

## Comparison of Developmental Pattern Change in Preschool Children with Spastic Diplegic and Quadriplegic Cerebral Palsy

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**Background:** This study compares the longitudinal change of developmental patterns in preschool children with spastic diplegic (SD) and spastic quadriplegic (SQ) cerebral palsy (CP).

**Methods:** Sixty children with spastic CP, aged 1-5 years ( $3.2 \pm 1.2$  years), were classified into 2 groups: SD ( $n = 29$ ) and SQ ( $n = 31$ ). Gross Motor Function Classification System (GMFCS) levels were classified during the initial assessments. Developmental profiles, including development quotients (DQs) of gross motor, fine motor, expressive language, concept comprehension, situation comprehension, self help, personal social and general development, were evaluated on initial and final assessments and an average of one year later. The DQ change index (%) was calculated as  $100\% \times (\text{final DQ} - \text{initial DQ})/\text{initial DQ}$ .

**Results:** Children with SQ had lower DQs in all developmental functions than those with SD on both assessments ( $p < 0.01$ ). The DQ distributions of developmental profiles were different in SD and SQ groups, although both groups displayed the lowest DQs in the gross motor domain. As indicated by the DQ change index, most DQs increased with age in children with SD; however, most decreased with age in children with SQ ( $p < 0.05$ ).

**Conclusion:** These findings suggest different CP subtypes demonstrate various development profiles. The evolution of developmental patterns with age was different in children with various CP subtypes.

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**Key words:** cerebral palsy, developmental function, outcome, longitudinal study

Cerebral palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances that occur in the devel-

oping fetal or infant brain.<sup>(1,2)</sup> Spastic CP is the most common type, accounting for 70% to 85% of all CP cases.<sup>(3)</sup> Spastic CP is further classified into diplegic, hemiplegic, quadriplegic, and monoplegic subtypes

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based on the topographic distribution of the affected areas of the body.<sup>(3)</sup> 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>(2)</sup> Furthermore, the accompanied disturbances in children with CP further influence developmental functions of the affected child.

Most researches have investigated the longitudinal study of functions in adolescents or adults with CP;<sup>(4-8)</sup> however, few researches investigate these functions in children with CP.<sup>(9-12)</sup> In adults with CP, there was a trend toward a deterioration of physical, social, and emotional well-being with increasing age.<sup>(4-7,13)</sup> The diminution in health status in adults with CP occurs concomitantly with a steady decline in the availability of comprehensive health services with age.<sup>(4-6)</sup> The previous work compared walking function in adults with cerebral palsy (CP) 7 years after an initial survey and found that the increased prevalence of deteriorated walking was significantly associated with bilateral spastic CP, pain, fatigue, and reduced balance.<sup>(7)</sup> In adolescents with CP, between 50% and 73% of the variance for overall quality of life scores was stable over 1 year.<sup>(8)</sup> Voorman et al observed that the gross motor function of some children with CP aged 9 to 15 years deteriorated, while that of the others improved or remained stable depending on their characteristics.<sup>(9)</sup>

Although some researchers have carried out longitudinal studies of motor functions and health status in children with CP,<sup>(9-12)</sup> as yet, no one has studied the longitudinal course of developmental profiles with a full spectrum of developmental functions in various domains in preschool children with CP. The clinical manifestations may evolve as children with CP mature.<sup>(14)</sup> We hypothesize that the course of developmental patterns in preschool children with CP evolves with their age depending on CP subtypes. This study compares the longitudinal change of developmental patterns in preschool children with spastic diplegic (SD) and spastic quadriplegic (SQ) CP.

