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Do facial morphology, posture and function change following glossectomy? A systematic review

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Abstract:

OBJECTIVES: To systematically investigate and critically appraise the quality of the currently available literature regarding the morphological, postural, and functional changes observed in individuals following glossectomy.

MATERIALS AND METHODS: A search without restrictions in eight databases (including grey literature) and hand searching from inception until March 2018 was performed. Data on morphological, postural, and functional changes after glossectomy were reviewed. Methodological quality was evaluated using the risk of bias in nonrandomized studies of intervention tool.

RESULTS: Out of 835 initially identified unique records, only three articles following patients for 1 year after glossectomy fulfilled the selection criteria. Overall, no significant morphological, postural, and functional changes were observed. Only the distance between the dorsum and the nasal line increased and the freeway space decreased significantly.

CONCLUSIONS: Overall, no significant differences were noted in the medium term, in terms of dentofacial structures adaptation and tongue function following glossectomy. Further research is warranted in order to elucidate the consequences of the altered oral environment.

Keywords:

Craniofacial development, glossectomy, macroglossia, tongue reduction surgery

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Introduction

Tongue position and volume are considered important elements that influence dentofacial biomechanics and morphological characteristics.^[1-3] The classical equilibrium theory suggests that resting pressures from the tongue, lips, and cheeks are crucial in determining the position of the teeth.^[4] A very large tongue is thought to constitute an important etiologic factor for deformities like open bite, bimaxillary protrusion, or spacing by exerting an expansive resting pressure on the dental arches or by being interposed between them.^[5-7] Tamari *et al.*^[8] demonstrated a positive relationship between the size of the mandibular arch and the volume of the

tongue. Other dentoalveolar findings in cases of an enlarged tongue include anterior or posterior cross-bite and protrusion of mandibular incisors.^[9-12]

It has been suggested also that tongue volume affects the vertical dimension of the face and the location of the chin and the symphysis.^[8,13-15] At the same time, a very large tongue may lead to disruption of physiologic functions including breathing, mastication, swallowing, and phonation.^[7] Consequently, the assessment of the size of the tongue has been held important in considering the etiology of the various types of malocclusion and the stability of orthodontic treatment outcomes.^[4]

Macroglossia is the term used to define tongue enlargement^[10] and might be classified

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into true, relative, and functional.^[16] True macroglossia is usually associated with a pronounced increase in tongue size due to a disease or syndrome,^[17,18] whereas in relative macroglossia, the tongue size is only slightly larger when compared with normal.^[19] The functional type of macroglossia occurs when the tongue does not adapt to the oral cavity after osteotomies, for example, following mandibular setback surgery.^[20] The most frequently reported treatment for macroglossia is glossectomy, that is, surgery to decrease the volume of the tongue.^[21,22] However, the information on the effect of glossectomy on dentofacial growth and function is limited. Similarly, it is not known whether pressure on the teeth actually changes.

The aim of the present study is to investigate and critically appraise the quality of the currently available literature regarding the morphological, postural, and functional changes observed in individuals following glossectomy.

Materials and Methods

A specific protocol following the guidelines outlined in the PRISMA-P statement^[23] and registered in PROSPERO (CRD42017079878) formed the basis for the present review. The Cochrane Handbook for Systematic Reviews of Interventions^[24] and the PRISMA statement^[25] were followed during conduct and reporting.

Eligibility criteria

The participants, intervention, comparison, and outcomes (PICO) acronym was used to define the eligibility criteria [Supplementary Table 1]. Studies evaluating facial morphology, posture, and function in individuals before and after any type of glossectomy, as a sole intervention or in combination with osteotomies, were reviewed. Subjects with tumors, clefts, syndromes, or congenital anomalies of the craniofacial region were excluded. Animal studies, noncomparative studies, systematic reviews, and meta-analyses were also not considered.

Information sources and search strategy

In total, eight databases (including grey literature) were searched since inception and up until March 2018. The first author developed detailed search strategies for each database. These were based on the strategy used for MEDLINE via PubMed but revised appropriately to consider the differences in controlled vocabulary and syntax rules [Supplementary Table 2].

