Different Radiologic Appearances of Giant Epidermoid Cysts at the Floor of the Mouth: Three Case Reports

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ABSTRACT

Epidermoid and dermoid cysts are benign lesions, which may occur anywhere on the body. Approximately 7% of these lesions occur in the head and neck region and 1.6% in the oral cavity. We present three cases of giant epidermoid cysts located on the floor of the mouth. Case 1 was a 22-year-old man referred to our clinic with a 10-year history of a submental mass, which began growing and causing pain in the throat. His physical examination revealed masses in both the submental and the sublingual regions. Magnetic resonance imaging (MRI) with contrast revealed a well-circumscribed isohypointense cystic mass with a 'sack of marbles' appearance. Case 2 involved a 23-year-old female referred to our clinic with difficulty in chewing and swallowing solid food for 1 week previously. Her physical examination revealed a sublingual mass displacing the tongue upward. Her MRI revealed a heterogeneous hyperintense cystic mass in the sublingual region. Case 3 was a 28-year-old man referred to our clinic with submental swelling, difficulty in breathing and swallowing, decreased tongue movements and snoring. His computerized tomography indicated a homogenous cystic mass in the submental region. The excision of the masses with an extraoral approach was performed in all the three cases. His histopathological examination revealed epidermoid cyst. In summary, epidermoid cysts may assume various appearances at radiological imaging.

Keywords: Dermoid cysts, epidermoid cysts, sack of marbles.

INTRODUCTION

Epidermoid and dermoid cysts are benign lesions, which may occur anywhere on the body. Approximately 7% of these lesions occur in the head and neck regions and 1.6% in the oral cavity (1). In 1955, Meyer divided dermoid cysts into three histological types. If the wall of the cyst contains only squamous epithelium, this is known as epidermoid cyst. Dermoid cysts contain skin adnexa, and teratoid cysts contain all three germ layers (2, 3). The diagnosis depends on clinical findings, and the imaging and histological examinations. The epidermoid cysts may produce different appearances at imaging scans. The treatment is surgery, which can be performed using the intraoral or extraoral approaches. The choice of surgical approach depends on the location and size of the mass (4). We described three cases of giant epidermoid cysts on the floor of the mouth.

CLINICAL REPORTS

Case 1

A 22-year-old man presented with a 10-year history of a submental mass which had begun growing 2 months previously. He also reported pain in his throat and difficulty in chewing, speaking and swallowing. His physical examination revealed a non-tender, fluctuant, soft, mobile mass in the submental region (Fig. 1A). At oral examination, a mass was observed in his sublingual region, displacing the tongue upwards (Fig. 1B). His ultrasonography revealed a hypoechoic cystic area containing multiple hyperechoic structures. His magnetic

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Fig. 1: The appearance of cystic mass. (A) At submental region. (B) At sublingual region.

resonance imaging (MRI) with contrast revealed a wellcircumscribed isohypointense cystic mass, 7.5×6.5 cm in diameter, resembling a 'sack of marbles' in appearance due to the coalescence of fat into the small nodules. The gadolinium-enhanced images also showed a cystic mass with peripheral rim-like contrast uptake (Fig. 2). The mass was totally excised using an extraoral approach. His histopathological examination revealed portions of an encapsulated specimen lined with a multilayered epidermal-type benign squamous epithelium (Fig. 3) and containing keratin in the lumen (Fig. 4). No adnexal structures in the cyst wall or evidence of malignancy was observed. The histopathological diagnosis of his mass was epidermoid cysts.

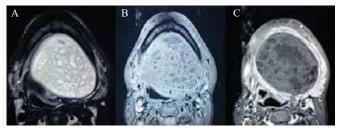


Fig. 2: (A) Axial T2-weighted images show cytic hyperintense mass with sack of marbles (coalescence of fat into the small nodules) in the floor of the mouth. (B) Axial T1-weighted images show izohypointense cystic mass in the mouth base. (C) Gadolinium-enhanced axial T1-weighted images show cystic mass with peripheral rim-like contrast enhanced.

Case 2

A 23-year-old female presented with difficulty in chewing and swallowing solid food over the previous week. She also had a 1-month history of a non-tender mass in the submental region. There was no history of trauma or surgery to the oral cavity or neck. Her physical examination revealed a sublingual mass displacing the tongue upward. Her MRI revealed a heterogeneous hyperintense cystic mass in the sublingual region, $4 \times 4 \times 5$ cm in size (Fig. 5). A surgical excision was performed on her under

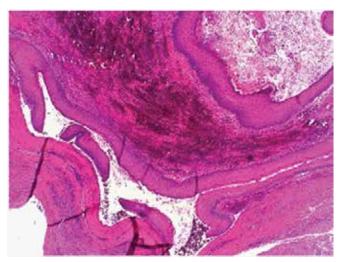


Fig. 3: (HE, ×50) Histological section shows the cyst containing keratinous material arranged in laminated layers and is lined by stratified squamous epithelium.

