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TRICHOEPITHELIOMA OF LEFT NASAL VESTIBULE :A UNIQUE CLINICAL PRESENTATION

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ABSTRACT Trichoepitheliomas are rare cutaneous benign lesion arising from the hair follicles. originate from the epithelial - mesenchymal cell having rare risk of malignant transformation. Trichoepitheliomas are mostly seen in the scalp, nose, forehead and upper lip.We report a 66 years old male presented with complaint of swelling in the left nasal vestibule area that was removed surgically with skin closure.

KEYWORDS : trichoepithelioma, nose, vestibule, excision

INTRODUCTION

Trichoepithelioma is a rare benign cutaneous tumor arising from hair follicle cells, characterized by a low risk of malignant transformation [1]. The tumor originates from epithelial-mesenchymal cell [2]. There are two clinical presentations of this condition: a hereditary sex-linked multiple form, which typically affects the face, scalp, and upper thorax of young adults; and a non-hereditary solitary form, which may occur on any part of the body (predominantly the face) in adults [3]. Typically, trichoepitheliomas present as solitary lesions, with an idiopathic etiology.

Histologically, trichoepithelioma is classified as a superficial trichoblastoma exhibiting prominent infundibulocytic differentiation [4] The main difference between trichoblastomas and trichoepitheliomas is the depth at which they arise in the dermis. Trichoblastomas are found in the deep dermis and subcutaneous tissue, whereas trichoepitheliomas are more superficial

This paper presents a case of a solitary trichoepithelioma located of nose, which was surgically excised. We discuss the diagnostic and therapeutic considerations related to this rare skin tumor.

CASE PRESENTATION

A 66-year-old male patient presented to the outpatient department with a 10-year history of a mass in the left nasal cavity and difficulty breathing for the past year. His medical history included hypertension, and he had undergone angioplasty in 2011.

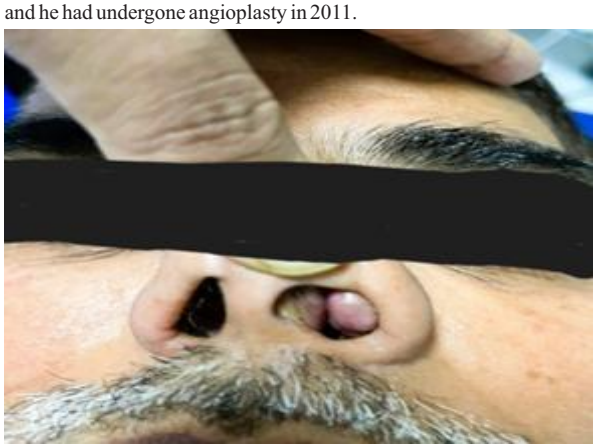

Figure 1: Mass in left nasal cavity

On examination, a firm to hard mass measuring 2 cm × 2 cm was noted in the left nasal cavity, arising from the vestibule. The mass was non-indurated and did not bleed on palpation. The remainder of the ENT examination was unremarkable.

Management: The patient was on antiplatelet medication, which was withheld for five days prior to surgery. He underwent surgical excision of the left nasal vestibule mass under local anesthesia with 2% lidocaine. A circumferential incision was made around the mass, and the lesion was excised in its entirety. The specimen was sent for histopathological evaluation and hemostasis was achieved prior to surgical closure.

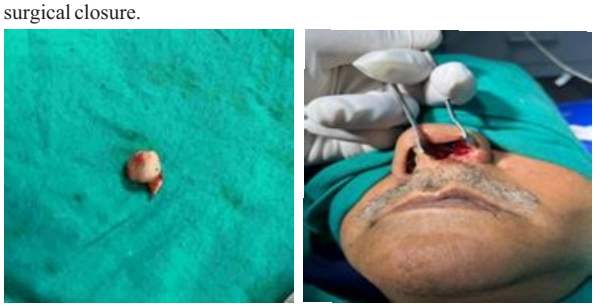

Figure 2: Nasal mass after excision

Histopathological Findings: The histopathological examination revealed a well-circumscribed dermal mass lined by stratified squamous epithelium. The dermis contained a tumor composed of superficial nests of basaloid cells exhibiting a leaf-like or frond-like architectural pattern, along with keratin horn cysts of varying sizes. Several cysts were ruptured, with associated multinucleated giant cells and keratin. The fibrous cellular stroma showed mononuclear cell infiltration.

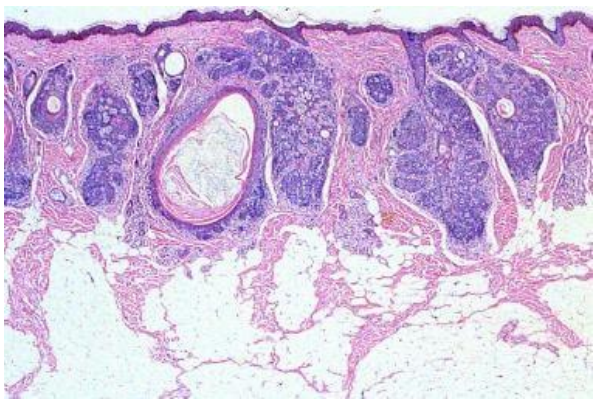

Figure 3: Histopathological slide showing tumor cells arranged in nests and islands separated by thick and thin fibrovascular stroma

The patient was followed up for six months postoperatively, during which no recurrence was observed.

