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Original Article

Effect of daily swallowing frequency on swallowing-related muscle mass in individuals with severe motor and intellectual disability

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ABSTRACT

Background: Among the causes of dysphagia in individuals with severe motor and intellectual disability (SMID), addressing disuse atrophy of swallowing-related muscles are expected to improve patient outcomes. Disuse atrophy is associated with a reduced frequency of muscle activity, and swallowing frequency may be associated with the atrophy of swallowing-related muscles.
Objective: We aimed to investigate the relationship between swallowing frequency and swallowing-related muscle mass in individuals with SMID.
Methods: This cross-sectional study included 62 individuals with SMID (31 men and 31 women; mean age: 46.9 [standard deviation, 13.2] years) admitted to our center. Swallowing frequency was measured using a laryngeal microphone, and swallowing-related muscle mass was estimated by measuring the cross-sectional area of the geniohyoid muscle (GHM) using an ultrasonic diagnostic device.
Results: Increased swallowing frequency and body weight corresponded to a larger cross-sectional area of the GHM. Moreover, women had a smaller GHM cross-sectional area than men. Multiple regression analysis showed that the swallowing frequency, body weight, and sex were significantly and independently associated with the GHM cross-sectional area ($p < 0.01$).
Conclusions: Swallowing frequency and body weight significantly affect swallowing-related muscle mass in individuals with SMID.

1. Introduction

Children with severe disabilities experience various comorbidities that affect each pathological condition prognosis. Respiratory diseases are the leading cause of death in children with disabilities, with aspiration pneumonia accounting for the majority of such cases [1,2]. The prevalence of dysphagia in children with disabilities is approximately 90% and is thus a major issue that requires urgent attention [3].

Dysphagia in individuals with severe motor and intellectual disability (SMID) has been attributed to the dysfunction and delayed acquisition of swallowing ability owing to postnatal developmental disorders, and preventive measures are taken to avoid complications [4,5]. However, the increase in survival rates owing to recent advancements in medical care has contributed to the increased number of older individuals with severe illnesses. The deterioration of the swallowing function has emerged as a concern in individuals with developmental disorders [6], although research on functional decline is ongoing. Possible causes of deterioration in swallowing function include the progression of age-related changes and disuse [7]. Although it is challenging to prevent the progression of age-related changes, disuse may be prevented. Moreover, it is important to implement measures to prevent disuse and to maintain the swallowing function of individuals with SMID.

Disuse is a physical function disorder induced by decreased activity,

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which causes muscular atrophy and bone atrophy [8]. Maintaining muscle activity is essential to counter disuse atrophy; and frequent muscle activity leads to increased muscle protein synthesis [9,10]. Therefore, the frequency of muscle activity (i.e., the frequency of daily swallowing) may be associated with the disuse atrophy of swallowing-related muscles. In this study, we measured the frequency of daily swallowing and mass of swallowing-related muscles in individuals with SMID with the aim of understanding their relationship and to thus counter the disuse of swallowing function, which is an important concern in individuals with SMID.

2. Subjects

This was a cross-sectional study of Japanese individuals with SMID who were identified based on their electronic health records and admitted to the Nishinomiya-Sunago Medical and Welfare Center for Children with Severe Motor and Intellectual Disabilities between October 2019 and November 2020. The exclusion criteria were as follows: (1) age <20 years, (2) presence of a progressive disease, (3) lack of consent from the family, (4) an unstable general condition during the study period, and (5) participation in the study deemed to be difficult by the primary care physician. This study was approved by the Ethics Committee of our University (approval number: R1-E48). Written informed consent was obtained from all participants, their families, and the person responsible for the facility. This study was conducted in accordance with the 1964 Declaration of Helsinki and its amendments.

3. Methods

3.1. Measurement of swallowing frequency

The swallowing frequency was measured using a laryngeal microphone (SH-12jk; Nanzu, Shizuoka, Japan) and an integrated chip recorder (ICR) (ICD-PX240; Sony, Tokyo, Japan) according to the method reported by Tanaka et al. [11]. First, laryngeal sounds were recorded on an ICR via a laryngeal microphone attached to the participant's neck. The obtained voice data of the laryngeal sound were imported and displayed as a voice waveform using voice editing software (Cool Edit 96; Syntrillium Software, Phoenix, AZ, USA). Auditory judgment was made using the voice data, and visual judgment was made using the voice waveform. The sound produced during swallowing was identified, and subsequently, the swallowing frequency was measured (Fig. 1).

