

Development of A Classification of Children with Developmental Coordination Disorders

Based on Clinical Subgroups

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A dissertation submitted in partial fulfillment of the requirements for the degree of

Doctor of Philosophy

University of Washington

2015

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Program Authorized to Offer Degree:

Rehabilitation Science

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Abstract

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Background.

Developmental coordination disorder (DCD) is a neurological disorder that is typically diagnosed in school-age children and affects approximately 5-10% of school-age children in US. Children with DCD often demonstrate a variety of motor concerns, including poor motor, sensory and postural control function compared to typical peers. They may show delayed and poor quality of fine motor or gross skills, especially higher-level motor skills, and experience challenges in their daily life activities. The presumed central nervous system (CNS) pathology of DCD has not been confirmed. Also, because these symptoms are relatively subtle, diagnosis and recommendations for intervention are difficult but critical. Better evaluation and classification of children's motor concerns is warranted.

Objectives.

To systematically determine the knowledge base of brain pathology in DCD, to examine new potential tests to determine children's ability within certain important and specific constructs that

are problematic for children with DCD, and to explore defining subgroups within children who have a DCD diagnosis.

Methods.

A systematic review was conducted to determine the knowledge base for the pathology in DCD by reviewing eight brain-imaging studies. Nineteen pairs of children with DCD and age-matched peers with typical development (TD) were tested to validate an assessment of motor planning, the Motor Planning Maze Assessment (Maze) and an assessment of gait coordination, three items from the Functional Gait Assessment that were modified for children (pediatric modified FGA, pmFGA). Paired-t tests and cross tables were used for statistical analysis. Children with DCD were also examined using tests across domains of fine motor, gross motor, balance, coordination, sensory processing, and intelligence. Through visual analysis using pattern recognition of test results portrayed by standardized percentile ranks, subgroups are proposed.

Results.

The systematic review revealed that pathologies of DCD related to motor function include many areas of the brain and several tracts in children with DCD. The validity of the clinical tests of motor planning and gait coordination were supported as assessments that differentiate motor function in a group of children with DCD and their peers with TD. Finally, via detailed examination of the children with DCD, it appears that clinical characteristics among several domains identified by standardized clinical assessments do suggest that subgroups of DCD exist.

Limitations.

Few studies exist examining the pathology of DCD. One psychometric parameter, construct validity, of the MAZE and pmFGA was examined. The sample size for sub-group analysis was small; therefore more robust statistical analyses could not be employed. Measurements used addressed some of the problematic domains in children with DCD, but other tests/measures are likely necessary.

Conclusions.

Preliminary data exists to define the pathology of DCD. The MAZE and pm FGA are two promising measures that could be used within the evaluation of children with DCD. Analysis of a group of standardized clinical assessments suggests that subgroups of DCD are identifiable. More reliable and valid statistical analysis with larger samples of children are needed in order to confirm the pathology, appropriate clinical measurements and identification of subgroups, which should better direct evaluation and intervention for children with DCD. Further research is warranted in all these areas.

Plain Language Summary

Developmental Coordination Disorder (DCD) occurs in 5-6% of children. It is a chronic condition where children may have subtle but identifiable problems in many activities such as large motor (skipping, catching balls), balance (hopping, bike-riding), and/or fine motor (tying shoelaces, handwriting) skills. These problems affect the success of children within their daily living, school, recreational and self-care tasks. This can lead to social and psychological problems. Children with DCD movement problems, however, are treatable. If we can identify children with DCD when they are young and provide them with the right kind of help, their outcome can be very good.

It is difficult to identify DCD because there is little information about its cause and because children with DCD have many symptoms. If we can find a way to better identify and classify children with DCD then we could better assist the children and their families. It is important to have a standard way to assess and classify children with DCD. The main goal of this study was to develop a way to assess children with DCD, from brain differences to movement abilities that could also identify subgroups of DCD. To do this, three things were done: (1) a review of studies about the brains of children with DCD; (2) an examination of two tests of motor planning (knowing how to organize movements before they occur) and coordination during walking of 19 children with DCD compared to 19 peers without DCD; and (3) an examination of the results of five standardized assessments to propose subgroups of DCD.

Findings: 1) From the review of studies that scanned the brains of children with DCD, the results indicated that the potential problems of DCD primarily relate to the parts of the brain that control motor function, balance, attention and management of thinking. 2) From the assessments of the children with DCD, the Motor Planning Maze Assessment (Maze) and the Functional Gait

Assessment (FGA) were useful in finding problems in motor planning and coordination of walking in the children with DCD. 3) Using the results of the assessment of the children with DCDs, it was possible to form subgroups of DCD based on the children's thinking abilities and specific motor and sensory functional abilities.

Interpretation: The review of studies about the brains of children with DCD suggest there are many possible ways the brains of children with DCD may be associated with their problems. Using clinical assessments, it was possible to pick up subtle problems in children with DCD. It was also possible to use a set of clinical assessments to suggest subgroups of children with DCD.

How May This Make a Difference?

1. As we learn more about how the brains of children with DCD work, we may find better ways to help children with DCD. Identifying subgroups of children with DCD may help us in doing that.

2. A set of clinical assessments was used in this study. These assessments are sensitive and can be used in clinical settings to help us learn more about the problems children with DCD and how best to help them.

Human Subjects approval number: **45014**

Institutional review board: **University of Washington**

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ACKNOWLEDGMENTS

I am very fortunate to have received support in so many ways during my doctoral studies

First I want to thank my amazing dissertation committee members *Dr. Deborah Kartin, Dr. Marcia Ciol, Dr. Tracy Jirikowic, Dr. Patricia Kramer* and the chair *Dr. Sarah Westcott McCoy*. Without their guidance and unconditional support, I would never have been able to finish my dissertation.

Sally, it was a great pleasure to be your student. Thank you so much for your tolerance with all my crazy ideas, and also for always keeping me on my toes. It was such an incredible experience learning experience that guide me closer to kind of researcher and PT I want to become. Thank you for always being with me.

Debbie, for your endless patience and wisdom to listen and help me through every tough moment in these five years and made these past years such an wonderful experience for me.

To *Mr. Bob Price* for creating everything I need for the studies and much more, for teaching me with great patience and for overcoming every technical difficulties.

To the DPT division for supporting my research, and giving me chance to share my knowledge.

To *Madisen Clark, SPT and Lizbeth Arias, DPT* for helping me with data collection and analysis.

To all children and families, the schools in the Seattle School District, therapists and social workers, who participated in my study, for making my research experience both enriching and enjoyable. I am fortunate, indeed, to have been able to conduct my research with such a remarkable group of people.

To all my peers in the PhD program and particularly my cohort for your being there for me during tough days and for allowing me to learn so much from you.

To *Laura Robinson* and *Vickie Corrin* who are always happy to help and are the only people I know who always have an answer for every question.

To the Walter C. and Anita C. Stolov Research award for funding this research project.

To Experiment.com and everyone helped me through the crowd funding for funding this research project from many different places around the world.

Finally, a lot of thanks to my families and all my friends in Taiwan and Seattle for always being supper supportive and encouraging me with their best wishes.

Thank you, 謝謝

Lin-Ya Hsu, PT, PhD

CHAPTER 1

Introduction

Developmental Coordination Disorder (DCD) is a neurological disorder that is usually first diagnosed in school-age children. The current diagnostic criteria for DCD depend on observed symptoms (Campbell, Palisano, & Vander, 2012; Henderson & Henderson, 2002; Wilson, Riddock, Smits-Engelsman, Polatajko, & Blank, 2013). However, because these symptoms are relatively subtle, diagnosis is difficult but critical. The commonly used diagnostic criteria are from the Diagnostic and Statistical Manual of Mental Disorders- fourth edition (DSM-V) (American Psychiatric Association[APA], 2013). The criteria include:

- A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) as well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike, or participating in sports).
- B. The motor skills deficit in Criterion A significantly and persistently interferes with activities of daily living appropriate to chronological age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure, and play.
- C. Onset of symptoms is in the early developmental period.
- D. The motor skills deficits are not better explained by intellectual disability (intellectual developmental disorder) or visual impairment and are not attributable to a neurological condition affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder) (APA, 2013, p. 74).

Because of these multiple criteria, it requires a long differential diagnosis process to confirm the diagnosis of DCD. This process requires both clinical and laboratory measures and also requires a long period of observation. Thus, the clinical feasibility of diagnosis is complicated. Also, the many questions about the underlying pathology of DCD also increase the difficulty of the diagnosis process.

Based on current literature, the pathology of DCD is still unknown (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker, Missiuna, Harris, & Boyd, 2012a). There are few studies in this area that address the possible affected areas in the central nervous system (CNS). There are two main challenges of finding the possible pathology of DCD: (1) children are usually not an appropriate population for brain imaging, and (2) there is a large variation among symptoms of DCD, so it is difficult to locate the exact area of the brain that is involved. In order to identify the pathology of DCD, we need to find an appropriate technology for children and we need to specify the symptoms of interest in DCD (Blank et al., 2012; Green, 2010; Zwicker et al., 2012a).

Overall, children with DCD can show a myriad of motor problems that affect all domains of the International Classification of Functioning, Disability and Health (ICF) (World Health Organization[WHO], 2001). At the activity level, compared to peers who are typically developing, children with DCD may show delayed and poor quality of fine or gross motor skills, especially with higher-level motor skills, such as writing, hopping, jumping, and ball skills (; Fong, Lee, & Pang, 2011; Green & Baird, 2006; Zwicker et al., 2012a). Body structure/function impairments may include delayed or poor postural control, such as poor feedback and feedforward control or poor coordination of postural muscles (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a). Besides the motor limitations, some children with DCD may have

problems with their sensory processing functions, such as inadequate information processing, poor visual perception, or poor motor planning. These body structure/function impairments not only influence their ability to carry out daily activities, but also impact their participation and quality of life (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker et al., 2012a).

Children with DCD may also have problems with their academic performance in school, which may lead to more restrictions in participation and may provoke psychological problems (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker et al., 2012a). Parents, teachers and medical professionals often overlook the challenges children with DCD face in their daily life. This is one of the main reasons that children with DCD may experience more stress and psychological issues (e.g. depression) when they become adolescents (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a). The motor problems of children with DCD are treatable. If we can identify these children in at an early age and provide appropriate intervention, their prognosis is very promising (Blank et al., 2012; Campbell et al., 2012;). Therefore, it is important to have a standardized process, which can efficiently and reliably diagnose children with DCD.

In order to solve the problems of the current DCD diagnostic process and improve the efficiency and accuracy of intervention planning and implementation for children with DCD and further identify the possible pathologies of DCD, it is important to systematically determine the knowledge base of brain pathology in DCD. It is also important to examine new potential tests to determine children's abilities within certain essential and specific constructs that seem to a problem for children with DCD. In addition it is important to determine if there are subgroups within DCD, so we may plan targeted interventions for these children. Furthermore, by

improving the clinical management of DCD, the risk of secondary impairments in children with DCD and the medical and social costs can be decreased.

In this dissertation, several research questions were addressed to improve the knowledge of the pathology and the development of an assessment system and the potential of clinical subgrouping of children with of DCD. The experimental setup and the results are presented in subsequent chapters. Chapter 2, titled **Central Nervous System Pathologies in Children with Developmental Coordination Disorder: A systematic review**, examined the evidence from brain imaging assessments of children with DCD, in order to summarize the potential CNS pathologies in children with DCD as this has yet to be done within the current literature. In Chapter 3, titled **Motor Planning and Gait Coordination Assessments for Children with Developmental Coordination Disorder**, the primary purpose was to examine the construct validity of the Motor Planning Maze Assessment (Maze) (May-Benson, 2006) and three items from the Functional Gait Assessment (FGA) (Wrisley, Marchetti, Kuharsky, & Whitney, 2004) that were modified for children (pediatric modified FGA, pmFGA), by comparing performance of children with DCD and age-matched peers who were typical developing (TD). The secondary purpose of this study was to examine the construct validity of total scores of the Dynamic Gait Index (DGI) (Lubetzky-Vilnai, Jirikowic, & McCoy, 2011) and the FGA (Wrisley et al., 2004), since currently there is no robust measure of these important constructs as they relate to children with DCD. Chapter 3 titled **Classification of Children with Developmental Coordination Disorders Based on Clinical Subgroups** presents a study focused on developing a clinical measurement system that identifies subgroups in children with DCD. Chapter 5 provides an overarching discussion and interpretation of the collective findings of the studies and outlines directions for future research.

CHAPTER 2

Central Nerve System pathologies in Children with Developmental Coordination Disorder:

A Systematic Review

Abstract

Developmental coordination disorder (DCD) is a neurological disorder that is typically diagnosed in school-age children and affects approximately 5-6% in school-age children in US . Children with DCD may often demonstrate poor motor, sensory and postural control function compared to typical peers. They may show delayed and poor quality of fine or gross motor skills, especially high-level motor skills, and experience challenges in their daily life activities. The presumed central nervous system (CNS) pathology of DCD has not been confirmed. The purpose of this systematic review was to examine the evidence from brain imaging assessments for children with DCD, and to summarize the potential CNS pathologies in children with DCD. A systematic review was conducted in PubMed, CINAHL, Web of Science and PsycINFO in September 2014. The evidence rating systems used in this study were the Oxford Centre for Evidence-based Medicine Levels of Evidence and a scale modified from the American Academy of Cerebral Palsy and Developmental Medicine (AACPD). From the eight articles reviewed, the results indicated that the potential pathologies of DCD related to motor function primarily include prefrontal lobe, frontal lobe, parietal lobe, and cerebellum. The corticospinal tract is involved and the posterior thalamic radiation may also be involved. The basal ganglia is involved related to the attention and executive functions, which can affect motor function as well. However, larger sample sizes and longitudinal studies are needed in order to further confirm these findings. For future research, brain imaging studies that test different motor functions and

explore the relationship between clinical measurements and brain imaging findings would be helpful to better understand the etiologies of DCD and to improve the diagnostic and intervention process for children with DCD.

Introduction

Developmental coordination disorder (DCD) is a neurological disorder that is usually first observed in school-age children. The current incidence of DCD is approximately 5-6% in school-age (5-12 years old) children in US (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker et al., 2012a). Overall, children with DCD may demonstrate poor motor and sensory function compared to typical peers and may show delayed and poor quality of fine or gross motor skills, especially higher-level motor skills, such as hopping, jumping, ball skills, and writing (Blank et al., 2012; Campbell et al., 2012;). They may also demonstrate delayed or poor postural control, such as poor feedback and feedforward control or poor coordination of postural muscles. In addition to the motor limitations, some children with DCD may have problems with their sensory processing functions, such as inadequate information processing (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Green, 2010; Green & Wilson, 2008). These functional limitations not only influence their ability to carry out daily activities, but also affect their participation and quality of life (Campbell et al., 2012; Blank et al., 2012; Green et al., 2011; Green, Baird, & Sugden, 2006; Green, Chambers, & Sugden, 2008).

Currently, the commonly used diagnostic criteria for DCD are from the Diagnostic and Statistical Manual of Mental Disorders- fifth edition (DSM-V) (APA, 2013). The criteria include:

- A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) as well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike, or participating in sports).

B. The motor skills deficit in Criterion A significantly and persistently interferes with activities of daily living appropriate to chronological age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure, and play.

C. Onset of symptoms is in the early developmental period.

D. The motor skills deficits are not better explained by intellectual disability (intellectual developmental disorder) or visual impairment and are not attributable to a neurological condition affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder) (APA, 2013, p. 74).

This definition was originally established in 1987 and revised in 2013 in place of past terms used for children presenting with poor motor coordination, such as ‘clumsy child’, ‘minimal brain damage’, ‘developmental dyspraxia’, etc (APA, 2013). However, as the DSM-V DCD criteria are all symptom dependent, the process to confirm a diagnosis of DCD is made through a relatively complex differential diagnosis process (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a).

The unknown pathology of DCD increases the difficulty of diagnostic process of DCD. The pathology of DCD is still unclear based on what is currently known, but results from clinical studies suggest that the poor motor and sensory performance of DCD may be due to CNS deficits (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a). Based on clinical performance, previous studies indicate that, the parietal lobe, cerebellum and basal ganglia may be involved in children with DCD (Groenewegen, 2003; van Waelvelde et al., 2006; Wilson et al., 2013). Although there are some hypotheses, the presumed CNS pathology of DCD has not been confirmed or systematically summarized. Therefore, the purpose of this systematic review was to

examine the brain imaging assessment evidence for children with DCD, and to summarize current knowledge of the potential CNS pathologies in children with DCD. Based on the findings of the review, suggestions and recommendations for future research are presented.

Methods

Search strategy

A systematic search was conducted in four electronic databases: PubMed, CINAHL, Web of Science and PsycINFO in September 2014. In each database, the search was performed with every possible combination among the following key words: (1) Developmental coordination disorder OR dyspraxia OR deficits in attention, motor control and perception OR coordination dysfunction OR sensory integrative dysfunction OR learning disability and motor impairment OR minimal brain dysfunction OR Asperger OR non-verbal learning disability OR clumsiness OR apraxia; (2) Brain image OR brain scanning OR magnetic resonance imaging OR computed tomography scan OR diffusion tensor imaging; (3) Children. This search was limited to articles written in English, human studies and studies including participants from birth to 18 years-of-age. Because the diagnostic criteria of DCD were defined in DSM-IV-TR or DSM-V, in order to gather specific and valid information for children with DCD, this search was also limited to articles published from January 2001 to September 2014.

Inclusion and exclusion criteria

To be included in this review, studies had to meet the following criteria: (1) the study population included children (≤ 18 years of age) who had DCD based on the DSM-V criteria for DCD (American Psychiatric Association, 2013), and (2) the study included at least one brain

imaging assessment either for the DCD group or for both DCD and a control/other clinical diagnostic group. Studies were excluded from review if the report was a review article, an animal study, or not written in English.

Evidence rating system

The evidence rating system used in this study was the Oxford Centre for Evidence-based Medicine (OCEBM) Levels of Evidence and a scale that was modified from the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) (American Academy of Cerebral Palsy and Developmental Medicine [AACPDM], 2008). As the target articles of this review were brain imaging assessment studies, the Diagnosis part of the OCEBM Levels of Evidence rating system was used. Based on this system, the studies were ranked on 5-point scale from Level 1 (systematic reviews of Level 1 diagnostic studies, validating cohort study with good reference standards or absolute SpPINs and SnNouts) to Level V (expert opinion without explicit critical appraisal or based on physiology, bench research or “first principles”) according to the description in the original scale. It also provides the grades of recommendation, which includes four grades: A (level 1 studies), B (level 2, level 3 or extrapolations from level 1 studies), C (level 4 or extrapolations from level 2 or 3 studies), and D (level 5 evidence or troublingly inconsistent or inconclusive studies of any level) (AACPDM, 2008).

