Palatin Tonsilin Epidermoid Kisti: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

Epidermoid Cyst of the Palatine Tonsil: A Case Report and Literature Review

Ayşe Nur UĞUR KILINÇ

Konya Eğitim ve Araştırma Hastanesi Patoloji AD, Konya, Türkiye

Geliş tarihi: 28.05.2020 Kabul tarihi: 15.06.2020 DOI: 10.17517/ksutfd.728417

Özet

Epidermoid kistler gelişimsel lezyonlardır ve baş ve boyun bölgesinde % 2 insidans oranı ile vücudun farklı organlarında görülebilir. Epidermoid kistler dilaltı, submental, submandibular ve bukkal mukozada ve nadiren baş ve boyun bölgesinde bademciklerde bulunur. Konjenital olarak fetal dönemde ortaya çıkarlar veya ameliyattan sonra travma veya implant nedeniyle edinilmiş bir lezyon olarak ortaya çıkarlar. Bu olgu sunumunda, kronik bademcik iltihabı olan 6 yaşındaki bir çocukta, bademciklerde gelişen nadir rastlantısal bir bulgu olan epidermoid kist tartışıldı. Sonuç olarak, bademcik genişlemesine, bademciklerde kistik gelişmeler de neden olabilir. Özellikle bademciklerin asimetrik genişlemesinde malignitenin ayırıcı tanısında kistik lezyonlar da klinikopatolojik olarak akılda tutulmalıdır.Tüm tonsillektomi materyalleri, tonsillit ve reaktif lenfoid hiperplaziye ek olarak bulunabilecek ek patolojiler açısından makroskopik ve histopatolojik olarak ayrıntılı bir şekilde incelenmelidir.

Anahtar Kelimeler: Tonsil, İnsidental bulgu, Epidermoid kist

Abstract

Epidermoid cysts are developmental lesions and may be seen in the head and neck area with an incidence rate of 2% as well as in different organs of the body. Epidermoid cysts are located in sublingual, submental, submandibular and buccal mucosa and rarely at tonsils in the head and neck area. They congenitally occur in fetal period or occur as an acquired lesion due to a trauma or implant after surgery. In this case report, we will discuss the epidermoid cyst, a rare incidental finding developing at tonsils, in a 6-year-old boy with chronic tonsillitis with literature review. In conclusion, tonsil hyperplasia may also be caused by cystic developments at the tonsils. It should clinically be kept in mind especially in differential diagnosis of malignancy in the asymmetry of tonsils. All tonsillectomy materials should be detailed examined gross and histopathologically for additional pathologies that may be found in addition to tonsillitis and reactive lymphoid hyperplasia.

Keywords: Palatine tonsil, Incidental finding, Epidermoid cyst

Yazışma Adresi: Ayşe Nur UĞUR KILINÇ, Konya Eğitim ve Araştırma Hastanesi Patoloji AD, Konya, Türkiye Tel: +90 (332) 221-0000/2154 Mail: aysenurugur@hotmail.com ORCID No: 0000-0002-0439-0101

INTODUCTION

Epidermoid cyst was defined by Roser in 1850 (1). Epidermoid cysts are lesions that are commonly found in several areas of the body and 1.6-6.9% of the epidermoid cysts occur in the head and neck area of the patients. Less than 0.01% of the epidermoid cysts in the head and neck areas develop in oral cavity (1, 2). They are located in sublingual, submental, submandibular, labial/lingual or buccal mucosa and rarely at palatine tonsils (2).They often appear as a painless and slowly growing mass without any symptoms.They may congenitally occur in fetal period or occur as an acquired lesion due to a trauma or implant after surgery (3).

In our report, we will evaluate epidermoid cyst, a rare incidental finding developing at tonsils,together with our

6-year-old boy child case, 12 epidermoid cyst of the palatine tonsil cases in the literature to date(1,3-13) .

CASE PRESENTATION

A 6-year-old boy with no previous disease or history of a surgery was admitted to our hospital with the complaints of snoring, sleeping with his mouth open and frequently having throat infection. In clinical evaluation, tonsillectomy and adenoidectomy were planned for the patient who had grade 2-3 tonsillar hypertrophy and adenoid vegetation. In whole blood and biochemical examinations, hemoglobin value was 14 g/dL and RBC was 5.26 10⁶/mm³ and no abnormality was found in other parameters. Material was sent to pathology laboratory for histopathologic examination. A cystic lesion of 2 cm was macroscopically recorded in tonsillar sections.

