



Ultrasonographic Evaluation of Swallowing Disorder in Children with Cerebral Palsy: Preliminary study

Serebral Palsili Çocuklarda Yutma Bozukluğunun Ultrasonografik Değerlendirmesi: Ön çalışma

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Abstract

Aim: Swallowing is a process influenced by many phases and complex neuromuscular mechanisms. Swallowing disorders are common in children with cerebral palsy (CP). The aim of this study is the dynamic evaluation of oral and pharyngeal phase changes of swallowing by ultrasound (US) in healthy and children with CP with swallowing disorder.

Material and Method: Sixteen children with CP (9 boys, mean age 45±21 months) in the patient group and 20 healthy children (11 boys, mean age 60±26 months) in the control group enrolled study. CP group was selected from orally fed children. In both groups, the measurements were performed with the transducer was placed under the chin and anterior part of the neck. During swallowing and rest distances between mandible symphysis and hyoid bone (MH) and between hyoid bone and thyroid cartilage (HT); frame rate of displays to determine swallowing times at same levels, tongue thickness and shear wave elastography for measuring tongue stiffness determined.

Results: The significant differences were found between tongue thickness, elastogram and screen frame rates at HT level of the two groups (respectively $p<0,002$, $p<0,002$, $p<0,003$). No significant difference was found between the patient and control groups in terms of other measurements. Muscle tone change was observed during rest in patients with CP with dysphagia. This tonus was high in spastic CP cases. Active tone was slightly decreased. The high tongue elastographic values during rest was consistent with this information in the literature. The most important change detected in children with CP is the increase in image frame rate, which indicates a long swallowing time in HT level.

Conclusion: Prolonged swallowing time, thickness of the tongue and tonus of the tongue muscles can be used to evaluate dysphagia in patients with CP. We think that pre- and post-treatment comparisons can be used to evaluate treatment effectiveness.

Keywords: Cerebral palsy, dysphagia, ultrasound, elastography

Öz

Amaç: Yutma birçok fazdan ve karmaşık nöromusküler mekanizmalardan etkilenen bir süreçtir. Serebral palsi (CP) olan hastalarda yutma bozuklukları yaygındır. Bu çalışmanın amacı, sağlıklı ve yutma bozukluğu gelişen CP'li çocuklarda ultrason (US) ile yutmanın oral ve faringeal faz değişikliklerinin dinamik olarak değerlendirilmesidir.

Gereç ve Yöntem: Hasta grubunda 16 CP'li (9 erkek, ortalama yaş 45 ± 21 ay) ve 20 sağlıklı çocuk (11 erkek, ortalama yaş 60 ± 26 ay) kontrol grubuna alındı. CP grubu oral yoldan beslenen çocuklardan seçildi. Her iki grupta da ölçümler dönüştürücü ile çene altına ve boynun ön kısmına yerleştirildi. Mandibula simfizi ile hyoid kemik (MH) arasındaki ve hyoid kemik ile tiroid kıkırdağı (HT) arasındaki yutma ve dinlenme mesafelerinde; aynı seviyelerde yutma sürelerini belirlemek için ekranların kare hızı, dil kalınlığı ve dil sertliğini ölçmek için kayma dalgası elastografisi belirlendi.

Bulgular: İki grubun HT düzeyinde dil kalınlığı, elastogram ve ekran kare hızları arasında anlamlı fark bulundu (sırasıyla $p < 0,002$, $p < 0,002$, $p < 0,003$). Hasta ve kontrol grubu arasında diğer ölçümler açısından anlamlı fark bulunmadı. Disfajili CP çocuklarda istirahat sırasında kas tonusunun değiştiği görüldü. Spastik CP vakalarında bu tonus yüksekti. Aktivitede tonus biraz azaldı. Dinlenme sırasındaki yüksek dil elastografik değerleri literatürdeki bu bilgilerle tutarlıydı. CP'li çocuklarda tespit edilen en önemli değişiklik, HT düzeyinde uzun bir yutma süresine işaret eden görüntü kare hızındaki artıştır.

