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## Perceived body distortion rather than actual body distortion is associated with chronic low back pain in adults with cerebral palsy: A preliminary investigation

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**Title:** Perceived body distortion rather than actual body distortion is associated with chronic low back pain in adults with cerebral palsy: A preliminary investigation

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**Key Word:** low back pain, body distortion, body image, adults with cerebral palsy

**Abstract**

**Objectives:** The aim of the present study was to investigate whether distorted body perception is a feature of the low back pain experience in people with Cerebral Palsy (CP) and whether any distortions noted are confounded by the presence of motor and postural impairments commonly seen in CP.

**Methods:** Forty-five individuals participated in this study: fifteen adults with CP with LBP (CP\_Pain group), fifteen adults with CP without LBP (CP\_noPain group), and fifteen age-matched adults with LBP but no CP (Pain group). Body perception was evaluated using the Fremantle Back Awareness Questionnaire (FreBAQ) and by assessing two-point discrimination (TPD) thresholds over the low back. A comprehensive assessment of motor function was also undertaken in the CP population and postural function was assessed in all three groups.

**Results:** Significant differences between the three groups were found for FreBAQ scores ( $p < 0.0001$ ). The TPD threshold in the low back of the CP\_Pain group was significantly larger than that of the CP\_noPain group ( $p = 0.01$ ), though we found no difference between the CP\_noPain group and the Pain group ( $p = 0.21$ ). We found no difference in motor or postural function between the two CP groups.

**Discussion:** The present results suggest that body image is disrupted in people with CP who experience low back pain. The disruptions in perception were similar to those seen in people with LBP and no CP suggesting the distortions maybe more related to the presence of pain than the presence of CP.

## INTRODUCTION

Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain.<sup>1</sup> The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication and behavior as well as secondary musculoskeletal problems.<sup>1</sup> Chronic pain is a common secondary impairment in adults with CP, with prevalence rates ranging from 63% to 83%.<sup>2,3</sup> 50% report pain in more than one body location,<sup>4</sup> though the back seems to be the most commonly affected area.<sup>2</sup> Some pain is likely contributed to by the movement impairments that characterize CP, most notably musculoskeletal factors<sup>5</sup> such as soft-tissue limitations,<sup>6</sup> joint deformity,<sup>7</sup> and spasticity.<sup>8</sup> However, recent studies have suggested that the pain experience in this population is more complex than simply a reflection of impaired musculoskeletal function, particularly in adults.<sup>9</sup> It has been shown that postural asymmetry,<sup>10</sup> gross motor function,<sup>11</sup> and spasticity<sup>12</sup> are not always associated with pain in individuals with CP. Therefore, it is

important to consider the contribution from non-musculoskeletal factors to chronic pain in adults with CP.

It is now well established that the low back pain (LBP) experience is associated with multiple factors,<sup>13,14</sup> including disruption of body image<sup>15</sup> and contemporary 'predictive processing' models of how the perception of pain emerges describe generative hypotheses about the state of the body as central to the emergence of pain.<sup>16</sup> Neuroimaging studies of people with chronic low back pain (CLBP) suggest structural and functional changes in cortical areas that are thought to subserve body perception.<sup>17,18</sup> Several studies have reported that people with CLBP feel a sense of alienation and rejection of the back,<sup>19,20</sup> represent the back differently when asked to draw how the back feels to them<sup>21,22</sup> and endorse questionnaire items associated with altered perceptual awareness of the back.<sup>23–26</sup> Furthermore, psychophysical findings consistent with disruption of the mechanisms that underpin body-image<sup>27</sup> such as decreased lumbar tactile acuity,<sup>28,29</sup> problems localising sensory input,<sup>30</sup> poor graphaesthesia performance,<sup>31</sup> spatially defined tactile processing deficits,<sup>32</sup> greater lumbar repositioning error,<sup>33</sup> decreased lumbar motor precision,<sup>34,35</sup> poor trunk motor imagery performance<sup>36,37</sup> and impaired visual recognition of actions specific to the back<sup>38</sup> also appear to be features of CLBP. Moreover, some data suggested that strategies targeting disturbed body perception could improve CLBP.<sup>39–41</sup>

We were interested in exploring if people with CP who complain of low back pain also present with distorted body perception specific to the low back, particularly given the alterations in trunk posture seen in this population and suggestions from previous research that people with CP display impaired tactile discrimination and proprioception.<sup>42,43</sup> We chose to measure self-reported body perception using a questionnaire as well as a test of lumbar tactile acuity, as this has been suggested as a simple clinical assessment that might reflect somatosensory cortex reorganization specific to the body part tested.<sup>44</sup> We were particularly

interested in investigating the influence that the alterations in motor ability and trunk posture and symmetry associated with CP might have on the emergence of distorted body perception, so we also undertook similar assessments in two control groups, people with CP but no back pain and people with back pain but no CP, as well as completing a battery of tests assessing motor and function in those participants with CP and postural capacity in all three groups.

## **METHODS**

This study was a cross-sectional case-control study. Ethical approval was obtained from the institutional ethics committee of Konan Women's University (ID: 2018011). Written informed consent was obtained from all participants before study commencement. The study was conducted in accordance with the Declaration of Helsinki.

