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Case Report

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Congenital mucocele of the ventral surface of the tongue: A case report

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Abstract

Congenital mucocele of the tongue is an extremely rare benign condition affecting the minor salivary glands. A 22-day-old male neonate was admitted with congenital mucocele of the ventral aspect of the tongue. His clinical presentation included a swollen tongue since birth. It was a renitent, immobile mass, with no associated inflammatory signs, occupying the ventral surface of the tongue, preventing oral closure and impeding feeding. His physical examination revealed no other morphological abnormalities. Facial computed tomography in 2-mm thin sections without and after injection of contrast medium confirmed the liquid aspect of the mass, well-circumscribed by a regular, thin clean wall which was enhanced after injection of contrast medium, without calcifications or partitioning. The diagnosis was compatible with a cystic lesion of the tongue, without being able to predict its etiology. Treatment consisted of complete removal of the cyst via the posterior surface of the tongue, with simple follow-up and no recurrence after 13 months. Histological examination confirmed the diagnosis of retentional mucocele. In conclusion, lingual mucocele is a benign tumor which may be responsible for swallowing and oral occlusion disorders; therefore, it requires prompt and appropriate diagnosis and management.

Keywords: Children, congenital mucocele, glands of Blandin-Nuhn, Lingual cyst, Niger.

Congenital mucoceles are uncommon malformations, with lingual localization being particularly rare. They result from damage to the minor salivary glands, and has typically two main types: extravasation mucoceles and mucus retention in the surrounding tissues of the lamina propria.[1,2] These lesions are usually located on the lips; localization on the ventral surface of the tongue is extremely rare and occurs sporadically.[3-5]

Clinical picture of lingual mucocele depends on its location and volume. Voluminous forms, rarely associated with ventral mucocele, may push back the tongue, leading to permanent opening of the mouth and difficulties in breastfeeding, or even life-threatening obstruction of the upper respiratory tract. These forms are rarely reported in the literature.[1] In this article, we report the first case of congenital mucocele of the ventral surface of the tongue in Türkiye and discuss diagnostic and therapeutic challenges of this condition.

CASE REPORT

A 22-day-old male neonate was admitted to our department with a large swollen tongue which had been progressing since birth. He was the third sibling of a non-consanguineous couple. Vaginal delivery was performed following a normal full-term pregnancy. On his clinical examination, the patient was in a good general condition, with a renal mass occupying the ventral aspect of the tongue and progressively increasing in volume. This mass pushed the tongue upwards, preventing mouth closure and hindering breastfeeding (Figure 1). There were no inflammatory signs, and the rest of the physical examination findings were normal. Computed tomography (CT) of the face in 2-mm thin sections without and with injection of contrast medium confirmed the liquid aspect of the mass, well-circumscribed by a regular, thin, clean wall that was enhanced after injection of

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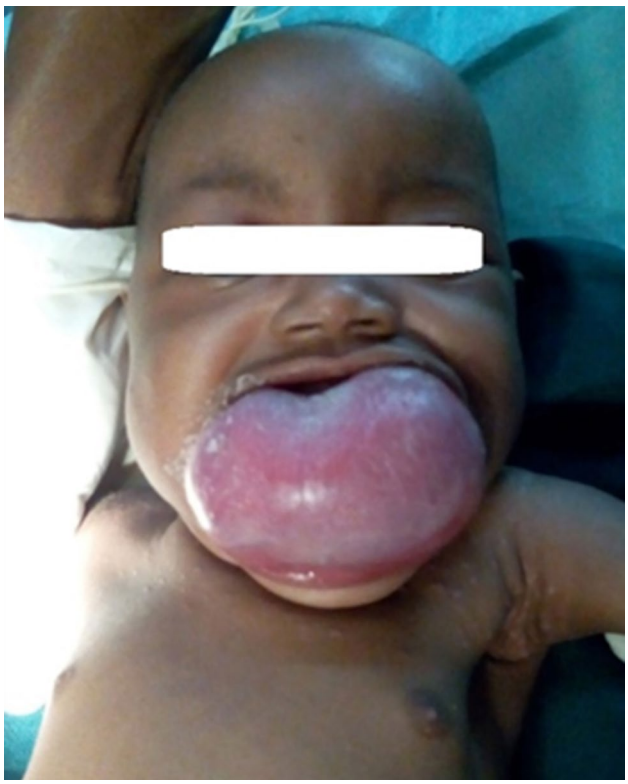

Figure 1. Clinical aspect. Newborn with macroglossia due to mucocele of the ventral surface of the tongue, preventing mouth closure and interfering with breastfeeding.

contrast medium, with no calcifications or partitions. Surgery consisted of a direct approach to the mass via a longitudinal incision on the ventral surface. After opening and aspiration of the liquid contents, the sac was dissected and resected (Figures 2 and 3). Histological examination confirmed the diagnosis of retentional mucocele. Postoperative management was uneventful, and with a 13-month follow-up, the result was satisfactory, with good oral occlusion and unproblematic suckling. A written informed consent was obtained from the parents of the patient.