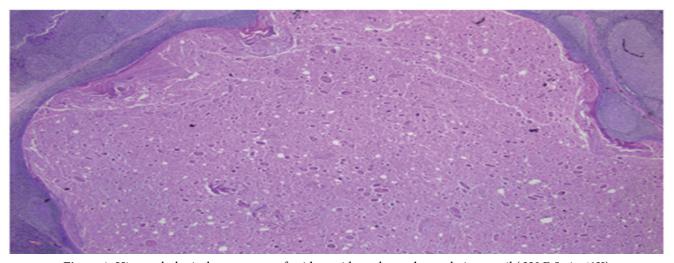


Figure 1. Histopathological appearance of epidermoid cyst located on palatine tonsil (H&E Stain 40X)

In microscopic examination, the inner surface of the cyst was lined with squamous epithelium and its lumen was filled with keratin material. No other cutaneous histological structures were observed in the cyst wall (Figure 1). Histopathological diagnosis was chronic tonsillitis with epidermoid cyst.

DISCUSSION

In the tonsils; several cystic lesions can develop, including tonsillar retention cyst, lymphoepithelial cyst, hydatid cyst, and the most common epidermoid cyst. Epidermoid cysts are cystic lesions histologically lined by maturing squamous epithelium. They do not have cutaneous adnexal and fibrous elements on their wall (6).

In the literature review, we found 12 cases until today **(Table 1)**. Increased frequency of tonsillar cysts reported in the literature in recent years may be associated with tonsillectomy materials undergoing routine histopathological examination.

Table 1. Comparison of data of epidermoid cyst of the palatine tonsil in literature and present case					
Case	Age/Sex	Localization	Size	Focality	Author (Reference)
1	42/F	Right	?	Unifocal	Erol K (2013)(3)
2	11/?	<u>;</u>	;	Unifocal	Swarnagowri BN (2014)(9)
3	17/M	<u>;</u>	;	Unifocal	Gulia PS (2015)(1)
4	40/F	Right	0,4-0,6cm	Multiple	Nikumbh B (2017)(8)
5	20/M	Right	2cm	Unifocal	Hsu-A (2017)(6)
6	47/F	Left	2 cm	Unifocal	Jain A (2017)(13)
7	59/F	Left	;	Unifocal	Gonzaga VLF (2018)(4)
8	28/F	Right	;	Unifocal	Boutaay R (2019)(7)
19	56/F	Right	;	Multiple	Mishra U (2019)(12)
10	32/F	Left	1,5cm	Multiple	Mathew AS (2019)(11)
11	8 /M	Right	;	Unifocal	Altındal A. (2019)(5)
12	6/M	Left	2 cm	Unifocal	Kılınc AU (2020)(Present)

Although epidermoid cysts have the same clinical and histological appearance, they may be classified as congenital and acquired (1, 2). While congenital cysts develop during the merger of embryonic tissues, acquired cysts are considered to be developing secondary to trauma and surgery. About the origin of the cyst, Remark and Bucy suggested the idea of the inclusion of ectodermal tissues during embryogenesis. In 1873, Wendt defended the idea of metaplastic transformation of non-keratinized squamous epithelial cells lining the cavity due to chronic infection secondary to prolonged irritation (14). Finally, Ewing suggested the idea of direct entry of these cysts during trauma as the implementation theory in 1928 (15).

They can be seen at any age from birth to the age of 72. The ratio of male to female in literature is 1:4 (3, 4, 5). In our literature review, the ratio of male to female was 4:7 and most of the patients were reported at adult ages (14, 16, 17).

Epidermoid cysts of the tonsils may generally be single, however, multiple cysts were monitored in especially 3 patients at adult ages. The diameter of the lesion in the patients was not generally reported in literature. In 3 patients with unifocal lesion with lesion diameters, the diameters were reported as 2 cm interestingly. In 2 patients with multifocal lesions with lesion diameters, the lesions had variable diameters and the larger diameter in 1 patient was reported as 1.5 cm and the larger parameter in the other patient as 0.8 cm. The lesion was located at the right tonsil in 6 out of 10 patients with reported localization and located at the left tonsil in 4 patients. Lesions in 2 patients under the age of 10 were located at the left tonsil and the patients were male. This suggests that congenital epidermoid cysts of tonsil may be associated with the left side and male gender. Eight of 9 patients with acquired epidermoid cyst seen at adult ages were female, which suggests that acquired epidermoid cysts may be associated with female gender.

As epidermoid cysts of the tonsil lead to macroscopic asymmetry they may clinically mimic malignancy (4,8).

Histopathologically epidermoid cysts can easily be diagnosed. In histopathological differential diagnosis, there are dermoid cysts containing adnexal elements on the wall, lymphoepithelial cysts containing large lymphoid tissue on the wall, and squamous cell carcinoma with dysplasia and invasion in the epithelium.

It is considered in some studies that epidermoid cysts are associated with Gardner syndrome caused by the adenomatous polyposis coli gene mutation or hereditary syndromes similar to Lowe syndrome which is an X-chromosomal oculo-cerebral- renal disorder caused by OCLR1-gene mutation (18,19). These associations should clinically be kept in mind in these patients. In the report of 12 cases in the literature, including our case, no morbidities and genetic disorders were reported.

