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Faculteit Revalidatiewetenschappen

master in de revalidatiewetenschappen en de kinesitherapie

Masterthesis

Auditory motor coupling and rhythm perception in typically developing children, children with autism spectrum disorder, attention deficit hyperactivity disorder and developmental coordination disorder

Janique Roufs
Annelise Vos

Eerste deel van het scriptie ingediend tot het behalen van de graad van master in de revalidatiewetenschappen en de kinesitherapie

PROMOTOR :

Prof. dr. Eugene RAMECKERS

BEGELEIDER :

Mevrouw Mieke GOETSCHALCKX

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How are auditory motor coupling and rhythm perception in TD, ASD, ADHD and DCD children assessed?

How do auditory motor coupling and rhythm perception differ in these populations?

Is there a link between auditory motor coupling and rhythm perception in these populations?

Highlights:

- *The MBEMA test is a valid and reliable method for assessing rhythm perception.*
- *Tapping tasks and the BAASTA test are applicable for measuring auditory motor coupling.*
- *ADHD and DCD children differ from TD children on rhythm perception and auditory motor coupling.*
- *ASD children do not differ from TD children on rhythm perception and auditory motor coupling.*
- *Rhythm perception is an important skill for performing auditory motor coupling tasks.*

Students : Roufs Janique & Vos Annelise

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Orientation

This master thesis frames within the research domain pediatric rehabilitation. Auditory-motor coupling and rhythm perception frequently appear in the daily functioning of humans, including children. Moving to the rhythm of a beat is inherent for humans. Rhythm perception is necessary for good auditory-motor coupling, without a decent perception synchronization will be poor.

Humans spontaneously move to the rhythm of a beat. It emerges in early childhood and continues to develop into adulthood. Recently more interest is gathered into the ability to perceive rhythms and to synchronize to these rhythms in various child populations, such as Autism Spectrum Disorder (ASD), Attention-Deficit Hyperactivity Disorder (ADHD) and Developmental Coordination Disorder (DCD). Because these populations have difficulty coordinating movements, it is interesting to know if impaired rhythm perception, and certainly auditory-motor coupling, contribute to their poor motor performance.

In other pathologies, such as cerebral palsy (CP), stuttering and learning disorders, rhythm perception and auditory motor abilities was assessed (Schweizer, Eylon, & Katz-Leurer, 2020). Research done by El Shemy and El-Sayed (2018) found that including a rhythmic auditory stimulation protocol in gait and gross motor training in ASD children leads to more improvements on the BOT-2 test compared to normal gait and gross motor training.

In the pediatric domain a limited amount of research has been done to examine rhythm perception and auditory-motor coupling in these child populations (Chang et al., 2021; Y. Y. Chen et al., 2013; Gaul & Issartel, 2018; Getchell, McMenamin, & Whitall, 2005; Kagerer & Clark, 2015; Khalil, Minces, McLoughlin, & Chiba, 2013; Lesiuk, 2015; Puyjarinet, Bégel, Lopez, Dellacherie, & Dalla Bella, 2017; Roche, Viswanathan, Clark, & Whitall, 2016; Tryfon et al., 2017; Whitall et al., 2008; Whitall et al., 2006). Nevertheless, no systematic review is available to compare ADHD, ASD and DCD with typically developing children. Furthermore, research to evaluate if poor rhythm perception is linked to poor auditory-motor synchronization is lacking.

This master thesis is part of an ongoing research project at the University of Hasselt. It regards a preparatory pilot study as part of the doctoral study of Goetschalckx Mieke with the title: “Rhythmic interlimb coordination in children with Developmental coordination disorder compared to typical developing children: the effects of individual, task and environmental

constraints". Test locations are: Hasselt (Fitlink, sporthal Runkst), Gent (GUSB), Mol and REVAL Diepenbeek. The children are tested in a nearby gymnasium of the residence of the children. The study started in September 2019, children's testing started in January 2021.

A central format was used. The research question was drafted by the doctoral student Goetschalckx Mieke. The students worked intensively to finish a successful literature study. The selection and assessment of the articles was performed by both students independently and discrepancies were discussed in consultation with each other. All the following parts of the study, such as drafting the inclusion- and exclusion criteria, data-extraction and discussion as well as writing the final review were performed by both students, in close cooperation. The final draft was read and approved by both students.

The research protocol was drafted by both master students, with the existing protocol of doctoral student Goetschalckx Mieke as guideline. The protocol was consulted for information about the methodology of the study.

Part 1: Literature study

Content

1. Abstract

Background: Children with Attention-Deficit Hyperactivity Disorder (ADHD), Developmental Coordination Disorder (DCD) and Autism Spectrum Disorder (ASD) all suffer from decreased motor performance.

Methods: A systematic literature search was performed identifying studies including rhythm perception and auditory motor synchronization in typically developing (TD) children and children with ASD, ADHD and DCD. This yielded 13 relevant studies and these were categorized into different population groups.

Results: DCD children were proven to be more variable in auditory motor synchronization compared to TD children. ADHD children performed worse on rhythm synchronization and perception tasks than TD children. For ASD children no significant differences were found. Multiple tasks were proven to be useful for assessing rhythm synchronization and perception. These tasks included rhythm perception tasks like MBEMA and rhythm synchronization tasks like the BAASTA assessment and tapping tasks. Psychometric properties of these tasks were not commonly reported.

Discussion and conclusion: TD children perform better than ADHD and DCD children, but not ASD children in rhythm discrimination or auditory motor synchronization tasks.

Objective of protocol: The aim of the study is to explore the performance of children with ADHD, DCD and ASD compared to TD children on auditory motor synchronization tasks and rhythm perception tasks.

Operationalization research question of protocol: Children aged 6 to 12 years old perform the abbreviated MBEMA and a tapping task to respectively evaluate rhythm perception and auditory motor synchronization.

Important keywords: Rhythm perception, auditory motor synchronization, children, Attention-Deficit Hyperactivity Disorder (ADHD), Developmental Coordination Disorder (DCD), Autism Spectrum Disorder (ASD), typically developing (TD) children.

2. Introduction

In daily life people spontaneously move to the beat of a rhythmic sound by tapping with their finger or foot or through walking (Leman et al., 2013). This spontaneous movement is a natural human behavior and emerges in early childhood and improves with age into adulthood (Damm, Varoqui, De Cock, Dalla Bella, & Bardy, 2020). Children older than 2,5 years can clap to a regular spontaneous tempo and can slow down the tempo to the auditory stimulation. By the age of 4, anticipation of sound and adjusting the motor action to the stimuli is reached (Provasi & Bobin-Begue, 2003). For the development of auditory-motor coupling the intent is more important than the auditory cueing for producing less variable limb patterns (Getchell, 2007). More, Moore, Cowan, Riley, Edmondson-Jones, and Ferguson (2011) found that until the ages of 7 to 9, auditory processing is more variable and has higher thresholds compared to adult norms.

Rhythm perception is the basis for auditory motor coupling, this is expressed as the ability to recognize and process a musical rhythm. Auditory motor coupling is the integration of an auditory stimulus and the motor output. For accurate coupling the ability to adapt motor performance to the auditory cue is necessary. Synchronizing and coupling movements to auditory cues is a difficult task, limbs are instructed to move at the same frequency as the stimulus and need to 'frequency lock'. During the entire task the limbs are expected to synchronize the movements with the beat. In order to achieve this, the limbs must additionally 'phase lock' (Getchell, 2007; Sternad, Dean, & Schaal, 2000). This occurs naturally in children during a dual motor task (Getchell et al., 2005; Getchell & Whitall, 2003). Auditory motor coupling is known to improve and become less variable with aging (Drewing, Aschersleben, & Li, 2006; McAuley, Jones, Holub, Johnston, & Miller, 2006).

Recent work has shown that some child disorders may have an impaired rhythm perception and/or auditory motor coupling that can affect their motor performance (Bhat, Landa, & Galloway, 2011; Chang et al., 2021; Y. Y. Chen et al., 2013; Chukoskie, Townsend, & Westerfield, 2013; Gaul & Issartel, 2018; Jamey et al., 2019; Kagerer & Clark, 2015; Khalil et al., 2013; Lesiuk, 2015; Puyjarinet et al., 2017; Roche et al., 2016; Rosenblum & Regev, 2013; Tryfon et al., 2017; Whitall et al., 2008; Whitall et al., 2006). Because rhythmic synchronization improves entrainment and performance on coordination tasks (Getchell, 2007; M. Thaut, Tian, & Azimi-Sadjadi, 1998; M. H. Thaut, McIntosh, Prassas, & Rice, 1992), there is an increased

interest in research of this subject. This review therefore distinguishes auditory motor coupling and rhythm perception in typically developing children, children with Developmental Coordination Disorder, children with Attention-Deficit Hyperactivity Disorder and children with Autism Spectrum Disorders.

Children with Developmental Coordination Disorder (DCD) typically have impairments in motor performance not explained by any neurological condition or IQ (American Psychiatric Association, 2013). DCD is found in 6% of the children between the ages 5 to 13 (American Psychiatric Association, 2013). Attention Deficit Hyperactivity Disorder (ADHD) is typically described as a neurodevelopmental disorder. Key findings in children with ADHD are according to the DSM-V impulsivity, inattention and hyperactivity (American Psychiatric Association, 2013). These symptoms are assumed to be related to motor performance. Difficulty with motor coordination and slow reaction time are presented (Kaiser, Schoemaker, Albaret, & Geuze, 2015). Motor problems occur in 30 to 50% of children with ADHD (Fliers et al., 2009; Gillberg et al., 2004; Goulardins, Marques, Casella, Nascimento, & Oliveira, 2013). According to the DSM-V criteria children with an Autism Spectrum Disorder typically have repetitive behaviors and impairments in social interaction (American Psychiatric Association, 2013). About 50 to 85% of children with ASD present impairments in motor performance (Bhat et al., 2011; Chukoskie et al., 2013). Children with ASD have difficulties in gross and fine motor tasks, impaired performance in sequential and imitation based tasks and difficulty coordinating both sides of the body in rhythmic tasks (Kaur, S, & A, 2018). In children with ASD, motor abnormalities may decrease with maturation, therefore age variability may influence the overall motor performance of children with ASD (Alsaedi, 2020).

Relatively little is known about the impact of a decreased rhythm perception or rhythm synchronization ability on motor performance. Because the previously mentioned disorders all suffer from decreased motor performance, it is interesting to know whether there is a link between a decreased rhythm perception or auditory motor coupling. We hypothesize that children with ASD, ADHD and DCD differ from typically developing children in their ability to perceive rhythms or to synchronize these rhythms or auditory sounds to their movements, known as auditory motor coupling. Furthermore, we accept that older children have a better performance than younger children, due to the continued development of rhythm synchronization.

To our knowledge no systematic review exists for this relatively new topic. Therefore, the aims of this review are (1) to summarize literature discussing auditory-motor coupling and rhythm perception, (2) to assess methodological quality of the relevant literature, (3) to summarize methods for assessing rhythm perception and/or auditory motor coupling, (4) to conclude how auditory motor coupling and rhythm perception differ in these child populations and (5) to conclude whether there is a link between auditory motor coupling and rhythm perception in these child populations.

3. Method

3.1 Research question

This literature search was used to answer the following questions: How are auditory-motor coupling and rhythm perception in children with DCD, ADHD, ASD and typically developing children assessed? How does rhythm perception and auditory motor coupling differ between TD children and children with DCD, ADHD and ASD? And is there a link between rhythm perception and auditory-motor coupling in children with DCD, ADHD and ASD?

3.2 Literature search

For the literature search the databases of PubMed and Web of Science (WOS) were used. Relevant terms were identified and consisted of 'auditory motor coupling', 'rhythm perception', 'motor timing', 'sensorimotor synchronization', 'synchronization', 'auditory-perceptual timing', 'auditory-motor', 'perceptual timing', 'beat perception', 'rhythmic auditory stimulation', 'sensorimotor coordination', 'sensorimotor coupling'. These terms were considered relevant for identifying the outcomes. The following terms were used to identify the relevant population: 'autism spectrum disorder', 'autism', 'attention deficit disorder with hyperactivity', 'attention deficit hyperactivity disorder', 'ADHD', 'developmental coordination disorder', 'DCD', 'developmental dyspraxia', 'motor skills disorder', 'motor skills disorder', 'perceptual-motor disorder', 'pediatric', 'school-aged children', 'typically developing children'. The key terms within both categories were combined using the Boolean operator 'OR' and between categories with the Boolean operator 'AND'. 'Sleep', 'speech', 'stutter', 'literacy', 'dyslexia' or 'cerebral palsy' were excluded using the Boolean operator 'NOT'. For an overview of the key terms used see appendix A. The search strategy was first performed in February 2021. In addition, the reference lists were scanned for other relevant studies. The search strategy was repeated in May 2021 to check if new studies were published regarding the subject of interest. All these new titles were excluded based on outcomes.

3.3 Inclusion and exclusion criteria

Inclusion criteria used in this systematic review were (1) children aged between 6 to 18 years old (2) children with (probably) Developmental Coordination Disorder (DCD), Autism Spectrum Disorder (ASD), Attention-Deficit Hyperactivity Disorder (ADHD) and/or children with a typical development, (3) studies that addressed auditory motor coupling and/or rhythm perception, (4) studies with a RCT, cohort, cross-sectional and/or case-study design and (5) the article's full text was published in English.

Studies were excluded if: (1) study population had a neurological disorder, musculoskeletal disorder, cardiorespiratory disorder, communication disorder, intellectual developmental disorder or a learning disorder, (2) the study included animal models, (3) the study population consisted of adults, defined as ages older than 18 years, (4) assessment of visual or social motor coupling, (5) usage of medication, (6) neuro-imaging and/or neurophysiological measurements, (7) conferences, meta-analysis, reviews, systematic reviews and practice guidelines, (8) studies in languages other than the Dutch or English language, (9) studies with low quality defined as a score less than 60% on the Downs and Black checklist and (10) studies without full-text available after contacting first authors.

3.4 Methodological quality

All included studies were assessed with the Downs and Black Checklist for Quality Assessment (D&B) (Downs & Black, 1998) for both randomized and non-randomized studies. The checklist consists of 27 questions addressing: reporting, internal validity - bias and confounding, external validity and power assessment. To emphasize the importance of double blinding in the study protocols, item 15 was modified to 'was an attempt made to blind investigators and assessors'. The checklist is originally developed for randomized and non-randomized studies. Items can be adjusted to fit the study's design. This checklist was chosen because it assesses both randomized and non-randomized studies and a high internal validity (0,89), a good interrater (0,75) and test-retest reliability (0,88) was reported for this checklist (Downs & Black, 1998). Assessing methodological quality is often done using the Cochrane's risk of bias tool (ROB). However, it was found that this tool is frequently implemented in a non-recommended way (Jørgensen et al., 2016). Moreover, the subitems of the ROB are less objective than those of the D&B checklist. Thus, for systematically scoring the methodological quality, the Downs & Black could better be used. For the assessment of the cross-sectional studies the following items were removed: reporting - item 8 and 9, external validity - item 13, internal validity; bias - item 14, 17 and 19, internal validity; confounding - 23 and 24. These items were not applicable to cross-sectional studies. Quality of the studies was categorized in three different grades: (1) good, defined as a score greater than or equal to 75%, (2) moderate, defined as a score greater than or equal to 60% but lower than 75%, (3) bad, defined as a score lower than 60% (Munn, Sullivan, & Schneiders, 2010) . For an overview of the methodological quality of the included studies, see table 1. The checklists and an analysis of strengths and weaknesses of all included studies can be found in appendix B.

3.5 Data-extraction

The following data was extracted from the included studies: participant characteristics, the method of intervention and/or assessment and the study results concerning rhythm perception or auditory motor coupling.

Table 1

Methodological quality of the included studies scored with the Downs and Black checklist

	Reporting								External Validity		Internal Validity - B				Internal Validity - C			Power	Score
	1	2	3	4	5	6	7	10	11	12	15	16	18	20	21	22	25	27	
TD children																			
Gaul & Issartel (2018)	y	y	n	y	p	y	y	n	utd	utd	utd	y	y	y	utd	utd	n	utd	47%
Kagerer & Clark (2015)	y	y	y	y	p	y	y	n	utd	utd	utd	y	y	y	utd	utd	utd	utd	53%
ASD children																			
Jamey et al. (2019)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	y	utd	y	utd	68%
Tryfon et al. (2017)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	y	utd	utd	utd	63%
ADHD children																			
Chen et al. (2013)	y	y	n	y	y	y	y	y	utd	utd	utd	y	y	y	n	utd	y	utd	63%
Khalil et al. (2013)	y	y	n	y	p	y	n	n	y	y	n	y	y	y	y	y	y	utd	68%
Lesiuk (2015)	y	y	n	y	p	y	y	y	y	y	utd	y	y	y	y	utd	y	utd	74%
Puyjarinet et al. (2017)	y	y	y	y	p	y	y	n	utd	utd	utd	y	y	y	y	utd	utd	y	63%
DCD children																			
Chang et al. (2021)	y	y	y	y	p	y	n	y	utd	utd	y	y	y	y	y	y	y	n	74%
Roche et al. (2016)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	n	utd	y	utd	63%
Rosenblum & Regev (2013)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	y	utd	n	utd	63%
Withall et al. (2006)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	n	utd	n	utd	58%
Withall et al. (2008)	y	y	y	y	p	y	y	y	utd	utd	utd	y	y	y	utd	utd	n	y	63%

Abbreviations: y: yes; n: no; utd: unable to determine

4. Results

Various possibilities to assess rhythm perception and/or auditory motor coupling are listed in section 4.3 and table 2. An overview of the results is presented in table 3. This table provides a summary of the study characteristics: participants, inclusion and exclusion criteria, study protocol and results. Section 4.4 describes the results of auditory motor coupling and rhythm perception in children with ADHD, DCD and ASD and whether this differs from typically developing, age-matched children. At last, the link between auditory motor coupling and rhythm perception in TD, DCD, ADHD and ASD children is described in section 4.5.

4.1. Study selection

The literature search revealed a total of 983 studies (310 Pubmed - 673 WOS). After removing duplicates 741 studies were screened based on title and abstract. Thirty-five studies were identified as possibly relevant, other studies were excluded based on outcome measures (n = 442), population (n=203) or design (n= 61). Thorough full-text analysis resulted in the exclusion of another 22 articles. Only assessments of rhythm discrimination consisting of multiple beats were included in contrast to timing discrimination. Timing discrimination, defined as tasks in which children were asked to discriminate between lengths of a stimulus and this in contrast to rhythm discrimination tasks, where children were asked to recognize a different rhythm. Timing discrimination was found not to be relevant for the purpose of this study. After rescreening in May 2021, WOS reported 21 new published articles, while Pubmed reported 11. After removing duplicates, ten articles were excluded based on title and abstract, one article was excluded after full-text analysis based on outcome measures. No new articles were included after rescreening.

Reference list of the remaining 12 articles were screened and yielded 1 extra result. Finally, 13 studies were included in this systematic review. All these 13 studies included in this review had a cross-sectional design. The included studies were categorized according study population: (1) (only) typically developing (TD) children (n= 2), (2) children with Autism Spectrum Disorder (ASD) (n= 2), (3) children with Attention-Deficit Hyperactivity Disorder (ADHD) (n= 4), and (4) children with Developmental Coordination Disorder (DCD) (n= 5).

