Tumor of Follicular Infundibulum; Eruptive Variant: A Case Report

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ABSTRACT

Tumor of follicular infundibulum (TFI) is a rare benign follicular tumor typically presenting as a solitary macule or papule on the head and neck area. Our case presented with multiple hypopigmented macules as an eruptive variant that should be differentiated from common skin lesions such as pityriasis versicolor, pityriasis alba and vitiligo. Histopathological study was necessary for definite diagnosis, which revealed horizontal plate – like proliferation of small monomorphic and pale staining epithelium with multiple slender connections with surface epidermis or upper part of hair follicles. TFI is chronic, persistent, and benign in nature, so the treatment should be conservative.

Keywords: Tumor of follicular infundibulum, isthmicoma, hypopigmented macule

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CASE REPORT

A 58 -year - old Thai male from Khon Kaen province presented with a 5 -year history of multiple asymptomatic hypopigmented macules on his lateral aspect of both cheeks and neck area. He denied the history of previous trauma, inflammation or erythematous rash. His past medical history was unremarkable. He did not take any medications. He was treated by a dermatologist with liquid nitrogen and carbon dioxide laser. The lesions showed some improvement, but still slowly increased in number.

One year ago, the patient noticed that those hypopigmented macules were more visible and gradually extended to the forehead, prompting him to come to Siriraj Hospital. Physical examination revealed multiple discrete irregular well - defined hypopigmented macules and patches on beard area, lateral aspect of cheeks, forehead and neck. (Fig 1) The clinical differential diagnoses were pityriasis versicolor, pityriasis alba and vitiligo. The potassium hydroxide mycological examination was done which yielded negative result. He was treated with 3% LCD and 0.1% triamcinolone cream for 1 month without any improvement. Then the skin biopsy was done.

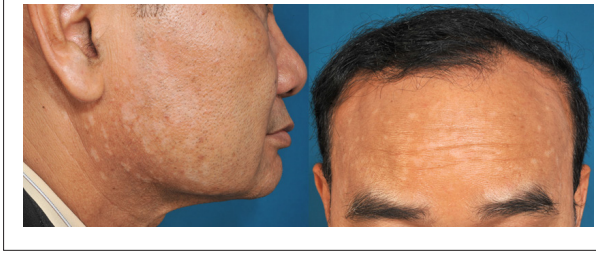

Fig 1. Multiple discrete irregular well - defined hypopigmented macules and patches on beard area, lateral aspect of cheeks and forehead.

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The histopathological study showed focal area of hyperplastic epidermis with elongated irregular shaped rete ridges, and focal area of horizontal epithelial plate connected to the overlying epidermis with multiple slender strands. These epithelial proliferations were composed of pale staining keratinocytes. (Fig 2A, 2B)

Melan - A staining showed the decrease of melanin and melanocytes at the base of the expanded rete ridges and epithelial plates. In contrast, in the area of normal epidermis, melanin and melanocytes were preserved. (Fig 3)

DISCUSSION

Tumor of follicular infundibulum (TFI) is a rare benign follicular tumor which was initially described by Mehregan and Butler in 1961.[1] TFI which was also known as isthmicoma, was proposed by Mc Calmont to highlight isthmic differentiation of tumor cells.[2] However, tumors originate from the region of follicular infundibulum, rather than from the isthmus part in all reported cases.

The reported frequency was 3 to 17 per 100,000 skin biopsy specimens.[1,3,4] It remains unclear whether the disease has male or female predominance. Most cases present as an asymptomatic solitary smooth or slightly scaly papule or nodule commonly located on head and neck area. TFI is probably under diagnosed because it might be clinically diagnosed as seborrheic keratosis, especially in elderly patients.[3,5] There are some less common clinical variations including an eruptive variant, in association with other diseases such as Cowden's disease, in association

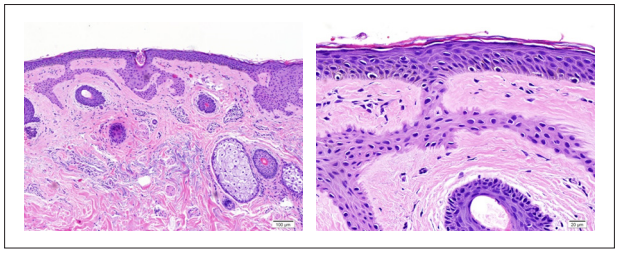

Fig 2A & 2B. Focal area of hyperplasic epidermis and horizontal plate-like proliferation of monomorphic pale staining epithelium with slender strand connected to the overlying epidermis (H&E) power 10X, 40X.

