

## Congenital Ranula

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*This article presents 3 cases of infants with congenital ranula, a rare salivary gland pathology. Pathophysiology is discussed, differential diagnosis, and different treatment choices are explained.*

**Keywords:** congenital ranula, marsupialization, infants, children.

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### INTRODUCTION

The most common lesions causing swelling of floor of the mouth in children are mucous retention cysts, dermoid cysts, epidermal cysts, hemangiomas, cystic lymphangiomas, and thyroglossal duct cysts.<sup>1,2</sup> Mucocoeles are defined as cavities filled with mucus. When they appear in the floor of mouth, they are called ranulas because the swelling resembles the vocal sac or air sac of frogs.<sup>3</sup> There are 2 kinds of ranulas. The simple ranula is the most common, and is limited to the oral cavity; the plunging,<sup>4</sup> or cervical, ranula, which has a low frequency, involves not only the floor of the mouth but also the submandibular space beyond the mylohyoid muscle.<sup>5,6</sup>

When there is no clinical pathology in the floor of the mouth and the lesion has only a cervical presentation, diagnosis becomes difficult.<sup>7</sup>

A ranula contains saliva<sup>8</sup> with electrolytes, glucose, proteins, and enzymes. Sodium, chloride, and glucose concentrations are lower than that of blood serum, but levels of potassium are much higher. Amylase concentration is highly variable.

Determination of these substances can lead us to the diagnosis of the salivary origin of this pathology.<sup>9</sup>



Figure 1. Congenital ranula.

Mucus is the exclusive product of minor salivary glands and the most important product of the sublingual gland. The mechanism of the ranula's development is extravasation or retention. The first refers to the escape of ductal and acinous fluid into the interstitial tissue, while the less frequent mechanism is obstruction of the salivary duct, impeding the exit of saliva, producing ductal dilation and swelling of the affected region.<sup>3</sup>

The clinical appearance of the ranula is a painless, soft swelling of bluish color due to vascular congestion and cyanotic tissue at the taut surface. In cases of significant swelling of the floor of mouth, Wharton's duct flow may be altered, with obstructive symptoms and increasing submandibular gland volume.

The most common sites of origin of this lesion are the deeper walls of the sublingual gland, followed by retention cysts of the duct of Rivinus.<sup>3</sup>

The histological difference between a ranula and a mucocoele compared with a mucous retention cyst, is that the former is lined by granulation tissue, present variable swelling and vascular congestion, while retention cysts presents ductal epithelium.<sup>3</sup>

Although the clinical behavior is typically benign, there is

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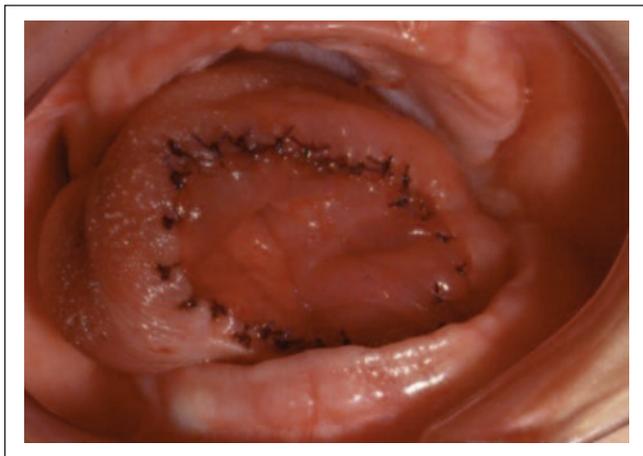
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a report of squamous cell carcinoma arising from a plunging ranula.<sup>10</sup>



**Figure 2.** Marsupialization of a congenital ranula.

Several approaches have been suggested for the treatment of oral and plunging ranulas, mainly because of their high rate of relapse.<sup>11,12</sup> Some treatments suggested by Akker *et al*<sup>13</sup> are 1) marsupialization, 2) intraoral excision of the sublingual gland, 3) marsupialization in combination with gland excision, 4) cervical excision of the ranula and the sublingual gland, and 5) radiation.