### **Participants**

Participants with CP and back pain (CP\_Pain group) and with CP but no back pain (CP\_noPain group) were recruited from an orthopaedics outpatient clinic, a child development support center, and a welfare service facility, whereas participants with back pain but no CP (Pain group) were recruited from an orthopaedics outpatient clinic. Inclusion criteria were: aged 18 years or older and cognitive level sufficient to complete the interview and questionnaires. Augmentative communication devices and information from parents and caregivers were used if it was necessary to facilitate data collection in subjects with communication difficulties. Individuals with CP who had any surgery or botulinum toxin injections in the neck, waist, and upper or lower extremities within 6 months before testing were excluded. People with low back pain but no CP who matched the age and gender of the enrolled patients with CP were recruited. Participants with back pain (CP\_Pain group and Pain group) were excluded from the study if they had signs or symptoms of nerve root pain,



evidence of specific spinal pathology (e.g., malignancy, fracture, infection, spinal canal stenosis), presented with an inflammatory, neurological or psychiatric disorder, or had undergone spinal surgery. Recruitment within the CP population was feasible with a recruitment rate of approximately 2 participants per month.

## **Procedure**

Data were collected in face-to-face interviews by using a standardized protocol that included questions about demographics, type of CP, use of medication, cognitive function, physical function, pain intensity, and psychological functioning. All participants were screened for cognitive impairment using the modified Mini-Mental State Examination.<sup>45</sup> As our sample included people with upper limb movement disorders we excluded items that required performance of upper limb tasks, namely, 1) Take this piece of paper in your right hand, fold it in half with both hands, and put it in your lap; 2) Please write a sentence; 3) Please copy this drawing. Individuals who scored 17 or higher out of a possible 25 points on the modified Mini-Mental State Examination were deemed appropriate to participate in this study.<sup>2</sup>

For the participants with CP, the level of gross motor impairment was determined by the Gross Motor Functional Classification System (GMFCS).<sup>46</sup> The level of fine motor impairment was determined using the Manual Ability Classification System (MACS).<sup>47</sup>

Sitting posture was evaluated in all participants using the sitting items of the Posture and Postural Ability Scale (PPAS).<sup>10,48</sup> The PPAS is the assessment tool designed to assess ability and quality of four kinds of postural tasks: supine, prone, sitting, and standing in adults with CP. Postural ability in sitting was rated on a 7-point ordinal scale ranging from unplaceable in an aligned posture (level 1) to, able to move into and out of position (level 7). Quality of posture in sitting was rated according to the position of the head, trunk, pelvis, legs (foot) and arms as well as weight distribution in the frontal plane and sagittal plane. Postural symmetry

and alignment gives 1 point for each item while asymmetry or deviation from midline gives 0 points. The total score of 0–6 points is calculated separately in the frontal and sagittal plane.

The PPAS has shown excellent inter-rater reliability, high internal consistency and construct validity for adults with CP.<sup>48</sup>

All participants were asked to indicate if they experienced any LBP and, if present, record on a body chart where the pain was distributed. For those experiencing LBP, pain intensity was recorded using three numerical rating scales (NRS) anchored 0 = ‘no pain’ and 10 = ‘pain as bad as you can imagine’ for present pain, average pain over the last week, and worst pain over the last week.<sup>49</sup>

Self-reported body-image of the low back region was evaluated using the Japanese-validated version of the Fremantle Back Awareness Questionnaire (FreBAQ) (0-36; higher scores indicate more disturbed perception). The FreBAQ is a validated scale used to assess back-specific body perception.<sup>15,25</sup> Participants were instructed that the questions should be answered in reference to the low back region as a whole and modifications were made to the instructions to account for pain free participants.<sup>23</sup> A five-point response scale (range: 0 = ‘never’ up to 4 = ‘always’) was used to enable quantitative assessment of any reported symptoms, the final score was obtained by summing the responses from each of the nine items.<sup>23</sup>

Measures of pain catastrophizing and kinesiophobia were completed on all participants. Pain-related catastrophizing was assessed using the Japanese version of the Pain Catastrophizing Scale (PCS)<sup>50,51</sup> (0-52; higher scores indicate more pain-related catastrophizing). Pain-related fear of movement was measured using the Japanese version of the Tampa Scale of Kinesiophobia (TSK)<sup>52,53</sup> (17-68; higher scores indicate more pain-related fear of movement).

Upon completion of all questionnaires participants were given a brief rest before TPD thresholds were determined. The lumbar TPD threshold was measured bilaterally according

to methods described by Moberg<sup>54</sup> and Luomajoki and Moseley<sup>55</sup>. A plastic caliper ruler with a precision of 1 mm was applied until the very first blanching of the skin. Subjects were instructed to say “one” when they perceived one point and “two” when they perceived two. Calipers were applied initially with 0 mm between the two tips, and the distance between the tips was increased by 5 mm increments until the subject was able to perceive two distinct points - this was considered a practice run and data from this run was not used for analysis. The calipers were then applied in a descending order of 5 mm increments starting from 100 mm until the two points were felt as one. A final ascending run was completed starting from 10 mm. Values for one descending run and one ascending run were averaged to obtain the final threshold value.<sup>55</sup> When testing over the back the calipers were aligned perpendicular with the spine and were centered on the transverse process of the most severe pain level for the CP\_Pain group and the Pain group, or the L3 level in the CP\_noPain group.<sup>55</sup> As there is some evidence that people with CP have a general deficit in tactile acuity<sup>56</sup> the cheek was used as a non-painful control site and testing was conducted according to the method described by Riquelme et al.,<sup>57</sup> with the caliper centered on the midpoint between the corner of the mouth and the ear canal. The testing protocol was identical to that described above for the low back except that the descending run commenced with a distance of 30 mm. For the two back pain groups lumbar spine TPD data from the most painful side only are reported. For the pain free group values from the left and right side were averaged for analysis. For all groups data from the left and right cheek were combined for analysis.

