Is there a relationship between upper limb function and swallowing function in children with cerebral palsy? A cross-sectional study

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Abstract

Objective: Despite the association between gross motor and swallowing functions in patients with cerebral palsy (CP), there have been no studies examining the relationship between upper limb functions and swallowing in detail. The aim of this study was to evaluate the relationship between upper extremity skills and swallowing function in children with CP.

Methods: The study included a total of 71 patients with CP who were attending the rehabilitation clinic. Upper limb functions were assessed using the Bimanual Fine Motor Function (BFMF) scale, and swallowing function with the Functional Oral Intake Scale (FOIS). The Nine-hole peg test (NPHT) was used to assess manual hand dexterity. Grip strength was measured with a Jamar hand dynamometer and pinchmeter. Correlation analysis was applied to outcome parameters. *Results:* The BFMF classification was level 1 in 6 patients (8.5%), level 2 in 22 patients (31.0%), level 3 in 27 patients (38.0%), level 4 in 12 patients (16.9%) and level 5 in 4 patients (5.6%). Nutrition was provided through oral intake in 59 (83.1%) patients and 12 (16.9%) were tube dependent. While a negative correlation was determined between swallowing function and BFMF and NHPT, a positive correlation was determined between swallowing function and grip strength values.

Conclusions: The findings of this study demonstrated that there is a relationship between swallowing functions and upper limb functions. These findings may help in predicting functional improvement in terms of swallowing and/or if the patient needs further intervention such as upper limb rehabilitation in addition to oral motor training to improve oral intake, and thereby nutritional intake.

Keywords: Cerebral palsy; upper extremity, swallowing, grip strength

INTRODUCTION

Cerebral palsy (CP) describes a group of permanent and non-progressive disorders of the development of movement and posture, causing activity limitations, which are attributed to non-progressive disturbances occurring in the developing fetal brain or within the first 2 years of life.¹

CP may cause spasticity, muscle contractures, weakness and coordination difficulties affecting the ability to control walking, swallowing and speech articulation.² Dysphagia is one of the common findings in children with cerebral palsy. Dysphagia in children with CP usually occurs in parallel with other developmental disorders such as impaired cognitive, and fine and gross motor skills.³ Previous studies have shown that swallowing difficulty or dysphagia is closely related to gross motor function.⁴ Poor gross motor function and manual ability have also been found to be strongly associated with poor oral motor functions (swallowing and speech).⁵ To date, there are no published studies that have examined the relationship between upper limb functions only and swallowing function.

Swallowing function and the upper extremities are closely related, as the action of swallowing starts with taking the food to the mouth with the hand and upper extremity movements.

For the reasons mentioned above, the aim

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of this study was to investigate the relationship between upper extremity skills and swallowing function in children with cerebral palsy.

METHODS

This cross-sectional study included a total of 71 patients with CP aged 6 - 12 years, who were attending the Physical Medicine and Rehabilitation (PMR) clinic. Those included were hemiplegic, diplegic and triplegic patients with a Gross Motor Function Classification System (GMFCS) classification between I and IV.

Patients with bilateral severe tetraplegic involvement, a history of orthopedic surgery or botulinum toxin injection in the past 6 months, epilepsy or any other disease and drug use that would interfere with physical activity were excluded from the study.

Written informed consent for participation in the study was obtained from the parents or legal guardian of each patient. Approval for the study was granted by the Local Ethics Board of the hospital (Approval date 21/04/2021, number E2-21-367) and all procedures were applied in accordance with the principles of the Helsinki Declaration.

Demographic data and clinical characteristics

A record was made for each patient of age (years), gender, height (cm), weight (kg), GMFCS level, type of CP and motor limb distribution (hemiplegia, diplegia and triplegia/quadriplegia). Body mass index was calculated using height and weight values. The characteristics of the patients were also recorded, including history of prematurity, multiple pregnancy, birth trauma and previous lung infection.

The primary caregiver was asked if the patient had mental retardation, epilepsy, hearing and visual impairment, dental problems, and speech problems such as aphasia/dysphasia/dysarthria accompanying cerebral palsy. Mental retardation was evaluated by a child psychiatrist, and hearing and visual impairments were evaluated by otolaryngologists and ophthalmologists and recorded as "present" or "absent". Speech problems were evaluated by a physiatrist with expertise in speech therapy with clinical knowledge to assess aphasia.

