Plunging Ranula Occurring Without its Oral Counterpart: A Case Report

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A ranula is a lesion that arises from mucin spillage from the sublingual or submandibular gland ducts, beneath the mucosa of the oral floor. A plunging or cervical ranula dissects the mylohyoid muscle and appears as a submandibular swelling. Rarely, a plunging ranula is encountered without its oral counterpart. Here, we report a rare case of a plunging ranula that occurred without its oral counterpart in a 11-year-old male patient. We have also discussed the pathogenesis and treatment options for the ranula.

Key words: *Cervical, Pathogenesis, Ranula, Surgery, Treatment, Ultrasonography*

INTRODUCTION

A ranula is a lesion caused by the escape and collection of mucus in the sublingual space, thereby producing a swelling in the floor of the mouth. The term "ranula" is derived from the Latin word "rana", which means frog. The appearance of this swelling in the floor of the mouth is comparable to the swollen and translucent underbelly of a frog, and is thus termed a "ranula". Ranulas mostly arise due to mucus escape from the ducts of the sublingual salivary gland. In addition to the lesion arising in the oral floor (i.e. a simple ranula), another variant of a ranula (i.e. cervical or plunging ranula) exists. A plunging ranula arises when the spilled mucin from the salivary gland dissects the mylohyoid muscle and produces a swelling in the neck, typically observed in the submandibular triangle. Most plunging ranulas clinically present with their oral counterpart; hence, they are easy to diagnose. However, a plunging ranula that presents only as a neck swelling without any oral counterpart is difficult to diagnose. For diagnosing such lesions, many similar entities must be ruled out. Radiological investigations such as contrast enhanced computed tomography or magnetic resonance imaging are mandatory for the diagnosis and management of such cases.

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Case Report

A 11-year-old male patient reported to a dental office with a painless swelling in the right submandibular region, which had been present for 3 years. The patient's parents reported an intermittent increase in the size of the swelling.

On extraoral examination, facial asymmetry due to swelling in the right submandibular region was observed. The swelling was roughly oval with ill-defined margins and measured approximately 4 and 2.5 cm in length and width, respectively. On palpation, the swelling was soft, nontender and fluctuant. No significant intraoral findings were observed.

Ultrasonography of the upper neck revealed anechoic loculated fluid collection medial to the right submandibular gland (Figure 1). The lesion was 5.7 mm deep to the skin surface. Contrast enhanced computed tomography showed a well-defined, non-enhancing cystic lesion involving the right submandibular region. The lesion was 28 mm in length and 14 mm in width with a craniocaudal extent of 28 mm (Figure 2).

On the basis of the clinicoradiological features of the lesion, a plunging ranula was provisionally diagnosed. Although less likely, dermoid cyst, thyroglossal duct cyst and cystic hygroma were considered as differential diagnoses.

Under general anesthesia, the lesion was enucleated in toto with excision of the right sublingual salivary gland by using cervical approach (Figure 3). The entire specimen was submitted for histopathological examination.

The specimen was dark brown, roughly oval, soft and fluctuant, measuring 30 and 20 mm in length and width, respectively. The lesion was bivalved; on cutting it open, the cystic lumen was visible, which was filled with a golden-yellow, jelly-like material (Figure 4). Microscopic examination revealed a cavity that contained eosinophilic material and was surrounded by fibrocellular connective tissue. Numerous mucinophages were observed within the cystic lumen and the adjacent part of capsule (Figure 5).

