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Transoral microscopic approach to epidermoid cyst in submandibular space

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Dermoid cysts are developmental anomalies that arise due to defect in the fusion of embryonic lateral mesenchymal approaches during fifth week of embryonic development. Dermoid cysts can be further subdivided into epidermoid , dermoid and teratoid cysts based on the lining epithelium and contents .We present an unusual case of 32 year old female who presented with a swelling in submandibular space, for whom an initial differential diagnosis of plunging ranula was made. The patient underwent an intraoral microscopic excision of tumor under GA following cytology and radiological investigations .Here we would like to stress the importance of multidisciplinary approach to the disease inorder to confirm the diagnosis, considering the manifold differentials and plan surgical approach. Furthermore, the benefits of undergoing an intra oral microscopic excision of the tumor, as it provides better cosmesis , functional outcome and reduced hospital stay.

Keywords: *Floor of mouth; Submandibular space; Microscopic excision; Epidermoid cysts; Dermoid cysts.*

Introduction

Dermoid cysts are benign slow-growing tumors, developmental cysts consisting of either squamous epithelium (epidermoid cysts), skin adnexa (true dermoid cyst), or from all three germ layers (ectoblastic, mesoblastic, and endoblastic - teratoid cyst) [1,2]. Based on origin, these cysts can be either congenital or acquired. It occurs in areas of defect of fusion of lateral pharyngeal arches (mainly the first and second) during embryonic development between the 3^{rd} and 4^{th} weeks.

Dermoid cysts constitute around 1.6 - 6.9 % of cysts appearing in the head and neck region [3]. The overall incidence of dermoid cysts appearing in the floor of the mouth is a mere 0.01 %, and the sites include sublingual, submaxillary, and submandibular area [1]. The usual presentation is a slow progressing painless swelling in the second or third decade. The swelling may progress and rarely presents with difficulty in speech, difficulty in swallowing, or difficulty in breathing [4,5]. Pain and superadded infection may lead to a draining sinus.

We present a rare case of epidermoid cyst presenting as a mass in submandibular space who underwent a transoral microscopic excision of the cyst and the benefits of better cosmesis and functional outcome inpatient care.

Case Report

A 32-year-old female presented to our ENT OPD with complaints of a soft, painless swelling in the submandibular area, which progressively increased in size to involve the floor of mouth during a five-month duration. (Fig1).No h/o trauma or discharge from the swelling. No h/o similar swelling in the past or any other part of the body. On examination, the swelling was soft, non-transilluminate, non-tender, and bimanually palpable, and a working diagnosis of 'plunging ranula' was made. No cervical lymphadenopathy was noted. Saliva could be expressed from the bilateral Wharton's duct. (Fig 2)

On USG, a hyperechoic lesion (40X25mm) with a peripheral cystic component in the right sublingual space with extension into the oral cavity was noted. No internal vascularity was noted, and an impression of plunging ranula made. The following MRI reported a 42x30x39mm lesion in the right sublingual region on the floor of the mouth extending up to the submandibular area, causing displacement of mylohyoid muscle medially. The lesion was hyperintense in T2/STIR and hypointense in T1, suggestive of plunging ranula. (Fig 3,4) With a working diagnosis of plunging ranula, a fine needle aspiration was performed intraorally, which revealed the presence of nucleated and anucleated squames, pointing to a diagnosis of epidermal inclusion cyst.

The patient was subsequently planned for a transoral excision of the tumor under GA. Surgical excision was done under a microscopic vision using Leica F 20 M525 microscope. A curvilinear incision was marked on the mucosa of the floor of mouth about 5 mm medial to the alveolus and lateral to the submandibular duct. Adrenaline (1:200,000) was infiltrated along the marking. The incision was given using a 15 no. blade. Mucosal flaps were raised medially and laterally. The submandibular vein and duct were retracted medially, and dissection was continued using bipolar cautery to reach the superior surface of the cyst. Few sublingual glands were excised in order to expose the superior surface. (Fig 5) Extracapsular dissection was continued on the medial surface, where the hypoglossal nerve was seen abutting the cyst and was carefully dissected. Inferiorly cyst was abutting the submandibular gland but had no attachments to it. Dissection continued laterally, and the cyst was delivered in toto. No significant bleeding was observed during the procedure, and bipolar cautery was used exclusively to achieve hemostasis of minor bleeders. Inverted mucosal sutures were put using 4-0 vicryl. (Fig 6)

In the immediate post-operative period, the patient was assessed for hypoglossal nerve function, which was normal. Lingual nerve function was evaluated on a post-operative day one and was found to be normal. Histopathological examination revealed a cyst grossly with a cut surface showing pultaceous material and a cyst wall of stratified squamous epithelium lined with keratin flakes, which confirmed the diagnosis of epidermal inclusion cyst. (Fig 7)

On post-operative follow-up, the surgical site was found to have healed well, and the patient reported no paraesthesia with good salivary gland functions and no other symptoms. The patient is in follow up for the past nine months and is symptom-free.

