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Original Article

# Trends observed in bilateral cerebral palsy during a thirty-year period: A cohort study with an ICF-based overview



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PEDIATRICS of NEONATOLOGY

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Key Words cerebralpalsy; epidemiology; ICF	<ul> <li>Background: To describe trends observed across thirty years in demographic and clinical characteristics and rehabilitation of patients with bilateral cerebral palsy.</li> <li>Methods: This retrospective study includes 464 (261 M and 203 F) inpatients with bilateral cerebral palsy, born from 1967 to 1997 and discharged from an outpatient rehabilitative facility from 1985 to 2015. Data concerning the health profile were collected from medical reports and organized in the domains of Body Functions and Structure; Activity and participation and Personal and Environmental factors as proposed in the International Classification of Functioning Disability and Health (ICF). The trend observed over the three birth decades was discussed. <i>Results:</i> The duration of the rehabilitative treatment decreased across decades approximately by two years per decade (from an initial 16.2 yrs to 12.3 yrs). Across the decades the rate of quadriplegia decreased, whereas rates of diplegia increased; spasticity was the prevalent observed motor type for all decades. The most frequent musculoskeletal disorder involved the middle inferior part of the body; among comorbidities a steady decrease in psychiatric disorders was found. With respect to the first decade a slight improvement was observed in the gross motor functioning and in the hand dexterity. No particular trend was observed concerning communication abilities. An increase in the use of pharmacological and surgical treatments for motor symptoms was observed.</li> <li>Conclusion: This study presents and describes the functioning of a large sample of Italian patients with bilateral CP on the basis of the ICF framework and it discussed the trend observed across decades.</li> <li>Copyright © 2021, Taiwan Pediatric Association. Published by Elsevier Taiwan LLC. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/ by-nc-nd/4.0/).</li> </ul>

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## 1. Introduction

Thanks to advances in neonatal care, a growing number of infants born preterm and/or with extremely low birth weight are now surviving. However, these infants are at an increased risk for neurodevelopmental disabilities such as cerebral palsy. The term Cerebral Palsy (CP) "describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems."<sup>1</sup>

The prevalence of CP appears to be stable over time at around 1.5–3 per 1000 live births,<sup>2</sup> but some changes concerning the health profile of CP have been found across years.<sup>2–7</sup>

CP has always been characterized as a disorder of movement and posture; however, more recent definitions allow clinicians to appreciate more than just the movement disorder.<sup>8</sup> Similarly, to maximize the efficacy of rehabilitation, interventions should be framed within the global functioning of the patients and should consider several factors including the occurrence of comorbidities, the role of family, the effect of social and cultural aspects, the occurrence of environmental factors that operate as facilitators or barriers and so on.

The aim of this paper is to describe the trend observed across three decades in the health profile of patients with bilateral CP, following the conceptual model of the International Classification of Functioning Disability and Health (ICF).<sup>9</sup> The ICF was published by the WHO in 2001. It is based on a biopsychosocial model and it describes disability and functioning as multidimensional concepts related to Body functions and Structures and Activities and Participation in daily living. It also describes contextual factors (both personal and environmental) that may influence these experiences.

## 2. Material and methods

Retrospective data of patients with bilateral CP born from June 1967 to June 1997 and admitted in a network of seven Children Rehabilitation Centers (Scientific Institute Eugenio Medea based at: Conegliano, Pieve di Soligo, Padova, Treviso, San Donà, Vicenza and Oderzo) were collected. These centers provide care for all children with disability in the circumscribed territory of three Local Health Units located in the Veneto Region, Italy.

Data concerning the health profile were collected from medical reports and organized in the domains of Body functions and structure; Activity and participation and Personal and Environmental factors as proposed in the ICF.

## 2.1. Participants

Participants were defined as eligible if all the following criteria were fulfilled: (1) diagnosis of bilateral CP (quad-riplegia and diplegia), (2) year of birth between June 1967 to June 1997, (3) treatment in one of the seven Children

Rehabilitation Centers, (4) date of treatment between 1985 and 2015 and (5) duration of treatment  $\geq$  3 years. The final sample consisted of 464 inpatients (261 M and 203 F).