Sonuç: CP'li çocuklarda disfajiyi US ile değerlendirmek için uzamış yutma süresi, dil kalınlığı ve dil kaslarının tonusu kullanılabilir. Bu noninvaziv ve tekrarlanabilir yöntemin tedavinin etkinliğini değerlendirmek için kullanılabileceğini düşünüyoruz.

Anahtar kelimeler: Serebral palsi, disfaji, ultrason, elastografi.



INTRODUCTION

The swallowing process depends on the integrity of a complex neuromotor mechanism. It involves a coordinated process that is divided into the following phases: oral preparatory, pharyngeal and esophageal.^[1] Dysfunction is one of these three phases which can result in dysphagia. This is a term for difficulty swallowing. It corresponds to a set of symptoms characterized by difficulty in propelling liquid or solid food from the oral cavity through the esophagus.^[2,3] During the deglutition, it is necessary to protect the airways in order to avoid aspiration pneumonia.

Cerebral palsy (CP) is a common cause of disability in childhood. Since different regions are affected during the development of the nervous system, clinical findings differ.^[4] Children with CP commonly have feeding disorders and swallowing problems (dysphagia).^[5] Dysphagia is associated with motor dysfunction in children with SP. Silent aspiration can be seen in CP cases. Aspiration is an important cause of developing acute and chronic lung diseases. Respiratory complications are important factors in increasing morbidity and mortality in CP.^[6]

Videofluoroscopic Swallow Study (VF) has been the most reliable method in the investigation of deglutition disorders.^[7] The primary method of imaging is barium passage radiographs. Ultrasound (US) can be used to assess more swallowing oral and pharyngeal phases.^[8] High spatial and temporal resolution, multiplanar evaluation capability, low cost, easily accessible, portability, repeatable, not requiring contrast material and ionizing radiation are some of the superior aspects of US.^[9] There are no definitive markers in the literature regarding the diagnostic value of US findings in assessment of dysphagia.

The examination by US has disadvantages compared with assessment by VF. The VF provides wide-field imaging with panoramic view, while US can perform low-range imaging. In US, transducer can compress tissues. It may be difficult to place and hold the transducer fixed. Therefore, the practitioner needs to be patient. Some structures in the study plan do not have anatomical markers. In our study, the transducer was placed in the midline of the neck without compression. The anatomical marker points we have identified have significant echo changes and they could easily be found in repeated studies.^[3,7,8]

In cases where the neuromotor mechanism such as CP is disrupted, the tongue, which is an important component of the swallowing process, should be evaluated. A decrease in the thickness of the muscle structures of the tongue may be indicative of inadequate stimulation. The degree of stiffness in the muscle layer can be evaluated by elastographic examination. In the studies aimed to elucidate the causes of dysphagia, elastography has not been used before.

Our purpose of the study is the dynamic evaluation of oral and pharyngeal phase changes of swallowing by US in healthy children and children with CP who developed swallowing disorder. We thought that tongue thickness and tongue elastographic values should differ due to insufficiency of excitations in neuromotor pathologies and we compared this data in the two groups.

MATERIAL AND METHOD

The children included in the study had no history of oral, cervical and thoracic operations, additional craniofacial anomaly and upper respiratory tract infection during the study. The patient group with CP was selected from orally fed children. Thus, it was assured that the swallowing mechanism was not affected for other reasons.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Parents of all participants were informed about the study protocol and their written consents were obtained. The child participants in the control group were informed face to face and what was asked of them was explained. After gaining the confidence of all participants, US examination was started using gel at room temperature.

Height and body weight of all participants were obtained. Body mass index (BMI) used to assist homogenization among the participants was calculated using the BMI formula ($BMI = \text{weight}/\text{height}^2$).

In the patient group, information was obtained about the difficulty of swallowing by talking with the parents of the child. These participants had no swallowing problems apart from solid food intake.

