(International Print/Online Journal)

SJIF IMPACT FACTOR: 5.565
PUBMED-National Library of Medicine ID-101739732

ISSN (Print): 2209-2870
ISSN (Online): 2209-2862

International Journal of Medical Science and Current Research (IJMSCR)
Available online at: www.ijmscr.com
Volume 5, Issue 4 , Page No: 506-510
July-August 2022

IJMSCR

Submandibular Epidermoid Cyst – A Rare Case

Dr. Avinash Kumar[1], Dr. Manjari Kishore[2], Dr. Garima Sinha[3], Dr. Silky[4]
[1,4]M.S.,[2]M.D., D.N.B, [3]M.D.,
[1,2,3]Assistant Professor, [4]Senior Resident,
[1,4]Dept. of E.N.T., [2]Department of Pathology,
[1,2,4]Noida International Institute of Medical Sciences (NIIMS),
Noida International University (NIU), Greater Noida, U.P
[3]Department of Anaesthesia & Critical Care Medicine, School of Medical Sciences and Research & Associated Sharda Hospital, Sharda University, Greater Noida, U.P.

***Corresponding Author:**
Dr. Manjari Kishore
Assistant Professor, Department of Pathology, Noida International Institute of Medical Sciences, NIIMS, Greater Noida, UP

Type of Publication: Case Report
Conflicts of Interest: Nil

Abstract

Epidermoid cysts are relatively common benign cysts that can be seen anywhere in the body. They are seen in approximately 7% cases in head and neck region. They can be seen in any age group with male preponderance. These cysts are usually painless, firm, subcutaneous nodules filled with keratinous debris. They can occur anywhere in the body, but are frequently seen on face, trunk, neck, extremities, and scalp. Histopathologically, they are lined by stratified squamous epithelium. Entrapment of epithelial remnants into deeper tissues during embryonic fusion is believed to be one of the etiologies of these cysts.

Herein we report a case of submandibular epidermoid cyst in a 38-year-old male patient. Histological examination confirmed the diagnosis of epidermoid cyst.

Keywords: Epidermal inclusion cyst, FNAC, salivary gland, histopathology

Introduction

Epidermoid cysts are benign cystic malformations.[1-2] They are slow growing, asymptomatic and painless lesions. These are variably termed as epidermal inclusion cysts or infundibular cysts.[2-3] These are common benign intradermal lesions, which can be noted virtually anywhere in the body. Several theories have been postulated for its occurrence. They can be primary in origin but are usually secondary to a traumatic event which implants the follicular epithelium into the dermis.[3-5] They are more commonly noted in male with site predilection for scalp, face, neck, trunk and back.[4-6]

The diagnosis is confirmed by histopathological evaluation with presence of stratified squamous epithelium surrounding the laminated layers of keratin.[1-4] Surgical enucleation is the treatment of choice with lowest rate of recurrence.[5-6]

Here we describe a case of epidermoid cyst of right submandibular gland which is uncommon location for this cyst to occur.

Case Report

A-38-year-old male presented with swelling in right submandibular region for the last 10 years. The patient was operated 4 times in rural hospital. The swelling was initially small in size and gradually progressed to its present size. There was slow enlargement of the mass. There was no history of diabetes mellitus, hypertension or any other comorbidities. The patient gave history of four

surgical intervention in rural hospital. Incision and drainage along with aspiration was done on all the occasions.

On examination, an approximately 8x3 cm swelling was seen in right submandibular region extending from 2 fingers below the angle of mandible to medial side of mandible [Figure 1 A-B]. The swelling was painless and mobile with extension to the medial side of the mandible. Intraorally, swelling was present on the floor of the mouth on right side and was elevated due to pushing effect of the swelling. The swelling was soft in consistency and mobile. It was non-tender, and no localized rise of temperature was noted.

