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An Uncommon Case of Inflamed Giant Sublingual Dermoid

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Abstract: *Sublingual dermoid are one of rare sites for dermoid cyst. A giant size dermoid of more than 5 cm diameter is very rare. The inflammation of a sublingual dermoid is uncommon. The inflammation can occur because of continuous friction due to mastication movements, resultant trauma and subacute infection. This dermoid was producing any interference with the tongue movements like speech, mastication and swallowing. The diagnosis was confirmed of MRI scan. The excision of giant size inflamed dermoid was done by oral route under general anaesthesia. Not much of difficulty was encountered during this surgery. The postoperative period was uneventful. We recommend intraoral route for giant size sublingual dermoid even if complicated by inflammation.*

Keywords: *Dermoid Cyst; Sublingual; Rare Dermoid, Infected Dermoid; Giant Dermoid*

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I. Introduction

Dermoid cysts are congenital lesions caused by a defect in fusion of the first and second pharyngeal arches. These are composed of tissues from different origins: ectoblastic, mesoblastic, or endoblastic· A true dermoid cyst is covered with epithelium showing keratinisation and dermal appendices [1]. It is located in the midline of the body and grows symmetrically [2].These may also appear at places where two embryonic processes meet, e.g. Outer angle of orbit, behind the pinna, below the tongue in midline. Swelling is round, smooth and margins yield to the pressure of finger and not slip away. The swelling remains free from skin and underlying structures.These may cause indentation of bone if underlying structure is bone. Dermoid cysts in the sublingual region are rarely observed [3]. When they develop, they do not appear until they grow large or are infected. Depending on the relationship with the mylohyoid muscle, these may induce an increase in the volume of sublingual or submandibular tissues.As the cyst grows, swelling of the floor of the mouth appears and the tongue is pushed up and backwards. They may reach a large size and involve more than one anatomical area, including that near the hyoid bone. These can occasionally cause serious problems with swallowing and speaking [4].

The differential diagnosis includes an abscess, ranula, or rarely malignancy. Diagnosis can be by CT scan or MRI. Treatment for dermoid cyst is complete surgical removal, preferably in one piece and without any spillage of cyst contents. Enucleation is done via intraoral or extraoral approach. Intraoral route is used for small or moderate dimension cyst above mylohyoid muscle. Extraoral approach is preferred for large size cysts [5]. Marsupialization, a surgical technique is inappropriate due to risk of malignancy. Superficial dermoid cysts on the face usually can be removed without complications. Removal of rare dermoid cysts requires special techniques and training. Dermoid cyst ablation is a minimally-invasive procedure in which a small needle and plastic sleeve are used to enter the dermoid cyst. We report a rare case of infected giant size dermoid cyst in sublingual area.

II. Case Report

A thirty-two years femalepatient presented to outpatient department with chief complaints of swelling in submental region for one anda half year. The swelling wasinsidious in onset and was gradually progressive in size reaching to the present size. It was not associated with any pain. There was nodifficulty in chewing and swallowing of solid food or anydifficulty in breathing. There was no history of any trauma or fever.

Extraoral examination revealed firm swelling of size 4×3 cm in submental region which was free from skin, well circumscribed, smooth surface, not mobile with protrusion of tongue or deglutition. On intraoral examination, there was a solitary swelling of size 4 × 3 cm in sublingual regionwhich was soft and nontender.

The swelling was well circumscribed, dome shaped,sessile, more on the left side than right side. The mucosa overlying the swelling was normal, free from swelling and there were no secondary changes. (FIG 1)There was no associated discharge from swelling. It was non pulsatile and cough impulse was absent. Tongue was normal and swelling didn't vary with movement of tongue. Transillumination test was negative. There was no clinical cervicallymphadenopathy.

FIG 1. Showing submental and intraoral swelling

On Ultrasonography a cystic lesion with internal echoes, debris and foci of macro calcification was present in sublingual region with sub-centimetriclymph nodes in upper cervical region.Contrast enhanced CT revealed peripherally enhancing hypodense predominantly cystic lesion measuring 5 × 4.2 × 3.5 cm in left sublingual space and floorof mouthwith multiple rounded faintnodular dependent calcification in it. (FIG 2a,b) Centimetric and sub-centimetric lymphnodes were seen in bilateral level Ib, level II, and level III. CT Features were suggestive of dermoid cyst or plunging ranula.

