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OLGU SUNUMU / CASE REPORT

A Rare Localisation For Cutaneous Ciliated Cyst On Nasal Sulcus: A Case Report

Nazal Sulkusta Nadir Lokalizasyonlu Silyalı Kutanöz Kist: Olgu Sunumu

[1] Hayriye KARABULUT

[1] Baran ACAR

[2] Servet GÜREŞÇİ

[1] Mehmet Ali BABADEMEZ

[1] Selcan BAYSAL

[1] Rıza Murat KARAŞEN

[1] Department of Otolaryngology, Ankara Kecioren Research and Training Hospital Ankara, Turkey

[2] Department of Pathology, Ankara Kecioren Research and Training Hospital Ankara, Turkey

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Corresponding Address /Yazışma Adresi:

Dr. Hayriye KARABULUT

Department of Otolaryngology Ankara Kecioren Research and Training Hospital

Ankara, Turkey

Tel: +90 0312 3569000

Fax: +90 312 3569002

e-mail:drhayriye@hotmail.com

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duzcetipdergisi@duzce.edu.tr

ABSTRACT

The cutaneous ciliated cyst (CCC) is an unusual lesion found in young women that typically occurs on the lower limbs. We report a case of cutaneous ciliated cyst of nasal sulcus in a 33 year-old woman, previously undescribed anatomic location for this lesion. Total excision provided healing and recurrence did not occur in five months following up. Here we also report immunohistochemical staining to identify the cyst.

Key words: Cutaneous ciliated cyst, nasal sulcus

ÖZET

Silyalı kutanöz kist genç kadınlarda, tipik olarak alt ekstremitelerde görülen, nadir bir lezyondur. Otuz üç yaşında bir bayan hastada, bu lezyon için daha önce tanımlanmamış bir anatomik lokalizasyon olan nazal sulkusta silyalı kutanöz kist olgusunu bildiriyoruz. Hastanın tedavisinde total eksizyon ile tam iyileşme sağlandı ve 5 aylık takipte nüks görülmedi. Burada ayrıca silyalı kutanöz kisti tanımlamada kullanılan immünhistokimyasal boyamalar da bildirilmektedir.

Anahtar kelimeler: Silyalı kutanöz kist, nazal sulkus

INTRODUCTION

The cutaneous ciliated cyst (CCC) is an unusual cystic lesion that usually affects the lower limbs of young females(1). Two theories have been proposed to explain the mechanism of lesion formation: Heteropia of the müllerian epithelium and metaplasia of the ecrine glands. While positive staning for amilase, sex steroid reseptors supports the first theory, positive immunohistochemical staining for carcinoembriyogenic antigen (CEA), epithelial membrane antigen (EMA) and periodic acid schiff (PAS) supports the second theory (2). To date 41 cases have been reported in the literature(1-7)

CASE REPORT

A 33 year-old woman presented with a mass on the right nasal sulcus that she recognized when it enlarged in the past two weeks. On physical examination the lesion was sore, tender and 3 cm in diameter extending from nasal sulcus to buccal area. The lump was aspirated two times before and after the cytological examination, and it was reported as a benign cyst. As it recurred within 1-2 months, the cyst was totally excised with nasal sulcus incision. Pathological examination revealed a multiloculated cyst measuring 2,7 cm in diameter. On microscopic examination the cyst wall was composed of loose connective tissue with papillary projections into the lumen lined by pseudostratified epithelium (figure 1A). The epithelium was ciliated in most of the parts (figure 1B) and was alternating with nonkeratinized squamous epithelium in focal areas. There were many scattered vacuolated cells. The cyst wall was infiltrated with lymphocytes and macrophages and was devoid of adnexa, mucous gland, cartilage or smooth muscle. By immunohistochemical examination epithelial cells showed positive reaction with EMA (figure 2A) and CEA (figure 2B). Immunostaining against smooth

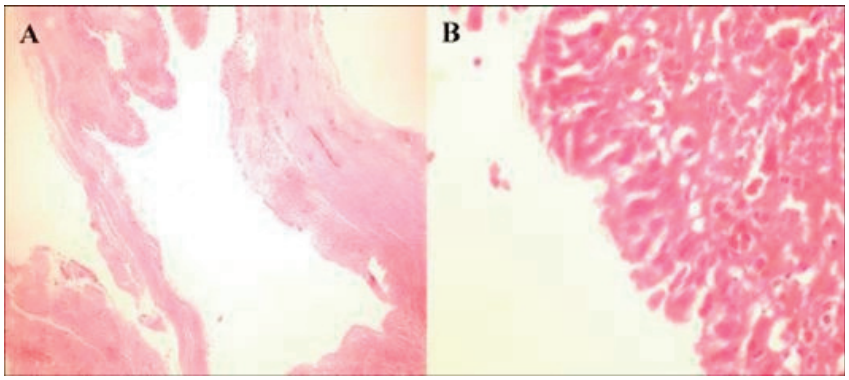

Figure 1. A: Cyst wall lined with stratified epithelium with papillary projections (HEx 40) B: Ciliated epithelium with high magnification (HEx400)