Motor function was determined with the Gross Motor Function Classification System (GMFCS). This system determines the best level of a child's existing abilities and limitations in gross motor function. The GMFCS levels range between I and V, where I= walks without limitation and V= has to be transported in a manual wheelchair.⁶

Evaluation of hand and upper limb

The dominant/non-dominant and affected/ unaffected hand sides of the patients were recorded. A record was made of the passive range of motion (ROM) for the shoulder (flexion/ extension, internal/external rotation, abduction/ adduction), elbow (flexion/extension, pronation/ supination), wrist (flexion/extension, radial/ ulnar deviation), thumb (flexion/extension for metacarpophalangeal and interphalangeal joint), and 2nd finger (flexion/extension for metacarpophalangeal, proximal/distal interphalangeal joint). The Modified Ashworth Scale with a score range of 0-4 was used for the assessment of spasticity.7 The Nine-hole peg test (NHPT) is a simple, fast, manual dexterity test based on performance (sec) with proven validity and reliability, which is particularly sensitive to changes in upper limb performance.8 The test material consists of 9 small, standard size bars and a nine-hole board on which to place them. The NHPT is performed while sitting. The patient takes the nine bars from one box on the table and places them in the holes of the other box as quickly as possible and removes them immediately after finishing. The time is measured with a stopwatch, starting from when the hand touches the first bar and finishing when the last bar is placed in the box.8

Grip strength was measured with a Jamar hand dynamometer. Measurements were taken with the subject sitting, and the elbow flexed as far as 90° and the wrist in semi-pronation with the thumb pointing upwards. The children were then instructed to make a gripping action with maximum force. In the affected and unaffected hand, 3 consecutive measurements were taken and the average values was recorded in kilograms. The lateral, palmar and fingertip grip strength values were evaluated in the same way with a pinchmeter.

The Bimanual Fine Motor Function (BFMF) scale was applied to assess hand function. The BFMF is a five-stage evaluation tool that considers each hand function separately. The higher the BFMF, the worse the motor hand function.⁹

The House hand functional classification scale (HHFCS) has nine subgroups (0 - 8), ranging from 0=does not use, to 8= active spontaneous use. Detailed functional levels are given to enable the identification of small functional improvements.¹⁰

Evaluation of swallowing function

The swallowing function was evaluated using the Functional Oral Intake Scale (FOIS). Scores range from 1-7, with higher scores indicating better swallowing function.¹¹

Study protocol

The relationship was investigated between the evaluation parameters of the intact and paretic extremities and the FOIS.

Statistical analysis

The power of the study was calculated using the G Power and Sample Size Statistical Program version 3.1.8 software. The minimum sample size was calculated as 34 for each group to provide 80% power, 0.20 effect size and set at 0.05 significance level. Data obtained in the study were analyzed statistically using the IBM Statistical Package for Social Sciences (SPSS) version 20.0 software (IBM Corporation, Armonk, NY, USA). The normal distribution of continuous variables was evaluated using the Kolmogorov-Smirnov test. Continuous variables (all variables are non-normal distribution) were expressed as median (minimum-maximum) values, and categorical variables as number (n) and percentage (%).Statistically significant differences in measurements between dominant and non-dominant extremities were evaluated with the Wilcoxon Signed Rank test. Relationships between upper limb outcome parameters and FOIS were analyzed using the Spearman Rho Correlation test. Correlation coefficients were rated as follows: 0.81-1.00 excellent, 0.61-0.80 very good, 0.41 0.60 good, 0.21-0.40 fair, and 0-0.20 poor. A value of p<0.05 was considered statistically significant.

RESULTS

The median age of the participants was 6.50 years (range, 6.00-11.00 years). The dominant hand was right side in 48 (67.6%) patients. Paresis was present in the left upper extremity in 48 (67.6%) patients. The clinical and demographic features of the patients are summarized in Table 1.

The BFMF classification was level 1 in 6 patients (8.5%), level 2 in 22 (31.0%), level 3 in 27 (38.0%), level 4 in 12 (16.9%) and level 5 in 4 (5.6%). A significant difference was found between the dominant and non-dominant extremities in terms of NHPT, grip strength and the HHFCS (p=0.001). (Table 2) The evaluation

results of the dominant and non-dominant upper extremities are presented in Table 2.

Nutrition was provided through oral intake in 59 (83.1%) patients (FOIS level 4-7) and 12 (16.9%) were tube dependent (FOIS level 1-3) (Table 3).

