

ATYPICAL CASE OF AN INFECTED EPIDERMOID CYST OF PAROTID GLAND: A CASE REPORT

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<p>Article Info Received 15/02/2015 Revised 27/03/2015 Accepted 12/04/2015</p> <p>Key words: Epidermoid cyst; Parotid swelling.</p>	<p>ABSTRACT Epidermoid cysts present as a nodular and fluctuant subcutaneous lesion beneath the skin and are most common in acne-prone areas of the head, neck and back. The cyst develops out of ectodermal tissue. however, primary epidermal cysts of salivary gland appear to be very rare, there are only very few case reports of epidermal cyst in parotid gland that have been published in world literature. We are presenting a case of a 22 year old female who presented with a swelling in right parotid region with multiple discharging sinuses for last 17 years. Patient received first and second line antitubercular treatment elsewhere on account of a highly misleading clinical picture. A diagnosis of infected epidermoid cyst was made on frozen section intraoperatively. Excision of the cyst and the involved skin along with a partial parotidectomy was performed and histopathology confirmed the above diagnosis. The cosmetic defect was repaired with the advice of the Plastic Surgeon.</p>
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INTRODUCTION

Epidermoid cysts are rare, slow-growing, benign, and 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%. Epidermoid cysts can occur at any age from birth to 72 years; they usually become apparent in patients between 15 and 35 years [1]. Primary epidermoid cyst of the salivary gland is extremely rare and has only been reported once in the parotid gland [2]. It could be easily mistaken for a salivary gland abscess, neoplasm and other cysts. Therefore, an excisional biopsy is necessary for a prompt diagnosis and confirmation [3].

Case History

A 22 year old female presented with a swelling in right parotid region since 17 years. Swelling was painful with multiple discharging sinuses over its surface. Past

history revealed that she was treated in many local and 7 tertiary care hospitals over the last 17 years. The right parotid swelling was first noticed at the age of 5. Along with the swelling, the patient also had pulmonary tuberculosis. The parotid swelling hence was considered to be tubercular in origin and treated with 9 months of anti-tubercular drugs. Pulmonary tuberculosis was completely cured but the parotid swelling persisted. Each time the swelling increased in size antibiotics were taken from local hospitals. In 2008 CT neck at the first tertiary care centre revealed a parotid abscess and the patient was treated with antibiotics. On further investigation, Mantoux was positive, culture revealed no growth. Patient was treated with 24 months of second line antitubercular drugs at a second tertiary hospital. Tests for AFB and TB culture were repeated in 2010 in another tertiary care hospital which was negative, doctors at this hospital suspected Actinomycosis as the diagnosis. Biopsy taken at this hospital revealed a chronic abscess. The patient visited yet another tertiary care hospital in June 2011, incision and drainage done here revealed epidermal inclusion cyst as



diagnosis. Patient took the opinion of senior doctors at the fifth tertiary care hospital in 2011 who repeated the Biopsy. The result of biopsy was inconclusive and TB culture was negative. At the sixth tertiary care centre in 2013, MRI revealed parotid abscess which was treated with antibiotics. At the seventh tertiary care hospital, treatment was continued with antibiotics. Our hospital was eighth tertiary care hospital that the patient visited. On examination there was a localised swelling in right infraauricular region about 4x3 cm in size. The skin over swelling was inflamed, adherent with multiple discharging sinuses. (Fig 1)

On palpation swelling was tender, firm in consistency. On deep palpation whitish material was expressed from sinus openings. (Fig 2)

This material from swelling was sent for cytology and culture and sensitivity which revealed methicillin sensitive staphylococcus aureus. Hence a diagnosis of botryomycosis was kept in mind. Cytology revealed chronic inflammatory cells. On the basis of culture report, patient was started on linezolid and vancomycin injection. Swelling decreased in size but was still persistent.

MRI parotid gland revealed multi lobulated cystic lesion in right parotid space. It showed intermediate signal on T1 and hyperintense signal on T2 weighted images. (Fig 3a-b). There was mild peripheral post contrast enhancement. The lesion is predominantly infra auricular

with multiple septations within superficial lobe of parotid gland. A provisional diagnosis of chronic granulomatous disease of parotid gland was kept in mind. Our differential diagnosis included actinomycosis, botryomycosis and tuberculosis.

Our plan of action included

1. Excision of infected skin, subcutaneous tissue, superficial lobe of parotid and level 1 and 2 lymph nodes with frozen section.
2. Plastic surgeon opinion for cosmetic correction of defect.

We modified the routine parotidectomy incision as in the picture to enable excision of involved skin. (Fig 4)

Frozen section revealed infected epidermoid cyst. Based on this report, complete cyst excision and involved part of superficial lobe of parotid gland was excised using operating microscope and facial stimulator.(Fig 5-8)

Level II lymph node sampling revealed chronic inflammation. Suturing was done with the help of a local advancement flap with suggestion of plastic surgeon. (Fig 9)

Postoperatively the healing was uneventful. (Fig 10)

After surgery, repeat MRI revealed that the previously seen lesion was no longer visualized. (Fig 11).