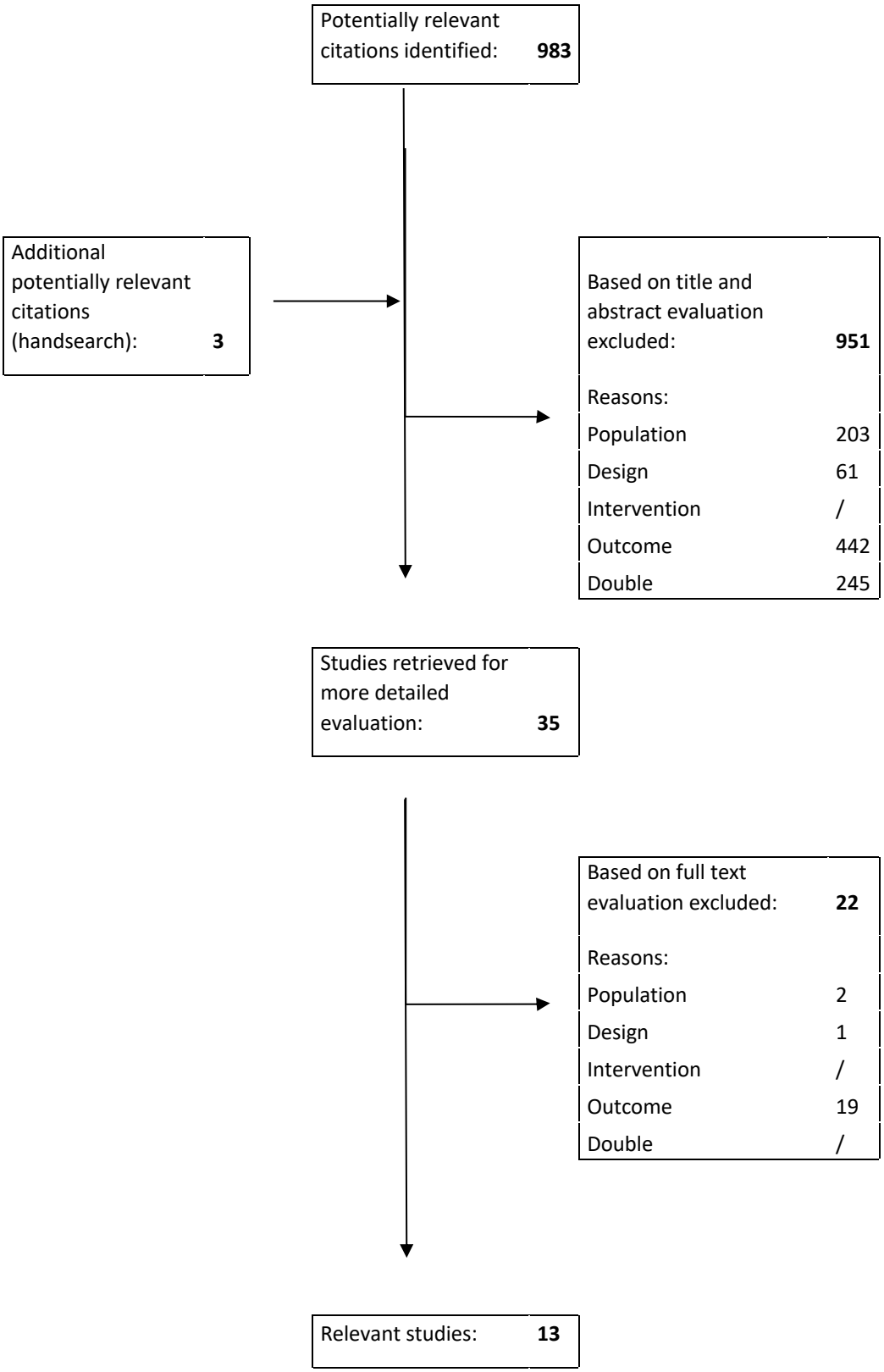

Figure 1. Flowchart of included studies

4.2. Methodological quality

The two studies with typically developing children (Gaul & Issartel, 2018; Kagerer & Clark, 2015) had a low methodological quality as assessed by the Downs and Black checklist. Description of the participants' characteristics was insufficient in Gaul and Issartel (2018). Additionally, the two studies did not report whether the included children were representative of the normal population and did not report the recruitment of their participants.

The included studies describing rhythm perception and auditory motor coupling in ASD children (Jamey et al., 2019; Tryfon et al., 2017) had moderate quality. They did not address whether the study population was representative of the entire population.

During the assessment of methodological quality of the included studies regarding ADHD children, three studies did not clearly describe the participants' characteristics (Y. Y. Chen et al., 2013; Khalil et al., 2013; Lesiuk, 2015). Additionally, in two of these studies (Y. Y. Chen et al., 2013; Puyjarinet et al., 2017) they did not report whether the study population was representative of the entire population, increasing the risk of selection bias. None of these studies reported blinding of the investigators. Puyjarinet et al. (2017) and Khalil et al. (2013) did not report to adjust confounders in their data-analysis.

Out of the five included studies assessing DCD children, none of these studies reported generalizability. Only Chang et al. (2021) reported blinding of the assessors and investigators. Whitall et al. (2006), Whitall et al. (2008), Chang et al. (2021) and Rosenblum and Regev (2013) did not adjust for confounders in their results. In addition, the five studies did not perform a power analysis.

The lack of describing participants' characteristics resulted in an increased risk of selection bias. The risk of selection bias increases when the study population is not representative of the entire population. During quality assessment, participants characteristics were underreported in most included studies. Of all included studies only Chang et al. (2021) reported blinding. Not mentioning blinding of assessors and investigators may increase the risk of confirmation bias (Tripepi, Jager, Dekker, & Zoccali, 2010).

4.3 Rhythm perception and auditory motor coupling tasks

4.3.1 Tapping tasks

Of the 13 studies included in this review, three used a tapping task to investigate auditory motor coupling in children. Tryfon et al. (2017) used an auditory motor synchronization task in which children with ASD were instructed to synchronize their tapping with variable rhythms. The task involved participants to listen to a woodblock rhythm and then tap back in synchrony with the rhythm a second time. They included three rhythms that were randomly presented at three different levels of increasing metrical complexity. The original task that was previously used in typically developing adults was adapted in order that it would be more suitable for children.

Roche et al. (2016) examined the abilities of children with and without DCD to perceive auditory changes in rhythm, and their modulation of bimanual finger tapping in response to these changing rhythms. They used a tapping task in which the participants tapped their index fingers alternately in time to the presented auditory signals. The task included two conditions: (1) gradual/subliminal, where the auditory stimuli were gradually ramped up in steps, and (2) abrupt, where the auditory stimuli were abruptly ramped up.

Finally, Whitall et al. (2008) used a similar task to examine the abilities of children with and without DCD to perceive auditory changes in rhythm, and their modulation of bimanual finger tapping in response to these changing rhythms/frequencies. The participants listened to the beat for five seconds, after these five seconds they were verbally cued to begin alternating tapping, one finger in time with each beat. To increase the attentional demands of this task a non-homologous pair of fingers (left index finger and right middle finger) was chosen.

Neither of these studies described the psychometric properties of their protocols.

4.3.2 Rhythm perception

Both Lesiuk (2015) and Chang et al. (2021) used a discrimination task to assess rhythm, tone or beat discrimination. Lesiuk (2015) used five music perception subtests for the evaluation of music perception abilities of children with and without executive function deficits. The study used a melodic tone discrimination test, pitch discrimination task, pulse count task, rhythm discrimination task and a duration discrimination task. In the melodic tone discrimination task participants had to identify the tone that was changed. In the pitch discrimination task participants had to indicate if the tones were higher or the same. In the pulse count task

participants had to count the number of pulses that would occur between two beep sounds. In the rhythm discrimination task participants had to indicate whether a presentation of two rhythmic phrases were the same or different. At last, in the duration discrimination task participants had to indicate whether the second tones were shorter or longer than the first.

Chang et al. (2021) used three behavioral tests to measure the auditory perceptual thresholds in children at risk for DCD and TD children. These tests assessed rhythm discrimination, duration discrimination and pitch discrimination. Similar to the study of Lesiuk (2015), participants had to indicate in the duration discrimination task whether a tone-pair was shorter in contrast with the other tones. In the rhythm discrimination task participants were instructed to select the sequence with a changing rhythm. At last, the pitch discrimination task was used as a control task to ensure that the potential differences found between the groups were not due to any issues with hearing, testing procedure or engagement. In this task participants were instructed to select the tone-pair with different tones or different pitch frequencies.

Neither of these studies described the psychometric properties of their protocols.

4.3.3 Battery for the Assessment of Auditory and Sensorimotor Timing Abilities (BAASTA)

Puyjarinet et al. (2017) assessed perceptual and sensorimotor timing skills of children with ADHD with the Battery for the Assessment of Auditory and Sensorimotor Timing Abilities (BAASTA), short version. This test consists of a set of perceptual and motor production timing tasks, of which this study selected five for assessing beat tracking skills in children with and without ADHD. Of the five tasks selected in the study, three were perceptual tasks and two were sensorimotor tasks. The perceptual tasks consisted of (1) anisochrony detection with tones, (2) anisochrony detection with music and (3) beat alignment task (BAT). The control task for the perceptual tasks consisted of a duration discrimination task. The sensorimotor tasks consisted of (1) paced tapping to tones and (2) paced tapping to music. The control task for the sensorimotor tasks consisted of unpaced tapping. The control tasks were performed to assess perception without beat tracking and motor variability without auditory stimuli. No psychometric properties of their abbreviated version were described.

4.3.4 Gross motor synchronization

Six of the included studies in this review used gross motor tasks to assess auditory-motor coupling or rhythm discrimination. None of the studies used the same method. Khalil et al.

(2013) assessed group rhythmic synchrony in TD children with a percussion instrument, based on the gamelan musical art. Participants, attempted to synchronize their percussions with an isochronous beat, played by the instructor over one minute. To measure synchronization the study calculated the synchrony between each player and the leader. This is the first time a study uses such a gamelan-like instrument to measure group synchronization. Furthermore, the study conducted no research to assess the psychometric properties of this methodology.

Y. Y. Chen et al. (2013) assessed the timing perception ability and motor coordination of ADHD and TD children in a series of rope jumping tasks involving different rates of rope jumping. Rope jumping skills involve good perception of time discrimination (Y. Y. Chen et al., 2013). The participants were instructed to jump at a constant speed of 100Hz, followed by jumping at variable speed where the participants had to switch between 80Hz, 100Hz or 120Hz. The three tempi were indicated by a sound played by a CD player. The study reported no research to assess the psychometric properties of their methodology.

A study by Gaul and Issartel (2018) assessed the sensory-motor integration of TD children through a rhythmic unimanual coordination task using a handheld pendulum. The study used a total of three stimuli, of which two were unimodal stimuli (auditory or visual) and one bimodal stimulus (auditory and visual). Participants were instructed to the endpoint of the movement with the visual or auditory endpoints. In the unimodal visual condition, participants were instructed to swing the pendulum to the side where the stimuli moved to. In the unimodal auditory condition participants were instructed to swing the pendulum to the left when the sound panned to the left ear with a high pitch and to the right when the sound panned to the right ear with a low pitch. The bimodal condition was similar to the unimodal condition, where the presentation of both auditory and visual stimuli appeared at the same time. The methodology of this study was previously used (Armstrong & Issartel, 2014), nevertheless, no research had been conducted to assess the psychometric properties of this methodology.

Whitall et al. (2006) used a dual motor task, in which TD children and children with DCD had to clap and march to a specific beat. They chose this method after reasoning that this dual-motor task paradigm would be the most useful in quantifying auditory-motor coupling in children with DCD because these children are more likely to have both coordination and stability problems with the task, even without adding an auditory cue (Whitall et al., 2006).

Hereby they maximize the likelihood of finding differences between TD and DCD children (Whitall et al., 2006). Participants were instructed to clap, using hand-held cymbals, and march at the same time to an auditory beat in four different conditions: 0.8Hz, 1.2 Hz, 1.6Hz and 2.0Hz. This methodology, the dual-motor task, was previously used in other studies (Getchell et al., 2005; Getchell & Whitall, 2003), nevertheless, no research had been conducted to assess the psychometric properties of this methodology.

A study by Rosenblum and Regev (2013) assessed the timing abilities of TD children and children with DCD through the interactive metronome (IM). The interactive metronome is a computer-based version of a traditional music metronome used to improve timing accuracy in musicians (Bartscherer & Dole, 2005). Participants were instructed to synchronize their movements with the rhythm dictated by the computer, while performing 14 various tasks, such as clapping hands, tapping both toes or heels on a footpad or tapping the right or left to or heel on a footpad (Rosenblum & Regev, 2013). The participant's timing score is the difference in milliseconds between the moment the beat sounds and the participant's motor response. A longer response time indicates a lower time management ability. The study by Rosenblum and Regev (2013) analyzed three of the seven categories, namely hands, feet and bilateral tasks. The analysis yielded high values of reliability: 0.87 for the category hands, 0.92 for the category feet and 0.81 for the category bilateral. The reliability of all 14 tasks was 0.95 (Rosenblum & Regev, 2013). The Interactive Metronome is therefore proven to be reliable. Other psychometric properties of this methodology are unknown.

At last, a study by Kagerer and Clark (2015) assessed auditory-motor integration in TD children. Blindfolded participants were instructed to move a pen towards the perceived sound source. Acoustic stimuli were presented in one of two speakers positioned at 45° and 135° relative to the home position. The study conducted no research to assess the psychometric properties of this methodology.

4.3.5 Montreal Battery for the Evaluation of Musical Abilities (MBEMA)

Jamey et al. (2019) used the abbreviated Montreal Battery for the evaluation of musical abilities, in short MBEMA, to assess a range of music perception abilities in ASD and TD children. The abbreviated MBEMA is an ideal test battery for this purpose because it is an objective, short and child-friendly test battery of music perception that can be used in both healthy and clinical populations and across cultures (Peretz et al., 2013). The stimuli in the

abbreviated MBEMA focusses on music discrimination, rather than emotional discrimination (Gosselin, Paquette, & Peretz, 2015), limiting potential confounding influence of emotional valence (Jamey et al., 2019). The abbreviated MBEMA includes three types of tests, namely melodic pitch, rhythm and memory. The task consists of 30 unfamiliar melodies, of which 20 are used for the melodic pitch and rhythm tests and 10 of those 20 melodies for the memory test plus the remaining 10 melodies. In the melodic pitch test participants were instructed to make a same-different judgement between two short melodies that may differ in the pitch of one note. In the rhythm test, equally to the melodic pitch test, participants were instructed to make a judgement between two short melodies that may differ in rhythmic grouping. At last, in the memory test participants were instructed to recall if a given melody was heard before or not. The study conducted no research to assess the psychometric properties of this methodology.

Table 2

Data overview of the included assessments

	Participants	Task	Subtests	Outcome	Psychometric properties
TD children					
Gaul & Issartel (2018)	71 TD	Handheld pendulum swing	Unimodal condition; Bimodal condition	Auditory motor coupling	Unknown
Kagerer & Clark (2015)	51 TD, 11 adults	Pointing to an auditory stimulus with a pen	/	Auditory motor coupling	Unknown
ASD children					
Jamey et al. (2019)	41 ASD, 32 TD	Abbreviated version of the MBEMA	Melodic pitch discrimination; Rhythm discrimination; Memory task	Rhyhtm perception	Unknown
Tryfon et al. (2017)	31 ASD, 23 TD	Tapping task	/	Auditory motor coupling	Unknown
ADHD children					
Chen et al. (2013)	10 ADHD, 10 TD	Rope jumping	Constant speed; Variable speed	Auditory motor coupling	Unknown
Khalil et al. (2013)	102 children	Playing a percussion instrument	/	Auditory motor coupling	Unknown
Lesiuk (2015)	29 EF difficulties, 42 TD	Discrimination task	Melodic tone discrimination; Pitch discrimination; Pulse count discrimination; Rhythm discrimination; Duration Discrimination	Rhythm perception	Unknown

Table 2 (continued)

	Participants	Task	Subtests	Outcome	Psychometric properties
Puyjarinet et al. (2017)	41 ADHD (22 ADHD only, 19 ADHD + DCD), 14 TD	BAASTA	Perception tasks: Anisochrony detection with tones; Anisochrony detection with music; Beat alignment test; Duration discrimination task (control task); Sensorimotor tasks: Paced tapping to tones; Paced tapping to music; unpaced tapping (control)	Rhythm perception and auditory motor coupling	Unknown
DCD children					
Chang et al. (2021)	20 rDCD (5 rDCD + pADHD, 15 rDCD - pADHD), 27 TD	Discrimination task	Rhythm; Duration; Pitch discrimination	Rhythm perception	Unknown
Roche et al. (2016)	24 DCD, 22 TD	Tapping task	Gradual condition; Subliminal condition	Auditory motor coupling	Unknown
Rosenblum & Regev (2013)	21 DCD, 21 TD	Interactive Metronome	Clapping; Tapping toes; Tapping heels; Tap right hand, left toe; Tap left hand, right toe; Balance feet; Repeat clapping	Auditory motor coupling	Reliability: hands - 0,87; Feet - 0,92; Bilateral - 0,81
Whitall et al. (2006)	10 DCD, 8 TD, 10 adults	Clap and march to a beat	/	Auditory motor coupling	Unknown
Whitall et al. (2008)	10 DCD, 10 TD, 10 adults	Tapping task	/	Auditory motor coupling	Unknown

Abbreviations: MBEMA: Montreal Battery of Evaluation of Musical Abilities; BAASTA: Battery for Assessment of Auditory Sensorimotor and Timing Abilities.

4.4 Auditory motor coupling and rhythm perception in various populations

4.4.1 TD children

4.4.1.1 Study characteristics

In this systematic review two studies were included investigating rhythm perception and/or auditory motor coupling in TD children. In the study by Gaul and Issartel (2018), a sample of 71 children had to perform a unimanual coordination task in which a pendulum had to move in synchrony with the auditory stimulus in various frequencies (preferred, -20% and +20%). All participants were right-handed, had normal or corrected vision and no known neuromuscular deficit. In the study of Kagerer and Clark (2015) 51 children between the ages of 5 and 12 participated. Like the previous study, all participating children were right-handed and had normal or corrected vision. Participants were asked to point to the auditory target. In addition, all children were tested with the Movement Assessment Battery for Children 2 (MABC-2). A score at or above the 20th percentile was necessary for children to be included. Children were asked to point to the source of an auditory stimulus, while being blindfolded. This was done in two different noise situations: with and without background noise.

4.4.1.2 Results

Gaul and Issartel (2018) reported significant differences between the multiple frequencies where the participants demonstrated lower coordination in the +20% frequency condition compared to the preferred frequency. Additionally, they found a significant effect for class. Fifth class children performed significantly better than 1st and 3rd class children. Kagerer and Clark (2015) reported findings for the initial direction error (IDE) in their task. Initial direction error was defined as the difference between the direction in which children pointed first and the actual direction of the source of the sound. They reported significant effects for direction and noise and also significant interactions between direction x age group and noise x age group. Children from all age groups presented higher IDE during both noise conditions. No significant difference was found for the variability of IDE between age groups or noise condition.

4.4.2 ASD children

4.4.2.1 Study characteristics

Two studies were included assessing children with ASD. Jamey et al. (2019) assessed musical perception using the abbreviated version of the Montreal Battery for the Evaluation of Musical Abilities (MBEMA). This study included 41 boys with ASD and 23 TD boys. ASD boys were

included when they were diagnosed by expert opinion and if this was supported by standard diagnostic measures (Autism Diagnostic Interview-Revised (ADI-R) and Autism Diagnostic Observation Schedule (ADOS)). Inclusion in the ASD group required a score on the Social Communication Questionnaire (SCQ) of minimum 13. For TD boys this score was maximum 12. Lastly, Tryfon et al. (2017) used a tapping task in which children were instructed to tap in synchrony to a woodblock rhythm. Thirty-one boys with ASD and 23 TD age-matched boys were assessed. ASD diagnosis was given by expert opinion and supported by standard diagnostic measures (ADI-R and ADOS). TD participants did not have a neurological or psychiatric illness in their history.

