

Diagnosis and management of recurrent unilateral mixed ranula: A pediatric case report

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Abstract

Plunging ranula which is also known as diving or cervical ranula usually occurs along with oral ranula. They commonly arise from the sublingual gland. Ranula is a nonepithelial-lined salivary gland cyst that is formed due to escape of mucus from sublingual gland and its successive herniation via the mylohyoid muscle into submandibular space and beyond. Here we present a case of unilateral plunging ranula in a 5-year-old child in which ultrasonography was used as a diagnostic tool and managed by a less invasive procedure without complication and recurrence.

Keywords: Plunging Ranula, Floor of the mouth, Painless Swelling.

Introduction

There are two types of ranulas, simple and plunging. An extravasation of saliva from the sublingual gland due to trauma or obstruction of the duct results in the formation of a plunging ranula. Fluid from the obstructed gland flows between the fascial planes and muscle of the base of the tongue to the submandibular space. The accurate prevalence of plunging ranula is unknown, however, these lesions are considered uncommon. Maximum number of plunging ranulas either accompany a swelling in the floor of mouth or have a history of treatment of intraoral ranula.¹ Sublingual ranulas are formed below the mucous membranes of the floor of the mouth above the mylohyoid muscle, whereas plunging ranulas extend below the mylohyoid into the submandibular or parapharyngeal space, giving rise to a neck swelling.^{2,3}

Numerous surgical techniques have been used to treat ranula, like marsupialization, incision and drainage, excision of the ranula only, excision of the ranula and the sublingual gland, and sclerotherapy. Some percentage of recurrence has been reported depending on the type of treatment.⁴

Case

A 5-year-old female patient reported to the department of oral medicine and radiology with a complaint of a painless swelling in the right floor of the mouth region below the tongue since 2 months which was treated by an oral physician and had subsided, but within a few days it has recurred and had slowly progressed to the present size with no other associated signs and symptoms. Her past medical history was noncontributory. On extra oral examination a solitary diffuse swelling was present in the right neck region extending super inferiorly around 2cm from the right lower border of mandible to the level of the thyroid and anteroposteriorly from the chin to the right angle of the mandible, on palpation it was soft and fluctuant and non-tender (Fig1). Intra oral examination revealed a solitary well defined, dome shaped translucent swelling in the right floor of the mouth measuring about 3x3 cm blueish in color with smooth surface lingual to 85 and on palpation it was soft, fluctuant and non-tender (Fig 2). On hard tissue examination full complement of deciduous teeth were present without any caries or any other deformities. A provisional diagnosis of Mixed (oral and plunging) Ranula of right side of the floor of the mouth was given and the patient was subjected to ultrasound.

Ultrasonography (Fig 3) of the cervical region revealed well defined cystic lesion with fine echoes in the right

sublingual space measuring about 4.1x1.2cm suggestive of Plunging Ranula.

Drainage and Marsupialization of the lesion was done and histopathology revealed presence of Para keratinized stratified squamous type of epithelium and connective tissue. Stromal tissue showed lined dilated duct to form a mucin filled cyst with granulation tissue. A brisk of inflammatory response, extravasated mucin and foamy histiocytes were seen and few areas showed phagocytosed mucin macrophages adjacent to minor mucous salivary glands suggestive of Mucous Extravasation Cyst of the right side of the floor of the mouth.

Discussion

A ranula is defined as a mucus filled cavity, a mucocele, in the floor of the mouth in relation to the sublingual gland. The term ranula is derived from the word 'rana', which means frog in Latin, as the swelling in the sublingual mucous membrane resembles the translucent underbelly of a frog it is termed as Ranula.^{1,4}

Sublingual glands are a pair of salivary glands situated in the floor of the oral cavity, inferior to the tongue and medial to the body of the mandible. These glands weigh about 2g shaped like an almond drain into a row of 12 to 20 short excretory ducts, known as ducts of Rivinus, which open along the summit of the sublingual fold into the floor of the mouth. Leaking of the saliva into the surrounding tissue due to trauma, obstruction or inflammation induces a fibroblastic reaction that seals the mucus in a connective tissue sac termed as extravasation pseudo cyst or ranula.^{4,5,6} In less than 10% of all ranulas, congenital obstruction, by either an imperforate salivary duct or an ostial adhesion, leads to the formation of a retention cyst.⁷ It was suggested that

ductal agenesis or hypoplasia or trauma during birth may be the cause for infantile or pediatric ranulas.⁸

Typically, a ranula is a slow-growing unilateral, very rarely bilateral painless swelling in the floor of the mouth. It can range in size from 4 to 10 cm.⁷ The frequent age of occurrence is second and third decade and in children younger than 10 years, it has a slight female predilection.^{4,9} They appear as a tense fluctuant dome-shaped vesicle, most of the time with a blue hue. The most common site is the lateral floor of the oral cavity.¹ Ranulas can present itself as oral ranulas which have only intraoral swelling, while a plunging ranula shows cervical swelling without swelling of the floor of the mouth and when plunging ranula is associated with intraoral swelling it is termed as mixed ranula just as seen in the present case,¹⁰ Usually the plunging ranulas are seen with the oral ranula.⁹ Intraoral ranulas were seen in 45% of the cases; 34% of the cases were said to be associated with plunging ranulas and 21% of plunging ranulas occur without oral swelling.⁷