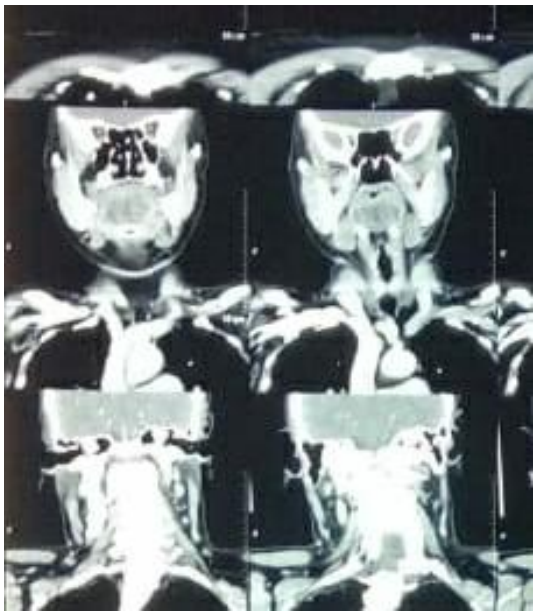

FIG 2a. CT showing sublingual dermoid

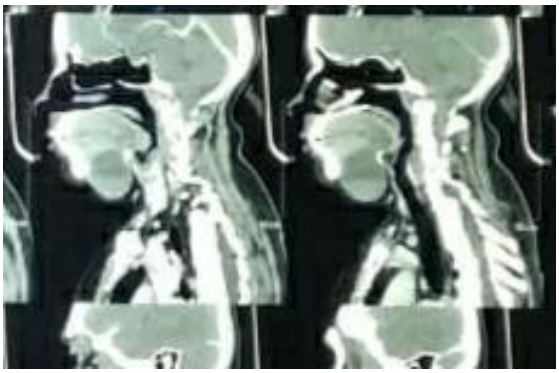

FIG 2b. CT showing sublingual dermoid

Patient was planned for surgery and excision was done under general anaesthesia with nasotracheal intubation. Para-median tongue stitches with 2-0 silk were applied. The tongue suture was used as traction sutures to achieve good plane of dissection right through the procedure. Intraoral horizontal incision was given in the floor of mouth over the most prominent part of cyst. Incision line was infiltrated with diluted lignocaine adrenalin solution. Sharp and blunt dissection was done to free the cyst from fibers of mylohyoid muscle. The muscle was split in the direction of the fibers using sharp dissecting scissors to minimize bleeding. Once capsule was reached, blunt dissection was done to free the capsule.Sublingual duct was identified and preserved. (FIG 3)

FIG 3. Intraoral excision

Cyst was removed per oral. (FIG 4) The resultant defect was closed in layers after ensuring hemostasis. Postoperatively,clear fluids were started on second post-operative day. Semisolid soft diet with meticulous oral hygiene was continued for 2 weeks. Cyst fluid was sent for pus culture and sensitivity which was found to be sterile. Cyst wall was sent for histopathological examination. (FIG 5)

FIG 4. Excised cyst

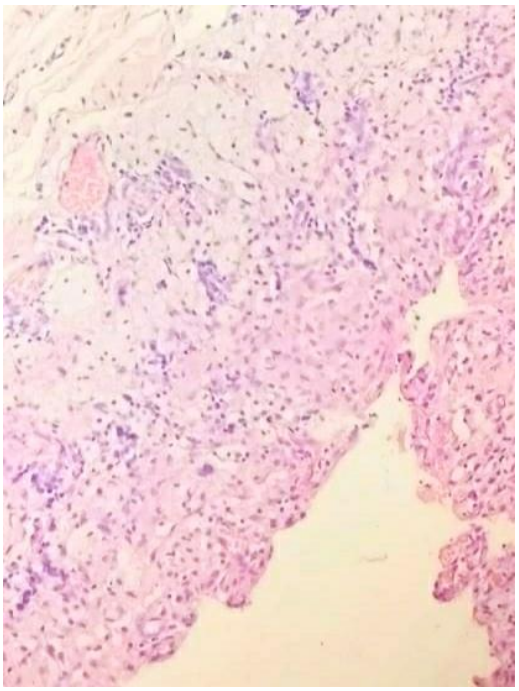

FIG 5. Microphotograph showing inflamed cyst wall

III. Discussion

Dermoid cysts are unusual swellings which present in childhood and can occur in every part of the human body like nose, sinuses, orbits, scalp, rectum, ovaries, abdomen, testicles, salivary glands, spinal cord brain etc. The floor of the mouth is the second most common site for dermoid cysts in the head and neck region after the lateral eyebrow. Dermoid cysts ofmouth are most frequently located on the median line of the floor and are caused by retention of germinal epithelium during the growth of mandible and hyoid branchial arches and are considered rare [6]. They represent 1.6% of all dermoid cysts. 25% of the cases occurring in the head and neck region occur in oral cavity [7]. When they occur in the oral cavity, dermoid cysts most commonly involve sublingual, submental, or submandibular regions, although other sites have been reported, including the lips, tongue, and buccal mucosa [8].According to the anatomic relationship, dermoid cysts can be designated as: submental/median geniohyoid (between geniohyoid and mylohyoid muscles), sublingual/median genioglossal (above mylohyoid and genioglossal muscles), and submandibular/lateral [9].Even when they are laterally positioned; their stalk can often be traced to the midline.The location of the cyst is important in determining the surgical approach.Clinically, the dermoid cysts usually present as a painless, well-circumscribed, thin-walled, unilocular, slow-growing mass at the sublingual, submental or submandibular region. As cyst grows, a swelling