## METHODS

### Participants

Children with CP from the Rehabilitation Department of our hospital were recruited for a lon-

gitudinal study. The inclusion criteria for the study were a diagnosis of CP with SD or SQ and an age of between 1 to 5 years. SD is defined as motor disability primarily involving the lower extremities, with upper motor neuron signs in the lower limbs.<sup>(15)</sup> SQ is defined as massive total motor disability involving all four limbs and trunk, with upper motor neuron signs in all limbs.<sup>(15)</sup> The exclusion criteria of the study included the presence of a progressive neurological disorder or a severe concurrent illness or disease not typically associated with CP, such as traumatic brain injury or active pneumonia. These children were studied further at follow-up visits. Ultimately, 60 children with spastic CP were enrolled. The children were classified into 2 groups based on CP subtypes: SD (n = 29) and SQ (n = 31). The Institutional Review Boards for Human Studies at Chang Gung Memorial Hospital approved this protocol, and all the participants' parents or caregivers provided informed consent.

### Assessment procedures

Developmental profiles and motor severity assessments were performed for all children during their initial visit. Further assessments were arranged at follow-up visits which were, on average, one year ( $1.1 \pm 0.3$  year) after the initial visits. Developmental profiles were assessed using Chinese Children Developmental Inventory (CCDI),<sup>(16)</sup> widely used to assess developmental profiles in children with developmental delay in Taiwan.<sup>(17-19)</sup> The CCDI, a 320-item questionnaire, consisted of statements describing the behavior of children to which parents/caregivers are asked to answer "yes" or "no" if the child has or has never performed the specified behavior. The CCDI contains a normative score, which yields age equivalents in eight domains of developmental function. These eight domains include gross motor (34 items), fine motor (44 items), expressive language (54 items), concept comprehension (67 items), situation comprehension (44 items), self help (36 items), personal social (34 items), and general development (131 items). The general developmental domains integrate 7 items and 124 items, which are introduced in the other 7 domains. The validity and reliability of the CCDI were greater than 0.83 and 0.88, respectively.<sup>(16)</sup>

The severity of the motor disturbance in CP was classified using the Gross Motor Function

Classification System (GMFCS).<sup>(20)</sup> The GMFCS grades the self-initiated movement of CP patients with particular emphasis on their functional abilities (sitting, crawling, standing, and walking) and their requirement for assistive devices (e.g., walkers, crutches, and canes), and wheeled mobility. The GMFCS employs a 5-point scale (I–V) ranging from “independent” (level I) to “dependent” (level V). The demographic data, including age and gender, were recorded.

### Data and statistical analysis

The development quotient (DQ) was determined as a percentage of the development age divided by the chronological age. Due to a high DQ variability among children with CP on their initial assessment, this study used the DQ change index to measure the change in developmental profiles on follow-up. The DQ change index (%) is calculated using the following equation:  $100\% \times (\text{final DQ} - \text{initial DQ}) / \text{initial DQ}$ . Positive values of DQ change index indicate that DQs on final assessments as compared with initial assessments have increased, while negative values of DQs change index indicate they have decreased.

An independent *t*-test was used to compare continuous data (age and DQ change index) between the

two groups. The repeated measure ANOVA was used to measure the DQ differences on initial and final assessments. The between factor denotes the group difference and within factor denotes both assessment differences. Differences in gender between the groups were determined via the Chi-square test. Differences in the GMFCS levels between the groups were determined via the Mann-Whitney test. A *p* value of *p* < 0.05 was considered to be statistically significant.

## RESULTS

There was no significant difference between the demographic data of the 2 groups (Table 1). The SD group had a worse GMFCS level than the SQ group (*p* < 0.01). Approximately 93% of the SD group children were categorized as having GMFCS levels I and II. However, approximately 81% of the SQ group children had a GMFCS level higher than level II.

In the SQ group, the DQs for all developmental functions were lower than those for the SD group on initial and final assessments (*p* < 0.001) (Table 2). However, the repeated measure ANOVA showed no significant differences on both assessments. The DQ distributions of developmental profiles were different in the SD and SQ groups on their initial and final

**Table 1.** Demographic Data, Follow-up Interval, and Motor Severity of Children with Spastic Diplegic and Quadriplegic Cerebral Palsy (CP)

Data	CP groups				<i>p</i>
	Diplegia (n = 29)		Quadriplegia (n = 31)		
	Mean (SD)	N (%)	Mean (SD)	N (%)	
Demographic					
Age	3.2 (1.2)		3.2 (1.3)		0.931
Sex: Male		18 (62.1)		20 (64.5)	0.844
Follow-up interval (years)	1.1 (0.4)		1.1 (0.3)		0.918
GMFCS					< 0.001
Level I		19 (65.5)		0 (0.0)	
Level II		8 (27.6)		6 (19.4)	
Level III		2 (6.9)		5 (16.1)	
Level IV		0 (0.0)		11 (35.5)	
Level V		0 (0.0)		9 (29.0)	

**Abbreviation:** GMFCS: Gross Motor Function Classification System.

Values are expressed as mean (standard deviation) or number (%).

