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Epidermoid Cyst of Maxilla- Rare and interesting Case Report.

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ABSTRACT:

Epidermoid cyst, a variant of dermoid cyst, is a benign lesion of epithelial origin, in which the cystic cavity is lined by epithelium without skin appendages. The head and neck sites most commonly affected with cutaneous cysts include the scalp (34%), neck (18%), periorbital area (17%), cheeks (including lips-16%), periauricular area (9%), and nasal area (including forehead-6%). Although a common cutaneous presentation in the head and neck region, epidermoid cysts of the maxillary sinus is an extremely rare occurrence. We report a case of epidermoid cyst of the maxillary sinus. The lesion being rare and asymptomatic often presents a diagnostic dilemma for the clinician.

Keywords: Epidermoid cyst, Dermoid cyst, Cholesteatoma

INTRODUCTION:

The occurrence of epidermoid cyst in the paranasal sinuses is rare. The frontal sinus being the most common site of involvement, followed by ethmoid sinus and the least common being maxillary sinus. Epidermoid cysts manifest between the second to the sixth decade with no specific sex predilection. In 1991, Hartman and Stankiewicz reported that they found only 20

cases of cholesteatoma of any paranasal sinus(1).In 1992, Storper and Newman found only 10 such cases(2).

CASE REPORT:

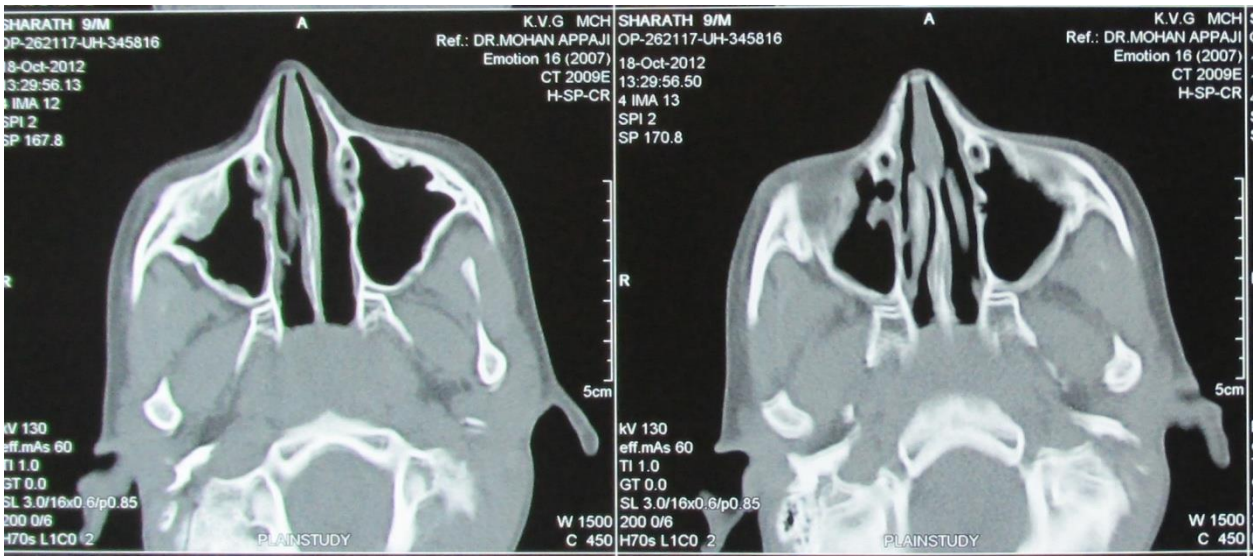
An eight year old apparently healthy boy, presented with history of a vague swelling below right eye and right malar region since five years. The lesion was initially small in size and had progressively grown to the present size. There was no history of discharge from the swelling, nor nasal obstruction or discharge. He did not give complaints of problems related to vision or ocular movements. History of any trauma or surgical intervention to face or head was ruled out.

