Relationships between Developmental Profiles and Ambulatory Ability in A Follow-up Study of Preschool Children with Spastic Quadriplegic Cerebral Palsy

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Background: To investigate the follow-up course of developmental profiles in preschool children with spastic quadriplegic (SQ) cerebral palsy (CP) who had varying ambulatory abilities.

Methods: Forty-eight children with SQ CP between 1 and 5 years old were classified into 2 groups, the ambulatory and non-ambulatory groups, based on Gross Motor Function Classification System (GMFCS) levels during the initial assessment. The developmental profiles, consisting of development quotients (DQs) of 8 domains, were evaluated during the initial assessment and the final assessment one year later. The DQ change index (%) was calculated as 100% × (final DQ – initial DQ)/initial DQ.

Results: The DQs of all developmental domains in the non-ambulatory group were lower than those in the ambulatory group on both initial and final assessments ($p < 0.01$). As indicated by the DQ change indices, most DQs in the ambulatory group decreased slightly, whereas those in the non-ambulatory group decreased considerably ($p < 0.05$). Furthermore, fine motor function increased proportionally with age in the ambulatory group, but not in the non-ambulatory group.

Conclusion: The DQs of the developmental profiles varied in preschool CP children with different ambulatory abilities. The course of developmental profiles in preschool children with SQ CP evolves with age and relates to the degree of ambulatory function. Knowledge of these developmental profiles may be helpful in understanding, predicting, and managing the developmental problems of these children.

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

Cerebral palsy (CP) is a common cause of childhood disability.[1-3] CP describes a group of disorders that affect movement and posture, cause activity limitation, and is attributed to non-progressive dis-

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turbances that occur in the developing fetal or infant brain.[4,5] The motor disorders of CP are often accompanied by disturbances in sensation, perception, cognition, communication, and behavior, and by epilepsy and secondary musculoskeletal problems.[5] On the basis of the predominant neuromotor abnormalities, CP is classified into several groups, such as spastic, dyskinetic, and ataxic.[6] On the basis of the anatomical classification, spastic CP is further classified into 4 subtypes, namely, diplegic, hemiplegic, quadriplegic, and monoplegic.[7] Previous classification schemes included only the affected extremities and required a subjective comparison of the severity of the disorder in the arms and legs. The Surveillance of Cerebral Palsy in Europe group (SCPE) recommended an alternative classification, unilateral versus bilateral motor involvement.[6] Spastic quadriplegic (SQ) CP is frequently associated with other problems, which further influence the developmental profiles and health status in these children.

Various clinical manifestations may be observed in CP, even with the same CP subtype. Although CP is caused by a non-progressive form of brain damage, its clinical manifestations may evolve as children with CP mature.[8] Problems associated with CP, secondary musculoskeletal problems, and environmental factors can influence the developmental course of children with CP. Furthermore, secondary musculoskeletal impairment and changes in motor function related to multiple factors may gradually occur in CP patients.[9-13] A previous investigation reported that the development of gross motor function in children with CP decreased with increasing age.[14] There is substantial within-stratum variation in gross motor development in children with CP, providing parents and clinicians with a means to plan interventions and to judge progress over time.[15] In one study, the rate of motor improvement and top achievements over the years differed according to the severity of the motor impairment.[16] The gross motor development reached a plateau at the age of 6-7 years.[16] Adults with CP display a trend of deterioration of physical, social, and emotional well-being with increasing age.[17-20] Although numerous longitudinal studies have been performed on developmental function in adolescents and adults with CP,[17-20] the complete spectrum of developmental function in children with CP has seldom been addressed.[14-16,21]

Thus far, no studies have been conducted on the longitudinal course of developmental profiles and the complete spectrum of developmental function in various domains, including the domains for motor, speech, personal-social skills, and self-care activities, in preschool children with CP. Elucidation of the longitudinal course of developmental profiles in children with SQ CP can enable clinicians to predict their developmental profiles and determine more flexible strategies for treating children with CP of varying degrees of severity. We hypothesize that the course of developmental profiles in preschool children with SQ CP evolves with age and depends on the degree of ambulatory function. Those with non-ambulatory SQ CP make less progress in developing skills than those with ambulatory SQ CP. This study investigated the follow-up course of developmental profiles in preschool children with SQ CP and varying ambulatory abilities.

