

An Interesting Localization of Trichoepithelioma: Case Report

İlginç Lokalizasyonlu Bir Trikoepitelyoma Olgusu

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ABSTRACT Trichoepithelioma is a benign cutaneous tumour originating from the hair follicles and it is most commonly found on the face and scalp. Its onset is usually in childhood or early adult life. Histologically, it contains horn cysts and abortive hair papillae. Although trichoepithelioma is usually encountered on cutaneous tissue, we present a rare case of trichoepithelioma localized on the hard palate. A 16-year-old man was referred to our clinic with complaints of a big swelling overlying left side of the palate for approximately 8 months. Submandibular, submental and neck examinations were free of lymphadenopathies. The lesion was removed surgically and it was diagnosed as a trichoepithelioma. Postoperative healing was uneventful.

Key Words: Palate, hard; oral surgical procedures

ÖZET Trikoepitelyoma saç folikülünden köken alan, sıklıkla yüz ve skalpta görülen benign kutanöz bir tümördür. Çocuklukta veya erken olgunluk döneminde oluşmaya başlar. Histolojik olarak "horn" (boynuz) kistleri ve abortif saç papilleri ihtiva eder. Trikoepitelyoma genellikle kutanöz dokuda görülmekle birlikte, bu çalışmada sert damakta lokalize nadir bir trikoepitelyoma olgusu sunulmuştur. On altı yaşındaki erkek bir hasta yaklaşık 8 aydır mevcudiyeti bilinen damak bölgesi sol kısımda lokalize büyük bir şişlik nedeniyle kliniğimize başvurdu. Submandibular, submental ve boyun muayenesinde lenfadenopatiye rastlanılmadı. Lezyon cerrahi olarak çıkarıldı ve trikoepitelyoma olarak tanılandı. İyileşme problemsizdi.

Anahtar Kelimeler: Sert damak; oral cerrahi prosedürler

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Trichoepithelioma is a benign cutaneous tumour originating from hair follicles and most commonly found on the face and scalp.^{1,2}

Trichoepithelioma occurs either as multiple or solitary lesions. Solitary trichoepithelioma occurs more commonly than multiple trichoepithelioma, is not inherited and consists of a firm, elevated, flesh-colored nodule usually less than 2 cm in diameter.^{1,2} Its onset is usually in childhood or early adult life. The presence of a solitary trichoepithelioma and an apocrine adenoma within the same tumour has been described.¹

Trichoepithelioma is usually encountered on the cutaneous tissue. We could not find any trichoepithelioma that was localized on oral mucosa in a review of the current literature. We are presenting the first case of trichoepithelioma localized on hard palate.

CASE REPORT

A 16-year-old man was referred to the Oral and Maxillofacial Surgery Clinic of Ataturk University, Faculty of Dentistry with complaints of a big swelling overlying the left side of the palate. The patient reported that this swelling had been noticeable for approximately 8 months. On examination, a solitary big nodule was found in the palate region. Palatine mucosa and the other oral tissue were intact. Dentition was within the normal limits. Submandibular, submental and neck examinations were free of lymphadenopathies. Routine laboratory investigations were within the normal limits.

Coronal computed tomography (CT) showed an approximately 2.5 x 2.5 cm, well-circumscribed solitary mass on the left hard palate. The mass did not lead to bony destruction or invasion (Figure 1). Our differential diagnosis for palatal lesion included: pleomorphic adenoma, low grade salivary gland malignancy, lymphoma and epidermoid carcinoma.

The patient was informed of the possible risks and benefits of the surgery. He signed an institutional consent form. A needle aspiration biopsy was performed to establish histological characteristic of the lesion. It was reported as benign. Under general anesthesia, the lesion was removed en block with 0.5 cm margins, including the overlying palatal mucosa, and the palatine vessels were ligated



FIGURE 1: Coronal CT view showing a non destructive mass on the hard palate.

