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Surgical treatment of plunging ranula: Report of three cases and review of literature

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ABSTRACT

Introduction: Plunging ranulas arise when a simple ranula extends beyond the floor of the mouth into the neck. These cysts usually arise from the sublingual salivary gland and rarely from the submandibular gland. They are either the result of mucus retention or they represent a mucus escape reaction occurring from disruption of the sublingual duct because of local trauma. Two variants have been described in the literature: a simple oral ranula and the deep diving or plunging ranula. A number of different modalities have been described for the treatment of ranulas and especially for plunging ranula.

Purpose: The aim of this study is to present the experience of surgical management in three cases of patients with plunging ranula using an extra-oral extension. In addition, clinical and radiographic findings of the patients along with the relevant review of the literature also refered.

Cases reports: Three cases of patients with specific diagnosis of plunging ranulas were treated by a cervical approach and excision of the cyst without any intraoral extension. Data of patients were recorded including gender and age, location of lesion and possible treatment.

Results: All cysts were ranulas and the cases presented indicate that this lesion can be managed by a less invasive procedure without complications and recurrence. At the first patient ranula grow to a large size and the treatment became by removal of the ipsilateral sublingual and submandibular glands, while at second and third patient by removal of sublingual gland only.

Conclusion: There is a consensus about the appropriate treatment of ranula. The best treatment for a plunging ranula is excision of the lesion along with the involved gland (usually sublingual gland).

1. Introduction

The name “ranula” is used to describe an extravasation cyst which is founds in the floor of the mouth. This lesion resembles the bulging underbelly of a frog and is derived from the Latin word “rana” meaning “underbelly of frog”. The most usual type which appears in the oral cavity is the mucous cyst and ranula is a mucus filled cavity, a formation of mucocele which specifically occurs in the floor of the mouth in relation with the ducts of the sublingual or submandibular gland [1,2]. Ranula is a retention cyst arising mainly from sublingual gland, which enlarges progressively and extends into the surrounding soft tissues [3]. It can be classified in two varieties: an oral or superficial ranula which is the most common type and a plunging or cervical ranula. Both varieties appear with different clinical behavior [1].

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Plunging ranula is known as diving, cervical or deep ranula and usually appears in conjunction with an oral ranula. It is a cystic extravasation mucocele that arise from the sublingual gland and usually from a torn duct of Rivinus [3,4]. The plunging ranula develops from extravasation of mucus saliva after trauma or obstruction of the associated sublingual or rarely submandibular salivary duct. Fluid from the obstructed gland penetrates between the fascial planes and muscle of the tongue's base to the submandibular space [1,5]. Cervical ranula has a prevalence rate of 0.2–0,9 cases for every thousand people and represent 6% of all oral sialocysts [4,6].

For the treatment of plunging ranulas a variety of surgical procedures have been described in the literature ranging from surgical removal of the ranula with or without exclusion of involved salivary gland. Despite invasive surgical procedures ranula has a tendency to recur [5,7].

2. Cases Reports

2.1. Case 1

A 42 year-old male patient reported to our center with a chronic swelling between left submandibular and sublingual region, which developed from both salivary glands (Fig. 1). The swelling was asymptomatic and the patient was in good general health without any history of systemic disorder. The patient reported that he had previously underwent surgical treatment through two unsuccessful intraoral procedures for this lesion, which was diagnosed as a ranula. Mastication and swallowing of the patient was difficult due to the large size of the swelling (which almost was 2,5 × 2 cm in size) and tongue movements were obstructed. On examination a soft diffuse swelling was present between left sublingual and submandibular region. A provisional diagnosis of a plunging ranula was made and the lesion properly differentiated from other swellings of the anterior neck region, as dermoid and epidermoid cyst. Axonic tomography (CT) showed a fluid cavity in left sublingual space extending to the left submandibular space beside mylohyoid muscle (Fig. 2).

