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Age-Related Changes of Pain Experience in Cerebral Palsy and Healthy Individuals

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Abstract

Objective. Pain is a serious problem for many individuals with cerebral palsy (CP). Pain and injury in early life may cause long-term changes in somatosensory and pain processing. Nevertheless, no information exists regarding the influence of age on pain reports and touch sensitivity among persons with CP or the influence of age on the quality of life in individuals with CP.

Design. The present cross-sectional study investigated pain characteristics, touch sensitivity, and quality of life in 86 individuals with CP and 115 healthy volunteers. Participants were grouped by age in children (6–10 years), adolescents (11–17 years), and young adults (18–30 years). Touch sensitivity at different body locations were tested by using von Frey monofilaments. Data about pain and quality of life were obtained from a semi-structured interview and questionnaires.

Results. Participants with CP reported more pain as well as more reduced touch sensitivity and quality of life than healthy controls. Neither pain reports nor touch sensitivity or quality of life were influenced by age in CP, whereas significant age-related changes were observed in healthy participants. Multiple regression analyses also showed that age was the best predictor of current pain intensity in healthy controls but not in individuals with CP.

Conclusion. These findings emphasize the importance of considering the presence of pain at very early ages in CP. Furthermore, these results provide clinicians and researchers with a new age-related psychosocial and psychophysiological perspective to investigate the mechanisms that could be involved in the presence and maintenance of pain in this population.

Key Words. Cerebral Palsy; Quality of Life; Child Disability; Adolescence

Introduction

Recently, there has been a growing recognition that pain represents a serious problem for many children and adolescents with cerebral palsy (CP). Indeed, pain is perceived as a secondary problem in 47–78% of children and adolescents with CP [1–4]. Pain in CP is frequently described as moderate and is located mostly in the abdomen and in the musculoskeletal system [2,3]. Pain also seems to limit daily activities and satisfaction with life in children and adolescents with CP [1,4]. Moreover, it has been suggested that pain is one of the most significant factors influencing the quality of life in children with CP [5]. However, although children with CP have significantly lower health status concerning physical function and bodily pain than age- and sex-matched typically developing children, they have similar quality of life [6]. To date, research on pain in CP has been oriented primarily to the analysis of its influence on the quality of life without considering age-matched healthy controls. Such a comparison group would allow for the control of environmental factors that could influence pain and quality of life.

Pain and injury in early life may cause long-term changes in somatosensory and pain processing [7]. Therefore, it seems plausible that the developing nervous system of children and adolescents with brain damage may respond differently to pain and somatosensory information in early and later life. In this sense, several studies have shown that children with CP had poorer touch sensitivity, stereognosis, and proprioception compared with healthy children [8–10]. However, despite age-related differences in many chronic pain conditions, virtually nothing is known about pain and touch sensitivity in children and adolescents with CP.

The present study included three age groups (children, adolescents, and adults) of individuals with CP and healthy controls to examine pain, touch sensitivity, and quality of life. Based on previous work indicating that pain may play a key role in children with CP [1–6], we

hypothesized that younger participants with CP would show differences in pain and somatosensory perception and that differences between individuals with CP and healthy controls would appear in both younger and older subjects. Moreover, consistent with previous results showing that pain represents the most significant factor influencing quality of life in CP [5], we also explored age-related changes on quality of life and other factors affecting pain intensity in individuals with CP and healthy controls.

Methods

Participants

One hundred fifty persons with CP (50 children aged 6–10 years, 50 adolescents aged 11–17 years, and 50 young adults aged 18–30 years) and 150 healthy persons were initially contacted through a letter explaining the details of the study. Participants with CP were identified by physiotherapists from specialized centers for CP, whereas non-CP participants were recruited by asking for volunteers in educational centers, from primary schools to universities. Inclusion criteria were an age of 6–30 years and a cognitive level that allowed for the understanding of simple yes/no questions.

Eighty six subjects with CP (27 children, 24 adolescents, and 35 young adults) and 115 healthy subjects (34 children, 35 adolescents, and 46 young adults) agreed to participate in the study. For participants under the age of 18, permission and written informed consent from the center and from their parents or legal tutors were obtained. Healthy adults provided written informed consent and verbalized willingness to participate. In the case of adults with CP, their parents or legal tutors provided written informed consent, and participants verbalized willingness to participate. The study was approved by the Ethics Committee of the Regional Government of the Balearic Islands.

Cerebral Palsy Assessment

Information about age, type of cerebral palsy, and orthopedic impairments in CP participants was obtained from their health history. The cognitive level of participants with CP was determined by psychologists from the specialized centers by using standardized neuropsychological tests (Wechsler Adult Intelligence Scale-Revised, Wechsler Intelligence Scale for Children-Revised, and Columbia Mental Maturity Scale). These tests were shown to be valid and reliable in neurological populations and have been previously used to assess cognitive functioning in persons with CP [11].

The level of motor impairment was determined by the Gross Motor Function Classification Scale (GMFCS) [12] at the time of the interview. Table 1 displays the clinical characteristics of participants with CP within each age group.

