

The relationship between different upper extremity patterns and independence level in individuals with spastic cerebral palsy from the ICF perspective

Spastik serebral palsili bireylerde farklı üst ekstremitte patternleri ve bağımsızlık seviyesi arasındaki ilişkinin ICF perspektifine göre incelenmesi

Hasan Bingöl^{1,2}, Hikmet Kocaman^{2,3}, Mintaze Kerem Günel⁴

¹ Vocational School of Health, Mus Alparslan University, Muş, Turkey

² Department of Physiotherapy and Rehabilitation, Institute of Health Sciences, Hacettepe University, Ankara, Turkey

³ Research Assistant, PT, MSc, Faculty of Health Science, Karamanoğlu Mehmetbey University, Karaman, Turkey

⁴ Faculty of Physiotherapy and Rehabilitation, Hacettepe University, Ankara, Turkey

ORCID ID of the author(s)

HB: 0000-0003-3185-866X

HK: 0000-0001-5971-7274

MKG: 0000-0001-6895-2495

Corresponding author / Sorumlu yazar:
Hasan Bingöl

Address / Adres: Muş Alparslan Üniversitesi Sağlık Hizmetleri MYO, Kulp Yolu 7. Km. Muş, Türkiye
E-mail: hesenbingol@gmail.com

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Abstract

Aim: Although various upper limb and hand involvement patterns in individuals with cerebral palsy (CP) have been defined, our knowledge about their functionality in daily life is still insufficient. The purpose of this study was to investigate upper extremity involvement patterns concerning the level of functionality in individuals with spastic CP.

Methods: A total of 101 individuals, aged 7 to 21 years, with spastic cerebral palsy (30% unilateral CP, 70% bilateral CP), and a total of 172 hand and upper limb patterns were evaluated in this study. To identify different spastic upper extremity patterns, two classification systems, one for the upper limb and one for the hand (Classification of Upper Limbs and Hand Patterns), were used separately. Then, the Manual Ability Classification System [MACS] and Functional Independency Measure [Wee-FIM] were utilized to quantify hand functions and functional independency level, respectively, in the activities of daily living.

Results: A strong correlation was found between MACSI and Simple Flex of Hand Pattern ($r=0.72$) while a moderate correlation was detected between MACSII and Simple Flex of Hand Pattern ($r_2=0.57$). Besides, the level of independence in daily living activities was consistent with Type Ia and Type Ic patterns of the upper limb ($r_1=0.56$ and $r_2=0.44$)

Conclusion: It was concluded in the light of the obtained data that Type Ia and Type Ic patterns of the upper limb, as well as Simple Flex of the hand pattern, are very efficient for functionality. Additionally, the pattern of Simple Flex Plus and Intrinsic Punching Hand were significantly related to bad capacity in hand functioning. The various upper limb and hand patterns affect the functionality or functional independence in daily living. Consequently, abnormal upper limb and hand patterns, which commonly occur in the upper extremities of children with CP, are quite different from each other in terms of functionality. Hence, it is recommended that before the application of BoNT-A or orthopedic surgery in managing spasticity, the client should be comprehensively evaluated in both terms of rigidity and functionality.

Keywords: Cerebral Palsy, Upper Limb, Pattern, ICF, ADL, Independency

Öz

Amaç: Serebral palsili (SP) bireylerde şu ana kadar çeşitli el ve kol tutulum patternleri tanımlanmış olsa da, günlük yaşamdaki işlevsellikleri hakkındaki bilgilerimiz hala yetersizdir. Bu çalışmanın amacı, spastik SP'li bireylerde üst ekstremitte tutulum patternlerini işlevsellik düzeyine göre araştırmaktır.