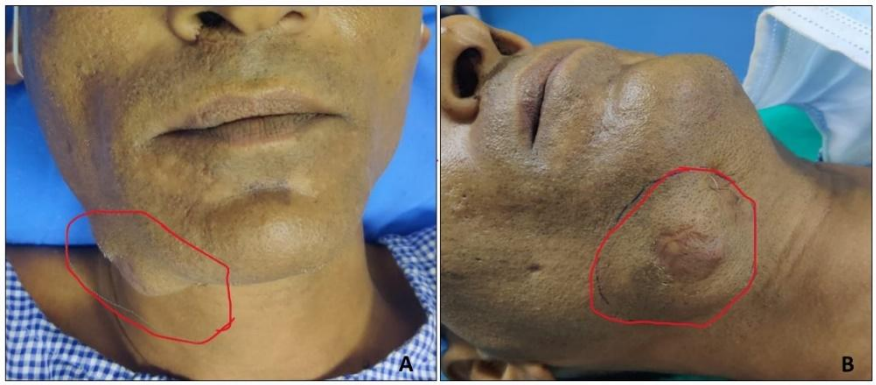

Figure 1A-B: Clinical images showing the right submandibular mass

Due to repeated aspiration and incision and drainage in rural hospitals skin over the most dependent part was rough and puckered. The patient presented with discharge from that area. However, in rest of the areas skin was normal in colour, texture and non-adherent. Contrast enhanced computed tomography (CECT) showed a well-defined, lobulated cystic lesion, measuring 6.3x3.7 cm, located anterior to submandibular gland under the platysma and extending to medial aspect of right hemi-mandible with no contrast enhancement. No bony erosion was present [Figure 2]. The lesion was intending the anterior portion of right submandibular gland and did not cross the midline. CECT features were suggestive of an underlying benign etiology.

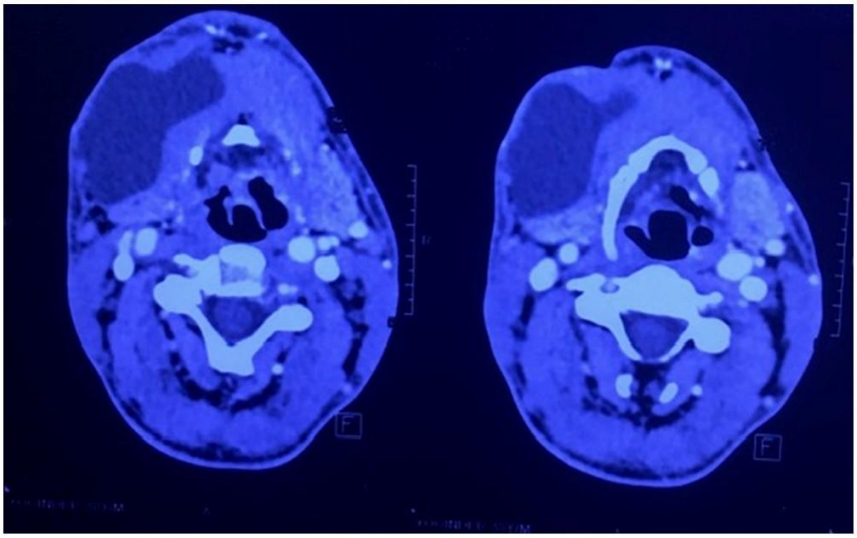

Figure 2: CECT images showing the right submandibular cystic lesion

Volume 5, Issue 4; July-August 2022; Page No 506-510

Fine needle aspiration cytology (FNAC) of the lesion was done. Papanicolaou & giemsa stained smears revealed numerous anucleate squames along with few mature squamous epithelial cells and mild inflammation. Cytological features were suggestive of Epidermal inclusion cyst [Figure 3A-B]. Excision & histopathological correlation was advised.