The Chi-Square or Mann-Whitney tests were selected for categorical data analysis, and an independent *t*-test was selected for continuous data analysis.

**Table 2.** The Developmental Functions Measured by Chinese Child Development Inventory (CCDI) on Initial and Final Assessments of Children with Spastic Diplegic and Quadriplegic Cerebral Palsy (CP)

CCDI	Initial assessment		Final assessment		<i>p</i> (between groups)	<i>p</i> (within assessments)
	CP groups		CP groups			
	Diplegia (n = 29) Mean (SD)	Quadriplegia (n = 31) Mean (SD)	Diplegia (n = 29) Mean (SD)	Quadriplegia (n = 31) Mean (SD)		
DQ (%)						
GM	52.4 (14.6)	23.7 (10.1)	51.2 (14.1)	20.9 (13.5)	< 0.001	0.158
FM	82.0 (19.4)	40.3 (23.4)	88.2 (26.1)	40.2 (26.8)	< 0.001	0.221
EL	84.8 (28.9)	53.7 (30.2)	84.7 (23.1)	49.1 (27.8)	< 0.001	0.270
CC	88.3 (31.2)	60.3 (35.4)	95.2 (32.4)	54.2 (32.3)	< 0.001	0.866
SC	75.2 (18.4)	39.9 (25.1)	77.3 (22.5)	37.9 (23.3)	< 0.001	0.968
SH	64.2 (17.3)	33.6 (15.4)	71.7 (20.5)	30.7 (18.6)	< 0.001	0.227
PS	70.8 (19.5)	42.2 (21.5)	71.3 (18.2)	36.5 (19.0)	< 0.001	0.176
GD	86.6 (22.5)	51.2 (24.5)	89.0 (22.7)	45.4 (24.5)	< 0.001	0.312

**Abbreviations:** GM: Gross Motor; FM: Fine Motor; EL: Expressive Language; CC: Concept Comprehension; SC: Situation Comprehension; SH: Self Help; PS: Personal-Social Development; GD: General Development.

Values are expressed as mean (standard deviation).

The repeated measure ANOVA was selected for data analysis.

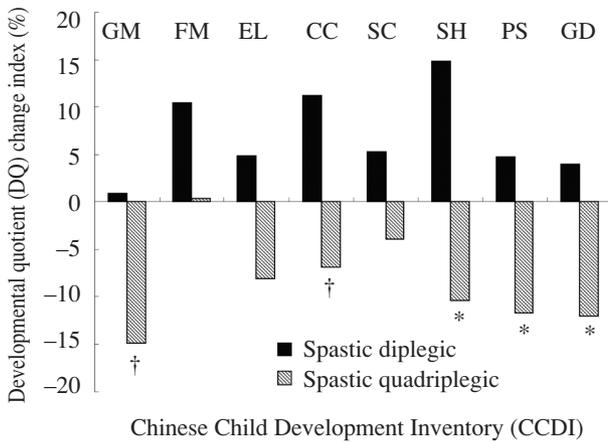
assessments, although both groups displayed lowest DQs in the gross motor domain. Development profiles in the SD group demonstrated lowest DQs in the gross motor domain (initial: 52%; final: 51%), and higher DQs in fine motor, expressive language, concept comprehension, and general developmental domains (initial: 82~88%; final: 85~95%) on both assessments. Developmental profiles in the SQ group displayed lowest DQs in the gross motor domain (initial: 24%; final: 21%), and higher DQs in expressive language and concept comprehension domains (initial: 54~60%; final: 28~32%) on both assessments. The DQs of gross motor and fine motor domains in SQ group were lower than those in the SD group by 29~30%, and 42~48%, respectively, on both assessments.