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Figure 1. *Clinical Photograph showing swelling in right submandibular region*

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Figure 2. *Clinical Photograph showing smooth swelling in right floor of mouth*

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Figure 3. *T1 weighted MRI image showing hypo intense swelling*

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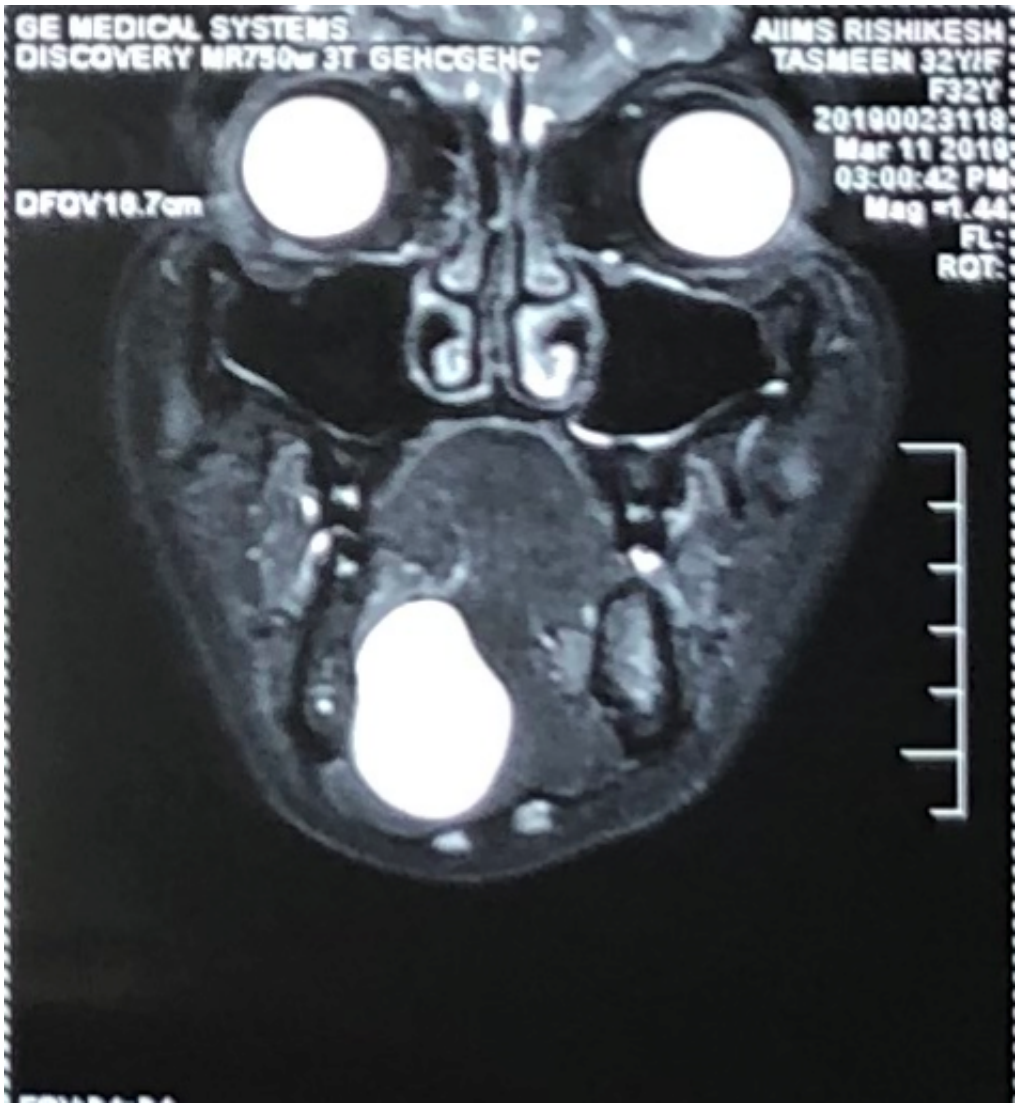