No restrictions were placed on the language, date, or status of publication. In addition, efforts to obtain additional studies were made and the reference lists in reviews included or excluded studies, as well as other

related articles were searched. The authors of studies were to be contacted in order to provide additional data if needed.

Study selection

The first two authors assessed for eligibility, independently and in duplicate, the titles and the abstracts of the retrieved records. They were not blinded to the identity of the authors, their institution, or the results of the research. Subsequently, they obtained and assessed again the full text of the records considered by either reviewer to meet the inclusion criteria. Disagreements were resolved by discussion or consultation with the last author. A record of all decisions on study identification was kept. As recommended, kappa statistics were not calculated to describe the extent to which assessments by the two authors were the same.^[24]

Data collection and data items

The same two authors performed data extraction independently and in duplicate; any disagreements were resolved by discussion or consultation with the last author. Predetermined and prepiloted data collection forms were used to record the following information: bibliographic details of the study, details on study design and verification of study eligibility, characteristics of the subjects, details on the intervention, outcomes assessed, and results.

The retrieved data on the investigated outcomes was categorized as follows:

- (a) Morphological and postural parameters
- (b) Functional parameters.

Risk of bias in individual studies

The risk of bias in individual studies was assessed independently and in duplicate by the first two authors using the ROBINS-I tool.^[26] Any disagreements were resolved by discussion or consultation with the last author.

Summary measures and synthesis of results

If deemed possible, the random effects method for meta-analysis was to be used to combine data. However, quantitative data synthesis was not carried out as planned because of the lack of an adequate amount of data regarding each of the assessed variables.^[24]

Risk of bias across studies and additional analyses

If a sufficient number of studies were identified, analyses were planned for “small-study effects” and publication bias. If deemed possible, exploratory subgroup analyses were planned according to participant and intervention characteristics. Finally, the quality of evidence for the statistically significant differences at the longest follow-up was assessed based on the grades

of recommendation, assessment, development, and evaluation (GRADE) approach.^[27]

Results

Study selection

The flowchart of records through the reviewing process is shown in Figure 1. Initially, 835 records were identified, 346 were identified as duplicates, and 478 more were excluded on the basis of their title and abstract. From the 11 full-text records assessed, 8 were excluded because they included patients with tumors. Finally, three reports were included in the systematic review.^[28-30]

Study characteristics

The characteristics of the studies included in the present systematic review are presented in Table 1. They were published between 1990 and 2013 and investigated the effect of glossectomy on (a) morphological and postural parameters: maxillomandibular relationships; position of the head, the cervical column, and the hyoid bone; rest position of the mandible and the tongue^[28,30] and (b) functional parameters: tongue pressure on the teeth,^[29] oral motor ability, and ability to recognize forms.^[28] The mean age of the patients varied from 15 to 21 years, approximately. Ingervall and Schmoker^[28] and Fröhlich *et al.*^[29] assessed patients before and approximately

6 and 12 months after glossectomy. Kawakami *et al.*^[30] followed upto 1 year two different groups: one subjected to mandibular setback and glossectomy, and one to osteotomy alone.

Risk of bias within studies

Table 2 presents the summary of the risk bias assessment. All studies presented serious risk of bias regarding confounding, as important parameters were not always appropriately controlled. These included gender, age and growth status, type of malocclusion, presence of fixed appliances, tongue volume removed, glossectomy technique, etc., On the contrary, the risk of bias in the selection of participants and classification of interventions was found to be low. The risk of bias in the measurement of outcomes was considered serious as either the assessors were aware of the intervention, or there was no information on blinding or concerns existed regarding the reliability of the assessments. Finally, the risk of bias in the selection of the reported result was moderate for all the included studies.