Yöntemler: Çalışmaya yaşları 7-21 yıl arasından değişen toplam 101 spastik SP'li tanımlı birey (%30 unilateral SP, %70 bilateral SP, toplam 172 el ve kol patterni) dahil edildi. Anormal el ve kol patternlerini tanımlamak için El ve Kol Patternleri Sınıflandırma Sistemleri ayrı ayrı kullanıldı. Sonrasında, çalışmaya dahil edilen hastaların el işlevsellikleri ve günlük yaşam aktivitelerindeki bağımsızlık düzeyleri El Becerileri Sınıflandırma Sistemi (EBSS) ve Fonksiyonel Bağımsızlık Ölçütü (FBÖ) kullanılarak belirlendi

Bulgular: EBSS I ve Simple Flex el patterni arasında yüksek düzeyde bir ilişki bulunurken ($r=0.72$) ve EBSS II ve Simple Flex el patterni arasında orta düzeyde bir ilişki tanımlanmıştır ($r=0.57$). Ayrıca, Tip Ia ve Tip Ic kol patternleri ile günlük yaşam aktivitelerindeki bağımsızlık seviyesi arasında orta derecede bir ilişki saptandı ($r_1=0.56$ ve $r_2=0.44$)

Sonuç: Elde edilen verilerin ışığında; bir yandan Tip Ia ve Tip Ic kol patternlerinin diğer yandan Simple Flex el patterninin fonksiyonellik açısından verimli olduğu sonucuna varıldı. Ek olarak, Simple Flex Plus ve Intrinsic Punching Hand el patternlerinin el işlevsellik açısından birbirlerinden farklıdır. Bundan Ötörü spastisite yönetiminde sıklıkla başvurulan yöntemlerden biri olan BoNT-A tedavisine karar vermeden önce hasta hem rijit hem de fonksiyonel açıdan kapsamlı bir şekilde değerlendirilmelidir.

Anahtar kelimeler: Serebral palsy, Üst ekstremitte, Pattern, ICF, GYA, Bağımsızlık

Introduction

Cerebral Palsy (CP) is a non-progressive yet permanent condition that occurs as a result of damage to the developing brain and is characterized by postural and movement disorders as well as activity limitations [1]. Upper motor neuron (UMN) lesion results in several forms of motor impairment ranging from neural-related ones, such as spasticity, to non-neural related ones, due to changes in the mechanical properties of muscles. These two interact with each other, causing muscle hypertonia, leading to increased resistance to passive motion in the upper limb [2]. Consequently, muscle hypertonia increases energy consumption during activities by interference with voluntary muscle movements [3].

Studies have shown that the increase of tone in upper limbs of children with CP, which occurs around the age of 10 years [4], and is mainly predominant in the shoulder (adductors and internal rotators), forearm (pronators), elbow, wrist and finger (flexors), leads to structural and functional contractures [5,6]. Hyperactivity of different muscle combinations in CP forces the affected extremity into abnormal posture and movement patterns [7]. Chaleat-Valayer et al. [8] developed two different classification systems by identifying different upper limb and hand patterns that were common in patients with CP. These abnormal postures or movement patterns that occur as a result of spasticity adversely affect upper extremity functions such as reaching, grasping-releasing and manipulating objects [7, 9]. Secondary body structure-function problems in children with CP affect their manual ability and have a direct impact on their performance in daily living such as feeding, dressing, toilet-bathing; and an indirect effect on their participation in the social environment, school and home activities.

The International Classification of Functioning, Disability and Health (ICF) is a conceptual framework that describes the effects of disability on individuals' daily living and social participation beyond physical injury and disability. In general, the ICF model is a conceptual framework that gives a dynamic idea about evaluation, goals and interventions [10]. Various rehabilitation approaches focus on ICF's activity and participation domains rather than on body structure/function domain. However, the relationship between the main domains of ICF is not yet fully understood [11]. The relationship between the main domains of ICF mostly relies on evaluation and is based on cross-sectional studies published previously [7,12,13]. In contrast, several clinical studies have shown that the relationship between different domains of ICF is complicated, and the improvements in activity and participation related to intervention are also influenced by individual and environmental factors [11,14,15]. These results make it difficult to think of the ICF framework for treatment and goals planning.

In their study, Kim and Park tried to show the results of spasticity on the main domains of ICF by using path analysis [12]. Since BoNT-A is a popular intervention to manage pediatric upper limb hypertonia and improving function in the spastic upper extremity [7], the professionals working in this field must have detailed information on all the spectrum of upper extremity impairment (related to all areas of ICF). Patterns caused by muscle hypertonia should be evaluated precisely

before deciding on BoNT-A injection as an adjunctive treatment method [16-18]. Also, according to the current consensus published in 2009, in non-focal conditions such as CP, body structures and functions (contracture and decreased range of motion), as well as the severity of the functional level (Gross Motor Classification System, Manual Ability Classification System) should be taken into account. The use of BoNT-A in children with CP must target a muscle group rather than a single muscle [19].

