

Factors Associated with Motor Speech Control in Children with Spastic Cerebral Palsy

Chia-Ling Chen^{1,2}, MD, PhD; Keh-Chung Lin³, PhD; Chia-Hui Chen¹, MEd;
Chih-Chi Chen¹, MD; Wen-Yu Liu⁴, PhD; Chia-Ying Chung^{1,2}, MD;
Chung-Yao Chen⁵, MD; Ching-Yi Wu⁶, ScD

Background: Speech production is often impaired in children with cerebral palsy (CP). This study investigated the factors associated with motor speech control in children with spastic cerebral palsy.

Methods: Thirty-three children with spastic CP who were able to speak were identified for this study. They were classified into two groups: group A (spastic diplegia or hemiplegia, n = 17) and group B (spastic quadriplegia, n = 16). Each child received various assessments, namely cognition, language, modified Verbal Motor Production Assessment for Children (VMPAC), speech intelligibility, CP subtype, and Gross Motor Function Classification System (GMFCS).

Results: Group A showed better cognition and language function, higher modified VMPAC scores and better GMFCS levels than group B ($p < 0.05$). However, the two groups did not significantly differ in speech intelligibility. Linear regression indicated that all modified VMPAC scores were negatively related to CP subtype (adjusted $r^2 = 0.51\sim 0.63$, $p < 0.001$). The average modified VMPAC scores had a positive relationship with global language scores (adjusted $r^2 = 0.63$, $p < 0.001$), and the modified VMPAC scores of the sequence subtests were positively related to the full intelligence quotients (adjusted $r^2 = 0.55$, $p < 0.001$).

Conclusion: The findings of this study suggest that motor impairment severity, cognition and language functions are associated with the motor speech control among children with CP.

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Key words: cerebral palsy, oro-motor control, motor speech control, speech intelligibility, motor severity

Cerebral palsy (CP) is a non-progressive movement and posture disorder caused by brain damage or dysfunction that occurred before, during or

shortly after birth.⁽¹⁾ CP with different classifications presents with various clinical manifestations. Spastic CP is by far the most common type and occurs in

From the ¹Department of Physical Medicine and Rehabilitation, Chang Gung Memorial Hospital at Linkou; ²Department of Physical Medicine and Rehabilitation, Chang Gung Memorial Hospital at Keelung, Chang Gung University College of Medicine, Taoyuan, Taiwan; ³Graduate Institute of Early Intervention, Department of Physical Therapy; ⁴Graduate Institute of Physical Therapy, Department of Physical Therapy; ⁵Graduate Institute of Clinical Behavioral Science, Department of Occupational Therapy, Chang Gung University, Taoyuan, Taiwan; ⁶Graduate Institute of Occupational Therapy, College of Medicine, National Taiwan University, Taipei, Taiwan.

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Correspondence to: Prof. Ching-Yi Wu, ScD, Department of Occupational Therapy, Chang Gung University, 259, Wunhua 1st Rd., Gueishan Township, Taoyuan County 333, Taiwan (R.O.C.) Tel.: 886-3-2118800 ext. 5761; Fax: 886-3-2118700;

E-mail: cywu@mail.cgu.edu.tw

70% to 80% of all cases. Spastic CP is further classified topographically according to affected region of the body; for instance, the affected areas of the body may have partial (e.g., diplegia) or total (e.g., quadriplegia) bodily involvement. Some children with CP also have other associated problems such as seizures, mental retardation or speech and behavioral problems. Speech and language disorders are one of the major associated impairments in children with CP.⁽²⁻⁴⁾ Impaired speech functions occur in 38% of children with CP.⁽⁵⁾ Even in children with CP who have an intelligence quotient above 70, as many as half still exhibit motor speech problems.⁽⁶⁾ Such problems impair the functional communication of children with CP.⁽⁷⁻¹¹⁾

Verbal communication is often modeled as a dynamic system.⁽¹²⁾ Speech production is a complex motor activity involving coordination of the respiratory, laryngeal and articulatory subsystems.⁽¹³⁾ How these fine articulating movements are controlled and coordinated for speech production in children with CP is not well understood. Studies of speech movement control have examined the central control of movement patterns, interarticulator coordination and the role of sensory feedback.^(13,14) In addition, motor speech problems are also associated with cognitive difficulties and language function in children with CP.⁽⁶⁾

Speech production is often impaired in children with CP.⁽¹⁵⁾ Poor speech production might be a direct result of motor impairment due to disturbed neuromuscular control of speech mechanisms.^(10,16-18) The more severe the CP, the greater the impairment of speaking and communicative ability.⁽¹⁹⁾ We hypothesized that motor speech control is correlated with motor impairment severity, cognitive impairment and language impairment in children with spastic CP. This study examined the factors associated with motor speech control in children with spastic CP.

