

## Chapter 8

### **General discussion**



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The aim of this thesis was to improve the understanding why children with cerebral palsy (CP) show low levels of physical activity as reported in literature, and to determine the effectiveness of a six-month program aimed at stimulating physical activity of children with CP. This physical activity stimulation program consisted of a lifestyle intervention, involving counseling and home physiotherapy, and a four-month fitness training program. In Chapter 2 to 4 the level of physical activity of children with CP was compared to that of typically developing children (TDC), using step activity monitors, as well as heart rate monitors. In Chapter 5 to 7 the development of the six-month physical activity stimulation program, and its effects on physical activity and mobility capacity, fitness, fatigue, attitude towards sports, social participation, self-perception and quality of life were described. In this general discussion, the results of these studies will be used to provide directions for further research aimed at improving physical activity in children with CP.

## **Physical activity**

As discussed in the general introduction of this thesis, physical activity contributes to optimal development of the musculoskeletal system and physical fitness. Since children with CP are known to have lower levels of total physical activity than TDC,<sup>1,2</sup> more insight into the nature of these differences is needed. For that purpose the characteristics that might influence the level of walking activity of children with CP were examined. Also, the walking activity of Dutch children with CP distributed over the day and week was compared to TDC and to American children with CP (Chapters 2 to 4).

By means of a step activity monitor Chapter 3 and 4 demonstrated that children with CP show less walking activity than TDC in both the Netherlands and the United States. Likewise, reduced energy expenditure has been previously reported for children with CP.<sup>1,2</sup> As shown in other studies, the gap between TDC and children with CP widened, in both the Netherlands and the United States, when walking problems aggravate (Chapter 2 and 3).<sup>3,4</sup> In line with previous work, it was found that walking activity was particularly low in older5 children with bilateral spastic CP (compared to those with unilateral spastic CP),<sup>2,6</sup> who use walking aids (Chapter 2).<sup>2-4</sup> This study was the first to demonstrate a positive association between sports club participation and walking

activity of children with CP. These results agree with the lower participation in physical activities, such as sports, reported in children with CP who have problems with walking.<sup>7</sup> Our findings emphasize that physical activity promotion is particularly important for children with bilateral spastic CP and those using walking aids. Thereby, older children showed less walking activity than younger children with CP, supporting that promoting physical activity should be initiated at a young age. Further on in this general discussion, the effects of a physical activity stimulation program on sports club participation will be discussed.

The assessment of the physical activity distribution during the day provided information about the nature of the reduced physical activity levels in children with CP. Chapter 3 demonstrated surprising differences between Dutch and American children with CP with similar mobility capacity: American children with CP walking without aids showed more walking activity, while American children with CP using walking aids showed less walking activity than their Dutch counterparts. These differences were particularly evident at the higher stride rates. Possibly, the two countries offer different environmental and social opportunities for walking, which will be discussed in more detail later in this chapter. Furthermore, Chapter 4 demonstrated that children with CP spent more time inactive and less time at each stride rate level than TDC. These results support the importance of reducing inactivity next to promoting physical activity. Although general physical activity is important in general, the focus on the distribution of physical activity over the day joins the recent developments in the physical activity assessment among TDC. Authors have recently demonstrated that multiple short-duration activity moments during the day seem to elicit more health benefits than just being active for one continuous time interval while being inactive during the rest of the day.<sup>8,9</sup> This may be advantageous for children with CP, since they often mention fatigue as limitation for long-lasting activities. Information about the distribution of physical activity seems particularly important for children with CP who use walking aids, since Chapter 4 showed that, compared to TDC, their effort of walking expressed as percentage of heart rate reserve for the different stride rate levels was higher, whereas it was similar for children with CP walking without aids. Consequently, children with CP who use walking aids may be advised to expand their physical activity by reducing inactivity and inserting more low-intensity activity

moments during the day, while children walking without aids may be prompted to participate in a sports club and expand their activities without restriction.

