B-ENT, 2014, **10**, 165-169

Submandibular lateral epidermoid cyst: imaging findings of a rare case

B. Erol[1], Ü. Osma[2], Ö. T. Selçuk[2], D. Süren[3], H. Eyigör[2], C. Gürses[1] and M. Köroğlu[1]

[1]Department of Radiology, [2]Department of Otolaryngology-Head and Neck Surgery, [3]Department of Pathology, Antalya Education and Research Hospital, Antalya, Turkey

Key-words. Epidermoid; sonoelastography; MR imaging

Abstract. *Submandibular lateral epidermoid cyst: imaging findings of a rare case*. Epidermoid cysts (EC) represent less than 0.01% of all oral cavity cysts. Lateral epidermoid cysts in the neck are very rare. A male patient aged forty-five had a complaint of painless swelling in the neck. A well-circumscribed hypo-echoic mass with internal echoes was detectedin the right submandibular regionby ultrasonography. There were round areas inside the cyst with acoustic shadowing. The tissue hardness and the internal nature of the mass were evaluated with sono-elastography. Magnetic resonance imaging showed the mass's location and tissue properties in more detail. Magnetic resonance images revealed a well-circumscribed mass – hyperintense on T2-weighted images, hypo-intense on T1-weighted images – in the right submandibular region that had displaced the submandibular gland and mylohyoid muscle. There was no contrast enhancement in the mass on the contrast-enhanced fat-suppressed T1-weighted MR images. In this case report, we present the imaging features of a rare lateral EC in the submandibular region.

Introduction

Epidermoid cysts are developmental pathologies which represent less than 0.01% of all oral cavity cysts.[1] Patients may present with clinical symptoms such as speech difficulties, swelling, pain, dysphagia and dyspnoea. These pathologies can be seen at any age and in both sexes. However, they usually tend to be seen between the ages of 15 to 35.[2] We describe a rare epidermoid cyst in the submandibular region in a 45-year-old male patient, with histopathological features. We describe magnetic resonance (MR) imaging and sono-elastography in addition to the ultrasonography (US) findings.

Case report

A 45-year-old male patient with a painless swelling in the neck was admitted to the otolaryngology clinic. The patient's clinical story indicated that the mass had been present for nearly 30 years. The patient had no history of previous surgery, trauma, smoking, alcohol use, weight loss or night sweats. The patient did not have any other clinical complaints such as pain, dysphagia or dyspnoea. Physical examination revealed a mobile mass in the right submandibular region. Other otolaryngological examination was normal. Nasopharyngoscopy and indirect laryngoscopy were normal. The patient was evaluated primarily with US to determine the internal structure of the mass. A well-circumscribed hypo-echoic mass approximately 79×48 mm in size, with two round, well-defined, hypo-echoic areas approximately 2 cm in diameter and with an acoustic shadow were observed in the right submandibular region adjacent to the submandibular gland in US (Figure 1A). Colour Doppler US showed no significant vascularity within the mass (Figure 1B). The hardness of the tissue mass is compatible with low colour-coded red and green in sono-elastography. Round hypo-echoic areas coded in blue were consistent with the high degree of tissue stiffness (Figure 2).

MR imaging was performed before surgery to investigate the exact location of the mass, the anatomical relationship with the submandibular gland, the extension to surrounding structures, possible malignancy, lymphadenopathy and the presence of associated pathologies. MR images showed a mass filling the right submandibular region and displacing the right submandibular gland posterolaterally and the mylohyoid muscle medially (Figure 3A and Figure 3B). On T2-weighted MR images the mass was hyperintense and the round areas were hypo-intense; on T1-weighted MR images, the mass was hypo-intense, and round areas were hyperintense (Figure 3A and Figure 3C). There was no contrast enhancement in