Results of individual studies

(a) Effect on morphological and postural parameters
Following glossectomy, no significant spatial changes were shown in the position of the head, the cervical column, and the hyoid bone, as well as the craniocervical angulation.^[28] Evaluation of tongue at the rest position showed that the distance between the dorsum and the nasal line increased significantly ($P < 0.01$). However, no significant differences were observed on the distance between the posterior part of the tongue and the pharyngeal wall. The freeway space decreased significantly between the presurgical and the postsurgical evaluations ($P < 0.01$), but the anteroposterior position of the mandible in the rest position did not change.

In the study by Kawakami *et al.*,^[30] no significant differences were noted between the glossectomy plus mandibular setback patients and the osteotomy only group regarding the ANB angle, the Frankfort mandibular plane angle, as well as the position of the hyoid bone and the tongue at 1 year after the operation. However, the researchers did not observe in the mandibular setback plus glossectomy group the narrowing of the airway width posterior to the tongue and the clockwise rotation of the mandible that occurred in the osteotomy-only patients.

(b) Effect on functional parameters

Fröhlich *et al.*^[29] tested the tongue pressure on the teeth (maxillary and mandibular incisors and molars) at rest, as well as during chewing and swallowing, following glossectomy. The pressures reported exhibited large variability. Even negative values were recorded at rest in many individuals, particularly in the maxillary

