Measuring physical fitness in persons with severe or profound intellectual and multiple disabilities
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Chapter 9

General Discussion
As this thesis is comprised of multiple studies related to psychometric characteristics of measurements concepts when testing for a multiple disabled target group, a solid overview of the results and their relations is a necessary and natural last step. This discussion is roughly divided into four parts. The first part touches upon general findings, which deal with classification, test setting and adjustments in formulas. Then, this discussion goes into more detail discussing results grouped per studied concept: body composition, functional exercise and aerobic capacity, balance, flexibility and physical activity respectively. Additionally, the third part deals with the methodological issues encountered during research; lack of standards and cut-off scores, small sample size and heterogeneity of the study population. The final part of this discussion seeks to give an overview of the implications of the different studies comprised in this thesis, as well as to indicate recommended areas of further research.

**General Findings**

**Classification and Test Setting**

The subjects of this thesis have severe or profound intellectual as well as visual disabilities and the study population is thus referred to as persons with severe or profound intellectual and multiple disabilities (SPIMD). The term ‘multiple’ includes locomotor disabilities, neurological problems, sensory disabilities, and/or problems with food ingestion.

An important topic that came up during the studies of this thesis [all chapters] is the importance of combining both severity of intellectual disabilities as locomotor levels (GMFCS) to classify the abilities of persons with severe or profound intellectual disabilities. Only if both classifications are used, the appropriate physical fitness tests can be individually selected. Contrary to common assumptions, persons with profound intellectual disabilities do not automatically have low locomotor levels. The ability to walk varies considerably in persons with severe intellectual disabilities as well as in those with profound intellectual disabilities. Specifically, 75% of the subjects with severe intellectual disabilities were able to walk, leaving 25% which were not able to walk. In contrast, 56% of the subjects with profound intellectual disabilities is able to walk, whereas 44% is not able to walk [1, 2]. Not testing for GMFCS levels thus means ignoring a big potential for both research as improvement of individual physical fitness.

Also, contrary to common beliefs, persons with SPIMD can become accustomed to test and measurement situations despite their severe or profound intellectual and multiple disabilities, if optimal test environment and conditions are created. Testing in persons with SPIMD in a feasible, valid and reliable way is possible -although challenging due to the limitations in intellectual functioning, adaptive behaviour [3] visual impairment [4], low motivation and adherent stress [5, 6].

This thesis made use of environmental cues to counter these limitations. Environmental cues included adaptations of existing test protocols, performing practice sessions with a familiarization protocol, testing at the regular physical activity hours in the regular sports hall accompanied by familiar gymnastic instructors, who were used to motivate the participants. In addition, the use of a treadmill to assess cardiorespiratory fitness is shown to be an effective environmental cue. Following theories about perception-action coupling [7, 8], these environmental cues may have facilitated the performance of the subjects by both reducing stress as enhancing motivation.

Technology could be used to develop other environmental cues such as auditive or other sensory stimuli, which may even further ameliorate the testing situation. Future research should focus...
on this. To sum up, testing in persons with SPIMD in a feasible, valid and reliable way is possible, if participants are classified according to both GMFCS levels and level of disability, and if optimal test environment and conditions are created.

**Adjusted Formulas**

This thesis has brought forth a couple of important findings regarding formulas or equations applicable to research on this specific target group. First of all, it proposes a new reliable equation to calculate body height from tibia length [1]. Second, a simple prediction equation is suggested for predicting standing waist circumference from supine waist circumference measurement [2]. The latter equation allows the comparison of supine measurements of waist circumference in disabled persons with international standards [9]. Third, we also found that the Fernhall’s formula [10, 11] for calculating peak heart rate for people with ID, systematically overestimates peak heart rate for people with SIMD [chapter 5]. The subheader “Functional exercise and aerobic capacity” discusses this finding in more detail.

Overall, whereas for other specific target groups such as the elderly, children with cerebral palsy, and persons with mild or moderate ID feasible and reliable fitness tests already were available [5, 12, 13, 14, 15], the feasibility and reliability of physical fitness measurements and tests for participants with SPIMD had so far not been established. Using the feasible and reliable fitness tests described in this thesis, physical fitness levels can now be objectively evaluated. Moreover, specific interventions aimed at promoting physical fitness of participants with SPIMD can be evaluated objectively as well.