Figure 1: Ultrasonogram showing anechoic loculated fluid collection in the right submandibular region

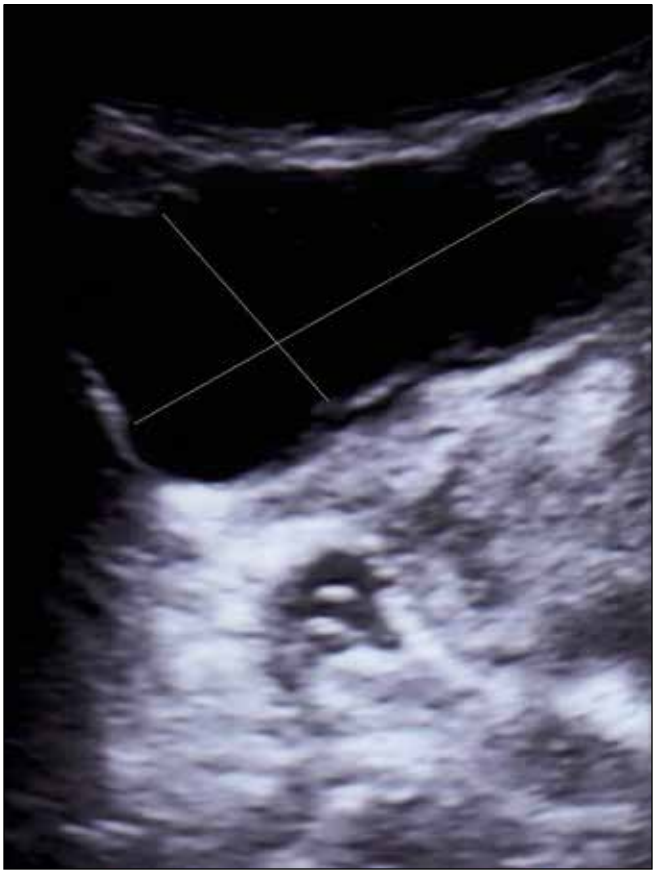


Figure 2: Contrast-enhanced computed tomography showing a non-enhancing cystic lesion involving the right submandibular region (arrow)

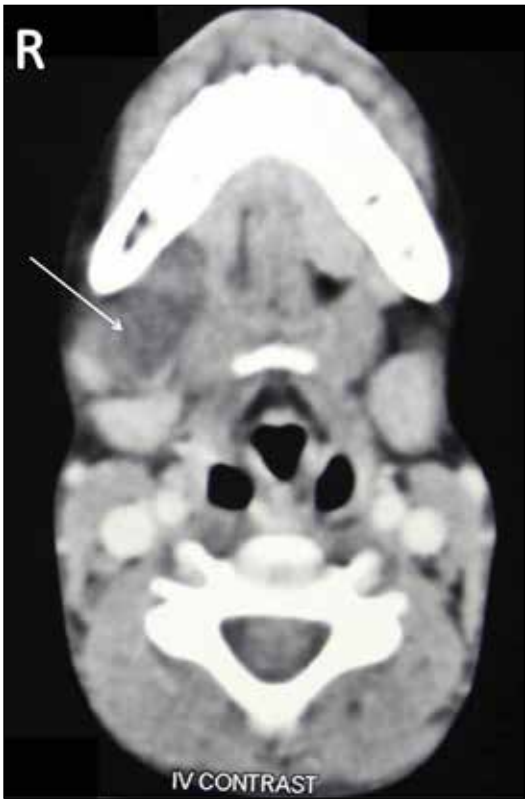


Figure 3: (A) Intraoperative image of subplatysmal dissection showing a superficial part of the ranula (B) Enucleation of the cyst following careful separation from the surrounding attachments

