

Clinical Note

Characteristics of Clinical and Imaging Findings of Epidermoid Cysts under the Skin of the Mental Region

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Abstract: Epidermoid cyst often arises in the floor of oral cavity; however, it is relatively rare to arise subcutaneously in the mental region. We reported characteristics of clinical and imaging findings of epidermoid cysts under the skin of the mental region. The patient was a 53-year-old male. He complained of a subcutaneous mass without pain in the left mental region. A palpable soft and movable thumb-sized mass without pain was observed at the first examination. Ultrasound examination revealed a well-demarcated mass with heterogenous hyper-echoic internal echo and posterior echo enhancement. On power Doppler sonography, a small amount of blood supply was found around the periphery of the mass. Plain CT scan revealed a globular soft-tissue mass with peripheral high and central low CT values area. The mass was removed under local anesthesia using intraoral approach. A histopathologic diagnosis was epidermoid cyst. Recurrence is not observed 2 years after surgery. There were only 3 reports of epidermoid cysts in the mental region, including our case in Japan, clinically characteristics of age ranging 20 to 66 years of age and the mean age of 46 years, with 2 males and 1 female. The size was from 20 mm to 12 mm in diameter and from the tip of a thumb up to the tip of a small finger. Ultrasound images of the inside of epidermoid cysts that we experienced this time showed enhancement of relatively uniform hyperechoic image and posterior echo, which was characteristically slightly different from findings of epidermoid cysts generally reported.

Key words: Epidermoid cyst, Mental region, Ultrasonography, CT

Introduction

Dermoid and epidermoid cysts are congenital or acquired cystic malformations. The congenital form is considered to originate from entrapped ectodermal tissue during the embryonic period, while the acquired form is considered to arise from entrapped epithelial tissue secondary to events such as inflammation and injury. They can develop in various parts of the body, including the oral and maxillofacial region. The most frequently affected sites in the oral and maxillofacial region include the midline of the floor of the mouth. To the best of our knowledge, subcutaneous epidermoid cysts in the mental region are extremely rare. Dermoid and epidermoid cysts in the mouth floor are classified into the sublingual type, mental type, and sublingual-mental type based on positioning relative to the mylohyoid muscles, and preoperative CT, ultrasound, and MRI examinations are useful to examine the positioning in detail. However, it is important to diagnose by closely examining imaging and clinical findings together in detail because these examinations exhibited no typical imaging findings in some cases. This time, we report a case of epidermoid cysts under the skin of the left mental region for characteristics of clinical and imaging findings for a 53-year-old male

Materials and Methods

A 53-year-old man presented with a mass with a fluctuating size

in the left mental region since 3 years. The patient had not received any treatment because of the absence of significant symptoms. In March 2012, he visited a dentist because the mass was showing progressive enlargement, and he was subsequently referred to our university hospital. Physical examination revealed a movable, painless, and relatively hard thumb-sized mass in the left mental region. No abnormalities were observed in the mucosa. His medical history included an episode of facial nerve palsy 20 years back. Regarding ethical issues, an additional description is made that the Declaration of Helsinki is observed and informed consent was obtained from the patient in this case examined.

Results

An ultrasound study showed a well-demarcated mass lesion in the left mental region with distinct posterior echo enhancement. The periphery of the mass was hypoechoic, while the central portion of the mass was hyperechoic with a relatively homogeneous echotexture in which small hypoechoic lesions were present (Fig.1A). Power Doppler sonography revealed limited blood supply in the periphery of the mass, while there was no significant blood flow within the mass (Fig.1B). Noncontrast computed tomography (CT; axial and coronal images) revealed a subcutaneous, globular soft tissue lesion in the left mental region, with a peripheral high-density area (CT value, 45–60) and a central low-density area (CT value, 14–20) (Fig.1 C, D). On the basis of the clinical and imaging findings, differential diagnoses of an ectopic lymph node and a salivary gland tumor were established. In April 2012, the patient underwent resection of the mass through an intraoral

