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CASE REPORT

Plunging ranula: surgical management of case series and the literature review

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Key Clinical Message

Plunging ranulas are rare; report of this condition is particularly limited in our environment. We present case series in children; with all cases having both oral and cervical components. It is important to note this type of presentation of plunging ranula and their appropriate management.

Keywords

Cosmetic dentistry, developmental biology, dysphagia, growth/development, imaging, maxillofacial surgery.

Introduction

Plunging ranula (PR) otherwise known as cervical ranula is a nonepithelial-lined salivary gland cyst that forms following mucus escape from sublingual gland and its subsequent herniation via the mylohyoid muscle into submandibular space and beyond. Although the precise prevalence of this condition remains unknown; they are nonetheless rare and surgical management is the first-choice therapy for them [1] . This report presents clinical findings for four cases of PR, their surgical management and review relevant literature.

Case Series Report

A summary of the four case series is shown in Table 1. In addition, medical, surgical, and family histories revealed no significant findings. Although investigations such as computer tomography (CT) scan and magnetic resonance imaging (MRI) could not be performed due to financial constraints, radiographs showed no involvement of the hard tissues. Diagnosis of PR was made based on the clinical and ultrasound reports. The associated sublingual gland was excised under general anesthesia via a transcervical approach for cases 1, 2, and 3 while an intraoral approach was employed for case 4. The accumulated saliva collection was evacuated during the surgery for all the cases. Postoperative histopathologic report in all cases indicated a finding consistent with extravasated mucocoele (PR). Patients are being followed up but with no sign of recurrence so far (Figs. 1–3).

Literature Review

A ranula is a swelling in the floor of the mouth caused by leakage of mucus from the sublingual or submandibular salivary gland. The word was coined from *rana*; a Latin word meaning frog because the typical swelling in the floor of mouth resembles the underbelly of a frog. Ranulas are mostly mucous

Table 1. Summary of four cases of plunging ranula

Case	Age (years)	Sex	Presentation	Clinical features	Ultrasonographic findings	Recurrence	Follow-up
Case 1	3	Female	Painless left submandibular and floor of mouth mass of 8-month duration (Fig. 1). Associated with difficulty in feeding	Diffused, soft to fluctuant, and nontender mass 6 cm by 8 cm in midline floor of mouth, raised tongues	Firmly circumscribed hypoechogenic, anterior neck, size 82 mm by 91 mm by 76 mm, uniform internal echotexture no mixed-internal echoes	No	3 years
Case 2	6	Female	Painless left submandibular mass, 1-year duration, and accompanying floor of mouth swelling of 6-month duration (Fig. 2)	Diffused, soft to fluctuant, and nontender swelling measures; 7 cm by 8 cm	Positive for plunging ranula	No	6 months
Case 3	4	Male	Painless floor of mouth swelling on and off from birth. Four years later with cervical component	Well-circumscribed fluctuant swelling in floor of mouth, 3.5 cm by 3 mm in size. Tongue is raised. Concomitant fluctuant cervical mass 4.5 cm by 4 mm in size	Positive for plunging ranula Aspiration of both oral and cervical swellings yielded a viscous straw colored fluid	No	4 months
Case 4	10	Female	Three-month duration painless swelling in left submandibular region accompanied by floor of mouth swelling (Fig. 3), has affected speech	Centrally located sublingual soft mass, bluish hue and thick overlying mucosa, elevating the tongue	Positive for plunging ranula	No	6 months