After informed consent was obtained from the participants, one investigator (HY) collected clinical and demographic details and administered all tests and questionnaires in the order outlined above. We checked regularly with participants during testing to gauge their level of fatigue and took breaks as needed. The total time of testing was between 40 and 60 minutes

depending on the number of breaks taken. All participants were able to complete all assessments in the single testing session and the protocol appears feasible and acceptable in the populations tested.

### **Sample Size**

The study was regarded as a preliminary investigation and no formal power calculation was undertaken. We planned to recruit between 12 and 15 participants per group based on the recommendation that preliminary studies for which little data exists to inform a formal sample size calculation should seek to recruit around 12 participants per group.<sup>58</sup>

### **Statistical Analysis**

Statistical analysis was performed using the Statistical Package for Social Sciences Version 25.0 (IBM SPSS Statistics for MAC, Version 25.0. Armonk, NY: IBM Corp.). The GMFCS and MACS were classified into two groups: a mild group (levels 1 to 3) and a severe group (levels 4 and 5). The proportion of participants classified into each group were compared between the CP\_Pain group and the CP\_noPain group using Fisher's exact test. Age, Mini-Mental State Examination, postural ability in sitting, the quality of sitting posture in the frontal and sagittal view, FreBAQ, TSK, PCS and TPD threshold of the cheek and low back were compared between three groups (the CP\_Pain group versus the CP\_noPain group versus Pain group) using the Kruskal-Wallis test and the Steel-Dwass test as post-hoc analyses. The proportion of female participants was compared between the three groups using Fisher's exact test. Difference in pain intensity and pain duration between the CP\_Pain group and the Pain group was compared using the Mann-Whitney U test. To help with interpretation of the results from this preliminary investigation effect sizes were calculated based on  $\eta^2$  (A large effect was defined as  $> 0.14$ , a moderate effect between 0.06 and 0.14 and a small effect  $< 0.06$ ),  $V$  (A large effect was defined as  $> 0.5$ , a moderate effect between 0.3 and 0.5 and a

small effect  $< 0.3$ ) or  $r$  (A large effect was defined as  $> 0.5$ , a moderate effect between 0.3 and 0.5 and a small effect  $< 0.3$ ) where appropriate (see table 2 for details). A univariate correlation was performed examining the relationships between the FreBAQ total score and present pain. The data were presented as medians with interquartile ranges. All p-values were adjusted using the Benjamini-Hochberg procedure for multiple tests. FDR-adjusted p-values are reported.

## RESULTS

### Clinical characteristics

Forty-five individuals participated in this study: fifteen adults with CP and LBP (eight females; mean age = 40.1, SD = 14.5), fifteen adults with CP and no LBP (nine females; mean age = 43.7, SD = 17.0), and fifteen age-matched participants with LBP but no CP (eight females; mean age = 41.5, SD = 17.7). Of the CP\_Pain group, four participants took nonsteroidal anti-inflammatory drugs and seven participants took anti-spastic medications. Amongst the CP\_noPain group, three participants took anti-spastic medications. In the Pain group, six participants took nonsteroidal anti-inflammatory drugs. None of the subjects in the study required augmentative communication devices. One person in the CP\_noPain group was using a hearing aid. All participants back pain was classified as non-specific low back pain by the assessing medical doctor. Individual demographic and clinical characteristics of the participants with CP are shown in Table 1. When investigating motor function we found no significant differences in GMFCS ( $p = 1.0$ ) and MACS ( $p = 1.0$ ) between the CP\_Pain group and the CP\_noPain group. Group level data for all participants can be found in Table 2. There were no significant differences in age ( $p = 0.98$ ) gender ( $p = 0.98$ ) and cognitive function ( $p = 0.07$ ) between the three groups. There were significant differences in the level of postural ability in sitting ( $p = 0.003$ ) and quality of posture in the frontal plane ( $p =$

0.0007) and sagittal plane ( $p < 0.0001$ ). For all three analyses we found no difference between the two CP groups, though the Pain group demonstrated significantly better function (all  $p < 0.05$ ) and postural form (frontal plane all  $p < 0.01$ ; sagittal plane all  $p < 0.01$ ) than the two CP groups. The effect sizes were large for the level of postural ability in sitting ( $\eta^2 = 0.30$ ), quality of posture in the frontal plane ( $\eta^2 = 0.39$ ) and sagittal plane ( $\eta^2 = 0.42$ ) and moderate for the Mini-Mental State Examination ( $\eta^2 = 0.14$ ) and small for the age ( $\eta^2 = 0.007$ ) and gender ( $V = 0.06$ ).

The pain related characteristics of all participants are summarized in Table 2.

### **Pain intensity**

There were no significant differences in pain intensity (present pain  $p = 0.98$ ; average pain over the last week  $p = 0.45$ ; worst pain over the last week  $p = 0.51$ ) or pain duration ( $p = 0.66$ ) between the CP\_Pain group and the Pain group. The effect sizes were small for the present pain ( $r = 0.00$ ), average pain over the last week ( $r = 0.20$ ), and worst pain over the last week ( $r = 0.17$ ).

### **FreBAQ**

Significant differences between the three groups were found for FreBAQ scores (CP\_Pain group: 14.0; CP\_noPain group: 4.0; Pain group: 12.0,  $p < 0.0001$ ). Post hoc analysis showed that there was no significant difference in FreBAQ between the CP\_Pain group and Pain group ( $p = 0.53$ ), though both had significantly greater levels of self-reported body perception disturbance than the CP\_noPain group (all  $p < 0.01$ ) (Figure 1). The effect size was large for the FreBAQ ( $\eta^2 = 0.47$ ). The FreBAQ score was significantly correlated with present pain ( $\rho = 0.60$ ,  $p < 0.01$ ).

