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Segmentally Arranged Hyperpigmented Basaloid Follicular Hamartoma

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Dear Editor:

Basaloid follicular hamartoma (BFH) is a rare malformation with characteristic histopathologic patterns. BFH consists of malformed hair follicles composed of anastomosing cords and strands of basaloid or squamoid cells[1]. Although BFH has been well described histologically, it occurs in the form of skin-colored papules, nodules, or plaques without specific clinical features[1]. Herein, we report a case of segmentally arranged hyperpigmented BFH in a 5-year-old girl.

A 5-year-old girl presented with asymptomatic, firm, hyperpigmented, grouped papules and comedones arranged segmentally on the left lateral aspect of her nose, left retroauricular area, and posterior neck (Fig. 1A, B). The lesions appeared when she was 6 months old, and the affected area had increased since then. She had no medical or family history. A skin biopsy of the retroauricular area demonstrated multiple islands composed of basaloid and squamoid cells restricted to the upper dermis, with branching cords and anastomosing strands (Fig. 2A, B). Some anastomosing strands were connected to the epidermis. No significant clefting between the tumor and stroma was observed, nor were there any atypical cellular or mitotic features. Based on clinical and histological findings, a diagnosis of BFH was made. Given the child's age and her parents' preferences, we decided to observe the lesions rather than treat them surgically or by laser.

The differential diagnosis of BFH, a benign adnexal tumor, includes infundibulocystic basal cell carcinoma (ICBCC), nevoid basal cell carcinoma syndrome (NBCCS), trichoepithelioma, and fibrofolliculoma. Of these, it is most important to rule out ICBCC, a malignant condition, to avoid unnecessary surgical excision. BFH is a superficial malformation of hair follicles and is composed of basaloid and

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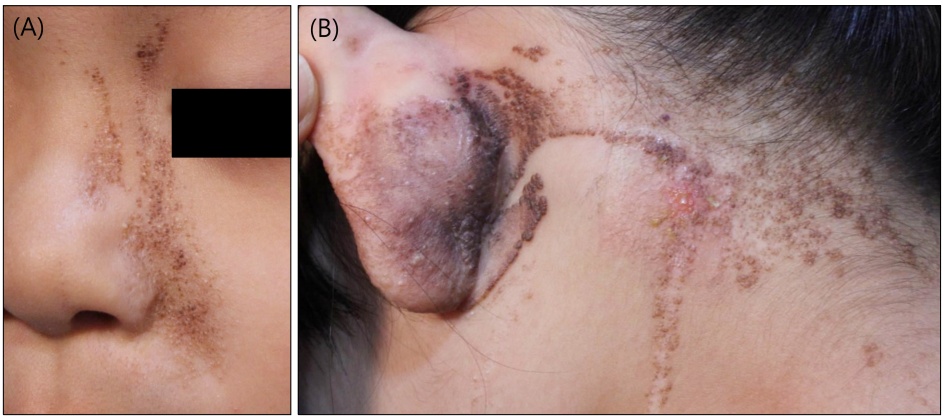

Fig. 1. (A) Segmentally arranged, hyperpigmented, grouped papules and comedones on the left lateral aspect of the nose, (B) left retroauricular area, and posterior neck.