## 2.2. Variables

The set of variables retrieved from the clinical records that were considered to determine the health profile of the patients are shown in Table 1.

Activity limitations for motor, manual and communication functioning were measured with the following scales: the Gross Motor Function Classification System – Expanded & Revised (GMFCS – E&R)<sup>10</sup>; the Manual Ability Classification System (MACS)<sup>11</sup> and the Communication Function Classification System (CFCS).<sup>12</sup>

The GMFCS was ideated and adopted from the end of 1990s, whereas the MACS and the CFCS were adopted even later (2006 for MACS and 2011 for CFCS); therefore, they were not available at the time of discharge of some patients. For the aim of this research, the scales were retrospectively filled on the basis of the clinical records by the same physician who met all the patients for the first assessment.

The GMFCS is a 5-level classification system that describes the gross motor function of children and youth with CP. Level I indicates that the patient performs gross motor skills with minimum limitations (on speed, balance and coordination) and it represents the highest level of functioning. Level V indicates that patient is impaired in all areas of motor functioning and cannot walk independently even if he/she is able to use powered mobility.

The MACS is a 5-level classification system that describes the ability on the use of hands to handle objects in everyday activities in children and youth with CP. Level I indicates that patient is able to handle objects easily and successfully. Level V is used when the patient cannot handle object and, due to his/her severely limited ability, needs total assistance.

The CFCS is a 5-level classification system that describes everyday communication performance in children and youth with any disability. Level I indicates that patient is able to communicate easily both as sender and receiver with most people in most environments. Level V indicates that patient is seldom effective to communicate even with familiar conversational partners.

Data concerning prevalent risk factors and comorbidities that may have interfered with the normal development of brain structure and functions were also considered. Risk factors were: hypoxia, preterm delivery (infants <36 weeks of gestation), hemorrhage, perinatal infection, congenital anomalies, exposure to toxic substances, epilepsy (defined as two or more unprovoked seizures after the neonatal period), other and unknown.

Moreover, in accordance with other studies<sup>4-13</sup> a score of overall disability, the Disab score, was computed following the method of Blair et al.<sup>14</sup> The Disab score is obtained by summing the score assigned to the following disabilities: the extent and severity of body impairment, the level of cognitive impairment and the occurrence of complications. It ranges from 0 to 12 (1–5 mild; 6–8 average; severe  $\geq$  9).

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ICF domains	Variables	Classification	Measurement
Body functions and structure	Gestational age at birth (weeks)	Term ( $\geq$ 37 weeks), moderately preterm (26 -36 weeks), extremely preterm (22-25 weeks)	Mean number of completed weeks
	Body weight at birth	Term ( $\geq$ 3000 g), Low (2999–998 g), extremely low ( $\leq$ 999 g)	Mean birth weight
	Part of the body affected	Diplegia or quadriplegia	Rates (%)
	Motor types	Spastic, ataxic or dyskinetic (including dystonia and athetosis)	Rates (%)
	Musculoskeletal disorders	Affecting the middle inferior part of the body (hip, thigh, knee, foot), the trunk, or the superior part of the body (elbow, wrist, hand)	Rates (%)
	Comorbidities	Disorders of visual function (Visual acuity loss, saccadic impairments, strabismus, oculomotor problems)	Rates (%)
		IDD	Mild (approximate IQ range 50–69), moderate (IQ range 36–49), severe or profound (IQ $\leq$ 35)
		Disorders of auditory function (Hearing acuity loss, auditory attention and orientation to stimuli)	Rates (%)
		Epilepsy, disease of the respiratory system, psychiatric disorders, dysphagia and communicative disorders	Rates (%)
Activity and	Mobility — walking and moving	GMFCS	Levels I–V
participation	Mobility — carrying moving and handling objects	MACS	Levels I–V
	Communication-receiving and producing	CFCS	Levels I–V
Personal factors	Age at the time of admission, age at the time of discharge		
Environmental	Total duration of treatment		
factors	Orthopedic surgery	Soft tissues or bone surgery	Rates (%)
	Type of rehabilitative interventions	Physical, speech, occupational and other therapies	Rates (%)
	Pharmacological treatment for motor symptoms		Rates (%)
	Surgical procedures	Tracheostomy, supportive technologies, percutaneous endoscopic gastrostomy	Rates (%)

 Table 1
 ICF domains, related variables, classification and measurement considered to report patient's health condition.

