Developmental Profiles of Preschool Children With Spastic Diplegic and Quadriplegic Cerebral Palsy

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Cerebral palsy (CP) is a disorder of movement and posture control with multiple impairments. The clinical manifestations of CP vary among children. The aim of this study was to compare the developmental profiles of preschool children with either of two types of CP: spastic diplegic (SD) CP and spastic quadriplegic (SQ) CP. Relationships between the children's various developmental functions were also investigated. We recruited 137 children with spastic CP, aged 1–5 years (mean age = 3.7 ± 2.1 years), and we classified them into two groups: SD ($n = 59$) and SQ ($n = 78$). The comparison group comprised 18 children with typical development. Developmental functions were assessed in all the children, using the Chinese Child Development Inventory with the updated norms. This scale addressed eight functional domains: gross motor ability, fine motor ability, expressive language ability, concept comprehension ability, situation comprehension ability, self-help ability, personal-social skills, and general development. A development quotient (DQ) was determined for each domain as a percentage of the developmental age divided by the chronological age. The developmental profiles of the CP subtypes were found to differ. Children with SQ were found to have lower DQs than those with SD ($p < 0.01$). There was also a difference in the distribution of DQs between the SD and SQ groups, although the lowest DQ in both groups was for the gross motor domain. An uneven delay in the development of gross motor function was found in both groups of children with CP. Motor functions, including gross motor and fine motor functions, were significantly related to self-help ability. Complex and significant correlations among developmental functions were also identified in children with CP. The findings in the present study may allow clinicians to anticipate the developmental profile of children with CP on the basis of whether they have the SD or SQ subtype. This, in turn, is likely to facilitate individual assessment, goal setting, and the planning of interventions in children with CP.

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Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, which limit physical activity, and are attributed to non-progressive disturbances that occur in the developing fetal or infant brain [1]. CP is also the most common cause of physical disability in children, with

a prevalence of two per 1,000 live births [2–4]. The motor disorders associated with CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, and by epilepsy and secondary musculoskeletal problems [1,5–7]. Accordingly, CP is considered to be a multiple developmental disorder and the overall effect of CP on an individual is determined by a complex interplay of functions and impairments.

There are many instruments for assessing the bodily functions, activities, and social interaction of children with CP. For example, the Child Health Questionnaire can be used to measure the physical and psychosocial health of children above 5 years of age [8]. The Lifestyle Assessment Questionnaire is a condition-specific measurement tool developed to assess the impact of disability on the lives of children with CP; this tool contains questions that address various domains: physical independence, clinical burden, mobility, schooling, economic burden, and social integration [9,10]. The Caregiver Questionnaire measures the difficulties and satisfaction associated with being a caregiver, covering personal care, positioning/transferring, comfort, and communication [11].

Most of the previous research on this topic has focused on motor functions in children with CP [10, 12–15]; only a few studies have comprehensively addressed the developmental profiles, and the full spectrum of developmental functions in various domains (e.g. motor, speech, and social skills). The Chinese Child Development Inventory (CCDI) was designed to assess the developmental profiles of children below the age of 6 years [16,17]. A previous study suggested there was a tendency for general development to decline across subtypes of CP, from spastic hemiplegia to spastic diplegia (SD), athetoid CP, hypotonic CP, and spastic quadriplegia (SQ) [18]. In general, assessment using the CCDI has identified gross motor function to be the most affected developmental domain of children with CP [18]. However, the abovementioned study was limited by its small sample size for each CP subtype. Another study reported that the development of gross motor function was delayed in 90% of children with CP, and that 65% of these children also had delayed development in their fine motor function [19].

While the motor disability of children with the SD subtype of CP primarily involves the lower extremities, that of children with the SQ subtype involves all four limbs and the trunk. Compared with children with SQ, those with SD have better motor functions and, therefore, more opportunities to explore their environment, to learn, and to interact socially. Because the clinical manifestations of CP vary among children, it is expected that there will be differences in the development of motor functions between children with SD and those with SQ. In addition to gross and fine motor functions, we hypothesized that the developmental profiles of preschool children with SD would differ from those with SQ, with regard to the domains of communication, concept comprehension, situation comprehension, self-help ability, personal-social ability, and general development. The aim of the present study was to investigate the developmental profiles, the full spectrum of developmental functions, of preschool children with spastic bilateral CP. The relationships among developmental functions were also investigated.

