

CASE REPORT

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A rare case of unilateral tonsillar epidermoid cyst

Gaurang Singhal, Pallika Kumar^{*} , Anjali Pathak and Poonam Rani

Abstract

Background: Epidermoid cyst is a developmental lesion with 1.6–6.9% cases found in the head and neck region. Intraoral epidermoid cyst is a rare finding and intratonsillar epidermoid cyst is even rarer, with less than 0.01% incidence.

Case presentation: We are reporting a case of 16 year old with complain of unilateral tonsillar enlargement since 5 months. She underwent tonsillectomy under general anesthesia, and the specimen was sent for histopathological examination. It was reported as multiple intratonsillar epidermoid cysts on final histopathological examination.

Conclusion: We would like to report such a case due to its rarity and to recommend the need for histopathological diagnosis after every case of tonsillectomy in order to differentiate various benign and malignant lesions for their proper management.

Keywords: Tonsil, Epidermoid cyst, Benign

Background

Tonsil is a lymphoid tissue covered situated in the lateral pharyngeal wall [1]. It plays an indispensable role in the immune function of the body [2]. There are various benign as well as malignant lesions described in the tonsil. Epidermoid cyst is one of such benign lesions which forms from the development of abnormal epithelial components of ectodermal tissue during the fetal life or from implantation of epithelium during trauma or surgery [3]. It was first described by Roser in 1850. They can be congenital or acquired. Around 1.6–6.9% of epidermoid cyst present in the head and neck region [4]. Out of which the most common intraoral sites include sublingual, submental, submandibular, lingual/labial, and buccal mucosa [5]. Intratonsillar epidermoid cyst is a rare manifestation with an incidence of less than 0.01% [6]. Among these patients, multiple epidermoid cysts are even more rare. They usually present as slowly growing painless mass [7].

Case presentation

A 16-year-old female patient presented in the ENT outpatient department with complaints of difficulty in swallowing for the last 5 months which has increased in the last 15 days, and it was associated with pain in swallowing. On local examination, there was a left tonsillar mass that was crossing the midline whereas the right tonsil appeared normal. There was no cervical lymphadenopathy. Preoperative contrast-enhanced computed tomography of the neck was done which showed enhancing soft tissue growth in the left tonsillar fossa measuring approximately 2.3×2.4 cm in size causing significant obliteration of the oropharyngeal lumen with areas of necrosis within (Figs. 1 and 2). The patient was diagnosed with a left tonsillar fossa mass (Fig. 3) and was then posted for left tonsillectomy under general anesthesia after getting routine investigation and preanesthetic workup done. Unilateral tonsillectomy was done (Fig. 4) and the mass was sent for histopathological examination (Fig. 5). The final histopathology report (Fig. 6) showed multiple sections with dilated crypts which are filled with keratinized debris, acute and chronic inflammatory cells, and foamy histiocytes. Few cystic structures are present lined by

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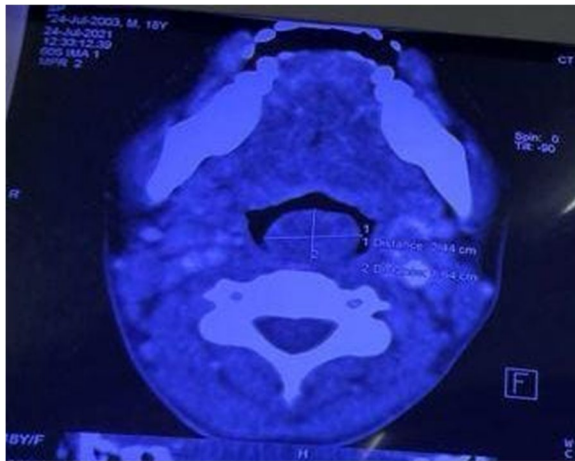


Fig. 1 CECT neck (Coronal cut) showing mass arising from left tonsillar fossa with necrosis

stratified squamous epithelium. The findings were suggestive of acute chronic tonsillitis with multiple epidermoid cysts. The postoperative period was uneventful and there are no signs of recurrence to date.