IDD: intellectual and developmental disabilities.

GMFCS: gross motor function classification system.

CFCS: communication function classification system.

To describe the trend in the health condition across the thirty-year study period, data were grouped into three decades of birth: from June 1967 to May 1977, from June 1977 to May 1988, and from June 1988 to June 1997.

The study has been reviewed and approved by the Ethics Committee (Prot. N. 61/17 - CE), the article is adherent to the committee's recommendations, all participants gave their written informed consent and the study was conducted in accordance to the ethical standards of the Declaration of Helsinki (1964).

## 3. Results

### 3.1. Participants

The final sample of 464 patients was as follows: 97 patients were born from 1967 to 1977 (age range at the time of the admission: 0-16 years; 55% males, 45% females); 158 were born from 1977 to 1988 (age range: 0-15 years; 53% males, 47% females) and 209 were born from 1988 to 1997 (age range: 0-15 years; 60% males, 40% females).

#### 3.2. Body function and structure

## 3.2.1. Gestational age at birth in completed weeks and body weight at birth

Data concerning the gestational age and the body weight were retrieved for 244 participants. Considering the available data, a prevalence of moderately preterm babies with a low body weight was observed in all decades. The mean gestational age was 33 weeks in the first (n = 33, range 24–43, SD = 4.17) and in the second (n = 89, range 20–42, SD = 4.88) decades and there was a little increase to 34 weeks in the last decade (n = 122, range 24–43, SD = 4.74).

The mean body weight was 2793 g (SD = 952,89) in the first decade which was similar to the third decade (2333, SD = 1006,46), but it reduced to 2313 g (SD = 927,99) in the second. An increase in the global severity (as measured by the Disab score) was related to reduced birth gestation (F(2, 245) = 5.59, p = .004) and weight (F(2, 247) = 6.28, p = 0.004)p = .002). The association between gestational age at birth, body weight at birth and cognitive abilities was not clear; indeed patients with a severe cognitive impairment had a mean gestational age and body weight at birth higher than those with no intellectual impairment, F(3,244) = 5.79, p = .001 and F(3,246) = 6.62, p < .001, respectively.

#### 3.2.2. Part of the body affected and motor types

The prevalence (in percentages) of different localizations of impairments and the prevalence of different motor impairment observed across decades are reported in Fig. 1.

#### 3.2.3. Deformities and comorbidities

The prevalence (in percentages) of deformities (both congenital and acquired) and occurrence of comorbidities across decades are reported on Table 2.

## 3.3. Activity and participation

#### 3.3.1. Scales GMFCS, MACS E CFCS

The trend of motor, manual and communication abilities is presented in Fig. 2.

#### 3.4. Personal and environmental factors

The age of admission was 2.5 years (SD = 2.9) from 1967 to 1977, slightly reduced to 1.9 years (SD = 2.9) in the decade from 1978 to 1988, growing again to 2.8 years (SD = 2.9) in the last decade. The age of discharge gradually decreased from 18.2 years (SD = 3.3) to 16.4 years (SD = 2.9) and finally to 14.6 years (SD = 4.1). The average length of treatment also decreased from an initial 16.2 years (SD = 3.6) to 15 years (SD = 3.6) and to 12.3 years (SD = 4.7) in the last decade. The frequency of orthopedic, pharmacological, rehabilitative and surgical interventions is reported in Table 3.

