

Clinical Factors Related to Upper Extremity Shortness in Children With Hemiplegic Cerebral Palsy

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Abstract

Objective : The purpose of this paper is to investigate the upper extremity shortness in people with hemiplegic cerebral palsy (HCP) compared with a normal control group, and evaluate the correlations between extremity shortness, muscle tone, and functional activity.

Methods : This study included 30 children with HCP (13 males, 17 females; mean age, 6.33 yr) and 17 normal children as a control (9 males, 8 females; mean age, 6.17 yr). The length of each limb segment composed of the humerus, ulna, radius, and the second and fifth metacarpal bones was measured for both the patients and control group using a radiograph. The discrepancy in side-to-side length was calculated as a percentage. The Modified Ashworth Scale (MAS) was used in the evaluation of spasticity, and the Quality of Upper Extremity Skills Test (QUEST) was used in the evaluation of functional ability.

Results : Upper extremity shortness differed significantly between the patients and control group ($p<.05$). The percentage differences in bone length correlated positively with the MAS point, and negatively with QUEST score in each limb segment ($p<.05$).

Conclusion : The hemiplegic upper limb length discrepancy correlated with the functional activity and spasticity of the upper limb. Further studies to evaluate the change in upper extremity shortness after functional improvement and spasticity management will help to demonstrate the effect of rehabilitative treatment and determine the method and intensity of therapy required.

Key words : Bones of upper extremity, Cerebral palsy, Hemiplegia, Motor activity, Muscle spasticity

I. Introduction

Hemiplegic cerebral palsy (HCP) is a type of cerebral palsy that occurs when part of the brain is damaged at any time before, during or after birth, sometimes in unilateral or asymmetric periventricular leukomalacia. Motor dysfunction in hemiplegia is most commonly of the spastic type. Affected body parts show abnormalities in functional activity, skeletal growth, bone maturity, muscle length and muscle tone (Miller, 2005).

Specialists in pediatrics, orthopedics and rehabilitation have primarily focused their interest on leg length discrepancy (LLD) because of skeletal deformities, musculoskeletal pains and increased energy expenditures, especially gait disturbance by LLD (Moseley, 1987). Although the arm is more involved than the leg in most children with HCP, upper limb length discrepancy seems to receive less attention than LLD (Miller, 2005). The reason for this may be that most daily life activities can be done by an unaffected upper limb. We have experienced many complaints of cosmetic problems, socio-emotional stress and functional impairments due to upper limb length discrepancy in HCP patients who have grown up to be adolescents or adults in our clinic. Several previous studies have documented delayed skeletal maturation and muscular underdevelopment in the affected upper limb of children with HCP (Flett, 2003; McDaid, Kozin, Thoder, & Porter, 2002; Mukherjee & Gaebler-Spira, 2007; Strecker & Stephens, 1983; Tizard, Paine, & Crothers, 1954; Uvebrant, 1988). However, the definition of abnormal upper limb shortness and clinical factors related to it have not been established yet.

The purposes of the present study are to investigate upper extremity shortness in HCP patients compared with normal controls and to evaluate its relationship with muscle tone and

functional activity. We chose muscle tone and functional activity as clinical factors related to limb shortness because these factors can be managed with comprehensive rehabilitation programs, such as physiotherapy, occupational therapy, neurolytic blocking agents, oral medication and reconstructive procedures. To demonstrate of correlations between upper extremity shortness, muscle tone and functional activity would be helpful in the treatment of limb discrepancy.