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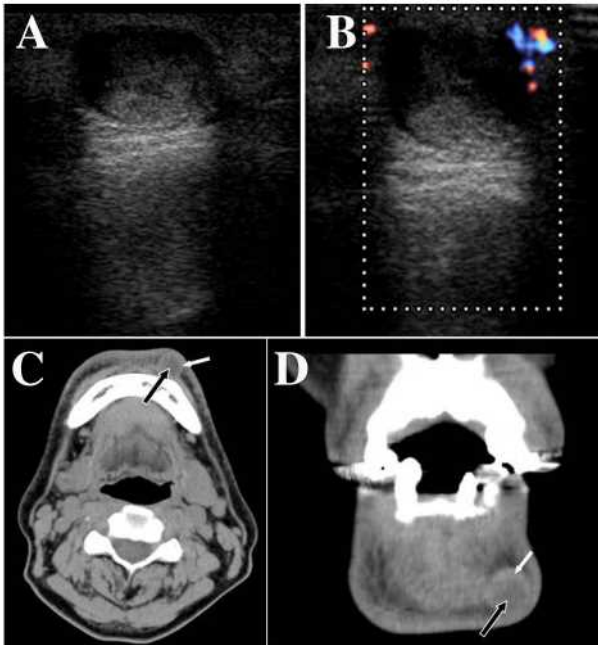


Figure 1. Ultrasound imaging.
 1A: A well-demarcated mass can be observed in the left mental region, with distinct posterior echo enhancement. The periphery of the mass is hypoechoic, while the central portion of the mass is hyperechoic with a relatively homogeneous echotexture in which small hypoechoic lesions are present. 1B: Power Doppler sonography shows limited blood supply in the periphery and no blood flow within the mass. 1C, D: Noncontrast computed tomography (CT; C: axial image, D: coronal image) A subcutaneous, globular soft tissue (black arrow) mass can be observed in the left mental region, with a peripheral high-intensity area (white arrow) and a central low-density area (black arrow).

incision under local anesthesia. The mass was bluntly dissected from the surrounding tissue and removed. It was partly adherent to the surrounding tissue and showed no communication with the oral cavity. Gross histopathological examination of the resected specimen revealed a cystic structure with a thin connective tissue wall that was filled with keratinized material (Fig.2 A). Histopathological examination revealed keratinized stratified squamous epithelium lining the inner wall of the cyst, while the subepithelial layer comprised loose connective tissues (Fig.2 B). The lining epithelium partly showed degeneration and deficit. Macrophages and foreign body giant cells were present in the cystic wall (Fig. 2 C). A histopathological diagnosis of an epidermoid cyst was established.

Discussion

Dermoid and epidermoid cysts develop from entrapped ectodermal tissue. Histopathologically, the cyst wall is covered by keratinized squamous epithelium. Meyer¹⁾ described three histopathological variants: epidermoid, true dermoid, and teratoid cysts. Epidermoid cysts do not contain any skin appendages, true dermoid cysts contain skin appendages, and teratoid cysts contain skin appendages (ectoderm), connective tissues (mesoderm), and respiratory and digestive organs (endoderm). Epidermoid cysts can develop in various parts of the body, such as the anus, rectum, ovaries, and oral and maxillofacial region²⁾.

The most frequently affected sites in the oral and maxillofacial region include the floor of the mouth, submental region, and submandibular region, while the rarely affected regions include the uvula and parotid region³⁾. The epidermoid cysts occurred under the skin of the mental region in this case, reports on such case are rare literally in Japan, and the number of cases reported in Japan was 3 cases^{4,5)} including our case (Table 1). Age ranged 20 to 66 years of age and the mean age was 46 years with 2 males and 1 female. The size was from 20 mm to 12 mm

