

An Unusual Case of Epidermal Inclusion Cyst of Maxilla

¹Vaidya Abhishek, ²Sharma Arpit, ³Dabholkar Jyoti, ⁴Raut Abhijit

¹Registrar, Department of Otorhinolaryngology and Head-Neck Surgery, Seth GS Medical College and King Edward Memorial Hospital, Mumbai, Maharashtra, India

²Lecturer, Department of Otorhinolaryngology and Head-Neck Surgery, Seth GS Medical College and King Edward Memorial Hospital, Mumbai, Maharashtra, India

³Professor and Head, Department of Otorhinolaryngology and Head-Neck Surgery, Seth GS Medical College and King Edward Memorial Hospital, Mumbai, Maharashtra, India

⁴Lecturer, Department of Radiology and Head-Neck Surgery, Seth GS Medical College and King Edward Memorial Hospital Mumbai, Maharashtra, India

Correspondence: Vaidya Abhishek, Department of ENT and Head and Neck Surgery, Seth GS Medical College and King Edward Memorial Hospital, Parel, Mumbai-400012, Maharashtra, India, Phone: + 91 9821278013
e-mail: abhishek.d.vaidya@gmail.com

Abstract

Introduction: Epidermal inclusion cyst is a relatively common condition usually associated with trauma, with characteristic presentation, and seen in the extremities. However, it is an uncommon entity in the head and neck region.

Objective: The purpose of this paper is to report the unusual presentation of epidermal inclusion cyst in the maxilla without any definitive history of trauma and to review the concerned English language literature.

Conclusion: In the absence of any definitive history of trauma, only a thorough clinical examination along with characteristic radiological imaging and histopathological findings will help in diagnosing epidermal inclusion cyst.

Keywords: Epidermal inclusion cyst; epidermoid cyst; maxillary cyst.

INTRODUCTION

Epidermal inclusion cysts are uncommon but not rare lesions. They may be congenital due to trapping of ectoderm at time of fusion of neural tube or other epithelial linings. They may also be secondary or acquired due to inclusion of epidermal elements into dermis post-traumatically or iatrogenically.¹ When seen in head and neck or skull region, such lesions are relatively rare, and usually trace back to a history of trauma or surgical procedure. We present a case of an epidermal inclusion cyst in the maxilla and zygoma in an elderly woman without a clear history of trauma or surgical procedure.

CASE REPORT

A 60-year old woman, otherwise healthy, presented to us with a ten month history of a vague swelling below her left eye and on her left malar region. The lesion had grown in size during the few months before presentation. There was

also a fifteen day history of an odorless cheesy discharge from the swelling. There were no problems relatable to vision or ocular movements. Neither did she have any complaints of nasal discharge or obstruction. Patient was a diabetic and hypertensive, well controlled on oral hypoglycemics and antihypertensives. There was no recent or remote history of any trauma or surgical intervention on face or head. The patient had only a vague recollection of a trivial blunt trauma to her face in the childhood.

On examination, a diffuse swelling was present on left malar region extending to just below the lateral canthus. It was firm and non-tender with a small opening which yielded cheesy discharge on expression. There were no local signs of acute inflammation, and the local cutaneous sensations were normal. A complete blood count and erythrocyte sedimentation rate were within normal limits. Chest radiograph did not reveal any abnormality. Having examined the patient our differential diagnosis included bony lesions such as tuberculous osteomyelitis (given the prevalence of



Fig. 1: CT scan (Coronal) showing expansile lytic mass lesion in the left maxilla extending, into zygoma with expansile erosion of the cortex



Fig. 2: T1 weighted MRI scan showing hypo to isointense signal in lesion

tuberculosis in this part of the world) and osteogenic sarcoma or a chronic inflammatory lesion like a foreign body granuloma, which were to be negated by imaging.

Computed tomography (CT) scan showed an expansile lytic mass lesion in the left maxilla extending into the left zygoma causing erosion of the cortex of the superior and lateral walls of the maxilla (Fig. 1). Extension of the mass was also observed in the bony orbit with mass effect of the inferior, lateral and inferior oblique muscles. Posterior wall of the maxilla showed erosion, however no extension in the

pterygopalatine fossa or masticator space was noted. There was no retro-orbital extension of the mass. On post contrast scan the mass showed peripheral enhancement. At magnetic resonance imaging (MRI), the mass was hyperintense on T2 weighted images and iso- to hypointense on T1 weighted images (Figs 2 and 3).

