Clinical exercise testing in pediatric rehabilitation

Children and young adults with a physical disability, such as cerebral palsy (CP), often experience problems with walking, including fatigue during or after walking, a reduced walking distance, and/or a reduced walking speed. These problems are likely to be primarily caused by their motor impairments that can cause a pathological gait. Adequate insight into the underlying physical mechanisms behind these walking problems can direct rehabilitation treatment. Physical examination and gait analysis are often used by rehabilitation practitioners to substantiate their treatment for individuals with these walking problems. However, studies have suggested that i) increased energy demands of walking and ii) decreased physical fitness values may also cause walking problems in children with CP. Nevertheless, assessment of these fitness parameters is not yet part of clinical routine. Clinical exercise tests can be used to reliably measure the energy demands of walking and physical fitness. Therefore, the work in this thesis focused on assessing the usefulness of clinical exercise tests for children and young adults with physical disabilities.

Clinical exercise testing encompasses many different tests. In this thesis we focus on three main exercise tests that were performed in a clinical laboratory. The energy demands of walking were assessed using a walking test at a self-selected speed. One of the primary outcomes of this walking test is the gross energy cost (EC), which is the amount of energy used per meter (during walking). The two components of physical fitness that were assessed are the aerobic and anaerobic fitness. Aerobic fitness is essential for daily physical activities and exercise at low to moderate intensity for longer durations. The primary outcome for aerobic fitness is the maximal oxygen uptake (VO\textsubscript{2peak}), and the secondary outcome is the anaerobic threshold, i.e. the point above which energy production is supplemented by anaerobic glycolysis. Both of these outcomes were measured during a maximal exercise test on a cycle ergometer. Anaerobic fitness is of major importance for short-duration high-intensity exercise. A 20-second Wingate test was performed to measure the primary outcome of the anaerobic fitness, i.e. the mean anaerobic power over 20 seconds.

In chapter 2, we investigated the proportion of individuals with deviated gross EC and VO\textsubscript{2peak} values among the children and young adults with a physical disability seeking medical advice for walking problems. First, cut-off values based on references values for typically developing peers were defined in order to distinguish between normal, mildly deviated and strongly deviated gross EC and VO\textsubscript{2peak}. A mild or strong deviation of either gross EC (> 2SD compared to typically developing peers) or VO\textsubscript{2peak} (< 25\textsuperscript{th} percentile
compared to typically developing peers of the same gender) indicates that an intervention to reduce walking problems by lowering the gross EC or increasing the $V_{O_{peak}}$ is required. A mildly increased (between 2SD and 3SD) gross EC was present in 13% and a mildly decreased (between 10th and 25th percentile) $V_{O_{peak}}$ was present in 12% of the individuals with physical disabilities. The majority even had a strongly (> 3SD) increased gross EC (51%) and a strongly (< 10th percentile) decreased $V_{O_{peak}}$ (60%).

Many individuals with physical disabilities who experience problems with walking thus had deviated gross EC and $V_{O_{peak}}$ values. Therefore, chapter 2 also examined whether patient characteristics (i.e. diagnosis, gender, age, body height, body mass index and walking speed) can be used to predict an increased gross EC or a decreased $V_{O_{peak}}$. Unfortunately, predicting these outcomes was not possible for individuals with CP classified as Gross Motor Function Classification System (GMFCS) levels I and II, and individuals with other neurological diagnosis. Therefore, clinical exercise tests are recommended in this population to guide treatment. Since all the individuals with CP, classified as GMFCS levels III or IV, had deviated gross EC and $V_{O_{peak}}$, lower motor functioning (i.e. a higher GMFCS level) is predictive of deviated gross EC and $V_{O_{peak}}$ for this population.

When individuals with physical disabilities have higher energy demands of walking and/or have a reduced $V_{O_{peak}}$ compared to their typically developing peers, their physical strain of walking is high. Physical strain provides information on the intensity of walking and is defined as the oxygen uptake of walking ($V_{O_{walk}}$) expressed as a percentage of $V_{O_{peak}}$. Chapter 3 describes the average physical strain of walking for children and adolescents with CP classified as GMFCS levels I, II and III. Individuals with CP had a significantly higher physical strain (GMFCS levels I: 55 ± 12%; II: 62 ± 17%; and III: 78 ± 14%) than their typically developing peers (40 ± 11%) during walking at a self-selected speed. According to training guidelines for typically developing individuals, walking at a self-selected speed is moderate exercise for individuals classified as GMFCS levels I and II, but can even be considered vigorous exercise for children classified as GMFCS level III. Furthermore, chapter 3 shows that 43% of these CP individuals even walk at or above their anaerobic threshold, compared to 10% in typically developing peers.