The US examinations were performed by a single pediatric radiologist with fifteen years of experience. Participants were comfortably seated on the examination table. None of the children were sedated. All of the children were convinced to talk for US exams. Parents stayed with their children throughout the examinations. Children with CP, who cannot sit, sat down leaning on their parents. We acted slowly and gently so that all children could adapt to the study. The examination time was an average of 5 minutes for healthy participants and 10 minutes for children with CP. When an effective examination could not be made, the examination was repeated with a pause in the required time.

All scanning was performed with real time scanner (Toshiba Aplio 500, Canon Medical System Corporation, Tokyo, Japan) employing a 5 MHz transducer. The measurement of the examination was recorded in hardware of scanner. To evaluate the oral phase of swallowing, the linguistic examinations were performed under the base of the mouth in the submental area. Sonographically, the mandibula, hyoid bone and thyroid cartilage, which can be identified as echogenic in the midline of the neck, were determined as the reference points. The cervical region was divided into two parts on anteriorly: Mandible to hyoid bone and hyoid bone to thyroid cartilage. Thus, laryngeal motion during swallowing was evaluated.

Transducer was positioned under the patient submental area in a longitudinal plane, floor of mouth, at the level of the mental protuberance of mandible and hyoid bone (**Figure 1**). Firstly, we measured the distances between mandible

to hyoid bone (MH) via US exam in resting period (MHrest) and swallowing its own spit (MHsw) (**Figure 2**). After the transducer was placed under the chin and reached the fixed position, it was instructed to swallow it a second time. Due to aspiration risk, the participants did not drink or eat anything during exams. Then, transducer was moved distally, region between hyoid bone and thyroid cartilage (HT). We measured the same length measurements again during rest (HTrest) and swallowing phases (HTsw). This data were recorded. The frame rate of the display, which shows the number of frames or images per second, was obtained during swallowing period and recorded for both regions. Subjects with CP and control group participants' tongue thicknesses on base of tongue in longitudinal image were made and recorded (**Figure 2**). Distance and thickness measurements were also made electronically in display. All length measurements were performed in millimeters. Then, at the rest stage in the longitudinal plan, tongue stiffness was determined using shear wave elastography (SWE) from glossal muscle structures near the tongue root. When calculating the SWE value, at least three measurements were made in the imaging area where the parallel lines were obtained as much as possible and their averages were recorded. The SWE data were performed in kiloPascal (kPa). The entire procedure was completed in 4 to 10 minutes. The procedure was repeated in cases where it was difficult to cooperate after the child was relieved by talking.

Statistical analyses were performed using the SPSS 22.0 for Windows (SPSS, Chicago, IL, USA). The Shapiro-Wilk test was used for examining the continuous variables, with normal and without normal distributions. An independent samples T-test was used for the between-group comparisons of the continuous variables with normal distributions. The data are expressed as the mean + standard deviations (SD). The Mann-Whitney U test was done for variables with a non-normal distribution. The data are expressed as median

and interquartile ranges. Categorical data was analysed by Pearson's chi-square test, and Fisher's exact test was done if the expected frequency was less than 5 in >20 % of all cells. Statistical significance was considered as $p < 0.05$.

RESULTS

There were 16 children with CP (9 boys and 7 girls) in the patient group and 20 healthy children (11 boys and 9 girls) in the control group. The mean age was 45 ± 21 months in the patient group and 60 ± 26 months in the control group. Body mass indexes (BMI) of the participants were calculated to ensure that children have similar body structure. BMI was 19 ± 5.9 kg/m² in the patient group and 18.2 ± 4.5 kg/m² in the control group (**Table 1**).