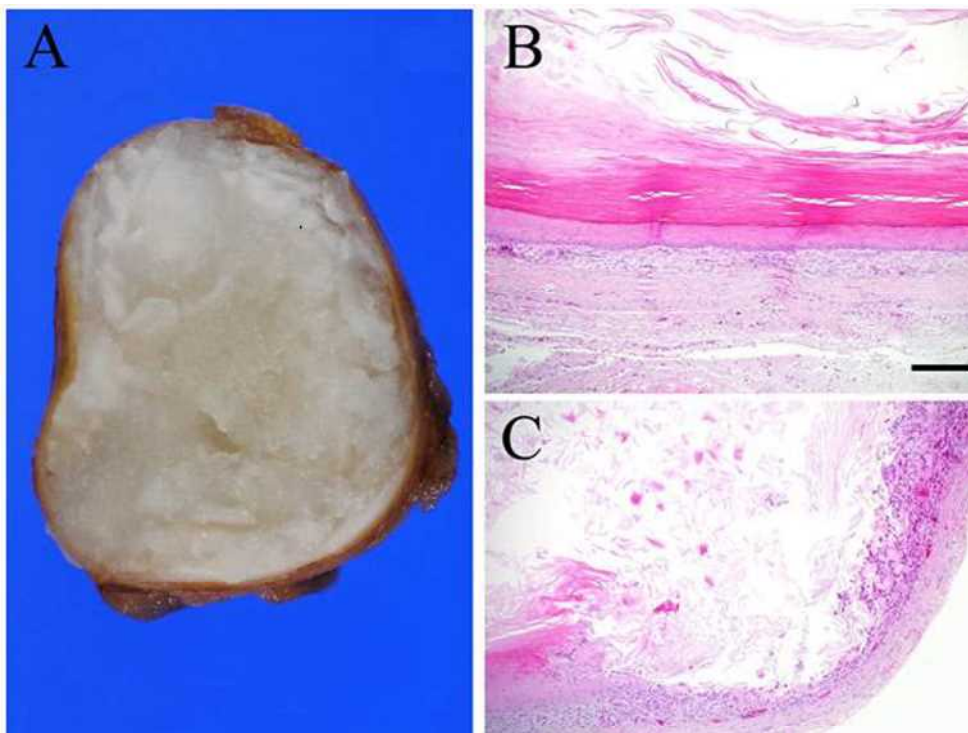


Figure 2. Pathologicalexaminationfindings.
 2A: A cross-section shows a cystic structure filled with keratinized material (scale bar: 5 mm). 2B: Histopathologically, the inner wall of the cyst is lined with keratinized stratified squamous epithelium, while the subepithelial layer comprises loose connective tissues (hematoxylin and eosin, scale bar: 200 µm). 2C: The lining epithelium partly shows degeneration and deficit. Macrophages and foreign body giant cells are present in the cystic wall (hematoxylin and eosin, scale bar: 200 µm).

Table 1. A reported case of epidermoid cysts in the mental region in Japan ⁵⁾

Case No	Authors	Age	Sex	Site	Magnitude	Histopathological diagnosis
1	Sasakura ⁴⁾ / 1996	20	female	mental region	10×12×18mm	epidermoid cyst
2	Kuraguchi <i>et al</i> ⁵⁾ / 2011	66	male	mental region	20×15mm	epidermoid cyst
3	Uchida <i>et al</i> / 2017	53	male	mental region	12×10×8mm	epidermoid cyst

Table 2. Imaging characteristics of cystic diseases in the head and neck region

Disease	Cyst pattern	Enhance effect (CT)	Inside of cyst	CT finding	MR findings
Dermoid cyst	Unicystic	Capsule is enhanced	Nonhomogeneous	Low density	Medium to high signal intensity on T1 Very high signal intensity on T2
Cystic lymphangioma	Multicystic	Septum is enhanced	Presence of septum	Low density	High signal intensity on T1 High signal intensity on T2
Adipoma	Unicystic	Capsule is less enhanced	Homogeneous	Low density	Very high signal intensity on T1 Very high signal intensity on T2

in diameter and from the tip of a thumb up to the tip of a small finger.

Several mechanisms have been proposed with regard to the congenital causes of epidermoid cysts in the head and neck region. Bergmann⁶⁾ explained that these cysts arise from entrapped epithelium during the midline fusion of the first and second branchial arches. Hasse⁷⁾ explained that they arise from the cervical vesicle, a vestige of the cervical sinus, that is embedded in the tissue. Colp⁸⁾ explained that the cysts arise from ectoderm entrapped within the mesoderm during the fusion of the first and second branchial arches. Acquired causes of epidermoid cysts have been proposed by Anderson⁹⁾ and Ackerman¹⁰⁾, who explained that these cysts arise from epithelium entrapped in tissue because of injury, inflammation, or surgery. We speculate that the epidermoid cyst in the present patient was congenital because he reported no history of injury or inflammation and was aware of the painless, palpable mass for a long period of time.

The majority of epidermoid cysts are congenital. Typically, the clinical symptoms appear after puberty, most frequently between 16 and 35 years of age^{11,12)}. Meyer¹⁾ explained that epithelial cells are more active during puberty, leading to the development of cysts. Our patient was as old as 53 years, and we assumed that there was a delay in treatment initiation because the cyst was asymptomatic; patients with a subcutaneous mass in the mental region may experience less impairment and symptoms compared with those having a mass in the floor of the mouth or buccal region.

