Original Article / Özgün Araştırma

Oral Health Status of Children with Cerebral Palsy who Have Dysphagia: A Comparative Study

Disfajisi Olan Serebral Palsili Çocuklarda Ağız Sağlığı Durumu: Karşılaştırmalı Bir Çalışma

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Abstract

Objective: This study aimed to compare oral health status of children with and without cerebral palsy (CP) who have dysphagia.

Materials and Methods: The study included two groups of children with and without CP who suffer from dysphagia. The parents filled a written questionnaire about demographic variables. The oral motor functions of the children, including mouth breathing, tongue thrust, lip closure, tongue posture, severity and frequency of drooling, swallowing, chewing- and eating-related functions, malocclusion and dental caries status were evaluated.

Results: Between-group comparisons showed a significant difference in swallowing functions, chewing and drooling, mouth breathing, lip closure, tongue thrust and malocclusion. However, no significant difference in oral hygiene habits was found between the groups. These results indicated that the orofacial structures of children with CP were more affected than those of children without CP.

Conclusion: Swallowing disorder in CP affects oral functions and oral health negatively as in other neurological diseases. Early diagnosis and management of dysphagia are important to improve the quality of life of children with CP. Therefore, a multidisciplinary approach, including dentists, is necessary for early diagnosis and treatment.

Keywords
Cerebral palsy, dysphagia, drooling, oral health, swallowing, swallowing disorders

Anahtar Kelimeler
Serebral palsi, disfaji, salya akıtma, ağız sağlığı, yutma, yutma bozuklukları

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Öz

Amaç: Bu çalışmanın amacı, disfajisi tanısı alan serebral palsili (SP) olan ve olmayan çocukların ağız sağlığı durumlarını karşılaştırmaktır.

Gereç ve Yöntemler: Çalışmaya disfaji tanısı alan SP'li olan ve olmayan iki grup çocuk dahil edilmiştir. Ebeveynler demografik değişkenler hakkında bir anket doldurmuştur. Ağız solunumu, dil itme, dudak kapanışı, salya akıtma şiddeti ve sıklığı, yutma, çiğneme ve yeme ile ilgili fonksiyonlar, malokluzyon ve diş çürükleri değerlendirilmiştir.

Bulgular: Gruplar arasındaki karşılaştırma sonucunda, yutma fonksiyonları, çiğneme ve salya akıtma, ağız solunumu, dudak kapanışı, dil itme ve malokluzyon açısından anlamlı bir fark saptanmıştır. Ancak gruplar arasında ağız hijyeni alışkanlıkları açısından anlamlı bir fark bulunmamıştır. Bu çalışmanın sonuçları, SP'li çocuklarda orofasiyal yapıların SP'li olmayan çocuklardan daha fazla etkilendiğini göstermiştir.

Sonuç: SP'de yutma bozukluğu, diğer nörolojik hastalıklarda olduğu gibi ağız fonksiyonlarını ve ağız sağlığını olumsuz etkilemektedir. Disfajinin erken teşhisi ve tedavisi SP'li çocukların yaşam kalitesini iyileştirmek için önemlidir. Bu nedenle, erken tanı ve tedavi diş hekimlerinin de dahil olduğu multidisipliner bir yaklaşımla yapılmalıdır.

Introduction

Dysphagia is defined as any disruption to the swallow sequence (1). Dysphagia is a symptom of multiple functional disorders associated with the central nervous system in children (1,2). This condition may be due to anatomical or neurological maturation abnormalities, sensory disturbance of the oral cavity or esophageal motility disorders (3). Although approximately 1% of children in the general population experience dysphagia, the incidence rate is much higher in children with neurological diseases (1,4).

