Case Report
Unilateral Dermoid Cyst of the Floor of the Mouth

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Abstract:

Dermoid cysts are the developmental cysts that occur in the head and neck.

The vast majority of dermoid cysts of the floor of the mouth (DCFOMs) are located in the midline; which is the second most common site of these cysts in the head and neck region.

This case report describes a unilateral dermoid cyst in the floor of the mouth in a 19 years old Saudi girl. This lesion appeared as a left sublingual doughy swelling of 2 months duration which elevated the tongue and caused difficulty in eating.

Surgical excision was done through oral approach. The histopathology examination confirmed the diagnosis of a dermoid cyst having characteristic stratified squamous epithelial lining and focal skin adenexa in the subepithelial tissue. The patient has been followed up for 28 months without any recurrence.

Key Words: Dermoid cysts, the floor of the mouth.

Introduction:

Dermoid cysts (DCs) result from defective embryonic development (dysontogenic cysts) and represent the cysts arising from the entrapped germinal epithelium during the closure of mandibular & hyoid branchial arches.[1,2] These have characteristic histology consisting keratinizing stratified squamous lining epithelium associated with many dermal adnexal structures comprising pilo-sebaceous units and sudoriferous glands in the cyst wall. Other dysontogenic cysts include the epidermoid cyst having the lining of keratinizing stratified squamous epithelium lacking any dermal adnexal structures and the teratoid cyst that has the structures/ epithelia derived from all the three embryonic germ layers[1-3].

The dermoid cyst in the floor of mouth (DCFOMs) is an uncommon lesion. Dermoid cysts at the mouth floor account for only 0.01% of all oral cysts, and 1.6 % of all body dermoid cysts. DCFOMs comprise 7% of all head and neck cysts[4-8]. However, the frequency of dermoid cysts occurrence in the floor of the mouth is the second most common in the head and

neck region after the lateral eyebrow site. DCFOMs[4-5] have been identified in patients in all ages, but they are found more frequently between ages of 15 to 35[7-9]. Their increased incidence & rapid growth in young persons may be due to the increased sebum production by sebaceous glands during puberty[10]. Other factors causing their rapid enlargement may be the complication of secondary infection or very rare development of carcinoma[11].

Dermoid cysts typically occur in the midline presenting symmetrical lesion, but lesions may affect one side only.[12]

Case Report:

A 19 years old, Saudi, female presented on December 2009 to oral & maxillofacial surgery department at King Fahad hospital, Hofuf, KSA with a complaint of swelling under her tongue with difficulty in eating. Swelling started small and has increased in size over 2 months. Her medical history was unremarkable.

Extra-oral examination revealed non-tender doughy swelling in the submental region without changes of skin color and no lymphoadenopathy (Fig.1)

Fig. 1: doughy swelling of the submental region.

Intra-oral examination showed a well-defined swelling 4x2 cm on left side of the floor of the mouth elevating the tongue without mucosal ulceration or color changes. Hard tissue examination revealed caries of teeth 17, 27, 36, 37, 46, 47 and missing tooth 26 (Fig 2).

Fig. 2: unilateral doughy swelling of left sublingual region.

Plain radiographs including (occlusal & OPG) were performed along with computed tomography with contrast (CT scan) which showed unilocular well defined radiolucency of the left side of the floor of the mouth and the absence of salivary calculi. The radiolucent lesion was homogenous and low in attenuation reflecting the presence of keratin (Fig 3).

