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Tongue thickness evaluation using ultrasonography can predict swallowing function in amyotrophic lateral sclerosis patients

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HIGHLIGHTS

- Tongue sonography is a useful tool for the quantitative evaluation of tongue atrophy in amyotrophic lateral sclerosis patients.
- Tongue thickness is associated with oral preparatory and transit time but not with pharyngeal transit time in amyotrophic lateral sclerosis patients.
- Amyotrophic lateral sclerosis patients show a progressive reduction in tongue thickness over time using ultrasonography.

ABSTRACT

Objective: Dysphagia is a critical issue in amyotrophic lateral sclerosis (ALS) patients. An evaluation of swallowing function is important for assessing the risk of aspiration. We investigated the validity of tongue sonography compared with videofluoroscopic examination for ALS patients.
Methods: We investigated 18 ALS patients. Nine subjects underwent repeated investigations. All of the subjects underwent tongue sonography and videofluoroscopic examination. Additionally, tongue sonography was evaluated in 18 age- and sex-matched healthy volunteers. To determine tongue thickness, we measured the vertical distance from the surface of the mylohyoid muscle to the tongue dorsum using ultrasonography.
Results: In the ALS patients, the tongue was significantly thinner than in healthy subjects. Tongue thickness was associated with body mass index and onset type in the ALS patients ($p = 0.006$). Temporal analyses of videofluoroscopy revealed that tongue thickness was associated with oral preparatory and transit time ($p = 0.032$) but not with pharyngeal transit time. Repeated measurement data revealed a decrease in tongue thickness over the course of the measurements ($p = 0.002$).
Conclusions: In ALS patients, reduced tongue thickness suggests disease progression and tongue dysfunction.
Significance: Tongue sonography is a useful modality for the non-invasive and quantitative evaluation of tongue thickness and dysphagia in ALS patients.

1. Introduction

Amyotrophic lateral sclerosis (ALS), one of the most severe neurodegenerative diseases, is characterized by progressive weakness, muscular atrophy, dysarthria, dysphagia and dyspnea. It is

Abbreviations: ALS, amyotrophic lateral sclerosis; VF, videofluoroscopic examination; %VC, % vital capacity; ALSFRS-R, ALS functional rating scale-revised; BMI, body mass index.

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important to prevent aspiration pneumonia and manage nutrition and rehabilitation to improve the prognosis and quality of life of ALS patients (Goyal and Mozaffar, 2014).

Dysphagia caused by bulbar dysfunction is a major factor that determines a patient's prognosis (del Aguila et al., 2003; Zoccolella et al., 2008). Evaluations of swallowing function are important for assessing the risk of aspiration and monitoring the progression of neurologic impairment. Such evaluations may help to determine feeding decisions, such as food texture, seating position and whether to introduce tube feeding, which can prevent aspiration pneumonia and improve the patient's nutritional state. Quantitative evaluation often requires specialized instruments. Among them, videofluoroscopic examination (VF) is one of the most reliable and standard methods for evaluating swallowing function. However, VF poses several problems for individuals with dysphagia because it requires extensive patient cooperation and carries the risk of aspiration. Aspiration pneumonia is associated with high mortality in ALS patients (Sorenson et al., 2007). It is difficult to perform VF in patients with severe ALS for whom obvious aspiration is predicted. Therefore, efforts have been made to establish an alternative and non-invasive method for evaluating dysphagia and the risk of aspiration.

Several simple and non-invasive methods have been developed for evaluating swallowing function (Yoshikawa et al., 2011; Tsuga et al., 2012; Weikamp et al., 2012). Tongue sonography may be an objective and non-invasive swallowing evaluation technique that carries no risk of aspiration. Because ALS results from neuronal death and causes muscle atrophy, including atrophy of the tongue muscle, tongue thickness may reflect the state of muscular atrophy and disease progression.

The aim of this study was to investigate the validity of tongue sonography as a tool for evaluating dysphagia and tongue condition in ALS patients. In particular, we investigated the concordance between VF and ultrasonography.

2. Methods

2.1. Ethics

The study protocols were approved by the ethics committee of each participating hospital and were performed according to the guidelines of the national government based on the Helsinki Declaration of 1964. Informed consent was obtained from all patients. All data analyses were blinded.

