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Update article

# Prevalence of pain in 240 non-ambulatory children with severe cerebral palsy

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ABSTRACT

*Background:* Several studies have given frequencies of pain in children with cerebral palsy, but comparing the findings is difficult. We aimed to estimate the prevalence of pain in non-ambulatory children with cerebral palsy and describe their characteristics by presence or absence of pain.
*Methods:* Data were extracted from an ongoing longitudinal national cohort following non-ambulatory children with severe cerebral palsy aged 3 to 10 years over 10 years. We described and compared data for the first 240 children at inclusion by presence or absence of pain. Pain was assessed by a visual analog scale and the Douleur Enfant San Salvadour scales and by investigator interview.
*Results:* Overall, 65 children experienced pain, for a prevalence of 27.1% (95% confidence interval 22–33%). All children experiencing pain had orthopaedic pain and 45.6% had pain from another origin. The main pain sites were hips (43.4%) and feet (26.9%). Joint mobilisation was the source of pain for 58.3% of children experiencing pain, and sitting was identified as painful for 10.3%. Pain was greater with scoliosis (43.1% vs 24.1% with and without pain; $P = 0.006$) and spasticity treatment (32.3% vs 17.2%; $P = 0.020$).
*Conclusions:* Children with cerebral palsy frequently experience pain and also early pain, mostly articular and orthopedic. The assessment of pain should be systematic because of its high prevalence. Interventions to prevent scoliosis, hip luxation, and foot deformities and to reduce spasticity, such as the use of analgesics before joint mobilization exercises, may reduce the prevalence of this pain.

## 1. Introduction

Cerebral palsy (CP) is a group of disorders that occurs in 2 to 3 per 1000 live births [1]. This group includes permanent disorders of motor function (movement, muscle control, posture, and/or balance) due to a non-progressive injury or malformation that affects the immature brain before, during, or immediately after birth. Among the children with CP who do not acquire the ability to walk (levels IV or V of the Gross Motor Function Classification System [GMFCS]) [2], one third would have a high risk of orthopaedic contractures, especially hip dislocation, scoliosis, knee flexum, and foot deformities (equinus, with or without varus or valgus) [3]. In these children, pain is frequent, sometimes triggered or aggravated by care, and may affect the quality of life of children and their families [3].

The assessment of pain in these children is encouraged and involves questioning children and their families. The Fondation Motrice, in partnership with family associations, carried out the first major national survey (ESPaCe; (http://www.fondationparalysiecerebrale.org/) to give children and adults with CP and their families the opportunity to discuss motor rehabilitation practices so as to appreciate their content and feelings, especially painful experiences.

Several studies have given frequencies of pain in people with CP, but comparing the findings is difficult. Indeed, the prevalence of pain in CP is not the same in young children as in teenagers or young adults: it increases with age [4,5] and disease severity [6,7]. In the current literature, the frequency of pain ranges from 14% to 70% depending on the characteristics of pain, how pain is defined, and the study population [8–14]. Specifically, some studies showed very different pain prevalence values because of wide age ranges: 11% of pain prevalence in 198 children aged 5 to 18 years [10], 62% in 153 children aged 8 to 18 [14], 56% in 73 children aged 4 to 18 [11], and 78% in 94 children aged 4 to 18

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[6]. As well, other studies had very wide-ranging disease severities (GMFCS levels): the whole spectrum of severity in studies by Kennes et al. [12] and Ramstad et al. [14] and GMFCS levels III to V in another study [10]. In the study by Kennes et al. [12], the prevalence in young children aged 5 to 13 years was 14%, but the study considered the whole spectrum of disease severity and the pain was declared by the parents. Finally, some studies gave prevalence values for small samples: 70% in only 20 children [8] or 34.9% in 19 children [9].

The aim of this present study was to estimate the prevalence of pain among a homogeneous population of young and non-ambulatory children with CP and to identify the factors associated with a high risk of pain.

## 2. Material and methods

### 2.1. Sample

A multicentre national cohort study was implemented by the *hospices civils de Lyon* in September 2009 and is ongoing (385 children are expected; study title "Étude longitudinale d'une population d'enfants atteints d'une forme bilatérale sévère de paralysie cérébrale : incidence et évolution des complications orthopédiques et des douleurs en lien avec ces complications"; ClinicalTrials.gov identifier: NCT01840930). This non-interventional study involves the follow-up of children aged 3 to 10 years old at inclusion, with CP GMFCS level IV (self-mobile with limitations, assisted transportation or self-mobility with power chairs) and V (extremely limited self-mobility even with an assistive technology) over a 10-year period, at a frequency of 1 visit per year. At each visit, the investigator uses a case report form to collect clinical, orthopaedic, radiological, and environmental data. Children can be recruited by the *centre hospitalo-universitaire*, *centre hospitalier général*, *Institut médico-éducatif*, *Institut* or *Centre d'éducation motrice*, *Service d'éducation spéciale et de soin et à domicile*, and *Centre d'action médico-sociale précoce*.

