

Basaloid follicular hamartoma: clinical, dermoscopic, and histopathological characteristics of case

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Abstract

Basaloid follicular hamartoma (BFH) is a rare benign adnexal tumor with variable clinical presentation. We report a case of a 64-year-old man, who presented with an incidental finding of a 3mm hyperpigmented macule on his cheek. Dermoscopy revealed a structureless blue lesion. Histopathology examination showed interconnecting lobules and cords of bland pigmented epithelial cells within the dermal stroma, with the presence of pseudohorn cysts. The lesional cells were faintly positive for Bcl2 on immunohistochemical staining. These findings were consistent with basaloid follicular hamartoma. Histological differential diagnoses include benign lesions such as trichoepithelioma, and malignant lesions such as basal cell carcinoma (BCC).

Keywords: basaloid follicular hamartoma; benign adnexal tumor

Introduction

Basaloid follicular hamartoma (BFH) is a benign adnexal tumor, with variable clinical appearance in the form of papules, nodules, or plaques, which may be skin-colored to brown in color. They can present as solitary lesions, or in a generalized form associated with systemic disease [1-4]. The diagnosis is made on histopathological examination.

Case Synopsis

A 64-year-old man presented with an incidental finding of a brownish macule on his right cheek of unknown duration. It was asymptomatic and

had not changed in appearance for the past several years. He had a past medical history of hypertension, hyperlipidemia, and atrial fibrillation on anticoagulation. There was no personal or family history of malignancy, autoimmune disorders, or similar skin lesions.

Physical examination revealed a 3mm linear hyperpigmented macule. There were no constitutional symptoms and systemic examination was unremarkable (**Figure 1**).



Figure 1. Hyperpigmented macule over cheek

Dermoscopy findings revealed a structureless bluish macule, suggestive of a tattoo (**Figure 2**). The patient denied any history of tattoo, deliberate or traumatic. A dysplastic melanocytic nevus was considered. An excisional punch biopsy was performed.

Microscopically on histopathological examination,

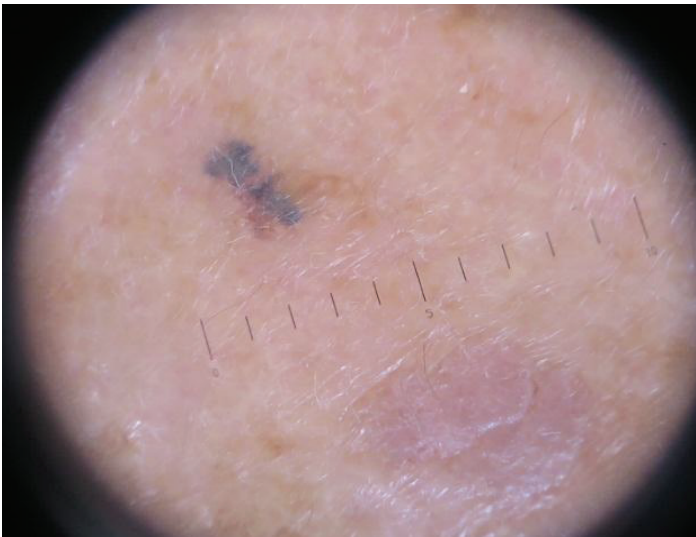


Figure 2. Dermoscopy showing structureless blue macule

the lesion was characterised by small interconnecting lobules and cords of bland epithelial cells, lying within sclerotic dermal stroma. The lesional cells appeared heavily pigmented. Mitoses were scarce. There was no peripheral palisading noted (**Figures 3, 4**).

The overlying epidermis showed mild acanthosis with pseudohorn cyst formation. There were also mild dermal solar elastosis and melanophages.

Immunohistochemical staining of lesional cells was negative for melan-A and EMA. The lesional cells showed weak positivity for Bcl2 only within the outermost cells.

The features were those of a benign adnexal tumor, most consistent with basaloid follicular hamartoma.

Case Discussion

The lesional cells in BFH are folliculocentric and limited primarily to the superficial dermis. Hair follicles are distorted, with branching cords of basaloid cells bridging the central pilosebaceous structures. As in our case, tumor cells have bland nuclei and show only rare mitoses.