Based on clinical and CT findings, treatment was planned for excision of the lesion with the involved sublingual and submandibular glands. Excision of the mass was carried out under general anaesthesia via an extra-oral submandibular incision. The lesion was located above the mylohyoid muscle and extended to the sublingual and submandibular spaces. Successful treatment was achieved by removal of the ipsilateral sublingual and submandibular glands with preservation of the marginal mandibular branch of the facial nerve (Fig. 3). The specimen was sent for histopathological examination, which confirmed the clinical diagnosis of a plunging ranula. The patient is free of the lesion after 4 years post-op.

2.2. Case 2

A 45 year-old female patient was referred to our department with a swelling in left sublingual region. The lesion arised from the sublingual salivary gland and extended through the floor of the mouth. The patient reported that the swelling enlarged during the last months. A CT scan showed a cystic lesion on the left side of the neck involving the floor of the mouth in the sublingual region and extending from chin to the hyoid bone and into the submental region (Fig. 4). The size of the lesion upon examination was approximately 2 × 2 cm and probably existed an ectopic sublingual salivary gland inferiorly mylohyoid muscle. A provisional diagnosis of plunging ranula was made.

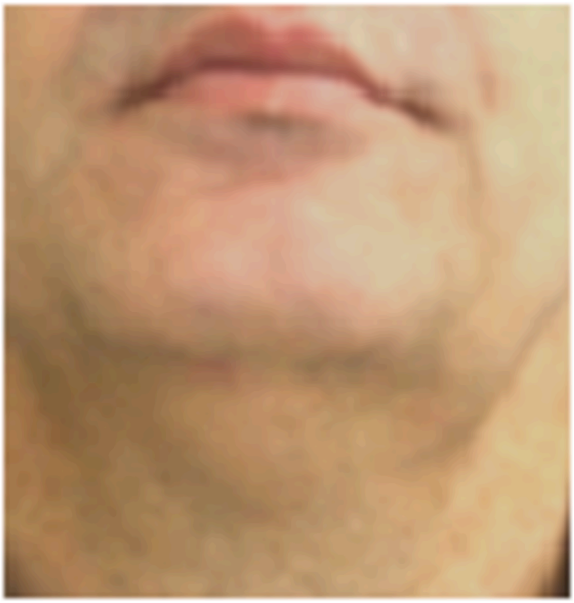

Fig. 1. Extra-oral view showing left sublingual and submandibular swelling.

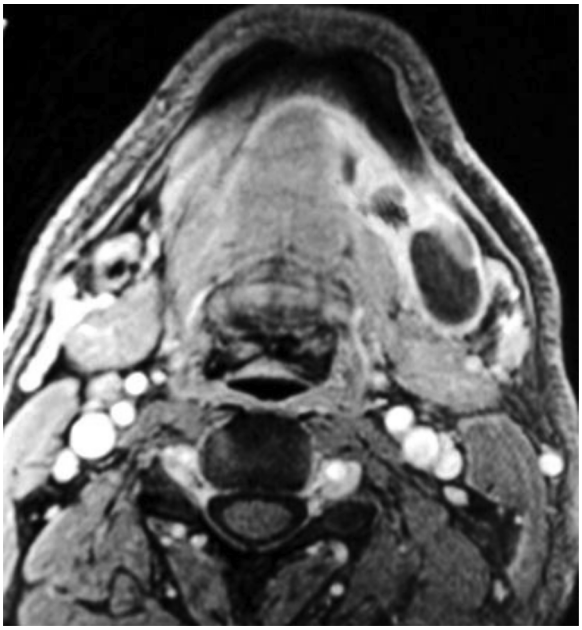

Fig. 2. CT showing the fluid cavity in left sublingual space extending to submandibular space.