The results of the correlation analysis of the FOIS and upper limb outcome parameters are shown in Table 4. The BFMF levels were determined to be correlated negatively with the FOIS score (r=-0.425, p=0.001).

DISCUSSION

This study investigated the relationships between upper limb function and swallowing function in 71 patients with CP. The study results revealed that poor upper limb function was associated with worse swallowing function. To the best of our knowledge, this study is one of the first to determine the association between upper limb function and swallowing function in patients with CP.

Dysphagia may be seen in individuals with CP for reasons such as oral motor control disorders and abnormal neurological development.¹² The prevalence of oropharyngeal dysphagia in children with cerebral CP is uncertain, but is estimated to be between 19% and 99% depending on the definitions and tools used.¹³ Oropharyngeal dysphagia is characterized by difficulties in one or more stages of swallowing, such as oral-preparatory, oral propulsive, or the pharyngeal phase.¹⁴ In this study, dysphagia was found in 71.8% of the patients, which was consistent with the literature.¹⁵ In this study sample, 12 (16.9%) of the patients were tube dependent (level 1-3), and 59 (83.1%) had oral intake (level 4-7).

In previous studies, gross motor function has been shown to be one of the best predictors of swallowing function. Reilly *et al.* showed that more than 90% of individuals with CP have clinically significant oromotor dysfunction regardless of severity, and individuals with more severe gross motor dysfunction have a correspondingly more severe oromotor dysfunction.¹⁶ In three different studies evaluating the gross motor functions of individuals with cerebral palsy with GMFCS, it was stated that the rate of dysphagia was higher in patients with worse GMFCS scores.^{4,17-18}

In the only study in the literature in which upper extremity function was evaluated, Goh *et al.* applied the Manual Ability Classification System (MACS), which is a form of gross motor function classification for upper limb functions, the incidence of dysphagia was found to increase

Table 1: Demographic data and disease characteristics

	n=71
Age median (minimum-maximum)	6.50 (6.00-11.00)
Gender n(%)	
Girl	36 (50.7)
Boy	35 (49.3)
Height (cm)	125.00 (99.00-151.00)
Weight (kg)	19.00 (12.00-33.00)
Body Mass Index	13.60 (8.70-20.80)
Caregiver n(%)	
Mother	65 (91.5)
Grandmother	4 (5.6)
Father	2 (2.8)
History $n(\%)$	
Prematurity	41 (57.7)
Multiple pregnancy	10 (14.1)
Birth Trauma	9 (12.7)
None	11 (15.5)
Comorbidities <i>n</i> (%)	
Mental retardation	26 (36.6)
Auditory impairment	4 (5.6)
Visual impairment	34 (47.9)
Dental problem	35 (49.3)
Urinary/Fecal incontinence	22 (30.9)
Speech & language impairment	48 (67.6)
History of lung infection n(%)	17 (23.9)
Type of CP $n(\%)$	
Hemiplegia	42 (59.2)
Diplegia	13 (18.3)
Triplegia	16 (22.5)
GMFCS levels	
Ι	7 (9.9)
II	21 (29.5)
III	35 (49.3)
IV	8 (11.3)
V	0

CP: Cerebral palsy GMFCS:Gross motor function classification system

as GMFCS and MACS levels increased.5

As stated above, upper extremity functions in the literature have been scored based on motor function only, such as GMFCS and MACS. However, the function of the upper extremity becomes more complex in relation to nutrition, and it is not possible to make evaluations with these scoring systems. Therefore, for the first time in the literature, upper limb functions were evaluated in more detail, using the BFMF classification, grip strength and NHPT.

The bilateral ROM and spasticity of the entire

upper extremity were recorded, and grip strengths with Jamar dynamometer and pinchmeter. The NHPT was used, which evaluates manual dexterity based on performance, recorded in (seconds), and the BFMF classification system, which specifically evaluates fine motor skills. While a negative correlation was determined between swallowing function and BFMF and NHPT, a positive correlation was determined between swallowing function and grip strength values and HHFCS. In contrast to these results, it has been argued in a previous study that dysphagia is related to the