Table 1 Clinical characteristics of persons with cerebral palsy (N = 86) for each age group

Clinical Variable	Children (N = 27)	Adolescents (N = 24)	Young Adults (N = 35)
Type of cerebral palsy			
Bilateral spastic	19	17	20
Unilateral spastic	1	1	2
Diskinetic	4	5	3
Ataxic	2	1	6
Mixed	1	0	4
Motor impairment (GMFCS)			
Level 1	2	3	8
Level 2	5	2	3
Level 3	4	7	3
Level 4	3	3	4
Level 5	13	9	17
Orthopedic impairment			
None	18	9	0
Hip sub-dislocation	7	6	10
Equinus	0	1	9
Scoliosis	2	8	16
Cognitive impairment			
None	7	10	7
Mild	5	2	3
Moderate	2	1	6
Severe	13	11	19

Number of persons classified according with type of cerebral palsy, orthopedic, cognitive, and motor impairment (following the Gross Motor Function Classification Scale [GMFCS]).

Pain-Related Questionnaires

Data about pain and quality of life were collected using a semi-structured interview. Augmentative communication devices and information from parents and caregivers were used as needed to facilitate data collection in subjects with communication difficulties. In addition, parents of all participants with CP completed the same written questions at home, unless they asked to do it in a face-to-face interview. Data from participants with CP who were unable to self-report (N = 43) were completed using their parents' data. This procedure has been used successfully in previous studies [2,4].

Pain was measured by using the following information from the interview:

1. whether they were experiencing chronic pain or not (yes/no response);
2. how many painful clinical treatments, such as surgery and stretching, did they receive;
3. ratings of current and worst pain in the last week by using a 11-point scale (0 = no pain, 10 = unbearable pain); and

4. the location of painful body regions by using a drawing of the human figure, and pain intensity ratings at each location by using a 4-point numerical scale (0 = no pain; 1 = mild; 2 = moderate; 3 = severe) (QL07/00 Pediatric Pain Questionnaire) [13]. Four pain scores were computed by dividing the sum of pain intensity ratings by the number of painful sites for each of the following body locations: head, upper limbs (shoulders, arms, elbows, hands), lower limbs (legs, hips, knees, ankles), and back. Pain scores were set to 0 in all participants with no pain.

The cerebral palsy quality of life (CP-QOL) questionnaire for children [14] was administered to evaluate subjective feelings (ranging from 1 = very sad to 9 = very happy) during several situations grouped in five areas: *social well-being and acceptance, functioning, participation and physical health, emotional well-being and self-esteem*, and *pain*. We decided to use this questionnaire for all the participants regardless of age to make data about quality of life comparable. The CP-QOL questionnaire was also completed by healthy volunteers, except those questions directly related to CP.

Assessment of Touch Sensitivity

Detection thresholds for mechanical stimuli were bilaterally measured at nine body locations (cheek, lip, ventral part of the lower arm, dorsum of the hand, thenar eminence, distal phalanx of the index finger, thoracic back, dorsum of the foot, calf of the leg) in a subgroup of participants (63 persons with cerebral palsy and 34 healthy controls). For this purpose, a kit of von Frey monofilaments (Somedic Sales AB, Sweden) consisting of 17 nylon hairs with increasing diameters (0.14–1.01 mm), constant lengths, and nominal force ranging from 26 mg to 110 g (manufacturer's data) were used. They were applied by pressing the filament at a 90° angle against the skin until it was bent. The filament was held in place for 1.5 seconds and then removed. Subjects were instructed to answer "yes" when a touch stimulus was perceived. After the task was explained, subjects were asked to wear a sleeping eye mask, and some practice trials were given to familiarize them with the assessment procedure. The task began with a randomly selected filament applied to one testing site. When a positive response to the stimulus was obtained, the same filament was applied two more times. If the touch sensation was positively felt in three consecutive trials, a thinner filament was applied to the testing site; if one negative response was given, a thicker filament was used. Null stimuli were also included to detect false positive responses. Responses with a delay greater than 3 seconds were considered as incorrect. Thus, the detection threshold for mechanical stimuli at one specific body location was defined as the lowest pressure perceived by the subject in three consecutive trials. The order for testing the 18 body locations was varied across the subjects, with the only restriction that the two sides of one body location were not assessed consecutively. An average touch sensitivity score was computed considering all body locations.

Statistical Analyses

Group differences on the presence of chronic pain and the number of painful clinical treatments were tested using chi-square and Mann–Whitney tests, respectively. Differences on pain reports (presence of chronic pain, number of painful clinical treatments, current and worst pain intensity, and pain scores at four body locations), touch sensitivity, and quality of life scores were tested by using analyses of variance (ANOVAs) with the between-subject factors GROUP (CP vs healthy controls) and AGE (children vs adolescents vs young adults). In the case of pain scores, an additional within-subjects factor BODY LOCATION (head vs upper limbs vs lower limbs vs back) was included to examine the spatial distribution of pain. ANOVA results were adjusted using Bonferroni corrections for post hoc comparisons and Greenhouse–Geisser corrections for the violation of sphericity assumptions. Pearson correlations were used to examine the relationship between pain reports, touch sensitivity, and age in CP participants and healthy controls. Finally, multiple regression analyses were used to test the contribution of age and quality of life to the current pain intensity in persons with CP and healthy controls.