Other treatments, such as cryosurgery, laser excision, sclerotherapy, fenestration, and continuous pressure have been reported.<sup>14</sup> The use of OK-432 has been demonstrated as an effective alternative, but is not easily available and is associated to adverse effects like fever and pain in the site of injection. The use Botulinum toxin type A as a non invasive treatment option has also been described.<sup>15</sup>

Conventional marsupialization has been associated with reports of recurrence in 61% to 89% of cases. This is because wound margins tend to remain in contact due to the closed space and movement of the tongue and floor of the mouth. Multiple attempts at eliminating the ranula might cause fibrosis of the surface, and a plunging ranula can occur as a result of the salivary spill. For this reason, Crysdale *et al* recommend that large ranulas over 1 cm in diameter be treated by sublingual gland removal.<sup>16</sup>

According to Mapfumo, when excision of the ranula and the sublingual gland is performed, the rate of recurrence is zero. This is similar to the Crysdale study, which reported 0% of relapse, and the Zhao study, reporting 1.55%.<sup>14,15</sup>

Sublingual gland excision could present several risks: Blood loss due from its lingual and sublingual irrigation; lingual nerve damage, as this nerve is located close to the posterior portion of the gland before it penetrates the tongue; Wharton's duct damage; and minor complications such as hematoma and infection.<sup>15</sup>

Baurmash, in his article opposing sublingual gland removal suggest that other lesions with a cystic like appearance are capable of causing swelling of the floor of the mouth but are not related to the sublingual gland; therefore,

its resection could be an oversight. It is possible to find mucocoeles that originate from the mucous secretion of minor glands in the anterior floor of the mouth, retention cysts from Wharton's duct, and single or multiple cysts from Rivinus's ducts. The author recommends unroofing the ranula and packing the cavity with gauze for 7 to 10 days. This applies pressure to the cavity, producing an inflammatory response, consequent fibrosis that seals the leaking fluid resulting in an acinar atrophy. With this procedure, a 10% to 12% recurrence reduction has been reported.<sup>17</sup>

For the treatment of plunging ranulas, the majority of authors agree to eliminate the sublingual gland.<sup>3,6,7,17</sup> Takagi *et al* recommend a less invasive technique, draining the saliva accumulated with a 1-cm incision in the floor of the mouth and letting it drain. Then, pressure is applied to the submandibular region using a dressing for 3 weeks to diminish the pathological space and preventing saliva accumulation.<sup>18</sup>

## CASE REPORTS

### Case 1

A 2-month-old healthy infant male, with no relevant prenatal history, presented to the hospital. At day 3, his mother identified a small mass that caused lingual protrusion that grew slowly during the first month. She reported that feeding and breathing were frequently interrupted.

At clinical examination, a mild lingual protrusion was found. Elevation of the tongue revealed the floor of the mouth to be elevated on the left side, with the oral mucosa of the same color as the adjacent mucosa and slightly translucent (Figure 1). The submandibular duct could not be identified, and cervical extension was not present. A diagnosis of ranula was made and scheduled for marsupialization.

Under general anesthesia with orotracheal intubation, aspiration of dense saliva was carried out with excision of the roof of the ranula, allowing complete evacuation of the saliva. Marsupialization was accomplished suturing with silk (4-0) all borders of the lesion. Simple silk sutures (2-0) were placed on all 4 sides of the lesion (Figure 2). The surgery was accomplished without complications. 1 day after the procedure the patient recovered favorably and was discharged. Feeding and the clinical aspect improved immediately. Follow-up was done weekly for 1 month, then monthly for 3 months. Any remaining sutures were removed allowing epithelialization around the silk to create new salivary ducts. Epithelialization of the multiple miniholes created by the suture allowed the exit of the salivary fluid into the oral cavity.

### Case 2

A 2 month old female without prenatal background was brought in for consultation for an elevation in the floor of the mouth. Patient had an uneventful birth.

A submandibular mass was identified during the second week, but was not until the end of the first month that an elevation was palpated in the floor of the mouth.



Figure 3. Intraoral aspect (Case 2).



Figure 5. Preoperative view (Case 3).

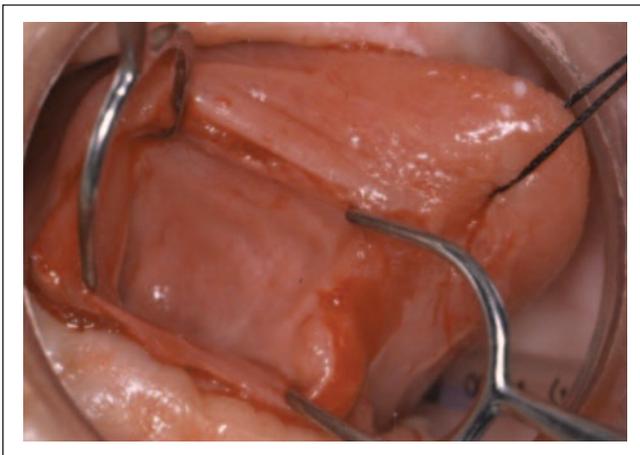


Figure 4. Intraoral aspect during surgery (Case 2).

Saliva was easily aspirated and marsupialization under general anesthesia was accomplished (Figure 4). A double suture, absorbable (Vicryl 3-0) internally and a simple silk suture (4-0) externally were used. After 3 months follow-up, the patient was discharged without relapse.