The rating original scale described by AACPDM, which was primarily focused on intervention studies. Studies were ranked on a 5-point scale from Level I (systematic reviews or large randomized clinical trials [$n > 100$]) to Level V (expert opinion) according to the description in the original scale. The components of the AACPDM rating system included: (1)

the extent to which inclusion/exclusion criteria were well-defined; (2) whether the approaches are well defined in both intervention and control group; (3) whether or not appropriate measures were used and if they were valid and reliable; (4) whether assessments were masked from the intervention processes; (5) if statistical evaluation was included; (6) if dropout rate was reported; and (7) if control of confounding variables was reported (AACPD, 2008). In order to make the scale more appropriate for the diagnostic studies in this review it was modified. Components 2 and 4, which are specific for intervention studies, were removed. For example, the question of component 2 is: Was the intervention well described and was there adherence to the intervention assignment? In addition, several sub-questions were added to the original seven components to clarify the validity and reliability. For example, in the original rating system, the third question is “Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?” In order to do more detailed rating, six sub questions were also answered under this question:

(2-1) Were the measures used clearly described?

(2-2) Did the authors report or describe the validity of the measures?

(2-3) Were the measures used valid?

(2-4) Did the authors report or describe the reliability of the measures?

(2-5) Were the measures used reliable?

(2-6) Were the measures used appropriate to represent the outcomes of interest?

The scoring of each sub-question was 0, 0.5, 1 or not available. The sub-question scores were then averaged and the sub-question average score was used within the sum of the total score (Appendix 1). Two authors (SWM and LYH) independently conducted the evidence evaluations.

Data extraction

Data extraction of this study focused on the brain imaging that was applied and imaging findings. The following headings were used to extract the data in the tables of evidence: inclusion and exclusion criteria, comorbidities of target population, participant age and gender, sample size available for analysis, drop-out attrition, details of brain imaging assessment (i.e. type of brain imaging, measurements, and task that participants completed for fMRI and Diffusion Tensor Imaging (DTI) studies), clinical measurements, location of studies (i.e. countries), results (i.e. including brain areas networks that involved, and statistical significance), clinical importance, study conclusions, and special comments (i.e. future recommendations). Data extraction was conducted by one author (LYH).

Results

The study selection process is illustrated in Figure 1. It was conducted by LYH and confirmed by SWM; eight studies were included in the review. The complete quality assessment of the eight included studies was conducted by LYH and SWM, and the result is presented in Table 1. In eight studies three different kinds of brain imaging techniques were used to examine the brain activity in 283 children (104 children with DCD; 17 children who were very preterm infants without DCD concerns; 23 children with ADHD without DCD concerns; 139 children who were peers with typical development) (Atkins et al., 2005; Debrabant, Gheysen, caeyenberghs, van Waelvelde, & Vingerhoets, 2013; de Kieviet et al., 2014; Kashiwagi, Iwaki, Narumi, Tamai, & Suzuki, 2009; Langevin, MacMaster, Crawford, Lebel, & Dewey, 2014; Querne et al., 2008; Zwicker, Missiuna, Harris, & Boyd, 2010; Zwicker, Missiuna, Harris, &

Boyd, 2011; Zwicker, Missiuna, Harris, & Boyd, 2012b; ;). The children ranged in age from 8 to 12 years. The descriptive data from these studies is presented in Table 2 and 3.

Study design

Based on the rating system, all eight articles were Grade B. Of these eight research reports, seven were level 2b evidence (i.e., an exploratory cohort study with good reference standards) (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b), and one was level 3b evidence (i.e., a cohort study without consistently applied reference standards) (Kashiwagi et al., 2009). As all eight were exploratory studies with a relatively small sample sizes for children with DCD (range from 7 to 23), none of these studies could be rated as a validating cohort study (level 1b).

Results of brain imaging

In these eight studies, three brain-imaging techniques were used: structural MRI, functional MRI (fMRI), and DTI. These are three different techniques that can actually be conducted using the same machine but with different protocols. Therefore, some studies attempted to collect all three types of imaging data. Others collected two types of data, usually structural MRI+ fMRI or structural MRI+DTI combinations, at the same time, in order to have a more comprehensive description of brain activity. Generally, the structural MRI provides the basic information (size, and shape) of a brain; the fMRI can provide the brain activation patterns and/or relationship and the DTI can provide more white matter information related to the tracts in a brain (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b).

For brain imaging use, MRI is good at providing a contrast between grey and white matter, so it is a good choice for many conditions of the CNS. The structural MRI provides the static brain images, and the functional MRI helps us to understand how different areas of the brain respond to external stimuli, such as functional tasks (e.g. trial-tracing task). Generally, the structural MRI has better resolution than the fMRI (e.g. structural MRI: 1mm; fMRI: 2-3 mm) (Poldrack, Mumford, Nichols, 2011). The resting images of structural MRI can be a standard reference in the fMRI or DTI study for calibration of the basic brain activities for each participant. Usually, in a MRI study, the measurements include volume/size of specific lobes or areas, the shape of the brain, etc. In an fMRI study, brain activation is the essential (Poldrack et al., 2011; Manenti et al., 2007). The blood oxygenation level dependent (BOLD) is a common fMRI measurement that estimates the hemodynamic response to transient neural activity resulting from a change in the ratio of oxyhemoglobin and deoxyhemoglobin. From the BOLD, areas that are more related to certain functions or tasks can be identified (Poldrack et al., 2011).

DTI is a MRI technique that measures the restricted diffusion of water in tissue in order to produce neural tract images instead of using these data solely for the purpose of assigning contrast or colors to pixels in a cross sectional image (Manenti et al., 2007). Therefore, DTI can usually identify the white matter and the tracts more clearly than with other techniques. DTI measurements include: fractional anisotropy (FA), mean diffusivity (MD), axial diffusivity (AD), and radial diffusivity (RD). FA is a scalar value between 0 and 1 that describes the degree of anisotropy of a diffusion process. Zero means that diffusion is isotropic, which means the water flow is unrestricted (or equally restricted) in all directions. One means that diffusion is fully restricted to occur only along one direction. FA is thought to reflect fiber density, axonal diameter, and myelination in white matter. MD is a measure of the total diffusion within a voxel

(the unit for the 3-dimensional image). AD is the diffusivity on the longest axial, and the RD is the average for the diffusivities on two minor axes (Manenti et al., 2007).

The statistically significant results of all eight studies suggest that compared to their typical peers, and peers without DCD concerns (who were preterm infants), children with DCD demonstrate different patterns of brain activity during motor or attention tasks (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b).⁸ In the three studies that children did fine motor tasks during fMRI assessment, the results show that children with DCD demonstrated statistically significant different patterns of brain activation in parietal lobe, frontal lobe, prefrontal lobe and/or cerebellum. In Kashiwagi's study, children with DCD had less activation in left posterior parietal cortex (i.e., superior parietal lobe and inferior parietal lobe) and postcentral gyrus while doing a fine motor task (Kashiwagi et al., 2009). They also found that there was a negative correlation between the task performance and the changes in left inferior parietal lobe. They found no significant difference between the DCD group and control group in the basal ganglia and cerebellum. However, Zwicker et al. (2010, 2011) found that the DCD group had significantly greater activation in the frontal, parietal, temporal and cerebellar regions (i.e., left inferior parietal lobe, right middle frontal gyrus, right supramarginal gyrus, right lingual gyrus, right parahippocampal gyrus, right posterior cingulate gyrus, right precentral gyrus, right superior temporal gyrus and right cerebellar lobe VI) when doing a tracing task. They also found that compared to the control group, the DCD group significantly under activated the left precuneus, left superior frontal gyrus, right superior temporal gyrus/insula, left inferior frontal gyrus and left postcentral gyrus. When examining the correlation to the motor performance, the activation in the right middle frontal gyrus was negatively correlated with the

number of tracing tasks completed. In a later study Zwicker et al. (2012b) compared brain-imaging variables not only for different groups, but also for pre and post skilled motor practice training. The results showed that the DCD group was under-activating the cerebellar-parietal and cerebellar-prefrontal network (i.e., right inferior parietal lobe, right lingual gyrus, right middle frontal gyrus, left fusiform gyrus, left inferior parietal lobe, right cerebellar crus I, left cerebellar lobe VI and left cerebellar lobe IX). Similar to Kashiwagi's study, there was no significant difference in basal ganglia activation between the DCD and control groups in these two studies (Kashiwagi et al., 2009).

Different from the previous studies using fine motor tasks, Querne et al. (2008) chose an attention task (go-no go task) during fMRI testing to assess the brain activity in children with DCD. The results show that in the right hemisphere, the anterior network connection between the middle frontal cortex (MFC) and the anterior cingulate cortex (ACC) was significantly higher in the control group than in the DCD group. The top-down connection from MFC to the inferior parietal cortex (IPC) was also significantly different between two groups. For the control group, the MFC-IPC tract was in a positive direction, whereas it was in a negative direction in DCD group. Furthermore, the posterior network connection between ACC and IPC was significantly higher in the DCD group than in the control group. The connection between striatum and IPC was close to zero in the DCD group. In the left hemisphere, the top-down network connection from MFC to IPC was negative and significantly higher in DCD group than in control group. Within the posterior network, the value of the path coefficient between ACC and IPC was close to one, indicating a very strong and significant connection in DCD group, but this was not observed in control group. Finally, no path coefficient difference was observed between striatum and IPC in either group.

In Zwicker's DTI study (2012b), the outcome variables were different from the fMRI studies. The results show that the DCD group demonstrated significant lower mean diffusivity in the corticospinal tract than control group. The DCD group also showed a trend of lower mean diffusivity in posterior thalamic radiation ($p < 0.06$) and lower axial diffusivity in both corticospinal tract and posterior thalamic radiation ($p < 0.06$) than control group. There was no significant difference of the fractional anisotropy in all tracts and mean diffusivity in superior and middle cerebellar peduncles between two groups. The correlation of brain imaging results to motor impairment showed that the mean axial diffusivities in the corticospinal tract and posterior thalamic radiation were significantly positively correlated with scores from the Movement Assessment Battery for Children 2 (MABC-2) (Henderson & Sugden, 2007).

Discussion

Study design

There were no level 1b (validating cohort study) studies to include in this review. All of the studies included were exploratory cohort studies with or without good reference standards. The lack of any validating cohort studies may exist due to the challenges of brain imaging for children and difficulties recruiting children with DCD and without other obvious medical comorbidities. With these possible constraints, validating cohort studies may not always be realistic. Given these considerations, conducting high quality, structured exploratory cohort studies may be more appropriate. In this review, all of the eight studies were Grade B by the recommendation rating, which indicates that there were some limitations in study design, such as small sample size, less well-defined measurements and tasks that were used. However, with

caution, the findings provide some direction for assessing CNS pathology in children with DCD and provide some information for clinical applications and future research.

Brain imaging

The statistically significant results of eight studies suggest that compared to children with typical development, children with DCD demonstrate different patterns of brain activity; however, the brain area, the network, and the level of activation were actually slightly different in different studies.

In all eight studies, the brain-imaging techniques used were described, including the operation system, the setting parameters, and the measurements. Five of them also include the analysis software and protocols that were used. In addition, all of these studies provided clear brain image figure samples to assist in explanation of the results. Authors of all of these studies had specific hypotheses as to which areas or tracts of brain they expected to be affected and thus focused on. Only the measurements of the corresponding brain areas tracts were reported. Therefore, we believe the validity and reliability of the brain imaging data collection and data reduction of these studies are good. However, none of these studies reported the motion correction information. Since the population is children, who may move their heads more during the testing process than adults, thus affecting the quality of the data, it would have been better to confirm the motion correction process or provide caution as related to the results.

In Zwicker's study (2010, 2011), children with DCD actually over activated more brain areas to accomplish the trail-tracing task compared to their typical peers. The results showed that the DCD group demonstrated greater activation in frontal, parietal and temporal regions than control group. The control group, instead, relied primarily on the precuneus to support their

motor performance. The activation of inferior parietal lobe has been linked to interpretation of sensory information. The activation of supramarginal gyrus and the lingual gyrus have been linked to visual-motor and visuospatial processing. The posterior cingulate gyrus has been linked to spatial attention. The precentral and parahippocampal gyri have been linked to spatial memory. The cerebellar lobe VI also has been linked to spatial processing (Zwicker et al., 2010, 2011). In summary, in this study, children with DCD relied heavily on visual and spatial processing to complete the trail-tracing task. They relied on information from their visual system to guide motor performance much more than their typical peers. This may be a compensation strategy for their poor sensory feedback from other sensory systems. The DCD group also showed activation in dorsolateral prefrontal cortex (middle frontal gyrus). This area is known as the area of attention control and initial stages of explicit motor learning. This supports that children with DCD might use greater cognitive effort to complete the fine motor task. Generally, children with DCD activate more brain regions, thus they appear to use more effort to accomplish motor performances than their peers. On the other hand, the control group showed greater activation in precuneus (i.e., visuospatial processing and initiation of movement programming), superior and inferior frontal gyrus (i.e., spatially oriented processing and inhibitory control over motor response), postcentral gyrus (i.e., motor control and motor learning), and insula (i.e., motor control and learning and error processing). This may suggest that children who are typically developing demonstrate a more efficient strategy for this visuomotor task (Zwicker et al., 2010,2011). In Zwicker's other fMRI study, which included three practice sessions between pre and post fMRI test, children with DCD also showed different activation patterns in the dorsolateral prefrontal cortex, the inferior parietal lobe and the cerebellum. Therefore, they

suggested that children with DCD might be under-activating the cerebellar-parietal and cerebellar-prefrontal network (Zwicker et al., 2010, 2011).

In contrast, in Kashiwagi's article (2009), children with DCD actually showed less activation in left posterior parietal cortex and postcentral gyrus than typical peers during their visuomotor task. The posterior parietal cortex plays the role to integrate multimodal information relevant to motor control. That is to say, children with DCD can have difficulties in their eye-hand coordination and sensory-motor integration during the visuomotor task. Children with DCD tended to under activate the brain area and showed poor accuracy in the visuomotor task. In this study, no cerebellum or basal ganglia activity was specifically addressed, so they concluded that the pathologies of DCD might be in the left posterior parietal cortex and postcentral gyrus, and not in the basal ganglia or cerebellum. This conclusion is in contrast to Zwicker et al.'s (2010, 2011), who explained that this difference might be due to different tasks that used in the studies. Both of these two research teams used a visuomotor task that used a joystick, but there were different levels of difficulty (Zwicker et al., 2010, 2011). In Kashiwagi's study, the tracking path was a straight line between two points, and there were also watching and resting conditions (Kashiwagi et al., 2009). In Zwicker's study, children traced a flower trail from the MABC-2, and there were no resting periods during testing (Zwicker et al., 2010, 2011, 2012b). Therefore, the demands for continual processing of visuospatial information might be different in these two studies. Since, microstructural changes in brain were observed for children with DCD, it is possible that different patterns of brain activity were due to the minor differences of tasks. In order to confirm the validity of patterns of brain activity, studies collecting several levels of task, such as different complexity levels of trial-tracing tasks, may be needed.

In Querne's study (2008), different from the previous studies that focused on motor tasks, their primary focus was on attention and executive functions. Their results suggest that children with DCD showed stronger path coefficients in the left hemispheric network than in the right hemispheric network. The inhibition of a proponent motor response predominantly involved the left hemispheric network in DCD group. This suggests that children with DCD demonstrated abnormal hemispheric lateralization for attention and inhibitory functions. The control group, however, actually showed a positive influence at the right striatum to IPC tract, which was not fully functioning in DCD group. This suggests that one of the pathologies of DCD may be within the basal ganglia. Furthermore, children with DCD may have impaired capacities to automatize motor behavior with practice and consequently continue to exert top-down control processes during behavior or tasks. Based on the evidence above, Querne et al. (2008) suggest that children with DCD could be characterized by an abnormal brain hemispheric specialization during development. The abnormal hemispheric specialization does not only impact their attention and executive functions. Since the attention control and executive function can also influence motor performance, the abnormal brain hemispheric specialization in children with DCD could be a factor for their poor motor performance. However, in order to confirm this hypothesis, a longitudinal study or a cross-sectional study that includes children with DCD at different ages is needed.

Different from the fMRI studies, Zwicker's DTI study can provide information about the integrity of motor, sensory and cerebellar pathways in children with DCD (Zwicker, et al., 2012b). The results of this study suggest that the DCD group showed significantly less water diffusion in the corticospinal tract. This result is actually contrary to findings in other pediatric populations with neurological impairments, such as cerebral palsy and traumatic brain injury.

Therefore, there appear to be different mechanisms underlying DCD, compared with these other neurological impairments (Querne et al., 2008; Kashiwagi et al., 2009; Zwicker et al., 2010, 2011, 2012b). The results also suggest that this lower diffusivity of the motor pathways was primarily driven by lower axial diffusivity. In other words, the differences between DCD and control groups may exist in the intrinsic characteristics of axons or in the extra-axonal/extracellular space. This low axial diffusivity phenomenon has also been found in children with Autism and children with developmental dyslexia. However, in those studies, the low axial diffusivity was driven by the lower fractional anisotropy (Zwicker et al., 2011, 2012b). In Zwicker's study (2012b), there was no significant difference in fractional anisotropy. Therefore, children with DCD may perform motor skills with similar difficulties to what children with Autism and children with developmental dyslexia do due to the low axial diffusivity of corticospinal tract, but the underlying mechanism may be different. Furthermore, the axial diffusivity was highly correlated to the scores on MABC-2 for all children in this study; therefore, reduced axial diffusivity may play an important role in DCD. Besides the corticospinal tract, Zwicker et al. (2012b) mentioned that even though there was no significant difference in mean diffusivity and axial diffusivity of the posterior thalamic radiation, there was a trend that values of the DCD group were lower than control group. Since the p value was 0.06, they claimed that this non-significant result might be due to the small sample size. This suggests that the sensory areas of the brain in children with DCD may also be involved, but further confirmation is necessary. There was no significant difference of the measurements in superior and middle cerebellar peduncles (Zwicker et al., 2012b). That is to say, there is no microstructural difference along the efferent and afferent pathways contained within these cerebellar peduncles. In Zwicker's fMRI study (2010, 2011), the results suggest that the

cerebellum was involved in children with DCD, suggesting that the cerebellar involvement may be related to the cerebellum itself, but not the connections with other brain areas.

Based on these eight studies, the sites of potential pathologies of DCD that relate to motor function primarily include prefrontal lobe, frontal lobe, parietal lobe, and cerebellum. Corticospinal tract is involved and the posterior thalamic radiation may also be involved, which may affect the sensory processing from the somatosensory system. Basal ganglia is involved related to the attention and executive functions, which can affect the motor function as well. However, larger sample sizes and longitudinal studies are needed in order to further confirm these findings (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b).

Limitations and Recommendations for future research

In this review, the major limitation of the included studies is that the small sample size may have been too small to validate the pathologies of DCD. A possible reason for small sample size may have been due to the difficulty identifying and recruiting children with a DCD diagnosis or perhaps even more, having them participate in brain imaging studies. Also, since the target population of DCD is children, developmental change is important to consider. Therefore, studies with larger sample sizes, longitudinal follow-up studies and/or studies including children with DCD of different ages are necessary to validate the current findings of brain imaging (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b).