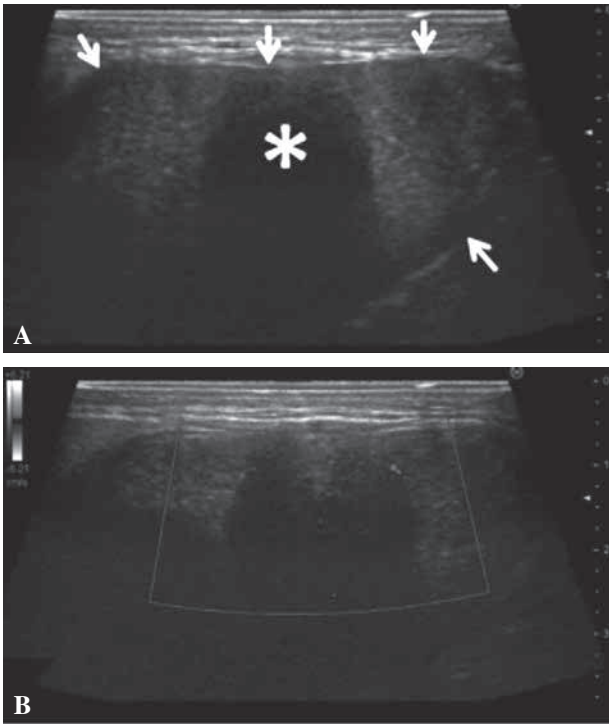


Figure 1
Ultrasonography revealed a hypo-echoic, well-circumscribed mass with internal echoes in the right submandibular region (arrows) and hypo-echoic, round, well-defined areas approximately 2 cm in diameter with an acoustic shadow (stars) (A). The colour doppler ultrasound image of the lesion was hypovascular (B).

the mass on the contrast-enhanced fat-suppressed T1-weighted MR images (Figure 3D). The hyper-intense component in T1-weighted MR sequences was seen as a hypo-intense lesion in fat-suppressed sequences and this suggests that it contains fat. In this region the patient's MR results were considered to indicate a dermoid or epidermoid cyst. Fine needle aspiration biopsy revealed non-neoplastic squamous cells and keratinous material. Cyto-pathological diagnosis was compatible with a benign cyst. Given the pre-operative findings, the total excision of the cyst, including the sub-mandibulary gland, was planned.

Under general anaesthesia, the mass adjacent to the right submandibular gland was totally excised with the gland. During the surgery, the cyst adhered strongly to the submandibulary gland.

Lamellar-type keratinous material in the lumen of the cyst, which was lined by a squamous epithelium, was observed pathologically (Figure 4A). A granular layer of squamous epithelium was evident (Figure 4B). There was no dysplasia in the squamous epithelium. No pilosebaceous unit attached to or adjacent to the cyst was observed. Pathological findings indicated an epidermoid cyst.

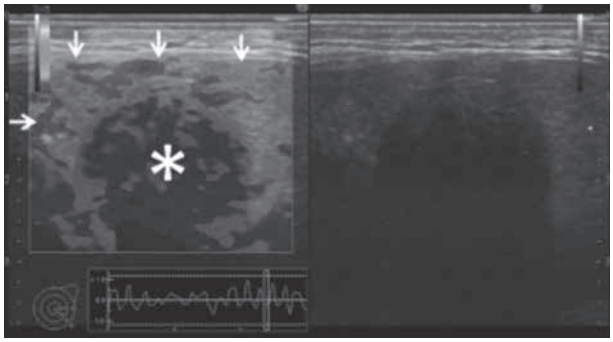


Figure 2
Sono-elastography identified low tissue stiffness indicated by the red and green colour-coded mass (arrows), with blue colour coding showing a high degree of tissue stiffness in the round area (asterisk).

