

AN EPITHELIOID HEMANGIOMA CAMOUFLAGED AS RADIAL ANEURYSM

Ferreira José^{1,2}, Tiago José¹, Soares Tony¹, Cabral Gonçalo¹, Costa Tiago¹, Rossello José¹, Cunha E Sá Diogo¹, Gerardo Gonçalo³

¹ Department of Vascular Surgery, Hospital Beatriz Ângelo, Loures, Portugal

² Department of Orthopedic Surgery, Hospital Beatriz Ângelo, Loures, Portugal

³ Department of Pathology, Hospital da Luz, Lisboa, Portugal

* Corresponding author: jose.nfs.ferreira@gmail.com

Abstract

Introduction: Epithelioid hemangioma (EH) is an uncommon benign vascular lesion usually present as subcutaneous nodules in the head and neck area. Sometimes, these lesions can occur in the peripheral arteries, and when they do, they can be mistaken for aneurysmal dilatations of that respective vessel. We report a case of a 43-year-old male who underwent surgical recession of a radial aneurysm, which after anatomopathological examination, revealed an EH.

Keywords: Epithelioid hemangioma, radial artery, radial aneurysm

INTRODUCTION

EH was first described in 1969 as benign subcutaneous lesions, histologically defined by capillary vessel proliferation, eosinophilic infiltration, and lymphoreticular hyperplasia, typically along a blood vessel¹. EH most commonly presents as red-to-purple painless nodules with a slight propensity on the head and neck, often in a periauricular location, in middle-aged adults with a female predominance^{2,3}. Although rare, they have also been described in the lower back³, penis^{3,4}, orbit^{3,4}, colon³, oral cavity^{3,4}, thigh³ and ankle³. Large vessel involvement is extremely rare^{4,5}, but cases are reported from the subclavian, axillary, brachial, ulnar, radial, facial, popliteal, common carotid, superficial temporal, posterior auricular and occipital arteries^{4,6}.

We describe a case of a male patient who underwent surgical excision of a radial artery EH, with an initial diagnosis of radial artery aneurysm.

CASE REPORT

We report a 43-year-old Caucasian male, with a medical history of Alpha-Thalassemia trait. He was referred to our angiology and vascular surgery consultation complaining of 7-8

months of evolution of a painless pulsatile mass in the right wrist. There was no history of trauma. On physical examination, the right radial artery had a pulsatile and expansile mass. There was an excellent amplitude ulnar pulse and neurological examination of the hand was normal. There were no other findings, including subcutaneous nodules on the face, neck or other anatomical areas.

Duplex ultrasound (DUS) scan suggested an aneurysm arising from the radial artery with a maximum diameter of 2 cm. There was minor anemia and no eosinophilia in the whole blood count.

At the patient's request, he underwent elective aneurysm repair. Under local anesthesia, the radial artery was identified and the mass was dissected and isolated from the remaining soft tissues (Figure 1). It was excised entirely. On macroscopic examination, the resected specimen consisted of a small smooth mass (Figure 2). We sent for histology analysis. Despite mobilization of the vessel ends, primary repair was impossible without excessive tension on the anastomotic site, for which it was decided to perform only the excision of the mass and ligation of the vessel. The patient's recovery was uncomplicated.

Contrary to our expectations, histology of the mass revealed proliferation of small capillary-sized vessels lined by



Figure 1 Identification and referencing of radial artery and isolation of the mass from adjacent soft tissues.

endothelial and epithelial cells with abundant eosinophilic cytoplasm surrounded by inflammatory infiltrate of eosinophils admixed with lymphocytes (Figure 3). This finding was compatible with an epithelioid hemangioma arising from the radial artery.

The patient was followed up for 3 months with complication-free and with normal hand circulation.

DISCUSSION

Many terms have identified EH lesions, including angiolymphoid hyperplasia with eosinophilia, intravenous atypical vascular proliferation, inflammatory angiomatous nodule, and histiocytoid hemangioma, which demonstrate the inconsistencies in nomenclature³. The development of EH inside a large muscular artery is rarely seen⁴ and even more rarely affect the wall of a vessel⁵. Usually, the lesion grows into the vessel's lumen developing occlusive symptoms⁴. The artery, in our case is also not a usual location. We found more 3 case of EH of radial artery described in the literature⁷⁻⁹.

When associated with a peripheral artery, almost always it is confused with an aneurysm, clinically and radiologically. DUS is the primary imaging modality to evaluate these pulsatile masses. Magnetic Resonance Imaging (MRI) is the next step if the mass appears solid on DUS⁶. However, even MRI can suggest



Figure 2 Radial artery excised mass

the swelling as an arterial aneurysm⁶. In laboratory findings, 20% of EH present increased eosinophils counts and serum IgE levels^{4,6}. In this case, the patient had no changes in blood tests and the DUS show an arterial enlargement with turbulent flow but no mural thrombus.