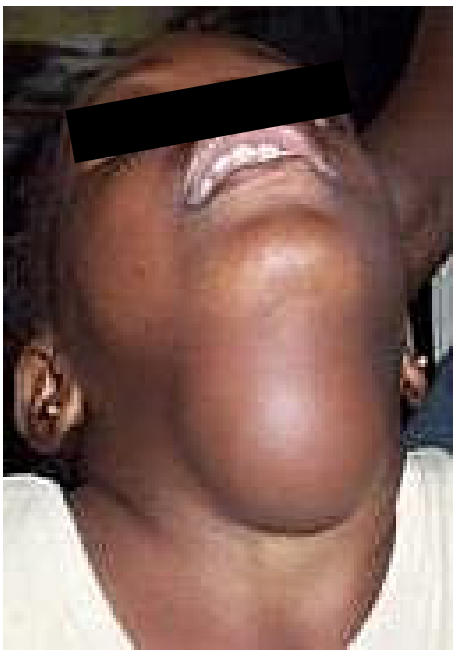

Figure 1. Picture of a 3-year-old girl with 6-month history of painless swelling in floor of month and left submandibular region.

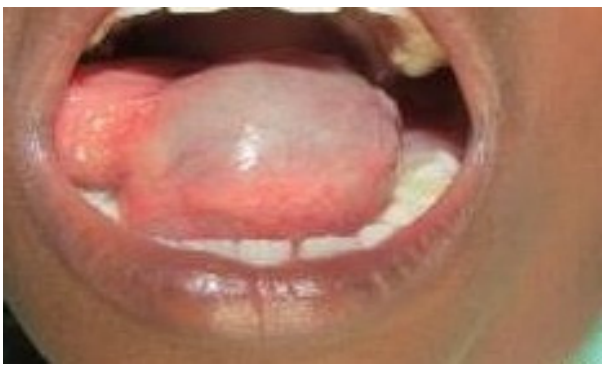

Figure 2. Picture of a 6-year-old girl with 1-year history of painless swelling on the left submandibular region; this was accompanied by floor of the mouth swelling of 6-month duration.

extravasation cysts of the sublingual gland with spillage of mucus into the submandibular space and/or adjacent structures [2]. They are anatomically grouped into three different types which are as follows: superficial or oral ranula, which occurs above the mylohyoid muscle, plunging, or cervical ranula which is found

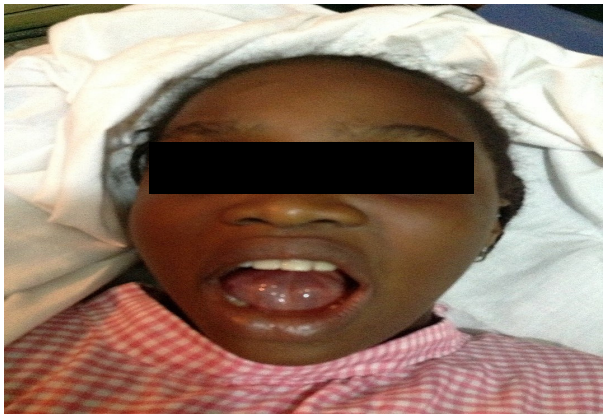

Figure 3. Picture of a 10-year-old girl with 3-month history of painless swelling on the left submandibular region accompanied by floor of the mouth swelling.

beneath the mylohyoid muscle, and mixed type which has both an oral and a cervical component [3]

The prevalence of ranula is around 0.2 cases of every 1000 individuals, accounting for 6% of intraoral cysts of the salivary glands [1]. Age range of patients is from 3 years to 61 years with children and young adults in the second decade and third decades of life been more are affected than others [2].

The etiology of this lesion can be described by either of two processes: the foremost is by incomplete blockage of the duct of a sublingual gland; this leads to the development of a true retention cyst lined by epithelium. About 10% of all ranulas develop in this manner demonstrated experimentally by ref. [4]. Secondly, the duct or deeper areas of body of the sublingual gland may become damaged by direct trauma thus initiating mucus to escape into the surrounding area with subsequent formation of a nonepithelial-lined cyst [1, 5, 6]. In the way, an obstructed duct secondary to trauma may cause back pressure to build up if severe enough this will rupture the acini and lead to mucus spillage. In contrast, submandibular and parotid ducts will not form ranula from ligation because they only secrete saliva when stimulated, unlike the sublingual that constantly discharges saliva even during the interdigestive phase.