In these eight selected studies, five of them used fMRI and three of them was using DTI to assess children with DCD (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014;

Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b). These two brain-imaging techniques actually provide different information about brain activity, thus are necessary to provide a more comprehensive picture of the pathologies of DCD. Since the results of the studies suggest that pathologies of DCD may be in the microstructural abnormalities in brain, studies that use different brain imaging techniques would be helpful to broadly examine the brain in children with DCD. The type of task that children are asked to do during brain imaging likely also affects the study results. In these four fMRI studies, three of them used visuomotor tasks and one of them used an attention task. These can test the fine motor function, attention and executive functions related to some of the concerns in children with DCD (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a). However, children with DCD also demonstrate poor gross motor and/or postural control function (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a).^{1-2,5-6} It would be very helpful, if tasks related to these functions could be undertaken during brain imaging testing. For example, in studies of other populations, participants were asked to do active ankle dorsi-flexion and plantar flexion during fMRI testing to gather data of brain activities during the pretend balance or walking tasks (Goble et al., 2011; Manenti et al., 2007;).

In other studies participants have been asked to mentally practice doing a gross motor or balance tasks (such as walking) during fMRI testing (Debrabant et al., 2013). In order to have more information about brain activity in children with DCD related to different functions, it might be beneficial to examine brain activity during these types of movements or mental practice.

Children with DCD also show problems with sensory and sensory integrated postural control.

Therefore, with the constraints of the techniques, brain imaging during tasks such as providing

visual, proprioceptive or vestibular stimulus to interfere with their sense of balance may reveal helpful information (Belforte & Eula 2012; Goble et al., 2011; Nutt, Horak, & Bloem, 2011).

In all Zwicker's studies (2010, 2011, 2012b), they used the < 5 percentile cutoff of MABC-2 to include children with DCD, but in Langevin and Debrabant's studies, they actually suggested that it would be worth to set the cutoff at < 15 percentile. Zwicker also recommended that the results of children whose MABC-2 scores are in 6 to 15 percentile might be worth evaluating separately (still defined as children with DCD concern by MABC-2). The motor function level of children with DCD actually demonstrates various symptoms. For example, some of them may have more impaired fine motor, and some of them may have more impaired postural control ability. Not every child with DCD has the same performance and challenges. Therefore, it may be helpful to have studies that explore the relationship between brain activity patterns and the children's clinical performance. Perhaps in the future, subtypes of DCD can be used to classify and study both clinical measurements and brain imaging confirmation (Green et al., 2008).

Clinical application

The results of this review suggest that children with DCD demonstrate different patterns of brain activity than children without DCD to accomplish several specific types of tasks. This suggests that for children with DCD, the most efficient motor and learning strategies may be different from strategies for typical developing children. For example, Zwicker (2010, 2011, 2012b) suggests that children with DCD rely more on information from the visual system to finish or learn a task and that children with DCD seem to exert great effort and may experience fatigue with motor-based activities. Therefore, for clinical practice, intervention programs should

consider including carefully planned visual cues, providing training of other sensory systems or adjusting the intensity and duration of an intervention session.

Conclusion

There is moderate to strong evidence to suggest that children with DCD actually demonstrate different patterns of brain activity compared to their typical peers during specific tasks. The potential pathologies of DCD include differences in activation of the prefrontal lobe, frontal lobe, parietal lobe, and cerebellum. The corticospinal tract shows low axial diffusivity, which is correlated with motor skill ability and the posterior thalamic radiation may also be involved, which may affect the sensory processing from the somatosensory system. The basal ganglia activity in children with DCD differs during attention and executive function tasks (Atkins et al., 2005; Querne et al, 2008). Studies with larger sample size, longitudinal studies, studies including participants of different ages or with different functional limitations, and studies using different types of tasks during brain imaging are needed to validate the pathologies of DCD.

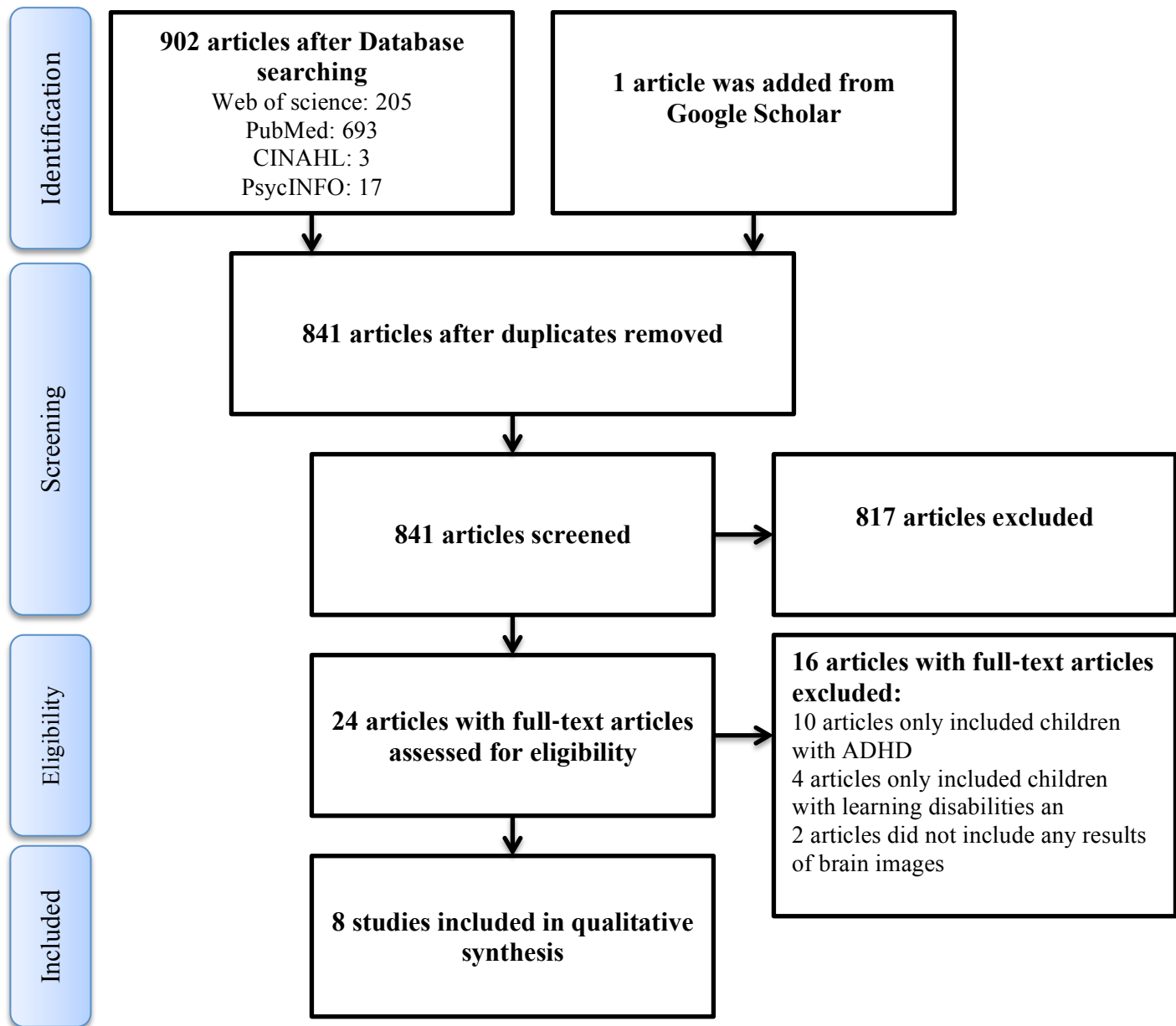


Figure 1. Flow Chart of Article Selection Progress

Table 1. Levels of Evidence and Quality Ratings of Extracted Studies

Study	Level of evidence (OCEBM)	Grades of Recommendation (OCEBM)	Quality scores (modified scale)
Querne et al., 2008	2b Cohort study with good reference standards	B	10.5
Kashiwagi et al., 2009	3b Cohort study without consistently applied reference standards	B	10
Zwicker et al., 2010	2b Cohort study with good reference standards	B	14.5
Zwicker et al., 2011	2b Cohort study with good reference standards	B	13
Zwicker et al., 2012b	2b Cohort study with good reference standards	B	13.5
de Kieviet et al., 2014	2b Cohort study with good reference standards	B	16
Debrabant et al., 2013	2b Cohort study with good reference standards	B	14.5
Langevin et al., 2014	2b Cohort study with good reference standards	B	11

Table 2. Extracted Research Evidence (Participants of studies)

Study	Sample size(n)	Age (year) Mean(SD)	Gender (M/F)	Comorbidities
Querne et al., 20	DCD: 9 Control: 10 Total: 19	DCD: 9.9 (1.8) Control: 10.0 (1.1)	DCD: 2F 7M Control: 3F 7 M	Did not mention
Kashiwagi et al., 2009	DCD: 12 Control: 12 Total: 24	DCD: 10.8(1.0) Control: 10.4(1.0)	DCD: 12M Control: 12M	ADHD: 3 Developmental dyslexia: 3 ADHD+ dyslexia: 2
Zwicker et al., 2010	DCD: 7 Control: 7 Total: 14	DCD: 10.8(1.5) Control: 10.9(1.5)	DCD: 1F 6M Control: 3F 4M	None (excluded ADHD)
Zwicker et al., 2011	DCD: 7 Control: 7 Total: 14	DCD: 10.8(1.5) Control: 10.9(1.5)	DCD: 1F 6M Control: 3F 4M	None (excluded ADHD)
Zwicker et al., 2012b	DCD: 7 Control: 9 Total: 16	DCD: 10.8(1.5) Control: 10.75(1.6)	DCD: 1F 6M Control: 3F 6M	None (excluded ADHD)
de Kieviet et al., 2014	DCD: 13 Very preterm no DCD: 17 Term control: 47 Total: 77	DCD: 8.6 (0.3) Very preterm no DCD: 8.7(0.3) Term control: 8.7(0.5)	DCD: 6F 7M Very preterm no DCD: 11F 6M Term control: 25F 22M	Did not mention
Debrabant et al., 2013	DCD: 17 Control: 17 Total: 34	DCD: 9.4 (0.6) Control: 9.2(0.9)	DCD: 3F 14M Control: 3F 14M	None (excluded ADHD)
Langevin et al., 2014	DCD: 9 DCD+ADHD: 23 ADHD: 23 Control: 26 Total: 85	DCD: 12.22 (2.68) DCD+ADHD: 11.39(2.89) ADHD: 11.78(2.99) Control: 11.58(3.18)	DCD: 2F 7M DCD+ADHD: 4F 19M ADHD: 3F 24M Control: 12F 14M	ADHD

Table 3. Extracted Research Evidence (Measurements, procedures and results)

Study	Brain imaging	Task that applied	Clinical measurements	Results
Querne et al., 2008	fMRI	Go-no go task	<ul style="list-style-type: none"> • Intelligence score: <ul style="list-style-type: none"> -WISC-III -K-ABC • Visuospatial score: <ul style="list-style-type: none"> -NEPSY • Executive function attention score: <ul style="list-style-type: none"> -Stroop • Sensorimotor score: <ul style="list-style-type: none"> -Rey figure -NEPSY -Grooved pegboard -Purdue pegboard 	<ul style="list-style-type: none"> • R't hemisphere: <ul style="list-style-type: none"> ○ DCD: <ul style="list-style-type: none"> ▪ MFC-ACC: weak ▪ ACC-IPC: middle ▪ IPC-MFC: important ○ Control: <ul style="list-style-type: none"> ▪ MFC-ACC: middle ▪ MFC-striatum: middle ▪ MFC-IPC: weak ▪ ACC-IPC: weak ▪ Striatum-IPC: weak; • L't hemisphere: <ul style="list-style-type: none"> ○ DCD: <ul style="list-style-type: none"> ▪ MFC-ACC: weak ▪ IPC-MFC: strong, positive ▪ MFC-IPC: strong, negative ○ Control: <ul style="list-style-type: none"> ▪ MFC-ACC: middle ▪ MFC-striatum: middle ▪ IPC-MFC: non-existent
Kashiwagi et al., 2009	fMRI	Visually guided tracking task: -Tracking condition -Watching condition -Resting condition	<ul style="list-style-type: none"> • RCPM • MABC • Soft neurological signs • Interview scores • WISC-III IQ 	<ul style="list-style-type: none"> • DCD: less activation in L't hemisphere: Posterior parietal cortex and postcentral gyrus; • Correlation: performance-maximal signal changes: Negatively correlated at left inferior parietal lobe ($r=-0.413$)

Zwicker et al., 2010	fMRI	Flower trail-tracing task using a joystick	<ul style="list-style-type: none"> • MABC-2 • DCDQ • KBIT-2 • CADS 	<p>Brain area activation:</p> <ul style="list-style-type: none"> • DCD>TD: <ul style="list-style-type: none"> ○ L't inferior parietal lobe ○ R't middle frontal gyrus ○ R't supramarginal gyrus ○ R't lingual gyrus ○ R't parahippocampal gyrus ○ R't posterior cingulate gyrus ○ R't precentral gyrus ○ R't superior temporal gyrus ○ R't cerebellar lobe VI • TD>DCD: <ul style="list-style-type: none"> ○ L't precuneus ○ L't superior frontal gyrus ○ R't superior temporal gyrus/insula ○ L't inferior frontal gyrus ○ L't postcentral gyrus; • Correlation: <ul style="list-style-type: none"> ○ DCD: activation in the right middle frontal gyrus negatively correlated with number of traces completed
Zwicker et al., 2011	fMRI	Flower trail-tracing task using a joystick (between two sessions of testing, there were 3 sessions of skilled motor practice: tracing task outside the scanner during sitting)	<ul style="list-style-type: none"> • MABC-2 • DCDQ • KBIT-2 • CADS 	<ul style="list-style-type: none"> • DCD: lower percent signal change at both practice and retention in: <ul style="list-style-type: none"> ○ R't inferior parietal lobe ○ R't lingual gyrus ○ R't middle frontal gyrus ○ L't fusiform gyrus ○ L't inferior parietal lobe ○ R't cerebellar crus I ○ L't cerebellar lobe VI ○ L't cerebellar lobe IX • Under-activation of the cerebellum at retention

Zwicker et al., 2012b	DTI	None	<ul style="list-style-type: none"> • MABC-2 • DCDQ • KBIT-2 • CADS 	<ul style="list-style-type: none"> • Fractional anisotropy: <ul style="list-style-type: none"> ○ No differences: <ul style="list-style-type: none"> ▪ Corticospinal tract ▪ Posterior thalamic radiation ▪ Cerebellar peduncles • Mean diffusivity: <ul style="list-style-type: none"> ○ DCD significantly < TD: <ul style="list-style-type: none"> ▪ Corticospinal tract ○ DCD < TD (p<0.06): <ul style="list-style-type: none"> ▪ Posterior thalamic radiation ○ No differences: <ul style="list-style-type: none"> ▪ Superior cerebellar peduncles ▪ Middle cerebellar peduncles • Axial diffusivity: <ul style="list-style-type: none"> ○ DCD < TD(p<0.06): <ul style="list-style-type: none"> ▪ Corticospinal tract ▪ Posterior thalamic radiation • Correlations with motor impairment: • Mean axial diffusivity correlated with M-ABC-2: <ul style="list-style-type: none"> ○ Corticospinal tract(r=0.56), ○ Posterior thalamic radiation(r=0.70)
de Kieviet et al., 2014	MRI DTI	None	<ul style="list-style-type: none"> • MABC • WISC-III 	<ul style="list-style-type: none"> • Fractional anisotropy: <ul style="list-style-type: none"> ○ No differences: <ul style="list-style-type: none"> ▪ Corticospinal tract ▪ Posterior thalamic radiation ▪ Cerebellar peduncles • Mean diffusivity: <ul style="list-style-type: none"> ○ DCD significantly < TD: corticospinal tract ○ DCD tend to be < than TD(p<0.06): posterior thalamic radiation ○ No differences: <ul style="list-style-type: none"> ▪ Superior cerebellar peduncles ▪ Middle cerebellar peduncles

Debrabant et al., 2013	fMRI	Predictive motor timing task	<ul style="list-style-type: none"> • MABC-2 • WISC-III 	<ul style="list-style-type: none"> • Axial diffusivity: <ul style="list-style-type: none"> ○ DCD tend to be < TD: ○ Corticospinal tract ○ Posterior thalamic radiation • Correlations with MABC-2: <ul style="list-style-type: none"> ○ Mean axial diffusivity: <ul style="list-style-type: none"> ▪ Corticospinal tract($r=0.56$) ▪ Postero thalamic radiation($r=0.70$) • TD: <ul style="list-style-type: none"> ○ Higher activation in the right dorsolateral prefrontal cortex (DLPFC) and right inferior frontal gyrus (IFG) for responses at unpredictable as opposed to predictive ISIs • DCD: <ul style="list-style-type: none"> ○ Non-differentiable. ○ Less activation than TD in the right DLPFC, the left posterior cerebellum (crus I) and the right temporo-parietal junction (TPJ) for this contrast. ○ Activation in the right temporo-parietal junction (TPJ) positively correlated with RT as an indicator of processing load in both groups.
Langevin et al., 2014	DTI	None	<ul style="list-style-type: none"> • MABC-2 • WASI • Developmental NEuroPSYchological Assessment II • The McCarron Assessment of Neuromuscular Development • Neurodevelopmental index 	<ul style="list-style-type: none"> • Fractional anisotropy: <ul style="list-style-type: none"> ○ Reduction in the frontal regions of the corpus callosum for ADHD ($p = .039$) ○ Reductions in regions of the corpus callosum underlying parietal brain regions for DCD ($p = .040$), as well as the left superior longitudinal fasciculus ($p = .026$). • White matter integrity was impacted in both frontal and parietal regions for DCD+ADHD ($p = .029$; $.046$).

CHAPTER 3

Motor Planning and Gait Coordination Assessments for Children with Developmental Coordination Disorder

Abstract

PURPOSE: The primary purpose of this study was to examine the construct validity of the Motor Planning Maze Assessment (Maze) and three items from the Functional Gait Assessment (FGA) that were modified for children (pediatric modified FGA, pmFGA), by comparing performance of children with DCD and age matched peers who are typical developing (TD). The secondary purpose of this study was to examine the construct validity of total scores of the entire Dynamic Gait Index (DGI) and the FGA.

METHODS: Nineteen pairs of children with DCD and TD, age from 5-12 years, participated in this study. The Movement Assessment Battery for Children, 2nd Ed (MABC-2) was used to verify the group membership. Children in both groups were tested on the Maze, pmFGA DGI, and FGA. Paired t-tests and agreement tables were used to compare the motor performances between children with DCD and TD.

RESULTS: Children with DCD showed significantly higher (less efficient) summary scores in the Maze ($p < 0.001$). They also took significantly longer to finish the first and second items of the Maze ($p = 0.01, 0.03$). For gait coordination, children with DCD also demonstrated significantly fewer steps ($p \leq 0.001$) while doing the pmFGA items. However, the quality scores demonstrated minimal differences between the two groups on all three pmFGA items. Children with DCD also showed significantly lower DGI and FGA total scores ($p < 0.001$).

CONCLUSION: The Maze, pmFGA, DGI, and FGA tests are easily administered in clinical settings and can differentiate motor planning and gait coordination between children with DCD and with TD, thus construct validity is supported.

