

CASE REPORT

Bilateral Plunging Ranula due to Habitual Etiology

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ABSTRACT

Plunging ranulas are pseudocysts in which mucous have extravasated either around or through the mylohyoid muscle to escape the confines of sublingual space to involve the submandibular and inferior aspect of parapharyngeal spaces. We describe a rare case of extensive bilateral plunging ranula in which involvement of multiple tissue spaces was well illustrated by magnetic resonance imaging. Clinical and histologic features and pathogenesis have been concisely discussed. The ranulas mostly occur unilaterally and to the best of our knowledge, the development of two discrete ranulas/bilateral presentations in the same patient is especially rare and very few cases have been reported until now. Although the cause of extravasations remains unclear, this case report is significant because of its characteristic possible habit-related etiology.

Keywords: Bilateral ranula, Plunging ranula, Ranula.

How to cite this article: Bishen KA, Singh A, Limaye M, Mishra K. Bilateral Plunging Ranula due to Habitual Etiology. *J Oral Health Comm Dent* 2017;11(1):19-22.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Ranula is a clinical term generally used to describe cystic lesions or mucocele specifically occurring in the floor of the mouth and arising in association with the ducts of submaxillary or sublingual gland.¹ According to the variation of its extension, they are categorized into two: "simple/sublingual type" and "plunging/diving type."^{1,2} A sublingual ranula develops in the floor of the mouth,

while plunging ranula develops as a soft cystic swelling in the submandibular space or in upper cervical region.³ The main differences between these two are elaborated in Table 1. Some authors identify a third variant called as "mixed type." The term mixed ranula (or the sublingual-plunging or sublingual-submandibular ranula) is used when a sublingual ranula has an extension beyond the mucous membrane of oral cavity into the floor of mouth into the cervical region through a hiatus of mylohyoid muscle.³ Out of these three, the majority is the sublingual type, followed by the plunging type and the mixed type. The ranulas mostly occur unilaterally and, to the best of our knowledge, the development of two discrete ranulas/bilateral presentations in the same patient is especially rare.⁴

The aim of presenting this article is to describe a case of extensive bilateral plunging ranula. The involvement of multiple tissue spaces was well illustrated by magnetic resonance imaging (MRI), and was not evident from the history, examination, plain radiography, or ultrasonography. Several methods of treatment of plunging ranulas have been reported. We treated this case only by marsupialization.

CASE REPORT

A 23-year-old male patient had a chief complaint of swelling in neck on both sides. He had been consulting a private dental practitioner since 5 weeks. He was on antibiotics, but his condition did not improve. The patient's history revealed that he was a habitual whistler. He used to put fingers of both his hands simultaneously in his

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Table 1: Differences between simple and plunging ranula

<i>Simple/sublingual type</i>	<i>Plunging/diving type</i>
Such a ranula is limited to the sublingual space	Such ranulas have extravasated either around or through mylohyoid muscle to escape the confines of sublingual space to involve the submandibular and inferior aspect of parapharyngeal spaces
Clinically seen as an intraoral mass lesion	Clinically seen as submandibular or neck mass with no apparent oral connection
Histologically, it is a true retention cyst possessing a mucosal lining (derived from ductal epithelium)	Histologically, it is an extravasation phenomenon (pseudocyst) with a lining composed of condensed connective tissue rather than true epithelial lining

