

Case Report

Plunging Ranula Presenting as a Giant Anterior Cervical Cystic Mass: A Case Report and Literature Review

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Keywords

Ranula · Plunging ranula · Sublingual gland · Management

Abstract

Plunging ranula, a subtype of ranula, commonly presents as a submandibular or submental cystic mass without oral counterpart, and its clinical management remains challenging. Herein, the authors report an extremely rare case of 30-year-old female patient with plunging ranula involving the root of the left anterior neck.

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Introduction

Typically, ranula manifests as a bluish translucent cystic mass in the floor of the mouth, which is caused by ductal obstruction or injury of the sublingual gland resulting in mucous extravasation and accumulation. The pseudocyst of spilled mucus may spread through the mylohyoid muscle to the upper neck, subsequently showing as a soft, painless, and fluctuant swelling, which is also known as plunging ranula. According to clinical symptoms, lesions can be classified into three types: oral ranula, plunging ranula, and mixed ranula [1]. In this paper, the authors report an exceedingly rare case of plunging ranula presenting as a giant anterior cervical cystic mass.

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Case Report

The patient was a 30-year-old Chinese woman who presented with a 6-year history of painless swelling in the left submandibular region. During the last 3 months, the lesion began to expand down to the root of the neck.

Physical examination revealed an approximately 12 cm × 6 cm mass in the left anterior cervical region without any evidence of swelling in the floor of the mouth. It exhibited a cystic lesion without tenderness. By color Doppler ultrasound, there was a well-defined anechoic loculated fluid collection in the left superficial anterior cervical region, from the edge of the mandibular angle to the suprasternal fossa, whose size was approximately 10.98 cm × 1.49 cm × 4.92 cm (Fig. 1). There was poor internal sound transmission and visible soft tissue separation in the irregularly shaped mass without obvious blood flow signal in color-flow Doppler imaging. This examination indicated a potential diagnosis of the left plunging ranula, while lymphatic malformation needed to be excluded. In addition, the extent of the lesion was confirmed by contrast-enhanced computed tomography (CT) (Fig. 2). After fine-needle aspiration was performed in the clinic, the egg white-like liquid discharge was found. Taken together, all the evidence confirmed the typical report of ranula.

Via a transoral approach, the surgery was performed under general anesthesia for this patient. The procedure included complete excision of the enlarged left sublingual gland and aspiration of the mucus. Then, blunt dissection was performed toward the anterior margin of the submandibular gland, and no significant cystic fluid was observed. The left submandibular region was compressed for 3 days. Histopathological examination of the specimen after surgery revealed dilated ducts and the mucous pool due to partial rupture of the acinar and mucous overflow (Fig. 3). One week after surgery, the patient came back to the clinic for further consultation because of the swelling in the left anterior cervical region, which was almost the same size as before the surgery (Fig. 4). And 15 mL of light, red clear fluid, instead of mucus, was extracted from the carotid triangle of the left neck. The patient declared that the swelling gradually recovered on its own without any intervention. No signs of recurrence were discovered after a 6-month follow-up.

Discussion

The possible pathogenesis of plunging ranulas includes duct variation, ectopic sublingual gland near the mandibular hyoid muscle, perforation or laceration of the mandibular hyoid muscle, leading to mucus accumulation in the submandibular or submental space [2]. Besides, iatrogenic causes, such as recurrence of ranulas, surgical complications involving sialolithiasis, Warton catheter displacement, implant implantation, and other genetic factors could also induce pathological changes of the disease [3]. In this case, the lesion initially spread into the left submandibular region for almost 6 years. Subsequently, it extended to the lower neck without any apparent inducement in the last 3 months. The cystic expansion observed in the superficial cervical space may be attributed to the long-term effect of gravity on mucus accumulation.

Presenting as a cervical cystic swelling without intraoral involvement, plunging ranula usually causes misdiagnosis. Due to the high protein concentration, leading to specific signal intensities, imaging examinations could be conducive to differential diagnosis, and measure the range of the cyst as well. Formed by the posterior border or a deficiency in the anterior section of the mylohyoid muscle, tail-sign could be a distinguishing feature of MRI [4]. And ranula appears on CT as an approximately oval cyst with a uniform central attenuation zone



Fig. 1. Ultrasonography. An echo-free fluid formation in the left anterior cervical region with a craniocaudal extent of 10.98 cm and anteroposterior dimension of 1.49 cm (the arrow points).

and a hardly visible wall [5]. Notably, when compared to MRI and CT, color Doppler ultrasound is a more convenient and economical imaging modality that avoids radiation exposure. In this particular case, although the patient did not undergo an MRI, both ultrasonography and CT declared consistent results.

More than 80% of ranula are extravasation cysts, histologically characterized by partial rupture of the glandular vesicles and pseudocysts filled with mucus, without epithelial lining. And a vascular fibro-connective tissue wall containing chronic inflammatory cells and macrophages filled with mucin could be observed [6]. In this case, histological findings are corresponding to typical manifestation with dilated ducts and overflow of mucus. Pathological examination is also the gold standard for distinguishing squamous cell carcinoma and papillary cystadenocarcinoma of the sublingual gland.

