Lateral Dermoid Cyst of Sub-Mandibular Region: A Case Report

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Abstract

Dermoid cysts located in the lateral cervical region are relatively rare. We describe a case of a lateral sublingual dermoidcyst in a 22 year-old girl. This girl presented with double chin for cosmetic reason. The lesion was a painless and slowly enlarging mass of the right submandibular region. Intraorally, there was no obvious swelling. There is always a difficulty of making a correct diagnosis of these lesions with clinical examinations and conventional radiography. To achieve a diagnosis and to develop correct surgical strategy specialized imaging such as Ultrasonography, Computed Tomography, Magnetic Resonance Imaging and histopathological examination should be carried out. In the present case, ultrasonography demonstrated a subcutaneous mass. FNAC showed lipomatous growth. It was found to be a dermoid cyst, and was documented to be lateral in position. Histopathology revealed a cyst containing a keratinizing stratified squamous epithelial lining. Based on the radiographic and clinicopathological findings, the patient was finally diagnosed as having a lateral dermoid cyst.

Key Words: Lateral sublingual dermoid cyst, Neoplasm, Cystic hygroma, Ranula.

Introduction

The dermoid cyst is an uncommon clinicopathological lesion of developmental origin which describes three histologically closely related lesions: dermoid cyst, epidermoid cyst and teratoma. Epidermoid and dermoid cysts are benign nature, which may occur anywhere in the body, but most predominantly in the ovary and scrotal regions[1]. Only about 7% are found in the head and neck region. The floor of the mouth is the second most common site in the head and neck region after the lateral eyebrow as these are the sites of embryonic fusion. The occurrence in oral cavity is approximately 1.6%. The floor of the mouth is one of the most commonly affected areas, however, these cysts can also be found in the tongue, lips, buccal mucosa and jaw bones[2,3]. Dermoid cyst is even rare in lateral sublingual situation. Very few cases have been reported. Dermoid cysts are derived from epithelial rests that are included during midline union of the first and second branchial arches.

The vast majority of dermoid cysts of the floor of mouth (DCFOM) are located in the midline (sublingual 52%, submental 26%), 16% involve more than one of the three possible spaces in the floor of the mouth region (submental, sublingual, submandibular), and only 6% are situated exclusively in the submandibular space where they appear to be lateral neck cysts[4]. However, the origin of lateral dermoid cyst remains somewhat of a riddle. Other common benign cystic lateral cervical masses are typically soft, slow-growing and painless, making clinical distinction difficult. An accurate diagnosis is of course necessary for the choice of treatment plan. Advanced imaging techniques such as magnetic resonance imaging are useful to achieving this end, leading to successful and uneventful management of such lesions. The final diagnosis of a dermoid depends on histologic investigation of its cyst wall.

This case report describes a relatively rare case of a true lateral sublingual dermoid cyst arising in the left submandibular region, and discusses the etiology, differential diagnosis and clinical management of a lateral dermoid cyst.

Case Report

A 22 year old female patient reported to our department with double chin for cosmetic reason in the midface to slightly left side since one and half years. Initially the swelling was small with gradual increase to recent size. Patient had no history of pain in the mass. On examination, obvious facial asymmetry was seen with extraoral painless swelling on left midline to submandibular region (Fig. 1A) with no associated swelling intraorally (Fig. 1B). The edges were indistinct, surface was smooth, soft in consistency, non tender, compressible, fluctuant in nature and overlying skin was normal in colour and there was no rise in temperature. The swelling was freely mobile and not attached to the underlying tissues. No sinus or fistula was evident. There was no cervical lymphadenopathy. Her past medical and dental history was not relevant.

Based on the clinical findings, the differential diagnosis of the lateral cervical mass included cystic masses like cystic hygroma, thyroglossal duct cyst, plunging ranula, branchial cleft cyst; benign or malignant salivary gland tumours, infectious or inflammatory processes, lipoma, lymphoma. Dermoid cyst was not considered in the differential diagnosis of lateral cystic mass. All routine laboratory investigations were normal. Secondly, FNAC was performed which revealed white granular cheesy material and the presence of epithelial remnants, desquamated tissue and cellular debris which pointed to be a diagnostic hypothesis of lipomatous mass.

Enucleation of cyst under local anesthesia was done by submandibular approach (Fig 4 and 5). The specimen received was yellowish in color; soft tissue mass with 4cm × 4m × 3 cm in dimension. On palpation it had dough like consistency. It had a thin walled capsule surrounding, with a cheesy white material inside. The excised specimen was sent for histopathological examination.

Microscopic examination revealed a cyst inside a thick, fibrous capsule lined by stratified squamous epithelium with marked orthokeratosis (Fig 6). A few sebaceous glands were seen lying in the subepithelial stroma, which established the diagnosis of dermoid cyst. No malignancy was seen.

On follow-up, the incision was healed well with good cosmetic result. Post-operative recovery was uneventful throughout the follow up period of one year and no recurrence was observed.

Discussion

Dermoid cysts are unusual, but well recognized lesions in the head and neck. In the region of the oral cavity, they are present as solitary, painless, slow growing mass in the floor of the mouth but they can be situated in submental and submandibular space[5,6].There are three theories of etiology of dermoid cyst

i. Congenital inclusion of dermal and epidermal elements of germ layers in deeper tissues along the embryonic lines of fusion,
ii. Acquired traumatic implantation of dermal and epidermal elements of surface epithelium which may proliferate and keratinize
iii. Growth from rest of totipotent cells displaced from the blastomere[7].