Cerebral palsy (CP) refers to a group of non-progressive disorders of movement and posture associated with an immature brain defect (5). It is one of the most common neurological diseases in children with a prevalence of 1.2-2.5 in 1,000 live births in industrialized countries (6). Despite the fact that the technological possibilities increase, the incidence of CP has increased over the years. The situation can be resulted from increasing opportunities for the survival of preterm, low birth weight infants and keeping better records of CP (7). It is a more common condition in Turkey with 4.4-8 at 1,000 live births (6,8). The high prevalence in Turkey is related with excess prevalence of consanguineous marriage; excessive infectious and febrile illnesses, inadequacy of nutrition in infants; negativity in birth conditions; inadequate baby care and diseases during pregnancy (2).

Dysphagia, drooling, tooth decay, enamel hypoplasia, erosion, gingival disease, orthodontic disorders, bruxism, eruption problems, trauma are some of the effects of CP on the oral region (9).

Dysphagia is a common problem for many children with CP (1). However, many children with other muscular disorders, acquired brain injuries, craniofacial or airway malformations, as well as those with respiratory, cardiac or gastrointestinal diseases, also experience the same problem (1). According to the literature, the prevalence of dysphagia in children with and without CP are 19.2-60% (9) and 25-45%, respectively (10).

Dysphagia may cause respiratory problems, insufficient nutrition, growth retardation, motor dysfunction, aspiration, chronic lung diseases and general health deterioration. Moreover, nutrition may be a stressful and undesirable process for all children and their parents who have negative experiences while eating. Dysphagia causes negative interactions between the family and the child. Feeding difficulty and the increased feeding time reduce the quality of life of families and increase the risk of depression (11,12). Motor dysfunction that causes dysphagia may result in mouth breathing. Therefore, dysphagia may adversely affect oral health, oral functions and occlusal development (13,14).

The aim of this study was to compare the oral health statuses of children with and without CP who have dysphagia. In the literature there is no studies on this subject in primary dentition. CP may cause oral health problems even in the primary dentition. It was thought that the quality of life of children with CP and their families can be increased with early measures.

The hypothesis of the study was that swallowing disorder in CP affects oral health and oral function negatively.

Materials and Methods

The sample was selected from among children who were admitted to the Dysphagia Research and Application Center. Considering the number of patients admitted to the center within a year, the sample size was determined to be at least 25 children in each group by using power analysis. As a result, the sample size of this study was determined as 53 children with CP and dysphagia (CP group) and 50 children without CP but with dysphagia [non-cerebral palsy (NCP) group]. Children with primary dentition were included. Hence, children with congenital anomalies that could affect their orofacial development (e.g. cleft lip/palate) were excluded from the study. Before the clinical examination, the parents singed an informed consent form. This study was approved by the Hacettepe University Ethics Committee for

Non-Interventional Clinical Studies (decision no: GO 14/550-12, date: 05.11.2014).

The study's primary outcomes were set to diagnose dysphagia by taking detailed anamnesis, clinical examination, evaluation of nutrition and imaging with videofluoroscopy and/or fiberoptic endoscopic evaluation of swallowing by a physiotherapist specialized in dysphagia. Additionally, the children's oral motor functions including swallowing, chewing and eating functions, as well as drooling, were determined by a pediatric dentistry specialist who has trained by a dysphagia physiotherapist, by using Oreland's scale (15).

Secondary outcomes included the oral health statuses of the children. To determine this, an interview was conducted with the parents who were asked about the medical history, feeding and oral hygiene habits of their children. Intraoral examination consisted of the eruption status of teeth which were recorded according to Logan and Kronfeld's scale (16). Furthermore, the World Health Organization diagnostic criteria were used to determine the dmft/s values (decayed, missing because of caries and filled tooth numbers/surfaces in primary teeth), and gingival health statuses were determined using the gingival, dental plaque and gingival enlargement statuses. Mouth breathing, tongue thrust, macroglossia, lip closure and tongue posture were assessed in each patient by using Oreland's scale (15).

At the end of the study, all children and their parents were informed about oral hygiene practices, and modifications were made for the patient according to their status of disability. Children with co-operation were treated for caries and gingivitis. Uncoordinated and un-cooperative children were referred to the same department for application of general anesthesia for the dental procedures.