Conditions were established to record measurements based on the participants' daily lives; hence, activity restrictions were not applied. However, because eating is an activity that clearly affects swallowing frequency in daily life and because bathing makes it impossible for the participants to wear the test equipment, they were prohibited from eating, drinking, and bathing for 1 h before measurement and during the measurement period. The measurement time was 1 h, between 13:00 and 15:00, and the swallowing frequency was defined as the number of times a participant swallowed during the 1-h period. For each participant, three measurements were recorded, that is, one measurement daily over the course of 3 days. During the measurements, the tester patrolled the testing area regularly to check the participant's condition and record the participant's behavior and condition. In addition, if a participant required the measurements to be discontinued owing to poor physical condition (such as the presence of convulsions and fever), the measurements were repeated on another day.

3.2. Measurement of swallowing-related muscle mass

We measured geniohyoid muscle (GHM) mass because it significantly contributes to the prevention of aspiration [12] and because evaluation using ultrasonography is a well-established measurement method [13]. A single dentist, highly proficient in clinical work related to dysphagia, used a unit of an ultrasonic diagnostic device (ARIETTA 70; Hitachi Aloka Medical, Ltd., Tokyo, Japan) to measure the cross-sectional area of the GHM at rest, which is proportional to muscle strength, based on the method devised by Kajisa et al. [13].

The measurement site was set as the midpoint between the mandibular mentum and maximum convexity of the thyroid cartilage. A 6.0-MHz linear transducer was used for measurement and was placed perpendicular to the skin of the participant's jaw, applying minimal pressure to avoid muscle deformation (Fig. 2).

Each participant was seated on a seating-support chair that they used regularly and that was set to a posture with a 60° reclining angle to avoid excessive tension in the cervical muscles, including the GHM to be measured.

3.3. Factors associated with swallowing-related muscle mass

Age, sex, body weight, and arm muscle area (AMA), which is an indicator of whole-body skeletal muscle mass, are factors, other than swallowing frequency, that may be related to the mass of swallowing-related muscles [14–17]. In this study, analyses including the above four variables were conducted to evaluate factors related to GHM muscle mass.

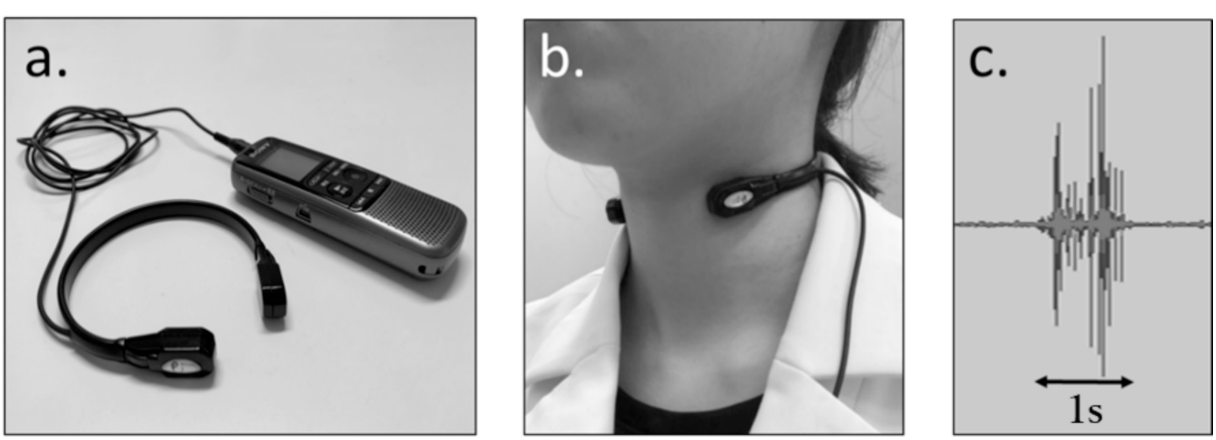

Fig. 1. Measurement of swallowing frequency. The number of swallows taken by participants with severe motor and intellectual disabilities was measured using a laryngeal microphone and integrated chip recorder (ICR). (a) Laryngeal microphone and ICR. (b) Representative image of a participant wearing the laryngeal microphone. (c) The sounds produced during swallowing were recorded on a personal computer.

