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CASE REPORT Open Access

# Unilateral auricular multiple trichoepitheliomas: a case report and review of literature

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## Abstract

**Background** Trichoepithelioma is a rare benign tumor. It could be an inherited or acquired condition. Trichoepithelioma has mostly involved the face with bilateral multiple lesions. Involvement of the auricle by this tumor is extremely rare encountered in clinical practice.

**Case presentation** A 36-year-old female presented to the Dermatology Clinic with painless right auricular papules 4 years ago. Examination revealed multiple non-tender, firm, rounded, dome-shaped, flesh-colored, pink, and shiny papules. There was mild bleeding from the lesions after trivial trauma. There was no family history of similar problem. No abnormality was found on other examinations. An excisional biopsy of one lesion was performed under local anesthesia. The histopathological evaluation confirmed the diagnosis of trichoepithelioma. The lesions under local anesthesia were excised with primary closure of the wound. No recurrence was seen during 2 years of follow-up.

**Conclusion** Only five cases of trichoepitheliomas that affected the auricle were reported in the PubMed database. Four of them are affecting both auricles as well as other parts of the body, particularly the face. The fifth case was with a unilateral single giant auricular lesion. Our case was with non-familial multiple unilateral auricular trichoepitheliomas. The tumors responded well on surgical excision and primary closure.

**Keywords** Trichoepithelioma, Auricle, Unilateral trichoepithelioma, Multiple trichoepitheliomas, Case report

## Background

Trichoepithelioma (TE) is a rare benign (harmless) tumor that originates from the walls of the hair follicle after puberty. Trichoepithelioma is mostly seen in the face, nose, scalp, and/ or upper lip. It is characterized by the benign proliferation of epithelial-mesenchymal origin cells [1]. TEs are categorized into three types: solitary, multiple (2 types), and desmoplastic [2, 3]. Multiple familial TE type 1 is an inherited disorder that generally progresses through adolescence and over time they grow larger and increase in number. The genetic disorder can be triggered by alterations in the CYLD gene or by changes in other genes which are quite unknown. Meanwhile, the multiple familial TE type 2 is an acquired mutation which is essential for the tumors to develop [4, 5]. The tumors affect women more frequently and severely than men, and they can be mutilating and cause psychological problems [3]. The tumors signify benign hamartomas of the pilosebaceous apparatus. TEs have tumor islands of basaloid cells, horn cysts, and immature hair papillae [5].

The rare incidence and absence of enough signs made TE management a controversial domain in dermatologic surgery. Few cases involving the auricle have been reported [6–9]. In all these reported cases, the distribution of the tumors was on both auricles as well as the

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involvement of the whole face, apart from the study by Genc et al. which reported a left giant single auricular TE. Hence, our case differs from the abovementioned reported cases in that the tumors were multiple and involved the right auricle only.

