Midline Submental Epidermoid Cyst: A Rare Case
A Pancholi, S Raniga, P Vohra, V Vaidya

Citation

Abstract
Epidermoid cysts of the neck are much less common than dermoid cysts in the head and neck, most often they are located in the submental region. Both are uncommon in the head & neck region. They present with midline, painless slow growing swelling. Imaging helps to determine the nature & contents of the lesion and the relationship of the lesion with the mylohyoid muscles which helps decide the surgical approach. Treatment is usually local excision. We will like to report a rare case of epidermoid cyst of the neck. The most striking point in the imaging was the presence of echogenic solid nodules in a cystic lesion on ultrasonography.

INTRODUCTION
The spectrum of teratoma includes true dermoid cyst, epidermoid cyst and teratoid cyst. Epidermoid and dermoid cysts are inclusion cysts lined by ectoderm. Epidermoids are devoid of skin appendages in their wall, while these are present in case of dermoids. Both are uncommon in the head & neck region and submental location of the dermoid represents 11.5% of head and neck dermoid [1]. Epidermoid cysts of the neck are much less common than dermoid cysts in the head and neck, most often they are located in the submental region [1]. They present with midline, painless slow growing swelling. Imaging helps to determine the nature & contents of the lesion and the relationship of the lesion with the mylohyoid muscles which helps decide the surgical approach. Treatment is usually local excision.

We will like to report a rare case of epidermoid cyst of the neck. The most striking point in the imaging was the presence of echogenic solid nodules in a cystic lesion on ultrasonography- only once reported before in the literature.

CASE REPORT
A 28 year old male presented with a midline painless swelling in the submental region of the neck for 3-4 months. Examination of the neck revealed a non-tender, soft, mobile, non-transilluminant swelling of 3 X 4 cm in the midline submental region. The swelling was becoming prominent on contraction of the mylohyoid muscle, and was not moving with swallowing or protrusion of the tongue. There was no evidence of cervical lymphadenopathy. Laboratory investigations including hemogram & ESR were normal.

Ultrasonography of the neck was done on AU-5 EPI, ESAOTE machine using 5-10 MHz linear probe. Ultrasonography through submental approach in coronal and sagittal planes revealed a 3 X 4 cm sized, well-circumscribed, encapsulated cystic lesion in the submental region (Between the anterior bellies of Diagastric muscles) [FIGRUE 1]; below the mylohyoid muscle [FIGURE 2], containing multiple echogenic coalescent nodules with central anechoic areas [FIGURE 1 AND 2]. The nodules were fixed in the lesion suggestive of thick cheesy fluid of the cyst. On ultrasonography, differential diagnoses of dermoid and epidermoid cyst were kept.
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**Figure 1**
Figure 1: Ultrasonographic oblique coronal image shows a well-encapsulated cystic lesion in the submental region between the anterior bellies of digastric muscles (marked with D).

**Figure 2**
Figure 2: An ultrasonographic oblique coronal image shows multiple echogenic nodules fixed in the cystic lesion. Myelohyoid muscle is displaced superiorly suggestive of submental location of the lesion.

The cyst was completely removed by surgical excision from cervical approach and sent for histopathological examination which revealed thin wall without any evidence of dermal appendages with lumen containing keratin material suggestive of epidermoid cyst. The striking sonographic appearance of multiple echogenic nodules in the lesion was caused by multiple spherical keratin formations.

**DISCUSSION**

The term “dermoid cyst” has been applied to several different pathologic entities, including benign cystic teratomas and various sequestration-type cysts. True dermoid cysts, epidermoid cysts, and teratoid cysts compose the spectrum of teratomas; all are covered by squamous epithelium [1]. Dermoids and epidermoids are ectoderm-lined inclusion cysts that differ in complexity. Epidermoid cysts have only squamous epithelium; dermoids contain hair, sebaceous and sweat glands, and squamous epithelium [1].

The presence of skin appendages within the wall of the dermoid cyst and the absence of these features in the epidermoid cyst is the basic difference between these two. A teratoid cyst may also contain tissue of other major organ...
systems (e.g., nervous, gastrointestinal, respiratory). Because of the squamous epithelium lining, all three of these cysts may have cheesy keratinaceous material within the lumen [1, 3, 4].

Most dermoid cysts in the oral cavity floor are derived from epithelial debris or rests, enclaved during midline closure of the bilateral first and second branchial arches. They may also result from ectodermal differentiation of multipotent cells, most likely pinched off at the time of anterior neuropore closure [5]. The causes of both epidermoids and dermoids include failure of surface ectoderm to separate from underlying structures, sequestration of surface ectoderm, and implantation of surface ectoderm. Most congenital dermoid and epidermoid cysts probably arise due to an embryologic accident during the early stages of development, between 3 and 5 weeks of gestation [5].

Epidermoid inclusion cysts have a thin squamous lining because of lack of dermal appendages, which only rarely contains calcifications. These cysts contain debris from the desquamation of their squamous epithelial lining. The debris consists of mostly keratin, a proteinaceous material, and some cholesterol. Epidermoids are often described as pearly tumors because of the shiny, smooth, waxy character of their “dry keratin” at gross inspection [1, 5].

