Journal of Plastic, Reconstructive & Aesthetic Surgery (2013) 66, e318–e320

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

CASE REPORT

A rare case of 'histiocytoid haemangioma' of the hand

Giuseppe Accardo*, G. Nele, F. Turrà, I. La Rusca, F. Schonauer

Department of Plastic Surgery, University of Naples "Federico II", Via Pansini 5, 80122 Naples, Italy

Received 2 January 2013; accepted 3 April 2013

KEYWORDS
Histiocytoid haemangioma;
Hand tumour;
Angiolymphoid hyperplasia with eosinophilia;
Tumour-like lesion

Summary Histiocytoid haemangioma has been identified by Rosai in 1979 as a group of vascular tumour-like lesions. This lesion can occur in a wide variety of sites. Surgical excision is the treatment of choice.

We present the case of a 35-year-old man with a swelling in the thenar region of his right hand. Preoperative X-rays, magnetic resonance imaging (MRI) and angio-computed tomography (angio-CT) demonstrated a lesion invading the radial digital artery to the index finger without any bone erosion. It was excised '*en bloc*' with the artery itself. The radial digital nerve to the index finger and the main digital artery to the thumb were preserved. No local recurrence was observed at the 3-year follow-up.

Histiocytoid haemangioma of the hand is a rare disease, and patients should undergo early surgical treatment to achieve complete excision of the lesion without any functional deficit.

Case report

A 35-year-old man presented to our clinic with a swelling in the thenar region of his right hand. The lesion onset dated 7 months with no history of trauma. There was no family history of cutaneous or vascular malformations.

On examination there was no pain, no flexor tendon involvement and no sensory deficits. Both thumb and index finger were well perfused.

X-rays showed no bony erosion. A magnetic resonance imaging (MRI) scan revealed a soft-tissue tumour-like lesion around and invading the digital artery to the index finger; an angio-computed tomography (angio-CT) scan revealed the vascular occlusion of this last vessel.

At surgery the mass appeared as a nodular, capsulated lesion of 2.8 × 1.5 × 1.1 cm size (Figure 1). It was excised '*en bloc*' with the radial digital artery of the index finger. The radial digital nerve to the index finger and the main

* Corresponding author. C.so Vittorio Emanule 21 B, 80059 Torre del Greco, Naples, Italy. Tel.: +39 3478283441; fax: +39 0818491105.
E-mail address: gaccardo@hotmail.it (G. Accardo).

1748-6815/$ - see front matter
http://dx.doi.org/10.1016/j.bjps.2013.04.027

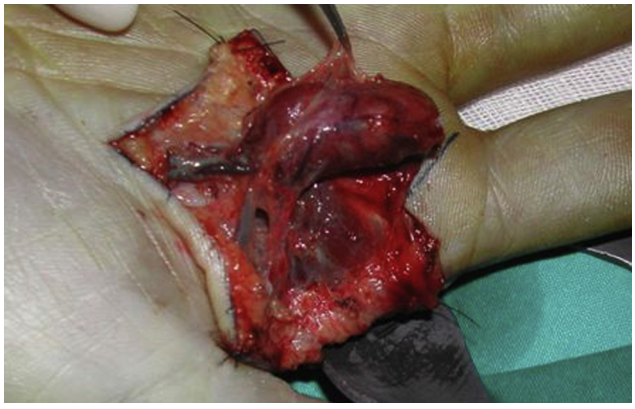

Figure 1 The lesion appearance at surgery: invasion of the radial digital artery to the index finger.

artery to the thumb were preserved (Figure 2). At histology, a haematoxylin and eosin-stained section showed a vascular lesion in the form of small vessels, some of which presented a prominent endothelium with a 'hob nail and histiocytoid' pattern. A rich inflammatory reaction with an important plasmacellular and lymphoid infiltration was associated. These aspects were all expression of a benign lesion, histiocytoid haemangioma, which has local recurrence capacity. The diagnosis of histiocytoid haemangioma was confirmed by an immunohistochemistry staining for an endothelial cell marker, CD34 (Figures 3 and 4).

At the 3-year follow-up the patient did not report any functional deficit or local recurrence.

Discussion

In 1979 Rosai et al.[1] first used the term 'histiocytoid haemangioma' to describe a group of skin, soft tissue, large vessels, bone and heart entities. According to the authors, all these lesions were characterised by the proliferation of a 'histiocytoid or epithelioid endothelial cell'. Immunochemical and ultrastructural findings suggest an endothelial origin for the lesion, which is characterised by well-formed