The lesion was approached via a subciliary incision. It was seen extending deep to the zygoma and also into the maxilla. During surgery, the pultaceous content of the mass could be appreciated. The lesion was completely excised along with the skin pore. The patient made an uneventful recovery (Figs 4 and 5).

On gross histopathological examination, the mass was greyish brown and had cheesy content. Microscopic examination showed skin with an underlying cyst. The cyst was lined by squamous epithelium and contained lamellated keratin. Surrounding the cyst was dense lymphocytic infiltrate with occasional foreign body giant cells. Chronic inflammatory reaction was seen, as were fragments of normal lamellar bone. Special stains for mycobacteria and fungi (AFB and GMS) were negative. A diagnosis of epidermal inclusion cyst was made (Figs 6 to 8).

DISCUSSION

Several different terms have been used to describe epidermal cysts. Epidermal inclusion cyst (EIC) refers to those cysts that are the result of the implantation of epidermal elements in the dermis. The more general term epidermoid cyst is



Fig. 3: T2 weighted MRI scan showing hyperintense signal in the lesion



Fig. 4: Postoperative CT scan (bone window) showing clearance of the affected part of maxilla and zygoma



Fig. 5: Postoperative CT scan (soft tissue window) shows clearance of lesion

avored by some. Sebaceous cyst is a misnomer, and should not be used because these cysts are not of sebaceous origin.

The cause of epidermoid cyst may include failure of the surface ectoderm to separate from the underlying structures, sequestration of surface ectoderm or implantation of ectoderm. Epidermoid cysts can thus be either congenital or acquired. The congenital ones develop during the fusion of the embryo when the ectodermal tissue gets trapped in the line of fusion.^{2,3} Such embryological 'accidents' usually take place during 3rd to 5th weeks of gestation. Congenital

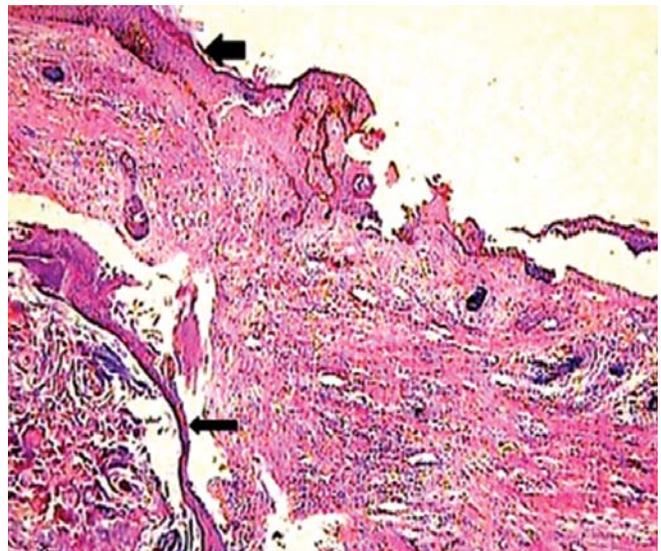


Fig. 6: Photomicrograph of excised specimen (magnification 4x). Thick black arrow points to stratified squamous epithelium of skin, thin black arrow points to epithelium of underlying cyst

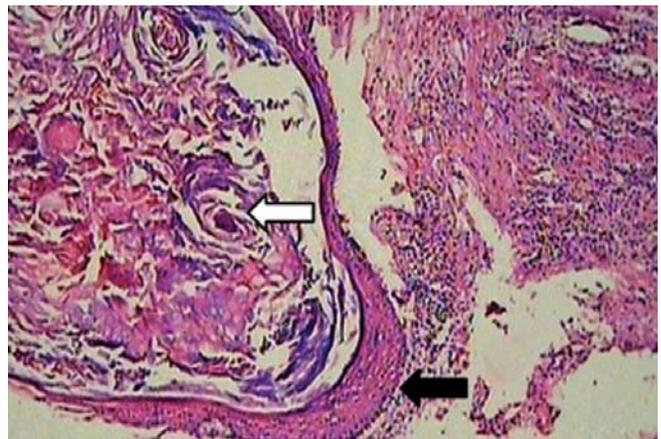


Fig. 7: Photomicrograph (10x, H and E) showing squamous epithelium of cyst (black arrow) and lamellated keratin (white arrow). Also note inflammatory infiltrate in the upper right side of the picture