Differential diagnoses of plunging ranula may include lymphatic malformation, venous malformation, thyroglossal duct cyst, branchial cleft cyst, and abscesses [1, 7]. Apart from history and clinical features, fine-needle puncture and analysis for its amylase, as a typical method, are vital to clarify the diagnosis. Since plunging ranula is caused by mucus extravasation in essence, puncture on the cervical swelling could draw out egg white-like mucus, from which the inflammatory cells, predominant histiocytes, high amylase, and protein content could be detected in the cystic fluid by biopsy and biochemical analysis. Different from plunging ranula, the presence of dark red blood in a fine-needle biopsy could indicate the diagnosis of venous malformation. Lymphatic malformation, cystic hygroma, shows clear yellowish watery liquid, and lymphocytes can be found by smear microscopic examination. The paracentesis of branchial cleft cyst removes clear yellow or brown fluid with or without cholesterol crystals, and dermoid cyst also has its characteristic secretion. However, paracentesis of thyroglossal duct cyst can also extract transparent, slightly turbid yellow, thin, or viscous fluid, which could be identified by negative staining of amylase and the movement of the mass with swallowing on the clinical examination.

Although therapeutic modalities for plunging ranulas remained controversial, their treatment options mainly include sclerotherapy and surgical treatment [8, 9]. Sclerotherapy, intracystic injection of OK-432, has been reported as an applicable, less-invasive treatment method for plunging ranulas. Being the only alternative therapy to surgery, injection of OK-432 has the benefit of requiring neither general anesthesia nor hospitalization, and having few and mostly self-limiting complications, but it showed the possibly deficient curative effect

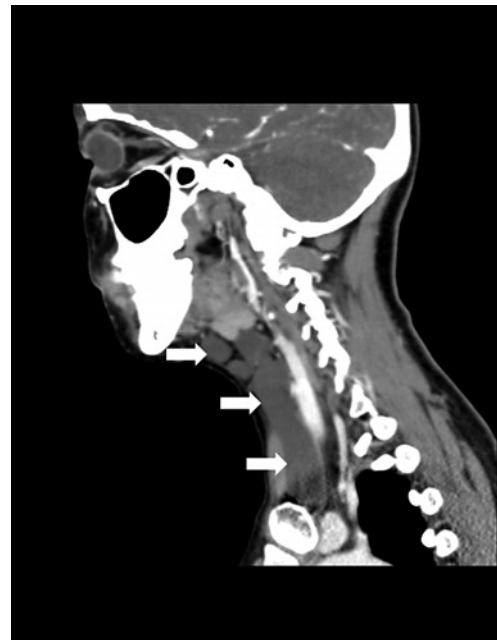


Fig. 2. Contrast-enhanced CT. A non-enhancing cystic lesion ranging from the submandibular region to the supraclavicular region (the arrows point).

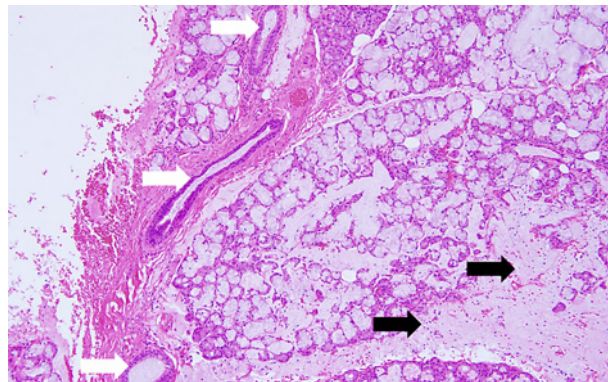


Fig. 3. Histological image. Sublingual gland with dilated ducts (the white arrows point) and the mucous pool (the black arrows point) due to partial rupture of the acinar and mucous overflow (HE, ×400).

(65.60%) that needs further treatment, such as surgery, to make up for it [10]. Without removing the sublingual gland, which could lead to recurrence, the treatment that excising the involved salivary gland and draining the cystic contents is sufficient. According to the different resection sites, the operation can be categorized into only resection of ranula, resection of the ranula and ipsilateral sublingual gland, and resection of ranula, ipsilateral sublingual gland, and submandibular gland. And the surgical approach is mainly divided into transoral and transcervical approaches, and the former is more common. The transcervical approach may not only leave skin scars and damage the marginal mandibular branch of the facial nerve, which is the most common complication, but also have some shortcomings such as limited visual field and insufficient exposure. For instance, the rate of recurrence and possible complications, such as lingual nerve dysfunction, is lower by transoral approach (3.50%) than transcervical approach (22.58%) [10]. Though 1 week after surgery, the swelling recurred in the left anterior neck with almost the same position and size as before surgery, no obvious mucus was extracted due to the ipsilateral sublingual gland radial excision in this case. There were no signs of recurrence in the follow-up period.



Fig. 4. Clinical photograph. The left anterior cervical swelling ranging from the submandibular triangle to the supraclavicular region (the arrow points).

Conclusion

In summary, plunging ranula is rare to extend from the edge of the mandibular angle to the root of the neck, presenting as a giant anterior cervical cystic mass. A comprehensive analysis of clinical presentation, imaging examination, and histologic findings is useful for the accurate diagnosis. Recently, surgery resection has been regarded as one radical cure method for ranula, including transoral removal of the ipsilateral sublingual gland and transcervical evacuation of the mucus at the same time. Of note, the crucial point in the treatment of plunging ranula is the radical excision of the sublingual gland. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see <https://doi.org/10.1159/000532010>).

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Statement of Ethics

This retrospective review of patient data did not require ethical approval in accordance with local guidelines. Written informed consent for publication was obtained from the patient of this case report and for any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Q.G. and Y.W. conceived and designed the study. F.J. and F.W. drafted the manuscript, and searched and reviewed the literatures. All authors have read and agreed to the published version of the manuscript. All authors contributed to the article and approved the submitted version.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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