4.4.2.2 Results

Jamey et al. (2019) showed that the accuracy scores on the MBEMA did not differ between the ASD and TD group. The accuracy was influenced by IQ and age but not by ASD symptom severity. In addition, Tryfon et al. (2017) found no between group differences for performance on the motor rhythm (tapping) synchronization task.

4.4.3 ADHD children

4.4.3.1 Study characteristics

Four studies were included focusing on children with ADHD or ADHD-like symptoms. Lesiuk (2015) included 71 children with (n= 29) or without (n= 42) executive function deficits measured with standardized executive function scores. ADHD is proven to be a disorder with executive function deficits, therefore this study was included in our ADHD subset (Yáñez-Téllez et al., 2012). Beat discrimination was assessed with the Music Perception Inventory. Puyjarinet et al. (2017) used the Battery for the Assessment of Auditory and Sensorimotor Timing Abilities (BAASTA) for measuring perceptual and sensorimotor timing. They included 55 children, of which 22 children with ADHD only, 19 children with ADHD and DCD and 14 TD control children. The ADHD-only group received their diagnosis based on the DSM-5 criteria, complemented by interviews with parents and validated parent and teacher reported questionnaires. The ADHD-DCD group performed below the 15th percentile on the MABC-2. In the study by Khalil et al. (2013), children were asked to synchronize playing on a gamelan-like instrument with a rhythm played by the group leader on a similar instrument. The study population consisted of 102 children from grades two to six and were tested on ADHD like symptoms using the Strength and Weaknesses of ADHD Symptoms and Normal Behaviour (SWAN) rating scale. The last study (Y. Y. Chen et al., 2013) assessed the ability of children to

synchronize their rope-jumping to a specific frequency produced by a CD player. Ten children diagnosed with ADHD and ten age-matched TD controls participated in the study.

4.4.3.2 Results

Lesiuk (2015) demonstrated that children with executive function deficits scored lower on all music perception subscores with significantly differences found on duration discrimination and rhythm discrimination. Puyjarinet et al. (2017) reported that children with ADHD, with and without DCD, showed general difficulties in perceiving durations and in tracking the beat in comparison to the control group. Children with ADHD found it difficult to judge whether a sound was misaligned or not with a musical beat. In the tapping task they found that children with ADHD-DCD synchronized worse to a beat than children with only ADHD. But children with ADHD only differed significantly from the control group. While TD controls tapped well to both tones and melodies, ADHD children had more difficulty when tapping to melodies than to tones. Khalil et al. (2013) demonstrated a significant correlation between the ability to synchronize with scores on the SWAN scale, indicating that better synchronizers are more attentive and show less ADHD-like behaviors. In the study by Khalil et al. (2013) the mean value of hand-foot deviation time was significantly greater in the ADHD group compared to the control group. Y. Y. Chen et al. (2013) reported that participants in the ADHD group fell more often during the rope jumping task and presented greater variation in timing in both the foot jumping and rope whirling cycle than the control group.

4.4.4 DCD children

4.4.4.1 Study characteristics

Five studies were selected regarding rhythm perception and auditory motor coupling in DCD children. Rosenblum and Regev (2013) included 21 children with DCD and 21 age- and gender-matched TD children. Children were included in the DCD group if the mABC-2 total scores were below the 15th percentile. Children in both DCD and TD group performed movement tasks to the Interactive Metronome (IM). Roche et al. (2016) used data from 24 DCD children and 22 age- and gender-matched TD controls for the determination of the auditory threshold using a staircase method where children were asked to discriminate between two rhythms. They used data from 19 children with DCD and 17 age- and gender-matched controls for the tapping experiment. Whitall et al. (2006) tested 28 participants in three groups. The DCD group consisted of 10 children who were included if motor performance was below the norm for age and intelligence assessed by a neurodevelopmental exam and a score at or below the 10th

percentile on the MABC-2. Ten age- and gender-matched children were included in the control group. The last group consisted of ten college-aged adults. The participants were instructed to clap and march along to a beat in different frequency conditions. Inclusion of participants in the study by Whitall et al. (2008) is similar as Whitall et al. (2006). They included ten DCD children, ten TD children and ten adults. DCD children were evaluated with the following criteria: (1) a diagnosis given by a pediatrician through a medical and developmental history and neurodevelopmental exam using the Neurodevelopmental Specialists Service (NESS), (2) a score on the mABC-2 of less than 15% and (3) normal cognitive functioning on the Woodcock-Johnson tests of Cognitive Abilities (W-J). Participants were tested using a tapping task. Chang et al. (2021) included 61 children aged between six and seven years old. (rDCD) children were scored based on the following criteria: (1) a score at or below the 16th percentile on the MABC-2, (2) evidence of impact on daily functioning, (3) IQ above the fifth percentile (4) no medical condition affecting motor functioning. Five participants in the rDCD/DCD group were identified with probable-ADHD. The auditory perceptual thresholds of the participants were tested using duration discrimination, rhythm discrimination and pitch discrimination tasks.

4.4.4.2 Results

In the first study (Rosenblum & Regev, 2013) including children with DCD, significant group differences were found for response timing in all categories of the Interactive Metronome task. This indicates that the response time of DCD children is longer in comparison to TD controls, thus DCD children perform worse in synchronizing their movements to the metronome. Roche et al. (2016) found no group effect in the perceptual threshold analysis and in the performance on the tapping task. DCD and TD groups differed significantly in the variability within a trial, this effect was seen in the perceptible condition and in the subliminal condition. Whitall et al. (2006) demonstrated that adults performed better than both groups of children when assessing absolute deviation of phasing their clap and march to an auditory metronome beat. Both groups of children did not differ significantly from each other. The variability of the s to the beat was significantly greater in the DCD group than the adult and TD groups. DCD children show a similar mean error compared to TD children but the synchronization is less stable. Whitall et al. (2008) found no significant group effect when assessing normalized intertap interval. They did find a significant lower performance when DCD children were asked to match the frequency of 0,8 Hz compared to adults and TD

controls. No significant differences were found during determination of how closely the participants matched their tap to the auditory signal. DCD children were significantly more variable than TD controls and adults in matching their taps to the auditory beat. This was seen as well when assessing variability within a trial. Lastly, statistically significant differences were found between the rDCD group and the TD group in the study by Chang et al. (2021). The rDCD group showed lower thresholds in duration discrimination and rhythm discrimination in comparison to the TD controls. There was also a trend for lower pitch discrimination thresholds but this did not reach statistical significance. These findings indicate that rDCD have inferior rhythm perception than TD controls.

4.5 Link between auditory motor coupling and rhythm perception in TD, ASD, ADHD and DCD children

Puyjarinet et al. (2017) was the only included study that performed a rhythm perception task and an auditory motor coupling task. Both tasks are part of the BAASTA test protocol. TD and ADHD children and adults were asked to tap regularly with and without an auditory beat. They found that when tapping without sound, children and adults with ADHD showed a significantly higher motor variability compared to TD controls. When controlled for this higher motor variability ADHD children and adults still showed lower synchronization performances than controls. Indicating that motor performance is not the only important variable for accurate rhythm synchronization and therefore, rhythm perception is an important skill for performing auditory motor coupling tasks.

Table 3

Data overview of the included studies

	Participants	Characteristics*	Age range*	Method	Results
TD children					
Gaul & Issartel (2018)	71 TD	Righthanded, normal or corrected vision and no known neuromuscular deficit	Unknown	Synchronizing unimanual movements of swinging a handheld pendulum to an auditory stimuli at a preferred frequency (PF), +20% and -20%.	Synchronization: 5th > 3rd class ($p<0,05$), 5th > 1st ($p<0,01$), 3rd > 1st ($p<0,05$); +20%: 5th > 3rd ($p<0,05$) and 5th > 1st ($p<0,01$); PF: 5th > 1st ($p<0,01$)
Kagerer & Clark (2015)	51 TD, 11 adults	Righthanded, normal or corrected vision and MABC ≥ 20th	5-12 yrs	Pointing blindfolded to an auditory beat presented in speakers standing in different corners	Initial Directional Error: 7-8yrs ≠ 9-10 yrs for direction of target ($p<0,05$); Variability: 7-8 yrs = 9-10 yrs
ASD children					
Jamey et al. (2019)	41 ASD, 32 TD	ASD diagnosis, score on SCQ > 13, gestational age older than 35 weeks and absence of hearing impairment	6-12 yrs; 10,1 ± 1,8	Rhythm and melodic pitch discrimination using the Montreal Battery for the Evaluation of Musical Abilities (MBEMA)	Accuracy: ASD = TD; no significant effect of age in ASD vs TD; in ASD only a significant effect of age on rhythm and melodic pitch discrimination; no significant effect of symptom severity on rhythm and melodic pitch discrimination
Tryfon et al. (2017)	31 ASD, 23 TD	ASD diagnosis, supported by ADI-R & ADOS, normal hearing, IQ > 70, gestational age older than 35 weeks, absence of a neurological disease	6,9-15,6 yrs; 11,5 ± 2,8	Synchronization of finger tapping to a woodblock rhythm	Intertap interval: ASD = TD; Onset asynchrony: ASD = TD; Non-absolute onset asynchrony: ASD = TD; Global accuracy: ASD = TD

Table 3 (continued)

	Participants	Characteristics*	Age range*	Method	Results
ADHD children					
Chen et al. (2013)	10 ADHD, 10 TD	ADHD diagnosis by local hospital, no combined syndromes	9,65 ± 1,27	Coordination of rope jumping to a metronome beat	Hand-foot deviation: ADHD > TD (p<0,01); variation in foot jumping and rope whirling cycle: ADHD > TD (p<0,05)
Khalil et al. (2013)	102 children	ADHD-like behavior measured with SWAN-C and SWAN-I	7-12 yrs	Synchronizing to a beat played by the instructor	Synchronization was significantly correlated by SWAN-I and SWAN-C (p<0,05)
Lesiuk (2015)	29 EF difficulties, 42 TD	Present executive function deficits measured with standardized EF scores (BRIEF)	9-11 yrs	Discrimination of melodic tone, pitch, pulse count, rhythm and duration using the Music Perception Inventory	Duration discrimination: no-EF > EF (p=0,007); rhythm discrimination: no-EF > EF (p=0,024)
Puyjarinet et al. (2017)	41 ADHD (22 ADHD only, 19 ADHD + DCD), 14 TD	ADHD diagnosis based on DSM5 criteria, IQ > 70, ADHD+DCD: MABC < 15th, no other comorbid conditions	8,7 ± 1,5	Perceptual and sensorimotor timing skills assessed with the Battery for the Assessment of Auditory and Sensorimotor Timing Abilities (BAASTA)	Beat perception: ADHD < TD (p<0,0001); tapping synchronization: ADHD < TD (p<0,00001), ADHD-DCD < ADHD (p<0,01);
DCD children					
Chang et al. (2021)	20 rDCD (5 rDCD + pADHD, 15 rDCD - pADHD), 27 TD	MABC ≤ 16th, no intellectual disability, absence of any medical condition affecting motor functioning	6,17-7,92 yrs; 6,88 ± 0,55	Behavioral tests measuring auditory perceptual thresholds for duration discrimination (DD), rhythm discrimination (RD) and pitch discrimination (PD)	DD: DCD > TD (p=0,028); RD; DCD > TD (p=0,037); PD: DCD = TD

Table 3 (continued)

	Participants	Characteristics*	Age range*	Method	Results
Roche et al. (2016)	24 DCD, 22 TD	Independent diagnosis of DCD, MABC score ≤ 5th, no neuromuscular or pervasive developmental disorder and no cognitive impairment	6-11 yrs; 9,29 ± 1,79	Rhythm perception task discriminating different auditory stimuli; Tapping task synchronizing taps to the auditory stimuli in subliminal and perceptible condition	Rhythm perception: DCD = TD; Tapping task: DCD = TD in subliminal and perceptible condition; variability DCD > TD in the subliminal (p<0,001) and perceptible condition (p=0,02)
Rosenblum & Regev (2013)	21 DCD, 21 TD	MABC ≤ 15th, 20 right-handed, 1 left-handed, 8 girls, 13 boys, no known neurotic/emotional disorders, autistic disorders, physical disabilities and neurological diseases	7-10 yrs; 9;9 ± 0;1	Synchronization of gross motor movements to an Interactive Metronome (IM)	Timing ability: DCD < TD (hands (p<0,0001), feet (p< 0,0001), and bilateral tasks (p< 0,0001)).
Whitall et al. (2006)	10 DCD, 8 TD, 10 adults	MABC ≤ 10th, normal cognitive functioning and no general medical disorder	7,09 ± 0,48	Coordination of rhythmic clapping and marching to an auditory beat	Absolute Deviation of Phasing: DCD = TD; Variability: DCD > TD/adults (p<0,001)
Whitall et al. (2008)	10 DCD, 10 TD, 10 adults	Independent diagnosis of DCD, MABC score ≤ 15%, normal cognitive functioning no pervasive developmental disorder and no motor difficulties due to a general medical condition	6,2-7,6 yrs; 7,04 ± 0,42	Synchronizing taps to an auditory stimuli in multiple frequencies using a non-homologous pair of fingers.	Intertap Interval: DCD = TD; Variability of phase: TD < DCD (p=0,0001); phase variability within a trial: TD < DCD (p=0,02)

*treatment group

Abbreviations: SCQ: Social Communication Questionnaire; ADI-R: Autism Diagnostic Instrument – Revised; ADOS: Autism Diagnostic Observation Scale; SWAN: Strengths and Weaknesses of ADHD behavior and Normal behavior; SWAN- C: Concentration items; SWAN- I: Inattention items; BRIEF: Behavior Rating Inventory of Executive Function; MABC: Movement Assessment Battery for Children

5. Discussion

The aim of this systematic review was to summarize literature discussing auditory-motor coupling and rhythm perception in children with ADHD, DCD and ASD in order to answer the following research questions: (1) How are auditory-motor coupling and rhythm perception in children with DCD, ADHD, ASD and typically developing children assessed? (2) How do auditory motor coupling and rhythm perception differ in these children's populations? And (3) is there a link between rhythm perception and auditory-motor coupling in children with DCD, ADHD and ASD?

In total, 13 articles could be included in the review. The methodological quality of two of the included studies was low. The other 11 articles had a moderate methodological quality. The studies by Gaul and Issartel (2018) and Kagerer and Clark (2015) were included in this review to be able to assess rhythm perception and auditory motor coupling in typically developing children. Otherwise, no comparison was possible.

Different tasks were used to assess auditory motor coupling and rhythm perception in children, namely: (1) tapping tasks, (2) rhythm perception tasks and (3) gross motor rhythm synchronization tasks. Two studies used a test battery for the evaluation of rhythm perception or synchronization. Puyjarinet et al. (2017) assessed rhythm synchronization with the BAASTA, Jamey et al. (2019) assessed rhythm perception with the MBEMA. Of the different tasks, only one was assessed for reliability. Rosenblum and Regev (2013) assessed the reliability of the Interactive Metronome, for the evaluation of a gross motor task, and found that the Interactive Metronome has good reliability to assess timing abilities of TD and DCD children. Based on the lack of research to the psychometric properties of the other tasks, no conclusion can be drawn in regards to the reliability, validity, sensitivity and specificity of these tasks.

Besides the different tasks that were used to assess auditory motor coupling and rhythm perception, different children's populations were examined, namely TD (two studies, 122 children), ASD (two studies, 72 children), ADHD (four studies, 182 children of which 19 with comorbid DCD) and DCD (five studies, 65 children and 20 with risk of DCD) children. Based on the 13 study results, is seems that children with ADHD or DCD perform differently in auditory motor coupling and rhythm perception in comparison to TD children. Children with DCD with comorbid ADHD perform even worse in rhythm synchronization and rhythm perception tasks

than only DCD or ADHD children. Up to now, no results are available to show a different auditory motor coupling or rhythm perception ability in children with ASD compared to TD. For ASD children no significant differences were found in rhythm synchronization and perception in comparison with TD children. Kagerer and Clark (2015) and Gaul and Issartel (2018) report contradictory findings in the TD children-sample when assessing the effect of age, on performance on rhythm perception and on auditory motor coupling tasks. Both studies had a low quality assessed with the Downs and Black checklist and did not report a power calculation or generalizability. Furthermore, Kagerer and Clark's protocol differs greatly from Gaul and Issartel's protocol, and therefore measures different outcomes. These are some of the probable causes for the discrepancy in the TD sample.

The findings for ADHD and DCD are in line with other pathologies. Compared to TD children it was reported that children with cerebral palsy had no significant different rhythm perception but were more variable and had more difficulties with matching their steps to a higher or lower frequency than their preferred step frequency (Schweizer et al., 2020). Similar results were reported for children with learning difficulties, they performed worse on tapping tasks than age and gender matched controls (Cumming, Wilson, Leong, Colling, & Goswami, 2015). Children who stutter had worse rhythm discrimination than TD children (Wieland, McAuley, Dilley, & Chang, 2015). Atypical rhythm perception was reported to be a risk factor for developing speech and language disorders (Ladányi, Persici, Fiveash, Tillmann, & Gordon, 2020).

When comparing the possible protocols and their psychometric properties for measuring rhythm perception and auditory motor coupling, no conclusion could be made regarding which test has the best psychometric properties or is best to implement. None of the studies assessed the reliability or validity of their test, except Rosenblum & Regev. Therefore, other literature has been taken into account. J. L. Chen, Penhune, and Zatorre (2008) stated that the tapping task, used by Tryfon et al. (2017), is sensitive to measure fine-grained tapping timing. The population did not consist of ASD children; therefore, it has to be stated that the performance characteristics of individuals with ASD on such tasks has not yet been well defined. Furthermore, Axelrod, Meyers, and Davis (2014) assessed different types of finger tapping tasks and found a specificity of approximately 90% and a sensitivity around 40%. Tapping tasks are also used for diagnosing psychogenic movement disorders (PMD), and

proved to be 89% specific and 77% sensitive for the diagnosis of PMD (Criswell, Sterling, Swisher, Evanoff, & Racette, 2010).

The rhythm perception tasks used, in the study of Lesiuk (2015) were previously used for testing children's musical aptitudes and are proven reliable, with a relatively high validity (Seashore, Lewis, & Saetveit, 1956). It is unknown if the tasks have been defined for ADHD children, therefore careful interpretation of the results of these tasks is needed.

Furthermore, research has been done to assess the BAASTA. The BAASTA is a test battery that originally consists of several tasks, including duration discrimination, anisochrony detection with tones and anisochrony detection with music. Furthermore, a beat alignment task is normally used. Production timing is also assessed with unpaced tapping, paced tapping to an isochronous sequence, paced tapping to music, synchronization-continuation and adaptive tapping. A study by Dalla Bella et al. (2017) proved that the battery is sensitive to poor perceptual and sensorimotor timing skills. In regards to these results and by keeping in mind that ADHD and DCD children have poor auditory motor coupling, the battery might be sensitive to measure differences between TD and DCD/ADHD children.

The second test-battery included in this review is the MBEMA, or the Montreal Battery for the Evaluation of Musical Abilities. Previous research done by Peretz, Champod, and Hyde (2003) concluded that the adult version of the MBEMA, the MBEA or the Montreal Battery of Evaluation of Amusia, is theoretically motivated and has good psychometric properties. It is proven to be sensitive, to have normally distributed scores and a good test-retest reliability. Nonetheless, the battery has only been validated for adults. Peretz et al. (2013) states that in their experience, the MBEA cannot be used with children younger than 10 years of age, but they suggest to adapt the task demands by reducing the length of the melodies from 10 notes to 7, reducing the number of test items from 30 to 20 and eliminating the metric test because a pilot study has proven that this test is too difficult for 6-year old's. Because of the impracticality, Peretz et al. (2013) constituted an abbreviated MBEMA and conclude that the MBEMA shows better sensitivity than the MBEA in children. In contrast to the full version, which took over 30 to 45 minutes to administer, the abbreviated MBEMA only takes 20 minutes. Furthermore, both versions, the MBEA and the MBEMA are freely accessible. The abbreviated MBEMA is used by Jamey et al. (2019) to assess ASD and TD children

Lastly, the only study that evaluated the psychometric properties of their task, the Interactive Metronome, was Rosenblum and Regev (2013). The study found a good reliability for three categories, ranging between 0.81 and 0.92. Kuhlman and Schweinhart (2004) assessed the internal reliability and concurrent validity between the Metronome's diagnostic items. The internal reliability was found to be 0.89. Furthermore, the concurrent validity for the school performance test was r =.264, p=.001.