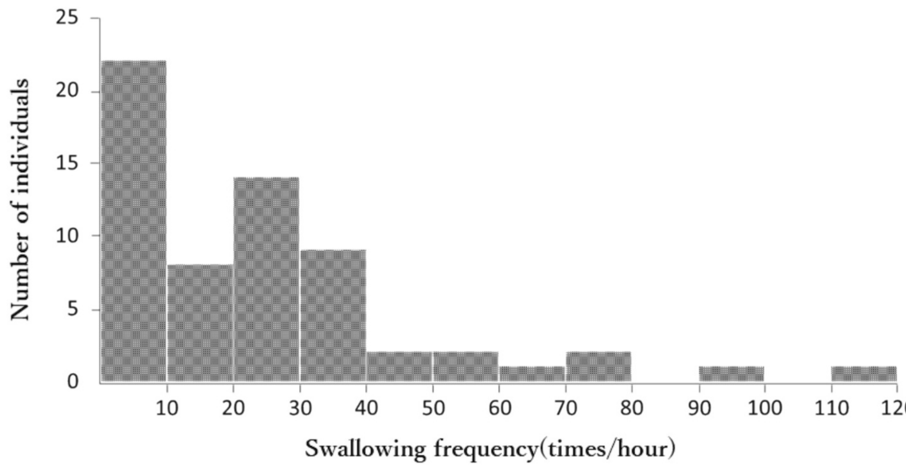

Fig. 2. Distribution of swallowing frequency. The frequency of swallowing varied considerably among individuals with severe motor and intellectual disability.

3.4. Statistical analysis

Multivariate analysis was conducted with the GHM cross-sectional area as the objective variable and swallowing frequency, age, sex, body weight, and AMA as explanatory variables. The method of statistical analysis was selected with the help of an expert, and multivariate analysis was conducted using the statistical analysis software SPSS Statistics version 22.0 (IBM Japan Ltd., Tokyo, Japan), with the statistical significance set at 5%.

4. Results

4.1. Summary

In total, 178 Japanese individuals with SMID were admitted to the Nishinomiya-Sunago Medical and Welfare Center for Children with Severe Motor and Intellectual Disabilities. After excluding those who met the exclusion criteria, 62 participants were included (men and women: 31 participants each, 20–69 years; mean age: 46.9 [standard deviation (SD): 13.2] years). None of the 62 participants dropped out over the course of the study.

The most common primary disease among the participants was cerebral palsy, affecting 56 (90%) participants, followed by congenital cerebral malformation in four (6%) participants, head trauma in three (5%) participants, cerebrovascular disorder in three (5%) participants, and chromosomal aberration in one (1%) participant. The most common medical complication was epilepsy ($n = 52$ [84%] participants).

4.2. Swallowing frequency in individuals with SMID

The average of the three measured swallowing frequencies was calculated, and the integer resulting from rounding off this average was considered as the swallowing frequency for each participant to determine the distribution of the swallowing frequency. The most frequent swallowing frequency per hour was 0–10 times, with a mean frequency of 22.9 (SD: 22.9), median frequency of 21, minimum frequency of 1, and maximum frequency of 111. Thus, there was significant variation in the swallowing frequency among participants (Fig. 3).

4.3. Factors affecting swallowing-related muscle mass

Multivariate analysis using the GHM cross-sectional area as the objective variable and swallowing frequency, age, sex, body weight, and AMA as explanatory variables revealed that the adjusted corrected multiple correlation coefficient was 0.60, and the adjusted corrected coefficient of determination was 0.36. The unadjusted corrected multiple correlation coefficient was 0.64, and the unadjusted corrected coefficient of determination was 0.41. Swallowing frequency, body weight, and sex were significantly correlated with the cross-sectional area of the GHM (Table 1). Swallowing frequency and body weight were positively correlated with the GHM cross-sectional area. Participants with lower swallowing frequency and body weight tended to have a smaller GHM cross-sectional area; women had a significantly smaller GHM cross-sectional area than men ($p < 0.01$). Swallowing frequency (standard partial regression coefficient = 0.34, t-value = 3.30) and body