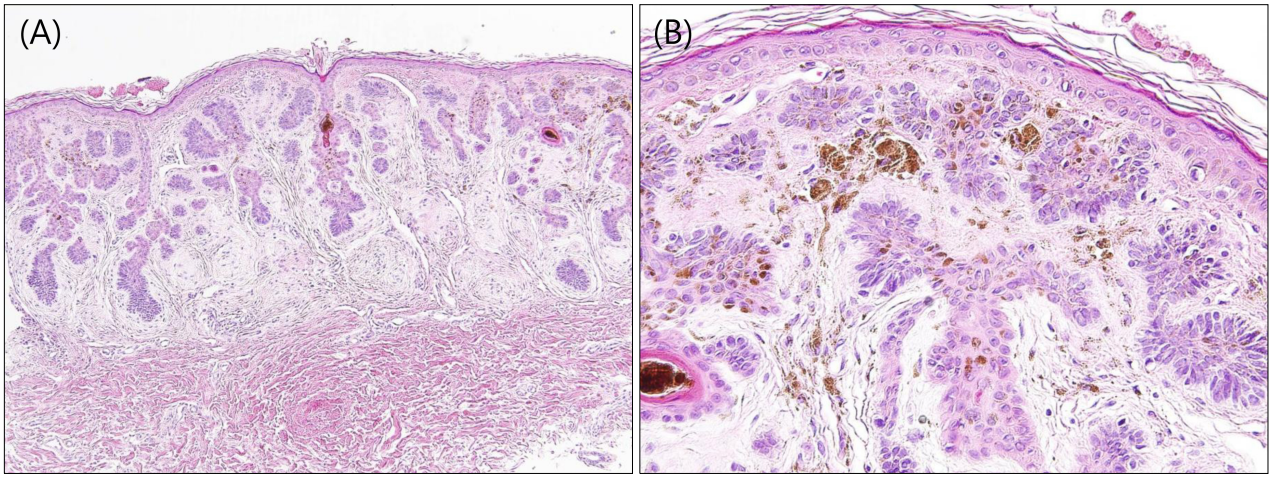

Fig. 2. (A) Multiple islands restricted to the upper dermis with branching cords and anastomosing strands (H&E, ×100). (B) Basaloid cell proliferation admixed with squamoid cells surrounded by loose stroma and melanophages. No cleft between the stroma and tumor cells was observed. Atypical cellular or mitotic features are absent (H&E, ×200).

squamoid cells, while ICBCC is composed of basaloid cells and can occasionally infiltrate into the deeper dermis, subcutaneous fat, or skeletal muscles. Additional characteristics of ICBCC include mitotic activity in the tumor cells and clefts within the stroma. Based on these histologic features, a diagnosis of BFH, rather than ICBCC, was made in this case.

NBCCS is hereditary, unlike BFH, and consists of multiple basal cell carcinoma (BCC) that manifest at an early age, along with skeletal anomalies, jaw cysts, and ectopic calcification.

Although the premalignant potential of BFH is currently undetermined, several cases of BCC within BFH have been reported[2,3]. Some evidence indicates that BFH is not a stable hamartomatous disease and that it might behave similarly to a sebaceous nevus, a lesion in which BCC can arise[3].

Twenty-nine cases of BFH have been reported thus far. In Korea, there have been several cases of skin-colored, localized or multiple, scattered BFH[4,5]. Our case is unique owing to its clinical features: hyperpigmented, segmentally, and linearly arranged lesions with unilateral distribution. Although BFH does not have well-recognized clinical features, a specific histologic pattern can be found, providing a diagnostic clue. Failure to differentiate BFH from BCC may result in aggressive surgical excision of a benign adnexal tumor. However, further studies are needed to elucidate the premalignant potential of hamartomatous lesions and the possible relationship between BFH and BCC.

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A Case of Cutaneous Metastatic Cholangiocarcinoma on the Percutaneous Transhepatic Biliary Drainage Catheter Insertion Site

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Dear Editor:

Cutaneous metastasis of cholangiocellular carcinoma (CCC) is a rare occurrence and the majority of such cases develop after the placement of a percutaneous transhepatic biliary drainage (PTBD) catheter[1]. Most cases of PTBD catheter-related cutaneous metastatic CCC that have been reported in the literature to date have rarely included a dermatologic manifestation[2]. Herein, we report a case of cutaneous metastatic CCC that developed after PTBD catheter insertion. A 74-year-old male patient who had an unresectable CCC visited our dermatology clinic for evaluation of a firm, 2 cm-sized, erythematous and hyperpigmented nodule on his right abdomen. The nodule had developed on the exit site of a former indwelling PTBD catheter. The catheter had been inserted one year prior and was removed after 5 months. A percutaneous transhepatic gallbladder drainage catheter had then been inserted near the exit site of the former PTBD catheter. The nodule had appeared 6 months after the removal of the PTBD catheter and it had gradually increased in size (Fig. 1). The patient had a medical history of malignancies associated with percutaneous indwelling catheters; therefore, we performed a skin biopsy on the nodule to determine if it was a possible skin metastasis or a hypertrophic scar. The histopathology results identified well-developed glands composed of atypical tumor cells between the sclerotic scar tissues (Fig. 2). Based on the clinical and histologic findings, we made a diagnosis of metastatic CCC resulting from PTBD catheter insertion. PTBD is commonly used in the treatment of malignant biliary obstruction and catheter-related cutaneous metastasis on the PTBD exit site has been only rarely reported since the 1980s[3]. Catheter-related metastasis can develop at all sites along the catheter tract, from the insertion site to the exit site[4]. Recently, Takahashi et al.[5] suggested that the incidence of cutaneous metastasis related to PTBD has been rather underestimated. They reported that catheter site metastasis may recur meta-

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