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CASE REPORT

Intraoral approach for the excision of a submandibular epidermoid cyst

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ABSTRACT

Epidermoid cysts are rare, slow-growing, benign, developmental cysts that arose from abnormally situated ectodermal tissue. They represent less than 0.01% of all oral cavity cysts and in particular those of the submandibular region are extremely rare. A detailed analysis of the patient's clinical features and a study of the extent, nature and exact location of the lesion provided by MRI are essential for appropriate preoperative surgical planning. The recommended treatment for epidermoid cysts of the mouth floor is surgical enucleation. The position of the mylohyoid muscle and the size of the cysts generally dictate the surgical approach. The authors present and discuss the management of the first case in literature of an epidermoid cyst extending in submandibular region through the mylohyoid muscle removed by an intraoral surgical approach without any resultant morbidities and with no evidence of recurrence up to the one-year follow-up. With this approach, we managed to avoid visible cutaneous scars provided the patient with excellent cosmetic and functional outcomes.

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KEY WORDS: Epidermal cyst; Neck muscles; Case reports.

Epidermoid cysts of the floor of the mouth are rare lesions attributable to the entrapment and subsequent growth of epithelial cells during the midline fusion of the first and second branchial arches in the third and fourth intrauterine weeks.¹ Such lesions may occur anywhere in the body, although about 7% are found in the head and neck and 1.6% are within the oral cavity.^{2,3} The midline region of the mouth floor is most affected, but cysts may also develop in the submandibular region. Usually, the first clinical manifestations appear between 15 and 35 years.⁴ These masses are soft, painless, and slow-growing, and they contain cystic or keratinous fluid.¹ If large enough, the cyst can potentially compromise the airway, speech, and swallowing.⁵ Histologically, the cysts are coated with stratified squamous epithelium.⁶ Preoperative diagnostic tests include ultrasound (US) and magnetic resonance imaging (MRI), but only histology is definitive.⁷ Recommended treatment is surgical excision through intraoral or extraoral approach, depending on the lesion's size and

location.^{5,8} The authors present the first case of a submandibular epidermoid cyst extending through the mylohyoid muscle removed by an intraoral approach.

Case report

A 19-year-old male was referred to our Division of Maxillofacial Surgery, University of Turin, Italy, for treatment of a painless, slowly enlarging swelling in the right submandibular region (Figure 1). He did not report any prior episode of inflammation, dysphagia, odynophagia, or fever. There was no previous history of surgery or trauma to the oral cavity or neck. Clinical examinations indicated that the swelling was mobile, soft to firm, painless, and non-pulsatile. The overlying skin was normal in color and texture and was pinchable. There were no visible changes to the oral floor. Preoperative MRI revealed a well-circumscribed oval mass extending in the right submandibular region. The mass displaced the ipsilateral submandibular



Figure 1.—Preoperative images show a right submandibular swelling: A) frontal view; and B) lateral view.

gland laterally (Figure 2A). Patient anamnesis, clinical examination and the radiological findings suggested an embryonic developmental cyst (dermoid or epidermoid). We decided to enucleate the lesion with the patient under

general anesthesia using an intraoral surgical approach. We incised the lingual vestibule, next to the lingual frenulum, and extended the incision along the ventral surface of the tongue as far as was necessary. Wharton's duct and the lingual nerve were identified and avoided. The anterior upper pole of the lesion laid in the midline of the mouth floor below the genioglossus muscle, while the posterior pole laid below the mylohyoid muscle. The cyst was bluntly dissected from surrounding tissue and the mylohyoid muscle was carefully displaced to reach the posterior cyst pole. The mass was completely enucleated without interrupting the muscle (Figure 3). The excised specimen assumed a dimension of 5×3×7 cm (Figure 4). Then the incision was sutured in layers. The specimen was sent for anatomopathological examination, and the microscopic aspects confirmed a cystic-type lesion lined with stratified

