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Case Report

Localized basaloid follicular hamartoma: A case report

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ABSTRACT

Basaloid follicular hamartoma (BFH) is a rare benign follicular tumor with varied clinical presentation. A 64-year-old male presented with a gradually enlarging plaque on the scalp of 10 years duration, the surface of which showed a unique cobblestone like appearance and dilated follicular openings discharging keratinous material. Histology was initially reported as trichofolliculoma. As the clinical picture was not suggestive of trichofolliculoma, the histopathology was reviewed. A final diagnosis of BFH was made based on the characteristic features such as distorted hair follicles with peripheral extensions of basaloid cells in a branching and anastomosing pattern. A periodic follow-up of such lesions is essential due to the risk of malignant transformation to basal cell carcinoma.

Keywords: Basaloid follicular hamartoma, Cobblestone, Scalp

INTRODUCTION

Basaloid follicular hamartoma (BFH), a rare hair follicle tumor that occurs on the face, scalp, and occasionally the trunk, is characterized histologically by lace like network of branching cords and strands of basaloid cells with follicular differentiation.[1] Clinically the localized forms can be mistaken for seborrheic keratosis, nevus sebaceus, appendageal tumors, or basal cell carcinoma (BCC). We report an acquired localized form of BFH on the scalp.

CASE REPORT

A 64-year-old male presented with a gradually enlarging asymptomatic plaque on the scalp of 10 years duration. There was no history of ulceration, oozing, or bleeding from the lesion. He did not have similar lesions elsewhere on his body. There was no history of similar skin lesions in the family members. On examination, there was a well defined hyperpigmented firm plaque on the vertex, of size 5 cm × 3 cm with the surface showing cobblestone like appearance and patulous follicular openings [Figure 1]. A dark keratinous material was expressed from the dilated follicular openings. There were no other cutaneous or mucosal lesions. The systemic examination was normal. Differential diagnoses of seborrheic keratosis, organoid nevus, and appendageal tumor were considered.

Hemogram, urine analysis, liver function tests, and renal function tests were normal. Histology was initially reported as trichofolliculoma. As the clinical picture was not suggestive of trichofolliculoma, the histopathology was reviewed. A final diagnosis of BFH was made based on the presence of distorted hair follicles with peripheral extensions of basaloid cells in a branching and anastomosing pattern in the mid and lower dermis [Figures 2a and b]. There were a few horn cysts and inflammatory infiltrate in the periphery of the lesion. There was no continuity with the surface epidermis.

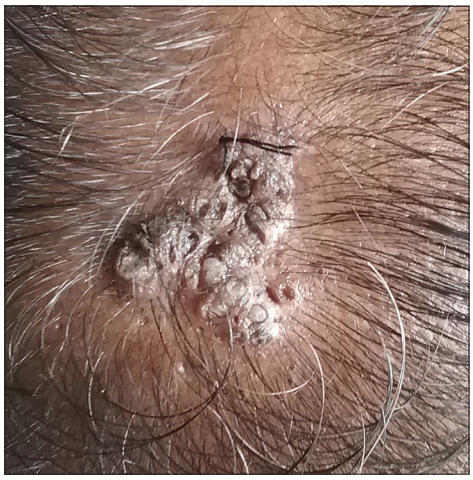

Figure 1: Hyperpigmented plaque with the surface showing a cobblestone like appearance and patulous follicular openings.

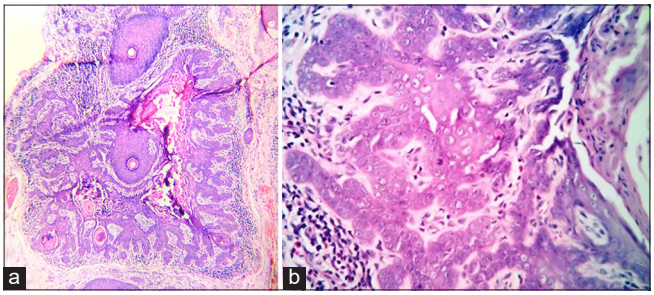

Figure 2: (a) Distorted hair follicles with peripheral extensions of basaloid cells in a branching and anastomosing pattern (H and E, x100). (b) Basaloid cells in a branching and anastomosing pattern (H and E, x400).

DISCUSSION

BFH is a rare, benign, superficial malformation of hair follicles first described in 1969 by Brown *et al*., in association with myasthenia gravis and diffuse alopecia (Brown-Crounse syndrome).[2] In 1985, the term BFH was coined by Mehregan and Baker, who reported a localized and solitary type of lesion without associated abnormalities.[3] The formation of BFH has been linked to a mutation in the patched gene (PTCH) on chromosome band 9q23 which is part of the same pathway implicated in nevoid BCC syndrome.[1,4]

BFH can be hereditary or acquired. The hereditary BHF may be generalized or unilateral (nevoid). The acquired type presents as localized or solitary forms. The cutaneous lesions may be macules, papules, plaques, or nodules in generalized, localized, or linear distribution.[5] Comedo-like and milia-like papules have been reported previously.[4]

The congenital and familial forms usually occur in association with other genodermatoses or systemic diseases such as systemic lupus erythematosus or cystic fibrosis.[6,7]

The localized forms of BFH can a present as solitary papules or plaques or linear unilateral lesions along the lines of Blaschko. Plaques with alopecia can often be mistaken for lesions of nevus sebaceus, seborrheic keratosis, lupus erythematosus, or sarcoidosis. Similar to most of the previous reports, a diagnosis of localized form of BFH was established only after histopathological examination.[1,8]

Solitary localized form of BFH was first described in 1992 as a smooth plaque or a papule appearing most commonly on the face or scalp. The localized types usually are not associated with systemic diseases. The histopathology of BFH is usually limited to the superficial and mid dermis.[9] BFH can mimic trichoepithelioma (TE), trichofolliculoma or infundibulocystic BCC histologically.[8] The main differentiating features are malformed and distorted hair follicles in BFH as opposed to normal follicular units in TE. The fibrocytic stroma is more prominent in TE than in BFH.[1,8] Trichofolliculoma characteristically shows dilated cystic follicle that communicates with the skin surface. The follicle is lined by squamous epithelium and surrounded by numerous secondary follicles, many of which contained a hair shaft.[10] In our case, BCC was excluded by the absence of cytologic atypia, mitosis, and artifactual separation between tumor cells and surrounding stroma. However, rare case reports of development of BCC over BFH suggest periodic follow-up in such cases.[11]

CONCLUSION

We report this case to highlight the unique clinical presentation of localized type of BFH with a cobble stone like appearance and dilated follicular openings extruding keratinous material. Furthermore, BFH needs to be considered in the clinical differential diagnosis of asymptomatic plaques on the scalp.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

Dr. Betsy Ambooken and Dr. N. Asokan are on the Editorial Board of the Journal.

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