

Early Stage of Pilomatricoma with an Empty Cyst Cavity and Unusual Clinical Appearance

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ABSTRACT

We present an unusual case of cystic pilomatricoma. A 13-year-old Japanese boy was referred to our department for evaluation of a 10-day history of a small reddish nodule on his right cheek. Histopathological examination showed a well-circumscribed cystic structure showing a central empty cavity in the dermis. The wall of the cyst was composed of basophilic cells. Inner layers of the wall were covered with flattened epithelial cells having trichohyalin granules. Based on these findings, a diagnosis of cystic pilomatricoma was made. To our knowledge, there has been no report of cystic pilomatricoma with an empty cavity. Dermatologists should be aware of pilomatricoma in the early stage showing a central empty cavity.

Key words basophilic cell; early stage; empty cyst cavity; pilomatricoma

Pilomatricoma is a benign cutaneous tumor derived from hair matrix cells. It is usually a solitary, skin-colored or bluish and firm nodule on the head, neck or upper extremities. Histopathologically, the tumor is comprised of basophilic cells and eosinophilic shadow cells. We present a case of cystic pilomatricoma in the early stage with unusual clinical and histopathological findings.

PATIENT REPORT

A 13-year-old Japanese boy was referred to our department for evaluation of a 10-day history of a small nodule on his face. Physical examination revealed a small reddish nodule, approximately 4 mm in diameter, located on the right cheek (Fig. 1a). Dermoscopic observation showed dilated vessels at the center of the nodule and a dark-purple structure in the lesion (Fig. 1b). Based on these findings, we suspected the lesion to be melanocytic nevus, dermatofibroma or trichoblastoma. To make a final diagnosis, the lesion was resected under local anesthesia. Histopathological examination showed

a well-circumscribed cystic structure in the dermis (Fig. 1c). There were hemorrhagic foci in the upper part of the cyst (Fig. 1c). The lower part of the wall of the cyst was composed of basophilic cells and few shadow cells (Fig. 1d). In addition, inner layers of the wall were covered with flattened epithelial cells having trichohyalin granules in part (Fig. 1e). On the other hand, the upper part of the wall of the cyst showed epidermoid keratinization and was indistinguishable from an epidermoid cyst (Fig. 1f).

DISCUSSION

Pilomatricoma is a benign cutaneous tumor derived from hair matrix cells.^{1,2} It is usually a solitary, skin-colored or bluish and firm nodule on the head, neck or upper extremities affecting young individuals.¹ In contrast, however, the lesion was a small reddish nodule with a dark-purple structure showing an unusual clinical presentation for pilomatricoma in our case. Histopathologically, the tumor is also commonly well-circumscribed and often surrounded by a connective tissue capsule. Characteristically, the tumor mass consists of both basophilic cells and eosinophilic shadow cells. As the lesion matures, basophilic cells are transformed into shadow cells.²

Another intriguing point of the tumor in our case is the clinical course. Kaddu et al. categorized pilomatricomas into 4 chronologically distinct stages from a histopathological perspective: early, fully developed, early regressive and late regressive.³ Although an early lesion shows a nodular structure of basaloid epithelium containing a central cystic cavity filled with keratinous and cornified material, fully developed lesions are large, oval and horizontally oriented neoplasms. Fully developed lesions are also comprised of peripheral basaloid epithelium and internal masses containing shadow cells without cyst formation. In addition, both early and late regressive lesions do not have cystic structures. In our case, the tumor was only 4 mm in diameter at his first visit to our hospital and only 10 days had passed since he found the tumor. Based on the staging system mentioned above as well as our unusual clinical presentation, our case can be categorized into early stage. We consider that early stage can include not only solid cavity type