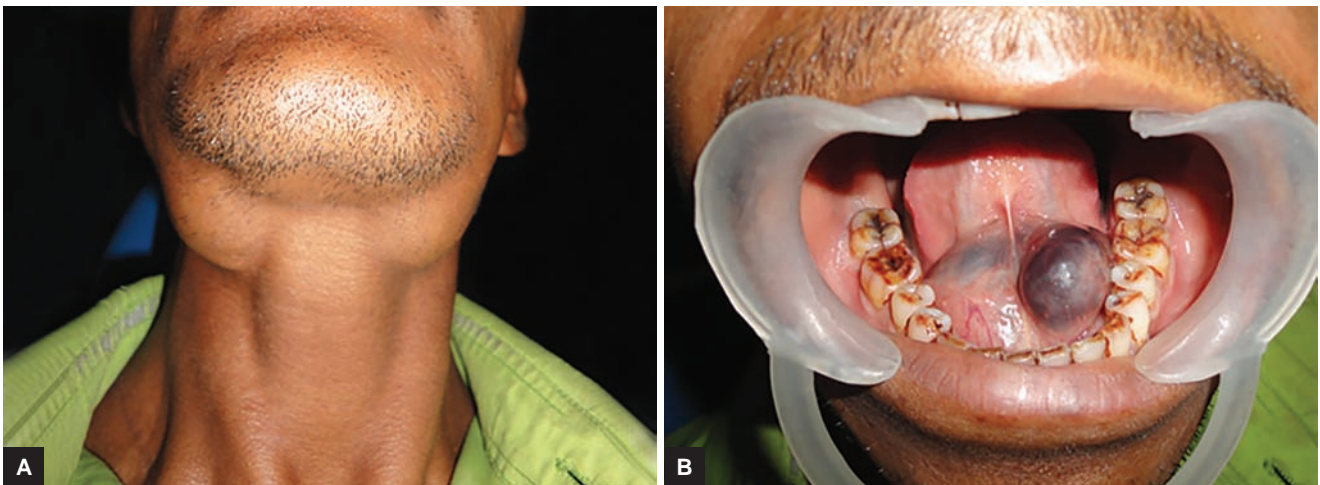
mouth, under the tongue to bend it up. The frequency of whistling was reported to be more than 30 per day. Other medical history was insignificant.

Extraoral clinical examination revealed that there was swelling bilaterally present over submandibular region (Fig. 1A). Intraorally, a soft bluish, fluctuant swelling was present on the left side of the floor of the mouth, while on right side, the floor was elevated with a whitish appearance (Fig. 1B). The patient reported that the size of swelling decreased in between meal times. Examination by lower occlusal radiograph was unremarkable. Following this, fine needle aspiration cytology was performed, and a diagnosis of benign epithelial cyst was made. For evaluating the extension of cyst and formulating a treatment plan, MRI of neck and floor of mouth was performed. T1- and T2-weighted images were taken in axial, coronal, and sagittal planes. This reported the extension of the cyst in bilateral sublingual and submandibular spaces. Anteriorly, it extended to the submental space (Figs 2A to C).

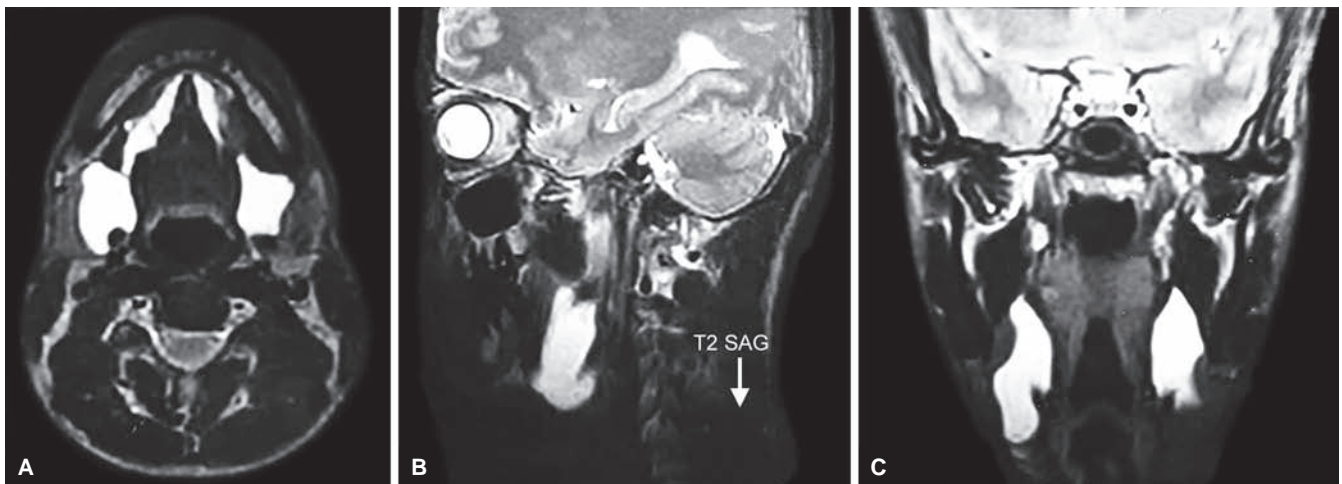
Under local anesthesia, the cyst was aspirated and blood-tinted fluid was collected. Then an incision was made over left-most elevated area of cyst; the remaining fluid was drained and the swelling was decompressed (Figs 3A and B). Barrel bandage was applied to avoid further fluid collection. Antibiotics and analgesics were prescribed. After 2 weeks, patient had an uneventful recovery.