II. Methods

1. Subjects

The present study included 30 children with HCP (13 males, 17 females; mean age 6.33 years, SD 2.24 years) induced by prenatal or perinatal etiology who participated in an inpatient or outpatient rehabilitation program at the Pediatric Rehabilitation Clinic, between March 2007 and July 2008. Prenatal, natal, and postnatal histories of the patients were investigated. Thirteen ischemic strokes, seven premature births with germinal matrix hemorrhage, six traumatic brain injuries and four vascular malformations with intracranial hemorrhage were included in patients group. All 30 children with HCP showed above 3 level in Gross Motor Function Classification System. The control group was comprised of 17 healthy children (9 males, 8 females; mean age 6.17 years, SD 1.76 years). All children in the present study had a normal nutritional status on the basis of the weight, height and serum laboratory studies including hemoglobin, electrolytes, iron and vitamins. Because all participants had to be exposed to X-irradiation, only children whose legal guardian wanted to know the bony growth and length discrepancy of their children through radiologic

evaluation were included in this study. Other exclusion criteria were as follows: serious contractures with difficult radiological examination; syndromic or familial causes of physical disabilities; inborn errors of metabolism; congenital infections; congenital abnormalities; past history of upper extremity fractures; peripheral nerve injury or upper extremity surgery.

2. Measures

1) Radiological Assessment

Radiographs the humerus, forearm, and hands were obtained on both involved and uninvolved sides in the patient group (or both dominant and nondominant sides in the control group) from each child, with a radiographic ruler placed adjacent to the extremity. Radiographs were taken by a single trained observer and then measured by a single examiner to avoid interobserver errors. All segmental bone lengths were measured from the proximal epiphyses to the distal epiphyses. Humeral, radial and ulnar lengths were measured from anteroposterior radiographs. Humeral length was measured from the head of the humerus to the end of the lateral condyle with the shoulder slightly adducted and the elbow flexed at 90°. The radiographs of the forearm were taken in the pronation position on an x-ray cassette attached to a wide plaster. Radial length was defined as the distance from the proximal end to the distal end of the radial styloid process. Ulnar length was defined as the distance from the proximal end of the olecranon process to the distal end of the ulna styloid. Hand lengths were defined as the distance from the proximal end to the distal end of the second and fifth metacarpal bones. Length discrepancy was recorded in millimeters and as a percentage of the uninvolved (or dominant) side. The percentage of shortness for each bone was calculated by the

following formula and expressed as a ratio (Demir, Oktay, Uysal, & Seluk, 2006):

The percentage of shortness for each bone in the patient group =

$$\frac{[\text{Uninvolved bone length} - \text{Involved bone length}] \times 100}{\text{Uninvolved bone length}}$$

The percentage of shortness for each bone in the control group =

$$\frac{[\text{Dominant bone length} - \text{Nondominant bone length}] \times 100}{\text{Dominant bone length}}$$

2) Modified Ashworth Scale

The Modified Ashworth Scale (MAS), which classifies muscle tone in 6 stages, was used for spasticity evaluation. A single trained examiner who was an occupational therapist with 15 years experience measured MAS points in hemiplegic upper extremities. Muscle spasticity was examined for limb segments as follows: shoulder adductor for the humerus; elbow flexor for the humerus, radius and ulna; wrist flexor for the radius, ulna and second or fifth metacarpal bone; finger flexor for the second or fifth metacarpal bone. For statistical analysis, each stage was rated on a scale of 0 to 5 points.

3) Quality of Upper Extremity Skills Test

Upper extremity function was quantitatively assessed with the Quality of Upper Extremity Skills Test (QUEST) by a single occupational therapist who was skilled in the use of this test (DeMatteo et al., 1993; Law et al., 1991). For QUEST scoring, sub-items in dissociated movements and grasps domains were chosen according to each limb segment: shoulder and elbow items according to humeral length; elbow and wrist items according to radial or ulnar length; wrist, finger and grasp items

according to second or fifth metacarpal bone length. We calculated functional severity score by the following formula (Stevenson, Roberts, & Vogtle, 1995):

severity score =

$$\frac{\text{QUEST score of each limb segment in involved arm (or nondominant arm)} \times 100}{\text{QUEST score of each limb segment in uninvolved arm (or dominant arm)}}$$

3. Procedure

Radiographs, MAS and QUEST were done on the same day. Radiographs were taken by a single trained observer and then measured by a single examiner, doctor in rehabilitation medicine, to avoid interobserver errors. MAS and QUEST were measured by a single occupational therapist who was skilled in the use of these tests.

The patients were divided into 2 subgroups with the reference point that was calculated from the mean value plus $2SD$ of percentage shortness in the control group: those without significant length discrepancy (subgroup 1, HCP–non–short) and those with (subgroup 2, HCP–short). We compared MAS point and QUEST score between the 2 subgroups. We also evaluated the relationship between the rate of each segmental shortness and age/MAS point /QUEST score in the patients.

4. Statistical analysis

SPSS for Windows release 17.0 was used to analyze the data. Significance levels for all analyses were set at $p < .05$ (two-tailed). Upper extremity length discrepancy was compared between the patient and control groups by using the Mann–Whitney U test. Also, the Mann–Whitney U test was used to compare MAS point and QUEST

score between the 2 subgroups. The correlation of the percentage shortness of segmental bone length with age, QUEST score or MAS point was evaluated by the Spearman correlation test.

5. Ethics statements

The study was conducted in full compliance with the amended Declaration of Helsinki, following approval from the institutional review board of Pusan National University Yangsan Hospital (IRB–05–2012–039). Informed consent was obtained from each legal guardian.

III. Results

1. Upper extremity length discrepancy in the control and patient groups

A total of 47 subjects completed the study (17 subjects in the control group and 30 subjects in the patient group). The results of the Mann–Whitney U test for comparison of percentages of upper extremity shortness in the patient and control groups are presented in Table 1. The average percentage of limb shortness ranged from 2.57% to 5.82% in the patient group, from 0.35% to 1.83% in the control group. The percentage of shortness of all upper extremity segments was significantly different between the control and patient group ($p < .05$).

2. Upper extremity shortness and related clinical factors in the patient group

The patients were divided into 2 subgroups, HCP–short and HCP–non–short, with the reference point that was calculated from the mean

value plus 2SD of the percentage of shortness in the control group. The reference point was 0.63% for the humeral segment, 1.00% for the radius, 1.02% for the ulnar segment, 2.92% for the second metacarpal segment and 3.45% for the fifth metacarpal segment.

QUEST scores were significantly lower in HCP-short subgroup than in HCP-non-short subgroup for the ulna, and second and fifth metacarpal bones ($p<.05$). However, for the humeral and radial segments, there were no

statistically significant differences in QUEST severity scores between the 2 subgroups ($p>.05$) (Table 2).

MAS points for elbow flexor, wrist flexor and finger flexor were significantly higher in HCP-short than HCP-non-short subgroup in all forearm and hand segments ($p<.05$) except that for wrist flexor ($p>.05$). MAS points for shoulder adductor and elbow flexor divided by humeral length discrepancy were not statistically significant between the 2 subgroups ($p>.05$) (Table 3).

Table 1. Upper Extremity Length Discrepancy in the Control and Patient Groups

	Percentage of Shortness (%)		<i>p</i>
	Control (<i>n</i> =17)	Patient (<i>n</i> =30)	
	<i>M</i> ± <i>SD</i>	<i>M</i> ± <i>SD</i>	
Humerus	0.35 ± 0.14	2.57 ± 2.22	<.001**
Radius	0.48 ± 0.26	3.05 ± 2.05	<.001**
Ulna	0.46 ± 0.28	3.21 ± 2.09	<.001**
Second metacarpus	1.46 ± 0.73	5.23 ± 5.05	<.001**
Fifth metacarpus	1.83 ± 0.81	5.82 ± 8.96	.0013*

* $p<.05$, ** $p<.001$

Table 2. QUEST Severity Scores in the Patient Group

(*N*=30)

Chosen items	Related segment	QUEST severity score (%)		<i>p</i>
		HCP-non-short	HCP-short	
		<i>M</i> ± <i>SD</i>	<i>M</i> ± <i>SD</i>	
shoulder and elbow items	Humerus	91.15 ± 16.35 (<i>n</i> =6)	86.49 ± 15.14 (<i>n</i> =24)	.465
elbow and wrist items	Radius	99.44 ± 1.24 (<i>n</i> =5)	81.85 ± 26.38 (<i>n</i> =25)	.139
elbow and wrist items	Ulna	107.14 ± 14.29 (<i>n</i> =4)	81.34 ± 24.56 (<i>n</i> =26)	.030*
wrist, finger, grasp items	Second etacarpus	95.78 ± 3.66 (<i>n</i> =12)	76.79 ± 19.97 (<i>n</i> =18)	.004*
wrist, finger, grasp items	Fifth metacarpus	97.66 ± 17.79 (<i>n</i> =12)	78.27 ± 21.29 (<i>n</i> =18)	.035*

* $p<.05$

There was no significant relationship between patient age and the percentage of shortness in the humeral ($r=.03$, $p=.88$), radial ($r=.18$, $p=.34$), ulnar ($r=.12$, $p=.51$), the second metacarpal ($r=.07$, $p=.70$), and the fifth metacarpal segments ($r=.03$, $p=.88$). In the patient group, the percentage of upper extremity shortness correlated negatively with the QUEST severity score in the radial ($r=-.44$, $p=.01$), ulnar ($r=-.47$, $p=.01$), the

second metacarpal ($r=-.66$, $p=.00$) and the fifth metacarpal segments ($r=-.45$, $p=.01$). However, in the humeral segment, no significant relationship between QUEST severity score and percentage shortness was noted ($r=-.17$, $p=.37$) (Figure 1). MAS points for elbow flexor, wrist flexor and finger flexor correlated positively with percentage of shortness of each segmental limb segment ($p<.05$) except the humeral segment ($p>.05$). MAS points

Table 3. MAS Points in the Patients Group

Measure	Related segment	MAS point (mean value)		<i>p</i>
		HCP-non-short	HCP-short	
Shoulder adductor	Humerus	0.83	1.29	.168
Elbow flexor	Humerus	1.50	2.33	.194
	Radius	0.80	2.44	.014*
	Ulna	0.75	2.34	.018*
Wrist flexor	Radius	1.20	2.92	.014*
	Ulna	1.50	2.80	.070
	Second metacarpus	1.75	3.22	.002*
	fifth metacarpus	2.00	3.06	.027*
Finger flexor	Second metacarpus	2.00	3.11	.007*
	Fifth metacarpus	2.00	3.11	.017*

* $p<.05$

Table 4. Correlation between the Upper Extremity Shortness and MAS points in the patient group

Measure	Related segment	<i>r</i>	<i>p</i>
Shoulder adductor	Humerus	.23	.22
Elbow flexor	Humerus	.28	.12
	Radius	.57	<.001**
	Ulna	.39	.03*
Wrist flexor	Radius	.45	.01*
	Ulna	.54	.002*
	Second metacarpus	.58	<.001**
	fifth metacarpus	.49	.006*
Finger flexor	Second metacarpus	.46	.01*
	Fifth metacarpus	.55	.002*

* $p<.05$, ** $p<.001$

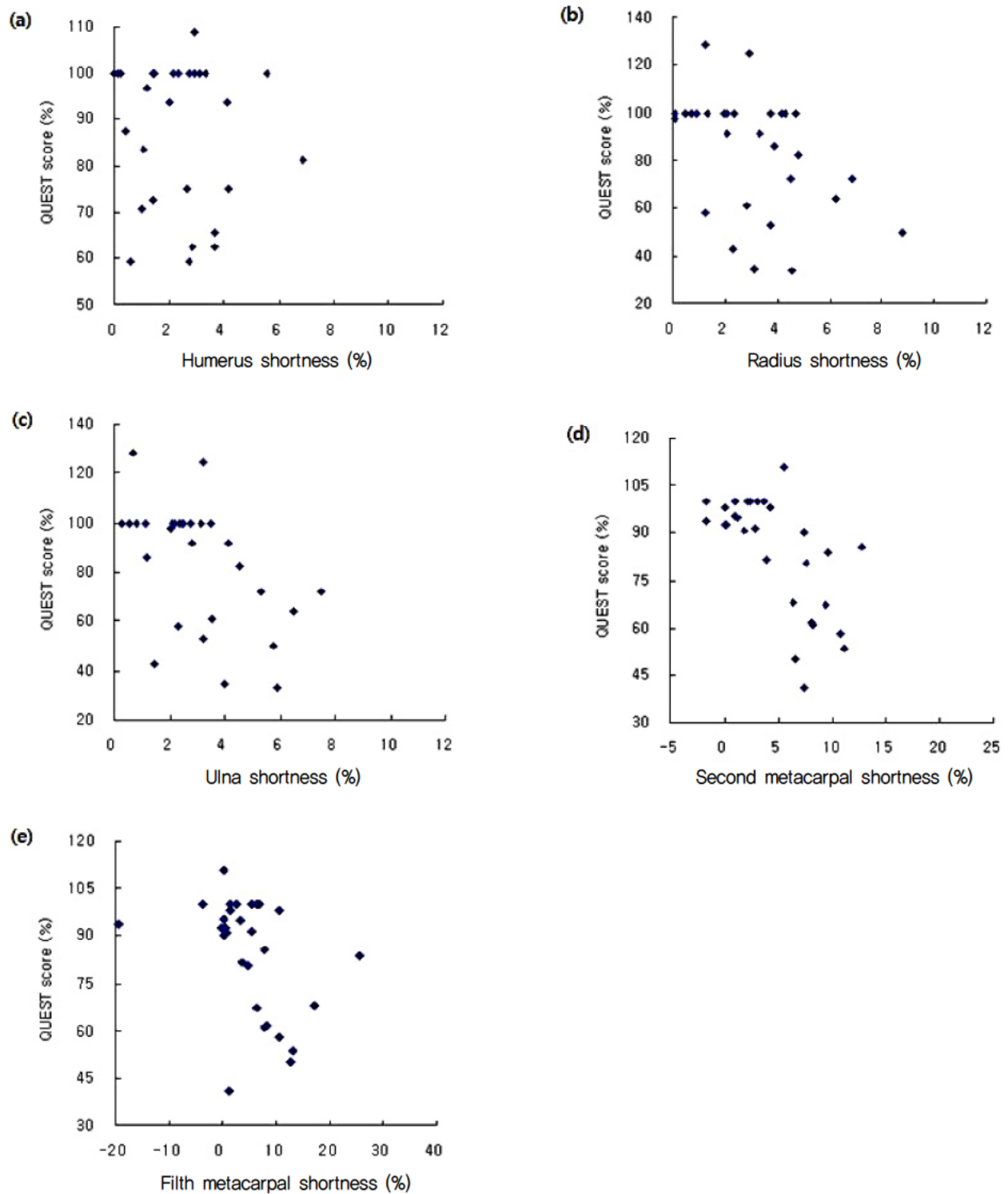


Figure 1. Correlation between upper extremity shortness and QUEST severity score in the patient group. The percentage of shortness in all segments, except the humeral segment, correlated negatively with QUEST severity scores (correlation coefficient: (a) -0.17 in the humeral segment, (b) -0.44 in the radial segment, (c) -0.47 in the ulnar segment, (d) -0.66 in the second metacarpal segment, (e) -0.45 in the fifth metacarpal segment).

for shoulder adductor was not significantly related to humeral length ($p>.05$) (Table 4). Children with a greater disparity in upper limb length tended to have a greater disparity in functional severity and muscle tone.

IV. Discussion

To clarify the correlation of upper extremity shortness in HCP with muscle tone and functional activity and to eliminate the confounding factors, it is necessary to review the literatures. The main variables causing residual limb shortening in children with HCP have been postulated to be nutritional factors and neurological or non-nutritional factors (Rempel, Colwell, & Nelson, 1988; Shapiro, Green, Krick, Allen, & Capute, 1986; Stevenson, Gressard, Cater, & Blackman, 1994). Nutritional factors include deficient nutritional intake, increased energy expenditure and decreased oromotor function, and they may have confounded the effects of non-nutritional factors on growth (Eddy, Nicholson, & Wheeler, 1965; Gisel & Patrick, 1988; Krick & Van Duyn, 1984). In this study, we chose children who were within normal stature and at a nutritional status in order to exclude such confounding factors. Neurological or non-nutritional factors causing poor growth in CP have been postulated to be either direct, through a negative neurotrophic effect on linear growth, or indirect, through the endocrine system, immobility or lack of weight-bearing (Jones, Mollano, Morcuende, Cooper, & Saltzman, 2004; Rempel et al., 1988; Tobis, Saturen, Larios, & Posniak, 1961).

