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Instrumented finger-to-nose test classification in children with ataxia or developmental coordination disorder and controls

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ABSTRACT

Background: During childhood, many conditions may impact coordination. Examples are physiological age-related development and pathological conditions, such as early onset ataxia and developmental coordination disorder. These conditions are generally diagnosed by clinical specialists. However, in absence of a gold phenotypic standard, objective reproducibility among specialists appears limited.
Methods: We investigated whether quantitative analysis of an upper limb coordination task (the finger-to-nose test) could discriminate between physiological and pathological conditions impacting coordination. We used inertial measurement units to estimate movement trajectories of the participants while they executed the finger-to-nose test. We employed random forests to classify each participant in one category.
Findings: On average, 87.4% of controls, 74.4% of early onset ataxia and 24.8% of developmental coordination disorder patients were correctly classified. The relatively good classification of early onset ataxia patients and controls contrasts with the poor classification of developmental coordination disorder patients.
Interpretation: In absence of a gold phenotypic standard for developmental coordination disorder recognition, it remains elusive whether the finger-to-nose test in these patients represents a sufficiently accurate entity to reflect symptoms distinctive of this disorder. Based on the relatively good results in early onset ataxia patients and controls, we conclude that quantitative analysis of the finger-to-nose test can provide a reliable support tool during the assessment of phenotypic early onset ataxia.

1. Introduction

Coordination is the process that allows motor performance through interactions of particular groups of muscles (Miller-Keane and O'Toole, 2003). This term can also be used to describe harmonious movement executions of several muscles (Farlex_Inc, 2004). Coordination involves the complex integration of motor and multisensory feedback signals by different body parts to perform smooth and efficient goal directed movements. Important aspects for accurate coordination are the knowledge of where and how the body is located in space (proprioception) and correct estimation of the intended end point of the movement (Groh, 2014). The cerebellum is involved in fine-tuning many of the processes related to coordinated movements. It evaluates, influences and modifies the information it receives from a vast number of multi-sensory inputs (Haines and Dietrichs, 2012). Important sensory input sources for coordination are the muscle receptors (informing about location, speed and orientation of muscles), otoliths and semicircular canals in the ear (informing about head position, which is important for balance) and visual-spatial information from the eyes (for estimation of distances of intended targets) (Bodranghien et al., 2015).

Impaired coordination is associated with many pediatric conditions such as ataxia, developmental coordination disorder (DCD) and physiologically immature coordination in young children. Children with physiologically immature coordination are considered healthy by clinicians and by their parents even if their coordination is sub-optimal compared to adults (Brandsma et al., 2014; Sival and Brunt, 2009). For optimal clinical diagnosis, surveillance and treatment evaluation, it is important to distinguish between these three underlying conditions and to obtain an objective reliable biomarker for quantitative assessment. Typical symptoms of ataxia that can be used for its diagnostic recognition are interruptions, exaggerated corrections and errors in position, direction, and velocity during goaldirected movements (D'Angelo and De Zeeuw, 2009; Manto et al., 2012). Due to abnormal sensory input or deficits in cerebellar fine tuning, goal directed

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movements, gait and kinetic function may show ataxic features such as overshooting, impaired timing, intention tremor and increased curvature of trajectories (Manto et al., 2012). These symptoms are not always clearly present in all domains of ataxic patients and, furthermore, features mimicking ataxia may be present in patients under other conditions, as well. These overlaps can hinder a correct diagnosis and indicate the need for consensus on symptom evaluation. The term early onset ataxia (EOA), is used for ataxia that starts before the 25th year of life (Chio et al., 1993; Harding, 1983). The diagnosis of DCD is often employed by rehabilitation specialists to specify a condition involving chronically impaired coordination, after exclusion of an explanatory medical diagnosis and/or major movement disorder (such as ataxia) (Jucaite et al., 2003). Children with DCD have difficulties in reaching motor milestones (such as grasping, sitting and standing), sensory-motor integration, postural control and visual-spatial planning (Wilson et al., 2013; Zwicker et al., 2009; Zwicker et al., 2012). As pediatric coordination data should be interpretable against healthy age-related reference values, it is important to discern these conditions from immature coordination (Brandsma et al., 2014; Sival and Brunt, 2009), which is explained by ongoing cerebellar growth and development continuing until 17 years of age (Brandsma et al., 2014; Tiemeier et al., 2010). In absence of a sufficiently reliable biomarker to differentiate between these underlying conditions, diagnostic methods depend on subjective recognition by clinical specialists. We have shown that the phenotypic inter-observer agreement by clinical experts of ataxia is of moderate strength (Fleiss kappa = 0.45) (Lawerman et al., 2015). Distinction between mild ataxia, DCD and physiological immature motor coordination can be very challenging as well.