Results

Subjective Pain Reports

The presence of chronic pain, defined as current pain lasting more than 3 months, was significantly more frequent in participants with CP (45.31%) than in healthy controls (18.40%) (chi-square = 15.37, $P < 0.001$). In addition, participants with CP underwent more painful clinical treatments than healthy controls (Mann–Whitney $U = -4.92$, $P < 0.001$). The univariate ANOVA on current pain intensity yielded a significant GROUP × AGE interaction effect ($F[2,205] = 4.40$, $P < 0.05$), indicating that current pain intensity was differentially modulated by age in participants with CP and healthy controls. Post hoc comparisons revealed that children with CP reported more enhanced pain intensity than healthy children ($P < 0.05$), whereas no group differences were observed in adults or adolescents. Furthermore, post hoc comparisons indicated that age-related differences on current pain intensity appeared in healthy controls (ps < 0.01) but not in participants with CP (Figure 1).

The ANOVA on worst pain intensity revealed a significant main effect of AGE ($F[2,205] = 5.74$, $P < 0.01$), indicating that in general, adults reported more pain than children ($P < 0.01$). In addition, the ANOVA on the number of painful locations showed significant main effects of AGE ($F[2,205] = 6.23$, $P < 0.01$) and GROUP ($F[1,205] = 32.1$, $P < 0.001$), indicating that persons with CP reported more painful locations than healthy controls ($P < 0.01$) and that adults had more painful locations than children ($P < 0.01$).

The spatial distribution of pain scores at several body locations for CP participants and healthy controls within

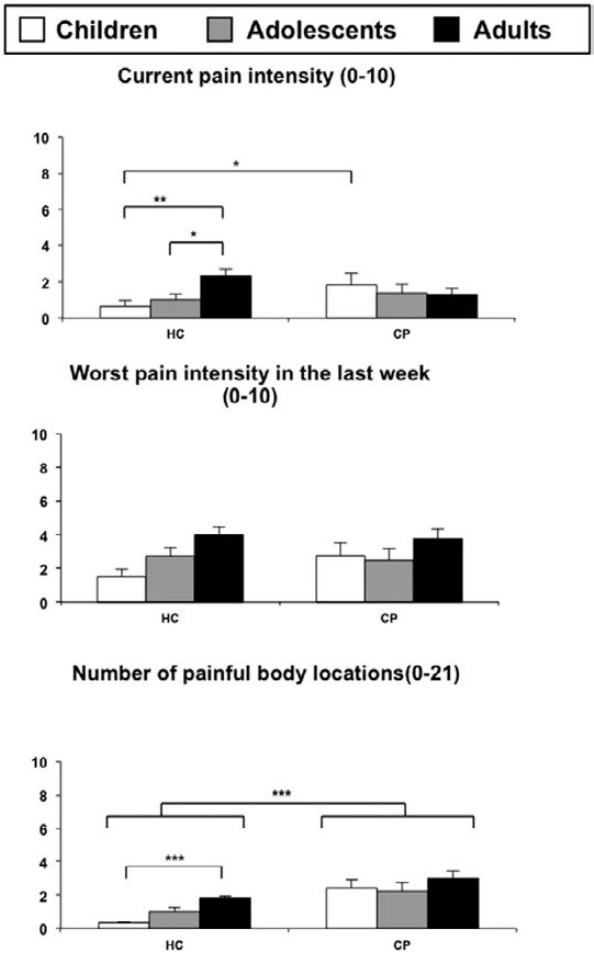

Figure 1 Pain ratings and number of painful body locations in persons with cerebral palsy (CP) and healthy controls (HC) for three age groups: children (6–10 years), adolescents (11–17 years), and young adults (18–30 years). Current and worst pain intensities were measured by using an 11-point scale (0 = no pain, 10 = unbearable pain). Asterisks indicate significant differences on post hoc comparisons (significance level: $^{*}P < 0.05$, $^{**}P < 0.01$, $^{***}P < 0.001$).

each age group is displayed in the upper panel of Figure 2a. The multivariate ANOVA of these scores revealed that pain intensity was significantly higher in persons with CP than in healthy controls (GROUP main effect: F[1,184] = 17.69, $P < 0.001$), higher in young adults than in adolescents and children (AGE main effect: F[2,184] = 3.86, $P < 0.01$), and higher in lower limbs than in the rest of body locations (BODY LOCATION main effect: F[3,552] = 19.19, $P < 0.001$). Moreover, a significant BODY LOCATION × GROUP interaction effect (F[3,552] = 13.02, $P < 0.001$) was found, indicating that differences on pain intensity appeared on specific body locations. Post hoc comparisons revealed that CP participants reported more enhanced pain scores than healthy controls on back ($P < 0.05$), lower limbs ($P < 0.001$), and upper limbs ($P < 0.01$) but not on head pain (Figure 3). Moreover, lower limb pain was more intense than both upper limb ($P < 0.001$) and head pain ($P < 0.001$) in CP participants, whereas back pain was more intense than both upper ($P < 0.001$) and lower limb pain ($P < 0.05$) in healthy controls.

