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Case Report

Bilateral plunging ranula: A case report ☆

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ABSTRACT

Plunging ranula is the extravasation of saliva from the sublingual gland caused by trauma or obstruction of the duct, extending through a defect in the mylohyoid muscle into the submandibular gland. The prevalence of plunging ranula is estimated to be about 2.6 per 100,000 cases with mostly unilateral lesions. Bilateral plunging ranula are rare, with only a few cases reported. This case report describes an 11-year-old boy who was diagnosed with bilateral plunging ranula by radiologic examination of ultrasound and CT scan. The patient underwent extirpation surgery with the result of ranula on histopathologic examination. Six-month follow-up after extirpation, the patient was asymptomatic and no sign of recurrence.

Introduction

Plunging ranula or cervical ranula is a nonepithelial salivary gland cyst that forms due to obstruction or trauma in the sublingual salivary gland duct, leading to mucus extravasation and herniation through the mylohyoid muscle within the sublingual gland and surrounding areas, extending to the submandibular gland [1,2]. The global prevalence of bilateral ranula is unknown; 1 study estimated it to be around 2.6 per 100,000 people per year. Ranula can occur at any age, but it is most commonly found in children and young adults, with a peak incidence in the second decade of life. Plunging ranula is commonly found on the left side only. Bilateral plunging ranulas are rare, with only 25 cases reported in the literature [1–3].

The etiology of ranula lesions is believed to be related to incomplete blockage of the sublingual gland duct, leading to the development of a retention cyst by the epithelium or due to damage to the duct or underlying tissue of the sublingual gland caused by direct trauma, causing mucus to leak into the surrounding area and form a nonepithelial cyst [1,3]. Ranula

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can be diagnosed through medical history and physical examination, identifying a mass in the neck accompanied by swelling in the floor of the mouth that may extend to the submental area, contralateral neck, nasopharyngeal region, including the base of the skull, retropharyngeal area, and upper mediastinum. Supporting examinations such as CT scans and MRIs can aid in the diagnosis of ranula. The first-line treatment for ranula is surgical management [1,2]. In this case report, the author will present a case of an 11-year-old boy with a mass under the tongue extended to the mandible as bilateral plunging ranula. The patient underwent surgical extirpation of the ranula and there was no recurrence or complications.

Case report

An 11-year-old male patient presented with chief complaint of a lump under his tongue 2 months before his hospital admission. The lump was located in the midline beneath the tongue, initially the size of a marble, bluish-purple in color, soft to fluctuant, and painless. The lump had reportedly grown to the size of an egg and was accompanied by a noticeable enlargement under the mid-jaw area 3 weeks prior to admission. The presence of the lump caused discomfort for the patient and affected his speech. The patient did not experience difficulties with swallowing, nausea, vomiting, fever, headaches, shortness of breath, or drastic weight loss. He was the first child, born at full term, with no family history of malignancy, similar complaints in the family, trauma, systemic diseases, or prior surgeries in the oral region. Physical examination revealed that the child was alert and oriented, with stable vital signs. Local examination indicated 2 related lumps under the tongue, bluish-purple in color, fluctuating in consistency, approximately 5 cm, immobile, and painless (Fig. 1).

Ultrasonography (USG) examination shows the presence of a cystic mass suggestive of a ranula, characterized by a hypoechoic mass with fine internal echo components. The mass has well-defined, regular edges, thin walls, and mea-