### **TPD thresholds**

No significant differences between the three groups were found for the TPD threshold at the cheek ( $p = 0.98$ ). Analysis of the TPD threshold over the low back found significant differences between the three groups ( $p = 0.02$ ). Post hoc analysis showed that there was no significant difference in lumbar TPD between the CP\_noPain group and Pain group ( $p = 0.21$ ), though the CP\_Pain group had significantly poorer tactile acuity than the CP\_noPain group ( $P = 0.01$ ) (Figure 1). The effect sizes were large for the TPD threshold in the low back ( $\eta^2 = 0.19$ ) and small for the TPD threshold in the cheek ( $\eta^2 = 0.002$ ).

### **PCS and TSK**

Analysis of the PCS and TSK scores demonstrated the same results. A main effect for group was seen for both PCS ( $p < 0.0001$ ) and TSK ( $p = 0.007$ ). Post hoc analysis showed that there was no significant difference in PCS ( $p = 0.07$ ), or TSK ( $p = 0.74$ ) between the CP\_Pain group and Pain group, though both had significantly greater levels of kinesiophobia and pain related catastrophizing than the CP\_noPain group (all  $p < 0.05$ ). The effect size was large for the PCS ( $\eta^2 = 0.56$ ) and TSK ( $\eta^2 = 0.25$ ).

### **DISCUSSION**

The main aim of the present study was to investigate if people with CP who complain of low back pain present with distorted body perception specific to the low back. We found that people with CP and low back pain endorsed questionnaire items related to distorted body image more frequently than people with CP who do not report low back pain. Furthermore the score on the FreBAQ questionnaire in the CP\_Pain population was no different to that seen in a matched group with low back pain and no CP and very similar to the results of previous investigations that have used the FreBAQ to assess body perception in general low

back pain populations.<sup>15,23,25,26</sup> This builds on work demonstrating that lumbopelvic self-perception is impaired in people with low back pain compared to matched control groups<sup>23,24,26,59,60</sup> and extends this finding to include people with low back pain and CP.

We were also interested in investigating the confounding effect postural and motor impairments might have on self-perception of the back in the CP population. We found no difference in gross or fine motor impairments or sitting posture function and quality between the CP\_Pain group and CP\_noPain group. These findings support previous studies that showed that movement impairments and sitting posture were not specific factors affecting LBP in individual with CP,<sup>10-12</sup> as well as suggesting that disrupted self-perception is related to the presence of back pain not the presence of CP. These results indicate that it is perceived rather than actual trunk distortion that is important in the genesis of low back pain in this population. In confirmation of this interpretation we found that the two low back pain groups (Pain and CP\_Pain) reported similar levels of lumbopelvic self-perception despite presenting with significant differences in sitting posture alignment. Together these findings point to a dissociation between perceived and actual body distortion and point to a greater importance of the perceived body in contributing to the pain experience.

We also investigated TPD thresholds over the lumbar spine, as tactile acuity is thought to represent a reasonable clinical correlate of the representation of that body part in primary somatosensory cortex<sup>44</sup> and as such, possibly provides insight into one of the central nervous system mechanisms that underpin body perception. Similar to previous research<sup>22,55</sup> we found that the precision of tactile discrimination is poorer over the lumbar spine in people with back pain compared to people without, at least in the CP population. Moreover, this impairment seems to be specific to the painful area as we found no difference in TPD thresholds over the cheek, a finding also consistent with previous low back pain research.<sup>61</sup> Contrary to previous research, we did not find a difference in TPD thresholds between the CP\_noPain group and



the Pain group. This might be a reflection of the small sample size or the fact that lumbar TPD is somewhat impaired in people with CP even in the absence of back pain. Some support for this idea can be seen in data from previous investigations of TPD over the lumbar spine in healthy participants which suggest a normative value somewhat less than reported for the CP\_noPain group in this study,<sup>62</sup> though a different measure of central tendency is reported.

These results may provide insight into potentially important factors contributing to the back pain experience that have not been previously investigated in adults with CP. Contemporary models of perception highlight the importance of cognitive modulation of sensory information in the emergence of perception.<sup>63</sup> Prior information about internal and external states is used to generate predictions about the causes of sensory information and perceptions, such as pain, can be viewed as the brain's best fitting model for the information entering the senses weighed against predictions about the causes of the information.<sup>63-65</sup> One important implication of this process for musculoskeletal pain problems is that perception of pain with action will always be influenced by factors that drive us to predict pain with action.<sup>16</sup> This implicates body representation as central to the emergence of pain, as prior beliefs about the state of the body and the risk to the body associated with a particular movement will create stronger and more precise predictions of pain and increase the likelihood of the emergence of pain with action.<sup>66</sup> Furthermore, updating of predictions away from one of pain towards one of a lower expectation of pain are partly driven by prediction error,<sup>65,66</sup> that is, receiving sensory inputs that diverge from the expectation of pain, such as non-noxious sensory inputs with action. However, divergent sensory information from the body that is noisy, ambiguous and imprecise can be 'explained away' and will less likely lead to an updating of pain expectation. Riquelme (2013)<sup>57</sup> reported that the increase of non-noxious somatosensory experiences provided by somatosensory therapy may have effects on pain processing and

may reduce pain perception in CP individuals, and the data presented here offer further support for similar approaches, though formal testing of these ideas is clearly needed.

