An Unusual Localization of a Trichofolliculoma: External Auditory Canal: Case Report

Trikofollikülomanın Sıra Dışı Yerleşimi: Dış Kulak Yolu

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ABSTRACT Trihcofolliculoma is an uncommon, benign hamartomatous neoplasm of the hair follicle, most commonly occurring in the head and neck region of the adults. It tends to locate on the face and scalp. Other rare sites of occurrence include vulva, eyelid and upper lip. Clinically, the lesions are small, range from 0.2 to 0.5 cm in diameter, and appear as whitish-red or pearly papules or as smooth nodules, sometimes with a central pore containing wool-like wisps of immature hair. External auditory canal is an extremely rare localization of trichofolliculoma, and to the best of our knowledge, only three cases have been reported in the literature until now. In this report, we presented an unusual case of trichofolliculoma localized in the external auditory canal leading to hearing loss. In this case, surgical excision was enough to treat external auditory canal mass which was a trichofolliculoma.

Key Words: Ear canal; hearing loss

ÖZET Trikofolliküloma, sıklıkla yetişkinlerin baş ve boyun bölgesinde ortaya çıkan, saç folikülünün nadir, iyi huylu hamartomatöz tümörüdür. Vulva, göz kapağı ve üst dudak seyrek olarak görüldüğü diğer bölgelerdir. Klinik olarak lezyonlar küçük, 0,2-0,5 cm çapında, beyazımsı kırmızı renkte veya sedefli papüller veya bazen immatür saçın ortası gözenekli yün demetleri benzeri yumuşak bir nodül olarak ortaya çıkmaktadır. Dış kulak yolu, trikofollikülomanın oldukça nadir görüldüğü bir bölgedir ve bilgilerimize göre günümüze kadar literatürde üç olgu bildirilmiştir. Bu çalışmada, işitme kaybına götüren dış kulak yolu yerleşimli sıra dışı bir trifolliküloma olgusunu sunmaktayız. Bu olguda trikofollikülomanın sebep olduğu dış kulak yolu kitlesinin cerrahi olarak çıkarılması tedavi için yeterli olmuştur.

Anahtar Kelimeler: Kulak kanalı; işitme kaybı

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Trichofolliculoma is an uncommon, benign hamartomatous neoplasm of the hair follicle, most commonly occurring in the head and neck region of the adults.[1,2] External auditory canal is an extremely rare localization of trichofolliculoma and to the best of our knowledge, only three cases have been reported in the literature until now.

In this report, we presented an unusual case of trichofolliculoma localized to external auditory canal leading to hearing loss.

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CASE REPORT

A 33-year-old female patient was admitted to our clinic with the complaint of left sided hearing loss and a mass in the left external auditory canal (EAC). The patient told that her complaints had started 4 months ago.

As it was concluded from her history, the initial lesion started in the form of an asymptomatic, painless papule, 2- 3 mm in diameter. In time, the lesion enlarged and occluded the ear canal. Except for the hearing loss, there were no symptoms such as pruritus, otorrhea, otalgia or tinnitus. On otomicroscopic examination, left external auditory meatus was obstructed by a smooth-surfaced firm mass (Figure 1). The left tympanic membrane could not be seen. Right EAC and tympanic membrane were normal. Tuning fork tests revealed that, Weber's test was lateralized to left, and Rinne test was negative at the left and positive at the right ear. Other otorhinolaryngologic and systemic examination findings were normal. Surgery was planned with a prediagnosis of ear canal polyp or a soft tissue tumor of EAC. The patient underwent mass excision from left EAC and primary suturation was enough to repair the wound site. The mass was 14x9x5 mm in size. The histopathologic evaluation of the mass demonstrated the diagnosis of trichofolliculoma (Figure 2). There was no recurrence after a period of 36 months in the follow-up.

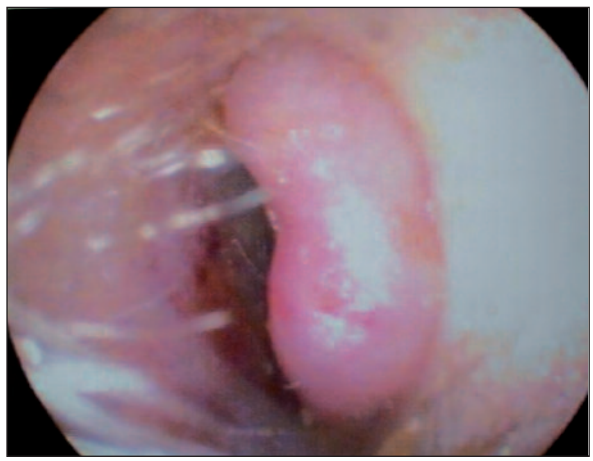

FIGURE 1: A firm mass occluding the left external auditory canal.
(See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