Dermoid cysts are circumscribed, encapsulated lesions lined with ectodermally derived squamous epithelium that contains a variable number of skin appendages (sebaceous glands, hair follicles, and sweat glands). They have a thicker lining, which may contain dystrophic calcifications. The lumen of the cyst is filled with a mixture of keratin, sebaceous material, and occasionally hair, and is often described as buttery or cheesy tumor. Curiously, dermoid cysts of the floor of the mouth are remarkably free of hair compared with dermoid cysts elsewhere [1, 5].

Dermoid and epidermoid cysts are developmental pathologies that occur in the head and neck with an incidence of 6.9-7% [1, 5, 6], with the lateral eyebrow being the most common location [1]. About 11.5% of dermoid cysts of the head and neck are in the floor of the mouth, the second most common location and most common cervical location [1], and they represent less than 0.01% of all oral cavity cysts [1]. Congenital dermoid cysts of the floor of the mouth are relatively rare but when they occur, they do so inevitably in the midline [1]. Epidermoid cysts of the neck are much less common than dermoid cysts in the head and neck, most often they are located in the submental region, although they sometimes can impose certain diagnostic problems [1, 6].

The most common clinical appearance of a dermoid cyst in the neck is a midline, suprahyoid, slowly growing mass, enlarging over years or decades, by the accumulation (within an enclosed space) of cutaneous products [1, 2, 3]. Typically, the mass is soft, mobile, and unattached to overlying skin. They have no intimate association with the hyoid bone and therefore do not move with tongue protrusion [1, 8, 9]. The size of the cyst ranges from a few millimeters to 12 cm.

Ultrasonographic features include solid and cystic structures within a heterogeneous mass, and calcifications are seen more frequently than cartilage and bone formation [10]. On CT scans, they are moderately thin walled, uninlocular masses filled with a homogeneous, hypodense (0–18 HU) fluid material with multiple hypodense fat nodules giving the “sack-of-marbles” appearance, which is virtually pathognomonic for a dermoid cyst in this location [1, 4]. Alternatively, the cyst may be heterogeneous on CT scans because of the various germinal components [4]. Fluid-fluid levels with supernatant lipid are possible. The rim of these cysts often enhances following administration of contrast material [4].

MR imaging in the coronal plane optimally depicts the relationship of these cysts to the mylohyoid muscle in the floor of the mouth and helps determine the surgical approach [11]. Dermoid cysts have variable signal intensity on T1-weighted images. They may be hyperintense (because of the presence of sebaceous lipid) or isointense relative to muscle on T1-weighted images. They are usually hypointense on T2-weighted images. The mass has a clearly demarcated rim but frequently has a heterogeneous internal appearance [11, 12].

Epidermoid cysts have fluid attenuation on CT scans and are hypointense on T1-weighted images and hyperintense on T2-weighted images, following the signal intensity of fluid (Fig 25). An epidermoid cyst located entirely within the sublingual space may be difficult to distinguish from other cystic lesions in the floor of mouth (eg, a simple ranula) on the basis of imaging criteria alone [11].

Treatment consists in complete surgical removal, trying not to rupture the cyst, as luminal contents may act as irritants to fibrovascular tissues, producing postoperative inflammation with excellent prognosis in cases free of complications [11]. Recurrence is unlikely after complete surgical excision.
Clinical signs and surgical approach are determined by the relationship of the cyst to the musculature of the floor of the mouth. Most dermoid cysts are located superior to the mylohyoid muscle in the sublingual space, only a small external swelling will be seen and will be removed with an intraoral approach. Less commonly, the lesion is inferior to mylohyoid muscles and there is usually an obvious submental swelling and the cyst is externally palpable will be removed with an external submandibular approach. Differentiation of midline submental neck mass into epidermoid and dermoid is more of academic interest than of any practical importance. Treatment of both the lesions is surgical excision. Both of the lesions are well-encapsulated and benign. The only important point that determines the treatment is the location of the lesion in relationship to the myelohyoid muscle.

Malignant transformation of oral cysts has not been reported. About 5% of dermoid cysts undergo malignant degeneration into squamous cell carcinoma.

CONCLUSION

This is a report on a rare case of epidermoid cyst in the submental region of neck. They often manifest as slow-growing masses and cause symptoms only after enlarging sufficiently or after infection. Ultrasonography is frequently used initially to confirm the cystic nature of the lesion. CT and MR imaging provide essential information on the cyst location that allows optimal preoperative planning. Ultrasonographic evaluation sometimes can give all the necessary information before surgical intervention, as in our case. Differentiation of epidermoid and dermoid preoperatively is more of academic interest. The only important issue is the relationship of the lesion to the myelohyoid muscle as it determines the surgical approach.

CORRESPONDENCE TO

Dr. Sameer Raniga M 81, Shantinagar, Tarsali Road Vadodara-390009. Contact Number: +91-9825525484 (MOBILE) +91-265-2647053. E mail: samhet10200@yahoo.com

References

Author Information

Alpesh Pancholi, M.B.B.S.
Second year resident, Department of Radiology, S.S.G. Hospital and Medical College

Sameer Raniga, M.D., D.N.B.
Assistant Professor, Department of Radiology, S.S.G. Hospital and Medical College

Prakash A. Vohra, M.D., D.M.R.D.
Professor and Head, Department of Radiology, S.S.G. Hospital and Medical College

Vijay Vaidya, M.D.
Assistant Professor, Department of Radiology, S.S.G. Hospital and Medical College