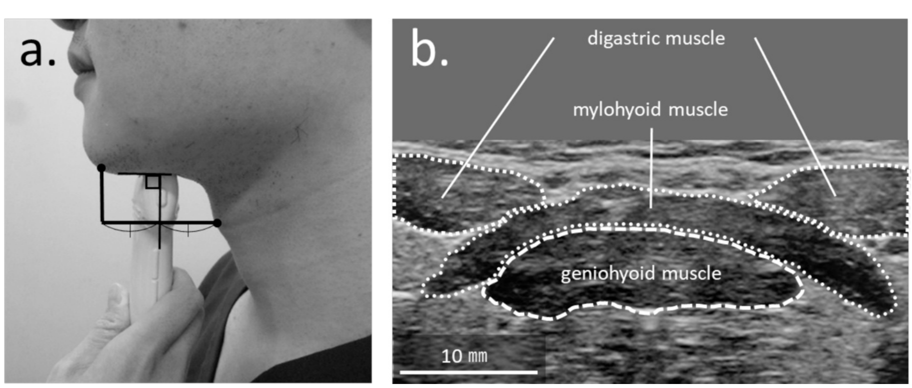

Fig. 3. Measurement of the geniohyoid muscle cross-sectional area. The cross-sectional area of the geniohyoid muscles in participants with severe motor and intellectual disabilities was measured using an ultrasonic diagnostic device. (a) Measurement position: A 6.0-MHz linear transducer was placed perpendicular to the submandibular skin at the midpoint between the participant's mandibular chin and maximum thyroid cartilage prominence. (b) Measurement of the cross-sectional area: The cross-sectional area of the geniohyoid muscle drawn in the deep layer of the coronal section was measured.

Table 1
Factors that may be associated with the mass of swallowing-related muscles.

R	Adjusted R	R^2	Adjusted R^2	
0.64	0.60	0.41	0.36	
Model	Unstandardized coefficients		Standardized coefficients	t	p-Value
	B	Standard Error	β		
Swallowing frequency	0.01	0.00	0.34	3.30	<0.01
Age	−0.01	0.00	−0.17	−1.62	0.11
Sex	0.25	0.09	0.30	2.85	<0.01
Body weight	0.02	0.01	0.34	3.11	<0.01
AMA	0.01	0.01	0.10	0.94	0.35

Data are presented as values with standard error. $P < 0.05$ was considered statistically significant.
Abbreviations: AMA, arm muscle area.

weight (standard partial regression coefficient = 0.34, t-value = 3.11) had the same degree of impact on the GHM cross-sectional area, followed by sex (standard partial regression coefficient = 0.30, t-value = 2.85). In contrast, age and AMA were not significantly correlated with the GHM cross-sectional area.

5. Discussion

5.1. Measurement of swallowing frequency

In this study, daily swallowing frequency was measured in participants at rest rather than while eating; the swallowing count is likely to vary depending on the meal content and is unlikely to be representative of the daily swallowing frequency [18]. Furthermore, meals increase the swallowing frequency; however, in healthy individuals, the number of swallows per meal is much lower than the daily number of swallows at rest [11]. Therefore, we measured the average resting swallowing frequency as representative of the daily frequency because it is unlikely that the count fluctuates: the resting frequency occupies a larger part of the day, and its total number is expected to be the higher.

5.2. Swallowing frequency in individuals with SMID

The mean swallowing frequency of individuals with SMID as measured in this study was 22.9 times/h (SD: 22.9). The mean swallowing frequency in healthy individuals measured by Tanaka et al. [19] using the same approach was 40.7 times/h (SD 19.5). The volume of saliva secreted [20,21], age, swallowing reflex threshold [19], and taste stimuli [20] may affect swallowing frequency. To the best of our knowledge, no studies have compared saliva volume and taste stimuli in healthy individuals and individuals with SMID, and their impact on swallowing frequency is unknown. In addition, the individuals with SMID analyzed in our study (mean age of 46.9 [SD 13.2]) were older than the healthy individuals (mean age of 26.5 [SD 3.5]) studied by Tanaka et al. [19]. Moreover, Arvedson et al. [22] reported that patients with cerebral palsy have delayed swallowing reflex initiation compared with healthy individuals, suggesting that individuals with SMID have a high swallowing reflex threshold. Although the impact of saliva volume and taste stimuli remains unknown, the results of our study indicate that differences in age and swallowing reflex threshold may have resulted in lower swallowing frequency of individuals with SMID in our study than of healthy individuals.

5.3. Factors affecting the mass of swallowing-related muscles

Age, sex, body weight, and AMA have all been reported as factors that affect the mass of swallowing-related muscles [14–16]. In this study, we evaluated the magnitude of the effect of swallowing frequency on the GHM cross-sectional area, including factors that affect the mass of swallowing-related muscles. Consequently, we found that swallowing frequency, body weight, and sex were associated with the GHM cross-sectional area. The factors that most significantly affected the muscle mass were swallowing frequency and body weight.

Yamaguchi et al. [14] and Baba et al. [15] also confirmed associations of sex and body weight with the GHM cross-sectional area, similar to the results of the present study. Therefore, sex and body weight should be considered factors affecting the mass of swallowing-related muscles, even in individuals with SMID.