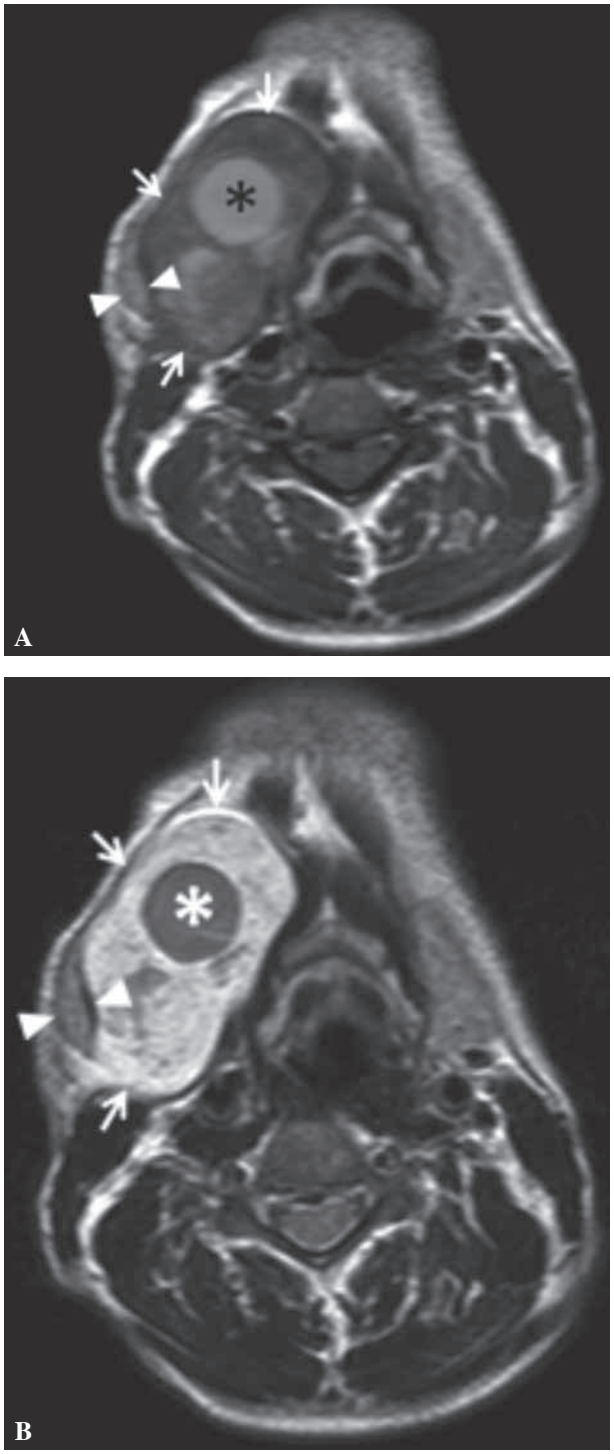
Discussion

ECs are keratinous cysts located in subcutaneous tissue that are frequently encountered in surgical pathology. ECs are benign lesions lined with a squamous epithelium that can grow anywhere. ECs are often seen in ovaries and testicles; only 7% of ECs occur in the head and neck, with 1.6% being found in the oral cavity.[3]

ECs may be congenital or acquired. In the third and fourth weeks of intra-uterine life, congenital cysts may arise as a result of the jamming of the ectodermal elements during the midline fusion of the first and second brachial arches, or from a tuberculumimpar between the two mandibular archs. Acquired cysts may develop after the iatrogenic or traumatic occlusion of sebaceous gland ducts or epithelial-cell inclusions.[4] Our patient had no history of surgery or trauma.

Although ECs are lined only by epithelial cells, dermoid cysts may contain skin elements such as hair follicles; sebaceous glands and teratoid cysts may contain other tissue elements such as muscle, cartilage, and bone.[2]

US is the initial imaging modality of choice to evaluate the internal structure of a mass. CT or MR imaging can be selected for the exact anatomical localisation of a massin relation to surrounding structures before drawing up the surgical inter-vention plan. On US, ECs have been seen to be hypo-echoic, or iso-echoic, homogeneous, uni-