The primary histological entity in the differential diagnosis to consider is that of a basal cell carcinoma (BCC). Infundibulocystic BCC (IFBCC) in particular may appear microscopically very similar to BFH. Although both consist of cords or basaloid cells in a fibrous stroma, IFBCC is not folliculocentric and may be seen in the interfollicular dermis. IFBCC may also

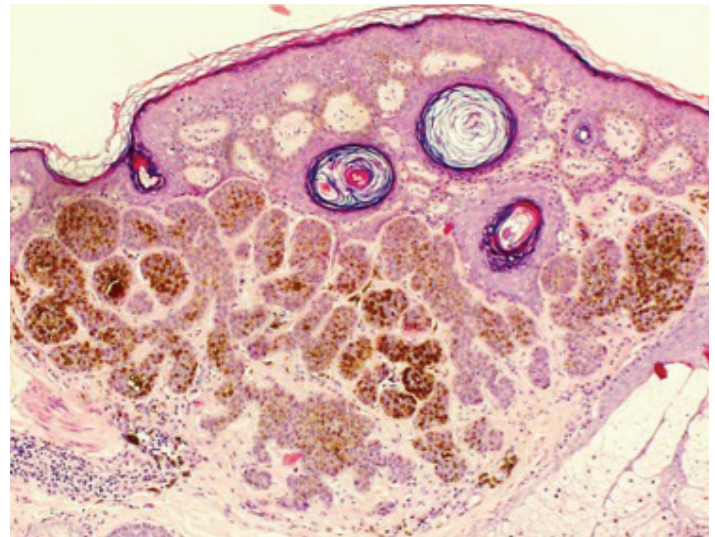


Figure 3. Pseudohorn cyst formation in the epidermis, with interconnecting small lobules of pigmented cells in close association with the overlying epidermis. (H&E, 40x).

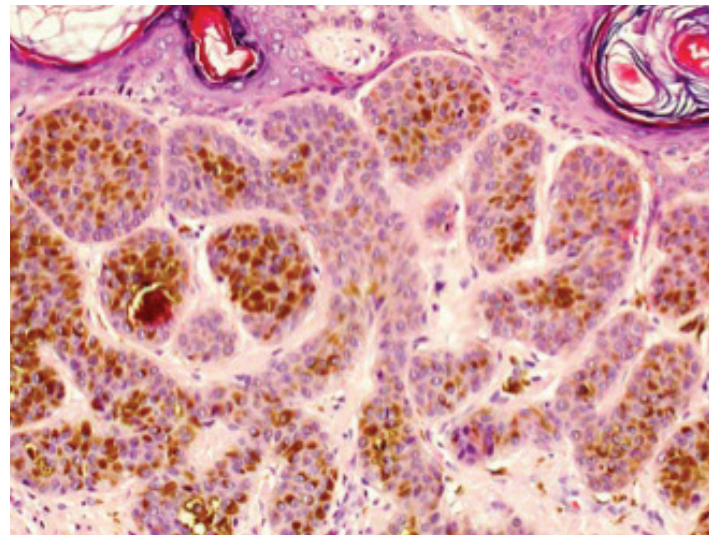


Figure 4. Heavily pigmented lesional cells with scarce mitoses. (H&E, 100x).

exhibit deep infiltration and ulceration [5].

Fibroepithelioma of Pinkus is another variant of BCC characterized by arborizing cords of basaloid cells arising from the epidermis. Microscopically, eccrine ducts and prominent fibrovascular stroma may be seen in fibroepithelioma of Pinkus [1, 3]. Peripheral palisading is characteristic of most variants of BCC but is usually less pronounced in BFH [1].

An important benign tumor in the differential diagnosis for BFH is that of trichoepithelioma. Compared to BFH, trichoepitheliomas tend to be larger in size and display a more nodular growth

pattern. Islands of basaloid cells are arranged in a lacelike pattern, with a more prominent cellular stroma and normal follicular bulb and papillae in trichoepitheliomas [1, 6]. The presence of papillary mesenchymal bodies and the absence of connection with the epidermis are other features more typical of trichoepitheliomas.

Folliculocentric basaloid proliferation (FBP) is a reactive lesion originating in clinically normal skin, often occurring adjacent to a BCC. It also consists of folliculocentric basaloid aggregates. However, there are no keratin cysts and no direct epidermal attachment unlike that in BFH [7].

Immunohistochemical staining can help in differentiating between BCC and BFH, but cannot distinguish between BFH and trichoepithelioma. Bcl2 is uniformly positive in BCC, but typically only reactive in the outermost tumor cells of BFH and trichoepithelioma. Other IHC markers that can be used include CD34, which is highlighted in the stromal cells of BFH and trichoepithelioma but not that of BCC [8-10].

The clinical presentation of BFH is diverse. Solitary BFH lesions are usually acquired, whereas multiple lesions may be hereditary without associated disorder, or associated with systemic manifestations, such as palmoplantar pitting, alopecia, and myasthenia gravis [1, 3, 11, 12]. There is also a risk of progression to BCC [3, 4]. Most case reports describe lesions as papules, predominantly skin to flesh colored. Our case is interesting in that it is the first to be described as a macule and is more heavily pigmented than other BFH lesions described. Despite the varied appearance and context, the histological features are similar.

Conclusion

Basaloid follicular hamartoma has a varied clinical presentation. In our patient, it resembled that of a dysplastic melanocytic nevus. However, the histological features, with the presence of both horn cysts and basaloid lobules of cells, bring to mind a number of other benign tumors in the differential diagnoses. This rare presentation of an uncommon tumor increased the diagnostic challenge involved in this case.

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