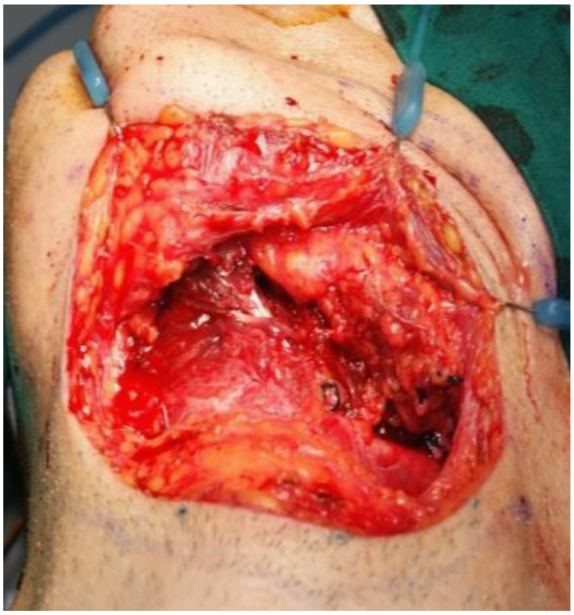

Fig. 3. Peri-operative excision of the ranula through an extra-oral. submandibular approach with both sublingual and submandibular salivary glands.

Following detailed clinical examination and radiological interpretation, surgical excision was performed along with the involved sublingual gland. During surgery, drainage of the extravasation cyst was done and the surgical treatment took place using an extra-oral approach under general anaesthesia. Removal of the ipsilateral sublingual gland was also done via a submental incision (Fig. 5) and all the tissue was subjected to histopathological evaluation. The excised mass confirmed the diagnosis of a plunging ranula in a mucous extravasation cyst.

2.3. Case 3

A 42 year old male patient reported to our clinic with a swelling in right sublingual region (Fig. 6). Initial diagnosis of a plunging

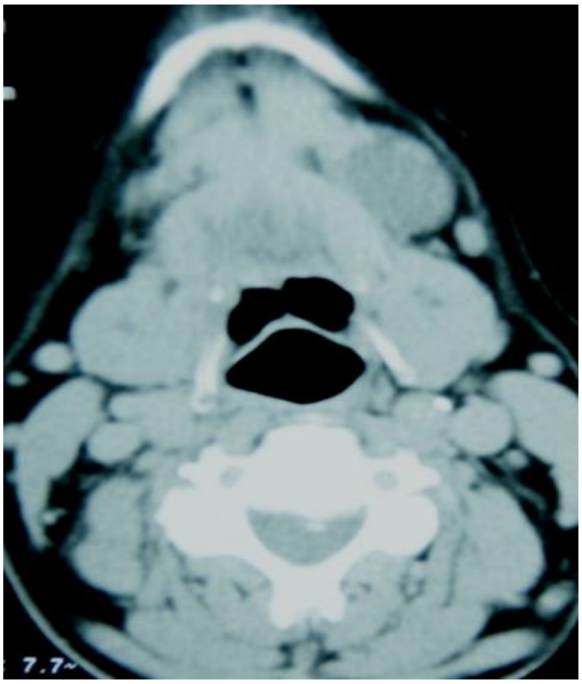

Fig. 4. Imaging presentation (CT) showing a cystic mass lesion in left. sublingual space (measured 2 × 2 cm in midline).

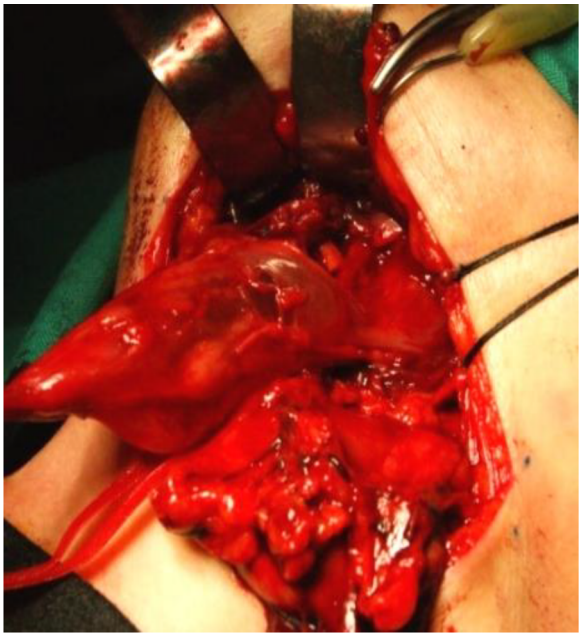

Fig. 5. Peri-operative presentation of ranula's excision with sublingual salivary gland.

ranula was made on the basis of clinical findings. Magnetic resonance imaging (MRI) showed a cystic lesion in the right side of the sublingual region and the size of the lesion was approximately 3,5 × 2,5 cm (Fig. 7). On examination the patient was asymptomatic.

