

A Case Report of a Lingual Cyst Lined by Respiratory Epithelium in a Child

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ABSTRACT: The lingual cyst lined by respiratory epithelium is a rare pathology. It probably appears from the default of undifferentiated cells of the foregut during embryonic growth. This pathology is seen more often in males and children; however, only 5 patients younger than 4 years old have been reported. The pathophysiology and the management of this cyst were described in a 4-year-old girl. She presented with a soft mass on the dorsum of the tongue covered by normal mucosa, which existed since her birth, causing difficulty in eating, breathing, and talking. The magnetic resonance described a hyperintense image with an anteroposterior diameter of 27 mm, craniocaudal of 19 mm, and transversal of 26 mm in the midline groove of the tongue; the scintigraphy showed normality. The enucleation of the lesion was performed, eradicating the capsule of the cyst and obtaining a complete cleavage. The histopathologic examination defined a cyst lined predominantly by respiratory epithelium. Unlike in other cases, in this case their cystic lining and capsular constituents were contemplated considering the current histological recommendations. It is relevant to differentiate this pathology from other cysts with similar histological findings.

KEYWORDS: Cysts, respiratory epithelium, tongue diseases, epithelium, respiratory

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Introduction

Lingual cysts are infrequent; they are possibly consequent from epithelia of the upper gastro-respiratory area. From a descriptive point of view, they would be termed as lingual cysts lined by gastrointestinal and/or respiratory epithelium. Embryonal remnants captured inside the developing tongue could suffer cystic degeneracy, creating a framework lined by the respiratory-form or the gastrointestinal-form epithelium.¹

The lingual cyst with respiratory epithelium is a rare pathology, more often in males² and children; however, only 5 patients younger than 4 years have been reported (4 females³⁻⁶ and 1 male¹). It is crucial to take into account that there was abundant disorientation concerning the terms used to depict these cysts until 1999, when Manor et al⁷ commended descriptive histologic wording. Thus, a considerable number of lingual cysts have been reclassified by contemplating their cystic lining and capsular constituents.³

This case describes the diagnosis and the management of a lingual cyst lined by respiratory epithelium in a 4-year-old girl.

Case Report

A 4-year-old girl consulted at the service of Stomatology and Maxillofacial Surgery at the San Vicente Hospital in Medellín, Colombia, presenting a soft mass in the midline groove of the tongue (Figure 1A), which had existed since her birth, causing difficulty in eating, breathing, and talking. Her mother reported that the mass had grown slowly until a year ago.

On inspection, the external appearance showed a 3 × 3 cm² soft, well-delimited, nontender mass, affecting the midline

dorsum of the tongue, covered by normal mucosa. A minor limitation of tongue movement was also observed. There were no additional physical alterations perceived. The magnetic resonance described a hyperintense image with an anteroposterior diameter of 27 mm, craniocaudal of 19 mm, and transversal of 26 mm in the midline dorsum of the tongue (Figure 1B and C). Besides, scintigraphy was requested to define the possibility of the presence of thyroid tissue in the tongue and the results showed normality. With this information, a presumptive diagnosis of a dermoid cyst was proposed, and surgery was scheduled to excise the lingual mass.

Previously to the incision on the dorsum of the tongue, the lesion was aspirated, obtaining a white liquid with a dense texture of approximately 10 mL (Figure 1D). A single midline incision of approximately 5 mm was performed on the dorsum of the tongue, and a thick-walled cystic mass was observed. The excision of the lesion was performed, eradicating the capsule of the cyst, conserving the contiguous muscle tissue, and obtaining a complete cleavage (Figure 2A and B). After the excision, the final repair measured approximately 7 cm by 5 cm. The suture was performed using 4-0 absorbable sutures for muscle planes, and the mucosa was sutured with interrupted stitches using 4-0 vicryl sutures.

Following the American Academy of Pediatrics recommendations,^{8,9} the child was treated with cefazolin (25 mg/kg/d IV for 3 days in fourth equally divided doses) and dexamethasone (0.1 mg/kg/d IV for 2 days, divided q12h).

The histopathologic examination defined a cyst lined predominantly by respiratory epithelium *supported by a dense*



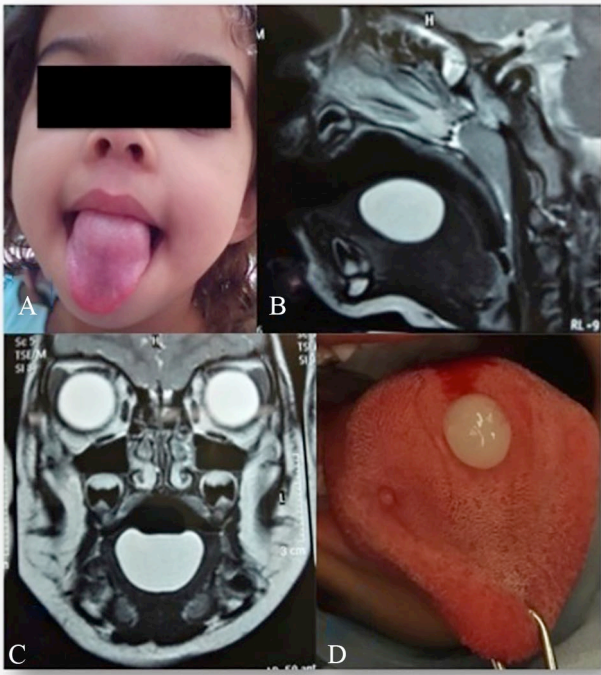


Figure 1. (A) Intraoral photograph presenting the cystic lesion affecting the lingual dorsum. (B) Sagittal view of the magnetic resonance displayed in a hyperintense image. (C) Coronal view of the magnetic resonance showing the cyst. (D) Aspiration of the cyst presenting a white liquid with a dense texture.