Introduction

Developmental Coordination Disorder (DCD) is a chronic condition involving impairment in gross motor, postural, and/or fine motor performance that affects children's ability to perform the skilled movements necessary for daily living, including the performance of academic and self-care tasks (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011). Children with DCD represent a large (5-6% of children) under-served population in the US (Campbell et al., 2012). Compared to other neurological diseases with obvious motor limitations (e.g. cerebral palsy), children with DCD usually demonstrate mild or subtle difficulties during daily activities. While these children are able to do typical functional movements, they have difficulty with complex motor activities that may lead to loss of social acceptance for participation in community sports programs and decreased physical activity, followed by increased risk for obesity, poor health fitness, and social isolation (Edwards et al., 2011; King, Harring, Oliverira, Clark, 2011; Zwicker et al., 2012a). Parents, teachers, and medical professionals often overlook the challenges children with DCD face in their daily life (Blank et al., 2012; Green, 2010). While their motor problems are treatable and the prognosis can be promising if intervention occurs early in their development, controversy exists on appropriate examinations and intervention techniques (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Missiuna, Mandich, Polatajko, & Malloy-Miller, 2001). Therefore, it is necessary to continue research related to identifying appropriate assessments and interventions for children with DCD. Assessments for children with DCD, due to their more subtle motor problems must include not only general motor examinations but also specific assessments for motor-related impairments, such as motor planning, postural control, motor coordination, and sensory integration (Edwards et al., 2011; Fong et al., 2011; Green et al., 2011; Zwicker et al., 2012a;).

The overarching goal of this research was to identify valid and sensitive measurements that may be beneficial for children with DCD, in order to better direct more efficient and effective intervention. Current challenges of assessing children with DCD include lack of information about the pathology and the large variety of subtle symptoms associated with DCD. The Movement Assessment Battery for Children (MABC-2) (Henderson & Sugden, 2007; Chow & Henderson, 2003) and the Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2) (Bruininks & Bruininks, 2005) are measurements widely suggested for use in children with DCD in order to identify their motor problems including fine motor, gross motor, balance, and coordination. However, since children with DCD usually demonstrate specific subtle differences in their motor performance, measurements focusing on single domains may be more helpful for identifying children's limitations and developing appropriate intervention plans. In this study, we focused on measurements in two domains in which some children with DCD demonstrate very subtle limitations: motor planning as tested by the Motor Planning Maze Assessment (Maze) (May-Benson, 2006) and coordination of gait under varied sensory conditions as tested by three different gait coordination tests; the Dynamic Gait Index (DGI) (Lubetzky-Vilnai et al., 2011), the Functional Gait Assessment (FGA) (Wrisley et al., 2004), and three items from the FGA that were modified for children (pediatric modified FGA, pmFGA).

The Motor Planning Maze Assessment (Maze) (May-Benson, 2006) is a measurement tool that examines children's upper extremity motor planning ability and spatial skills. It includes three different tasks in which children are asked to manipulate a grommet on a wire to make the grommet move across three different shapes of the wire (a Maze) to complete the task. The Maze test is scored by both quality of the movements and speed (the actual movement time) to address children's motor planning ability. The Maze test was found to be an acceptable and quick clinical

test for children with motor problems and children with sensory integration dysfunction from 4 to 13 years old (May-Benson, 2006). However, it has not been widely examined or used in children with DCD.

The Dynamic Gait Index (DGI) (Lubetzky-Vilnai et al., 2011) and the Functional Gait Assessment (FGA) (Wrisley et al., 2004) are two measurements that have been used to test coordination of gait under varied sensory conditions in adults. Both tests include measurements of walking under different conditions such as walking with different speeds and walking while turning the head. One previous study suggested that the DGI could differentiate between children with fetal alcohol spectrum disorder (FASD) with mild motor limitations and typical peers (Lubetzky-Vilnai et al., 2011). However, based on further as yet un-published results within our research team, we found that the DGI may not be sensitive enough to pick up subtle motor limitations during walking and change across time or after therapy in children with FASD. Therefore, in addition to using the complete DGI and FGA, we chose to examine three challenging items from the FGA, which had been modified for use in children (pmFGA). Within this research we evaluated whether or not the DGI, the FGA and three items of pmFGA were sensitive enough to identify gait coordination problems in children with DCD as compared to TD. The three pmFGA items selected were: 1) narrow-base (heel-toe on a line) walking (NBW), 2) walking forward with eye closed (ECW), and 3) backward-walking (BW). (Lubetzky-Vilnai et al., 2011; Wrisley et al., 2004;)

The primary purpose of this study was to examine the construct validity of the Maze and pmFGA by comparing performance of children with DCD and their typical peers. We hypothesized that the Maze and the pmFGA would differentiate motor planning performance and gait coordination between children with DCD and their typical developing peers. We expected

children with DCD would have higher Maze quality scores (less efficient) and longer time to finish the Maze items, and lower pmFGA quality scores (less efficient) with fewer steps before loss of balance, compared to their typical developing peers. A secondary purpose of this study was to further examine the total scores of the DGI and the FGA. We hypothesized that the DGI and the FGA would not be significantly different between the two groups.

Method

Sample

A convenience sample of 19 pairs of children (19 children with DCD and 19 age-matched peers with typical development (TD)) participated. The study was reviewed and approved by the university Institutional Review Board. All parents and children signed approved consent and assent forms, respectively, prior to participating in the study.

Because DCD is not routinely diagnosed in the US currently, we recruited participants in two ways. We contacted local therapy centers and asked therapists to inform any of their clients who matched our inclusion criteria about the study. We also screened elementary school-age children within a local school district in order to locate and recruit potential participants for the study. We used the DCD Questionnaire (DCDQ) (Wilson et al., 2009) to screen children for potential DCD concerns. The DCDQ has reported sensitivity and specificity of 84.6% and 70.8%, respectively (Wilson et al., 2009). The DCDQ is a standardized questionnaire of 15 questions, in which parents provide their impression of their child's sensorimotor abilities during various activities. Once scored, the DCDQ is used to identify children who may need further testing to discern a diagnosis of DCD.

Inclusion criteria for children with DCD were: 1) DCDQ scores that indicated DCD concerns (5-7 years: < 47 points; 8-9 years: < 56 points; 10 -12 years: < 58 points) (Wilson et al., 2009) or a medical diagnosis of DCD; 2) identification of motor problems via the Movement Assessment Battery for Children, 2nd Ed (MABC-2) (percentile rank lower than 15% in at least one composite of MABC-2); and 3) age 5 years 0 months to 12 years 11 months (elementary school age) (Henderson & Sugden, 2007). For their peers with TD, we recruited children without any motor problems based on DCDQ screening and matching their age (+/- 6 months) with children with DCD. Children in both groups were excluded if they had an IQ <60, other motor related medical diagnosis (e.g. cerebral palsy), a history of serious head injury or seizures, a visual acuity impairment not corrected by glasses, or a report of any lower limb or back injury within the previous six months.

Procedures

After the screening, children in both groups participated in a 1.5 hours test session in either the human motion analysis lab at the university or the clinics or institutions from which they were referred. Child tests were administered in the same order for all participants and included: 1) Growth parameters and sensorimotor screening measures (for general descriptive purposes); 2) Movement Assessment Battery for Children (MABC-2) (Henderson & Sugden, 2007) (gross/fine motor ability to verify DCD diagnosis and TD); 3) the Motor Planning Maze Assessment (Maze) (May-Benson, 2006) (motor planning); 4) gait assessments (including the three pmFGA items selected from FGA (gait coordination), the complete Dynamic Gait Index (DGI), the complete Functional Gait Assessment (FGA) (Lubetzky-Vilnai et al., 2011; Wrisley et al., 2004;).

Instrumentation

1. Growth parameters and Sensorimotor Screening Measures: Height and weight were recorded using a tape measure of the top of the head marked on a wall and a standard weight scale, respectively. For the brief sensory screening, we applied light touch, light pressure and examined proprioception as clinical measures of peripheral sensation. While sitting on a chair with eyes closed, the child was asked to report the sensation in response to a series of light touches (cotton ball) and light pressure (examiner finger) on each lower leg. For proprioception, the examiner completed a limb position test by placing one leg in a position and having the child match the position with the other leg and a limb movement test where the examiner moved one of the child's legs in a pattern and the child attempted to match the movement with other leg. The clinical screening of strength, range of motion, and posture, were tested while the child was in supine and prone. The child was asked to move his/her legs in all planes of movement. If full range was not achieved volitionally, the examiner applied gentle overpressure to determine if full range of joint movement was available. The child was scored as having typical or atypical strength/range for each movement. Standing posture was observed and the presence or absence of scoliosis, kyphosis, lordosis, leg length difference, and standing foot pronation was noted (Kaufman & Schilling, 2007; Shumway-Cook & Woollacott, 2007;).

2. Movement Assessment Battery for Children-2nd edition (MABC-2): The MABC-2 was designed to detect motor skill impairments in children aged 3-16 years. The child is asked to do 8 common child gross and fine motor activities, such as balance on one foot or trace a line through a path on a piece of paper. Test results yield scores for Total Impairment, Manual Dexterity, Ball Skills, and Balance (static and dynamic). Reliability and validity of the MABC-2 is considered

moderate to good based on studies reported in the test manual (test-retest reliability: ICC= 0.49-0.91; inter-rater reliabilities: ICC= 0.92- 1.00) (Henderson & Sugden, 2007).

3. The Motor Planning Maze Assessment (Maze): This test measures the motor planning in children from 4 years old to teens. Motor planning abilities are operationally defined as the child's ability to manipulate a maze (wire pattern on a handle or handles) to move a grommet from the beginning to the end of the wire pattern. The test consists of three items, each consisting of a specific maze that is scored based on the child's task completion time and demonstration of errors (quality). The first two mazes are completed using the child's dominant hand, and the third maze is completed using both hands (May-Benson, 2006). (See measurement scales in Appendix 2.)

4. Dynamic Gait Index (DGI): This test involves walking under eight conditions to test dynamic balance during challenging vestibular stimulation. Walking 20' while turning head to right and left, stepping over obstacles, walking then pivoting around quickly are three representative items on the test. The DGI is scored using a 0-to-3 scale. Reliability and validity have been demonstrated in children with FASD (test-retest reliability: $r=0.96$; inter-rater reliability: $r=0.96$) (Lubetzky-Vilnai et al., 2011). (See full test in Appendix 3.)

5. Functional Gait Assessment (FGA): This test involves ten functional walking skills during challenging conditions. There are seven items from the DGI, but the scaling criteria are different. FGA includes more detailed parameters in the scoring criteria, such as the distance a person travels before deviations occur. In addition, the FGA includes some more challenging items than the DGI including walking with closed eyes, walking within a narrow space, and walking backwards. The FGA was designed for adults with neuromotor diseases, such as stroke and

lower limb amputation. To the author's knowledge, it has not been used for children with significant motor problems (Wrisley et al., 2004). (See full test in Appendix 4.)

6. Pediatric-modified Functional Gait Assessment (pm-FGA): In order to develop an efficient and sensitive gait assessment for children with DCD, three challenging items from FGA were selected and modified for use with children in this study. The three selected items were: (1) narrow base walking (NBW), (2) walking with eyes closed (ECW), and (3) walking backward (BW). Children were asked to walk with a comfortable speed under these specific situations. We calculated how many steps children could walk before loss of balance or inability to follow the commands (e.g. did not walk heel to toe). We also recorded a quality score from 0 to 3 (Wrisley et al., 2004). (See Appendix 5 for scoring details.)

Analysis

Descriptive statistics were used to describe the clinical presentation and sensorimotor behaviors of children with DCD and their age-matched peers with TD. Paired t-tests were used to examine the differences of the clinical presentation between two groups. Paired t-tests or agreement tables, as appropriate based on the level of measurement and score distributions, were used to compare the time and quality scores for the Maze, the pmFGA, the total DGI and total FGA between children with DCD and with TD.

Results

Table 1 shows demographic information, sensorimotor screening measures, and the motor performance (MABC-2) by group. Overall there were no statistically significant differences

between the groups on the screening measures for strength, range of motion and sensation.

Children showed statistically significant differences in all parts of MABC-2.

Maze (Motor planning)

We separated the Maze outcomes into three different types of measurement: the speed (time to complete each item), the quality score (scores that describe the quality of movement), and the total summary score from the original design of the Maze. Table 2 shows the descriptive data of all measurements for both groups. The speed of Maze revealed statistically significant differences between the two groups in the items that required one hand to manipulate the grommet on the wire (item 1, item 2: $p=0.01, 0.03$). Children with DCD took a longer time to finish the items than children with TD. For the third item, which required children using both hands to manipulate the bead on the wire, there was no significant difference ($p=0.43$) between groups. For the quality score, on all three items the percentage of scores of children with DCD was higher than scores of TD group (item 1, item 2, item 3: 94.73%, 78.95%, 84.21%). Children with DCD also demonstrated statistically significantly higher (less efficient) summary scores in Maze compared to the TD group ($p<0.001$).

Figure 1a-1c shows the individual quality score differences within each DCD-TD pair of children (scores of TD – scores of DCD) for all three items of Maze. These figures present an interpretation of the construct validity of the Maze items to differentiate children with DCD and TD. Most of the differences between DCD and TD pairs fall above the 0 line, indicating that most children with DCD had higher scores (indicating lower quality) compared to their peers with TD, especially in the first two items of Maze (Figure 1a-1b).

pmFGA, DGI, FGA (Gait coordination)

For the gait assessments, we also have several different types of measurement: pmFGA- STEPS (steps children could do before losing balance or deviating from the specific movements, such as not walking heel-to-toe in NBW), pmFGA QUALITY (scores that describe the quality of movement), and the total summary score from the originally designed DGI and FGA. Table 2 shows the descriptive data of all the measurements for both groups.

The pmFGA-STEPS showed statistically significant differences between the two groups in all three items (NBW, ECW, BW: $p=0.001$, $p<0.001$, $p<0.001$). Children with TD were able to walk farther without errors than children with DCD. However, for the quality score for each item, only about half of the children with TD showed better scores than children with DCD (percentage of children with TD who had higher scores than children with DCD: NBW (52.6%), ECW (63.2%), BW (52.6%)).

Figures 1d-1f display the individual score differences within DCD-TD pairs of children (scores of TD – scores of DCD) for all three items of pmFGA. These figures present an interpretation of the construct validity of the pmFGA items to differentiate children with DCD and TD. A slight majority of the differences between DCD and TD pairs fell above the 0 line. However, a number of pairs showed no difference (fell on the 0 line).

On the total scores of the DGI and the FGA, children with DCD demonstrated significantly lower summary scores compared to the TD group (DGI: $p<0.001$; FGA: $p<0.001$).

Discussion

As our primary research question, we explored the construct validity of a motor planning test, the Maze, and a gait coordination test, the pmFGA, to differentiate children with DCD and children with TD. The results supported our hypothesis that generally, both the Maze test and the

pmFGA items are sensitive enough to differentiate between the two groups of children in our study. Compared to children with TD, children with DCD performed with slower and had lower quality motor planning skills on the Maze. Children with DCD demonstrated less coordinated control during the pmFGA. We also evaluated whether or not the total DGI and FGA tests were sensitive enough to differentiate between the groups. Unlike our hypothesis, both the DGI and the FGA demonstrated differences between the children with DCD and TD in our study.

Maze (Motor planning)

Children with DCD showed significantly slower speed than peers with TD while doing the items that required only their dominant hands. There was no significant speed difference in the item 3 maze that required bilateral manipulation. This may reflect that fine motor limitations in children with DCD are more obvious in unilateral tasks, such as writing. It may also imply that assisting with the other hand, or a bilateral approach may be a good strategy to compensate or improve children's fine motor abilities. However, the bilateral manipulation item was the third item tested of the Maze. Therefore, it is possible that there was a learning effect from the testing order. Potentially, because of the learning experience, the speed differences between groups were lower in the last item. Overall, our findings suggest that the Maze items have some construct validity support, and should be considered for inclusion in evaluation of motor planning in children with DCD.

Gait coordination

pmFGA

Children with DCD demonstrated significantly fewer steps before loss of balance than children with TD in all three walking conditions. However, only about half of children with TD

demonstrated better quality scores compared to children with DCD during all three walking conditions. This implies that the number of steps is a better measurement to differentiate the gait coordination in children with DCD and TD than the quality rating used for this measure. This suggests that even though children with DCD showed more problems in gait coordination, some of them could present quality of movement similar to children with TD before they lost their balance. The recording the number of steps while doing these challenging gait items is recommended for inclusion in an evaluation of complex gait coordination in children with DCD.

DGI, FGA

The summary scores of both the DGI and FGA tests were sensitive enough to differentiate gait coordination between the children with DCD and children with TD in this study. The maximum summary score for DGI is 24 points for eight items. In a previous study using the DGI on children with FASD with relatively minor motor problems, researchers also found that the DGI was sensitive enough to show children's motor problems (Lubetzky-Vilnai et al., 2011). However, in our unpublished data on intervention in children with FASD, the DGI items were not sensitive enough to reflect differences pre and post intervention, that were found using other motor assessments. There were only 2 children with FASD (out of 23) having lower than 20 points on the DGI before the intervention. This raises concerns about the difficulty level of DGI and a potential ceiling effect for children with minor motor dysfunction. In this study, all children with TD scored over 20, and eleven scored 24 points. In the DCD group, there were 15 children scoring over 20, and three of them received the maximum 24 points. This again indicates that the DGI may not be challenging enough for children with subtle motor issues. Therefore, DGI should be used with caution in children with minor motor problems.

Compared to DGI, the FGA adds the more challenging items that we examined further (pmFGA). The maximum summary score for FGA is 30 points for eight items (Wrisley et al., 2004). This may then decrease a ceiling effect and increase the sensitivity for reflecting change after intervention. In this study, all children with TD scored over 26, and ten scored 30 points. In the DCD group, there were 7 children scoring over 26, and only one of them received the maximum 30 points. There were four children with DCD receiving points under 20. This indicates that compared to the DGI, the FGA may be challenging enough to differentiate children with subtle motor issues. Our results also support that the full FGA can significantly differentiate the gait coordination differences between children with DCD and children with TD. Nonetheless, the FGA was designed for adults and the scoring criteria were set for the adult population. Therefore, prior to future use of the FGA in children further modification and development needs to be considered.

Limitations

There were several study limitations. We had 19 pairs of children in this study age 5 to 12 years, which may make our sample too small and diverse to fully answer the question of construct validity of the Maze and Gait coordination tests for all children with DCD. These assessment tools are not standardized or age-corrected. In this study, we did consider our results within three different age bands based on the MABC-2 age bands (5-6 years, 7-10 years, 11-12 years) (Figure 1a-f, different symbols representing the three age groups). Within this small sample, there was no obvious difference among the age groups. However, it is still possible that the assessment validity varies with age. Therefore, further research with larger samples and examination of different age groups is recommended. Due to our smaller sample size, we did not

use the Kappa test to test the statistical hypothesis of the quality scores for the measurements. Therefore, the reported sensitivities (percentage of differentiation between two groups) are uncorrected for chance. Finally the rater was not blind to the children's diagnosis. Further studies with larger samples are recommended to confirm differences between children with DCD and TD and explore other psychometrics of these two promising measurements.