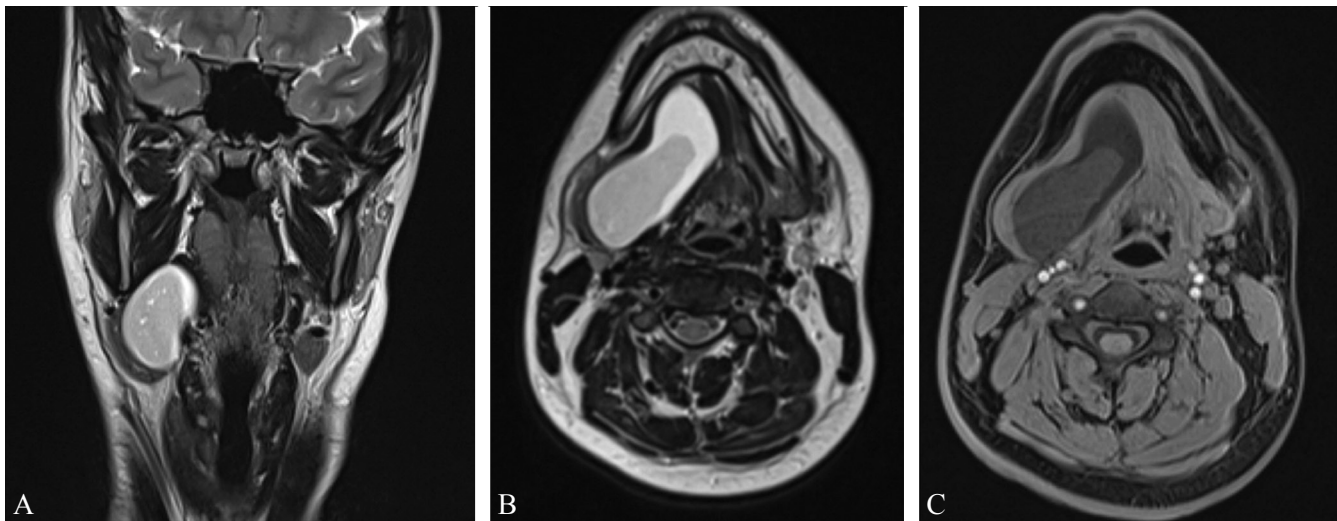


Figure 2.—Preoperative MRI: A) T2-weighted-coronal scan – well-defined cystic mass with hyperintense signal; B) hyperintense T2-weighted axial scan; and C) hypointense T1-Weight Axial scan: anterior upper cyst pole laying in the midline of the floor of the mouth.

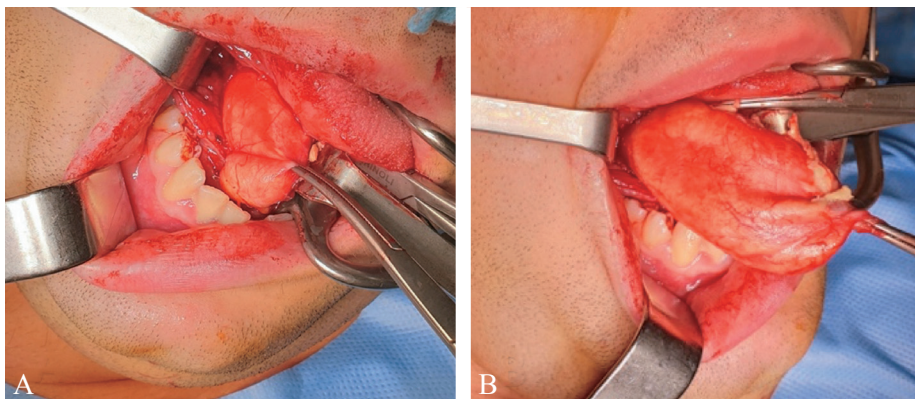


Figure 3.—Intraoperative images: A) lateral vestibule incision; and B) enucleation of the cyst.