Final histopathology report confirmed diagnosis of epidermoid cyst.

Figure 1. Infra auricular swelling with multiple discharging sinuses



Figure 2. Whitish material expressed from the sinus openings on deep palpation



Figure 3a. T1 lesion shows intermediate signal : b - T2 lesion shows hyperintense signal

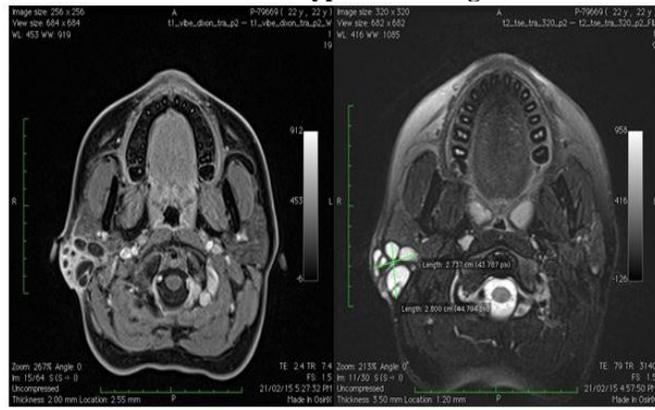


Figure 4. Illustrating the skin incision planned



Figure 5. Involved skin excised



Figure 6. Whitish material in the cyst



Figure 7. Cyst wall



Figure 8. Illustrating the use of facial stimulator



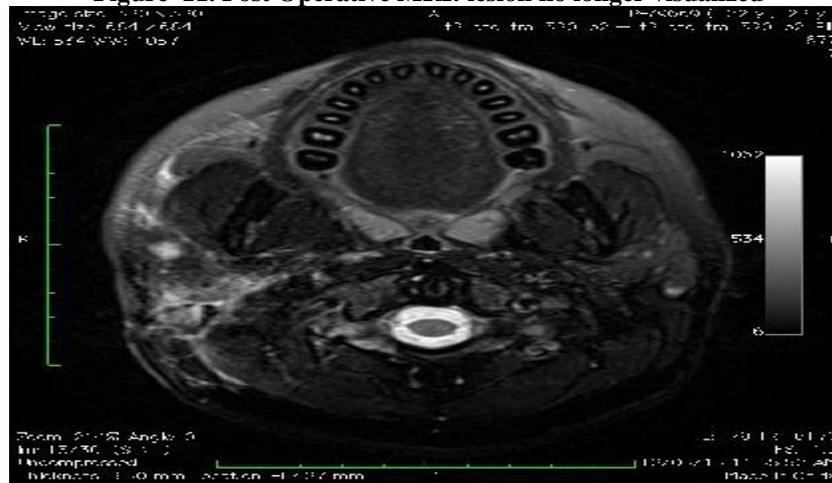
Figure 9. Suturing with help of local advancement flap



Figure 11. 1 month post operative



Figure 11. Post Operative MRI: lesion no longer visualized



DISCUSSION

Epidermoid cysts are relatively less common in the head–neck region, hence are liable to be misdiagnosed [4]. Epidermoid cysts of parotid gland are of congenital origin. Although these may exist since birth, they are slow growing and generally not identified until adulthood. The exact histogenesis of salivary epidermal cyst is unknown, but it may have arisen from developmental branchial pouch analogue epithelium which can occur in salivary gland or could be due to obstruction in salivary duct within the substance of gland leading to epithelial lining cavity filled with viscous semisolid epithelial degradation products [5]. If the cyst stays for longer time, it may get infected forming sinuses or fistulas [6].

Histologically, epidermal cysts have a lining of stratified squamous epithelium and are usually filled with cheesy material or keratin [6,7]. Pathologically they are considered to be identical to middle ear congenital cholesteatomas [8].

It can be diagnosed by ultrasound and FNAC usually [9] but diagnosis may be challenging in infected cysts and may require CT scan and further confirmation with excisional biopsy.

Cysts in parotid gland may be diagnosed with relative ease but infection complicates the diagnosis. Our emphasis in this case is how infection in a benign epidermoid cyst can be misleading. Chronic non healing

lesions are often attributed to tuberculosis in our country. Features favouring diagnosis of tuberculosis in this case included concurrent presence of parotid swelling and pulmonary tuberculosis in childhood. Mantoux positive, lesion not responding to antibiotics based on culture report, clinical picture of non-healing multiple discharging sinuses. When the patient failed to respond to first line of tuberculosis treatment, multi drug resistant tuberculosis was kept in mind and 24 months of second line drugs was tried. Failure to respond to this treatment necessitated the need to keep a differential diagnosis in mind. Decision to post the patient for excisional biopsy and frozen section was made as all the other investigations were inconclusive. Diagnosis of epidermoid cyst was concluded on basis of histopathology report. Hence we emphasize the role of excisional biopsy in our case.

CONCLUSION

Epidermoid cysts of parotid gland are rare lesions with only few cases mentioned in literature. Infection in these cysts increases dilemma in the diagnosis as in our case. Hence, an excisional biopsy must be planned to confirm the diagnosis.

Conflict of Interest

None Declared.

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