Conclusion

Our results suggest that construct validity was supported for the Maze test, the pmFGA, the DGI, and the FGA in children with DCD, based on different performances between children with DCD and age-matched children with TD. These tests are quick clinical assessments that can be simply applied in various settings. Although, more accurate construct validity in different age bands and larger samples of children are needed in order to confirm the result, it is recommended that the Maze and FGA may be helpful in identifying motor planning and gait coordination deficits in children with DCD. Further research is warranted.

Table 1: Personal and Demographic Characteristics by Group

CHARACTERISTIC	DCD (n=19)	TD (n=19)
Age in months, Mean (SD)	104.8 (28.1)	103.6 (26.2)
Sex, % of females	36.8	36.8
Body height (cm), Mean (SD)	136.7 (15.8)	132.4 (12.6)
Body weight (lb), Mean (SD)	74.9 (33.7)	67.3 (17.6)
Posture problems, %		
Foot pronation	5.3	0.0
Range of motion limitation, %	0.0	0.0
Muscle weakness, %	5.3	0.0
Peripheral sensation problems, %	0.0	0.0
M-ABC2 scale scores, Mean (SD)		
Manual dexterity	4.6 (2.0)*	9.3 (1.7)*
Aiming and catching	8.7 (3.8)*	12.4 (3.3)*
Balance	6.5 (3.6)*	11.1 (2.0)*
Total test score	5.1 (2.7)*	11.2 (2.3)*

*: p-value < 0.05, by paired t-test

Table 2 Maze and pmFGA Scores by Group

	DCD (n=19)	TD (n=19)
Maze Quality Scores, Median (1st, 3rd Quintile)		
Item 1	1 (1, 2.5)	0 (0, 0.5)
Item 2	3 (2, 5)	1(0, 2)
Item 3	2 (1, 2.5)	0 (0, 0)
Maze Time (s), Mean (SD)		
Item 1	4.7(2.7)*	2.6(1.1)*
Item 2	14.7(8.9)*	9.9(2.2)*
Item 3	7.8(4.6)	6.8(2.7)
Maze Summary Scores, Mean (SD)	7.3(4.4)*	1.5(1.4)*
pmFGA Quality Scores, Median (1st, 3rd Quintile)		
Narrow Base Walking	1 (0.5, 3)	3 (3, 3)
Walking with Eyes Closed	2 (1.5, 3)	3 (3, 3)
Walking Backward	2 (0.5, 3)	3 (3,3)
pmFGA Steps, Mean (SD)		
Narrow Base Walking	7.7(5.5)*	13.3(3.3)*
Walking with Eyes Closed	8.3(3.5)*	14.4(1.8)*
Walking Backward	9.1(4.6)*	14.3(2.1)*
DGI Summary Scores, Mean (SD)	20.7(2.6)*	23.5(0.6)*
FGA Summary Scores, Mean (SD)	23.2(4.9)*	29.1(1.4)*

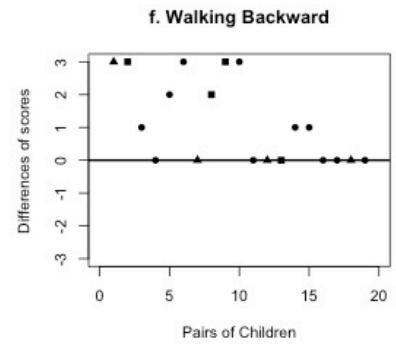
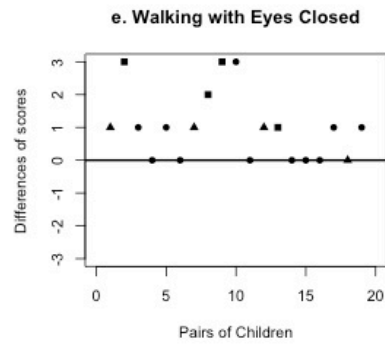
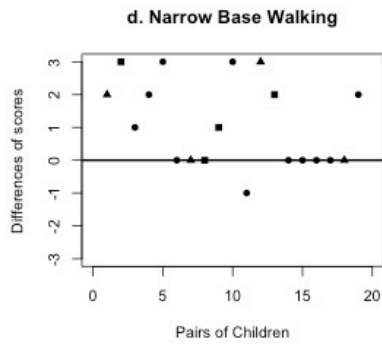
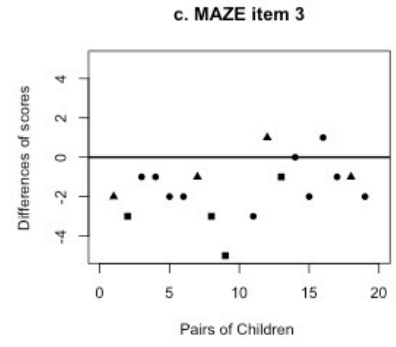
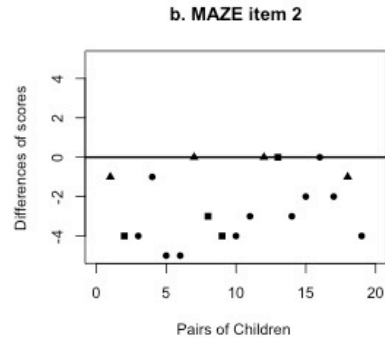
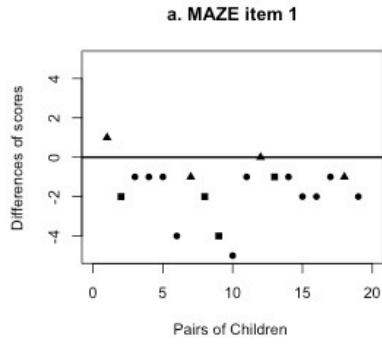
*: p-value < 0.05, by paired t-test

Figure 1. Individual Score Differences on the Maze and the pmFGA

Figure 1a-1c shows the individual score differences within each pair (scores of TD – scores of DCD) for all three items of Maze to interpret the construct validity of the Maze items. The negative differences of scores indicate the item can differentiate the child with DCD and the child with TD in the same pair. The 0 values indicate children from both groups showed no differences in the item. The positive values indicate the child with DCD had better performance than the child with TD in that item.

Figure 1d-1f shows the individual score differences within each pair (scores of TD – scores of DCD) for all three items of pmFGA to interpret the construct validity of the pmFGA items. The positive differences of scores indicate the item can differentiate the child with DCD and the child with TD in the same pair. The 0 values indicate children from both groups showed no differences in the item. The negative values indicate the child with DCD had better performance than the child with TD in that item.

For all figures, the solid squares indicate children in age band 1 (5-6 years); the solid circles indicate children in age band 2 (7-10 years); the solid triangles indicate children in age band 3 (11-12 years)



CHAPTER 4

Classification of Children with Developmental Coordination Disorders Based on Clinical Subgroups

Abstract

PURPOSE: The purpose of this study was to develop a preliminary classification system using standardized clinical tests to explore the identification of subgroups within children with DCD. The hypotheses were that there would be distinct sensorimotor subgroups in children with DCD and children in different subgroups would demonstrate different functional limitations based on the results of clinical tests.

METHODS: Nineteen children with DCD participated this study. The Diagnostic and Statistical Manual of Mental Disorders- fifth edition (DSM-V) criteria were used to verify the group membership. All children were examined using standardized tests across domains of fine motor, gross motor, balance, coordination, sensory processing, and intelligence. Descriptive statistics were used to describe the clinical presentation and sensorimotor behaviors of children with DCD. Through visual analysis via pattern recognition by standardized percentile ranks scores, subgroups were suggested.

RESULTS: The results suggested that children with DCD could be classified into three groups according to intelligence and overall performance: (1) Overall-limited type with no cognitive concerns (Overall-NC); (2) Overall-limited type with cognitive concerns (Overall-C); and (3) Partially-limited type with no cognitive concerns (Partial-NC). Within the Partial-NC group, two subgroups were suggested by other specific domain test results: with balance and body

coordination problems (Partial-NC-BC), and with fine motor and visual motor integration problems (Partial-NC-F).

CONCLUSION: Subgroups of DCD are identifiable by using standardized clinical tests.

Identifying subgroups may be helpful for evaluation and intervention in children with DCD.

However, more reliable and valid statistical analysis with larger samples of children is needed to confirm the subgroups.

Introduction

Developmental Coordination Disorder (DCD) is a chronic condition involving impairment in gross motor, postural, and/or fine motor performance that affects a child's ability to perform the skilled movements necessary for daily living, including the performance of academic and self-care tasks. As a childhood disorder, DCD involves impairments in sensory and motor control of coordination for volitional movement and postural control. The challenges of diagnosing DCD include lack of information about the pathology of DCD and the large variety of symptoms associated with DCD. If subgroups of movement syndromes could be determined under the larger umbrella diagnosis of DCD, then targeted and potentially more efficient and effective interventions could be recommended for children and their families (Blank et al., 2012; Compbell et al., 2012; Green et al., 2011; Missiuna et al., 2001; Zwicker et al., 2012a;).

Currently, the commonly used diagnostic criteria are from the Diagnostic and Statistical Manual of Mental Disorders- fifth edition (DSM-V) (APA, 2013). The criteria include:

- A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) as well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike, or participating in sports).
- B. The motor skills deficit in Criterion A significantly and persistently interferes with activities of daily living appropriate to chronological age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure, and play.

C. Onset of symptoms is in the early developmental period.

D. The motor skills deficits are not better explained by intellectual disability (intellectual developmental disorder) or visual impairment and are not attributable to a neurological condition affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder) (APA, 2013, p. 74).

This definition was originally put in place in 1987 and revised in 2013 replacing previously used terms to describe children with poor motor coordination, such as ‘clumsy child’, ‘minimal brain damage’, ‘developmental dyspraxia’, etc (American Psychiatric Association, 2013). However, these criteria are all symptom-dependent which results in a relatively complicated differential diagnosis process to confirm the diagnosis of DCD (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker et al., 2012a).

Children with DCD represent a large (5-6% of children) under-served population in the United States (US). Due to in part to needing input from multiple health professionals, the diagnosis of DCD is just beginning to occur in the US. While children with DCD are able to perform typical functional movements, they have difficulty with complex motor coordination which may lead to loss of social acceptance in community sports programs and decreased physical activity, which may further put the child with DCD at risk for obesity, poor health fitness, and social isolation (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011; Zwicker et al., 2012a). Parents, teachers and medical professionals often overlook the challenges children with DCD face in their daily life (Blank et al., 2012; Green, 2010). However, their motor problems are treatable and the prognosis can be promising if appropriate intervention is provided early in their development (Blank et al., 2012; Campbell et al., 2012; Zwicker et al., 2012a). Interventions from different theoretical perspectives have been developed to improve

motor abilities and tested with varying success. Previous research also suggests that with appropriate intervention, children with DCD may have the potential to catch up with their peers by the time they are 15-17 years old (Cantell, Smyth, & Ahonen, 2003). This has led to the hypothesis that there are likely subgroups under the umbrella DCD diagnosis that potentially require different types of intervention for maximum improvement of motor abilities. Research related to identifying sub-groups is just beginning (Ghanizadeh, 2010; Green et al., 2008; Peleg, Asbeh, Kuflik, & Schertz, 2009; Vaivre-Douret et al., 2011; Visser, 2003).

In 2003, Visser (2003) reviewed previous studies on differences between children with DCD and controls. The results suggested that DCD is not a uniform disorder, and there appear to be different subtypes of disabilities based on the comorbidities children have, such as Attention Deficit Hyperactivity (ADHD) and dyslexia. This review also suggested that it is important to define subtypes under DCD in order to improve the diagnosis and treatment of children with DCD. Later, in 2008, Green et al. (2008) recruited a large clinical sample of 100 children with DCD from 5 to 13 years old in the United Kingdom. They used the Movement Assessment Battery for Children (MABC), the Developmental Test of Visual-Motor Integration and Supplementary test (VMI), and the Clinical Observations of Motor and Postural Skills (COMPS) to test the sensorimotor abilities in children with DCD. Factor analysis was applied to identify subtypes under the DCD diagnosis. Five clusters appeared after the analysis: (1) weak kinesthesia, (2) poor static balance, (3) weak static and dynamic balance, (4) poor manual dexterity and perceptual skills, and (5) generalized poor performance on all measures. Their results also suggested that children with different subtypes of DCD might need different intervention approaches.

Further development of classification methods to optimize clinical decision making for children with DCD is critical for improving the effectiveness and efficiency of physical therapist management. Therefore, the purpose of this study was to explore the development of a classification system using multiple standardized clinical tests across many potential domains in order to identify subgroups within children with DCD. The hypotheses were that there would be distinct sensorimotor subgroups in children with DCD and children in different subgroups would demonstrate different functional limitations based on the results of standardized clinical tests. Based on previous research, we already know that there is large variability in functional limitations in children with DCD (Blank et al., 2012; Campbell et al., 2012; Ghanizadeh, 2010; Green et al., 2008; Green et al., 2011; Tsai, Pan, Cherng, & Wu, 2009; Vaivre-Douret et al., 2011; Visser, 2003; Wuang, Su, & Su, 2012; Zwicker et al., 2012a). Children with DCD may demonstrate difficulties in gross motor functions, fine motor functions, postural control, or sensory processing. Therefore, under this aim, we expected that children would fall into subgroups of DCD, which included gross motor, fine motor, postural control, sensory processing and mixed, based on the results of our clinical tests.

Methods

Sample

This is a descriptive study. A convenience sample of 19 children with DCD participated. The study was reviewed and approved by the university Institutional Review Board. All parents and children signed approved consent and assent forms, respectively, prior to participating in the study.

Because DCD is currently not routinely diagnosed in the US, we recruited participants in two ways. We contacted local therapy centers and asked therapists to inform any of their clients who matched our inclusion criteria about the study. We also screened elementary school age children within one of the local school districts in order to locate and recruit potential participants for the study. We used the DCD Questionnaire (DCDQ) (Wilson et al., 2009) to screen children for potential DCD concerns. The DCDQ is a standardized questionnaire of 15 questions, in which parents provide their impression of their child's sensorimotor abilities during various activities. The reported sensitivity and specificity of the DCDQ are 84.6%, and 70.8%, respectively (Wilson et al., 2009). Once scored, the DCDQ is used to identify children who may need further testing to discern a diagnosis of DCD.

Inclusion criteria, relating to all areas of the DSM-V (APA, 2013), for children with DCD were: 1) DCDQ scores that indicated DCD concerns or a medical diagnosis of DCD; 2) identification of motor problems via the Movement Assessment Battery for Children, 2nd Ed (MABC-2) (Henderson & Sugden, 2007) (percentile rank lower than 15% in at least one composite of the MABC-2); 3) parents or therapists report having problems in academic work and daily life participation; and 4) age 5 years 0 months to 12 years 11 months (elementary school age). Children were excluded if they had an IQ <60, other motor related medical diagnoses (e.g. cerebral palsy), a history of serious head injury or seizures, a visual acuity impairment not corrected by glasses, report of any lower limb or back injury within the previous six months.

Procedures

After the screening, all children participated in a 2.5-hour test session in the human motion analysis laboratory at the university or at the referring physical therapy clinics. The results of tests were used to: (1) verify the clinical DCD diagnosis and (2) explore subgroups of DCD. For verifying the clinical DCD diagnosis, the child tests included: (1) Growth parameters and Sensorimotor screening measures (for general descriptive purposes), and (2) Movement Assessment Battery for Children (MABC-2) (Henderson & Sugden, 2007) (gross/fine motor/balance ability). Parent report questionnaires were: 1) Demographic questionnaire (for medical history review and participation and academic work report); 2) MABC checklist (Henderson & Sugden, 2007) (motor coordination during home/community activities); and 3) Attention-deficit hyperactivity disorder (ADHD) checklist (Barkely & Murphy, 1998) (presence of ADHD during home/community activities).

The clinical standardized tests used with the children included: 1) MABC-2 (Henderson & Sugden, 2007) (gross/fine/balance motor ability); 2) Beery-Buktenica Developmental Test of Visual-Motor Integration, 6th Edition (Beery & Beery, 2004) (BEERY VMI) (visual perceptual ability); 3) Kaufman Brief Intelligence Test-2nd edition (K-BIT) (Kaufman & Kaufman, 2004) (cognitive ability); and 4) Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2) coordination composites (Bruininks & Bruininks, 2005) (manual coordination and body coordination). Parents were asked to complete the Sensory Processing Measure (SPM) (Parham & Ecker, 2007) (reaction to sensory input during home/community activities), a parent report questionnaire.

Instrumentation

Clinical Measures

1. Growth parameters and Sensorimotor Screening Measures: Height and weight were recorded using a tape measure of a floor to top of the head measurement marked on a wall and a standard weight scale, respectively. For the brief sensory screening, we applied light touch, light pressure, and examined proprioception as clinical measures of peripheral sensation. While sitting on a chair with eyes closed, the child was asked to report the sensation to a series of light touches (cotton ball), light pressure (examiner finger) on each lower leg. For proprioception, the examiner completed a limb position test (i.e., leg put in a position and child matches position with other leg) and limb movement test (i.e., leg moved in a pattern and child matches movement with other leg). The clinical screening of strength, range of motion, and posture, was tested while the child was lying in supine and prone. The child was asked to move the legs into all planes of movement, and when full range had not been achieved volitionally, the examiner applied gentle overpressure to determine if full range of joint movement was available. The child was scored as having typical or atypical strength/range for each movement. Standing posture was observed to note the presence or absence of scoliosis, kyphosis, lordosis, leg length difference, and standing foot pronation (Kaufman & Schilling, 2007; Shumway-Cook & Woollacott, 2007;).

2. Movement Assessment Battery for Children-2nd edition (MABC-2): The MABC-2 was designed to detect motor skill impairments in children aged 3-16 years. The child is asked to do 8 common child gross and fine motor activities, such as balance on one foot or with a pencil trace a line through a path on a piece of paper. Test results yield scores for Total Impairment, Manual Dexterity, Ball Skills, and Balance (static and dynamic). Reliability and validity of the MABC-2 is considered moderate to good based on studies reported in the test manual (test-retest reliability: ICC= 0.49-0.91; inter-rater reliabilities: ICC= 0.92- 1.00) (Henderson & Sugden, 2007).

3. Beery-Buktenica Developmental Test of Visual-Motor Integration, 6th Edition (VMI): The VMI is a developmental test to measure visual motor integration function (eye-hand coordination). The internal consistency of the VMI is moderate ($r = 0.88$), and inter-rater reliability is high ($r = 0.92$) (Beery & Beery, 2004).

4. Kaufman Brief Intelligence Test-2nd edition (K-BIT) : This test measures verbal and non-verbal intelligence and was used as a brief measure of cognitive status. The child completes cognitive problem solving activities such as answering riddles and solving matrices problems. The test is designed for ages 4-90 years. For children 4 to 18 years, internal reliability ($\alpha = .86-.92$), test-retest reliability ($r = .83 - .91$) and construct validity are supported (Kaufman & Kaufman, 2004).