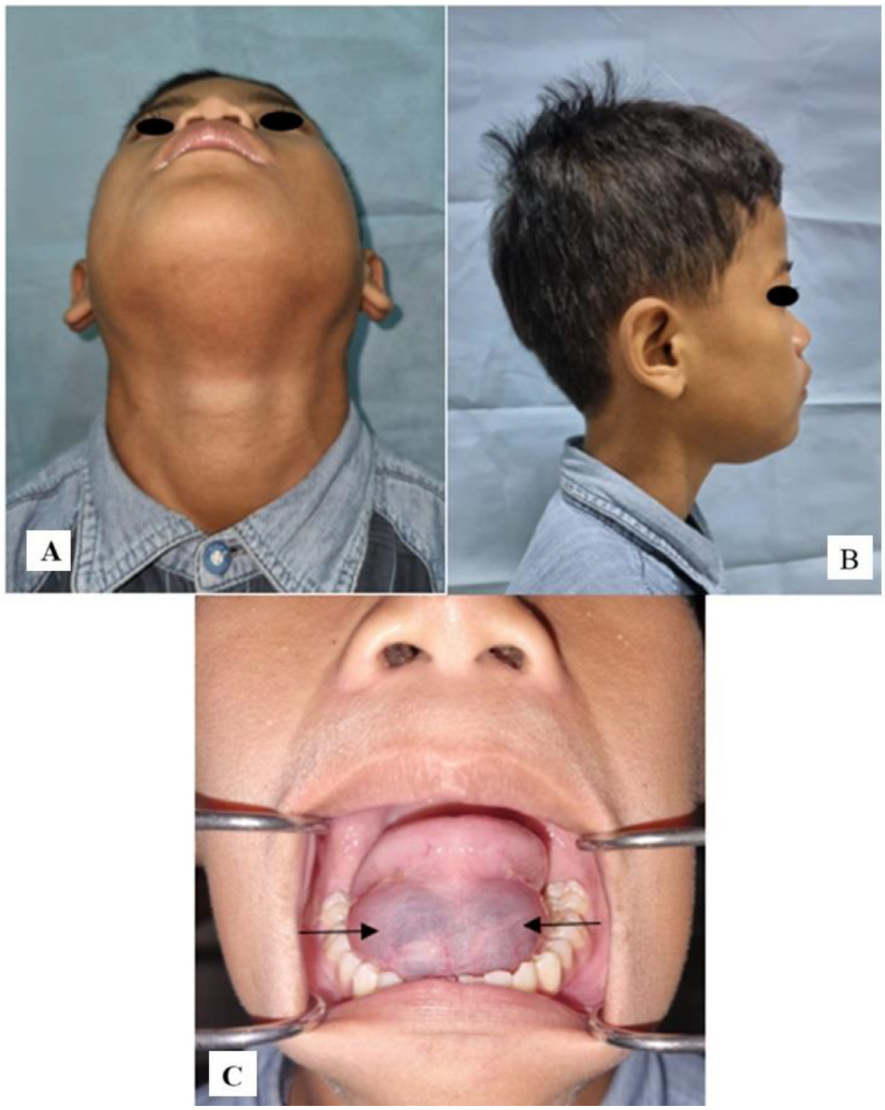

Fig. 1 – Clinical image. (A) Anterior and (B) lateral of bilateral mass in the floor of the mouth and submandibular area. (C) Bluish-purple color, fluctuating, and immobile mass (arrow).

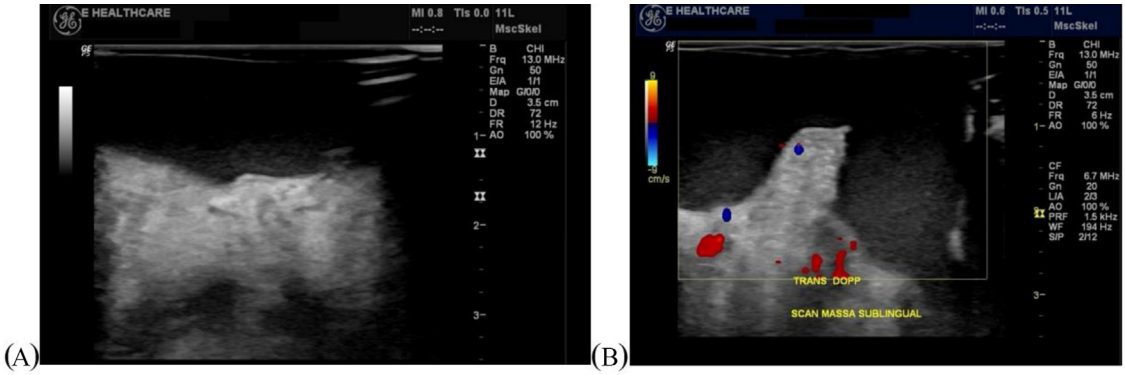

Fig. 2 – Grey scale (A) and Color Doppler ultrasound (B) of the sublingual area show thin-walled cystic mass containing fine internal echo in the sublingual region without mass vascularization.

sures 5.5 × 3 × 2 cm in the sublingual region. Color Doppler examination does not reveal any mass vascularization (Fig. 2).

