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Large Submental Epidermoid Cyst: A Rare Case Report

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Abstract:

Epidermoid cyst, specially in large size is one of the rare lesions of maxillofacial region, affects submental and sublingual region. The cyst develops from the entrapment of pluripotent ectodermal and mesodermal primordia, in the embryonic lines of fusion, when first and second branchial arches of each fuse in the midline, between third and fourth weeks of intrauterine life. After entrapment, this tissue shows gradual proliferation and ultimately transform to a cyst. A 19-year-old-boy presented with such a midline painless large swelling in the submental and sublingual region with diagnosis of epi dermoid cyst. After enucleation, the patient didn't show any recurrence after 2 years follow up.

Keywords: Epidermoid cyst, submental region, sublingual region.

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Introduction:

Epidermoid cysts represent cystic malformations, has no sex predilection and commonly affect the people, between the ages of 15 & 35 years[1,2]. This lesion is relatively uncommon in head and neck area, incidence ranging from 1.6 to 7%[1,2] and represent less than 0.01% of all oral cavity cyst[2]. Other swelling in this region may be due to ranula, lymphangioma, hemangioma, lipoma etc. Generally, lesion is symptomless, slowly enlarge can cause obstruction with consequent dysphagia, dysphonia, at last dyspnea[2]. The size of dermoid cyst is very variable[2]. This congenital lesion develops in the midline due to retention of germinal epithelium, during growth of branchial arches and lower jaw[3]. Histopathologically, these lesions are classified as epidermoid cyst (lined only by stratified squamous epithelium with a fibrous wall and no skin adnexa), dermoid cyst (lined by stratified epithelium with skin adnexa) and teratoid (cystic or solid featured with other tissues such as muscle, cartilage, or bone)[4,5]. Recurrence of dermoid cysts is not expected and malignant transformation is very rare but has been reported in 5% cases[6]. Literatures indicate that these lesions can be developed from acquired factors such as trauma or surgical implantation that forces epithelial cells into deep tissue[7].

Case Report:

A 19-year-old Bangladeshi male patient presented with a painless swelling in the midline of neck just below chin & tongue. Patient noticed the swelling for 7-8 months, which was increasing very slowly. He noticed water like discharge through oral cavity after compression & repeated reduction in size of swelling periodically. He didn't give any history of dyspnea, dysphagia, dysphonia. His tongue movement was normal and presented no other swelling in other parts of body. Patient wass free from any comorbidities. He just presented the swelling for aesthetic purpose.

Clinical examination revealed a large oval swelling in FOM and submental region (Figure 1). Overlying skin & mucosa had no changes in color and texture. The swelling was non tender, about 7 cm X 7 cm in size, doughy consistency, smooth surface, movable bimanually, free from overlying & underlying structures. Swelling didn't move during protrusion of tongue & deglutition. On instruction, patient could open his mouth properly and could move tongue in all directions. His dentitions were in normal alignment without any mobility

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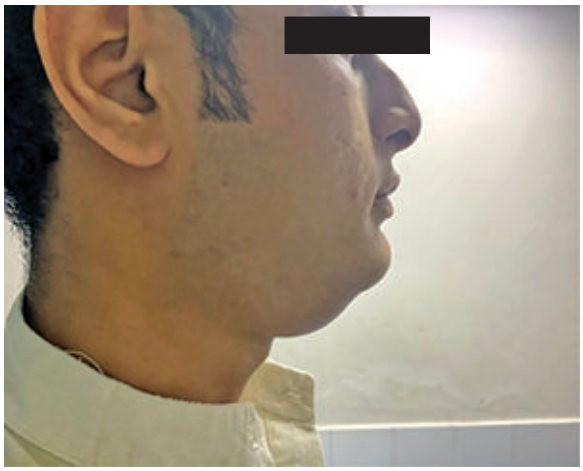

Fig.-1: *Preoperative presentation of submental swelling*

and displacement of teeth. There was no pulsation over the swelling and there was no cervical lymphadenopathy.

The clinical presentation was that of a submental and sublingual dermoid cyst, while the differential diagnosis included a plunging ranula, epidermoid cyst, thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, lymph node enlargement, teratoid cyst, sublingual salivary gland infection, lipoma, lymphoma.

Diagnostic modalities included OPG, CT scan of maxillofacial region, USG of thyroid, hematological test and FNAC. All hematological values were in normal ranges. OPG showed no dento-alveolar pathologies and bony lesions. USG revealed a big hypoechoic mass (40.5X38.1 mm) in size with solid part (27.4 X 2.7.0 mm) in submental region with normal both lobes of thyroid (Figure 2). CT scan revealed a lesion (6.9 ✕ 5.6 5.1cm) with globules of fat (-6 to -8HU) in a high-density fluid medium (21-22 HU) in the sublingual space and descent to submental space splaying the genioglossus muscles laterally (Fig 3). No significant enhancement was seen in post contrast scans. FNAC showed a number of foamy macrophages & degenerated cells. The background showed amorphous eosinophilic material.