METHODS

Participants

This study enrolled thirty-three children with CP (seventeen boys and sixteen girls), aged 4-12 years old. These children were undergoing treatment in the rehabilitation department at this hospital. The inclusion criteria were as follows: (1) spastic CP with diplegia, hemiplegia or quadriplegia from birth; (2)

ability to perform the tasks needed for this study; (3) good cooperation during examination; and (4) the ability to speak and understand the verbal commands required for this analysis. Exclusion criteria were any history of the following within the previous three months: (1) significant medical problems such as active pneumonia or a urinary tract infection; (2) significant visual or hearing impairment; (3) any major surgical treatment such as orthopedic surgery or neurosurgical surgery; (4) treatment by nerve or motor point blockage such as a botulinum toxin injection; and (5) any dental procedure such as tooth extraction. Children with a history of visual/hearing impairment or facial palsy were also excluded from this study. The enrolled subjects were grouped according to topographic distribution of limb involvement: group A [spastic diplegia (SD) or hemiplegia (SH), $n = 17$] and group B [spastic quadriplegia (SQ), $n = 16$]. The Institutional Review Board for Human Studies at Chang Gung Memorial Hospital approved this protocol, and all subjects provided informed consent.

Assessment procedures

All children underwent cognition, language, motor speech and motor impairment severity assessments. Demographic data, including age, body weight (BW), body height (BH) and gender, were also recorded.

Cognition was evaluated by the Wechsler Preschool and Primary Scale of Intelligence-Revised (WPPSI-R) for preschool children and Wechsler Intelligence Scale for Children,⁽²⁰⁾ Third Edition (WISC-III) for school children.⁽²¹⁾ The Verbal Intelligence Quotient (VIQ) and Performance Intelligence Quotient (PIQ) contribute to the full scale IQ (FIQ). The VIQ and PIQ are designed to examine specific skills, while the FIQ can be used to evaluate global intelligence. The WPPSI-R and WISC-III assessments yield intelligence quotients (IQs), VIQ, PIQ, and FIQ.

To assess language proficiency, we use the Preschool Language Impairment Scale (PLIS)⁽²²⁾ for preschool children and the Language Impairment Scale (LIS)⁽²³⁾ for school children. Both PLIS and LIS are individually administered tests consisting of two subtests, namely receptive and expressive abilities for spoken language. For the PLIS and LIS, a 2-point scale was used to analyze verbal comprehen-

sion, verbal expression and global language. The child's score are computed by adding the total number of items for which the child receives credit on each scale. A global language score is then obtained by adding the two subtests. Performance is reported as percentile ranks of the auditory comprehension subtest, the verbal expression subtest and the global language test. The PLIS consists of 30 items in auditory comprehension subtest and 32 items in verbal expression subtest. Test-retest reliability and concurrent validity of the PLIS has been found to have ranges of 0.84-0.92 and 0.76-0.85, respectively.⁽²²⁾ The LIS consists of 30 items for each subtest. Test-retest reliability and concurrent validity of the LIS has been found to have ranges of 0.75-0.80 and 0.53-0.72, respectively.⁽²³⁾

Motor speech control was evaluated using the modified Verbal Motor Production Assessment for Children (mVMPAC).⁽²⁴⁾ This test provides data for the differential diagnosis of children with motor speech disorders.⁽²⁵⁾ The 3-point VMPAC scale (0: incorrect, 1: partly incorrect, and 2: correct) is designed to assess the neuromotor integrity of the motor speech system in children who have speech production disorders.⁽²⁴⁾ The test items are systematically organized into three main areas and two supplemental areas, including connected speech, language control and speech characteristics. Only the main areas, including global motor control, focal oromotor control, and sequencing areas, were analyzed in this study due to cultural differences; specifically the differences between the populations for which the instrument was validated and the study population in Taiwan. The global motor control area, consisting of global motor control and oromotor integrity subtests, addresses neuromotor innervation to those peripheral muscles in the torso, neck, head, and oro-facial region required for the efficient production of speech.⁽²⁴⁾ The focal oromotor control area, consisting of both non-speech and speech oromotor movements subtests, is utilized to assess the volitional oromotor control for mandibular, labial-facial, and lingual control.⁽²⁴⁾ The sequencing area, consisting of both non-speech and speech oromotor movement sequences subtests, is designed to assess the ability to produce non-speech and speech movements in the correct sequential order.⁽²⁴⁾ The percentage score for each area was defined as the raw score divided by the sum of the corresponding item scores of each area. The

