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Epithelioid Hemangioma of the Humeral Artery: Case Report

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Abstract

A 32-year-old right-handed male presented with a lump on the right middle arm that had been present for 3 to 4 months. The MRI reports an 8 mm nodular formation in contact with the humeral artery and vein between the middle and distal third of the right arm that undergoes enhancement after administration of venous contrast in close contact with branching of the radial artery. In surgery it was observed a pedunculated lesion from the humeral artery, in principle compatible with a pseudoaneurysm which compromised the integrity of the vascular lumen, so a bypass was performed with a basilic branch graft. The anatomopathological result showed a highly vascularized tumor proliferation and hemorrhagic suffusions made up of epithelioid cells that suggest the differential diagnosis between epithelioid hemangioendothelioma, pseudomyogenic hemangioendothelioma or epithelioid angiosarcoma among other vascular tumors. Finally, a second surgery was performed because there were doubts about having free margins with a new bypass with the ipsilateral saphenous vein. In molecular studies CD34, CK, ERG and FOSB were +++ so an epithelioid hemangioma was the definitive diagnosis. Although rare, vascular tumors should be considered in the differential diagnosis when a mass is associated with an artery. Pathological diagnosis and obtaining free margins given the possibility of malignant recurrence of this tumor are essential.

Introduction

This case report describes a rare vascular tumor (epithelioid hemangioma) in the humeral artery of a young male patient, treated by excision and bypass grafting.

Case Presentation

A 32-year-old right-handed male presented with a lump on the right middle arm that had been present for 3 to 4 months. He hadn´t suffered any accident before. Clinically, there was a palpable tumor without pulse. An ultrasound from another center reported a possible schwannoma or a peripheral sheaths tumor. The first diagnostic imaging came from an MRI that reports an 8 mm nodular formation in contact with the brachial artery and vein between the middle and distal third of the right arm that undergoes enhancement after administration of venous contrast in close contact with branching of the radial artery. A saccular aneurysmal dilation of the brachial artery cannot be ruled out. With these results, a new ultrasound with a high-frequency linear probe was performed. It shows a hypoechoic nodular lesion with well-defined edges, measuring 8.5 mm × 5.5 mm × 13 mm in size, in contact with the posterior wall of the brachial artery in which it imprints and generates slight compression, as well as with slight contact with the posterior part of the median nerve. In surgery it was observed a pedunculated lesion from the brachial artery, in principle compatible with a pseudoaneurysm which compromised the integrity of the vascular lumen, so a bypass was performed with a basilic branch graft (Figure 1). The resection piece was sent to pathology, but not the extension of the margins made before the bypass. The anatomopathological result showed a highly vascularized tumor proliferation and hemorrhagic suffusions made up of epithelioid cells that suggest the differential diagnosis between epithelioid hemangioendothelioma, pseudomyogenic hemangioendothelioma or epithelioid angiosarcoma among other vascular tumors (Figure 2). When in doubt about having tumor margins, a new MRI of the entire upper limb was performed to rule out focality and residual macroscopic lesion. Although normal results, we decided to widen the margins and a new bypass with the ipsilateral saphenous vein (Figure 3). The diagnosis of the piece described a residual focus of tumor proliferation of 1 mm with free borders as well as practically total occlusion of the arterial lumen. In molecular studies CD34, CK, ERG and FOSB were ++++ (Figures

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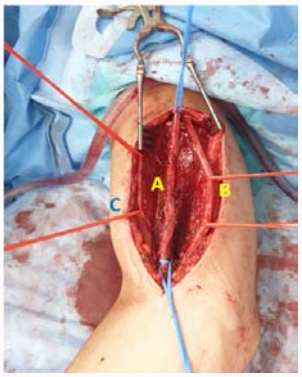

Figure 1: Intra-operative photography of the second intervention shows the first bypass in the brachial artery with the branch of the basilic vein (A). To its right is the median nerve (B) and to its left is the medial antebrachial cutaneous nerve (C).

