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Prevalence and predictors for the ability to run in children and adolescents with cerebral palsy

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ABSTRACT

Background: Running is a fundamental movement skill and a prerequisite for children to participate in numerous daily activities. The prevalence of the ability to run in people with Cerebral Palsy and the role of their impairments on running ability are unknown. Therefore, the aim of this study is to determine the prevalence of the ability to run and to identify contributing factors.
Methods: In this study, 280 children and adolescents with spastic Cerebral Palsy, Gross Motor Function Classification System level II were included. The ability to run was defined by instrumented running analysis. Runners and non-runners were compared regarding their clinical measures of spasticity, weakness, and postural control. Logistic regression was applied to identify the most important predictors for the ability to run.
Findings: The ability to run was significantly higher in unilateral (67%) than in bilateral (55%) affected patients. Significant differences between runners and non-runners were found for spasticity, BMI and postural control, but not for muscle strength. Lower *M. rectus* femoris spasticity, higher m gastrocnemius spasticity and enhanced postural control appear to be the best predictors for being able to run.
Interpretation: Patients with Gross Motor Function Classification System level II represent a large group in the gait laboratory and the functional impairment within this group differs greatly. Therefore, for clinical decision making we suggest to separate patients in this group based on their running ability. Spasticity and postural control affect the ability to run and needs to be accounted for in intervention programs.

1. Introduction

Running is an important skill for children in everyday life. It is a prerequisite for many recreational activities and a necessity in order to keep up with their peers. Consequently, limitations in activity and motor function lead to restriction in everyday mobility, education and social relationships (Beckung and Hagberg, 2002). Adequate physical activity prevents secondary impairments and chronic health conditions not only in normally developed children but also in people with Cerebral Palsy (CP) (Brunton and Bartlett, 2010). Being able to walk without restrictions is not only desirable for social and community participation (Palisano et al., 2009), but it has also been proven that running improves the participation of students with CP in school environments (Gibson et al., 2017). Furthermore, the ability to participate in sports positively influences the quality of life (Groff et al., 2009). Young people with CP are less active and spend less time doing sports activities than normally developed youth (Bjornson et al., 2007). Identifying the factors that contribute to and influence the ability to run is therefore key to improve the lives of people affected with CP.

As a fundamental movement skill, running is assessed as part of the Gross Motor Function Classification System (GMFCS) which is the mainstay in the description of motor function in CP (Palisano et al., 2008). In detail, GMFCS level I children “perform gross motor skills such as running and jumping, but speed, balance, and coordination are limited”, whereas, level II “children have at best only minimal ability to perform gross motor skills such as running and jumping”. Accordingly, some patients rated as GMFCS II may have the ability to run, while others do not, which makes this group especially interesting, since the line is drawn between the ability and inability to run. In addition to the GMFCS level, patients with CP are categorized based on their limb involvement as uni- and bilateral (Cans et al., 2007). It can be expected that the prevalence of the ability to run is higher in unilateral involved patients, since these patients profit from a higher functioning unaffected limb partly compensating for the impaired side during walking

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(Damiano et al., 2006a, 2006b).

From a body's structural and functional level, the ability to run may be limited by spasticity, weaknesses, contractures and deficient postural control. Spasticity, which is defined by a velocity-dependent exaggeration of stretch reflexes (Young, 1994), has proven to prevent affected patients from achieving faster walking velocities (Damiano et al., 2006a, 2006b), and is therefore likely to have an influence on the ability to run. Especially, the spasticity of the rectus femoris muscle and its restricting effect on the knee flexion has been described as a central issue in CP (Jonkers et al., 2006). During the swing phase of running, knee flexion ability gets even more important (Böhm and Döderlein, 2012; Davids et al., 1998), exaggerating the detrimental effects of the rectus spasticity. Contrary, spasticity of the calf muscle may help to generate a higher vertical stiffness of the leg during the stance phase of walking (Hösl et al., 2016). Since CP children have lower muscular strength available, spasticity may have positive effects in terms of leg stiffness and elastic energy retrieval, partly compensating for the weakness (Fonseca et al., 2004).

Muscle weakness is another pathological impairment in CP (Mockford and Caulton, 2010). Since running demands higher ankle plantarflexion, knee and hip extension, as well as flexion moment generation (Böhm and Döderlein, 2012; Davids et al., 1998), a minimum of muscle strength may be required for the ability to run.

In contrast, contractures appear to play a minor role when it comes to the ability to run. Studies have found more closely related kinematic joint patterns of children with CP during running, compared to walking of normally developed children. This leads to the conclusion that the pathologically increased hip, knee and ankle plantar flexion is less detrimental for running mechanics (Böhm and Döderlein, 2012; Davids et al., 1998).