Not only does this study thus contribute to existing literature, it also, quite practically delivers tools for the work floor. For instance, the fitness tests that were evaluated in chapters 2 to 8 of this thesis have proven to be feasible, valid and reliable for persons with severe or profound intellectual and multiple disabilities, and could thus immediately be implemented in daily practice. What follows is a more detailed overview of the results per studied fitness concept.

**Body composition**

Anthropometric measurements are widely used to reliably quantify body composition and to estimate risk of overweight in both healthy subjects and patients. Body composition of persons with ID is described in several studies [16, 17, 18]. However, information about the reliability of anthropometric measurements in persons with SIMD had so far not been subject to research. In 45 participants with SIMD, body mass index, waist circumference, skinfolds, and tibia length were measured twice [1]. The results of this study show that for individuals with SIMD and GMFCS levels I and II, feasibility and test-retest reliability for body composition measurements were acceptable, with exception of the skinfold measurements. Body Mass Index is calculated using body height and body weight. However, body height is difficult to measure for those subjects unable to stand upright. Therefore, a study was set up in order to calculate body height from tibia length. This study [1, chapter 2] revealed a new reliable equation to calculate body height from tibia length. Measuring tibia length is more feasible, accurate, and reliable than measuring the total body height of participants with SIMD- both for those able to stand as for those not able to do so.

Waist circumference as an indicator of abdominal fat is an important predictor of health risks and is usually measured in standing position. We found that measuring waist circumference in participants with SIMD who are able to stand upright is feasible and reliable. However,
due to motor disabilities, many individuals with PIMD and GMFCS levels IV and V are unable to stand straight or to stand at all. When dealing with these participants, waist circumference can only be measured with the participant lying in a supine position. It had so far been unknown whether measuring waist circumference of a participant in a supine position is valid and reliable. This issue is particularly relevant when international standards [10] for healthy individuals are applied to the disabled. The results of a test-retest study [2, chapter 3] performed in 43 participants with PIMD and GMFCS levels IV and V indicated that supine waist circumference can be reliably measured for this target group. Our validity study [2, chapter 3] performed in 160 healthy participants, compared waist circumference obtained in both standing and supine positions. This study shows that supine waist circumference is biased towards higher values (1.5 cm) when compared with standing waist circumference. A simple equation could be put forward, which predicts standing waist circumference based on supine measurements. Such an equation allows for the comparison of waist circumference measurements of disabled persons with the international standards [10]. Based on BMI and waist circumference values, 10% of the female SPIMD subjects are obese and 39% are abdominal obese, while 0% of the male persons are obese and only 7% are abdominal obese. Thus, women with SPIMD are at a higher risk for developing overweight related health problems compared to SPIMD classified men. Compared to outcomes of measurements in persons with mild or moderate ID, persons with SPIMD show a more healthy body composition status [18, 19, 20]. Further research into the explaining mechanisms behind this interesting observation is recommended.

Another issue related to body composition stems from the fact that children and adults who have SPIMD and GMFCS level IV or V are often fed by stomach tube [21]. Tube feeding may increase body weight mainly through fat deposition [22]. Children and adults with severe CP have relatively high body-fat/muscle-content ratio. When the relatively low energy expenditure is also taken into account, we see a potential risk of overfeeding [23]. To establish a healthy body weight in persons with PIMD, it is necessary to take into account both BMI and waist circumference. Rieken et al [24] suggest to use bioelectrical impedance analysis as an alternative measurement of body composition and present a preliminary clinical guideline on diagnosing undernutrition and overnutrition in children with severe neurological impairment and ID. We recommend that a similar guideline will be designed for adults with SPIMD and GMFCS levels IV and V, so as to prevent over- or undernutrition in this target group.

