

A Case of Infundibulocystic Basal Cell Carcinoma Clinically Mimicking a Melanocytic Nevus Associated with Epidermal Cysts

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A 52-year-old Japanese woman presented with a 1.5-cm black, glossy, flat, pediculated lump that clinically mimicked a melanocytic nevus on the left temporal side of her head. The subcutaneous tumor beneath the nodule was elastic and hard. A histological examination showed that the tumor was well circumscribed with an exo- and endophytic growth 2.4 × 1.9 cm in size. The lesion contained several keratinous cysts and was composed of funicular fascicles containing squamoid cells. Excessive mucinous material deposition was observed around the tumor periphery and a palisading arrangement of nuclei in the tumor periphery was seen in some areas. Based on these findings, a diagnosis of infundibulocystic basal cell carcinoma (IFC-BCC) was made. This report presents a case of IFC-BCC that clinically mimicked a melanocytic nevus and was also associated with epidermal cysts.

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Key words: follicular differentiation, funicular fascicles, infundibulocystic basal cell carcinoma, melanocytic nevus-like appearance, squamoid cells

Introduction

Infundibulocystic basal cell carcinoma (IFC-BCC) was first reported in 1987 as a new variant of BCC with follicular differentiation¹. This type of BCC was first described by Tozawa and Ackerman based on the results of 15 biopsy specimens of BCC. Their report generated considerable controversy in the literature, mainly concerning the difference between this type of BCC with follicular differentiation and trichoepithelioma^{2,3}.

Follicular differentiation itself is not unique to IFC-BCC⁴; however, the manner of follicular differentiation observed in IFC-BCC is unique because lower follicular structures such as follicular bulbs and papillae are reportedly absent within the tumor, unlike in trichoblastoma and trichoepithelioma^{4,5}. Another characteristic pathological finding of BCC with follicular differentiation is that the tumor cell bundles composed of squamoid cells spread in either a reticular or funicular shape. Three years later, Walsh and Ackerman finally named this new

variant of BCC with follicular differentiation as IFC-BCC and classified it as a subtype belonging to the clinicopathological classification of BCC⁶.

The typical clinical features of IFC-BCC include the fact that they are small, well-circumscribed, skin-colored, dome-shaped, observed as superficially located nodules on the face, and remain small for a long time, as previously reported^{4–9}. Since its initial description, additional reports have been described^{8–13}. The histopathological differences between IFC-BCC and benign conditions such as basaloid follicular hamartomas, folliculocentric basaloid proliferation, trichoblastoma, and trichoepithelioma have been debated⁹.

Here, we report a case of a 52-year-old woman with IFC-BCC of the scalp that clinically mimicked a melanocytic nevus associated with epidermal cysts.

Case Report

A 52-year-old Japanese woman presented with a rubbery-

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firm, blackish, flat, pediculated lump in the left temporal area of her head. A firm, painless, enlarging subcutaneous tumor was found under the lump. The tumor was initially noticed a few years earlier, but the patient reported that she had never experienced either an injury or a burn. Her medical and family histories were unremarkable. The lesion was a solitary, glossy, black-colored, flat, 1.5-cm nodule (Fig. 1). An elastic, firm, subcutaneous tumor, free of galeal fascia, was noted beneath the nodule. It was suspected to be an epidermal cyst with nodular



Fig. 1 Clinical presentation at the age of 52 years. An infundibulocystic basal cell carcinoma on the head in the left temporal area. A firm, painless, enlarging subcutaneous tumor is seen under the lump. The lesion is a solitary, glossy, black-colored, flat, 1.5-cm nodule.

BCC and was simply excised under local anesthesia. The tumor was easily removed because of its sharp circumscription. Cornified components within the tumor leaked out during the surgical procedure. The middle half of the tumor was not evaluated because the lower part of the tumor was suspected to be an epidermal cyst because of its cornified components.

Histopathologically, a well-circumscribed tumor was observed that consisted of two epidermal cysts (10 mm and 7 mm in diameter) in the fixed section with an epithelial lining on the bottom of the tumor (Fig. 2a, 2b). The tumor cell bundles spread in a solid or funicular shape and were composed of squamoid cells (Fig. 2c). In addition, a palisading arrangement of trichoblastic cells was observed in other areas. Alcian blue-periodic acid-Schiff staining showed mucinous material deposition around the tumor periphery. No follicular bulbs or papillae were observed within the tumor (data not shown). The vertical surgical margin of the tumor was focally positive. As a result, a second total extirpation was performed.

