CASE REPORT

A Large Lateral Neck Swelling in an Elderly Patient: A Case Report

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Abstract

Cystic lesions are common in the head and neck. The most common are the cutaneous cysts, which are referred to as epidermoid cysts. These cysts present as nodular and fluctuant subcutaneous lesions and they are seen most commonly in the acne- prone areas like the head, neck and the back. However, the presence of benign cystic lesions in the salivary glands is rare. Here, we present a case of a 59 year old male, who presented with a benign large swelling of the neck and floor of mouth.

Keywords: *Epidermoid, cyst, neck, floor of mouth.*

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Introduction

Epidermoid cysts are rare, slow-growing, benign, developmental cysts that are derived from abnormally situated ectodermal tissue. It is defined as a simple cyst lined with stratified squamous epithelium and lumen is filled with cystic fluid or keratin and no other specialized structure[1]. Epidermoid cysts may grow anywhere on the body and about 7% of them are located in the head and neck, with the oral cavity accounting for only 1.6%. Intraorally, it is a benign slow growing and painless entity, which is usually located in the submandibular, sublingual, and submental region[2,3]. Several synonyms exist for epidermoid cysts: epidermal cysts, epidermal inclusion cysts, infundibular cysts and keratin cysts. The epidermal inclusion cysts more specifically refer to the implantation of epidermal elements into the dermis. The infundibular cysts originate from the infundibular portion of the hair follicle. The presence of epidermoid cysts in the salivary gland is a rare entity and these cysts require surgical interventions[4]. Hence, it is very essential to have a pre-operative diagnosis for the workup of the patients. Here, a case of epidermoid cyst of submandibular gland with informed consent of the patient & approval from the institutional ethical committee is presented.

Case Report

A 59 year old male patient reported to our dental hospital with the chief complaint of swelling on right side of the neck from past 7 years. There was no history of pain associated with swelling, difficulty in swallowing, breathing or hoarseness of voice and fever. Patient did not have any tobacco habit and any past history of surgery or trauma in the same region. Past medical history was non-contributory.

Extraorally, there was a large diffuse swelling measuring 10x7 cm on right side of the neck with double chin appearance involving submandibular, submental region and crossing midline. No movement of the swelling was observed during swallowing. On palpation it was non-tender, soft and fluctuant in consistency. Intraorally, a diffuse sessile swelling 4.5 x 1.5 cm was present on right side of floor of mouth pushing the tongue to the contralateral side (Figure-1).

Fine needle aspiration showed presence of eosinophilic proteinaceous material and few inflammatory cells. On ultrasound, it appeared as cystic lesion with echogenic contents in the right submandibular region. Magnetic resonance post contrast T2 weighted images revealed a large cystic lesion with hyperintense signal in the right submandibular region. It measured

9.5x 5.2x 6.4 cm with mild enhancement of wall and restricted diffusion on diffusion weighted sequences suggestive of epidermoid cyst (Figure- 2).

Figure- 1: Extraoral & intraoral extension of swelling in right submandibular region

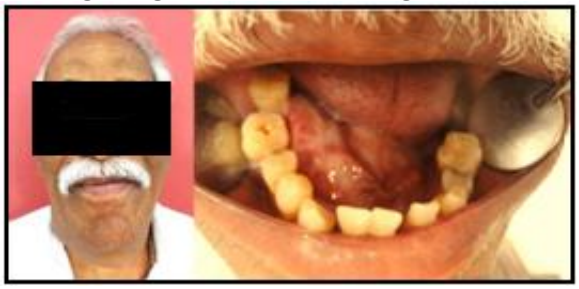


Figure- 2: Magnetic resonance T2 axial section showing large cystic lesion with hyperintense signal post-contrast

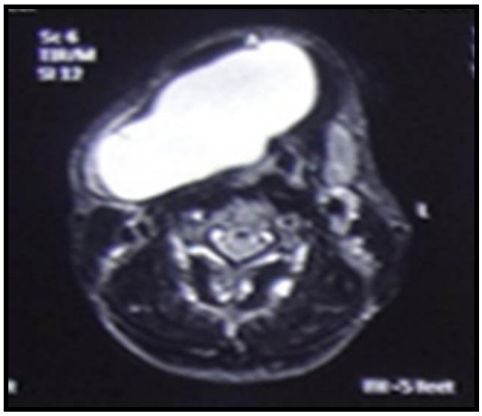
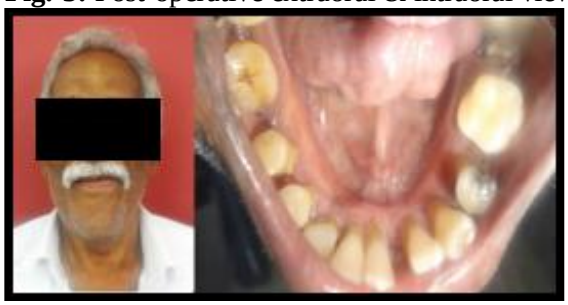


