A proliferative vascular tumour of the skin in a kidney-transplant recipient (recurrent pyogenic granuloma with satellitosis)

Y. Le Meur¹, C. Bedane², P. Clavère³, P. Peyronnet¹, and C. Leroux-Robert¹

Service de ¹Néphrologie, ²Dermatologie, ³Radiothérapie, Centre Hospitalier Universitaire Dupuytren, Limoges, France

Introduction

As in the acquired immunodeficiency syndrome, cutaneous vascular tumours have been described in transplant recipients. Kaposi’s sarcoma is a well known complication of immunosuppressive therapy in these patients [1]. More recently cutaneous bacillary angiomatosis due to rochalimaea has also been reported, presenting as an unusual cutaneous vascular neoplasm [2]. We present the case of a kidney-transplant patient with a cutaneous vascular proliferation which proved to be a recurrent pyogenic granuloma with multiple satellitosis (Warner–Wilson Jones syndrome). This condition has never previously been reported in an immunodeficient patient.

Case report

In October 1994, a 53-year-old man received a cadaveric renal transplant (original renal disorder, polycystic kidney disease). The immunosuppressive regimen was quadruple and sequential consisting of antithymocyte globulin for 11 days, associated azathioprine and corticosteroids. Cyclosporin A (CsA) was introduced on day 9. The initial course was complicated by numerous problems: serum sickness occurred from days 15 to 18, there were herpetic gingival lesions on day 24, a staphylococcal wound infection on day 45 which was treated with antibiotics (pristinamycin, teicoplanin) and surgical drainage on day 55, and Pneumocystis carinii pneumonia treated by intravenous sulfamethoxazole-trimethoprim on day 60.

On the 70th post-transplant day, the patient developed a small wart-like skin lesion of the right thumb after a recent injury. The lesion grew rapidly over a 2-month period. It appeared vascular in nature with superficial necrosis, bled easily, and measured 2 × 2 × 3 cm. Arteriography of the hand confirmed it was a vascular tumour (Figure 1). A 3-mm punch biopsy specimen showed vascular proliferation of the dermis, which was interpreted as being a pyogenic granuloma. Since the lesion continued to enlarge, a resection of the tumour with a skin graft was performed 15 days later.

Macroscopically the tumour was pedunculated and erupted through a breach in the epidermis. The surface of the lesion was necrotic with polynuclear cells, leukocytoclastic debris, and clumps of bacteria. The dermis demonstrated stromal oedema and a lobular vascular proliferation (Figure 2). The vessels were small, and lined by plump endothelial cells protruding into the lumen. Rare mitotic figures were seen, but there was no endothelial cell atypia or necrosis. An inflammatory infiltrate existed between the vessels. The Warthin–Starry stain was negative for the cat-scratch disease bacillus. Transmission electron-microscopy showed that the proliferative vessels had normal architecture and revealed no bacilli within blood vessel walls or the intracellular spaces.

Soon afterwards the lesion abruptly reoccurred with multiple satellite lesions, completely destroying the skin graft. In 6 weeks, the tumour measured 5 × 5 × 3 cm. Treatment with doxycycline for 4 weeks was unsuccessful and the tumour continued to enlarge (7 × 6 × 3 cm) (Figure 3). Radiotherapy was administered at a dose of 9 Gray in 6 fractions over 18 days, which arrested the tumour proliferation and led to its regression and disappearance in 2 months. Two years later it was still in remission.

Discussion

Patients with immunodepression have a high incidence of cutaneous Kaposi’s sarcoma [1]. Skin lesions that clinically resemble Kaposi’s sarcoma have been noted in patients with bacillary angiomatosis, especially with the acquired immunodeficiency syndrome [3,4]. Some angiosarcomas are occasionally difficult to diagnose. The discovery of a proliferative vascular tumour in a transplant recipient requires an accurate diagnosis to distinguish between these different disease processes.

Histologically our patient’s lesions were quite suggestive of a pyogenic granuloma. There was a large proliferation of small blood vessels with plump endothelial cells protruding into the lumen. These
histological features are not found with Kaposi’s sarcoma, but could correspond to cutaneous bacillary angiomatosis, in which a superficial biopsy frequently shows the features of a pyogenic granuloma [3,4]. Nevertheless the Warthin–Starry stain was negative and no bacteria were detected with electron-microscopy. We therefore conclude that our patient’s tumour belongs to the disease spectrum of pyogenic granuloma.

Pyogenic granuloma (also called lobular capillary haemangioma) is a benign vascular neoplasm that can develop spontaneously or in response to physical trauma [5]. It appears as a sessile or pedunculated smooth-surfaced red nodule, that may easily bleed, crust or ulcerate. The lesions are usually located on the face, arms or hands. This vascular tumour is usually solitary and varies between 5 and 10 mm in size. In rare cases, multiple satellite lesions can occur around the primary tumour (Warner–Wilson Jones syndrome) especially after surgical treatment of the original pyogenic granuloma [6]. The satellite lesions usually develop in young people (<25 years old), and commonly occur on the trunk. These satellite lesions usually measure 1 mm to 1 cm in diameter. Our patient’s tumour was atypical in terms of age, location, and especially size of the lesion.

Successful treatment of pyogenic granuloma with satellitosis has been obtained with excision, light freezing with solid carbon dioxide, or diathermy [6–8]. In some patients, however, spontaneous involution occurs within 6–12 months. Because of the size and the expansive proliferation of our patient’s tumour we decided to perform radiotherapy, which was successful within 2 months. To our knowledge this treatment has never been employed for this disease entity. Nine Gray were administered in six fractions similar to treatment for other benign vascular tumours such as angioma.

The cause of pyogenic granuloma is unknown.
However, physical trauma is probably an important factor in the pathogenesis of this lesion. Recurrence with multiple satellites often occurs after surgical treatment of the primary tumour. It could be due to direct trauma or an angiogenic substance released by the primary tumour [9]. With these assumptions, it is pertinent that our patient was under immunosuppressive treatment, which could explain why the tumour growth was so extensive and rapid. Furthermore we believe our patient’s high degree of immunosuppression was consistent with the multiple opportunistic infections encountered in the initial period after transplantation.

Acknowledgements. We thank Richard Smoot for his assistance in the preparation of the manuscript.

References


Received for publication: 20.1.97
Accepted: 24.1.97