Based on the above premise, the present study aimed to examine different upper limb and hand patterns in children with spastic CP and investigate these patterns concerning functionality in daily living. The hypothesis of this study was that in children with spastic CP, upper limb and hand patterns differentiate depending on the severity of involvement and these abnormal patterns adversely affect the functionality of the upper limbs and hands.

Materials and methods

The study protocol was approved by the Scientific Research and Ethical Board of Mus Alparslan University with the decision number E.2065 on 2/13/2018. Before inclusion into the study, informed consent was obtained from the parents or participants. A total of 101 patients (children and adults) with unilateral and bilateral CP (aged 7-21 years) were enrolled in this current study to investigate the relationship between the independence in daily living and the abnormal upper limb and hand patterns. The study also aimed to establish a resource document to serve for decision making of BoNT-A injection in the spastic upper limb(s) of the patients. The inclusion criteria included (1) being diagnosed with spastic CP, (2) having had no previous surgical interventions such as muscle lengthening, (3) having had no previous BoNT-A treatment, and (4) being older than seven years of age. Based on the static posture positions of the upper limbs and hands, patterns of each were classified according to the Classification of Upper Limb Patterns and Classification of Hand Patterns (with the permission of R. Bard-Pondarre), respectively. Considering the unilateral or bilateral involvement of participants enrolled in this study, a total of 172 upper limb and hand patterns were evaluated. Detailed clinical observations and classification of abnormal spastic patterns of the patients were performed while the subjects were in sitting or standing positions without being exposed to any activity. In patients with bilateral spastic CP, patterns of each upper extremity were evaluated separately considering the asymmetric involvement.

Manual performance of each subject was measured based on how the children (CP) use their hands in daily life or children's designated hand function of capacity in a clinical setting. Functional ability in daily living (what a child with a disability does) were exhibited for the tasks of such as self-care, eating, dressing, mobility, and communication skills.

Outcome Measures

Upper Limb and Hand Patterns Classification System

This is based on a previous study on the classification of spastic upper limb and hand patterns in adult stroke patients [16]. Chaleat-Valayer et al. [8] developed a similar classification for

patients with CP. In their study on intra- and inter-rater reliability of both classification systems, the same researchers concluded that both these classification systems had good reproducibility in children with CP [8].

Manual Ability Classification System

The Manual Ability Classification System (MACS) is used to quantify the severity of upper limb involvement. It classifies how children with cerebral palsy use their hands to handle objects in daily activities [20]. This system includes five levels of hand function, from Level I, indicating that the child is capable of easily manipulating objects without restrictions in daily living activities, to Level V, indicating that the child requires full assistance to handle objects. [21]

Functional Independence Measure

Functional Independence Measure (Wee-FIM) comprises rating that best describes the child’s level of function related to self-care, mobility and cognitive skills and can be used for all children with developmental disorders ranging from 6 months to 21 years of age. It consists of 18 items, and each item is scored between 1 and 7. Of these scores, 6 and 7 represent “INDEPENDENT”, 3 to 5 represent “ASSISTANCE REQUIRED”, and 1 and 2 indicate “DEPENDENT”. Taking this classification into consideration, we can sub-categorize the total scores as following: Any score between 18-36 represents “DEPENDENT”, between 37-90 represents “ASSISTANCE REQUIRED”, and between 91-126 indicates “INDEPENDENT”.

Mapping Instruments by Using ICF-CY as A Reference Framework

ICF is a classification system or framework in which tools that assess body structure and functionality, as well as activity and participation are mapped. This broad framework organizes assessments based on their content and enables them to focus on selecting relevant aspects of functionality and disability during evaluation [22]. The outcome measures were categorized according to ICF main domains consisting of components from body structure/impairment, activity and participation [10]

Statistical Analysis

Statistical analyses were performed using the IBM SPSS Statistics v.22. Hand patterns, according to upper limb patterns, manual ability level according to hand patterns, and upper limb patterns according to Wee-FIM levels, were given using cross-tabulations. The calculated correlation coefficient (r) or the strength of the relationship was related to how similar the values of the two variables were. In contrast, the direction of the association was determined by analyzing whether the increase or decrease in the values of one variable was in the same direction with the increase or decrease in the values of the other variable. The relationship between the two variables –in percentage– was interpreted as follows: Values of <20% were considered small, 40-59% were deemed moderate, and 60- 79% and above were considered large [23].

