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A Rare Case of Trichoepithelioma on Unusual Location

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Abstract:-

- ***Purpose:***

Trichoepithelioma is a very rare benign tumor of the skin. Trichoepitheliomas originate in hair follicles and occur in the head and neck. It is found on the scalp, nose, and forehead, but very rarely on the hairless part of the upper lip. Therefore, removal of trichoepithelioma was performed to improve facial aesthetics, which was the patient's primary concern.

- ***Material& methods/ Case description:***

An adult male patient reported to the department with the solitary nodular lesion on the non-hair bearing area of left upper lip.

- ***Result:***

Post-op after 3 months there was no scar formation or any complication.

- ***Conclusion:***

Trichoepithelioma arises from a benign proliferation of cells of epithelial-mesenchymal origin. They can be classified as solitary trichoepithelioma, multiple familial trichoepithelioma, and desmoplastic trichoepithelioma. It very commonly presents as multiple lesions with autosomal dominant inheritance. The case highlights the importance of differential diagnosis that not every lesion is mucocele (trauma). They can also be diagnosed as Basal cell carcinoma or trichoepithelioma. Therefore, proper clinical diagnosis and treatment planning is very important in such case while keeping in mind about malignant transformation in recurrence of the lesion. However, Trichoepithelioma are more common in females thus we present a unique and rare case of trichoepithelioma in adult male patient.

- ***Clinical relevance:***

Most of the upper lip and lower lip lesions having small mass in their mucosa, are clinically diagnosed as mucocele generally caused due to trauma. The case highlights the importance of differential diagnosis that not every lesion is mucocele (trauma). They can also be diagnosed as Basal cell carcinoma or trichoepithelioma. However, they are more common in females thus we present a unique and rare case of trichoepithelioma in adult male patient.

***Keywords**:- Trichoepithelioma, Upper-Lip, Hair Bearing Area.*

I. INTRODUCTION

Trichoepithelioma is an uncommon, atypical benign neoplasm of skin, emerging from the hair follicles. It is most commonly seen in the hair bearing field of scalp, nose, and forehead.[1] It can be classified into desmoplastic trichoepithelioma, solitary trichoepithelioma & multiple familial trichoepithelioma.

In 1901, solitary trichoepithelioma was first described which is frequently mistaken with basal cell carcinoma. It appears round, papule or nodular like lesion measuring anywhere between 3mm to 3cm. It is a slow growing neoplasm which is non-tender in nature. It usually occurs during adulthood.[4]

Multiple trichoepithelioma mostly occur during puberty. It appears as “symmetrically distributed crops” most commonly in mid-face region. When individually seen it appears as nonhereditary solitary trichoepithelioma but with time it gradually increases in size and become pedunculated.

- *Case Report*

A 26-year-old male patient presented to the department with the main complaint of an asymptomatic nodular lesion on the left side of the upper lip.

Patient was apparently well 2.5 years back when he notices a pea size swelling on upper lip, resembling the colour of lip. Because of this nodular lesion patient inbuilt a habit of biting his upper lip, due to which he experienced slight pain on the same site. Gradually patient notices slow increase in the size of the lesion with time. After 2 years patient started getting conscious of this nodular swelling, and due to aesthetic concern patient wanted the removal of the same. Patient does not give any history of systemic disease, family history and trauma to upper lip.

Clinical examination revealed a 1 x 1 cm nodular lesion in the hairless area of the left upper lip, approximately 2cm away from the left corner of mouth corresponding to maxillary left lateral incisor & canine, involving outer skin and mucosa. It is a hard, mobile lip mass located on the hairless area of the left middle portion of upper lip. Lesion was closer to the mucous membranes than to the skin. (Fig-1)

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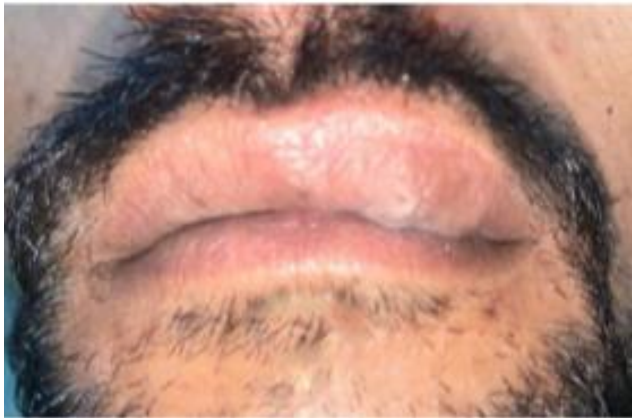

Fig 1 Pre-Operative Picture

Further, under local anaesthesia nodular lesion was transmucosally excised, maintaining 5mm margin and the lip skin was also preserved. (fig.2a&b)