Figure 4. *T2 weighted MRI image showing hyperintense swelling*

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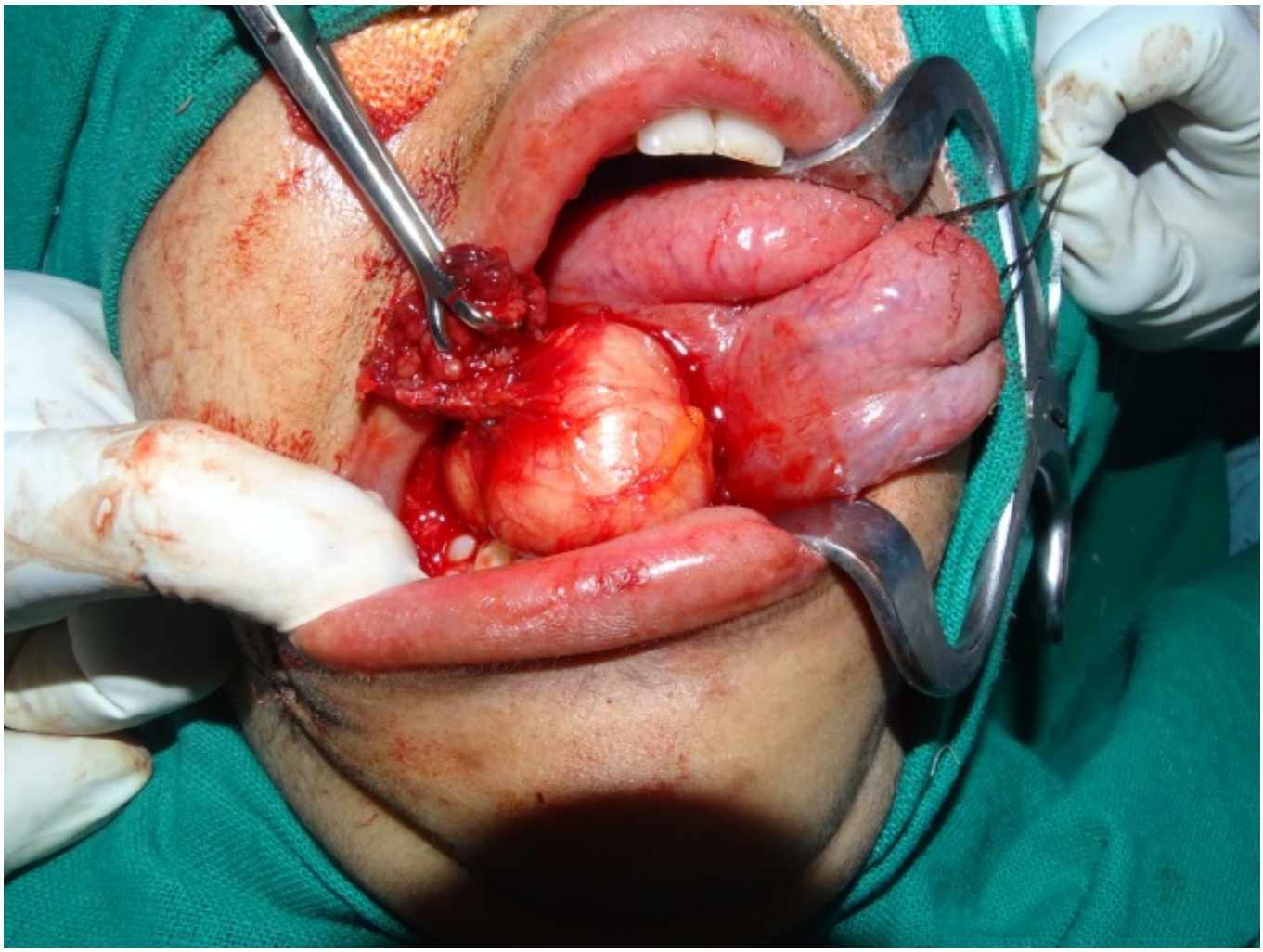

Figure 5. *Intra oral exposure of tumour after elevating mucosal flaps*

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Figure 6. *Excised epidermoid cyst with sublingual gland*