Table 1. The table showing measurements and intergroup relationships in both groups

	CP group	Control group	p
Age (months)	45.0± 21.0	60.0±26.0	-
BMI (kg/m ²)	19.0±5.9	18.2±4.5	p>0.05
TT in rest (mm)	14.0±3.7	20.4±3.7	p<0.02
MHsw (mm)	9.8±3.5	11.0±1.7	p>0.035
MHrest (mm)	14±2.8	16±2.2	p>0.065
HTsw (mm)	6.6±2.2	6.8±1.5	p>0.05
HTrest (mm)	10±3.1	11±1.1	p>0.405
TSWE in rest (kPa)	11.4±4.9	6.5±1.7	p<0.02
MH frame rates (image/sc)	13.5±3.6	12.2±2.2	p>0.179
HT frame rates (image/sc)	11.8±4.6	8.2±1.4	p<0.03

BMI: Body mass index, TT in rest: Tongue thickness in rest, MHsw: Distance between mandible to hyoid bone in swallowing, MH rest: Distance between mandible to hyoid bone in rest, HTsw: Distance between hyoid bone to thyroid cartilage in swallowing, HTrest: Distance between hyoid bone to thyroid cartilage in rest, TSWE in rest: Tongue Shear Wave Elastogram in rest, MH frame rates: Frame rate in mandible to hyoid bone level, HT frame rates: Frame rate in hyoid bone to thyroid cartilage level

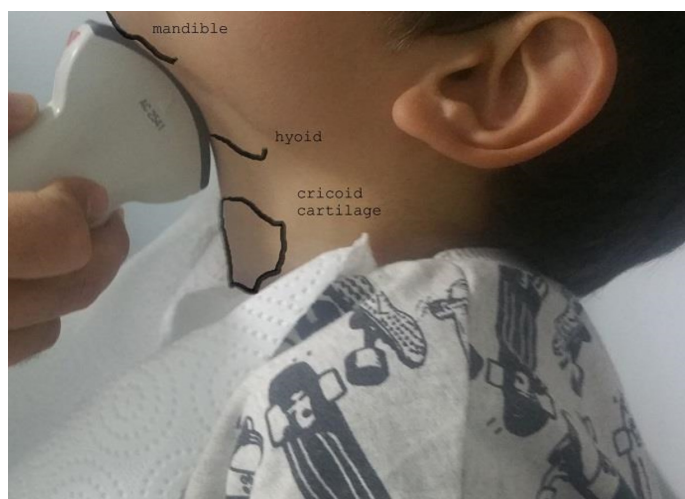


Figure 1. During the ultrasound (US) examination, the participant was in a sitting position. The anatomical landmark relationship with the transducer placed under the chin is shown.

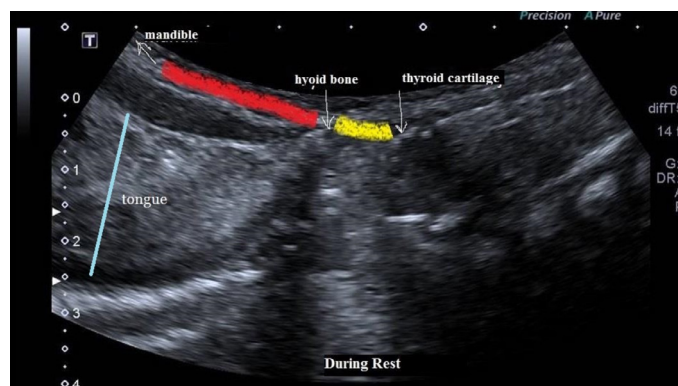


Figure 2. In healthy 7-year-old boy, US image shows distances between mandible and hyoid bone (red line), hyoid bone and thyroid cartilage (yellow line) during rest. In swallowing phase, the hyoid bone appears to move minimally upwards. Due to this, the distance between the hyoid bone and the mandible narrows. As the hyoid bone returns to its normal position, the distance between the hyoid and thyroid cartilage also narrows. Muscle thickness measurements taken from the area close to the root of the tongue were measured through longitudinal section (blue line). Anatomical control of the axial plane can be performed to confirm the measurement area (not shown).

