General discussion

MAIN FINDINGS

In this thesis, we presented the results of a comprehensive study of aspects of physical fitness in children and adolescents, aged 2-18 years, with moderate to severe levels of intellectual disabilities (ID). Although this is a relatively small and difficult to assess population, thanks to a close and positive collaboration with parents as well as the management and professionals of seven specialized children's day program centers, we were able to include and test a relatively large study group of 130 Dutch children and adolescents aged 2 to 18 years. Based on a systematic review of the literature and a pilot study, we selected a battery of five tests that appeared feasible and reliable for this specific group of children and adolescents.

Our next step was to measure physical fitness in a large sample of these children and adolescents. The results demonstrated that overweight or obesity was present in 34-40% of the children, and in most children - varying per test from 71 to 91% -, scores on muscular strength, muscular endurance and cardiorespiratory fitness were lower than the 5 or 10 percent of reference values of typically developing (TD) children.

The fitness outcomes were significantly associated with the children's motor development and physical activity. More than half of them (53%) were not active enough to meet the Dutch physical activity guideline of daily 60 minutes of moderate to vigorous intensity. As was to be expected, children with low motor development had significantly lower physical activity levels than those with more developed motor skills.

Separately, in collaboration with the research group of Dr. Fernhall of the University of Illinois at Chicago, we retrospectively studied the highest achieved heart rates obtained during a maximal exercise test (HR_{peak}) of 210 children with and without ID (8-17 years) in an existing dataset. We investigated what the best equation would be to predict HR_{peak}. Formerly, this had not been investigated in a sample of only children and adolescents with ID. HR_{peak} turned out to be not related to age in this group, and was best predicted by just the average HR_{peak} of the group. For youth with ID caused by Down syndrome (DS), the average HR_{peak} was 172 beats·min^{-1}; for youth with other causes of ID it was 188 beats·min^{-1}. High measurement error remained; therefore, caution is warranted using this method to predict individual HR_{peak}.

After our methodological reflections, consequences of all findings will be discussed below.

METHODOLOGICAL CONSIDERATIONS

Bias

We have recruited our participants through one large care provider in the Netherlands, Reinaerde. This may have led to a selection bias regarding the population studied. Even though we included participants from seven different specialized day program centers, all participants did receive care and support from the professionals of Reinaerde. At Reinaerde

a lifestyle program called 'Reinaerde Fit' was being implemented during the execution of the current study, which could have led to higher awareness of the importance of physical activity, which could have positively influenced the results.

Children with more severe ID and/or low motor development had difficulties performing the muscular strength and muscular endurance tests. The tests' cognitive and motor demands were not matching some of the participants' possibilities. Due to this selective drop-out, children with more severe ID and/or low motor development were underrepresented in the results. The results are therefore likely to be an overestimation of what we expect for children and adolescents with more severe ID and very low motor development.

Testing issues

For this study, we selected field-based physical fitness tests. Lab-based tests provide generally more objective and accurate data, but are less feasible for the population of interest here. Children and adolescents with moderate to severe ID need functional tests that can be conducted in their own familiar environment. The advantage of field-based tests is that the results are easier to generalize to the daily living situation. However, due to the functionality of the field-based tests, we presumably tested motor skills as well. This overlap most likely occurred in the muscular strength and muscular endurance tests, since in these tests a notable selective drop-out occurred. Despite these limitations, we argue -based on our experience- that these tests are the most appropriate tests for this population.

The validity of the used fitness tests had not been determined in children and adolescents with moderate to severe ID. Previous studies did indicate sufficient validity in youth with mild to moderate ID [1-4], and we discussed the face validity of the tests with physical therapists before the start of the pilot study. Based on this information, we considered the validity of the tests to be sufficient. However, this should ideally be confirmed by a validity study. The assessment of criterion validity will be difficult, because gold standard tests are often complicated lab-based tests. Construct validity could be a feasible alternative.

The test instructors were physical therapists with longstanding working experience in this population. They received training on how to conduct the tests and were instructed to motivate the participants to maximize their performance. Because of the inevitable heterogeneity of the participants, standardization of motivation in this population is not feasible. Therefore, motivation style was dependent on the preferences of the participant. The test instructors also had to estimate whether the participant performed the tests according to protocol and at their maximal effort. We diminished subjectivity of this evaluation by having two test instructors conducting the tests.

