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Localized Grouped Basaloid Follicular Hamartoma on the Chest of an Adult

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Dear Editor:

A 30-year-old man presented with a 6-year history of multiple asymptomatic papules on the chest. He did not have any particular medical history. None of his family members complained of similar symptoms. Physical examination revealed multiple-grouped, skin-colored to erythematous papules near the left areola (Fig. 1). Skin biopsy was performed on one of the papules. Hematoxylin-eosin staining revealed basaloid cells forming cords and strands that were confined to the superficial dermis, with epidermal attachment and a horn cyst (Fig. 2A, B). In the dermis, the stroma was myxoid and mildly fibroblastic (Fig. 2B). There was neither nuclear pleomorphism nor cleft formation between the tumor and the stroma. Alcian blue staining was strongly positive in the stroma (Fig. 2C). On immunohistochemical study, staining for Ki-67 showed weak positivity (Fig. 2D), and Bcl-2 staining revealed positivity only in the outermost tumor cells (Fig. 2E). Moreover, stromal cells adjacent to the basaloid cells showed CD34 positivity (Fig. 2F). Accordingly, the patient was diagnosed with basaloid follicular hamartoma (BFH). After our recommendation to excise the lesion to avoid cosmetic problems, the patient decided to remain under observation.

BFH is a rare benign folliculocentric tumor with anastomosing cords and strands of the basaloid cells. Clinically, various forms have been reported that can be categorized as generalized, localized, and solitary types[1]. The generalized type can be further divided into sporadic form pre-

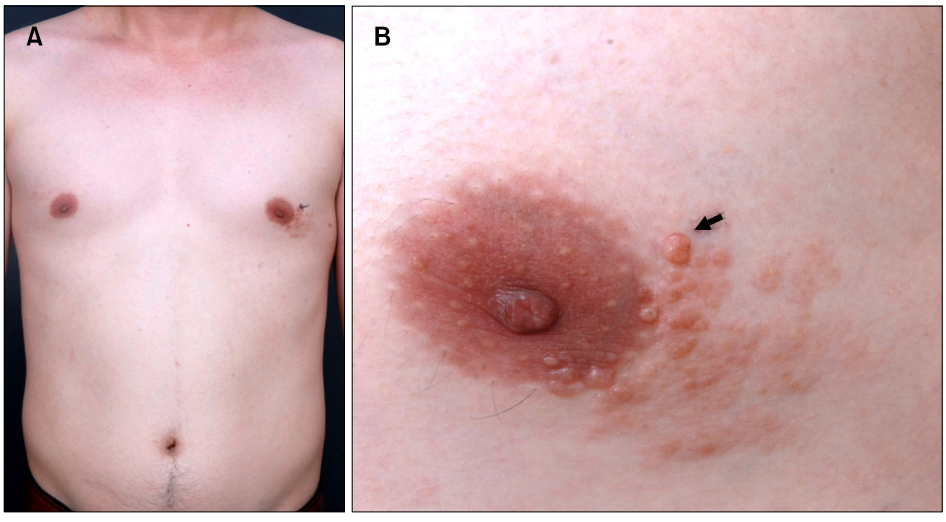

Fig. 1. (A) Multiple grouped skin-colored to erythematous papules near the left areola in a 30-year-old man. (B) Skin biopsy was performed on one of the papules (black arrow).

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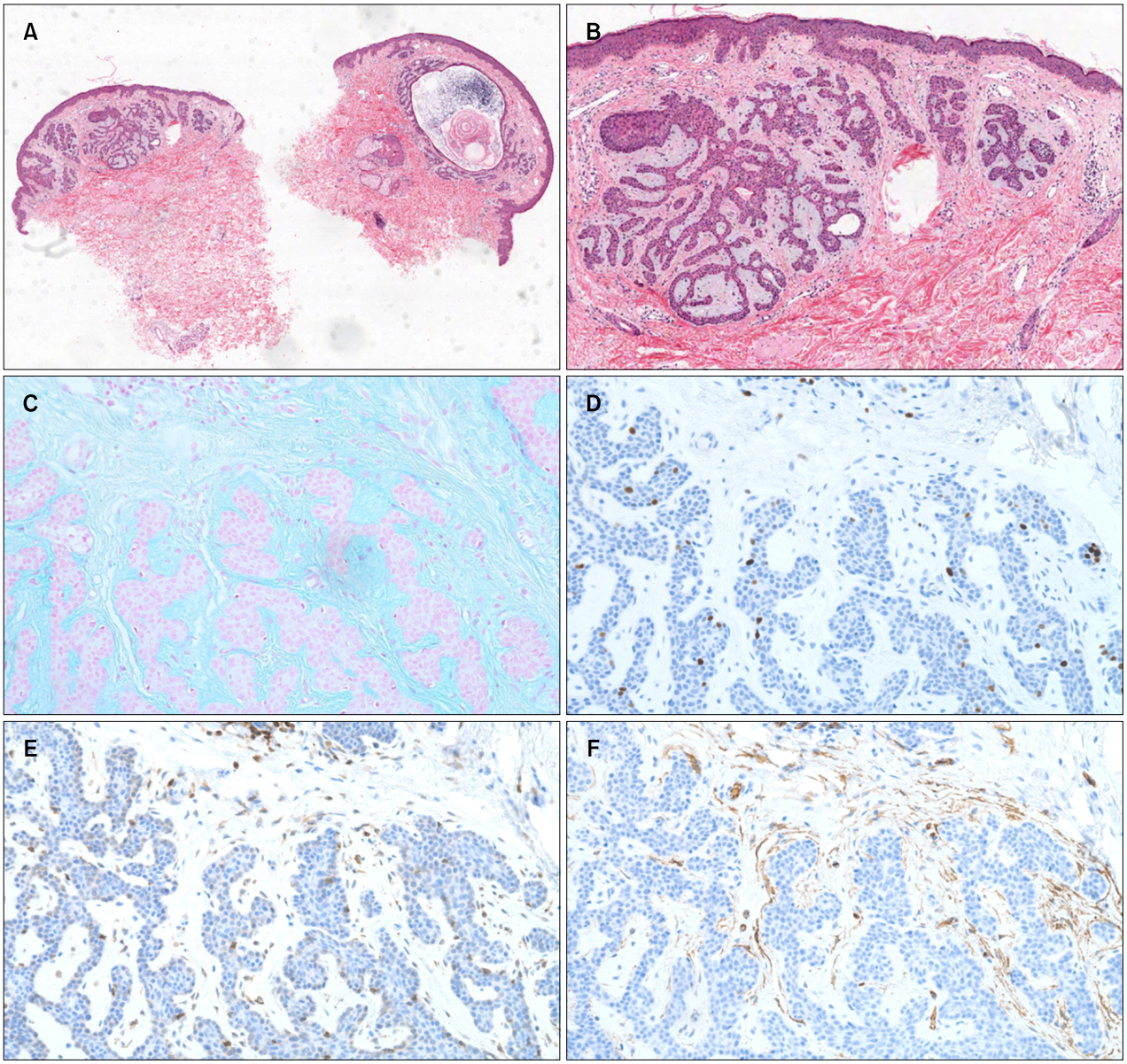

