Influence of Chronic Pain in Physical Activity of Children with Cerebral Palsy

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ABSTRACT

BACKGROUND: Children with cerebral palsy (CP) perform less physical activity than their typically developing peers (TDP). Pain, important comorbidity in children with CP, restrains levels of physical activity.

OBJECTIVE: This study aims at exploring the influence of chronic pain in physical activity of children with CP and TDP.

METHODS: 24-hour heart rate was registered in four groups of children: children with CP and TDP, with and without chronic pain. Heart rate based indexes of physical activity (MET percentages, energy expenditure) were computed. A self-reported diary of activities rated activities pain and fatigue intensity.

RESULTS: Children with CP and chronic pain reported more painful activities and higher pain than their TDP with chronic pain. Moreover, children with CP and chronic pain presented higher time and periods of light activity and less sedentary activity

than their TDP with chronic pain. No differences were found between CP and TDP without chronic pain.

CONCLUSION: Children with CP regulate physical activity differently than TD children in the presence of chronic pain. The maintenance of light levels of physical activity in children with CP may suggest efficient pain coping strategies and perseverance in participation. These findings encourage the implementation of programs to improve fitness in this population.

Keywords: *Cerebral palsy, children, chronic pain, MET, physical activity.*

1. BACKGROUND

Cerebral palsy (CP) is a neurodevelopmental condition mainly characterized by motor impairment affecting motor performance and physical activity. It has been reported that 89% of adolescents with CP were not physically active enough (van Eck et al., 2008), participating in 48 minutes/day less in physical activities and 80 minutes/day more in sedentary activities and performing mostly low intensity physical exercise compared with their typically developing peers (TDP) (Nooijen et al., 2014Balemans et al., 2013; Carlon et al., 2013; Maher et al., 2007). Physical activity at moderate-vigorous intensity has been associated with high cardiorespiratory fitness (Ryan et al., 2015). Thus, although children and adolescents with CP need higher energy expenditure and showed higher aerobic responses, such as oxygen consumption, to perform the same physical activities than their peers (Peri et al., 2015; Verschuren et al., 2014; Brunton & Rice, 2012; Dallmeijer et al., 2011; Maltais et al., 2005), daily energy expenditure and cardiorespiratory parameters remain lower than in TDP, due to adaptative strategies leading to minimize physical strain and keep oxygen cost sostenible (Balemans et al., 2014, Sison-Williamson et al., 2014; Verschuren et al., 2014; Verschuren & Takken, 2010). Nevertheless, although energy efficiency has an important influence in activity limitation, it appears that is not associated with participation restriction or health status in individuals with CP (Kerr et al., 2008). Therefore, to adequately plan physical intervention in individuals with CP, other factors should be considered.

Pain is one of the most important secondary problems in individuals with CP and could be one of the most relevant factors influencing both their physical activity and participation in daily activities. More than 50% of individuals with CP have pain from moderate to severe intensity on a daily basis and at multiple body locations

(Alriksson-Schmidt et al., 2016; Riquelme et al., 2011). Pain negatively affects participation (Dang et al., 2014; Ramstad et al., 2012; Berrin et al., 2007) and quality of life in individuals with CP (Findlay et al., 2016; Colver et al., 2015; Badia et al., 2014; Riquelme et al., 2011), deteriorating health perception (Ramstad et al., 2012). On the other hand, chronic pain has been related to a reduction of physical activity levels, an increase of sedentary activities and a decreasing of aerobic conditioning (Higuchi, 2016; Gueddari et al., 2014; Hallman et al., 2014; van der Velde et al., 2011). In addition, Hadrevi et al. (2013) have suggested that chronic pain develops metabolic processes related to energy utilization that could perpetuate the pain disorder, and some metabolic substances associated with cardiorespiratory responses at physical exercise have been related to pain sensitivity and pain tolerance (Scioli-Salter et al., 2016). Thus, exercise promoting programs have being effective in reducing pain intensity and increasing pain tolerance through different physiological mechanisms, such as improving muscle oxygenation or normalizing the muscle contraction cycle (Jones et al., 2014; Hadrévi et al., 2013; Sogaard et al., 2012; Andersen et al., 2010).

Despite extensive research on physical activity and cardiorespiratory fitness in children with CP, there are no studies examining the role of pain on physical activity and aerobic parameters within this population. The present study aims at comparing physical activity between children with CP and typically developing peers (TDP), with and without chronic pain, through the monitorization of heart rate during a typical weekday (24 hours).