From practical experience we know that alertness, fatigue and other circumstances may influence test outcomes in children with ID considerably. In the pilot study, this variation could have caused the moderately reliable outcomes for some tests, which were sufficient to use the tests at group-level, but less suited for use at the individual level due to high

measurement errors. Further research into field-based physical fitness tests for reliable use at the individual level in this population is needed.

To interpret the physical fitness outcomes, we used norm-referenced cut-off scores for TD children which were available for ages up to 10 or 11 year. Comparing the outcomes of our older participants was therefore difficult. Conservatively, we compared these outcomes with the reference values of the nearest age group available, which meant comparison with a - sometimes much - younger age group. This has most likely resulted in an underestimation of the percentage of adolescents that score under the lowest percentile (P5 or P10).

Even though accelerometers are the preferred option to measure physical activity [5, 6] Reilly et al., 2008), it comes with limitations. They cannot record water based activities , and the influences of data processing decisions can be substantial [7]. We provided detailed description of the data processing methods to address this issue and to enable replication of our results.

The gross motor scale of the Bayley Scale of Infant Development, third edition (BSID-III) was used to study gross motor development. This test was developed for TD children up to 42 months; no validation studies were conducted for children or adolescents with ID. However, the BSID is often used by physical therapists in clinical practice to evaluate the gross motor development in children and adolescents with ID. Other motor development tests were validated in children with mild ID but assume high motor skills, like the Test of Gross Motor Development (TGMD)[8, 9] or the Bruininks-Oseretsky Test (BOT, [10, 11]. These tests would have likely provided very low scores in the majority population and thereby a bottom effect. Using the gross motor scale of the BSID-III, the outcomes were acceptably distributed (score range 42 to 72) with only one participant reaching the maximum score (data not shown).

CLINICAL IMPLICATIONS

Testing physical fitness

One of the reasons to conduct this study was the call of physical therapists for physical fitness tests for children and adolescents with moderate to severe ID. The tests we selected based on our pilot study demonstrated to be feasible and reliable, however not all tests were reliable enough for use at the individual level. Body composition measures and the stair climbing test were, but -according to the ICC values- the overarm throwing, and modified 6-minute walk test (6MWT) were not. Since we are not aware of feasible and reliable alternatives, we would advise professionals to use these tests to evaluate the physical fitness of a child or adolescent with moderate to severe ID, but to take the large minimal detectable change (MDC) into account. This means that a score needs to change more than the MDC, before one can say it is an actual improvement or decline, instead of a random effect caused by the measurement error.

We found a more appropriate way to predict the HR_{peak} in children with ID than the existing prediction equations, but large measurement errors remained. Estimating the HR_{peak} by using the average value for the group of children and adolescents with ID caused by DS (172 beats·min^{-1}), and another value for the children and adolescents with ID but without DS (188 beats·min^{-1}) will lead to overestimation or underestimation in a part of the population. Therefore, these values can only be used to indicate a range of the aimed training intensity, not a specific value. Furthermore, it is important to monitor the children's physical signs of exertion, like sweating, gasping and blushing, to prevent overtraining or undertraining.

Increasing physical fitness

This thesis emphasizes the need for increasing the physical fitness of children and adolescents with moderate to severe ID, in order to support and maximize their health and participation (Figure 1). As physical activity and motor development appear to be associated with several fitness components, it stands to reason that increasing physical activity and improving motor development first are potential effective strategies as a basis for increasing physical fitness in this specific population. In order to do that, caregivers and therapists need to take the following specific considerations into account.