Table 2:	Evaluation	of hand	and	upper	extremity

	Dominant Hand	Non-dominant Hand	р
ROM median (min-max)			
Shoulder			
Flexion	180 (90.0-180.0)	180.0 (80180.0)	0.168
Extention	30.0 (10.0-30.0)	30.0 (10.0-30.0)	0.159
Internal rotation	90.0 (35.0-90.0)	90.0 (30.0-90.0)	0.482
External rotation	90.0 (30.0-90.0)	90.0 (30.0-90.0)	0.604
Abduction	180.0 (90180.0)	180.0 (90180.0)	0.761
Adduction	30.0 (30.0-30.0)	30.0 (30.0-30.0)	0.517
Elbow			
Flexion	135.0 (100.0-135.0)	135.0 (90.0-135.0)	0.143
Extention	0.0 (-25.0-0.0)	0.0 (-60.0-0.0)	0.092
Pronation	90.0 (20.0-90.0)	90.0 (10.0-90.0)	0.326
Supination	90.0 (90.0-90.0)	90.0 (90.0-90.0)	0.810
Wrist			
Flexion	80.0 (80.0-80.0)	80.0 (30.0-80.0)	0.145
Extention/	70.0 (45.0-70.0)	70.0 (40.0-70.0)	0.332
Thumb			
MCP joint flexion	70.0 (70.0-70.0)	70.0 (70.0-70.0)	0.719
MCP joint extention	0.0 (0.0-0.0)	0.0 (-5.0-0.0)	0.120
IP joint flexion	90.0 (90.0-90.0)	90.0 (70.0-90.0)	0.218
IP joint extention	30.0 (30.0-30.0)	30.0 (10.0-30.0)	0.272
2nd finger			
MCP joint flexion	90.00 (90.0-90.0)	90.0 (90.0-90.0)	0.157
MCP joint extention	45.00 (45.0-45.0)	45.00 (40.0-45.00)	0.255
PIP joint flexion	100 (100.0-100.0)	100.0 (90.0-100.0)	0.113
PIP joint extention	0.0 (0.0-0.0)	0.0 (-5.0-0.0)	0.546
DIP joint flexion	90.0 (90.0-90.0)	90.0 (90.0-90.0)	0.759
DIP joint extention	0.0 (0.0-0.0)	0.0 (-5.0-0.0)	0.228
Modified Aschworth Scale Shoulder			
Flexor muscle	0.0 (0.0-3.0)	0.0 (0.0-4.0)	0.128
Internal rotation muscle	0.0 (0.0-2.0)	0.0 (0.0-4.0)	0.128
Adductor muscle	0.0 (0.0-2.0)	0.0 (0.0-4.0)	0.647
Elbow	0.0 (0.0-4.0)	0.0 (0.0-4.0)	0.047
Flexor muscle	0.0 (0.0-3.0)	0.0 (0.0-4.0)	0.221
Pronator muscle	0.0 (0.0-3.0)	0.0 (0.0-4.0)	0.335
Wrist	0.0 (0.0-5.0)	0.0 (0.0-4.0)	0.555
Flexor muscle	0.0 (0.0-3.0)	0.0 (0.0-4.0)	0.167
Thumb	0.0 (0.0-5.0)	0.0 (0.0-4.0)	0.107
Flexor muscle	0.0 (0.0-2.0)	0.0 (0.0-3.0)	0.319
Adductor muscle	0.0 (0.0-1.0)	0.0 (0.0-3.0)	0.348
2nd finger	0.0 (0.0 1.0)	0.0 (0.0 5.0)	0.510
MCP joint flexor muscle	0.0 (0.0-1.0)	0.0 (0.0-2.0)	0.566
PIP joint flexor muscle	0.0 (0.0-1.0)	0.0 (0.0-3.0)	0.351
DIP joint flexor muscle	0.0 (0.0-1.0)	0.0 (0.0-2.0)	0.574
NHPT	```` /	. ,	
None	2 (2.8)	23 (32.4)	
Normal (<20 seconds)	0	0	0.001
Abnormal (≥ 20 seconds)	69 (97.2)	48 (67.6)	
Jamar dynamometer (kg)	4.0 (0.0-17.00)	0.0 (0.0-18.00)	0.001
Lateral grip strength (kg)	2.0 (0.0-11.00)	0.0 (0.0-8.00)	0.001
Palmar grip strength (kg)	2.0 (0.0-10.00)	0.0 (0.0-9.00)	0.001
Fingertip grip strength (kg)	1.0 (0.0-6.00)	0.0 (0.0-5.00)	0.001
House's functional classification	8.0 (1.0-8.0)	3.0 (0.0-8.0)	0.001
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ROM: Range of motion, MCP: Metacarpophalengeal, IP: Interphalangeal, PIP: Proximal nterphalangeal, DIP: Distal interphalangeal, NHPT:Nine Hole Peg Test, min:minimum, max:maximum

