Dermoscopy of Linear Basaloid Follicular Hamartoma

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A 25-year-old male presented with asymptomatic tiny eruptions in a linear pattern on the chest since seven years. Examination revealed multiple skin-colored grouped follicular papules in a band-like pattern extending downwards from the right sternoclavicular joint till the left sternal margin at the level of the areola [Figures 1 and 2]. There was no family history of such lesions and the rest of the cutaneous and systemic examination was unremarkable. Clinical differential diagnoses of segmental localized forms of trichoepithelioma and hair follicle nevus were considered. Dermoscopy using DermLite™ DL3 (3Gen, San Juan Capistrano, CA, USA) under polarized mode revealed whitish globules of varying sizes with some exhibiting brownish clods or smaller globules and one to two fine hairs in the centre. The intervening area showed dense brown pigment network and eccrine openings. The latter were surrounded by the dense pigment network and a few by pigmentation lines [Figure 3]. Histopathologically, clusters and strands of basaloid cells in relation to a dilated hair follicle were seen in the upper dermis. The clusters were separated from one another by fibrous stroma. The overlying epidermis showed increased melanization of the basal layer [Figure 4]. Localized linear basaloid follicular hamartoma was diagnosed based on the clinical and histological findings.

Basaloid follicular hamartoma (BFH) is a rare benign follicular tumor due to mutation in the *PTCH* gene bearing close resemblance to infundibulocystic basal cell carcinoma (BCC) histologically. It occurs in varied clinical forms – inherited or sporadic, congenital or acquired, and solitary or multiple [Table 1]. Multiple lesions can be generalized or localized (linear, unilateral and nevoid forms). Clinical lesions may be comprised of papules, plaques, or patches of alopecia frequently involving the scalp and face.[1] The linear form of the disorder usually presents at birth or develops in early childhood. Occurrence at a later age, as in our case, is also described.[1,2] The lesions are characterized by multiple skin-colored to pigmented follicular papules or plaques

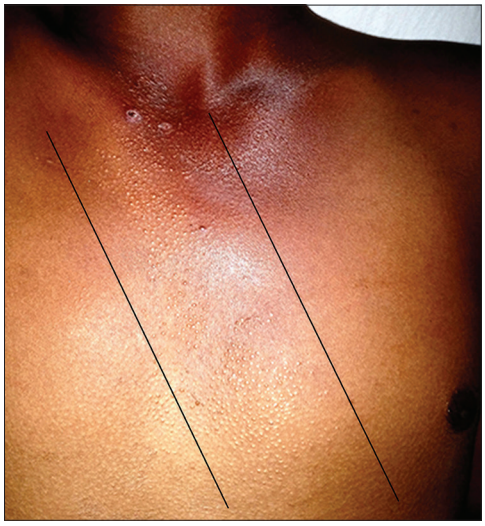

Figure 1: Grouped skin-colored follicular papules in a band-like configuration extending downwards from the right sternoclavicular joint till the left sternal margin at the level of areola

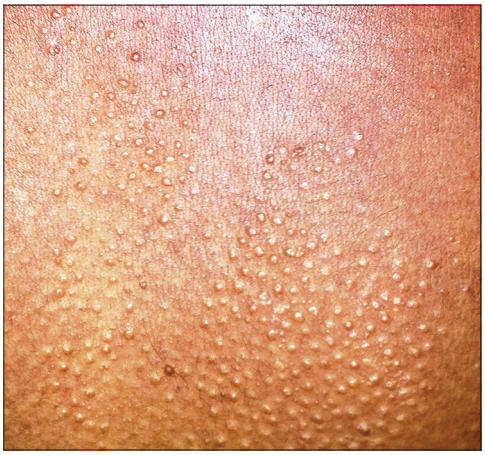

Figure 2: Close-up view of grouped skin-colored papules at the level of the left areola

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in a band-like configuration along the Blaschko's lines frequently involving the face, trunk, and extremities. Linear BFH generally occurs in isolation although association with various disorders [Table 1] and development of BCC in the lesions have been described as well.[2,3] Clinical differential diagnoses of linear BFH include unilateral nevoid BCC syndrome, linear forms of epidermal and eccrine nevi, and Blaschkoid variants of trichoepithelioma, eccrine spiradenoma, eccrine poroma, and syringoma.[2]

All forms of BFH microscopically demonstrate multiple clusters, branching cords and lace-like network of bland, and uniform basaloid epithelial cells separated by fibrous stroma in the papillary dermis in relation to a dilated hair follicle.[4] Histopathologically, BFH bears quite a resemblance with BCC, and hence, immunohistochemistry may be essential in unequivocally differentiating the two.[5]

In dermoscopic analysis of our case, the whitish globules correspond to the individual papules clinically and to the dilated hair follicles histologically. The brownish clods within whitish globules represent the proliferation of basaloid cells in a fibrous stroma in relation to the dilated follicles. The intervening dense pigment network is attributable to the patient's skin color corresponding histologically to the increased basal layer melanization. Dermoscopic characterization of BFH with similar clinical and morphological attributes to our case is not available in the literature to enable comparison or assertion of our findings. Although dermoscopic features of a solitary BFH has been documented, they represent that of a different morphological type.[6] Our dermoscopic observations are preliminary (in Fitzpatrick type V skin) and further studies are required to delineate any reproducible features.