Touch Sensitivity

Figure 2b displays a topographical distribution of touch sensitivity thresholds on the nine body locations (cheek, lip, ventral part of the lower arm, dorsum of the hand, thenar eminence, distal phalanx of the index finger, thoracic back, dorsum of the foot, calf of the leg) in participants with CP (N = 63) and healthy controls (N = 34) for each age group. Due to the small sample sizes, a mean threshold index was calculated by averaging the threshold values across all nine body locations. The ANOVA on the average thresholds revealed a significant interaction effect between GROUP and AGE (F[2,91] = 5.1, $P < 0.01$) and a GROUP main effect (F[1,91] = 8.94, $P < 0.01$). Post hoc comparisons indicated that CP adults and young adults were less sensitive to touch than healthy adults and young adults (ps < 0.01). Moreover, post hoc comparisons also revealed that healthy children had more reduced touch sensitivity than healthy adults ($P < 0.05$) (Figure 4).

Quality of Life

Figure 4 displays the mean scores on the five domains of the CP-QOL in persons with CP and healthy controls within each age group (children, adolescents, and young adults). Participants with CP and healthy controls differed in all domains of the CP-QOL. In particular, CP reported lower scores than healthy controls in *social well-being and acceptance* (GROUP main effect: F[1,195] = 11.11, $P < 0.01$), *participation and physical health* (GROUP main effect: F[1,195] = 20.26, $P < 0.001$), *emotional well-being and self-esteem* (GROUP main effect: F[1,195] = 16.31, $P < 0.001$), and *functioning* (GROUP main effect: F[1,195] = 21, $P < 0.001$). Moreover, CP participants had higher scores in the domain *Pain* (GROUP main effect (F[1,195] = 8.64, $P < 0.01$) than healthy controls. In addition, main effects of AGE were found in *participation and physical health* (F[2,195] = 12.03, $P < 0.001$) and *emotional well-being and self-esteem* scores (F[2,195] = 22.46, $P < 0.001$), indicating that children had higher scores than adolescents and young adults in both groups (all ps < 0.05). No significant interaction effects between GROUP and AGE were yielded in any of the CP-QOL domains (Figure 5).

Relationship Between Pain Reports, Age, and Touch Sensitivity

Significant positive correlations were found between different pain reports (current and worst pain intensity,

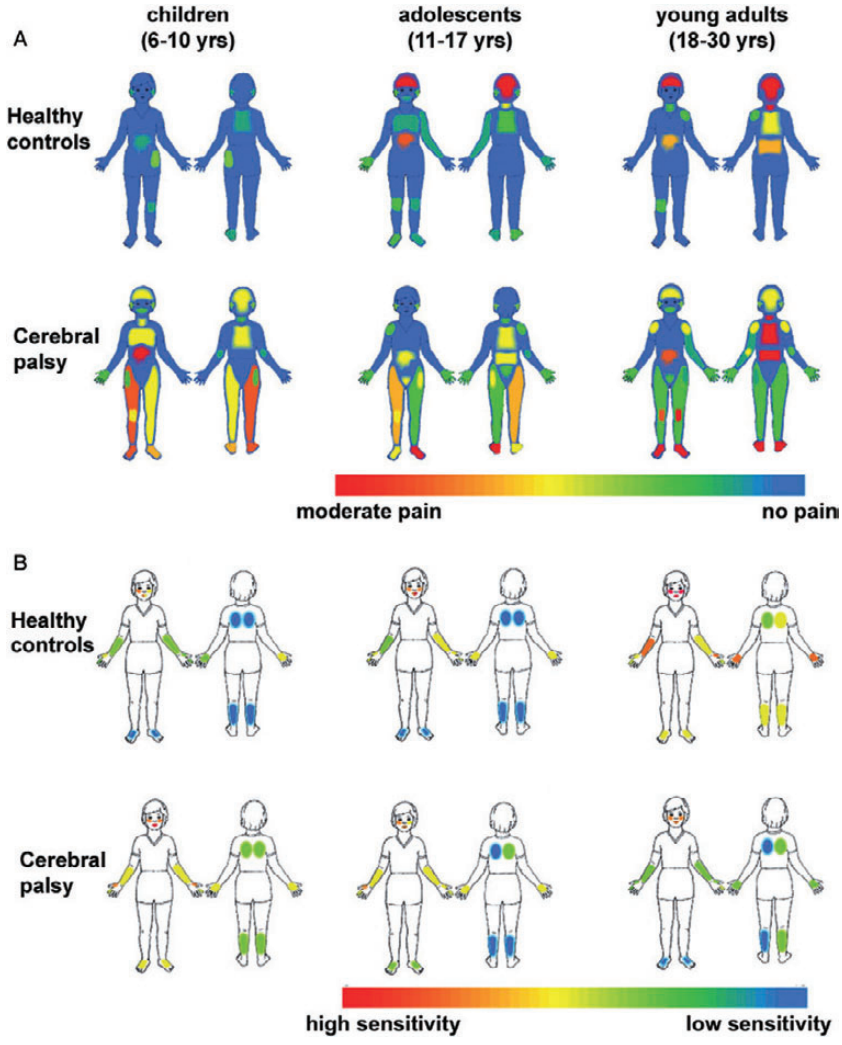

Figure 2 Topographical distribution of pain intensity scores and touch sensitivity thresholds on several body locations. (Panel A) Pain was obtained from participants' self-reports by rating the intensity of pain sensations at each body region with a 4-point numerical scale (0 = pain, 1 = mild, 2 = moderate; 3 = severe). The color-based scale ranges from 0 (when all the people reported no pain) to 1 (equivalent to moderate pain). (B) Touch sensitivity was measured bilaterally applying von Frey monofilaments at nine body locations (cheek, lip, ventral part of the lower arm, dorsum of the hand, thenar eminence, distal phalanx of the index finger, thoracic back, dorsum of the foot, calf of the leg).

