

Lingual lymphatic malformation: A combination of sclerotherapy and sirolimus

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Sir,

Lymphatic malformations are rare, benign, slow-flowing, congenital malformations composed of abnormal cystic dilatations. Although congenital, they are not always apparent at birth. Approx. 90% of the lesions are diagnosed before the age of five and may be subject to inflammatory flare-ups in response to local infectious or traumatic events. They may involve the skin, mucous membranes, or underlying tissues, then called superficial lymphatic malformations, or involve underlying organs, then called deep lymphatic malformations.

A seven-year-old child had presented, since the age of two years, with a swelling at the base of the tongue with a nipple-like surface and a sessile base (Fig. 1a) with an MRI appearance in favor of lymphangioma. He was put on sirolimus at a dose of 0.8 mg/m^2 and three sessions of sclerotherapy with bleomycin (Fig. 1b).

A three-year-old child had macroglossia present since birth with cystic formations on the ventral side of the tongue (Fig. 1c) with an MRI appearance in favor of lymphangioma treated with sirolimus at a dose of 0.8 mg/m^2 and three sessions of sclerotherapy with bleomycin (Fig. 1d).

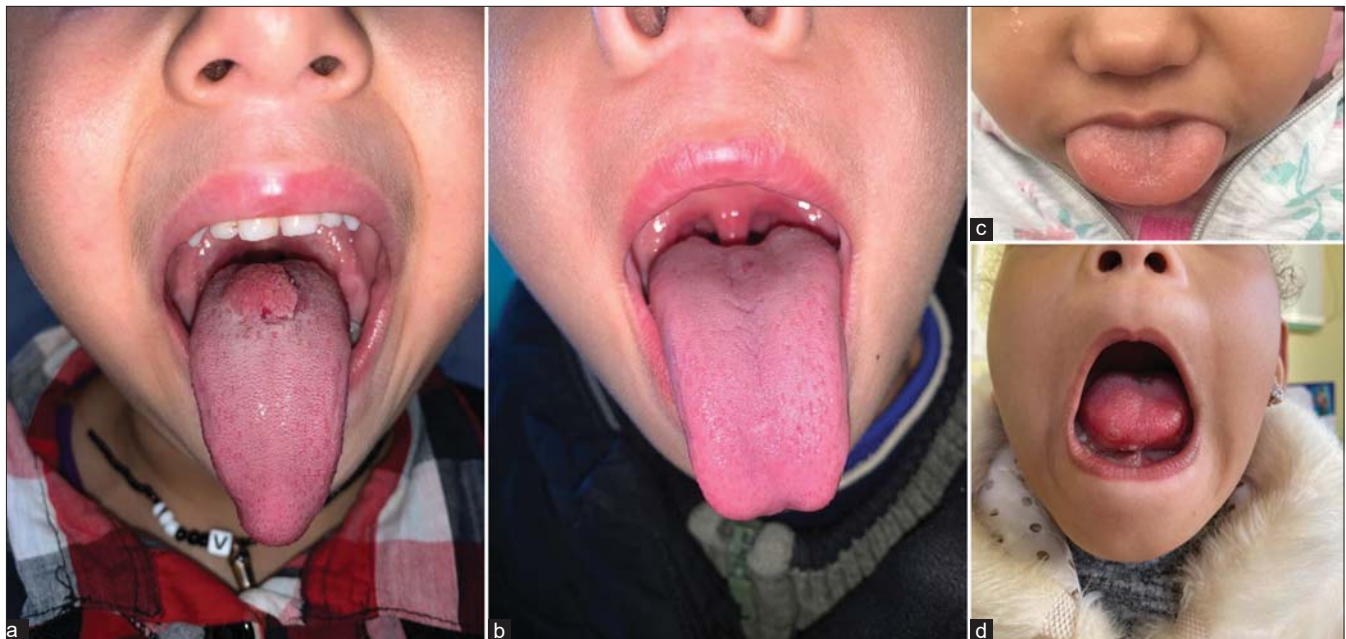


Figure 1: (a) Swelling at the base of the tongue with a nipple-like surface and sessile base. (b) Result after treatment. (c) Macroglossia with cystic formations on the ventral side of the tongue. (d) Result after treatment.

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mTOR is an enzyme of the serine/threonine kinase family that regulates proliferation, growth, cell mobility and survival, protein biosynthesis, and transcription. Sirolimus, by directly inhibiting mTOR, has anti-proliferative, immunosuppressive, and anti-angio and lymphangiogenic properties.

Bleomycin sclerotherapy, which consists of injecting a product causing an inflammatory reaction followed by fibrosis responsible for the retraction of the cysts, after puncturing and aspirating these, has a significant role in the therapeutic management of lymphangiomas, particularly macrocystic lymphangiomas.

A combination of sirolimus at a dose of 1.6 mg/m² per day in two doses and sclerotherapy with bleomycin with an average of two sessions per patient allowed a clear decrease in the size of the malformation in our two patients.

Two cases reported in the literature on lymphangioma of the tongue treated by sclerotherapy with bleomycin concluded to the effectiveness and good tolerance of this molecule [1,2].

Sirolimus is prescribed for patients with tongue lymphangioma as a second-line treatment in the case of failure or an incomplete response to precursor treatments, such as surgery and sclerotherapy. Sirolimus was prescribed at a dose of 1.6 mg/m² taken twice a day with the result that the size of the malformation decreased significantly, especially in microcystic lymphangiomas, with a suspensive effect [3,4].

Sclerotherapy with bleomycin and sirolimus are treatments of choice for lymphatic malformations of the tongue with their combination giving quicker results with good tolerance without side effects.

Consent

The examination of the patient was conducted according to the principles of the Declaration of Helsinki.

The authors certify that they have obtained all appropriate patient consent forms, in which the patients gave their consent for images and other clinical information to be included in the journal. The patients understand that their names and initials will not be published and due effort will be made to conceal their identity, but that anonymity cannot be guaranteed.

REFERENCES

1. Kataria P, Passey JC, Agarwal AK. Lymphangioma circumscriptum of the tongue: successful treatment using intralesional bleomycin. *J Laryngol Otol.* 2009;123:1390-2.
2. Chakravarti A, Bhargava R. Lymphangioma circumscriptum of the tongue in children: Successful treatment using intralesional bleomycin. *Int J Pediatr Otorhinolaryngol.* 2013;77:1367-9.
3. Akyüz C, Ataş E, Varan A. Treatment of a tongue lymphangioma with sirolimus after failure of surgical resection and propranolol. *Pediatr Blood Cancer.* 2014;61:931-2.
4. Pandey V, Tiwari P, Sharma SP, Kumar R, Panigrahi P, Singh OP, et al. Development of a biomarker of efficacy in second-line treatment for lymphangioma of the tongue: A pilot study. *Br J Oral Maxillofac Surg.* 2019;57:1137-42.

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