(Figure 2, 3). Gauze iodoform pack and preoperatively prepared palatal plate were inserted to the defect. The packing was replaced biweekly. Postoperative recovery was uneventful. After six months, clinical and radiographic examination of the patient was carried out, and a good healing was seen in the operation site.

Specimen was prepared for microscopic examination after fixation with 10% formalin, dehydration with alcohol and embedded in paraffin blocks. Histopathological examination showed an entirely subepithelial and well circumscribed tumor (Figure 4). Horn cysts were the most characteristic histologic feature. They consist of a fully keratinized center surrounded by basophilic cells.

Tumour exhibited anastomosing cords of epithelium and one or a few layers of cells with eosinophilic cytoplasm and large, oval, pale, vesicular nuclei situated between the basophilic and horn cysts. Some of the basophilic cells were arranged in an adenoid network (Figure 5). Occasionally, they were found as solid aggregates. Tumour islands showed peripheral palisading of cells and surrounded by either a myxoid or a hyalinized stroma. The epithelial cells did not exhibit nuclear atypia or mitotic figures. In addition, they were observed in some dystrophic mineralization within intact horn cysts (Figure 6).

Histochemical and immunohistochemical staining results were as follows; positive for keratin and for vimentin; negative for mucicormine, CEA, S-100, and actin. Thus, the lesion was diagnosed as a trichoepithelioma.

DISCUSSION

Trichoepithelioma is a benign skin tumour originating from hair follicles. It may occur as a solitary non-familial type. The lesion is seen on the face, forehead and upper lip. Occasionally, lesions are seen on the scalp, neck and upper trunk. Trichoepitheliomas are skin-coloured and frequently are traversed by telangiectases. In case of numerous trichoepitheliomas, inheritance is through an autosomal dominant trait. Solitary trichoepithelioma is not inherited.¹



FIGURE 2: Intraoral photograph shows the hard palate after excision of the mass.



FIGURE 3: View of the resected mass.

In our literature review, we could not encounter any case of trichoepithelioma localized on oral mucosa. However we found a case called benign keratotic squamous epithelial neoplasm reported by Mangarano et al.³ It was reported that this case looked most likely to trichoadenoma but it was not a perfect match.³ Although mixed tumor or pleomorphic adenoma often occurs in the palate, it involves both major and minor salivary glands and it

is not related to the skin or the hair follicles.⁴

A cutaneous tumour seen on oral mucosa originating from hair follicles can be explained by following two theories:

One theory is that ectopic adnexal structures were embryologically trapped in the palatal mucosa and underwent neoplastic change. One adnexal structure that is routinely found in the oral cavity is the sebaceous glands.³

Another possibility is that there was a metaplastia in the surface epithelium, creating cells capable of forming the present lesion.³

Histologically, the trichoepithelioma may also be confused with basal cell carcinoma, trichoblastoma, trichoadenoma, syringoma and microcystic adnexal carcinoma. Trichoepithelioma is associated nearly always with some evidence of fol-

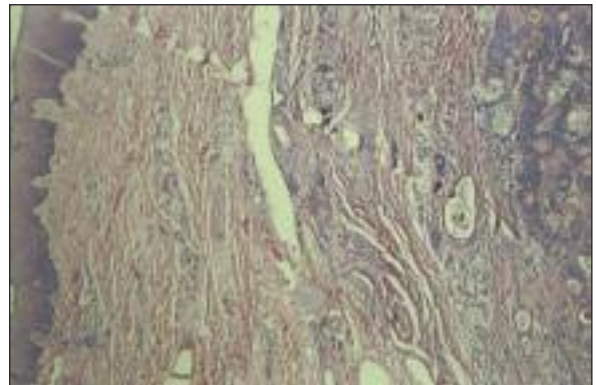


FIGURE 4: Trichoepithelioma is localised under the epithelium (Hematoxylin-Eosin stain original magnification x 100).