Our results also show that catastrophisation about pain and kinesiophobia in the CP\_Pain group was significantly higher than the CP\_noPain group, but not significantly different from the Pain group. This finding may indicate that catastrophisation about pain<sup>67</sup> and kinesiophobia,<sup>68</sup> which are considered to contribute to the pain experience in the general population with LBP, also contribute to the pain experience in adults with cerebral palsy with LBP, though more longitudinal data are needed. This association between catastrophisation and pain related clinical status has been noted previously in people with CP<sup>69</sup> and cognitive-behavioural models of care may be useful for the management of low back pain in people with CP.<sup>70</sup> This would seem particularly important in this population as previous work has shown that adults with CP reported relatively high rates of use of more passive and marginally effective treatments for pain such as medications, modalities and massage.<sup>2</sup>

Several limitations of our study should be considered. Participants were not consecutively sampled which introduces some selection bias and the sample size is relatively small so it is possible that we lacked power to detect some differences between groups. Data collection was not blinded, while this is likely to introduce minimal bias for the self-reported measures, the assessments of tactile acuity and motor and postural function are potentially subject to some measurement bias. Furthermore, lumbar TPD thresholds were not measured at the exact same site for all participants as it depended on the distribution of back pain, though we know of no data that suggests TPD thresholds differ significantly within the lumbar spine. Finally, the study was cross-sectional which precludes any clear conclusions being made regarding the causal relationship between body perception and back pain in people with CP. Despite

these limitations, the data support previous findings and expand the study of chronic LBP in adults with CP.

## **CONCLUSION**

The findings of the present study suggest that body image is disrupted in people with CP who experience low back pain. Interestingly, though perception of the trunk was disrupted, we found no difference in actual trunk posture between CP patients with and without back pain.

This dissociation between perceived and actual body distortion was confirmed by comparison between the two back pain populations. Both back pain groups reported the same level of perceived disruption despite large differences in actual trunk posture. This suggests the perceptual distortions maybe more related to the presence of pain than the presence of CP and any associated postural abnormalities. Disrupted body perception has been suggested as a target for treatment for numerous musculoskeletal pain problems and these ideas may be worth testing in people with CP who experience low back pain.

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## **CONFLICTS OF INTEREST**

The authors state that they don't have any conflict of interest.

## REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl.* 2007 Feb;109:8-14.
2. Hirsh AT, Kratz AL, Engel JM, Jensen MP. Survey results of pain treatments in adults with cerebral palsy. *Am J Phys Med Rehabil.* 2011 Mar;90(3):207-16.
3. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Physical and mental components of health-related quality of life and musculoskeletal pain sites over seven years in adults with spastic cerebral palsy. *J Rehabil Med.* 2011 Apr;43(5):382-7.
4. Andersson C, Mattsson E. Adults with cerebral palsy: a survey describing problems, needs, and resources, with special emphasis on locomotion. *Dev Med Child Neurol.* 2001 Feb;43(2):76-82.
5. Murphy KP. Cerebral palsy lifetime care - four musculoskeletal conditions. *Dev Med Child Neurol.* 2009 Oct;51 Suppl 4:30-7.
6. Sheehan FT, Babushkina A, Alter KE. Kinematic determinants of anterior knee pain in cerebral palsy: a case-control study. *Arch Phys Med Rehabil.* 2012 Aug;93(8):1431-40.
7. Flynn JM, Miller F. Management of hip disorders in patients with cerebral palsy. *J Am Acad Orthop Surg.* 2002 May-Jun;10(3):198-209.
8. Lundy CT, Doherty GM, Fairhurst CB. Botulinum toxin type A injections can be an effective treatment for pain in children with hip spasms and cerebral palsy. *Dev Med Child Neurol.* 2009 Sep;51(9):705-10.
9. Vogtle LK. Pain in adults with cerebral palsy: impact and solutions. *Dev Med Child Neurol.* 2009 Oct;51 Suppl 4:113-21.
10. Rodby-Bousquet E, Czuba T, Hägglund G, Westbom L. Postural asymmetries in young adults with cerebral palsy. *Dev Med Child Neurol.* 2013 Nov;55(11):1009-15.

11. Doralp S, Bartlett DJ. The prevalence, distribution, and effect of pain among adolescents with cerebral palsy. *Pediatr Phys Ther.* 2010 Spring;22(1):26-33.
12. Baker JA, Pereira G. The efficacy of Botulinum Toxin A for spasticity and pain in adults: a systematic review and meta-analysis using the Grades of Recommendation, Assessment, Development and Evaluation approach. *Clin Rehabil.* 2013 Dec;27(12):1084-96.
13. Rabey M, Smith A, Beales D, Slater H, O'Sullivan P. Differing Psychologically Derived Clusters in People With Chronic Low Back Pain are Associated With Different Multidimensional Profiles. *Clin J Pain.* 2016 Dec;32(12):1015-1027.
14. Nijs J, Goubert D, Ickmans K. Recognition and Treatment of Central Sensitization in Chronic Pain Patients: Not Limited to Specialized Care. *J Orthop Sports Phys Ther.* 2016 Dec;46(12):1024-1028.
15. Wand BM, Catley MJ, Rabey MI, O'Sullivan PB, O'Connell NE, Smith AJ. Disrupted Self-Perception in People With Chronic Low Back Pain. Further Evaluation of the Fremantle Back Awareness Questionnaire. *J Pain.* 2016 Sep;17(9):1001-12.
16. Ongaro G, Kaptchuk TJ. Symptom perception, placebo effects, and the Bayesian brain. *Pain.* 2019 Jan;160(1):1-4.
17. Kregel J, Meeus M, Malfliet A, et al. Structural and functional brain abnormalities in chronic low back pain: a systematic review. *Semin Arthritis Rheum.* 2015 Oct;45(2):229-37.
18. Wand BM, Parkitny L, O'Connell NE, et al. Cortical changes in chronic low back pain: current state of the art and implications for clinical practice. *Man Ther.* 2011 Feb;16(1):15-20.