On examination, a diffuse swelling was present on right malar region, extending to just below right lateral canthal region. On palpation, there were no local signs of acute inflammation, and local cutaneous sensation was normal. The swelling was firm in consistency and non tender. with no discharge. A complete blood count and erythrocyte sedimentation rate were within normal limits. Chest radiograph did not reveal any abnormality. An initial differential diagnosis of chronic inflammatory lesion like foreign body granuloma or osteogenic sarcoma was arrived at, after examining the patient, which was negated by imaging.

FIGURE 1: Epidermoid cyst presenting as swelling on right malar region, extending to just below right lateral canthal region.

Computed tomography (CT scan) showed soft tissue lesion involving the inferior orbital margin with pressure erosion of bony inferior orbital wall with small calcific focus within, suggestive of epidermoid cyst. There was no mass effect on intraorbital muscles and no erosion of maxilla or extensions into pterygo-palatine fossa or masticator space or retroorbital extension.

FIGURE 2: CT scan showing soft tissue lesion involving the inferior orbital margin with pressure erosion of bony inferior orbital wall with small calcific focus within, suggestive of epidermoid cyst.



The lesion was approached via a subciliary approach. The lesion was seen extending deep to zygoma and into maxilla. Pressure erosion of inferior orbital margin was noted. The lesion was completely excised and the irregular infra-orbital margins was rasped. The patient made an uneventful postoperative recovery.

On gross histo-pathological examination the mass was grayish brown and had cheesy material. Microscopic examination showed skin within the underlying cyst. The cyst was lined by squamous epithelium and contained lamellated keratin. Surrounding the cyst was lymphocytic infiltrate with occasional foreign body giant cells. Chronic inflammatory reaction was seen. A diagnosis of epidermal inclusion cyst was made.