Fig. 2. (A, B) Hematoxylin-eosin staining revealing basaloid cells forming cords and strands confined to the superficial dermis, with myxoid stroma. Neither nuclear pleomorphism nor cleft formation between the tumor and the stroma was seen. (C) Staining for Alcian blue is strongly positive in the stroma. (D) Staining for Ki-67 is weakly positive. (E) Staining for Bcl-2 is positive only in the outermost tumor cells. (F) Staining for CD34 is positive in the stromal cells adjacent to the basaloid cells (all virtual slides).

senting multiple BFHs without systemic disease, acquired form associated with alopecia and autoimmune diseases, familial form inherited in an autosomal dominant manner, and congenital form associated with alopecia and cystic fibrosis. Localized forms present as linear unilateral lesions along the lines of Blaschko[2,3] or as plaques with alopecia[4]. Solitary forms appear as a smooth plaque or papule appearing most commonly on the face or scalp, with and without associated diseases. Presentation as multiple-grouped, skin-colored to erythematous papules on the chest as in this case has so far not been reported.

Histopathologically, on hematoxylin-eosin staining, thin anastomosing strands and branching cords of undifferentiated basaloid cells form distorted hair follicles within loose fibrous stroma. In addition, the follicles may or may not be connected to the epidermis, and peripheral palisading of basaloid cells can be present but less prominent than in basal cell carcinoma. Neither pleomorphism nor mitotic activity is seen[5]. On immunohistochemical staining, the outermost basal cells in BFH stain positive for

Bcl-2, and stromal cells next to tumor cells stain positive for CD34. Moreover, Ki-67, a proliferative marker associated with mitosis, shows relatively weak nuclear positivity[1]. These features help differentiate BFH from basal cell carcinoma, especially infundibulocystic type, in that this presents with deeper infiltration, strong nuclear positivity for Ki-67, negativity for CD34, and prominent cytoplasmic positivity for Bcl-2. Other differential diagnoses include trichoepithelioma, which shows more abundant and highly fibrocytic stroma, and frequently involves follicular bulbs and papillae[5].

In conclusion, we report a case of BFH with clinical presentation of localized grouped papules on the chest of an adult that has not been reported.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

REFERENCES

1. Mills O, Thomas LB. Basaloid follicular hamartoma. Arch Pathol Lab Med 2010;134:1215-1219.
2. Kim TH, Oh SJ, Kim YC, Roh MR. Segmentally arranged hyperpigmented basaloid follicular hamartoma. Ann Dermatol 2015;27:218-220.
3. Lee MW, Choi JH, Moon KC, Koh JK. Linear basaloid follicular hamartoma on the Blaschko's line of the face. Clin Exp Dermatol 2005;30:30-34.
4. Mehregan AH, Baker S. Basaloid follicular hamartoma: three cases with localized and systematized unilateral lesions. J Cutan Pathol 1985;12:55-65.
5. Requena L, Fariña MC, Robledo M, Sangueza OP, Sanchez E, Villanueva A, et al. Multiple hereditary infundibulocystic basal cell carcinomas: a genodermatosis different from nevoid basal cell carcinoma syndrome. Arch Dermatol 1999;135:1227-1235.

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p16^{INK4a} Expression in Porokeratosis

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Dear Editor:

Porokeratosis is a chronic skin disorder of aberrant epidermal keratinization, clinically manifesting as patches with an elevated peripheral keratotic ridge that corresponds histologically to the cornoid lamella. The lesions have a tendency toward peripheral expansion[1]. The cornoid lamella is a column of tightly packed parakeratotic cells with pyknotic nuclei. In the cornoid lamella, the granular layer is usually lost and the keratinocytes beneath the parakeratotic column show vacuolated or eosinophilic degenerative cytoplasm in association with mild superficial dermal mononuclear cell infiltration[1]. According to the number, size, and distribution of the lesions, at least six clinical variants have been described, including disseminated superficial porokeratosis (DSP) and porokeratosis of Mibelli (PM)[1]. The malignant transformation of porokeratosis into Bowen's disease and squamous cell carcinoma has been described, with a reported incidence of

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