Methods

Participants

The study sample group comprised children with CP, who were recruited from the rehabilitation clinic of a tertiary hospital. The inclusion criteria for the study were a diagnosis of spastic bilateral CP (either SD or SQ) and an age from 1 to 5 years. SD is a motor disability primarily involving the lower extremities, with upper motor neuron signs in the lower limbs [20]. SQ is a more complete motor disability that affects all four limbs and the trunk, and there are upper motor neuron signs in all limbs [20]. The criteria for exclusion from the study were if the children had spastic unilateral CP (e.g. hemiplegic) or another type (e.g. dyskinetic or ataxic), or if they had either a progressive neurological disorder or a severe concurrent illness or disease not typically associated with CP, such as traumatic brain injury or active pneumonia. Children who were excluded were nevertheless examined at follow-up visits. A total of 137 children with CP were recruited for this study (75 boys and 62 girls, mean age = 3.7 ± 2.1 years). They were classified into two CP subtype groups: SD ($n = 59$) and SQ ($n = 78$). The same physician performed both the diagnosis and classification of CP. To compare, we selected 18 children with typical development (TD) (13 boys and 5 girls), who were matched for age and sex with the CP group. The study protocol was approved by

Institutional Review Board for Human Studies of Chang Gung Memorial Hospital, and informed consent for participation was provided by the parents or caregivers of each child.

Assessment procedures

The developmental profiles of all the participants were assessed using the CCDI with updated norms [16]. The CCDI is a modification of the Minnesota Child Development Inventory [21], which was used in Chinese populations and commonly used with developmentally delayed children in Taiwan [18,19, 22–24]. The CCDI with the updated norms is a questionnaire containing 312 items; the items are divided into eight domains, each evaluating a different developmental function. Parents or caregivers complete the CCDI by indicating which of the listed behaviors they have observed in their child. The CCDI produces a normative score and a scaled score, yielding an age equivalent for each of the eight developmental domains. A development quotient (DQ) can be calculated for each domain as the percentage of the developmental age in the corresponding domain divided by the chronological age. The eight domains assessed by the CCDI are: (1) gross motor ability (locomotion, balance, and coordination movements); (2) fine motor ability (finger dexterity, accuracy of release, and visual–motor coordination); (3) expressive language ability (expressing oneself in interpersonal relationships); (4) comprehension-conceptual ability (comprehension of language and abstract concepts); (5) situation-comprehension ability (comprehension of non-language-based situations); (6) self-help ability (ability to manage personal daily activities); (7) personal-social skills (interpersonal relationships in social life); and (8) general development. The validity and reliability of the original CCDI have been shown to be greater than 0.83 and 0.88, respectively [17].

The demographic data (age and gender) and birth history (gestational age, birth weight, and delivery modes) were recorded. Delivery modes consisted of natural delivery and Cesarean section.

Statistical analysis

A one-way analysis of variance was used to compare the continuous variables—age, birth weight, gestational age, and DQ—among the three groups of children (SD, SQ, and TD). The *post hoc* Dunnett's T3 test was used for multiple comparisons among groups. The χ^2 test was used to compare gender ratios and mode of delivery among the groups. The Pearson's correlation was used to determine the relationships among DQs for different developmental functions. For all analyses, $p < 0.05$ was considered to be statistically significant.

RESULTS

Analyses of the demographic data revealed no significant differences in age or gender ratio among the SD, SQ, and TD groups (Table 1). Children with CP recorded significantly lower values for gestational age and birth weight than those in the TD group ($p < 0.01$) (Table 1). However, there were no significant differences in gestational age and birth weight between the SD and SQ groups (Table 1). There were also no significant differences among the three groups with regard to delivery modes.

Table 1. Demographic and birth data of children with spastic diplegic and quadriplegic cerebral palsy, and children with typical development*

	Children with CP		Children with TD ($n = 18$)	$p^{\dagger}$
	SD ($n = 59$)	SQ ($n = 78$)		
Demographics				
Age (yr)	3.9 ± 2.0	3.6 ± 2.0	4.0 ± 0.7	0.650
Sex, male	35 (59.3)	40 (51.3)	13 (72.2)	0.239
Birth history				
Gestational age (wk)	33.7 ± 5.4	35.0 ± 5.1	38.9 ± 2.8	< 0.001
Birth weight (g)	2,190 ± 961	2,359 ± 988	3,227 ± 661	< 0.001
Delivery mode, natural	35 (59.3)	42 (53.8)	6 (33.3)	0.153

*Data presented as the mean ± standard deviation or *n* (%); $^{\dagger}\chi^2$ tests (categorical data) or one-way analysis of variance (continuous data). CP = cerebral palsy; SD = spastic diplegia; SQ = spastic quadriplegia; TD = typical development.

