Lymphangioma of the tongue associated with open bite: case report

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Abstract

Background Lymphangioma of the tongue is a rare condition related to congenital malformations of the lymphatic system. It may have different implications such as macroglossia. This condition could exacerbate in aesthetic abnormalities and functional problems such as maxillofacial structural deformities, dysphagia, airway obstruction and speech difficulties. The aim of this paper is to describe this disease by means of a case report.

Case report A 4-year-old patient was referred to our department presenting macroglossia, functional difficulty during swallowing and mastication, speech disturbances, airway obstruction, and deformities of the maxillofacial structures. Diagnosis of lymphangioma was confirmed by biopsy. After surgical removal of the lesion, the patient was treated with rapid palatal expansion, functional appliance with lingual grid and fixed multibracket appliance. After treatment, improvement in function, indicated by the gradual increase in occlusal force, could be taken as a positive sign of stability.

Conclusion The 3-step treatment protocol described in this article proves to be effective in controlling the malocclusion in the three planes of the space in a patient affected by lymphangioma of the tongue.

KEYWORDS Difficult chewing; Lymphangioma of the tongue; Macroglossia; Open bite; Swallowing difficulty.

Introduction

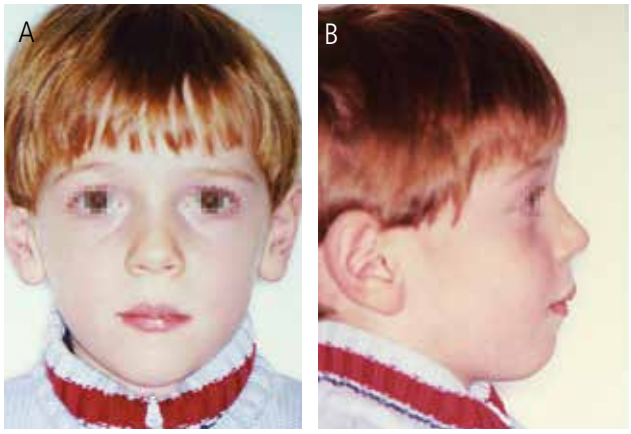
Lymphangioma of the tongue is a rare condition related to congenital malformations of the lymphatic system. It is caused by the formation of benign tumors on the dorsum of the tongue, but it could also occur in the palate, buccal mucosa, gingiva, and lips [Iamaroon et al., 2003]. It may have different implications such as macroglossia [Guelmann and Katz, 2003]. This condition is essentially painless but it could exacerbate in aesthetic abnormalities and functional problems such as maxillofacial structural deformities, dysphagia, airway obstruction and speech difficulties. The diagnosis usually occurs during infancy and early childhood. Treatment is usually multidisciplinary.

Case report

A Caucasian, 4-year-old male patient presented to the attention with a chief complaint of unaesthetic appearance related to macroglossia, functional difficulty during swallowing and mastication, speech disturbances, airway obstruction, and deformities of maxillofacial structures (Fig. 1a-d).

Clinical history revealed the absence of relevant systemic conditions. No other mass was detected in the head and neck region. Panoramic radiograph showed no anomalies in number and position of the teeth. The frontal cephalometric radiograph revealed a slight mandibular asymmetry. Lateral cephalometric analysis showed a skeletal Class II open bite with increased lower facial height (Fig. 1e-g). Cephalometric values before treatment are shown in Figure 2a.

The main treatment objectives were to re-establish a proper occlusion and function as early as possible. Initially, the tongue-reduction surgery at the age of 5 years was performed. Incisional biopsy under local anaesthesia was performed on the dorsal portion of the tongue for histopathologic examination. A definitive diagnosis of lymphangioma was then established. Surgical removal represents the treatment of choice when the lymphangioma of the tongue interferes with function or aesthetics [Jian, 2005].



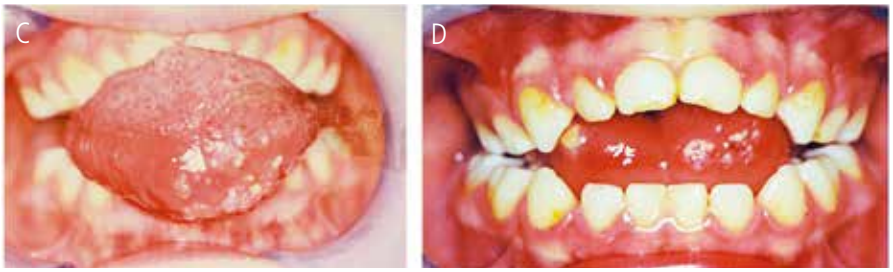


FIG. 1 Initial situation of the patient before treatment.
Extraoral examination of the patient (A, B).
Intraoral examination during swallowing and at rest (C, D).
Radiographic examinations prescribed for the case study and diagnosis (E-G).