After comparing the possible protocols and their psychometric properties for measuring rhythm perception and auditory motor coupling, taking into account research assessing the psychometric properties, the abbreviated MBEMA was deemed the most fit for evaluating rhythm perception in children. For the assessment of rhythm synchronization, other tasks like tapping tasks, the BAASTA and the Interactive Metronome can be used. Whereas the BAASTA and tapping tasks are proven to be sensitive, the IM is proven to be reliable.

In Puyjarinet et al. (2017) a link between auditory motor coupling and rhythm perception was made. They stated that accurate synchronization between an auditory cue involved perceptual and motor components. Thus, rhythm perception is essential for performing auditory motor coupling tasks. These findings are in line with Sowiński and Dalla Bella (2013). Sowiński and Dalla Bella (2013) included various non-musicians that were deemed as poor synchronizers when performing a tapping task. They concluded that poor synchronizers perform worse on rhythm perception tasks compared to good synchronizers. Furthermore, they state that poor synchronization cannot be merely ascribed to motor performance, because good and poor synchronizers were similarly accurate when tapping to a spontaneous beat.

Recommendations for future research consists of performing high quality RCTs for determining therapeutic importance. Indications for using rhythm perception and auditory motor tasks in treatments are already reported for different neurological pathologies. Crosby, Wong, Chen, Grahn, and Patterson (2020) found that rhythm-based treatments are a potential consideration for treating post stroke individuals. Their findings are supported by Gonzalez-Hoelling, Bertran-Noguer, Reig-Garcia, and Suñer-Soler (2021), they reported that including rhythmic auditory stimulation (RAS) in therapy improves walking ability in subacute stroke patients. Similar results were reported in a systematic review and meta-analysis done by Yoo

and Kim (2016), they support the application of rhythmic auditory cueing in rehabilitation of stroke patients.

In ASD children the effect of auditory rhythmic cueing on gross motor skills was assessed by El Shemy and El-Sayed (2018). A RAS intervention protocol was used, this consisted of gait training using a metronome to match the child's step pattern. Significant improvements were found in all four subtests of the BOT-2 test compared to a conventional physical therapy program. The results look promising but more research is needed for providing a practice guideline.

Additionally, further research is needed for determining the psychometric properties of the BAASTA and tapping tasks for evaluating rhythm synchronization in a child population.

5.1 Limitations and strengths

For this systematic review both authors performed the literature search and data analysis, therefore no blinding could be done. This makes the review susceptible to biases like confirmation bias. Another possible bias of this systematic review is selection bias. Although a systematic search was performed, there is a chance that not all useful studies are included because of the many synonyms for rhythm perception and auditory motor coupling. To minimize this risk as many synonyms as possible were included in the search strategy. Generalizability of the results is limited, not many studies reported their generalizability, only few articles were used for data analysis per pathology and many studies used 'ADHD like characteristics' or 'probable DCD' or 'risk of DCD'. Therefore, careful interpretation is needed for all pathologies. Moreover, this systematic review does not provide new insights but gives a summary of existing research.

A distinct strength of this review is that an overview of the whole literature is given and their shortcomings, uncertainties and contradictions are discussed.

6. Conclusion

Rhythm perception is essential for accurately performing auditory motor coupling tasks. DCD and ADHD children have an impaired rhythm perception and/or auditory motor synchronization, no differences were found for ASD children. Various possibilities exist for measuring auditory motor coupling and rhythm perception in children, the MBEMA test was considered to be a valid and reliable method for assessing rhythm perception, whereas tapping tasks and the BAASTA test were considered applicable for measuring auditory motor coupling.

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8. Appendix

8.1 Appendix A

Search strategy Pubmed and WOS

	Key words in Web of Science	Hits February 2021	Hits May 2021
#1	AB= (autism spectrum disorder OR autism OR attention deficit hyperactivity disorder OR ADHD OR developmental coordination disorder OR DCD OR developmental dyspraxia OR motor skills disorder OR perceptual-motor disorder OR pediatric OR school-aged children OR typically developing children)	271.640	279.907
#2	AB=(auditory motor coupling OR rhythm perception OR motor timing OR sensorimotor synchronization OR synchronization OR auditory perceptual timing OR auditory-motor OR perceptual timing OR beat perception OR rhythmic auditory stimulation OR sensorimotor coordination OR sensorimotor coupling)	87.737	89.489
#3	TI=("autism spectrum disorder" OR autism OR "attention deficit hyperactivity disorder" OR ADHD OR "developmental coordination disorder" OR DCD OR "developmental dyspraxia" OR "motor skills disorder" OR "perceptual-motor disorder" OR pediatric OR "school-aged children" OR "typically developing children")	247.954	253.505
#4	TI=("auditory motor coupling"OR "rhythm perception" OR "motor timing" OR "sensorimotor synchronization" OR synchronization OR "auditory perceptual timing" OR "auditory-motor" OR "perceptual timing" OR "beat perception" OR "rhythmic auditory stimulation" OR "sensorimotor coordination" OR "sensorimotor coupling")	39.707	40.366
#5	TS=("autism spectrum disorder" OR autism OR "attention deficit hyperactivity disorder" OR ADHD OR "developmental coordination disorder" OR DCD OR "developmental dyspraxia" OR "motor skills disorder" OR "perceptual-motor disorder" OR pediatric OR "school-aged children" OR "typically developing children")	462.978	473.833
#6	TS=("auditory motor coupling"OR "rhythm perception" OR "motor timing" OR "sensorimotor synchronization" OR synchronization OR "auditory perceptual timing" OR "auditory-motor" OR "perceptual timing" OR "beat perception" OR "rhythmic auditory stimulation" OR "sensorimotor coordination" OR "sensorimotor coupling")	129.263	131.496
#7	#1 AND #2	332	344
#8	#3 AND #4	62	63
#9	#5 AND #6	664	685
#10	#7 OR #8 OR #9	664	685
#11	#10 NOT AB=(sleep OR speech OR literacy OR stutter OR dyslexia OR "cerebral palsy") AND LANGUAGE: (English)	652	673

	Key words in Pubmed	Hits February 2021	Hits May 2021
#1	'autism spectrum disorder'[MeSH Terms] OR 'autism spectrum disorder'[Title/Abstract] OR autism[Title/Abstract] OR 'attention deficit disorder with hyperactivity'[MeSH Terms] OR 'attention deficit hyperactivity disorder'[Title/Abstract] OR 'ADHD'[Title/Abstract] OR 'developmental coordination disorder'[MeSH Terms] OR 'developmental coordination disorder'[Title/Abstract] OR DCD[Title/Abstract] OR 'developmental dyspraxia'[Title/Abstract] OR 'motor skills disorder'[Title/Abstract] OR 'motor skills disorder'[MeSH Terms] OR ’perceptual-motor disorder’[Title/Abstract] OR 'pediatric'[Title/Abstract] OR ‘school-aged children’[Title/Abstract] OR ‘typically developing children’[Title/Abstract]	385.409	394.996
#2	'auditory motor coupling'[Title/Abstract] OR 'rhythm perception'[Title/Abstract] OR 'motor timing'[Title/Abstract] OR 'sensorimotor synchronization'[Title/Abstract] OR 'synchronization'[Title/Abstract] OR 'auditory-perceptual timing'[Title/Abstract] OR 'auditory-motor'[Title/Abstract] OR 'perceptual timing'[Title/Abstract] OR 'beat perception'[Title/Abstract] OR 'rhythmic auditory stimulation'[Title/Abstract] OR 'sensorimotor coordination'[Title/Abstract] OR ‘sensorimotor coupling’[Title/Abstract]	25.270	25.711
#3	#1 AND #2	359	372
#4	#3 NOT (Sleep[Title/Abstract] OR Speech[Title/Abstract] OR Literacy[Title/Abstract] OR Stutter[Title/Abstract] OR Dyslexia[Title/Abstract] OR ‘Cerebral Palsy’[Title/Abstract])	304	315
#5	#4 Filter Language: English	299	310

8.2 Appendix B

Overview of the strength and weaknesses analysis

Article	Strengths	Weaknesses
Jamey et al. (2019)	Well defined hypothesis and aim of the study Clear in- and exclusion criteria Use of test batteries or scales for outcome measurements Avoiding confounding bias Statistical analysis adjusted for multicollinearity	Selection bias (rekrutering) No reported power calculation No reported blinding
Tryfon et al. (2017)	Exclusion criteria well defined well defined hypothesis and aim of the study Task adapted to study population	Inclusion criteria not wel defined: does not report age range Performance on the tapping task has not yet been well defined, no norm values. No reported blinding selection bias (lack of reporting recruitment method) No reported adjustment for confounders No reporting of external validity No reported power calculation
Lesiuk (2015)	Well defined hypothesis and aim of the study Usage of a validated test battery and scale for outcome measures Good external validity	No clear defined in- and exclusion criteria No reported blinding Selection bias (lack of reporting recruitment method) No reported adjustment for confounders No reported power calculation
Chang et al. (2021)	Well defined hypothesis and aims of the study Clear in- and exclusion criteria Reporting of time of recruitment subgroups of DCD+ADHD (common recurring comorbidities) Adjustment for confounders in method and statistical analysis Double blinding	Selection bias (lack of reporting recruitment method) No reported validity of the tests used No reporting of external validity No reported power, instead reported limited power
Puyjarinet et al. (2017)	Well defined hypothesis and aim of the study Clear in- and exclusion criteria Usage of a validated test battery Adjustment for confounders in method and statistical analysis	No reported age range Selection bias (lack of reporting recruitment method) No reported blinding No reported power calculation.

Article	Strengths	Weaknesses
Khalil et al. (2013)	Well defined hypothesis and aim of the study usage of validated test for assessing ADHD symptoms and behavior adjustment for confounders in study method Accounted for the extreme data bias/outliers.	No clear in- and exclusion criteria No clear reported age range No reported validity of instruments used to asses beat synchrony Selection bias (lack of reporting recruitment method) No reported blinding
Y. Y. Chen et al. (2013)	Well defined hypothesis and aim of the study Adjustment of confounders in the study method	Small study sample No clear in- and exclusion criteria No reported age range No reported validity of the test used Selection bias (lack of reporting recruitment method) No reported blinding No reported power calculation
Gaul and Issartel (2018)	Well defined hypothesis and aim of the study	No clear in- and exclusion criteria No reported age range No reported validity of the tests used No reported adjustment for confounders Selection bias (lack of reporting recruitment method, righthanded children only) No reported blinding No reported power calculation
Roche et al. (2016)	Well defined hypothesis and aim of the study Adjustment for confounder: age and gender matched controls - auditory threshold clear in- and exclusion criteria usage of valid tests for participants' characteristics	Selection bias No reported validity of tapping task No reported blinding No reported power calculation
Whitall et al. (2006)	Well defined hypothesis and aim of the study Clear in- and exclusion criteria Usage of valid test for participants' characteristics Age and gender matched control group	Small study sample No reported validity of synchronization task Selection bias No reported blinding No reported adjustment for confounders Usage of a double gross motor task No reported power calculation

Article	Strengths	Weaknesses

Whitall et al. (2008)	Well defined hypothesis and aim of the study Age and gender matched controls Clear in- and exclusion criteria	Small study sample No reported validity of the tapping task No reported blinding Selection bias No reported adjustment for confounders Reported power influenced by small study sample
Rosenblum and Regev (2013)	Well defined hypothesis and aim of the study Age and gender matched controls Clear in- and exclusion criteria Usage of a valid test battery	No reported blinding Selection Bias No reported adjustment for confounders No reported blinding No reported power calculation
Kagerer and Clark (2015)	Well defined hypothesis and aim of the study Clear inclusion criteria and patients' characteristics	No clear exclusion criteria No reported validity of the synchronization task Selection bias No reported adjustment for confounders No reported blinding No reported power calculation

8.3 Appendix C

Downs and Black checklists per pathology

TD children

Gaul and Issartel (2018)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	No = 0
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	No = 0
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0

12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		
15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Unable to determine = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	No = 0

Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 9/19

Kagerer and Clark (2015)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	No = 0
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Unable to determine = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Unable to determine = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 10/19

ASD children

Jamey et al. (2019)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0

Internal validity - bias		
15	*Was an attempt made to blind investigators and assessors*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Yes = 1
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 13/19

Tryfon et al. (2017)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind those measuring the main outcomes of the intervention?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Unable to determine = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 12/19

ADHD children

Y. Y. Chen et al. (2013)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	No = 0
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Yes = 2
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0

Internal validity - bias		
15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	No = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Yes = 1
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 12/19

Khalil et al. (2013)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	No = 0
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	No = 0
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	No = 0
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Yes = 1
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Yes = 1
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	No = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Yes = 1
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Unable to determine = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 12/19

Lesiuk (2015)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	No = 0
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Yes = 1
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Yes = 1
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Yes = 1
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 14/19

Puyjarinet et al. (2017)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	No = 0
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Unable to determine = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 11/19

DCD children

Chang et al. (2021)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	No = 0
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Yes = 1
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes = 1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Yes = 1
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Unable to determine = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 13/19

Roche et al. (2016)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	No = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	Yes = 1
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 12/19

Rosenblum and Regev (2013)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Yes =1
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	No = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 12/19

Whitall et al. (2006)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1
5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially = 1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	No = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	No = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 11/19

Whitall et al. (2008)

Item	Criteria	Possible Answers
Reporting		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1
2	*Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.	Yes = 1
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.	Yes = 1
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5	*Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.	Partially =1
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests which are considered below).	Yes = 1
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the interquartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
10	*Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1
External validity		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.	Unable to determine = 0
12	*Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.	Unable to determine = 0
Internal validity - bias		

15	*Was an attempt made to blind the investigators and assessors?*	Unable to determine = 0
16	*If any of the results of the study were based on "data dredging", was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.	Yes = 1
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the question should be answered yes.	Yes = 1
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrates the outcome measures are accurate, the question should be answered as yes.	Yes = 1
Internal validity - confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case-control studies where there is no information concerning the source of patients included in the study.	Unable to determine = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.	Unable to determine = 0
25	*Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusions of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomized studies if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.	No = 0
Power		
27*	*Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?* Sample sizes have been calculated to detect a difference of x% and y%.	Unable to determine = 0

*Item has been modified.

Score = 11/19

8.4 Appendix D

E-mails sent to first authors

Dear Prof. R. Roche

On Web Of Knowledge we have read the abstract of your papers:

- Roche, R., Horn, C., Chang, T. Y., Viswanathan, P. and Whitall, J. 2004 Auditory motor processing in Typically developing children: a cross-sectional study Journal of Sport & Exercise Psychology
- Roche, R., Viswanathan, P., Clark, J. E. and Whitall, J. 2010 Auditory motor adaptation in children with developmental coordination disorder Journal of Sport & Exercise Psychology

We are working on our master thesis entitled: auditory motor coupling and rhythm synchronization in children with Attention Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD) and Developmental Coordination Disorder (DCD).
I'm very interested to read the full text of your papers. You would do me a great favour if you could send it to me by email.

Yours sincerely,
Annelise Vos and Janique Roufs
Hasselt University
Faculty of Rehabilitation sciences and Physiotherapy
Belgium

Dear Prof. T. Lesuik

On Web Of Knowledge we have read the abstract of your paper:
Lesiuk, T. (2015). Music perception ability of children with executive function deficits. Psychology of Music, 43(4), 530-544. doi:10.1177/0305735614522681

We are working on our master thesis entitled: auditory motor coupling and rhythm synchronization in children with Attention Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD) and Developmental Coordination Disorder (DCD).
I'm very interested to read the full text of your paper. You would do me a great favour if you could send it to me by email.

Yours sincerely,
Annelise Vos and Janique Roufs
Hasselt University
Faculty of Rehabilitation sciences and Physiotherapy
Belgium

Dear Prof. C. Morimoto,

On Web Of Knowledge we have read the abstract of your paper:
Morimoto, C. N., S.: Kobori, S.: Kaneko, F.: Muratake, S.: Okamura, H. (2020). Easy assessment of individuals with various severities of autism spectrum disorder, focusing on motor timing control. Research in Autism Spectrum Disorders, 79. doi:10.1016/j.rasd.2020.101682

We are working on our master thesis entitled: auditory motor coupling and rhythm synchronization in children with Attention Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD) and Developmental Coordination Disorder (DCD).
I'm very interested to read the full text of your paper. You would do us a great favour if you could send it to me by email.

Yours sincerely,
Annelise Vos and Janique Roufs
Hasselt University
Faculty of Rehabilitation sciences and Physiotherapy
Belgium

Dear Prof. B. Murphy,
On Pubmed we have read the abstract of your paper:
Hession, C. E., Law Smith, M. J., Watterson, D., Oxley, N., & Murphy, B. A. (2019). The Impact of Equine Therapy and an Audio-Visual Approach Emphasizing Rhythm and Beat Perception in Children with Developmental Coordination Disorder. *Journal of alternative and complementary medicine (New York, N.Y.)*, *25*(5), 535–541. https://doi.org/10.1089/acm.2017.0242
We are working on our master thesis entitled: auditory motor coupling and rhythm synchronization in children with Attention Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD) and Developmental Coordination Disorder (DCD).
I'm very interested to read the full text of your paper. You would do us a great favour if you could send it to me by email.
Yours sincerely,
Annelise Vos and Janique Roufs
Hasselt University
Faculty of Rehabilitation sciences and Physiotherapy
Belgium

Dear Prof. R. Sartori

On Pubmed we have read the abstract of your paper:
Sartori, R. F., Valentini, N. C., Nobre, G. C. and Fonseca, R. P. 2020 Motor and verbal inhibitory control: development and validity of the go/No-Go app test for children with development coordination disorder Appl Neuropsychol Child

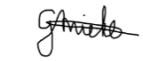
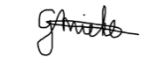
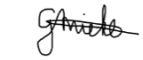
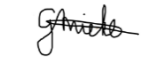
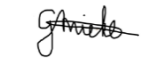
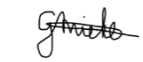
We are working on our master thesis entitled: auditory motor coupling and rhythm synchronization in children with Attention Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD) and Developmental Coordination Disorder (DCD).
I'm very interested to read the full text of your paper. You would do us a great favour if you could send it to me by email.

