

Pilomatricoma, also known as calcifying epithelioma of Malherbe, is the most common adnexal tumour in children and young adults. It typically occurs on the head, neck or upper limb. Here, we report a case of bullous variant of pilomatricoma. A male adult presented a firm nodule covered by a sequent bulla on the left shoulder. Pathological examination showed the tumour was composed of irregularly shaped epithelial islands which included two cell types: basophilic cells and eosinophilic shadow cells. No cellular atypia or abnormal mitotic figures were observed. These findings were consistent with a diagnosis of pilomatricoma. The bulla was attributed to lymphatic obstruction that resulted in the dilation of lymphatic vessels. Our pilomatricoma had the unusual feature of a cross-shaped, giant bulla measuring 10 cm × 5 cm overlying the tumour. To our knowledge, it was the largest bullous pilomatricoma in the literature. After surgical resection, there was no recurrence.

Key words: pilomatricoma, bullous appearance, adnexal tumour, lymphatic obstruction.

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Pilomatricoma with a bullous appearance

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Case report

A 24-year-old Chinese man visited our clinic in March 2008 with a firm nodule that he had had on the left shoulder for 4 months. It had been growing gradually and had been covered with a bulla for 3 months. The contained fluid of the bulla had been removed 2 months ago, but quickly relapsed. There was no history of particular local trauma or chronic irritation at the site of the lesion. Skin examination revealed a cross-shaped, translucent, thick-walled, giant bulla measuring 10 cm × 5 cm on the left shoulder. The underlying nodule had a demarcated borderline with the size of about 2.5 cm. It was protuberant and hard (Fig. 1). Laboratory studies, including blood cell count, urinalysis and serum biochemistry, were all within normal limits. No other significant findings were observed.

The nodule was surgically removed under local anaesthesia. When the bulla was opened at the beginning of the resection, a large amount of serohemorrhagic liquid was secreted from the cut surface of the bulla. The resected nodule had a rough surface (Fig. 2). It was elastic, hard and the content of the nodule was pultaceous. Pathological examination showed that the tumour was composed of irregularly shaped islands of epithelial cells consisting of two cell types: basophilic cells and eosinophilic shadow cells (Fig. 3). Upon total sectioning of the excised lesion, we observed neither cellular atypia nor abnormal mitotic figures. These findings were consistent with a diagnosis of pilomatricoma. There were no recurrences after 6 months of follow-up.

Discussion

Pilomatricoma, also known as calcifying epithelioma of Malherbe, is the most common adnexal tumour in children and young adults. Our case was also a young adult. It typically occurs on the head, neck or upper limb. The



Fig. 1. A cross-shaped, translucent bulla measuring 10 cm × 5 cm on the left shoulder, with a hard nodule on the central region



Fig. 2. Macroscopic appearance of the resected nodule revealing a rough surface

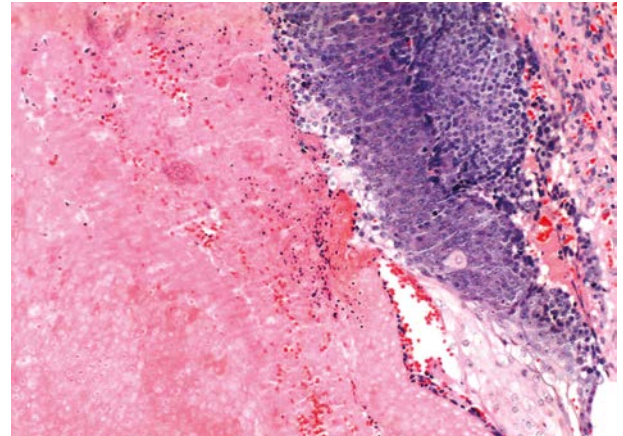


Fig. 3. The tumour was composed of islands of basophilic cells, shadow cells and transitional cells in a fibrotic stroma (H&E, × 100)

tumour may show a wide variety of signs [1–4] and may be subcutaneous, ulcerating, perforating, pigmented (with haemosiderin), keratotic, vascular, anetodermic, bullous or telangiectatic and resembling basal cell carcinoma [1, 5, 6]. Bullous pilomatricoma is an uncommon lesion, and only a few cases of this variant exist in the literature [1, 7, 8]. The bullous appearance was attributable to lymphatic obstruction that resulted in the dilation of lymphatic vessels. In the literature the accompanied bulla most frequently had round or oval shape and the size varied from 0.5 to 5 cm [1, 5, 9–12]. However, the pilomatricoma we reported had the unusual feature of a cross-shaped, giant bulla measuring 10 cm × 5 cm overlying the tumour. To our knowledge, it was the largest bullous pilomatricoma in the literature.

Considering the healing of the wound, we excavated the nodule by fenestration operation without excising the covered skin. The bulla disappeared and did not relapse. Consequently, the histopathological examination showed a dermal tumour consisting of irregular aggregations of basophilic cells at the periphery of islands of ghost cells, and did not reveal the extraordinary dilation of lymphatic vessels in the overlying dermis.

Authors declare no conflict of interest.

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