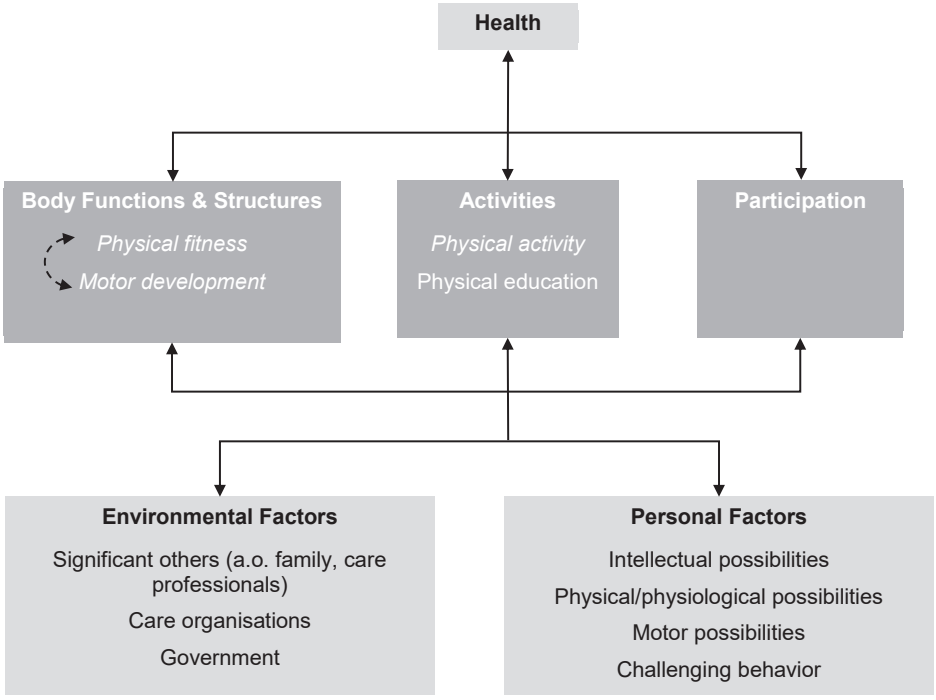

Figure 1. The relationship between the main topics in the discussion displayed in the ICF framework.

Support from significant others.

Children with moderate to severe ID are highly dependent on their social and professional environment, like parents, professional care givers, physical therapists, psychologists, and physicians, to become and stay physically active [12, 13]. Support from these significant others is needed, since personal characteristics of the children and adolescents, like cognitive, physical and behavioral challenges, can be a barrier to become physically active [12].

Enough time and opportunities

Children and adolescents with ID need more time and opportunities to learn new skills than TD peers [14]. Positive experiences of physical activities facilitate being active during childhood and later on in life [12]. In the Dutch legal framework for primary schools, it is compulsory to include physical education in the educational program, aimed at learning the basic forms of exercise and increasing social skills [15]. In addition, during physical education, other domains, like social interaction can be addressed as well.

Specific expertise

Besides the proper amount of attention and time that must be allocated in the day programs, specific expertise of professionals is essential to support the development of physical fitness, physical activity and motor skills [15]. Since in general, children and adolescents with ID have a delay in motor development, more musculoskeletal problems and other health conditions [16], the urge for specific expertise is even higher. Professional care givers, with a general pedagogical or medical training and background, do not necessarily have the specific knowledge and expertise to support the development of for example motor skills in this complicated and vulnerable population. Professionals, like physical activity experts, are needed to offer suitable adapted physical education or activities to these children and adolescents. These physical activity experts should collaborate with other professionals at the day program centers, like care givers and physical therapists.

Opportunities during activities of daily living

The urge for experts does not mean that persons without a physical education background cannot support these children in their physical activities. Physical activity throughout the day is very important. Care professionals and family members are the designated persons to make sure the children and adolescents with ID are not only active during predetermined physical activities, but can also expand their physical activities and possibilities in daily living, like walking stairs, dress independently or playing outside. The right information and education can help these significant others to turn perceived barriers into facilitators [17].

Address the full potential

Sometimes, it seems that overweight, low strength and endurance, low levels of physical activity and low motor skills are more easily taken for granted by the parents and professional caregivers because of the existing intellectual, physical and/or behavioral constraints. However, physical fitness and motor skills are conditional to perform activities of daily living, like carrying a bag, or participate in games with peers. An important question that surfaces is: At which level is the bar set? Is it sufficient if a child can walk independently for 5 minutes? Are 'we' satisfied if he/she can perform all fundamental motor skills? Of course, it is all depending on the physical possibilities of the individual, and the capacity to develop. But it is the responsibility of the social and professional environment to help the child or adolescent reach his/her full potential, including working on physical fitness and motor skills.

National guidelines for physical education

In the Netherlands, the guideline for amount of physical education per week at primary schools is 2 times 45 minutes, organized and supervised by professionals with specific expertise [15]. For secondary schools, the government does not provide guidelines for the amount of physical education, but it still is compulsory to offer physical activity and sports orientation [18]. Since day program centers are not considered part of the education system, these national guidelines do not formally apply. Because of this, the management of the day program center or care organization are to decide which developmental areas are prioritized, and which expertise is engaged.