Postural control is the ability to control the body position in order to achieve orientation and stability, which demands complex interactions between sensory system, central nervous system, and muscle skeletal system (Woollacott and Shumway-Cook, 2005). In patients with cerebral palsy (CP), these interactions are known to be affected, which may be a reason why postural control is impaired and the maintenance of stability is critical (Woollacott and Shumway-Cook, 2005). The single support phase during running poses higher demands on the stability than the double support phase during walking, so that running may require a certain level of postural control.

The prevalence of the ability to run in patients classified as GMFCS II and the role of structural and functional CP related impairments on the ability are unknown today. Therefore, the objective of this study is to determine the prevalence of the ability to run in children and adolescents with spastic CP and to identify contributing factors.

Hypotheses are:

1. In patients rated as GMFCS II, > 50% are able to run. In addition, patients with unilateral involvement have a higher prevalence.
2. Predictors of the ability to run are spasticity, weaknesses, and postural control. Rectus spasticity is negative and gastrocnemius spasticity positive for the ability to run. Furthermore, a greater strength in the leg muscles as well as a higher postural control is required for being able to run.

2. Method

2.1. Participants and data collection

Data were retrospectively analyzed from all people with infantile spastic CP who had been referred from physical examination to the Gait Analysis Laboratory for evaluation of their gait and assessment of possible orthotic or orthopedic interventions between January 2009 and April 2018. All children provided written consent and the study was approved by the local ethics committee.

For inclusion, participants had to be classified as GMFCS level II and were aged between 6 and 17 years. Excluded were patients with additional syndromes, pain induced limitation to run, missing compliance, profound visual impairments or obesity according to the age dependent body mass index threshold (Cole et al., 2000). Patients that had undergone surgical interventions within the last two years were excluded so that all patients had sufficient recovery time from their intervention. Trials with shoes or orthotics were excluded and all trials were performed barefoot. If patients were analyzed multiple times, the first sessions that met the inclusion criteria was chosen.

Kinematics was captured using an eight-camera Vicon MX system (Vicon Inc., Oxford, UK) and the Plug-in-Gait model of the lower extremity (Davis et al., 1991). Kinetics were measured simultaneously by two force plates (AMTI, Watertown, MA). The participants were first instructed to walk over the 13 m walkway with a self-selected speed. Afterwards the patients had time to recover and were then asked to run at a comfortable self-selected speed, according to their capabilities. Running was defined by a phase of double float, where neither foot touches the ground (Novacheck, 1998). This phase of double float had to be shown bilaterally for at least three gait cycles. The gait events touch down and take-off were automatically defined by the event detection algorithm of the Nexus 1.8 software (Vicon, UK). A double float phase existed when the take-off event of the contralateral leg occurred before the touch-down event of the ipsilateral leg. Five consistent trials, determined by visual inspection of the kinematic and kinetic wave forms, were used for further data procession.

Subsequent to the gait analysis, a standardized clinical examination was performed by trained gait laboratory staff with at least two years of experience. To test the postural control, patients were asked to do as many vertical single leg jumps as possible and to perform a single leg balance test as long as possible, with eyes open and arms on the hips.

To examine the muscle weakness, the testing scheme by Kendall was used (Kendall et al., 2005). Maximal active strength of the hip flexion, knee extension and flexion and ankle plantar and dorsiflexion was tested in seated position with the pelvis stabilized. Hip extension strength was tested in prone position with the knees extended.

Muscle spasticity of the rectus femoris was tested in prone position using the Duncan Ely test (Bohannon and Smith, 1987). Spasticity of the calf muscles was tested supine in extended and flexed knee position to examine gastrocnemius and soleus spasticity respectively. Spasticity was rated according to the Modified Ashworth Scale (MAS) (Bohannon and Smith, 1987).

2.2. Data processing and statistical analysis

The patients were divided into groups of uni- and bilateral involvement and within these groups into runners and non-runners. In unilateral patients the involved side was analyzed. In bilateral patients the Gait Profile Score (GPS) (Baker et al., 2009) was utilized to divide the legs into a more and a less affected side with a greater and smaller GPS respectively. Because of the interlimb dependence (Sangeux et al., 2013), only the side with the greater GPS was analyzed.

Chi-square test was used to evaluate differences in prevalence of running ability between patients with uni- or bilateral CP and the number of patients with previous surgeries. A two factor ANOVA on the factor laterality (uni- and bilateral) and running ability (non-runners and runners) was performed on the parameters shown in Table 1. Parameters that were found to be significantly different between the runners and non-runners groups underwent a predictor analysis using logistic regression to reveal the contribution on the probability being able to run. The statistical significance level was set to $p = 0.05$.

3. Results

In total 280 patients met the inclusion criteria. Overall, prevalence of running in children and adolescents with CP and GMFCS II was 58%. The ability to run was significantly higher in uni- than in bilaterally