); ** Consistency was rated by comparing the variance in point estimates across studies, the overlap of the 95% CIs and heterogeneity (I^2 value); *** Directness was rated through differences in population (compliance with DCD criteria and the comparative definition of the TD group combined with the differences in outcome measures).

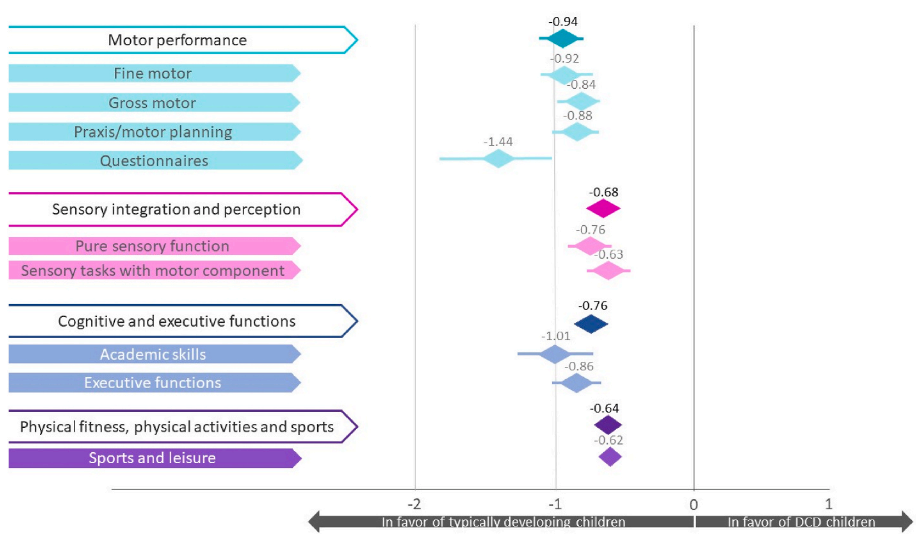

Fig. 1. Comparison of total standardized mean differences between typically developing children and children with developmental coordination disorder for which moderate evidence was found.

with a considerable level of heterogeneity among the studies suggesting variations in outcomes that extend beyond chance. The overall effect should, therefore, be interpreted with caution, and this emphasizes the need for exploring other contributing factors such as the composition of the sample (e.g. DCD criteria, and the presence of comorbidities).

While most studies demonstrated significant differences regarding the pooled SMD for sensory processing and perception, it is noteworthy that 38.5% of comparisons were statistically insignificant. Five studies [28,117–119,166] indicated that children with DCD did not perform statistically worse than the TD group regarding their perception skills as measured by the Beery-Buktenica Developmental test of Visual Motor Integration or the Test of Visual Perceptual Skills. This challenges the notion of a uniform sensory profile as a general deficit in children with DCD.

When examining the pure sensory functions (using tasks that do not require motor action), children with DCD exhibited a substantial deficit [Table 2]. The most pronounced differences were found in depth perception [Table 5] [120].

Regarding the sensory processing and perception tasks that require motor action, children with DCD also displayed deficits. The most significant differences were observed in copying items of the various test batteries that were used, indicating challenges in visuo-motor integration skills and eye-hand coordination skills.

The identified sensory processing and perception deficits that were found in this study underscore the intricate relationship between sensory processing, perception, and motor coordination deficits in children with DCD. These findings indicate that items that examine copying should be included in the assessment to profile children with DCD. Future research endeavors should focus on unravelling the sources of heterogeneity, addressing the nature of sensory processing and perception challenges that are implicated in children with DCD. Additionally, understanding the impact of perceptual visuospatial, oculomotor, depth perception, visuomotor, and tactile perception on motor skill development warrants further investigation.

4.4. Cognitive functions

Based on 228 comparisons, it is clear that in children with DCD cognitive as well executive functions are advised to be included in the assessment profile. Although the overall methodological quality of the included studies was sufficient, less than half of the studies clearly defined DCD according to all DSM-5 criteria, and only slightly more than half controlled for confounders. This poses the question of whether cognitive and executive function problems are key aspects of DCD or a mere reflection of the known overlap between DCD and other developmental disorders. Nonetheless, clinicians should be aware that DCD is not just a motor problem [6]. Specifically, when tailoring an intervention to a specific child, knowledge about co-occurring executive dysfunction or cognitive problems is essential. For example, if that child cannot integrate what the therapist demonstrates into their own action, and observational learning is at the base of your intervention, it will not be effective.