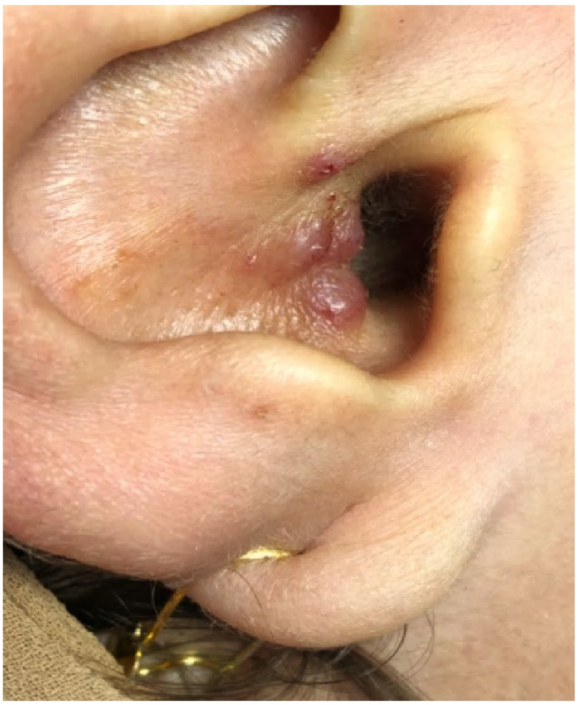

**Fig. 1** Multiple papules that incompletely fill the outer ear canal

## Case presentation

A 36-year-old healthy woman presented to the Dermatological Private Clinic with right auricular papules of 4 years duration. Physical examination showed multiple asymptomatic firms, rounded, dome-shaped, flesh-colored, pink, and shiny papules that increase in number. It involved only the right side outer ear canal (Fig. 1). The condition is sometimes associated with slight bleeding after mild trauma. There was no family history of the same condition. The rest of the ear, nose, throat, and systemic examination were normal. We put the following as a differential diagnosis of these lesions: benign trichoblastomas, trichofolliculomas, trichoadenomas and trichoepitheliomas, and basal cell carcinoma. An excisional biopsy of a single lesion was performed and sent for histopathological examination. The report shows keratinized stratified squamous epithelium strands of the epidermis consisting of a superficial nest of small basaloid cuboidal cells overlying the dermis. Many keratinous cysts have a marginal border of basaloid cuboidal cells, and keratinous cysts are enclosed by thick fibrous stroma (Fig. 2). Therefore, we reached the diagnosis of non-inherited multiple TEs of the auricle. Surgical excision of the lesions with primary closure was performed by an otolaryngologist under local anesthesia. No recurrence was observed at a 2-year follow-up. Informed consent was obtained from the patient for case publication with the related images.

## Discussion

TEs are rare benign tumors initiated from the pilosebaceous follicle. Typically, it affects females in early childhood or early adolescence. In spite of that, several cases of non-familial multiple TEs reported the involvement of

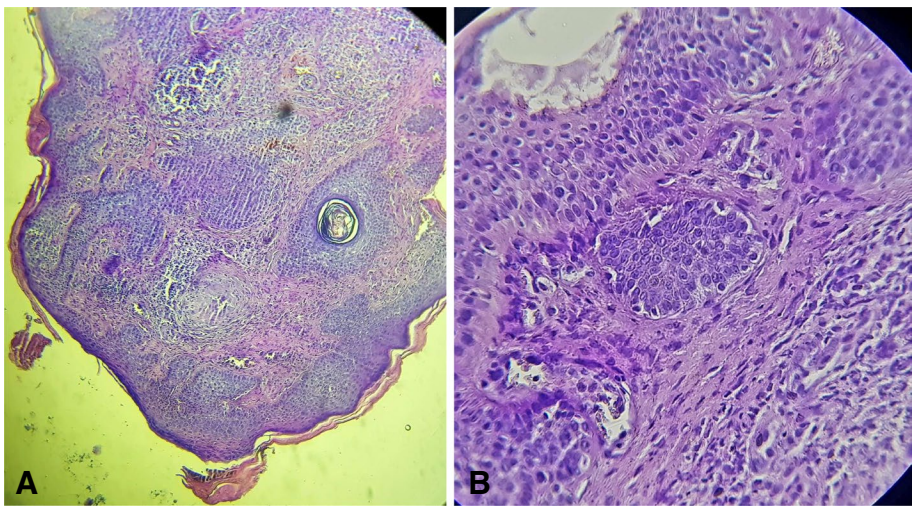


**Fig. 2** Tumor consisting chiefly of a nest of basaloid cells with horn cyst (H&E; **A** with × 100, **B** with × 400)

**Table 1** Reported cases of trichoepithelioma of the auricle in PubMed from 1990 to 2022

| Authors | Year | Country | Age per years | Gender | Duration | Family history | Side | Singles or multiple | Involvement | Deafness | Treatment modality | Follow-up period | Recurrence |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Bibi et al. [6] | 1990 | Israel | 53 | Female | 10 years | Positive | Bilateral | Multiple | Whole face | Conductive 50 dB | Excision with a split skin graft | 5 years | No |
| D'Souza et al. [7] | 1994 | India | 48 | Male | 30 years | Positive | Bilateral | Multiple | Face, both pinna, scalp, neck, shoulders, upper back, and limbs | No | Excision with a split skin graft | 1 year | No |
| Kaluskar [8] | 2005 | Ireland | 52 | Female | 5 years | Negative | Bilateral | Multiple | Not mentioned | Conductive | KTP/532 Laser | 9 months | No |
| Genc et al. [9] | 2012 | Turkey | 86 | Female | Not mentioned | Negative | Left | Single | No | Conductive | Excision and primary closure | 6 months | No |
| Sehrawat et al. [12] | 2016 | India | 35 | Female | 9 years | Positive | Bilateral | Multiple | Face, scalp, neck, and left forearm | No | Not mentioned | Not mentioned | |
| Our case | 2023 | Iraq | 36 | Female | 4 years | Negative | Right | Multiple | No | No | Excision with a split skin graft | 2 years | No |