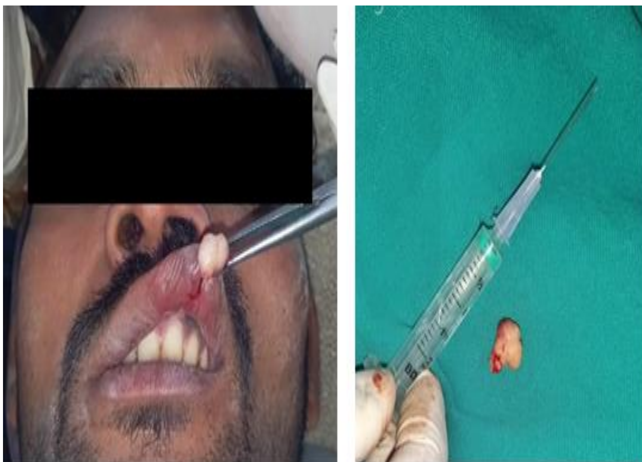

Fig 2 A) Intra-Operative Picture During Excision and B) Excised Mass

Primary closure of the wound with 3-0 prolene was done and wound left for primary healing.

Post-operatively after 3 months there was no scar mark found in the region of the upper lip and no discontinuity could be seen in the excised region. (Fig-3)

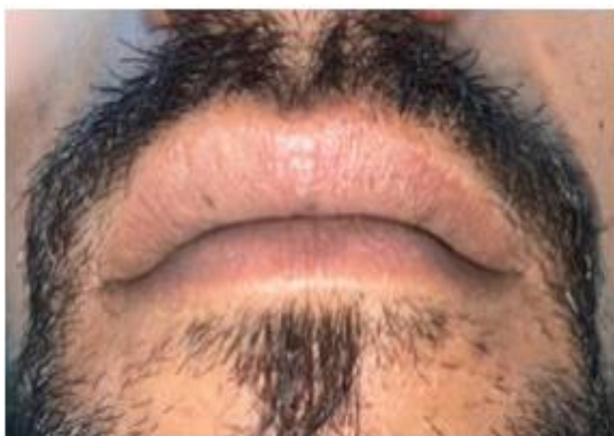

Fig 3 Post- Operative Photograph after 3 Months

- *Histopathological*

The sample was further sent for biopsy and Submitted H/E-stained tissue sections shows well circumscribed dermal nodule composed of uniform, basaloid cells showing leaf-like or frond-like architectural pattern and few central cysts filled with loose lamellar keratin. Peripheral palisading of the epithelial islands can be noted. Fibrous cellular stroma is closely seen associated with the epithelial components with no peripheral clefting between epithelial and stromal components. Few areas are also showing papillary-mesenchymal body (fibroblastic aggregate resembling abortive follicular papillae). Overall, features are suggestive of Trichoepithelioma. (Fig-4)

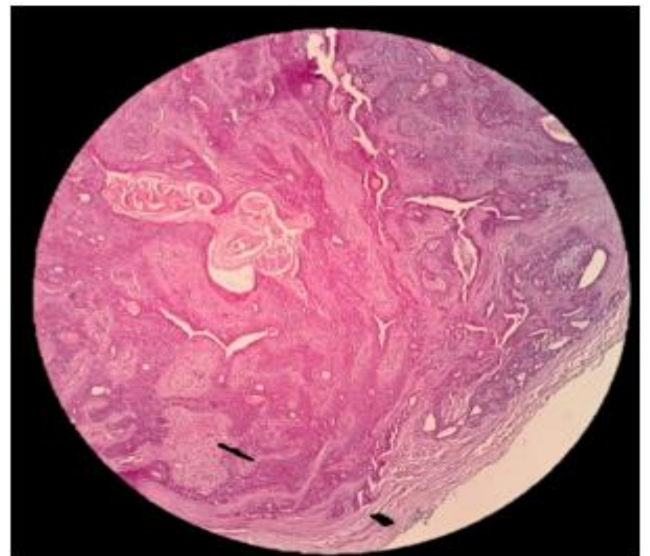

Fig 4 Histopathologic Image Indicative of Trichoepithelioma

II. DISCUSSION

The vermilion border of the lips consists of a modified mucous membrane consisting of non-hair- bearing area, immensely vascularized, non-keratinized stratified squamous epithelium. This membrane is 3 to 5 layers thick, unlike facial skin, which has 16 layers. Therefore, lips lack the typical dermal appendages of the dermal lip, with no hair follicles or glands of saliva & sweat. (Fig-5)