Yours sincerely,
Annelise Vos and Janique Roufs
Hasselt University
Faculty of Rehabilitation sciences and Physiotherapy
Belgium

8.5 Appendix E

Progress form and self-evaluation

VOORTGANGSFORMULIER WETENSCHAPPELIJKE STAGE DEEL 1

DATUM	INHOUD OVERLEG	HANDTEKENINGEN
04-11-20	Bespreking start van de masterproef, overlopen van de contracten. Afspraken: doorsturen van planning, laten weten wanneer periodes stagevrij zijn. Eind deze maand een gesprek plannen om scooping, planning en specifieke onderzoeksvraag/zoekstrategie overlopen (4-12 13:00-14:00)	Promotor: Eugene Rameckers Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs
04-12-20	Situering van de masterproef besproken. Beginnende zoekstrategie besproken. Enkele struikelblokken aangekaart met betrekking tot verandering in stagevrije periodes. Afspraken: nieuwe planning doorsturen, mailen indien de volledige zoekstrategie gevonden is.	Promotor: / Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs
23-2-21	Zoekstrategie en geïncludeerde artikels besproken. Plan van aanpak voor kwaliteitsbeoordeling overlopen. Afspraken: uitvoeren van kwaliteitsbeoordeling, maken van een sterkte- en zwakteanalyse en initiële data-extractie uitvoeren	Promotor: / Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs
22-3-21	Geïncludeerde artikels, uitgevoerde kwaliteitsbeoordeling en data-extractie overlopen. Overleg over inclusie van studies met lage kwaliteit. Deze bleken nuttig genoeg om te includeren maar oppassen met interpretatie	Promotor: / Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs
5-5-21	Overlopen plan van aanpak voor de discussie, evaluatie van de geschreven delen van de masterproef en overlopen van het protocol.	Promotor: / Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs
4-6-21	**<u>Niet-bindend advies:</u>** De promotor verleent hierbij het advies om de masterproef WEL te verdedigen.	Promotor: Eugene Rameckers Copromotor: Mieke Goetschalckx  Student(e): Annelise Vos Student(e): Janique Roufs

BEOORDELING VAN DE WETENSCHAPPELIJKE STAGE-DEEL 1

Wetenschappelijke stage deel 1 (Masterproef deel 1- MP1) van de Master of Science in de revalidatiewetenschappen en de kinesitherapie bestaat uit **twee delen**:

1) De literatuurstudie volgens een welomschreven methodiek.
2) Het opstellen van het onderzoeksprotocol ter voorbereiding van masterproef deel 2.

Omschrijving van de **evaluatie**:

1) 80% van het eindcijfer wordt door de promotor in samenspraak met de copromotor gegeven op grond het product en van het proces dat de student doorliep om de MP1 te realiseren, met name het zelfstandig uitvoeren van de literatuurstudie en het zelfstandig opstellen van het onderzoeksprotocol, alsook de kwaliteit van academisch schrijven.
2) 20% van het eindcijfer wordt door de interne jury gegeven op grond van het ingeleverde product en de mondelinge presentatie waarin de student zijn/haar proces toelicht.

In de beoordeling dient onderscheid gemaakt te worden tussen studenten die, in samenspraak met de promotor, een nieuw onderzoek uitwerkten en studenten die instapten in een lopend onderzoek of zich baseren op voorgaande masterproeven of onderzoeksprojecten. Van deze laatste worden bijkomende inspanningen verwacht zoals bv. het bijsturen van de eerder geformuleerde onderzoeksvraag, de kritische reflectie over het onderzoeksdesign, het uitvoeren van een pilotexperiment.

Beoordelingskader:

Beoordelingskader: criteria op 20	
18-20	Excellente modelmasterproef
16-17	Zeer goede masterproef
14-15	Goede masterproef
12-13	Voldoende masterproef
10-11	Zwakke masterproef
≤ 9	Onvoldoende masterproef die niet aan de minimumnormen voldoet

Onderstaand zelfevaluatierapport is een hulpmiddel om je wetenschappelijke stage -deel 1 zelfstandig te organiseren. Bepaal zelf je deadlines, evalueer en reflecteer over je werkwijze en over de diepgang van je werk. Check de deadlines regelmatig. Toets ze eventueel af bij je (co)promotor. Succes!

ZELFEVALUATIERAPPORT WETENSCHAPPELIJKE STAGE - DEEL 1 RWK

LITERATUURSTUDIE	Gestelde deadline	Behaald op	Reflectie
De belangrijkste concepten en conceptuele kaders van het onderzoekdomein uitdiepen en verwerken	11/11/2020	11/11/2020	Voldoende
De belangrijkste informatie opzoeken als inleiding op de onderzoeksvraag van de literatuurstudie	11/11/2020	11/11/2020	Voldoende
De opzoekbare onderzoeksvraag identificeren en helder formuleren in functie van de literatuurstudie	NVT	NVT	NVT
De zoekstrategie op systematische wijze uitvoeren in relevante databanken	21/12/2020	09/02/2021	Niet gehaald omwille van stage
De kwaliteitsbeoordeling van de artikels diepgaand uitvoeren	26/02/2021	24/02/2021	Voldoende
De data-extractie grondig uitvoeren	26/03/2021	23/03/2021	Voldoende
De bevindingen ïntegreren tot een synthese	30/04/2021	25/05/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage

ONDERZOEKSPROTOCOL	Gestelde deadline	Behaald op	Reflectie
De onderzoeksvraag in functie van het onderzoeksprotocol identificeren	16/04/2021	2/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
Het onderzoeksdesign bepalen en/of kritisch reflecteren over bestaande onderzoeksdesign	16/04/2021	2/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
De methodesectie (participanten, interventie, uitkomstmaten, data-analyse) uitwerken	16/04/2021	2/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage

ACADEMISCHE SCHRIJVEN	Gestelde deadline	Behaald op	Reflectie
Het abstract to the point schrijven	07/05/2021	02/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
De inleiding van de literatuurstudie logisch opbouwen	16/04/2021	28/04/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
De methodesectie van de literatuurstudie transparant weergegeven	26/03/2021	23/03/2021	Voldoende
De resultatensectie afstemmen op de onderzoeksvragen	30/04/2021	25/05/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
In de discussiesectie de bekomen resultaten in een wetenschappelijke tekst integreren en synthetiseren	30/04/2021	25/05/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage

Het onderzoeksprotocol deskundig technisch uitschrijven	16/04/2021	02/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage
Referenties correct en volledig weergeven	07/05/2021	02/06/2021	Niet gehaald omwille van verhoogde werkdruk door groepswerken en stage

ZELFSTUREND EN WETENSCHAPPELIJK DENKEN EN HANDELEN	**Aanvangsfase**	**Tussentijdse fase**	**Eindfase**
Een realistische planning opmaken, deadlines stellen en opvolgen	Verliep vlot	Verliep moeizaam	Verliep moeizaam
Initiatief en verantwoordelijkheid opnemen ten aanzien van de realisatie van de wetenschappelijke stage	Verliep vlot	Verliep vlot	Verliep vlot
Kritisch wetenschappelijk denken	Verliep vlot	Verliep vlot	Verliep vlot
De contacten met de promotor voorbereiden en efficiënt benutten	Verliep vlot	Verliep vlot	Verliep vlot
De richtlijnen van de wetenschappelijke stage autonoom opvolgen en toepassen	Verliep moeizaam	Verliep vlot	Verliep moeizaam
De communicatie met de medestudent helder en transparant voeren	Verliep vlot	Verliep vlot	Verliep vlot
De communicatie met de promotor/copromotor helder en transparant voeren	Verliep vlot	Verliep vlot	Verliep vlot
Andere verdiensten:	/	/	/

8.6 Appendix F

Approved request for defending the master thesis

Eugene RAMECKERS
aan Annelise, mij, Mieke

Dag Anneliese en Janique

hierbij de benodigde getekende formulieren.

ook een paar feedback punten in de thesis en protocol.

goed geschreven en helder lezend.
ik mis de motivatie van de keuzes in beide stukken wel eens

maar goed geschreven\

gr eugene
Prof. Dr. Eugene Rameckers
Pediatric Rehabilitation
Faculty Rehabilitation Science.
+32(0)11269305 Mobiel +31641671909
www.uhasselt.be
Hasselt University - campus Diepenbeek
Agoralaan Gebouw A B-3590 Diepenbeek

Op do 3 jun. 2021 om 22:49 schreef Annelise Vos <annelise.vos@student.uhasselt.be>:

Inschrijvingsformulier verdediging masterproef academiejaar 2020-2021,
Registration form jury Master's thesis academic year 2020-2021,

GEGEVENS STUDENT - *INFORMATION STUDENT*

Faculteit/School: **Faculteit Revalidatiewetenschappen**
Faculty/School: ***Rehabilitation Sciences***

Stamnummer + naam: **1745938 Roufs Janique**
Student number + name

Opleiding/*Programme*: **1 ma revalid. wet. & kine**

INSTRUCTIES - *INSTRUCTIONS*

Neem onderstaande informatie grondig door.

Print dit document en vul het aan met DRUKLETTERS.

In tijden van van online onderwijs door COVID-19 verstuur je het document (scan of leesbare foto) ingevuld via mail naar je promotor. Je promotor bezorgt het aan de juiste dienst voor verdere afhandeling.

Vul luik A aan. Bezorg het formulier aan je promotoren voor de aanvullingen in luik B. Zorg dat het formulier ondertekend en gedateerd wordt door jezelf en je promotoren in luik D en dien het in bij de juiste dienst volgens de afspraken in jouw opleiding.
Zonder dit inschrijvingsformulier krijg je geen toegang tot upload/verdediging van je masterproef.

Please read the information below carefully.

Print this document and complete it by hand writing, using CAPITAL LETTERS.

In times of COVID-19 and during the online courses you send the document (scan or readable photo) by email to your supervisor. Your supervisor delivers the document to the appropriate department.

Fill out part A. Send the form to your supervisors for the additions in part B. Make sure that the form is signed and dated by yourself and your supervisors in part D and submit it to the appropriate department in accordance with the agreements in your study programme.
Without this registration form, you will not have access to the upload/defense of your master's thesis.

LUIK A - VERPLICHT - IN TE VULLEN DOOR DE STUDENT
PART A - MANDATORY - TO BE FILLED OUT BY THE STUDENT

Titel van Masterproef/*Title of Master's thesis:* AUDITORY MOTOR COUPLING AND *

X behouden - *keep*

O wijzigen - *change to*:

* RHYTM PERCEPTION IN TYPICALLY DEVELOPING CHILDREN, CHILDREN WITH AUTISM SPECTRUM DISORDER, ATTENTION DEFICIT HYPERACTIVITY DISORDER..

AND DEVELOPMENTAL COORDINATION DISORDER

/:

O behouden - *keep*

O wijzigen - *change to*:

In geval van samenwerking tussen studenten, naam van de medestudent(en)/*In case of group work, name of fellow student(s)*: ANNELISE VOS

☒ behouden - *keep*

O wijzigen - *change to:*

LUIK B - VERPLICHT - IN TE VULLEN DOOR DE PROMOTOR(EN)
PART B - MANDATORY - TO BE FILLED OUT BY THE SUPERVISOR(S)

Wijziging gegevens masterproef in luik A/*Change information Master's thesis in part A:*

O goedgekeurd - *approved*

O goedgekeurd mits wijziging van - *approved if modification of:*

Scriptie/*Thesis:*

O openbaar (beschikbaar in de document server van de universiteit)- *public (available in document server of university)*

O vertrouwelijk (niet beschikbaar in de document server van de universiteit) - *confidential (not available in document server of university)*

Juryverdediging/*Jury Defense:*

De promotor(en) geeft (geven) de student(en) het niet-bindend advies om de bovenvermelde masterproef in de bovenvermelde periode/*The supervisor(s) give(s) the student(s) the non-binding advice:*

O te verdedigen/*to defend the aforementioned Master's thesis within the aforementioned period of time*

O de verdediging is openbaar/*in public*

O de verdediging is niet openbaar/*not in public*

O niet te verdedigen/*not to defend the aforementioned Master's thesis within the aforementioned period of time*

LUIK C - OPTIONEEL - IN TE VULLEN DOOR STUDENT, alleen als hij luik B wil overrulen
PART C - OPTIONAL - TO BE FILLED OUT BY THE STUDENT, only if he wants to overrule part B

In tegenstelling tot het niet-bindend advies van de promotor(en) wenst de student de bovenvermelde masterproef in de bovenvermelde periode/*In contrast to the non-binding advice put forward by the supervisor(s), the student wishes:*

O niet te verdedigen/*not to defend the aforementioned Master's thesis within the aforementioned period of time*

O te verdedigen/*to defend the aforementioned Master's thesis within the aforementioned period of time*

LUIK D - VERPLICHT - IN TE VULLEN DOOR DE STUDENT EN DE PROMOTOR(EN)
PART D - MANDATORY - TO BE FILLED OUT BY THE STUDENT AND THE SUPERVISOR(S)

Datum en handtekening student(en)
Date and signature student(s)

03-06-2021

Datum en handtekening promotor(en)
Date and signature supervisor(s)

04-06-2021

eugene rameckers

UHASSELT
KNOWLEDGE IN ACTION

Inschrijvingsformulier verdediging masterproef academiejaar 2020-2021,
Registration form jury Master's thesis academic year 2020-2021,

GEGEVENS STUDENT - *INFORMATION STUDENT*

Faculteit/School: **Faculteit Revalidatiewetenschappen**
Faculty/School: ***Rehabilitation Sciences***

Stamnummer + naam: **1747198 Vos Annelise**
Student number + name

Opleiding/*Programme*: **1 ma revalid. wet. & kine**

INSTRUCTIES - *INSTRUCTIONS*

Neem onderstaande informatie grondig door.

Print dit document en vul het aan met DRUKLETTERS.

In tijden van van online onderwijs door COVID-19 verstuur je het document (scan of leesbare foto) ingevuld via mail naar je promotor. Je promotor bezorgt het aan de juiste dienst voor verdere afhandeling.

Vul luik A aan. Bezorg het formulier aan je promotoren voor de aanvullingen in luik B. Zorg dat het formulier ondertekend en gedateerd wordt door jezelf en je promotoren in luik D en dien het in bij de juiste dienst volgens de afspraken in jouw opleiding.
Zonder dit inschrijvingsformulier krijg je geen toegang tot upload/verdediging van je masterproef.

Please read the information below carefully.

Print this document and complete it by hand writing, using CAPITAL LETTERS.

In times of COVID-19 and during the online courses you send the document (scan or readable photo) by email to your supervisor. Your supervisor delivers the document to the appropriate department.

Fill out part A. Send the form to your supervisors for the additions in part B. Make sure that the form is signed and dated by yourself and your supervisors in part D and submit it to the appropriate department in accordance with the agreements in your study programme.
Without this registration form, you will not have access to the upload/defense of your master's thesis.

LUIK A - VERPLICHT - IN TE VULLEN DOOR DE STUDENT
PART A - MANDATORY - TO BE FILLED OUT BY THE STUDENT

Titel van Masterproef/*Title of Master's thesis:* AUDITORY MOTOR COUPLING AND RHYTHM PERCEPTION IN TYPICALLY DEVELOPING CHILDREN, CHILDREN WITH AUTISM SPECTRUM DISORDER, ATTENTION DEFICIT HYPERACTIVITY DISORDER AND DEVELOPMENTAL COORDINATION DISORDER

☒ behouden - *keep*

O wijzigen - *change to*:

/:

O behouden - *keep*

O wijzigen - *change to*:

In geval van samenwerking tussen studenten, naam van de medestudent(en)/*In case of group work, name of fellow student(s)*: JANIQUE ROUFS

☑ behouden - *keep*

O wijzigen - *change to:*

LUIK B - VERPLICHT - IN TE VULLEN DOOR DE PROMOTOR(EN)
PART B - MANDATORY - TO BE FILLED OUT BY THE SUPERVISOR(S)

Wijziging gegevens masterproef in luik A/*Change information Master's thesis in part A:*

O goedgekeurd - *approved*

O goedgekeurd mits wijziging van - *approved if modification of:*

Scriptie/*Thesis:*

O openbaar (beschikbaar in de document server van de universiteit)- *public (available in document server of university)*

O vertrouwelijk (niet beschikbaar in de document server van de universiteit) - *confidential (not available in document server of university)*

Juryverdediging/*Jury Defense:*

De promotor(en) geeft (geven) de student(en) het niet-bindend advies om de bovenvermelde masterproef in de bovenvermelde periode/*The supervisor(s) give(s) the student(s) the non-binding advice:*

O te verdedigen/*to defend the aforementioned Master's thesis within the aforementioned period of time*

O de verdediging is openbaar/*in public*

O de verdediging is niet openbaar/*not in public*

O niet te verdedigen/*not to defend the aforementioned Master's thesis within the aforementioned period of time*

LUIK C - OPTIONEEL - IN TE VULLEN DOOR STUDENT, alleen als hij luik B wil overrulen
PART C - OPTIONAL - TO BE FILLED OUT BY THE STUDENT, only if he wants to overrule part B

In tegenstelling tot het niet-bindend advies van de promotor(en) wenst de student de bovenvermelde masterproef in de bovenvermelde periode/*In contrast to the non-binding advice put forward by the supervisor(s), the student wishes:*

O niet te verdedigen/*not to defend the aforementioned Master's thesis within the aforementioned period of time*

O te verdedigen/*to defend the aforementioned Master's thesis within the aforementioned period of time*

LUIK D - VERPLICHT - IN TE VULLEN DOOR DE STUDENT EN DE PROMOTOR(EN)
PART D - MANDATORY - TO BE FILLED OUT BY THE STUDENT AND THE SUPERVISOR(S)

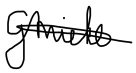
Datum en handtekening student(en)
Date and signature student(s)

31061 2021

Datum en handtekening promotor(en)
Date and signature supervisor(s)

04-06-2021

eugene rameckers



Part 2: Researchprotocol

Content

1. Introduction

Auditory motor coupling and rhythm perception are important in children's daily living. The main part of a good auditory motor coupling is based on a good functioning auditory timing perception, which makes it possible to coordinate limbs relative to each other and to adapt movements to the external environment or task demands. To be able to adapt, plan or automatize movements, a decent (timing) prediction must be made and one must learn from errors (Trainor, Chang, Cairney, & Li, 2018). One can speak about sensory predictions, these exist when a person extracts regularities from an upcoming event or context or from memory- or motor predictions, based on the internal modeling system. The theory behind the internal modeling system states that inverse models are used in combination with forward models. The inverse model generates a motor command based on the desired motor outcomes. On the contrary, the forward model predicts the desired motor outcome used by the inverse model. When both models are used together, an efference copy of the motor command output from the inverse model can be used as an input to a following forward model for further predictions. This results in rapid online motor control. Sensory and motor predictions work together simultaneously for sensorimotor synchronization, of which auditory motor coupling is a part of. Sensory predictions guide the inverse model, hereby guiding the internal modeling system. At last, perceived errors in motor or sensory domains are used to update the sensory and motor predictions.

Rhythm perception is expressed as the ability to recognize and process a musical rhythm. Auditory motor coupling is the integration of an auditory stimulus and the motor output, by using the internal modeling system and sensory predictions. For accurate coupling the ability to adapt motor performance to the auditory cue is necessary. Synchronizing and coupling movements to auditory cues is a difficult task, limbs are instructed to move at the same frequency as the stimulus and need to 'frequency lock'. During the entire task the limbs are expected to synchronize the movements with the beat. In order to achieve this, the limbs must additionally 'phase lock' (Getchell, 2007; Sternad, Dean, & Schaal, 2000). This occurs naturally in children during a dual motor task (Getchell, McMenamin, & Whitall, 2005; Getchell & Whitall, 2003; Sternad et al., 2000). Auditory motor coupling is known to improve and become less variable with aging (Drewing, Aschersleben, & Li, 2006; McAuley, Jones, Holub, Johnston, & Miller, 2006).