DISCUSSION

Mucoceles are rare benign cystic tumors of the oral cavity, constituting the most frequent lesion observed in tumoral pathology of the oral cavity in children.[6] They develop on the minor salivary glands. In humans, there are three types of minor salivary glands on the lingual mucosa: Weber's glands lining the lateral edges of the tongue, Von Ebner's glands surrounding the circumvallate papillae, and Blandin's and Nuhn's glands located on the ventral surface of the tongue.[7] Based on their histological characteristics, mucoceles can be divided into two groups: mucoceles with mucus extravasation surrounded by granulation tissue, by far the most frequent, and retentional forms with epithelial mucosa, which account for less than 10% of mucoceles.[1,2,4,5] The latter form corresponds to our case report and is most often due to secondary obstruction of the excretory duct of the salivary gland; however, the etiology of this obstruction may also be of malformative origin (i.e., atresia of an excretory duct or channeling defect).[1,2] Mucoceles can be found ubiquitously in the oral cavity. However, the lower lip is the preferred site, particularly for extravasation mucoceles; other sites, notably the

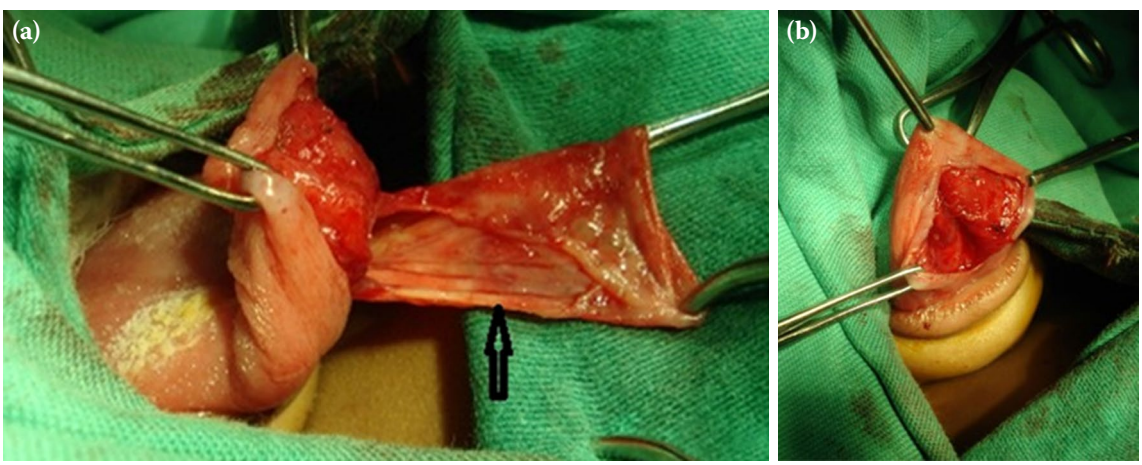

Figure 2. Intraoperative view. **(a)** Dissection of the mucocele sac after evacuation of contents. **(b)** Intraoperative appearance after complete excision of the mucocele sac.