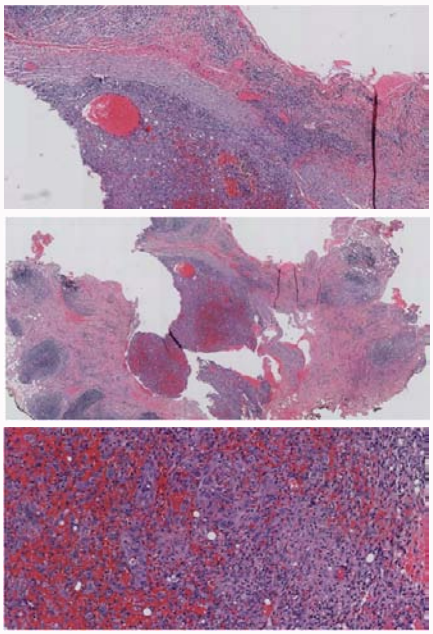

Figure 2: Pathological analysis showed a highly vascularized tumor proliferation and hemorrhagic sub-fusions made up of epithelioid cells.

4-6). The presence of EGFL7-FOSB fusion gene was demonstrated by Archer's PLEX fusion NGS panel. The definitive diagnosis was epithelioid hemangioma. In the one-year follow-up, there was no local recurrence and the bypass was functional.

Discussion

Epithelioid hemangioma is a benign vascular neoplasm composed of well-formed blood vessels lined by plump, epithelioid (histiocytoid) endothelial cells, with abundant eosinophilic cytoplasm and a variable eosinophilic infiltrate. As many as half of the cases show recurrent gene fusions in the FOS and FOSBS genes. Cutaneous and soft tissue lesions are most commonly located in the head and neck region, especially the forehead, preauricular region, and scalp, followed by the distal extremities and trunk. The penis is uncommonly located involved. Visceral occurrences are exceedingly rare [1]. Some arise in large vessels [2,3]. Epithelioid hemangioma occurs over a wide age range, with a peak incidence in the fourth decade of life and no sex predilection. The etiology is unknown. Superficial epithelioid hemangiomas are usually 0.5 cm to 0.2 cm in size, only rarely exceeding 5 cm. Most have a nonspecific nodular appearance. On MRI imaging intravascular presentation of epithelioid hemangioma is always in the form of an arterial aneurysm [4]. Yu-Ching Lin et al. consider that TWIST MR angiography (a non-invasive time-resolved 3-dimensional MR angiography technique, which can yield dynamic angiographic images from early arterial to late venous phase in high temporal and spatial resolution, comparable to conventional catheter angiography) [5], is beneficial in the evaluation of vascular lesions [5] as assisted pathologists to differentiate epithelioid hemangioma from Masson's hemangioma and Kimura disease (lesions with similar histologic features). Furthermore, TWIST MR angiography also clearly delineated the vascular anatomic relationship of the lesion, which is beneficial for pre-operative planning [6]. Typically exhibits a proliferation of well-formed small blood vessels lined by plump and epithelioid endothelial cells with abundant eosinophilic cytoplasm and enlarged round nuclei. The tumor is typically well-demarcated and has a distinctive lobulated growth at low power. Moreover, it shows increased maturation of the vascular lumina at the periphery of the lesion. The stromal component is often edematous, and the tumor may show hemorrhagic changes within the solid component. A subset of cases may display centrifugal growth around a central small artery and evenly complete intravascular growth. So the essential diagnostic criteria is a lobular architecture with vasoformation; lining endothelial cells are epithelioid, with eosinophilic cytoplasm and enlarged round nuclei; loose hemorrhagic stroma, often with eosinophilia. Demonstration of FOS/FOSB rearrangements or expressions may be helpful in selected cases. There are various treatment methods for epithelioid hemangioma that includes steroid