2. METHODS AND MEASURES

2.1. Participants

Children with CP and TDP were recruited from educational centers established in the island of Majorca (Spain) in 2015 using an informational letter explaining the details of the study. Inclusion criteria were: (*a*) age between 4 and 16 years, and (*b*) cognitive level that allows understanding the instructions and procedure (i.e., to report when they felt pain in an activity). Augmentative communication devices and information from caregivers were used as needed to facilitate data collection in subjects with communication difficulties.

Twenty-six children with CP [7 females; mean age = 9.12 (3.82)] and 26 TDP [13 females; mean age = 9.27 (3.07)] matched in age, sex and chronic pain (defined as pain or repetitive pain experiences lasting more than 3 months) decided to participate in the study. Subjects or their parents reported their age, sex, height, weight and presence of chronic pain. The type of cerebral palsy, gross motor function and cognitive level were obtained from the health history in children with CP. According to the presence of chronic pain, four subgroups of children (children with CP with chronic pain, children with CP without chronic pain, TD children with chronic pain, and TD without chronic pain) were formed for the analyses. Table 1 displays clinical characteristics of participants with CP.

Parents/legal tutors signed informed consents and participants gave their written or verbal approval to participate in the study. Participants' rights were protected. The study protocol was approved by the Ethics Committee of the Regional Government of the Balearic Islands (Ref. IB 2388/14 PI).

2.2. Instruments and procedure

Heart rate was recorded along 24-hour ambulatory daily life activities. Heart rate was recorded by means of a device (Bodyguard 1, Firstbeat Technologies Ltd., Finland) attached by pressure clips to two adhesive hipoalargenic electrodes (Ambu BlueSensor, Ambu L., Denmark) placed on the skin: one on the right subclavicular area and another one on the left submamilar area. Heart rate data was recorded every second, stored and analyzed offline at the end of the experiment. Heart rate based indexes (i.e. *energy expenditure*, *exercise intensity according to MET thresholds*) were calculated offline by Firstbeat software (for methodology, see Firstbeat Technologies white papers). Heart rate based indexes during rest and active activities have shown to be reliable when calculating energy cost in children with CP with a broad range of motor function, even at low levels of physical activity (Bratteby Tollerz et al., 2011; Bar-Haim et al., 2008; Suzuki et al., 2001; van den Berg-Emons et al., 1996).

The measurement instrument was placed and removed 24-hour later, by a trained member of the research team. Prior to placement of heart rate sensors, the researcher gave participants and their relatives information on the equipment and specific instructions about their use. The device was small, light and comfortable and any participant reported any disconfort during the experiment, except some transitory skin irritation in some children when electrodes were removed.

In addition, a diary of activities of the monitoring period was completed by the child or their parents. The participants were instructed to write in the diary all the activities performed during the monitoring period, their time of beginning and end, including pauses. Every activity had to be rated with self-reports of pain and fatigue experienced during the activity in an 11-point faces scale. At the end of the

experiment, when recording devices were removed, the researcher reviewed with the participants the diary of activities to clarify the notes and avoid blanks. This procedure has been previously used in to explore physical activity in patients with chronic pain (Hallman et al., 2014; Hallman & Lyskov, 2012; Duque et al., 2000).

2.3. Statistical analyses

Analyses of variance (ANOVA) including the between-subject factors GROUP (children with CP versus typically developing peers) and PAIN (chronic pain versus non-chronic pain) were performed. ANOVA results were adjusted by using Bonferroni corrections for post-hoc comparisons. Significant levels were set at $p<0.005$.

3. RESULTS

3.1. Self-reports

Chronic pain in children with CP was reported to be caused by musculoskeletal problems or constipation, and located in lower limbs, back and abdomen. Chronic pain (lasting more than 3 months) in TDP was associated with musculoskeletal injuries, such as fractures or strains, headache, or to digestive and upper respiratory health conditions and was mostly located in upper or lower limbs, head, abdomen, throat and mouth.

Figure 1 displays pain and fatigue intensity reported by each of the groups. Main effects of GROUP ($F_{1,48}=6.35$, $p=0.015$) and PAIN ($F_{1,48}=7.03$, $p=0.011$) on ***number of painful activities*** indicated that children with CP reported more painful activities than their TDP, and that children with chronic pain reported more painful activities than children without chronic pain. In addition, significant interaction GROUP x PAIN ($F_{1,48}=10.63$, $p=0.002$) further showed that children with CP and

chronic pain reported more painful activities during the 24-hour period than TDP with chronic pain (post-hoc, all $p<0.001$), whereas no significant differences were found between children with CP and TDP without chronic pain. Post-hoc comparisons also showed than only children with CP had significant differences between children with and without chronic pain in these reports (children with CP and pain reported more painful activities than children with CP but without chronic pain, $p<0.001$).