The DQ change indices of all developmental domains, except those of the fine motor and situation comprehension domains, between the two groups differed significantly ( $p < 0.05$ , Fig. 1). The DQs change indices of all developmental functions were positive values (0.9~14.8%) in the SD group; however, except for fine motor domain, they were negative values (-3.9~-14.9%) in the SQ group (Fig. 1). The

DQ change indices of fine motor in the SQ group and those of gross motor in the SD group were 0~1% (Fig. 1). As indicated by DQs change indices, the DQs of the expressive language, situation comprehension, personal social, and general development domains increased by 4~5%, and those of the concept comprehension, fine motor, and self help domains increased by up to 10~15% DQ in the SD group. However, the DQs of the expressive language, concept comprehension, and situation comprehension domains decreased by 4~8%, and those of the gross motor, self help, personal social, and general development domains decreased up to 10~15% in the SQ group (Fig. 1).

## DISCUSSION

The evolvement of developmental patterns with age was different in children with various CP subtypes. As indicated by the DQ change index, most DQs increased with age in children with SD; however, most decreased with age in children with SQ. Namely, developmental functions did not increase proportionally with increasing age in children with



**Fig. 1** The development quotient (DQ) change index measured using the Chinese Child Development Inventory (CCDI) in children with spastic diplegic and quadriplegic cerebral palsy. \*:  $p < 0.01$ ; †:  $p < 0.05$ . The DQ change index (%) is calculated according to the following equation:  $100\% \times (\text{final DQ} - \text{initial DQ}) / \text{initial DQ}$ . Abbreviations used: GM: Gross Motor; FM: Fine Motor; EL: Expressive Language; CC: Concept Comprehension; SC: Situation Comprehension; SH: Self Help; PS: Personal-Social Development; GD: General Development.

SQ, unlike the results obtained for children with SD. This result may have various pathogenesis in different CP subtypes.<sup>(21,22)</sup> The pathogenesis that caused CP was complex, and involved factors such as extreme prematurity, infection, and hypoxia-ischemia damage.<sup>(23)</sup> The vulnerability of different brain structures and types of disability associated with CP are strongly influenced by the gestational age at which development is disturbed.<sup>(24)</sup> Brain maldevelopments and grey matter lesions were more often seen in term than in preterm born children with CP, while periventricular white matter lesions occurred significantly more often in preterm than in term-born children.<sup>(25)</sup> Children with SD were often associated with white matter disorders in premature infants.<sup>(21)</sup> Children with SQ often had periventricular leukomalacia in preterm children, term-type lesion and brain anomalies in full term children.<sup>(26,27)</sup> Accordingly, the brain damage in SQ CP was more complicated and diffuse than that in SD CP. These findings may enable clinicians to anticipate the long-term course of developmental patterns in children with CP having CP subtypes.

Different CP subtypes demonstrated various

developmental profiles. In this study, children with SQ had lower DQs in all developmental functions than those with SD. The DQ distributions of developmental profiles were different in SD and SQ groups, although both groups displayed the lowest DQs in the gross motor domain. Brain damage in CP determines the CP subtypes, which is, in turn, associated with various developmental functions. Furthermore, various CP subtypes were related to associated problems, such as cognition, speech, and social problems. Previous studies indicated quadriplegia was associated with poor motor functions,<sup>(9)</sup> and limb distribution was associated with self-care and domestic life in children with CP.<sup>(28)</sup>

Gross motor development could proportionally increase with age in children with SD CP, while it could not increase proportionally with age in children with SQ CP. In this study, as indicated by the DQ change indices, the DQ of gross motor showed nearly no change in children with SD on follow-up assessment, while they were reduced by 15% in children with SQ. A previous study has shown that gross motor functions of severely impaired children tended to decrease,<sup>(9)</sup> while that of the mildly impaired children increased or remained stable.<sup>(9)</sup> This difference could be attributed to the fact that children with SQ had greater secondary motor impairments and associated problems, such as cognition, speech, and social problems, and seizure disorders, than those with SD. The secondary musculoskeletal impairments and changes in motor functions may occur over time in individuals with CP.<sup>(29)</sup>

