Multiple Brownish Macules on both Palms in a Patient Who Underwent Kidney Transplantation

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Case History

A 50-year-old man presented with multiple macules on his left palm. He was diagnosed with end-stage renal disease of unknown cause 30 years prior and underwent haemodialysis through a left-sided arteriovenous (AV) fistula, as well as kidney transplantation 13 years before presentation. He was taking immunosuppressive agents including prednisolone, tacrolimus and mycophenolate mofetil. Dermatological evaluation revealed multiple ill-defined painful brownish macules on his left palm [Figure 1]. A punch biopsy was done on one of the palmar lesions. Histological examination revealed dermal proliferation of irregular anastomosing vascular channels lined by atypical cells with large hyperchromatic nuclei and frequent mitosis [Figure 2a and b]. Immunohistochemistry was positive for CD31 and CD34 indicating a vascular tumour origin [Figure 2c and d]. Human herpesvirus-8 (HHV-8) staining was negative.

(Question) What is your diagnosis?

Diagnosis:

Cutaneous angiosarcoma.

Discussion

Angiosarcoma (AS) is a rare, aggressive malignant neoplasm of the vascular endothelial cells that presents as an enlarging ulcerative or haemorrhagic red nodule with a poorly demarcated margin.[1] It is an aggressive tumour arising in either blood or lymphatic vessels that has a poor prognosis. According to the Surveillance, Epidemiology, and End Results (SEER) programme, which recorded 434 cases of cutaneous angiosarcoma from 1973 to 2007, 5- and 10-year survival of cutaneous angiosarcoma was 34% and 14%, respectively.[2]

AS predominantly affects the face, scalp and neck of elderly men. Although AS usually presents as an ulcerated nodule, a variety of clinical presentations can exist including cellulitis, and facial and eyelid angioedema.[3] Histologically, AS shows irregular vascular channels in the dermis, which are lined by atypical endothelial cells having variably enlarged nuclei and nuclear pleomorphism. CD31 and CD34 staining can highlight those endothelial cells for differentiation of AS from other diagnoses.

Chronic immunosuppression can cause a variety of diseases and the association between the development of AS and chronic immunosuppression is well established. However, AS after kidney transplantation is rarely observed, and most cases present at the site of the AV fistula.

There were 32 reported cases of angiosarcoma after kidney transplantation in English literature. The most common site of occurrence was AV fistula (n = 19), followed by cutaneous (n = 9), allograft (n = 2) and heart (n = 1). However, none of them were on the hand without preceding arteriovenous fistula (AVF) lesions.

Treatment methods for this aggressive tumour include a combination of surgery and radiation therapy; in some cases, the addition of chemotherapy and immunotherapy with recombinant interleukin-2 prolongs survival.[4] However, despite these aggressive treatments, AS shows a poor prognosis with a high tendency toward local recurrence and metastasis.

Factors suggesting a favourable prognosis in patients with AS include younger age (<50 years), localized tumour, complete excision of the tumour, smaller size (≤5 cm) and lesions of trunk and extremities.[2]

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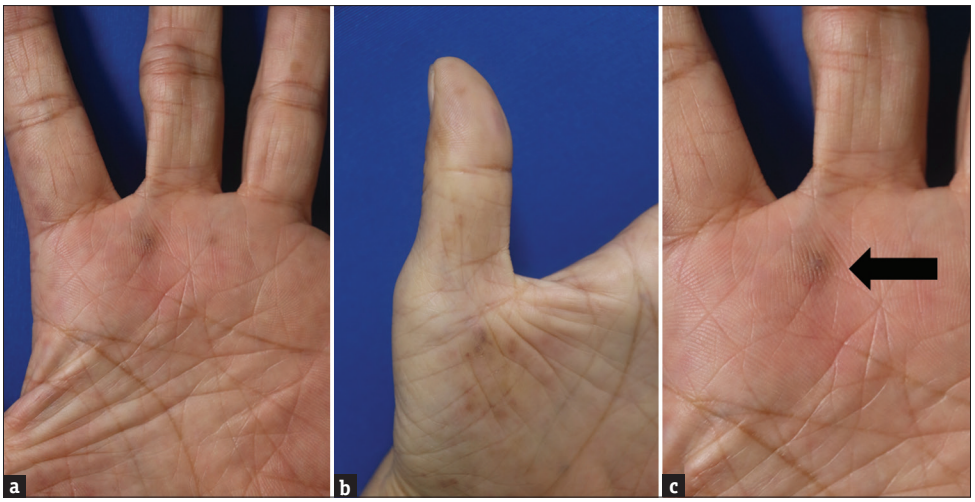


Figure 1: (a-c) The patient presented with multiple painful brownish macules on his left palm. Punch biopsy of one of the palmar lesions was performed

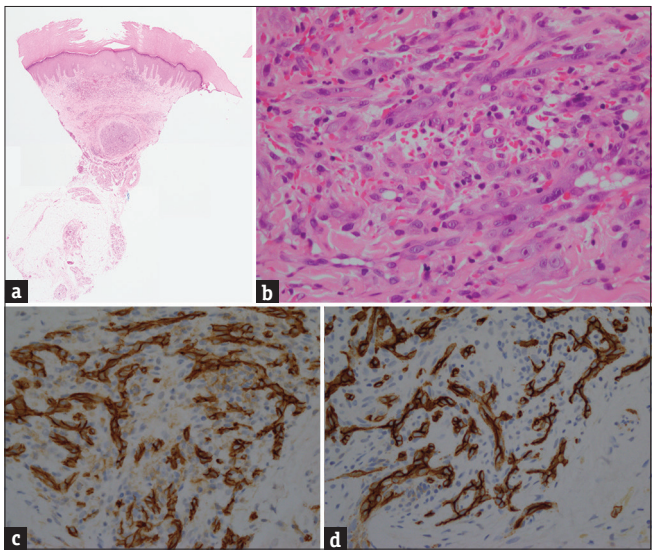


Figure 2: (a) Skin biopsy revealed nodular infiltration of the lower dermis (A: H&E stain, X100). (b) Irregularly shaped anastomosing vascular channels, a lining of endothelial cells with nuclear atypia, and increased mitosis with large hyperchromatic nuclei were observed (B: H&E stain, X400). (c, d) Immunohistochemical staining showed diffuse CD 31 and CD 34 positivity, indicating a vascular origin of the tumor cells (c: CD31 stain, X400, d: CD34 stain, X400).

Since angiosarcoma has a poor prognosis, clinical suspicion and prompt diagnosis are essential. Its various clinical presentations include discolouration of the overlying skin, poorly defined bruise-like lesions that progress to nodular or ulcerating lesions with proximal or distal neurovascular symptoms and sudden onset of pain.

Learning points

- Angiosarcoma is a rare, aggressive malignant tumour arising from the vascular endothelial cells.
- Angiosarcoma predominantly affects elderly men in the face, scalp and neck and has a poor prognosis.
- Chronic immunosuppression is associated with the development of angiosarcoma, and it is rarely observed after kidney transplantation, mainly at the site of AVF.
- The treatment of cutaneous angiosarcoma involves a combination of surgery, radiation therapy and in some cases, chemotherapy and immunotherapy.
- Recognizing that cutaneous angiosarcoma has a variable clinical presentation is critical for prompt diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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