The treatment for these lesions can be medical or

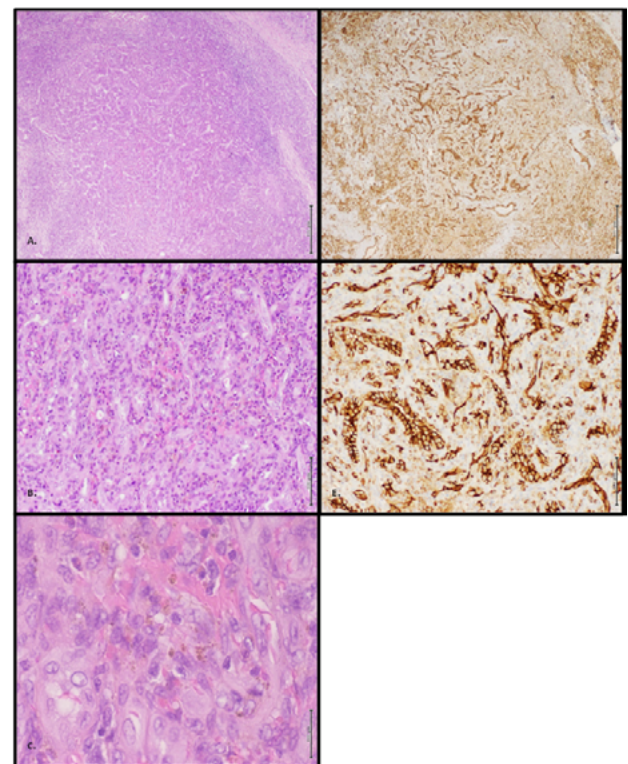


Figure 4 Epithelioid haemangioma is a well demarcated lesion characterized by a proliferation of small capillary-sized vessels, lined by plump, epithelioid endothelial cells, as seen in hematoxylin and eosin staining with 20x (A), 100x (B) and 400x magnification (C); the epithelioid endothelial cells are immunoreactive for CD31 highlighted in 20x (D) and 100x (E) magnification

surgical^{4,6}, however, surgical excision with or without arterial reconstruction is the preferred treatment modality^{3,6}. Even after surgical excision, EH can have a local recurrence of 33%, especially when the lesion is incompletely excised^{3,4,6}.

Histologically, these lesions are characterized by proliferation of blood capillaries lined by plump epithelial endothelial cells or even solid sheets of epithelial cells^{3,6}. An infiltrate with prominent eosinophilia is common; lymphoid follicles are usually present at the periphery of the lesion^{3,6}.

In this case, perhaps several mistakes were made, like performing surgery based only on DUS or failure to perform arterial reconstruction with a vein graft. However, the treatment of this patient was successful, with no recurrence of the disease at 3-month follow-up. Nevertheless, more research and higher quality education is necessary to differentiate between similar vascular anomalies to those cases like the one we are reporting, with incorrect initial diagnosis, do not happen again.

CONCLUSIONS

Upon presentation and evaluation of a pulsatile mass in a peripheral artery, EH and other vascular tumours should be considered as a differential diagnosis. The case we report here shows us that benign vascular tumours, like EH, are often confused with aneurysms before histological confirmation. Therefore, these tumors should be suspected when a young patient without an antecedent history of trauma presents with a lesion suggestive of aneurysmal dilatation.

REFERENCES

1. Wells GC, Whimster IW. Subcutaneous Angiolymphoid Hyperplasia With Eosinophilia. *Br J Dermatol*. 1969 Jan;81(1):1–15.
2. Ko JS, Billings SD. Diagnostically Challenging Epithelioid Vascular Tumors. *Surg Pathol Clin*. 2015 Sep;8(3):331–51.
3. Wiggins CJ, Dibbs RP, Bartlett EL, Ashton DJ, Maricevich RS. Atypical presentation and management of an epithelioid hemangioma: a case report and review of the literature. *Ann Pediatr Surg*. 2020 Dec;16(1):53.
4. Ragazzi M, Falco G, Valli R, Rocco N, Bordoni D, Cadenelli P, et al. Epithelioid hemangioma of brachial artery: report of a case and review of the literature. *Open Med*. 2015 Jan;1:10(1).
5. Barros MB, Lozano FS, Flores T, Antunez P. True Radial Artery Aneurysm Secondary to Haemangioma—Case Report and Literature Review. *EJVES Extra*. 2003 Dec;6(6):115–6.
6. Gunawardena T, Kanagasabapathy S, Cassim R, De Silva C, Wijeyaratne M. An epithelioid haemangioma of the ulnar artery masquerading as an aneurysm. *Ann Vasc Surg*. 2021 Aug;15:19-15.
7. Khaira, H.S., Deshmukh N.S., and Vohra R.K., Angiolymphoid hyperplasia presenting as a radial artery aneurysm. *Eur J Vasc Endovasc Surg*, 1999. 17(2):178-9.
8. Morton, K., Robertson A.J., and Hadden W., Angiolymphoid hyperplasia with eosinophilia: report of a case arising from the radial artery. *Histopathology*, 1987. 11(9):963-9
9. Sandbank, J., et al., Angiolymphoid hyperplasia with eosinophilia (epithelioid hemangioma). *J Cardiovasc Surg (Torino)*, 1991. 32(3):370-2.