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Giant tufted angioma successfully treated with radiation

Dear Editor,

We report the case of a 52-year-old man with Pick's disease who presented painful erythema from the right breast to the abdomen of 1 month's duration. He had no history of trauma. The erythematous induration had irregular borders and was 20 cm in diameter at the time of the first consultation (Fig. 1a). Although multiple red to dark brown papules and purpura were observed in the erythematous induration, bleeding, erosion, ulceration, hyperhidrosis and hypertrichosis were not. The findings of routine laboratory tests were normal including platelet count of $196 \times 10^3/\mu L$, prothrombin time (PT) and activated partial thromboplastin time (APTT). A chest computed tomography scan showed subcutaneous swelling from the right breast to the abdominal wall (Fig. 1b). Histological findings of a biopsy specimen taken from the lesion revealed multiple separated cellular lobules scattered throughout the dermis, resulting in a cannonball appearance (Fig. 1d). Each lobule was surrounded by dilated crescent-shaped capillary lumina and was composed of aggregated endothelial cells forming small capillary lumina. Cellular atypia was not observed. A diagnosis of tufted angioma was made on the basis of these findings. The lesion rapidly expanded downward by 8 cm within 1 month after the first consultation (Fig. 1c); the patient complained of intolerable pain and tenderness, especially in the area of expansion. Non-steroidal anti-inflammatory drugs had no effect. The laboratory tests including PT and APTT were normal and the platelet count of $199 \times 10^3/\mu L$ was unaltered from the first consultation. Histological findings of a skin sample obtained from the expanding area also showed tufted angioma without hemorrhage. Radiation therapy with X-ray was provided; 30 Gy at the front and 30 Gy at the right lateral side (total, 60 Gy). After radiation therapy, the induration softened, erythema reduced (Fig. 2) and the pain resolved. No recurrence was observed for at least 20 months after the therapy.

Tufted angioma, also known as angioblastoma, is a rare and benign cutaneous vascular tumor characterized histologically by circumscribed angiomatous tufts and lobules throughout the dermis with a cannonball appearance.[1] Erythema or red subcutaneous nodules (diameter, 2–5 cm) are commonly seen. It is important to differentiate between tufted angioma and malignant neoplasms such as Kaposi's sarcoma and angiosarcoma. These malignant neoplasms exhibit atypia and usually lack a lobular morphology, and this can aid in diagnosis. In 70% of the cases, tufted angioma occurs when the patient is below 10 years of age; however, a few cases of adult-onset tufted angioma have also been reported.[1] Pain is reported by 40–80% of the patients,[2,3] but is usually not severe. The intolerable pain and tenderness in our patient was resistant to any non-steroidal anti-inflammatory drugs.

Various treatment options are available, including surgical excision,[4] radiation,[5,6] cryosurgery, pulsed

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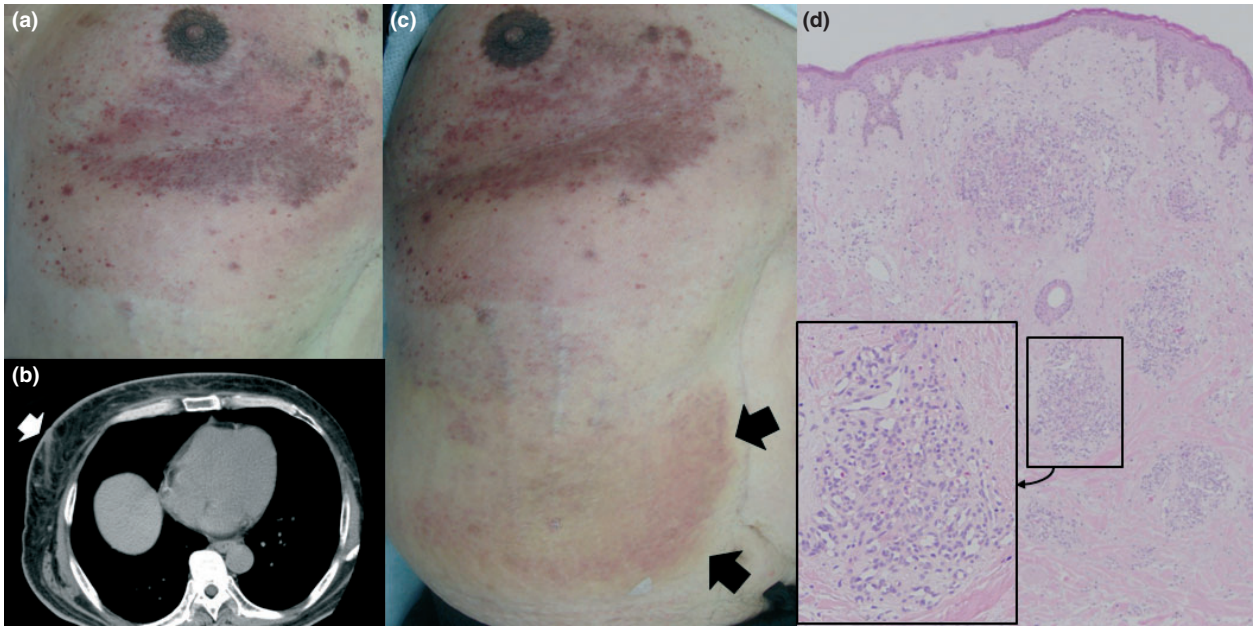