A significant main effect PAIN ($F_{1,48}=7.19$, $p=0.010$) on ***pain intensity average during 24 hours*** (Fig. 1) was also found. In addition, an interaction GROUP x PAIN ($F_{1,48}=4.42$, $p=0.041$) further showed higher pain intensity only in children with CP with chronic pain compared with children with CP without chronic pain, whereas no significant differences were found in the TDP group.

A significant interaction GROUP x PAIN ($F_{1,48}=13.07$, $p=0.001$) on ***pain intensity during painful activities*** was found, showing that children with CP and chronic pain reported higher pain intensity than TDP with chronic pain during painful activities ($p=0.001$). In addition, children with CP and pain reported higher pain intensity at painful activities than children with CP without chronic pain ($p<0.001$). No differences were found in TDP children.

The ***type of activities producing pain*** was also different in TD and CP children: whereas pain in TDP children was mainly due to musculoskeletal injuries related with accidents (e.g. burning, fractures, strains) or abdominal, throat and tooth pain, pain in CP children included these causes, but also others directly related with their pathology (e.g. spams, seizures or rehabilitation). Furthermore, whereas TD children reported pain mainly during meals or at waking up, CP children reported pain during sleep, standing or static positions such as sitting or lying, parental managing (e.g. transferring from wheelchair to bed), washing or clothing.

A significant main effect of GROUP on ***number of activities causing fatigue*** was found ($F_{1,48}$=5.17, p=0.027), indicating that CP children reported fatigue in more activities than their TDP. ***Fatige intensity during activities causing fatigue*** and ***the fatige intensity average during 24 hours*** did not show significant differences among the groups. CP children experienced fatigue in the same ***type of activities*** than their TDP (eg. walking to the school, climbing stairs, sports, homework), but reported fatigue also in daily activities such as eating, clothing, washing, standing, and in specific activities such as motor rehabilitation, respiratory physiotherapy, spams and seizures.

3.2. Heart rate based indexes

Figure 5 displays the heart rate during the 24-register period for one typical child of each group. Heart rate based indexes showed significant main effects of GROUP on ***time of light physical activity*** (intensity between 20%-30% of MET maximal, in minutes) and ***time of moderate/vigorous physical activity*** *(*intensity > 30% MET maximal) (all F>6.80, all p<0.014, CP>TDP) (Fig. 2). Significant interactions of GROUP x PAIN on ***time of sedentary physical activity*** *(*intensity < 20% of MET maximal; $F_{1.48}$=5.49, p=0.24), *time of light physical activity* ($F_{1.45}$=5.38, p=0.025) and *time of moderate/vigorous physical activity* (F(1.48)=5.45, p=0.024) indicated that children with CP and chronic pain performed more time of light and moderate/vigorous physical activity (both p<0.003), whereas spent less time sedentary (p=0.05) than their TDP with chronic pain; no significant differences were found for children without pain. When analysing the ***time of periods lasting over 4 minutes*** within an specific physical activity intensity, a significant main effect of GROUP ($F_{1,48}$=12.10, p=0.001, CP>TDP) and GROUP x PAIN interaction were found

only on light physical activity ($F_{1,48}$=8.83, p=0.005). Post-hoc analyses showed more periods of light activity in CP with chronic pain than in TDP with chronic pain (p<0.001) and than in children with CP without pain (p=0.008). The ***average MET maximal*** showed a significant main effect GROUP ($F_{1,48}$=7.96, p=0.007, CP>TDP) and a significant interaction GROUP x PAIN ($F_{1.48}$=9.82, p=0.003) indicating higher MET maximal in CP with chronic pain than in TDP with chronic pain (p<0.001) and than in TDP, children with chronic pain displayed lower MET maximal than children without chronic pain (p=0.01). Figure 3 displays the percentage of MET maximal during the 24-register period for one typical child of each group.