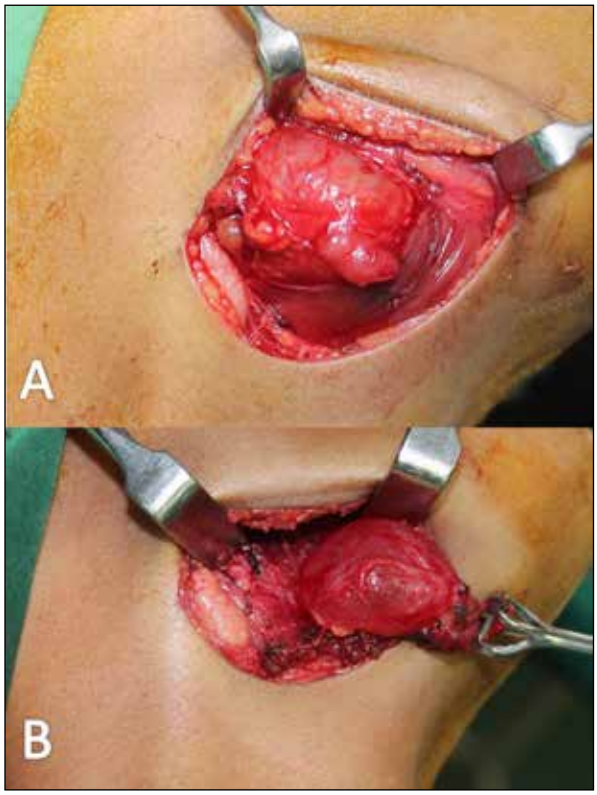


Figure 4: (A) Gross specimen of the ranula along with the offending right sublingual gland (B) Cystic lumen showing a golden-yellow, jelly-like material

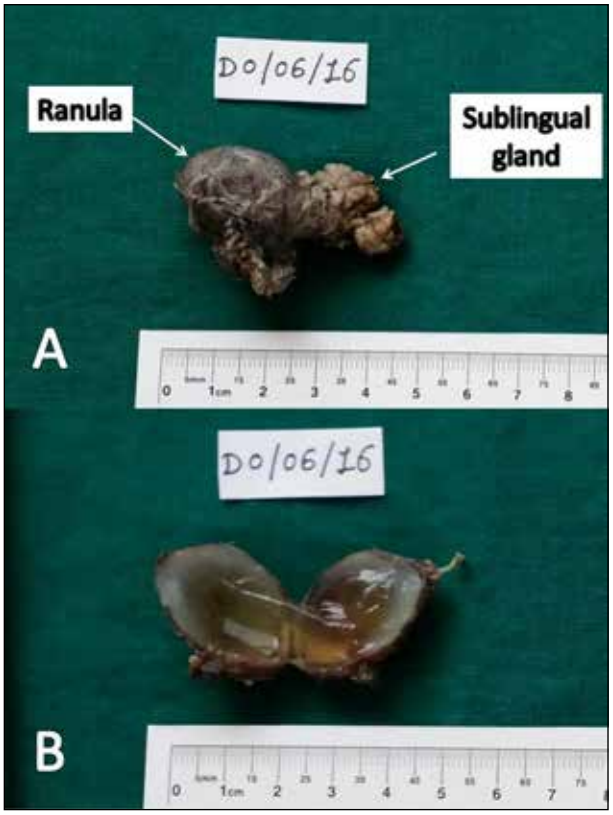
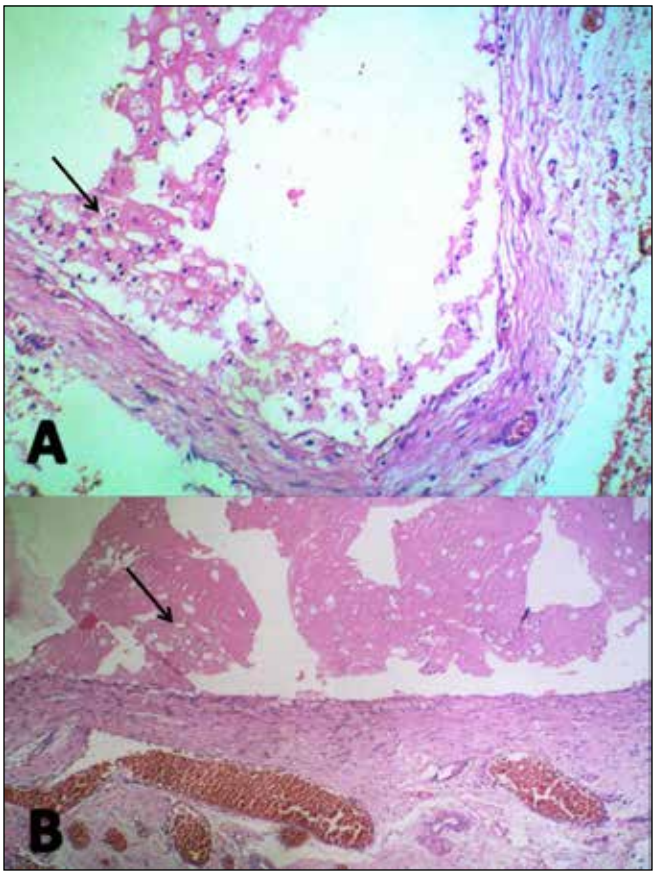