#### 3.5. Global disability

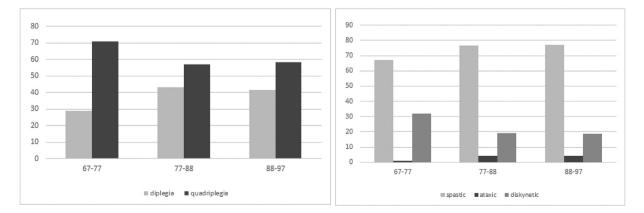
The overall Disab score indicated that most patients (44%, n = 206) had a severe disability and fell into the third band ( $\ge$ IX). One third of patients fell into the moderate band (VI–VIII; 30%, n = 140) and the remainder fell into the mild band (I–V; 25%, n = 118). The distribution of these bands is represented in Fig. 2. The worst outcome in terms of mean Disab score was associated to the presence of hemorrhage and perinatal infections, followed by hypoxia and congenital anomalies (Fig. 3).

#### 4. Discussion

This study presents data on a large group of patients with bilateral CP admitted to a network of rehabilitative centers of the North East of Italy from the end of 1960s to the end of 1990s.

Patients started rehabilitation at an average age of 2 years with little variation across decades. In accordance with Kim et al.,<sup>15</sup> the average rehabilitation starting age of 2 years remained stable until the early 2000's. The number of admitted patients gradually increased across decades.

Quadriplegia was the most frequently observed impairment. Across the decades, it gradually decreased, whereas a concurrent increase in rates of diplegia was observed. The trend of increasing diplegia was also reported in Sweden from 1979 to 1982.<sup>7</sup> Overall, this suggested a decrease



**Figure 1** Rates of diplegia and quadriplegia (on the left) and rates of spastic, ataxic and dyskinetic disorders (on the right) observed across decades.

	67–77	78–88	89—97
Acquired and/or congenital de	formities		
Middle inferior part of the b	ody		
Foot (e.g., clubfoot,	68	68	63
bunion, flat feet)			
Hip (e.g., dislocation,	20	20	14
subluxation)			
Thigh	38	32	24
Knee (e.g., genu	55	49	52
valgum)			
Spine (e.g., scoliosis,	18	19	21
spondylolisthesis)			
Superior part of the body	4	2	2
(e.g., elbow deformities,			
wrists deformities)			
Intellectual disability			
None	44	34	34
Mild	12	15	13
Moderate	42	19	20
Severe	22	33	32
Psychiatric disorders			
None	78	80	85
Anxiety	7	7	4
Intermittent explosive	5	3	3
Depression	—	1	1
Obsessive compulsive	5	2	1
Psychotic disorders due to	4	5	4
medical condition			
Anxiety and depression	1	1	1
Other comorbidities			
Breathing disorders	21	16	17
Dysphagia	14	6	15
Disorders of visual function	76	75	65
Disorders of auditory	5	3	73
function			
Communication disorders	31	23	25
Epilepsy	37	35	32

Table 2Prevalence (%) of deformities (both congenital<br/>and acquired), intellectual disability, psychiatric disorders<br/>and other comorbidities.

in motor severity in patients referred to the rehabilitation Centers. Our results differ substantially from observations in non-European countries (Australia) during the same time period. Indeed, Stanley and colleagues found an overall increase of quadriplegia and a decrease in diplegia.<sup>4</sup>

In our sample, spasticity was the most frequently observed motor type, accounting for between 67% and 77% of all cases, followed by dyskinesia (reported in 19%–32%) and ataxia (in less than 5%). This is in accordance with  $SCPE^{16}$  that showed a prevalence of spasticity (85%) followed by dyskinesia (6%) and ataxia (4%). A prevalence of spasticity was also described in non-European countries.<sup>4</sup>

After the first decade, the rate of spasticity gradually increased from 67% to 77%, as did rates of ataxia, which increased from 1% to 4%. Rates of dyskinesia almost halved (from 32% to 19%) which could be explained by the reduction of neonatal hyperbilirubinemia that was frequently associated with dyskinetic CP.<sup>17</sup>

Congenital and acquired anomalies were frequently observed and mainly involved the middle inferior part of the body, followed by the spine and the superior part of the body. In the last decade their frequency slightly reduced (except for anomalies involving the knees).