Statistical Analysis

The data were analyzed by the SPSS statistics software, version 20.0. Descriptive statistics were calculated as frequency/percentage (n/%) for the qualitative data and mean±standard deviation for the quantitative data. Using chi-squared and t-tests, comparisons between the independent and dependent variables were made in 95% confidence intervals. The odds ratio was assessed for statistically significant outcomes.

Results

The distribution of the children according to age and sex is given in Table 1. The mean age of the CP group was 43.67 months [standard deviation (SD): 13.06], and the mean age of the NCP group was 41.68 months (SD: 13.64). No significant difference was found between the groups in terms of age (p=0.669) and sex (p=0.588).

When the first encountered about dysphagia was asked, most children with CP (41.5%) were observed to have symptoms of dysphagia immediately after birth. In the control group, most of the children (48%) showed signs of dysphagia in transition to solid food intake. Additionally, dysphagia was noticed during feeding in 43 (81.1%) and 29 (58%) children with and without CP, respectively (Table 2).

The results of the questionnaire about breast feeding showed that 74 (71.8%) children were breast-fed. The number of the children who were breast-fed for less than 6 months was 59 (57.3%). No significant difference was found in terms of the duration of breast-feeding or bottle-feeding in the comparison of the children with and without CP (p>0.05).

The eruption status of teeth in 72 (69.9%) children were normal according to Logan and Kronfeld's scale. No significant difference was found about teeth eruption status between the two groups.

As shown in Table 3, DMFT and DMFS scores and in Table 4, gingival health status, presence of dental plaque and gingival overgrowth of the children with and without CP. However, the children with

Table 1. Distribution of children based on age and sex

	Cerebral palsy group		Non-cerebral palsy group		Total	
	n	Month	n	Month	n	Month
Female	24	43.67	20	41	44	42.45
Male	29	43.86	30	42.13	59	42.98
Total	53	43.77	50	41.68	103	42.75
Standard deviation	-	13.06	-	13.64	-	13.39

and without CP showed no statistically significant differences in terms of the mean values of dmft, dmfs and gingival health and plaque indices. There was a statistically significant difference in gingival overgrowth between the children with and without CP (p<0.05).

A significant difference was determined in terms of chewing and swallowing functions, drooling frequency and severity between the two groups (p<0.05). A negative relationship was determined between lip closure (p<0.000) and drooling frequency (p=0.000) and severity (p=0.000), mouth breathing (p=0.000), lip closure, macroglossia (p=0.000), tongue thrust (p=0.000) and tongue position (p<0.005). The differences between the two groups were also found for occlusal problems including anterior open bite and high palate (Table 5).

Discussion

Feeding histories indicated that a large proportion of the children had swallowing disorders within the first years of life which preceded the diagnosis of CP in

Table 2. Feeding problems of the children

		Cerebral palsy group		Non-cerebral palsy group		Total		p
		n	%	n	%	n	%	
When did you notice the swallowing disorder in your child?	When he/she was born	22	41.5	17	34	39	37.9	0.010
	While switching to solid food	10	18.9	24	48	34	33	
	Passed after infection	12	22.6	6	12	18	17.5	
	Sequelae after seizure	9	17	3	6	12	11.Jul	
How did you notice the swallowing disorder in your child?	While feeding (cannot swallow, vomiting, retching, bruising, refusing food)	43	81.1	29	58	72	69.9	0.011 (OR: 0.321)
	Cannot chew	10	18.9	21	42	31	30.1	

OR: Odds ratio

Table 3. Caries status of children

	Cerebral palsy group			Non-cerebral palsy group			p
	n	mean	Standard deviation	n	mean	Standard deviation	
DMFT	53	3.60	5.365	50	4.04	5.525	0.685
DMFS	53	9.68	20.562	50	7.98	15.787	0.641

Table 4. Oral health status of children

		Cerebral palsy group		Non-cerebral palsy group		p
		n	%	n	%	
Gingiva health status	Healthy gingiva	6	11.3	3	6	0.339
	Inflame gingiva	47	88.7	47	94	
Plaque	Present	50	94.3	47	94	0.660
	Absent	3	5.7	3	6	
Degree of gingival enlargement	0	18	34	33	66	0.010
	1	16	30.2	8	16	
	2	14	26.4	8	16	
	3	5	9.4	1	2	