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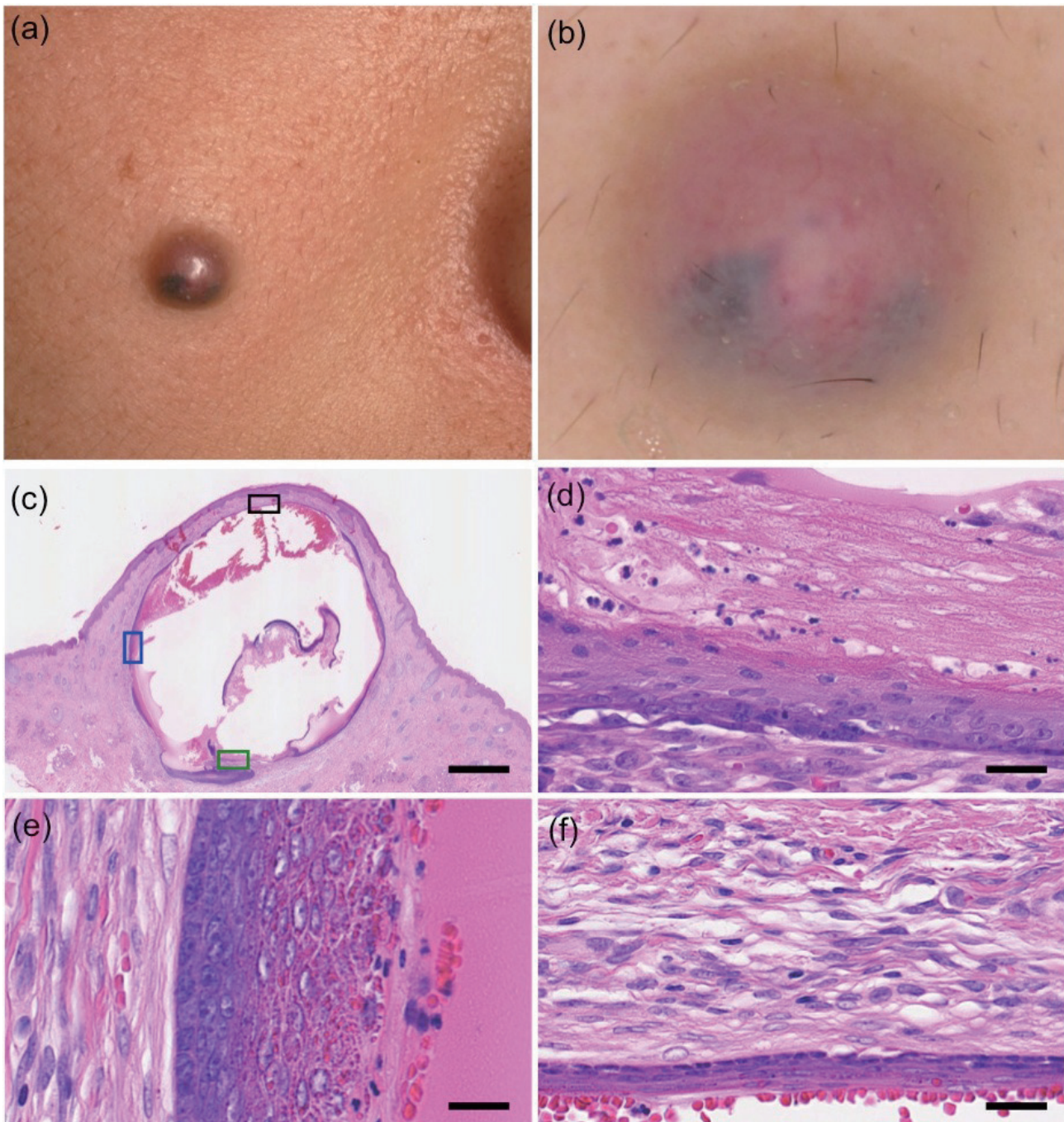


Fig. 1. (a) A reddish nodule on the right cheek. (b) A dark-purple structure and a dilated vessels by dermoscopy. (c) Lower magnification of the resected specimen (hematoxylin and eosin). Bar = 1 mm. (d) The wall of the cyst composed basophilic cells in green square of Fig. 1a (hematoxylin and eosin). Bar = 25 μ m. (e) Trichohyalin granules in the cell wall of blue square of Fig. 1a (hematoxylin and eosin). Bar = 25 μ m. (f) The wall of the cyst composed epidermoid keratinization in black square of Fig. 1a (hematoxylin and eosin). Bar = 25 μ m.

filled with cornified or keratinous material in the center but also empty cavity type as seen in our case. Empty cavity type seems to be characterized by the presence of cells having abundant trichohyalin granules without cornified material. In our case, however, the cyst had an epidermoid keratinization as well (Fig. 1f).

It has been reported that hybrid cyst is a combination of infundibular and trichilemmal cyst.^{4,5} Therefore, hybrid cyst should be included in a histopathological differential diagnosis of an early stage of pilomatricoma as seen in our case. To our knowledge, there has been no report of cystic pilomatricoma with an empty cavity.

Dermatologists should be aware of pilomatricoma in the early stage showing a central empty cavity.

The authors declare no conflict of interest.

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