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Case Report

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Sublingual dermoid cyst: a case report and review of literature

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ABSTRACT

Dermoid cysts in the floor of the mouth are relatively uncommon developmental lesions. They generally arise in the midline along the lines of embryonic fusion of the facial processes containing ectodermal tissue. We report a case of 24 year old lady with a Sublingual dermoid cyst which was surgically excised.

Keywords: Dermoid cyst, Floor of the mouth, Developmental, Surgical excision

INTRODUCTION

Dermoid cysts of the oral cavity are relatively rare in the head and neck region. Among cysts of the oral cavity, <0.01% of the oral cysts are dermoid cysts.[1] The incidence of dermoid cysts in a study done by New et al showed only 1.6% were in the floor of mouth out of 1495 patients.[2] They are thought to arise as a result of a defective development along embryonic lines of fusion containing both ectodermal and endodermal elements. We report an unusual case of sublingual dermoid cyst.

CASE REPORT

A 24 year old female was referred with history of progressively increasing swelling in the floor of mouth for 3 years which was also extending to below the chin. The patient did not report any swallowing difficulties, but her speech was mildly affected. She had no dyspnoea or pain. There was no history of previous surgery or trauma to the oral cavity or neck. Clinical examination revealed a 4x5 cm large symmetrical, midline, soft, non-tender and non-pulsatile swelling in the floor of the mouth, with normal covering mucosa (Figure 1). There was mild displacement of the tongue superiorly and posteriorly. The swelling was seen appearing externally in the submental region measuring 5 × 4 cm which was soft, non-tender and non-pulsatile swelling. Investigations ultrasonography neck revealed hypoechoic lesion with well-defined margins measuring 4 × 4.39 × 2.43 cm noted in the submental region with foci of calcification with in suggestive of lingual dermoid cyst.

Magnetic resonance imaging (MRI) neck demonstrated well defined cystic lesion with multiple septa within seen epicentred at the sub lingual space predominantly on the right side measuring 4.7 × 4.1 × 3.3 cm extending superiorly to base of the tongue and genioglossus muscle, inferiorly limited to mylohyoid muscle. The lesion appeared hyperintense on T2WI and iso to hyperintense on T1WI and showed restriction of DWI, suggestive of sublingual epidermoid cyst (Figure 2). The progressively growing nature of the cyst and its size raised the suspicion of potential compromise to the airway. Surgical excision was therefore advised.

Under general anaesthesia, the patient underwent surgical excision of the cyst via an intraoral approach. Sublingual mucosal incision was given on the right side of floor of mouth extending till midline. Blunt dissection was done and mylohyoid muscle retracted laterally and cyst was carefully dissected from surrounding tissues en bloc and excised in toto. Excised specimen sent for histopathological examination (Figure 3 and 4) and the

wound was closed primarily. Her postoperative recovery has been uneventful. Histopathological examination was suggestive of a epidermoid cyst.

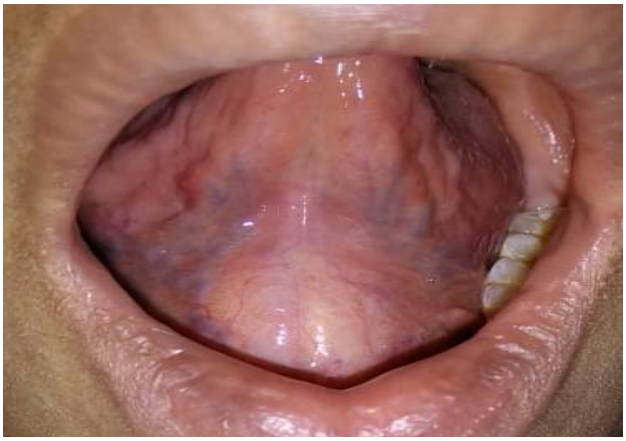

Figure 1: Pre-operative, dome-shaped, symmetrical sublingual swelling.