Fig.-3: Post-operative extraoral & intraoral view



Surgical excision of the entire swelling was performed under general anesthesia at a medical hospital and was subjected for histopathological analysis. The specimen excised was yellowish brown soft tissue mass 8cm × 7 cm × 5 cm in dimension. On grossing, it had a thin walled capsule surrounding, with a cheesy white material inside. Histologically, cyst was lined by stratified squamous epithelium and lumen filled with lamellated keratin confirming the diagnosis of epidermoid cyst. Post-operatively patient was followed up for 10 months and no recurrence was noted (Figure- 3).

Discussion

Swelling in the submandibular region of neck is commonly seen in practice. This can be due to enlargement of submandibular gland or lympadenopathy in this area. Submandibular gland enlargement can be due to sialoadenitis or occasionally, benign or malignant tumor. These are usually firm to hard lesions. Cystic lesions of the submandibular gland are relatively uncommon. These can range from epidermoid cyst to submandibular mucoceles[5].

Most clinicians and researchers believe that epidermoid cysts that appear in the floor of the mouth are a result of entrapped ectodermal tissue of the first and second branchial arches, which fuse during the third and fourth weeks in utero. A second theory suggests that midline epidermoid cysts may be a variant of the thyroglossal duct cyst with predominating ectodermal elements[6]. Even if they are congenital, the diagnosis is commonly possible in the second to fourth decade of life. Growth of the cyst may be constrained by hormonal stimulus during puberty, producing a hyper secretion of fat, which would explain the greater incidence in the adult stage. It may also occur due to obstruction in the main salivary duct within the substance of the gland leading to epithelial lined cavity, filled with viscous or semi solid epithelial degradation products[5-7].

Anatomic classification divides the epidermoid cysts of the floor of the mouth into three groups according their relation to the muscles of the floor of the mouth: firstly, sublingual or median genioglossal cysts located above the geniohyoid muscle; secondly, median geniohyoid cysts located in the submental region between the geniohyoid and the mylohyoid muscles and lateral cysts located in the submaxillary region[6]. Epidermoid cyst arising in the lateral neck region involving submandibular gland, as in our case, is extremely rare.

Clinically, they can occur at any age from birth to 72 years but usually become apparent between 15 to 35 years. Males are more commonly affected and may present as small or large masses. Intraorally, it is a benign slow growing and painless entity which is usually located in the submandibular, sublingual and

submental region. They can cause symptoms of dysphagia, dyspnea and dysphonia due to upward displacement of tongue, 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[1,8]. Fine needle aspiration cytology reveals thick, yellowish white, granular fluid containing exfoliated keratin.

Differential diagnosis includes ranula, obstructive or infective sialadenitis, lymphatic or arteriovenous malformation, lymphadenitis, branchial cleft cyst, submandibular cellulitis or abscess and benign or malignant tumours of salivary glands[9].

Diagnosis becomes a dilemma because of non-specific clinical, fine needle aspiration and imaging findings. Definitive diagnosis is based only on histology. Histologically, it consists of thin connective tissue wall, lined by stratified squamous epithelium and desquamated keratin filling the cystic cavity. No dermal appendages are found within the underlying connective tissue[9].

Epidermal cysts which do not cause functional or cosmetic problems are normally not treated. When a cyst is ruptured, infected or inflamed, injections of corticosteroid or incision and drainage have been attempted. Surgical excision appears to be the mainstay of treatment; however, the extent of removal is dictated by adherence of the tumour capsule to the surrounding vital structures. It comprises of total surgical excision without any rupture as spillage of the cystic contents to the underlying fibrovascular structures can cause post-operative inflammation. In cases where pus and blood are excreted, hydrogen peroxide gel can be used to dry out the cyst[10].

Recurrences and rate of malignant transformation to squamous carcinoma and basal cell carcinoma are very rare; however, a 5% rate of malignant transformation of the teratoid variety of oral epidermoid cysts has been quoted in the literature[4,10].

Conclusion

Epidermoid cysts of submandibular salivary gland origin in an elderly patient are quite a rare entity. Here, we reported a case of epidermoid cyst, which clinically appeared as lateral neck swelling extending intraorally upto floor of mouth. Aetiology in this case is unclear, with no history of trauma to the face or intraoral operative procedures. Therefore, oral physician can consider differential diagnosis of epidermoid cyst of submandibular gland in cases of dilemma regarding benign lesions of neck.

Conflict of Interest: *None declared*
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Ethical Permission: *Obtained*

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