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Original articles

Electrical activity of the masseter and suprahyoid muscles and aspects of swallowing in children and adolescents with osteogenesis imperfecta

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ABSTRACT

Purpose: to characterize the electrical activation of the masseter and suprahyoid muscles at rest and during swallowing tasks, to compare it with clinical aspects of swallowing.

Methods: a cross-sectional study, divided into mild osteogenesis imperfecta and moderate-to-severe osteogenesis imperfecta groups. Surface electromyography was performed on the masseter and suprahyoid muscles at rest and during swallowing tasks. The Orofacial Myofunctional Evaluation with Scores assessment form was used to assess clinical aspects of swallowing.

Results: moderate-to-severe osteogenesis imperfecta participants presented a higher percentage of masseter activation than mild osteogenesis imperfecta ones. Regarding the clinical aspects of swallowing, the total sample presented 40.9% normal lip occlusion or with slight effort; 59.1% demonstrated tongue protrusion and 50% showed two other signs of atypical function. Furthermore, the higher the score for lip activity during swallowing, the lower the activation of the suprahyoid muscles at rest.

Conclusions: the activation of the suprahyoid muscles while swallowing saliva and during consecutive swallows of liquid was similar, and activation during different tasks was higher in the moderate-to-severe osteogenesis imperfecta group. The better the labial myofunctional condition during swallowing, the lower the electrical activation of the suprahyoid muscles at rest.

Keywords: Osteogenesis Imperfecta; Stomatognathic System; Electromyography; Speech Therapy

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INTRODUCTION

Osteogenesis imperfecta (OI) is considered a rare genetic disease with an estimated prevalence of 1:10,000 to 1:20,000 births. It comprises a heterogeneous group of collagen disorders, with the most prominent clinical feature being the risk of fractures. However, several skeletal and extra-skeletal manifestations are common, including short stature, skeletal deformities, a bluish coloration of the sclera, hearing loss in young adults, and joint hypermobility[1,2].

These signs and symptoms comprehend those directly related to the stomatognathic system and its functions. Craniofacial abnormalities in maxillary and mandibular development, dental and occlusal changes, second molar retention, swallowing and sucking disorders, sleep-disordered breathing and temporomandibular disorders have been reported in recent studies[3-10]. However, little research has been published characterizing the stomatognathic muscle and functional issues in this population.

A thorough investigation of their orofacial biomechanics, such as the muscular strength of oral structures, is essential to understand the orofacial functional changes caused by OI, as well as the repercussions for the craniomandibular complex[11-13]. Surface electromyography (sEMG) is valuable technology that has the advantage of being non-invasive and easy to use. Techniques using sEMG can quantitatively assess the functioning of orofacial muscles and other structures[14-16]. This type of assessment can be added to clinical orofacial myofunctional examinations to better understand some of the biomechanical characteristics of a patient's stomatognathic system.

Thus, this study aimed at characterizing the electrical activities of the masseter and anterior suprahyoid muscles in children and adolescents with OI while swallowing and at rest. The secondary objective was to compare the electromyographic results with the clinical findings of the swallowing tests.

METHODS

This was a cross-sectional study approved by the research ethics committees of the Universidade Federal do Rio Grande do Sul (UFRGS) and the teaching Hospital de Clínicas de Porto Alegre (HCPA), Porto Alegre, Rio Grande do Sul, Brazil, report number 3.526.427, and followed resolutions 466/12 and 510/16 of the National Research Council. All the participants and their guardians signed informed consent forms.

Participants were recruited from the renowned special center reference center for Osteogenesis Imperfecta at HCPA in southern Brazil. They were included in the study if they had a medical diagnosis of OI and were between 6 and 19 years of age. Subjects were excluded if they had undergone speech therapy over the previous six months, or had a medical history of surgery, tumors or trauma in the head or neck.

Convenience sampling was based on the inclusion criteria. The sample size estimate, based on the number of registered patients, showed that seventy patients were eligible to participate in the study. Among these patients, 67.14% (47) were type 1 and 32.85% (23) were types 3, 4 and 5. The OI cases were grouped according to severity: the mild OI (MOI) group included participants with a clinical classification of OI type 1, while the moderate-to-severe (MSOI) group included participants with a clinical classification of OI types 3, 4 and 5[2].

To calculate the sample size, the estimated prevalence for both groups (MOI vs. MSOI), a 5% significance level (α), and a sampling error (d) of 0.16 were used. As this is a rare disease, a finite population correction factor was also used. There were 22 cases in the final minimum sample size: 15 MOI and 7 MSOI.

After routine consultation at the outpatient clinic, patients who agreed to participate were referred to the Faculty of Dentistry at UFRGS for orofacial myofunctional and sEMG assessments, with a focus on swallowing to meet the objectives of the study.

For the clinical evaluation, the OMES orofacial myofunctional assessment scoring form[17] was used. Its domains verify different aspects regarding the appearance and posture of structures, the mobility of orofacial structures and orofacial functions. For the purposes of this research, only the item concerning swallowing was analyzed.

Special items on the OMES form address swallowing, such as a specific assessment of lip and tongue activity, as well as other signs of changes in functional efficiency. During the entire swallowing evaluation, each patient remained seated with their feet flat on the floor, about 1 meter away from a video camera mounted on a tripod. The same evaluator, who had previous training and experience with the OMES assessment form, examined all the participants. Patients were offered 100mL of a thin liquid (water) in a glass, and a sandwich cookie (Bono®). Patients were given the following instructions: "Please drink the water as you normally do" and "Please eat the cookie as you normally do"

To record the EMG evaluations, we used Miotec® equipment, Miotool software, four analog input channels, an A/D converter with 14 bit-resolution to acquire EMG signals, electrical isolation to withstand 5000 volts, and a maximum acquisition capacity of 2,000 samples/s/channel. A 20Hz high pass filter and a 500Hz low pass filter were similarly used. The signals were captured and saved on a portable computer with no connections to electrical networks. The pairs of disk-shaped Kendall™ pediatric electrodes were spaced 20 mm apart, and came with a fixed amount of conductive gel provided by the manufacturer. These were glued to the bellies of the right and left masseter muscles, as well as the right and left anterior bellies of the temporal muscle. In order to avoid electromyographic interference, the reference electrode connected to the ground wire was also placed on the styloid process of the ulna and radius, on the participants' wrists. Before having the electrodes applied, the skin sites were cleaned with 70% alcohol to reduce impedance. Participants with beards were asked to remove them. Surface ElectroMyoGraphy for the Non-Invasive Assessment of Muscles (SENIAN) recommendations were followed[18].

The electromyographic signal for the masseter muscles was normalized using a maximum voluntary contraction (MVC), which is considered 100% electrical muscle activation. The Pernambuco et al. (2013)[19] method was used to capture the masseter muscle signals as follows:

a) We had the patient rest for at least ten seconds by saying "Stay still without moving or clenching your teeth";

b) We asked the patient to perform a maximum voluntary contraction with two 10 mm thick cotton rolls positioned between the premolars and molars, taking care not to touch the vestibular mucosa. They were asked to do so three times, for five seconds each try. There was a rest interval of 10 seconds between each contraction. The patients were previously trained to clench their teeth for the stipulated time. The verbal command to begin this test was "Squeeze your teeth together as hard as you can", and participants were encouraged with further verbal stimuli such as "Squeeze, squeeze, squeeze". The mean value was determined using 100% amplitude.

The electromyographic signals for the anterior suprahyoid muscles were similarly normalized using MVC, because of the 100% electrical muscle activation. The Pernambuco et al. (2013)[19] method was used to capture the signals from the anterior suprahyoid muscles engaged in an incomplete swallowing task, as follows:

a) We had the patient rest for at least ten seconds by saying "Stand still without moving any part of your face". Then, the participant was asked to perform an incomplete swallow of saliva. This was repeated three times, with ten-second intervals. Participants were trained prior to this test. The mean value was determined using 100% amplitude.

After performing MVC, participants were also asked to do the following tasks which were later assessed:

a) Rest for 10s: each participant was instructed to remain completely still for 10s. Additional guidance was given so that they did not move their tongues or swallow saliva;

b) Swallowing saliva: each patient was asked to swallow saliva three times. They received the following instruction: "Hold the saliva in your mouth and swallow when I say so". This task was repeated three times with ten-second intervals;

c) Swallowing a comfortable volume of liquid: swallowing 5mL of room temperature water in a single sip. Each participant was told: "Take this water into your mouth and hold it. Swallow it when I say so." This task was repeated three times with ten-second intervals;

d) Consecutive swallowing of liquid (100mL): each participant was asked to swallow 100mL of room-temperature water in a continuous manner, as they normally would.

All tests and interpretations were performed in the same environment and by the same evaluator who had been previously trained in clinical and research practice. To read and analyze the electromyographic signals of the consecutive swallowing task, the average (% MVC) of the entire interval of swallowing the 100 mL volume of liquid was considered. As for the single swallow task, with a comfortable volume of liquid (5mL), as well as the saliva swallowing task, the two-second interval that comprised the peak of the electromyographic signal of each of the three repetitions was analyzed, and the average of the three repetitions was used. The average (% MVC) of the recorded electrical activation with the normalized electromyographic signal was recorded[19]. All the values of the electromyographic tests were normalized and the percentage values were based on an MVC value of 100%. The EMG tasks were also simultaneously recorded with a video camera to help confirm the elevation times of the hyolaryngeal complex for analysis.

The data were statistically treated with the help of the SPSS® statistical program, version 20.0 for Windows®. A significance level of 5% was used for all tests. That is, the null hypothesis was rejected when the p-value was less than or equal to 0.05. Results were presented in absolute and relative distributions (n - %), as well as measures of central tendency (mean and median) and variability (standard deviation and amplitude), with a Shappiro-Wilk symmetry test. The Mann Whitney U test was used to compare continuous variables between two independent groups. Linearity relationships were estimated using Spearman's correlation coefficient.

The Wilcoxon test was performed to determine whether there was a statistical difference between the right and left masseter muscles, or the right and left anterior suprahyoid muscles during different tasks. Pairs that did not show a statistically significant difference were grouped on the right and left sides for the correlation between electrical activation and swallowing aspects. Only the right and left masseter muscles could not be grouped (Z= -1.997; p=0.046) for the consecutive swallowing task (100mL).

No age groups were established, so a simple linear regression model was applied. The model was calculated using the normalized values of the consecutive swallowing task (100mL) and the percentages were based on an MVC value of 100%. The arithmetic mean was calculated using the results of the right and left sides.

RESULTS

Table 1 presents the sample characterization for the total sample and groups. Table 2 shows the results for the total sample. They include the measures of central tendency and variability for the electrical activation in the anterior suprahyoid and masseter muscles during the tasks of swallowing saliva, single swallows of a controlled volume (5mL of liquid), consecutive swallowing (100mL of liquid) and 10s intervals of rest. Figure 1A demonstrates that the activation in the anterior suprahyoid muscles while swallowing saliva and performing consecutive swallows of liquid (100mL) swallowing is similar. Figure 1B demonstrates that the masseter muscles in the MSOI group presented a higher percentage of muscle activation than in the MOI group. Figure 2B demonstrates that the MSOI group showed a higher percentage of muscle activation than the MOI patients, except for the anterior suprahyoid muscle results at rest (2B) and the 5mL single swallows (2F).

Table 1. Sample characterization

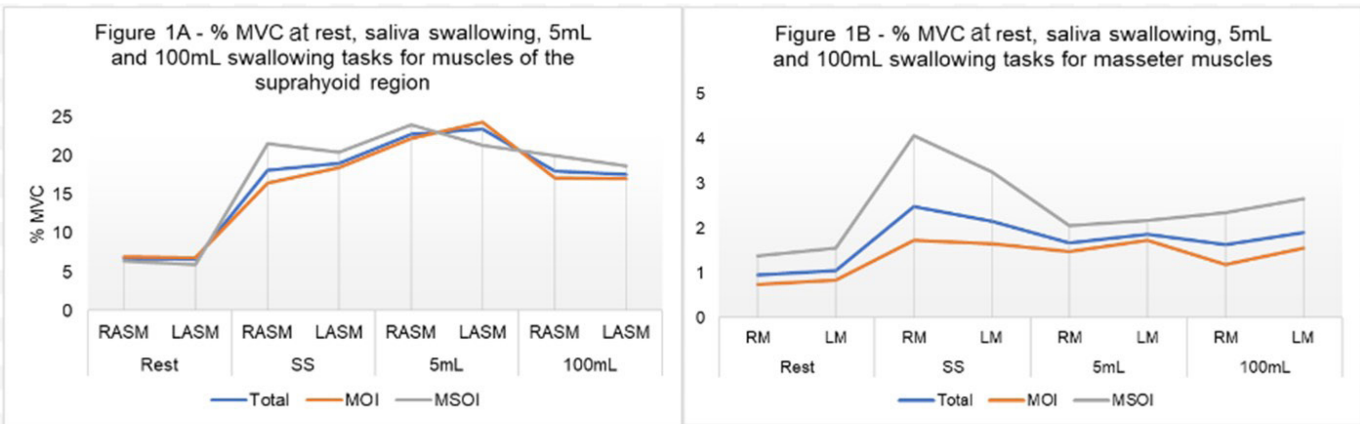
OI Groups	n	Mean age (SD)	Gender (%)
Total sample	22	12.09 (4.3)	Females 12 (54.6) Males 10 (45.4)
MOI	15	12.87 (3.6)	Females 6 (40) Males 9 (60)
MSOI	7	10.43(5.5)	Females 6 (85.7) Males 11 (4.3)

Captions: OI: osteogenesis imperfect; MOI: mild osteogenesis imperfecta; MSOI: moderate-to-severe osteogenesis imperfecta; SD: standard deviation.

Table 2. Measures of central tendency and variability of the electrical activity of the masseter and anterior suprahyoid region at rest (10s), while swallowing saliva and during single (5mL) and consecutive (100mL) swallows of water

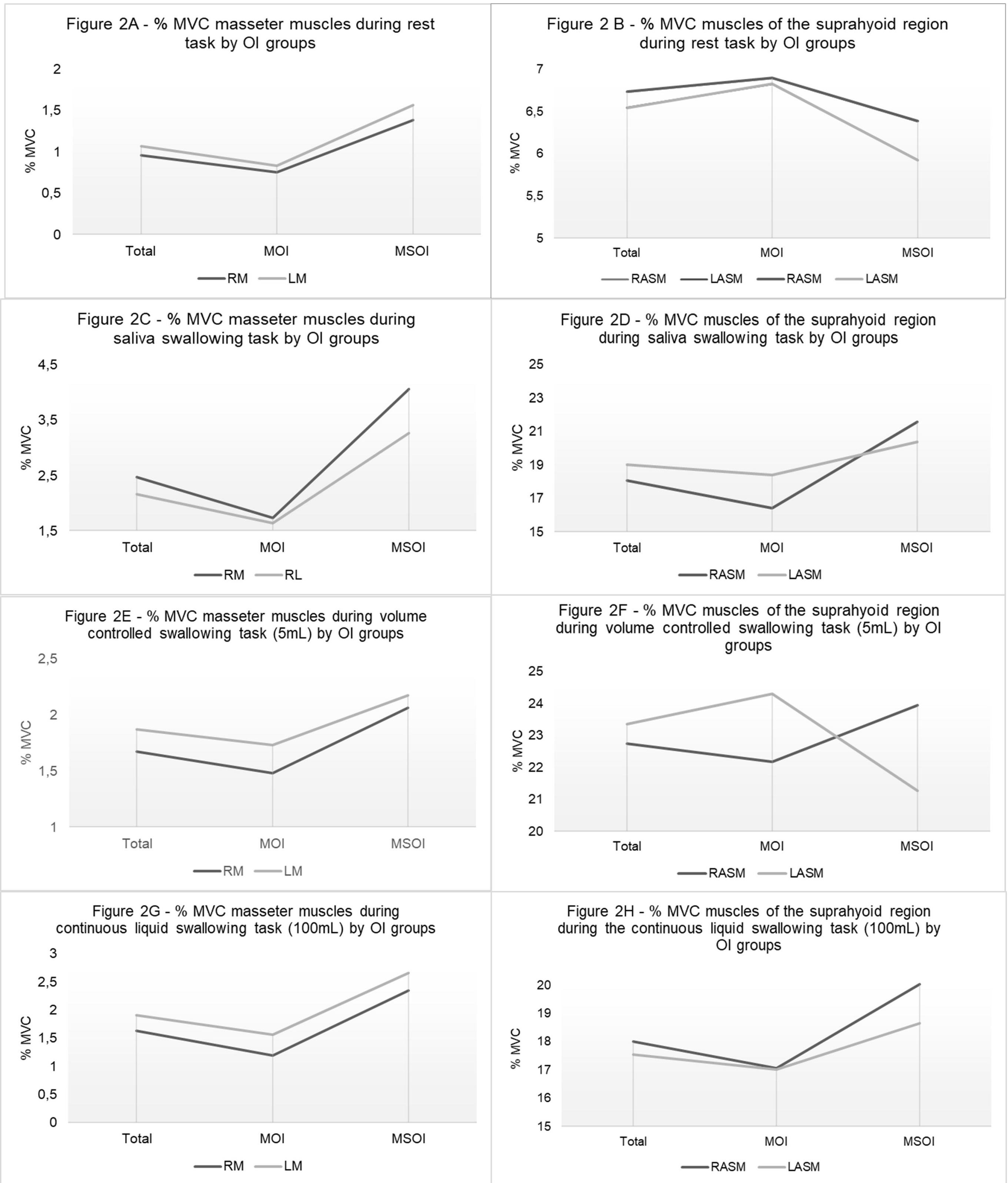
Variables A		Descriptive measures							
		n	Mean	SD	Amplitude		Quartile		
					Minimum	Maximum	1º	2º	3º
EMG at rest (% MVC)	RASM	22	6.73	4.62	1.88	16.90	3.02	4.96	10.48
	LASM	22	6.54	3.7	2.39	14.35	3.58	5.06	9.84
	RM	22	0.95	0.58	0.23	1.99	0.42	0.83	1.42
	LM	22	1.06	0.72	0.28	2.69	0.43	0.72	1.60
EMG swallowing saliva (% MVC)	RASM	22	18.04	7.05	2.66	32.65	13.49	19.66	22.24
	LASM	22	19.01	7.62	3.05	31.05	14.43	17.36	27.35
	RM	22	2.47	2.58	0.27	12.10	1.10	1.53	2.98
	LM	22	2.16	1.58	0.28	6.85	1.11	1.72	2.64
EMG single swallows 5mL (% MVC)	RASM	22	22.74	16.16	8.89	67.47	14.08	16.56	25.00
	LASM	22	23.34	12.93	10.44	55.57	16.00	18.55	26.50
	RM	22	1.67	1.00	0.52	3.91	0.96	1.34	2.35
	LM	22	1.87	1.26	0.53	4.63	0.91	1.46	2.50
EMG consecutive swallows 100mL (% MVC)	RASM	22	18.00	10.65	3.86	45.60	10.85	13.73	22.39
	LASM	22	17.53	9.10	2.55	38.76	11.37	16.07	22.55
	RM	22	1.63	1.07	0.40	4.76	0.81	1.23	2.16
	LM	22	1.90	1.16	0.46	4.27	1.03	1.54	2.87

Captions: A: asymmetrical distribution of variables (Shappiro Wilk; $p<0.05$); *MD: missing data: n=1(4.5%); SD: standard deviation; RASM: right anterior suprahyoid muscle; LASM: left anterior suprahyoid muscle; RM: right masseter muscle; LM: left masseter muscle; MVC: maximum voluntary contraction; mL: milliliters.



Captions: RASM: right anterior suprahyoid muscle; LASM: left anterior suprahyoid muscle; RM: right masseter muscle; LM: left masseter muscle; MVC: maximum voluntary contraction; MOI: mild osteogenesis imperfecta group; MSOI: moderate-to-severe osteogenesis imperfecta group; SS: saliva swallowing.

Figure 1. Percentage of maximum voluntary contraction in the tasks of rest for 10s, saliva swallowing, volume controlled liquid swallowing (5mL) and continuous liquid swallowing (100mL)



Captions: RASM: right anterior suprahyoid muscle; LASM: left anterior suprahyoid muscle; RM: right masseter muscle; LM: left masseter muscle; s: seconds; mL: milliliters; MVC: maximum voluntary contraction; MOI: mild osteogenesis imperfecta group; MSOI: moderate-to-severe osteogenesis imperfecta group.

Figure 2. Percentage of maximum voluntary contraction in tasks at rest for 10s, saliva swallowing, volume controlled liquid swallowing (5mL) and continuous liquid swallowing (100mL) in the total sample and by osteogenesis imperfect groups

Table 3 presents the measures of central tendency and variability for the scores regarding the clinical aspects of swallowing. With reference to the relative distribution of the clinical aspects of swallowing, in the total sample, 40.9% presented normal occlusion of the lips or occlusion with slight effort, while 18.2% demonstrated moderate effort. Regarding tongue activity while swallowing, 40.9% kept their tongues within the oral cavity in the correct swallowing position, while 59.1% showed adaptive or dysfunctional tongue protrusion. There were other types of behavior and signs indicative of atypical swallowing, such as head movements, facial muscle tension and leaking food or liquid. As for items with a below-average score on the assessment form, 4.5% had one item, 50% had two items and 45.5% had three items.

Regarding the relative distribution of the clinical aspects of swallowing, in the MOI group 46.7% presented normal lip occlusion, 40% occlusion with slight effort and 13.3% occlusion with moderate effort. With respect to tongue activity while swallowing, 46.7% kept their tongues within the oral cavity in the correct swallowing position, while 53.3% showed adaptive or dysfunctional tongue protrusion. There were other types of behaviors and signs indicative of atypical swallowing, and 60% had two items with below-average scores while 40% had three items.

Regarding the relative distribution of clinical aspects of swallowing, in the MSOI group, 28.6% presented normal lip occlusion, 42.9% occlusion with slight effort and 28.6% moderate effort. As for tongue activity while swallowing, 28.6% kept their tongues within the oral cavity in the correct swallowing position, while 71.4% presented adaptive or dysfunctional tongue protrusion. With respect to other types of behavior and signs of changed swallowing, 14.3% had one item with below-average scores, 28.6% had two and 57.1% had three.

Table 3. Measures of central tendency and variability for the scores of the clinical aspects of swallowing for the total sample and each osteogenesis imperfect group

Variables	Total (n=22)			OI Groups					
				MOI (n=15)			MSOI (n=7)		
	Mean	SD	Median	Mean	SD	Median	Mean	SD	Median
Swallowing: lip activity	3.2	0.8	3	3.3	0.7	3	3	0.8	3
Swallowing: tongue activity	2.4	0.5	2	2.5	0.5	2	2.3	0.5	2
Swallowing – other behavior and signs of atypical activity: head movements, tense facial muscles and leaking food and/or liquids	2.4	0.6	2	2.4	0.5	2	2.4	0.8	3
Total Swallowing Result	14	1.4	14	14.1	1.4	14	13.7	1.4	14
Number of swallows during consecutive swallows (100mL)	15.8	6	-	14.7	4.9	-	19.6	6.7	-
Consecutive swallowing time (100mL)	21.5	7.4	-	20.7	7.1	-	23.2	8.3	-

Captions: SD: standard deviation; OI: osteogenesis imperfect; MOI: mild osteogenesis imperfecta; MSOI: moderate-to-severe osteogenesis imperfect.

Table 4 compares the OI groups and the electrical activation results of the 10s rests, saliva swallowing, single swallows (5mL of liquid) and consecutive swallows (100mL of liquid). A statistically significant difference was noted between groups for the tasks of consecutive swallows (100mL of liquid), and 10s rests. The means of the MSOI group, for right and left masseters, were significantly higher than those of the MOI group.

Table 4. Electrical activity of the masseter and anterior suprahyoid muscles at rest (10s), while swallowing saliva and during single (5mL) and consecutive (100mL) swallows of liquid for the total sample and each osteogenesis imperfect group

Variables [A]	Total (n=22)					OI Groups										p¥
						MSOI (n=7)					MOI (n=15)					
	Mean	SD	Quartile 1st	Quartile 2nd	Quartile 3rd	Mean	SD	Quartile 1st	Quartile 2nd	Quartile 3rd	Mean	SD	Quartile 1st	Quartile 2nd	Quartile 3rd	
At rest (% MVC)																
RASM	6.73	4.62	3.01	4.96	10.48	6.89	4.11	2.79	5.34	11.86	6.38	4.53	3.64	4.57	7.71	0.972
LASM	6.54	3.69	3.58	5.06	9.83	6.82	3.64	4.06	5.39	10.31	5.92	4.02	3.31	4.73	6.63	0.647
RM	0.95	0.58	0.42	0.83	1.42	0.75	0.48	0.34	0.54	1.14	1.38	0.57	0.71	1.49	1.94	0.017
LM	1.06	0.74	0.43	0.71	1.61	0.83	0.60	0.39	0.50	1.35	1.56	0.81	0.78	1.68	2.41	0.012
Swallowing saliva (% MVC)																
RASM	18.04	7.05	13.49	19.66	22.24	16.41	7.58	10.39	17.39	21.54	21.54	4.35	17.72	22.12	25.40	0.053
LASM	19.01	7.63	14.43	17.36	27.35	18.38	8.08	14.17	16.39	27.35	20.37	6.93	14.52	18.33	27.50	0.751
RM	2.47	2.58	1.10	1.53	2.98	1.73	1.01	0.95	1.48	2.52	4.06	4.07	1.13	2.57	6.69	0.180
LM	2.16	1.58	1.11	1.72	2.64	1.64	0.84	0.91	1.47	2.41	3.26	2.23	1.26	2.76	5.20	0.053
Single swallows of liquid 5mL (% MVC)																
RASM	22.74	16.16	14.08	16.56	25.00	22.17	14.79	15.10	16.81	27.38	23.95	20.03	10.73	16.31	24.21	0.860
LASM	23.34	12.93	16.00	18.55	26.50	24.30	13.11	16.16	20.84	29.66	21.27	13.31	14.18	16.33	22.61	0.459
RM	1.67	1.00	0.96	1.34	2.35	1.48	0.93	0.75	1.16	2.00	2.06	1.10	1.14	1.43	3.17	0.113
LM	1.87	1.26	0.91	1.46	2.50	1.73	1.30	0.79	1.07	2.34	2.17	1.21	1.44	1.68	2.97	0.192
Consecutive swallows of liquid 100mL (% MVC)																
RASM	18.00	10.65	10.85	13.73	22.39	17.06	9.94	10.48	13.72	19.97	20.01	12.64	10.97	13.73	25.62	0.549
LASM	17.53	9.11	11.37	16.07	22.55	17.01	8.99	10.98	15.39	24.09	18.64	9.99	11.50	18.47	21.59	0.972
RMM	1.63	1.07	0.81	1.23	2.16	1.19	0.77	0.73	1.11	1.76	2.34	1.32	1.17	2.06	3.29	0.026
LMM	1.90	1.16	1.03	1.54	2.87	1.56	0.96	0.71	1.17	2.21	2.65	1.26	1.28	2.80	4.05	0.038

Captions: ¥: Mann Whitney U Test comparing MOI x MSOI; A: asymmetrical distribution of variables (Shappiro Wilk; $p<0.05$; SD: standard deviation; RASM: right anterior suprahyoid muscle; LASM: left anterior suprahyoid muscle; RMM: right masseter muscle; LMM: left masseter muscle; s: seconds; mL: milliliters; MVC: maximum voluntary contraction; OI: osteogenesis imperfect; MOI: mild osteogenesis imperfecta; MSOI: moderate-to-severe osteogenesis imperfect.

Table 5 demonstrates the correlations between electrical activation during different tasks. In the evaluation of electrical activation during all tasks, a significant positive correlation was identified between the muscle pairs of the anterior suprahyoid region and the masseter muscles, indicating that high muscle activation on the right side correlated with high muscle activation on the left side of the muscle pair. Furthermore, this behavior was also observed between the anterior suprahyoid muscles and the left masseters. During the volume-controlled liquid swallowing task (5mL), there was a significant positive correlation in comparisons between the muscle pairs, as well as between the anterior suprahyoid and masseter muscles.

Table 5. Analysis of the correlation between the electrical activity of different tasks

Task Comparisons			Spearman´s Correlation r	p
At rest (% MVC)				
RASM	X	LASM	0.841	0.000
RASM	X	RM	0.361	0.098
RASM	X	LM	0.606	0.003
LASM	X	LM	0.477	0.025
LASM	X	RM	0.356	0.104
RMM	X	LM	0.805	0.000
Swallowing saliva (% MVC)				
RASM	X	LASM	0.621	0.002
RASM	X	RM	0.116	0.608
RASM	X	LM	0.422	0.051
LASM	X	LM	0.478	0.025
LASM	X	RM	0.267	0.230
RMM	X	LM	0.862	0.000
Single swallows of liquid 5mL (% MVC)				
RASM	X	LASM	0.781	0.000
RASM	X	RM	0.510	0.015
RASM	X	LM	0.477	0.025
LASM	X	LM	0.531	0.011
LASM	X	RM	0.510	0.015
RMM	X	LM	0.863	0.000
Consecutive swallows of liquid 100mL (% MVC)				
RASM	X	LASM	0.740	0.000
RASM	X	RM	0.092	0.684
RASM	X	LM	0.119	0.597
LASM	X	LM	0.477	0.025
LASM	X	RM	0.002	0.994
RMM	X	LM	0.913	0.000

Correlation classification – Very weak correlation |0.000| to |0.199|; weak correlation |0.200| to |0.399|; moderate correlation |0.400| to |0.699|; strong correlation |0.700| to |0.899|; very strong correlation |0.900| to |1.00|(A).
Captions: EA: electrical activity; RASM: right anterior suprahyoid muscle; LASM: left anterior suprahyoid muscle; RMM: right masseter muscle; mL: milliliters; MVC: maximum voluntary contraction.

Table 6 presents the correlated analysis between electrical activation and swallowing aspects. It was noted that the longer the time - and also the greater the number of consecutive swallows (100mL) - the greater the electrical activation in the masseter muscles during all evaluated tasks. In addition, the higher the score for lip activity while swallowing, the lower the electrical activation in the anterior suprahyoid muscles at rest.

Table 6. Analysis of the correlation between the electrical activity presented during tasks and aspects of the swallowing assessment

Comparisons		Spearman's Correlation							
		r	p	r	p	r	p	r	p
		At Rest		Swallowing Saliva		Single swallows of liquid 5mL		Consecutive swallows of liquid 100mL	
EA RLM (% MVC)	OMES – total swallowing	-0.147	0.514	-0.97	0.668	-0.196	0.381	-	-
	OMES – lip activity while swallowing	-0.143	0.527	-0.222	0.320	-0.163	0.468	-	-
	OMES – tongue activity while swallowing	-0.386	0.076	-0.051	0.822	-0.357	0.103	-	-
	OMES – other signs while swallowing	0.155	0.490	0.075	0.740	0.018	0.938	-	-
	Swallowing time 100mL	0.618	0.002	0.510	0.015	0.664	0.001	-	-
	Nº of swalllows 100mL	0.568	0.006	0.437	0.042	0.638	0.001	-	-
EA RLASM (% MVC)	Total OMES swallowing results	-0.146	0.516	-0.041	0.858	0.067	0.766	0.088	0.696
	OMES – total swallowing	0.070	0.757	-0.121	0.591	0.077	0.732	0.080	0.722
	OMES - lip activity while swallowing	-0.474	0.026	-0.051	0.822	-0.168	0.456	-0.197	0.380
	OMES - tongue activity while swallowing	-0.020	0.929	0.110	0.625	0.110	0.625	0.238	0.286
	Swallowing time 100mL	0.272	0.221	0.179	0.425	0.358	0.102	0.130	0.563
	Nº of swalllows 100mL	0.334	0.128	0.391	0.072	0.403	0.063	0.151	0.502
EA RM (% MVC)	OMES – total swallowing	-	-	-	-	-	-	-0.098	0.664
	OMES - lip activity while swallowing	-	-	-	-	-	-	-0.155	0.490
	OMES - tongue activity while swallowing	-	-	-	-	-	-	-0.240	0.281
	OMES - other signs while swallowing	-	-	-	-	-	-	0.160	0.476
	Swallowing time 100mL	-	-	-	-	-	-	0.561	0.007
	Nº of swalllows 100mL	-	-	-	-	-	-	0.421	0.051
EA LM (% MVC)	OMES – total swallowing	-	-	-	-	-	-	-0.123	0.585
	OMES - lip activity while swallowing	-	-	-	-	-	-	-0.174	0.438
	OMES - tongue activity while swallowing	-	-	-	-	-	-	-0.226	0.312
	OMES - other signs while swallowing	-	-	-	-	-	-	0.110	0.625
	Swallowing time 100mL	-	-	-	-	-	-	0.609	0.003
	Nº of swalllows 100mL	-	-	-	-	-	-	0.499	0.018

Correlation classification – Very weak correlation |0.000| to |0.199|; weak correlation|0.200| a |0.399|, moderate correlation |0.400| to |0.699|; strong correlation |0.700| to|0.899|; very strong correlation |0.900| to |1.00|(A).
Captions: EA: electrical activity; RLM: right and left masseter muscles; RLASM: right and left anterior suprahyoid muscles; RM: right masseter; LM: left masseter; mL: milliliters; MVC: maximum voluntary contraction.

The linear regression model indicated coefficient of determination (R Squared) was 0.003 for the anterior suprahyoid muscles. This indicates that only 0.3% of the results for electrical activation in anterior suprahyoid muscles can be explained by age. For the masseter muscle, the coefficient was 0.314. This indicates that 31.4% of the results for electrical activation in the masseter muscle can be explained by age.

DISCUSSION

This study characterized the electrical activity of the masseter and anterior suprahyoid muscles in children and adolescents with OI during swallowing and at rest and also compared the electromyographic results with the clinical findings of swallowing tests. The quantitative assessment of muscles and orofacial structures allows the understanding of some biomechanical characteristics of the stomatognathic system of the population with OI and helps to understand the oromyofunctional repercussions of the disease and also to provide better intervention.

In this sample, the activation of the anterior suprahyoid muscles while swallowing saliva and performing consecutive swallows of liquid (100mL) was similar. The literature, however, points out that the intensity of the activity of the muscles involved in swallowing is influenced by factors such as volume, viscosity and flavor[20,21]. One three-dimensional study using an electromagnetic articulator to evaluate the swallowing of water and saliva also observed that the movements of certain portions of the tongue were influenced by the volume and the type of element to be swallowed, and that the amplitude of the movement was directly proportional to the volume of water swallowed[22].

Though it is possible that the swallowing of saliva was the most changed swallowing function captured by electromyography, these distorted results may be due to interference from the command to swallow[23]. Swallowing saliva occurs spontaneously as a result of its accumulation in the mouth. Spontaneous swallowing happens unconsciously during wakefulness and also during sleep, and is a type of protective reflex action. Voluntary swallowing is part of eating behavior, and results from the harmonized and orderly activation of the perioral, lingual and submental striated muscles in the oral phase. This process is largely bypassed during spontaneous swallowing, although partial excitation may occur. Physiologically, the main difference between voluntary and spontaneous swallowing is the origin of the swallowing trigger[24]. The switch from spontaneous to voluntary function in a population with craniofacial abnormalities could justify the greater activity in the anterior suprahyoid muscles, as an attempt to organize the swallowing movement with saliva.

Regarding the number of swallows and the time for swallowing 100mL of water, in the literature we consulted there were no studies concerning children and adolescents that could be reliably compared to the present study, not even for references about the clinical parameters in this population. However, the literature does report a swallowing speed of 10mL/s as a reference parameter for the adult population. Swallowing speeds of <10mL/s would indicate an abnormal swallowing index. To calculate swallowing speed (mL/s) however, one must use 150mL of water as the base intake[25]. A study carried out with a sample of participants aged between 8 and 80.11 years in an Indian population found normative values of swallowing speeds above 10mL/s, in all age groups[26]. In our study, we were not able to calculate the swallowing speed, as the amount of water ingested (in mL) was different. However, it drew our attention that the time taken to ingest 100mL seemed to be faster. If we calculated the speed based on the 100mL ingested, we would find values of 4.65mL/s, 4.83mL/s and 4.31mL/s for the total sample, in the MOI group and MSOI group, respectively. The literature also reports that drug-induced esophageal injury may occur as a result of the oral intake of bisphosphonates. This is especially true for elderly patients who present generally reduced conditions, motility disorders or anatomical changes in the esophagus. Among the main symptoms are globus sensation, sensation of a foreign body and odynophagia. Patients also ingest more fluids when swallowing solid consistencies[27]. In our study, dysphagia-related symptoms were not collected. It is recognized that, due to bone changes, many patients with moderate-to-severe OI use a wheelchair and undergo drug therapy with bisphosphonates.

Muscle activation during the different tasks was higher in the MSOI group. The start of a swallow involves the pressing of the tongue against the hard palate while simultaneously contracting the submental muscles. Contracting jaw elevator muscles and holding the teeth closed for approximately 1 second stabilizes the mandible which, in turn, helps the suprahyoid muscles lift the tongue against the palate and propel the bolus from the oral cavity to the oral level of the pharynx[20]. OI types III and IV present greater skeletal deformities than type I[2] and, as mandibular stabilization in the skull requires the isometric contraction of the mandibular elevator muscles[28], greater activation in them can be justified by the attempt to maintain the synergistic balance of the mandible[29].

In the correlations between electrical muscle responses during all tasks, high muscle activation on the right side was correlated with high muscle activation on the left side for both muscle pairs. A study with high-density surface electromyography that assessed normal swallowing functions found a pattern presented by submental and infrahyoid muscle complexes, with symmetrical muscle activation on the left and right sides[30]. Another study that also used high-density electromyography on healthy adult participants observed that the facial muscles showed almost symmetrical patterns of muscle activation during tasks, and also at rest[31]. However, even in healthy participants, a certain degree of muscle asymmetry can be found and is considered physiologically compatible with adequate function[32,33].

Considering the correlation between electrical responses and swallowing aspects, it was noted that the longer the time and the greater the number of swallows for 100mL of liquid, the greater the electrical activation of the masseter muscle, which plays an important role in mandibular stabilization in the oral phase of swallowing[28,29]. A study with healthy individuals without TMD or swallowing disorders confirmed that, during swallowing, the masseter and anterior temporal muscles were activated at the same time as the submental and sternocleidomastoid muscles. The increase in electrical potential while swallowing followed the mandibular stabilization provided by the isometric contraction of the recorded muscles. Furthermore, the study found that the activity of the masseter and anterior temporal muscles in the group that swallowed with occlusal contact was significantly higher than the muscle activity of the group that presented no occlusal contact while swallowing[28]. Thus, the participation of this musculature in swallowing tasks that require more mandibular stabilization can be justified.

Also, the higher the score for lip activity while swallowing, the lower the electrical activation of the anterior suprahyoid muscles at rest. During normal swallowing, the lips must be closed with no excessive contraction. The jaw elevator muscles do contract to help stabilize the jaw and elevate the tongue against the palate[34]. Thus, it was observed that participants with better myofunctional competence in the lips presented lower electrical activation in the anterior suprahyoid muscles at rest; a finding that suggested their tongues were in their proper resting position – which must be contained by the dental arches, with the anterior portion elevated and touching palatal rugae[34]. This is when muscle activity is expected to decrease.

According to the Orofacial Myofunctional Evaluation with Scores (OMES)[17], the higher the score, the better the performance. Regarding the clinical aspects of swallowing in our study, the patients' scores were similar to those of the healthy young population. In a study[35] that included participants aged 18 to 40 years, the partial score for swallowing function resulted in a mean of 14.81 ± 1.12 for men and 15.17 ± 0.80 for women. Although the study separated the analysis by sex, the scores did not differ significantly between them, suggesting that individuals can be considered as a single group for research purposes[35].

Performing different assessments on significant samples of populations with rare diseases is a particularly difficult task. Ours is the first study we are aware to perform quantitative assessments of the swallowing in children and adolescents with OI. The absence of a control group and the sample size were limitations to our research. Further investigations with a larger sample and a control group are suggested.

CONCLUSIONS

In this sample, the activation of the anterior suprahyoid muscles while swallowing saliva and during consecutive swallows of liquid (100mL) was similar. Muscle activation during different tasks was higher in the MSOI group. High muscle activation on the right side was correlated with high muscle activation on the left side for both muscle pairs. Regarding the clinical aspects of swallowing function, the scores were close to those of the healthy young population. The comparison of the electromyographic and clinical swallowing results showed that the longer the time and the greater the number of swallows to drink 100mL of liquid, the greater the electrical activation of the masseter muscle. Moreover, the higher the score for lip activity while swallowing, the lower the electrical activation of the anterior suprahyoid muscles at rest. Further specific studies regarding the signs and symptoms of swallowing among this population are recommended.

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Author contribution:

ACCO: Conceptualization; Data curation; Methodology; Project administration; Investigation; Writing - Original draft; Writing - Review & editing.

EG: Conceptualization; Project administration; Writing - Review & editing.

TMF: Resources; Project administration, Writing - Review & editing.

HJS: Writing - Review & editing.

Data sharing statement:

Regarding the data from the research "Biomechanical characterization of the orofacial musculature in osteogenesis imperfecta" the authors declare that no research data will be shared, as the research consent from assured participants that "The results will be presented jointly".