

Plunging Ranula in Children – A Case Report

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Abstract

Few cases of plunging ranulas occur during childhood and the lesions are frequently misdiagnosed. Plunging ranula is an uncommon condition in children under 10 years of age. Here a plunging ranula in a child is reported along with its management. A seven year old male patient visited to the department of oral and maxillofacial surgery complaining of swelling in right side of floor of mouth from 3 months. Patient gave history of difficulty in mastication and speech. On palpation a diffuse soft fluctuant non tender and fluid containing mass was observed. The suspicion plunging ranula was performed by ultrasound. The diagnosis was confirmed. The ranula and the sublingual gland excision was done. Differential diagnosis depends on clinical examination and ultrasonography. In paediatrics the key to success of the treatment depends on the radical excision of the cyst and sublingual gland via an intraoral approach.

Keywords: Plunging Ranula, Children, Surgical procedure.

Introduction

Ranula is derived from the latin word “Rana” meaning frog, as it resembles underbelly of frog¹. It is a mucous filled cyst like mass that usually originates from extravasation of mucus from the sublingual gland (intraoral ranula). It is referred to as a plunging ranula when it affects the submandibular space and adjacent structures in the neck. Plunging ranula is an uncommon condition, which presents itself as a fluctuating swelling in the neck. Ranulas superior to the mylohyoid muscle appear as a translucent bluish swelling under the tongue. The exact etiology of plunging ranula is still unknown but, an association with trauma, congenital anomalies and sublingual glandular diseases have been described². The ranula formation is thought to be secondary to blockage of the sublingual gland ducts, which leads to backpressure of

mucin and subsequent extravasation of mucus, through a hiatus in the mylohyoid muscle^{3,4}.

Plunging ranula is usually centered in the submandibular area, but can extend into the submental space, both cervical areas, the retropharyngeal space or the supraclavicular area². This poses diagnostic challenge to differentiate from other neck swellings especially from cystic hygroma⁵.

Treatment of ranula includes marsupialization, excision of sublingual salivary gland, excision of the ranula with or without excision of the sublingual salivary gland, and sclerotherapy. Excising the sublingual salivary gland is the key to minimize recurrence⁶.

A case of plunging ranula in a seven year old male is discussed along with its management.

Case Report

A seven year old male patient visited to the department of oral and maxillofacial surgery with chief complaint of swelling in right side of the floor of mouth from 3 months. Swelling was sudden in onset which was initially of peanut size and has been increased and attained the present size which was not associated with pain. Patient gave history of difficulty in mastication and speech. On systemic examination there was no abnormality detected.

On extraoral examination, a diffuse soft fluctuant non tender swelling about 2x3 cm in size was present in submandibular region (fig 1,2). On intraoral examination, a solitary dome shaped, bluish swelling measuring 3x3 cm was present on the right side of floor of mouth which was non tender, non compressible and fluctuant (fig3).



Fig.1



Fig.2



Fig.3

A provisional diagnosis of sublingual plunging ranula and differential diagnosis of thyroglossal duct cyst, cystic hygroma was made after correlating the history and clinical findings. The patient was then subjected to occlusal radiographic examination which revealed no signs of obstruction.

Ultrasonographic evaluation of the swelling showed large collection of fluid with internal echoes measuring 1.8x2.2 cm in right paramedian submental region. FNAC was performed under topical anesthesia and yielded collection of thick yellow mucus like aspirate.

After all preoperative evaluation, the case was posted for surgical excision of the sublingual gland along with epithelial lining of ranula under general anesthesia.

Surgical Procedure

The surgical technique is as follows: Intra oral mucosal incision was given in the floor of mouth lateral to lingual frenum taking care of Wharton's duct (fig 4), blunt dissection was carried out. Cystic swelling containing thick mucoïd material was seen (fig 5), entire mucous material was drained out. Blunt dissection was carried out taking care of lingual nerve and Wharton's duct (fig. 6). Sublingual salivary gland was freed and gland was removed along with epithelial lining of cyst (fig 7). After excision of sublingual salivary gland, mucosal closure was done with 3-0 vicryl suture (fig 8).

The ranula along with sublingual gland were sent for histopathological examination. Histopathology of the excised lesion showed mucin collection in the lumen lined by connective tissue with inflammatory cell and confirmed our preoperative diagnosis of ranula.