A CT scan of the head and neck with contrast reveals a hypodense mass with well-defined borders and regular margins, largest measures approximately 2.08 × 5.78 × 2.71 cm in left side, located in the bilateral sublingual space between the mylohyoid muscle and genioglossus muscle. This mass extends into the bilateral submandibular space, particularly on the left side, presenting a "tail sign." Postcontrast imaging shows no significant enhancement or uptake (Hounsfield Units precontrast: 3-10, postcontrast: 6-12). The findings from the CT scan suggest the presence of a cystic mass due to bilateral plunging ranulas (Fig. 3).

Based on the clinical, ultrasound, and CT scan findings, treatment was planned for extirpation ranula. The lesion is approached intraorally through a mucosal incision followed by drainage of fluid with a syringe and then extirpation of the ranula (Fig. 4).

The extirpated specimen was sent for histopathological examination. Tissue weighing 1.3 grams, with a brownish-white color, was sent for macroscopic and microscopic examination. The histopathological examination of the sublingual area covered with squamous stratified epithelium within normal limits nuclei. The subepithelial connective tissue is edematous, lined with inflammatory cells and lymphocytes with dilated blood vessels. Additionally, tubular seromucous gland structures lined with ciliated epithelium and normal nuclear appearance were seen. Muscle tissue was also observed, and there were no signs of malignancy. Among these, structures resembling cysts without an overlying epithelium were noted. The lumen contained extracellular mucin with macrophage cells present. The microscopic examination concluded the presence of a sublingual ranula (Fig. 5).

There were no specific complaints postoperatively and the patient was discharged 1 days postoperatively. The patient had no complaints of lumps, speech impairment, and no recurrence during the follow-up period.

Discussion

A ranula is a cavity filled with mucus or a mucocele located at the floor of the mouth, associated with the sublingual glands. The term "ranula" comes from the Latin word "rana," meaning frog, as the swelling resembles a water-filled sac or the translucent underside of a frog's belly. A ranula is characterized by the presence of dome-shaped, fluctuating, and tense vesicles, typically large (over 2 cm), and sometimes bluish in color. It is most commonly found in the lateral portion of the floor of the mouth [3,4].

A ranula can be classified into simple and plunging (diving/cervical) forms. A simple ranula is confined to the sublingual space, while a plunging ranula can extend from that area and is often found as swelling in the neck. Plunging ranulas are typically observed alongside simple ranulas [3,4]. Approximately 45% of cases present initial symptoms of a ranula as swelling in the oral cavity. Plunging ranulas are associated with oral cavity swelling in 34% of cases, whereas only 21% of cases occur without oral involvement [4].

Plunging ranula is the extravasation of saliva from the sublingual gland due to trauma or obstruction of the duct. Fluid from the obstructed gland escapes between the fascial plane and muscles at the base of the tongue, moving into the submandibular space. Plunging ranula occurs when there is pressure from mucin fluid entering through a perforation in the mylohyoid muscle within the submandibular cavity. Rupture of the excretory duct leads to extravasation and accumulation of saliva in the surrounding connective tissue, resulting in the formation of a pseudocyst without an epithelial lining [3,4]. Therefore, it can be concluded that there are 3 mechanisms for the occurrence of plunging ranula in the neck: the presence of sublingual glands projected through the mylohyoid muscle, ectopic sublingual glands located on the cervical side of the mylohyoid muscle explaining the possibility of plunging ranula without oral components, cysts penetrating through the mylohyoid muscle, and the ducts of the sublingual gland merging with the submandibular gland, allowing access to the neck behind the mylohyoid muscle [4].