Based on the clinical and MRI findings, surgical treatment was planned via an extra-oral submandibular and sublingual approach under general anaesthesia (cyst didn't approach transoral because the size was rather large and extended beyond sublingual space). Excision of the lesion was performed along with the associated sublingual gland via a submental and submandibular incision (Fig. 8). Closure of surgical trauma was done in layers and the surgical specimen was sent for histopathological examination, which was consistent with the clinical diagnosis of a plunging ranula. There was no sign of recurrence and the patient is free of the lesion after 4

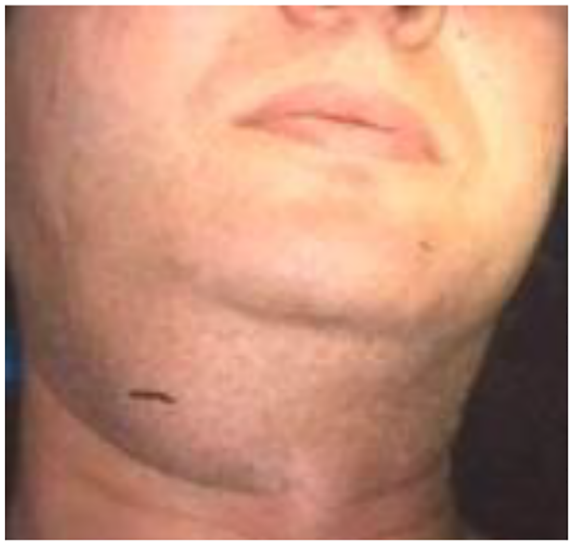

Fig. 6. Extra-oral view showing a large swelling in the right side of the sublingual region.

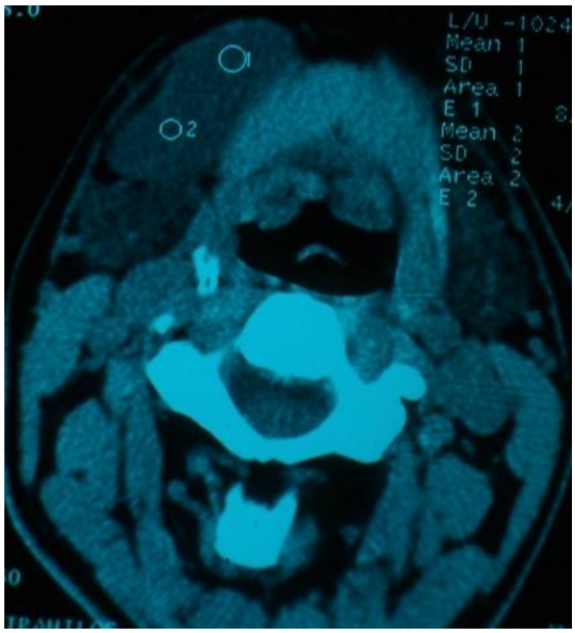

Fig. 7. Pre-operative MRI showing a cystic lesion in right sublingual region.

year of follow-up.

3. Discussion

Ranula is an extravasation cyst and may develops from extravasation of mucus after trauma to the sublingual gland and rarely submandibular gland or obstruction of salivary ducts [1,8]. Many theories for the origin and pathogenesis of these cysts have been postulated by Hippocrates, who described that ranulas are the effect of local inflammation of sublingual salivary gland [4]. The lesion forms due to extravasation of mucus and subsequent formation of a pseudocyst [3]. Another theory for this anatomical lesion is inflammation causing atrophy and mucous degeneration of the sublingual gland [4].