With a diagnosis of Dermoid Cyst, patient was treated by enucleation of the cyst under general anesthesia through extraoral transcervical approach over skin crease line (Fig 4,5,6).

Dissected capsulated cystic lesion contained creamy white fluid with white multiple friable jelly balls with some fine hairy structures (Fig 7)

Histopathological examination of the excised specimen showed cyst wall lined by squamous epithelium supported by fibrous tissue. The epithelium was ulcerated showing foreign body giant cell reaction. Lumen contained keratin flakes (Fig 8). Hence, it was conclusively diagnosed as an epi dermoid cyst. After enucleation, the patient didn't show any recurrence after 2 years follow up.

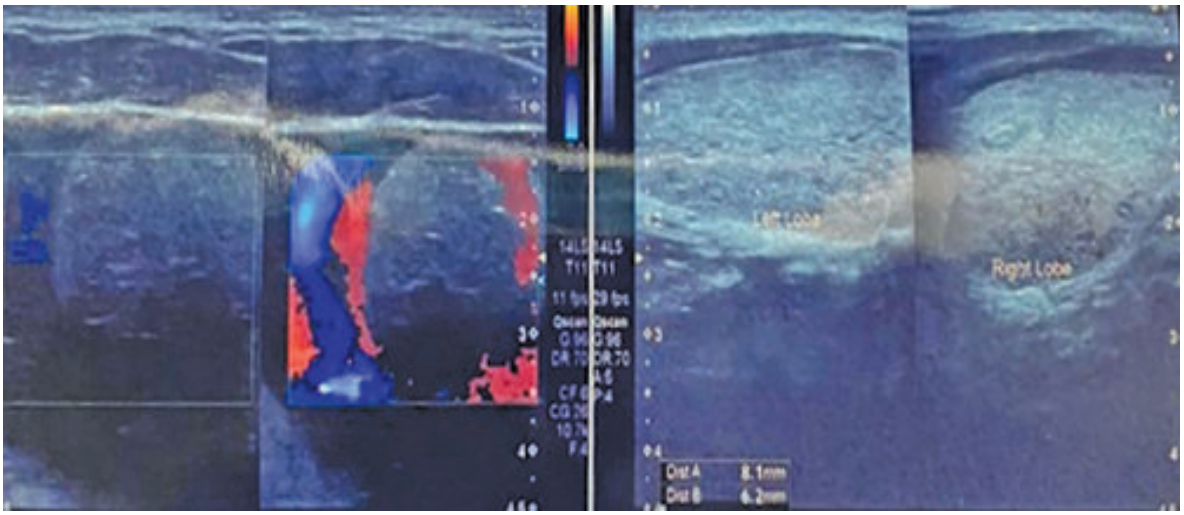

Fig.-2: *USG of thyroid reveals normal both lobes of thyroid with presence of submental mass*