In the CP group, the MH distance was 14 ± 2.8 mm and the HT distance was 10 ± 3.1 mm at rest. In the control group, these measurements were 16 ± 2.2 mm and 11 ± 1.1 mm, respectively. The MH and HT distance during swallowing were 9.8 ± 3.5 mm and 6.6 ± 2.2 in the CP group and 11 ± 1.7 mm and 6.8 ± 1.5 mm in the control group, respectively. At rest, tongue thickness at the root level was 14 ± 3.7 mm in the patient group and 20.4 ± 3.7 mm in the control group (**Figure 3**). In the same stage, the tongue muscle stiffness obtained with SWE near the tongue root was 11.4 ± 4.9 kPa in children with CP and 6.5 ± 1.7 kPa in the control group (**Figure 4**).

The screen frame rates throughout the swallowing action were calculated as 13.5 ± 3.6 images/second in MH and 11.8 ± 4.6 images/second in HT in children with CP. In the control group, these values were found to be 12.2 ± 2.2 images/second in MH and 8.2 ± 1.4 images/second in HT.

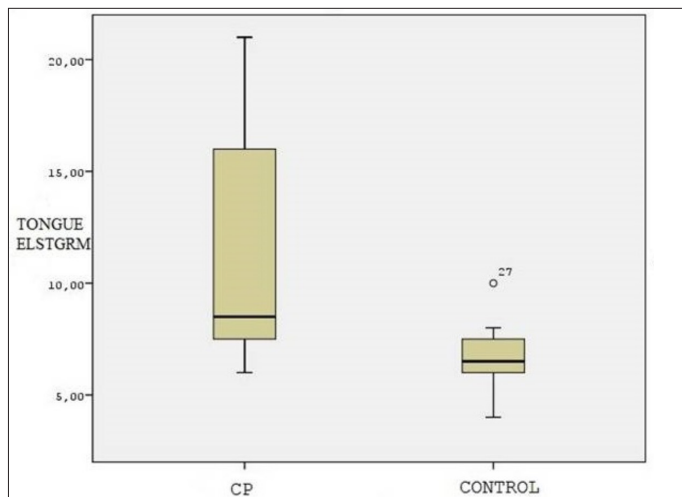


Figure 3. The graph shows that tongue thickness in children with CP is thinner than in the control group

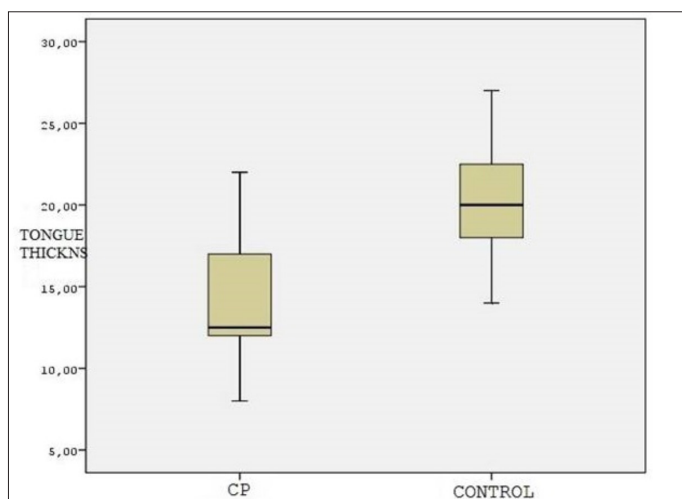


Figure 4. The graph shows that the tongue elastogram values during resting are higher in children with CP than in the control group. The graph shows that the tongue elastogram values during resting are higher in children with CP than in the control group

There was no significant difference between the CP and control groups in terms of MH and HT measurements at the resting and swallowing stages ($p > 0.005$). The significant differences were found between tongue thickness, elastogram and screen frame rates at HT level of the two groups (respectively $p < 0.002$, $p < 0.002$, $p < 0.003$) (**Table 1**). We calculated cut-off values of tongue elastogram, tongue thickness and frame rate at HT level for CP patients. The cut-off values, specificity and sensitivity ratios for these measurements are given in **Table 2**.