number of painful locations, head pain score, back pain score) and age in healthy controls, whereas no significant correlation were found between pain reports (except for upper limb pain) and age in participants with CP (Table 2). Moreover, worst pain intensity was negatively correlated with touch sensitivity thresholds in healthy controls, indicating that enhanced pain was associated with enhanced touch sensitivity. By contrast, head, lower limb, and back pain scores were positively correlated with touch sensitivity thresholds in participants with CP, indicating that enhanced pain was associated with reduced touch sensitivity. Finally, back pain was positively correlated with the number of orthopedic problems in CP.

To further analyze the contribution of age and psychosocial variables to current pain in healthy controls and individuals with CP, multiple regression analyses were performed separately for each group of participants using *Current pain intensity* as the dependent variable and age and quality of life scores as predictor variables. The analyses showed that different predictors accounted for a significant proportion of the variance in individuals with CP

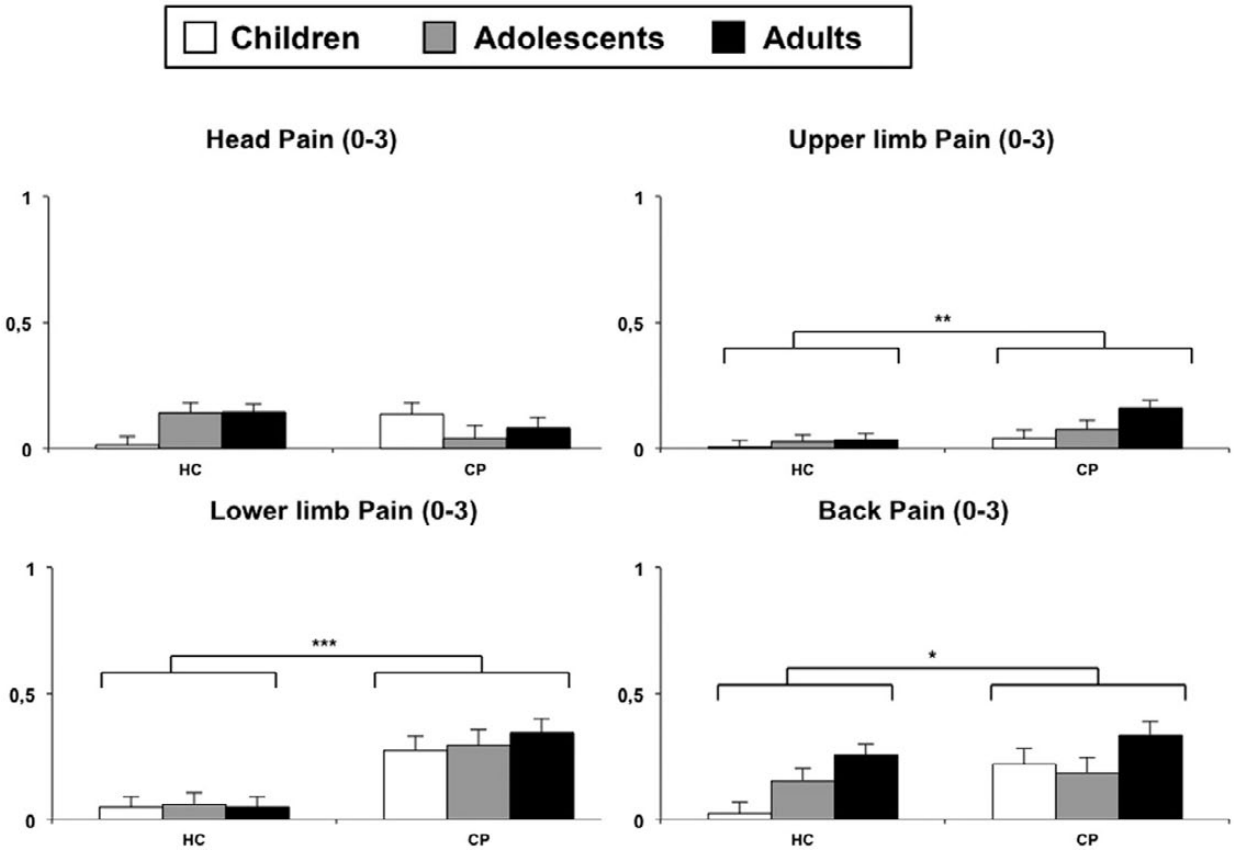

Figure 3 Pain scores on four body locations in persons with cerebral palsy (CP) and healthy controls (HC) for three age groups: children (6–10 years), adolescents (11–17 years), and young adults (18–30 years). Pain scores were computed by dividing the sum of pain intensity ratings by the number of painful sites for each of the following body locations: head, upper limbs (shoulders, arms, elbows, hands), lower limbs (legs, hips, knees, ankles), and back. Asterisks indicate significant differences on post hoc comparisons (significance level: $*P < 0.05$, $**P < 0.01$, $***P < 0.001$).