Table 5. Oromotor dysfunction						
		Cerebral palsy		Non-cerebral palsy group		p
		n	%	n	%	
Swallowing	Normal	2	3.8	10	20	0.014
	Infantil	12	22.6	6	12	
	Presence of swallowing reflex	28	52.8	30	60	
	Absence of swallowing reflex	11	20.8	4	8	
Chewing	<3	40	75.5	28	56	0.017
	3-5	9	17	7	14	
	6-10	1	1.9	10	20	
	>10	3	5.7	5	10	
Mouth breathing	Present	38	71.2	9	18	0.000
	Absent	15	28.8	40	80	
Lip closure	Present	18	34	41	82	0.000 OR:0.116
	Absent	35	66	9	18	
Tongue position	Ahead	33	62.3	19	38	0.017 OR: 0.383
	Behind	20	37.7	31	62	
Tongue thrusting	Present	34	64.15	5	10	0.000
	Absent	19	35.85	45	90	
Macroglossia	Present	25	47.17	4	8	0.000 OR: 0.101
	Absent	28	52.83	46	92	
Severity of drooling	Dry	5	9.4	32	64	0.000
	Mild	10	18.9	2	4	
	Middle	9	17	7	14	
	Severe	7	13.2	2	4	
	Too severe	22	41.5	7	14	
Frequency of drooling	Never	5	9.4	32	64	0.000
	Sometimes	15	28.3	6	12	
	Frequently	12	22.6	5	10	
	Always	21	39.6	7	14	
Open bite	Present	22	41.5	7	14	0.002
	Absent	31	58.5	43	86	
High palate	Present	39	73.6	17	34	0.000
	Absent	14	26.4	33	66	
OR: Odds ratio						

many cases (17). According to the results of the study, dysphagia occurred in 41.5% of the children with CP and in 34% of the children without CP just after birth. 74% of the children in the NCP group had no diagnosed chronic diseases, the others had diseases that do not constitute a basis for swallowing disorders

such as heart murmurs and asthma. In other words, children in the NCP group had an acquired swallowing disorder, not a congenital.

In 81.1% of the children with CP and 58% of the NCP children, it was noticed as a result of neglecting eating, coughing or vomiting. Prematurity plays an important role in the etiology of CP (18). No difference was detected about prematurity between the two groups. It is therefore difficult to rule out the possibility that prematurity could have had a role in the oromotor dysfunctions of the two groups.

The influence of swallowing pattern on development of malocclusion has been a subject of studies, and a correlation between the type of swallowing and several malocclusion symptoms has been suggested (19-21). In this study, 22.6% of the CP and 12% of the NCP children had immature swallow, and 20.8% of the CP and 4% of the NCP children did not have any swallowing reflexes. Comparison of the malocclusion frequency of the CP and NCP groups revealed that 75.5% of the CP and 54% of the NCP children had any occlusion anomalies. The etiological factor of malocclusion might be attributed to the swallowing pattern. Melsen et al. (22) evaluated the sucking habits in healthy children with permanent dentition and stated that tongue thrust and teeth apart swallow increased frequency of distal occlusion, extreme maxillary overjet and open-bite. In our study, the common finding in both groups was having open-bite and high palate with a statistically significant difference (open-bite by 41.5% and 14% and high palate by 73.6% and 34% in the CP and NCP groups, respectively). The other findings in both groups was having under-jet, deep-bite, cross-bite, diastema, crowding and midline deviation with no statistically significant difference. The muscles of the face and the oral cavity play an important role in facial growth and occlusal development (23). Hence, the disturbances of the facial, masticatory and tongue musculature cause abnormal facial growth and increase the incidence of malocclusion (23). A number of studies reported greater prevalence of malocclusion in those without CP (24-30). The manifestation of malocclusion in CP has been attributed to the low tonicity of the facial muscles and the uncoordinated movement of the lip and tongue (24,25,31,32). Jackson suggested that disturbances of the facial, masticatory and tongue musculature are the cause of the increasing incidence of orthodontic problems and showed children diagnosed with CP at primary dentition to have more normal or minor malocclusions than older patients with CP whose deranged neuromuscular complexes had a longer period of time to bring about maldevelopment (33).