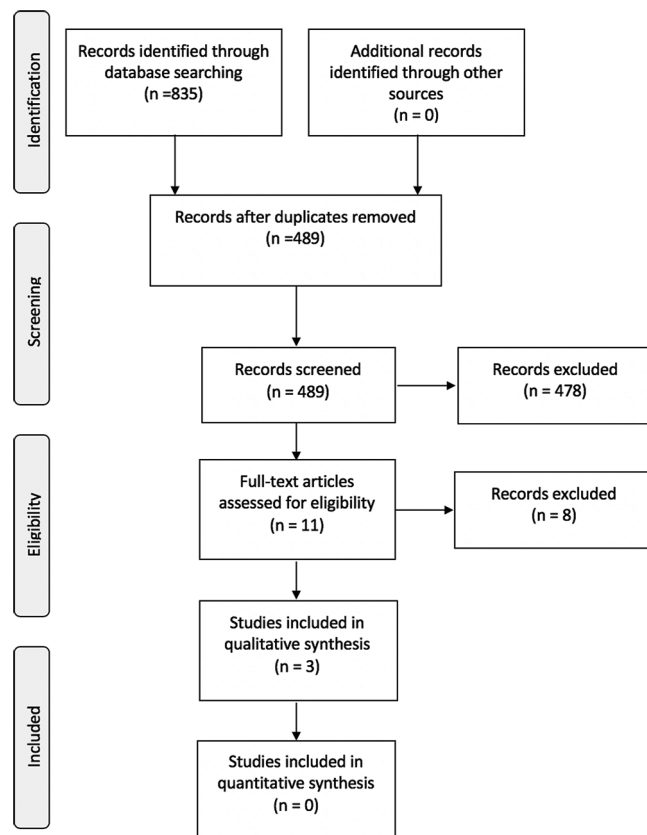


Figure 1: Flow of records through the reviewing process

Table 1: Characteristics of the studies included in the systematic review

Study	Inclusion and exclusion criteria Sample and intervention characteristics	Methodology and outcomes of assessment Additional information
Ingervall and Schmoker [1990]	Inclusion criteria: clinical diagnosis of macroglossia 27 patients: 16 M, 11 F; age at Pre-op: 15 y 8 m Intervention: glossectomy according to Schmoker (1985)	Assessments: Pre-op, Post-op 6 m, Post-op 12 m Cephalometric evaluation (natural head position): position of the head, the cervical column, the hyoid bone and the tongue, craniocervical angulation, free space and difference in the sagittal position of the mandible Oral ability to recognize forms: time needed to identify 12 test bodies placed in the mouth Oral motor ability: time needed to assemble 2 halves of a test piece in the mouth Power calculations: No; Method error: No
Fröhlich <i>et al.</i> [1993]	Inclusion criteria: clinical diagnosis of macroglossia Exclusion criteria: presence of palatal appliance, surgery 21 patients: 11 M, 10F; age at Pre-op: 15y3m Intervention: glossectomy according to Schmoker (1985)	Assessments: Pre-op, Post-op 6 m, Post-op 12 m Tongue pressure on teeth: measurements performed with an open cannula embedded in a small custom-made acrylic shield attached to on the lingual surfaces of the teeth Power calculations: No; Method error: Yes, certain problems were observed
Kawakami <i>et al.</i> [2004]	Inclusion criteria: Md setback surgery Exclusion criteria: 2-jaw surgery and/or craniofacial anomaly 40 patients: 10 M, 30 F; age at Pre-op: 20y10m Intervention: Group 1: Md setback + glossectomy (Egyedi-Obwegeser/Mixter) Group 2: Md setback	Assessments: Pre-op, Post-op 6 m, Post-op 12 m Cephalometric evaluation (Frankfort horizontal parallel to the floor): ANB, FMA, position of the hyoid bone and the tongue Power calculations: No; Method error: Yes, no results reported

FMA – Frankfort mandibular plane angle; Md – Mandibular; Pre-op – Before operation; Post-op – After operation; m – Months; y – Years

Table 2: Summary of the risk of bias assessment

Domain of bias	Ingervall and Schmoker, 1990	Fröhlich <i>et al.</i> , 1993	Kawakami <i>et al.</i> , 2004
Due to confounding	Serious	Serious	Serious
In selection of participants	Low	Low	Low
In classification of interventions	Low	Low	Low
In measurement of outcomes	Serious	Serious	Serious
In selection of the reported result	Moderate	Moderate	Moderate
Overall	Serious	Serious	Serious

incisors. In general, none of the measurements at 12 months differed significantly from the preoperative recordings. Moreover, glossectomy exerted a minor effect on patients' ability to recognize forms and overall oral motor ability.^[28]

Risk of bias across studies and additional analyses

As it was not possible to retrieve a sufficient number of trials, we were not able to conduct analyses for "small-study effects" and publication bias.^[24] Overall, the confidence in the observed estimates was not strong [Supplementary Table 3].

Discussion

Despite clinical reasoning that tongue volume influences not only the position of the maxillary and mandibular dentition, but also the posture of the mandible and the vertical height of the face, there is limited information about the effects of altering tongue volume on craniofacial growth and dental arch formation. Based on the data

provided in the present systematic review, overall, no significant differences were observed in the medium term, in terms of dentofacial structures adaptation and tongue function following glossectomy. However, the quality of retrieved evidence gives an insight on the strength of the relevant estimates.

From the initially identified records, only three full-text studies evaluating the changes resulting from glossectomy were included in this systematic review, reflecting the scarcity of relevant research. No significant effects on morphological and postural parameters, like the spatial position of the head, the cervical column, and the hyoid bone, or the craniocervical angulation, were noted.^[28] However, it seems that the operation resulted in the tongue occupying less space in the oral cavity than before, this possibly being the reason for the observed decrease in the freeway space. Liu *et al.*,^[3] employing a young animal model, reported that a surgical decrease in tongue volume slows craniofacial skeletal growth in the mandibular symphysis regions

and anterior dental arch expansion during periods of accelerated growth. In addition, decreases in bone mineral density and content were noted, especially in the anterior mandibular region.

When glossectomy was applied as an adjunct to mandibular setback, no significant differences were noted between the glossectomy plus setback patients and the osteotomy-only group regarding the ANB angle, the Frankfort mandibular plane angle, as well as the position of the hyoid bone and the tongue, 1 year after the operation.^[30] It would seem logical that when the mandible is set back, the tongue would also move posteriorly and narrow the upper airway. However, the cervical hyperflexion that usually characterizes these patients postsurgically^[31] as well as subsequent alterations in hyoid bone position^[32] may compensate and prevent airway obstruction. The small effect of tongue reduction on mandibular relapse also supports the idea that large tongue volume is not inherent in patients with mandibular prognathism^[33] and that adjunctive tongue resection is rarely necessary in the mandibular prognathism patients without the existence of a disproportionate tongue size.^[34,35] The reported results could also be related to the methodology used. The use of lateral cephalometric radiography limits the accuracy of airway measurements.^[36] The two-dimensional images allow only assessments in the sagittal plane and fail to provide a full-scaled view of the upper airway. Cone beam-computed tomography (CBCT), by allowing a 3D registration of pre- and post-treatment data, and magnetic resonance imaging (MRI) appear to be superior to cephalometric radiography in estimating the size of the pharynx and the hypopharynx.^[37]