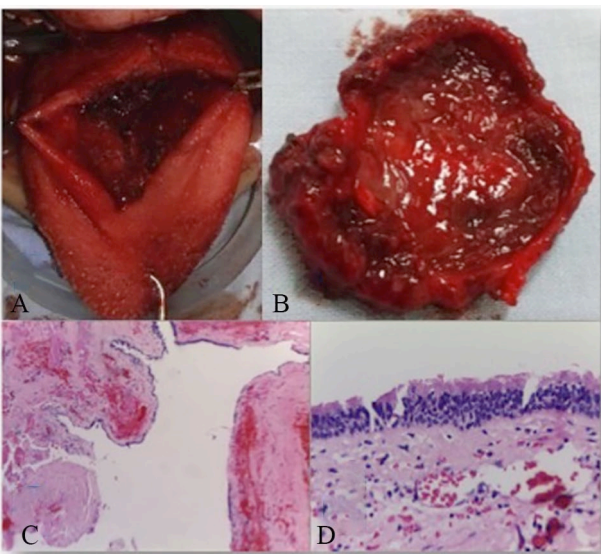


Figure 2. (A) Excision of the lesion. (B) The cut surface of the cyst. (C) Cyst lined uniformly by ciliated columnar epithelium (hematoxylin and eosin). (D) Magnification illustrating the ciliated, columnar type epithelium (hematoxylin and eosin).

connective tissue capsule (without inflammatory cells) with scarce zones presenting non-keratinized stratified squamous epithelium. No dermal appendages were appreciated in the connective tissue (Figure 2C and D). This conducted to the concluding diagnosis of a lingual cyst lined by respiratory epithelium.

Figure 3A presents the suture on the dorsum of the tongue, and Figure 3B shows the postoperative after 4 days. The child was followed up for 6 months, and there was no recurrence.

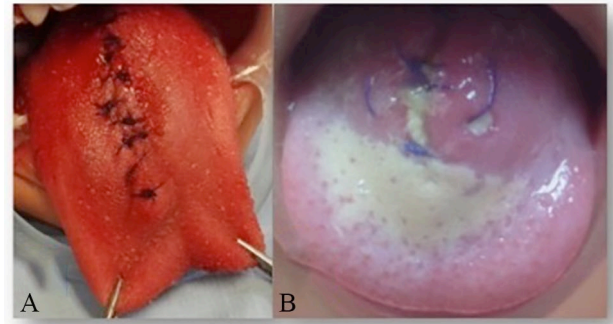


Figure 3. (A) Suture on the dorsum of the tongue. (B) Postoperative after 4 days.

The parents of the child permitted to publish images and all the procedures signing the informed consent.

Discussion

The lingual cyst lined by respiratory epithelium appears from the default of undifferentiated cells of the foregut during embryonic growth.¹⁰ The name of this cyst was proposed to differentiate it from other cysts with similar histological findings.¹ Unlike other cases, this case contemplated their cystic lining and capsular constituents considering the current histological recommendations.³

Contrary to this report, the literature describes a minor masculine preference; only 5 females with lingual cysts lined by respiratory epithelium have been informed, and most of them have been labeled in the first 2 years of their lives¹⁰; however, it has been occasionally reported in teenagers.¹¹ As was described in other works, the cyst reported here was localized in the lingual dorsum¹⁰⁻¹²; however, other locations, such as the floor of the mouth^{3,5} and the ventral tongue, were observed.^{2,11-13}

Figure 2C and D shows that the lingual cyst lined by respiratory epithelium characterizes a pathology demarcated histologically by the existence of ciliated, columnar respiratory epithelium and the nonappearance of any additional arrangement inside the cyst wall. This type of cyst may be diagnosed considering the morphology solely, following recommendations previously reported.³ It is essential to highlight that the epithelium would be diffusely positive for CK7 if the immunohistochemical analysis is performed¹⁴; for this reason, immunohistochemical analysis is not usual.³ Considering the histologic presentation, the differential diagnosis of the lingual cysts must embrace epidermoid, dermoid, teratoid, and thyroglossal duct cysts, lingual thyroid, hemangioma/lymphangioma, mucocele or ranula, and lymphoepithelial cysts. Therefore, these pathologies must include some structures, such as squamous epithelium and lumen filled with keratin; skin appendages in the wall of the cyst; derivatives of the ectoderm, endoderm, and mesoderm; columnar or stratified squamous epithelium with thyroid tissue in the cyst wall; mass of thyroid tissue; endothelium-lined spaces; extravasated mucin and granulation tissue; and lymphoid aggregates in the cyst wall.^{2,10,15}

As performed in the present case, the selection process for these cysts is surgical excision.^{2,8,16} The resection method considers the approachability to the mass, magnitude, and care for postoperative airway impediment secondarily to lingual inflammation.¹⁷ Besides, antibiotics and steroids are recommended.¹⁸

Author Contributions

C-M Ardila: writing and revision of the paper, collection, and making the data and/or figures; E Alvarez-Martinez: supervision for the description of pathology and surgery.

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