SAGITTAL MEASURES	**SAGITTAL MEASURES**
SNA: 83,3°	SNA: 81,1°
SNB: 70,8°	SNB: 75,7°
ANB: 12°	ANB: 5,3°
A-NFH 0,2 mm	A-NFH -0,8 mm
Po-NFH -2,1 mm	Po-NFH -2,7 mm
Wits: 0,8 mm	Wits: 0,6 mm
DENTOALVEOLAR MEASURES	**DENTOALVEOLAR MEASURES**
Is-APo 0,8 mm	Is-APo 0,8 mm
Ii-APo 0,3 mm	Ii-APo 0,3 mm
Is-MAX 115,8°	Is-MAX 115,8°
Ii-MAND 104,2°	Ii-MAND 104,2°
VERTICAL MEASURES	**VERTICAL MEASURES**
SN^Go-Gn: 41,5°	SN^Go-Gn: 41,5°
FMA 33,2°	FMA 33,2°
MM: 30,8° mm	MM: 30,8° mm

(Landmarks: S, sella; N, nasion; Go, gonion; A, Point A; B, Point B; Pog, pogonion; Gn, gnathion; Me menton; PNS, posterior nasal spine; U, tip of soft palate; Ptm, pterygomaxillary fissure; Ba, basion).

FIG. 2 Cephalometric values before (A)and after (B) treatment.

FIG. 3 First phase of treatment. Rapid maxillary expansion with Hyrax rapid palatal expander.

The patient was periodically re-evaluated and the lesion did not show tendency to enlarge.

Progressive open bite closure was observed after the glossectomy. By the time the patient was 6 years old, the treatment plan consisted in a first phase of orthodontic treatment with rapid maxillary expansion by a Hyrax expander cemented on the upper primary second molars (Fig. 3) to obtain transverse increase of the upper jaw. At the age of 8, laterolateral and posteranterior teleradiographs and a new panoramic radiograph were prescribed for further evaluation. At the age of 12, the patient was re-examined for the second phase of orthodontic treatment with the use of a functional appliance with lingual grid (Fig. 4) in order to control the skeletal Class II growth pattern and the tongue position. The third phase of treatment involved the use of orthodontic multi-bracket straight-wire appliance (Fig. 5).

After a period of 28 months, overbite and overjet improved, resulting in Class I molar and canine relationships. Cephalometric values after treatment are shown in Figure 2b. Maxillary anterior proclination was corrected, the dental and facial midlines were improved, both arches were aligned and the open bite was successfully managed (Fig. 6, 7).

FIG. 4 Second phase. Functional appliance with lingual grid.

FIG. 5 Third phase. Orthodontic multi-bracket straight-wire appliance.

FIG. 6 Extraoral examination after treatment (A-C). Final occlusion after the orthodontic appliance removal (D, E, F).

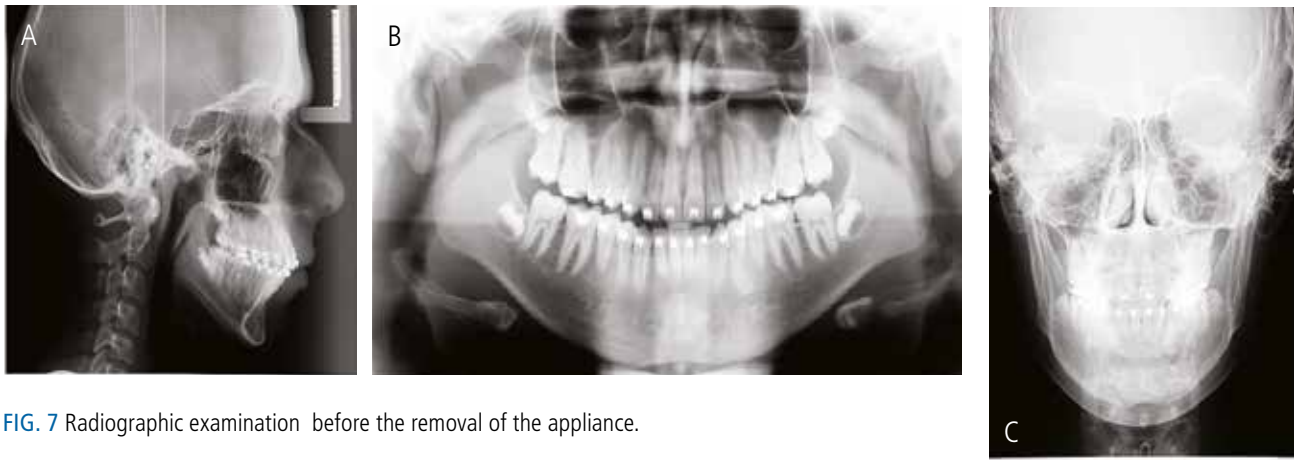


FIG. 7 Radiographic examination before the removal of the appliance.