Pathogenesis of ranula is associated with sublingual and rarely the submandibular gland and mainly appears in the floor of one side of the oral cavity or rarely bilaterally [1,2,5]. Ranulas fluid is clear and the swelling which are caused is soft [9]. It is characteristically large (>2 cm) and most reported ranulas are 4–10 cm in size and rarely bigger than 10 cm [3]. When ranula become larger, it acquire a

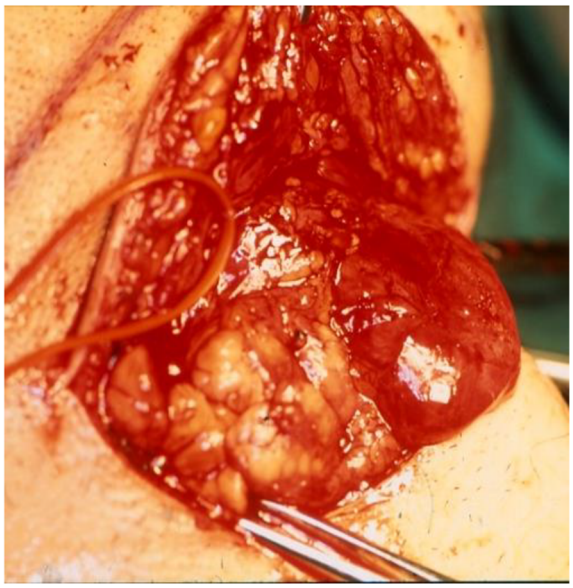

Fig. 8. The ranula with associated sublingual salivary gland.

blue colour and resemble frog's belly Big sized ranulas may cause deviation of the tongue with associated difficulties in speech and mastication [4,6,10]. They can present at any age (usually occur in children and young adults), but they usually exist since birth. Presentation most frequent in the second and third decades of life and the reported male to female ratio is 1:1.3 [10–12]. Ranula has been described in association with congenital anomalies, such as duct agenesis, hypoplasia of the sublingual gland and trauma causing direct damage to the sublingual gland [5,9]. The most common factor is that trauma causes direct damage to the duct of the sublingual gland [7,9]. However, different studies - which took place in cats - accounted that ranula may be created after ligation of the sublingual salivary duct [2].

Superficial ranulas arise more often in the left side of the oral cavity, while plunging ranulas are located more often in the right side [13]. Plunging ranula develop from the base of the sublingual salivary gland, when a superficial ranula extends beyond the floor of the mouth and penetrate through mylohyoid and genioglossal muscle of the submandibular or submental space into the neck in the cervical connective tissue. Sometimes, mylohyoid muscle isn't a barrier between the sublingual and submandibular spaces and sublingual gland may be located through mylohyoid muscle beyond the sublingual space and also beyond mandibular muscle forward the lingual nerve and submandibular salivary gland [14, 15]. Another explanation to the creation of plunging ranula may be the anatomical variation of an ectopic sublingual salivary gland inferiorly to the mylohyoid muscle. Therefore, some ranulas extend through the mylohyoid [4]. When a duct from the sublingual gland join the submandibular gland or its duct allow ranula to form in continuity with the submandibular gland [6]. Generally, plunging ranula may be possible to arise as a result of extravasation of saliva from the sublingual gland in the mylohyoid muscle through a hiatus. This herniation is estImated to be about 36%–45% of the general population. Cervical ranula appears as an asymptomatic lesion (associated with oral swelling in 34% of cases), but may cause dysphagia or airway obstruction (Toru [3,14,15]. When plunging ranulas extend forward to the mylohyoid muscle and hyoid bone, they appear as submandibular, parapharyngeal or whiplash swellings [4].

Diagnosis of plunging ranula is difficult even with modern imaging techniques, as mimic other neck lesions. It is usually determined by a combination of history, clinical examination, imaging studies and many times after suction of saliva with a syringe (FNA) [3,4]. Sialography, ultrasonography, computed tomography and magnetic resonance may also be of assistance [11,16]; Toru [8,15]; R [17]. The differential diagnosis of plunging (cervical) ranula is made among some other neck swellings and especially cystic hygroma [12]. Ranulas must also be differentiated from branchial cleft cysts, thyroglossal duct cysts, dermoid cyst, cystic or neoplastic thyroid diseases, parathyroid or cervical thymus cyst, laryngocele, lipoma, submandibular sialadenitis, tumours of the salivary glands (e.g pleomorphic adenoma) and infections cervical lymphadenopathy [3,8,18]. Also from intramuscular hemangioma of floor of the tongue and benign teratoma [19]. Differential diagnosis from these neck swellings is difficult without CT scan [3,4,20]. For example, a case of squamous cell carcinoma in the wall of a ranula arising from a sublingual salivary gland has been reported [21]. Generally, squamous cell carcinoma with arising from the cyst wall and papillary cystadenocarcinoma of a sublingual gland maybe present as ranula [16, 22].

