Epidermal Inclusion Cyst with Melanoma-like Melanophagic Proliferation

Sir,

In a review of 128 cutaneous epithelial cysts in the past year, we found 6 cysts that were pigmented, of which 3 were also associated with a granulomatous reaction. Among these, there was an unusual specimen which merits detailed description.

CASE REPORT

A 26-year-old male had had a painless right gluteal swelling for the past 10 years. It measured $5 \text{ cm} \times 4 \text{ cm}$ and was soft and non-fluctuant. The swelling was excised.

The surgical specimen measured $4 \text{ cm} \times 3 \text{ cm} \times 3 \text{ cm}$ and appeared as an irregular mass of adipose tissue. The cut surface revealed a bilocular cystic lesion (Fig. 1). The larger cyst (*A*) measured 2 cm in diameter and had a thick fibrous wall with a smooth, pearly white inner lining. This showed a focal black pigmented area of 0.6×0.4 cm. The smaller cavity, (*B*), was collapsed, irregular and completely surrounded by black tissue.

Microscopically, the larger cyst was lined by keratinizing stratified squamous epithelium with marked hyperpigmentation of the keratinocytes. Melanocytes arranged in the basal layer showed enlargement, but no nuclear atypia. The pigment also stained the luminal keratin flakes. The focal pigmented area in the wall included a collection of heavily pigmented cells, obscuring the cytologic morphology (Fig. 2). The pigment was identified as melanin by the bleaching reaction. The cells were seen to be oval- to spindle-shaped, with moderate cytoplasm and round-to-oval vesicular nuclei. Immunohistochemistry showed no evidence of S100 or HMB 45 positivity in these cells, indicating that they were melanophages.

The smaller cavity, which lacked an epithelial lining, was lined by a very prominent foreign body giant cell reaction. This was surrounded by similar oval-to-fusiform pigmented cells. The giant cells also showed melanin within the cytoplasm. A few of the sections showed a tract of communication lined by granulomatous tissue between the two cavities.

DISCUSSION

The present case showed features of a ruptured pigmented epidermal cyst. The unusual feature is its association with an exuberant melanophagic proliferation, which has not been previously reported. The cells were plump oval-to-fusiform and markedly pigmented. The nuclei showed no atypia. Our initial impression was either a naevus or melanoma in association with an epidermal cyst. Immunohistochemical stains revealed S100 and HMB to be negative, leading to the inference that these were melanophages. We presume that chronic irritation of this subcutaneous epidermal cyst was responsible for the rupture and increased production of melanin by melanocytes, which provoked a granulomatous reaction and melanophagic proliferation. Another unusual feature was that the rupture of the cyst led to formation of an irregular cavity, giving this lesion a "double-deckered" appearance.

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Fig. 1. Bilocular lesion showing larger cyst (A) with a pigmented area (*arrow*) and smaller cavity (B) surrounded by black tissue.



Fig. 2. Pigmented area in cyst wall composed of heavily pigmented oval-to-fusiform cells; similar cells also surrounded the cavity.