main modified VMPAC score (%) was calculated as the mean percentage score of the three main areas. High intra-rater reliability ($r = 0.56-0.90$, $p < 0.01$) and inter-rater correlations ($r = 0.93-0.99$, $p < 0.01$) have been previously confirmed.⁽²⁴⁾

The CP subtypes and the Gross Motor Function Classification System (GMFCS) grades were used to measure the motor severity of CP.⁽²⁶⁾ The GMFCS grades the self-initiated movement of CP patients with particular emphasis on their functional abilities (sitting, crawling, standing and walking) and their need for assistive devices (e.g., walkers, crutches, canes and wheelchairs). The GMFCS employs a 5-point scale (I-V) from "independent" (level I) to "dependent": (level V). Demographic characteristics, including age, body weight, body height, and gender, were also recorded.

Experimental setup for measuring speech intelligibility

The subject was seated in a quiet room. The recording system used to measure speech intelligibility consisted of an IBM ThinkPad 570E laptop with a model CS 4280 soundcard and one external microphone. The microphone was placed on a table approximately 15 cm from the mouth of the subject. The subjects were shown pictures or text printed on cards and asked to read them aloud in a normal voice. If the subject encountered an unfamiliar word, the experimenter would explain the word or ask the subjects to read it with the assistance of phonetic transcription. The experimenter did not model the correct sound production or provide other assistance. The speech recording tasks were assessed by a 4-point speech intelligibility scale (0: 0-25% intelligibility; 1: 25-50% intelligibility; 2: 50-75% intelligibility; 3: 75-100% intelligibility). The speech recording tasks included sixty-seven picture-cards for preschool children and 140 word-cards for school children. Speech samples from ten participants with CP were randomly selected to measure the reliability. The speech intelligibility tests showed high intra-rater reliability ($r = 0.89$, $p < 0.01$) and inter-rater correlations ($r = 0.76$, $p < 0.01$).

Statistical analysis

Differences in the continuous data (age, BH, BW, modified VMPAC, cognition and language functions) between groups were compared by an

independent t-test. Gender differences between groups were determined by Chi-square test. The Mann-Whitney U test was used to determine group differences for the GMFCS levels. The Pearson correlation test was used to clarify relationships between motor speech control (modified VMPAC) and other clinical measurements (CP subtypes, GMFCS levels, cognition, and language functions) as well as the demographic data. A stepwise linear regression analysis was used to characterize the relationships between motor speech control (modified VMPAC) and other clinical measurements and demographic data. A *p* value of < 0.05 was considered statistically significant.

RESULTS

The two groups did not significantly differ in demographic characteristics (Table 1). Approxi-

mately 70% of the children in group A had GMFCS levels lower than 3, while 88% of the children in group B had GMFCS levels higher than 2 (*p* < 0.001, Table 1).

Speech intelligibility did not significantly differ between the two groups (Table 2). However, group A had better global motor control, focal oro-motor control, sequencing and average modified VMPAC scores than group B (*p* < 0.01, Table 2). Comparison of the VIQ, PIQ and FIQ scores indicated that group A had better cognition than group B (*p* < 0.01, Table 2). Language function measured by the verbal comprehension and global language functions were also better in group A than in group B (*p* < 0.05, Table 2). However, there were no significant differences in the verbal expression function between the two groups.