- Accepted Article
19. Osborn M, Smith JA. Living with a body separate from the self. The experience of the body in chronic benign low back pain: an interpretative phenomenological analysis. *Scand J Caring Sci.* 2006 Jun;20(2):216-22.
  20. Crowe M, Whitehead L, Gagan MJ, Baxter GD, Pankhurst A, Valledor V. Listening to the body and talking to myself - the impact of chronic lower back pain: a qualitative study. *Int J Nurs Stud.* 2010 May;47(5):586-92.
  21. Moseley GL. I can't find it! Distorted body image and tactile dysfunction in patients with chronic back pain. *Pain.* 2008 Nov 15;140(1):239-43.
  22. Nishigami T, Mibu A, Osumi M, et al. Are tactile acuity and clinical symptoms related to differences in perceived body image in patients with chronic nonspecific lower back pain? *Man Ther.* 2015 Feb;20(1):63-7.
  23. Wand BM, James M, Abbaszadeh S, et al. Assessing self-perception in patients with chronic low back pain: development of a back-specific body-perception questionnaire. *J Back Musculoskelet Rehabil.* 2014;27(4):463-73.
  24. Beales D, Lutz A, Thompson J, Wand BM, O'Sullivan P. Disturbed body perception, reduced sleep, and kinesiophobia in subjects with pregnancy-related persistent lumbopelvic pain and moderate levels of disability: An exploratory study. *Man Ther.* 2016 Feb;21:69-75.
  25. Nishigami T, Mibu A, Tanaka K, et al. Validation of the Japanese Version of the Fremantle Back Awareness Questionnaire in Patients with Low Back Pain. *Pain Pract.* 2018 Feb;18(2):170-179.
  26. Janssens L, Goossens N, Wand BM, Pijnenburg M, Thys T, Brumagne S. The development of the Dutch version of the Fremantle Back Awareness Questionnaire. *Musculoskelet Sci Pract.* 2017Dec;32:84-91.

27. Tsay A, Allen TJ, Proske U, Giummarra MJ. Sensing the body in chronic pain: a review of psychophysical studies implicating altered body representation. *Neurosci Biobehav Rev.* 2015 May;52:221-32.
28. Catley MJ, O'Connell NE, Berryman C, Ayhan FF, Moseley GL. Is tactile acuity altered in people with chronic pain? A systematic review and meta-analysis. *J Pain.* 2014 Oct;15(10):985-1000.
29. Adamczyk W, Luedtke K, Saulicz E. Lumbar Tactile Acuity in Patients With Low Back Pain and Healthy Controls: Systematic Review and Meta-Analysis. *Clin J Pain.* 2017 Mar 21.
30. Wand BM, Keeves J, Bourgoin C, et al. Mislocalization of sensory information in people with chronic low back pain: a preliminary investigation. *Clin J Pain.* 2013 Aug;29(8):737-43.
31. Wand BM, Di Pietro F, George P, O'Connell NE. Tactile thresholds are preserved yet complex sensory function is impaired over the lumbar spine of chronic non-specific low back pain patients: a preliminary investigation. *Physiotherapy.* 2010 Dec;96(4):317-23.
32. Moseley GL, Gallace A, Iannetti GD. Spatially defined modulation of skin temperature and hand ownership of both hands in patients with unilateral complex regional pain syndrome. *Brain.* 2012 Dec;135(Pt 12):3676-86.
33. Laird RA, Gilbert J, Kent P, Keating JL. Comparing lumbo-pelvic kinematics in people with and without back pain: a systematic review and meta-analysis. *BMC Musculoskelet Disord.* 2014 Jul 10;15:229.
34. Hadizadeh M, Mousavi SJ, Sedaghatnejad E, Talebian S, Parnianpour M. The effect of chronic low back pain on trunk accuracy in a multidirectional isometric tracking task. *Spine (Phila Pa 1976).* 2014 Dec 15;39(26):E1608-15.

- Accepted Article
35. Pranata A, Perraton L, El-Ansary D, et al. Lumbar extensor muscle force control is associated with disability in people with chronic low back pain. *Clin Biomech (Bristol, Avon)*. 2017 Jul;46:46-51.
  36. Bray H, Moseley GL. Disrupted working body schema of the trunk in people with back pain. *Br J Sports Med*. 2011 Mar;45(3):168-73.
  37. Bowering KJ, Butler DS, Fulton IJ, Moseley GL. Motor imagery in people with a history of back pain, current back pain, both, or neither. *Clin J Pain*. 2014 Dec;30(12):1070-5.
  38. de Lussanet MH, Behrendt F, Puta C, et al. A body-part-specific impairment in the visual recognition of actions in chronic pain patients. *Pain*. 2012 Jul;153(7):1459-66.
  39. Wand BM, O'Connell NE, Di Pietro F, Bulsara M. Managing chronic nonspecific low back pain with a sensorimotor retraining approach: exploratory multiple-baseline study of 3 participants. *Phys Ther*. 2011 Apr;91(4):535-46.
  40. Wand BM, Tulloch VM, George PJ, et al. Seeing it helps: movement-related back pain is reduced by visualization of the back during movement. *Clin J Pain*. 2012 Sep;28(7):602-8.
  41. Wand BM, Abbaszadeh S, Smith AJ, Catley MJ, Moseley GL. Acupuncture applied as a sensory discrimination training tool decreases movement-related pain in patients with chronic low back pain more than acupuncture alone: a randomized cross-over experiment. *Br J Sports Med*. 2013 Nov;47(17):1085-9.
  42. Wingert JR, Burton H, Sinclair RJ, Brunstrom JE, Damiano DL. Tactile sensory abilities in cerebral palsy: deficits in roughness and object discrimination. *Dev Med Child Neurol*. 2008 Nov;50(11):832-8.
  43. Wingert JR, Burton H, Sinclair RJ, Brunstrom JE, Damiano DL. Joint-position sense and kinesthesia in cerebral palsy. *Arch Phys Med Rehabil*. 2009 Mar;90(3):447-53.