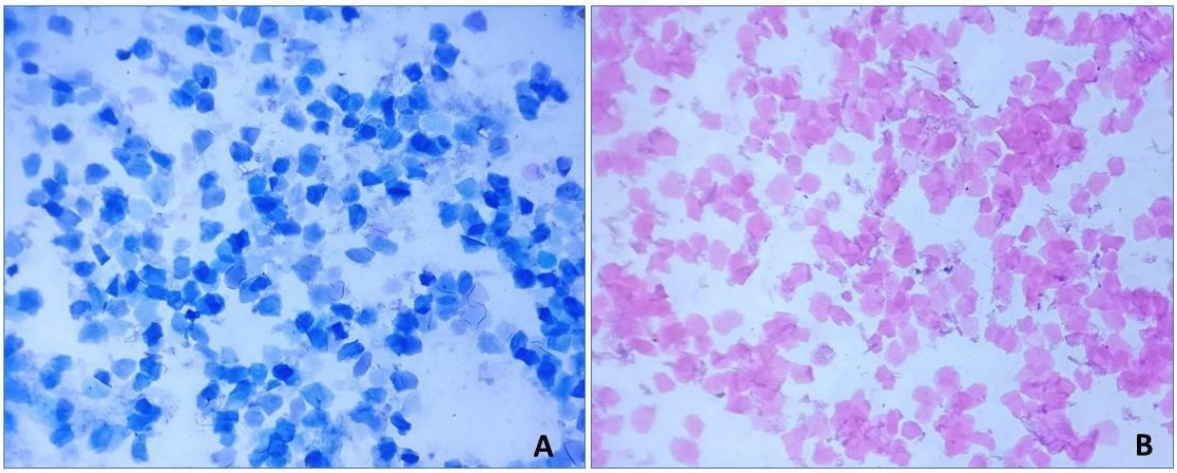

Figure 3A-B: Papanicolaou [A] & Giemsa [B] stained smears revealing sheets of anucleate squames [A: Pap, 20X, B: Giemsa, 20X]

The baseline blood investigations were within normal limits. Surgical excision was planned under General nasotracheal anaesthesia. The patient was accepted for surgery under ASA class I risk status.

Standard NPO orders were followed. Before shifting to OT, nebulization of the patient with 3ml of 4% lidocaine, otrivin nasal spray to the more patent nare and gargling with 2% lidocaine viscous was done.

Awake fibre-optic nasotracheal intubation with 7.0mmID cuffed portex tube was done after adequate premedication (inj. Glycopyrrolate 4mcg/kg i.v., inj. Midazolam 0.5mg i.v.) and mild sedation (inj. Dexmedetomidine 1mcg/kg i.v. over 30 min.) of the patient after blocking glossopharyngeal, superior and recurrent laryngeal nerves. Anaesthesia was maintained with inhalational and i.v. agents.

Excision of the submandibular gland with enucleation of the cyst was done [Figure 4A]. Adhesion of the cyst with submandibular gland was present and hence it was not possible to preserve the gland. About 75ml blood loss was replaced with crystalloids. After anaesthetic reversal and extubation of the patient, post-operative course was uneventful. Drain was applied which was removed after 48 hours.

The excised submandibular gland and cyst was sent for histopathological examination. Grossly, a cut-open cyst was received, measuring 3x3 cms along with a grey-brown soft tissue mass labelled as submandibular gland, measuring 6.5x2 cm [Figure 4B]. Cut section of cyst revealed presence of thick pultaceous material. Cut section of glandular tissue showed grey-brown areas.