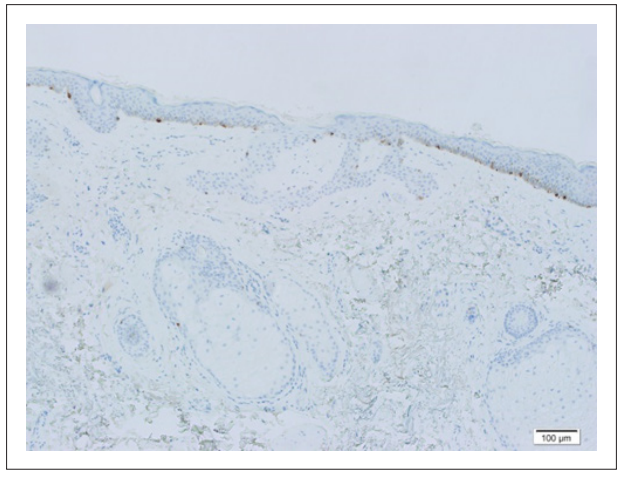

Fig 3. A decrease of melanin and melanocytes in the tumor area compares with normal epidermis. (Melan-A) power 40X.

with hamartoma such as nevus sebaceous and TFI-like reactive epidermal change.[4]

The clinical appearance of an eruptive variant consists of multiple asymptomatic well – demarcated hypopigmented macules, similar in size, located on face, neck and upper trunk. Lesions gradually increase in number. Sun exposure can make them more visible and aggravate pruritus in some cases.[3,4] Due to the rarity of the disease, it should be differentiated from other common skin diseases including pityriasis versicolor, pityriasis alba, vitiligo, post inflammatory hypopigmentation and idiopathic guttate hypomelanosis. Our patient clinically presented as eruptive variant, so histopathological study was done for definite diagnosis.

The histopathological features of TFI have some differences among clinical variants. However, our case showed the classic histopathological features, which consist of a horizontal plate – like proliferation of small monomorphic nuclei and pale staining epithelium with multiple slender connections with surface epidermis or upper part of hair follicles. Thin columns of the cells are interconnected. Occasionally, some cases show small horn microcysts with keratohyaline granules and/or small tubular structures resembling sebaceous ducts within the lesions. Some reports show an increase of elastic fibers surrounding the tumor with an Orcein (elastic) stain.[3,4,6] The only difference between solitary and eruptive TFI is the decrease of epidermal melanin in the tumor area

of the eruptive variant, which can be correlated with clinical hypopigmentation.[3,7]

TFI is generally considered as a chronic and benign adnexal tumor. Eruptive TFI is not inherited, or associated with other systemic disease or malignancy.[8] Transformation to basal cell carcinoma has been reported in one case of a solitary lesion.[9] Treatments of TFI include topical corticosteroids, keratolytic agents, retinoic acid and imiquimod or excision. There was a single reported case of recurrence occurring 10 months after the primary lesion was excised.[4] For multiple lesions, cryotherapy and ablative laser should be considered.[10] In our patient, he was previously treated with liquid nitrogen and carbon dioxide laser, but the result was unsatisfactory. According to the reasons mentioned above, the patient should be informed of that fact and regular clinical follow up is recommended.

To our knowledge, hypopigmented macule is one of the common clinical presentations found in our practices. If the patient does not respond to the treatment recommended based on the original diagnosis, TFI should be considered, especially in patients with hypopigmented lesions on head and neck area. Histopathological study is a key to establish the diagnosis.

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