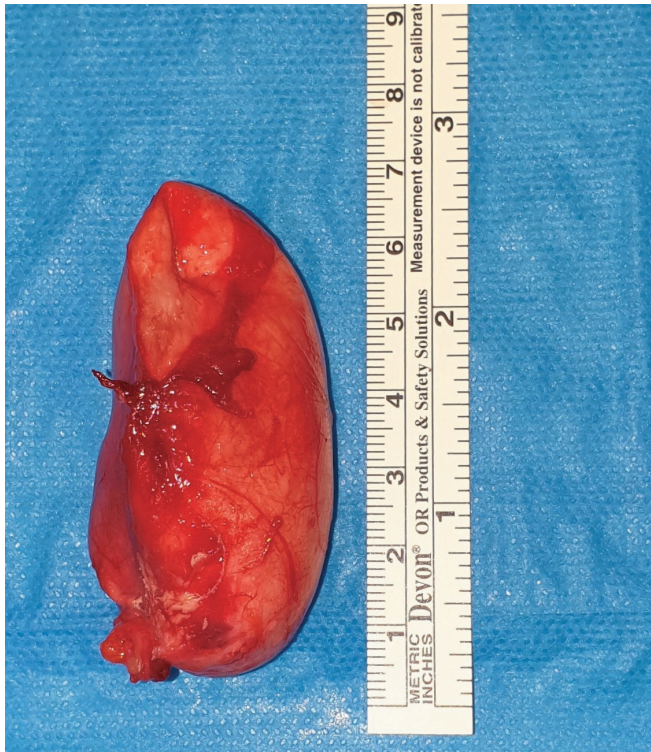


Figure 4.—Macroscopic image of the surgical specimen measured 5×3×7 cm.

squamous epithelium and filled with keratin, suggestive of epidermoid cyst (Figure 5). The postoperative course was uneventful, and the patient was discharged the day after surgery. There was no evidence of recurrence up to the 1-year follow-up. The aesthetic results were excellent and judged satisfactory by the patient (Figure 6).

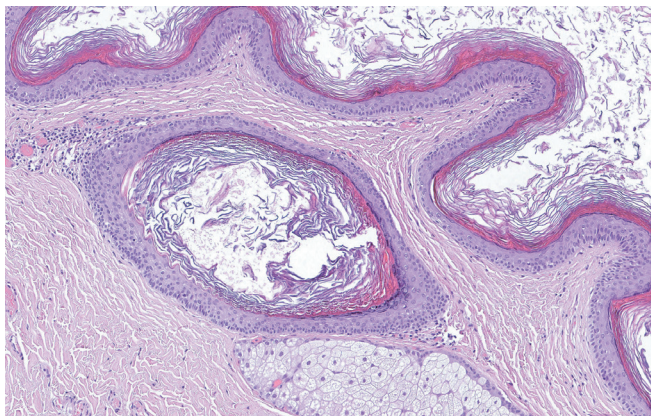


Figure 5.—Histopathological characteristics of the surgical specimen: cystic-type lesion lined with stratified squamous epithelium. H&E original magnification 30×.



Figure 6.—One-year postoperative images of the patient: A) frontal view; and B) lateral view.

Discussion

Dermoid cysts in the floor of the mouth are uncommon entities. They constitute less than 0.01% of all oral cavity cysts. They are most common along the midline of the oral floor (78%) and those of the submandibular region are especially rare 16% involve more than one space and only 6% are in the submandibular space.^{1, 7, 9} Dermoid cysts may be classified as congenital or acquired, even if there is no difference between the two on presentation or histologically. Congenital cysts are dysembryogenetic lesions that arise from ectodermic elements entrapped during the midline fusion of the first and second branchial arches between the third and fourth weeks of intrauterine life.¹⁰ Acquired forms are derived from either iatrogenic or traumatic inclusion of epithelium and skin appendages.¹¹ Meyer *et al.*⁶ classified: 1) floor of mouth dermoid cyst into three variants based on histopathology which was epidermoid, dermoid and teratoid variants – epidermoid cyst is lined with simple squamous epithelium with a fibrous wall and no adnexal structures; 2) true dermoid cyst – an epithelial-lined cavity with keratinization and with skin appendages; 3) teratoid cyst – lined with a range of epithelia, from simple squamous epithelium to ciliated respiratory type, containing derivatives of ectoderm, mesoderm, and endoderm.⁶ Of the three variants, epidermoid is the most common, followed by dermoid and teratoid, respectively. There is a bimodal age distribution that can first present during infancy and later during puberty when hormone change leads to increased sebum production, but they can occur at any age. Depending on the anatomical relation to the muscles of the floor of the mouth, epidermoid cysts are classified as supramylohyoid (above mylohyoid and genioglossus muscles) presented as swelling in the floor of the mouth; inframylohyoid (between the mylohyoid and geniohyoid muscles) presented as swelling below the chin, and peri- and trans-mylohyoid.^{4, 12} In the presented case, the anatomical location of the cyst was suggestive of an origin in the medial region, followed by lateral ex-