Recent work has shown that some child disorders have an affected rhythm perception and/or auditory motor timing that can affect their motor performance (Chang et al., 2021; Y. Y. Chen et al., 2013; Gaul & Issartel, 2018; Kagerer & Clark, 2015; Khalil, Minces, McLoughlin, & Chiba, 2013; Lesiuk, 2015; Puyjarinet, Bégel, Lopez, Dellacherie, & Dalla Bella, 2017; Rosenblum & Regev, 2013; Trainor et al., 2018; Whitall et al., 2008; Whitall et al., 2006). Because rhythmic synchronization improves entrainment and performance on coordination tasks (Getchell, 2007; M. Thaut, Tian, & Azimi-Sadjadi, 1998; M. H. Thaut, McIntosh, Prassas, & Rice, 1992), there is an increased interest in research of this subject. Children with Developmental Coordination Disorder (DCD) typically have impairments in motor performance and show higher variability in movement patterns, not explained by any neurological condition or IQ (American Psychiatric Association, 2013). Trainor et al. (2018) proposed that auditory perceptual timing deficits might be a core symptom of DCD, leading to higher variability in movement patterns. Secondly, children with Attention-Deficit Hyperactivity Disorder (ADHD) also show difficulty with motor coordination, furthermore slow reaction time is presented (Kaiser, Schoemaker, Albaret, & Geuze, 2015). Motor problems occur in 30-50% of children with ADHD (Fliers et al., 2009; Gillberg et al., 2004; Goulardins, Marques, Casella, Nascimento, & Oliveira, 2013). DCD shows high comorbidity with ADHD (Gomez & Sirigu, 2015) and both show deficits in auditory perceptual and sensorimotor timing (Noreika, Falter, & Rubia, 2013). Lastly, 50-85% of children with Autism Spectrum Disorder (ASD) also report impairments in motor performance (Bhat, Landa, & Galloway, 2011; Chukoskie, Townsend, & Westerfield, 2013). Children with ASD have difficulties in gross and fine motor tasks, impaired performance in sequential and imitation based tasks and difficulty coordinating both sides of the body in rhythmic tasks (Kaur, S, & A, 2018). ASD has, equal to ADHD, high comorbidity with DCD (Albajara Sáenz et al., 2021). Research done by El Shemy and El-Sayed (2018) found that including a rhythmic auditory stimulation protocol in gait and gross motor training in ASD children leads to more improvements on the gross motor functioning compared to normal gait and gross motor training. More research is necessary to evaluate the therapeutic impact of rhythmic auditory synchronization training in children with DCD, ADHD and ASD.

Relatively little is known about the impact of a decreased rhythm perception or rhythm synchronization ability on motor performance. Because the previously mentioned disorders all suffer from decreased motor performance, it is interesting to know if there is a link between

a decreased rhythm perception or auditory motor coupling. According to our systematic review children with DCD were proven to be more variable in auditory motor synchronization compared to TD children. Secondly, ADHD children performed worse on rhythm synchronization and perception tasks than TD children. Lastly, no significant differences were found for ASD children compared to TD children. Currently no study has been executed that distinguishes between TD, ADHD, ASD and DCD children. Therefore, the purpose of this study is to distinguish auditory motor coupling and rhythm perception in typically developing children (TD), children with Developmental Coordination Disorder (DCD), children with Attention-Deficit Hyperactivity Disorder (ADHD) and children with Autism Spectrum Disorder (ASD).

2. Purpose

2.1 Research question

The primary research question: “What is the difference between the performance of children with ADHD, DCD or ASD on auditory motor coupling/ rhythm perception tasks, compared to TD children?”

2.2 Hypothesis

For the primary research question:

- Null hypothesis (H_0): “There are no differences in rhythm perception and auditory motor synchronization between TD, ADHD, ASD and DCD children.”
- Alternative hypothesis (H_A): “There are differences in rhythm perception and auditory motor coupling between TD, ADHD, ASD and DCD children.”

If the alternative hypothesis (H_A) is accepted, we will further analyze between which groups these differences exist.

3. Methods

3.1 Research design

A case-controlled blinded research protocol is developed to assess rhythm perception and auditory motor synchronization of children with ASD, DCD and ADHD in comparison with TD children. The participants will be divided into four groups based on their DSM-V diagnosis: namely DCD-group, ADHD-group, ASD-group. Children without any disorder will be grouped as TD group. Participants are mostly aware of their diagnosis and therefore their subgroup, this makes double blindness very hard to achieve. Assessors and statistical analysts will be blinded in this research design.

3.2 Participants

A visualization of the inclusion criteria per subgroup is given in figure 1.

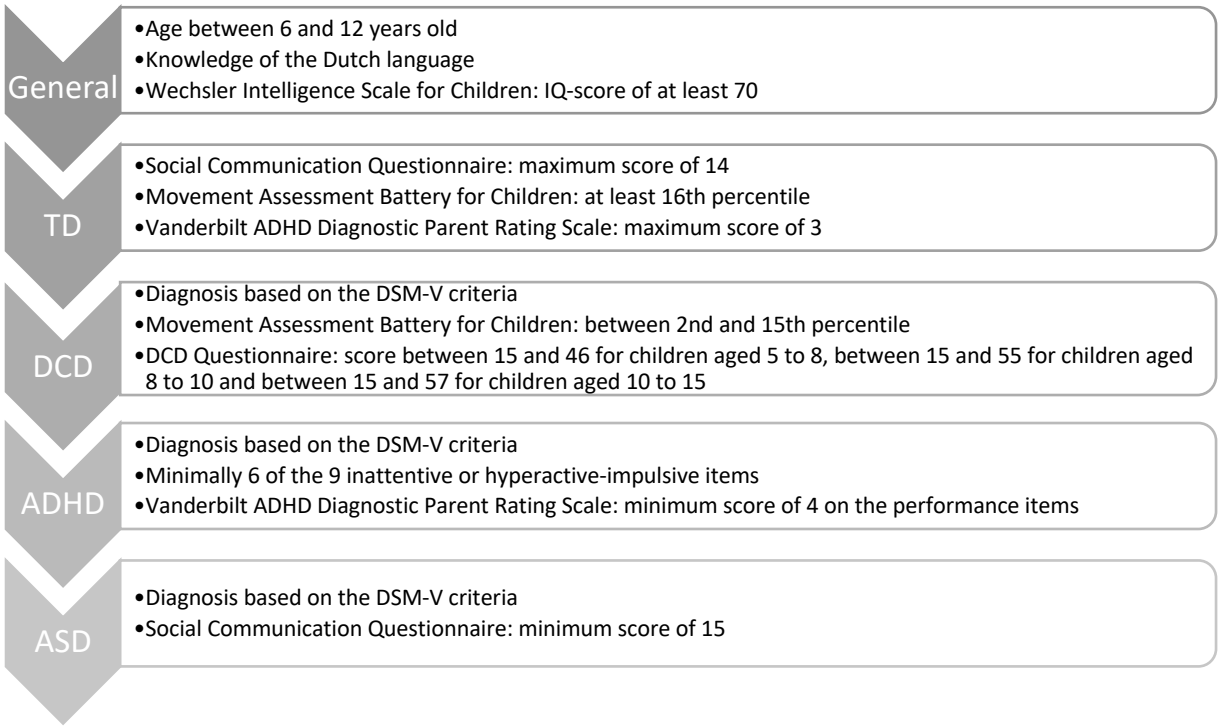


Figure 1. Visualization of inclusion criteria per subgroup, *abbreviations:* Diagnostic and Statistical manual of Mental disorders-5th edition.

3.2.1 Inclusion criteria

Inclusion criteria in this study protocol depends on the subgroup, standard inclusion criteria are (1) children aged between 6 and 12 years old, (2) children with knowledge of the Dutch

language to understand, speak and read instructions and (3) typically developing children or children with the diagnosis of ASD, ADHD or DCD (see 3.2.1.1 – 3.2.1.4).

3.2.1.1 Autism Spectrum Disorder

Inclusion criteria specifically for the ASD subgroup are (1) an (expected) diagnosis of ASD by psychologists or neuropsychiatrists, (2) a diagnosis based on the DSM-V criteria for ASD and (3) a score on the social communication questionnaire with a minimum of 15.

3.2.1.2 Attention-Deficit Hyperactivity Disorder

Inclusion criteria for the ADHD subgroup are (1) an (expected) diagnosis of ADHD by psychologists or neuropsychiatrists, (2) a diagnosis based on the DSM-V criteria for ADHD, (3) a score of at least two or three on six subitems of the nine inattentive or nine hyperactive-impulsive items, or both, and (4) a score of at least four on two subitems or a score of five in one subitem of the performance items of the Vanderbilt ADHD Diagnostic Parent Rating Scale (Becker, Langberg, Vaughn, & Epstein, 2012). This questionnaire is filled in by the children's parents.

3.2.1.3 Developmental Coordination Disorder

Inclusion criteria for the DCD subgroup are (1) an (expected) diagnosis of DCD, (2) a diagnosis based on the DSM-V criteria for DCD, (3) a score on the Movement ABC between the 2nd and 15th percentile and (4) a score between 15 and 46 for children aged 5 to 8, between 15 and 55 for children aged 8 to 10 or between 15 and 57 for children aged 10 to 15 on the DCD Questionnaire.

3.2.1.4 Typically developing children

Inclusion criteria for the TD subgroup are (1) no known neuromuscular disorder, (2) a score on the social communication questionnaire with a maximum of 14, (3) a score lower than four on the Vanderbilt ADHD Diagnostic Rating Scale and (4) a score of at least the 16th percentile on the Movement ABC.

3.2.1.5 Comorbidities

Children with other comorbid disorders than ASD, ADHD and DCD will be excluded from this research. Comorbid ASD, ADHD or DCD is included, but will be analyzed differently depending on the number of children with comorbidities. When less than three included children have

identical comorbid disorders, the comorbidities will be controlled in statistical analysis. A new subgroup will be formed when four children or more with identical comorbidities are included.

3.2.2 Exclusion criteria

Standard exclusion criteria for all subgroups are (1) impaired hearing, (2) an IQ-score lower than 70, measured with the Wechsler Intelligence Scale for Children-V-NL and (3) neurological disorders, musculoskeletal disorders, cardiorespiratory disorders, communication disorders, intellectual developmental disorders or learning disorders.

3.2.3 Recruitment

Participants for this study are recruited from schools all around Flanders. All included children will have to come to the research facilities close to UHasselt (Hasselt). Recruitment methods consist of mouth-to-mouth information and flyers. Physical therapists around Hasselt are asked to refer any children they deem fit to participate in this study. All included children will perform all diagnostic measures to find out in which subgroup they can be included and whether they check all inclusion and exclusion criteria. A pilot study will be performed to quantify the number of children needed in each subgroup to achieve a power of 80%. Tryfon et al. (2017) performed a similar task and included between 23 and 31 children per subgroup. Thus, we expect to include a minimum of ± 20 children per subgroup.

3.3 Medical ethics

All participants are given a detailed description of this study's protocol and are asked to sign an informed consent. The Medical Ethics Committee gave permission to perform the original study. This study is registered at clinicaltrials.gov (Identifier: NCT04891562) and has the code B115202000000. For these amendments, new permission of the Medical Ethics Committee Hasselt needs to be applied for.

3.4 Intervention

Participants in all subgroups are tested using the Social Communication Questionnaire (SCQ), the DCD Questionnaire, the Vanderbilt ADHD Diagnostic Rating Scale and the Movement ABC. IQ is assessed with the Wechsler Intelligence Scale for Children. All four subgroups will receive the abbreviated version of the Montreal Battery of Evaluation of Musical Abilities (MBEMA) (Peretz et al., 2013) and a tapping task. Children diagnosed with ADHD need to be withhold from taking methylphenidate the days of the sessions. This medication is typically

administered to reduce inattention, hyperactivity and impulsivity, symptoms commonly reported in the ADHD population. Previous research proved the beneficial effect of methylphenidate on timing abilities (Ben-Pazi, Shalev, Gross-Tsur, & Bergman, 2006; Rubia et al., 2009). Therefore, it could affect the results of this study.

3.4.1 Social Communication Questionnaire (SCQ)

The Social Communication Questionnaire will be administered to assess, if children included in this study, belong to the subgroup "children with (expected) autism spectrum disorder". Children with a score of 15 of more will be included in this given subgroup.

The Social Communication Questionnaire is a screening tool based on the Autism Diagnotic Interview – Revised (ADI-R). The questionnaire scores the children's communication skills and social functioning. It consists of 40 questions to be filled in by the child's parents and takes about ten minutes. With a cut-off score of 15, there are only 28% of false-negatives and 38% of false-positives. Overall sensitivity is 0,71. False -negative ASD children seem to be somewhat higher functioning ASD children, therefore, the SCQ seems to be a useful tool for identifying children at risk (Eaves, Wingert, Ho, & Mickelson, 2006).

3.4.2 Developmental Coordination Questionnaire – 2007 version (DCDQ'07)

The Developmental Coordination Questionnaire will be administered to assess, if children included in this study, belong to the subgroup "children with (expected) developmental coordination disorder". The children are included in the DCD subgroup if: children between the age of 5 and 8 have a score between 15 and 46, children between the age of 8 and 10 have a score between 15 and 55 or children between the age of 10 and 15 have a score between 15 and 57 on the DCD Questionnaire.

The Developmental Coordination Questionnaire is a psychological assessment tool for children aged 5 to 15 years old to screen for coordination disorders. This assessment is filled in by the child's parents. The DCDQ 2007 version consists of 15 items divided into three categories, namely "control during movement", "fine motor and handwriting" and "general coordination". The questionnaire takes about 10 to 15 minutes to complete. The overall sensitivity for the DCDQ'07 is 0.84, the specificity is 0.70. Results of the study by Wilson et al. (2009) provide evidence that the DCDQ 2007 version is a valid screening tool for DCD.

3.4.3 Vanderbilt ADHD Diagnostic Rating Scale (VADRS)

The Vanderbilt ADHD Diagnostic Rating Scale will be administered to asses, if children included in this study, belong to the subgroup “children with (expected) autism spectrum disorder”. Children with a score of four or more will be included in this ASD subgroup.

The Vanderbilt ADHD Diagnostic Rating Scale is a psychological assessment tool for parents of children aged 6 to 12 years old. The test consists of two checklists, one to be filled in by the child’s parents and the other to be filled in by the child’s teacher. The checklists consist of 55 items that cover four aspects: inattention, hyperactivity, conduct/oppositional problems and anxiety/depression problems. The teacher’s rating scale (VADTRS) has a sensitivity of 0,69 and a specificity of 0,84. Positive prediction is only 0,32, indicating the need of assessment by various observers (Wolraich, Bard, Neas, Doffing, & Beck, 2013). The parent’s rating scale (VADPRS) was found to be consistent with the DSM-V diagnostic criteria and other ADHD measurements like the teacher rating scale (Wolraich et al., 2003). In the present study only the parent rating scale will be used.

3.4.4 Movement Assessment Battery for Children -2 (MABC-2)

The Movement Assessment Battery for Children will be administered to assess if children, included in this study, belong to the subgroup “children with (expected) developmental coordination disorder”. Children with a score between the 2nd and 15th percentile will be included in this DCD subgroup.

The Movement Assessment Battery for Children-version 2 consists of a performance test and a 30 items checklist that measures manual dexterity, ball skills and balance. The MABC-2 can be used in children from 3 to 16 years old. Necessary time to fill in the checklist is ten to fifteen minutes; the motoric test takes about twenty to forty minutes. The MABC-2 is proven to be a clinically useful instrument for identifying children with impairments in motor performance (Brown & Lalor, 2009). Part one of the test, the checklist, will be filled in by the parents of the children. Part two of the test, the motoric test, will be filled in by a physiotherapist by observing the children.

3.4.5 Weschler Intelligence Scale for Children – V- NL (WISC-V-NL)

The Wechsler Intelligence Scale for Children will be administered to assess the intelligence quotient of the children included in this study. Children with an IQ-score below 70 will be excluded from this study.

The Wechsler Intelligence Scale for Children is an IQ test and can be performed with children from 6 to 16 years old. The test checks the verbal understanding skills, visual spatial awareness, fluid reasoning, working memory and processing speed in various tasks. This task takes one to two hours to complete, but short breaks in between are allowed. The WISC-V-NL is proven to be reliable with a score of 0.95 for the total IQ-score. Furthermore, norm values are available for children from Flanders (Pearson). The test will be administered by a (neuro-) psychiatrist.

3.4.6 Evaluation of rhythm perception: Montreal Battery of Evaluation of Musical Abilities

For the evaluation of rhythm perception, the abbreviated MBEMA is used. The MBEMA is an adaptation of the Montreal Battery for Evaluation of Amusia (MBEA), the latter is used for evaluating musical abilities in adults. Peretz et al. (2013) states that the adult version is not suitable for children below the age of 10. The abbreviated version of the MBEMA consists of three tests: melody, rhythm and memory. Each test consists of 20 unfamiliar tonal melodies. In the melody tasks one of the rhythms is slightly different from the others, children have to identify the one that is different. During the rhythm task one melody has an altered duration of two adjacent tones, children need to find this rhythm. Lastly in the memory task, children are asked whether they have heard a melody before and answer yes or no.

Evidence describing validity, sensitivity and reliability of the MBEMA is scarce. However, according to Peretz et al. (2013), the abbreviated MBEMA has a better sensitivity than the MBEA for adults. Therefore, the study stated that this version is suitable for identifying individual differences in music perception in children.

3.4.7 Evaluation of auditory motor coupling: Tapping task

A tapping task is used to evaluate auditory motor coupling. In this task children are asked to synchronize their finger taps to a metronome beat and three different woodblock rhythms. The children tap a computer mouse with the index finger of their preferred hand.

The woodblock rhythms are based on the auditory-motor synchronization task used in Tryfon et al. (2017). The rhythms are given subsequently in a random order to reduce learning-effect. The three rhythms are composed of an increasing metrical complexity. The first rhythm is strongly metric, meaning that there is a certain fixed order. The second and third rhythms are respectively medium and weakly metric, meaning that there is no fixed order. Participants complete six blocks of nine trials per block, all blocks have a different order of rhythms and will be given in a random order. Table 1 shows a representation of the randomized blocks and trials. Two practice blocks are included to ensure that the participants comprehend the task. During the practice trials they will be given a strongly metric rhythm that differs from the rhythms used in the intervention. J. L. Chen, Penhune, and Zatorre (2008) stated that this protocol is sensitive to measure fine-grained tapping timing.

Table 1.

Representation of blocks and trails for woodblock rhythms

Block	Trial
Block #1	#1#1#1-#2#2#2-#3#3#3-#2#2#2-#1#1#1-#3#3#3-#3#3#3-#2#2#2-#1#1#1
Block #2	#1#1#1-#3#3#3-#2#2#2-#2#2#2-#3#3#3-#1#1#1-#3#3#3-#1#1#1-#2#2#2
Block #3	#3#3#3-#2#2#2-#1#1#1-#1#1#1-#2#2#2-#3#3#3-#2#2#2-#1#1#1-#3#3#3
Block #4	#3#3#3-#1#1#1-#2#2#2-#1#1#1-#3#3#3-#2#2#2-#2#2#2-#3#3#3-#1#1#1
Block #5	#2#2#2-#1#1#1-#3#3#3-#3#3#3-#2#2#2-#1#1#1-#1#1#1-#2#2#2-#3#3#3
Block #6	#2#2#2-#3#3#3-#1#1#1-#3#3#3-#1#1#1-#2#2#2-#1#1#1-#3#3#3-#2#2#2

#1; strongly metric rhythm #2; moderate metric rhythm #3; weakly metric rhythm

No standard protocol for tapping tasks exists, limiting evidence regarding psychometric properties. However, Axelrod, Meyers, and Davis (2014) assessed different types of finger tapping tasks and found a specificity of approximately 90% and a sensitivity around 40%. (Criswell, Sterling, Swisher, Evanoff, & Racette, 2010).

3.4.8 Division of sessions

The assessments for the determination of the subgroup takes place in one session, whereas measurements for determining rhythm perception and auditory motor coupling skills are performed in a second session. Two sessions are provided to minimize fatigue and to maintain motivation and attention during the test sessions. In the first session all the demographics and participant characteristics are assessed by using the questionnaires and tests described above

(see section 3.4.1-3.4.5). Parents of the children fill in the VADPRS, the checklist of the MABC-2, the DCDQ'07 and the SCQ, this takes about one hour to complete. In the meantime, the child will perform the MABC-2 and WISC-V-NL. This session will take about an average of three hours. There will be a one-week period between session one and session two. In the second session rhythm perception is assessed using the abbreviated version of the MBEMA, auditory-motor coupling is assessed by the different tapping tasks. The MBEMA has a duration of approximately 20 to 40 minutes. Between the MBEMA and the tapping tasks a rest period of 10 minutes will be provided. Between the different blocks of the tapping tasks a rest period of three minutes is allowed. These sessions will take about an average of two hours. Figure 2 represents the study design.