Actinomyces we relatively commonly see in patients with chronic tonsillitis has been reported to accompany to the lesion in literature.

It is known that rarely squamous cell carcinoma can develop from epidermoid cysts (20). However, in these 12 cases in the literature, dysplasia and malignancy were not observed in the epithelium.

In conclusion, asymmetrical tonsillar enlargement may also be caused by cystic developments at the tonsils. Children and especially adults it should clinically be kept in mind in differential diagnosis of malignancy in the asymmetry of tonsils.All tonsillectomy materials should be detailed examined gross and histopathologically for additional pathologies that may be found in addition to tonsillitis and reactive lymphoid hyperplasia.

Funding: None

Conflict of Interest: None

Parental permission was obtained for this case.

REFERENCES

- Gulia SP, Lavanya M, Kamidi V, Kumar A. Epidermoidc yst of the tonsil: an incidental finding. Case Reports. 2015;2(12):777– 779.
- Calderon S, Kaplan IJJoo, surgery m. Concomitant sublingual and submental epidermoid cysts: a case report. J Oral Maxillofac Surg. 1993; 51 (7):790-792.
- 3. Erol K, Erkan KM, Tolga D, Bengu C. Epidermoid cyst localized in the palatine tonsil. J Oral Maxillofac Pathol. 2013;17(1):148.
- Fernandes V, Khandolkar P, Pillai V, Sukhthankar I, Chari A. Squamous Inclusion Cyst in the Palatine Tonsil Mimicking a Tumor. Int J Otolaryngology and Head & Neck Surgery. 2018;7:249–253.
- Altindal AS, Unal N. A Rare Case Report of Epidermoid Cyst in Palatine Tonsil. Turkiye Klinikleri J Case Rep. 2019;27(1):34–7.
- Hsu T, Yang C, Huang Y, Wang C, Shih Y, Wang C. Cystic Tonsillar Tumor: Epidermoid Cyst at Palatine Tonsil. E-Da Medical Journal. 2017;4(3):28–31.
- 7. Rachida B, Sondos J, Nouha A, Jamel KJTPAMJ. Epidermoid cyst of the tonsil: a rare finding. 2019;34.
- Nikumbh Dhiraj B, Nikumbh Roopali D, More Hemant. Intratonsillar multiple epidermal inclusion cysts-A tumor mimic. Archives of Cytology and Histopathology Research. 2017;2(1):18– 20.
- 9. Swarnagowri B, Suba G, Prabhakaran NJSJoMCR. Epidermal inclusion cyst in palatine tonsil: a case report. 2014;2:83-4.
- Dutta M, Saha J, Biswas G, Chattopadhyay S, Sen I, Sinha R. Epidermoid Cysts in Head and Neck: Our Experiences, with Review of Literature. Indian J Otolaryngol Head Neck Surg. 2013;65(1):14–21.
- 11. Mathew AS, Gowda BC, Sridhar A. Multiple Epidermoid Cysts in the Palatine Tonsil Causing Referred Otalgia: A Case Report.
- Takasaki K, Kitaoka K, Kaieda S, Hayashi T, Abe K, Takahashi H. A case of actinomycosis causing unilateral tonsillar hypertrophy. Acta Otolaryngol. 2006 Sep;126(9):1001-4.
- Jain A, Saeed N, Maheshwari V, Kamal M. Epidermoid Cyst of The Tonsil: a rare finding. Int Arch BioMed Clin Res. 2017;3(1):14–15.
- Fernández JLV, Rojas JL, Fernández JA, Quevedo MSJAO. Dermoid cyst of the floor of the mouth. Acta Otorrinolaringol Esp. 2007;1(58):31-3.
- Rozario JP, Appaji M, Abhilash AM. Epidermoid Cyst of Maxilla- Rare and interesting Case Report. Otolaryngology online journal. 2013;3(4):163–170.
- Shivakumar M, Yogesh T, Nagaraj T, Sinha PJIJoM, Reports DC. Epidermal inclusion cyst of buccal mucosa: A rare case report. 2015:1-3.
- Lima SM, Chrcanovic BR, de Paula A, Freire-Maia B, Souza LN-JTSWJ. Dermoid cyst of the floor of the mouth.ScientificWorld Journal. 2003;3:156-62.
- Koh K-J, Park H-N, Kim K-AJIsid. Gardner syndrome associated with multiple osteomas, intestinal polyposis, and epidermoid cysts. Imaging Sci Dent. 2016;46(4):267-72.
- Ikehara S, Utani AJTJod. Multiple protrusive epidermal cysts on the scalp of a Lowe syndrome patient. J Dermatol. 2017;44(1):105-7.

 Faltaous AA, Leigh EC, Ray P, Wolbert TT. A Rare Transformation of Epidermoid Cyst into Squamous Cell Carcinoma: A Case Report with Literature Review. Am J Case Rep. 2019;3;20:1141-1143.

KSU Medical Journal 2020;15(3) : 111-114