Chewing, oral food transport and swallowing constitute a continuum (24). These processes, taken together, are often considered to represent the entirety of the feeding process (34). Chewing efficiency, defined as the ability to grind a certain portion of a test food during a given time, is closely related to the number of occluded teeth (35). Shwartz et al. (36) found a significant correlation among chewing efficiency, age and number of posterior teeth in patients with CP. They stated that foods must be broken into smaller pieces through chewing, and it is dealt with based mostly on the efficiency with a full complement of molar teeth. The results of this study showed that the percentages of nonoccluded molar teeth in the CP and NCP children were 40.1% and 38%, respectively. Poor chewing efficiency is associated with few occlusal contacts (37). However, other factors including more extracted teeth, unrestored teeth, poorer oral hygiene and gingival health might contribute to the availability of posterior teeth for chewing (35,38,39).

Delayed tooth eruption is a common finding in children with general developmental delays that involve the oral musculature. Pope and Curzon (40), Moslemi et al. (38) and Rodrigues dos Santos et al. (41) reported delayed time of permanent teeth eruption in children with CP. However, Wessels (42) stated that the eruption status of both primary and permanent teeth in children with CP did not differ significantly from healthy children. In this study, 37.7% of the CP and 30.1% of the NCP children showed delayed primary tooth eruption. The results of this study revealed that, apart from the number of occluded teeth, delayed tooth eruption might also contribute to swallowing disorders in both CP and NCP children.

Unfortunately, no study was in literature about children with dysphagia and CP in primary dentition to compare with our study about caries statues or periodontal diseases. However, in relation to oral health, children with CP are more prone to caries and periodontal diseases than healthy children are (23,43). The results of this study revealed that

children in both groups had high incidence of caries with no statistically significant difference between the two groups (DMFt=3.60 and DMFS=9.68 in the CP group, DMFT=4.04 and DMFS=7.98 in the NCP group). There was also no statistically significant difference in relation to gingival health. Oral health problems occurring as a result of dysphagia are the main reason of orofacial disorders and absence of oral hygiene habits. Therefore, children with dysphagia and their parents should be instructed about practicing oral hygiene and the necessity of routine dentistry follow up.

Conclusion

According to the results of this study, swallowing disorder in CP did not affect oral health negatively and swallowing disorder in CP affected oral function negatively.

The results of this study showed that the risk of children with dysphagia to develop orofacial disorders, dental caries and gingival problems is worrisome. Considering that these problems will increase with age, functional factors disrupting orofacial development must be identified and eliminated as soon as possible. Moreover, long-term preventive measures and dental treatment strategies should be planned. Thus, early diagnosis and treatment should involve a multidisciplinary approach including dentists.

Ethics

Ethics Committee Approval: This study was approved by the Hacettepe University Ethics Committee for Non-Interventional Clinical Studies (decision no: GO 14/550-12, date: 05.11.2014).

Informed Consent: Before the clinical examination, the parents singed an informed consent form.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Concept: S.Ö., N.D., Design: N.D., Supervision: S.S.A., M.T., Fundings: İ.M.G., Materials: İ.M.G., Data Collection or Processing: İ.M.G., Analysis or Interpretation: M.T., S.S.A., Literature Search: S.Ö., Critical Review: S.Ö., N.D., Writing: İ.M.G., M.T., S.S.A.

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Questionnaire

EXAMINATION FORM TO EXAMINE THE EFFECT OF SWALLOWING DISORDER IN CHILDREN WITH AND WITHOUT CEREBRAL PALSIED CHILDREN DURING PRIMARY DENTITION ON TOOTH AND JAW STRUCTURES

Date: .../.../20..

Child's name and surname: .....