epidermoid cysts may result from failure of ectoderm to separate from underlying neural tube, or due to abnormal sequestration of surface ectoderm along the embryologic sites of dermal fusion along the eyes, ears, and face.^{4,5} The acquired cysts, which are known as epidermal inclusion cysts, arise from inclusion of epidermal structures in the dermis and other deeper tissues after trauma.^{2,3,6} The terms traumatic epidermoid cysts and EIC describe the same phenomenon, and both indicate a traumatic etiology.⁶

Thus, an epidermal inclusion cyst (EIC) can be described as a dermal cystic enclosure of keratinizing squamous epithelium that is filled with keratin debris.⁷ EICs are usually secondary to trauma, and mainly observed in the fingers, palms, and soles.^{1,2} EICs also occur in other areas and in

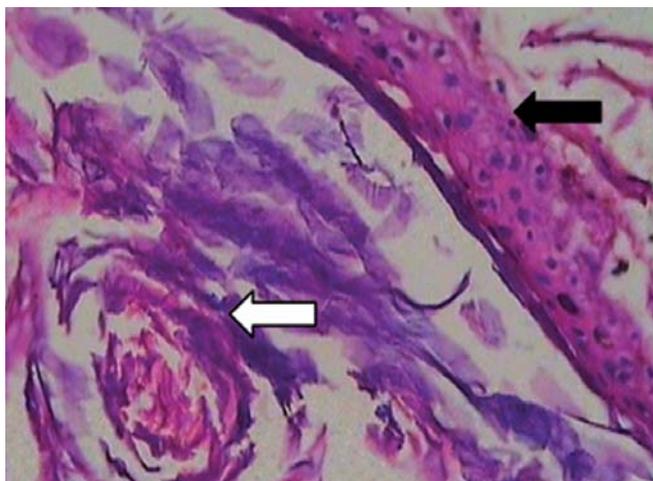


Fig. 8: High magnification (40x, H and E) photomicrograph showing squamous epithelial cyst lining (black arrow) and lamellated keratin (white arrow)

tissues deeper than the dermis which may be secondary to biopsy or surgery.^{3,6}

The most accepted theory of etiopathogenesis is the 'epithelial implant theory', according to which the epidermal elements being driven into deeper tissues is the cause of cysts. The trapped epidermis acts like a skin graft, becomes independent, and continues to grow in its new location producing keratin thus forming a cyst.^{1,2,8} Implantation may be due to a blunt or a penetrating trauma, including lumbar puncture, needle biopsy or surgery.¹

The fingers, toes, palms and soles are commonest sites for EICs. The presumable reason for this is that these sites are very prone to trauma.^{1,2} More likelihood of being traumatized is also probably the reason that men are affected more than women.^{9,10}

While some cases of post-traumatic EICs of the head and neck region and skull bones have been described, to the best of our knowledge, ours is the only case of EIC in maxilla and zygoma without a precedent surgery or clear history of trauma described in English literature. In the absence of a history of trauma or intervention, the cause in our case may be attributed to a remote history of blunt trauma which was elicited only on detailed questioning, and which the patient had only a vague recollection of. A congenital cause in our case was not entertained due to the late age of presentation; the commonest age of presentation of a congenital cyst being first four decades.¹¹

Clinically, these cysts present as painless, slow growing, well circumscribed swellings.² A history of trauma or intervention is usually elicited. The time lag between inciting event and appearance of symptoms may vary from as early

as 6 months to 20 years.^{2,12} Usually EICs are slow growing.¹³ They are usually solitary, but occasionally more than one may be present. The symptoms depend on the location and size of tumor, and the pressure it exerts on the surrounding structures.¹ They are usually small and a size of more than 5 cm may be unusually encountered.^{8,14}

On radiology, the CT attenuation and MRI signal density of epidermoid cyst is quite similar to that of cerebrospinal fluid. Occasionally, the cyst may show slightly negative attenuation values at CT and hyperintense signal at T1-weighted MR imaging.^{11,15} The MRI findings in our case were thus consistent with literature.

