General discussion
The aim of this thesis was to develop and evaluate the effectiveness and feasibility of a multidisciplinary and multicomponent intervention We12BFit!. We12BFit! aims to improve physical fitness (PF) and motivation for physical activity (PA) in children with developmental coordination disorder (DCD). The steps of treatment theory as defined by Whyte et al. were used to guide the development and ensured a sound basis in theory and scientific evidence. This general discussion will summarise the individual chapters and discuss the methods and implications of this thesis.

1. Main findings

First, in preparation for the development of We12BFit! we performed a systematic review to gain insight into the characteristics of PA interventions effective in improving cardiopulmonary fitness (CRF) (Chapter 2). As 13 out of 23 studies were effective, we showed that not every training effort automatically leads to success. Interventions with medium to high effect sizes focused on improving PF than PA behaviour more often, had slightly higher frequencies of activities and had a shorter duration than less effective interventions. Moreover, interventions with larger effect sizes appeared to be more controlled, as they usually included protocolled training sessions and relied on smaller sample sizes than interventions with lower effect sizes. Defining and applying pre-set relative training intensity, as well as monitoring and adjusting relative training intensity, seem to be important aspects of successful interventions. Based on this systematic review, a duration of at least six weeks and a frequency of three to four times a week are recommended to successfully improve CRF.

Second, by integrating the available knowledge from the literature and expertise from rehabilitation professionals we made specific choices with regard to the targets, essential ingredients, parameters and participant characteristics of We12BFit! (Chapter 3 and 4). We12BFit! consists of two intertwined parts: We12BFit!-PF which targets CRF, muscle strength and anaerobic power, and We12BFit!-Lifestyle PA which targets motivation for PA. We12BFit!-PF is a 10 week group training with two 60 minute training sessions a week. In week 6 of We12BFit!-PF, We12BFit!-Lifestyle PA is introduced. We12BFit!-Lifestyle PA starts with a parent meeting, the provision of pedometers and an information booklet for parents, followed by individual coaching of the child and his parents. We12BFit! is designed to suit the needs and abilities of 7 to 12 year old children with DCD. Specific adjustments to the target group included defining the intensity relative to each individual child’s starting level, selecting activities that they were expected to be able to perform, allowing for time to get used to the training regimen, providing the training in small groups and allowing for individual questions during the coaching sessions.

Third, we showed that children with DCD improved their CRF, anaerobic power and motivation for PA (Chapter 5). As no control group was included in this study, it could
It cannot be ascertained whether the effects can be solely attributed to We12BFit!. However, it seems unlikely that the improvements can be attributed to spontaneous development during the study period, as previous research showed that CRF and anaerobic power of both children with motor coordination problems and typically developing (TD) children declined over time.\textsuperscript{10,29,30} Indirectly, the children’s social emotional well-being, enjoyment of activities, motor skills and weight also improved after We12BFit!. The group aspect of We12BFit!-PF and the high intensity combined with the positive motivational climate of We12BFit!-PF may have improved children's self-efficacy and reinforced their motivation for PA. Not just the children benefitted from We12BFit! as parents reported that they themselves also increased their activities, extended their strategies to motivate their child to be active and improved their knowledge on PA and PF. We12BFit! had high attendance rates and was shown to be feasible. Key aspects identified for improving the effectiveness and feasibility of We12BFit! were: improving strength exercises to involve the right muscle groups and to be performed in a group, adding variety to the training sessions to make the exercises more enjoyable and establishing a connection face-to-face between the coach, the child and his parents before conversing over video or phone call for the coaching sessions.

Fourth, triggered by the indirect effects of We12BFit! on participation in PA, we aimed to extend our insight on the impact of DCD on participation and to find out in what recreational and leisure activities the participation involvement and attendance of children with DCD possibly differed from TD children (Chapter 6). Findings on participation involvement, operationalised as enjoyment, were inconclusive. Participation attendance was operationalised as diversity, frequency, duration and intensity of participation. Diversity and frequency were lower for a number of domains in children with DCD compared with TD children, this was mostly the case for physical, skill-based, self-improvement and play activity subdomains. In addition, children with DCD spent less time in activities of moderate to vigorous intensity, these activities are important for improving and maintaining PF. For some measures, diversity and frequency seemed to decrease with age in children with DCD, whereas enjoyment seemed to increase. Possibly, with increasing age children with DCD only participate in the activities they enjoy, thereby limiting their participation diversity and frequency. To allow children with DCD to select enjoyable physical activities as they become older, they should be offered a variety of physical activities at a young age.