DISCUSSION

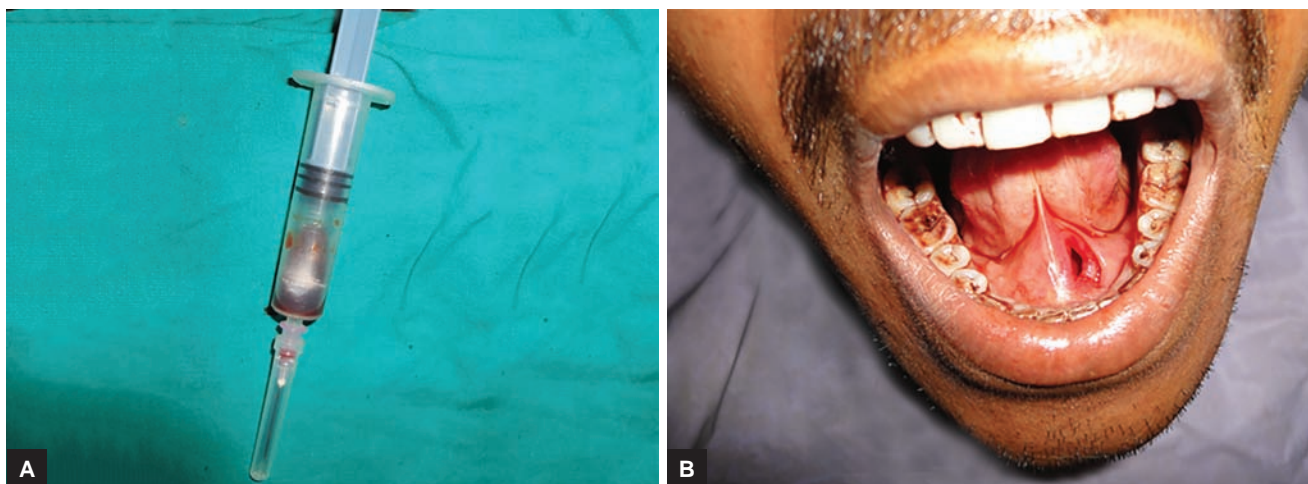
The classic ranula arises as a slowly enlarging painless swelling on the floor of the mouth. If the lesion is superficial, it may have an appearance of a translucent blue domed swelling, resembling the “belly of a frog,” hence, the name ranula – which is derived from the Latin word “rana” meaning frog. If the lesion is a deep-seated one (plunging type), the overlying mucosa is normal in appearance,⁵ and such plunging ranulas manifest as



Figs 1A and B: Clinical photograph of (A) bilateral swelling present over submandibular region; and (B) intraoral soft bluish, fluctuant swelling



Figs 2A to C: T2-weighted MRI images in (A) axial; (B) sagittal; and (C) coronal planes reflecting the extension of ranula bilaterally in sublingual and submandibular spaces and extending anteriorly into the submental space



Figs 3A and B: (A) Blood-tinted fluid aspirated from the cyst; and (B) marsupialized, drained, and decompressed swelling

only a swelling in neck and may have no visible intraoral involvement, making the diagnosis difficult.⁶

The true incidence of ranula is not known, but the age range for occurrence is from 2 to 61 years. It is more commonly found in females, with a female-to-male ratio being 1.3:1.⁶ The site of occurrence of ranulas, as discussed, is the floor of the mouth and neck, and they can develop into large masses that are many centimeters in diameter, fill the floor of the mouth, and elevate the tongue. They are usually located lateral to midline – a feature helping to distinguish it from midline dermoid cysts.⁷ The anterior crossing of oral ranula beneath the frenulum to the contralateral sublingual space can be encountered when it becomes significantly large.² The ranulas usually occur unilaterally; bilateral occurrence is quite rare and very few cases have been reported till now.