Ranula occurs due to partial obstruction of the sublingual gland, resulting in the formation of epithelial retention cysts, and this condition is found in less than 10% of cases [3]. Meanwhile, post-traumatic ranula is caused by trauma to the sublingual gland, leading to a mucous escape reaction (MER) and the development of a pseudocyst [4]. Congenital ranula may arise from ostial adhesions or imperforate salivary

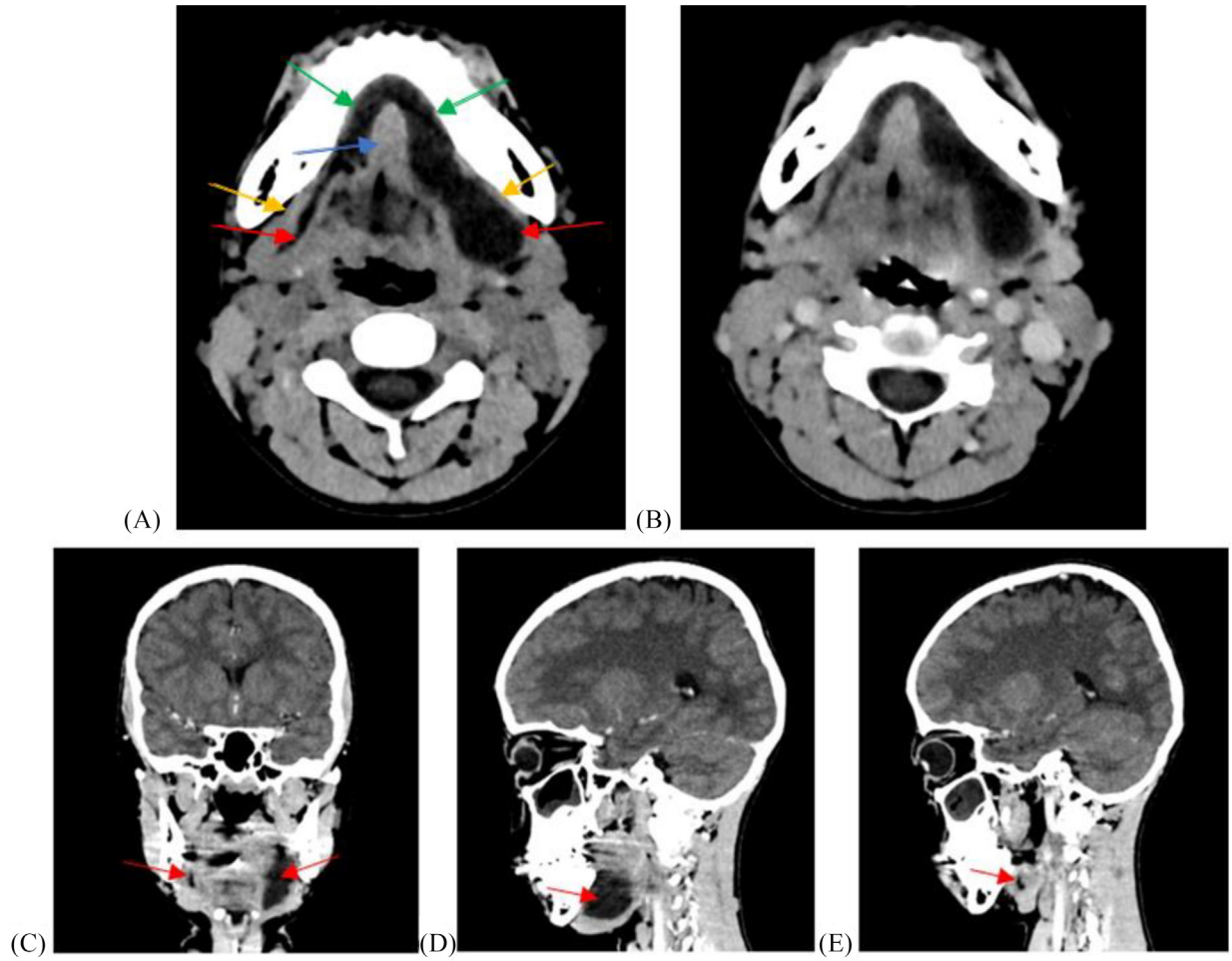

Fig. 3 – The axial precontrast (A), axial postcontrast (B), and reformatted coronal (C), left sagittal (D), and right sagittal (E) computed tomography (CT) images show nonenhancing bilateral sublingual mass (green arrow) that extends into the bilateral submandibular region (red arrow), passing through the mylohyoid (orange arrow) and genioglossus muscles (blue arrow).

ducts and is a very rare condition that can spontaneously resolve [3,4].