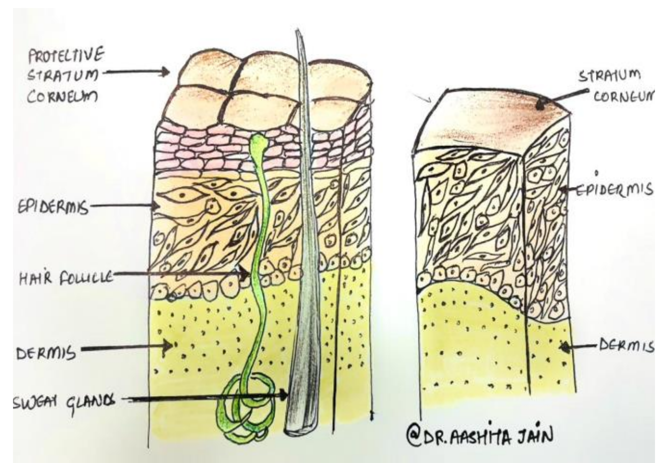

Fig 5 Illustrative Image of Skin with Hair Follicles and Without Hair Follicles

In particular, there is no visible skin lesion instead of a protruding growth pattern with elevation of the upper epidermis, which develops deeply to become symptomatic mucosal irritation. Indeed, it was initially diagnosed as mucous cyst as most of the upper lip and lower lip lesions having small mass in their mucosa, are clinically diagnosed as mucocele generally caused due to trauma. and the differential diagnosis was given as lipoma.

This solitary trichoepithelioma has very rare, uncommon & unusual features. Clinically and histologically, trichoepithelioma might be difficult to distinguish from basal cell carcinoma. Trichoepithelioma is caused by the benign proliferation of cells of epithelial-mesenchymal origin. In most cases this pathology occurs as multiple autosomal dominant lesions in the hairy skin. Therefore, multiple lesions are often found and a solitary nodular lesion is less commonly found.

Incidence of desmoplastic trichoepithelioma of 1 in 5000 has been addressed in a cohort of British adults. Of all cutaneous neoplasms DTE represents less than 1%.[5]

This nodular lesion shares familiar properties of BCC. This lesion resembles to other skin diseases in that there are papules and nodules. It is It is more common in women than in men.[3] Other differential diagnosis are syringoma, milia, sebaceous hyperplasia, angiofibroma, trichoadenoma. This is also associated with certain syndromes that include Brit-hogg-dube syndrome, cowden syndrome, bazex syndrome, rombo syndrome.

Trichoepithelioma has a tendency to reoccur and leads to occasional transformation to high- grade carcinomas such as BCC and Trichoblastic carcinoma (trichoblastic carcinoma is very rare and poorly understood with potentially aggressive behaviour) and mixed epithelial or sarcomatous tumors. And such malignant transformations have different treatment modalities than benign lesion. Hence, in present scenario making proper clinical diagnosis is important to prevent further complications.

Solitary trichoepitheliomas can be best treated by surgical excision of the lesion. In the case of multiple tumours, definitive surgery is impractical and cannot predict the emergence and growth of new carcinoma cells. As a temporary cosmetic procedure, curettage, dermabrasion or electrocautery can be used to reduce tumor volume and improve its shape.[4] Complications of surgical treatment include lesion recurrence, pain, bleeding and scarring.

There are some effective non pharmacological treatments that include carbon di oxide, erbium: YAG and diode laser.

Certain pharmacological treatments are also there which are cost effective as well as easy to use as primary treatment such as Srolimus 0.2% gel, rapamycin 1% cream, imiquimod 5% cream, Vismodegib 150mg.

III. CONCLUSION

The case highlights the importance of differential diagnosis that not every lesion is mucocele (trauma). They can also be diagnosed as Basal cell carcinoma or trichoepithelioma. Therefore, proper clinical diagnosis and treatment planning is very important in such case while keeping in mind about malignant transformation in recurrence of the lesion. However, Trichoepithelioma are more common in females thus we present a unique and rare case of trichoepithelioma in adult male patient.

REFERENCES

[1]. Yiltok et al. Multiple familial trichoepithelioma: a case report and review of literature. Nigerian Journal of Clinical Practice :June 2010 Vol. 13(2):230-232
[2]. Bedi T.R., L.K. Bhutani, Kandhari K.C. trichoepithelioma. Indian journal of dermatology and venereology.1970.
[3]. Arthur Book et al(Ed 1968), Text Book of Dermatology
[4]. J. D. Osguthorpe and t. C. Calcaterra. Trichoepithelioma: report of an unusual case and review of the literature. Head & neck surgery 1~461-464 1979
[5]. Rahman Jawaria et al. Desmoplastic Trichoepithelioma:Histopathologic and Immunohistochemical Criteria for Differentiation of a Rare Benign Hair Follicle Tumor From Other Cutaneous Adnexal tumors.Cureus.2020Aug;12(8): e9703