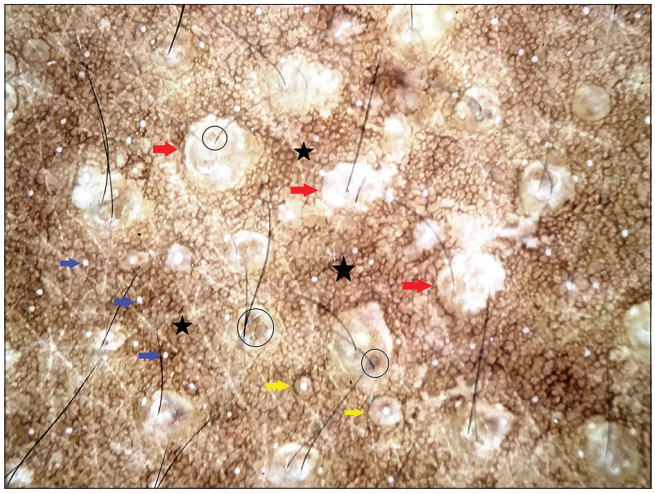

Figure 3: Non-contact dermoscopy under polarized mode using DermLite™ DL3 showing multiple whitish globules with one to two fine hairs in the centre (red arrows). Some of these globules also show central brownish clods (black circles). Dense brown pigment network (black stars) and eccrine openings are seen in the intervening area. The eccrine openings are surrounded by the pigment network (blue arrows) and a few of them by pigmentation lines (yellow arrows). [Original magnification x10]

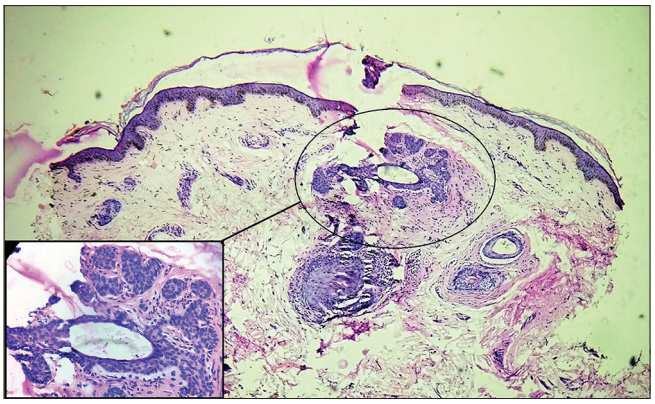

Figure 4: Photomicrograph showing a folliculocentric lesion (black circle) in the upper dermis. Islands of proliferated basaloid cells separated by stroma are seen around a dilated hair follicle. In set: Higher magnification showing clusters and branching strands of basaloid cells with an intervening organized fibrous stroma in relation to a dilated hair follicle. [Hematoxylin and Eosin, Original magnification x5 (scanner view) and x40 (in set)]

Table 1: Classification of multiple basaloid follicular hamartoma

Type	Features
Inherited	
Familial multiple BFH	Inherited as an autosomal dominant disorder characterized by multiple lesions on face, trunk and extremities
Generalized BFH syndrome	Inherited autosomal dominantly due to mutation in PTCH gene. Manifestations include generalized congenital or childhood onsed multiple BFH with multiple milia, comedo-like lesions, hypotrichosis, hypohidrosis, and palmar and/or plantar pits
Congenital	
Congenital generalized BFH	Generalized BFH associated with alopecia and cystic fibrosis
Acquired	
Linear unilateral BFH	Multiple grouped lesions in a band-like fashion along the lines of Blaschko. This form of BFH has been described with extensive trichoblastoma, Graves' disease, congenital dorsal scoliosis and Happle-Tinschert syndrome
Generalized BFH with associated anomalies	Acquired generalized BFH may be associated with myasthenia gravis, alopecia and systemic lupus erythmatosus
Generalized BFH without associated anomalies	Multiple acquired BFH rarely occur sporadically

BFH = Basaloid follicular hamartoma

Dermoscopically, other benign hair follicle tumors such as trichoepithelioma and trichoadenoma show somewhat identical features with multiple circumscribed white structures (as clods and milia-like cysts), and hence, clinical and histological correlation of dermoscopic findings is essential.[7] Considering the close resemblance to BCC, we believe dermoscopy could possibly be a useful adjunct in clinically differentiating BFH from BCC as there are defined dermoscopic criteria for the latter.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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