The global prevalence of plunging ranulas is unknown. One study in New Zealand showed an incidence rate is 2.6 per 100,000 person-years. Ranula can occur in patients ranging from 3 years to 61 years of age, with children and young adults in their second and third decades of life being more frequently affected. The plunging ranula variant often appears later in the third decade. The male-to-female ratio for ranula occurrence is 1:1.3. Plunging ranula is generally found on the left side only. Bilateral plunging ranulas are rare, with only 25 cases reported in the literature [1-4]. This case is an 11-year-old child with bilateral plunging ranula, which is a rare case.

Diagnosis of plunging ranula is established through medical history, physical examination, and supportive tests. Cervical ranulas are often asymptomatic, presenting as progressively enlarging and fluctuating masses. Case reports indicate that ranulas can vary in size from 4 to 10 cm. The surrounding skin is usually intact. The mass is fluctuating, movable, and painless, unrelated to the thyroid gland or lymph nodes. Large ranulas may cause dysphagia or airway obstruction [1,3]. In this case the patient showed a blue-purple lump under the tongue which had enlarged to the mandible. On physical examination the lump is fluctuating and painless.

Ultrasound is typically inconclusive in demonstrating the sublingual glands in relation to their location. It aids in detecting cystic lesions in the submandibular region. The advantage of this examination is that it is noninvasive and readily available in healthcare facilities. Additionally, the technique can assess the status of the mylohyoid muscle and differentiate between a plunging ranula, which enters through the posterior border of the mylohyoid muscle into the submental space, and a rarer type of ranula that experiences herniation through a defect in the mylohyoid muscle into the submental cavity [1,3,5]. In this case, the ultrasound examination showed well-defined hypoechoic cystic mass with fine internal echo without mass vascularization in the sublingual area.

Meanwhile, the CT scan examination reveals an ovoid cyst with a homogeneous central attenuation of 10-20 HU. The cyst wall is usually very thin or not visible, and the lesion is lo-

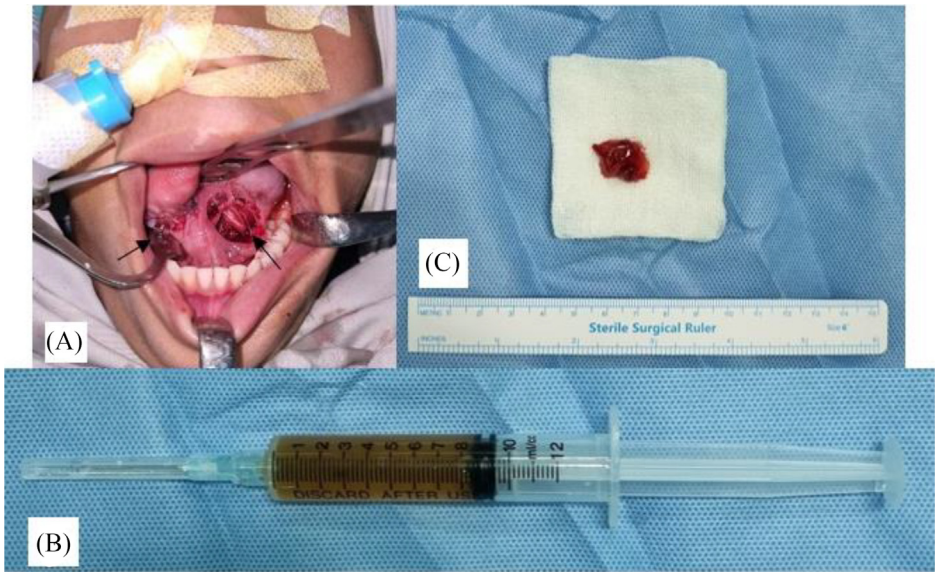

Fig. 4 – Transoral surgical approach. (A) Extirpation of bilateral ranula (arrow). (B) Yellowish fluid of ranula. (C) Macroscopic ranula.

cated laterally to the genioglossus muscle and above the mylohyoid muscle. A ranula may also extend to the anterior area, behind the mandibular symphysis, and above the genial muscle. A plunging ranula often infiltrates the surrounding tissue and extends inferiorly and dorsally toward the submandibular gland region, although it rarely affects the gland internally. A characteristic appearance of a small tail widening in the sublingual cavity can be observed [1,3]. This appearance aligns with cases presenting a "tail sign" due to interrupted continuity of the cervical portion of the ranula within the sublingual cavity, which is considered pathognomonic for a plunging ranula.