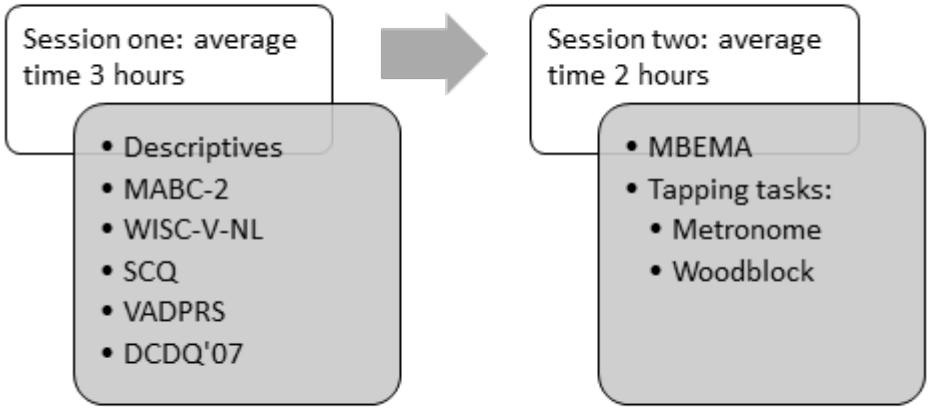


Figure 2. Study design, *abbreviations*: Movement Assessment Battery for Children Second edition (MABC-2), Wechsler Intelligence Scale for Children (WIS-C), Social Communication Questionnaire (SCQ), Vanderbilt ADHD Diagnostic Parent Rating Scale (VADPRS), Developmental Coordination Questionnaire – version 2007 (DCDQ'07), Montreal Battery of Evaluation of Musical Abilities (MBEMA)

3.4.9 Equipment

For the assessment of the demographics and participant characteristics questionnaires and test forms are used. For the assessment of the tapping task a computer is necessary, the participant taps on a computer mouse connected to the corresponding computer. A headphone (Sennheiser RS 127-8) is used for transferring the sounds used for the MBEMA and the tapping tasks. By using a headphone ambient noise is reduced.

3.5 Outcome measures

3.5.1 Demographics and participant characteristics

- Age (ratio: 6-12 years old)
- Gender (nominal: male/female)
- Disorder (incl. co-morbidities) (nominal: TD, ASD, ADHD and DCD)
- Medication (ordinal: none/ some (and what medication))
- Score of the Social Communication Questionnaire (interval: 0 - 40)
- IQ-score of the Wechsler Intelligence Scale for Children (interval: 0- 35)
- Score of the Movement Assessment Battery for Children (interval: percentile 0-100)
- Score of the Vanderbilt ADHD Diagnostic Rating Scale (interval: inattentive items: 0 – 36; hyperactive items: 0- 36; oppositional – defiant items: 0- 32; conduct items: 0- 56; performance items: 0- 40)
- Score of the DCD Questionnaire – version 2007 (interval: 15-75)

3.5.2 Primary outcome measures

Primary outcome measures for all subgroups are rhythm perception and rhythm synchronization/ auditory motor coupling. Rhythm perception using the MBEMA task assesses accuracy calculated as the percentage of correct responses in each task. Rhythm synchronization is measured using the tapping task by assessing mean intertap intervals and mean phase variability. Mean intertap interval is defined as the mean time interval between the onset of taps during a trial in milliseconds. The mean phase variability shows the difference between the onset of the auditory signal and the onset of a finger tap. These outcome measures have a ratio for measurement level.

3.5.2 Secondary outcome measures

A study by Enokizono et al. (2020) assessed the effect of age and gender on tapping tasks. The study revealed a significant effect of age on both tapping speed and regularity, meaning that both speed and regularity improve with age. Furthermore, the analysis revealed no significant effect of gender, however a small effect was present. Because of these previous results, gender and age will be taken into account in the statistical analysis.

3.6 Data analysis

Baseline characteristics of all subgroups are identified and a descriptive analysis is performed. After data collection, individual within-group results are assessed with a t-test suspecting normal distribution and homogeneity of the results. When within-group results are not normally distributed, a signed rank test will be used. When no homogeneity is found, a Welch ANOVA will be used. For assessing differences between rhythm frequencies within a group, a mixed model will be used. To examine differences in the MBEMA and tapping task performances between groups (ASD, ADHD and DCD) multiway ANOVA will be used, suspecting normal distribution and homogeneity of the results. When no normal distribution or homogeneity is found, a transformation can be performed, but this needs careful interpretation. Data analysis is performed using the JMP 14.2 software. Significance level is 0,05 with a confidence interval of 95%.

No existing study examined all four populations that will be examined in this study, therefor no results could be used to calculate a sample size. Because of this, a pilot study will be performed to quantify the number of children needed in each subgroup to achieve a power of 80%.

4. Time planning

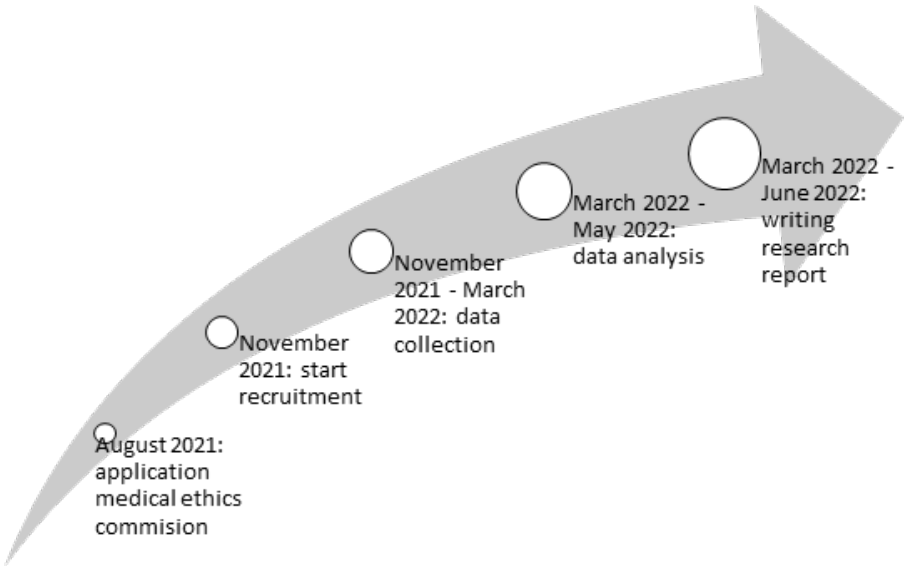


Figure 3. Visualization of time planning

The protocol will be implemented from July 2021 to June 2022 in a pilot study. The pilot study will consist of less children in each subgroup, ten to fifteen maximum. Medical ethics will be written in July 2021 and applied for in August. When approval is achieved recruitment starts in November 2021, data collection starts right after in November 2021 until March 2022. Data analysis starts in March 2022 after finishing data collection. At the same time, the methodology of the study will be written down in the research report. When all data is analyzed, results and discussion will be written and the research report will be finished. Figure 3 visualizes the time planning.

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6. Appendix

PDF's of the included assessments and questionnaires

D3 NICHQ Vanderbilt Assessment Scale—PARENT Informant

Today's Date: _____ Child's Name: _____ Date of Birth: _____

Parent's Name: _____ Parent's Phone Number: _____

Directions: Each rating should be considered in the context of what is appropriate for the age of your child. When completing this form, please think about your child's behaviors in the past 6 months.

Is this evaluation based on a time when the child ☐ was on medication ☐ was not on medication ☐ not sure?

Symptoms	Never	Occasionally	Often	Very Often
1. Does not pay attention to details or makes careless mistakes with, for example, homework	0	1	2	3
2. Has difficulty keeping attention to what needs to be done	0	1	2	3
3. Does not seem to listen when spoken to directly	0	1	2	3
4. Does not follow through when given directions and fails to finish activities (not due to refusal or failure to understand)	0	1	2	3
5. Has difficulty organizing tasks and activities	0	1	2	3
6. Avoids, dislikes, or does not want to start tasks that require ongoing mental effort	0	1	2	3
7. Loses things necessary for tasks or activities (toys, assignments, pencils, or books)	0	1	2	3
8. Is easily distracted by noises or other stimuli	0	1	2	3
9. Is forgetful in daily activities	0	1	2	3
10. Fidgets with hands or feet or squirms in seat	0	1	2	3
11. Leaves seat when remaining seated is expected	0	1	2	3
12. Runs about or climbs too much when remaining seated is expected	0	1	2	3
13. Has difficulty playing or beginning quiet play activities	0	1	2	3
14. Is "on the go" or often acts as if "driven by a motor"	0	1	2	3
15. Talks too much	0	1	2	3
16. Blurts out answers before questions have been completed	0	1	2	3
17. Has difficulty waiting his or her turn	0	1	2	3
18. Interrupts or intrudes in on others' conversations and/or activities	0	1	2	3
19. Argues with adults	0	1	2	3
20. Loses temper	0	1	2	3
21. Actively defies or refuses to go along with adults' requests or rules	0	1	2	3
22. Deliberately annoys people	0	1	2	3
23. Blames others for his or her mistakes or misbehaviors	0	1	2	3
24. Is touchy or easily annoyed by others	0	1	2	3
25. Is angry or resentful	0	1	2	3
26. Is spiteful and wants to get even	0	1	2	3
27. Bullies, threatens, or intimidates others	0	1	2	3
28. Starts physical fights	0	1	2	3
29. Lies to get out of trouble or to avoid obligations (ie, "cons" others)	0	1	2	3
30. Is truant from school (skips school) without permission	0	1	2	3
31. Is physically cruel to people	0	1	2	3
32. Has stolen things that have value	0	1	2	3

Adapted from the Vanderbilt Rating Scales developed by Mark L. Wolraich, MD.

Revised - 1102

HE0350

D3 NICHQ Vanderbilt Assessment Scale—PARENT Informant, continued

Today's Date: _____ Child's Name: _____ Date of Birth: _____

Parent's Name: _____ Parent's Phone Number: _____

Symptoms (continued)	Never	Occasionally	Often	Very Often
33. Deliberately destroys others' property	0	1	2	3
34. Has used a weapon that can cause serious harm (bat, knife, brick, gun)	0	1	2	3
35. Is physically cruel to animals	0	1	2	3
36. Has deliberately set fires to cause damage	0	1	2	3
37. Has broken into someone else's home, business, or car	0	1	2	3
38. Has stayed out at night without permission	0	1	2	3
39. Has run away from home overnight	0	1	2	3
40. Has forced someone into sexual activity	0	1	2	3
41. Is fearful, anxious, or worried	0	1	2	3
42. Is afraid to try new things for fear of making mistakes	0	1	2	3
43. Feels worthless or inferior	0	1	2	3
44. Blames self for problems, feels guilty	0	1	2	3
45. Feels lonely, unwanted, or unloved; complains that "no one loves him or her"	0	1	2	3
46. Is sad, unhappy, or depressed	0	1	2	3
47. Is self-conscious or easily embarrassed	0	1	2	3

Performance	Excellent	Above Average	Average	Somewhat of a Problem	Problematic
48. Overall school performance	1	2	3	4	5
49. Reading	1	2	3	4	5
50. Writing	1	2	3	4	5
51. Mathematics	1	2	3	4	5
52. Relationship with parents	1	2	3	4	5
53. Relationship with siblings	1	2	3	4	5
54. Relationship with peers	1	2	3	4	5
55. Participation in organized activities (eg, teams)	1	2	3	4	5

Comments:

For Office Use Only

Total number of questions scored 2 or 3 in questions 1–9: _____

Total number of questions scored 2 or 3 in questions 10–18: _____

Total Symptom Score for questions 1–18: _____

Total number of questions scored 2 or 3 in questions 19–26: _____

Total number of questions scored 2 or 3 in questions 27–40: _____

Total number of questions scored 2 or 3 in questions 41–47: _____

Total number of questions scored 4 or 5 in questions 48–55: _____

Average Performance Score: _____

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THE DEVELOPMENTAL COORDINATION DISORDER QUESTIONNAIRE 2007© (DCDQ'07)

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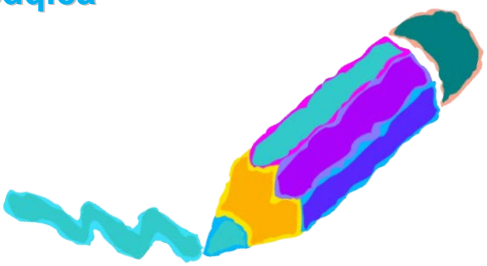

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March 2012

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COORDINATION QUESTIONNAIRE (REVISED 2007)

	Year	Mon	Day
Today's Date:			
Child's Birth:			
Child's Age:			

Name of Child: _____

Person completing Questionnaire: _____

Relationship to child: _____

Most of the motor skills that this questionnaire asks about are things that your child does with his or her hands, or when moving.

A child's coordination may improve each year as they grow and develop. For this reason, it will be easier for you to answer the questions if you think about other children that you know who *are the same age as your child*.

Please compare the degree of coordination your child has with other children of the same age when answering the questions.

Circle the one number that best describes your child. If you change your answer and want to circle another number, please circle the correct response twice.

If you are unclear about the meaning of a question, or about how you would answer a question to best describe your child, please call _____ at _____ for assistance.

	Not at all like your child 1	**A bit like your child 2**	**Moderately like your child 3**	**Quite a bit like your child 4**	**Extremely like your child 5**

1. Your child *throws a ball* in a controlled and accurate fashion.

   1 2 3 4 5

2. Your child *catches* a small *ball* (e.g., tennis ball size) thrown from a distance of 6 to 8 feet (1.8 to 2.4 meters).

   1 2 3 4 5

3. Your child *hits* an approaching *ball* or *birdie* with a bat or racquet accurately.

   1 2 3 4 5

4. Your child *jumps* easily *over* obstacles found in garden or play environment.

   1 2 3 4 5

5. Your child *runs* as fast and in a *similar* way to other children of the same gender and age.

   1 2 3 4 5

6. If your child has a *plan* to do a motor *activity*, he/she can organize his/her body to follow the plan and effectively complete the task (e.g., building a cardboard or cushion "fort," moving on playground equipment, building a house or a structure with blocks, or using craft materials).

   1 2 3 4 5 **(OVER)**

	Not at all like your child 1	A bit like your child 2	Moderately like your child 3	Quite a bit like your child 4	Extremely like your child 5

7. Your child's printing or *writing* or drawing in class is *fast* enough to keep up with the rest of the children in the class.

1 2 3 4 5

8. Your child's printing or *writing* letters, numbers and words is *legible*, precise and accurate or, if your child is not yet printing, he or she *colors and draws* in a coordinated way and makes pictures that you can recognize.

1 2 3 4 5

9. Your child uses appropriate *effort* or tension when printing or writing or drawing (no excessive *pressure* or tightness of grasp on the pencil, writing is not too heavy or dark, or too light).

1 2 3 4 5

10. Your child *cuts* out pictures and *shapes* accurately and easily.

1 2 3 4 5

11. Your child is interested in and *likes* participating in *sports or active* games requiring good motor skills.

1 2 3 4 5

12. Your child learns *new motor tasks* (e.g., swimming, rollerblading) easily and does not require more practice or time than other children to achieve the same level of skill.

1 2 3 4 5

13. Your child is *quick and competent* in tidying up, putting on shoes, tying shoes, dressing, etc.

1 2 3 4 5

14. Your child would ***never*** be described as a *"bull in a china shop"* (that is, appears so clumsy that he or she might break fragile things in a small room).

1 2 3 4 5

15. Your child does ***not*** *fatigue easily* or appear to slouch and "fall out" of the chair if required to sit for long periods.

1 2 3 4 5

Thank you.

COORDINATION QUESTIONNAIRE (DCDQ'07): SCORE SHEET

Name: _____ **Date:** _____

Birth Date: _____ **Age:** _____

	Control During Movement	Fine Motor/ Handwriting	General Coordination
1. Throws ball			
2. Catches ball			
3. Hits ball/birdie			
4. Jumps over			
5. Runs			
6. Plans activity			
7. Writing fast			
8. Writing legibly			
9. Effort and pressure			
10. Cuts			
11. Likes sports			
12. Learning new skills			
13. Quick and competent			
14. "Bull in shop"			
15. Does not fatigue			

TOTAL _____ / 30 Control during Movement + _____ / 20 Fine Motor/ Handwriting + _____ / 25 General Coordination = _____ / 75 **TOTAL**

For Children Ages 5 years 0 months to 7 years 11 months
15-46 indication of DCD or suspect DCD
47-75 probably not DCD

For Children Ages 8 years 0 months to 9 years 11 months
15-55 indication of DCD or suspect DCD
56-75 probably not DCD

For Children Ages 10 years 0 months to 15 years
15-57 indication of DCD or suspect DCD
58-75 probably not DCD

 www.dcdq.ca

Administration and Interpretation of the DCDQ'07

Overview

The *Developmental Coordination Questionnaire (DCDQ)* is a parent report measure developed to assist in the identification of Developmental Coordination Disorder (DCD) in children. Parents are asked to compare their child's motor performance to that of his/her peers using a 5 point Likert scale. It provides a standard method to measure a child's coordination in everyday, functional activities. As reported in 2000[1], the internal consistency of the DCDQ is high and the results from discriminant function analyses were appropriately strong for a screening tool.

Developmental Coordination Disorder is a DSM-IV[2] diagnosis. An indication of DCD based on the score of the DCDQ fulfills the requirement for Criterion B of this diagnosis. However, the questionnaire cannot be used alone for this purpose. Diagnosis must be made based on the results of several reports and tests. The questionnaire is labeled "The Coordination Questionnaire" to avoid parents becoming concerned that a medical condition is being diagnosed.

The *DCDQ`07* presented here is considered to have stronger psychometric properties than the 2000 version because it was developed with a population-based sample and has a larger age range[3] . The research took place between 2004 and 2006, involving 287 typically developing children, as well as 232 children who were reported to have motor coordination difficulties or who were more likely to have DCD. This revised version is appropriate for use with children ages 5 to 15.

The *DCDQ'07* consists of 15 items, which group into three distinct factors. The first factor contains a number of items related to motor control while the child was moving, or while an object was in motion, and is labelled "Control during Movement". The second factor contains "Fine Motor and Handwriting" items and the third factor relates to "General Coordination". These factor scores alone do not provide an indication of whether the child may have DCD. However, when the scores of each of the factors are examined relative to the scores of the other factors and are then compared with formal and informal assessment results, support for the identification of particular motor strengths and challenges a child is experiencing may be provided.

Prior to Administration

Before copying for clinical or research use, it is recommended that a name and phone number be written into the space on the first page so that parents can call if they have questions about the meaning of an item. This contact person should be knowledgeable about the condition of DCD, or know who to refer the question to if questions of this nature arise. The validity of the results will be increased if parents have the opportunity to clarify the intent of an item.

It is recommended that the 2 page questionnaire be copied double sided. The Score Sheet on the 4th page should be kept separate from the questionnaire itself. It is not recommended that parents be given the Score Sheet.

Respondents
This questionnaire was developed for parents, as parents know their children the best and can reliably report developmental problems. In addition, only the data from parent report was used to develop the scoring system. This DCDQ is therefore intended to be used with parents. However, some clinicians and researchers are experimenting with having both parents (or one parent and the child's primary teacher) complete it. Sometimes two or more respondents have completed the questionnaire separately, but in other situations they have conversed while completing one form. Subjectively, the results appear to be satisfactory but no one has yet studied this approach.