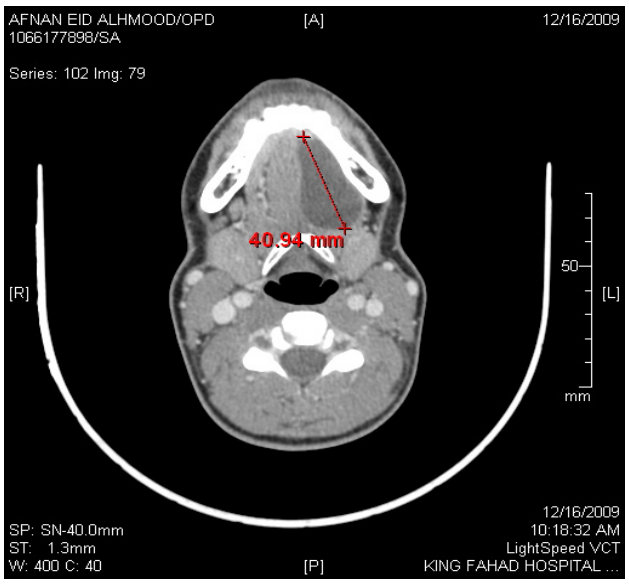

Fig. 3: Axial C.T. scan shows unilocular well defined radiolucency of left side of the floor of the mouth

CT scan confirmed the sublingual location of this lesion above the mylohyoid muscle.

Intra-oral surgical approach was planned for the sublingual cyst removal. Excision of left sublingual salivary gland and cystic lesion were done with identifying of the lingual nerve and Wharton's duct through sublingual incision under general anesthesia. The cyst was well defined per-operatively and did not show any attachment with the sublingual salivary gland or salivary duct.

Histological examination of the excised lesion showed a keratin filled cyst lined by keratinizing stratified squamous epithelium; many skin adnexa including hair follicles & few atrophic sebaceous gland acini were present in the subepithelial fibrous tissue wall; conforming to the diagnosis of a dermoid cyst (Fig.5).

Patient was referred to dental center for restorative assessment and management of deeply carious teeth no 17, 27, 36, 37, 46 and 47. Patient has been followed up for 28 months with uneventful progress and no evidence of recurrence. (Fig.6)

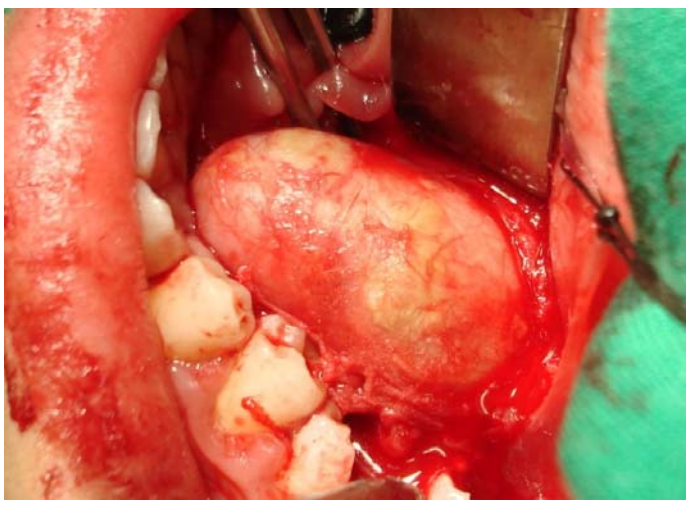

Fig. 4: yellowish cystic lesion excised through sublingual incision.

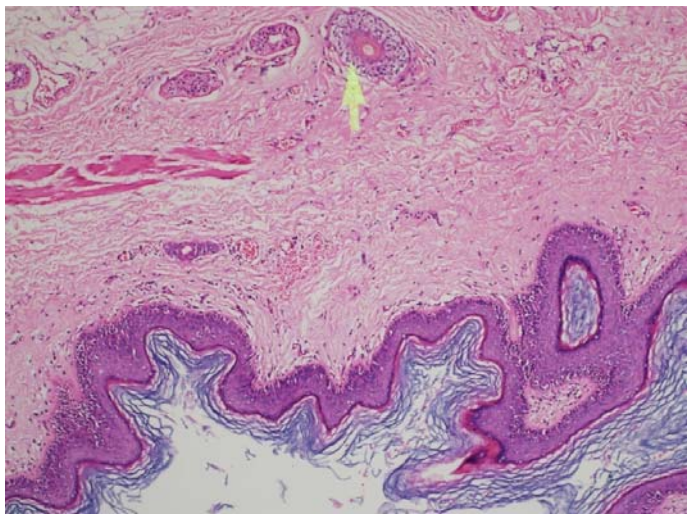

Fig. 5: Dermoid cyst wall with epidermal type epithelium & focal skin adenexa (arrow) in the subepithelial tissue. H & E: 100x.