MRI is the most sensitive examination for evaluating the sublingual glands. The MRI appearance of a ranula is characterized by high fluid content, which results in low density on T1-weighted images and high signal intensity on T2-weighted images. Therefore, the appearance of a plunging ranula can resemble lymphangioma, lateral thyroglossal duct cysts, and inflamed lymph nodes. Additionally, the signal intensity of the ranula may also depend on the protein content or concentration [3].

Histopathological, a cervical ranula resembles the picture of mucus extravasation, with a biopsy of the lateral neck showing an amorphous substance with sparse inflammatory cells and a predominance of histiocytes, staining positively for mucin. Additionally, peripheral fibrosis may be found, lined with nonkeratinized stratified squamous epithelium, alongside inflammatory cells, mucin, and mucinophages visible in hematoxylin-eosin staining. Biochemical analysis indicates viscous fluid accompanied by mucin, inflammatory cells, proteins, and salivary amylase, suggesting its source is the salivary glands [3,6]. Differential diagnosis for cervical ranula includes thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, submandibular sialadenitis, intramuscular hemangioma, cystic or neoplastic thyroid disease, infected cervical lymphadenopathy, hematoma, lipoma, laryngocele, and dermoid cysts. Dermoid and epidermoid cysts contain keratin and protein-rich and/or lipid substances, while lipomas demonstrate low attenuation. This can be differentiated from plunging ranula by their high mucin fluid content. Thyroglossal duct cysts are located in the midline, while branchial cleft cysts typically lie medially to the anterior border of the sternocleidomastoid muscle. Cystic hygroma can be differentiated from plunging ranula as it often contains septa [4]. In this case, histopathological examination reveals a cystic structure without epithelium, containing extracellular mucin in the lumen with an appropriate number of inflammatory cells corresponding to the presentation of a sublingual ranula.

Until now there is no consensus that explains the management of intra oral ranula or plunging ranula specifically, but there are several management options in the form of surgical and nonsurgical methods. Nonsurgical management performed as ranula management is sclerotherapy using OK-432 (picibanil). Sclerotherapy has several advantages such as the administration of this injection causes minimal pain and short procedure duration so that it can be tolerated by children, without local anesthesia, secondary infection and bleeding are rare, nerve injury and cosmetic problems can be avoided. The disadvantage of this method is that the success with 1 injection is only 33.4%, so multiple injections are required [5].

Surgical therapies include incision and drainage, marsupialization, ranula extirpation, sublingual gland extirpation with intra oral approach [6].

Incision and drainage, and marsupialization are the techniques of choice for the management of ranula smaller than 2 cm [7]. This technique can be performed under topical anesthesia and the procedure does not require a long time and there is no tissue damage or severe inflammation. However, the disadvantages of this technique are the high recurrence