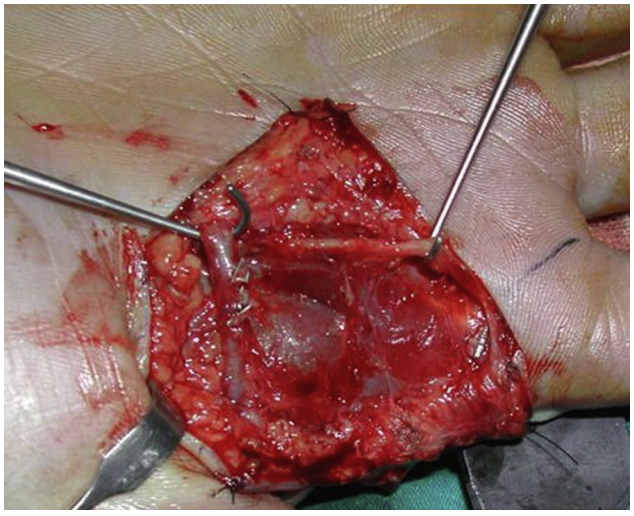

Figure 2 The radial digital nerve to the index finger and the main artery to the thumb that were both preserved.

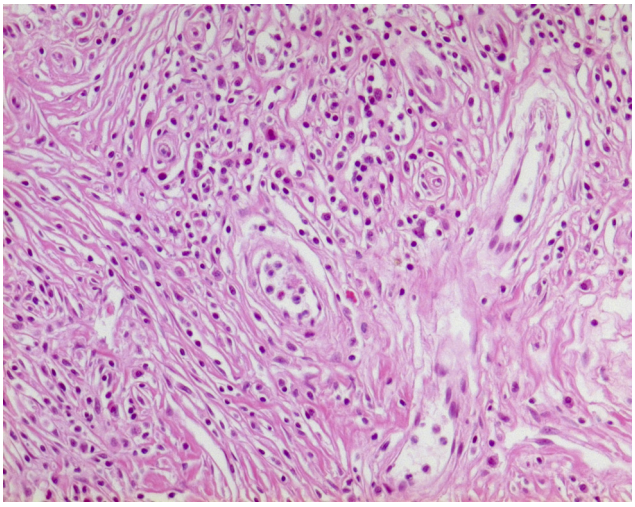

Figure 3 The prominent endothelial cells of histiocytoid hemangioma can be appreciated at haemathoxyline and eosin staining. Scattered inflammatory cells with predominance of plasmacellular and lymphoid infiltration and rare eosinophils can be seen. H&E, 20×.

capillary-sized vessels lined by histiocytoid or epithelioid endothelial cells and often followed by secondary inflammatory infiltrate.[2]

Histological diagnosis is often difficult as epithelioid endothelial cells can be observed in a variety of vascular lesions, ranging from benign and reactive to the neoplastic and fully malignant. Allen et al.[3] proposed in 1992 a classification of 'histiocytoid haemangioma-like' lesions that included angiolymphoid hyperplasia with eosinophilia, histiocytoid haemangioma of the testis, epithelioid haemangioendothelioma and probably spindle cell haemangioendothelioma.

At the moment, histiocytoid haemangioma can be considered an uncommon benign vascular proliferative lesion of unknown origin with no reported incidence of metastatic disease and a good prognosis. However, these

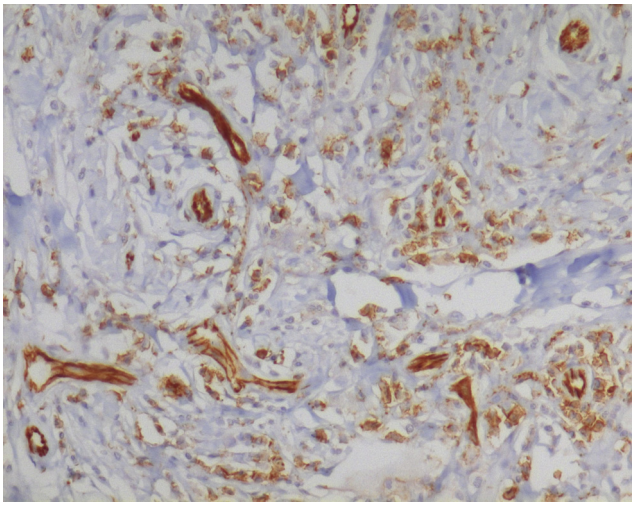

Figure 4 Immuhistochemistry staining for an endothelial cell marker like CD34 is markedly positive. CD34 Immunohistochemistry, 20×.

kinds of lesions could have local recurrence and simultaneous multifocal presentations, even in different organs, as described by Dannaker et al.[4] in 1989. They reported the case of a 31-year-old man with multiple histiocytoid haemangiomas involving skin, subcutaneous tissue and bone limited to the left arm and hand.