Results

Age, gender, type of CP, GMFCS and MACS levels of the participants are presented in Table 1.

Type Ia (36.04%) and Type Ib (17.6%) accounted for the majority of the upper limb patterns of 172 included in the

study, and a small proportion showed Type III b (2.3%) pattern. Besides, Simple Flex and Simple Flex Plus yielded proportions of 41.1% and 22.3%, respectively (Table 2).

When we look at the distribution between manual ability levels (MACS) and hand patterns of the participants (See Table 3), the majority of 18 hand patterns with MACS level I were associated with Simple Flex hand pattern (72.2%), whereas 5.5% and 22.2% were related to Total Flex and Intrinsic Punching Hand patterns, respectively. According to these results, there was a strong relationship between MACS level I and Simple Flex Hand pattern. Likewise, there was a moderate relationship between MACS level II (somewhat reduced quality of handling objects) and MACS level III and the Simple Flex Hand pattern (56.9% and 50% respectively). As a result, MACS levels I and II, in which manual ability does not limit independence in daily living activities, are associated with high and moderate levels of Simple Flex Hand pattern. On the other hand, there was a very weak relationship between MACS levels IV and V, and Simple Flex Hand pattern (13.6% and 12.2% respectively).

Table 1: Characteristics of the Participants

Age	Gender		Type of CP		GMFCS					MACS				
	Female	Male	Bilateral Dominant	Unilateral Dominant	I	II	III	IV	V	I	II	III	IV	V
11.62 (4)	45	56	71	30	22	19	26	24	10	10	39	11	12	29

MACS: Manual Ability Classification System, GMFCS: Gross Motor Classification Systems

Table 2: Upper Extremity Pattern Distributions, n, number of upper limb or hand patterns; %, percent for each pattern

Type of Upper Limb Patterns	n	%	Type of Hand Patterns	n	%
Type Ia	62	36.04	Simple Flex	70	41.1
Type Ib	30	17.6	Total Flex	12	7.01
Type Ic	10	5.2	Simple Flex Plus	38	22.3
Type IIa	21	12.2	Total Flex Plus	8	5.2
Type IIb	32	18	Intrinsic Punching Hand	32	18.8
Type IIc	6	3.4	Superficial Punching Hand	3	1.7
Type IIIa	7	4.06	Profound Punching Hand	9	5.2
Type IIIb	4	2.3	-	-	-
Total	172	100		172	100

Table 3: The Relationship between Manual Ability Level and Hand Patterns

MACS Levels	Hand Patterns						n
	Simple Flex	Total Flex	Simple Flex Plus	Total Flex Plus	Intrinsic Punching Hand	Superficial Punching Hand	
I	13(72.2)	1(5.5)	-	-	4(22.2)	-	18
II	37(56.9)	3(4.6)	9(13.8)	3(4.6)	11(16.9)	-	65
III	9(50)	2(11.1)	3(16.6)	1(5.5)	2(11.1)	-	18
IV	3(13.6)	2(9)	9(40.9)	1(4.5)	3(13.6)	2(9)	22
V	6(12.2)	4(8.1)	17(34.6)	4(8.1)	12(24.4)	2(4)	49

MACS: Manual Ability Classification System, n: number of hand patterns for each MACS level, % percentage of each defined hand patterns

The majority of the total of 49 patterns related to MACS level V, which corresponds to the bad capacity associated with the manual ability where objects cannot be grasped, or total help is required, are associated with Simple Flex Plus and Intrinsic Punching Hand patterns (34.6% and 24.4% respectively). The relationship between MACS level I, II, III and Total Flex hand pattern is remarkably close to 0. More patterns are needed to determine the relationship between these variables.