We have found in clinical practice that children with HCP have the limb length discrepancy of their upper limbs as well as their lower limbs. In their hemiplegic upper limbs, there were the impairments in function and muscle tone that are important

elements for daily life activities. Many studies have been conducted on length difference and the cause of upper limb discrepancy, but there have been few studies on the association of hypofunction and increased muscle tone in the upper limb on the affected side with shortening (Gisel & Patrick, 1988; Jones et al., 2004; Krick & Van Duyn, 1984; Rempel et al., 1988).

Thus, we first evaluated the degree of the shortness of the affected upper limb in children with HCP. Since there are no acceptable criteria for the evaluation of the difference in length and the lengths of the upper limbs continue to grow in children, it is more reasonable to examine the ratio of the length of the involved upper limb to that of uninvolved upper limb than to simply compare the difference in length between both upper limbs (Demir et al., 2006; Stevenson et al., 1995). If a certain measurement value exceeds 2 standard deviations from the mean percentage of upper extremity shortness in the control group, there would be a significant discrepancy in the length of the upper limb. Also, it is possible to investigate the difference in the length of the upper limb in children with HCP by specifically examining the shortness of the humerus, ulna, radius, second metacarpal and fifth metacarpal bone, which represent the arm, forearm and hand. In this study, a significant shortening was observed in each segment in the patient group compared to the control group. The measurements exceeded the mean percentage of upper limb shortness in the control group in 18~24 (60%~80%) of 30 patients, which is consistent with the result of the studies by Stevenson et al.(1995) and Van Heest, House and Putnam(1993). However, in this study, the upper extremity shortness did not show a significant correlation with patient ages, which is inconsistent with the result of the earlier study by Van Heest et al.(1993). Since the ages of our patients narrowly ranged from 4.25 to 8.87

years, it was inappropriate to analyze upper extremity shortness in terms of age.

In this study, the correlation between upper extremity shortness, spasticity and function in the involved upper limbs in children with HCP was examined. It seems likely that the increased muscle tone or the decreased function of the upper limbs shown in HCP patients may be attributed to non-nutritional factors as well as the underlying disease itself.

Spasticity in cerebral palsy has both neurophysiological and musculoskeletal components. The former includes abnormalities in muscle tone, primitive reflexes and movement and/or postural control responses, whereas the latter includes abnormal changes in connective tissue, muscle and bone, and in children the rate of bone growth relative to muscle growth. Other studies have shown that mechanical stress is essential to the developmental biology of bones (Carter, 1987; Carter, Orr, Fyhrick, & Schurman, 1987). Endochondral ossification is inhibited by a high compressive-dilational force and accelerated by an intermittent shear stress in vivo. Bone formation and growth are products of biomechanical demand placed across growing bone. In our results, spasticity with elbow, wrist and finger flexor correlated positively with the percentage of shortness of each limb segment. Consequently, it can be postulated that in children with HCP, intense spasticity preventing active movement may decrease mechanical stress which is critical to early skeletal development.

Carter et al.(1987) have demonstrated that skeletal morphogenesis fails to progress in the absence of mechanical stress. Whalen, Carter and Steele(1988) have indicated that activity level and exercise are predominant factors in the regulation of bone density or bone mass. In our study, QUEST was used to assess motor functions of the patients.