Indeed, the acquisition of motor skills is highly dependent on intact executive functions, which are also important for planning and organizing daily life activities. Our meta-analysis confirms the results of Subara-Zukic et al. [5], in showing clear problems with executive functioning in children with DCD. The largest effect size was found for flexibility/shifting abilities, particularly using the WCST [108]. For example, children with DCD needed approximately twice as many trials to complete the first category. The WCST is a test that can be easily assessed by a digital device and is hardly dependent upon motor proficiency, making it a suitable item to include in the pre-intervention assessment profile. A recent review showed significant correlations between executive function and the M-ABC [167]. So, even during the assessment, the intertwinement of symptoms needs attention. Interestingly, hot executive function (EF) was assessed by only one study [140] and differences were not significant. However, the (probable) DCD sample was small and not clearly defined. Since there is overlap between DCD and ASD [5], studying hot EF in children with DCD may be worthwhile.

4.4.1. Academic skills

The symptoms of children with DCD are known to interfere with their academic achievement. Decreased language skills as well as decreased math skills have been reported [168]. Of the nine studies that reported on academic skills, only two studies focused on mathematical skills [125,126]. Further insight into the extent of math problems in children with DCD is needed. Our meta-analysis showed pronounced differences for basic reading skills, which urge early recognition and additional educational support to limit the impact of these deficiencies on their academic career. Although the vast majority of existing studies have focused on children in the primary school age range, more recent studies have revealed the impact of childhood DCD on academic achievement in secondary school. For example, Harrowell et al. [169] reported a significantly lower educational level for adolescents with DCD than their peers at age 16, even when controlling for IQ. Importantly, they also reported that many of the participants with DCD did not receive any additional formal education support [169]. These findings underline the still hidden nature of the disorder and the importance of early recognition, not only of motor issues but also of cognitive and learning problems [6].

4.5. Behavioral and emotional problems and quality of life in DCD

Likewise, the analysis of the included studies on behavioral and emotional outcomes underlines that DCD is a heterogeneous disorder. In six out of seven studies, children with DCD had significantly more problems with attention and/or hyperactivity [26,48,82,131,152]. Externalizing problems also occur in children with DCD, such as rule-breaking and aggressive behavior. However, few studies addressed this topic, but those who did revealed more signs of externalizing behavior in both preschool [153] and primary school children [48,152]. So far, it is unknown which factors mediate the presence of externalizing behavior in DCD. Clinicians should be aware of the possible occurrence of behavioral problems, and screening of these problems should be part of the assessment to make up a profile of a child with DCD.

In addition, children with DCD experienced significantly more internalizing problems, i.e. depression and anxiety, than TD children in all six studies included [26,48,152,106,155]. According to the Environmental Stress Hypothesis (ESH), both psychological stressors (e.g. stress, low self-concept) and psychosocial stressors (e.g. lack of social support by parents, peers and teachers) mediate the link between DCD and internalizing problems [170–172]. Stressors are challenging circumstances that exceed the coping ability of a child, leading to arousal, tension and worrying [173]. Until now, only one study investigated the presence of signs of stress in children with probable DCD, with more signs of stress reported by children in the p-DCD group than in the TD group [83].

Negative self-concept also is a stressor related to internalizing problems [174]. Self-concept is the evaluation of competence once compared to internal standards or in relation to others [175]. Generally, different self-concept domains are discerned, such as perceived physical and cognitive competence and social acceptance [176]. A lower perceived physical competence was found in children with DCD compared to TD children in five out of six studies [18,62,107,148,177,178]. In addition, self-efficacy was lower in leisure activities, self-care and schoolwork. Intriguingly, not all children with DCD experience stress or develop lower perceived motor competence, lower social acceptance and/or internalizing problems [174]. An unexpected finding was that children with DCD regularly perceived themselves not (yet) to have poor motor competence, as almost half the comparisons reported (48.4%) revealed insignificant results. Research should focus on which factors induce resilience and protect children against stress and the development of low perceived competence and internalizing problems. This understanding can guide the development of interventions aimed at preventing the emergence of these stressors. In addition, assessment of the possible occurrence of these problems by clinicians needs to be incorporated in a profile of a child with DCD, in order to induce timely management of these problems by providing support and increasing public understanding [179].