Discussion

Histopathologically, our patient had multiple epidermal cysts with an infundibular epithelium-like epithelial lining. The epithelial cords spread in a reticular or funicular shape and were composed of squamoid cells. A proliferation of trichoblastic cells of the tumor with mucinous material deposition around the tumor periphery was observed in another area, and the tumor cells had never differentiated toward a lower follicular structure. These histopathological findings were consistent with the crite-

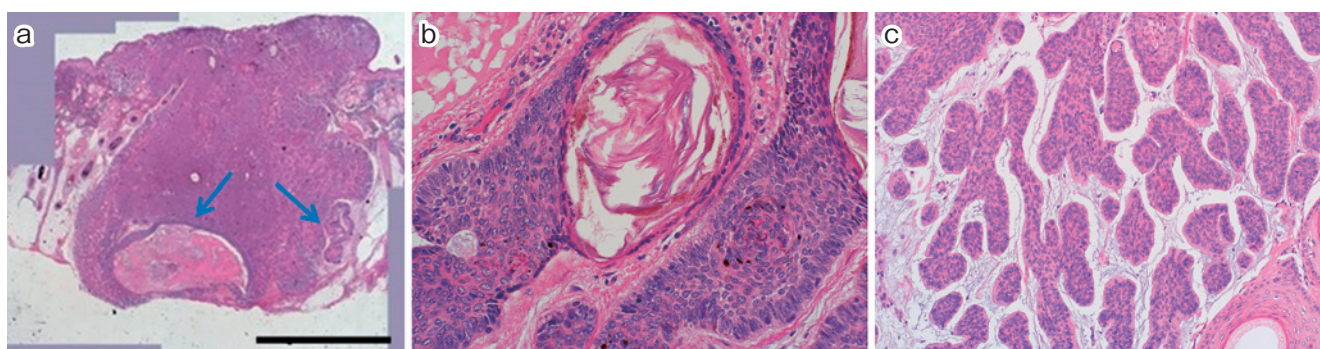


Fig. 2 Histopathological findings

- (a) A whole view of the tumor shows a well-circumscribed tumor consisting of two small keratinous cysts (H&E stain; $\times 4$) (bar indicates 10 mm; arrows: 10 mm and 7 mm in diameter).
- (b) Magnification of the central part of the tumor shows a keratinous cyst with an epithelial lining. In addition, a palisading arrangement composed of trichoblastic cells is seen in the other area. Magnification from the left upper part of the tumor shows the tumor cell bundles spread in either a solid or funicular shape, composed of squamoid cells (H&E stain; $\times 200$).
- (c) Lower follicular structures such as follicular bulbs or papillae are not seen within the tumor (H&E stain; $\times 100$).

ria for IFC-BCC described by Walsh and Ackerman^{4,6}. Benign neoplasms such as trichoepithelioma, trichoblastoma, and basaloid follicular hamartoma were excluded because they have follicular bulbs and papillae with an abundant amount of fibrocytic stroma^{6,9}. Moreover, trichoepithelioma and trichoblastoma were excluded because they do not contain mucinous material deposition between the neoplasm and stroma due to their fibroepithelial unit components.

In terms of clinical presentation, an IFC-BCC has been described as a small and slow-growing neoplasm that is well-circumscribed and located in the superficial dermis, with a clinically less aggressive biological behavior⁶⁻⁹. In our case, the neoplasm seemed round and well-circumscribed at the bottom of the tumor and was easily removed. However, the neoplastic cells extended vertically throughout the dermis into the subcutis, beyond the tumor margin, and the vertical surgical margin of the neoplasm was focally positive on the galea. As a result, a second total extirpation was needed. We considered this aggressive biological behavior of the neoplasm in our case to be unique. Walsh and Ackerman described that the extension of the IFC-BCC through the dermis into the skeletal muscle was a malignant characteristic of this neoplasm, however this clinical manifestation was rarely recognized⁴. Hida et al. described a case of IFC-BCC that showed a rapid increase in tumor size and number during chemotherapy¹⁰. However, our patient did not undergo chemotherapy. The aggressive biological behavior observed in this case also led us to exclude other benign neoplasms such as trichoepithelioma, trichoblastoma, and basaloid follicular hamartoma⁹.

Ansai reported that IFC-BCC in Japan tended to be misdiagnosed clinically as melanocytic nevi on the basis of a survey on all BCCs in Japan¹⁴. In our case, the neoplasm was black with a rubbery-firm consistency that mimicked a large melanocytic nevus in its clinical appearance. Our patient's lesions were clinically different from those previously described in the English literature, which were small, with an average size of 1-10 mm, and dome shaped⁶⁻⁹. The size of the tumor in our patient (24 mm in diameter) is unusually large, as it is the second largest IFC-BCC ever reported¹⁻¹³. The largest tumor, described by Arakawa A et al. in Japan, measured 40 mm in diameter on the lower abdomen of an 88-year-old woman¹³. The neoplasm in our patient may have grown to this size largely because it was hidden by the patient's hair. Both of the epidermal cysts in the bottom of this

neoplasm grew without recognition into what seemed to be one epidermal cyst as noted during the surgical procedure. Ansai also reported the frequency of IFC-BCC among all BCCs in Japan¹⁴. They were identified in 5.7% of a total 1,227 cases of BCCs and the incidence of an IFC-BCC occurring on the head was extremely rare (0.16%)¹⁴. In the English literature, IFC-BCC occurring on the head has not been reported to date.

Herein, we report the case of a 52-year-old woman, who had a tumor on the scalp with unusual clinical features, which was diagnosed as IFC-BCC that clinically mimicked a melanocytic nevus and was associated with epidermal cysts. This unique case suggests that IFC-BCC has a wide range of clinical presentations.

Conflict of Interest: The authors report no conflicts of interest.

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