Pearson correlation analysis indicated the all modified VMPAC scores, including global motor control, focal oro-motor control, sequencing and average scores, were correlated negatively with the

Table 1. Comparison of the Demographic Data and Severity of Motor Impairment between the Two Groups of Children with Cerebral Palsy

Data	Group A (n = 17) Diplegia and hemiplegia	Group B (n = 16) Quadriplegia	<i>p</i>
Demographic			
Age (years)	6.1 ± 1.7	7.0 ± 2.3	0.230
Sex: Male	12 (70.6)	8 (50.0)	0.226
Body height (cm)	19.8 ± 4.8	20.6 ± 7.5	0.721
Body weight (kg)	108.9 ± 11.7	110.3 ± 14.5	0.762
Motor impairment severity			
CP subtypes			
Diplegia	13 (76.5)	0 (0.0)	
Hemiplegia	4 (23.5)	0 (0.0)	
Quadriplegia	0 (0.0)	16 (100.0)	
GMFCS			
			< 0.001*
Level 1	9 (52.9)	0 (0.0)	
Level 2	3 (17.7)	2 (12.5)	
Level 3	4 (23.5)	9 (56.3)	
Level 4	1 (5.9)	4 (25.0)	
Level 5	0 (0.0)	1 (6.2)	

Abbreviations: GMFCS: Gross Motor Function Classification System; *: *p* < 0.01.

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

Table 2. Comparison of Cognition, Language, and Motor Speech Functions between the Two Groups of Children with Cerebral Palsy

Data	Group A (n = 17) Diplegia and hemiplegia	Group B (n = 16) Quadriplegia	<i>p</i>
Intelligence quotient (IQ)			
Verbal IQ	98.3 ± 21.1	78.0 ± 17.3	0.005*
Performance IQ	78.8 ± 16.1	62.3 ± 16.6	0.007*
Full IQ	87.5 ± 18.2	67.3 ± 15.4	0.002*
Language (percentile)			
Verbal comprehension	59.6 ± 33.5	28.9 ± 26.3	0.007*
Verbal expression	53.8 ± 36.0	41.7 ± 41.3	0.374
Global	54.4 ± 30.4	35.8 ± 30.8	0.048
Motor speech			
Speech intelligibility	2.85 ± 0.21	2.69 ± 0.43	0.348
Motor speech control (VMPAC, %)			
Global motor control	88.2 ± 9.7	57.8 ± 22.2	< 0.001*
Focal oro-motor control	92.9 ± 5.9	72.6 ± 13.1	< 0.001*
Sequence	96.2 ± 8.9	72.2 ± 24.2	0.003*
Average	92.5 ± 5.9	67.1 ± 15.9	< 0.001*

Abbreviations: VMPAC: Verbal motor production assessment for children; *: *p* < 0.01.

Values are expressed as mean ± SD.

CP subtype ($r < -0.57, p < 0.01$, Table 3) and motor impairment severity ($r < -0.56, p < 0.01$). That is, children with diplegic and hemiplegic CP had better modified VMPAC scores than children with quadriplegic CP. All modified VMPAC scores correlated positively with intelligence quotients: VIQ ($r > 0.38, p < 0.05$, Table 3), PIQ ($r > 0.36, p < 0.05$) and FIQ ($r > 0.44, p < 0.05$). Most modified VMPAC scores were strongly associated with the language functions of verbal comprehension ($r > 0.46, p < 0.01$, Table 3), verbal expression ($r > 0.40, p < 0.05$) and global language function ($r > 0.39, p < 0.05$).

The stepwise linear regression analysis results in Table 4 show the regression equations of all modified VMPAC scores and related factors (adjusted r^2 : 0.51~0.63, $p < 0.01$). All modified VMPAC scores, including global motor control, focal oro-motor con-

trol, sequencing and average scores were correlated negatively with CP subtype (Table 4). The average modified VMPAC scores correlated positively with global language score (Table 4). The modified VMPAC scores of the sequence subtests correlated positively with FIQ (Table 4). The global motor scores correlated negatively with age (Table 4).

DISCUSSION

Motor impairment severity was found to be associated with the motor speech control in children with CP. In this study, all modified VMPAC scores correlated negatively with CP subtype according to the stepwise linear regression test, although all modified VMPAC scores correlated negatively with CP subtype and GMFCS level according to Pearson cor-

Table 3. Correlations of Motor Speech Control with Demographic and Clinical Data among the Children with Cerebral Palsy