adult women patients [10]. The exact prevalence of TEs is not yet known. TEs are characterized by a slowly progressive increment in the size and number of the lesion for long periods, and the usual presentation of the tumors is for a cosmetic reason [11]. This case report presented a 36-year-old woman lacking family history, complaining of right auricular TEs for 4 years duration. Table 1 shows the reported cases of TEs of the auricle.

The patient in the current study seeks medical advice because of their increased number of lesions and frequent bleeding with slight trauma, unlike previous cases that were presented to the medical clinics usually for cosmetic goals and to recover their outlook. Others showed asymptomatic dense deformed tumors that cause psycho-social problems [13, 14].

Basal cell carcinoma (BCC) shares similar features to the TE except that it tends to affect older age group individuals with a single ulcerative pigmented nodule as well as the histology illustrates the presence of the mitotic figure, apoptotic cells, retraction between epithelium and stroma, and ulceration. The present work shows a young age patient with non-ulcerated multiple pink papules. While the histological study was characterized by basaloid cells which develop from undifferentiated germinative cells of the follicular-sebaceous unit and keratinous cysts enclosed by thick fibrous stroma, these findings give a base for the diagnosis of TEs. Besides, there are immunohistochemical methods that may be worth distinguishing the two [15, 16]. However, this modality is not present in our city as well as it is costly.

At the other end, the TE shares histological features with trichoadenoma. However, this benign tumor originates on the lines of the infundibular portion of the pilosebaceous unit due to the existence of keratinized epidermoid cells in the cyst wall with keratohyalin granules. Clinically, trichoadenoma presents as a single grayish tumor, measures up to 15 mm in diameter, and affects the face or buttocks [17].

TEs show a major cosmetic worry and the treatments are challenging particularly in patients with multiple auricular lesions similar to the presented case. Numerous modalities have been useful in treating TEs such as surgery, electro destruction, ablative resurfacing, obliteration by CO2 laser or Erbium-YAG laser, and topical medications like imiquimod and rapamycin [18].

Surgical excision of the TEs has been proven to be effective and prevent the deep damage of underlying tissue. But TEs involving auricles had been reported in 3 cases that undergone surgical removal that caused high rates of scarring and poor cosmetic outcome. The laser irradiation that offers less pain and fast recovery was used for the fourth patient but may give a bad outcome and scaring of the cartilage of the auricle. However, management of multiple TEs is an extremely hard attempt and all treatment methods carry a risk of side effects such as reddens swelling, hyperpigmentation, recurrence, and sometimes scarring [11].

Although TEs are benign, they can quite rarely experience malignant transformation to basal cell carcinoma or trichoblastic carcinoma. The histopathological findings and immunohistochemistry if needed have been considered for the differentiation [19].

## Conclusion

TEs of the auricle are seldom to be seen in daily clinical practice. Few cases of TEs involved the auricle that were reported in the literature. All of them are bilateral and associated with other lesions in the body except one case which was single, giant, and on one auricle. The presenting case was with non-hereditary multiple TEs on one auricle and without other lesions neither in the face nor other parts of the body. Surgical excision of the lesion with primary closure was successful in the presenting case.

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#### Authors' contributions

Kubaisi TA analyzed and interpreted the patient data regarding the clinical and pathological finding as well as writing the basic manuscript draft. Al-Ani RM formatted the references and writing the final manuscript draft. All authors read and approved the final manuscript.

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#### Availability of data and materials

All patient's data was presented in the article.

### Declarations

#### Ethics approval and consent to participate

This study was approved by the ethical committee of the University Of Anbar. Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

#### Consent for publication

The patient gave written informed consent for the publication of the data and materials contained within this study.

#### Competing interests

The authors declare that they have no competing interests.

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## Publisher's Note