Figure 1. (a) Clinical features of erythematous induration that extended from the right breast to the abdomen observed at the time of the first consultation. (b) Computed tomography scan of the chest at the time of the first consultation. A subcutaneous lesion with an irregular border was observed (arrow). (c) Clinical features 1 month after the first consultation. The erythematous induration had expanded downward by 8 cm (arrows). (d) Histological findings of the induration (hematoxylin–eosin, original magnification ×40). The square shows a high magnified image (original magnification ×200).

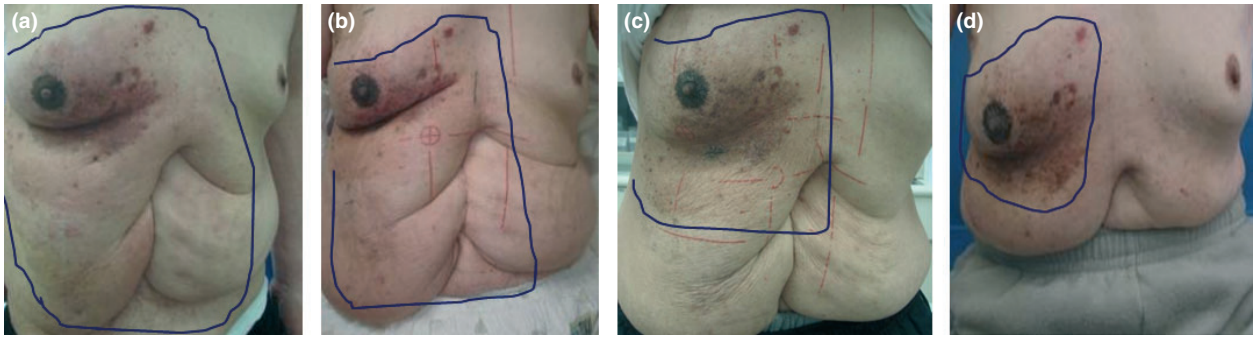

Figure 2. Clinical course of the lesion after radiation therapy. (a) Before radiation therapy, (b) after receiving 30 Gy and (c) after 60 Gy of radiation, and (d) 3 months after radiation therapy (d). The areas marked with blue circles indicate the induration.

dye laser,[7] interferon (IFN)-α therapy[8] and corticosteroids.[9] However, the efficacies of these therapies are still controversial. Surgical excision is often followed by recurrence, and the results obtained with cryosurgery, pulsed dye laser, s.c. IFN-α injection, systemic corticosteroid therapy and intralesional corticosteroid injection are variable. Moreover, spontaneous regression has been observed in some patients.[2,6] Therefore, in some cases, treatment is provided only if the lesion is symptomatic or cosmetically disfiguring. However, there are reported cases in which the condition has persisted for more than 50 years.[1]

Tufted angioma spreads gradually over a period of months or years, and the lesion remains unchanged unless it is treated. However, in some cases, the lesions grow rapidly,[10] as observed in our patient. There are two possibilities in the rapid expansion of the lesion in the present case: tufted angioma growth itself and hemorrhage within the tumor. Several cases have been reported as a tufted angioma with

Kasabach–Merritt phenomenon.[11] However, hemorrhage was not observed histologically in the biopsy specimens obtained from the expanding area 1 month after the first consultation. In addition, platelet counts, PT and APTT were within normal range through the clinical course. Thus, it is likely that expansion of the lesion was mainly caused by tumor growth itself. The size of the lesion in our patient was too large to be treated by surgical excision or intralesional IFN-α injection. Kimura *et al.*[5] had demonstrated the efficacy of soft X-ray for treating this disease. Hence, we decided to use radiation therapy. Further, Ishikawa *et al.*[2] reported disease recurrence 2 months after electron beam therapy with a 20-Gy dose. Therefore, the radiation dose was set at 30 Gy each for the front and lateral sides. Although, in our patient, recurrence was not observed for 20 months after radiation therapy, the condition may recur in the future. It is thus necessary that regular follow-up examinations are conducted.

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Spiradenocylindroma: Rapid increase in size attributed to hemorrhage

Dear Editor,

Spiradenomas and cylindromas are benign, slowly growing, cutaneous adnexal neoplasms. Occasionally, neoplasms with features of spiradenoma and cylindroma within the same lesion are reported in the published work.[1–4] A patient with a rapidly enlarging tumor which histopathologically showed foci of both spiradenoma and cylindroma with an organized hematoma is presented.

A 52-year-old man presented with a 25-year history of a painless solitary tumoral mass of approximately 4 cm on the extensor surface of the left arm. The

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