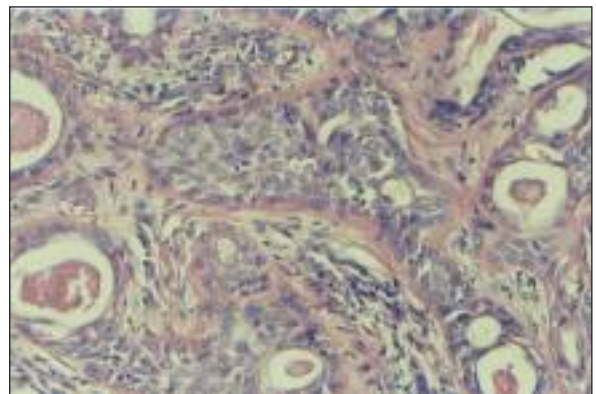


FIGURE 5: The basophilic cells is arranged in an adenoid network. (Hematoxylin- Eosin stain original magnification x 400).

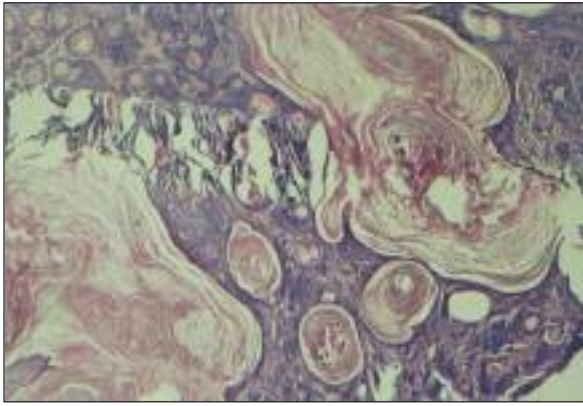


FIGURE 6: Horn cysts are the most characteristic histologic feature in the trichoepithelioma (Hematoxylin- Eosin stain original magnification x 100).

licular differentiation in the manner of follicular germs and papillae at least, whereas basal cell carcinoma practically never exhibits follicular germs and papillae. Instead, it shows signs of follicular differentiation in the form of follicular germs alone at distal ends of columns and cords of pink neoplastic cells and of tiny infundibular cysts. The stroma in trichoepithelioma is almost always prominent and consists of bundles of collagen that resemble those of the perifollicular sheath. In contrast, practically no stroma is associated with a basal cell carcinoma. Lastly, trichoepithelioma is usually constructed of neoplastic basaloid cells arranged in cribriform pattern whereas neoplastic squamoid cells of basal cell carcinoma arrayed in an anastomosing pattern.^{1,3,5-7}

Histopathologic features of trichoblastoma included aggregations formed mostly by follicular germinative cells, nuclei of neoplastic cells at pe-

ripheries of nodules are often columnar and in palisaded array. Identifiable follicular germs and papillae are present, nodules of neoplastic cells extent throughout the dermis and subcutaneous fat and even into skeletal muscle. Sometimes, two-toned nodules comprise of a large central eosinophilic zone and a narrow peripheral basophilic rim. The stroma is highly fibrocystic, abundant mucin saturates the stroma at times, and individual necrotic keratinocytes are scattered within aggregations of neoplastic cells.^{1,5}

Trichoadenoma has numerous cystic components. Cystic structures are distributed uniformly throughout the lesion. Cyst wall thickness vary little from cyst to cyst. Trichoadenomas are not tubular structures.^{1,3}

The resemblance to microcytic adnexal carcinoma may be considerable, it shows horn cysts, strands of basaloid cells and a dense desmoplastic stroma. It differs, by showing ductal structures and a deeply infiltrating growth.¹

The horn cysts near the epidermis in syringoma resemble those occurring in trichoepithelioma, and their presence in syringoma was formerly misinterpreted as the occurrence of both types of tumours within the same lesion. Although trichoepithelioma shows solid strands of basophilic epithelial cells and horn cysts, it lacks ductal structures.⁶

As a result, this case shows that trichoepithelioma should also be considered in the differential diagnosis of palatal masses.

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