Figure 5: (A) Photomicrograph showing the cystic lumen, surrounded by compressed fibrovascular connective tissue. Mucinophages are seen in the lumen (arrow; hematoxylin & Eosin stain, magnification: 100x) (B) The cystic lumen showing pooled mucus and numerous mucinophages (arrow; hematoxylin & Eosin stain, magnification: 100x)



On the basis of the clinicoradiological features and microscopic findings, a plunging ranula involving the right sublingual gland was diagnosed. The patient was followed up at monthly intervals. No signs of recurrence were observed for 6 months postoperatively. Thereafter, the patient was lost to follow-up.

DISCUSSION

A plunging ranula usually presents with its oral counterpart. Approximately 21% of plunging ranulas occur without a corresponding intraoral swelling.[1] The fundamental developmental mechanism of ranula involves the traumatic severance of a salivary duct, followed by the extravasation of mucus, resulting in localized mucin collection within the connective tissue beneath the mucosa of the oral floor.

Various etiologies have been suggested for the development of a plunging ranula, such as the presence of an ectopic sublingual gland on the cervical side of mylohyoid muscle; presence of a perforation or dehiscence in the mylohyoid muscle, through which the mucin may pass and accumulate in the submandibular space; and Iatrogenic causes, such as the recurrence of an oral ranula, a complication of surgeries involving the floor of the mouth (i.e. removal of a sialolith), transposition of the Warton duct, and dental implant placement.[2,3,4,5] Ductal variation has recently been identified as a predisposing factor for the development of a ranula.[6] The occurrence of bilateral ranulas and ranulas in siblings has suggested a role of genetic abnormalities in the pathogenesis of such cases.[7] In the present case, the patient presented with a swelling involving the right submandibular region but without any oral counterpart. Similar to previous case reports, ultrasonography was nondiagnostic in our case.[1] Magnetic resonance imaging is considered the most sensitive investigation for diagnosing ranulas. However, contrast enhanced computed tomography was sufficient for diagnosing the ranula in our case. Various treatment modalities for plunging ranulas have been described, such as sclerotherapy with OK-432, excision of the ranula alone, excision of the ranula along with the ipsilateral sublingual gland, excision of the ranula along with the ipsilateral sublingual and submandibular glands and excision of the ipsilateral sublingual gland with the evacuation of cystic contents.[8] Any modality that does not involve the excision of the offending sublingual or submandibular gland, may be a predisposing factor for recurrence. Excision of the offending salivary gland alone, along with the evacuation of cystic contents, is sufficient for treatment. However, this approach will not provide any specimen of the lesion for microscopic examination. In addition, the pathological tissue will be left in situ. Thus, excision of the ranula, along with the offending sublingual or submandibular gland will provide diagnostic accuracy and a reduced likelihood of recurrence. In the present case, we followed this line of treatment, which we recommend for such cases.

CONCLUSION

A plunging ranula presenting without its oral counterpart may be difficult to diagnose. A step-by-step investigative approach of investigations will aid in the accurate diagnosis of lesions. Surgical excision by using a cervical approach, along with the removal of the ipsilateral sublingual or submandibular gland is a favorable treatment option for a plunging ranula occurring without its oral counterpart.

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