Energy expenditure at light activity (Fig. 4) showed a significant main effect of GROUP ($F_{1,48}$=5.18, p=0.028, CP>TDP) and a significant interaction of GROUP x PAIN ($F_{1,48}$=4.78, p=0.034), indicating higher energy expenditure in children with CP and chronic pain than in TDP with chronic pain (p=0.005), whereas no differences were found for children without chronic pain. No significant differences were observed for *total energy expenditure* or *energy expenditure at sedentary* or *moderate/vigorous* physical activity.

4. DISCUSSION

The aim of the present study was to compare physical activity between children with CP and their typically developing peers (TDP) with and without chronic pain, by using self-report and heart rate based indexes of physical activity during a typical week day (24 hours). Our data showed that children with CP and chronic pain presented higher light activity and less sedentary activity than their TDP with chronic pain, whereas no differences were found between CP and TDP without chronic pain.

Our results on physical activity in TDP without pain are similar to those from

previous studies (Garriguet et al., 2016; Zarrouk et al., 2009; Wong, 1994). In addition, our data showed a significant reduction of global physical activity in children with chronic pain, both children with CP and TDP. This is in accordance with previous research reporting a reduction of physical activity frequency, intensity and duration in children presenting other painful conditions, such as headache, back pain or stomach ache (Swain et al., 2016; Sollerhed et al., 2013) or in children with disabilities, such as juvenile idiopathic arthritis or hypermobility syndrome (Gueddari et al., 2014; Limenis et al., 2014; Schubert-Hjalmarsson et al., 2012). Nevertheless, physical activity was different in children with CP and TDP when chronic pain was present. Thus, CP children with chronic pain showed longer time of light and moderate physical activity, more time in long periods of light physical activity and less time of sedentary activity, as well as higher energy expenditure at light physical activity, than their TDP with chronic pain. This seems to be in contrast with the effects of pain in other childhood disabilities, such as juvenile idiopathic arthritis or pediatric sickle cell disease, which associated pain to a reduction of time dedicated to moderate physical activity and an increment of sedentary activity (Hallman et al., 2014; Perruchoud et al., 2014). The maintainance of at least light levels of physical activity in children with CP with chronic pain occurred even when they reported more frequent pain events and higher pain intensity during daily life activities than their TDP with chronic pain. As suggested by Perruchoud et al. (2014), the relationship between pain and physical activity is complex and can be related to the ability to cope with pain. The chronicity of the CP condition may expose children to high levels of pain in a daily basis from their early childhood (Alriksson-Schmidt et al., 2016; Findlay et al., 2016; Riquelme et al., 2011), which may have emphasized the development of effective pain coping strategies in these children, such as task persistence and activity

perseverance (Brunton & Bartlett, 2013; Jensen et al., 2011, 2006; Dudgeon et al., 2006), in contrast with the shorter-lasting pain experience of TD children. In this sense, feelings of pain control and higher self-efficacy perception has shown to be related with higher physical activity in children with juvenile idiopathic arthritis (Higuchi, 2016; Norgaard et al., 2016). As pain coping is an integral part of physical activity (Sollerhed et al., 2013), children with CP that have learnt to cope with unpleasant body sensations during years, may maintain unpleasant physical activity which assure them daily life participation. Thus, pain coping strategies should be taken into account for the design of future studies on this topic. As chronic pain has been associated to aerobic deconditioning and disability (Doury-Panchout et al., 2014) and children with CP present low physical activity levels that would not allow achieving the healthy fitness level (van Eck et al., 2008), the perseverance in physical activity shown by the children with CP in our study might facilitate the adaptation of daily activity levels from light to moderate intensity (Suzuki et al., 2001) or the implementation of physical activity programs in this population despite the presence of pain. On the other hand, children in chronic pain from neurologic/MSK causes may benefit from movement as a pain relief strategy and this may contribute to our findings, as no differences in physical activity were found between children without pain. As sedentary behaviour and low cardiorespiratory fitness has been shown to increase the likehood of pain conditions, pain intensity and analgesic consumption in children (Vierola et al., 2016), physical activity programs may not only improve physical condition but also reducing pain in children with CP.

This reseach has some limitations that must be taken into account for the correct interpretation of the results. Due to the small sample size, effects of gross motor function impairment in physical activity and pain could not be explore in

children with CP, despite of having been closely related to physical activity levels and energy expenditure (Trost et al., 2016; Bania et al., 2014; Kamp et al., 2014). The characteristics of the sample should be taking into account for generalization of results. Moreover, the use of self and surrogate daily activities and pain and fatigue reports may have decreased the reliability of the data. Nevertheless, our study lays a scientific basis for future studies exploring the complex relationships between two important comorbid conditions in children with cerebral palsy: pain and low physical activity.