($r = 0.447$, $F[2,83] = 10.37$, $P < 0.001$) and healthy controls ($r = 0.238$, $F[1,113] = 7.39$, $P < 0.01$). In persons with CP, two CP-QOL subscales (*Functioning* and *Emotional well-being and self-esteem*) were significant predictors of *Current pain intensity*, whereas *age* was the best predictor of pain intensity in healthy controls (Table 3).

Discussion

The major aim of the present study was to analyze age-related differences on pain and touch sensitivity in persons with CP and healthy controls. Basically, we found that participants with CP reported more pain, lower touch sensitivity, and more reduced quality of life than healthy controls. Moreover, it was observed that differences on pain reports and touch sensitivity were mediated by age in healthy controls but not in persons with CP. In addition, analyses of the relationship between pain and touch sensitivity revealed that increased pain intensity was associated with increased average touch sensitivity in healthy controls, whereas increased ratings of head, lower limb, and back pain were associated with reduced touch sensitivity in participants with CP.

In the present study, 43% of participants with CP experienced pain as chronic and underwent more painful clinical treatments than healthy controls. These findings are in agreement with previous reports indicating that around 60% of persons with CP experience recurrent pain of a moderate-to-severe intensity on a daily or weekly basis that significantly interferes with daily activities [2,4,15–17]. Moreover, our data suggest that pain reported by participants with CP was significantly above the prevalence rate of pain among the general population. However, contrasting with the extensive data about pain in adults, little is known about pain characteristics among children and adolescents. To date, almost all epidemiological studies have focused on the presence of specific pain syndromes

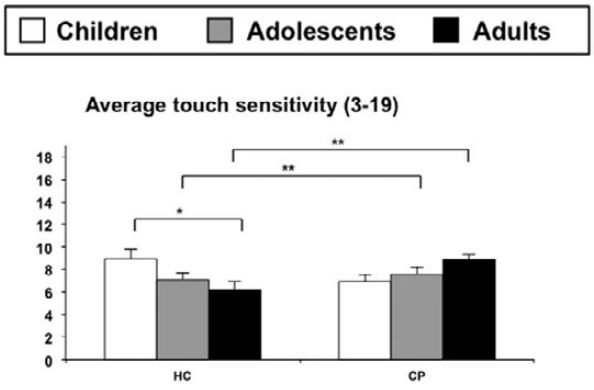

Figure 4 Average touch sensitivity in persons with cerebral palsy (CP) and healthy controls (HC)for three age groups: children (6–10 years), adolescents (11–17 years), and young adults (18–30 years). Touch sensitivity was computed as the average threshold of nine body locations (cheek, lip, ventral part of the lower arm, dorsum of the hand, thenar eminence, distal phalanx of the index finger, thoracic back, dorsum of the foot, calf of the leg). Asterisks indicate significant differences on post hoc comparisons (significance level: $^{*}P < 0.05$, $^{**}P < 0.01$, $^{***}P < 0.001$).

(e.g., juvenile chronic arthritis, knee pain, fibromyalgia, low back pain, cancer pain, migraine, headache), rather than on specific clinical manifestations of chronic pain. In this context, our data on the prevalence of pain among healthy children and adolescents seem to be in agreement with previous population-based surveys. Thus, recent cross-sectional studies revealed that around 25–40% of school-children reported chronic pain [18,19], although only 5.1% had moderate or severe pain problems. In addition, according with other epidemiological surveys of the general population [18–21] our data show a significant enhancement of pain with increased age among healthy controls. A further survey among children (5–16 years of age) with different chronic diseases (arthritis, cancer, enuresis, and headache) and healthy showed that presence of chronic pain differed depending on the health condition, with the lowest rates (4–7%) in healthy and children with enuresis or headache and the highest rates (78%) in children with arthritis [21]. Thus, it seems that chronic pain in CP appears to have similar rates than in other chronic diseases.

A further relevant finding of the present study was that pain and quality of life were not influenced by age in CP, but significant age-related differences appeared in healthy controls. We observed that healthy children reported lower pain scores and a better quality of life compared with healthy young adults. In contrast, the three age groups of participants with CP did not differ in pain ratings (except for upper limb pain) or quality of life. Moreover, age was the best predictor of pain intensity in healthy controls but not in participants with CP. These findings are in agreement with previous surveys in healthy adult samples indicating that the prevalence of chronic pain increases with aging and that age influences pain perception [16,17,22]. Moreover, our data further suggest that the influence of age on pain would extend to children and adolescents among the general population. Pain in CP appears to be a relevant phenomenon from an early age, showing no age-related changes over the life span. Therefore, it seems that the presence of pain should be specifically addressed in rehabilitation programs for children and adolescents with CP.