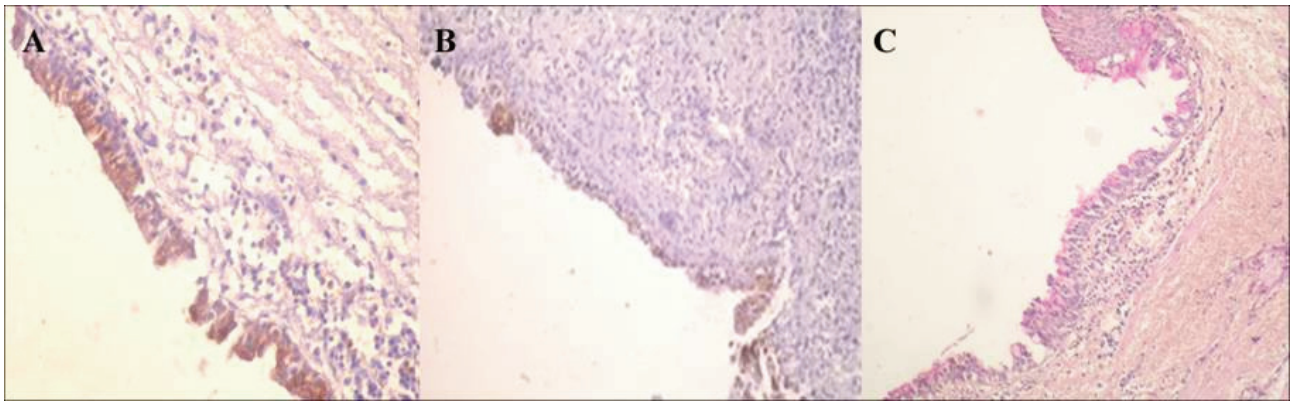

Figure 2A: Immunoperoxidase staining for EMA (x200) B: Immunoperoxidase staining for CEA (x100) C:PAS positive vacuolated cells (PAS x200)

muscle actin (SMA), S-100, estrogen and progesterone receptor were negative. The vacuolated cells were PAS positive (figure 2C) but alcian blue negative.

DISCUSSION

The cutaneous ciliated cyst (CCC) is a rare lesion that is found in young women, and typically occurs on the lower limbs. Although the pathogenesis of CCC is unclear, two major theories have been proposed to explain the mechanism: the theory of sequestration and migration (heteropia) of the ciliated epithelium from the müllerian epithelium, and ciliated metaplasia of the eccrine glands. The first theory was supported with the similarities between the fallopian tube epithelium and CCC. The occurrence of CCC on the lower extremities and buttocks was explained by the heterotopia of müllerian duct cells during early embryogenesis. Since it has been reported firstly in 1890 by Hess (3) there have been 41 reported cases of CCC (4,5), but only 2 of them were on face. This rare location can be explained by metaplasia theory that the embryonic ecrine glands contain cilia in their secretory and ductal epithelia (6). We reported our case because of its rare anatomic location.

For the proof of sequestration and migration theory, the previous studies have reported positive staining for fallopian tube markers as amylase (7), and sex steroid receptors (1). In support of the metaplasia of the ecrine glands, monoclonal antibodies for SMA, CEA and EMA were used in previous studies (2). Our study supports the epithelial theory oppose the ecrine theory for some reasons. Firstly the cyst localization is on face, away from lower extremities. Secondly the appearance of the cyst is not related with hormonal irregularity periods like puberty or pregnancy. Thirdly positive immunohistochemical staining for CEA, EMA and PAS that would be expected in ecrine glands, supports the theory. Negative staining for estrogen and progesterone receptors suspends from müllerian theory.

CCC should also be considered in the differential diagnosis of subcutaneous lesions like sebaceous cyst, epidermal cyst. This case is the third one as a CCC on face reported until today. Recurrence has not been seen in the fifth month of the surgical excision in our case. Information about following up has not been reported in the previous studies.

In conclusion, CCC can also occur on face and total excision provides healing.

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