Histologically, the ranulas being pseudocysts lack an epithelial lining. Characteristically, an early lesion consists of a moderately well-defined and, occasionally, circumscribed cavity within the soft tissue, i.e., filled with an eosinophilic material that stains positive for mucin. Within the eosinophilic material, an admixture of acute and chronic inflammatory cells and foamy histiocytes may be found. The tissue surrounding the cavity consists of compressed fibrovascular (which on low magnification may be misinterpreted as an epithelial lining). Variable number of polymorphonuclear leucocytes, lymphocytes, and plasma cells are seen in the surrounding connective tissue. Salivary gland acini or ductal elements adjacent to the lesion are frequent findings. As the lesion matures, granulation tissue progressively grows into the cavity as an attempt of the body to repair the lesion and slowly obliterates the defect. Changes are also seen in salivary glands adjacent to primary lesion. These include a generalized chronic sialadenitis, distension of ducts, atrophic changes in acini of gland, and variable degree of

interstitial fibrosis within the salivary gland lobules. In addition, the overlying epithelium may show thinning and flattening of rete ridges as a result of circumferential expansion of the lesion.^{5,7}

There had been considerable debate in the literature in regard to the pathogenesis of ranula.³ There are primarily two different schools of thoughts regarding the pathogenesis. One is a true cyst within an epithelial lining formed due to ductal obstruction, and the other is a pseudocyst without an epithelial lining, formed due to ductal injury and extravasation of mucous from the sublingual gland.¹ The vast majority of ranulas are now recognized as extravasation cysts arising from sublingual gland. In particular, the plunging ranulas are almost exclusively extravasation cysts.

The cause of extravasation remains unclear.³ However, ranula formation has been speculated to be associated with congenital anomalies and anatomic variation of sublingual gland and/or trauma, resulting in severance of a duct with resultant mucous extravasation into the ambient tissue.⁴ Accidental creation of ranula as a result of intraoral removal of submandibular sialolith has been reported.⁸ Thus, it is possible that a second lesion may be caused by traumatic injury from a first surgery for a sialolith or a ranula or that the lesion may be recurrent. Or else, the bilateral presence may indicate a latent susceptibility of the patient to development of plunging ranulas. Lastly, anatomic variation of sublingual gland may be involved in formation of ranulas – as was reported in a case where the plunging ranula was actually a part of sublingual gland, which had extended to the submandibular region, facilitated by mylohyoid muscle herniation.³

Thus, the etiopathologic factors involved and their role in formation of plunging ranulas are:

- Anatomic variation of the sublingual gland
- Congenital anomaly of the gland

- Latent susceptibility of the patient for the development of plunging ranulas [which may be genetic or due to factors (i) and (ii)]
- Trauma – leading to severance of the gland duct
- Iatrogenic – injury from a first surgery for a sialolith or a ranula
- A sadomasochistic habit (e.g., whistling by inserting finger in mouth under the tongue) in a person with latent susceptibility for development of ranula would have caused the severance of the sublingual gland duct, as would have been the situation in the present case.

As the pathogenesis of ranula was not clear, exact treatment protocol was not established. Several methods of treatment of plunging ranulas have been reported, including injections of botulinum toxin type A into the lesion,⁸ marsupialization, excision of the pseudocyst, and sublingual or submandibular gland. The prognosis is same except that some operators may prefer to only unroof the lesion rather than to excise it totally. We treated this case only by marsupialization, and the patient had an uneventful recovery.

CONCLUSION

We present a rare case of bilateral plunging ranula in which the possible pathogenesis could have been a latent susceptibility of the patient for the development of plunging ranulas along with a habit of frequent whistling, which would have caused the rupture of duct. The

extension of the cyst bilaterally into the sublingual and submandibular spaces and anteriorly into the submental space was confirmed by MRI. For treatment, the ranula was marsupialized, fluid was drained, and the swelling was decompressed. Barrel bandage was applied to avoid further fluid collection and the patient had an uneventful recovery.

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