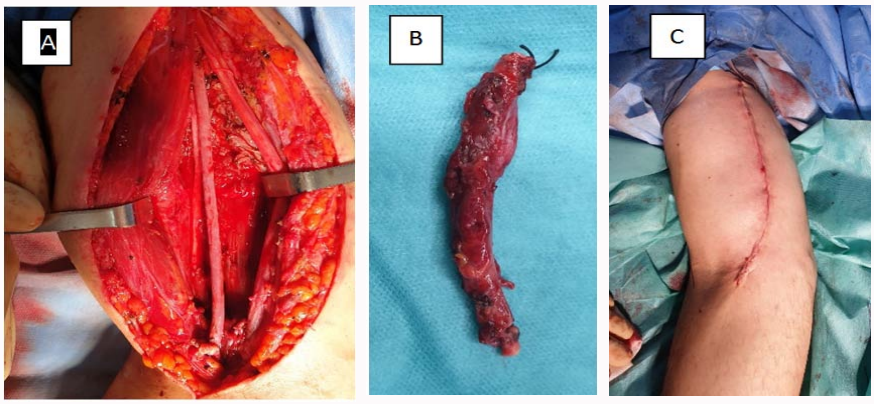

Figure 3: A. Second surgery showing bypass of the brachial artery with the saphenous vein. B. New extended excision sent to pathology. C. Direct suture.

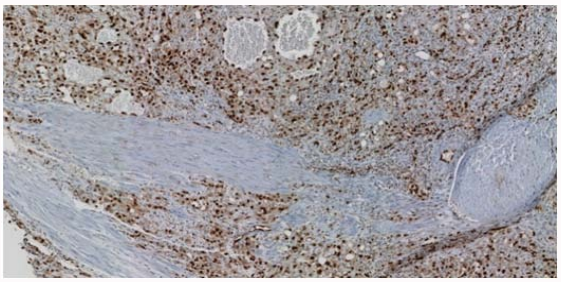

Figure 4: Inmuno CK+.

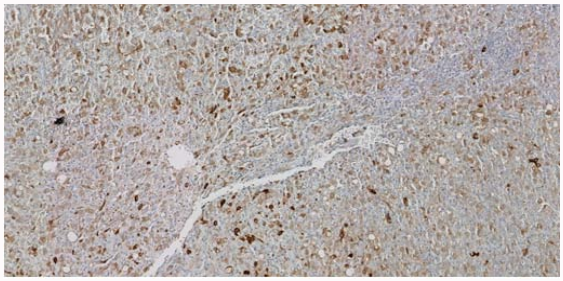

Figure 5: Inmuno ERG+.

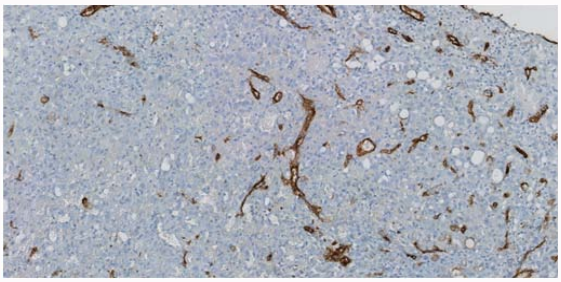

Figure 6: Inmuno CD34+.

injection, radiotherapy, cryotherapy, laser therapy and excision [7]. Surgical excision is considered the optimal treatment choice [7]. The prognosis is as many as one-third of patients experience local recurrence, related to incomplete excision or multicentricity. Most of the recurrences are indolent and can be cured by re-excision, although very rare recurrences can be locally aggressive. Rare lymph node metastasis may occur. To date, no patients have developed distant metastases.

Conclusion

Although rare, vascular tumors should be considered in the differential diagnosis when a mass is associated with an artery. In our experience, the definitive diagnosis of epithelioid hemangioma is pathological, since imaging is inconclusive. It is of the utmost importance to perform a complete resection of the tumor, analyzing all the samples to ensure that free borders are obtained, given the risk of tumor recurrence and malignancy.

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