Correspondence

Acquired progressive lymphangioma in the groin area successfully treated with surgery

doi: 10.1111/j.1365-2230.2009.03286.x

Acquired progressive lymphangioma (APL), also called benign lymphangioendothelioma, is a rare, benign proliferation of lymphatic vessels.[1] A search of the English literature in the PubMed electronic database found 35 reported cases. Clinically, an APL usually arises on the trunk and limbs as a solitary, asymptomatic erythematous patch or plaque, and most commonly occurs in adolescents and young adults.[1] We report a case of an APL on the right inguinal area of a patient with a favourable result after surgical excision.

A 33-year-old man presented with a 1-year history of frequent drainage of clear fluid from his right groin area, sufficient to wet his clothing (Fig. 1a), and resulting in a need for frequent changes of clothes. On physical examination, a lesion measuring 10 × 2.0 mm in diameter was found in the groin.

A wide excision and histological examination of the lesion was performed (Fig. 1b). Immunohistochemically, the tumour cells stained positively for Factor VIII, CD31, CD34 and D2-40 (Fig. 2), indicating that the channels were lymphatic vessels. Staining was negative for Kaposi's sarcoma-associated virus and human herpesvirus 8. Based on the clinical and histopathological findings, the patient was diagnosed as having an APL. The patient was