METHODS

Participants

We recruited preschool children with SQ CP from the rehabilitation department of a tertiary hospital for the follow-up study. The diagnosis and classification of CP was performed by the same physiatrist. The inclusion criteria were as follows: (1) diagnosis of SQ CP; and (2) age between 1 and 5 years old. SQ is defined as massive total motor disability involving all four limbs and the trunk, with upper motor neuron signs in all limbs. The exclusion criteria included the presence of dyskinetic or ataxic CP, spastic hemiplegia or diplegic CP, a progressive neurological disorder, severe concurrent illness, or disease not typically associated with CP, such as traumatic brain injury or active pneumonia; these children were examined in each assessment. Ultimately, 48 children with spastic CP (mean age, 3.1 y; SD, 1.0 y) were enrolled. On the basis of the Gross Motor Function Classification System (GMFCS) levels, the children were classified into 2 groups:[22] ambulatory (GMFCS levels II–III, n = 20) and non-ambulatory (GMFCS levels IV–V, n = 28) groups. The study protocol was approved by the Institutional Review Board for Human Studies at our hospital, and all participants' parents or caregivers provided informed consent.

Assessment procedures

Developmental profiles and motor severity assessments were performed for all children during their initial visit. The developmental profiles were reassessed at follow-up visits, i.e., at an average of approximately 1 y (mean, 1.1 y; SD, 0.2 y) after the initial visits. The motor severity assessment for CP was performed by the same physiatrist. The developmental profiles were assessed using the Chinese Child Development Inventory (CCDI),[23] which is widely used for children with developmental delay in Taiwan.[24-27] This inventory is used as a modification of the Minnesota Child Development Inventory (MCDI) in Chinese populations.[28]

The CCDI is a standardized instrument; it comprises a 320-item questionnaire that is divided into 8 scales that evaluate the 8 domains of functional development. Parents or caregivers complete this questionnaire by indicating which of the listed behaviors they have observed in their child. The CCDI contains a normative score, which yields age equivalents of 8 domains of the developmental functions. The development quotient (DQ) in each domain is calculated as the percentage of the developmental age in the corresponding domain divided by the chronological age. The 8 domains include gross motor ability (34 items), fine motor ability (44 items), expressive language ability (54 items), concept-comprehension ability (67 items), situation-comprehension ability (44 items), self-help ability (36 items), personal-social skills (34 items), and general development (131 items). The general development domain contains 124 items which are selected from the other 7 domains. The gross motor domain is useful in the evaluation of locomotion and related balance and coordination movements; the fine motor domain in visual-motor coordination; the expressive language domain in the ability to express self in interpersonal relationships; the concept comprehension domain in the ability to comprehend language and abstract concepts, the situation comprehension domain in the comprehension of certain situations (language is not included), the self help domain in the development of the ability to manage personal daily activities, and the personal-social domain in interpersonal relationships in social life. The validity and reliability of the CCDI were greater than 0.83 and 0.88, respectively.[23]

The motor severity of CP was classified using the GMFCS.[22] The GMFCS grades the self-initiated movement of CP patients, particularly emphasizing functional abilities (sitting, crawling, standing, and walking) and requirements of patients in terms of assistive devices (e.g., walkers, crutches, and canes) and wheeled mobility. Furthermore, the GMFCS employs a 5-point scale (I–V) ranging from "independent" (level I) to "dependent" (level V). Baseline data on the children, including age, and gender, were recorded.

Data and statistical analysis

Because of the high DQ variability among children with CP during the initial assessment, this study used the DQ change index to measure the change in developmental profiles on follow-up. The DQ change index (%) was calculated using the following equation: 100% $\times$ (final DQ – initial DQ)/initial DQ. Positive values for the DQ change index indicate that the DQ increased at the time of final assessment from that at the time of initial assessment, while negative values indicate that the DQ decreased. An independent *t*-test was used to compare continuous data (age and DQ change index) between the two groups. Repeated measures ANOVA was used to measure the DQ differences on initial and final assessments. The between-subject factor denoted the group differences and within-subject factor denoted time (both assessments) differences. Differences in gender between the groups were determined via the chi-square test. A p value <0.05 was considered statistically significant.