5. CONCLUSION

Children with CP may regulate physical activity in a different way than TD children in the presence of chronic pain. The maintenance of at least light levels of physical activity suggests efficient pain coping strategies and perseverance in participation in daily activities in children with CP and chronic pain. This information may be useful to the implementation of higher levels of physical activity, both to improve fitness conditioning and reduce pain in children with CP. Further research is warranted to elucidate the bidirectional association between physical activity and pain in individuals with CP and its changes along the lifespam.

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Conflict of interest

The authors certify that they have no affiliations with or financial involvement in any organization or entity with a direct financial interest in the subject matter or materials discussed in the article.

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TABLE 1. Clinical characteristics of individuals with cerebral palsy. GMFCS= Gross motor function classification system (Palisano et al., 1997); it describes the gross motor function according to 5 levels: 1=walks without limitations, 5=transported in a manual wheelchair.

TABLE 1

Clinical variable	*Children (n)*	
Chronic pain	*Yes*	*No*
	11	15
Type of cerebral palsy		
Bilateral spastic	6	13
Unilateral spastic	0	2
Diskinetic	5	0
Ataxic	0	0
Motor impairment (GMFCS)		
Level I	1	1
Level II	2	3
Level III	0	4
Level IV	1	1
Level V	7	6
Cognitive impairment		
None	6	6
Mild	4	6
Moderate	1	3
Severe	0	0