Volume 5, Issue 4; July-August 2022; Page No 506-510

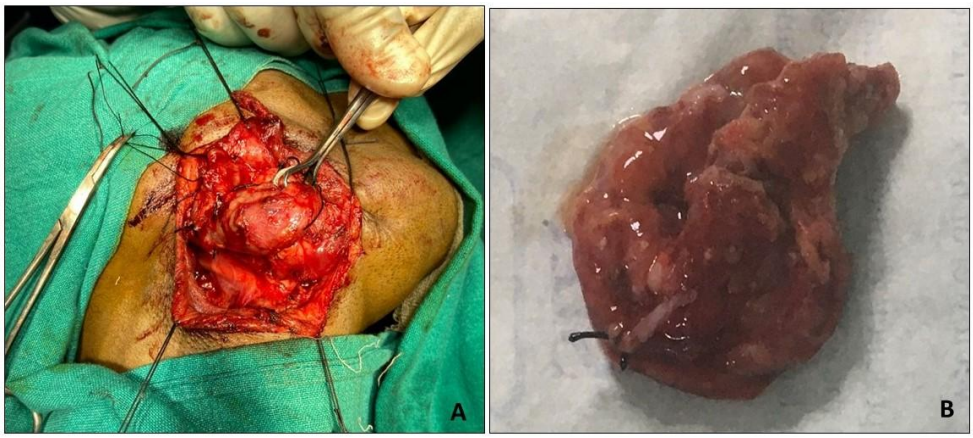

Figure 4A-B: A- Intraoperative image of submandibular cystic lesion; B- Excised mass sent for histopathological examination

Sections showed a cystic cavity lined by keratinizing stratified squamous epithelium [Figure 5A]. Moderate chronic inflammation along with marked congested vessels were noted in the sub-epithelial tissue. Sections from the submandibular gland tissue showed normal histology of salivary glands with few dilated ducts and congested vessels [Figure 5B]. A definitive diagnosis of epidermal inclusion cyst of submandibular gland was made.

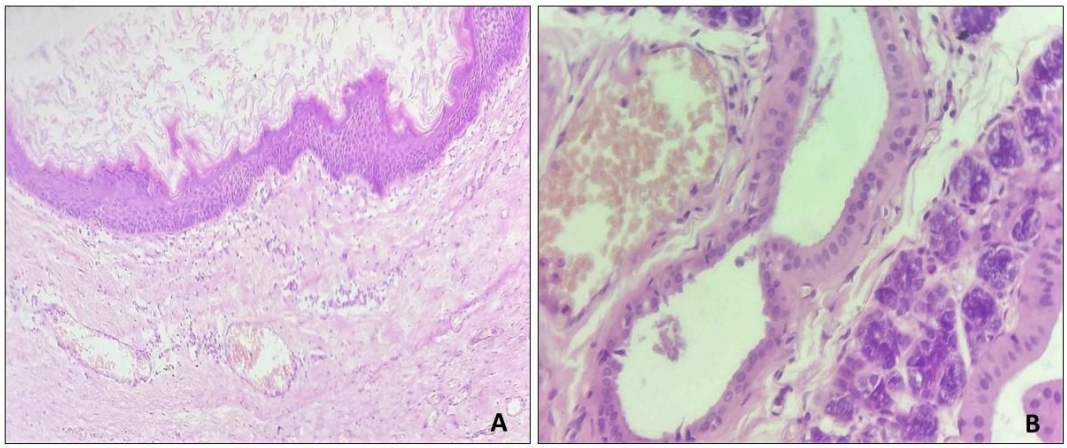

Figure 5A-B: A- A cystic cavity lined by keratinizing stratified squamous epithelium with luminal keratin flakes [H&E, 20X]; B- Attached submandibular gland showing few dilated ducts & clusters of acinar cells [H&E, 40X]

Discussion

Epidermoid cysts are rare benign skin lesions seen all over the body with 7 % in head and neck area.[1-3] They can be categorized as congenital or traumatic. In congenital cases, embryonal aberration of residual ectodermal tissues is responsible for its etiology.[2-3] Traumatic entrapment of surface epithelium is the other etiology.[3-4] They can be seen in any age group but are more common in 15– 45 years with male preponderance.[2-5]

Epidermoid cysts have been described in submandibular region, oral and pharyngeal region but are rare.[5-7] Epidermoid cysts arising in the medial side of submandibular gland is extremely rare.[5-8] In our case, because of repeated aspirations and incision and drainage in rural hospitals, there was fibrosis along with skin puckering and sinus formation.