Considering the high physical strain of individuals with CP during walking, it is not surprising that complaints of fatigue are frequently reported by this population. Therefore, chapter 4 investigated whether fatigue in children/young adults with physical disabilities is related to the energy demands of walking and to the physical fitness. Two dimensions of fatigue were assessed. First, general fatigue, i.e. the experience of feeling
tired, weak or lacking energy, as measured with the subscale ‘subjective fatigue’ of the Checklist Individual Strength (CIS8R) questionnaire. Because general fatigue does not relate to any of the outcomes of clinical exercise tests, we argue that general fatigue probably has other mental and physical causes. Second, walking-induced fatigue was measured by asking individuals the rate of perceived exertion after 6 minutes of walking at a self-selected speed with the OMNI (OMNIwalk). In the whole sample, a higher net EC (i.e. gross EC corrected for resting energy consumption) was weakly related to an increased OMNIwalk. However, in individuals aged 12 – 22 years low anaerobic fitness and high physical strain values were moderately related to high OMNIwalk scores. Therefore, interventions aimed at increasing anaerobic fitness and reducing physical strain might reduce walking-induced fatigue in individuals aged 12 – 22 years with physical disabilities.

Energy demands of walking can be used to evaluate treatment in children with CP. In typically developing children, the energy demands of walking decrease as they get older (even up to adult age) and it is assumed that this is similar for individuals with CP. Therefore, the cross-sectional study in chapter 5 assessed the association between the energy demands of walking and age for children and young adults with CP, compared to their typically developing peers. Three outcomes for the energy demands of walking were used: 1) the gross EC, 2) the net EC, and 3) the net nondimensional (NN) EC, which is corrected for resting energy consumption, leg length and gravity. Both the net EC and NN EC were corrected to control for the effects of growth. The results showed that both the typically developing and the individuals with CP walk more efficiently when they are older, as indicated by a decline of all three energy cost outcomes with increasing age. The gross EC showed the largest decline (-3% per year), whereas the net and NN EC showed a similar decline (-1.5% per year). Therefore, correcting for this decline is necessary for all three outcomes when evaluating individual changes after treatment.

Research has identified a need for reliable and valid tests to assess physical fitness in children with CP classified as GMFCS levels III and IV. While the above-described laboratory tests are indicated for research purposes and clinical decision-making, field tests are easier to perform. Therefore, chapter 6 evaluated the test-retest reliability of a novel 6-Minute RaceRunner Test (6MRRT) for individuals aged 5 – 19 classified as GMFCS levels III and IV. The racerunner is an alternative walking and/or training device for individuals with limited walking abilities. This device consists of a 3-wheeled frame, handlebars, a saddle, and a trunk support. Individuals can propel themselves forward by stepping their feet on the ground while seated. Because the intraclass correlation coefficient (ICC) is 0.89 for individuals classified as GMFCS level III and 0.91 for
individuals classified as GMFCS level IV, the 6MRRT is a reliable test for measuring the distance covered with the racerunner. However, because the smallest detectable difference (SDD) is large, children and young adults with CP need to improve more than 37% (GMFCS level III) or 52% (GMFCS level IV) in distance before one can rule out the influence of measurement error. Therefore, a single 6MRRT measurement is only useful for individual evaluation when large (> 188 m) improvements are expected. Another important finding is that 25 of the 38 individuals reached a heart rate of 180 beats per minute or higher, which means that these individuals reached a (near) maximum effort during the test. This indicates that the racerunner could be a promising device to use during a fitness training and as a device to measure the VO_{peak} during a maximal exercise test.

Chapter 7 presents an extensive discussion of our studies and their clinical implications for (pediatric) rehabilitation. The main findings of this thesis indicate that both high gross EC and low VO_{peak} values are common in children and young adults with physical disabilities. One or both of these deviated values result in high physical strain values of walking. It is important to distinguish between these different causes, because each cause requires a different intervention strategy. When the outcomes of both tests are used, the recommended treatment for individuals can be better targeted to their walking problems. In sum, appropriate use of the outcomes of clinical exercise tests improves the diagnostic evaluation of walking problems in children and young adults with physical disabilities. However, whether walking problems will be reduced by improvement of the physical strain by i) either lowering the energy demands of walking, or ii) improving the aerobic fitness or anaerobic threshold, or iii) improving the anaerobic fitness for children and young adults with physical disabilities, remains to be determined in future research.