Data	Modified VMPAC (%)			
	Global motor control	Focal oro-motor control	Sequence	Average
Demographic				
Age	-0.421 [†] (-0.677, 0.091)	-0.370 [†] (-0.632, 0.031)	-0.303 (-0.585, 0.404)	-0.395 [†] (-0.650, 0.060)
Sex [‡]	-0.084 (-0.415, 0.267)	0.014 (-0.330, 0.355)	-0.258 (-0.737, 0.226)	-0.121 (-0.445, 0.231)
Clinical data				
Motor impairment severity				
CP subtypes [§]	-0.679* (-0.829, -0.438)	-0.726* (-0.855, -0.510)	-0.576* (-0.767, -0.291)	-0.746* (-0.867, 0.542)
GMFCS	-0.580* (-0.923, 0.716)	-0.601* (-0.782, -0.325)	-0.561* (-0.758, -0.27)	-0.654* (-0.814, 0.041)
Intelligence quotient (IQ)				
Verbal IQ	0.393 [†] (0.058, 0.648)	0.382 [†] (0.045, 0.641)	0.698* (0.467, 0.84)	0.575* (0.289, 0.766)
Performance IQ	0.365 [†] (0.025, 0.629)	0.461* (0.14, 0.694)	0.595* (0.317, 0.779)	0.537* (0.238, 0.743)
Full IQ	0.441 [†] (0.116, 0.681)	0.476* (0.159, 0.704)	0.718* (0.498, 0.851)	0.629* (0.369, 0.755)
Language (percentile)				
Comprehension	0.466* (0.147, 0.697)	0.508* (0.2, 0.724)	0.550* (0.225, 0.751)	0.583* (0.3, 0.711)
Expression	0.247 (-0.105, 0.544)	0.31 (-0.037, 0.59)	0.487* (0.173, 0.711)	0.406 [†] (0.073, 0.657)
Global	0.396 [†] (0.061, 0.65)	0.449* (0.125, 0.686)	0.566* (0.277, 0.761)	0.541* (0.243, 0.745)

Abbreviations: GMFCS: Gross Motor Function Classification System; VMPAC: Verbal motor production assessment for children; *: $p < 0.01$; †: $p < 0.05$; ‡: Sex: 1: male, 2: female; §: CP (cerebral palsy) subtypes: 1: diplegia or hemiplegia, 2: quadriplegia. Values are expressed as coefficients (95% confidence interval).

Table 4. Association of Motor Speech Control with Demographic and Clinical Data of the Children with Cerebral Palsy

VMPAC	Independent variable	Coefficient	Partial correlation	Adjusted r^2	p
Global motor control	CP subtypes [†]	-0.62	-0.60	0.511	< 0.001*
	Age	-0.29	-0.28		
Focal oro-motor control	CP subtypes	-0.73	-0.73	0.511	< 0.001*
Sequence	CP subtypes	-0.29	-0.25	0.546	< 0.001*
	Full intelligence quotient	0.57	0.49		
Average	CP subtypes	-0.64	-0.60	0.634	< 0.001*
	Global language function	0.32	0.34		

Abbreviations: VMPAC: Verbal motor production assessment for children; *: $p < 0.01$; †: CP (cerebral palsy) subtypes: 1: diplegia or hemiplegia, 2: quadriplegia.

relation analysis. Children with SD or SH had better motor speech control than those with SQ. A possible reason is that neuromotor impairment due to the brain damage that caused CP resulted in different effects on motor severity and verbal motor impairment. The VMPAC may measure motor speech control.⁽²⁵⁾ The motor severity may thus be reflected in CP subtype and GMFCS level.⁽²⁶⁾ Previous studies have reported that the severity of functional motor limitations is related to motor speech problems.^(6,19) Some aspects of speech and communication seemed to be improved when there is motor function improvement after some children receive intrathecal baclofen therapy.⁽²⁷⁾

Cognition and language functions were also found to be associated with verbal motor function in children with CP. Regression analysis revealed that some modified VMPAC scores correlated positively with verbal expression or FIQ in this study. The motor processes of speech are shaped by multiple intrinsic factors such as cognitive, linguistic and sensorimotor maturation and extrinsic factors such as auditory and visual stimulation as well as perceptual saliency factors.⁽²⁸⁾ Pirila et al. also reported that cognition is related to language and motor speech problems in children with CP.⁽⁶⁾ Pirila et al. demonstrated that half of all CP children with normal intelligence exhibit impairments, primarily in the motor speech

domain, whereas children with additional cognitive difficulties reveal impairments in both language and motor speech skills.⁽⁶⁾ However, another study found that increases in articulation rate are not necessarily accompanied by improvements in memory span among children with CP.⁽²⁹⁾