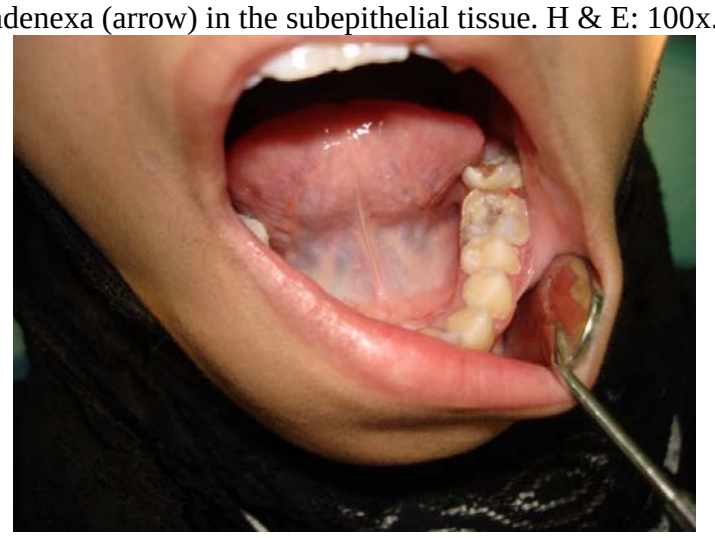

Fig.6: healing 2 months post-operatively.

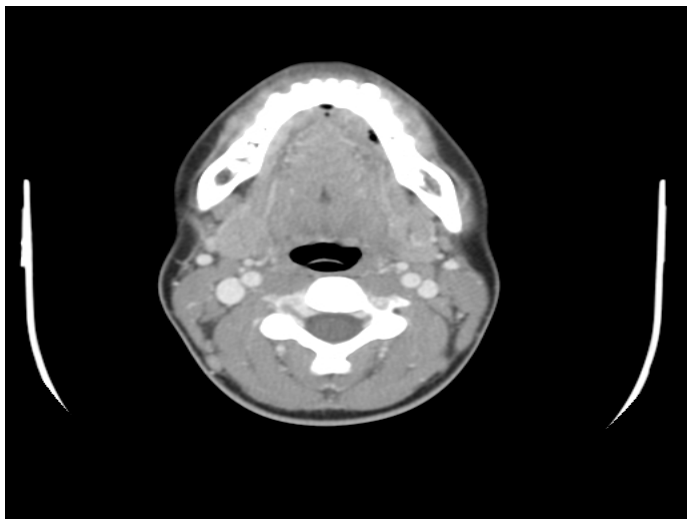

Fig. 7: Axial C.T. scan 28 months post-operatively showed healing of left sublingual region.

Discussion:

The management of lesions in the floor of mouth is much varied; as the differential diagnosis of such lesions include infectious process; salivary gland cysts and neoplasms, thyroglossal duct cyst, cystic hygroma and uncommon mesenchymal neoplasms including lipoma and schwannoma[1, 2, 9, 13, 14 & 15]. Clinical & radiological evaluation is helpful in evaluating the inflammatory lesions but cannot completely exclude an infected dermoid cyst. CT scan evaluation further helps in differential diagnosis; distinguishing homogenous hypoattenuating uniloculated Dermoid & epidermoid cysts from multiloculated ill defined cystic hygromas and from the isoattenuating thyroglossal duct cyst; such evaluation also helps in identifying any lesional attachment or extension with the salivary gland[13 & 14].

Such clinic-radiological evaluation can help in planning the management; however the final identification still rests with the tissue diagnosis by biopsy. Some reports of successful preoperative identification of the sublingual lesions by Fine needle aspiration have been described; but occasionally authors have described difficulty of getting sufficient material from the sublingual dermoid cyst due to the thick cyst contents[1 - 2].