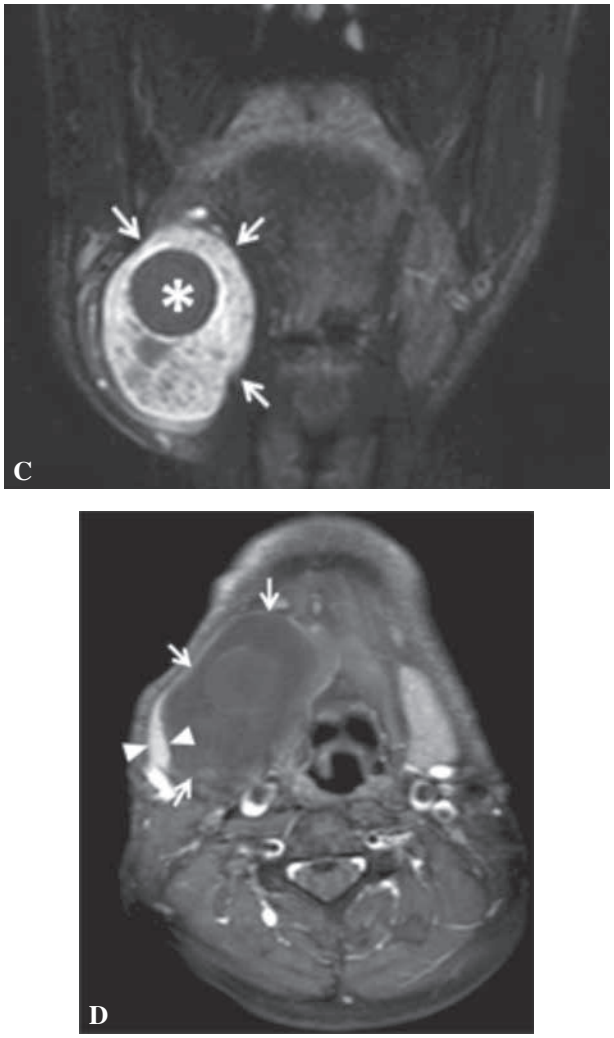


Figure 3
MR images showed a mass (arrow) filling the right sublingual and submandibular regions and displacing the right submandibular gland posterolaterally (arrowheads) and the mylohyoid muscle medially. Axial T1-weighted MR images showed a hypo-intense mass (arrows), and round hyperintense areas (asterisk) (A). In axial (B) and coronal (C) T2-weighted MR images, the mass was hyperintense (arrows) and the round areas were hypo-intense (asterisks). There was no contrast enhancement in the mass in the axial contrast-enhanced T1-weighted MR images (D).

locular masses with an internal echo.[5,6] Sonoelastography shows increased tissue stiffness in EC or dermoid cysts.[7] Similary; in our case, high tissue stiffness in the circular structures in the cyst detected. A low-density unilocular mass was detected on CT. The mass was hyperintense on the T2-weighted MR images, and signal intensity was low or intermediate in the T1-weighted MR images. Contrast material may be stained with the cyst wall.[6] MR imaging findings in our case were similar and no enhancement was observed with contrast material.

The floors of mouth cysts are divided into three groups depending on the location: sublingual or median genioglossal cysts on the geniohyoid muscle, median geniohyoid cysts in the submental region between the geniohyoid muscle and

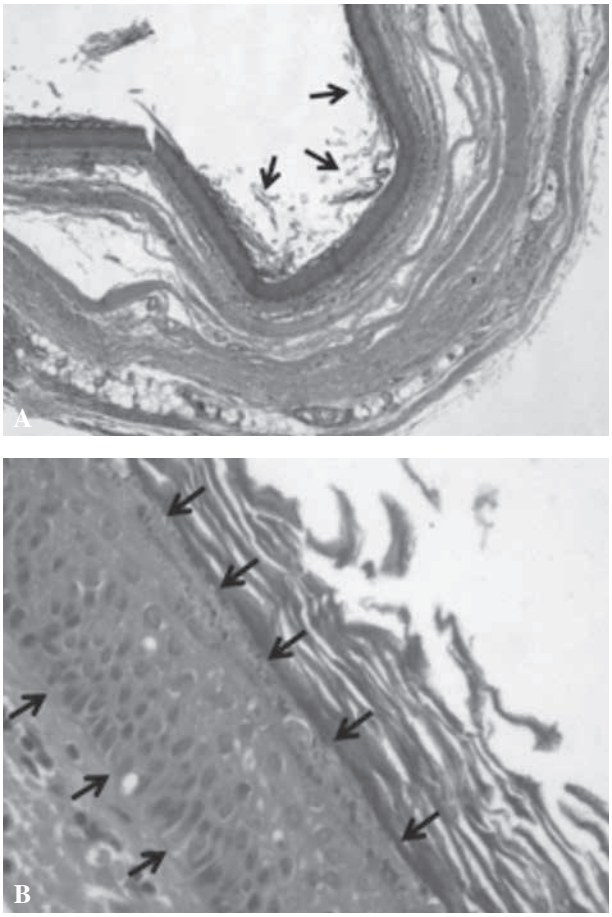


Figure 4
The structure of the cyst containing keratinous material (arrows) in the lumen (H & E ×40) (A). There was a significant granular layer of squamous epithelium lining the cysts. Squamous epithelium including the granular layer (arrows) lining the cyst (H & E ×400) (B).

mylohyoid muscles and, finally, lateral cysts in the submandibular area.[8] Our patient's cyst was a lateral epidermoid cyst in the submandibular region.

The major differential diagnosis in the case of epidermoid cysts is dermoid cyst. Dermoid and epidermoid cysts are similar in imaging findings. CT and MR images usually identify a thin-walled unilocular mass. Although epidermoid cysts are hypo-intense in T1-weighted MR images, signal intensity varies in dermoid cysts. Both are hyper-intense in T2-weighted MR sequences.[9] However, the liquid matrix, with the appearance a small sack of marbles made up of small nodules containing oil, is pathognomonic in the case of a dermoid cyst. It also has a heterogeneous inner structure. However, it may not be possible to distinguish between some features with imaging findings.[9]