Table 2. Development quotients for each of the eight developmental domains assessed by the Chinese Child Development Inventory in children with spastic diplegic and quadriplegic cerebral palsy, and children with typical development*

CCDI development quotient (%)	Children with CP		Children with TD ($n=18$)	$p^{\dagger}$
	SD ($n=59$)	SQ ($n=78$)		
Gross motor	47.4±21.2	25.4±16.8	92.5±10.9	<0.001
Fine motor	64.6±26.0	35.0±23.3	95.7±11.7	<0.001
Expressive language	66.3±30.5	43.8±26.0	93.9±10.7	<0.001
Concept comprehension	68.3±28.7	45.6±22.7	96.2±11.2	<0.001
Situation comprehension	63.9±28.4	33.7±19.3	97.1±11.8	<0.001
Self-help	57.6±21.6	35.7±17.7	92.8±16.6	<0.001
Personal-social	66.3±29.7	39.2±21.9	96.4±15.5	<0.001
General development	62.2±24.4	38.2±19.5	92.8±8.0	<0.001

*Data presented as the mean±standard deviation; †one-way analysis of variance. CP=Cerebral palsy; SD=spastic diplegia; SQ=spastic quadriplegia; TD=typical development; CCDI=Chinese Child Development Inventory.

As seen in Table 2, both the SD and SQ groups had lower DQs than the TD group for all developmental domains ($p<0.01$). The DQs for all developmental domains were significantly lower (by 22–30%) for the SQ group than for the SD group ($p<0.01$) (Table 2). In addition, the DQs for all developmental domains were significantly lower (by 28–45%) for the SD group than for the TD group ($p<0.01$) (Table 2).

As represented by the distribution of DQs, there was a difference in the developmental profiles of the SD and SQ groups (Figure). Within the SD group, DQs were the lowest for the gross motor domain (47%), higher for the self-help domain (58%), and the highest for the other developmental domains (62–68%) (Figure). Within the SQ group, DQs were also the lowest for the gross motor domain (25%), higher for the situation comprehension, fine motor, self-help, general development and personal-social domains (34–39%), and the highest for the other developmental domains (44–46%) (Figure).

As shown in Table 3, there were significant correlations among the different developmental domains ($r=0.665$–0.931, $p<0.01$). Gross motor function was strongly correlated with self-help ability, fine motor function and situation-comprehension ability ($r=0.831$–0.845, $p<0.01$), and fine motor function was most strongly correlated with situation-comprehension ability ($r=0.919$, $p<0.01$).

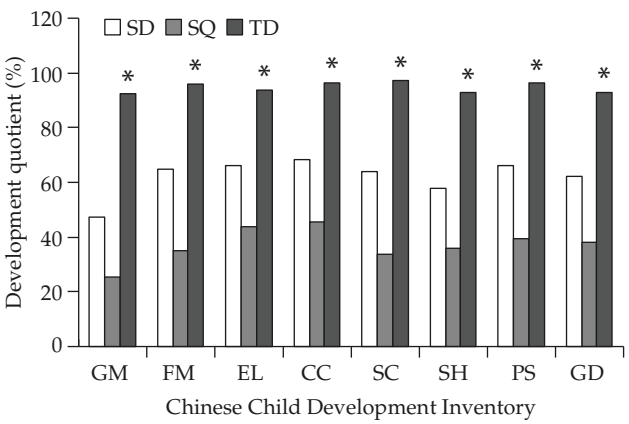

Figure. *Development quotients for the eight domains assessed by the Chinese Child Development Inventory in children with spastic diplegic and quadriplegic cerebral palsy, and children with typical development. *p<0.001. GM=Gross motor; FM=fine motor; EL=expressive language; CC=concept comprehension; SC=situation comprehension; SH=self-help; PS=personal-social; GD=general development; SD=spastic diplegia; SQ=spastic quadriplegia; TD=typical development.*

Discussion

In the present study, we found that children with different subtypes of CP had different developmental profiles. Children with SQ had lower DQs for all developmental domains than those with SD. The distribution of DQs across domains also differed between the SD and SQ groups, although in both groups, DQs were the lowest for gross motor function. Our results may reflect differences in the pathogenesis of CP between the various subtypes of this condition [25,26]. The nature of brain injury suffered determines the type of CP, which itself affects a particular range of developmental functions. The pathogenesis of CP is complex and can involve prematurity, infection, hypoxic-ischemic damage, and congenital malformations [27,28]. The gestational age at which brain development is disturbed is strongly associated with the

