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# Tumor of follicular infundibulum in beard area: A case report and literature review

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**Abstract**
**Background:** Tumor of follicular infundibulum (TFI) is an uncommon, benign adnexal neoplasm without distinctive clinical features. It usually presents as a solitary lesion on the head, neck, or trunk. Histologically, it demonstrates a plate-like epithelial proliferation with thin epidermal connections. Although benign, TFI has clinical importance due to associations with basal cell carcinoma and Cowden's syndrome.
**Case Presentation:** We report a case of TFI in a 42-year-old male with multiple hypopigmented lesions over the beard area. Histopathology revealed a well-circumscribed lesion with epithelial strands arranged parallel to the epidermis, peripheral palisading of basaloid cells, and focal glycogenation without atypia. Immunohistochemistry was negative for BCL-2 and Ber-EP4, with a Ki-67 index <2%, helping to exclude basal cell carcinoma.
**Conclusion:** Recognition of TFI is important for appropriate management and for excluding malignant mimics. Surgical excision remains curative.

**Keywords:** Tumor of follicular infundibulum, infundibuloma, basal cell carcinoma, hypopigmented lesions

## Introduction
Tumor of follicular infundibulum (TFI), also referred to as infundibuloma, is a rare benign cutaneous neoplasm of follicular differentiation, first described by Mehregan and Butler in 1961 [1]. It demonstrates differentiation towards the follicular isthmus/outer root sheath and is occasionally termed isthmicoma.
Clinically, TFI usually appears as a solitary hypopigmented macule, papule, plaque, or nodule, most often <1 cm in diameter, and may mimic a scar or atrophic lesion [2]. Pigmented and scaly presentations have been reported, although less common [3]. The condition is more frequent in middle-aged and elderly women, predominantly involving the head, neck, and trunk [4]. Rare associations include Cowden syndrome, nevus sebaceous, and Schopf-Schulz-Passarge syndrome [5].
Histologically, TFI is characterised by a plate-like horizontal proliferation of pale-staining squamous epithelial cells, with interconnecting strands forming a reticulated or fenestrated pattern. Keratocysts at the lesion base and an eosinophilic basement membrane surrounding tumor islands are frequent findings [6].
We report a case of multiple TFI lesions over the beard area in a middle-aged male, with emphasis on clinicopathological features and differential diagnosis.

## Case Report
A 42-year-old male patient presented with progressively persistent, hypopigmented, raised, and irregularly shaped lesions of varying sizes (1.5 x 0.5 cm), symmetrically distributed over the beard area. The lesions were predominantly asymptomatic, accompanied by occasional itching, and did not exhibit surface changes such as scaling or atrophy upon examination. A subsequent biopsy, followed by histopathological analysis, revealed a well-circumscribed lesion characterised by tumour-forming strands oriented parallel to the epidermis. The findings included peripheral palisading of basaloid cells and mild lymphocytic infiltrate surrounding and infiltrating the tumor. Furthermore, certain epithelial cells displayed signs of glycogenation without evidence of atypia.

Immunohistochemical analysis using markers such as BCL-2, Ber-EP4, CD10, Ki-67, and CK20 yielded negative results for both BCL-2 and Ber-EP4, while the Ki-67 index was determined to be less than 2%, effectively ruling out the diagnosis of basal cell carcinoma.

**Discussion**

Follicular infundibular tumors are relatively rare, comprising only 0.02% of all skin biopsies. This account pertains to the multiple or eruptive form of TFI, characterised by asymptomatic, gradually progressive, hypopigmented macules symmetrically distributed in the patient's beard area and accompanied by a history of itching. Typically, the lesions manifest as macular forms, although they may also appear as papules or depressed lesions. Instances of pigmented maculopapular and scaly reticulated patches are uncommon presentations of these lesions. Notably, the histopathological differential diagnoses encompass basal cell carcinoma, clear cell seborrheic keratosis, and eccrine syringofibroadenoma [7].

The aetiology of this neoplasm remains a subject of contention. Few researchers postulate that TFI (trichilemmal carcinoma with fibroplasia) may represent an epidermal reactive process to underlying dermal fibrosis. While the prevailing architectural and histopathological characteristics point towards an infundibuloma, contemplation of a collision tumor is warranted [8]. The presence of numerous keratocysts at the base of the lesion raises the prospect of a collision with a trichoadenoma, a tumor demonstrating lineage towards the infundibular segment of the hair follicle. Notably, the absence of a granular layer, as anticipated in keratocysts or trichoadenoma, and the observation of an abrupt type of keratinisation challenge this consideration. In addition to infundibuloma, the histopathological differential diagnosis may encompass a superficial variant of basal cell carcinoma; Weyers *et al.* propose TFI as belonging within the spectrum of basal cell carcinoma (BCC) and possibly progressing to a more aggressive variant of BCC. Furthermore, fibroepithelioma of Pinkus may mimic the fenestrated pattern of TFI; however, the latter lacks the distinctive budding of basaloid cells along the interconnected epithelial strands, which is indicative of the germinal differentiation of the former entity, despite the shared fibrotic stroma. Basaloid follicular hamartoma also warrants consideration [9].