Table 4 and Figure 1 show the distributions of upper limb patterns at different levels of independence in daily life and the Wee-FIM score range of each upper limb pattern, respectively. According to Table 4, while there was a moderate relationship between Type Ia and Type Ic upper limb patterns and the “INDEPENDENT” level in daily life (56.4% and 44.4% respectively), Type Ib, Type IIb, Type IIc and Type IIIb upper limb patterns had a weak correlation with the level of independence in ADL. In contrast, there is no relationship between Type IIa and Type IIIa upper limb patterns and the

“INDEPENDENT” level in daily life. More patterns are needed to determine the relationship. While the relationship between “ASSISTED” level in daily living and Type IIc of upper limb pattern was very strong (83.3%), Type IIb had a lower than moderate relationship (41.9%), and Type Ia had a weak relationship (35.4%).

Table 4: The relationship between Upper Limb Patterns and Wee-FIM Levels

Upper Limb Patterns		Wee-FIM Levels			Total
		INDEPENDENT	ASSISTED	DEPENDENT	
Type Ia	n	35	22	5	62
	%	56.4%	35.4%	8.06%	43.2%
Type Ib	n	8	8	14	30
	%	26.6%	26.6%	46.6%	21.6%
Type Ic	n	4	2	3	9
	%	44.4%	22.2%	33.3%	5.7%
Type IIa	n	1	4	16	21
	%	4.7%	19.04%	76.1%	11.4%
Type IIb	n	5	13	13	31
	%	16.1%	41.9%	41.9%	12.5%
Type IIc	n	1	5	0	6
	%	16.6%	83.3%	0.0%	2.3%
Type IIIa	n	0	2	5	7
	%	0.0%	28.5%	71.4%	2.3%
Type IIIb	n	1	2	3	6
	%	16.6%	33.3%	50%	1.1%
Total	n	55	58	59	172
	%	100.0%	100.0%	100.0%	100.0%

n: number of upper limb patterns for each independency level, % percent; Wee-FIM: Functional Independency Measure

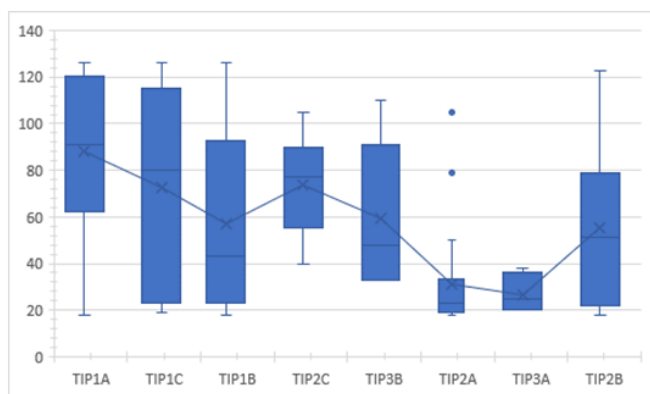


Figure 1: Wee-FIM score distribution according to upper limb patterns

Discussion

This study sought to explore different upper limb and hand involvement patterns observed in children with spastic CP concerning functionality in daily activities. Overall, MACS I, MACS II, and MACS III were associated with Simple Flex Hand pattern. That is, MACS I-III levels corresponding to moderate-minor problems in hand function were compatible with the Simple Flex Hand pattern at a decreasing rate. Based on these distributions, it can be concluded that Simple Flex Hand pattern is efficient in daily activities. Besides, a moderate correlation was found between Type Ia and Type Ic patterns of the upper limb and functional independence in daily living.

On the other hand, a moderate-high relationship was found between Type IIa, Type IIIa, and Type IIIb of upper limb patterns and dependent level in daily living. Additionally, our results indicated that Type Ia of upper limb pattern was moderately functional; instead, Type Ic upper limb pattern is slightly functional. Conversely, it was found out that Type IIa and Type IIIa upper limb patterns were associated with significantly poor functionality.