RESULTS

No significant differences were observed in the baseline data between the 2 groups (Table 1). Approximately 55% and 45% of the children in the ambulatory group were categorized as having GMFCS levels of II and III, respectively. However, approximately 46% and 54% of children in the non-ambulatory group were classified as having GMFCS levels of IV and V, respectively (Table 1).

Repeated measures ANOVA showed significant differences in the DQs on both assessments of both CP groups ($p < 0.05$, Table 2). In the initial and final assessments, the DQs of all developmental functions in the non-ambulatory group were lower than those in the ambulatory group ($p < 0.001$, Table 2). The

Table 1. Baseline and Follow-up Data in Ambulatory and Non-Ambulatory Children with Spastic Quadriplegic Cerebral Palsy (CP)

Data	CP groups Ambulatory (n = 20)	CP groups Non-ambulatory (n = 28)	*p*
Age	3.2 ± 1.1	2.9 ± 1.0	0.252
Sex			0.096
Male	8 (40.0)	18 (64.3)	0.096
Female	12 (60.0)	10 (35.7)	
Follow-up interval (years)	1.1 ± 0.2	1.1 ± 0.2	0.659
GMFCS			< 0.001
Level II	11 (55.0)		
Level III	9 (45.0)		
Level IV		13 (46.4)	
Level V		15 (53.6)	

Abbreviation: GMFCS: Gross Motor Function Classification System.

Values are expressed as mean ± SD or n (%).

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

DQ distributions of the developmental profiles were similar in both the ambulatory and non-ambulatory groups, i.e., they were lowest in the gross motor and self-help domains and highest in the expressive language, concept-comprehension, and general development domains (Table 2).

The DQ change indices of all development domains except those of the expressive language, concept-comprehension, and personal-social domains differed significantly between the 2 groups ($p < 0.05$, Fig. 1). The DQ change indices of most development domains except the fine motor domain were –2.7~ –10.3% in the ambulatory group, however, the indices of all developmental functions in the non-ambulatory group were –12.1 ~ –27.2% (Fig. 1). As indicated by the DQ change indices, the DQ of the gross motor and self-help domains in the ambulatory group decreased by 3-10%, while those in the non-ambulatory group decreased by 23–27% ($p < 0.05$, Fig. 1). The DQs of the situation-comprehension and general development domains in the ambulatory group decreased by 6-7%, while those in the non-ambulatory group decreased by 21-22% ($p < 0.05$, Fig. 1). Furthermore, the DQ of the fine motor domain increased by 1% in the ambulatory group,

Table 2. Developmental Profiles Measured by the Chinese Child Development Inventory (CCDI) during the Initial and Final Assessments of Ambulatory and Non-Ambulatory Children with Spastic Quadriplegic Cerebral Palsy (CP)

CCDI	Initial assessment		Final assessment		*p*	*p*
	CP groups		CP groups			
DQ (%)	Ambulatory (n = 20)	Non-ambulatory (n = 28)	Ambulatory (n = 20)	Non-ambulatory (n = 28)	(group)	(within)
GM	32.6 ± 11.8	19.4 ± 9.3	28.7 ± 13.5	13.4 ± 5.3	< 0.001	< 0.001
FM	65.9 ± 24.8	28.4 ± 18.1	63.4 ± 19.8	22.1 ± 15.1	< 0.001	0.013
EL	81.4 ± 29.5	42.6 ± 25.6	75.1 ± 24.4	37.4 ± 27.8	< 0.001	0.007
CC	85.5 ± 30.4	43.0 ± 24.2	81.0 ± 26.7	38.1 ± 26.0	< 0.001	0.017
SC	66.0 ± 28.4	30.5 ± 25.8	57.2 ± 19.9	21.6 ± 12.5	< 0.001	0.003
SH	49.0 ± 16.9	28.5 ± 17.5	46.4 ± 16.8	20.8 ± 10.9	< 0.001	0.002
PS	63.3 ± 19.5	30.4 ± 16.8	56.7 ± 14.9	23.5 ± 14.0	< 0.001	< 0.001
GD	72.2 ± 17.8	39.5 ± 19.5	66.6 ± 15.6	31.0 ± 16.9	< 0.001	< 0.001

Abbreviations: DQ: developmental quotient; GM: gross motor; FM: fine motor; EL: expressive language; CC: concept-comprehension; SC: situation-comprehension; SH: self-help; PS: personal-social; GD: general development.