**Case 3**

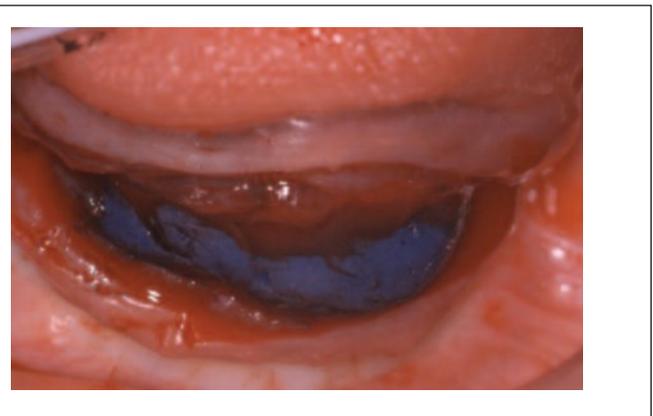
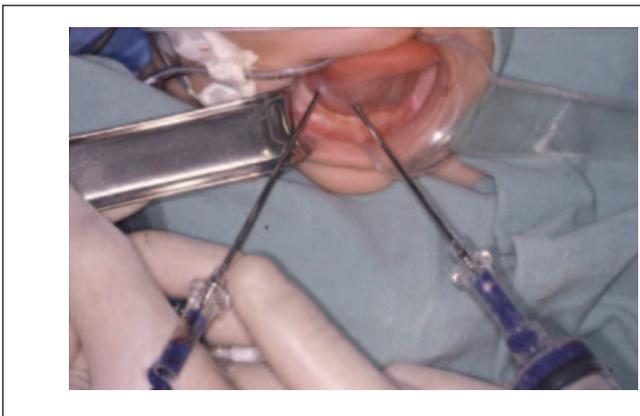
In this case, the parents identified the lesion during

infant's first week. Clinically, the ranula presented a bilateral aspect (Figure 5). When the patient turned 1 month old, he was admitted for surgery.

Its management was different from that of the 2 previous cases. Before exposing the ranula and at the time of saliva aspiration, alginate was infiltrated slowly through a catheter, such that the alginate displaced the saliva and delimited the size and relationships of the lesion (Figures 6 and 7). A few minutes after the material had set, the roof of the ranula was excised, so that suturing (with resorbable Vicryl and silk (3-0)) the inner epithelium to the oral mucosa became easier. The postoperative period was uneventful, and 3 months later the patient was discharged (Figure 8).

**DISCUSSION**

The etiology and pathophysiology of congenital ranula is not clear.<sup>16</sup> The 2 main theories are 1) mucous extravasation covered by inflammation and connective tissue and 2) mucous retention as a result of ductal atresia or acinus dilatation, covered by epithelium.<sup>19,20</sup> Sometimes there is no evidence of mucous extravasation, so a direct association with the sublingual gland cannot be found,<sup>2,3</sup> nor should traumatic origin be ruled out, especially in patients with a history of dyspnea at birth that required aspiration and intubation.<sup>21</sup> Some authors succeeded in causing acinary degeneration and



Figures 6 and 7: Alginate injection at time of saliva aspiration. Surgical view of the material (Case 3).



Figure 8. Postoperative view (Case 3).

atrophy<sup>21</sup> while others obtained injuries similar to mucoceles.<sup>22</sup> Despite those efforts, Zhao *et al* found a history of trauma in the floor of the mouth in only 2.7% of patients with ranulas.

Congenital ranula appears clinically as a fluctuant, painless, elevation in the floor of the mouth causing the tongue to move upward and forward and whose mucosa may appear from white to violet. Presentation may be unilateral or bilateral.<sup>22</sup> Identifying the salivary duct and verifying its permeability is not always possible. During aspiration, a large quantity of saliva can be obtained, usually more viscous than usual.

Marsupialization can be done as a simple and rapid procedure. Silk suture allows epithelialization around the material, so the suture should be left in place as long as possible. Recovery is fast and the patient should be followed up on an ambulatory basis.

Differential diagnosis should always include lymphangioma with cervical extension (cystic hygroma), floor of the mouth infection, and dermoid cyst. Lymphangiomas are multilocular and are lined with epithelium having lymphatic content. They are usually infiltrative, occupying adjacent aponeurotic spaces, including nerves, muscles, and blood vessels.

Fifty percent of all hygromas can be identified at birth, and almost 90% can be identified by the second year. They are usually compressible, painless, and sometimes translucent. They can grow quickly after upper airway infections. Complete surgical excision is the treatment of choice.<sup>23</sup>

## CONCLUSIONS

Congenital ranula is an uncommon lesion requiring early diagnosis and treatment to improve the patient's feeding and airway. Marsupialization is the treatment of choice because it is a simple procedure, that avoids any important anatomic structures in the floor of mouth.

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