This induced the question whether semi-quantitative rating scales could support the phenotypic recognition of the underlying disorders for coordination impairment. In children, the most frequently applied ataxia rating scales are: the Scale for Assessment and Rating of Ataxia (SARA) (Schmitz-Hübsch and Montcel, 2006) (a recently developed, reliable, rating scale for the assessment of coordination in the domains of gait, upper and lower limbs and speech) and the International Co-operative Ataxia Rating Scale (ICARS) (Trouillas et al., 1997). These scales are designed to quantify the severity of ataxia during different tasks. In a recent study we analyzed movement execution of the same group of children during one of these tasks, gait, using a similar methodology (Mannini et al., 2017). The correct identification of these conditions using a single motor task in one domain is highly unlikely or even not possible. However, future integration of the results of objective assessments of different tasks in gait, posture and kinetic domains can lead to a more accurate evaluation. Although the SARA was originally designed to evaluate ataxia severity, Brandsma et al. showed that the scale also reflects other causes of coordination impairment (such as chorea, myoclonus and dystonia) (Brandsma et al., 2016). However, until now, it is unknown whether quantified parameters of SARA and ICARS, (including the finger-to-nose test), could differentiate between different disorders of coordination impairment.

This study is part of a larger project that intends to quantify the different tasks described in the SARA. A reliable identification of conditions such as EOA or DCD based on only one task is very challenging or even impossible as the symptoms affecting the patient may appear only during certain tasks and this can differ between patients with similar disorders. Analogous to our study on the automatic classification of SARA-gait (Mannini et al., 2017), we aimed to evaluate whether automatic classification of the SARA finger-to-nose test could also discern between EOA, DCD and physiological immaturity. For this purpose, we use machine learning techniques. To train our model we used individual task executions labeled with the diagnosis of the participant. Since each participant was asked to perform the task at least ten times, and the majority was able to complete the test, we obtained a dataset of > 300 samples. We based our analysis on the description of the assessment of the finger-to-nose test of SARA and ICARS and on the remarks of pediatric neurologists. We believe that, to achieve a reproducible tool that will be accepted and can be used in clinical practice, it is important to follow the guidelines established by clinical neurologists. Setting aside the consensus achieved across a large number of stakeholders to develop SARA and ICARS and creating custom evaluations and custom features, might lead to a tool unfamiliar and unacceptable to clinical evaluators and that might therefore not be usable in clinical practice. Based on this, we focused our analysis on the evaluation of dysmetria. We compared the results of the objective classification against the phenotypic assessment of two clinicians based only on the finger-to-nose test videos. In general, we expect the finger-to-nose test trajectories in control children to be more even and uniform, having fewer interruptions and abrupt changes than the ones of EOA and DCD children of the same age. Under the premise that EOA and DCD diagnoses concern different visually discernible categories of coordination impairment, we expect that the automatic analysis of the finger-to-nose test will provide reliable information that, in conjunction with the objective analysis of other evaluation tests, can lead to a reliable automatic classification. If so, automatic classification of the finger-to-nose test performances would provide an objective biomarker for phenotypic and quantitative coordination assessment in young children. Moreover, the quantification of movement performance could help to monitor motor control development during follow-up evaluations.

2. Methods

2.1. Participants

The study was performed in accordance with the research and integrity codes of the UMCG. The Medical Ethical Committee of the UMCG provided a waiver for ethical approval because the clinical finger-to-nose test was performed as part of routine clinical assessment in patients, has been shown to be harmless and painless to the child and its execution did not involve any risks. After informed consent by the parents, we included nine EOA, seven DCD and 16 healthy age-matched control children. The inclusion criteria for EOA were a clinical diagnosis of pediatric ataxia and/or recognition of ataxia as primary movement disorder, independently assessed by three pediatric neurologists with expertise in movement disorders. The inclusion criteria for DCD were exclusion of a movement disorder by a pediatric neurologist and an officially established diagnosis of DCD in a rehabilitation center. All pediatric patients performed the finger-to-nose test as part of their routine clinical SARA evaluation. We recruited healthy controls by advertisement. Healthy young children were not diagnosed with a neurological or orthopedic disorder, and were declared to be healthy by their parents. The children did not receive medication with a known negative side-effect on their coordination.

2.2. Clinical assessment

We videotaped the SARA performances of included patients and healthy controls. Prior to phenotypic assessment, video-recordings were stripped of identity tags for anonymous phenotypic and semi-quantitative (SARA) assessment. Two pediatric neurologists independently phenotyped the anonymous videotapes in random order, not aware of the underlying diagnosis. The pediatric neurologists indicated whether they observed ataxia as primary movement disorder, or DCD, or neither during the assessment of the upper limb coordination tests. In all children, we assessed SARA according to the official guidelines (Schmitz-Hübsch and Montcel, 2006). We compared differences between age, total SARA scores and finger-to-nose scores between the three subgroups by one-way ANOVA test in case of normally distributed variables and by the Kruskal-Wallis test (with a post hoc Mann-Whitney comparison when significant) in case of non-normally distributed variables. We assumed a significance level of $\alpha = 0.05$.