Interestingly, our findings showed that age-related changes in pain perception were mirrored by age-related changes in touch sensitivity (detection of light pressure against the skin using the von Frey monofilaments) in healthy controls but not in subjects with CP. Moreover, enhanced persistent pain was associated with reduced touch sensitivity in healthy controls, but a reduced sensitivity among individuals with CP was observed. In addition, significant differences on touch sensitivity were found only between healthy controls and persons with CP in young adults and adolescents but not in children. These findings are in agreement with previous reports indicating that touch and pressure sensitivity are less impaired in children with CP than other measures of sensitivity such as the two-point discrimination [23]. Our data extend those findings to show that touch sensitivity is impaired in young adults and adolescents with CP but not in children compared with age-matched healthy controls. Increased sensitivity to non-noxious stimulation (allodynia) measured by von Frey monofilaments is also a characteristic of patients suffering from chronic pain, particularly of those with neuropathic pain [24]. In this sense, the positive relationship observed between pain and touch sensitivity in healthy controls might be the result of plastic changes associated with the persistence of pain over time [17]. In contrast, reduced touch sensitivity together with enhanced pain observed in CP may indicate the existence of different psychophysiological mechanisms for the maintenance of pain. The implications of these age-related changes in touch sensitivity and pain for the development of chronic pain in healthy people and in persons with CP should be addressed in future studies using several measures of experimental pain sensitivity.

Our finding of a high level of pain compared with healthy controls is also of special importance because it has been suggested that pain reports in childhood and early adolescence could be associated with increased pain [25] and affective disorders in adulthood [26]. Moreover, repeated painful experiences during periods of neurological development may cause relevant changes in pain thresholds and tolerance throughout a person's lifetime [27]. In this sense, it has been suggested that the regular participation of children with CP in chirurgic and rehabilitation procedures (stretching, electrical stimulation, functional mobility

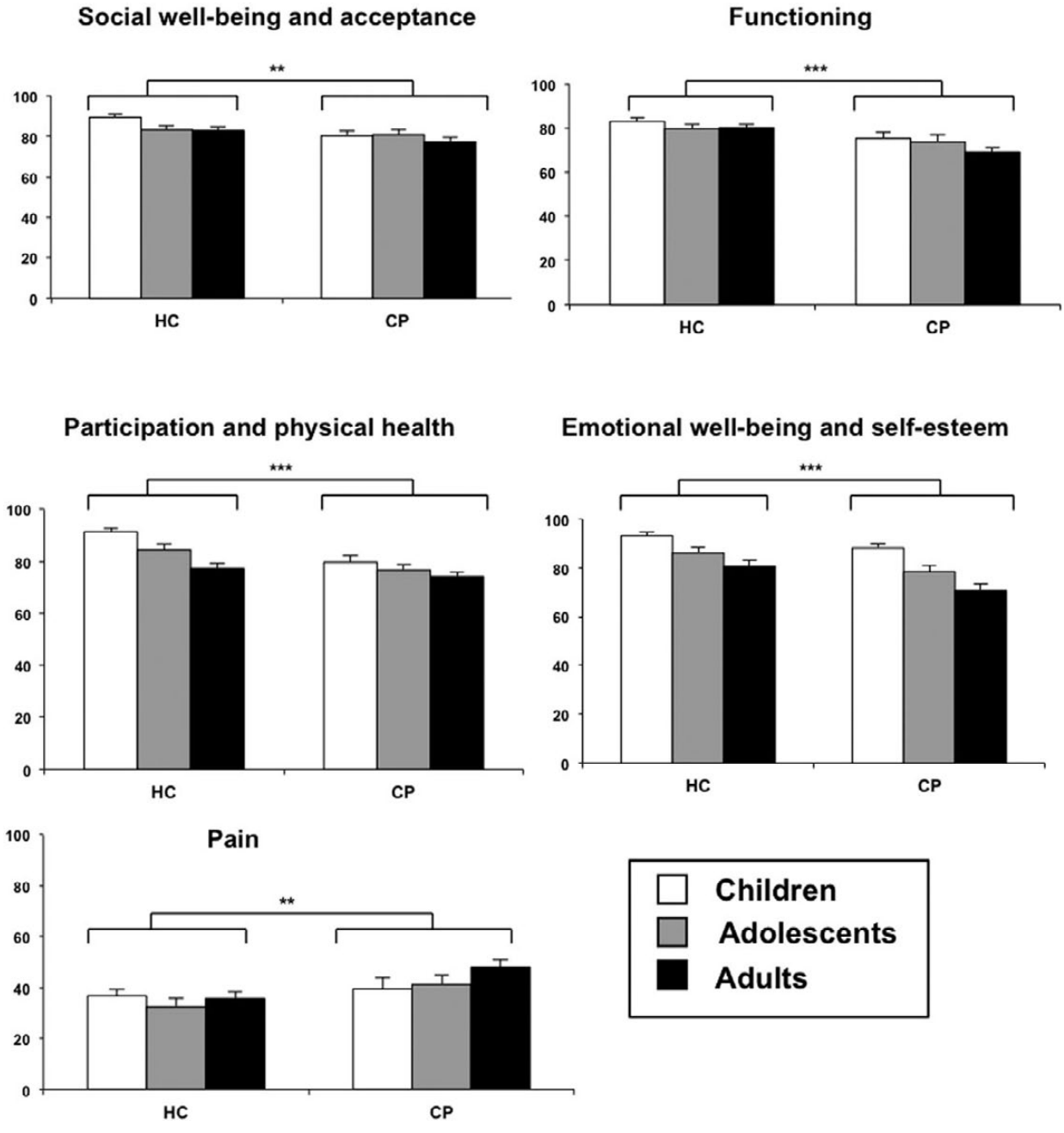

Figure 5 Scores on the cerebral palsy quality of life (CP-QOL) questionnaire in persons with CP and healthy controls (HC) for three age groups: children (6–10 years), adolescents (11–17 years), and young adults (18–30 years). Asterisks indicate significant differences on post hoc comparisons (significance level: $^{*}P < 0.05$, $^{**}P < 0.01$, $^{***}P < 0.001$).