1. Gender: M....
   F.....

2. Date of birth: ..../..../.....
3. Was your child born on due date?
   0. No (At what week of pregnancy was the child born?.....)
   1. Yes
4. Have you had any illnesses during your pregnancy?

   0. No
   1. Yes (Note.....)
5. Are there any chronic diseases of him/her diagnosed by the doctor?
   0. No
   1. Yes (Note.....)

6. Is there any medication he/she uses regularly?
   0. No
   1. Yes (Note.....)

7. When and how did you first notice the swallowing disorder in your child?

8. How long your child had just breast milk?
   0. Never breastfed
   1. …… months
   2. I don't know / don't remember

9. Does your child use a pacifier or baby bottle?
   0. Never used
   1. He/she used for a while (How long did he/she use? …….. months).
   2. Still using
10. Are your child's teeth cleaned?
    0. No
    1.Yes (with……)
11. Does your child receive fluoride?
    0. No
    1.Yes (in what way .....? What is the frequency and dosage? .....)

12. Has your child ever had a dental examination?

    0. No
    1. Yes

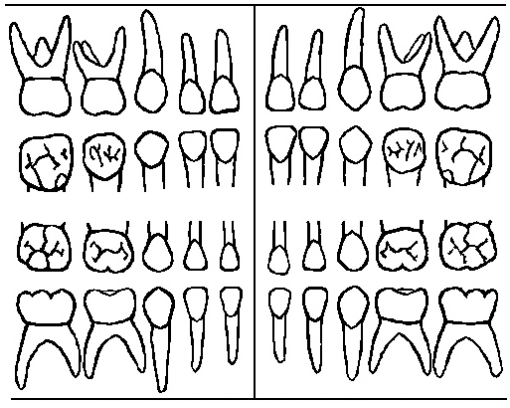
Intraoral Examination

13. Oral photos:
    0. Taken
    1. Not taken

14. Dental development status:
    0. Chronologically behind by age
    1. Normal
    2. Chronologically ahead by age
15. Number of teeth to close:
    0. 0-2
    1. 3-4
    2. 5-6
    3. 7-8
16. Wear condition in canine and molar teeth:
    0. No
    1. Mild (limited to enamel)
    2. Moderate (reached dentine)
    3. Severe (reached dentine and loss of occlusal anatomy)

17. Malocclusion status:
    0. No
    1. Yes

a. Overjet
b. Underjet
c. Deepbite
d. Openbite
e. Crossbite (anterior, posterior, unilateral, bilateral)
f. Crowding
g. Diastema
h. Midline deviation
i. High palate
j. Other (……..)



DMFT: …….
DMFS: …….

21. Gingival Health Status:
    0. Normal
    1. Inflammation
22. Presence of plaque:
    0. No plaque
    1. There is plaque
23.The severity of gingival enlargement:
    0. No gingival enlargement
    1. Gingival enlargement only involves the interdental papilla
    2. Gingival enlargement involves the papillae and the gum edge

3. Gingival growth covers ¾ of the crown or more

Evaluation of Oral Functions (Oreland 1989):

24. Swallowing:
    0. Normal
    1. Infantile
    2. There is a swallowing reflex
    3. No swallowing reflex
25. Chewing:
    0. <3
    1. 3-5
    2. 5-10
    3. >10
    4. Unable to cooperate
26. Mouth breathing:
    0. None
    1. Yes
    2. Cannot be determined
27. Lip closure:
    0. No
    1. Yes
    2. Cannot be determined
28. Tongue posture:
    0. Front
    1.behind
29. Tongue thrusting:
    0. Yes
    1. None
30. Macroglossia
    0. Yes
    1. None

31. Drolling:
    Violence
    Dry (no drooling)
    Mild (lips only)
    Medium (wet lips and chin)
    Severe (dresses get wet too)
    Very violent (clothes, hands, toys…)

    Frequency
    Never wet
    Occasionally
    Often
    Always

    32. Nutritional assessment:
    0. Liquid
    1. Puree
    2. Thick
    3. Mix

    Reliability level of information:

    Person filling out the form: