

# Tongue pressure in children and adolescents with osteogenesis imperfecta

## Pressão de língua em crianças e adolescentes com osteogênese imperfeita

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### ABSTRACT

**Purpose:** To investigate and correlate the lingual pressure of the anterior and posterior region in isometric contraction and during saliva swallowing of individuals with Osteogenesis Imperfecta (OI). **Methods:** This was an observational cross-sectional study, 22 subjects participated, with an average age of 12.09 years, divided into mild OI (OIL) (type 1) (n=15) and moderate/severe OI (OIMG) (types 3, 4 and 5) (n=7). The Iowa Oral Pressure Instrument (IOPI) was used and tongue isometry pressure was measured in the anterior region, posterior region and during swallowing. Statistical analyses was done with the SPSS program using the Mann Whitney test, Spearman correlation and simple linear regression model. The significance level of  $p < 0.05$  was used. **Results:** Pressure in tongue isometry was greater in the anterior region than in the posterior region in the total sample and in the OIMG group. The pressure of the anterior region during saliva swallowing was higher in the OIL group. Greater tongue pressure was also observed in the posterior region in the OIL group. **Conclusion:** Greater pressure in the anterior region of the tongue is correlated with greater pressure in the posterior region of the tongue, but there is no significant correlation between the isometric tongue capacity and the tongue pressure during the saliva swallowing function.

**Keywords:** Speech-language pathology; Stomatognathic system; Tongue; Osteogenesis imperfecta; Child; Adolescent

### RESUMO

**Objetivo:** Investigar e correlacionar a pressão lingual da região anterior e posterior em contração isométrica e durante deglutição de saliva de indivíduos com osteogênese imperfeita. **Métodos:** Estudo transversal observacional, do qual participaram 22 sujeitos, com média de idade de 12,09 anos, divididos em osteogênese imperfeita leve (tipo 1) (n=15) e osteogênese imperfeita moderada-grave (tipos 3, 4 e 5) (n=7). O *Iowa Oral Pressure Instrument* foi utilizado e foi aferida a pressão em isometria de língua da região anterior, da região posterior e durante deglutição. As análises estatísticas foram realizadas no programa SPSS, por meio dos testes Mann Whitney, correlação de Spearman e modelo de regressão linear simples. O nível de significância de  $p < 0,05$  foi utilizado. **Resultados:** A pressão em isometria de língua foi maior na região anterior do que na região posterior na amostra total e no grupo com osteogênese imperfeita moderada-grave. A pressão da região anterior durante deglutição de saliva foi maior no grupo com osteogênese imperfeita leve. Também foi observada maior pressão de língua da região posterior nesse grupo. **Conclusão:** Maior pressão da região anterior de língua correlaciona-se à maior pressão da região posterior de língua, porém, não há correlação significativa entre a capacidade isométrica de língua e a pressão de língua durante a função de deglutição de saliva.

**Palavras-chave:** Fonoaudiologia; Sistema estomatognático; Língua; Osteogênese imperfeita; Criança; Adolescente

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## INTRODUCTION

Osteogenesis imperfecta (OI) is a rare genetic disease with a prevalence of 1:15,000 to 20,000 individuals, affecting connective tissue synthesis, with wide phenotypic and molecular heterogeneity. The most common consequence associated with this condition is bone fragility, which can lead to multiple fractures and progressive bone deformities, including bowing of long bones and scoliosis<sup>(1,2)</sup>. Other extraskelatal manifestations have also been described, such as bluish discoloration of the sclera, hearing loss in young adults, and joint hypermobility<sup>(3,4)</sup>.

Muscle changes – including those in the stomatognathic system and its functions – have also been reported among extraskelatal manifestations<sup>(5)</sup>. Recent studies have observed disorders in swallowing and sucking functions, sleep-disordered breathing, and temporomandibular joint disorders<sup>(6-8)</sup>. However, the literature on this population’s muscular and functional stomatognathic issues is still scarce. Considering that multidisciplinary management improves the quality of life of OI patients<sup>(2)</sup>, studies addressing these individuals’ oral myofunctional characteristics are greatly important.

The effect of abnormal connective tissue on the upper airway of OI patients, associated with the disproportionate relative size between their tongues and oral cavities, has already been suggested as a risk factor for sleep apnea<sup>(7,9)</sup>. The tongue plays an essential role in all stomatognathic functions, and its anterior and posterior regions have different fiber and connective tissue compositions, possibly related to the different ways in which these functions are performed<sup>(10,11)</sup>. The Iowa Oral Performance Instrument (IOPI) is considered the gold standard for quantitatively measuring tongue pressure<sup>(12,13)</sup>. No studies were found in the consulted literature assessing tongue pressure in OI patients. Thus, this study aimed to investigate and relate the anterior and posterior tongue pressures in isometric contraction and during saliva swallowing in children and adolescents with OI.

## METHODS

This observational cross-sectional study was approved by the Research Ethics Committees of the Federal University of Rio Grande do Sul (UFRGS), CAAE 04448218.7.3001.5347, and of the Clinics Hospital of Porto Alegre (HCPA), CAAE 04448218.7.0000.5327, under evaluation report no. 3.526.427. All participants and their parents/guardians signed an informed consent form.

Study participants were recruited from the OI Reference Center at the HCPA, in Southern Brazil. Inclusion criteria were having a medical diagnosis of OI and being 6 to 19 years old. Exclusion criteria were a history of speech-language-hearing therapy in the previous 6 months and head and neck surgery, tumors, or trauma.

Convenience sampling followed the inclusion criteria. Based on the registered patients, the sample size calculation estimated that 70 patients could participate in the study, 67.14% (47) with OI type 1, and 32.85% (23) with OI types 3, 4, and 5. OI cases were grouped as follows: mild OI (Mi-OI group) – participants clinically classified with OI type 1; and moderate and severe OI (MS-OI group) – participants clinically classified with OI types 3, 4, and 5<sup>(3)</sup>.

The sample calculation used an estimated prevalence base in both groups (Mi-OI vs. MS-OI), setting the significance at 5% ( $\alpha$ ) and the sampling error (d) at 0.16. Since this is a rare disease, a formula was also used to correct for a finite population. This resulted in a minimum sample size of 22 cases, implying 15 cases in the Mi-OI group and seven cases in the MS-OI group.

After a routine consultation at the outpatient clinic, patients who agreed to participate were referred for study assessment. The IOPI, a portable device with a pressure transducer connected to an air-filled bulb, assessed tongue pressure. When the tongue presses the bulb against the hard palate, the resulting change in pressure is captured and displayed on its liquid crystal display (LCD)<sup>(14)</sup>. The procedure followed biosafety criteria, taking three measurements for each test, with a 1-minute rest interval between them. The highest values in each test were considered for subsequent analysis – except for the swallowing test, which used the mean of the three values<sup>(15)</sup>. The following tests were performed<sup>(15,16)</sup>:

- a) Pressure on the anterior region of the tongue (Figure 1): Raising the tongue towards the anterior palate and pressing the bulb positioned in that region. After the evaluator positioned the bulb in the anterior region of the participant’s palate, the participant was instructed to “press the air bulb with the tip of your tongue as much as possible”. After 2 seconds, the evaluator signaled that the pressure should be stopped. The sequence was repeated three times, with a 1-minute interval between repetitions<sup>(15)</sup>.
- b) Pressure on the posterior region of the tongue (Figure 2): Raising the back of the tongue towards the palate and pressing the bulb positioned in that region. After the evaluator positioned the bulb on the palate over the participant’s posterior tongue region, the participant was instructed to “press the air bulb with the middle of your tongue as much as possible, as I showed previously”.



**Figure 1.** Bulb position during pressure tasks in the anterior tongue region and saliva swallowing.  
Source: The author



**Figure 2.** Bulb position during pressure tasks in the posterior tongue region.

Source: The author

After 2 seconds, the evaluator signaled that the pressure should be stopped. The sequence was repeated three times, with a 1-minute interval between repetitions<sup>(16)</sup>.

- c) Swallowing (Figure 1): Swallowing saliva in the usual way, with the bulb positioned in the anterior palate, equivalent to the anterior tongue pressure test. After the evaluator positioned the bulb on the participant's anterior palate, the participant was instructed to "swallow the saliva". This action was repeated three times, with a 1-minute interval between repetitions<sup>(15)</sup>.

Data were statistically analyzed using the SPSS® statistical program, version 20.0 for Windows®. The significance level for all tests was set at 5% – i.e., the null hypothesis was rejected when the p-value was less than or equal to 0.05. The results were presented using absolute and relative distributions (n - %) and measures of central tendency (mean and median) and variability (standard deviation and range). The Shapiro-Wilk test studied their symmetry; the Mann-Whitney U test compared continuous variables between two independent groups; and Spearman's correlation coefficient estimated linear relationships. The study did not determine age groups; therefore, it applied the simple linear regression model.

## RESULTS

The sample comprised 22 individuals – 15 in the Mi-OI group and 7 in the MS-OI group. The mean age of the total sample was  $12.09 \pm 4.3$  years –  $12.87 \pm 3.6$  in the Mi-OI group and  $10.43 \pm 5.5$  in the MS-OI group. Altogether, 12 participants (54.6%) were females – six (40%) in the Mi-OI group and six (85.7%) in the MS-OI group.

The anterior tongue region had the highest mean pressure in the total sample and the MS-OI group. In the Mi-OI group, the posterior tongue region had the highest mean pressure. The means in the posterior tongue region differed between the

groups, indicating that the Mi-OI group had a higher mean tongue pressure in the posterior region than the MS-OI group ( $p = 0.007$ ). Both groups had similar mean anterior tongue pressure in saliva swallowing ( $p = 0.881$ ) (Table 1).

Anterior tongue pressure was moderately correlated with posterior tongue pressure ( $p = 0.001$ ), showing that the greater the pressure of the anterior region, the greater the pressure of the posterior region. Anterior tongue pressure was not correlated with anterior tongue pressure during saliva swallowing ( $p = 0.988$ ), indicating that, in this sample, the anterior tongue pressure capacity was not correlated with the anterior tongue pressure exerted when swallowing saliva. Posterior tongue pressure was moderately correlated – without any difference ( $p = 0.087$ ) – with the anterior tongue pressure during swallowing. Hence, this sample's posterior pressure capacity tended to relate to the anterior pressure exerted when swallowing saliva (Table 2).

The simple linear regression model results showed a 0.195 coefficient of determination ( $R^2$ ) for anterior tongue pressure, indicating that age can explain 19.5% of the results. The coefficient of determination ( $R^2$ ) for tongue pressure during saliva swallowing was 0.176 – i.e., age can explain 17.6% of the results. The coefficient of determination ( $R^2$ ) for posterior tongue pressure was 0.285 – i.e., age can explain 28.5% of the results.

## DISCUSSION

A study found that healthy young adults had anterior tongue pressure values of 63.94 kPa for men and 50.27 kPa for women<sup>(15)</sup>. These data, as well as the weighted average for young adults reported by the IOPI manual<sup>(16)</sup>, are higher than the present study findings. In contrast, another study found that a group of children aged 6 to 10 years not needing orthodontic treatment had 37.80 kPa, a value lower than those observed in the present sample<sup>(18)</sup>. Similar to the data of the current research, a study found an average of 48.7 kPa for the anterior region among healthy young adults<sup>(19)</sup>. Thus, OI patients with a mean age of 12 years performed like young adults in the Brazilian population.

A study observed that the posterior tongue pressure in healthy young adults was 38.4 kPa, a value lower than that of the present sample<sup>(19)</sup>. However, another study observed a higher value in participants aged 20 to 29 years; the authors found a posterior tongue pressure of 56.15 kPa<sup>(20)</sup>. A study with children and adolescents aged 3 to 16 years observed that those aged 6 years onwards had values generally higher than in the present study; only in the age groups of 6 years and 10 years – boys – were the values close to those of the current study. Thus, posterior tongue strength in children increased with age, without any significant overall difference between the sexes. Only one trend was observed, as girls had greater tongue strength than boys at 10 years old, and boys surpassed girls with greater tongue strength at 14 and 16 years old<sup>(21)</sup>.

Regarding the swallowing task, data from the literature indicate tongue pressure values higher than the present findings. A study found dorsum pressure during swallowing of 53.73 kPa in children aged 4 to 9 years<sup>(22)</sup>. It is important to mention that the position of the bulb in that study<sup>(22)</sup> was different from the one used in the present sample. Therefore, the studies could not be directly compared – although their results were quite different from the values observed in OI patients<sup>(22)</sup>.

**Table 1.** Tongue pressure in the anterior region, tongue pressure in the posterior region, and tongue pressure during saliva swallowing for the total sample and per group with osteogenesis imperfecta

Variables <sup>A</sup>	OI Groups											p-value <sup>y</sup>						
	Total (n = 22)			MI-OI (n = 15)			MS-OI (n = 7)			SD								
	Mean	SD		1	2	3	Mean	SD		1	2		3					
IOPI – bulb in the anterior region (kPa)	49.00	14.62		37.75	50.50	63.00	52.00	14.30		45.00	52.00	66.00	42.57	14.15	37.00	38.00	53.00	0.168
IOPI – bulb in the posterior region (kPa)	47.82	12.18		37.75	51.00	57.50	52.80	8.45		48.00	54.00	60.00	37.14	12.55	24.00	38.00	50.00	0.007**
IOPI – bulb in the anterior region with saliva swallowing (kPa) <sup>*MD</sup>	23.00	10.74		14.50	23.00	31.50	23.64	10.57		14.75	21.50	33.50	21.71	11.83	12.00	23.00	24.00	0.881

<sup>y</sup>Mann-Whitney U test; <sup>A</sup>variables with asymmetrical distribution – Shapiro-Wilk test; p < 0.05 \*\* statistically significant values (p ≤ 0.05) – Mann-Whitney U test

**Subtitle:** n = number of participants; \*MD = missing data; SD = standard deviation; OI = osteogenesis imperfecta; MI-OI = mild osteogenesis imperfecta; MS-OI = moderate-severe osteogenesis imperfecta; IOPI = lowa Oral Pressure Instrument; kPa = kilopascal – unit of pressure

**Table 2.** Correlation analysis between tongue pressures during tasks

Task: Comparisons		Spearman's correlation	
		r	p-value
<b>Tongue pressure</b>			
Positioned in the anterior region (kPa)	Positioned in the posterior region (kPa)	0.663	0.001**
Positioned in the anterior region (kPa)	Anterior region during saliva swallowing (kPa) *MD	-0.004	0.988
Positioned in the posterior region (kPa)	Anterior region during saliva swallowing (kPa) *MD	-0.382	0.087

\*\*statistically significant values ( $p \leq 0.05$ ) – Spearman's correlation test

**Subtitle:** kPa = kilopascal – unit of pressure; \*MD = missing data; r = 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))

The literature shows that people with OI may have limited temporomandibular joint movement<sup>(8)</sup>. A mean of 34.333 kPa was observed during swallowing in participants with temporomandibular disorder aged 18 to 28 years, with the bulb located on the incisive papilla, and the individuals swallowing saliva. The authors also observed that tongue pressure during protrusion and saliva swallowing correlated negatively with swallowing function in individuals with temporomandibular disorder. Thus, individuals with temporomandibular disorder and reduced tongue pressure have greater difficulty in swallowing adequately<sup>(23)</sup>.

Pediatric swallowing disorders have already been reported in children and adolescents with OI<sup>(6)</sup>. These data and the tongue pressure values during swallowing among the present study participants may suggest that they had difficulty in performing the swallowing function adequately or resorted to adapted swallowing.