training, splinting and orthotic fabrication, serial casting, training for use of adaptive equipment, and utilization of standing frames and other positioning devices) could be associated with a high prevalence of pain [25]. Furthermore, it has been noted that parents of children with CP identified assisted stretching as the most frequent painful activity of daily living [28] and that one of the most salient negative memories of childhood in adults with CP is pain related to stretching and bracing in physical therapy [29]. Thus, it appears that ongoing rehabilitation efforts would lead to increasing anxiety, fear, frustration, withdrawal, or distress about these interventions and facilitate the

Table 2 Relationship between several measures of pain and age, touch sensitivity, and motor impairments in participants with cerebral palsy and healthy controls

	Current Pain Intensity	Worst Pain Intensity	Number of Painful Locations	Head Pain	Upper Limb Pain	Lower Limb Pain	Back Pain
Persons with cerebral palsy (N = 86)							
Age	–0.109	0.087	0.102	–0.035	0.292*	0.096	0.150
Touch sensitivity threshold	0.155	0.164	0.117	0.317*	0.159	0.353**	0.299*
Motor level (GMFCS)	0.087	0.001	–0.194	–0.225	–0.060	–0.122	–0.150
Number of orthopedic problems	0.041	–0.197	0.097	0.114	0.325	0.035	0.434*
Healthy controls (N = 115)							
Age	0.238**	0.230***	0.457***	0.309**	0.120	0.030	0.336**
Touch sensitivity threshold	–0.163	–0.218*	–0.331	–0.227	–0.136	–0.107	–0.212

* $P < 0.05$, ** $P < 0.01$, *** $P < 0.001$.
GMFCS = gross motor function classification scale.
Current and worst pain intensities were measured by using an 11-point scale (0 = no pain, 10 = unbearable pain). The head, upper/lower limb, and back pain scores were obtained by using a 4-point scale (0 = no pain; 1 = mild; 2 = moderate; 3 = severe) (upper limb pain scores were computed by averaging the pain scores for shoulders, arms, elbows, and hands; lower limb pain scores were computed by averaging the pain scores for legs, hips, knees, and ankles).

Table 3 Multiple regression predictors of current pain intensity in persons with cerebral palsy and healthy controls

Predictors	Beta	Adj R2	R2
(a) Individuals with cerebral palsy (N = 86)			
Age	–0.104		
Social well-being and acceptance	–0.021		
Functioning	–0.502**		
Participation and physical health	–0.233		
Emotional well-being and self-esteem	0.296*		
Pain	0.159		
		0.181	0.200
(b) Healthy controls (N = 125)			
Age	0.238*		
Social well-being and acceptance	–0.002		
Functioning	–0.006		
Participation and physical health	–0.033		
Emotional well-being and self-esteem	–0.063		
Pain	–0.076		
		0.049	0.057

* $P < 0.01$, ** $P < 0.001$.

establishment and maintenance of pain memories [19,30]. In this sense, the present study highlights the importance of considering the presence of pain at very early ages in CP.

Nevertheless, our study has some limitations that should be taken into account for the interpretation of the results. Although our sample of persons with CP was selected from educational and occupational settings in our community, it is small, and the response rate was low. Moreover, it is noteworthy that the selected sample displays the different characteristics concerning motor and cognitive impairments and the type of CP compared with other epidemiological studies. The use of questionnaires, although adequate for this explorative purpose, has some important methodological bias such as the use of the same questionnaires by persons with diverse cognitive and developmental abilities that may cause some distortions. The use of pediatric questionnaires in an adult population and the lack of the proven validity of some instruments in healthy samples, such as the CP-QOL measure, may have introduced some methodological biases in the study. Moreover, the use of self and surrogate pain reports may have decreased the reliability of the data. Finally, the cross-sectional design of the present study represents a further limitation. Although our study does not provide information about how pain experience changes over time in CP, it lays a scientific basis for the

implementation of a longitudinal design and it guides the selection of appropriate outcome measures for future studies.

In conclusion, it appears that pain experience in CP participants was not influenced by age in a similar way as it happened in the healthy population. This suggests that different psychosocial and psychophysiological mechanisms may be involved in the maintenance of pain over the life span.

Competing Interests

The authors declare that they have no competing interests.

Author's Contribution

PM and IR conceived of the study, participated in its design and coordination. IR, IC, and PM performed the statistical analysis and executed the drafting of the manuscript. IR carried out the interviews and sensitivity tests.

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