FIGURE 1

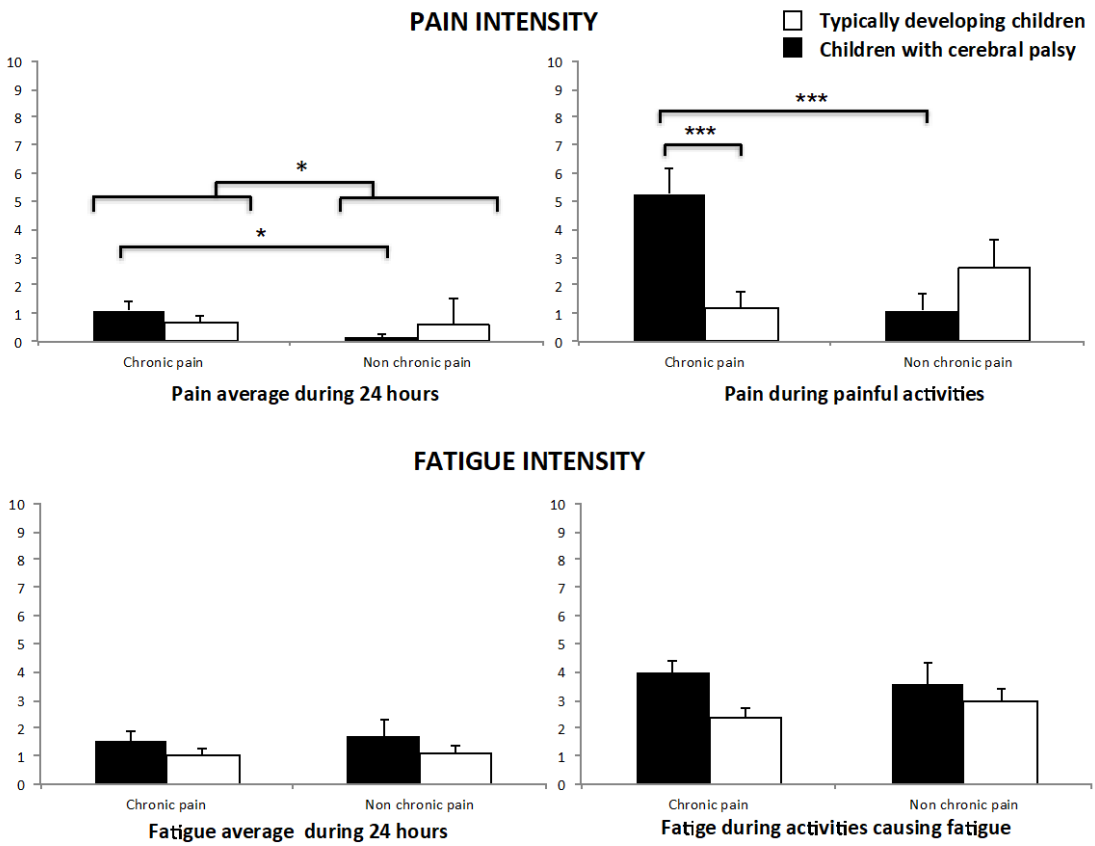

Fig. 1. Self-reports of pain and fatigue intensity for the four groups of children (cerebral palsy with and without chronic pain, and typically developing peers with and without chronic pain). Pain and fatigue during the 24-hour register = average of all activities pain intensity; Pain and fatigue during the painful activities = average of activities rated >0 in pain or fatigue intensity. Ratings were measured by using a11-point numerical scale: 0= No pain, 10= Unbearable pain). Results are displayed as mean ±SE. * $p < 0.05$, *** $p < 0.001$.

FIGURE 2

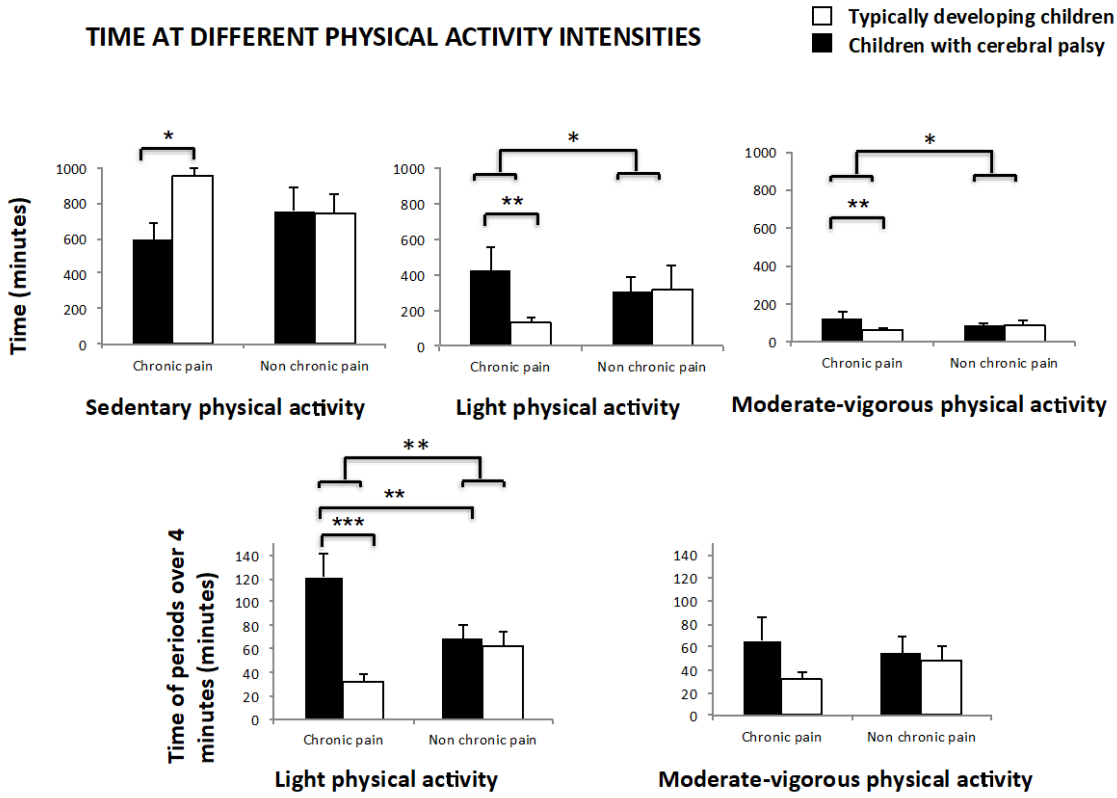

Fig. 2. Physical activity at different intensities for the four groups of children (cerebral palsy with and without chronic pain, and typically developing peers with and without chronic pain). Results are displayed as mean ±SE. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