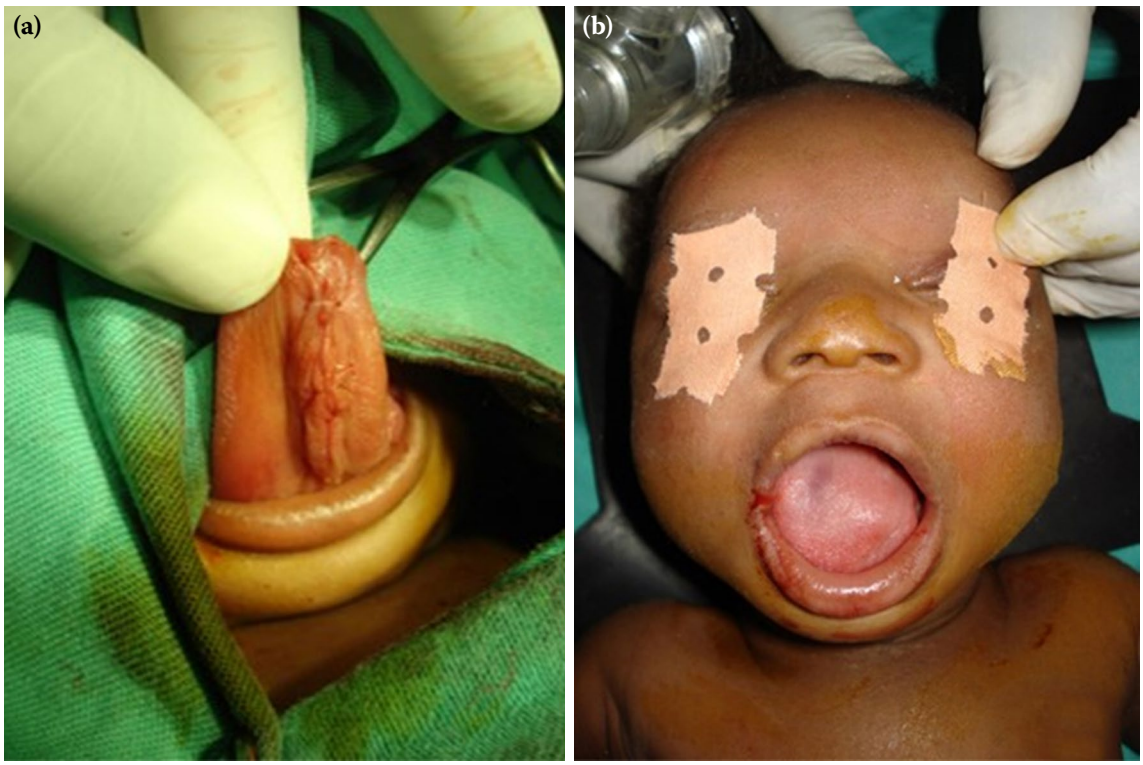

Figure 3. Postoperative view. **(a)** Sutured appearance of the ventral surface of the tongue. **(b)** Immediate postoperative appearance after complete excision of the cyst.

ventral surface of the tongue, are much rarer.[8,9] In this location, the mucocele is also known as Blandin-Nuhn mucocele.[10,11] Clinically, mucoceles are tumors, most often asymptomatic. Their clinical expression depends on their volume. In voluminous forms, these tumors can be responsible for feeding difficulties and/or airway obstruction, which can be life-threatening. In the latter case, pre-natal diagnosis is of vital importance, so that the necessary measures can be taken to ensure delivery in a facility with the human and material resources required for optimal care.[1,4] Fetal ultrasound can reveal the cystic lesion and even assess its impact on swallowing; however, fetal magnetic resonance imaging (MRI) remains the most optimal tool for antenatal diagnosis.[1] In our case, antenatal diagnosis was not made, and our case was diagnosed after birth. Congenital mucocele of the tongue presents as a swelling of variable volume, translucent and painless. It may be confused with certain cystic lesions of the ventral surface of the tongue, such as dermoid and epidermoid cysts, lymphangiomas and hemangiomas.[2,12] Diagnosis of certainty is provided by histology, which specifies mucous or serous content with a wall made of salivary-type glandular epithelium, and rules out a malignant process.[13]

The therapeutic approach to congenital mucocele depends on its volume and clinical expression. For mucocele of small volume and without any discomfort or clinical expression, some authors recommend expectoration, as cases of spontaneous resolution or rupture during feeding have been reported.[3,14,15] However, the possibility of volume increase, with the risk of obstructive complications, calls for an interventionist attitude from the moment of diagnosis for other authors. To date, several therapeutic options have been proposed for the management of congenital mucoceles including marsupialization, complete removal of the cystic mass, laser treatment, cryosurgery, sclerotherapy and needle aspiration.[1,4,7,8] The principle of this surgery is to completely resect the cyst to avoid recurrence. From this point of view, complete surgical excision of the cyst and the glandular components involved appears to be the procedure of choice.[7,14,16,17] However,

laser treatment of mucoceles in children seems to offer a number of advantages such as reduced postoperative pain, less bleeding, shorter operating time and good tolerance in children.[18,19] The cost and availability of this procedure limits the use of laser surgery, particularly in resource-limited settings such as ours. Our case benefited from a conventional surgical excision with simple after-effects, a satisfactory cosmetic result and no recurrence. Marsupialization presents a higher risk of recurrence than complete surgical removal of the cyst by conventional or laser surgery.[1,2] Cryosurgery with liquid nitrogen or carbon dioxide is a safe and effective alternative to surgical removal, particularly in preschool children.[20] Evacuatory puncture is a transitional solution in the case of voluminous forms and carries an increased risk of recurrence; Nohuz et al.[1] used it twice before delivery to reduce the risk of upper airway obstruction.

In conclusion, congenital mucocele is a rare benign tumor of the oral cavity. Its location on the ventral surface of the tongue is unusual. In its enlarged form, it can interfere with feeding and even be life-threatening due to obstruction of the upper airways; therefore, it requires prompt and appropriate diagnosis and management.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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