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### ABSTRACT

The purpose of this study was to report a case of bilateral swelling on the floor of the mouth of a 7-month-old patient. The lesion was congenital and had started to cause feed-ing problems. After the clinical diagnosis of ranula was made, the lesion was marsupialized. The obtained specimen was submitted for histopathological examination, which revealed an epithelial-lined cystic lesion. These results led to the final diagnosis of mucus retention cyst. After an 8-year follow-up period, the patient is in good general health with no recurrences.

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anula is the clinical term generally used to describe cystic lesions on the mouth's floor or the tongue's underside.<sup>1</sup> The word "ranula" derives from the Latin word "rana," which means frog, and is used to describe these lesions because some resemble a frog's underbelly and may present as a blue translucent swelling.<sup>2</sup>

Ranulas can occur in 2 forms: (1) oral or simple; and (2) plunging or cervical. A simple ranula, which is the most common form, is a collection of mucus on the mouth's floor. The plunging ranula is a lesion in which mucus pools below the mylohyoid muscle, in the cervical region, which may or may not be associated with a lesion in the mouth.<sup>3</sup> To various authors, a simple ranula can either be a mucous

retention cyst covered with epithelium or, more commonly, a pseudocyst caused by mucus extravasation<sup>1,4</sup> and lined with granulation tissue. To other authors, however, the term is more adequately used to describe the mucoceles that appear on the mouth's floor, without an epithelial lining.<sup>5</sup>

Congenital ranulas are relatively rare.<sup>6,7</sup> When they do not resolve spontaneously, leading to difficulty in feeding, breathing, or speaking, they should be surgically treated.<sup>6</sup>

The purpose of this report was to describe the case of an infant with sublingual ranula treated with marsupialization and followed up for 8 years.

# **CASE REPORT**

#### **HISTORY**

A 7-month-old female patient was taken to the Multidisciplinary Center for Oral Diseases (CEMDOB), School of Dentistry, São Paulo State University (UNESP), São José dos Campos, São Paulo, Brazil. Her parents complained that she had been born without a tongue and had difficulty suckling. The baby was delivered via caesarean section after a full-term gestation. Prenatal history provided no information relevant to the case. There was no report of trauma to the site.

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The mother reported that she had visited a pediatrician when her daughter was 1 month old because she was not able to see the child's tongue. The pediatrician referred her to an otorhinolaryngologist who, based on the diagnostic hypothesis of ranula, drained the lesion. After the procedure, according to the mother's report, the child had a fever of 40°C and was treated with a sulfa drug, that she did not remember the name or manufacturer. The lesion recurred after 3 days. The patient returned to the otorhinolaryngologist, who performed a new drainage bilaterally. The lesion, however, recurred after 1 week. Six months later, after reporting the case to his dentist, the father was told to take his daughter to the School of Dentistry of São José dos Campos, São Paulo, Brazil.

#### CLINICAL FEATURES, DIAGNOSIS, AND OUTCOME

Clinical examination revealed that the patient was in good general health. Intraoral examination revealed swelling of the mouth's floor, causing a superior and posterior displacement of the tongue (Figure 1). The oral mucosa's color was normal. Bimanual palpation revealed the presence of fluid in the submucosa, but no obvious calcification or spreading of the lesion to deeper structures. There was no pain. Based on the diagnostic hypothesis of ranula, an ultrasound of the region was ordered (Figure 2). The ultrasound showed a cystic image consistent with the clinical diagnosis. The lesion was marsupialized under local anesthesia in the outpatient clinic. During the procedure, a brown mucous fluid drained from the lesion. Histopathological examination revealed fragments of fibrous connective tissue covered with pseudostratified columnar epithelium (Figure 3) or stratified, squamous and nonkeratinized epithelium in some areas. There were areas with accumulation of mucoid material, as well as mononuclear inflammatory infiltrate. The final diagnosis was mucous retention cyst.

After 5 years, the patient returned to the Multidisciplinary Center for Oral Diseases (CEMDOB), School of Dentistry, São Paulo State University (UNESP), São José



Figure 1. Initial clinical aspect, showing bilateral swelling on the mouth's floor, with posterior displacement of the tongue.

dos Campos, São Paulo, Brazil, presenting with an opening on the mouth's floor (Figure 4). The mother reported that her daughter felt uncomfortable about it because her schoolmates kept asking what it was. The child, however, did not want corrective surgery and the mother did not authorize the intervention.

Currently, the child is now 8 years old and in good health without recurrence. Her parents remain opposed to any corrective procedure.

This case report was approved by the Research Ethics Committee of São José dos Campos School of Dentistry, São Paulo State University (UNESP), São José dos Campos, São Paulo, Brazil.

## DISCUSSION

Some theories have been proposed to explain the etiopathogenesis of the lesions generically referred to as ranulas. These theories include obstruction of a salivary duct, mainly of the sublingual gland, leading to the formation of a cyst lined with epithelium; injury to the secretory duct, leading to the formation of a pseudocyst; rupture of the acini caused by hypertension secondary to the obstruction of the main secretory duct; and traumatic destruction of the glandular parenchyma.<sup>1</sup> Congenital ranulas may be caused by atresic or imperforate ducts in the sublingual or submandibular glands<sup>6,8,10</sup> or by the duplication of the salivary duct,<sup>8</sup> leading to the formation of a true cyst.