Histopathologically, the terms "dermoid" or "dermoid cysts' have been used as umbrella titles to describe the three subtypes of these congenital cysts containing keratinous squamous material. Epidermoid cysts or epidermal inclusion cysts are lesions lined with a simple squamous epithelium with no adnexal structures. True dermoids are stratified squamous epithelial-lined cysts that contain skin adnexal structures, including hair, hair follicles, sebaceous and sweat glands. Finally, teratoid cysts are masses lined with a variety of

epithelia, including stratified squamous and ciliated respiratory epithelia, and contain elements of ectodermal, endodermal and/or mesodermal origin[8]. Leveque et al[9] regard lateral Dermoid Cysts as merely median Dermoid Cysts that have migrated during their expansion because such cysts frequently have fibrous attachments to deep structures or those surrounding the midline of the mandible such as genioglossus, geniohyoid, mylohyoid, digastric, and platysma muscles; these attachments mark the path taken in cyst migration. In fact, a lot of reported cases have been considered to have migrated from median DCs, suggesting that a true lateral DC does not exist in reality[8,9,10,11,12]. As acquired DCs arise from epithelium implanted during trauma, it is more plausible that they can occur at sites away from the midline. However, in our case, the whole of the lesion lay in the submandibular region without traces of migration from the medial region. Likewise, some investigators have reported true lateral DCs existing in the submandibular region[13,14,15] Generally, DCs are classified into 3 types by their anatomical position[8,10] : (1) median; developing between the genioglossus muscle, (2) lateral sublingual; developing between the genial muscles and mylohyoid, (3) true lateral; developing between the genioglossus and hyoglossus medially and the mylohyoid laterally. According to this classification, the cyst in our case was type (3) spreading laterally inferior to the mylo-hyoid muscle to its present position in the submandibular region. Although the origin of true lateral DCs is somewhat of a riddle, it has been thought that they probably arise from the ventral end of first pharyngeal pouch or from the extreme ventral end of the first branchial cleft[11,16]. The differential diagnosis of a lateral submandibular mass should include cystic masses like cystic hygroma, enteric duplication cyst, thyroglossal duct cyst, branchial cleft cyst, neoplasms, odontogenic or mucus extravasation masses, infectious or inflammatory processes as well as salivary gland pathology[13,14].

Fine needle aspiration cytology, ultrasound, CT and MRI provide essential information on the cyst location that allows optimal preoperative planning.

Ultrasonographic findings comprise solid and cystic structures within a heterogeneous mass[17]. On CT scans, the dermoid cyst appear as a moderately thin walled, unilocular masses filled with an homogeneous, hypoattenuating fluid substance with numerous hypoattenuating fat nodules giving the pathognomonic "sack-of marbles" appearance[18]. MRI of dermoid cysts give variable signal intensity on T1-weighted images and are usually hyperintense on T2-weighted images and are of considerable importance in depicting the relationship of cystic mass and muscles of floor of the mouth. MRI has been reported to be superior to other imaging modalities in demonstrating the exact location and extent of cystic lesions. This in turn aided surgical planning. While FNAC biopsy of dermoid cysts may provide sufficient diagnostic material, this method is complicated by potential sampling bias, given the copious keratinaceous cyst contents and relatively scarce epithelium-lined cyst wall. Needle biopsies of a dermoid cyst will often yield inconclusive, variable or non-diagnostic results. Imaging modalities are valuable to distinguish these lesions for a provisional diagnosis. In the case report described here, ultrasonography and FNAC are inconclusive.

Treatment comprises of surgical excision. The lesions can be exposed by intraoral or extraoral approach depending upon its location in relation to mylohyoid muscle. Lateral Dermoid cyst of the submandibular or sublingual space and are most conveniently excised via submandibular approach[13]. Large lesions may require both intraoral and extraoral incisions to provide direct visualization. In our case, surgical excision was done under local anaesthesia via submandibular approach. Recurrences are unusual after total surgical excision[18,19]. The present case was approached surgically by a submandibular approach.

Intraoperative it was lateral in position. Histologic examination established a diagnosis of Dermoid Cyst. All true dermoid cysts are lined by epidermis with the presence of adnexal structures such as sweat glands, sebaceous glands, hair follicles. In our case it was lined by sebaceous glands confirming the diagnosis. 5% rate of malignant transformation of the teratoid variety of oral dermoid cysts has also been reported in literature[18]. In our case though the lesion was long standing, there was no evidence of malignant transformation. The patient is under review since last one year without any recurrence.

Conclusion

Laterally situated dermoid cysts are rare lesions but should nevertheless be considered in the differential diagnosis of any lateral oral cavity or cervical lesion in both adult and pediatric patients. FNA interpretation is limited due to the cystic nature of these masses. Ultrasonography may be inconclusive. Radiologic imaging, in particular MRI, may provide nearly pathognomonic xenotypic findings that are valuably diagnostic and may influence surgical planning. Surgical excision is curative, and recurrences are rare.

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Legends

Fig 1 A. Pre-operative picture showing swelling in left submandibular swelling.
Fig 1 B. with no obvious swelling intraorally.
Fig 2. Outline of lesion.
Fig 3. Ultrasonography shows subcutaneous mass of appr.
Fig 4. Submandibular incision.
Fig 5 Cyst floor.
Fig.6: Macroscopic view of excised specimen.

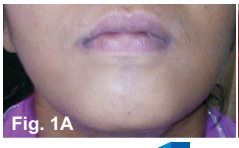
Fig. 1A


Fig. 1B


Fig. 2

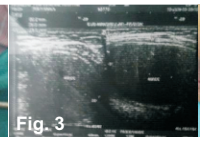
Fig. 3


Fig. 4


Fig. 5

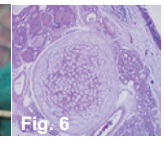
Fig. 6