The primary objective of this cohort is to measure the incidence of orthopaedic complications (scoliosis and hip joint) according to age. The secondary objectives are to describe the sequences over time of these complications and the related pain; to explore the impact of nutrition, surgery, asymmetric postures and environmental factors; and to describe the medical and rehabilitative follow-up of these children.

This prospective study was carried out in accordance with the ethical standards of the 1994 Declaration of Helsinki and was approved by the medical ethics committee of Lyon Sud Est 2. Informed consent was obtained from the parent for the inclusion and follow-up of all children.

In this present study, we extracted data from the inclusion visit for the first 240 children included in the cohort at the 51 investigator sites between September 2009 and September 2014.

## 3. Methods

The present study focused on pain. First, the criterion "presence of pain" was determined by a positive answer to either of the 2 following questions during the baseline visit:

- Is the child experiencing pain today or has he/she been experiencing pain during the last months?
- Does the child experience pain during joint mobilisation?

During the visit, the investigator directly posed these questions to the child's family and to the child, if the child could communicate and was able to understand the questions. The investigator could also question the child's physiotherapists to take into account their experience. We also asked whether the child was given analgesics occasionally, which represented a way to better detect the criterion "presence of pain."

Second, for children with the criterion "presence of pain," the investigator was asked to measure the level of pain: 1) with a visual analog scale (VAS) for children able to communicate or 2) by questioning the caregivers of the other children and completing the Douleur Enfant San Salvadour (DESS) scale, a specific scale for non-communicating children [15].

The study also collected data related to the origin and site of pain and treatments. These data are recorded at the visit by the investigator, according to the investigator's experience and that of the caregivers, and according to the family/child interview (when applicable). The pain to be recorded included osteoarticular pain (spine, hip, knee, foot), other pain (muscular/contractures, other than neuro-orthopedic) and painful joint mobilisations, painful sitting position.

We also extracted the GMFCS level (IV or V), the ability to communicate, and the presence of seizures, hip subluxation > 50%, scoliosis (clinical scoliosis, as evaluated by the investigator), prematurity (according to the diagnosis completed by the investigator at the inclusion visit), gastrostomy, and treatment for spasticity (lioresal [by mouth/intrathecally], rivotril, dantrium, valium, myolastan, other).

### 3.1. Statistical analysis

The characteristics of children are described by the mean (SD) for quantitative variables and number (%) for categorical variables. The prevalence of pain was estimated by the proportion of children identified as experiencing pain, with an exact 95% confidence interval (CI). The characteristics of children were studied in the entire cohort and by those with and without pain by Fisher's exact test. Statistical analyses involved use of SAS v9.3. $P < 0.05$ was considered statistically significant.

## 4. Results

The study population included 240 children (133 boys [55.4%]) with mean age 6.79 ± 1.93 years (median 6 years and 8 months). Most children (n = 211; 87.9%) presented a spastic form of CP, and 69.2% (n = 166) were non-communicating children. Overall, 65 children experienced pain, for an estimated pain prevalence of 27.1% (95% CI 22–33%); 114 children (47.5%) were occasionally given analgesics (Table 1).

Among the 65 children with pain, the pain could be measured in 44 (67.7%) with a VAS in 8/17 (47.1%) children able to communicate and the DESS in 34/48 (70.8%) unable to communicate (Table 2). Pain measured by the VAS and DESS could not be measured in 8/17 children (47.1%) able to communicate and in 13/48 (27.1%) unable to communicate.

**Table 1**
General characteristics of 240 non-ambulatory children with severe cerebral palsy.

| | |
|---|---|
| Sex | |
| Girls | 107 (44.6) |
| Boys | 133 (55.4) |
| Age (years) (*n* = 239) | |
| Mean | 6.79 (1.9) |
| [Min–max] | [3.21–10.98] |
| Spastic cerebral palsy | 211 (87.9) |
| No oral language | 166 (69.2) |
| Presence of pain | 65 (27.1)<br>95% CI (22–33%) |
| Occasional analgesic treatment | 114 (47.5) |

Data are *n* (%) unless indicated.

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