FIGURE 3

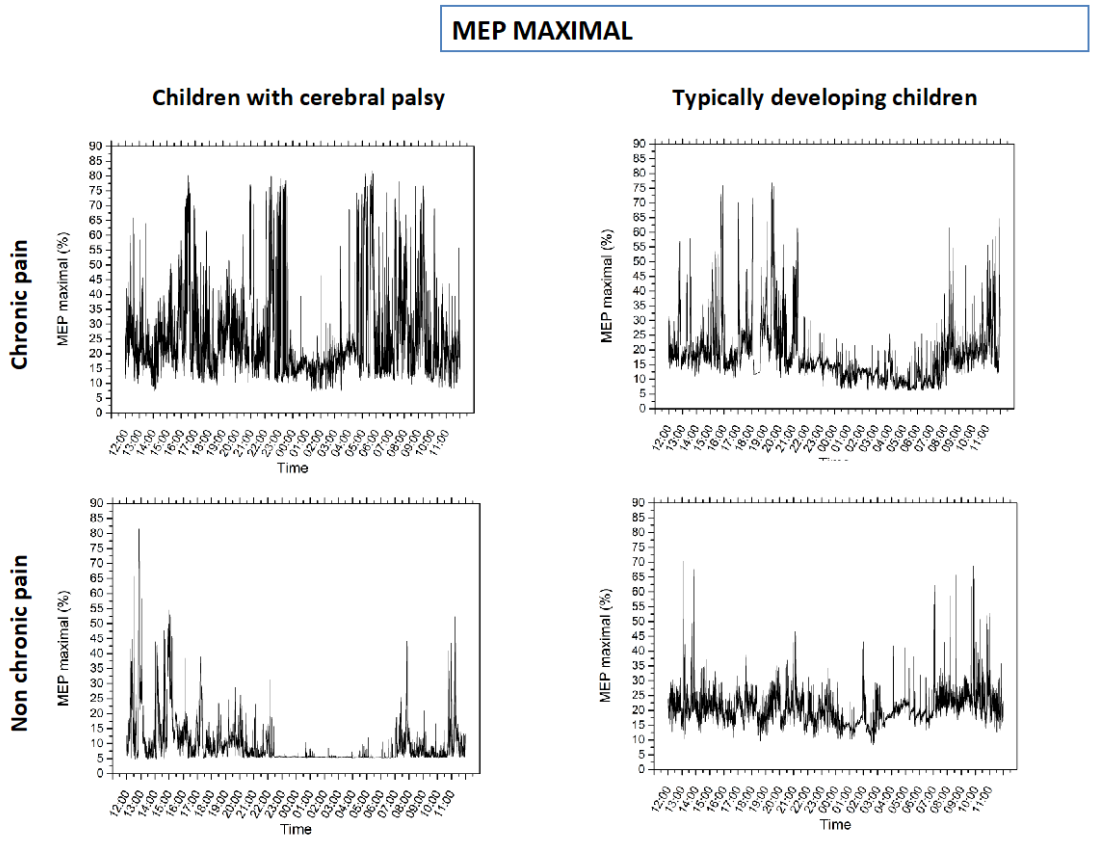

Fig. 3. Curves of MET maximal during the 24-hour register for one typical child from the four groups (cerebral palsy with and without chronic pain, and typically developing peers with and without chronic pain).