DISCUSSION

Trichoepithelioma is also known as Brooke's tumour Epithelioma adenoides cysticum. Solitary trichoepitheliomas are non-hereditary benign trichogenic tumors of very low incidence that affect adults[3] characterized by a low risk of malignant transformation[1]. The tumor

originates from epithelial-mesenchymal cell[2]. We found only a few cases of solitary nasal trichoepitheliomas in the literature[5,6]

Trichoepithelioma can be divided into 3 subgroups[7]:

1. Solitary Non-Hereditary trichoepithelioma: Idiopathic.
2. `Multiple Familial Trichoepithelioma (MFT): Autosomal dominant inheritance.

Associated with Brooke-Spiegler syndrome, which is caused by mutations on chromosome 16q21, specifically in the CYLD tumor suppressor gene.

Desmoplastic trichoepithelioma

Trichoepithelioma is a rare cutaneous condition and its exact prevalence is unknown. MFT seems to affect females more, probably due to smaller expressivity and chromosomal penetration in males[8]. Furthermore, these lesions can lead to social and psychological challenges due to their facial involvement. Inadequate treatment may result in incomplete removal and recurrence of the disease, while overly aggressive interventions can increase the risk of cosmetic and functional complications[2].

Patients with MFT primarily seek treatment for cosmetic reasons due to the disfiguring nature of the lesions, which can lead to social isolation. Currently, there are no known preventive measures. Excisional surgery is the primary treatment, while non-pharmacological options include laser resurfacing, electro-surgery, cryosurgery, and dermabrasion. Laser treatments, particularly erbium and CO2 lasers, have shown lower recurrence and scarring rates compared to conventional methods.

Recurrence was associated with superficial ablation while deeper ablation can cause complications such as hypopigmentation and atrophy[9]. Topical imiquimod is a Toll-like receptor 7 agonist, and it is capable of stimulating the innate immunity and the cellular response of the adaptive immunity, so it is known to have potent antiviral, antitumor and immunoregulatory effects[10] . Many clinical trials have shown its efficacy for treating cutaneous infections, epidermal neoplasm, autoimmune disorders and angiomatous lesions without the drawbacks of surgical excision or locally destructive methods such as electrocoagulation or cryotherapy. These drawbacks are scar, hyperpigmentation or hypopigmentation.

Genetic Associations: Two genetic mutations have been identified to be associated with MFT. The first one discovered is located on the chromosomes 9p21[11]. Later, another mutation in the cylindromatosis tumor suppressor gene (CYLD) was found on the chromosome 16q12- q13.

Solitary trichoepithelioma is not distinctive clinically and may be confused with basal cell carcinoma (BCC), intradermal nevus, dermatofibroma, trichofolliculoma, and sebaceous hyperplasia. Histopathology provides the final diagnosis[12]. Differentiating between basal cell carcinoma (BCC) and trichoepithelioma is clinically significant, as BCC typically requires excision with a 3 to 4 mm margin of healthy tissue[13] whereas trichoepithelioma may only necessitate a shave biopsy or minimal resection[14].

	Trichoepithelioma	basal Cell Carcinoma
Clinical features	- Typically presents as a skin-colored papule or nodule in children and young adults	Most common non-melanoma skin cancer - Pearly skin-colored papule with telangiectasias on sun-exposed areas in older individuals
histopathologic Features	- Islands of basaloid cells that do not interact with the epidermis - Papillary mesenchymal bodies - Horn cysts - Fibroblastic stroma - No high-grade atypia - Few to no mitoses -	- Basaloid islands that may connect with the epidermis - Clefting between tumor and stroma - Peripheral palisading of basaloid cells - Central cell necrosis - Myxoid stroma - Mitotic figures
Immunohistochemical Features	Bcl-2: peripheral epithelial expression CD10: stains stromal cells, especially around the tumor cells CD34: stains fibroblastic stroma around basaloid cells	Bcl-2: diffuse expression CD10: stains stromal and tumor cells CD34: no expression

The main difference between trichoblastomas and trichoepitheliomas is the depth at which they arise in the dermis. Trichoblastomas are found in the deep dermis and subcutaneous tissue, whereas trichoepitheliomas are more superficial.

Trichoadenoma and desmoplastic trichoepithelioma are composed of cords of epithelial cells and cornifying cysts embedded in sclerotic stroma. In trichoadenoma the cystic component predominates, while desmoplastic trichoepithelioma is a mostly solid neoplasm.

Trichofolliculomas are characterized by the intermediate differentiation of hair follicle nevus, which is indicated by simple hyperplasia of hair follicles, and trichoepithelioma, which lacks mature hair follicles. If hair is not observed in a trichofolliculoma, misdiagnosis of a sebaceous cyst, nervus, molluscum contagiosum, or basal cell carcinoma is possible[15]

TRICHOFOLLICULOMA	TRICHOEPITHELIOMA
Beningn hamartomatous cutaneous adnexal tumor	rare benign cutaneous tumor arising from hair follicle cells
No malignant transformation	Chance of malignant transformation
Present as solitary dome shaped papule with an enlarged central pore, through which multiple vellus hair emerge	Present as hereditary sex-linked multiple form, non-hereditary solitary form,flesh colour papule or nodule
Excellent prognosis after excision Rarely recurred	Recurrence can be seen

In this case report, we treated the patient with surgical excision and primary closure. After a 6-month follow-up, the patient is cosmetically satisfied and has shown no signs of recurrence.

CONCLUSION

Solitary trichoepitheliomas are extremely rare benign tumors that should be included in the differential diagnosis of basal cell carcinoma when a solitary solid nodule or papule is observed on the face. Histologically, trichoepitheliomas bear similarities to basal cell carcinoma but possess a very low risk of malignant transformation. The condition is most commonly seen in young to elderly women, which may lead to significant social and psychological challenges. Confirmation via excisional biopsy is essential for accurate diagnosis and subsequent treatment.

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