5. Bruininks-Oseretsky Test of Motor Proficiency-2 (BOT-2) coordination composites (manual coordination and body coordination): This test measures coordination, including body and bilateral coordination, and was used as a measure of motor coordination status for this study. The child completed the two coordination subtests in the BOT-2. The test is designed for ages 4-21 years. Overall, the inter-rater reliability has been shown to be high in children age from 4-18 ($r = 0.86-0.99$). The test-retest reliability generally is moderate to high (Age 4-7, 8-11, 12-21: $r = 0.63-0.91, 0.49-0.95, 0.46-0.90$) (Bruininks & Bruininks, 2005).

Parent Questionnaires

1. Health/Demographic/Activity History: This was a parent questionnaire to report current demographic information (e.g. grade level, income level, family structure), history of motor therapies, recent illnesses or hospitalizations, and other health concerns or diagnoses.

2. MABC checklist: This is a parent questionnaire about movement difficulties in everyday situations in which the child has to function (i.e., self-care, classroom, recreational activities). Movement behaviors are evaluated in three sections: 1) movement in static or predictable environments, 2) movement in a dynamic or unpredictable environment and 3) non-motor behaviors that affect movement. Motor behaviors in each section are rated on a 4-point scale (0 indicates the child performs very well and “3” indicates the child is not close to performing the skill successfully). The sum of raw scores for each category yields a total motor score raw score that is classified as typical, at-risk or atypical (Henderson & Sugden, 2007).

3. Attention-deficit hyperactivity disorder (ADHD) checklist: This is a parent questionnaire that asks questions about the child’s behavior and the problems they may sometimes have. Parents are asked to answer this questionnaire based on their child’s behavior over the past 6 months. Several factor scores can be calculated: attention factor, executive function factor, hyperactivity factor and impulsivity factor. The factor scores can be compared to the normative sample (Barkely & Murphy, 1998).

4. Sensory Processing Measure (SPM): The SPM is a standardized behavioral rating scale for children ages 5-12 years. It asks questions about a child’s sensory processing abilities and their effect on the child’s daily life. Parents respond to questions about their child’s behaviors relative to scales that examine responses to touch, balance and motion, body awareness, visual and auditory input and motor planning. A total t-score is obtained along with t-scores for each subtest. Internal reliability (Cronbach’s $\alpha = .77-95$), interrater reliability ($r > .94$) and evidence of good construct validity are reported in children age 5 -12 (Parham & Ecker, 2007).

Analysis

Descriptive statistics were used to present the clinical presentation and sensorimotor behaviors of children with DCD. The means and standard deviations for standard scores for each test are showed in Table 1. The percentile ranks were used to compare among all tests/measures. In this study, we set the 5th and the 25th percentiles as the cut-offs. Children who demonstrated \leq 5th percentile of a test domain was identified as having significant limitation in the domain measured. Children who demonstrated $>5^{\text{th}}$ and $\leq 25^{\text{th}}$ percentile of a test domain were identified as with concerns in the domain measured. For some previous studies, the 15th percentile was used as a cutoff for indicating concerns (Green et al., 2011; Zwicker et al., 2010, 2012b). In this study, in order to include more children with concerns and subtle problems, we selected 25th percentile instead. Through visual analysis via pattern recognition (by percentile rank test scores), subgroups were identified. Plots were used to display the subgroups of DCD based on the data from the standardized clinical measurements.

Results

We received 92 DCDQs during the screening phase from therapists and through school screening. Of them 27 scored as having DCD concerns. From these 27 with DCD concerns, we were able to contact the families of 24 children. Five children were excluded during the initial phone screening based on the DSM-V criteria due to symptoms primarily caused by another diagnosis. In the end, 19 children with presumed DCD participated. Table 1 shows demographic information, sensorimotor screening measures, medical histories, and the results of all clinical measurements for the children in the study. Of the 19 children with presumed DCD, there were two who had a medical diagnosis of DCD and were receiving intervention from physical and occupational therapists for their motor coordination problems. Three other children were

receiving either physical or occupational therapy for their sensorimotor concerns. Via examination of the social economic status, there was one child with DCD from a “high-risk” family based on low socioeconomic status. For this child, the parent-reported questionnaires were completed by the social worker and the schoolteachers, and then confirmed by the child’s mother. All children that participated could communicate by English, but there were two from Spanish-speaking families. The parent-reported questionnaires were completed by using the Spanish version or verbally translated by a bilingual (English and Spanish) speaker.

From the sensorimotor screening, no child appeared to have concerns based on the general sensory screening. There were two children with pronated feet, and one of them showed decreased muscle strength in bilateral dorsi-flexors. All children showed lower scores in all parts of MABC-2 (Fine motor, Gross motor, and Balance) compared to the norm, as expected.

Subgroups by intelligence

Figure 1 shows the individual percentile ranks over all measurement domains, including body coordination (BOT-2); balance, gross motor, fine motor, and manual coordination (MABC-2); visual motor integration (VMI); sensory processing (SPM); verbal intelligence and non-verbal intelligence (K-BIT). Note in the figure that the 5th (significant limitation) and 25th (concerns) percentile cut-off lines are indicated. This figure first presents the possible subgrouping based on the intelligence level for children with DCD. Children in either Group 1 or Group 3 were children without cognitive concerns. Children in Group 2 showed concerns in verbal and/or in non-verbal intelligence. Further, children in Group 1 and Group 2 showed limitations over most of the testing domains, but children in Group 3 only demonstrated limitations in several domains, such as the balance and the fine motor domains. Based on the

plotted data, we suggest classification of children into Group 1: Overall (with almost all measures percentiles below the cut-off)-limited sensorimotor function with no cognitive concerns (Overall-NC), Group 2: Overall-limited sensorimotor function with cognitive concerns (Overall-C), and Group 3: Partially (several measures percentiles below the cut-off)-limited sensorimotor function with no cognitive concerns (Partial-NC).

Partially-limited type with no cognitive concerns (Partial-NC)

Figure 2 shows the individual performance based on percentile rank over all measurement domains for just the children in the Partial-NC group. The figure presents the variations of limitations in these children. We suggest two subgroups under the Partial-NC category: (1) children primarily with balance and body coordination problems (Partial-NC-BC), and (2) children primarily with fine motor, visual motor integration and sensory processing problems (Partial-NC-F/SM). There was one child with DCD having significant limitations in balance, body coordination and fine motor. We suggest sorting this child into both groups due to the overlap (Figure 4).

Discussion

Our primary research question was to explore the existence of subgroups of DCD based on an extensive standardized clinical testing protocol that included many domains: gross motor, body coordination, balance, fine motor, manual coordination, visual motor integration, sensory processing, and intelligence. We suggest there are identifiable clinical subgroups under the DCD diagnosis, which may assist with determination of appropriate intervention. From the results, we propose the following three subgroups of DCD: Overall-limited type with no cognitive concerns

(Overall-NC), Overall-limited type with cognitive concerns (Overall-C), and Partially-limited type with no cognitive concerns (Partial-NC) based on their intelligence and overall performance across the domains. Furthermore, the results may suggest two additional subgroups under the Partial-NC group: children with balance and body coordination problems (Partial-NC-BC), and children with fine motor and visual motor integration problems (Partial-NC-F/SM).

The results of this study are similar in some respects and different in others in comparison to other research on the potential for subgroups of children with DCD. In Green et al.'s study, they used factor analysis to determine five clusters based on their three measurements: (1) weak kinesthesia, (2) poor static balance, (3) weak static and dynamic balance, (4) poor manual dexterity and perceptual skills, and (5) poor on all items. Similar to Green's study, our sample suggested groups with overall poor abilities (Overall-C and Overall-NC), a subgroup more related to fine motor deficit (Partial-NC-F) and a subgroup related to balance and body coordination problems (Partial-NC-B). This supports that clinical subgroups of DCD exist, and sensorimotor function domains may be useful to categorize children with DCD into more specific groups. Different from the Green's study in which they used scores of individual items from a test composite (e.g. static balance item and dynamic balance item from the balance composite of the MABC) (Green et al., 2008), we only used standardized total scores of composites for identifying subgroups (e.g. total standard score of balance composite of the MABC-2), which have higher reliability and validity than individual item scores. This approach eliminated the possibility of having more specific groups; we could only show groups with more general categories. However, since we only had 19 children with DCD in this study and were using visual analysis instead of a reliable statistical analysis, we were more conservative about our conclusions. We decided it would be more unbiased and accurate for us to use the original

composite scores represented as normative scores. Therefore, in our study, the possibilities of clinical subgroups are more limited, but generally, we observed similar patterns of subgroups as in Green's study.

In Visser's review paper (2003), subgroups associated with the comorbidities, such as ADHD, were identified. In this study, we recorded medical histories with learning disabilities, ADHD, Autism Spectrum Disorders, Fetal Alcohol Spectrum Disorders, and other neurological concerns. We also collected data from the ADHD screening. However, the results did not show any obvious relationship between the clinical performance and the comorbidities. In order to better include children with clinical DCD, we strictly adhered to the DSM-V criteria (APA, 2013) to rule out any possibilities of other medical conditions that might affect children's sensorimotor performance. Hence, the inclusion criteria might decrease the potential influence from the comorbidities. For future research with larger sample size, it would be important to confirm the effect of comorbidities and/or the severity of other medical conditions in children with DCD, and to determine if different subgroups categories based on comorbidities need to be created.

Other than the comorbidities, in our study, personal and environmental issues, such as the social economic status (SES) and the primary languages of the families were two factors that may have influenced the clinical subgroups. Of the 19 families that participated, there was one family with low SES and severe family and environmental problems. There were also two families, in which parents primarily spoke Spanish and had lower SES compared to other families in this study. All three children demonstrated overall limitations of sensorimotor and intelligence performance and were identified in the Overall-C group. However, by parents', teachers' and the social worker's report, these children had less exposure and knowledge related to US culture. Furthermore, they lacked opportunities and experiences for participating in

activities with their peers. This may imply that the results of testing may underestimate their intelligence, and learning abilities. Therefore, their sensorimotor limitations may not be purely due to DCD. For example, if these children had regular exposure of physical activities, sensory and cognitive stimuli, they might have had fewer problems and may not even be qualified as having DCD. The focus of intervention for these children may be very different from other children in the same subgroup. Accordingly, we suggest, for future study, the SES and language/culture should be important factors to consider for subgroups and the intervention approach for these children.

Previous physical/occupational therapy intervention that the children had is another factor that might influence the results. In this study, there were six children who had previous therapy. Two of them already had a medical DCD diagnosis before this study and had received physical and occupational therapy for their motor and sensory problems for 3 and 7 years. These two children both were in the Partial-NC group, and they did not show significant limitations ($\leq 5^{\text{th}}$ percentile), rather concerns ($> 5^{\text{th}}$ and $\leq 25^{\text{th}}$ percentile) in some of the testing domains. However, based on the parents' and therapists' report, they had struggled with considerable impairments in many domains, such as gross motor, fine motor, and balance at earlier ages. They continue to have difficulty with learning new high-level complex activities, such as cliff climbing, skiing, and playing basketball. The other four children who received therapy did not have a previous DCD diagnosis. They were having services for their ADHD, sensory integration disorder, or fitness problems. Unlike the two children with a confirmed DCD diagnosis as reported by their parents, the therapy did not show an obvious effect for the subgroups of DCD. They were having different problems and were identified into different subgroups. These results may reflect that children can change in their general subgroup designation due to early intervention. This

information also supports previous interventional studies, which suggest that these symptoms are treatable and the prognosis can be promising if we can provide appropriate and specific intervention in their early life (Blank et al., 2012; Campbell et al., 2012; Green et al., 2011).

While we were able to suggest subgroups, we likely do not have enough detailed information to use the classification to currently direct appropriate therapy interventions. We may need to add another layer of assessments on top of the current battery, which can identify more specific problems, such as difficulties in motor planning, and sensory-related balance control, to classify these children into more specific subgroups. For example, in the previous chapter, we tested the Functional Gait Assessment (FGA) (Wrisley, 2004) and the Maze motor planning assessment (Maze) (May-Benson, 2006) in children with DCD and their peers with TD. The results demonstrate the validity of these two clinical tests in differentiating gait coordination and motor planning limitations in children with DCD compared to children with TD. Therefore, exploring additional clinical tests that may help to further identify subtle limitations in other domains may be useful in recognizing other subgroups under a DCD diagnosis.

Limitations

There were several study limitations. We had only 19 children with DCD/DCD concerns in this study ranging in age from 5 to 12 years, making our sample too small and diverse to fully answer the question of subgrouping children with DCD. Given this, we did not use statistical analyses such as cluster analysis to identify the subgroups of DCD. The reported subgroups are based on visual classification. For future studies, it will be necessary to use more robust statistical analysis with a larger sample size. Authors of a previous study suggested that under appropriate intervention approaches and appropriate environment for development, children with

DCD might catch up their peers by the time they are 15-17 years old (Cantell et al., 2003). That is to say, in addition to the intervention, maturation may be another factor we should consider for this population. In this study, we tried to sort children by the three different MABC-2 age bands (5-6 years, 7-10 years, 11-12 years) (Henderson & Sugden, 2007), but with this small sample, there was no obvious difference among these age groups. It is still possible, however, that age may be an important factor if applying cluster analysis with a larger sample. Therefore, further examination of different age groups is recommended.

Conclusion

Our results suggest that subgroups of DCD are identifiable by using a set of standardized clinical measurements. Within this small sample, we categorized children into three major subgroups by overall performance across multiple domains and intelligence level, and two smaller subgroups by individual test domains. The clinical assessments were all standardized and can be administered in various settings. Although, more reliable and valid statistical analysis with larger samples of children are needed to confirm the subgroups, it is recommended that DCD is conceptualized as an umbrella diagnosis and identification of subgroups may be helpful for directing evaluation and intervention in children with DCD. Further research is warranted, and more personal and environmental factors may need to be considered.

Table 1: Personal and Demographic Characteristics by Group

CHARACTERISTIC	Overall- NC (n=5)	Overall- C (n=6)	Partial- NC (n=8)	Partial- NC-BC (n=3)*	Partial- NC-F (n=4)*
Age in months, Mean (SD)	92.6 (9.1)	93.2 (23.3)	120.9 (32.3)	135.7 (10.0)	112.8 (37.4)
Sex, % of females	60.0	83.3	62.5	33.3	75.0
Ethnicity/Race, %					
Caucasian	80.0	33.3	87.5	66.7	100.0
Asian	20.0	16.7	0.0	0.0	0.0
Spanish/Hispanic	0.0	33.3	0.0	0.0	0.0
African American	0.0	16.7	0.0	0.0	0.0
Caucasian + Spanish/Hispanic	0.0	0.0	12.5	33.3	0.0
Parent education level, %					
Less than high school	0.0	33.3	0.0	0.0	0.0
High school diploma	0.0	16.7	0.0	0.0	0.0
Some college	0.0	0.0	12.5	33.3	0.0
College or Professional degree	100.0	50.0	87.5	66.7	100.0
Annual household income					
< \$25,000	0.0	33.3	0.0	0.0	0.0
\$25,000-50,000	0.0	16.7	12.5	33.3	25.0
\$50,000-75,000	20.0	16.7	0.0	0.0	0.0
>\$75,000	80.0	33.3	87.5	66.7	75.0
Body height (cm), Mean (SD)	131.3 (3.3)	129.8 (13.4)	145.4 (19.0)	153.8 (8.8)	142.8 (23.0)
Body weight (lb), Mean (SD)	74.4 (27.4)	68.0 (22.4)	88.0 (34.1)	113.3 (38.7)	88.0 (50.1)
Foot pronation, %	20.0	16.7	0.0	0.0	0.0

Muscle weakness, %	20.0	0.0	25.0	33.3	50.0
MABC Checklist, % of Red zone (<5%)	100.0	66.7	62.5	33.3	100.0
Medical history					
Sensory Integration Disorder	0.0	16.7	0.0	0.0	0.0
Autism Spectrum Disorder	0.0	16.7	0.0	0.0	0.0
Learning Disabilities	40.0	16.7	0.0	0.0	0.0
ADHD					
Inattentive	20.0	66.7	25.0	33.3	33.3
Hyperactive	20.0	16.7	12.5	25.0	25.0

* There is one child in both the Partial-NC-BC group and the Partial-NC-F group.

Table 2: Clinical Measurements by Group

CHARACTERISTIC	Overall- NC (n=5)	Overall- C (n=6)	Partial- NC (n=8)	Partial- NC-BC (n=3)*	Partial- NC-F (n=4)*
MABC-2 Standard Score, Mean (SD)					
Manual Dexterity	4.6 (1.1)	3.0 (1.1)	5.8 (2.3)	5.3 (1.2)	4.3 (0.5)
Aiming and Catching	5.2 (1.1)	7.3 (2.7)	11.9 (3.0)	12.0 (0.0)	11.3(1.7)
Balance	4.4 (2.9)	5.0 (3.1)	8.9 (3.1)	6.3 (2.5)	8.5 (3.3)
Total	3.4 (1.1)	3.5 (2.2)	7.4 (2.1)	6.0 (2.0)	6.3 (1.7)
SPM T-Score ^a , Mean (SD)	63.4 (4.0)	64 (3.9)	60 (6.2)	58.3 (5.5)	62.3 (6.9)
VMI T-Score ^a , Mean (SD)	43.8 (8.4)	35.3 (8.7)	45 (5.8)	46.3 (4.2)	42 (5.6)
K-BIT 2 Standard Score^b, Mean (SD)					
Verbal	116.8 (16.8)	81.8 (8.8)	119.75 (14.3)	112.3 (4.9)	115 (12.4)
Non-verbal	114.4 (14.2)	92.3 (4.6)	117.9 (11.2)	120.0 (4.0)	118.75 (10.8)
Total	118.4 (5.3)	85.0 (5.1)	121.8 (12.6)	119.0 (3.6)	119.75 (12.8)
BOT-2 Standard Score^c, Mean (SD)					
Manual	34.6	39.3	42.8	40.7	38.8
Coordination	(8.6)	(5.1)	(11.0)	(6.1)	(8.2)
Body Coordination	32.8 (3.5)	29.0 (4.7)	35.1 (11.1)	30.0 (2.6)	30.8 (10.9)

* There is one child in both the Partial-NC-BC group and the Partial-NC-F group.

a: T-Score: Mean=50, SD=10

b: K-BIT 2 Standard Score: Mean=100, SD=15

c: BOT-2 Standard Score: Mean=50, SD=10

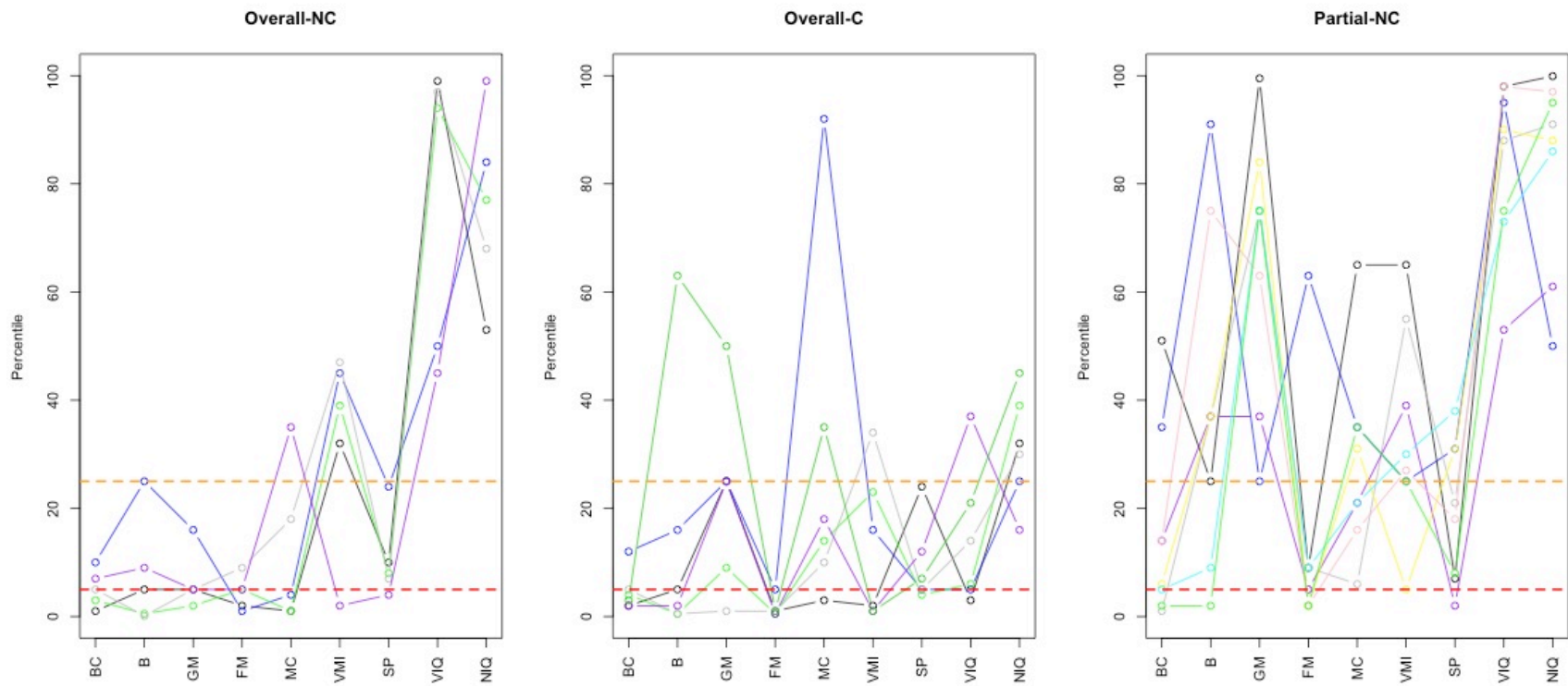


Figure 1. Individual Percentile Ranks for All Clinical Measurements of Subgrouping

Figure 1 shows the individual percentile rank for all clinical measurements of verifying DCD subgroups. The red dash lines represent the 5th percentile, and the yellow dash line represent the 25th percentile. BC: Body coordination; B: Balance; GM: Gross motor; FM: Fine motor; MC: Manual coordination; SP: Sensory processing; VIQ: Verbal IQ; NIQ: Non-verbal IQ.