The ethnic variation in incidence of PR has been documented by some studies, where it was noted to be generally more prevalent in people of Asian origin especially the Chinese, natives of New Zealand and Eskimos. This racial predilection could be attributed to a genetic alteration in people of this race, perhaps predisposing them to either develop areas of deficiencies within the mylohyoid muscle or an anatomically altered variant of the sublingual gland with easy extravasation of mucus following minor trauma [7, 8].

The most common clinical presentation of a ranula is as a blue-colored swelling located beneath the tongue, which may elevate it. On palpation, ranula is fluctuant, freely movable, and nontender with most reported cases measuring about 4–10 cm. The submandibular space is their usual location, and they are covered by an intact overlying skin [4].

Plunging ranula a non-epitheliallined cyst that forms when mucus escapes from the sublingual gland and via the mylohyoid muscle herniates into the submandibular space and beyond [9, 10]. Other names for this condition are as follows: cervical ranula, diving ranula, deep ranula, and oral ranula with cervical extension [11]. Most PRs have oral components and four mechanisms have been described by which they may arise. The first claims that part of the sublingual gland may move through the mylohyoid muscle, or an aberrant sublingual gland may be present below this muscle; this mechanism describes cases that without an oral part. On the other hand, the lateral part of the anterior two-thirds of the mylohyoid muscle may have a deficiency through which mucus from the sublingual gland may slip to the submandibular area. Deficiency in anterior two-thirds of mylohyoid muscles has been reported in about 27–45% of cadavers; these sites create a path through which tissues may pass through the muscle. Surgical manipulation during the removal of a ranula may mutilate and block its superior surface, and should such recur, a cervical ranula develops as this new lesion slips through the only available space which is the deficient area in the mylohyoid muscle. In addition, the sublingual gland duct may join that of the submandibular gland, thus a ranula may form in this line between the two glands. Plunging ranulas are rare compared to ranulas and only about 100 cases have been described in the English literature [11].

Presently, PR is known to develop from escape of mucus saliva from the sublingual gland and retention of this mucus and its extension into neighboring tissues [6].

The cervical ranula clinically presents as an asymptomatic, progressively expanding neck mass that may or may not have an intraoral component [12]. When an oral component is present, squeezing the neck mass may increase the size of swelling in floor of mouth. Plunging ranulas have been found to spread into the submental area, the contralateral side of neck, the nasopharyngeal region, including the skull of base, retropharynx, and sometimes to the upper mediastinum [1, 11].

Imaging studies to investigate PRs include the following: CT scan and MRI. The classical CT scan finding for PR is a lesion with a small tail that extends into the sublingual space [13]. In the absence of this sign, the diagnosis of a PR should be made if an identical cyst is seen in

the submandibular or parapharyngeal space with connection to the sublingual space [11]. Ultrasonography has been demonstrated to be quite successful in evaluating cystic lesions of submandibular region in young people, with particular utility for PR. This high-resolution ultrasound is a noninvasive test with no known biologic cost [13]. The status of the mylohyoid muscle can also be established by this test [4, 11]. This technique will also differentiate PRs that have plunged behind the posterior border of mylohyoid into the submental space and the less common ranula that has herniated through a mylohyoid muscle defect into submental space. The use of ultrasound suffices to investigate these lesions has been recommended as the referred test [12].

Lesions that may present as neck swelling and are thus differential diagnosis of PR are lymph node enlargement, abscesses, laryngocele, thyroglossal duct cyst, dermal cysts, cystic hygroma, thymic cyst, cysts of endocrine glands of the neck, benign salivary gland tumors, and some benign mesenchymal tumors [4, 9, 11, 14, 15].