44. Moseley GL, Flor H. Targeting cortical representations in the treatment of chronic pain: a review. *Neurorehabil Neural Repair*. 2012 Jul-Aug;26(6):646-52.
45. Folstein MF, Folstein SE, McHugh PR. "Mini-mental state". A practical method for grading the cognitive state of patients for the clinician. *J Psychiatr Res*. 1975 Nov;12(3):189-98.
46. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol*. 1997 Apr;39(4):214-23.
47. Eliasson AC, Krumlinde-Sundholm L, Rösblad B, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Dev Med Child Neurol*. 2006 Jul;48(7):549-54.
48. Rodby-Bousquet E, Agústsson A, Jónsdóttir G, Czuba T, Johansson AC, Hägglund G. Interrater reliability and construct validity of the Posture and Postural Ability Scale in adults with cerebral palsy in supine, prone, sitting and standing positions. *Clin Rehabil*. 2014 Jan;28(1):82-90.
49. Von Korff M, Jensen MP, Karoly P. Assessing global pain severity by self-report in clinical and health services research. *Spine (Phila Pa 1976)*. 2000 Dec 15;25(24):3140-51.
50. Sullivan MJ, Bishop SR, Pivik J. The pain catastrophizing scale: development and validation. *Psychol Assess*. 1995;7:524-32.
51. Matsuoka H, Sakano Y. Assessment of cognitive aspect of pain: Development, reliability, and validation of Japanese version of Pain Catastrophizing Scale. *Jpn J Psychosom Med*. 2007; 47:95–102.

52. Vlaeyen JW, Kole-Snijders AM, Boeren RG, van Eek H. Fear of movement/(re)injury in chronic low back pain and its relation to behavioral performance. *Pain*. 1995 Sep;62(3):363-72.
53. Kikuchi N, Matsudaira K, Sawada T, Oka H. Psychometric properties of the Japanese version of the Tampa Scale for Kinesiophobia (TSK-J) in patients with whiplash neck injury pain and/or low back pain. *J Orthop Sci*. 2015 Nov;20(6):985-92.
54. Moberg E. Two-point discrimination test. A valuable part of hand surgical rehabilitation, e.g. in tetraplegia. *Scand J Rehabil Med*. 1990;22(3):127-34.
55. Luomajoki H, Moseley GL. Tactile acuity and lumbopelvic motor control in patients with back pain and healthy controls. *Br J Sports Med*. 2011 Apr;45(5):437-40.
56. McLaughlin JF, Felix SD, Nowbar S, Ferrel A, Bjornson K, Hays RM. Lower extremity sensory function in children with cerebral palsy. *Pediatr Rehabil*. 2005 Jan-Mar;8(1):45-52.
57. Riquelme I, Zamorano A, Montoya P. Reduction of pain sensitivity after somatosensory therapy in adults with cerebral palsy. *Front Hum Neurosci*. 2013 Jun 24;7:276.
58. Julious SA. Sample size of 12 per group rule of thumb for a pilot study. *Pharm Stat*. 2005;4:287–291.
59. Wand BM, Elliott RL, Sawyer AE, et al. Disrupted body-image and pregnancy-related lumbopelvic pain. A preliminary investigation. *Musculoskelet Sci Pract*. 2017 Aug;30:49-55.
60. Ehrenbrusthoff K, Ryan CG, Grüneberg C, Wand BM, Martin DJ. The translation, validity and reliability of the German version of the Fremantle Back Awareness Questionnaire. *PLoS One*. 2018 Oct 4;13(10):e0205244.

61. Peters ML, Schmidt AJ. A comparison of two-point discrimination threshold of tactual, non-painful stimuli between chronic low back pain patients and controls. *Pain*. 1991 Jan;44(1):57-60.
62. Wand BM, Catley MJ, Luomajoki HA, et al. Lumbar tactile acuity is near identical between sides in healthy pain-free participants. *Man Ther*. 2014 Oct;19(5):504-7.
63. Friston K. The free-energy principle: a unified brain theory? *Nat Rev Neurosci*. 2010 Feb;11(2):127-38.
64. Van den Bergh BRH, van den Heuvel MI, Lahti M, et al. Prenatal developmental origins of behavior and mental health: The influence of maternal stress in pregnancy. *Neurosci Biobehav Rev*. 2017 Jul 28. pii: S0149-7634(16)30734-5.
65. Wiech K. Deconstructing the sensation of pain: The influence of cognitive processes on pain perception. *Science*. 2016 Nov 4;354(6312):584-587.
66. Vlaeyen JWS, Maher CG, Wiech K, et al. Low back pain. *Nat Rev Dis Primers*. 2018 Dec13;4(1):52.
67. Wertli MM, Eugster R, Held U, Steurer J, Kofmehl R, Weiser S. Catastrophizing-a prognostic factor for outcome in patients with low back pain: a systematic review. *Spine J*. 2014 Nov 1;14(11):2639-57.
68. Crombez G, Vlaeyen JW, Heuts PH, Lysens R. Pain-related fear is more disabling than pain itself: evidence on the role of pain-related fear in chronic back pain disability. *Pain*. 1999 Mar;80(1-2):329-39.
69. Engel JM, Schwartz L, Jensen MP, Johnson DR. Pain in cerebral palsy: the relation of coping strategies to adjustment. *Pain*. 2000 Dec 1;88(3):225-30.
70. Engel JM, Jensen MP, Schwartz L. Coping with chronic pain associated with cerebral palsy. *Occup Ther Int*. 2006;13(4):224-33.