Table 3. Pearson's correlation coefficients for inter-relationships among the development quotients for all eight domains assessed by the Chinese Child Development Inventory in children with spastic diplegic and quadriplegic cerebral palsy

	Correlation coefficient							
CCDI	GM	FM	EL	CC	SC	SH	PS	GD
GM								
FM	0.836*							
EL	0.678*	0.811*						
CC	0.665*	0.773*	0.868*					
SC	0.845*	0.919*	0.864*	0.819*				
SH	0.831*	0.832*	0.747*	0.836*	0.848*			
PS	0.781*	0.868*	0.892*	0.821*	0.937*	0.803*		
GD	0.796*	0.891*	0.918*	0.931*	0.923*	0.890*	0.922*	

*$p < 0.01$. CCDI = Chinese Child Development Inventory; GM = gross motor; FM = fine motor; EL = expressive language; CC = concept comprehension; SC = situation comprehension; SH = self-help; PS = personal-social; GD = general development.

extent to which different brain structures are compromised, the CP subtype that develops, and the distribution of affected limbs [29,30]. Potential factors underlying these associations are age-related changes in the extent to which different glutamate receptor subtypes are expressed in the brain [30]. In full-term infants, the most common CP subtypes are hemiplegia and quadriplegia, both of which are associated with brain maldevelopment or grey matter lesions. In contrast, in preterm infants, the predominant CP subtype is SD, and it is characterized by periventricular white matter lesions [29,31]. A previous study reported that the way in which developmental functions are affected by CP depend on the particular subtype involved [18]. For example, the gross and fine motor functions of children with SQ were both worse than those of children with other CP subtypes [18]. Significant differences between CP subtypes with regard to physical functioning, bodily pain, general health, parental impact time, and physical health have also been reported [32], as has an association between the distribution of affected limbs and self-care and domestic life [33]. Findings like these may allow clinicians to anticipate the developmental profile of a child with CP on the basis of a particular subtype.

In the present study, we found differences in the developmental profiles between children with SD and SQ; these differences exceeded those associated with motor skills. Children with SQ had moderate to severe developmental delays in all the domains assessed, while those with SD had only a moderate delay in the gross motor domain and mild delays in the other domains. There are many potential reasons for this discrepancy, including differences in molecular pathways that mediate brain injuries or anatomical regions affected by brain damage [30], or in the cumulative effects of prenatal, perinatal, and postnatal events and various developmental limitations. Compared with children with SD, those with SQ have damage in more areas of the brain and greater motor disability. Children with SQ also often have difficulties with eating, speaking, and in the acquisition of other precursor skills that support learning and concept development, such as language, motor planning, and sensory processing. This affects developmental progress in children of preschool age and may almost completely account for delays in sensory processing, the ability to manipulate objects, language, and social interaction. More severe levels of motor disability are known to be associated with a greater number of accompanying impairments [5,7]. Previous studies have revealed that the number of accompanying impairments differs significantly between the CP subtypes, with the highest prevalence being in children with SQ [7,34,35].

Developmental delays in children with SQ and SD did not occur evenly across the various functions assessed in this study. Gross motor function was particularly impaired in both groups. It is not surprising that in both groups, the lowest DQ was for the gross motor domain as motor impairment is a core feature of CP. The relatively high level to which self-help ability was also impaired may reflect the strong relationship between gross motor function and self-help ability. Our findings are supported by a previous study in which the development of gross motor

functions and self-help abilities was most delayed in children with CP [18].

In the present study, motor functions, including gross motor and fine motor functions, were significantly related to self-help ability. This may be explained by the motor impairments in children with CP, including poor muscle strength, limited range of motion, increased spasticity, poor selective motor control, and compromised coordination, which limit the extent to which they can manage their personal daily activities. The self-help ability—as evaluated by the CCDI with items addressing grooming, dressing, and eating—was highly dependent on the extent to which manual functions could be performed. Fine motor skills involve the subtle and organized movement of small muscles that are required for tasks such as grasping, releasing, and hand-to-hand transfer. Skills like these are a cumulative result of numerous factors: cognitive, sensory, perceptual, cultural, and motor. Previous studies have also reported more severe levels of gross motor disability associated with a reduced accomplishment in self-care activities [36]. Accordingly, the Gross Motor Function Classification System could be used to predict everyday functioning, particularly in terms of self-care and social functions [36]. Upper limb skills are reported to be correlated significantly with self-care ability, mobility, and overall functional skills in children with CP [37]. Therefore, treatment strategies for enhancing the self-care ability of children with CP should focus on integrating fine and gross motor interventions.