The dermoid cysts at the floor of mouth are manifested during the second or third decade of life as cystic, slightly yellowish masses in the oral floor midline or as a swelling below the chin.[15] The vast majority of DCFOMs are located in the midline (sublingual 52%, submental 26%), 16% involve more than 1 of the 3 possible spaces in the floor of the mouth region (submental, sublingual, submandibular), and only 6% are situated exclusively in the submandibular space (ie, appear to be lateral neck cysts)[16].

Histologically, the dermoid cyst is lined by a thin, uniform orthokeratinized stratified squamous epithelium that can be ulcerated and associated with secondary inflammatory reactions including focal foreign body type granulomatous reaction to the keratin contents. The cyst contains keratin rich sebum material admixed with occasional hair; the dermoid cyst wall characteristically contains skin adnexal structures including hair follicle, sebaceous & sweat glands. In the experience of some authors; the occurrence of hairs in the dermoid cysts of floor of mouth are distinctly uncommon in comparison to the dermoid cysts at the other body site[1, 2, 7, 9, 14-16].

Such histology distinguishes a dermoid cyst from the thyroglossal duct cyst which lacks the skin adnexal structures and instead contains variable amount of thyroid tissue; the cystic hygroma is distinguished by multiple vascular spaces contining lymphoid tissue in the wall. The mucocele ‘ranula’ which is the most common lesion in the floor of mouth is distinguished by mucinous contents and a cuboidal to columnar epithelial lining variably replaced by granulation tissue. Other pertinent differential diagnostic categories including salivary gland tumors and soft tissue tumors possess sufficient distinguishing morphological features and do not enter routinely into the differential diagnosis of dermoid cyst microscopically[9, 13-15].

The treatment for dermoid cysts consists of surgical excision and the approach depends on the cyst’s location; which has been traditionally described as sublingual or genioglossal dermoid cyst if situated above the mylohyoid muscle; and geniohyoid dermoid cyst if it occurs below the mylohyoid muscle (between mylohyoid muscle & neck’s cutaneous muscle) [2,15]. This distinction is aided by the coronal CT-scan evaluation. The sublingual dermoid cyst are best removed via intraoral approach as was done in the present case. An external submental approach is recommended for the submylohyoid (geniohyoid) cysts. Large peri- and trans-mylohyoid cysts may require both intraoral and extraoral incisions[17-19].

The prognosis for the dermoid cyst remains good and any recurrence after a complete surgical excision has not been reported[1, 2, 15-19].

Conclusion:

In conclusion, a case of unilateral dermoid cyst of the floor of the mouth is presented. The occurrence of such a lesion in the floor of mouth raises many differential diagnoses; the radiological evaluation by computed tomography (C.T scan) is very useful to narrow down the differentials as well as for further surgery planning and follow up. Histology makes the final diagnosis; although reports of fine needle aspiration cytology of these lesion has been promising. Surgical excision is the treatment of choice and may be performed intraorally or extraorally depending on the size and location of the lesion with no recurrence expected.

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الكيس الجلدي أحادي الجانب في قاع الفم

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الملخص:

الكيس الجلدي ونظير الجلدي عبارة عن أكياس تكوينية تتكون في منطقة الرأس والرقبة، ويعد قاع الفم ثاني منطقة تتكون فيها هذه الأكياس في الرأس والرقبة، وتتكون معظمها في خط المنتصف.

الهدف من هذا التقرير الطبي هو عرض كيس جلدي أحادي الجانب لمريضة عمرها 19 سنة، أظهر الفحص الإكلينيكي وجود ورم لين تحت الذقن وقاع الفم لجهة اليسار بعمر شهرين مقاسه الطولي 4 سم مما أدى إلى رفع اللسان وصعوبة الأكل.

وقد تم استئصاله من داخل الفم وأظهرت النتائج المخبرية تشخيصه ككيس جلدي وقد تم متابعة المريضة لمدة 28 شهرا دون ظهور آثار لرجوع الكيس.