In this study, children with SD or SH had better modified VMPAC scores than those with SQ. These findings suggest that motor speech control and coordination, such as global motor control, focal oro-motor control and sequencing, are more severely impaired in children with SQ than in those with SD or SH, even if speech intelligibility is good. Therefore, appropriate verbal motor therapeutic interventions are needed for children with CP irrespective of the CP subtype. The therapeutic interventions on speech and communication should be given on the basis of an assessment of speech, language and communication. The goal of motor speech treatment intervention must be focused on the integration of all aspects of voluntary speech control and needs to be based on assessments of the neuromotor system, motor speech system and the interaction among the various articulators.⁽³⁰⁾

The findings of this study are limited because of its design. The design of the study was limited by sample size and the analyzed subject's characteristics. The enrollment criteria were an ability to speak

and show good cooperation. Subjects with severe impairments were not recruited in this study. Therefore, the study results cannot be generalized to all cases of CP. Despite this restriction, this study convincingly demonstrates that there is a correlation between motor speech control and severity of motor impairment, cognition and language function in children with CP.

Conclusion

In conclusion, all modified VMPAC scores were negatively related to CP subtype. Children with spastic quadriplegic CP had more severe verbal motor impairment than those with diplegic or hemiplegic CP. The average modified VMPAC scores had a positive relationship with global language scores, and the modified VMPAC scores of sequence subtests were positively related to FIQ. Our findings suggest that severity of motor impairment, cognition and language functions are associated with motor speech control among children with CP. Planning appropriate verbal motor therapeutic interventions is therefore vital, especially for children with SQ CP. Future research needs to investigate the mechanisms of motor speech control in children with CP through kinematic and electromyographic analysis; furthermore, there is a need to examine the strategies available for the treatment of motor speech dysfunctions among children with CP.

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痲痺型腦性麻痺兒童語言動作控制的相關因子

陳嘉玲^{1,2} 林克忠³ 陳嘉惠¹ 陳芝琪¹ 劉文瑜⁴ 鍾佳英^{1,2} 陳仲堯⁵ 吳菁宜⁶

背景： 本篇研究主要是探討痲痺型腦性麻痺兒童語言動作控制的相關因子。

方法： 33 位有說話能力的痲痺型腦性麻痺兒童分為兩組：A 組 17 位 (雙邊麻痺或半側偏癱) 及 B 組 16 位 (四肢麻痺)。所有個案皆接受認知、語言、改良式兒童口語動作評估 (Modified verbal motor production assessment for children, VMPAC)，包含整體動作控制、嘴部動作控制及連續分項測驗；語言清晰度；腦性麻痺種類及腦性麻痺兒童粗大動作功能分類表 (Gross motor function classification system, GMFCS) 的評估。

結果： A 組相較於 B 組有較好的認知、語言、VMPAC 分數及 GMFCS 等級 ($p < 0.05$)，然而，兩組的語言清晰度並沒有明顯差異。線性迴歸顯示所有的 VMPAC 分數與腦性麻痺類型呈現負相關 (adjusted $r^2 = 0.51\sim 0.63$, $p < 0.001$)，VMPAC 的平均分數與整體語言分數呈現正相關 (adjusted $r^2 = 0.63$, $p < 0.001$)，而 VMPAC 中的連續分項測驗與總智商呈現正相關的關係 (adjusted $r^2 = 0.55$, $p < 0.001$)。

結論： 這個研究的結果可以推測動作障礙的嚴重性、認知及語言功能和腦性麻痺兒童的語言動作控制有關。

(長庚醫誌 2010;33:415-23)

關鍵詞： 腦性麻痺，嘴部動作控制，語言動作控制，語言清晰度，動作嚴重度

¹長庚醫療財團法人林口長庚紀念醫院，²長庚醫療財團法人基隆長庚紀念醫院 復健科；長庚大學 醫學院 ³早期療育研究所，⁴物理治療學系暨研究所，⁵職能治療學系暨研究所；⁶台灣大學 職能治療學系暨研究所

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通訊作者：吳菁宜教授 長庚大學 職能治療學系。桃園縣333龜山鄉文化一路259號。Tel.: (03)2118800轉5761;

Fax: (03)2118700; E-mail: cywu@mail.cgu.edu.tw