Considering the sub-classification of upper limb patterns made according to elbow postures [8], it can be observed that the pronator teres and biceps brachii muscles are mainly affected. Studies have shown that these muscles or

supination-pronation movements of forearm affect hand patterns and therefore, manual ability [7]. According to the results of the study conducted by Kane et al. [24], wrist movements are positively associated with the complex bone-soft tissue structures, forearm rotations influence carpal kinematics, effective use of wrist and hand requires good coordination of the wrist and forearm, and rotation of forearm around its long axis for various degrees of pronation or supination need normal range of motion of the wrist and forearm (This is essential in nearly all daily activities, from simple movements such as turning keys and opening doors to complex movements such as throwing a baseball).

MACS Level I and II, which correspond to the effective use of the hand, were more compatible with the Simple Flex Hand pattern. Studies have shown that such a posture is consistent with the functional use of the hand [25]. MACS Level IV and V, which require continuous or total help to perform part of the activity, are often associated with "Simple Flex Plus" and "Intrinsic Punching Hand" patterns. This can be explained based on the fact that these two patterns are more complex postures where both the intrinsic and extrinsic structures of the hand are affected, according to the definition of the related classification resources: "Wrist flexion is associated with swan neck deformity or dinosaur hand (exclusion of the index finger during grasping)". Studies investigating hand patterns have concluded that the atypical posture in hand is often accompanied by ulnar deviation and that the increased wrist-finger flexion prevents functional use of the hand [26].

Normal selective hand and finger movements, free from the arm movements, which enhance the development of graduated movements required for the high quality of grasping and releasing objects, are not easy to perform. As some authors have suggested, hand functions do not consist solely of the functional sensory activation or intrinsic structures of the hand, so it can be concluded that the effective use of the hand in daily life is related with the upper limb posture as well as hand postures. It is in this context that rehabilitation approaches focusing on proximal segments such as the elbow and forearm responsible for positioning and orientation of the hand, or rehabilitation approaches focusing on increasing active ROM, may improve hand functions.

Considering the data on the relationship between upper limb patterns and independency levels in daily living, the highest level of independence in daily living was associated with Type Ia pattern. In contrast, the highest dependency was more related to Type IIa. In this regard, our results about the suitability of these two different patterns in terms of functional use are supported by several studies. In this context, various researchers examining the relationship between manual ability and the upper limb concluded that the maximum reach amount was directly related to the joint range of motion in the shoulder [27]. Another study reported that when there was not enough movement in the shoulder joint, a significant increase in trunk movements was observed during reaching as a compensation mechanism [28]. Similarly, the relationship between the amount of supination and pronation of the forearm and hand functions was examined in another study, and it was concluded that while the forearm rotates around its long axis, various degrees of pronation or

supination allows the orientation of movements leading to the effective use of the hand [24].

Limitations

A limitation of our study was the lack of CP population with similar topographic involvement. Although considered, creating a homogenous group would have caused a smaller sample size. Thus, further research is needed to investigate the functionality of the upper limb and hand involvement patterns in a more homogenous and larger population. However, despite this limitation, our study results are the first and precious because of its unique findings.

Conclusion

One of the primary purposes of orthopedic surgeons working with CP is to improve muscle hypertonia based on the assumption that this will increase motor functions. Now the question is whether spasticity is a problem and whether it always needs to be treated. This question can be answered based on the fact that in any case of muscle weakness in the lower extremities, the factor that enhances standing upright is skeletal muscle spasticity in favor of extension. However, the situation may be slightly different for the upper extremities, which often exhibit functionality in the open kinetic chain. Here, muscle weakness associated with spasticity should also be considered. As a result, both hypertonia and hypotonia in upper extremity muscles compromise the effectiveness of hand use in daily activities (e.g. eating, bathing, and dressing). Therefore, before deciding on BoNT-A injection or orthopedic surgery for the upper limb muscles, which are the most frequently used methods in the management of spasticity, the patient should be evaluated comprehensively in terms of rigidity and functionality. That is, when deciding on BoNT-A injection or orthopedic surgery for the upper limb muscles, it would be more appropriate to evaluate the patient's activity or participation estimation as well as a range of motion, estimated muscle tone, and pain. Otherwise, the injection may adversely affect the level of independence in daily living by disrupting the existing upper extremity posture, which is already functional for the patient. Consequently, it was concluded that Type Ia and Type Ic patterns for the upper limb, and Simple Flex Hand pattern for the hand were the most convenient patterns in terms of functional use.

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