On histology, EICs have a squamous epithelial lining, which only rarely shows calcification. They contain debris rich in keratin and lipids formed by desquamation of their squamous epithelial lining.⁶ A granulomatous foreign body giant cell reaction may occur due to rupture of the cyst.^{1,16} The cyst may have connection to skin in form of keratin filled pores. Unlike a dermoid, EICs lack appendages like hair, sweat glands, etc.¹⁷

The treatment of the cysts is complete excision, along with the attached skin pore when present. Incomplete resection may lead to recurrence.¹ Surgery should be deferred during an active infection.

CONCLUSION

Most epithelial inclusion cysts owe their origin to trauma. The precipitating trauma may be trivial and may be evident only on thorough enquiry. We present a case of EIC in maxilla and zygoma which is the only such described case in English literature to the best of our knowledge. The congruity of our histopathological and radiological findings with literature helped us confirm the diagnosis.

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SUMMARY

- Epidermal inclusion cyst is an uncommon clinical entity to be seen in facial bones.
- A history of trauma is usually elicited in the absence of which, a high index of suspicion must be maintained in view of other corroborative findings

- EIC has a characteristic radiological and histopathological findings making the diagnosis easier.
- Complete surgical excision including the skin pore is curative.

REFERENCES

1. Acarturk TO, Stofman GM. Post-traumatic epidermal inclusion cyst of the deep infratemporal fossa. *Ann Plast Surg Jan* 2001;46(1):68-71.
2. Epstein WL. Epithelial cysts in buried human skin. *Arch Dermatol* 1957;76:437-45.
3. Papanayotou PH. Epidermoid implantation cysts of the lower lip: Report of a case. *J Oral Surg* 1977;35:585-86.
4. Bnownstein MH, Helwig EB. Subcutaneous dermoid cysts. *Arch Dermatol* 1973;107:237-39.
5. Pear BL. Epidermoid and dermoid sequestration cysts. *AJR* 1970;110:148-54.
6. Fitzpatrick TB, Johnson RA, Wolff K, et al. *Color atlas and synopsis of clinical dermatology*. 3rd ed. New York: McGraw Hill 1997;162-64.
7. Sullivan DP, Liberatore LA, April MM, Sassoon J, Ward RF. Epidermal inclusion cyst versus thyroglossal duct cyst: Sistrunk or not? *Ann Otol Rhinol Laryngol* 2001;110:340-44.
8. Ulku CH, Uyar Y, Kocaogullar Y, Avunduk MC. Iatrogenic epidermal inclusion cyst of the parapharyngeal space: Unusual complication of ear surgery. *Skull Base*. Feb 2004;14(1):47-51;discussion 51.
9. Kotcamp WW. Epidermoid cyst: A case report. *J Bone Joint Surg Am* 1962;44A:377-79.
10. Carroll RE. Epidermal (epithelial) cysts of the hand. *Am J Surg* 1953;85:327-34.
11. Smirniotopoulos JG, Chiechi MV. Teratomas, dermoids, and epidermoids of the head and neck. *Radiographics* 1995;15:1437-55.
12. Fanti PA. Subungual epidermoid inclusions: report of 8 cases. *Dermatologica* 1989;178:209-12.
13. Thompson AC. Iatrogenic epidermoid cyst of the parotid region following ear surgery. *J Laryngol Otol* 1991;105:227-28.
14. Lever WF, Shaumburgh-Lever G. *Histopathology of the skin*. 5th ed. Philadelphia: JB Lippincott; 1975:1540.
15. Lee VS, Provenzale JM, Fuchs HE, Osumi A, McLendon RE. Post-traumatic epidermoid cyst: CT appearance. *J Comput Assist. Tomogr* Jan-Feb 1995;19(1):153-55.
16. McCarthy J, ed. *Plastic surgery*. Philadelphia: WB Saunders 1990;3569-72.
17. Cotran RS, Kumar V, Robbins SL. *Pathologic basis of disease*. 5th ed. Philadelphia: WB Saunders 1994;1182-83.