**Functional exercise and aerobic capacity**

Cardiorespiratory fitness can be divided into functional exercise and aerobic capacity [35]. Several tests are available for measuring functional exercise and aerobic capacity, including the six-minute walking test (6 MWD) and the shuttle run test (SRT). However, it was unclear whether these tests are feasible and reliable when testing subjects with SIMD. Our studies [10, chapter 4] showed that the 6 MWD is feasible and reliable for testing participants with SIMD and GMFCS levels I and II. An adapted SRT (aSRT) performed overground as well as on a treadmill has proven to be feasible and reliable for a target group with SIMD and GMFCS level I [chapters 4 and 5]. Moreover, the aSRT performed on a treadmill [chapter 5] appeared to be more valid for determining peak heart rate than the aSRT performed overground for people with SIMD and GMFCS level I. We also found that the Fernhall’s formula [11] for calculating peak heart rate of subjects with ID systematically overestimates peak heart rate of subjects classified with SIMD. Thus,
for future research it is recommended to adjust this equation so as to ensure a valid prediction of the maximal heart of this specific group.

Furthermore, we compared the achieved mean distance in the 6MWD of our participants with values reported in other studies [25, 26, 27]. This comparison indicated that persons with SIMD perform poorer on the 6MWD than those with other specific (chronic) health conditions including heart failure or COPD. The poor 6MWD results we observed suggest that the low functional exercise capacity of individuals with SIMD is a serious health problem, which in turn can negatively affect their independence in day-to-day activities.

Based on this result, further research should be directed towards developing, implementing and evaluating interventions aimed at increasing functional exercise and aerobic capacity of SIMD classified subjects thereby reducing related health problems.

Balance

Sufficient balance is necessary to perform daily activities. However, individuals with SIMD are particularly at risk when it comes to the development of deficits in locomotor skills [28] and are likely to have decreased balance. Yet, the feasibility and reliability of balance tests for subjects with SIMD had so far not been subject of research. The Berg Balance Scale (BSS) is a widely used test to quantify balance. Our study [29, chapter 6] revealed a sufficient percentage of successful measurements by the modified BBS (mBBS), indicating this test to be a feasible instrument for testing subjects with SIMD and GMFCS levels I and II. In addition, with exception of two out of the twelve mBBS tasks to be performed by the participants, reliability for test and retest appeared to be sufficient.

In the BBS, a score of 80% (45 points) indicates sufficient balance [30]. As its modifications influence the cut-off value, it was impossible to apply the same cut-off value to the mBBS. A comparison with scores in other target groups was hence not possible. Future research should be directed towards establishing accurate cut-off values.

Flexibility

Muscle flexibility is a relevant part of physical fitness and can be measured using the Modified Ashworth Scale (MAS) and the Modified Tardieu Scale (MTS). The results of our study [31, chapter 7] showed the feasibility of the MAS and MTS to be sufficient for testing participants with PIMD. For both inter and intrarater reliability, measurements of the MAS revealed acceptable agreement. However, the measurements of the MTS showed insufficient agreement for both test-retest and interrater reliability.

For our target group the MAS showed a better test-retest and intrarater reliability than the MTS, which contradicts the results of both Gielen [32] and Mehrholz et al. [33] who found the opposite to be true for subjects with respectively ID and adult patients with severe brain injury. In their studies the psychometric properties of the MTS were better than those of the MAS. Our measurements of the MTS however, put forward LOAs not narrow enough to indicate agreement between the two measurements of the same subject, leading us to conclude the LOAs for the MTS measurements to be clinically unacceptable.

Other authors [34] have suggested the MTS to be a more appropriate clinical measure of spasticity than the MAS. We thus assessed whether the LOA of the MTS measurements of our subjects with spasticity was narrower than that of our subjects without spasticity.
Yet, no difference was found between these groups. For our target group, including those suffering from spasticity, MAS seems the preferred measurement instrument. This preference may be accounted for by the interaction between multiple disabilities in our study population, which may differ in that respect from the study populations those of Mehrholz [34] and Gielen [33].

However, the validity of the aforementioned instruments for measuring spasticity and muscle tone is beyond the scope of this study. We recommend the validity of the MAS to be examined in future studies, with a focus on measuring the quality of daily movement rather than measuring spasticity, as the MAS may be more suitable for measuring the first.

**Physical activity**

Daily physical activity contributes directly to health [35] and is an important factor in maintaining and improving physical fitness. Monitoring daily physical fitness in persons with SPIMD is thus an important step in identifying those at health risks.