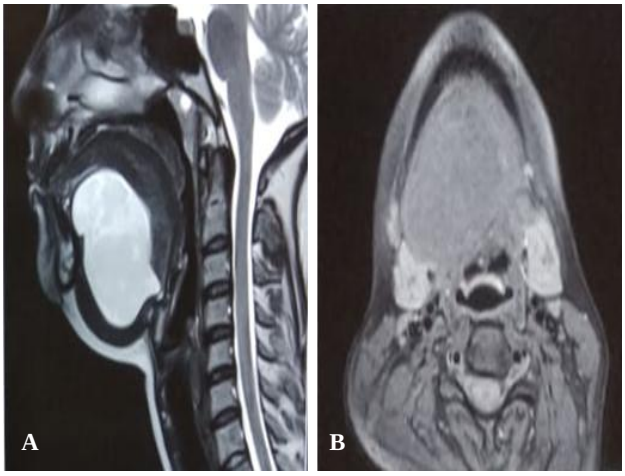

Figure 2: (A) Sagittal and (B) axial MRI neck scans, 5 cm well circumscribed sublingual cystic mass suggestive of sublingual dermoid cyst.

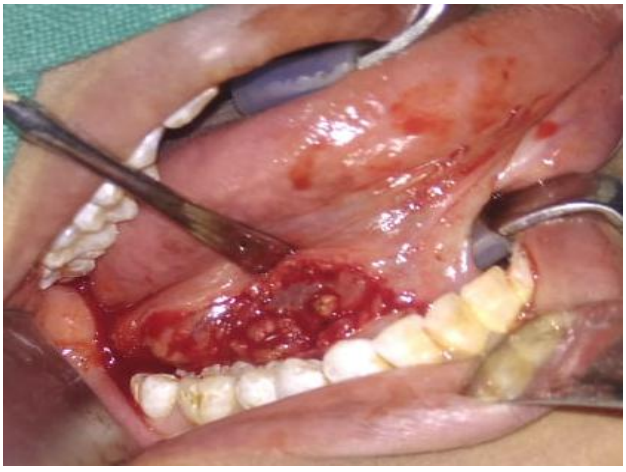

Figure 3: Intra oral approach used for excision of sublingual dermoid.

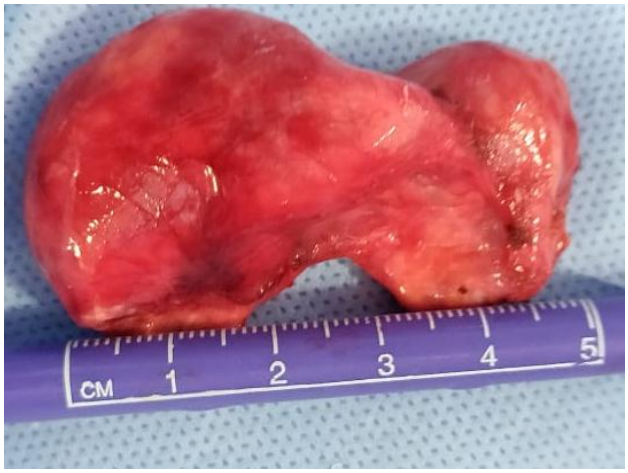

Figure 4: Excised specimen measuring 4.5 × 4 × 3.1 cm.

DISCUSSION

Dermoid cysts of the oral cavity are relatively rare in the head and neck region. In fact, Dermoid cysts occur primarily in the testes and ovaries, and the most common location in the head and neck is the external third of the eyebrow.[1] New et al found that only 6.94 per cent of dermoid cysts occurred in the head and neck, of which 11 per cent were located in the floor of the mouth.[2] The floor of the mouth is the second most common site for the presentation of dermoid cysts in the head and neck region, after the periorbital region.[2] A significant portion of the dermoid cysts in the floor of mouth are situated in the midline. 52% are in the sublingual space and 26% in the submental space. 6% are in the submandibular space while the rest involved more than one of the three possible spaces.[3] There is no apparent sex predilection.[3] Dermoid cysts of the floor of mouth are associated with a bimodal age distribution. Most of the cases are detected during the teenage years with a smaller peak during the first year of life.[4] However, lesion has been detected in individuals as old as 77 years and as young as 7 months.[4-7]

Dermoid cysts of the floor of mouth usually present clinically as a painless, soft and compressible lesion.[8] Meyer divided cysts of the mouth floor into three categories based on histology: dermoid, epidermoid and teratoid.[9] Gordon et al modified Meyer's classification of the cysts using new terminology, referring to the cysts as congenital germline fusion cysts.[9,10] Congenital germline fusion cysts have traditionally been referred to as dysontogenic cysts. The following classification is specific to those cysts within the floor of the mouth, epidermoid – stratified squamous epithelium lining with no dermal appendages; dermoid – stratified squamous epithelium lining with dermal appendages, present within associated underlying connective tissue, including sebaceous glands, hair follicles and sweat glands; and teratoid – stratified squamous epithelium lining with elements of all three germ layers, ectoderm, mesoderm

and endoderm, in the underlying tissue. In our case, the cyst in the floor of mouth was epidermoid cyst with stratified squamous epithelial lining with no dermal appendages.