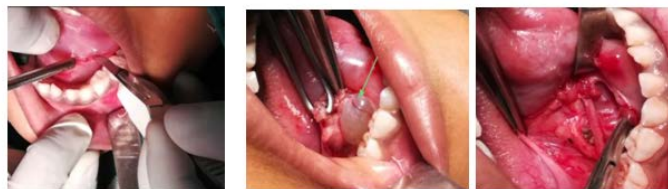


Fig. 4

Fig. 5

Fig. 6

Mucosal incision

Cystic swelling with mucus material

Whorton's duct and lingual nerve

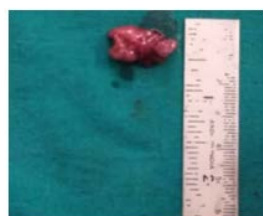


Fig. 7

Sublingual salivary gland excised



Fig. 8

Mucosal closure

Discussion

Pediatric plunging ranula is rare. There are several reported cases of plunging ranula in the literature. Although majority of these occurred in patients under the age of 30 years., only few occurred in children³. These represent for 6 percent of all oral sialocysts. There is no specific sex predilection for ranulas, but Neil et al., in their case report and review of literature involving plunging ranula stated that ranulas are more common in females⁵.

A ranula by definition is a mucus filled cavity, in the floor of the mouth in relation to the sublingual gland. Ranulas are characteristically large (>2 cm) and appear as a tense, fluctuant and dome shaped vesicle, sometimes with a bluish hue. The most common site is the lateral floor of the oral cavity as in the present case. Plunging ranula

occurs when the fluid pressure of the mucin dissects through a perforation in the mylohyoid muscle in the submandibular space⁶.

Arunachalam P.etal., in 2010 reported 2 cases of recurrent plunging ranula and dated that ranula is an extravasation cyst in the floor of mouth which develops from extravasation of mucus after trauma to the sublingual gland or obstruction of the duct⁷. The pathophysiology involved in extravasation is hypertension in the duct due to obstruction leading to acinar rupture in the salivary gland and then extravasation of the mucus. The initial stage is a traumatic rupture of the excretory duct and the second stage is the extravasation and subsequent accumulation of saliva within the tissue.

Plunging ranulas arise in the neck by one of the following four mechanisms.

1. The sublingual gland may project through the mylohyoid, or an ectopic sublingual gland may exist on the cervical side of mylohyoid. This may explain the plunging ranulas that exist without an oral component.
2. A dehiscence or hiatus in the mylohyoid muscle may occur. This defect is observed along the lateral aspect of the anterior two-thirds of the muscle. Through this defect, the mucin from the sublingual gland may penetrate to the submandibular space.
3. Approximately 45% of plunging ranulas occur iatrogenically after surgery to remove oral ranulas or secondary to surgical procedures for sialolith removal, duct transposition and implant placement.
4. A duct from the sublingual gland may join the submandibular gland or its duct, allowing ranulas to form in continuity with the submandibular gland. Therefore, the ranula accesses the neck from behind the mylohyoid muscle⁶.

Clinically, a Plunging ranula presents as a painless, fluctuant, gradually enlarging lateral neck swelling which does not usually change with swallowing or eating. It is most commonly centered on the submandibular triangle and averages 4-10 cm in size. It can extend superiorly in to the parapharyngeal space as far as the skull base, inferiorly to the supraclavicular area, posteriorly into the retropharyngeal space or across the midline anteriorly². Macdonald A.J. et al., in 2003 reported a giant ranula of the neck and stated that giant ranula is one that significantly involves the parapharyngeal space in addition to the submandibular space which makes differentiation from other cystic neck masses, particularly cystic hygroma difficult⁸.

Plunging ranulas generally appear in conjunction with oral ranula; but at least 20% can arise independently of an oral component. In the absence of oral swelling, the clinical diagnosis of ranula may not be suspected and a differential diagnosis may be difficult. The differential diagnosis of plunging ranula include thyroglossal duct cyst, intramuscular hemangioma, lipomas, cystic/neoplastic thyroid disease, branchial cyst, submandibular sialadenitis, laryngocele, dermoid cyst, lymphatic or vascular malformations and infectious cervical lymphadenopathy like tuberculosis, Epstein-Barr virus, cervical thymic cysts, dermoid cysts, cystic hygroma and benign teratoma².