To further elucidate the results, the field is in need of a clear operationalisation of the definition of participation enabling comparison of participation measurements. Attention should be given to the nature of participation that goes beyond the subjective experience and the objective being there: what is the role of the child during participation, is the child part of the interactions or is he a bystander? Moreover, as children with DCD form a heterogeneous group with regard to their motor difficulties and comorbidities, participation involvement and attendance may also differ between children with DCD. To inform future intervention development we need to gain insight in the characteristics of children with DCD that do and do not participate less in activities than TD children.
2 Strengths and limitations of methods

2.1 Treatment theory

An important strength of this thesis is the use of treatment theory for synthesizing the literature on PA interventions (Chapter 2) and the development and evaluation of We12B-Fit! (Chapter 3-5). Treatment theory guides the development of an intervention. First, treatment theory prescribes that targets, not aims, should be selected for intervention. As targets can only be directly improved, We12B-Fit! focused on specific components of PF and motivation for PA instead of the broader, more indirect aims such as PA, participation and lifelong PF. This focus on direct targets guides the process of co-creation between different professions, helps to restrict the number of targets and to subsequently arrange the appropriate ingredients.

Second, looking into the mechanism of action of the intervention makes developers aware of all relevant aspects of their treatment ingredients. In the case of We12B-Fit!-PF, this led to a strong focus on frequency, intensity, time, type of activities and continuous monitoring and adjustment of intensity. Despite the importance of intensity, monitoring and subsequent adjustment of intensity throughout the training sessions as applied during We12B-Fit!, is rarely performed in other interventions (Chapter 2). For We12B-Fit!-Lifestyle PA, looking into the mechanism of action led to a behavioural approach using the trans-theoretical model of change (TTM). A strength following from the use of the TTM, is that it allowed us to flexibly tailor the intervention to the stage of change of the child and his parents, which increases the chance of success. In selecting behavioural change techniques, we followed the taxonomy of Michie et al. This taxonomy allows for comparison of behaviour change techniques in the literature and enabled us to select the most effective techniques. A researcher in psychology once argued that using someone else’s theory seems to be seen as using someone else’s toothbrush. Considering the wide range of interventions focusing on PF and PA available in the literature the same seems to be going on in our field. However, the focus within treatment theory on the mechanism of action and selecting evidence-based ingredients encourages researchers to build on the available knowledge instead of repeatedly reinventing the wheel. In this thesis this was reflected in the fact that we did not just select single strategies from effective interventions but also larger elements from effective interventions such as the high intensity interval training protocol and the information booklet for parents.

Third, treatment theory carefully distinguishes between ingredients that are essential to bring about a change in the target and other ingredients that may mediate the effect, for instance by improving adherence. Reporting ingredients accordingly improves the comparability of studies and has the potential of increasing our collective insight on the effectiveness of different ingredients.
2.2 Co-creation

As coined by Aristotle, the whole is greater than the sum of its parts. This is exactly what co-creation aims for. Co-creation is “a process, in which teams of diverse stakeholders are actively engaged in a mutually empowering act of collective creativity with experiential and practical outcomes.” Choices made within the steps of treatment theory were informed by both the literature and advice from professionals (Chapter 3 and 4). These two perspectives were largely complementary but also challenged us to come up with solutions that had not been explored if the perspectives had not been combined. On the one hand, the literature provided information about the background of children with DCD and the requirements for an effective intervention. Complementarily, professionals contributed with their experience with the target group, practical considerations and important values for intervention such as the importance of a client-oriented approach.

However, as noted, where two perspectives meet and new options for intervention are explored, information is not solely complementary and may lead to some discordance which needs resolving. As internationally promoted, practitioners generally embrace a client- and task-oriented approach in their work. This entails that they prefer a highly individually tailored, integrated approach, simultaneously addressing different functional problems that their patients encounter at the moment of intervention (Chapter 3). This includes both goals classified as targets and the more indirect aims such as participation. On the other hand, science and approaches for intervention development such as treatment theory, advocate a focused, evidence-based intervention which is applied in a transparent and standardised way which can be evaluated properly. A narrow focus of intervention is especially relevant for newly developed interventions, as it allows for evaluation of the effectiveness of specific intervention ingredients.

An example of a discordance encountered during the development of We12BFit! concerns the desire of practitioners to include individual motor coordination as a target in We12BFit!-PF. This makes sense in the light of working in an integrative and task-oriented way. Improving children’s motor skills might for example indirectly improve their PA and consequently their PF and participation. However, this individual focus seems to be difficult to reconcile with the concept of a group training, chosen for its anticipated beneficial motivational effect and for the sake of minimising costs. Consequently, the question arose to what level a group intervention could be tailored to each individual patient. We opted to include children with similar requests for help regarding PF and PA, and basing exercises on the starting level of each individual child. Combining tailoring and standardisation, each child would perform the exercises at their own level but the intensity and difficulty would be increased following the same decision rules and steps in all children. Moreover, adding coordinative exercises at this stage of intervention development may jeopardise meeting the required parameters for improving PF: children may get fatigued and no longer meet the required intensity or spend too little time on PF exercises. This was resolved by
including children who had already completed individual physical therapy to remediate their motor difficulties. In addition, exercises were selected to be functionally relevant but not too challenging and cognitive motor dual tasks were avoided.

Another example of resolving differences between the literature and practical considerations, concerns choices regarding the duration and frequency of training. In chapter 2, a duration of at least six weeks and a frequency of three to four times a week was recommended for improving CRF. However, professionals anticipated that training three to four times a week would not be feasible. Therefore, we based the training of CRF on a protocol with a lower frequency, which had been proven effective despite its lower frequency.\(^7\) Where the frequency of this protocol was lower than recommended in chapter 2, the intensity was high and the duration of the intervention was longer than the lower limit of six weeks. In addition, tailoring the intervention to children with DCD we extended the duration of the original protocol of seven weeks to ten weeks.

Is the sum complete, did we include all parts? To maximise the benefits of co-creation, at least one other very important stakeholder should be included: the target population.\(^4,11\) During the initial intervention development (Chapter 3 and 4), two parents of a child with DCD were interviewed on their preferences for the intervention and two children with DCD pilot tested the training. During the evaluation parents provided their opinions and suggestions however the children’s input for the evaluation was merely indirect as they were obtained through trainer, coach, student and parent observations and reports.

### 2.3 Methods for evaluation

In this thesis we relied on a single-arm design using mixed-methods to evaluate both the effectiveness and feasibility of We12BFit!. The single-arm design allowed for detecting changes over time within participants. Although we were able to show that the improvements in PF and motivation for PA were of value as longitudinal studies showed that in general PF and PA of children with DCD declined over time, the inclusion of a randomised control group would facilitate the interpretability of the results. To guarantee sufficient power to detect significant changes in the primary outcome measure on CRF at least 19 participants were needed. This requirement was met by the inclusion of 20 participants, of which 19 provided sufficient data to assess CRF. Other measures, in particular the accelerometer based measures, were underpowered to reveal potential significant differences over time.

Whereas We12BFit! was tested in an intervention group only and a relatively small group of participants, we sought to maximise the amount and richness of the information we collected by using mixed-methods. Mixed-methods, combining both quantitative and qualitative methods to retrieve information on effectiveness and feasibility, provided us with a comprehensive image of what did work, what did not work and why. Within this mixed-methods design, the quantitative measures for measuring PF included maximal fitness testing. It should be noted that in children with DCD, the performance on these
tests is at least partly influenced by their motor coordination problems and lower perceived adequacy.\textsuperscript{12,13} Therefore, the improvements in CRF and anaerobic power may be partly due to improvements in motor coordination and perceived adequacy. To limit this confounding effect we selected tests that required minimal coordination, especially in the case of the health-related PF components CRF and muscle strength. In addition, during the 20 meter Shuttle Run Test (to measure CRF) we made an effort to overcome problems with pacing and motivation by having a therapist accompany the children throughout the test. Although we showed that CRF and anaerobic power were maintained until 12 weeks after We\textsuperscript{12}BFit!-PF, all measurements pertained the period of intervention. As laid out in the TTM it may take at least six months to integrate new behaviours into ones routines and to establish whether the maintenance stage of behaviour change has been reached. Unfortunately the current design did not allow for establishing the long term effects of We\textsuperscript{12}BFit! on PF and motivation for PA.

3 Implications of this thesis

3.1 Implications for research

Intervention development is an iterative process. The intervention development, execution and evaluation described in this thesis actually form the onset for further development. The methods for further development and consequent evaluation should be improved in a number of ways.

First, the target group, along with other stakeholders, should have a more pronounced role in the process of co-creation. So far, the role of the target group was rather modest. Directly involving parents and children in determining targets and ingredients may empower them, increase the relevance of the intervention and improve their adherence to the intervention.\textsuperscript{11,14}

Second, implementing an intervention is an intervention in itself. The process from developing an intervention to actually implementing it, should be planned for carefully. This goes for relatively simple but even more for complex interventions like We\textsuperscript{12}BFit! with its interacting components provided by a multidisciplinary team. Co-creation may be just the first of many strategies needed to implement an intervention. A stepwise approach such as intervention mapping\textsuperscript{15} or the behaviour change wheel\textsuperscript{16} may be helpful in identifying the desired (behaviour) change, the relevant processes from diagnosing to referring and follow-up that need to be addressed, the determinants of behaviour and corresponding strategies, and modes of delivery. Determinants of behaviour of rehabilitation professionals for example may concern beliefs regarding the abilities of children with DCD, attitude towards having children train at a high intensity, skills to manage groups of children and time to prepare sessions. Corresponding strategies may include enhanced or repeated instruction, education, intervisio and facilitation for the professionals involved in the treatment.
Third, the methods for evaluation should be improved by adding a control group if possible, adding follow up measures on PA and possibly PF at least 6 months after completion of the intervention, and extending the size of the sample. Additional measurement instruments might be included to quantify the stage of change regarding PA and to monitor the potential mediating effect of self-efficacy. Moreover, when aiming to additionally quantify the effects of We12BFit! on participation, instruments for this may need to be validated in children with DCD (Chapter 6). When evaluating the effects in a larger sample it would be recommended to compare the characteristics of children that do and children that do not benefit from the intervention.

In addition to extending the methods for further development of We12BFit! a number of improvements should be made regarding the content of We12BFit! (Chapter 5). Important improvements for We12BFit!-PF concern the strength exercises and the enjoyability of the exercises. Strength exercises need to be more specific to the targeted muscle groups and should be adjusted to be trained in groups. Although trainers were provided with suggestions to vary the high intensity training by adding game elements, this did not seem to suit their need and in between training sessions they had no time to prepare the games. To improve this the suggestions should be reconsidered, preferably in co-creation between the trainers and children, and the improved suggestions should be explicitly discussed during the instruction of the trainers. In addition, the trainers requested advice for enhancing children’s motivation during the training sessions. Effective strategies to do this should be extracted from the literature and included in the training/instruction of the trainers. This could be complemented by having the coach observe some of the training sessions and provide advice on this, during intervision the trainers might also help each other with this. Involving the coach in the training sessions may in turn help the coach to establish a connection with the participants face-to-face before conversing over video or phone call. Suggestions for improvement also concerned specific adjustments to the setting of implementation and elements that were received well and should be built upon further such as increasing the opportunity for interaction between parents and the interaction between parents and coaches or trainers, extending the booklet to include information for children as well and to integrate strategies to improve children’s social emotional skills. These suggestions should be carefully considered for the further development of We12BFit!.

Finally, although We12BFit! is specifically tailored to children with DCD it may be worthwhile to investigate whether We12BFit! is also beneficial for children with obesity, Cerebral Palsy (Gross Motor Function Classification Scale I), neurofibromatosis or delayed motor development. These children may experience motor problems in a similar range of severity as children with DCD and hence encounter similar problems with PF and PA. In addition to having more children benefit the intervention this would also allow for making We12BFit! more accessible; We12BFit! could be offered more often and at more locations than when provided solely for children with DCD.
3.2 Implications for practice and society

3.2.1 Moving does matter

This thesis underlines the value of comprehensive treatment for children with DCD. First, mapping the problems of children with DCD according to the International Classification of Functioning and Disability Children and Youth (ICF-CY) stresses the need for intervention: the problems that children with DCD experience do not just affect their motor functioning but have a substantial impact on their PF, PA and participation. These secondary problems play a crucial role in functioning, development and health in the short term and, easily overlooked but at least equally important, in the long term.

Second, We12BFit! shows the value of intervention. Intervening pays off: children with DCD who have already received physical therapy to remediate their motor problems, improved their CRF, anaerobic power and motivation for PA, and indirectly seemed to improve their motor coordination, self-efficacy and participation. Therefore, rather than trying to indirectly remediate all problems that children with DCD encounter by solely focusing on motor coordination, a direct approach targeting PF and motivation for PA should be added to the rehabilitation of children with DCD. This direct approach increases the chances of successfully improving PF and motivation for PA. Moreover, it is strongly recommended to combine targeting PF and PA, as high intensity group training in a positive motivational climate may improve self-efficacy and may consequently enhance the effect of behavioural intervention on motivation for PA. When stimulating PF and PA, trainers and coaches should focus on facilitating the experience of enactive mastery (i.e. progression and success), vicarious experience by providing the opportunity for children with DCD to see other children with similar problems putting in sustained effort and succeed, and providing verbal persuasion to let the child explore and push his physical boundaries. In addition, parents of children with DCD need to be given an active role in promoting their child’s PF and PA. Like the children, parents may benefit from experiencing success (in themselves or in their child), vicarious experience by seeing the abilities and progression of other children with DCD and their parents, and experiencing what it is like to try to become more physically fit and active themselves. Parents need to be provided with information about being active and DCD, both in writing and in person, and parents may benefit from the contact with other parents of children with DCD as well.

3.2.2 Matters to be moved

Ultimately We12BFit! would be no longer needed. Fortunately, there are a number of things that can be put in place to at least reduce the need for interventions like We12BFit!. First, early identification of children with DCD might prevent children with DCD from becoming inactive and deconditioned. The low PF and PA of children with DCD is assumed to be part of a negative spiral where poor motor skills lead to reduced participation in physical activity and consequently lower PF. When children with DCD are diagnosed early, treatment for motor coordination may start earlier and appropriate guidance can be
provided to prevent them from dropping out from activities. Children with DCD should be offered a variety of physical activities at a young age to enable them to select physical activities that they enjoy and that they continue to participate in as they become older. In providing guidance, practitioners should be aware that although children with DCD may report to participate in activities they may be active at a lower intensity than their peers. In the Netherlands the average time from the first meeting with a care professional to diagnosis is 33.5 months. This period may be shortened by increasing the awareness of DCD among care professionals, teachers and parents.

Second, sports should be made more approachable for children with DCD. The convention on the rights of persons with disabilities, issued by the United Nations, states that persons with disabilities should be enabled to participate on an equal basis with others in recreational, leisure and sporting activities. The convention instructs state parties to do this, among others, by taking the following measures: 1) encouraging and promoting participation in mainstream sporting activities and 2) by ensuring that “persons with disabilities have an opportunity to organise, develop and participate in disability-specific sporting and recreational activities.” For children with DCD, participation in mainstream sporting activities may be promoted by providing information about DCD to trainers and the general public. This might help them to get a better understanding of the behaviour and needs of children with DCD, and encourage them to think about solutions to include children with DCD successfully. Solutions may be different for each child: keeping the child involved with the team by giving the child a different role every now and then, focusing on sportsmanship rather than winning and fostering understanding within the team. Exploring the second measure proposed in the convention, there are a number of options. It is important to note though that tailored sports may be difficult to realise, especially in areas with a lower population density. Therefore, it may be advisable to think of initiatives that are suitable not just for children with DCD but that also appeal to other children. For children with DCD, the often competitive nature of sports may form a barrier for enjoying sports. One solution might be to have them train and compete with other children that have a similar level of ability. Another solution may lie in redirecting our focus from competitive activities to activities with a different or more varied focus of motivation. Originating from game research, we might be inspired by Bartle’s taxonomy of player types. Bartle distinguishes explorers, socialisers, achievers, and killers. Explorers are motivated by surprises, secrets and discovering things. Socialisers enjoy activities for the contact with others, they like to collaborate, make plans together, construct something together or make up stories. Achievers are motivated by the rewards in the game, completing the game and killers like to compete with others. Building on this new initiatives may be developed.

The Dutch government ratified the convention of the United Nations and for instance installed so-called “neighbourhood sport coaches” (in Dutch called “buurtsportcoaches”). These local coaches form an important link between sports and exercise providers and
other sectors such as care, welfare, youth care and childcare and education. The neighbourhood sport coach may play an important role in facilitating the participation of children with DCD in mainstream sports. Trainers and coaches of We12BFit! should be encouraged to collaborate with initiatives like this more closely. In addition, the Dutch government plans for a positive sports climate which entails training referees to take on a more guiding role and improving trainers’ pedagogical knowledge. These initiatives may be of great value for improving the participation in activities of children with DCD in the future. Finally, we need awareness of what being active, or actually lifestyle PA, means. First, being active is more than participating in sports. To quote a parents’ insight gained during We12BFit! “being active is not always in the big things, but maybe even more in the small things.” These small things may make a big difference when they become part of one’s everyday life. Examples are the default choice for cycling or walking to get from A to B, taking the stairs, getting children active (video) games and unwinding by undertaking activities. Second, the global recommendations on PA are based on maintaining a certain level of PF to prevent non-communicable diseases later in life. Being active and being physically fit should therefore not just be valued for their beneficial effects on functioning in the present but also for its importance regarding health in the future: being active is a way of life to live a long and healthy life.
References


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