in the floor of the mouth appears, and the tongue is pushed up and backward causing difficulty in mastication and speech. These well encapsulated lesions typically feel "dough like" on palpation, although they may be fluctuant and cyst like based on consistency of the luminal contents, that may range from a cheesy, sebaceous to liquefied substance. Depending on the size of lesion, it can cause dysphagia, dysphonia, and dyspnea. In the case of lower localization, they present with a characteristic double chin[5]. These can attain a large size before presentation. However, there can be sudden increase in the size of these lesions due to the onset of puberty when there is an increase in the secretion of sebum from the sebaceous glands or may be due to secondary infection of the cyst contents either by blockage of salivary glands involved in the cyst or by implantation of oral microbials into the cyst through trauma causing pain, trismus, fever, dysphagia, odynophagia and cervical lymphadenopathy.The bimanual palpation, plain radiographs and orthopantomogram are not of much help. The diagnosis is made on computerized tomography, magnetic resonance imaging and ultrasonography. Ultrasonography represents the first choice of imaging technique because it is reliable, economical, and without x-ray exposure, so it is easily suitable for young patients also. It may be helpful in differentiating between solid vascular and cystic lesions.

Histologically, midline dermoid cysts of the floor of the mouth are classified according to Meyer's classification into three groups: epidermoid cysts, which consist of an epithelial-lined wall that may be partly keratinized; dermoid cysts, which are epidermoid-like cysts but show evidence of skin appendages, such as hair follicles, hair, sweat, and sebaceous glands [10].Dermoid cysts tend to manifest earlier, usually in the second or third decadeand form slowly expanding, unilocular masses.and teratomas, which contain, in addition to skin appendages, mesodermal elements such as bone, muscle, respiratory and gastrointestinal tissues, and a fibrous capsule.

They may present during infancy or during the second to third decade and form multiloculated masses [11]. The latter type is the only variety that may have a malignant change. All 3 types contain a cheesy keratinous material.

The definite management of these lesions is complete surgical excision with very low recurrence rate. There are no rules regarding the timing for operation as dermoid cysts are mainly congenital, they can appear in every age of life, so the time when they appearis generally the right time to operate on them. Surgical enucleation is facilitated by the fibrous capsule surrounding the cyst that makes it easy to be enucleated. Intraoral approach is used for the sublingual cysts less than 6 cm whereas the extraoral approach is usually chosen for submandibular and submental space involvement with size more than 6cm or in infection process that may interfere to patient's airway[12]. In intraoral approach, a midline vertical mucosal incision is performed along the ventral surface of the tongue; however, only small cysts can be enucleated using this kind of incision but it leads to very good cosmetic and functional results.

Caution should be taken not to rupture the cyst, as cystic contents may act as irritants to fibrovascular tissues, causing postoperative inflammation. Lingual nerve and the submandibular ducts should be preserved.The extraoral incision is made in a natural skin crease, but some degree of scarring is inevitable. There is a risk of damage to the marginal mandibular branch of the facial nerve. The reason for extraoral choice is the fear of not being able to obtain adaquate exposure of the surgical field or incomplete removal of the cyst. Large cysts that perforate the mylohyoid may require both intraoral and extraoral incisions to provide direct visualization of important adjacent structures. Life-threatening complications such as hemorrhage from the lingual vessels and hematoma formation which can lead to significant swelling and edema of the floor of mouth and tongue, resulting in respiratory distress and airway obstruction from elevation of the tongue against the palatal vault can occur in surgery [13]. The cystic fluid can be aspirated to reduce the mass of the lesion. Although it cannot be the definitive treatment, it is useful to facilitate control of the deeper pole of the cyst [14]. Marsupialization may be another treatment option for very large cysts. Recurrences are unusual after absolute surgical excision [15].

IV. Conclusion

In this patient, a giant size dermoid cyst was removed by intraoral route. Clinically the cyst was not infected but cyst was inflamed on histopathological examination. So, the intraoral route is an easy route for excision of giant size sublingual dermoid

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