Biochemical analysis of fluid from ranula shows that there is a high amylase and protein content [3,5,7]. Saliva concentration in the sublingual gland includes amylase in a percent which is the same as with blood's serum, while into the submandibular salivary gland is higher percent than in blood's serum. Thus, the chemical analysis of fluid may assist to the ranula confirmation [1].

Histopathologically the plunging (cervical) ranula appears identical to the mucus extravasation and reveals a connective tissue with inflammatory cells. It is a fibrous connective soft tissue cyst and above it there is squamous or circular epithelium [7,14]. Many researchers detail that ranulas are surrounded from thick and fibrous boll and report them as pseudocysts. Plunging ranulas may

appear with this histological picture [7,9].

Treatment of ranula accomplish through different surgical techniques and traditional treatment involves enucleation of the cyst from the neck [8,17,18]. Despite this invasive surgery ranula tends to recur. Recurrence of the lesion is still a problem [5]; R [17]. However, surgery remains the main treatment modality of ranula and is usually achieved with the following methods:

- Surgical removal of the lesion and extra-oral also exclusion of the involved salivary gland [7, 28, 32] with or without biopsy of cystic wall [5,23].
- Removal only of the cyst with an extra-oral approach and ligating the branch of sublingual or submandibular gland duct [3,16].
- Endoscopic excision of the involved salivary gland only [11,24].
- Transoral approach with excision of sublingual gland alone could be the least invasive approach with minor complications, because there is no risk of damage to the marginal mandibular branch of the facial nerve, no cutaneous scarring and the submandibular gland is not injuring [25,26].
- Marsupialization with or without intra- or extra-oral excision of the involved salivary gland (mainly used for superficial ranulas) [27,28]. Marsupialization with packing of the cyst cavity may reduce the recurrence [5].
- Drainage and marsupialization of the lesion without excision of the involved salivary gland provides a high success rate with minimal complications and indicates when ranula has diameter lower than 2 cm and the transoral approach is the treatment of choice [25,27].
- Ranula have also been managed by cryosurgery and electrosurgery [12,27,28].
- Simple ranula have also been excisioned by laser (CO_2 and ER-YAG) [13,29].
- For pseudocyst of plunging ranula the treatment is OK-432 and sclerotherapy [30].
- Radiation therapy may be used in rare cases of deeply plunging ranula [6].

Generally, ranula treatment is achieved by the removal of the cyst along with the sublingual salivary gland and rarely the submandibular gland [3,28], as went in our cases. This treatment is good for surgery, but may create risks for paresis and paralysis of marginal mandibular nerve [5,30]. Drainage and marsupialization of the lesion via a transoral approach with or without cauterization, cryosurgery e. t.c. using as substitute operation technique [3,25,27,28]. Marsupialization is the most conservative method of surgical treatment for many surgeons [1,27,31], but recurrences occurring after marsupialization are usually located behind the floor of the mouth. In our days the better technique is removal of the ranula and excision of the involved salivary gland via a cervical approach in order to avoid subsequent recurrence from residual lingual tissue remaining, as many researchers report [5,8,28,31,32]. The tactic of surgically removing the associated salivary gland seems to account for almost zero recurrence, as reported in the literature [6,9,32].

4. Conclusion

The correct selection of surgical treatment plays an important active role. The surgical removal of the associated salivary gland, usually the sublingual, via a cervical approach is the management of choice for ranula and is a common treatment (especially for us, as we indicated). Removal of the sublingual gland must be the preferred treatment when there is history of recurrence or trauma and follow-up is required.

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