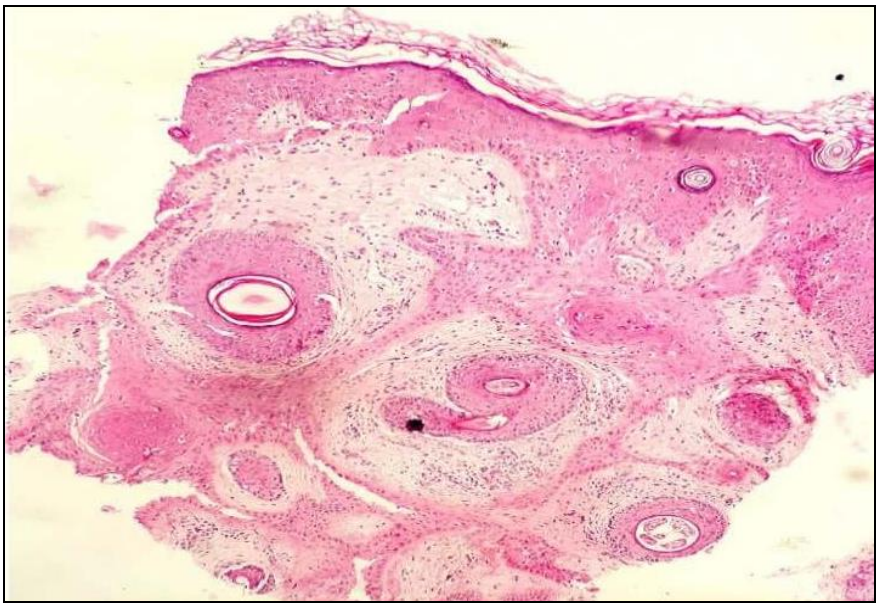

**Fig 1:** A well-circumscribed lesion with tumour-forming anastomosing strands (H & E, 10x)

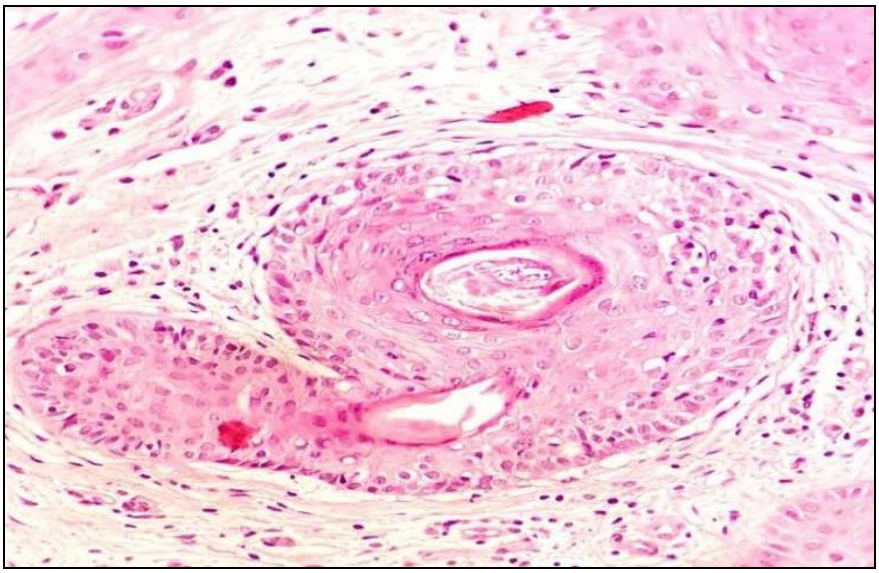

**Fig 2:** There is peripheral palisading of basaloid cells. (H & E, 40x)

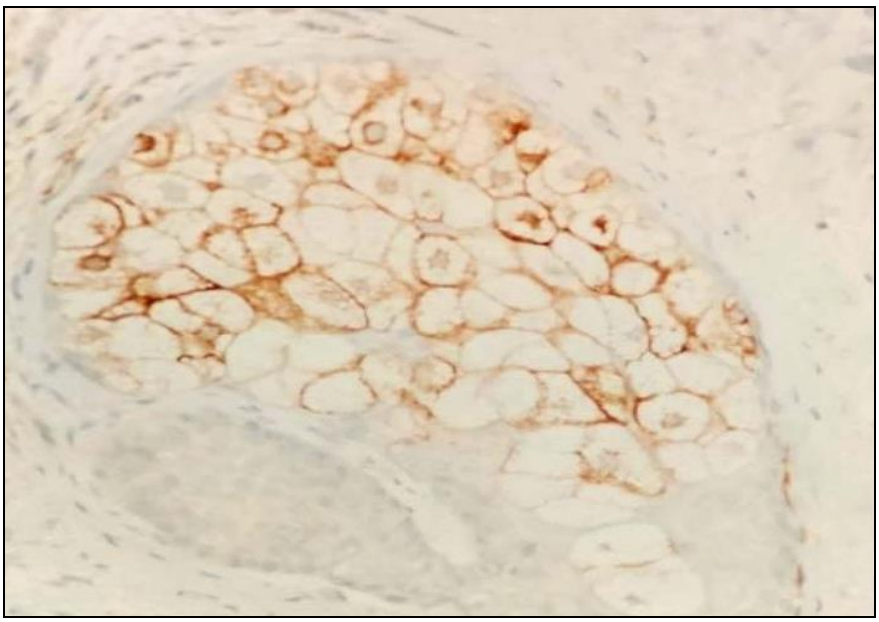

**Fig 3:** Differentiated keratinocytes highlighted by CD 10 immunohistochemistry (10x)

## Conclusion

Please be aware of the following information: Lesions described as basal cell hamartoma with follicular differentiation may be considered a variant of infundibuloma, as they exhibit structures similar to hair follicle papillae, akin to those seen in a documented case of typical solitary trichofolliculoma [6]. Histologically, this lesion can be distinguished from an adenoid type of seborrheic keratosis by its reticulated pattern of interconnecting epithelial strands and the presence of horn pseudocysts. Clinically, the differential diagnosis for this rare tumor in this location may include lichen planus, lichen sclerosus, lichen simplex chronicus, and pemphigus. Surgical excision remains the definitive treatment for this benign lesion. Notably, this tumor is underrepresented in the literature, likely due to its rarity and benign nature.

**Conflicts of interest:** There are no conflicts of interest.
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