Functional oral intake scale levels $n(\%)$	n=71
1	0
2	2 (2.8)
3	10 (14.1)
4	14 (19.7)
5	14 (19.7)
6	11 (15.5)
7	20 (28.2)

Table 3: Distribition of Functional Oral Intake Scale (FOIS) results

Table 4: Correlation analysis of FOIS and upper limb function outcomes

	Dominant hand		Non-dominant Hand	
-	r	р	r	р
ROM				
Shoulder				
Flexion	0.245	0.039	0.242	0.042
Extention	0.032	0.443	0.230	0.154
Internal rotation	0.270	0.023	0.226	0.059
External rotation	0.082	0.497	0.255	0.103
Abduction	0.243	0.041	0.129	0.283
Adduction	0.192	0.109	0.152	0.057
Elbow				
Flexion	0.308	0.009	0.218	0.046
Extention	0.195	0.196	0.131	0.212
Pronation	0.251	0.042	0.235	0.048
Supination	0.127	0.037	0.219	0.032
Wrist				
Flexion	0.233	0.094	0.102	0.466
Extention	0.079	0.575	0.146	0.298
Thumb				
MCP joint extention	0.066	0.639	0.239	0.084
IP joint extention	0.051	0.692	0.049	0.725
2nd finger				
MCP joint extention	0.120	0.330	0.133	0.341
PIP joint extention	0.246	0.075	0.138	0.325
DIP joint extention	0.105	0.455	0.056	0.692
Modified Ashworth Scale				
Shoulder				
Flexor muscle	-0.266	0.025	-0.168	0.162
Internal rotation muscle	-0.275	0.020	-0.207	0.083
Adductor muscle	-0.113	0.347	-0.119	0.321
Elbow	01110	01017	01115	01011
Flexor muscle	-0.062	0.607	-0.177	0.140
Pronator muscle	-0.244	0.044	-0.169	0.160
Wrist			0.107	0.100
Flexor muscle	-0.268	0.027	-0.082	0.298
Thumb	0.200	V•V#1	0.002	0.270
Flexor muscle	-0.248	0.037	-0.201	0.093
Adductor muscle	-0.252	0.040	-0.025	0.837
2nd finger	-0.434	v.v - v	-0.025	0.057
MCP joint flexor muscle	-0.113	0.149	-0.031	0.796
PIP joint flexor muscle	-0.347	0.003	-0.031 -0.324	0.790
DIP joint flexor muscle	-0.242	0.003	-0.102	0.397
•				
NHPT	-0.242	0.042	-0.066	0.584
Jamar dynamometer (kg)	0.274	0.021	0.223	0.061
Lateral grip strength (kg)	0.184	0.125	0.142	0.237
Palmar grip strength (kg)	0.219	0.066	0.198	0.091
Fingertip grip strength (kg)	0.042	0.731	0.043	0.720
House's functional classification	0.457	0.001	0.401	0.001
220 and 5 Functional clubbilication	0.101	0.001	011VI	0.001

r:correlation coefficient, ROM: Range of motion, MCP: Metacarpophalengeal, IP: Interphalangeal, PIP: Proximal nterphalangeal, DIP: Distal interphalangeal, NHPT:Nine Hole Peg Test

level of involvement of gross motor functions rather than upper limb functions.⁵

There were some limitations to this study. First, the low number of participants. Further evaluations with a larger number of children with CP are needed to analyze more significant differences in swallowing function between groups of different upper extremity skills. Another limitation of the present study was the very limited number of children with poor swallowing function, compared to the number of children with good swallowing function. Furthermore, the lack of reliability of dysphagia scales for pediatric patients was another limitation of this study.

In conclusion, the findings of this study demonstrated that swallowing function was worse in CP patients with poor upper limb function. Raising awareness of this relationship may help with early intervention. The findings of this study may help to predict functional improvement in terms of swallowing and/or if the patient needs further intervention such as upper limb rehabilitation in addition to oral motor training to improve oral intake, and thereby, nutritional intake. Nevertheless, further studies with larger sample sizes are required to confirm the relationship between dysphagia and upper limb functions, particularly fine motor skills.

DISCLOSURE

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