Regarding the recordings on functional parameters, such as tongue pressure on teeth, great variability was noted corroborating previous investigations in young adults.^[6,38] Twelve months after glossectomy, no measurement was significantly different from the preoperative assessment at rest, as well as during chewing and swallowing.^[29] Liu *et al.*^[4] showed in an animal study that immediately following glossectomy the overall strains from the tongue during mastication decreased and their orientation changed. However, this effect was less pronounced in the posterior mandible and palate. In the mandibular lingual symphysis, no strain change was observed. Four weeks after glossectomy, tongue strain during chewing increased again, particularly on the mandible.^[39] These findings are in contradiction to the hypothesis that lower loads would be exerted on the anterior oral cavity during function. The restructuring and healing processes in the tongue together with alterations in muscle contraction modes and kinematics during chewing could explain these observations.^[40-42] However, according to the classical equilibrium theory,

functional pressures are not as critical in determining tooth position as resting pressures.^[2]

Finally, glossectomy exerted a minor effect on patients' oral motor ability and ability to recognize forms, possibly due to the fact that partial tongue reduction does not significantly affect overall oral proprioception.^[28] In general, individuals performing well before the operation continued to do so later, corroborating previous longitudinal observations in a young adult population.^[43] Learning effects have been reported in subjects non-subjected to glossectomy^[43,44] that was not observed in the population under investigation. This finding could possibly be related to the fact that more patients wore fixed orthodontic appliances after surgery.

Strengths and limitations

The strengths of the present review include the use of a methodology which followed well-established guidelines. To our knowledge, there has been no other systematic review conducted on craniofacial changes after glossectomy. Moreover, the search strategy employed was both exhaustive, covering electronic, manual, and gray literature material up to March 2018, and comprehensive including every relevant study. Every effort to decrease bias in the methodology employed was made. Screening, verification of eligibility, abstraction of information, assessment of risk of bias, and of the quality of evidence were all performed in duplicate, and any disagreement was resolved by discussion or consultation until a final consensus was achieved.

There are also some limitations to the present review; these arise mainly from the nature and the characteristics of the data retrieved during the review process. The absence of contemporaneous control groups followed for similar time periods constitutes another significant methodological limitation. Furthermore, exploratory subgroup analyses for "small-study effects" and publication bias,^[24] could not be carried out, even though they were incorporated as possibilities according to the review protocol. Finally, the lack of appropriate tools to measure specific parameters following glossectomy, for example, Cone Beam CT for airway measurements may account for the observed results.

Recommendations for future research

Further research is warranted in order to elucidate the consequences of the altered oral environment following glossectomy on craniofacial growth, development, and function, especially in growing subjects.

It has been suggested that well-designed and properly executed randomized control trials provide the best

evidence.^[45] Since random allocation of subjects might be unethical under certain situations, it would be advisable to conduct at least well-controlled prospective nonrandomized studies.^[26] Particular importance should be placed on possible ways to control bias due to confounding and bias in the measurement of outcomes. Moreover, long-term evaluation of outcomes would be valuable concerning a variety of malocclusions and clinical situations such as open-bite cases and tongue thrust. Also, using more advanced tools for assessment, like Cone Beam CT, could be beneficial to assess morphological adaptations and airway volume changes in three dimensions after glossectomy. Finally, it would be of great interest to further investigate the effect of the different surgical techniques, as well as the effect on other outcomes including speech, taste, and sensitivity.