Values are expressed as mean ± SD.

An independent *t*-test was selected for continuous data analysis.

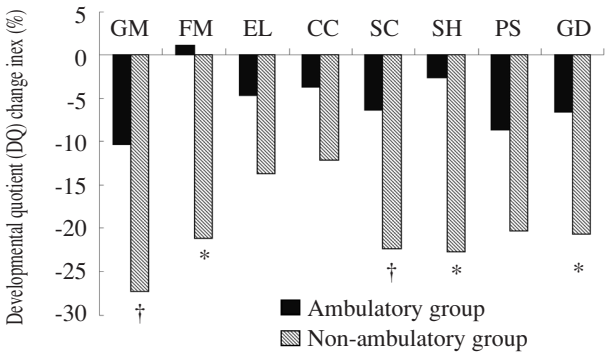

Fig. 1 The DQ change index measured by the Chinese Child Development Inventory in children with spastic quadriplegic cerebral palsy. *: $p < 0.01$; †: $p < 0.05$. The DQ change index is calculated according to the following equation: 100% × (final DQ – initial DQ) / initial DQ.
Abbreviations used: DQ: developmental quotient; GM: gross motor; FM: fine motor; EL: expressive language; CC: concept-comprehension; SC: situation-comprehension; SH: self-help; PS: personal-social; GD: general development.

but decreased by 21% in the non-ambulatory group ($p < 0.01$, Fig. 1). The DQs of the expressive language, concept-comprehension, and personal-social domains decreased by 4-9% in the ambulatory group, and 12–20% in the non-ambulatory group, although these DQ change indices did not significantly differ between the 2 groups (Fig. 1).

DISCUSSION

The developmental profiles of preschool children with SQ CP evolve with age and relate to the degree of ambulatory function. In this study, most developmental functions did not increase proportionally with increasing age in children with SQ CP, particularly in the children in the non-ambulatory group. As indicated by the DQ change index, most DQs decreased slightly with age in the ambulatory group but decreased considerably with age in the non-ambulatory group. Therefore, the non-ambulatory group made fewer developmental gains than the ambulatory group. This result may have occurred because of varying brain damage in children with CP with different ambulatory abilities. The brain damage in CP determines the disease severity and ambulatory function. Previous work demonstrated that typically developing children change, on average, one year in development per calendar year.[29] However, children with disabilities almost always change less than that amount, so that they appear to 'fall behind' other children as time goes on when their progress is adjusted for calendar time.[29] Our findings suggest children with SQ CP, especially non-ambulatory children, lose progress in most developmental functions when calendar time is included in the calculation of the results.

Fine motor development increased proportionally with age in the ambulatory group of children with SQ CP, but not in the non-ambulatory group. In this study, the DQs of the fine motor domains increased slightly in the ambulatory group, while those in the non-ambulatory group decreased markedly. This difference may result from the fact that the ambulatory group had better GMFCS levels than the non-ambulatory group. Children with GMFCS levels IV and V had more restricted hand function. Furthermore, children in the ambulatory group explored their environment to a greater extent than those in the non-ambulatory group; this is because motor, cognition, and speech function were better in the former. Therefore, fine motor skill development in the ambulatory group increased proportionally with age because this group had greater practical experience in object manipulation and fine motor skill practice through learning, education, participation in social and school activities, and self-care activities. Our findings suggest ambulatory children with SQ CP usually make progress in developing fine motor skills, while non-ambulatory children lose progress in developing these skills when calendar time is included in the calculation of the results.