ID	BC	B	GM	FM	MC	VMI	SP
1	Yellow	Green	Green	Red	Yellow	Green	Yellow
2	Yellow	Green	Green	Red	Yellow	Green	Red
3	Yellow	Green	Green	Red	Green	Red	Green
4	Red	Red	Green	Red	Green	Yellow	Yellow
5	Red	Yellow	Green	Yellow	Yellow	Green	Green
6	Red	Green	Green	Yellow	Yellow	Green	Yellow
7	Green	Yellow	Green	Yellow	Green	Green	Yellow
8	Green	Green	Yellow	Green	Green	Yellow	Green

Figure 2. Individual Cut-offs for All Clinical Measurements in the Partial-NC Group

Figure 2 shows the cut-offs for all clinical measurements of verifying DCD subgroups from children in the Partial-NC group. The red cells represent the $\leq 5^{\text{th}}$ percentile (with significant limitations), the yellow cells represent the $\leq 25^{\text{th}}$ percentile (with concerns), and the green cells represent $\geq 26^{\text{th}}$ percentile (with no concerns). BC: Body coordination; B: Balance; GM: Gross motor; FM: Fine motor; MC: Manual coordination; SP: Sensory processing.

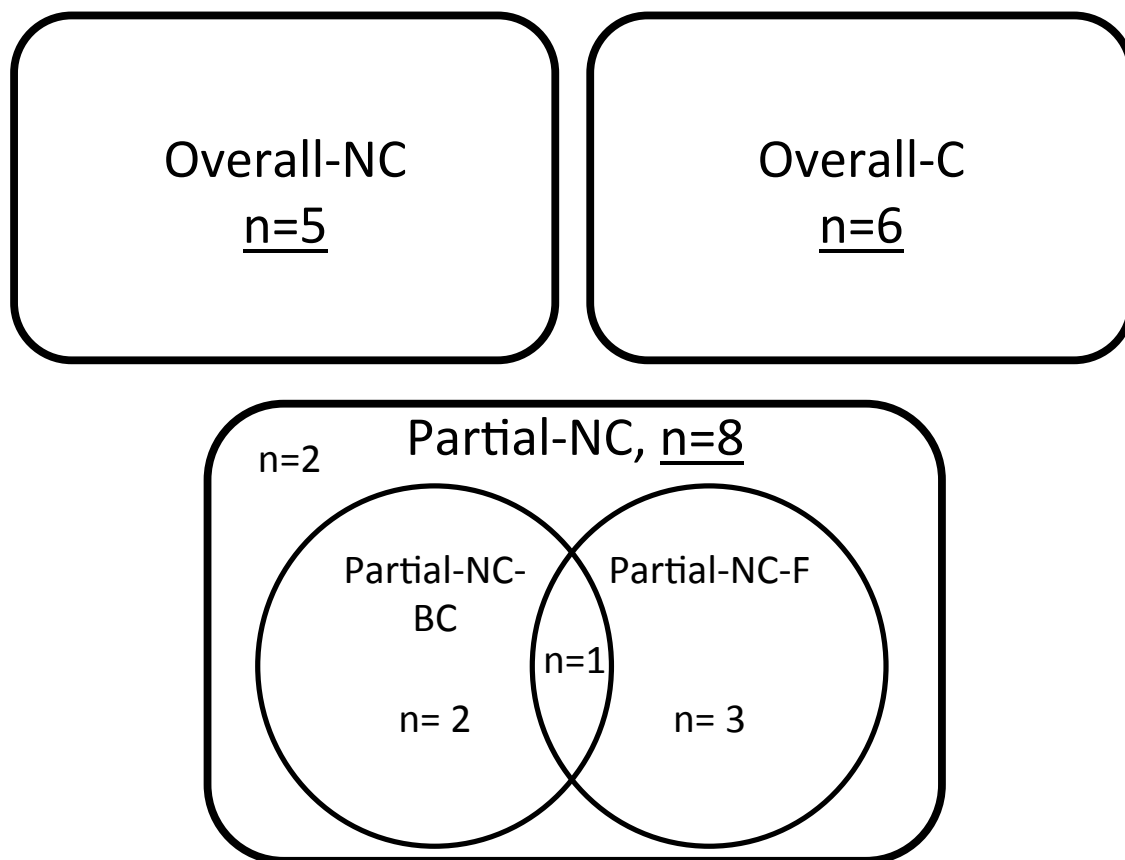


Figure 3. DCD Clinical Subgroups

Figure 3 shows the relationship and number of children with DCD sorted in to subgroups by clinical measurements. Overall-NC: Overall-limited type with no cognitive concerns; Overall-C: Overall-limited type with cognitive concerns; Partial-NC: Partially-limited type with no cognitive concerns. Within the Partial-NC group, Partial-NC-BC: children with balance and body coordination problems; Partial-NC-F: children with fine motor and visual motor integration problems. There were 2 children identified in the Partial-NC group, but not included in either the Partial-NC-BC or the Partial-NC-F group. Based on the results, these two children had some concerns areas, but did not demonstrated obvious impaired functions in any domains that tested.

CHAPTER 5

Discussion

In this dissertation, first, a systematic review has been done to summarize the literature related to the possible brain pathologies in children with DCD. Second, the validity of clinical tests of motor planning and gait coordination were examined as assessments to differentiate motor function in a group of children with DCD and their peers with TD. Finally, the group of children with DCD was examined in more detail to evaluate clinical characteristics among several testing domains, to suggest identification of subgroups by standardized clinical tests. Our purposes were to have more information about the variations in children with DCD in both the pathology and clinical performance, and to start the process of developing a clinical measurement system to identify subgroups in children with DCD. Following is a discussion of overall results and suggestions for future research.

Central nerve system pathologies in children with DCD

We found eight studies to include in this review and none were validating cohort studies. All of the studies included were exploratory cohort studies with or without good reference standards. The lack of any validating cohort studies may exist due to the challenges of brain imaging for children and difficulties recruiting children with DCD and those without other obvious medical comorbidities. With these possible constraints, validating cohort studies may not always be realistic. Given these considerations, conducting high quality, structured exploratory cohort studies may be more appropriate. With caution, the findings provide some direction for assessing CNS pathologies in children with DCD and provide some information for clinical applications and future research.

Using the literature available, the results of this review suggest that compared to children with typical development, children with DCD demonstrate different patterns of brain activity; however, the identified brain area, network, and level of activation varied in the different studies. The sites of potential pathologies of DCD that relate to motor function primarily included prefrontal lobe, frontal lobe, parietal lobe, and cerebellum. Corticospinal tract was involved and the posterior thalamic radiation may also be involved, which may affect sensory processing from the somatosensory (proprioception and cutaneous) system. Basal ganglia were also implicated, related to the attention and executive functions, which can affect the motor function as well (Atkins et al., 2005; Debrabant et al., 2013; de Kieviet et al., 2014; Kashiwagi et al., 2009; Langevin et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011, 2012b).

The results of this review suggest that children with DCD demonstrate different and varied patterns of brain activity than children without DCD to accomplish several specific types of tasks. This suggests that for children with DCD, the most efficient motor and learning strategies may be different from strategies for children who are typical developing. The varied pathological findings also suggest that there may be distinct subgroups under the umbrella diagnosis of DCD.

Motor planning and gait coordination assessments for children with DCD

Because there is a need for sensitive and discriminative tests and measures of the constructs of motor planning and gait coordination, we explored the construct validity of a motor planning test, the Maze, and a gait coordination test, the pmFGA, by comparing the performance of children with DCD to that of children with TD. The results supported that both the Maze (May-Benson, 2006) test and the pmFGA items (Wrisley et al, 2004) are sensitive enough to

differentiate between the two groups of children in our study. Compared to children with TD, children with DCD performed the activities more slowly and had lower quality motor planning skills on the Maze. Children with DCD demonstrated less time where they had coordinated control during the pmFGA. Our pmFGA results suggested that the number of steps is a better measurement to differentiate the gait coordination in children with DCD and TD than the quality rating previously used for this measure in adult populations.

We also evaluated whether or not the total DGI (Lubetzky-Vilnai et al., 2011) and FGA (Wrisley et al, 2004) tests were sensitive enough to differentiate between the groups. Unlike our hypothesis, both the DGI and the FGA demonstrated differences between the children with DCD and TD in our study. Even though we differentiated between the two groups of children using the DGI total score, there are still concerns about the difficulty level of DGI and a potential ceiling effect for children with minor motor dysfunction. Therefore, we suggest that the DGI should be used with caution as an evaluative measure in children with minor motor problems. Compared to DGI, the full FGA adds the more challenging items that we examined further (pmFGA) and our results suggest that the full FGA can significantly differentiate the gait coordination differences between children with DCD and children with TD. Nonetheless, the FGA was designed for adults and the scoring criteria were set for the adult population. Prior to future use of the FGA in children further modification and development needs to be considered.

These tests are quick clinical assessments that can be simply administered in various settings for the purpose of identification of motor planning and gait coordination constructs. Although, further study in different age bands and with larger samples of children is needed to evaluate validity and reliability related to using these tests in a discriminative manner, it is recommended that the Maze and FGA may be helpful in identifying motor planning and gait

coordination deficits in children with DCD. If one wishes to utilize these tests for evaluative purposes, then further testing to determine the standard error of the measurement, minimal detectable change and minimal clinically important difference will be necessary.

Classification of children with DCD based on clinical subgroups

The results of our descriptive analysis supported that subgroups of DCD are identifiable based on a standardized testing protocol that included five measures across gross motor, body coordination, balance, fine motor, manual coordination, visual motor integration, sensory processing, and intelligence domains. From the results, we suggest that one can identify groups based on their intelligence and overall performance of Overall-limited type with no cognitive concerns (Overall-NC), Overall-limited type with cognitive concerns (Overall-C), and Partially-limited type with no cognitive concerns (Partial-NC). Furthermore, the results may suggest two more subgroups under the Partial-NC group: children with balance and body coordination problems (Partial-NC-BC), and children with fine motor and visual motor integration problems (Partial-NC-F).

Our results show some similarity to two other studies examining the existence of subgroups of children with DCD. Similar to Green's study, our sample showed groups with overall poor abilities (Overall-C and Overall-NC), a subgroup more related to fine motor deficit (Partial-NC-F) and a subgroup related to balance and body coordination problems (Partial-NC-B) (Green et al., 2008). This further validates that clinical subgroups of DCD likely do exist, and sensorimotor function domains can be the categories to sort these children into more specific groups. Different from the Green's study, in which they used scores of individual items from a test composite, we only used total standardized scores of composites to identify subgroups. This

approach may decrease the possibility of having more specific subgroups (Green et al., 2008). Different from Visser's report (2003), indicating that subgroups could be associated with comorbidities, such as ADHD, we did not show any obvious relationship between children's clinical performance and the presence of these comorbidities. In order to include children with specific clinical DCD, we did follow the DSM-V criteria (APA, 2013) very seriously to rule out any possibilities of other medical conditions that might affect children's sensorimotor performance. Hence, the recruitment process might have decreased the potential influence from the comorbidities.

Other than the comorbidities, in our study, personal factors, social economic status (SES), and the primary languages of the families were factors that may have influenced the clinical subgrouping for several children. Some children may have had a lack of opportunities and experiences for participating in activities with their peers, which may have resulted in an underestimation of their intelligence and learning abilities. If these children could have regular exposure to physical activities, sensory and cognitive stimuli, they might have had fewer problems. Previous intervention might also have influenced the results. In this study, children who had the medical DCD diagnosis and had received therapy for their motor and sensory problems were identified as in the Partial-NC group. They had fewer concerns in some testing domains that surprised their parents. This result may reflect effects from intervention, which suggests that these symptoms are treatable and the prognosis can be promising if we can provide appropriate and specific intervention in their early life.

In summary, our results suggest that subgroups of DCD are identifiable by using standardized clinical tests. Although, more reliable and valid statistical analysis with larger samples of children is needed in order to confirm the subgroups, it is recommended that DCD is

a umbrella diagnosis and identifying subgroups may be helpful for directing evaluation and intervention in children with DCD.

Directions for Future Research

There are several directions and recommendations for the future research related to measurements and subgrouping of children with DCD. First, there are several recommendations related to research design. In this study, we had 19 pairs of children with and without DCD in this study, who ranged in age from 5 to 12 years, which makes our sample relatively small and potentially diverse to fully answer our research questions about the validity of measurements and the clinical subgroups under DCD. For future research, enlarging the sample size and separating the children into age groups may be necessary to better validate the measurements and to identify clinical subgroups. Also, due to the small sample size, we did not use the optimal statistical analysis to answer our questions of measurement validation and identification of clinical subgroups. For measurement validation, we showed the data of agreement instead of using the Kappa test. For the clinical subgroups identification, we used visual analysis instead of cluster analysis. These are more robust statistical approaches to answer our questions. Therefore, for future research with appropriate sample size, these statistical analyses are recommended.

For the Measurement development for subgrouping children with DCD, the assessment tools we used to identify subgroups of DCD were all standardized clinical tests. There are some domains that cannot be covered by them, such as motor planning and sensory integration during walking. However, these may be important domains we should test on children with DCD in order to capture their subtle sensorimotor problems therefore tests such as the Maze and FGA need further psychometric testing within the DCD population and development of norms within

the typically developing population. Besides, motor planning and gait coordination, there may be other domains within DCD that need be tested, such as sensory-integrated balance control. Once new tests have more psychometric support, we would want to add another layer of assessments on top of the current suggested testing system, which can potentially pick up more specific problems for classification of these children.

Beyond the recommendations related to test development and subgrouping based on clinical tests, further exploration of the brain pathology of DCD is another direction of future research. It would be helpful for evaluation and intervention in children with DCD to better define the neurological pathology of DCD. Further, if we can complete brain imaging and clinical tests in samples of children with DCD, then examine correlations between the two, we may be able to further delineate DCD subgroups. Therefore, conducting brain-imaging studies in combination with clinical assessments is recommended for future research.

After determination of subgroups of DCD, the development of intervention protocols for specific subgroups should be considered. There are some researchers recommending different treatment strategies for children with DCD (Cantell et al., 2003; Green et al., 2008; Piek, Dawson, Smith, & Gasson, 2008; Visser, 2003). Since children with DCD show diverse clinical performance, the classification into clinical subgroups may be helpful to direct therapists to the optimal treatment strategies for children. Therefore, for future research, comparison of intervention protocols for different subgroups, or furthermore developing clinical prediction rules for interventions for children with DCD is recommended.

Appendix 1. Questions for Systematic Review and the Scoring Sheet

Questions	Yes (1)	Partial (0.5)	No (0)	NA	Total points	Percentage	Comments
1. Were inclusion and exclusion criteria of the study population well described and followed?							
1-1 Were inclusion criteria of the study population well described and followed?							
1-2 Were exclusion criteria of the study population well described and followed?							
2. Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?							
2-1 Were the measures used clearly described?							
2-2 Did the authors report or describe the validity of the measures?							
2-3 Were the measures used valid?							
2-4 Did the authors report or describe the reliability of the measures?							
2-5 Were the measures used reliable?							
2-6 Were the measures used appropriate to represent the outcomes of interest?							
3. Did the authors conduct and report appropriate statistical evaluation including power calculations? Both parts of the question need to be met to score 'yes'.							
3-1 Did the authors conduct appropriate statistical evaluation? (Reflect to research questions.)							
3-2 Did the authors report appropriate statistical evaluation? (Explanations in results and discussions.)							
3-3 Did the authors conduct and report power calculations?							
4. Were dropout/loss reported and less than 20%? For 2-group designs, was dropout balanced?							
4-1 Were dropout/loss to follow-up reported?							
4-2 Were dropout/loss to follow-up good?	<20 %	<50%	>50 %				

potential biases used?							
5-1 Were appropriate methods for controlling confounding variables used?							
5-2 Were appropriate methods for limiting potential biases used?							
5-3 Were potential limitation and biases reported?							
Total							

Appendix 2. The Motor Planning Maze Assessment (Maze) and the Scoring Sheet
(May-Benson, T. A. Motor Planning Maze Assessment. (2006). Retrieved from
<http://www.tmbeducationalenterprises.com/pdf/MazeManual.pdf>)



Motor Planning Maze Assessment Teresa A. May-Benson, ScD, OTR/L	
Name:	Date:
1. Level I Maze – Rectangle	Score
Time: (record actual time)	
0 - ≤ 5 seconds	
1- 6-10 seconds	
2- ≥ 11 seconds	
Observations: (Score 1 for each observation)	
1. Child shakes maze to move grommet	
2. Child changes the maze to other hand	
3. Child uses other hand to move grommet	
4. Child uses whole arm or body instead of wrist	
	Total Item Score :
5. Other observations:	
2. Level II Maze – Single Hand Maze	Score
Time: (record actual time)	
0 - < 10 seconds	
1- 11-15 seconds	
2- 16-20 seconds	
Observations: (Score 1 for each observation)	
1. Child shakes maze to move grommet	
2. Child changes the maze to other hand	
3. Child uses other hand to move grommet	
4. Child uses whole arm or body instead of wrist	
	Total Item Score :
4. Other observations:	
3. Level III Maze – Two-Hand Maze	
Time: (record actual time)	
0 - < 10 seconds	
1- 11-15 seconds	
2- 16-20 seconds	
Observations: (Score 1 for each observation)	
1. Child shakes maze to move grommet	
2. Child removes one hand from maze	
3. Child uses one hand to move grommet	
4. Child uses whole arm or body movements	
	Total Item Score :
4. Other observations:	
© T. May-Benson, 2008	
Total Test Score:	

Appendix 3. Dynamic Gait Index (DGI)

(Lubetzky-Vilnai, A., Jirikowic, T. L., & McCoy, S. W. (January 01, 2011). Investigation of the Dynamic Gait Index in children: a pilot study. *Pediatric Physical Therapy : the Official Publication of the Section on Pediatrics of the American Physical Therapy Association*, 23(3), 268-273.)