Discussion

Children are most commonly affected by chronic tonsillitis. Chronic inflammation can be present in both chronic tonsillitis and tonsillar hypertrophy [1]. Unilateral tonsillar hypertrophy is most commonly seen in peritonsillar abscess, tonsillar cyst, tonsillolith, parapharyngeal space tumors, malignancy of tonsil, and tonsillar artery aneurysm [8].

Epidermoid cyst is lined by squamous epithelium only and it differs from dermoid cyst as it does not contain skin and adnexal structures [7].

According to literature, epidermoid cyst can be congenital or acquired. Congenital epidermoid cysts are found where the embryonic remnants fuse while acquired usually forms secondary to trauma or surgery. Remark and Bucy in 1854 proposed that the inclusion of ectodermal tissue during embryogenesis is the cause of the development of an epidermoid cyst. Later on, Wendt in 1873 proposed metaplastic theory according to which chronic infection causes metaplastic changes in the non keratinized stratified squamous epithelium lining the tonsil. Lastly, in 1920, Ewing proposed implantation theory, according to which the epithelial tissue is directly implanted during the trauma [9].

Mean age of presentation is 10–35 years with female preponderance having male to female ratio of 1:4 [7, 10]. In our case, the age of presentation was 16 years in a female patient. Epidermoid cysts are found to be associated with Gardner syndrome (APC gene mutation) or

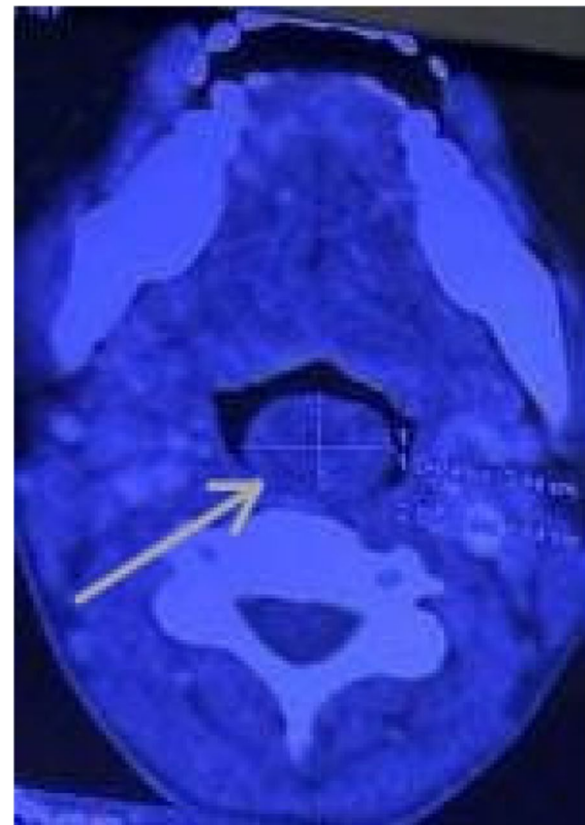


Fig. 2 CECT neck (Axial cut) showing mass arising from left tonsillar fossa with necrosis