Conclusions

Overall, no significant differences were noted in the medium term, in terms of dentofacial structures adaptation and tongue function following glossectomy. Further research is warranted in order to elucidate the consequences of the altered oral environment.

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Conflicts of interest

There are no conflicts of interest.

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Supplementary Table 1: Eligibility criteria for the present systematic review

Domain	Inclusion criteria	Exclusion criteria
Participants	Individuals of any age, gender, and ethnicity having undergone glossectomy.	Subjects with tumors, clefts, syndromes, or congenital anomalies of the craniofacial region.
Interventions	Any type of glossectomy as a sole intervention or combined with other osteotomies.	
Comparisons	Comparison between the measurements before and after glossectomy. Comparison with a contemporaneous, no glossectomy group of individuals followed for a similar period of time (if applicable).	
Outcomes	a. Morphological and postural parameters: skeletal and soft tissue morphology, occlusion, upper airway, mandibular position, etc. b. Functional parameters: oral ability to recognize forms, oral motor ability, tongue pressure on the teeth, etc.	
Study design	Experimental and observational studies comparing the outcomes of interest in individuals before and after glossectomy.	Animal studies. Non-comparative studies (case reports and case series). Traditional reviews. Systematic reviews and meta-analyses.

Supplementary Table 2: Strategy for database search (up to March 2018)

Database	Search strategy	Hits
PubMed	((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket))	302
Cochrane Central Register of Controlled Trials	((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket)) in Title, Abstract, Keywords in Trials'	13
Cochrane Database of Systematic Reviews	((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket)) {Including Limited Related Terms}	10
Scopus	TITLE-ABS-KEY (((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket)))	316
Web of Science™	TOPIC: (((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket))) Timespan: All years. Search language=Auto	162
Arab World Research Source	((tongue AND reduc*) OR (tongue AND resec*) OR glossectomy OR macroglossia) AND (("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket))	29
ClinicalTrials.gov	(orthodontic OR orthodontics) AND ((tongue AND reduction) OR (tongue AND resection) OR glossectomy OR macroglossia)	0
ProQuest Dissertations and Theses Global	ti(((tongue AND reduc*) OR glossectomy) AND ("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket)) OR ab(((tongue AND reduc*) OR glossectomy) AND ("fixed appliance" OR orthodon* OR "fixed orthodontic" OR bracket* OR multibracket))	3

Supplementary Table 3: Quality of available evidence

Studies	Quality assessment					Subjects	Effect Absolute	Quality ^a
	Risk of bias	Inconsistency	Indirectness	Imprecision	Other			
Distance between tongue dorsum and the nasal line [mm comparison before and 12 months after the operation]								
1	Serious ¹	Not serious	Not serious	Serious ²	No	27	Median 2.6 mm greater, <i>P</i> <0.01	⊕⊕⊕○, very low
Freeway space [mm comparison before and 12 months after the operation]								
1	Serious ¹	Not serious	Not serious	Serious ²	No	27	Median 0.7 mm less, <i>P</i> <0.01	⊕⊕⊕○, very low
Ability to recognise forms level 3 test bodies time [secs comparison before and 12 months after the operation]								
1	Serious ¹	Not serious	Serious ³	Serious ²	No	27	Median 3.2 secs higher, <i>P</i> <0.01	⊕⊕⊕○, very low
Ability to recognise forms level 1 test bodies misidentifications [number comparison before and 12 months after the operation]								
1	Serious ¹	Not serious	Serious ³	Serious ²	No	27	Mean 1.44 higher, <i>P</i> <0.01	⊕⊕⊕○, very low
Ability to recognise forms level 2 test bodies misidentifications [number comparison before and 12 months after the operation]								
1	Serious ¹	Not serious	Serious ³	Serious ²	No	27	Mean 1.52 higher, <i>P</i> <0.01	⊕⊕⊕○, very low

CI – Confidence interval. ^aQuality of evidence rating started from low because of the non-randomized study design. ¹Studies were considered as being of serious risk of bias. ²The results are based only on one study. ³The results are based only on one level of difficulty