TABLE I.—*Differential diagnosis of submandibular masses.*

Inflammatory masses	Tuberculous lymphadenitis, granulomatous lymphadenitis, abscess
Mucous extravasation phenomena	Ranula
Congenital masses	Brachial cleft cyst, epidermoid, dermoid and teratoid cyst, cervical lymphoepithelial cyst
Benign masses	Lipoma, pleomorphic adenoma
Malignant masses	Lymphoma, adenoid cystic carcinoma

pansion into the neck through the mylohyoid muscle. This lateral expansion was also reported in other cases of lateral congenital developmental cysts.¹³ Kudoh *et al.*⁹ reported the only case of an alleged epidermoid cyst arising in submandibular region. When approaching a cystic lesion of the submandibular region, several conditions should be considered for the differential diagnosis,¹⁴ as summarized in Table I. Imaging plays an important role in making a correct diagnosis and a reliable preoperative surgical planning. MRI is the gold standard in assessing the extent, nature, and exact location of the lesion in relation to the surrounding neck structures, given its excellent soft tissue contrast and multiplanar imaging capability. Epidermoid cysts are hypointense on T1-weighted images and hyperintense on T2-weighted images, following the signal intensity of fluid¹⁵ as shown in Figure 2. The definitive diagnosis is always given by the postoperative histological examination. Surgical enucleation is the only effective treatment for epidermoid cysts of the mouth floor. The position of the mylohyoid muscle and the size of the cysts dictate the surgical approach: the intraoral approach is recommended for a small to a medium sublingual cyst which is less than 6 cm in size and above the mylohyoid muscle, whereas an extraoral approach is appropriate for large cysts over 6 cm located or transgress below the mylohyoid muscle.^{11, 14} The cases of infra-mylohyoid submandibular epidermoid cysts reported in literature have all been treated through extraoral surgical approaches, with a latero-cervical incision^{1, 7, 9, 14} Gulati *et al.*¹² described a case of “potentially fatal supramylohyoid sublingual epidermoid cyst” treated by a combined intraoral and extraoral approach, while Klibngern *et al.*⁵ reported the case of a 26-year-old girl with a large sublingual cyst extended above the mylohyoid muscle removed by the intraoral approach to avoid aesthetic problems. However, during the excision of the cyst it was decided to remove the sublingual gland to increase the visibility on part of Wharton’s duct, lingual nerve, and medial pterygoid muscle, thus sacrificing an important anatomical structure. Ohta *et al.*¹¹ described a case of sublingual dermoid cyst extending to the mylohyoid muscle removed by intraoral surgical approach. They reported that an appropriate traction and countertraction

of the cyst might help to remove the cyst from mylohyoid muscle without extraoral incision. In the case presented, following a careful preoperative planning, we decided for the intraoral approach to reach the posterior cyst pole of the cyst without damaging the mylohyoid muscle and other surrounding anatomical structures. With this approach we managed to avoid visible cutaneous scars, provided the patient with excellent cosmetic and functional outcomes.

Conclusions

In conclusion, epidermoid cysts of the submandibular region are extremely rare. A detailed analysis of the patient’s clinical features and a study of the extent, nature and exact location of the lesion provided by MRI are essential for appropriate preoperative surgical planning. This is the first case of an epidermoid cyst extending in submandibular region through the mylohyoid muscle removed by an intraoral surgical approach without any resultant morbidities and with no evidence of recurrence up to the one-year follow-up. In agreement with other authors^{5, 11} we believe that, if properly performed and in selected cases, intraoral surgical enucleation of these lesions is a feasible and useful procedure with an excellent cosmetic outcome.

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