In this work, the DQs of comprehension, speech, and social functions, and self care domains increased with age in children with SD; however, they decreased with increasing age in children with SQ. The reasons for this difference could be the variations in multiple factors between children with SD and SQ, such as the differences in a child's characteristics, family support, participation, environmental factors, and health services. Children with SQ experienced greater difficulties in exploring their environment, communicating, learning, receiving education, participating in social interactions, and taking care of themselves due to severe motor limitations and associated problems than children with SD. A previous study revealed that the physical functioning of children with CP declined with time; however, all other subscales of the Child Health Questionnaire were

stable over the course of 1 year.<sup>(10)</sup> The accompanying problems, such as cognitive or mobility impairment, were associated with self-care and domestic life in children with CP,<sup>(28)</sup> and the lack of involvement in social relationships could have contributed to the poor development of social skills and social isolation.<sup>(6)</sup>

### Limitations

The limitations of this study include the study design and subject characteristics. The normative score (DQ), not raw score (DA) was used in this study. Children with disabilities often lag behind with increasing age when using the normative score even if they made progress when using the raw data. The etiology of CP, social economic status, and intervention strategies were not analyzed in this study. Not all children with CP received the brain imaging studies in our hospital. Children with CP in both groups received varied amounts of rehabilitation program and / or combinations of other complementary and alternative medicine. The enrollment criterion was spastic bilateral CP. Subjects with the other CP subtypes, such as spastic hemiplegia, or ataxia, were not recruited in this study. Therefore, the results of this study can not be generalized to all cases of CP. Despite this limitation, this study convincingly demonstrated the developmental pattern changes in children with spastic bilateral CP of various CP subtypes.

### Conclusion

As indicated by the DQ change index, most DQs increased with age in children with SD; however, most decreased with age in children with SQ. Children with SQ had lower DQs in all developmental functions than those with SD. The DQ distributions of developmental profiles were different in SD and SQ groups, although both groups displayed the lowest DQs in the gross motor domain. These findings suggest the evolvement of developmental patterns with age was different in children with various CP subtypes. Different CP subtypes demonstrated various developmental profiles. Therefore, the CP subtypes could be employed to identify preschool children with spastic CP who are at risk of experiencing deterioration of DQs. Future researches shall focus on the long-term study of the developmental patterns and therapeutic strategy planning for chil-

dren with CP.

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## 比較痲痺型雙邊麻痺與四肢麻痺腦性麻痺學前兒童之發展改變

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**背景：**以縱貫性研究比較痲痺型雙邊麻痺與四肢麻痺腦性麻痺學前兒童之發展改變。

**方法：**本研究以 60 位一至五歲 (3.2 ± 1.2 歲) 痲痺型腦性麻痺學前兒童分為雙邊麻痺型 (29 位) 和四肢麻痺型 (31 位)。初次評估時，以粗大運動功能分級系統 (Gross Motor Function Classification System) 將各兒童分為不同等級。另於初次及追蹤評估時測量各兒童的發展面向，平均追蹤時間約一年，評估面向包括粗動作、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，並以發展商數 (development quotient) 量化各面向的發展程度，以發展商數改變指數 (developmental quotient change index, %) 評估前後兩次發展商數的差異。

**結果：**四肢麻痺型兒童在初次及追蹤評估時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，但兩組兒童在各面向的發展商數分佈仍有不同。由發展商數改變指數發現，雙邊麻痺型兒童大部份的發展商數隨年齡增長而增加，而四肢麻痺型兒童大部份的發展商數隨年齡增長而減少 ( $p < 0.05$ )。

**結論：**本研究發現不同類型痲痺型腦性麻痺兒童呈現不同的發展功能，發展功能的改變在不同類型痲痺型腦性麻痺孩童隨年齡增長亦有不同程度的改變。  
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**關鍵詞：**腦性麻痺，發展功能，預後，縱性追蹤研究

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