The differential diagnosis of a swelling of the mouth's floor in neonates should include lymphatic malformations, teratoma, dermoid cyst, and thyroglossal duct cyst.<sup>9</sup> In the present case report, the clinical features and medical history ruled out diagnoses other than ranula. Nevertheless, various complementary examinations may be useful to reach a diagnosis (ie, sialography, fine needle aspiration cytology,<sup>11</sup> computed tomography,<sup>11,12</sup> magnetic resonance imaging,<sup>12,13</sup> and ultrasonography).<sup>12</sup> Some cases of congenital lesions are prenatally diagnosed, and ex utero intrapartum treatment

may be essential for airway management during labor and prevention of complications resulting from perinatal asphyxia.<sup>10,13</sup>

The treatment of ranulas includes incision and drainage, marsupialization,<sup>4,14-16</sup> micromarsupialization,<sup>17</sup> excision of the lesion with or without removal of the ipsilateral salivary gland,<sup>18,19</sup> cryosurgery, carbon dioxide laser excision,<sup>19</sup> the use of sclerosing agents (commonly OK-432),<sup>11,20</sup> and homotoxicological remedies representing the interaction between conventional medicine and classical homeopathy.<sup>21</sup> Spontaneous resolution may occur in children, and some authors have recommended a 5-month follow-up before treatment.<sup>19</sup> Steelman et al<sup>6</sup> reported a case of congenital ranula that spontaneously resolved, possibly due to rupture during feeding. Nevertheless, there is still no consensus regarding the best treatment option.19



Figure 2. Ultrasound of the area, showing a cystic lesion consistent with the clinical diagnosis of ranula.



Figure 3. Histopathological features of the specimen obtained during marsupialization. A cystic lesion is seen covered with pseudostratified columnar epithelium and a capsule of connective tissue with chronic inflammatory infiltrate.



Figure 4. Clinical aspect of the patient at 5 years old. A large opening is seen on the mouth's floor.

In the present case, the parents sought the Multidisciplinary Center for Oral Diseases (CEMDOB) after the patient had undergone 2 unsuccessful drainage treatments.

In 1988, Crysdale et al<sup>18</sup> recommended that ranulas larger than 1 cm be treated with excision of the sublingual gland due to the high recurrence rate after marsupialization. Baurmash<sup>14</sup> suggested that pseudocysts be packed with gauze after marsupialization to prevent the collapse of the granulation tissue wall, since this collapse may lead to fibrosis and hinder salivary flow, which may cause recurrence or the appearance of plunging ranulas. In 2001, Baurmash<sup>15</sup> also advised caution when considering the excision of the sublingual gland, since marsupialization is usually successful in cases of true cysts and recurrence is not usually observed. Injury to the lingual nerve<sup>19</sup> and rupture of the lesion are possible complications<sup>4</sup> during the surgical removal of ranulas and the sublingual gland. As treatment of choice, other authors have recommended marsupialization and, in cases of recurrence, the excision of both the gland and the cystic lesion.4,16 When surgically treating plunging ranulas, excising the sublingual gland is always recommended.<sup>4</sup> Some studies have reported that treatment with OK-432, a lyophilized streptococcal preparation originally developed as an immunotherapeutic agent for the treatment of cancer, is a safe and effective treatment for ranulas in children.11,20

The cystic fluid's color provides some information on the inflammation level and the lesion's duration. A clear liquid is seen when inflammation is minimal and the lesion is relatively new. On the other hand, a darker liquid, amber or brown, indicates a chronic lesion, with an inflammation level ranging from mild to severe.<sup>15</sup> In the present case, a brown cystic fluid was observed during marsupialization, which confirmed the lesion's previous manipulation.

There are cases of very large unilateral ranulas on the mouth's floor that give the false impression of being bilateral lesions. In these cases, the ranula's size causes a posterior displacement of the tongue, as well as difficulty in speaking and swallowing.<sup>17</sup> In cases of congenital ranulas caused by abnormali ties in the submandibular duct,<sup>8,9</sup> a bilateral manifestation is not the most common.<sup>8</sup> Pownell et al<sup>8</sup> successfully treated a bilateral congenital ranula with marsupialization and ductoplasty of the terminal portions of the submandibular ducts. In the present case report, it was not possible to clinically confirm whether or not there was atresia of the submandibular duct.

The patient presented with swelling of the entire mouth's floor, with superior and posterior tongue displacement, leading to difficulty in sucking and swallowing. The lesion went into remission with a single marsupialization procedure.

The correct diagnosis and timely treatment of congenital ranulas affecting the mouth's floor can improve the quality of life of both the child and parents, and may prevent breathing and feeding problems. It is extremely important to raise awareness of the dental surgeon's role in diagnosing and treating ranulas, which are often treated by medical surgeons only.

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