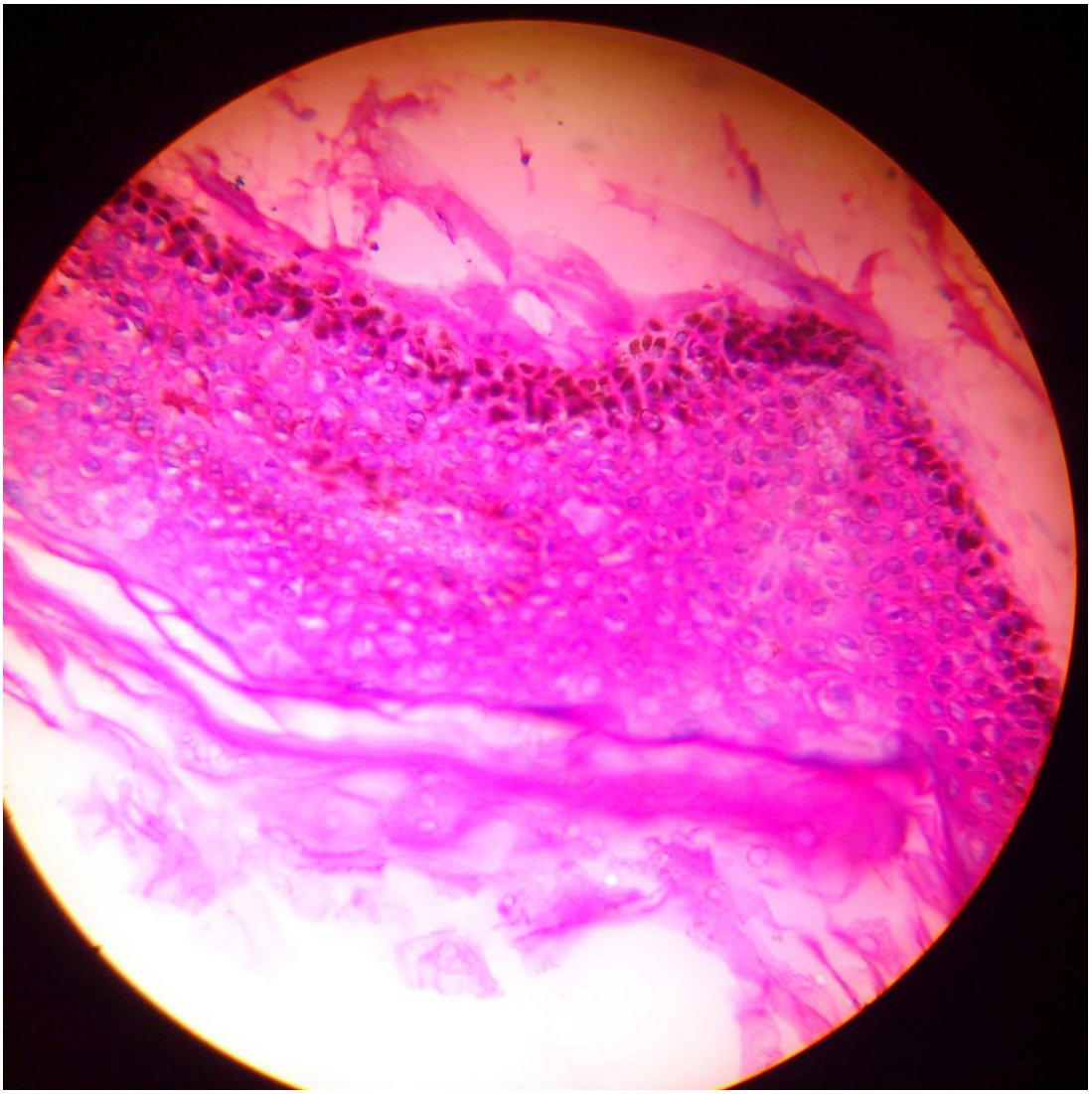


FIGURE 3: Histopathological examination demonstrating the cyst was lined by squamous epithelium and contained lamellated keratin with few lymphocytic infiltrate and occasional foreign body giant cells.

Figure 4: Postoperative image of right malar region (lateral view).

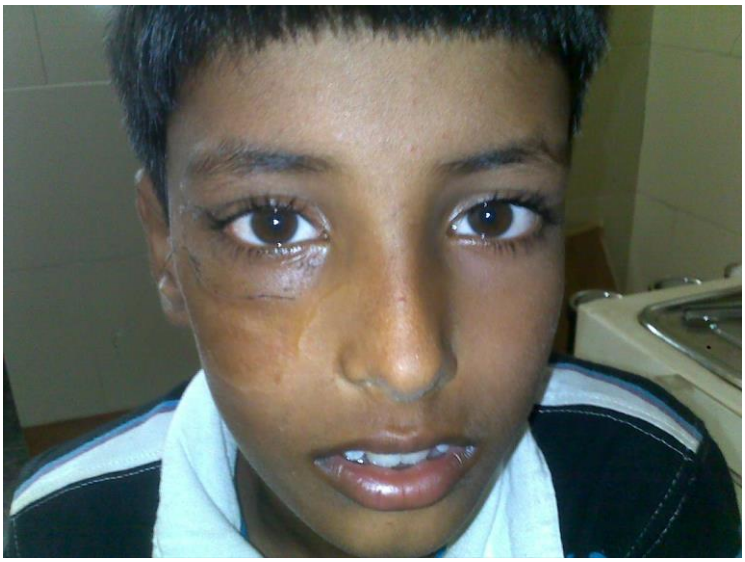


Figure 5: Postoperative image of right malar region (frontal view).