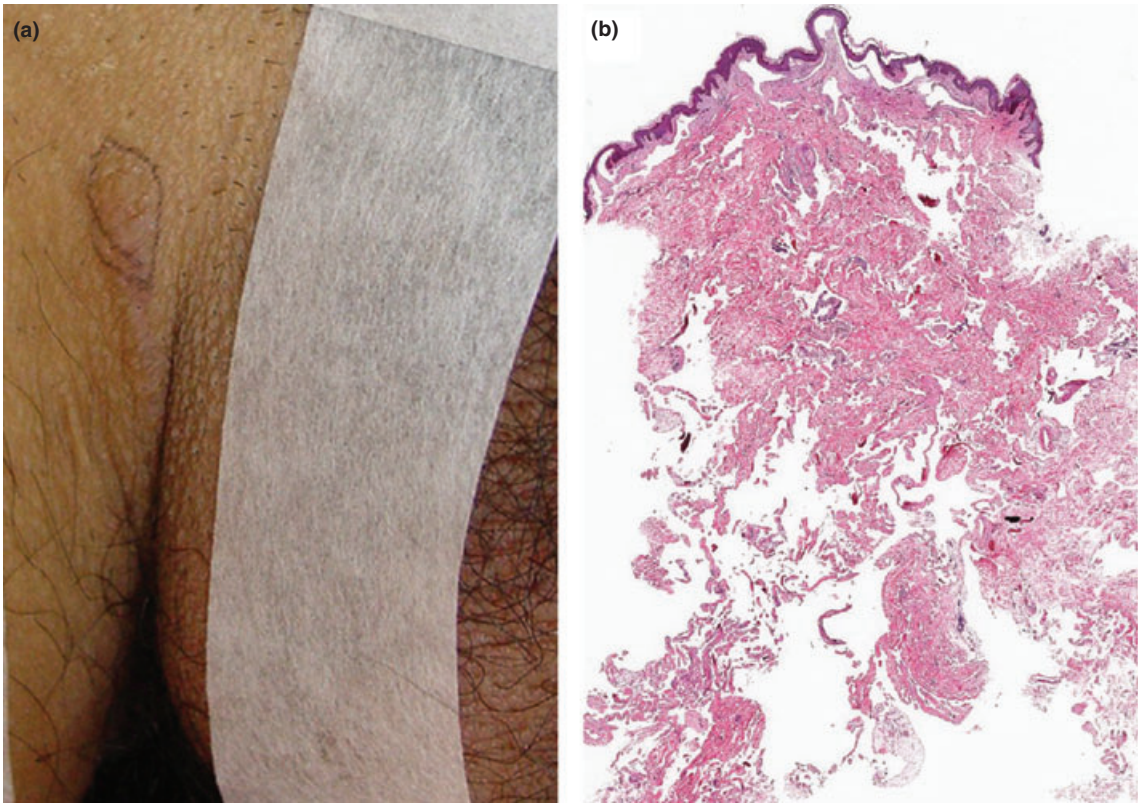

Figure 1 (a) A soft, fluctuant subcutaneous nodule located in the right groin area (circled). (b) Many irregularly dilated vascular channels were seen throughout the dermis and dissecting the collagen bundles. No cellular atypia or mitotic figures were seen (haematoxylin and eosin staining, original magnification × 40).

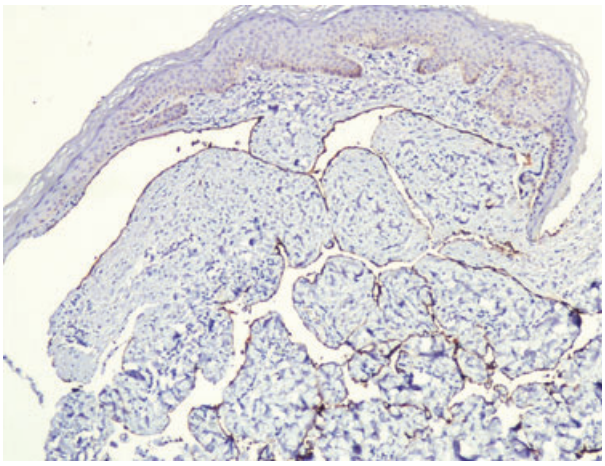

Figure 2 Interlacing vascular channels lined by a single layer of bland-looking endothelial cells which positively stained for D2-40 (original magnification × 100).

symptom-free 9 months after the operation, and follow-up is ongoing.

APL has been reported to develop after femoral arteriography, trauma and radiation therapy for breast carcinoma.[1–3] Those reports suggested that a reactive process plays a role in the development of APL. Careful differential diagnosis of a medial, infra-inguinal groin mass before a surgical approach is important to exclude other potentially dangerous gastrointestinal or genitourinary conditions, such as cystic hygroma, hernia, hydrocoele of the spermatic cord, or torsion of an undescended testicle.

Histopathologically, APL exhibits many dilated, tortuous, thin-walled lymphatic channels dissecting collagen bundles in the dermis. The vascular channels may have scanty proteinaceous material and some red blood cells. Immunohistochemically, the endothelial cells of an APL are variably positive for Factor VIII, *Ulex enropaeus* agglutinin I, CD31 and CD34. In our patient, positive staining for D2-40, a highly sensitive and specific marker of lymphatic endothelium,[4] further confirmed the vascular channels to be of lymphatic origin. Well-differentiated angiosarcoma and the patch stage or lymphangiomatous variant of Kaposi's sarcoma should be considered in the differential diagnosis of APL, especially when it occurs in high-risk populations prone to developing such malignant vascular neoplasms, including patients with human immunodeficiency virus or those with a history of radiotherapy or chronic lymphoedema.[3,5] Histological features such as a lack of cellular atypia and mitotic figures and scanty inflammatory cells can exclude a diagnosis of an angiosarcoma. Clinicopathologically, postradiation lymphatic anomalies of the skin may somewhat resemble an APL, but the pathological features of these anomalies show smaller, better-formed vascular structures and a less infiltrating growth pattern.[6]

Among the 35 reported cases, 23 patients (66%) received surgical excision with an excellent prognosis and only one local recurrence was observed at 7 months of follow-up.[6] Sclerotherapy, advocated in the management of other types of lymphangioma but not yet in APL, may be considered an alternative treatment.

In conclusion, we achieved a satisfying result with surgical intervention for an APL. This case serves to remind physicians of the rare disease condition of APL and the possible confusion with malignant vascular tumours if the clinical features are not carefully correlated with the histopathological features.

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Conflict of interest: none declared.
Accepted for publication 27 October 2008

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