Our research [chapter 8] has shown that daily physical activity can be reliably monitored through measuring heart rate patterns. Yet, further exploration of other influential factors such as emotions [36] is still recommended. Time of day and age have considerable influence on heart rate patterns. However, the analysis of heart rate patterns suggests other, probably more personal factors to have a significant influence on heart rate patterns [37] which can be explored in future research. Finally, our study shows that based on measuring heart rate patterns, individuals with PIMD do not meet the standards of sufficient activity as proposed by the guidelines of the American College of Sports Medicine [38]. Future research can shed a light on the suitability and validity of these guidelines for a population with PIMD or SPIMD.

**Methodological issues**

The first methodological issue we encountered had to do with the lack of a golden standard. As validity can only be examined against a golden standard, it would have been convenient and desirable for a golden standard to be available for every single instrument measuring a certain concept. Yet realistically, golden standards for our research population are unfortunately simply not available. If feasible, we attempted to assess validity, for example by performing a supra maximal block test on the treadmill.

Moreover, often neither standard values nor cut-off scores are available, as was the case for the aSRT nor the mBBS.

Another methodological issue is related to statistical power. Due to both the relatively small group of persons with SPIMD and a number of exclusion criteria, a rather small number of subjects were able to participate in the studies described in this thesis. However, despite the relatively small study groups the test-retest reliability of mBBS, walking tests, bodycomposition measurements and the MAS were comparable to those of other and larger target groups. In future research, more care facilities and thus subjects should be included and classified on both of level of ID and locomotor skills. This may improve sample size and thereby power. Then it might be possible to formulate golden standards, group specific standard values and cut-off scores, with which the validity of tests can be examined and the testscores can be compared.

Despite the fact that only cross-sectional designs are used in this research, we will touch upon the problems of heterogeniety in samples with SPIMD in the framework of intervention
studies. Randomized Controlled Trials (RCT’s) are considered to be the most reliable and valid way to perform intervention studies in various populations. Comparison between groups is thought to reflect differences in effect of the interventions. The required sample size in RCT’s depends partly on the variation between participants: a large variation calls for a larger sample size. Therefore, many studies include homogeneous groups. However, due to the variety of their co-morbidities, it is difficult to compose a homogeneous study population with sufficient power composed of persons with SPIMD. Neither is it possible to compose a large heterogeneous SPIMD study population. As a consequence, both the experimental and control group will consist of participants with much variation in their co-morbidities, resulting in a wide array of responses to the same intervention. In such circumstances detecting significant effects of an intervention is rather difficult. An anecdote from practical experience will illustrate this.

To subject three participants with SPIMD to passive or assistive active movements, powered exercise equipment (Shapemaster®, Barth Fidder, Shapemaster Benelux) was used. These machines are fitted with motors and gearboxes, and controlled by microchip technology. The machines automatically move selected levers and handles at pre-determined speeds through a pre-determined range of motion. Each machine provides multi-function movements. The outcome measures are body composition, muscle tone, heart rate, oxygen saturation and alertness [39]. At the individual level, relevant improvements were found for the different outcome measures.

1. A woman of 38 years old, with profound ID, GMFCS level IV, no spasticity, totally blind, epilepsy, and being overweight, participated in the study. Her BMI before the intervention was 27.7 kg/cm², after the intervention 26.2 kg/cm², which means a difference of 1.5 kg/cm². Her waist circumference decreases from 89 cm, which means abdominal obesitas, to 83 cm, which is indicating ‘healthy weight but attention needed’ [7]. Oxygen saturation during and after moving on the machines increased from 95% before the intervention program, to 99 % after 20 weeks. However, muscle tone, alertness and hart frequency showed no differences.

2. A girl of 17 years old, with profound ID, GMFCS level V, severe partially sighted, with spasticity, epilepsy and orthopedic defects also participated in the study. Her muscle tone in the legs decreased in 20 weeks with two points on the six point scale of the Modified Ashworth Scale. After every increase in intensity in a five weeks period, her heart rate increased first one or two heart rate zones during moving, but after three weeks, heart rate decreased again to the first level. This might indicate a training effect. However, saturation, alertness, BMI and waist circumference showed no differences.