The QUEST is an objective standardized measure with good inter-rater reliability and validity that evaluates upper extremity function in children with CP aged from 18 months to 8 years. It evaluates 4 domains that included 34 items: dissociated movement, grasp, weight bearing and protective extension (DeMatteo et al., 1993; Law et al., 1991). Upper limb motor function correlated with the lengths of the forearm and hand bones in our patients. This result does not correspond with that of the earlier study by Stevenson et al.(1995) which reported that only hand length correlates with functional severity. This difference may be because they performed only the prehension subtest of the QUEST which reflects the severity of neurological function of the hand, but not of the arm, whereas we assessed selected items for each limb segment: shoulder and elbow items for humeral length; elbow and wrist items for radial or ulnar length; wrist, finger and grasp items for the second or fifth metacarpal bone length. However, in our study, there is the question of why the percentage of shortness of the humeral segment did not show any significant correlation of the degree of spasticity during shoulder adduction and elbow flexion with QUEST scores measured for the shoulder and elbow items. Further studies are needed to determine whether the humeral segment is less affected by mechanical stress or whether our result is caused by measurement errors or the small number of patients.

The limitation of this study lies in the fact that we did not study the relation between the percentage of upper extremity shortness and other important clinical factors such as bone age or sensory function. Roberts, Vogtle and Stevenson(1994) have documented, in a study of 19 HCP children aged a mean of 8.6 years that the upper limbs of hemiplegia have a degraded skeletal maturation compared to the uninvolved side and that

there is a significant correlation between functional severity and bone age. Uvebran(1998) reported a relationship between the severity of undergrowth and sensory deficits as determined by the presence of stereognostic dysfunction. However, Van Heest et al.(1993) did not find a linear relationship between stereognosis deficits and undergrowth in the affected limbs. We could not evaluate sensation because the patients in our study were too young (median age, 6.3 years) to reliably assess sensory deficits.

V. Conclusion

Our study showed that children with HCP had some degree of upper limb length discrepancy on the affected side compared with the control children. Hemiplegic upper limb length discrepancy correlated with upper limb function. Moreover, there was a significant correlation between spasticity and upper extremity shortness. Further research is needed to evaluate changes in upper extremity shortness and function after spasticity management. This understanding of muscle tone and motor function influencing limb length discrepancy in children with HCP will lead to a proper clinical care and may contribute to a decrease in socio-emotional stress and to a better quality of life.

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국문초록

편마비성 뇌성마비 환자의 상지길이 단축과 이와 연관된 임상적 요소

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목적 : 본 연구는 편마비성 뇌성마비 환자들의 편마비측 상지길이 단축의 정도를 평가하고, 길이 단축과 근경직도 및 운동기능과의 연관성을 알아보고자 하였다.

연구방법 : 30명의 편마비성 뇌성마비 환자(남아 13명, 여아 17명, 평균연령 6세 4개월)와 17명의 대조군(남아 9명, 여아 8명, 평균연령 6세 2개월)을 대상으로 하였다. 상지의 상완골, 척골, 요골, 두 번째와 다섯 번째 중수골의 길이를 단순방사선 촬영을 통하여 길이 측정을 하였고, 건측(대조군에서는 우성측) 대비 편마비측(대조군에서는 비우성측)의 단축의 정도는 백분율로 나타내었다. 근경직도는 Modified Ashworth Scale (MAS)를 이용하였고, 상지의 운동기능은 Quality of Upper Extremity Skills Test (QUEST)를 사용하였다.

결과 : 대조군에 비하여 환자군에서는 편마비측 상지길이의 백분율이 의미있게 단축되어 있었다 ($p<.05$). 이러한 상지길이의 단축 정도와 근경직도의 증가 및 운동기능의 저하는 의미있는 연관성을 보였다($p<.05$).

결론 : 편마비성 뇌성마비 환자에서 보이는 편마비측 상지길이의 단축은 근경직 및 운동기능과 의미있게 연관되어 있다. 추후 이러한 환자들에게서 근경직도의 완화 및 운동기능의 향상을 도모하는 재활치료 후 변화되는 상지길이의 단축 정도를 연구한다면 재활치료의 효과를 좀더 객관적으로 증명하고, 치료의 방법과 정도를 정립하는데 도움이 될 것이다.

주제어 : 근경직도, 뇌성마비, 상지길이, 운동기능, 편마비