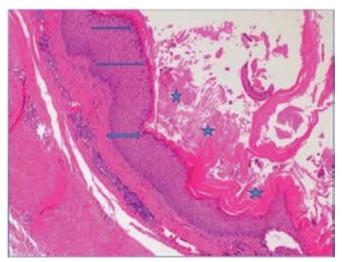


Fig. 4: (HE, ×100) Higher magnification showing that the cyst wall is composed of several layers of stratified squamous epithelium including the granular layer (arrows) and the cyst containing keratinous material (asterisks).

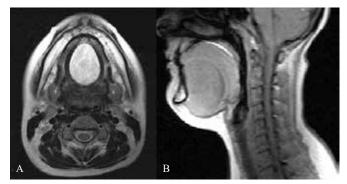


Fig. 5: T1-weighted MR images show heterogeneous hyperintense cystic mass at sublingual region. (A) Axial, (B) sagittal.

general anaesthesia using an extraoral approach. The mass lying under her mylohyoid muscle and extending towards the base of her tongue at a deeper location was excised. At her histological examination, the diagnosis was epidermoid cysts.

Case 3

A 28-year-old man was referred to Department of Otolaryngology, Ataturk University Faculty of Medicine, Erzurum, Turkey, with dyspnoea, shortness of breath and a painless, growing mass in the submental region. The patient had been monitored and treated for obstructive sleep apnoea syndrome (OSAS) for 2 years prior to his referral to our clinic due to his typical OSAS symptoms such as snoring and daytime sleeplessness. At his physical examination, a painless, mobile, soft mass was observed in the submental region. His computerized tomography (CT) indicated a homogenous cystic mass in the submental region, beginning from the corpus of the hyoid bone and passing through the base of the tongue, $5 \times 5 \times 6$ cm in size (Fig. 6). His surgical excision was performed under general anaesthesia using an extraoral approach. His histopathological examination revealed epidermoid cysts. A considerable alleviation of his OSAS findings was achieved after the surgical intervention.



Fig. 6: CT images show homogenous cystic mass at submental region. (A) Sagittal, (B) axial.

DISCUSSION

Numerous disorders may affect the submandibular region and the floor of the mouth. These include congenital lesions such as dermoid cysts, lymphoepithelial cysts and thyroglossal cysts, benign or malignant tumours of the salivary glands or mesenchymal tissues, and infections such as periapical abscess, perichondritis or sialadenitis (4).

Epidermoid cysts may be congenital or acquired. The pathogenesis of these cysts is unclear. Congenital cases arise from the ectodermal residual tissues, while the acquired lesions arise from trauma, inflammation or surgery (5). The epidermoid cysts are generally diagnosed at young ages, especially in the second or third decades (4). Our patients were aged 22, 23 and 28 years, respectively.

Physical examination alone is not enough to diagnose diseases of this region. On CT scans, dermoid cysts appear as thin-walled, unilocular masses with a central cavity. The central cavity is filled with a homogenous, hypoattenuating (0-18 HU) fluid material with multiple marbles. The 'sack of marbles' appearance resulted from the coalescence of fat into the small nodules and was virtually pathognomonic for dermoid cysts in this location (6). Epidermoid cysts exhibited fluid attenuation at CT and can also produce the sign known as 'sack of marbles' (7). At MRI, epidermoid cysts had the same signal intensity as fluid (7). Due to these various appearances at imaging, CT and MRI are not sufficient to distinguish epidermoid cysts from other cystic lesions. However, these methods are still important in assisting the surgeon in choosing the appropriate surgical approach.

The specific diagnosis depends on the histopathological examination after the surgical excision (8). Epidermoid cysts are known as 'pearly tumours' because of their shiny, smooth and waxy character at their macroscopic examinations. Histologically, epidermoid cysts are lined by a thin squamous epithelium which is rarely calcified. The cysts contain debris from desquamating lining of the epithelium, keratin and cholesterol (9).

The only effective treatment of these lesions is surgical enucleation. The surgical approach employed varies according to the size and location of the mass. Although intraoral approaches give better cosmetic and functional results, it is preferable to remove small and middle-sized tumours intraorally. Extraoral approaches were recommended for larger cysts (1, 8). Extraoral approaches were employed in our cases due to the size of the cysts.

The prognosis was good. Recurrence or relapse is extremely rare after surgery. No recurrence was observed in our cases after the 6-month follow-up.

Epidermoid cysts are rare lesions of the head and neck region, which are generally asymptomatic until the increased mass causes pressure symptoms. As in case 3, epidermoid cyst may rarely lead to OSAS. The presence of epidermoid cyst should be investigated in patients with OSAS complaints. Epidermoid cysts may exhibit various appearances on imaging studies. We describe three cases of giant epidermoid cysts on the floor of the mouth with different appearances at radiological imaging.

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