Intellectual development disorders (IDD) were present in more than half of patients. This is an important element because intellectual impairment can be considered the best predictor of survival.<sup>15</sup> In the first decade, when IDD was present, it was mainly moderate, followed by severe and mild forms. In the following two decades, the picture changed since a growing number of patients with severe IDD was registered. Although this could be explained by the general decrease observed in the body weight at birth considered in all the patients across decades, the correlation between gestational age, body weight at birth and cognitive abilities is unclear. Indeed, our results showed that patients with a severe IDD had a mean gestational age and body weight at birth higher than those with no intellectual impairment. These results are in accordance with other studies performed on children with bilateral spastic CP, which found a prevalence of severe IDD in children born at term with respect to those born preterm and found that birth weight was unrelated to cognitive impairment.<sup>18</sup>

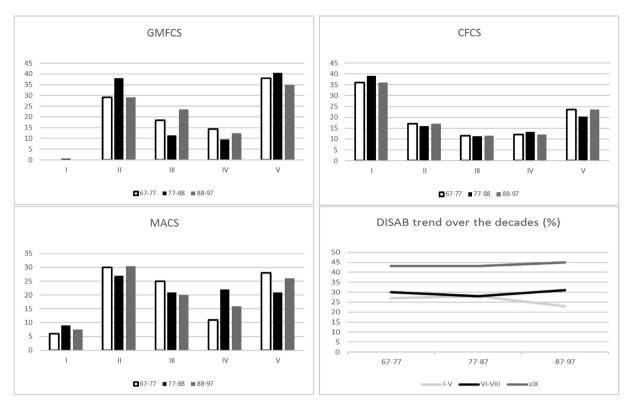
The rate of severe IDD (range 22%–33%) was similar to the rate reported by the SCPE.  $^{16}$ 

The rate of psychiatric disorders, mainly related to the occurrence of anxiety and to psychotic disorders, decreased across decades from about 20% to a final 15%. This rate is in accordance with the frequency reported by Bjorgaas et al.<sup>19</sup> This finding is relevant due to its implications on the CP global severity interacting as comorbidity, on the activities of daily living and on the quality of life from patients' and caregivers' perspectives.

The prevalent comorbidity in our sample was the visual perceptual deficit that occurred in more than half of our patients, followed by epilepsy and communication disorders. The rate of epilepsy in our sample was higher when compared to data of the SCPE<sup>16</sup> (more than 30% vs. 21%). This difference could have occurred because we considered patients with a diagnosis of epilepsy who did not necessarily have active seizures.

In all decades, the most represented GMFCS level was Level V ("Transport with manual wheelchair") followed by Level II ("Walk with limitations"). On the whole more than one third of all patients were classified as Level V and were therefore at an increased risk of mortality with respect to those classified as Levels I–IV.<sup>20</sup> Except for Level I which was almost absent, Levels from II to V generated a U-shape trend. With respect to both previous decades, in the last one a slight improvement in motor abilities was observed with a gradual decrease in participants' scoring Level V and an increase in patients scoring Level III. Patients' scoring Levels II and IV remained stable with respect to the first decade. This improving trend is in accordance with data found in a later period (from 1999 to 2010) explored by Hollung.<sup>2</sup>

Concerning manual abilities, the MACS Level II was most represented, followed by the Level V. This suggests that most patients had either good residual abilities or was completely impaired on manual functionality. Regarding communication functioning, most of patients were effective



**Figure 2** Rates across decades of different levels of scales measuring motor (GMFCS), manual (MACS) and communicative abilities (CFCS). On the bottom right of the picture the representation of the trend in global disability score over the decades.