Of the five studies measuring HRQoL, all showed that total HRQoL [158] or specific domains of HRQoL were affected in the DCD group, in particular the physical and psychosocial domains [37,157,84]. In one study [157], only children with combined DCD and ADHD suffered from lower HRQoL, children with DCD only did not. More research is needed to investigate which factors moderate or mediate lower HRQoL in children with DCD. The presence of stress and stressors can have an impact on sleep, resulting in fatigue. According to the two studies that addressed sleep or fatigue in children with DCD, parents reported their children to have more sleep problems [153,156], and to suffer from daytime sleepiness and cognitive fatigue, i.e. lack of concentration and memory problems. Physical fatigue was not reported [156].

4.6. Implications for clinical practice

The fact that the meta-analyses revealed significant differences in all reported domains between children with DCD and their TD peers, confirms that they are at risk of presenting difficulties at different levels of functioning. However, the magnitude of the differences indicates that, although they can, not all will exhibit the same problems. Clinicians should be aware that motor skills, i.e. fine, gross and praxis, other than those evaluated with the diagnostic general motor tests (e.g. MABC-2, BOT-2), can be deficient. Therefore, focusing on these specific motor skills can provide deeper insight into the challenges children are experiencing. Preferably, the selection of such skills can be facilitated with sensitive motor tasks (e.g. side jump, letter formation in handwriting or producing gestures to imitation or verbal command) combined with specific questionnaires regarding skill execution (e.g. DCD-Daily) and is based on the request for help to shape the task-oriented intervention. Furthermore, executive functions should not be overlooked in this context. Children with DCD struggle with learning and executing motor skills. Planning, shifting, working memory, inhibition can be targeted in motor intervention as part of making tasks more complex. The domains that either have smaller SMDs or for which the evidence is still weak, should also be considered by the treating professionals. The pre-intervention multi-dimensional assessment profile can be built using the most sensitive items for each domain, after they have been carefully mapped during the anamnesis.

A recent study advocated for more awareness regarding DCD among professionals in both health care and education [6]. Our meta-analysis confirms the wide-spread difficulties in children with DCD, insights that are much needed, not only for professionals to know which aspects should be tackled in therapy, but also which professionals can and may even need to be involved to ensure the best care for the child. For instance, formal educational support at school, psychological support (together with the family) and preferably in an interdisciplinary context, where different professionals can have a cumulative positive effect in the child's treatment.

5. Conclusion

The insights gained in the analyses are critical to create awareness in clinicians and researchers about the widespread impact of the disorder and the need to identify the key aspects of that are causing children with DCD to experience difficulties in daily life.

Despite the fact that DCD is defined by marked impairment in motor skills, a unified agreement on key tasks for assessing the disorder as a whole is lacking. Hence, one of our primary objectives was to identify (optimal) motor tasks that best capture varying levels of motor proficiency in individuals and can be considered as key features for profiling children with DCD. Importantly, we showed that while motor tests discriminate at least moderately well, there remains substantial heterogeneity in the pattern of motor performance on these "core" measures across studies. When motor measures other than those in screening tests are considered, the level of heterogeneity increases again. Further, our results suggest a need for motor assessments that more closely mirror real-life activities (i.e. ecological validity) across different performance domains.

For non-motor domains, group differences were not as large as for the motor domain. There was moderate discrimination between groups for executive/cognitive functions, sensory and perceptual processing, and sports and leisure activities indicating these aspects need to be incorporated in the multi-dimensional assessment profile. However, the very poor evidence for aspects of psychosocial function, QOL, behavior, physical fitness and physical activity hints towards their potential importance, but needs further investigations, until then they can be considered but are not required as such. The diverging nature and severity of the primary motor problems as well as the presence of problems in other domains emphasizes the diversity present within DCD children and provides a rationale for explaining the heterogeneity in this clinical group. Yet, they highlight the potential involvement of these domains in children with DCD and warrant clinicians to be alert for and assess other aspects than merely motor skill difficulties.

A number of glaring deficiencies in the reported research were identified. Our review could not provide definitive information about the prevalence of co-occurring disorders, as these were poorly evaluated or reported in the included studies. There is also a need for more studies with clinical samples of DCD children as they formed only 1/5 of the total DCD sample of the meta-analysis. Consequently, the depiction presented here may be somewhat optimistic, since it is predominantly derived from convenience samples where the application of DSM criteria was assessed retrospectively.

In conclusion, there is a definite need to develop motor assessments that more closely mirror real-life activities (i.e. ecological validity) across a range of performance domains, highlighting the broader significance of this principle in motor assessment practices. Our data provide evidence which key aspects or outcomes should be included in a profile, which after piloting can be established for clinical use. It also confirms that clinicians have a role to play as part of an interdisciplinary team, tackling the difficulties encountered by children with DCD from a holistic point of view.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bj.2024.100768.

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