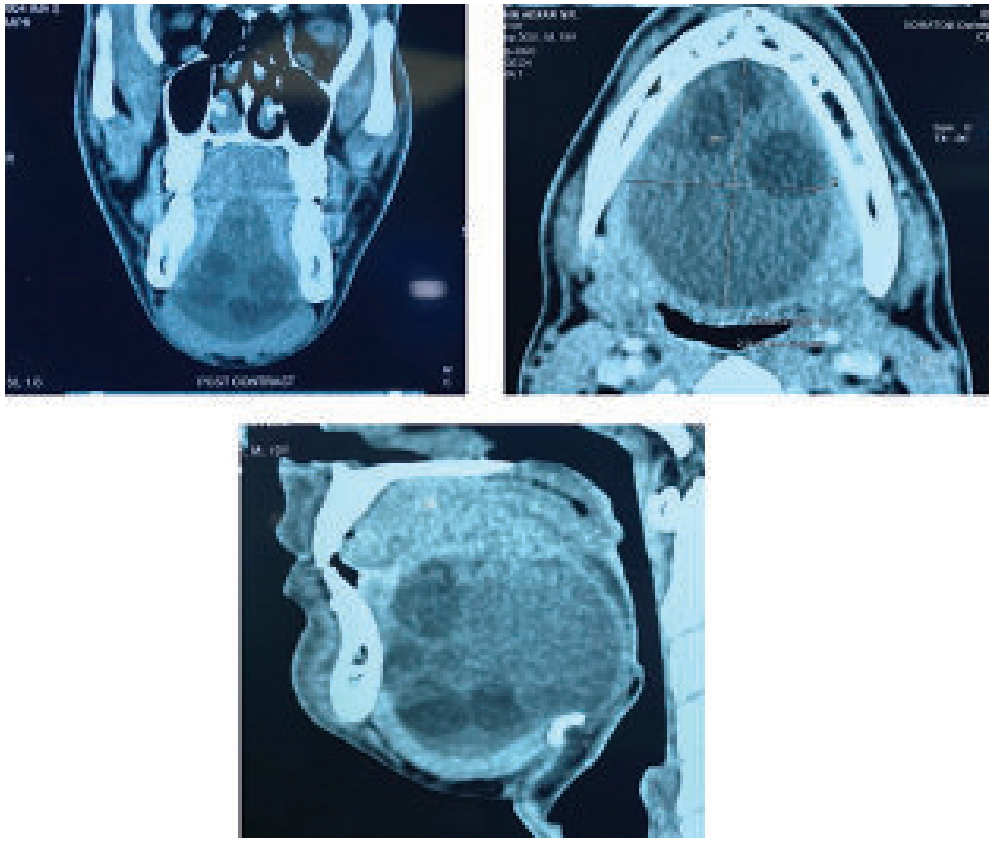

Fig.-3: *CT scan (coronal, axial and sagittal views) show a lesion (6.9 5.6 5.1cm) with globules of fat (-6 to -8HU) in a high-density fluid medium (21-22 HU) in the sublingual space and descent to submental space splaying the genioglossus muscles laterally with no significant enhancement in post contrast scans*