Grading: Mark the lowest category which applies. Repeat if doubt.

Demonstration: All items except for 1 and 8 should be demonstrated.

1. Gait Level Surface _____ *Instructions:* Walk and pick up the toy. (Place toy 3' after the 20' mark.) Grading: Mark the lowest category that applies.

(3) Normal: Walks 20', no assistive devices, good speed, no evidence for imbalance, normal gait pattern.

(2) Mild impairment: Walks 20', uses assistive devices, slower speed, mild gait deviations.

(1) Moderate impairment: Walks 20', slow speed, abnormal gait pattern, evidence for imbalance.

(0) Severe impairment: Cannot walk 20' without assistance, severe gait deviations, or imbalance.

2. Change in gait speed _____ *Instructions:* Begin walking at your normal pace (for 5'), when I tell you "go," walk as fast as you can (for 5'). When I tell you "slow," walk as slowly as you can (for 5').

(3) Normal: Able to smoothly change walking speed without loss of balance or gait deviation. Shows a significant difference in walking speeds between normal, fast, and slow speeds.

(2) Mild impairment: Able to change speed but demonstrates mild gait deviations, or no gait deviations but unable to achieve a significant change in velocity, or uses and assistive device.

(1) Moderate impairment: Makes only minor adjustments to walking speed, or accomplishes a change in speed with significant gait deviations, or changes speed but has significant gait deviations, or changes speed but loses balance but is able to recover and continue walking.

(0) Severe impairment: Cannot change speeds, or loses balance and has to reach for wall or be caught.

3. Gait with horizontal head turns _____ *Instructions:* Begin walking at your normal pace. When I tell you to "look to the side," keep walking straight, but turn your head to the side. Keep looking to the side until I tell you "look to the other side," then keep walking straight and turn your head to the other side. Keep your head to the side until I tell you, "look straight," then keep walking straight but return your head to the center.

(3) Normal: Performs head turns smoothly with no to slight change in gait velocity.

(2) Mild impairment: Performs head turns smoothly with moderate change in gait

velocity (i.e., minor disruption to smooth gait path or uses walking aid).

(1) Moderate impairment: Performs head turns with large change in gait velocity, slows down, staggers but recovers, can continue to walk.

(0) Severe impairment: Performs task with severe disruptions of gait (i.e., staggers outside 15° path, loses balance, stops, reaches for wall).

4. Gait with vertical head turns _____ *Instructions:* Begin walking at your normal pace. When I tell you to "look up," keep walking straight, but tip your head and look up. Keep looking up until I tell you "look down," then keep walking straight and turn your head down. Keep looking down until I tell you, "look straight," then keep walking straight but return your head to the center.

(3) Normal: Performs head turns with no to slight change in gait velocity.

(2) Mild impairment: Performs task with moderate change in gait velocity (i.e., minor disruption to smooth gait path or uses walking aid).

(1) Moderate impairment: Performs tasks with large change in gait velocity, slows down, staggers but recovers, can continue to walk.

(0) Severe impairment: Performs task with severe disruption or gait (i.e., staggers outside 15° path, loses balance, stops reaches for wall).

5. Gait and pivot turn _____ *Instructions:* Begin walking at your normal pace. When I tell you to "stop and turn," turn as quickly as you can to face the opposite direction and stop.

(3) Normal: Pivot and turns safely within 3 seconds and stops quickly with no loss of balance.

(2) Mild impairment: Pivot turns safely in >3 seconds and stops with no loss of balance.

(1) Moderate impairment: Turns slowly, requires verbal cueing, requires several small steps to catch balance following turn and stop.

(0) Severe impairment: Cannot turn safely, requires assistance to turn and stop.

6. Step over obstacle _____ *Instructions:* Begin walking at your normal speed. When you come to the shoe box, step over it, not around it, and keep walking.

(3) Normal: Able to step over box without changing gait speed; no evidence for imbalance.

(2) Mild impairment: Able to step over box, but must slow down and adjust steps to clear box safely.

(1) Moderate impairment: Able to step over box but must stop, then step over. May require verbal cueing.

(0) Severe impairment: Cannot perform without assistance.

7. Step around obstacles _____ *Instructions:* Begin walking at your normal speed. When you come to the first cone (about 6' away), walk around the right side of it. When you come to the second cone (6' past first cone), walk around it to the left.

(3) Normal: Able to walk around cones safely without changing gait speed; no evidence of imbalance.

(2) Mild impairment: Able to step around both cones, but must slow down and adjust steps to clear cones.

(1) Moderate impairment: Able to clear cones but must significantly slow speed to accomplish task, or requires verbal cueing.

(0) Severe impairment: Unable to clear cones, walks into one or both cones, or requires physical assistance.

8. Stairs _____ *Instructions:* Walk up these stairs as you would at home. At the top, turn around and walk down.

(3) Normal: Alternating feet, no rail.

(2) Mild impairment: Alternating feet, must use rail.

(1) Moderate impairment: Two feet to stair, must use rail.

(0) Severe impairment: Cannot perform safely.

Appendix 4. Functional Gait Assessment (FGA)

(Wrisley, D. M., Marchetti, G. F., Kuharsky, D. K., & Whitney, S. L. (January 01, 2004). Reliability, internal consistency, and validity of data obtained with the functional gait assessment. *Physical Therapy*, 84(10), 906-918.)

Appendix.

Functional Gait Assessment^a

Requirements: A marked 6-m (20-ft) walkway that is marked with a 30.48-cm (12-in) width.

1. GAIT LEVEL SURFACE

Instructions: *Walk at your normal speed from here to the next mark (6 m [20 ft]).*

Grading: Mark the highest category that applies.

- (3) Normal—Walks 6 m (20 ft) in less than 5.5 seconds, no assistive devices, good speed, no evidence for imbalance, normal gait pattern, deviates no more than 15.24 cm (6 in) outside of the 30.48-cm (12-in) walkway width.
- (2) Mild impairment—Walks 6 m (20 ft) in less than 7 seconds but greater than 5.5 seconds, uses assistive device, slower speed, mild gait deviations, or deviates 15.24–25.4 cm (6–10 in) outside of the 30.48-cm (12-in) walkway width.
- (1) Moderate impairment—Walks 6 m (20 ft), slow speed, abnormal gait pattern, evidence for imbalance, or deviates 25.4–38.1 cm (10–15 in) outside of the 30.48-cm (12-in) walkway width. Requires more than 7 seconds to ambulate 6 m (20 ft).
- (0) Severe impairment—Cannot walk 6 m (20 ft) without assistance, severe gait deviations or imbalance, deviates greater than 38.1 cm (15 in) outside of the 30.48-cm (12-in) walkway width or reaches and touches the wall.

2. CHANGE IN GAIT SPEED

Instructions: *Begin walking at your normal pace (for 1.5 m [5 ft]). When I tell you "go," walk as fast as you can (for 1.5 m [5 ft]). When I tell you "slow," walk as slowly as you can (for 1.5 m [5 ft]).*

Grading: Mark the highest category that applies.

- (3) Normal—Able to smoothly change walking speed without loss of balance or gait deviation. Shows a significant difference in walking speeds between normal, fast, and slow speeds. Deviates no more than 15.24 cm (6 in) outside of the 30.48-cm (12-in) walkway width.
- (2) Mild impairment—Is able to change speed but demonstrates mild gait deviations, deviates 15.24–25.4 cm (6–10 in) outside of the 30.48-cm (12-in) walkway width, or no gait deviations but unable to achieve a significant change in velocity, or uses an assistive device.
- (1) Moderate impairment—Makes only minor adjustments to walking speed, or accomplishes a change in speed with significant gait deviations, deviates 25.4–38.1 cm (10–15 in) outside the 30.48-cm (12-in) walkway width, or changes speed but loses balance but is able to recover and continue walking.
- (0) Severe impairment—Cannot change speeds, deviates greater than 38.1 cm (15 in) outside 30.48-cm (12-in) walkway width, or loses balance and has to reach for wall or be caught.

3. GAIT WITH HORIZONTAL HEAD TURNS

Instructions: *Walk from here to the next mark 6 m (20 ft) away. Begin walking at your normal pace. Keep walking straight; after 3 steps, turn your head to the right and keep walking straight while looking to the right. After 3 more steps, turn your head to the left and keep walking straight while looking left. Continue alternating looking right and left every 3 steps until you have completed 2 repetitions in each direction.*

Grading: Mark the highest category that applies.

- (3) Normal—Performs head turns smoothly with no change in gait. Deviates no more than 15.24 cm (6 in) outside 30.48-cm (12-in) walkway width.
- (2) Mild impairment—Performs head turns smoothly with slight change in gait velocity (eg, minor disruption to smooth gait path), deviates 15.24–25.4 cm (6–10 in) outside 30.48-cm (12-in) walkway width, or uses an assistive device.

- (1) Moderate impairment—Performs head turns with moderate change in gait velocity, slows down, deviates 25.4–38.1 cm (10–15 in) outside 30.48-cm (12-in) walkway width but recovers, can continue to walk.
- (0) Severe impairment—Performs task with severe disruption of gait (eg, staggers 38.1 cm [15 in] outside 30.48-cm [12-in] walkway width, loses balance, stops, or reaches for wall).

4. GAIT WITH VERTICAL HEAD TURNS

Instructions: *Walk from here to the next mark (6 m [20 ft]). Begin walking at your normal pace. Keep walking straight; after 3 steps, tip your head up and keep walking straight while looking up. After 3 more steps, tip your head down, keep walking straight while looking down. Continue alternating looking up and down every 3 steps until you have completed 2 repetitions in each direction.*

Grading: Mark the highest category that applies.

- (3) Normal—Performs head turns with no change in gait. Deviates no more than 15.24 cm (6 in) outside 30.48-cm (12-in) walkway width.
- (2) Mild impairment—Performs task with slight change in gait velocity (eg, minor disruption to smooth gait path), deviates 15.24–25.4 cm (6–10 in) outside 30.48-cm (12-in) walkway width or uses assistive device.
- (1) Moderate impairment—Performs task with moderate change in gait velocity, slows down, deviates 25.4–38.1 cm (10–15 in) outside 30.48-cm (12-in) walkway width but recovers, can continue to walk.
- (0) Severe impairment—Performs task with severe disruption of gait (eg, staggers 38.1 cm [15 in] outside 30.48-cm [12-in] walkway width, loses balance, stops, reaches for wall).

5. GAIT AND PIVOT TURN

Instructions: *Begin with walking at your normal pace. When I tell you, "turn and stop," turn as quickly as you can to face the opposite direction and stop.*

Grading: Mark the highest category that applies.

- (3) Normal—Pivot turns safely within 3 seconds and stops quickly with no loss of balance.
- (2) Mild impairment—Pivot turns safely in >3 seconds and stops with no loss of balance, or pivot turns safely within 3 seconds and stops with mild imbalance, requires small steps to catch balance.
- (1) Moderate impairment—Turns slowly, requires verbal cueing, or requires several small steps to catch balance following turn and stop.
- (0) Severe impairment—Cannot turn safely, requires assistance to turn and stop.

6. STEP OVER OBSTACLE

Instructions: *Begin walking at your normal speed. When you come to the shoe box, step over it, not around it, and keep walking.*

Grading: Mark the highest category that applies.

- (3) Normal—Is able to step over 2 stacked shoe boxes taped together (22.86 cm [9 in] total height) without changing gait speed; no evidence of imbalance.
- (2) Mild impairment—Is able to step over one shoe box (11.43 cm [4.5 in] total height) without changing gait speed; no evidence of imbalance.
- (1) Moderate impairment—Is able to step over one shoe box (11.43 cm [4.5 in] total height) but must slow down and adjust steps to clear box safely. May require verbal cueing.
- (0) Severe impairment—Cannot perform without assistance.

(Continued)

Appendix.
Continued

7. GAIT WITH NARROW BASE OF SUPPORT

Instructions: *Walk on the floor with arms folded across the chest, feet aligned heel to toe in tandem for a distance of 3.6 m [12 ft]. The number of steps taken in a straight line are counted for a maximum of 10 steps.*
Grading: Mark the highest category that applies.

- (3) Normal—Is able to ambulate for 10 steps heel to toe with no staggering.
- (2) Mild impairment—Ambulates 7–9 steps.
- (1) Moderate impairment—Ambulates 4–7 steps.
- (0) Severe impairment—Ambulates less than 4 steps heel to toe or cannot perform without assistance.

8. GAIT WITH EYES CLOSED

Instructions: *Walk at your normal speed from here to the next mark (6 m [20 ft]) with your eyes closed.*

Grading: Mark the highest category that applies.

- (3) Normal—Walks 6 m (20 ft), no assistive devices, good speed, no evidence of imbalance, normal gait pattern, deviates no more than 15.24 cm (6 in) outside 30.48-cm (12-in) walkway width. Ambulates 6 m (20 ft) in less than 7 seconds.
- (2) Mild impairment—Walks 6 m (20 ft), uses assistive device, slower speed, mild gait deviations, deviates 15.24–25.4 cm (6–10 in) outside 30.48-cm (12-in) walkway width. Ambulates 6 m (20 ft) in less than 9 seconds but greater than 7 seconds.
- (1) Moderate impairment—Walks 6 m (20 ft), slow speed, abnormal gait pattern, evidence for imbalance, deviates 25.4–38.1 cm (10–15 in) outside 30.48-cm (12-in) walkway width. Requires more than 9 seconds to ambulate 6 m (20 ft).
- (0) Severe impairment—Cannot walk 6 m (20 ft) without assistance, severe gait deviations or imbalance, deviates greater than 38.1 cm (15 in) outside 30.48-cm (12-in) walkway width or will not attempt task.

9. AMBULATING BACKWARDS

Instructions: *Walk backwards until I tell you to stop.*

Grading: Mark the highest category that applies.

- (3) Normal—Walks 6 m (20 ft), no assistive devices, good speed, no evidence for imbalance, normal gait pattern, deviates no more than 15.24 cm (6 in) outside 30.48-cm (12-in) walkway width.
- (2) Mild impairment—Walks 6 m (20 ft), uses assistive device, slower speed, mild gait deviations, deviates 15.24–25.4 cm (6–10 in) outside 30.48-cm (12-in) walkway width.
- (1) Moderate impairment—Walks 6 m (20 ft), slow speed, abnormal gait pattern, evidence for imbalance, deviates 25.4–38.1 cm (10–15 in) outside 30.48-cm (12-in) walkway width.
- (0) Severe impairment—Cannot walk 6 m (20 ft) without assistance, severe gait deviations or imbalance, deviates greater than 38.1 cm (15 in) outside 30.48-cm (12-in) walkway width or will not attempt task.

10. STEPS

Instructions: *Walk up these stairs as you would at home (ie, using the rail if necessary). At the top turn around and walk down.*

Grading: Mark the highest category that applies.

- (3) Normal—Alternating feet, no rail.
- (2) Mild impairment—Alternating feet, must use rail.
- (1) Moderate impairment—Two feet to a stair; must use rail.
- (0) Severe impairment—Cannot do safely.

TOTAL SCORE: _____ MAXIMUM SCORE 30

^a Adapted from Dynamic Gait Index.¹ Modified and reprinted with permission of authors and Lippincott Williams & Wilkins (<http://lww.com>).

Appendix 5. pediatric-modified Functional Gait Assessment (pmFGA)

(Modified from Wrisley, D. M., Marchetti, G. F., Kuharsky, D. K., & Whitney, S. L. (January 01, 2004). Reliability, internal consistency, and validity of data obtained with the functional gait assessment. *Physical Therapy*, 84(10), 906-918.)

Grading: Mark the lowest category, which applies. Repeat if doubt.

Demonstration: All items should be demonstrated.

Scoring: For each item, record the total steps before lose of balance of not following the instructions. Then, record the quality score based on the criteria. There is a maximum of 15 steps for each item.

1. Walking with narrow base support _____ *Instructions:* Walk on the floor with arms folded across the chest, feet aligned heel to toe. Begin walking at your comfortable speed.

(3) Normal: Able to ambulate for 15 steps heel to toe with no staggering and without body-sway over 45 degree from the center.

(2) Mild impairment: Able to ambulate 10-15 steps heel to toe, but with staggering or with body-sway over 45 degree from the center.

(1) Moderate impairment: Able to ambulate 4-9 steps heel to toe, with staggering or with body-sway over 45 degree from the center.

(0) Severe impairment: Ambulates less than 4 steps heel to toe without assistance.

2. Walking with eyes closed _____ *Instructions:* Walk at your comfortable speed from here to the next mark (6m(20ft)) with your eyes closed.

(3) Normal: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 15.24cm (6in) outside 30.48cm(12in) walkway width.

(2) Mild impairment: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 15.24-25.4cm (6-10in) outside 30.48cm(12in) walkway width.

(1) Moderate impairment: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 25.4-38.1cm (10-15in) outside 30.48cm(12in) walkway width.

(0) Severe impairment: Unable to perform 15 steps without assistance or deviates greater than 38.1cm (15in) outside 30.48cm (12in) walkway width.

3. Walking backwards _____ *Instructions:* Walk backwards until I tell you to stop.

(3) Normal: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 15.24cm (6in) outside 30.48cm(12in) walkway width.

(2) Mild impairment: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 15.24-25.4cm (6-10in) outside 30.48cm(12in) walkway width.

(1) Moderate impairment: Ambulates 15 steps without assistance, consistent speed, normal gait pattern, no evidence of imbalance, deviates no more than 25.4-38.1cm (10-15in) outside 30.48cm(12in) walkway width.

(0) Severe impairment: Unable to perform 15 steps without assistance or deviates greater than 38.1cm (15in) outside 30.48cm (12in) walkway width.

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