There are few mechanisms to explain the formation of plunging ranula, the sublingual gland may project through the mylohyoid, or an ectopic sublingual gland may exist on the cervical side of mylohyoid, this explains most plunging ranulas that exist without an oral component. Secondly, 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. Around 45% of plunging ranulas were reported to have occurred after surgical excision of oral ranulas. Lastly, 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 muscle.¹ Surgical procedure for sialolith removal, duct transposition and implant placement may also result in plunging ranulas.^[11-13]

Thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, submandibular sialadenitis, intramuscular hemangioma, cystic or neoplastic thyroid disease, infectious cervical lymphadenopathy (Epstein-Barr virus, cat scratch disease, tuberculosis), hematoma, lipoma, laryngocele, and dermoid cyst can be considered in the differential diagnosis of plunging ranula.¹

Sialography, ultrasonography, computed tomography and magnetic resonance is helpful in the diagnosis of Ranula. Sialography is the best preoperative diagnostic method for demonstrating a plunging ranula.¹⁴ CT, findings show ranula as roughly ovoid-shaped cyst with a homogeneous central attenuation of 10 to 20 HU. The cystic wall is either very thin or not seen, and the lesion is usually placed lateral to the genioglossal muscles and above the mylohyoid muscle. MRI is the most sensitive imaging modality to evaluate the sublingual gland and its states. On MR imaging, due to high water content ranula appears as a low T1-weighted, an intermediate proton density, and high T2-weighted signal intensity. In case of plunging ranula, this feature may be similar to that of a lymphangioma and lateral thyroglossal duct cyst

But, if the protein concentration of the ranula's contents is high, the signal intensities will often be high on all imaging sequences. In such cases, the MR differential diagnosis includes dermoids, epidermoids, and lipomas.¹⁵

Sonographically, the sublingual glands are difficult to image because of their location. A plunging ranula is more easily visualized because it is more superficial, it

appears as an anechoic, avascular structure with thin walls, incomplete septae, and sometimes fine internal echoes are visible suggesting debris. High-resolution sonography determines the extension of a plunging ranula, demonstrating the extension of the ranula through a mylohyoid defect into the submental region.⁴

Histopathology, of the cervical ranula reveals only rare inflammatory cells and predominant histiocytes, which stains positive for mucin.¹⁶ Biochemical analysis of fluid shows high amylase and protein content.

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 ranula.¹

Treatment options for ranula includes excision of the oral portion of ranula with the associated sublingual salivary gland and excision of the lesion via cervical approach with the sublingual gland can be done for cervical ranula. Marsupialization with or without cauterization of the lesion lining can be done, excision of the ranula with the associated sublingual salivary gland is the most accepted method with low recurrence rate.¹⁷ Risk for paresis and paralysis of the marginal mandibular nerve is the most common complication following surgical therapy of ranula.¹⁸

Radiation therapy of low doses of 20–25 gray, Intracystic injection of the streptococcal preparation, OK432, has been used to treat this lesion in a few reported cases.¹

Besides surgical management, CO₂ laser has been used to vaporize ranulas.¹⁹

When ranula has diameter less than 2cm drainage and marsupialization of the lesion without excision of the involved salivary gland has high success rate with

minimal complications just like in the above case.²⁰ Marsupialization with packing of the cyst cavity may reduce the recurrence.²¹

Conclusion

The cases of plunging ranula in children are moderate in frequency and have been reported to have high recurrence if not treated properly. Thus, proper radiological and histopathological investigations should be carried out for all cases of probable plunging ranulas for early diagnosis and affective treatment. Though surgical removal of the sublingual gland with the ranula have shown less recurrence, in children Marsupialization with drainage and packing of the cyst cavity just like in the above case also has shown less recurrence rate.

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Legends Figures



Fig 1: Solitary diffuse swelling present in the right neck region.



Fig 2: Dome shaped translucent swelling in the right floor of the mouth.

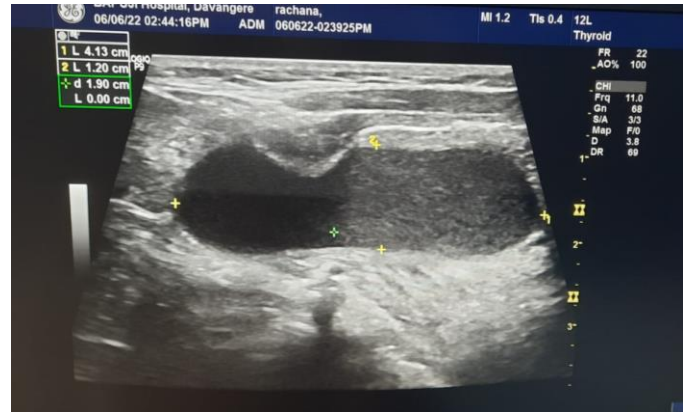


Fig 3: Ultrasonography

Well defined cystic lesion with fine echoes in the right sublingual space measuring about 4.1x1.2cm