Fig. 3 Intraoperative image



Fig. 4 Postoperative image

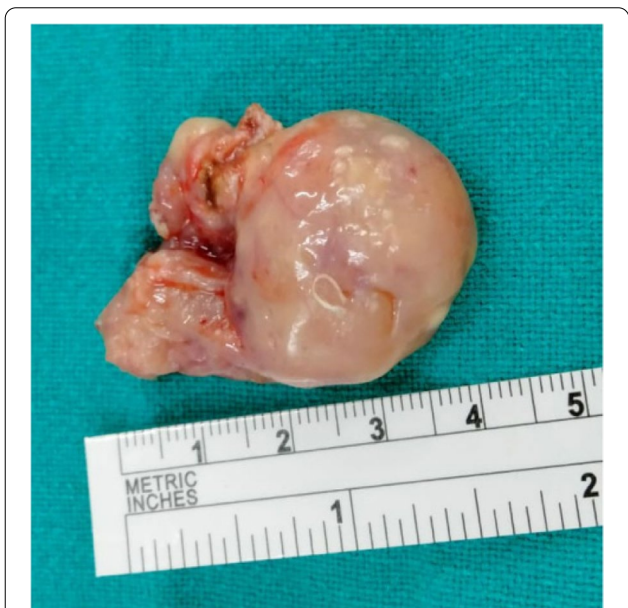


Fig. 5 Tonsillar mass

hereditary syndromes similar to Lowe syndrome (X chromosomal OCLR-1 gene mutation) [11, 12].

The patient usually presents with painless slowly growing mass however in our case patient presented with pain and the mass increased in size in last 15 days [7]. The

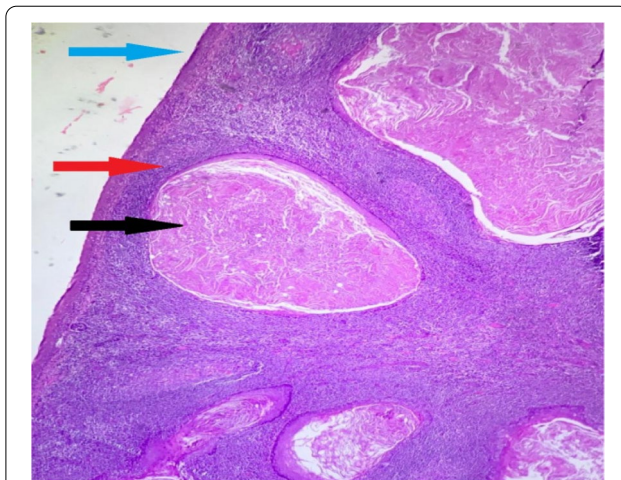


Fig. 6 Tonsillar epidermoid cyst on HPE. Black arrow—dilated cysts filled with keratinized debris, inflammatory cells. Red arrow—cyst lined by keratinized stratified squamous epithelium. Blue arrow—palatine tonsil lined by non-keratinized stratified squamous epithelium

reason could be the acute inflammation on chronic tonsillitis as seen in histopathology report. Out 7% cases of epidermoid cyst found in head and neck less than 0.01% are seen in tonsil. In our case report too, the cyst was found in the tonsil which is very rare and only few such cases have been described in the past.

Treatment of such lesions is surgical excision without opening of the cyst as the contents may cause an irritating effect on the surrounding tissue [13]. Diagnostic unilateral tonsillectomy is done since it carries a potential risk of malignancy. Similarly, we did the left tonsillectomy of the patient without opening the tonsillar contents, and the sample was sent for histopathological examination in toto as our patient also presented with unilateral left-sided tonsillar mass. Generally, single epidermoid cysts are seen however few reported multiple cysts; we also found multiple epidermoid cysts in our case which is even more rarer. Recurrence after surgery is rare in such cases. A similar finding has been seen in our case, as after 2 months of follow-up, there are no symptoms and signs of recurrence [14].

It has been reported that rarely squamous cell carcinoma can develop from epidermoid cysts however no such case has been reported in the past in cases of intratonsillar epidermoid cysts, similar to our case [15].

Conclusion

The aim of this paper is to report such a rare entity of multiple epidermoid cysts in the tonsil and the need for the histopathological diagnosis after every case of tonsillectomy to differentiate various benign and malignant lesions followed by their proper management.

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Authors' contributions

GS wrote the case report. AP edited it. PK performed the surgery. PR edited photographs. The authors read and approved the final manuscript.

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Not applicable.

Consent for publication

Written informed consent has been obtained from the parent of the patient.

Competing interests

The authors declare that they have no competing interests.

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