This results in big differences among day program centers for children and adolescents with ID. This became visible in our survey among a sample of 20 of these day program centers in the Netherlands [19]. None of the consulted centers organized physical education according to the Dutch guidelines for primary schools (2 times 45 minutes, guided by an expert). Weekly physical education was organized at 15-day program centers, often supervised by physical education experts (n=12). At the other centers, physical education was supervised by trainees (n=2) or the professional caregivers (n=1). Most of the centers (n=13) organized physical education lessons once a week ranging from 30 to 90 minutes. All respondents indicated that physical activity was considered important and was initiated and organized by professional caregivers, in forms of outdoor or indoor games and being physically active during activities of daily living. But the frequency, intensity, and quality of the activities was diverse.

A formal framework or at least nationwide guidelines for physical education at specialized day program centers would help to create uniformity and equality in the amount, quality and focus of physical education for this population that needs support in developing motor skills, physical fitness and become and remain physically active. The framework or guidelines for children and adolescents with ID should at least be comparable to that of their TD peers: 2 times a week 45 min, organized and guided by a professional in physical education, aimed

at learning the basic forms of exercise and work on goals to support the development of fitness, activity and motor skills.

DIRECTIONS FOR FUTURE RESEARCH

Further research is necessary to expand the body of knowledge on how to measure physical fitness in this population. Especially for children with severe ID, suitable tests for muscular strength and muscular endurance are needed for specific muscle groups. For cardiorespiratory fitness, a field-based test which demands maximal performance would be more accurate, instead of a submaximal performance. Information is missing on the validity of most of the physical fitness tests in this specific group of children and adolescents. Furthermore, the feasibility of a maximal lab-based performance tests in children and adolescents with more severe levels of ID has never been studied. This would give us useful information on physiological response on vigorous exercise, and might help us in finding more suitable ways to predict de HR_{peak} in this population.

Besides the above-mentioned knowledge gaps in measuring physical fitness in this population, future research also needs to address the issues in measuring physical activity and motor development. This includes the cross-validation of the intensity cut-points of accelerometers specific for children and adolescents with ID, and existing motor tests should be validated for older children with more severe levels of ID.

Research should also focus on ways to improve physical fitness in this challenging population. Previous research has shown that increasing physical fitness levels in children and adolescents with ID is possible [20, 21]. Several interventions have been evaluated, such as aerobic exercise programs, weight training programs or circuit training programs. These studies were mainly conducted in children and adolescents with mild to moderate ID, and the methodological quality of the studies was mostly low, due to lack of control groups or no concealed group allocation, and small sample sizes. Therefore, selection of the most effective interventions for children and adolescents with moderate to severe ID is difficult. Further research needs to evaluate intervention studies with good methodological quality, in which not only the short-term intervention effects are studied, but also the long-term effects. The association we found between physical fitness, physical activity and motor development provides a starting point for such interventions. Future research should indicate which of these components is most effective to increase physical fitness. It would be interesting to study the effect of interventions focusing on motor development alone, on physical activity alone, and on both combined. The effects should be studied in subgroups. For example, young children might benefit more from interventions focused on motor development than older children or adolescents.

Furthermore, it is necessary to set up a longitudinal study on the development of physical fitness, activity and motor development, in relation to health and participation in this vulnerable population. This will provide insights in the importance of personal and environmental factors for the development of physical fitness, physical activity and motor skills in this specific group of children. A longitudinal study would also provide an opportunity to study the timeframe in which gross motor skills can be developed. In TD children, preschool years are a critical period to develop functional motor skills [22]. A longitudinal study in 7-11 year old children with mild ID has shown that locomotor skills did not improve over a 3 year period, while ball skills did improve with a plateau around 10 years [23]. Unfortunately, nothing is known on the amount of physical activity or physical education during these years. Conducting such a longitudinal study, requires a large study sample. For this, care organizations in the Netherlands should collaborate. This will make it more feasible to include participants and makes the results more generalizable to the Dutch population of children and adolescents with moderate or severe ID. Ideally, measuring physical fitness, motor development and physical activity becomes a part of the regular care. For this, we first need to reach consensus among professionals on which tests to use.

CONCLUSION

This thesis emphasizes the need to increase the physical fitness of children and adolescents with moderate to severe ID. Even though we still have unanswered questions with regards to measurement and interventions, significant others, like parents, caregivers and other professionals need to start now with the available practice-based and evidenced-based knowledge. These youngsters need our support now to become and remain fit for the future.

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