Table 1 Demographics of the participating adults with cerebral palsy

	ID	Sex	Age	Type of cerebral palsy	GMFCS	MACS	Epilepsy
CP_Pain							
	1	M	48	BS	4	5	Yes
	2	M	73	BS	5	5	No
	3	M	27	BS	4	3	No
	4	M	43	BS	5	4	Yes
	5	M	21	D	5	5	No
	6	M	23	BS	2	2	No
	7	F	48	D	4	5	No
	8	F	34	US	2	2	No
	9	M	54	D	3	3	No
	10	F	57	D	1	2	No
	11	F	47	BS	4	5	No
	12	F	27	D	2	2	No
	13	F	32	BS	2	1	No
	14	F	29	BS	1	1	No
	15	F	38	BS	2	1	No
CP_noPain							
	1	F	61	D	5	4	No
	2	F	31	BS	3	2	No
	3	M	22	US	1	2	No
	4	F	33	US	2	3	Yes
	5	F	59	D	4	4	No
	6	M	58	D	5	4	No
	7	F	68	BS	4	3	No
	8	M	19	US	1	2	No
	9	M	50	D	3	3	No
	10	F	55	D	3	4	No
	11	M	22	US	1	3	No
	12	F	26	BS	5	5	No
	13	F	40	US	1	1	No
	14	M	48	D	5	5	Yes
	15	F	63	D	5	5	No

M, male; F, female; BS, bilateral spastic; US, unilateral spastic; D, dyskinetic

Table 2 Sample characteristics, pain parameters, FreBAQ, TPD threshold, PCS, TSK, and sitting postural ability

	CP_Pain (n = 15)	CP_noPain (n = 15)	Pain (n = 15)	Benjamini-Hochberg adjusted P value	Effect size
Mean Age (years) (SD)	40.1 (14.5)	43.7 (17.0)	41.5 (17.7)	0.98	$\eta^2 = 0.007$
Gender (female)	8	9	8	0.98	$V = 0.06$
modified Mini-Mental State Examination	23.0 (22.0 – 25.0)	25.0 (22.0 – 25.0)	25.0 (25.0 – 25.0)	0.07	$\eta^2 = 0.14$
NRS (present pain)	4.0 (2.0 – 5.0)	—	3.0 (2.0 – 6.0)	0.98	$r = 0.00$
NRS (average pain over the last week)	4.0 (3.0 – 6.0)	—	3.0 (2.0 – 4.0)	0.45	$r = 0.20$
NRS (worst pain over the last week)	5.0 (5.0 – 7.0)	—	5.0 (3.0 – 6.0)	0.51	$r = 0.17$
Pain duration (months)	72.0 (36.0 – 240.0)	—	96.0 (60.0 – 240.0)	0.66	$r = 0.12$
FreBAQ	14.0 (11.0 – 20.0) **	4.0 (2.0 – 6.0)	12.0 (8.0 – 15.0) **	< 0.0001	$\eta^2 = 0.47$
TPD threshold in the cheek	15.0 (11.2 – 17.5)	15.0 (11.2 – 17.5)	15.0 (12.5 – 18.7)	0.98	$\eta^2 = 0.002$
TPD threshold in the low back	65.0 (55.0 – 72.5) *	50.0 (47.5 – 61.2)	52.5 (50.0 – 67.5)	0.02	$\eta^2 = 0.19$
PCS	30.0 (24.0 – 40.0) **	6.0 (3.0 – 15.0)	26.0 (16.0 – 31.0) **	< 0.0001	$\eta^2 = 0.56$
TSK	40.0 (36.0 – 47.0) **	29.0 (27.0 – 37.0)	39.0 (35.0 – 42.0) *	0.007	$\eta^2 = 0.25$
Level of postural ability in sitting	7.0 (2.0 – 7.0) †	6.0 (2.0 – 7.0) ‡	7.0 (7.0 – 7.0)	0.003	$\eta^2 = 0.30$
Quality of posture in frontal view	3.0 (1.0 – 6.0) ‡	2.0 (0 – 6.0) ‡	6.0 (6.0 – 6.0)	0.0007	$\eta^2 = 0.39$
Quality of posture in sagittal view	3.0 (1.0 – 5.0) ‡	2.0 (0 – 6.0) ‡	6.0 (6.0 – 6.0)	< 0.0001	$\eta^2 = 0.42$

\* Differences are significant ( $p < .05$ ) compared with CP\_noPain group.

\*\* Differences are significant ( $p < .01$ ) compared with CP\_noPain group.

† Differences are significant ( $p < .05$ ) compared with Pain group.

‡ Differences are significant ( $p < .01$ ) compared with Pain group.

NRS: numerical rating scales; FreBAQ: Fremantle Back Awareness Questionnaire; TPD: two-point discrimination; PCS: Pain Catastrophizing Scale; TSK: Tampa Scale of Kinesiophobia, effect sizes ( $\eta^2$ : A large effect was defined as  $> 0.14$ , a moderate effect between 0.06 and 0.14 and a small effect  $< 0.06$ .  $V$  and  $r$ : A large effect was defined as  $> 0.5$ , a moderate effect between 0.3 and 0.5 and a small effect  $< 0.3$ .)