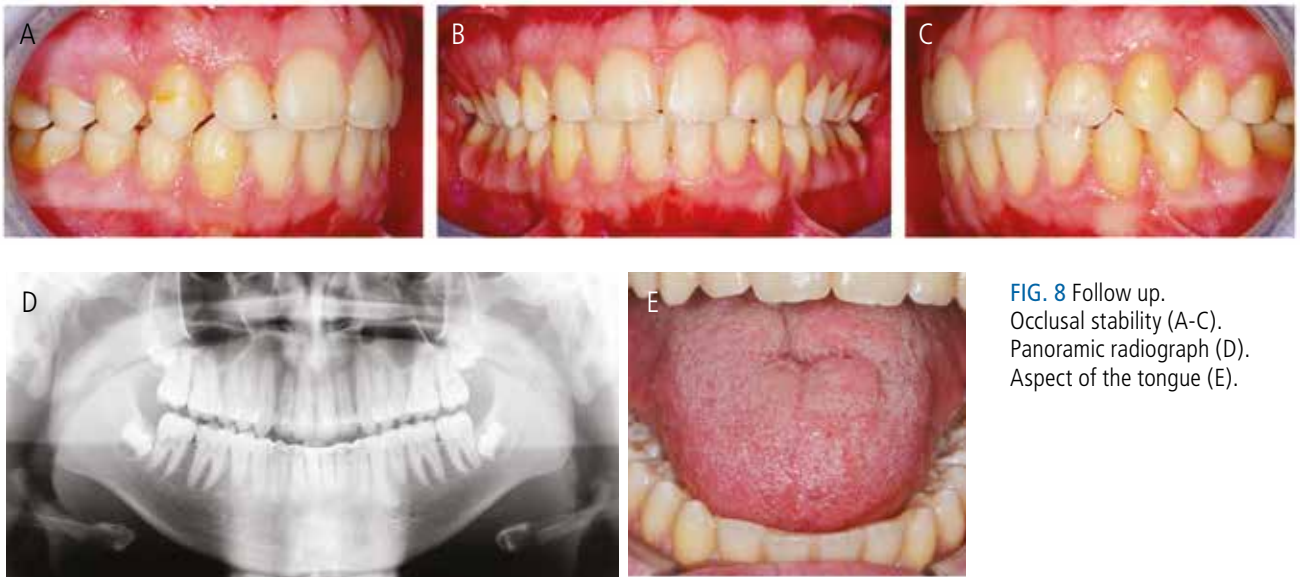


FIG. 8 Follow up.
Occlusal stability (A-C).
Panoramic radiograph (D).
Aspect of the tongue (E).

Discussion

Macroglossia as consequence of lymphangioma can cause skeleteal and facial deformities during childhood. In this case report, the main treatment objectives were to establish a dental Class I relationship, the closure of the open bite and the achievement of a dental Class I occlusion with an ideal overjet and overbite. By considering the objectives of treatment, reduction of excess tongue volume was of paramount importance to obtain the recovery of all the oral multifunctions [Farronato et al., 2012; Maspero et al., 2014]. Orthodontic treatment was divided in three phases according to the growth of the patient. The final occlusion was well maintained regardless of the remaining vertical growth (Fig. 8). Improvement in function indicated by the gradual increase in occlusal force could be taken as a positive sign of stability. However, considering the patient's skeletal age, careful follow-up was necessary.

Conclusions

Macroglossia caused by lymphangioma can be a major cause of an open bite malocclusion. Thorough evaluation of the aetiology, proper diagnosis, and timely intervention favourable functional outcomes were obtained.

The 3-step treatment protocol described in this article proved to be effective in controlling the transverse, vertical and sagittal dimensions of the patient. In particular:

- Macroglossia caused by a lymphangioma may be associated with skeletal and facial deformities as open bite.
- Surgical excision should be followed by orthopaedic and orthodontic treatment.

In order to avoid relapse it is important to begin the treatment in the early phases of infancy and to continue it to the end of the growth. Compliance and multidisciplinary approach management are the keys for stable results.

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