Epidermoid cysts grow slowly and tend to be left untreated until the patient develops subjective symptoms. The most frequently reported symptoms include painless swelling, articulation difficulty, mastication difficulty, dysphagia, dysarthria, and lingual movement disorder. These symptoms increase with enlargement of the mass^{12,13)}. Although rare, dyspnea and sleep apnea have also been reported¹⁴⁾. Our patient exhibited no impairment and a painless mass¹⁴⁾. A previous study reported that the mean time interval between the first symptom and treatment initiation was as long as 5 years¹⁵⁾. Our patient sought treatment approximately 3 years after he noticed the mass in the mental region because he did not experience difficulties in daily life.

The differential diagnoses for epidermoid cysts include true dermoid cyst, cystic lymphangioma, and adipoma. The imaging characteristics of these lesions are shown in (Table 2). Preoperative imaging studies include ultrasound, CT, and magnetic resonance imaging (MRI). These instruments are useful for assessment of the size and anatomical location of the cysts and for establishing a differential diagnosis. For

epidermoid cysts it appear as well-demarcated, relatively homogenous, hypoechoic masses on ultrasound; on CT, these cysts appear as well-demarcated, low-density masses; MRI shows low signal intensity on T1-weighted imaging and high signal intensity on T2-weighted imaging; contrast CT also shows well-demarcated masses because of enhancement of the capsules¹⁶⁾. For dermoid cysts, it show heterogeneous echo when ectodermal components are concomitantly present; CT may exhibit a mixed pattern of high- and low-density areas, and/or may present with calcification; T1-weighted images show adipose tissue with high signal intensity. Ultrasound of the cyst in the present patient showed a relatively homogeneous internal echo and posterior echo enhancement. Although the imaging findings for this patient were slightly different from those reported in the literature, the CT values within the mass (14–20) were close to those of water. We therefore considered that the imaging findings of the mass were typical of epidermoid cysts. For such patients, enhancement of the capsule using contrast-enhanced CT and evaluation of the signal intensity within the mass using MRI are important for diagnosis. We believe there is a need for improved imaging studies for cystic diseases arising in soft tissues. CT and MRI images of cystic lymphangioma show multicystic masses. On CT, the lesion shows the same absorption as water^{17,18)}. On T1-weighted MRI, the lesion shows septae and the same or slightly lower signal intensity compared with that of muscles¹⁶⁾. The differentiation of epidermoid cysts from adipomas is relatively easy because CT images of adipomas show very low density, while MRI shows high signal intensity on both T1 and T2-weighted images.

Additionally, it is important to know the thickness of the cyst walls enough for extraction before surgery by ultrasonography. Particularly, it is important to perform a preoperative examination with carefully changing the direction of scanning by ultrasonography in a case of thin space between the skin and cyst walls.

Epidermoid cysts are basically treated by complete removal of the lesion, because residual cyst walls may lead to recurrence. Surgery can be performed using an intraoral approach, extraoral approach, or both. The most appropriate approach is determined on the basis of the size and location of the cyst. Given that epidermoid cysts have no opening to the skin and are less likely to adhere to the overlying skin, an intraoral approach is usually employed considering the cosmetic benefits, as observed in our patient, who showed no facial deformity after surgery. With regard to the prognosis, the recurrence rate of epidermoid and dermoid cysts was reported to be 2%¹²⁾. According to that report, the

cyst walls in the patients with recurrence had ruptured during initial surgery, leading to recurrence within a year after surgery. Our patient showed favorable clinical outcomes with no recurrence for 2 years.

In conclusion, we reported a rare case involving a 53-year-old man with a subcutaneous epidermoid cyst in the left mental region that was successfully excised via an intraoral approach. There were few reports of epidermoid cysts in the mental region in Japan, particularly, ultrasound images exhibited no typical imaging findings in our case, and it was important to comprehensively diagnose with clinical CT or MRI imaging findings. Additionally, it was considered that it was also important to examine imaging or clinical findings of some epidermoid cysts in detail in the future.

Conflict of Interest

No Conflict of Interest (COI) exists regarding this medical paper.

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