Because of its extension to sublingual region, the patient can have symptoms of dysphagia, dyspnea and dysphonia due to upward displacement of tongue. Hence, early diagnosis helps in early

Volume 5, Issue 4; July-August 2022; Page No 506-510

intervention and prevent any unwanted complications. On palpation, it gives a consistency of "dough-like" but can be fluctuant and cyst-like based on its luminal contents, which can be cheesy material or any liquified substance.

Differential diagnosis can be submandibular gland tumor like pleomorphic adenoma, ranula, lymphangioma, hemangioma and branchial cyst.[6-8] The diagnosis is difficult using imaging modalities and clinical findings alone and therefore surgical excision with histopathology is important for its diagnosis. On histopathology, the classical features i.e., presence of stratified squamous epithelium with underlying keratin flakes help in making a definitive diagnosis.

Ranula or mucus extravasation cyst represent a pseudocyst cavity containing mucin, abundant epithelioid foamy histiocytes, neutrophils and granulation tissue. Branchial cysts are lined by stratified or ciliated columnar epithelial lining and shows fibrotic wall with lymphoid follicles resembling lymph node or tonsil.

Pleomorphic adenoma would show biphasic elements with presence of epithelial and myoepithelial components. Lymphangioma shows large lymphatic channels in loose connective stroma along with aggregates of lymphoid cells. Hemangioma is a benign tumor which shows variably dilated vascular channels; lobules of capillary sized vascular channels in capillary hemangioma and ectatic channels in cavernous hemangioma. A detailed histological examination helps in delineating the above-mentioned differential diagnosis.

Complete Surgical excision is the mainstay of treatment.[7-10] The extent of excision depends on its adherence to surrounding vital structures. Ideally, excision should be done when cyst is intact and not inflamed to avoid friable tissue ensuring its complete removal. There are very less chances of recurrences in these cysts.

Conclusion

Epidermoid cysts of head and neck are very rare and medially located epidermoid cyst with adherence to submandibular gland is extremely rare. Here we report such a case of a submandibular epidermoid cyst where complete surgical excision was done and follow up after 3 months showed no recurrence.

References:

1. Patel K, Bhuiya T, Chen S, Kenan S, Kahn L. Epidermal inclusion cysts of phalanx: a case report and review of literature. Skeletal Radiol 2006;35(11):861-3.
2. Pancholi A, Raniga S, Vohra PA, Vaidya V. Midline submental epidermoid cyst: A Rare Case. Internet J Otorhinolaryngol. 2006;2.
3. Koca H, Seckin T, Sipahi A, Kaznac A. Epidermoid cyst in the floor of the mouth: Report of a case. Quintessence Int. 2007;38:473-7.
4. Ozan F, Polat HB, Ay S, Goze F. Epidermoid cyst of the buccal mucosa: A case report. J Contemp Dent Pract. 2007;3:90-6.
5. Kandogan T, Koc M, Vardar E, Selek E et al. Sublingual epidermoid cyst: A case report. J Med Case Rep. 2007;1:87.
6. Janarthanam J, Mahadevan S. Epidermoid cyst of submandibular region. J Oral Maxillofac Pathol. 2012;16(3):435-7.
7. Passi D, Singh G, Mehta G, Singhal D. Unsually large submandibular epidermoid cyst: A case report, differential diagnosis and therapy. Contemp Clin Dent. 2014;5(2):252-5.
8. Datta G, Yada A. Swelling in the floor of the mouth: A diagnostic dilemma. Ann Maxillofac Surg. 2020;10(1):258-61.
9. Santos HB, Rolim LS, Barros CC, Caalcante IL, Freitas RD et al. Dermoid & epidermoid cysts of the oral cavity: a 48-year retrospective study with focus on clinical and morphological features and review of main topics. Med Oral Patol Cir Bucal. 2020;25(3):e364-e369.
10. Dammak N, Chokri A, Slim A, Bellalah A, Bouguezzi A et al. Epidermoid cyst of buccal mucosa: Case report and literature review. Clin Case Rep. 2021;9(9):e04853.

Volume 5, Issue 4; July-August 2022; Page No 506-510