3. A man of 43 years old, with profound ID, GMFCS level V, totally blind, spasticity, epilepsy, and orthopedic defects, also participated in the study. His muscle tone in both arms and legs decreased in 20 weeks with one point on the six point scale of the Modified Ashworth Scale. Oxygen saturation during moving on the machines increased from 91% before the intervention program, to 95 % after 20 weeks. BMI decreased after the intervention with 0.5 kg/cm², but before and after the intervention he already had a healthy BMI. Alertness increased during intervention with one point on a four point scale [39]. However, heart rate showed no differences during and after the intervention period.

As shown, individuals benefit from the intervention but not in the same way nor to the same extent. Individual differences in characteristics of locomotor skills, visual impairment, co-morbidities, and baseline measurements of the outcome measures account for this result. Yet, there were benefits, albeit different ones for different subjects. In group comparison studies the relevant individual benefits can be overlooked. Consequently, next to traditional research designs,
alternative research designs such as multiple case studies or program evaluation and adherent statistical analysis [40, 41] should be used for intervention studies aimed at participants with SPIMD.

**Implications**

In the introduction we presented an integration of models and concepts describing quality of life, participation, physical well-being, physical fitness, physical activity and health, so as to illustrate their mutual relatedness (figure 1).

**Figure 1.** Integration of models and concepts of participation, quality of life, physical well-being, physical activity, physical fitness, and health [3, 35, 42, 43, 44].

As we put forward in the introduction, out of the concepts introduced in this model, this thesis mainly focused on the measurement of physical activity and fitness, operationalized by body composition, functional exercise and aerobic capacity, balance, muscle flexibility and heart rate patterns.

Now that physical fitness and physical activity can be evaluated in persons with SPIMD, the next step to take in future research is to look at the relation of physical activity and fitness with the concepts on the right hand side of the figure, including health, physical well-being, and quality of life. Also, further research should elaborate on the relation between physical fitness and the concepts in the left hand side of the figure, including participation. This however can be a hard nut to crack, as evidence on the concept of participation is lacking. The direct relation between physical activity / fitness and participation in persons with SPIMD has so far been unknown as the level of participation is difficult to define and quantify for this specific group [45]. The question is whether persons with SPIMD are really able to participate in daily activities. Can they take part in or have influence on situations and contexts important to them personally? A complicating factor in this respect is the fact that these persons are often not able to verbalize what is important to them and, what's more, to verbalize what to them ‘taking part’ or ‘having influence’ actually means. It is often observed that a person merely sitting in a wheelchair placed in an activity room with others, is considered to ‘take part’ in that activity or society [45]. Or does “participation”
of these individuals require more? It is imperative and essential to investigate to which extent a person with SPIMD really participates in living habits, ‘work’, leisure activities, sports etc. However, so far knowledge on this topic is not available neither are instruments aimed at investigating the concept of participation in persons with SPIMD.

**Recommendations**

The vast majority of recommendations for future research have been mentioned previously—either under the studied concepts or in general findings. These however all amount to a call for tailored interventions. Interventions aiming at promoting physical fitness are generally carried out with participants suffering from mild or moderate ID. Performing similar interventions with participants classified as having severe or profound ID is thought to be more difficult, firstly because of the assumption that most of these persons are not able to walk, and secondly because these persons have more problems understanding the tasks required of them. We recommend developing, performing and evaluating tailored interventions geared to promote physical activity and fitness in persons with severe or profound ID who may or may not be able to walk. As environmental cues facilitate performance in persons with SPIMD, examining environmental cues provided by technological adaptations, like auditive or other pleasant sensory stimuli can be of significant help.

As knowledge on the concept of participation of individuals with SPIMD is lacking, further research should aim on feasible instruments to identify and quantify outcome measures of participation. Furthermore, research aimed on exploring the relations between physical fitness and physical activity on one side, and participation on the other, is recommended.

Previous paragraphs have discussed the shortcomings, questions and suggestions that came up during the various studies comprised by this thesis. This thesis functions as a basic first step to enhance and strengthen the role of physical activity and fitness for this target group, by enabling sound academic testing of physical fitness. We strongly advocate to consider the major role physical activity and fitness can play in well-being.
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