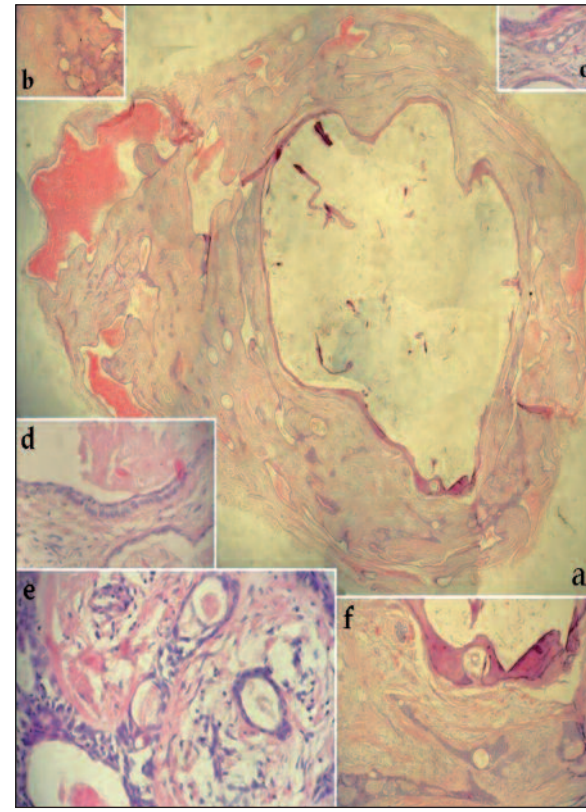

FIGURE 2: a. Primary follicle lined by infundibular, stratified squamous epithelium, 40xHE. **b.** Smaller infundibula radiating from the epithelial wall of the infundibular cystic space. **c, d.** Follicular structures and associated stroma 400xHE. **e.** Hair structures within the immature hair follicle lumen 400xHE. **f.** Secondary and tertiary follicles 100xHE.
(See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

DISCUSSION

Trichofolliculoma, first described in 1944 by Miescher, is an extremely rare benign hamartoma of the hair follicle tissue.[1] Clinically, the lesions are small, ranging from 0.2 to 0.5 cm in diameter, and appear as whitish-red or pearly papules or as smooth nodules sometimes with a central pore containing wool-like wisps of immature hair.[2-5] It is a tumor of adulthood with an exceptional report of congenital trichofolliculoma. There is no associated family history, or other systemic abnormalities suggesting that it is acquired rather than being a part of a syndrome. Our patient also did not have an associated family history or another abnormalities.[4,6]

Trichofolliculoma tends to locate on the face and scalp.[1,2] Other rare sites of occurrence include vulva neck, eyelid and upper lip.[6-8] One of the ex-

tremely rare localizations of trichofolliculoma is EAC. To our knowledge, only three EAC trichofolliculoma cases have been reported up to date.[9-11] The first EAC trichofolliculoma case was described in 1979 by Srivastava and Ajwani.[9] Two years after, O'Mahony reported the second case of EAC trichofolliculoma presenting as a fleshy mass occluding the left external auditory meatus and leading to hearing loss and tinnitus.[10] Our case also had conductive hearing loss on the left ear, but she did not have any complaint of tinnitus. In 1986, Cohen and Davis reported a young girl who developed multiple trichogenic adnexal tumours as well as two polypoid ear lesions caused by trichofolliculoma.[11]

The diagnosis of trichofollicolma was mostly made by histopathologic examination. The differential diagnosis includes all external auditory canal masses. The pre-diagnosis of our case was external auditory canal polyp. Treatment by excision and primary suturation could not be enough if the treatment was delayed.

Histopathogically, trichofolliculoma usually consists of a centrally dilated epithelium with keratinized stratified epithelium with keratohyaline granules, and numerous small secondary and tertiary follicles radiating from the keratin-filled primary follicle. The differential diagnosis of trichofolliculoma includes keratoacanthoma, basal cell carcinoma, trichoepithelioma, and syringoma.[4,6]

Trichofolliculoma may be seen in multiple localizations in the same patient and it may also be found with other adnexial tumors such as trichogenic myxoma.[11] Our patient has a solitary trichofolliculoma of left EAC without any other adnexial tumors.

Our case illustrates a peculiar localization and clinical manifestation of trichofolliculoma.

In conclusion, trichofolliculoma should be considered in the differential diagnosis of masses that occlude EAC, such as aural polyp and basal cell carcinoma.

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