DISCUSSION:

First described by Cruveilhier in 1829 as a “pearly tumor”, epidermoid cysts are painless, slowly enlarging, asymptomatic, expansile tumors and rare lesion. In 1838, Muller coined the term cholesteatoma to describe the presence of cholesterol crystals in a cavity lined with squamous epithelium and filled with layers of dense, squamated keratin. However, in 1854, Virchow noted

that cholesterol is not a necessary component of this entity. In 1928, Critchley and Ferguson proposed that the term 'epidermoid' more suitably conveys the epithelial character of this growth. **(3)**

Histologically dermoid cysts have been classified into three categories namely the Epidermoid cyst – where the cyst wall is devoid of any dermic annexes(**4)**,Dermoid cysts – where dermic annexes such as sebaceous glands, sudoriferous glands or hair follicles are present(**5)**, and Teratomas - which contain structures derived from all three germ layers- ectoderm, endoderm and mesoderm(**6)**.

Epidermoid cyst of the maxillary antrum the respiratory epithelium lining the sinus is partially or totally replaced by a hyperkeratotic squamous epithelium which eventually leads to the formation of epidermoid cyst within the sinus.(7)Etiopathogenesis of epidermoid cysts of the maxillary sinus has been controversial and a matter of debate for years with the entity being deemed either congenital or acquired. Several hypotheses have been advanced to explain the source of squamous epithelium within the sinus. (**8)**

The most prevalent theory, of congenital epithelial rests, was proposed by Remark and Bucy in 1854, which suggests the inclusion of ectodermal tissues during embryogenesis. The respiratory epithelium lining the sinus is partially or totally replaced by a hyper-keratotic squamous epithelium, between 3rd and 5th week of gestation, which eventually leads to the formation of epidermoid cyst within the sinus. The theory of metaplasia, proposed by Wendt in 1873, states that the non-keratinizing squamous epithelium lining the cavity undergoes metaplastic changes as a response to prolonged irritation, as a result of chronic infection. Habermann in 1888, proposed the immigration theory, which suggests the possibility of migration of keratinizing squamous epithelium into an area where it is not found normally. It was in 1928 that Ewing proposed the Implantation theory, which states that these cysts are a result of direct entry of epithelium into a site during trauma.(**9)**

Radiologically an epidermoid cyst appears as a sharply circumscribed bony defect with smooth marginal sclerosis, which closely mimics a maxillary mucocele. On CT, the entity appears as a non-enhancing, expansile, homogenous lesion.On magnetic resonance imaging, such lesions exhibit fairly low signal intensity on Tl-weighted imaging and high intensity on T2-weighted imaging **(10)**.

The differential diagnosis of a maxillary sinus epidermoid cyst includes mucocele, mucus retention cyst, pseudo cyst, and pyocele. (11) Certain neoplastic lesions like papilloma, mucin impaction tumor, schwannoma, chondroma, hemangioma, chordoma, juvenile nasal angiofibroma, and fibrous dysplasia can be considered as differential diagnosis. Malignant lesions include squamous cell carcinoma, esthesioneuroblastoma, salivary gland tumor, sarcoma, and ameloblastoma (10)**.** Histological examination is the main investigation to make a definitive diagnosis.

It is not uncommon that epidermoid cysts of maxillary antrum can elicit complications .The lesion may expand to erode the anterolateral wall of the maxillary antrum to present a facial swelling or may erode the floor of the orbit with clinical presentation of proptosis, restricted

extraocular movements, chemosis etc. (12) The tumor may erode the palate inferiorly to present as a palatal swelling or may involve osteomeatal complex causing nasal obstruction or rhinorrhea. **(3)**

After histopathological diagnosis is established, the choice of treatment is cyst enucleation, making sure that no trace of cystic lining is left behind to avoid recurrence. Although no cases of recurrence have been reported till date, it is very important to have recall visits and periodic follow up.

Conclusion

Epidermoid cyst, is described as a dermal cystic enclosure of keratinizing squamous epithelium that is filled with keratin debris. They are usually secondary to trauma and mainly observed over fingers, palm or soles. The rare occurrence in paranasal sinuses provides a diagnostic dilemma.

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