Swallowing frequency, sex, and body weight were associated with GHM cross-sectional area; however, age and AMA were not. Regarding age, Baba et al. [15] reported contrasting results to our study and found a correlation between age and GHM cross-sectional area in healthy participants. This difference could be explained by the fact that Baba et al. [15] analyzed healthy individuals, whereas our study included individuals with SMID. Furthermore, Baba et al. [15] evaluated this association using a simple regression analysis. In the present study, considering that the swallowing frequency greatly affected the GHM cross-sectional area, the correlation between age and the GHM cross-sectional area may have been weakened when evaluated considering multiple factors, including the swallowing frequency, even in healthy participants.

We hypothesized that, when whole-body muscle mass decreases owing to factors such as malnutrition or chronic inflammation, the mass of swallowing-related muscles may also decrease. Furthermore, in a study on an older population, Tamura et al. [16] found a significant positive correlation between AMA, which is used as an indicator of whole-body muscle mass, and tongue muscle mass, which is a swallowing-related muscle. Therefore, in the current study, we added AMA as an explanatory variable and evaluated whether whole-body muscle mass affected the mass of swallowing-related muscles. Consequently, we did not find any association between AMA and the cross-sectional area of the GHM. These findings suggest that the decreased mass of a swallowing-related muscle in individuals with SMID cannot necessarily be linked to a loss of whole-body muscle mass and is affected more by swallowing frequency and body weight. The decrease in swallowing frequency and body weight causes disuse of swallowing-related muscles; it has been shown that preventing the decline in swallowing frequency may be effective in preventing disuse atrophy.

5.4. Limitations

Our results indicate that swallowing frequency tends to have a strong effect on the mass of swallowing-related muscles in individuals with SMID. Individuals with SMID included in this study had various diseases; hence, the effects of disease-derived characteristics on the mass of swallowing-related muscles were not evident from the results of this study alone. To consider preventive measures against the disuse of swallowing-related muscles due to the causative disease, it is necessary to evaluate the factors affecting the mass of the swallowing-related muscles for each disease. Although this was not a multicenter study and the results cannot be completely generalized, the primary disease in 90% participants was cerebral palsy; hence, a general tendency for cerebral palsy was noted. However, to investigate this aspect in detail, it will be necessary to increase the number of participants and perform an analysis according to the causative disease. Furthermore, if no significant changes in swallowing frequency or the mass of swallowing-related muscles are observed in the same subjects over the course of their development, it is possible that not only muscle disuse but also underdevelopment may be contributing factors. In addition to the results of this cross-sectional study, prospective longitudinal studies are needed to evaluate this possibility.

6. Conclusion

To explain the effect of daily swallowing frequency on the mass of swallowing-related muscles in individuals with SMID, we performed multiple regression analyses, including multiple factors that may be related to the mass of swallowing-related muscles. Consequently, swallowing frequency, body weight, and sex were associated with the cross-sectional area of the GHM, with swallowing frequency and body weight having particularly large impacts. In contrast, age and AMA were not associated with the cross-sectional area of the GHM.

The results of this study indicated that the cross-sectional area of the GHM in critically ill children was not related to age or whole-body muscle mass. This suggests that, when assessing the disuse of swallowing-related muscles, it is important to evaluate the swallowing-related muscles themselves rather than making judgments based on age or overall muscle mass loss. In contrast, the GHM cross-sectional area was affected by muscle mass, sex, body weight (a measure of nutrition), and swallowing frequency (a measure of muscle activity). In support of this, Feng et al. [11] demonstrated that the GHM cross-sectional area was smaller in patients with aspiration on swallowing endoscopy than in those without aspiration, suggesting that the maintenance of GHM mass may be important to prevent aspiration. Therefore, it was hypothesized that maintaining swallowing frequency in addition to maintaining body weight may lead to the prevention of GHM disuse and, ultimately, to the prevention of swallowing function deterioration.

Data statement

Data supporting the findings of this study are available from the corresponding author upon reasonable request.

CRediT authorship contribution statement

Chisato Uota: Conceptualization, Methodology, Formal analysis, Investigation, Writing – original draft. **Kanji Nohara:** Conceptualization, Writing – original draft. **Nobukazu Tanaka:** Conceptualization, Methodology. **Nami Fujii:** Formal analysis, Investigation. **Aya Obana:** Formal analysis, Investigation. **Katsuji Tanaka:** Methodology. **Takayoshi Sakai:** Supervision.

Informed consent

Written informed consent was obtained from all participants, their families, and the person responsible for the facility. Consent has been obtained from the individuals photographed in Figs. 1 and 3 for the inclusion of their photos in the paper.

Ethics approval

This study was approved by the Ethics Committee of the University of Osaka (approval number: R1-E48).

Declaration of generative AI and AI-assisted technologies in the writing process

For a portion of the paper, I used a proofreading service that employs AI to improve the readability of the English text.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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