Histologic appearance of a PR is typically that of a pseudocyst without epithelial lining, it consists of a fibrovascular stroma which contains chronic inflammatory cells including macrophages filled with mucin [4]. Histiocytes or foamy macrophages predominate in the pseudocyst wall and mucin is also often observed. Occasionally, partial epithelial linings are seen. High amylase and protein are seen on biochemical analysis of the cystic fluid [1].

Many methods have been used for the treatment of cervical ranulas and this includes surgical removal of lesion, use of cryosurgery, marsupialization, excision of oral portion and associated sublingual gland or, rarely, submandibular gland. Others are excision of sublingual gland via intraoral approach, and drainage of lesion, and use of cervical approach to excise the lesion sometimes this is combined with excision of sublingual gland [1]. Recurrences have been known to occur even after these treatments [6]. However, when the sublingual gland together with the associated lesion is excised, a low recurrence rate is recorded [16, 17].

The oldest and most widely used therapy for oral ranula is marsupialization; this involves the removal of the roof of the cyst and attachment of its borders to surrounding tissues. This procedure recorded a very high recurrence rate of 61–89% within 6 weeks to 12 months of surgery. Early closure of the surgery site due to compression of the tongue on the cyst has been known to increase the recurrence rate. Therefore, packing the cavity with gauze for 7–10 days reduces the recurrence rate. Marsupialization and filling the cavity with gauze improves the success rate. Micromarsupialization involves the placement of a silk suture (Seton) for a minimum of 7 days during which an epithelial tract forms to allow for mucus drainage between the surface and the underlying salivary glandular tissue. This procedure is simple with minimal to nonexistent minimal to nonexistent; however, its recurrence or treatment failure is the primary complication [11].

Although considered experimental, sclerosing agents have been employed in the treatment of PR [18, 19]. Bleomycin and OK-432 have been successfully used to manage ranulas [19]. Even so, OK-432 has been shown to collapse and cause adhesion of the pseudocyst wall of a PR [18]. In a study of 32 cases of PR, 31 (97%) achieved a marked decrease in size of lesion with injection of OK-432. While about 50% of all cases experienced local pain or fever, this, however, resolved after some days [11]. Rho et al. thus advocate that sclerotherapy is a safe and potentially curative procedure that may be used as a primary treatment before considering surgery for PR.

Carbon dioxide laser has also been used as a treatment options for PR; this has shown good success with a decrease in recurrence rate [20]. In very few patients who cannot endure the stress of surgery, a worthwhile substitute would be the used of radiotherapy, and low doses, from 20–25 grays (Gy), have been known to be effective. The attendant complications of radiotherapy especially xerostomia can be circumvented using of low doses and protecting the contralateral parotid gland with a shield. The danger of malignancies developing as a result of treatment with radiation is very low [5].

In the management of PR, the sublingual gland could be excised either be via the intraoral or the transcervical technique. Excision of the gland through the intraoral method and drainage of the associated cervical content is enough to give cure and is presently considered as the treatment of choice [6]. With the transcervical approach, total excision of the sublingual gland is challenging because this method requires the painstaking division of the mylohyoid muscle in the floor of mouth [11]. However, this method is recommended for review cases and when the PR is large in size [21].

Removal of the gland by a transoral approach after drainage of the cyst has also been advocated. Should this method fail, then the cyst should be excised via a transcervical approach. Cases restricted to the neck are also best treated by the transcervical approach [11].

Complications common to ranula are also associated with PR, with the most common being; recurrence of the lesion, lingual nerve injury, which may result in decrease in sensation of tongue and damage to the duct of submandibular gland. Other complications which are generally not as common are hematoma, infection, and dehiscence of wound. Complications such as marginal mandibular nerve paralysis tend to occur because of the

proximity of this nerve to the cyst, proper identification prior to surgery can prevent damage to this nerve [22].