Table 2. In the table, diagnostic threshold values, sensitivity and specificity of screen frame rate in TT, TSWE and HT levels are given.

	Cut-off value	sensitivy	spesivity
TT	16.5 mm	75%	85%
TSWE at rest	7.5 kPa	75%	75%
HT frame rate	8.5 image/second	75%	70%

TT: Tongue thickness, TSWE: Tongue elastogram values, HT level: Hyoid bone to thyroid cartilage distances

In both groups, no statistically significant difference was found between all measurements and gender ($p > 0.082-0.904$ in children with CP and $p > 0.083-0.644$ in control group).

DISCUSSION

Children with CP have commonly feeding disorders and swallowing problems (dysphagia) which are at risk in many instances for aspiration with oral feeding, with potential pulmonary consequences.^[10] Pooled prevalence estimates determined by meta-analyses are as high as 50% for swallowing problems in patients with CP.^[11] Oropharyngeal dysphagia may be characterized by problems in any or all phases of swallowing.^[12] The types of oral and pharyngeal problems that children with CP have include reduced lip closure, poor tongue function, tongue thrust, exaggerated bite reflex, tactile hypersensitivity, delayed swallow initiation, reduced pharyngeal motility and drooling. Impaired oral sensorimotor function can result in drooling that in turn results in impaired hydration.^[11,13] Fluid-related problems are common and are often associated with a timing gap at the onset of delayed pharyngeal swallowing. Children with CP frequently need more time to complete feeding tasks, but caution is urged as fatigue and may become a factor, as well as reduced attention to the task.^[12] There is a risk of food aspiration and hypoxia during feeding. This risk in children with CP can decrease over time as developmental gains are made. The management of swallowing problems requires the investigation of the deglutition physiology by means of diagnostic imaging methods.^[14] Barium lumen radiographs can be used to evaluate the passage, to reveal lumen compression, and to examine the peristalsis of the esophagus. Still VF is considered the gold standard, a suitable method for diagnosing swallowing dysfunctions. In this study, we tried to analyze the changes in the oropharyngeal swallowing movement of the USA in children with CP, using a noninvasive and easily repeatable method.

In the study by Kenny et al.^[15] with a limited number of children with CP, tongue movement in the oral phase of swallowing was found to be insufficient in the posterior segment. In the same study, the movement of the hyoid bone during swallowing was found to be slow and inadequate. In another study, 20 children with spastic CP and 20 neurologically normal children were monitored by US imaging of the oral cavity during fluid and solid bolus tasks. In children with CP, the oral phase and total swallowing times for fluid bolus were longer than those of neurologically normal children. According to Casas et al.^[16], prolonged swallowing time was found as a result of pharyngeal motor impairment in children with CP. Yang et al.^[17] developed a descriptive scoring system for US observation of the oral stage of swallowing, as well as for items including tongue musculature bolus control, initiation, and coordination of tongue and hyoid movement. They significantly observed lower scores in 32 malnourished children with long-term neurological disability and severe feeding difficulties, when compared with 27 normal children. It indicated that US can detect impaired tongue movement in oropharyngeal dysphagia. In our study, instead of evaluating the movement of the tongue, muscle tone and muscle thickness of the tongue were examined. Tongue thickness decreased in children with CP with swallowing disorder compared to healthy children. SWE values of the tongue muscles were higher in children with CP. These findings may be related to an increase in overall muscle tone in CP group. According to another result we obtained, the longer frame rates at HT level in children with CP indicate longer swallowing time.

In a study to show the faringoglottal relationship in newborns, simultaneous pharyngoesophageal manometry, plethysmography, electromyography and glottal US were used.^[17] According to this study, glottal adduction during deglutition occurs in any respiratory phase, thus ensuring airway protection before and during deglutition. It has been reported that the laryngeal glottic closure reflex prevents aspiration during deglutition. It is reported that the investigation of the pharyngoglottal relationship by using noninvasive methods may be more acceptable for patients and is applicable to all ages. In current study, we evaluated age group children who could perform swallowing command. Therefore, no supportive examination method other than US was used.