Gross motor, self-help, situation-comprehension, and general development function did not increase proportionally with age in children with SQ CP, especially in those from the non-ambulatory group. As indicated by the DQ change index, the DQs of these four domains in the ambulatory group decreased slightly, while those in the non-ambulatory group decreased markedly. Previous studies have demonstrated that gross motor function in children with severe motor impairment tended to decrease,[14] while those of children with mild motor impairment increased or remained stable.[14] These differences could be attributed to the fact that the non-ambulato-

ry group had worse GMFCS levels, greater secondary motor impairment, such as secondary musculoskeletal complications, and associated problems, including cognition, speech, and social problems. Secondary musculoskeletal impairment and changes in motor functions could possibly occur because of motor control impairment, abnormal biomechanical forces, and even changes in the physical and social environment.[9-13] Children with better GMFCS levels frequently exhibit better selective motor control, muscle strength, and range of motion and less spasticity than those with worse GMFCS levels.[14]

The DQs of the developmental profiles varied in CP children with different ambulatory abilities. In this study, the DQs of all developmental domains in the non-ambulatory group were lower than those in the ambulatory group. The differences in DQ between the 2 groups were not the same for all developmental domains. However, the DQ distributions of developmental profiles were similar in the groups, i.e., the distribution was lowest in the gross motor and self-help domains and highest in the expressive language, concept-comprehension, and general development domains. This difference may result from differences between the groups with regard to several factors, including body function and structure, activities, participation in social and school activities, personal characteristics, and environmental factors. The non-ambulatory group displayed greater limitations in physical function, daily living activities, and participation in social activities and family support than the ambulatory group. Therefore, the developmental function in the non-ambulatory CP group was worse than that in the ambulatory CP group, not only in the motor domains but also in other domains, such as comprehension, language, and personal-social function. This occurred because of a lack of participation in activities by children in the non-ambulatory CP group. These findings may help clinicians predict the long-term course of developmental profiles in children with CP. Therefore, early intervention programs should be planned based on the developmental patterns of children with SQ CP.

Limitations

The limitations of this study include the study design and subject characteristics. The normative score (DQ), not raw score, was used in this study. Children with disabilities often lag behind with increasing age when using the normative score, even when the raw data shows they have made progress. The enrollment criterion was SQ CP with upper motor neuron signs in all limbs. Subjects with the other CP subtypes 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 follow-up course of developmental profiles of children with SQ CP.

Conclusions

The DQs of all developmental functions in the non-ambulatory group were lower than those in the ambulatory group. However, the DQ distributions of developmental profiles were similar in the groups. Most DQs decreased slightly with age in the children with SQ CP in the ambulatory group, but decreased considerably with age in the non-ambulatory group, especially the DQs for gross motor, self-help, situation-comprehension, and general development function. Furthermore, fine motor function increased proportionally with age in the ambulatory group, but not in the non-ambulatory group. These findings suggest that the DQs of developmental profiles vary in children with SQ CP with different ambulatory abilities. The course of the developmental profiles of preschool children with SQ CP was found to relate to ambulatory ability. Knowledge of these developmental profiles may be helpful in understanding, predicting, and managing the problems faced by these children. Future studies will focus on long-term study of the developmental profiles of children with CP of various subtypes.

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學齡前四肢痙攣型腦性麻痺兒童發展輪廓與行走能力之追蹤研究

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背 景： 縱向追蹤學齡前具有不同行走能力的四肢痙攣型腦性麻痺兒童其發展輪廓之進程。

方 法： 將 48 位年齡在 1-5 歲的四肢痙攣型腦性麻痺兒童，根據最初的粗動作功能分類評估量表評估分成具行走能力與不具行走能力兩組。最初以及最終 (一年後) 分別給予發展輪廓評估：含有 8 個面向的發展商數 (DQs)。並計算發展商數改變指標 (%)，等於 100% × (最終 DQ — 最初 DQ) / 最初 DQ)。

結 果： 不具行走能力組其發展商數各項功能面向皆比具行走組爲低 ($p < 0.01$)。在發展商數改變指標方面，具行走能力組其大部分發展商數有輕微降低的情形，但是不具行走組則發現有顯著下降的情形 ($p < 0.05$)。此外，具行走能力組其精細動作功能隨年齡等比增加，但不具行走能力組則無此現象。

結 論： 學齡前四肢痙攣型腦性麻痺兒童不同行走能力其發展輪廓不同。隨著年齡發展其發展進程和行走能力有關。對腦性麻痺兒童發展輪廓知識之建立有助於了解、預測及處理這些兒童之相關發展問題。

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關鍵詞： 腦性麻痺，發展功能，預後，行走，縱向研究

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