Parekh et al. reviewed 139 surgeries of PR in 89 patients and recorded a recurrence rate of over 50% for cases where the sublingual gland was not excised. Meanwhile, a recurrence rate as low as 2% or less was recorded when the sublingual gland was excised alone or in association with other treatments. They thus advocate that the sublingual gland should at all times be removed to improve the prognosis of this condition [9]. In addition, in the 15 patients treated with sublingual gland excision by ref. [3], only two cases (13%) recurred after a 2-year follow-up period. This further substantiates the suggestion that the sublingual gland be removed at all times.

Essentially, the best practice for the treatment of PR has been showed to be the excision of the sublingual gland, as damage to it has been established to be the source of the condition. Thus, partial or total removal with or without the excision of the accumulated saliva would result in cure [16, 23] and although a transoral approach is ideal, cervical incision is recommended when the pseudocyst is of considerable size [21].

Discussion

The observation that patients in this series are young children and the fact that one of the cases gave a positive history as well as evidence of existence of the mass from birth could be a pointer that perhaps PR is congenital as proposed by some authors [7, 8].

The patient in our first case presented with a swelling in the anterior neck area while for the other cases, the swelling was in the submandibular region. All patients were completely asymptomatic and history including clinical examinations was suggestive of PR with oral component. For all our cases, neck ultrasonographic findings indicated a firmly circumscribed hypoechogenic mass occupying the anterior area of the neck; strongly suggestive of PR. Although, CT scan and MRI are the utmost imaging techniques recommended for the investigation of PR [24], unfortunately these could not be performed due to financial constrain. Surgeries were performed under general anesthesia; the accumulated saliva and associated sublingual gland were removed via a transcervical approach in three cases while treatment of the 4th case was performed intraorally. The transcervical approached was employed for most of our cases as this provided adequate access for removal of the sublingual gland as well as to evacuate the large accumulation of extravasated saliva. The small body mass of the younger children in this series warranted this approach thus we were able to avoid any inadvertent injury to deep structures in the floor of mouth.

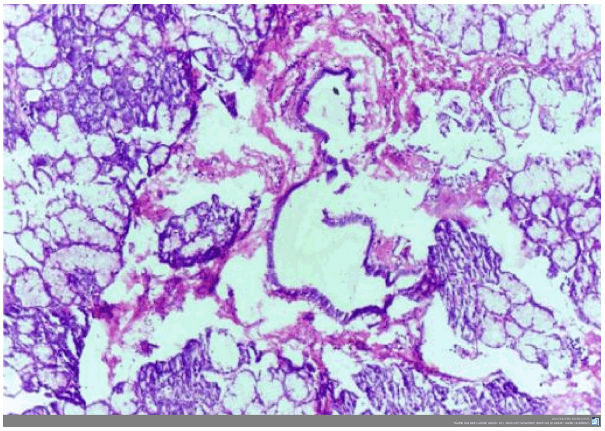

Figure 4. Histopathology of excised tissue showing distended acini and ruptured duct with the presence of mucin in connective tissue (H&E ×40).

No recurrence has been recorded so far. The excised tissues were sent for histopathologic evaluations and reported as pieces of soft tissue which in areas present distended acini and ruptured duct of salivary gland tissue. There was the presence of mucoid connective tissue surrounded by varying amounts of histiocytes laden with clear materials (Fig. 4). Thus, postoperative histopathologic report indicated findings consistent with extravasated mucocoele.

Conclusion

In conclusion, PRs are rare, although cases have been documented with moderate frequency especially in some ethnic groups; report of this condition is particularly limited in our environment. We present four cases of PR diagnosed in children; all cases had both oral and cervical components, and all were treated with surgical removal under general anesthesia.

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Conflict of Interest

None declared.

Authorship

ACOO: collected the clinical data, generated the manuscript, and responded to reviewers comments. OMO: reviewed the histopathologic slides, revised the

manuscript, and responded to reviewers comments. ICE: involved in writing the manuscript. RAA: supervised writing of the manuscript. MME: reviewed the histopathologic slides. OMG, AOA, and SBO: involved in patients follow-up, obtained patients consent, and involved in the literature search.

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