FIGURE 4

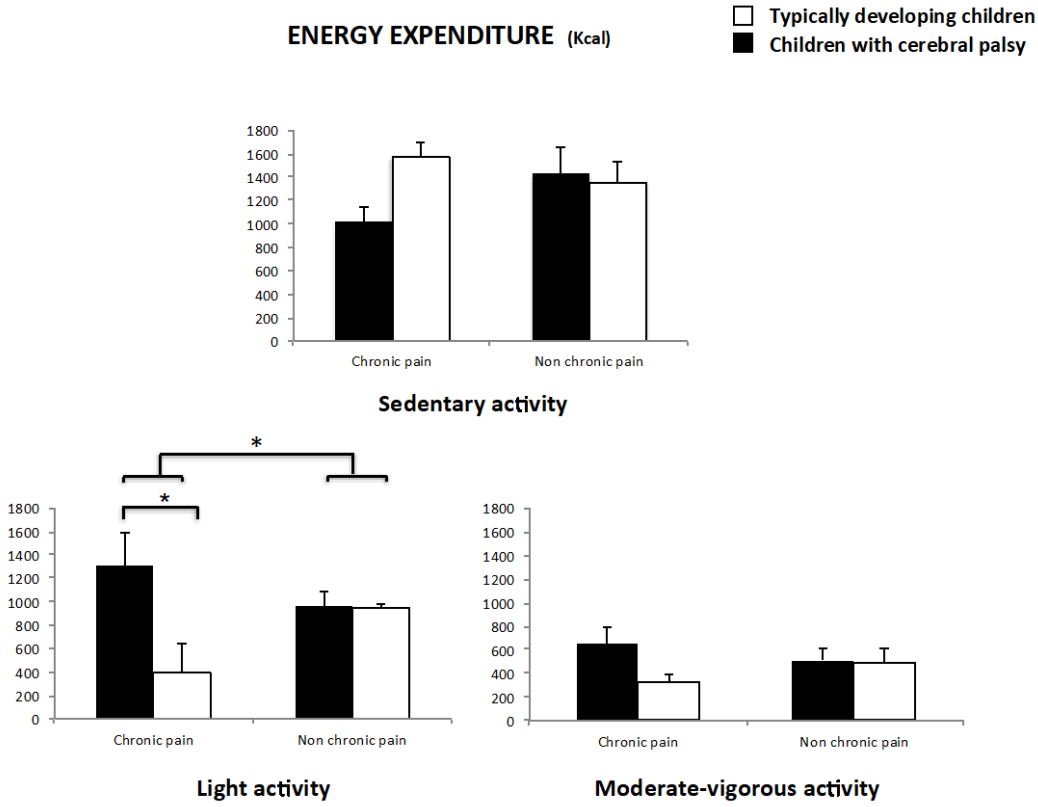

Fig. 4. Energy expenditure at different physical activity intensities for the four groups of children (cerebral palsy with and without chronic pain, and typically developing peers with and without chronic pain). Results are displayed as mean ±SE. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

FIGURE 5

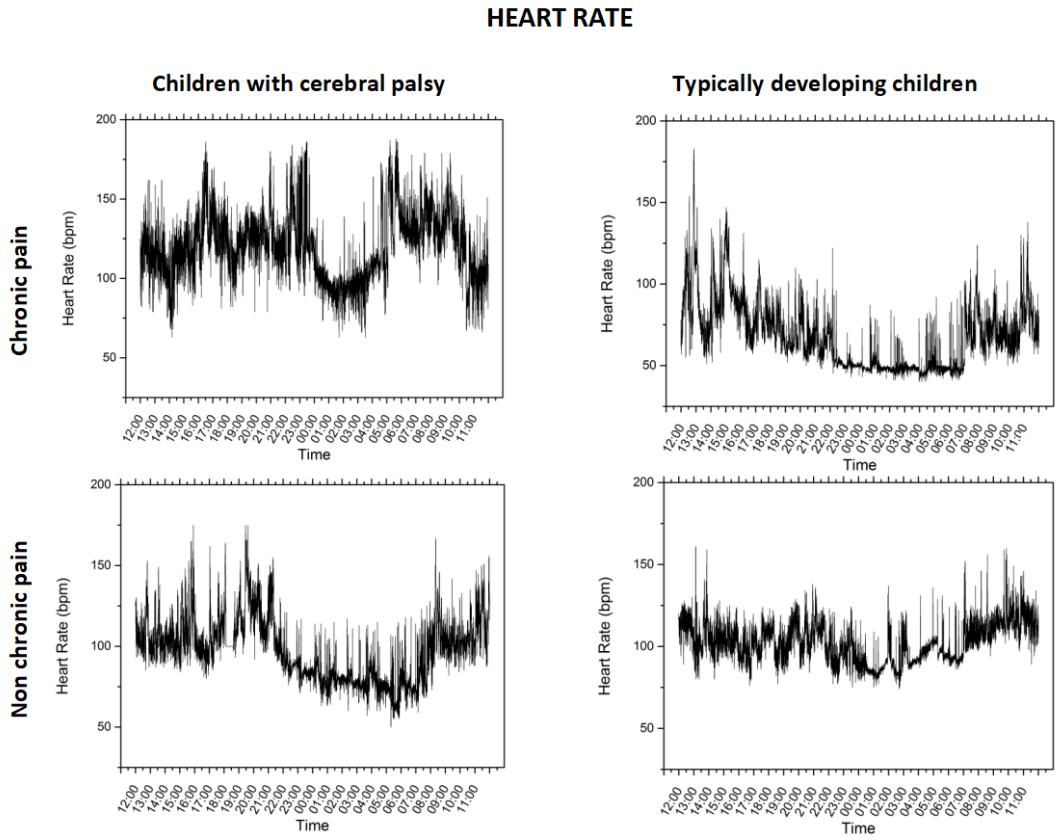

Fig. 5. Heart rate during the 24-hour register for one typical child from the four groups (cerebral palsy with and without chronic pain, and typically developing peers with and without chronic pain).