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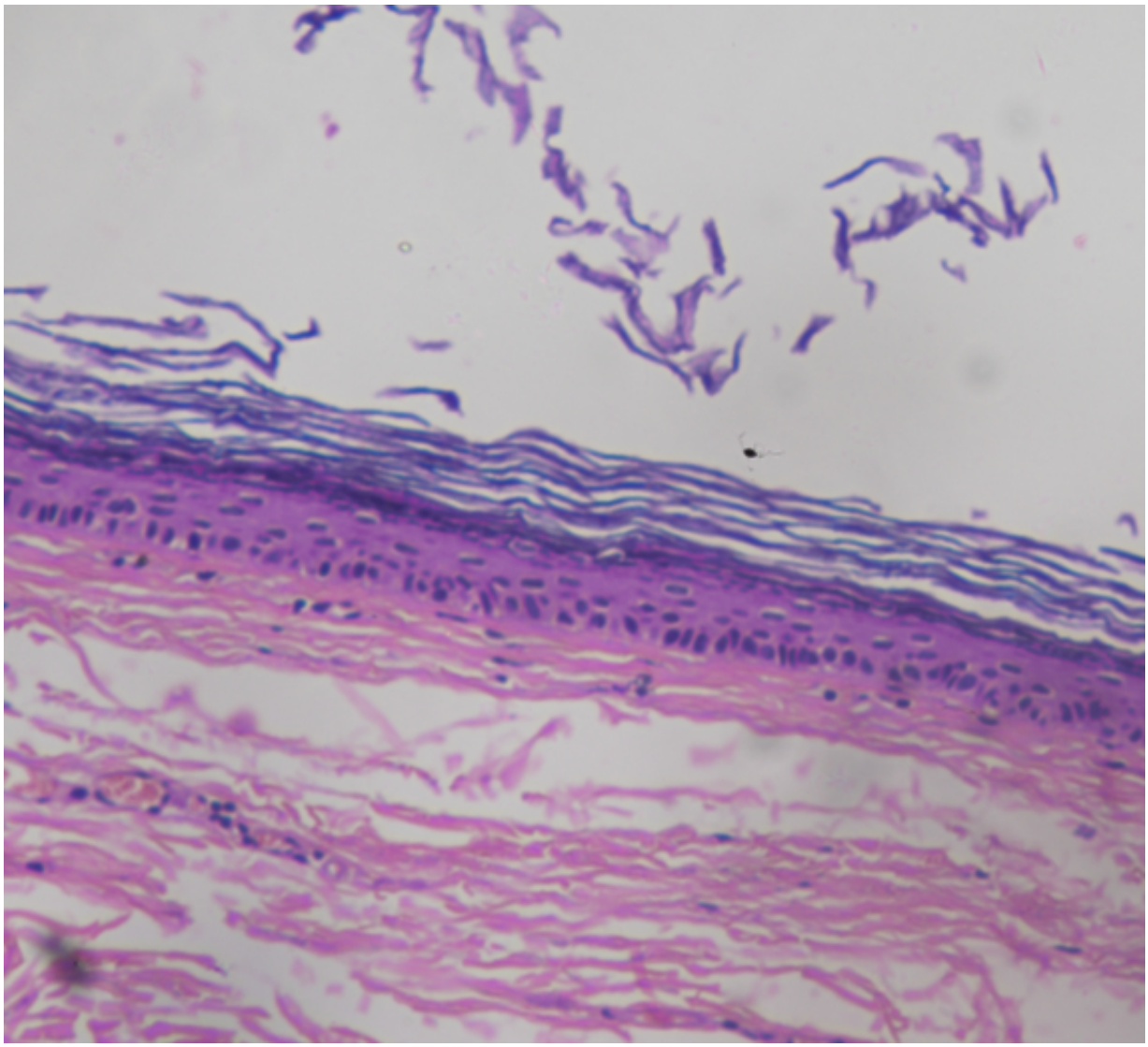

Figure 7. *Cyst wall of stratified squamous epithelium lined with keratin flakes*

Discussion

Dermoid cysts are benign slow-growing tumors, developmental cysts involving soft tissues of the body. Although epidermoid cysts in the form of cutaneous lesions are common in sites of face, neck, and scalp in the head and neck region, their incidence in the floor of the mouth is considered rare [3]. New and Erich reported 1.6% (24 cases out of 1495) cases of dermoid cyst in 1937 [6]. The usual presentation is a slowly progressing painless swelling without lymphadenopathy in the second or third decade. The swelling may progress and rarely presents with difficulty in speech, difficulty in swallowing, or rarely difficulty in breathing. Pain and superadded infection may lead to a draining sinus.