Some authors suggested that traumatic events may stimulate the onset of histiocytoid haemangiomas.[3] This lesion can occur in adults and in children. It may concern a wide variety of sites such as skin, soft tissue, oral mucosal membrane,[5] nails,[6] testis,[7] bone,[3] colon, heart and even the epididymis.[8]

Clinical diagnosis is often difficult as the tumour occurs as a progressively tender swelling next to a vessel course with no relevant symptoms in most of the cases. Skin discolouration may be appreciated especially in districts with a terminal circulation such as the hand. Pain and/or functional symptoms may rarely appear because of tumour compression on adjacent structures (nerves, vessels or tendons). Imaging can be useful to study tumour extension, the vascular involvement and any other simultaneous presentations; but histology is mandatory to obtain a certain diagnosis, especially when confirmed by a immunohistochemistry for an endothelial cell marker such as CD31 or CD34.

Surgical excision is the treatment of choice for histiocytoid haemangioma. Other therapies reported in the literature especially for angiolymphoid hyperplasia with eosinophilia are laser-assisted excision for smaller and superficial lesions[9], pharmacological treatment with pentoxifylline[10] and indomethacin farnesil[11], chemotherapy, intralesional steroid injections, low-dose irradiation, cryotherapy[9] and even interferon alpha.[12] Early diagnosis is very important for efficient eradication of these lesions. As there is no risk of metastasis, biopsies are recommended once histiocytoid haemangioma is suspected.

Kraphol et al.[13] had already described a rare localisation of the angiolymphoid hyperplasia with eosinophilia which involved the fourth and fifth rays, with complete occlusion of the ulnar artery, and a small lesion at the level of the metacarpophalangeal joint of the index finger. Our case is a rare localisation in the palm arising from the radial digital artery of the index finger never described before. The technical challenge consisted in preserving sensation to the radial side of the index finger and, more importantly, preserving vascularity to the thumb without interruption of its main artery.

In conclusion, histiocytoid haemangiomas group a variety of rare vascular lesions that can originate from different organs, even the hand as reported here. Diagnostic investigations should include MRI in combination with angiography, angio-MRI or angio-CT scan to determine the extent of the lesion and the vascular involvement. Once the diagnosis of histiocytoid haemangioma is suspected, early and complete surgical excision of the lesions should be undertaken.

Conflict of interest statement

There are no conflicts of interest or funding in relation to this report.

Informed consent for this report was obtained from the patient and the research was done following the code of ethical conduct of our hospital.

References

1. Rosai J, Gold J, Landy R. The histiocytoid hemangiomas. A unifying concept embracing several previously described entities of the skin, soft tissue, large vessels, bone, and heart. *Hum Pathol* 1979;**10**:707—30.
2. Allen PW, Ramakrishna B, MacCormac LB. The histiocytoid hemangiomas and other controversies. *Pathol Annu* 1992; **27**(Pt 2):51—87.
3. Dannaker C, Piacquadio D, Willoughby CB, Goltz RW. Histiocytoid hemangioma: a disease spectrum. Report of a case with simultaneous cutaneous and bone involvement limited to one extremity. *J Am Acad Dermatol* 1989 Aug;**21**(2 Pt 2):404—9.
4. Fetsch JF, Weiss SW. Observations concerning the pathogenesis of epithelioid hemangioma (angiolymphoid hyperplasia). *Mod Pathol* 1991 Jul;**4**(4):449—55.
5. Misselevich I, Podoshin L, Fradis M, Boss JH. Angiolymphoid hyperplasia with eosinophilia of oral mucous membrane. *Ear Nose Throat J* 1995 Feb;**74**(2):122—5.
6. Tosti A, Am Peluso, Fanti PA, Torresan F, Solmi L, Bassi F. Histiocytoid hemangioma with prominent fingernail involvement. *Dermatology* 1994;**189**(1):87—9.
7. Mazzella FM, Sieber SC, Lopez V. Histiocytoid hemangioma of the testis: a case report. *J Urol* 1995 Mar;**153**(3 Pt 1):743—4.
8. Gutiérrez Mínguez E, Serrano Bartolomé JM, Claver Criado M. Histiocytoid hemangioma of the epididymis. A very unusual entity. *Actas Urol Esp* 2000 Jan;**24**(1):70—1.
9. Metze D, Neumann R, Chott A. Angiolymphoid hyperplasia with eosinophilia. Histologic study and successful therapy with the argon laser. *Hautarzt* 1991;**42**:101—6.
10. Person JR. Angiolymphoid hyperplasia with eosinophilia may respond to pentoxifylline. *J Am Acad Dermatol* 1994;**31**: 117—8.
11. Nomura K, Sasaki C, Murai T, Mitsuhashi Y, Sato S. Angiolymphoidhyperplasia with eosinophilia: successful treatment with indomethacin farnesil. *Br J Dermatol* 1996;**134**:189—90.
12. Rampini P, Semino M, Drago F, Rampini E. Amgiolymphoid Hyperplasia with eosinophilia: successful treatment with interferon alfa 2b. *Dermatology* 2001;**202**:343.
13. Krapohl BD, Maches H-G, Reichert B. Mailander. A rare vasoproliferative lesion: amgiolymphoid hyperplasia with eosinophilia of the hand. *Br J Plast Surg* 2003;**56**:168—70.