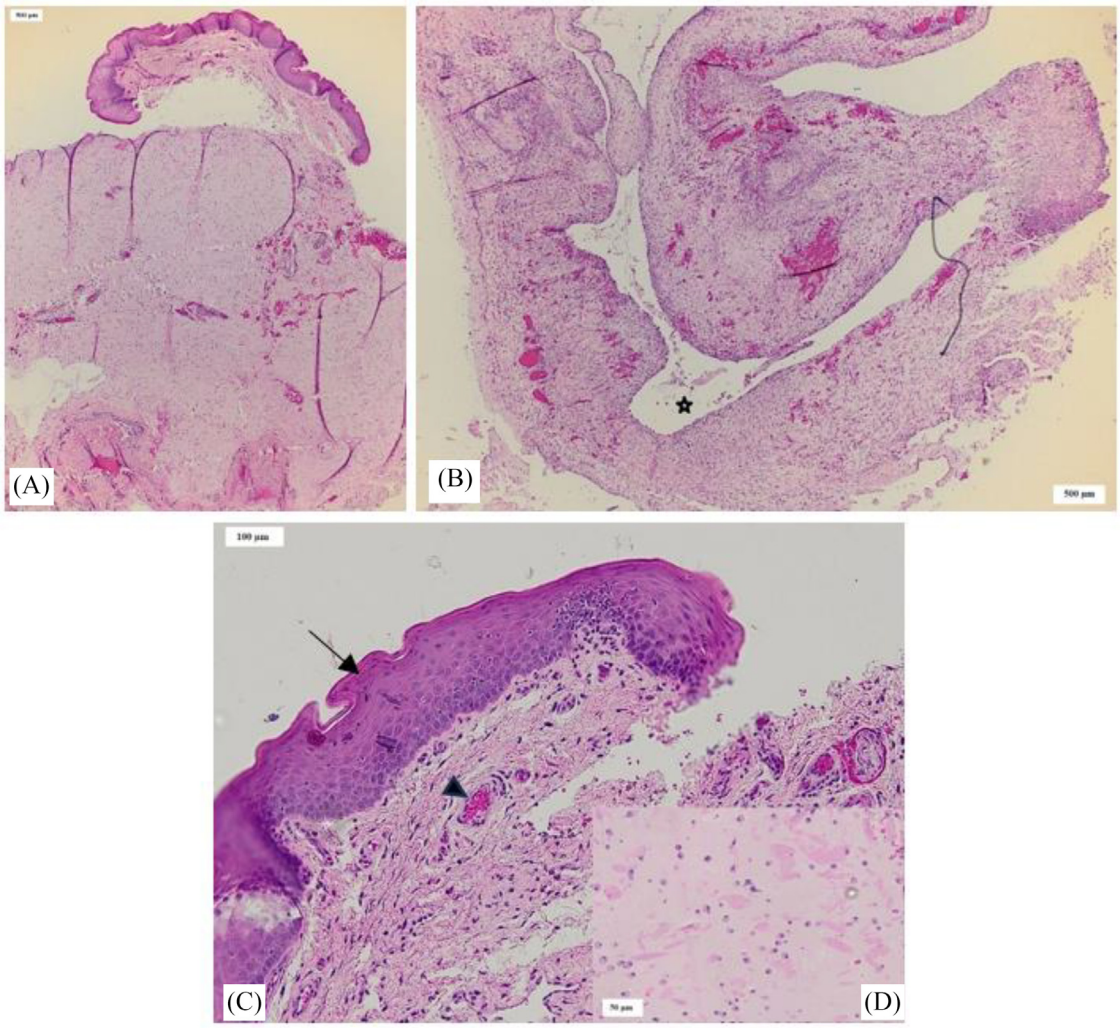

Fig. 5 – Histopathological features of ranula. (A) Histopathological features of ranula, magnification 40x. (B) Cyst formation without epithelial layered, filled by extracellular mucin (asterisk), magnification 40x. (C) Squamous stratified epithelium with normal nuclei (arrow), subepithelial tissue consists of fibrocollagenous stroma with vascular dilatation (arrow head), magnification 100x. (D, insert) Inflammatory infiltrates, which include plasma cells, lymphocytes, and macrophages within edematous stroma, magnification 400x.

rate within 6 weeks to 12 months after surgery and the difficulty in biopsy sampling. Ranula extirpation has a fairly low recurrence rate with a possible risk of paraesthesia of the tongue and infection. Sublingual gland extirpation with intra oral approach has the lowest recurrence rate but has the risk of lingual nerve injury, Wharton's duct, hematoma, and bleeding. The reported recurrence rates after various treatment modalities are incision and drainage (70%-100%), marsupialization (36.4%-80%), excision of ranula only (18.7%-85%), and excision of ranula along with sublingual salivary gland (0%-3.8%) [1,3,5,6]. In this case, the patient performed tumor incision and then drainage of the cystic fluid followed by ranula extirpation with the aim of minimizing the risk of complications and recurrence rates. After 6 months of surgery the patient no longer feels a lump on the tongue, speech disorders, or surgical complications such as tongue paraesthesia and infection.

Conclusion

Bilateral plunging ranula is a rare condition. In this case report, the author presents a case of bilateral plunging ranula in a child with both oral and cervical components. Ultrasound, CT scans of the head and neck, and histopathologic examination assisted in supporting the diagnosis. Surgical extirpation of the ranula was performed as the treatment for this case.

Patient consent

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

Author contributions

All the author contribution to Conceptualization, Validation, Writing Original Draft, Writing Review and Editing, Visualization, and Supervision.

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