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Case Report

Surgical Treatment of Plunging Ranula. A Case Report

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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 a girl with plunging ranula using an intra-oral extension. In addition, clinical and radiographic findings of the patients along with the relevant review of the literature also referred.

Case report

A patient , female, 10 years old, with specific diagnosis of plunging ranula was treated by a intra-oral approach and excision of the cyst without complications.

Results

All cysts were ranula and the case presented indicates that this lesion can be managed by a less invasive procedure without complications and recurrence. At the patient ranula grow to a large size and the treatment became 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).

Keywords

Soft tissue cysts, Ranula, Surgical Treatment, salivary gland diseses.

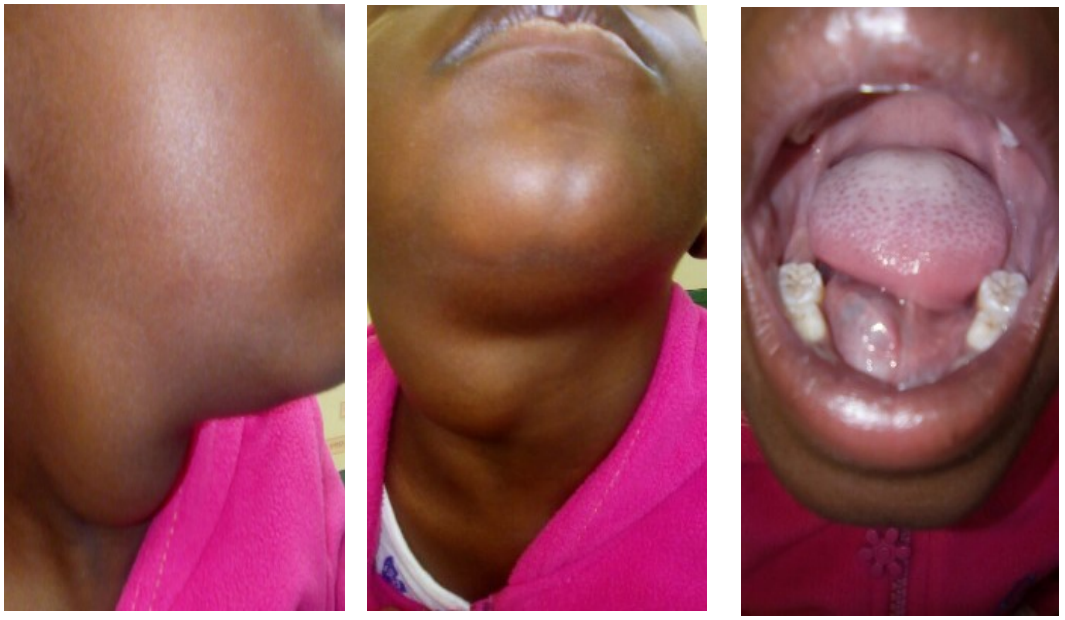
Introduction

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].

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 facial 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 ranula 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]. We reported a case female 10 years old with a plunging ranula, objective was to describe patient management.

Case Report

A 10 year-old female patient was referred to our department with a swelling in right sublingual region. The lesion arises from the sublingual salivary gland and extended through the floor of the mouth. The patient reported that the swelling enlarged during the last months. An Ultrasound showed a cystic lesion on the right 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 sub mental region (Fig. 1 A,B,C) extraoral and intraoral Ranula. The size of the lesion upon examination was approximately 2.5× 2.5 cm. A provisional diagnosis of plunging ranula was made.



A **B** **C**

Figure 1 (A,B,C). extraoral and intraoral ranula

Following detailed clinical examination and radiological (ultrasound) 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 intra-oral approach under general anaesthesia. Removal of the ipsilateral sublingual gland was also done via a floor of the mouth incision (Fig. 2) and all the tissue was subjected to histopathological evaluation. The excised mass confirmed the diagnosis of a plunging ranula in a mucous extravasation cyst. There was no sign of recurrence and the patient is free of the lesion after 2 year of follow-up (Fig. 3).