Apart from the fact that disease and personal characteristics were found to influence walking activity (Chapter 2), the results in Chapter 2 and 3 provided indications for the importance of the environment on walking activity, particularly in children with CP. Firstly, children with CP demonstrated less walking activity during the weekend than during schooldays, whereas TDC did not (Chapter 2). Secondly, Dutch and American children with comparable limitations in activities (as categorized by the Gross Motor Function Classification System [GMFCS]) and comparable gross motor capacity had different walking activity in both countries, whereas walking activity of TDC did not differ between countries (Chapter 3). Previous work has also demonstrated that daily walking performance differs among children with the same GMFCS level,<sup>10</sup> as well as across different settings.<sup>10-12</sup> Apparently, walking activity of children with CP is influenced by environmental barriers that can be of either social, attitudinal or physical nature. Pratt and colleagues expect that adapting the physical environment (e.g. improved transport options and better walkability of a neighborhood) may have large effects on improvement of physical activity.<sup>13</sup> Positive associations between urban planning and physical activity have already been demonstrated among TDC.<sup>14</sup> Improved insight into the relationship between physical activity and the physical environment (e.g. urban planning) in children with CP may provide guidelines for policy makers responsible for optimizing the accessibility of the environment for people with a disability.

Although policies may improve the accessibility of the physical environment, changes may take a long time to be effectuated. Therefore, it may be beneficial to concentrate on short-term realistic opportunities for the stimulation of physical activity by focusing on improving the social environment. It is known that social environmental influences, such as being allowed to play outside, parental activity rules, and family encouragement, are related to enhanced physical activity of TDC.<sup>15</sup> Similar social environmental barriers were observed during the counseling module in the physical activity stimulation program described in Chapter 5, 6, and 7. Some parents indicated that having no friends to play with in the neighborhood was a reason for their child to stay indoors. Other parents did

not encourage their child to play outside, because they were afraid of their child being bullied. Furthermore, parents indicated that the busy school days, and the logistics of bringing and fetching children to and from school made them adhere to a less tight schedule during the weekend, resulting in less active weekend days ('pyjama days'). Further research should examine whether parental influences, such as setting activity rules, and having friends in the neighborhood to play with, can influence physical activity of children with CP.

## **Physical activity stimulation program**

### **Rationale and effectiveness of the physical activity stimulation program**

The six-month 'LEARN 2 MOVE 7-12' physical activity stimulation program was developed to break down the vicious cycle of inactivity and possible deterioration of mobility, by focusing on improving fitness and promoting physical activity (Chapter 5, 6, and 7). The promotion of physical activity occurred through motivational interviewing (MI), as well as by practising gross motor skills in a home situation. MI is an interview style that can be used to elicit behavioural change in people in different stages of readiness for change and that offers strategies to deal with resistance to change.<sup>16</sup> It was hypothesized that the different components in the physical activity stimulation program would reinforce each other and would have added value for improving physical activity in comparison to regular pediatric physiotherapy. No statistically significant improvements in physical activity or any of the other measured outcomes (mobility capacity, fitness, fatigue, attitude towards sports, social participation, self-perception, and quality of life) were observed (Chapter 6 and Chapter 7). Nevertheless, clinically relevant trends for improvement were found for gross motor capacity and social participation in domestic life, as determined by a better accomplishment of activities in and around the home. These findings may provide directions for future physical activity programs.

### **Physical activity**

The study showed that objectively assessed walking activity did not change after the physical stimulation program. However, a short-term positive trend was found for subjective parent-reported physical activity (Chapter 6). As addressed in Chapter 6, the lack of an intervention effect on physical activity might be a result of insufficient

contrast between groups. Probably, participating families and physiotherapist were all interested in (increasing) physical activity and fitness. Moreover, all participants knew that physical activity was the outcome of interest, which might have raised awareness of the importance of physical activity. Also, the intervention period may have been too short to elicit a physical behavioural change: a recent review has indicated that successful behavioural interventions in TDC should last at least one year.<sup>17</sup> Likewise, previous work among young people with CP has only identified trends for improving physical activity after two months of internet-counseling,<sup>18</sup> or eight months fitness training.<sup>19</sup> As behavioural changes take time, future work should examine whether a longer duration of the physical activity stimulation program (i.e. for more than one year) can establish effects on physical activity.