Sialogram, ultrasonography, Magnetic resonance imaging (MRI), CT, and aspiration cytology can be helpful for diagnosis. Takimoto stated a simple radiographic technique for preoperative diagnosis of plunging ranula that involves injecting a contrast media in sublingual space. A sialogram performed on a patient with a sialocyst reveals smooth displacement of the glandular ducts around the mass. No direct communication with the

ductal system is demonstrated. However, the best method of demonstrating a communicating cyst is by sialography. Ultrasonography is usually inconclusive to study sublingual glands due to their location. CT scan can help to understand the origin and extent of this cystic lesion. Although a plunging ranula may extend into the submandibular triangle and displace the submandibular gland, it does not intrinsically affect this gland. MRI is the most sensitive study to evaluate the sublingual gland and its states⁶.

Clinicians have been using several different methods for the treatment of cervical ranulas. These include excision of the ranula only, cryosurgery, marsupialization with or without cauterization of the lesion lining, excision of the oral portion of the ranula with the associated sublingual salivary gland or, rarely, the submandibular gland, intraoral excision of the sublingual gland and drainage of the lesion, and excision of the lesion via a cervical approach, sometimes combined with excision of the sublingual gland⁶.

The most common advocated technique for the management of ranula in earlier days was marsupialisation. It had high recurrence rate of 61%-89% as the cyst was not completely excised. In some instances, it served as a precursor for plunging ranula. The packing of the ranula cavity with gauze after marsupialisation was found to be effective in curtailing the recurrence rate. It is generally recommended for cyst less than 2 cm in diameter.

In order to avoid this problem, Yang Y and Hong K recommended removing the cyst along with the sublingual gland. Removal of submandibular gland was recommended in plunging ranula, extending into the parapharyngeal space. The surgical excision can be carried out via transcervical or transoral approach.

Transcervical approach is commonly employed in en bloc resection of the cyst, as it is difficult to obtain a substantial cervical extension through intraoral approach. Probability of injury to the marginal mandibular, lingual and hypoglossal nerves are common in transcervical approach. Also, there is a risk of orocervical fistula formation and cervical scar in the cervical approach. The choice of incision should be based on the extension of cyst to avoid unwanted complications¹.

Besides surgical management, CO2 laser has been used to vaporize ranulas. In few cases, radiation therapy or intracystic injection of the streptococcal preparation, OK-432, has been used to treat this lesion. The use of this sclerosing agent as a treatment approach for the cervical ranula is considered experimental. A recent study found orally administered Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations(D10/D30/D200), a homotoxicological agent to be an effective treatment modality for ranulas⁶.

The most common complications is the recurrence of the lesion (5.78%) and sensory deficit of the tongue (4.89%), followed by damage of Whartons duct (1.82%). Postoperative hematoma, infection, or dehiscence of the wound were seldom seen. Excision of the ranula with the associated sublingual salivary gland is the most accepted method with low recurrence rate¹. A biopsy of the cystic wall is recommended not only for histologic confirmation, but also to rule out presence of squamous cell carcinoma arising from the cyst wall and papillary cystadenocarcinoma of the sublingual gland, which may present as ranula⁶.

Conclusion

Plunging ranula is an uncommon condition in young children. The differential diagnosis depends on clinical examination and ultrasonography. Although the treatment

of plunging ranula is still controversial, the key to success remains the radical excision of cyst and ipsilateral sublingual gland, via an intra oral approach or submandibular approach, with low morbidity and absence of recurrence.

References

1. Saraniya Packiri, Deepa Gurunathan. Management of pediatric oral ranula: A Systematic Review. Journal of clinical and Diagnostic Research. 2017 Sep, Vol-11(9).
2. Veronica Carlini, Noemi Pasqua. Plunging ranula in children: Case report and literature review. Pediatrics Reports 2016; volume 8: 6576
3. Mahadevan M, Vasan N. Management of pediatric plunging ranula. Int J Pediatr Otorhinolaryngol 2006; 70: 1049-54.
4. Zhi K, Gao L, Ren W. What is new in management of pediatric ranula? Curr Opin Otolaryngol Head Neck Surg 2014; 22: 525-9
5. Abdul Bagi Mustafa, Kamran Bokhari. Plunging ranula: An interesting case report. Open Journal of Stomatology, 2013,3,118-121.
6. Sunil Kumar Sharma, Gaurav Singhal. A Cervical Ranula: A Case report. International Journal of Medical Research and Health Sciences, 2016, 5, 7: 109-113.
7. Arunachalam, P. and Priyadarshini, N. (2010) Recurrent plunging ranula. Journal of Indian Association of Pediatric Surgeons, 15, 36-38.
8. Macdonald, A.AJ, Salman, K.L. and Harnsberger, H.R. (2003). Giant ranula of the neck: Differentiation from cystic hygroma. American Journal of Neuroradiology, 24, 757-761.