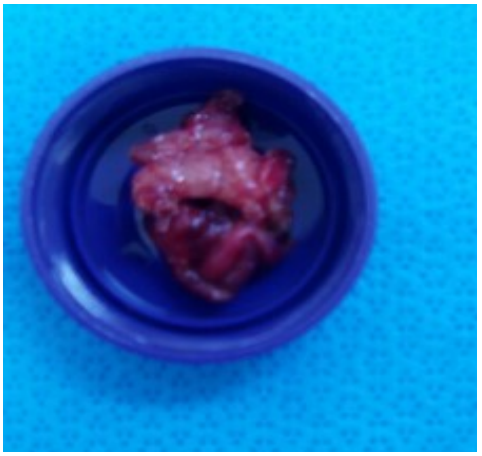


Figure 2. Sublingual gland was removal via a floor of the mouth

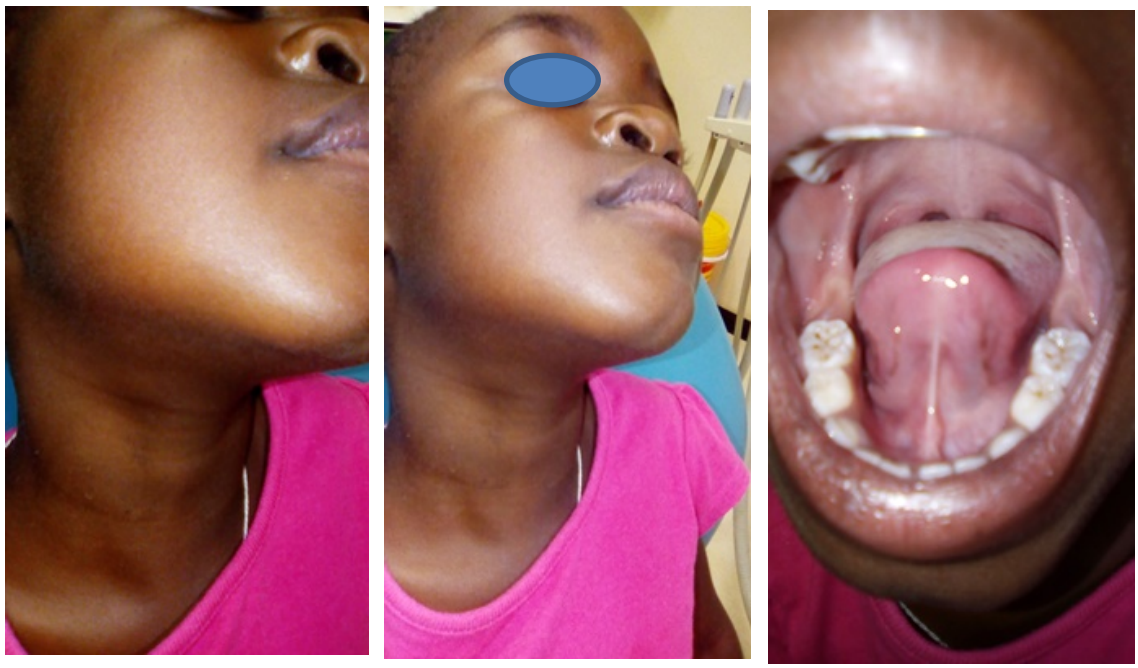


Figure 3. Post-operative result extra and intraoral

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]. Ranula fluid is clear and the swelling which are caused is soft [9]. It is characteristically large (>2 cm) and most reported ranula are 4–10 cm in size and rarely bigger than 10 cm [3]. When ranula become larger, it acquire a blue colour and resemble frog's belly Big sized ranula 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], [11], [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 ranula arises more often in the left side of the oral cavity, while plunging ranula are located more often in the right side [13]. Plunging ranula develops 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 sub mental 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 ranula 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 [3,14,15]. When plunging ranula 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]. Ranula 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 cyst adenocarcinoma of a sublingual gland maybe present as ranula, but is not frequent [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 ranula are surrounded from thick and fibrous boll and report them as pseudocysts. Plunging ranula 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]. 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 ranula) [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 (CO2 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].

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 intra-oral 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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