The cysts can be further classified according to their site of development during embryogenesis: group I develop along the nasolacrimal groove, and give rise to periorbital cysts; group II develop within the prenasal space, giving rise to cysts over the dorsum of the nose; group III develop along the midline of the first and second pharyngeal arches, giving rise to cysts within the sublingual, submental and submandibular regions; and group IV develop in the ventral or dorsal midline, typically in the thyroidal, suprasternal or suboccipital regions.[11] In our case report, according to site of development during embryogenesis, it was group III cyst developed along the midline of first and second pharyngeal arches.

Depending on the location of the dermoid cyst in relation to the mylohyoid muscle, there may also be extra-oral signs. Cysts deep to the mylohyoid often just present with sublingual swelling with or without displacement of the tongue.[12,13] In comparison, cysts that are superficial to the mylohyoid will present with submental swelling or fullness, resulting in a double chin appearance.[14] In our case report, swelling was superficial to mylohyoid and presented with submental swelling. The submental swellings are usually asymptomatic but may also cause dysphagia, dysphonia.[15] Another reason for sudden enlargement is due to secondary infection of the cyst contents with associated fever, pain, trismus and cervical lymphadenopathy.[16]

Three modes of pathogenesis have been described: acquired implantation, congenital teratoma and congenital inclusion.[13] However, as highlighted by Gordon et al, a teratoma is a solid neoplasm and should be considered separate to a teratoid variant germline fusion cyst.[12] In view of this, there are only two true modes of pathogenesis: acquired implantation and congenital inclusion. Acquired implantation is the method by which a small sample of epithelium is separated and implanted within the underlying tissues, typically following surgery or trauma.[17] Congenital inclusion cysts are formed during embryogenesis, where disruption occurs during the fusion of the embryological components, causing in-folding of the epithelium.[18] In our case, cyst appears to be congenital inclusion cyst.

Radiography is an important adjunct in the diagnosis and management of dermoid cysts. Ultrasound, computed tomography (CT) and MRI have all been reported in the literature. Ultrasound may be helpful in differentiating between solid, vascular and cystic lesions.[19] It is generally accepted that CT and MRI imaging allow more precise localisation in comparison to ultrasound. A high signal on T2- and a low signal on T1-weighted imaging typically indicate the presence of cystic structures.[20]

Aspiration cytology may be useful in differentiating between types of cysts.[21] However, the high-density fluid contents of dermoid cysts do not easily aspirate, limiting the usefulness of this technique.[22] Histological examination is essential to confirm the diagnosis.

Treatment for sublingual dermoid cysts is almost always surgical. The choice of surgical approach depends on size of cyst. An intraoral approach is more suitable for sublingual cysts of up to 6 cm in size, whereas an extraoral approach may be advantageous for those cysts greater than 6 cm.[23] In our study, we excised the cyst by intra oral approach as size of cyst was less than 6 cm. There are, however, case reports that detail the removal of very large dermoid cysts carried out by intraoral approach, which claim a cosmetic advantage. Boko et al reported on the intraoral surgical excision of a dermoid cyst measuring 13 cm in length.[24] Longo et al reported that extraoral incision is mandatory if the cyst is located beneath the mylohyoid muscle.[25] In our case, the cyst was located superficial to mylohyoid muscle.

Prognosis following surgical removal is excellent, with a low incidence of relapse. Malignant changes have been recorded in dermoid cysts, by New and Erich, but not in the mouth floor.[2] However, a 5 per cent rate of malignant transformation of the teratoid type or oral dermoid cysts has been reported by other authors.[26]

CONCLUSION

Dermoid cysts in the mouth floor are relatively uncommon developmental lesions which accounts for only 0.01% of all oral cysts. Due to its scarcity and relatively non-distinctive clinical presentation it is commonly misdiagnosed. Differential diagnosis is of paramount importance, as recommended surgical techniques vary depending on lesion size and anatomical position. Prompt diagnosis and treatment can prevent potential airway compromise. Treatment is surgical excision. Approach depends on size of cyst and involvement of mylohyoid muscle.

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