Both ECs and dermoid cysts have a cyst structure lined with squamous epithelium. However, dermoid cysts contain pilosebaceous units that adhere to the epithelium.[10] Teratoid cysts contain other heterologous tissue components such as muscle, cartilage and bone in addition to the morphology observed in dermoid cysts.[2] In our case, the cysts were lined with stratified squamous epithelium without any heterologous elements or pilosebaceous units associated with the epithelium. Ranula, thyroglossal duct cyst, cystic hygroma, brachial cleft cysts, heterotopic gastro-intestinal cyst, foregut duplication cystsand epidermal inclusion cysts are also found in the differential diagnosis of epidermoid cyst.[2,6] Epidermal inclusion cysts produce similar pathological findings to epidermoid cysts but patients have a history of previous surgery or trauma. Our patient had no prior history of surgery or trauma.

Depending on the size and location of the EC, treatment may involve intra-oral or extra-oral surgical excision.[8] In our case, with the horizontal incision in the submandibular region, the EC adjacent to the submandibular gland was totally excised without any complication.

The most common complications associated with epidermoid cysts are the compression effect of the cyst and the rupture of the cyst because of an infection. Facial paralysis due to the infection of a cyst located in the parotid region has been described in the literature.[11] We observed no complications in our patient.

The most important issue to be considered was whether there was a malignant transformation of squamous epithelium. All epidermoid cysts should therefore be totally excised and subjected to careful histopathological examination to determine the presence or absence of nuclear atypia and stromal invasion potential.[12,13]

Conclusion

We presented the imaging features of a rare lateral EC in the submandibular region. Each of these imaging features helped to establish the correct diagnosis.

References

1. Ozan F, Polat HB, Ay S, Goze F. Epidermoid cyst of the buccal mucosa: a case report. *J Contemp Dent Pract*. 2007; 8(3):90-96.

2. Patil K, Mahima VG, Malleshi SN. Sublingual epidermoid cyst: a case report. *Cases J*. 2009;2:8848.
3. Turetschek K, Hospodka H, Steiner E. Case report: epidermoid cyst of the floor of the mouth: diagnostic imaging by sonography, computed tomography and magnetic resonance imaging. *Br J Radiol*. 1995;68(806): 205-207.
4. Kandogan T, Koç M, Vardar E, Selek E, Sezgin O. Sublingual epidermoid cyst: a case report. *J Med Case Rep*. 2007;1:87.
5. Dutt SN, Hock YL, Saleem Y, Bhat N, East DM. Epidermoid cyst of the submandibular gland. *Indian J Otolaryngol Head Neck Surg*. 2000;52(4): 378-379.
6. Ege G, Akman H, Senvar A, Kuzucu K. Case report: Sublingual dermoid cyst. *Tani Girisim Radyol*. 2003;9(1): 57-59.
7. Bhatia KS, Rasalkar DD, Lee YP, Wong KT, King AD, Yuen YH, Ahuja AT. Real-time qualitative ultrasound elastography of miscellaneous non-nodal neck masses: applications and limitations. *Ultrasound Med Biol*. 2010; 36(10):1644-1652.
8. Longo F, Maremonti P, Mangone GM, De Maria G, Califano L. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. *Plast Reconstr Surg*. 2003;112(6):1560-1565.
9. Koeller KK, Alamo L, Adair CF, Smirniotopoulos JG. Congenital cystic masses of the neck: radiologic-pathologic correlation. *Radiographics*. 1999;19(1):121-146.
10. Saylam G, Tanrikulu S, Dursun E, Iriz A, Eryilmaz A. A mass at fat density in the parotid gland: dermoid cyst or lipoma. *B-ENT*. 2009;5(1):43-45.
11. Streppel M, Thomas JP, Stennert E, Guntinas-Lichius O, Wagner M. Infected epidermoid cyst as cause of peripheral facial palsy. A case report [in German]. *Laryngorhino-otologie*. 2001;80(10):617-619.
12. Bhatt V, Evans M, Malins TJ. Squamous cell carcinoma arising in the lining of an epidermoid cyst within the sublingual gland – a case report. *Br J Oral Maxillofac Surg*. 2008;46(8):683-685.
13. Ozcan KM, Dere H, Ozcan I, Gun T, Unal T. An epidermal cyst in the parotid gland following ear surgery: a case report. *B-ENT*. 2006;2(4):193-195.

Ömer Tarık Selçuk, M.D.
Department of Otolaryngology-Head and Neck Surgery
Antalya Education and Research Hospital
Antalya, Turkey
Tel.: +90 242 2494400
GSM: +90 533-3254432
E-mail: omertarikselcuk@yahoo.com