The concept of classifying dermoid cysts into three histological variants based on cystic wall and contents was put forward by Meyer in 1955 [2]. Epidermoid cysts consisted of a stratified squamous epithelial lining. True dermoid cysts had an epithelial lining comprised of elements from skin adnexa, namely sebaceous or sweat gland and occasionally hair with or without follicle. Teratoid cysts contained elements from all the germ layers, ectoderm, mesoderm, and endoderm, namely, bone, muscle, or squamous or ciliated mucosa. The typical age group of the presentation was between 10 and 35 years [7]. The male to female ratio 3:13., suggesting a higher

predominance in females [8].

Based on origin, these cysts can be either congenital or acquired. Congenital cysts develop during the 3rd or 4th week of embryonic development due to entrapment of ectodermal tissues between the 1st and 2nd branchial arch [9].Entrapment of epidermis secondary to penetrating /blunt injury, infection of pilosebaceous follicle leads to acquired dermoid cyst [10].Another theory put forward was epidermoid and dermoid cysts arising as a variant of thyroglossal cysts with predominant epithelial content [11].

Epidermoid cysts presenting in the floor of the mouth can be anatomically classified into three groups based on the relation with underlying muscles and hence the difference in clinical presentations. It can be sublingual or median genioglossal cysts (above geniohyoid), median geniohyoid (in the submental region, between geniohyoid and mylohyoid), and lateral cyst(submaxillary area) [12]. Lesions above mylohyoid present as difficulty in speech, mastication and enlarged tumors, difficulty in breathing. If the lesions arise below the mylohyoid muscle, they present as neck swelling in the submental or submandibular area.

The differentials of swellings present in the floor of the mouth are an infectious cause, plunging ranula, cystic hygroma, dermoid cysts, thyroglossal cysts, lymphoepithelial cysts and pleomorphic adenoma. Rare possibilities include lipoma, neurofibroma, hemangioma [13].

Diagnosis of swelling appearing in submandibular space is made after a thorough clinical examination, cytology, and radiological investigation. In our case, an MRI was also required to delineate the lesion from surrounding tissue suggesting an epidermoid cyst.

The surgical approach to an epidermal inclusion cyst is based on its location and size. The process may be intra oral or transcervical [14]. In some instances, a combined approach might be needed.

While giving an intraoral incision, special care should be taken to preserve bilateral Wharton's duct. Injuries to lingual vessels and nerve, sublingual and deep lobe of the submandibular gland, and even hypoglossal and glossopharyngeal nerve should be avoided. Extensive dissection in these areas may lead to post-operative hematoma or airway compromise, which may require a tracheostomy.

The transcervical approach to cysts in submandibular space involved an incision along with the cyst, and blunt dissection was done. In this method, injury to the marginal mandibular nerve can occur in a few cases. Post-operative scar and duration of hospital stay were prolonged when compared to the transoral approach.

The benefits of cyst excision under the microscope are multiple, unlike conventional transcervical approaches or even a transoral approach. Bilateral Wharton's duct openings could be properly identified. Using a microscope, better delineation of the tumor from surrounding tissue and hypoglossal nerve in our case was helpful. Furthermore, fine dissection of the sublingual gland was done using a microscope, by which we could prevent complications like retention cysts in the future. Lingual vessels and lingual nerve, along with the glossopharyngeal nerve, could be identified and preserved. Dissection using bipolar cautery also helped in minimal dissection. The postoperative recovery of the patient was satisfactory as we had no edema, and the patient's speech was not affected. Furthermore, we could offer better cosmesis, fewer hospital stays, and no paraesthesia.

On conducting a web search using Google scholar, Pub med index, Research Gate, Directory of open access journals, Microsoft academic, Scopus preview, wolfram alpha with keywords like transoral microscopic approach to submandibular space swellings, no articles could be found, hence introducing a novel technique for better patient care.

Conclusion

Epidermoid cysts in the head and neck region are relatively uncommon. The importance of clinical signs, along with the radiology like USG, MRI, and fine-needle aspiration aids to diagnosis. Further, the role of MRI in choosing the surgical approach is crucial. Surgical excision is the treatment of choice. An intraoral approach, under microscopic vision, is a useful and novel technique as it leads to the excellent cosmetic and functional outcome for the patient.

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