The comparisons between OI groups showed greater posterior tongue pressure in the Mi-OI group. A study with cephalometric analysis in young adults evaluated the effects of tongue and lip pressure on dentofacial morphology. The anterior tongue pressure increases as the posterior base of the skull increases and the overjet decreases. In addition, the greater the distance from the palatine plane to the dorsum of the tongue, the lower the posterior tongue pressure<sup>(24)</sup>. Cephalometric analyses in subjects with OI found lower sagittal and vertical linear measures than those observed in the control groups – the anterior base of the skull was even smaller in OI types 3 and 4 than in type 1. On the other hand, the posterior base of the skull did not have compensatory elongation<sup>(9)</sup>. Thus, greater linear measures in OI type 1 may explain the greater anterior pressure.

On average, 57% of the muscle fibers in the body of the tongue (the region from the circumvallate papillae to the frenulum)<sup>(10)</sup> are slow-twitch fibers<sup>(11)</sup>, in contrast with 46% in the blade of the tongue<sup>(11)</sup> (the region anterior to the frenulum)<sup>(10)</sup>. A study found, among other differences, that the blade also had looser connective tissue<sup>(11)</sup>, while the medial side of the body and the base had less connective tissue, among other characteristics<sup>(11)</sup>.

The reason for these differences is not yet fully understood. However, small muscle fibers and a large amount of loose connective tissue are seemingly related to the degree to which these areas can modify their shape. Lacking a bony skeleton, muscles provide the framework on which they interact mechanically. Hence, movement in one segment requires support from another, probably requiring tonic contraction (performed by slow muscle fibers)<sup>(11)</sup> and rearrangement (enabled by connective tissue). Although it is not yet clear how the interconnections of the extracellular matrix with muscles transmit muscle contractile forces, abnormal type-I collagen

in OI probably disrupts the connective tissue organization and interactions critical for transmitting contractile force<sup>(25)</sup>. This could also explain the difference in greater posterior tongue pressure in the Mi-OI group than in the MS-OI group. It is assumed that the tongue region with less connective tissue in milder OI cases can achieve greater pressure, as slow-twitch fibers sustain this generated force, and the other tongue regions can carry out the necessary rearrangement. On the other hand, this reorganization is compromised in more severe cases, hindering the sustained contraction of a region with less loose connective tissue available.

Greater anterior tongue pressure was correlated with greater posterior pressure<sup>(26)</sup>. A study evaluated whether exercises to strengthen anterior tongue muscles would affect the strength of posterior ones – it found that the posterior tongue strength increased with the increase in strength in anterior tongue muscles, due only to the exercise to strengthen the latter. The authors justify that, thanks to the special tongue muscle structure, the anterior and posterior tongue muscles cannot contract independently, as they share a complicated contraction interconnection<sup>(26)</sup>.

The present study found no significant correlation between greater anterior pressure and the pressure during swallowing. Similarly, posterior pressure was not significantly correlated with swallowing – agreeing with data from the literature in a similar analysis, which likewise did not find statistically significant differences between anterior and posterior tongue pressure and tongue pressure during swallowing<sup>(20)</sup>. The latter is considered a tongue task that does not reach the maximum force that the tongue can achieve, with greater execution pattern variability<sup>(20,27)</sup>. The effect of age on maximum tongue strength has been described, although tongue strength during swallowing remains stable throughout most of life<sup>(20,28)</sup>. Thus, the oral structure during this function should be assessed, rather than only its ability to perform a certain task that does not correspond to the function.

Different assessments in significant samples with rare disease populations are a particularly difficult task. This is the first study to perform quantitative swallowing assessments in children and adolescents with OI. The absence of a control group and the sample size were limitations of this study. Research with a larger sample divided by age and a control group may help to understand the findings in this population.

## CONCLUSION

Greater anterior tongue pressure was correlated with greater posterior tongue pressure. The isometric capacity of the anterior

and posterior tongue was not significantly correlated with tongue pressure during saliva swallowing.

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