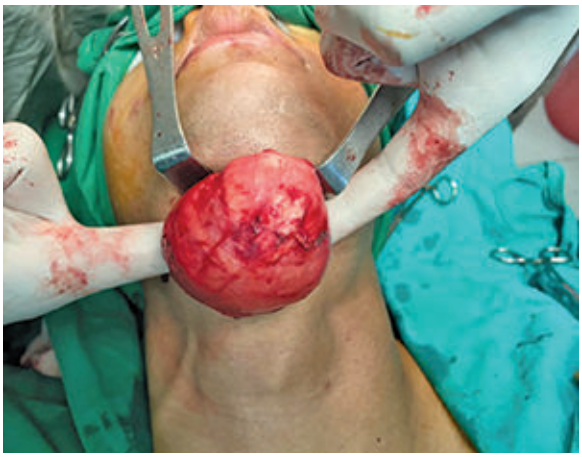

Fig.- 4: *Lesion during removal of mass (8 65.5 cm)*

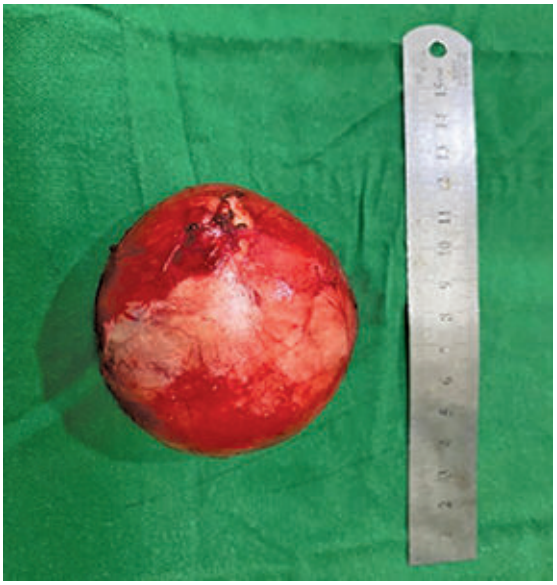

Fig.-5: *Resected specimen after enucleation approximately (8 65.5 cm)*

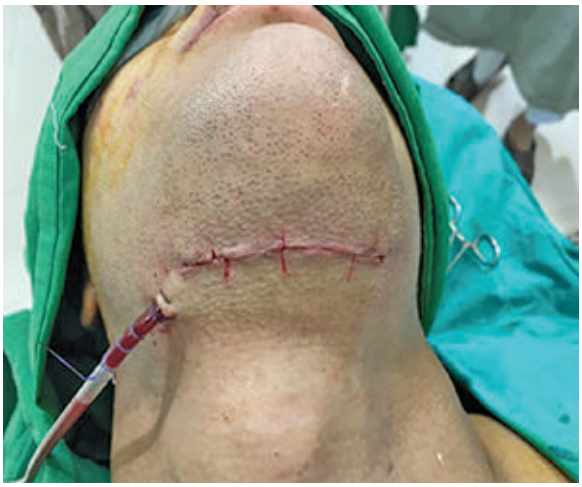

Fig.-6: *postoperative surgical site*

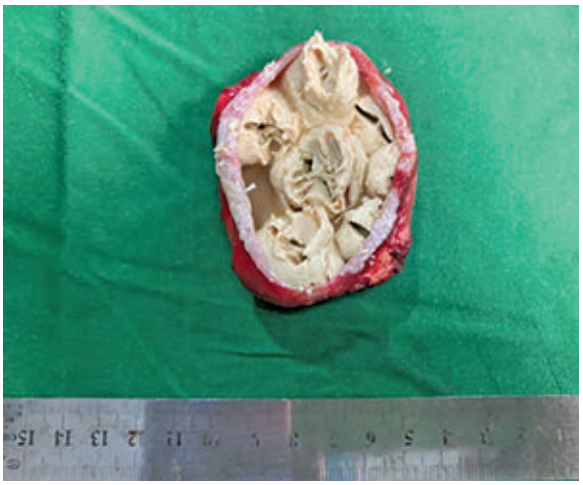

Fig.-7: *Dissected capsulated cystic lesion containing creamy white fluid with white multiple friable jelly balls with some fine hairy structures*

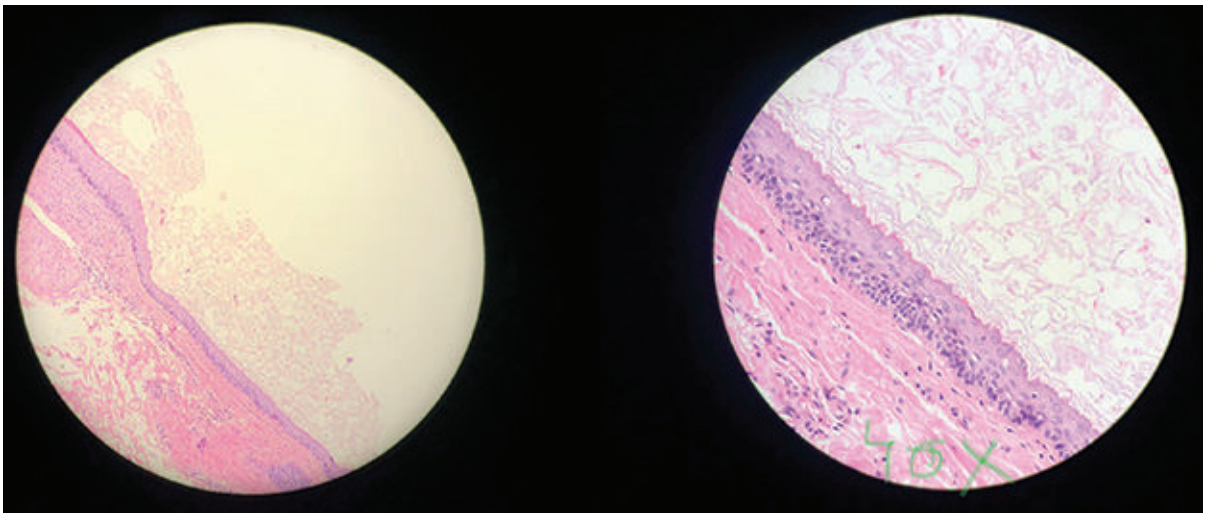

Fig.-8: *shows cyst wall lined by squamous epithelium supported by fibrous tissue. The epithelium is ulcerated showing foreign body giant cell reaction. Lumen contains keratin flakes*

Discussion:

The word "dermoid cyst" is a generic term which has been used to encompass & describe three different histological verities. Meyer classified the congenital FOM cysts into three different categories e.g., Epidermoid cyst lined by stratified squamous epithelium with no skin appendages; Dermoid cysts lined with keratinizing stratified squamous epithelium with skin adnexa; Teratoid cysts, cystic cavity in addition to skin appendages also encloses mesodermal derivatives such as bone, muscle, GI and respiratory tissues[1,3].

These epidermoid cysts are developmental benign conditions that have potential to be found anywhere in the body, however majority of the cases reported in the midline of the body commonly reported in ovaries & testes, the head & neck area much less involved[3]. The origin of epi dermoid cyst is thought to be congenital in most cases, but they also can be acquired[4,5]. Recurrence of epi dermoid cyst is not expected & malignant transformation through rare, has been reported in 5% cases[6].

On CT scans, it appears as moderately thin walled, unilocular mass filled with a homogenous, hypoattenuating fluid substance with numerous hypoattenuating fat nodules giving the pathognomonic "Sack of marbles" appearance[8]. The reported case also showed globules of fat in a high-density fluid medium which is almost similar to other reports[9,10]. The patient is under follow up of more than two years and has not shown any recurrence. But long term follow up is planned to exclude any recurrence and malignant transformation, which can be a possibility, though rare as known from literature[6,11].

Conclusion:

Epidermoid cysts are comparatively less common congenital lesions. Clinically it shows less sufferable symptoms. Epidermoid or Dermoid cyst can be considered as differential diagnosis of any slow growing sublingual and submental swelling. Preoperative CT scan and FNAC are helpful indicators for surgical planning. Long term follow up can exclude recurrence and malignant transformation.

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