The application of US in assessing the pharyngeal phase of swallowing is less common compared with that of assessing the oral phase. Previous studies have used US for observing lateral pharyngeal wall motion, thyroid-hyoid bone approximation, and hyoid bone displacement.^[9,19-21] Kuhl et al.^[9] and Huang et al.^[19] used US to measure hyoid bone-larynx approximation by using the result as a parameter to estimate larynx elevation. They found that the thyroid larynx approximation of the dysphagic stroke group was significantly less than that of the nondysphagic stroke group and the healthy group. They also reported similar measurement results between US and VF in 10 of the patients. Chi-Fishman.^[8] and Sonies.^[22] reported

that hyoid bone movement can be observed in all stages of swallowing with US. These studies were conducted with adult participants with swallowing dysfunction due to neurological reasons. In this study, we measured the distances between mandible and hyoid bone, hyoid bone and thyroid cartilage to evaluate larynx movement in children with CP. We concluded that there was no statistically significant difference in hyoid bone movement between healthy and children with CP. We found that the laryngeal elevation deficiency, which was found in adults with dysphagia, was not in children with CP.

In the study of Yabunaka et al.^[23] they used US to evaluate the movement of the hyoid bone in healthy adult patients and the changes caused by aging during deglutition. Their results were that as age increased, the average swallowing time increased and the movement of the hyoid bone decreased. In other words, as the age increased, the laryngeal phase of the swallowing deteriorated. In our study with a limited participant who could be considered as homogenous in the pediatric age group, we could not evaluate age-related changes. We did not find any gender related changes in healthy and CP children. In the presented study, the motion of the hyoid bone was interpreted with the same imaging method. According to our results, there was no significant difference between the two groups. Using parameters not included in Yabunaka's study, we showed an increase in swallowing time.

The muscle tonus is high in spastic CP group. Active tonus is normal or slightly decreased, primitive reflexes may be strong and persistent.^[5] According to the result we have reached, in children with CP with dysphagia, muscle tone varied at rest. The high tongue elastographic values during rest is consistent with this information in the literature.

Although we have not reached similar results in studies with adult groups, our common finding is that swallowing disorders can be identified by US. This method is portable and can be used at the bedside. It is cost effective, noninvasive method and has no radiation. Another advantage of US examination is dynamic method. Finally, other abnormalities in the oral stage, such as inadequate bolus control, premature oral leakage, impaired tongue propulsion, and multiple swallowing, can be observed using US. The major disadvantage of US is that it is operator dependent.

This study had some limitations. First, there was a lack of gold standard measurement for US results. Second, the total sample size is small. The number of patients in both groups was low. In order to confirm the results of our study, studies with larger patient populations in each age group are needed. Thirdly, we could not provide liquid or semi-solid food to prevent the risk of aspiration during swallowing in patients with CP. The examination was performed when the patient swallowed his/her own saliva in both groups. A certain amount of liquid, semi-solid and solid food intake and swallowing dynamics could not be evaluated. Lastly, insufficient swallowing sometimes caused a repeated examination, which led to prolonged US examination.

CONCLUSION

Oral and pharyngeal phases of swallowing function can be evaluated with US, which is a noninvasive method, in children with CP with swallowing problems. Duration of swallow, tongue thickness and tonus of tongue muscles can be used to evaluate dysphagia in patients with CP. We think that, pre- and post-treatment comparisons can be used to evaluate treatment effectiveness. US can be used as a quantitative method to clinically evaluate the oral and laryngeal phases of swallowing. Defining thyroid cartilage, tongue movement and muscle structure; the motion of the hyoid bone can be measured accurately and reproducibly with US. In addition, US can be used as a rapid examination tool to screen high-risk patients and monitor swallowing function..

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of Local Ethics Committee of Celal Bayar University medical School (permission granted: 28.03.2018, decision No: 20.478.486

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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