### **Mobility capacity**

As addressed in Chapter 6, no statistically significant effect was found for mobility capacity, expressed as gross motor capacity, walking capacity and functional strength. Nevertheless, the positive short-term trend found for gross motor capacity seems to be clinically relevant (Chapter 6).<sup>20</sup> Although the design of the study restricts us from concluding which component of the intervention was responsible for this positive observation, it seems likely that the individually tailored home-based physiotherapy and the gross motor activities practised during the fitness training contributed to this trend. The focus of the home-based physiotherapy on improving mobility capacity in the own environment (e.g. the child's home or neighborhood) in combination with the fitness training may have resulted in more practice of gross motor activities than regular physiotherapy.<sup>21</sup> This supports that children with CP should practise gross motor activities as often as possible in order to maintain and improve them ('use it or lose it'). To avoid life-long practising of activities in the physiotherapy practice, there may be an opportunity for low-frequent counseling and home-based physiotherapy to encourage children to perform all kinds of gross motor activities in regular daily life. This is in line with the lower physiotherapy frequency in a meaningful context proposed by Darrah et al.<sup>22</sup> The set-up of our low-frequent counseling and home-based physiotherapy may guide the development of future interventions aimed at improving mobility capacity of children with CP. Whether improving mobility capacity can lead to improvement of

physical activity should be studied in future research.

### **Social participation**

The study described in Chapter 5 to 7 was the first to evaluate the effect of a lifestyle intervention and fitness training on social participation in domestic life, showing a trend towards an increased accomplishment of activities in and around the home. This was in line with our expectations, since the home-based physiotherapy and counseling were focused at increasing activities in the own home. Better accomplishment of activities in and around the home may be a starting point for expanding daily activities in the future. Due to frequently mentioned barriers for sports, such as a lack of energy, transportation to and availability of sports clubs,<sup>23</sup> expanding daily physical activities is assumed to be more realistic for children with CP than expanding recreational activities outside the home. Accordingly, Van der Ploeg et al. showed that counseling focused on expanding daily activities was successful in increasing self-reported physical activity (particularly daily activities) of adults in rehabilitation centers, whereas counseling focused on sports participation was not.<sup>24,25</sup>

Recent research has pointed out that more frequent physical activity at low intensity may have more beneficial effects on inactivity-related health problems, than sporadic physical activity at moderate to high intensity in healthy adults.<sup>26</sup> Furthermore, an increased risk for all-cause mortality as a result of a sedentary lifestyle seems to occur independently from activity level among healthy adults.<sup>27</sup> In a health perspective, more short bouts of activities at low intensity have been suggested to be more beneficial than one hour at high intensity while the rest of the day is spent inactive.<sup>28,29</sup> For children with CP reducing the number of prolonged sedentary moments may be more feasible to achieve than increasing physical activity. This supports the idea that counseling should focus on expanding daily activities distributed equally over the day, while emphasizing that these activities may be performed at low intensity. Whether better accomplishment of activities in and around the home results in a reduction of inactivity and more low-intensity physical activity in the own environment should be confirmed by future research.



## **Potential for physical activity stimulation**

The discussion about physical activity, and the clinically relevant trends on gross motor capacity and the accomplishment of activities in and around the home, demonstrate that there may be potential for a long-lasting lifestyle intervention with periodical attention to decreasing inactivity. The observation during the lifestyle intervention that some children with CP are as active as their typically developing peers supports that improving physical activity is feasible in this population. Such an approach requires commitment of child and parents, as well as an intervention that is tailored to each family's needs. An individually tailored approach with involvement of the family may be advantageous for the child's and parents' self-management and motivation, and suits the shift towards family-centred care in a relevant context within pediatric rehabilitation.<sup>22</sup> Thereby, the chronic nature of CP results in children (with CP) facing various challenges with activities and social participation throughout childhood. Potentially, periodical coaching by the physiotherapist in the form of a lifestyle intervention can help to improve self-efficacy, and to detect pitfalls and opportunities for physical activity in each stage of life. This may prevent additional health care costs on the long-term.<sup>30</sup> Promising effects of lifestyle interventions targeting the family unit have been demonstrated among children with obesity.<sup>31</sup> Furthermore, positive effects of health-coaching in adults with a chronic illness have been reported on self-management, and the readiness for behavioural change.<sup>32</sup> Future research should examine whether a long-lasting (i.e. longer than one year) physical activity stimulation program can elicit improvement in physical activity and a reduction of sedentariness. Although there are indications for the potential of a low-frequent individually-tailored family-centred lifestyle intervention, more insight into the contribution of the separate components of these programs is needed to understand how they influence physical activity, by varying merely one component at the same time. Furthermore, it should be studied whether improvements in mobility capacity and accomplishment of activities lead to increases in physical activity on the long term.

## **Practical implications for implementing a lifestyle intervention**

Researchers investigating the effectiveness of lifestyle interventions have to consider a few aspects. The physiotherapist who performed the counseling in the present

study had additional training in motivational interviewing, so additional education on counseling might be necessary for physiotherapists. Furthermore, physiotherapists might experience barriers for executing the home-based physiotherapy, since home visits require more time than treatment at the physiotherapist's practice.<sup>22</sup> Although it is necessary to visit the child at home to assess opportunities for improving the performance of activities in the home situation, internet-based training methods, telephone calls, or internetlogs can be used afterwards for remote support decreasing the frequency of face-to-face therapy,<sup>13,18,33</sup> which may lower the burden for the physiotherapist.

## **Methodological issues**

### **Physical activity assessment problem in CP**

The assessment of physical activity in children with CP appeared to be challenging, since the different methods to assess physical activity used in this thesis provided different kinds of results (Chapter 2, 4 and 6). Children with CP showed a lower walking activity (activity monitoring), but no differences in daily exercise intensity (heart-rate monitoring) compared to TDC (Chapter 2, and 4). In addition, the children with CP who participated in the LEARN 2 MOVE physical activity stimulation program (Chapter 6) did not increase their walking activity but showed a trend to improve parent-reported physical activity as compared to the control group. An explanation for these discrepancies might be that the step activity monitor used in this study has been primarily designed to register walking, and was not calibrated for running. Heart rate monitors and parent-reports can capture all kinds of activities but have other disadvantages, such as the reaction of heart rate on excitement and stress, and the problem of recall-bias and socially-desirable answers of subjective parent-reports.<sup>34</sup> Similar discrepancies between the objective and subjective assessment of physical activity have been demonstrated among TDC.<sup>35</sup> Methods for objectively assessing physical activity of children with CP should be improved. The results in Chapter 4 show that a step activity monitor is preferred to assess community walking at low to moderate intensity, but that a heart rate monitor is recommended to capture the high intensity activities, like sports. Additionally, a different activity monitor, of which the ability to register walking has recently been determined in children with CP, might have the potential to capture other activities apart from walking.<sup>36</sup> There are

also positive notes about the use of Global Positioning System (GPS) data for quantifying social participation of children with CP,<sup>37</sup> but the added value of these systems for the assessment of physical activity are not clear yet.

### **Study population**

There is a chance that particularly families who are interested in (increasing) physical activity participated in the study. This is a likely possibility, because the baseline attitude towards sports of both child and parents was already very positive in both groups. This might have negatively influenced our results, since families with an already good attitude towards sports have less room for improvement. It also decreases the generalizability of our results for families who are less interested in physical activity. This exactly indicates the challenge of physical activity stimulation programs. Especially families that lack the interest in physical activity are supposed to have the lowest physical activity level, being the target population for physical activity stimulation programs. However, these families may not come up with issues in the area of physical activity, and may not apply for a physical activity stimulation program themselves, since they might not realize that it is important for their child to be physically active. When physical activity stimulation is integrated within regular physiotherapeutic care, also families who are less interested in physical activity may be reached.

### **Key message**

The reduced walking activity levels of children with CP especially during the weekend do demonstrate that physical activity promotion is needed. Interventions for children with CP should preferably be initiated at a young age, and particularly target children with bilateral CP and those using walking aids. Determining environmental barriers for increasing physical activity should be part of physiotherapy treatment, in order to explore opportunities for expanding physical activity and reducing inactivity. The stimulation of sports club participation and promotion of low-intensity daily activities are both advised.

Although the physical activity program described in this thesis did not elicit improvements in amount of physical activity, it did point to the need and potential to enhance mobility

capacity and accomplishment of activities in the domestic environment. Future research should examine whether a long-lasting (i.e. longer than one year) physical activity stimulation program can elicit improvement in physical activity and reduction of sedentariness. A low-frequency individually-tailored lifestyle intervention with periodical attention to physical activity and practising of gross motor activities in the home environment may have potential for improving physical activity and more cost-effective care, which should be confirmed in future studies.

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