We found significant correlations among various developmental functions in children with CP. These correlations are likely to hint at complex interactions among developmental functions. Compared with children with SD, those with SQ experience greater difficulties in exploring their environment, communicating, learning, receiving education, participating in social interaction, and taking care of themselves. These difficulties arise from severe motor limitations and associated problems, such as cognitive impairment and epilepsy, which deleteriously affect communication, self-care, and domestic and social life [32]. The lack of social interaction retards the development of social skills, thereby deepening social isolation [38]. Quantitative correlations have been found to exist between each of gross motor function, fine motor function, mental retardation, and three factors predictive of restricted participation: mobility, education, and social relations [34]. The relationships among different developmental functions are complex. Nevertheless, it is clear that gross motor function dominates all other factors in influencing the general development of children with CP. The health status of children with CP is significantly affected by the extent to which their gross motor functions are impaired, particularly in relation to physical functioning, bodily pain, behavior, general health, family activities, and physical health [32,39]. A number of studies have reported that the participation of children with CP in daily activities decreases with an increase in the severity of their gross motor impairment [10,12,34,36]. It has also been shown that more severe levels of motor disability are associated with less favorable progress of gross motor function in the future [40].

The limitations of the present study are associated with participant characteristics and the study design. We recruited participants with either the SD or SQ subtype of CP, but excluded those with hemiplegic or other subtypes. Our results can therefore not be generalized to all cases of CP. In addition, because of the relatively small sample size, we could not reasonably further classify children within the SD and SQ groups in terms of condition severity. Another limitation is our measurement tool. There is a ceiling effect which appeared in each domain of the CCDI with the updated norms. Despite the limitations, the present study convincingly demonstrates the developmental profiles of children with SD and SQ. Our findings may be of assistance in the individual assessment of and goal setting and planning of interventions for children with CP.

In conclusion, CP is a disorder of movement and posture control with multiple impairments. The developmental profiles of children with CP differed between the SQ and SD subtypes. An uneven delay in the development of gross motor function was found in children with SQ and SD. In addition, motor functions, including gross motor and fine motor functions, were significantly related to self-help ability. Complex and significant correlations were found among a number of developmental functions in children with CP. These findings may allow clinicians to anticipate the developmental profiles of children with CP on the basis of whether they have the SD or SQ subtype. This, in turn, is likely to facilitate individual assessment, goal setting, and the planning of interventions for children with CP.

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痙攣型雙邊麻痺與四肢麻痺腦性麻痺學前兒童之發展輪廓

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腦性麻痺是一種運動和姿勢控制失調的問題，同時合併有許多的障礙，其臨床上的表現也有很多變化。本研究的目的是在比較痙攣型雙邊麻痺與四肢麻痺腦性麻痺學前兒童之發展，並且分析各種不同發展功能間的相關情形。本研究以 **137** 位一至五歲（**3.7** ± **2.1** 歲）痙攣型腦性麻痺學前兒童分為雙邊麻痺型（**59** 位）和四肢麻痺型（**78** 位）。另取 **18** 位人口學資料與實驗組相近的正常兒童為對照組。並使用中華兒童發展量表及其新常模來評估八個行為項目的發展：粗動作、精細動作、溝通表達、概念理解、環境理解、身邊處理、人際社會、及一般發展。發展商數是以發展年齡除以實際年齡的百分比來計算。研究資料分析的結果顯示，不同類型的腦性麻痺有著不同的發展輪廓。四肢麻痺型兒童在各面向的發展商數均低於雙邊麻痺型兒童（$p < 0.01$）。雖然兩組兒童的粗動作發展商數都呈現最低分，但在其他面向的發展商數分佈仍不同。兩組兒童同時呈現特別嚴重的粗動作發展遲緩。粗動作和精細動作的運動功能皆和身邊處理能力有顯著的相關性。除此之外，腦性麻痺兒童的各種不同發展功能間有著複雜且顯著的相互關係。本研究的結果可以提供給臨床專業人員作為預估不同類型腦性麻痺兒童發展輪廓的參考，並且可以用來協助腦性麻痺兒童的評估、目標設定、及治療計畫的訂定。

關鍵詞：腦性麻痺，腦性麻痺類型，發展功能，運動功能

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