When the perspective of two adults gives a more complete or more accurate evaluation of the child's motor performance, this practice is likely to increase the validity of the score. However, it must be remembered that the scores were developed solely on parent response, so if the respondents have divergent opinions on the child's performance, or if the two forms have very different scores, the parent's score should be the one reported. The fact that others who know the child score the items differently can be noted, but it would be inappropriate to use the score of a teacher or coach alone (for example) in interpreting the results of the DCDQ.

Time to Complete
The DCDQ usually takes parents about 10-15 minutes to complete. As much as possible, arrange for the parent completing the questionnaire to do so in a non-distracting environment.

Administration - Written or Verbal
The DCDQ was designed to be self-administered by parents. In the reference sample of the development of the original DCDQ, however, parents were given the choice of completing a paper version of the questionnaire independently or of completing it over the phone while reading a paper copy along with the interviewer. In the study for the revised *DCDQ'07*, most parents completed a paper copy independently but a small proportion completed it with an occupational therapist following administration of the standardized motor tests. Either method of completion is acceptable.

Missing Items
When the questionnaire is completed or returned, review it for missed items or items where more than one item is circled. Ask the parent who completed it for clarification. **Note:** a total score can only be calculated if all items are scored. Missing one score will prevent you from obtaining a total score and having an indication of DCD or not.

If the parent does not know how to grade an item, or has not seen their child in a particular activity, ask them if there is anyone else who would know (e.g., the other parent, a caregiver, a teacher or a coach). You may inquire if the parent can make arrangements to ask that person, or if they will give you permission to do so.

Computing the Chronological Age

Enter the date that the DCDQ was completed and the child's Date of Birth (D.O.B.) on the first page of the questionnaire. Compute the chronological age by subtracting (first) the days, then the month and finally the year of birth. For example, if the questionnaire was completed on March 21, 2007, and the child was born on February 2, 2000, the child's chronological age would be calculated as shown in the first table:

	Yr	Mon	Day
DCDQ completion	2007	03	21
Child's D.O.B.	2000	02	02
Chronological age	7 yrs	1 mon	19 day

	Year	Month	Day
DCDQ completion	~~2007~~ 2006	14 ~~02~~ ~~03~~	51 ~~21~~
Child's D.O.B.	2000	06	28
Chronological age	6 years	8 month	23 days

If the day of the month in which the child was born is larger than the day of the month of questionnaire completion, add 30 days to the day of testing and subtract one month from the month of testing. Similarly, if necessary, a month of testing can be borrowed by adding 12 months to the month of testing and subtracting one year from the testing year, as shown above in the table on the right.

Computing a Total Score

Re-enter the numbers circled for all items of the questionnaire onto the Score Sheet (4th page).

Total each column to compute the 3 Factor Scores, and add all Factor Scores to compute a Total Score. *Double check your addition.*

Interpretation of Scores on the DCDQ

Using the child's chronological age at the time the questionnaire was completed, find the appropriate age grouping on the left column of the table below. Scan across that row to find the range of scores which the child's score falls within. This range will indicate whether the child's score is an "Indication of, or Suspect for, DCD", or "Probably not DCD".

Age Group	Indication of, or Suspect for, DCD	Probably not DCD
5 years *to* 7 years 11 months	**15 - 46**	**47 - 75**
8 years 0 months *to* 9 years 11 months	**15 - 55**	**56 - 75**
10 years 0 months *to* 15 years	**15 - 57**	**58 - 75**

Reporting of DCDQ`07 results
As outlined above, the DCDQ cannot be used alone to identify DCD. When using the questionnaire in a verbal or written report about a child, the terms ``indication of possible DCD``, ``suspect for DCD``, or ``probably not DCD`` should be used, as this test alone cannot be used to diagnose DCD.

Sensitivity and Specificity
It is sometimes desirable, especially when a diagnosis is not clear, to report the sensitivity and specificity of the test scores. The most accurate predictive values of the *DCDQ`07* are reported in the table below according to the different age ranges. If overall values for the questionnaire are required, however, the overall sensitivity is 84.6% and the specificity is 70.8%.

Age Group	Sensitivity and Specificity
5 years *to* 7 years 11 months	Sensitivity=75.0% Specificity=71.4%
8 years 0 months *to* 9 years 11 months	Sensitivity=88.6% Specificity=66.7%
10 years 0 months *to* 15 years	Sensitivity=88.5% Specificity=75.6%

The purpose of a screening instrument is to identify whether a child has a particular condition. Rarely is a screening tool alone 100% accurate in identifying all children with a condition while at the same time not falsely identifying any children who do not. When evaluating a screening tool such as the *DCDQ`07*, the degree of accuracy in identifying children with possible DCD (sensitivity) must be compared to the accuracy in correctly identifying children who do not have the condition (specificity). This "trade off" is common to all diagnostic tests because when one of these predictive values increases, the other decreases. By design, the *DCDQ'07* is most accurate in identifying children who may have DCD. It may identify children who do not have the condition, but further motor testing should reveal whether DCD is indeed present.

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Movement Assessment Battery for Children - 2

Checklist

Naam van kind:	Geslacht: jongen / meisje
Geboortedatum kind:	
Naam van ouder/leerkracht	
School:	Groep / Klas:
Adres:	Land: Nederland / België /
Datum Checklist ingevuld:	
Ingevuld door: moeder / vader / verzorger / leerkracht / therapeut / anders nl.	

Movement ABC-2 Test Afgenomen?

Ja / Nee

Deel A totaal:

Deel B totaal:

Totaal deel A + B =

De Checklist

De Checklist bestaat uit drie delen. Deel A en B zijn gericht op de motorische competentie van het kind. Deel C is gericht op eigenschappen van het kind die van invloed kunnen zijn op de motoriek, zoals het plezier in bewegen, de mate van overbeweeglijkheid of afleidbaarheid.

Instructie

Het scoren van **deel A en B** van de Checklist gaat in twee stappen. Eerst moet u bedenken of het kind de taak wel (score 0 of 1) of niet (score 2 of 3) kan uitvoeren. Als het kind de taak wel kan uitvoeren, kan gekozen worden uit '0: heel goed uitvoerbaar' of '1: net uitvoerbaar'. Als het kind de taak niet kan uitvoeren wordt gekozen tussen '2: net niet uitvoerbaar' of '3: nog lang niet uitvoerbaar'. Elk item krijgt een afzonderlijke score. Is informatie over een item niet bekend, dan moet geprobeerd worden om door observatie of met informatie van anderen (leerkrachten of ouders) deze informatie toch zo goed mogelijk in te schatten. Het kan zijn dat een kind bepaalde activiteiten niet laat zien. Vaak komt dit omdat het kind de vaardigheid niet beheerst. Scoor dan een 3. Mocht u het gevoel hebben het kind hier onrecht mee te doen, vul dan een 'N' in (maximaal drie keer voor deel A en B samen). Dit betekent: 'Niet gezien'.

Niet-motorische factoren die de prestatie kunnen beïnvloeden

In **deel C** worden enkele gedragskenmerken genoemd. De beoordelaar geeft aan of hij denkt dat het omschreven gedrag van toepassing is op het kind. Deze vragen worden met 'ja' of 'nee' beantwoord. Omcirkel na het invullen van de Checklist, het antwoord op de onderstaande twee vragen dat het meest van toepassing is.

(a) Denkt u dat de factoren zoals omschreven in deel C invloed hebben op wat het kind op gebied van motoriek werkelijk zou kunnen?
helemaal niet / enigszins / veel invloed

(b) Hoe belangrijk denkt u dat deze factoren zijn voor het opstellen van een (be)handelingsplan?
helemaal niet / enigszins / heel belangrijk

Deel A: Bewegen in een stilstaande (statische) voorspelbare omgeving

0 = goed uitvoerbaar	1 = net uitvoerbaar
2 = net niet uitvoerbaar	3 = nog lang niet uitvoerbaar
N = niet gezien	

A1 Zelfverzorging

		Score
A.1.1	Blijft in evenwicht tijdens het staand aankleden *(bijv. broek, rok)*	
A.1.2	Trekt kledingstukken over het hoofd aan *(bijv. T-shirt, trui)*	
A.1.3	Maakt knopen zelf dicht *(bijv. op blouse of jas)*	
A.1.4	Wast en droogt de handen	
A.1.5	Schenkt drinken in *(bijv. van een fles/pak in een beker)*	

A2 Schoolse vaardigheden

		Score
A.2.1	Werkt netjes met kleine voorwerpen *(bijv. blokjes, kralen, papier)*	
A.2.2	Maakt lettervormen met een potlood of pen	
A.2.3	Gebruikt een schaar om papier mee te knippen	
A.2.4	Loopt het klaslokaal/een kamer door zonder tegen iets of iemand aan te lopen	
A.2.5	Draagt voorwerpen door een ruimte *(boeken, kopjes of pennen)* zonder deze te laten vallen	

A3 Sport- en spelvaardigheden

		Score
A.3.1	Springt met twee voeten tegelijk tijdens de afzet en landing	
A.3.2	Hinkelt op zowel het linker- als het rechterbeen	
A.3.3	Gooit met een pittenzakje of bal, zodat een ander stilstaand kind kan vangen	
A.3.4	Gebruikt vaste speel/gymtoestellen *(bijv. klimrek, glijbaan)*	
A.3.5	Ontwijkt bij het oversteken van bijvoorbeeld speelplaats of gymzaal, stilstaande objecten of personen	

Deel A Totaal .....

Extra informatie *(optioneel)*:

Van dit kind is bekend dat hij/zij kampt met een leerprobleem		Ja / Wordt onderzocht / Nee
	Aandachtsproblemen	Ja / Wordt onderzocht / Nee
	Spraak- en/of taalproblemen	Ja / Wordt onderzocht / Nee
	Leesproblemen	Ja / Wordt onderzocht / Nee
	Sociale problemen	Ja / Wordt onderzocht / Nee

Deel B: Bewegen in een bewegende (dynamische) en onvoorspelbare omgeving

0 = goed uitvoerbaar	1 = net uitvoerbaar
2 = net niet uitvoerbaar	3 = nog lang niet uitvoerbaar
N = niet gezien	

B1 Zelfverzorging/Schoolse vaardigheden

		Score
B.1.1	Blijft in evenwicht staan of zitten als evenwichtsaanpassingen vereist zijn *(bijv. zit op een bank en schuift op als anderen erbij komen zitten; staat in een rij met bewegende kinderen)*	
B.1.2	Beweegt door een drukke ruimte om voorwerpen te verzamelen of uit te delen *(bijv. boeken of pennen)*	
B.1.3	Draagt een dienblad of kopje door een ruimte en ontwijkt hierbij andere bewegende personen *(bijv. in een zaal of kantine)*	
B.1.4	Blijft in de maat van de muziek tijdens het klappen in de handen of stampen met de voeten	
B.1.5	Beweegt zich op de maat van de muziek of met andere mensen *(bijv. marcheren in een rij, dansen in een groep)*	

B2 Balvaardigheden

		Score
B.2.1	Vangt een bal met twee handen	
B.2.2	Raakt/slaat een bewegende bal *(bijv. met een stick, knuppel of racket)*	
B.2.3	Gooit al rennend een bal, zodat een ander kind de bal kan vangen	
B.2.4	Houdt een stuitende bal onder controle	
B.2.5	Doet mee met teamsporten of spel waarin gooien, vangen, slaan of schoppen aan bod komen	

B3 Sport- en spelvaardigheden

		Score
B.3.1	Fietst zonder zijwieltjes	
B.3.2	Doet mee aan achtervolgspelletjes *(bijv. tikkertje, vangertje)*	
B.3.3	Blijft in evenwicht tussen andere kinderen in het water *(bijv. staan in het zwembad)*	
B.3.4	Gebruikt zonder hulp bewegende speel/gymtoestellen *(bijv. schommel, karretjes passend bij de leeftijd zoals driewielers of skelters)*	
B.3.5	Ontwijkt bij het oversteken van gymzaal/speelplaats bewegende objecten of personen	

Deel B Totaal .....

Algemeen *(omcirkel het meest passende antwoord)*:

Heeft dit kind een bewegingsprobleem?			JA / NEE
Indien 'JA', hebben deze problemen een negatieve invloed op:			
■ Schoolse vaardigheden:	helemaal niet	enigszins	veel invloed
■ Sport- en spelvaardigheden:	helemaal niet	enigszins	veel invloed
■ Zelfvertrouwen:	helemaal niet	enigszins	veel invloed
■ Sociale interactie:	helemaal niet	enigszins	veel invloed

Deel C: Niet-motorische factoren die de prestatie kunnen beïnvloeden

Geef hieronder aan of er (gedrags)factoren zijn die naar uw oordeel van invloed zijn op de motorische prestaties van het kind. De kopjes (met voorbeelden) worden als richtlijn gegeven. U mag ook de positieve aspecten benoemen, ook al worden de negatieve aspecten meer benadrukt.

		Ja	Nee
C.1	**Ongeorganiseerd** – bijv. verspreid liggende kleding vertraagt het aankleden na de gymles; trekt kleding in verkeerde volgorde aan		
C.2	**Vergeetachtig/Twijfelend** – bijv. begint langzaam aan complexe bewegingen; vergeet wat er gedaan moet worden in het midden van een reeks		
C.3	**Passief** – bijv. weinig geïnteresseerd; heeft veel aanmoedigingen nodig om deel te nemen		
C.4	**Timide** – bijv. bang voor activiteiten als springen en klimmen; vraagt constant om assistentie		
C.5	**Gespannen** – bijv. lijkt nerveus, bibbert; raakt in een stressvolle situatie in de war		
C.6	**Impulsief** – bijv. begint voordat de instructies/demonstraties voltooid zijn; ongeduldig		
C.7	**Snel afgeleid** – bijv. kijkt te veel in het rond; reageert op irrelevante geluiden		
C.8	**Overbeweeglijk** – bijv. wiebelt en draait; beweegt constant wanneer het aan het luisteren is naar de instructies; friemelt met kleren		
C.9	**Overschat eigen kunnen** – bijv. probeert de opdracht te veranderen door het moeilijker te maken; probeert de dingen te snel te doen		
C.10	**Onderschat eigen kunnen** – bijv. zegt dat de opdrachten te moeilijk zijn; verzint smoesjes waarom het de opdracht niet goed uit kan voeren		
C.11	**Gebrek aan doorzettingsvermogen** – bijv. geeft het snel op; is snel gefrustreerd		
C.12	**Van streek bij mislukken** – bijv. krijgt tranen in de ogen; weigert poging te herhalen		
C.13	**Lijkt geen plezier te beleven aan succes** – bijv. reageert niet als het geprezen wordt		
	Overige:		

Dank u voor het invullen van deze vragenlijst. Deze lijst graag terugsturen naar/inleveren bij:

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15/4199.05

Social Communication Questionnaire (SCQ) – Current
PC Answer Sheet

Michael Rutter, M.D., F.R.S., Anthony Bailey, M.D., Sibel Kazak Berument, Ph.D.,
Catherine Lord, Ph.D., and Andrew Pickles, Ph.D.

Name of Subject: _____ D.O.B. _____ Interview Date _____ Age: ____

Gender: ○ F ○ M Name of Respondent: _____ Relation to Subject: _____

Directions: Thank you for taking the time to complete this questionnaire. Please answer each question by selecting *yes* or *no*. A few questions ask about several related types of behavior; please select *yes* if *any* of these behaviors were present during the past 3 months. Although you may be uncertain about whether some behaviors were present or not, please answer *yes* or *no* to every question on the basis of what you think.

Item		**Yes**	**No**
1.	Is she/he now able to talk using short phrases or sentences? If *no*, skip to question 8.	○	○
2.	Do you have a to and fro "conversation" with her/him that involves taking turns or building on what you have said?	○	○
3.	Does she/he ever use odd phrases or say the same thing over and over in almost exactly the same way (either phases that she/he hears other people use or ones that she/he makes up?	○	○
4.	Does she/he ever use socially inappropriate questions or statements? For example, does she/he ever regularly ask personal questions or make personal comments at awkward times?	○	○
5.	Does she/he ever get his/her pronouns mixed up (e.g., saying *you* or *she/he* for *I*)?	○	○
6.	Does she/he ever use words that she/he seems to have invented or made up her/himself; put things in odd, indirect ways; or use metaphorical ways of saying things (e.g., saying *hot rain* for *steam*)?	○	○
7.	Does she/he ever say the same thing over and over in exactly the same way or insist that you say the same thing over and over again?	○	○
8.	Does she/he have things that she/he seems to do in a very particular way or order or rituals that she/he insists that you go through?	○	○
9.	Does her/his facial expressions usually seem appropriate to the particular situation, as far as you can tell?	○	○
10.	Does she/he ever use your hand like a tool or as if it were part of his/her own body (e.g., pointing with your finger or putting your hand on a doorknob to get you to open the door)?	○	○
11.	Does she/he ever have any interests that preoccupy her/him and might seem odd to other people (e.g., traffic lights, drainpipes, or timetables)?	○	○
12.	Does she/he ever seem to be more interested in parts of a toy or an object (e.g., spinning the wheels of a car), rather than in using the object as it was intended?	○	○
13.	Does she/he ever have any special interests that are *unusual* in their intensity but otherwise appropriate for his/her age and peer group (e.g., trains or dinosaurs)?	○	○
14.	Does she/he ever seem to be *unusually* interested in the sight, feel, sound, taste, or smell of things or people?	○	○
15.	Does she/he ever have any mannerisms or off ways of moving her/his hands or fingers, such as flapping or moving her/his fingers in front of her/his eyes?	○	○
16.	Does she/he ever have any complicated movements of her/his whole body, such as spinning or repeatedly bouncing up and down?	○	○

Item	Yes	No
17. Does she/he ever injure her/himself deliberately, such as by biting her/his arm or banging her/his head?	○	●
18. Does she/he ever have any objects (*other* than a soft toy or comfort blanket) that she/he *has* to carry around?	○	●
19. Does she/he have any particular friends or a best friend?	○	●
20. Does she/he ever talk with you just to be friendly (rather than to get something)?	○	●
21. Does she/he ever *spontaneously* copy you (or other people) or what you are doing (such as vacuuming, gardening, or mending things)?	○	●
22. Does she/he ever spontaneously point at things around her/him just to show you things (not because she/he wants them?	○	●
23. Does she/he ever use gestures, other than pointing or pulling your hand, to let you know what she/he wants?	○	●
24. Does she/he nod her/his head to indicate *yes*?	○	●
25. Does she/he shake her/his head to indicate *no*?	○	●
26. Does she/he usually look at you directly in the face when doing things with you or talking with you?	●	●
27. Does she/he smile back if someone smiles at her/him?	○	●
28. Does she/he ever show you things that interest her/him to engage your attention?	○	●
29. Does she/he ever offer to share things other than food with you?	○	●
30. Does she/he ever seem to want you to join in her/his enjoyment of something?	○	●
31. Does she/he ever try to comfort you if you are sad or hurt?	○	●
32. If she/he wants something or wants help, does she/he look at you and use gestures with sounds or words to get your attention?	●	●
33. Does she/he show a normal range of facial expressions?	○	●
34. Does she/he ever spontaneously join in and try to copy the actions in social games, such as *The Mulberry Bush* or *London Bridges Is Falling Down*?	○	●
35. Does she/he play any pretend or make-believe games?	●	●
36. Does she/he seem interested in other children of approximately the same age whom she/he does not know?	○	○
37. Does she/he respond positively when another child approaches her/him?	○	○
38. If you come into a room and start talking to her/him without calling her/his name, does she/he usually look up and pay attention to you?	○	○
39. Does she/he ever play imaginative games with another child in such a way that you can tell that each child understands what the other is pretending?	○	○
40. Does she/he play cooperatively in games that need some form of joining in with a group of other children, such as hide-and-seek or ball games?	○	○