2.2. Subjects

We investigated 18 patients who were diagnosed with definite, probable or probable laboratory-supported ALS according to the revised El Escorial criteria (Brooks, 1994). Patient characteristics, including the time from onset (months), % vital capacity (%VC) and serum albumin (g/dl), were evaluated. We evaluated disease severity using the revised Amyotrophic Lateral Sclerosis Functional Rating Scale (ALSFRS-R) (Cedarbaum et al., 1999). The ALSFRS-R is divided into 5 domains, and we focused on the bulbar-related items (3 items: speech, salivation and swallowing). All of the subjects underwent tongue sonography and VF at the same time. Additionally, tongue sonography was evaluated in 18 age- and sex-matched healthy volunteers. Nine of the ALS patients underwent repeat tongue sonography two or three times within 15 months of the first measurement.

2.3. Physical examination

Three neurologists (MO, HM, YI) performed the neurological examinations. We performed systemic neurological evaluations that included muscle atrophy, tonus, deep tendon reflexes and pathological reflexes. Using visual examination, we independently evaluated the tongue atrophy and fasciculation of each subject. When two or three of the neurologists made a positive evaluation, the patients were diagnosed as with tongue atrophy or fasciculation, as appropriate.

2.4. Tongue sonography

The ultrasound examinations were performed by a neurosonologist (MN) using an SSA-770A imaging system (Toshiba, Tokyo, Japan). Tongue thickness was measured using a 3.5-MHz convex array transducer according to a previously reported method with some revisions (Tamura et al., 2012). The subjects were examined in a 30° leaning position while seated. The tongue thickness was determined by measuring the distance between the upper and lower surfaces of the lingual muscles in the center of the plane perpendicular to the Frankfurt horizontal plane of the frontal section (Fig. 1A). This perpendicular plane intersected the distal surfaces of the bilateral mandibular second premolars. The vertical distance was measured from the surface of the mylohyoid muscle to the tongue dorsum (Fig. 1B). This measurement was performed three times, and the mean value was defined as the tongue thickness for each subject. We confirmed the reliability of tongue ultrasonography by calculating the interday variability. We measured the tongue thickness of the normal subject three times per day. Their mean value was defined as the tongue thickness for the day. These measurements were repeated for ten days, and the resulting coefficient of variation was 1.59%. Furthermore, fasciculation of the tongue muscle was evaluated using a 7.5-MHz linear array transducer with the patient in the same position. The patient was observed for two minutes without moving the probe to detect fasciculations of the tongue muscle. While the patients were relaxed, we diagnosed any involuntary twitching of small parts of the tongue muscle as fasciculation and confirmed that the muscle fired irregularly, as reported previously (Misawa et al., 2011). The total tongue sonography examination took at most five minutes.

2.5. Videofluoroscopic examination (VF)

Each subject underwent VF using 3 g of yogurt mixed with the contrast agent Baricon meal (Horii Pharmaceutical Ind., Ltd., Osaka, Japan) after confirmation that no severe aspiration had occurred. The subjects were seated in a relaxed posture during the meal. The VF images were analyzed in slow motion and in single-frame mode using the playback capability of QuickTime software (QuickTime 7.3, Apple, Inc., Cupertino, CA). The oral preparatory and transit time and the pharyngeal transit time were measured to quantitatively evaluate each swallowing motion. Temporal analysis was performed according to a standardized method, as reported previously (Lazarus et al., 1996; Yoshikawa et al., 2005). Oral preparatory and transit time was defined as the time interval between the first posterior movement of the bolus, including multiple tongue pumps or mandible movement to initiate swallowing, until the head of the bolus reached the point where the lower edge of the mandible crossed the tongue base. Pharyngeal transit time was defined as the time elapsed from the moment when the head of the bolus reached the point where the lower edge of the mandible crossed the tongue base to the moment when the tail of the bolus passed through the cricopharyngeal region or the pharyngoesophageal segment. Quantitative observations were recorded by two dentists (AH, MY) with experience evaluating videofluorographic records; these two observers discussed their observations and reached a consensus for each observation or measurement.

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