	67–77	78–88	89–97	
Orthopedic surgery and pharmacological treatment				
Soft tissues surgery	61	57	42	
Bone surgery	27	20	10	
Pharmacological	1	2	16	
treatment				
Rehabilitative intervention	on			
Physical therapy	100	99	99	
Speech therapy	32	32	39	
Occupational therapy	73	61	62	
Other (psychotherapy,	1	1	2	
psychological or				
psychomotor				
therapy)				
Surgical procedures				
Tracheostomy	0	0	2	
Ventilator	1	0	1	
Percutaneous	1	0	5	
endoscopic				
gastrostomy				

Prevalence (%) of orthopedic, pharmacological,

Table 3

senders and receivers of information with familiar and unfamiliar partners. The CFCS scores showed no change over the decades.

Interestingly, across decades, the duration of treatment showed a steady reduction. This could be due to an amelioration in the efficacy of treatments and of health care services as well as to the increased number of multidisciplinary alternatives available to the patients in the territory.

The rate of surgery performed on soft tissues and bones decreased in favor of the pharmacological treatment. This was true especially for interventions on bones with an important reduction perhaps due to an increase in noninvasive care and use of assistive devices. Across decades an increase in the use of pharmacological treatment was observed and, reasonably, represented a noninvasive alternative for managing symptoms in these children. On the other hand, when considering all types of surgical interventions (including gastrostomy and tracheostomy), surgical procedures increased, and the increase was mainly associated to percutaneous endoscopic gastrostomy and tracheostomy. This could have played a role in improving the quality of life even in more severe patients.

Rehabilitative interventions provided were constant over the years and were mainly represented by physical, occupational and speech therapies. This is in accordance with data collected in non-European countries by Kim and colleagues,<sup>15</sup> who found a prevalence of physical therapy and occupational therapy (about 80% and 60%, respectively). Interventions on motor, verbal and activities of daily living (ADL) represent the priority in the developmental age. There could be a shift to psychiatric and/or psychological interventions at the time of transition to adult age as demonstrated elsewhere.<sup>21</sup>

Concerning the DISAB score, the rate of severe disability was highest throughout the whole period. During the first

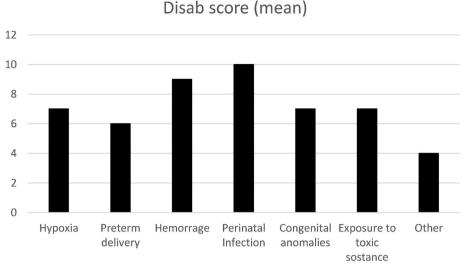


Figure 3 Relationship between Disab score and CP etiologies.

two decades a homogeneous distribution of mild and average levels of disability was observed, whereas in the last decade an increase in the rate of average disability and a concurrent decrease in mild disability were observed, thus suggesting a progressive impairment of the global severity. This mirror-trend is in accordance with the trend observed in motor abilities.

This study provides a comprehensive overview of the CP in a multicentric study across three decades. It presents to our knowledge the largest sample size of Italian population of bilateral CP published so far and it is comparable to European and non-European studies. It includes a heterogeneous sample of patients with different clinical characteristics and severity based on different points of view. Data were collected across a long period of time, which is an appropriate trend-providing factor. It includes results based on multiple outcome measures that provide a global picture of the clinical status including information such as motor, communicative, manual functioning, intellectual abilities, comorbidities and environmental factors.

The main limitations are its lack of epidemiological strength, with an evident reporting bias for CP patients being referred, with no information on the remaining population not involved in the study. Another point of weakness is the retrospective nature of the study and the lack of certain critical baseline information such as birthweight.

This study provides an overview based on the global picture of bilateral CP and how it changes over the years. It could be of interest to study the evolution of the same sample in follow up study.

## Authorship

Conception and design of study: Martinuzzi, Pellegri. Acquisition of data: Pellegri, Pizzighello.

Analysis and/or interpretation of data: Pizzighello, Vavla, Minicuci.

Drafting the manuscript: Pizzighello, Vavla, Minicuci, Martinuzzi.

Revising the manuscript critically for important intellectual content: Pellegri, Vavla, Martinuzzi.

## Financial disclosure

The authors have indicated they have no financial relationships relevant to this article to disclose.

#### Declaration of competing interest

The authors have no conflict of interests.

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