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Kasuistika | Case report

Spontaneous primary pseudo-aneurysm of brachial artery on an adult patient mis-diagnosed for 8 years: Case report

Fadi A. Hadidi[b,e], Azmy M. Hadidy[a,e], Moaath M. Al Smady[c,e], Osama Samara[a,e], Nosaiba T. Al Ryalat[a,e], Rasha Al Khattab[a,e], Lina Hadidi[d,e]

[a] *Department of Radiology and Nuclear Medicine, Jordan University Hospital, Amman, Jordan*
[b] *Department of Orthopedic Surgery, Jordan University Hospital, Amman, Jordan*
[c] *Department of Vascular Surgery, Jordan University Hospital, Amman, Jordan*
[d] *School of Medicine, The University of Jordan, Amman, Jordan*
[e] *Faculty of Medicine, Jordan University Hospital, The University of Jordan, Amman, Jordan*

ARTICLE INFO

Article history:
Received: 1 March 2014
Received in revised form:
25 April 2014
Accepted: 29 April 2014
Available online: 24 May 2014

Klíčová slova:
Pažní tepna
Primární
Pseudoaneurysma
Spontánní

Keywords:
Brachial artery
Primary
PSA: pseudo-aneurysm
Spontaneous

SOUHRN

Spontánně vzniklé primární pseudoaneurysma v pažní tepně je vysoce ojedinělý nález; v dostupné literatuře bylo popsáno pouze několik takových případů, vždy na podkladě známé etiologie. Naše kasuistika jako první popisuje případ dospělého pacienta se spontánně vzniklým primárním pseudoaneurysmatem v pažní tepně, které bylo po dobu osmi let chybně diagnostikováno a následně řešeno chirurgicky bez jakýchkoli komplikací.

ABSTRACT

Brachial artery PSA is extremely rare with only few cases reported in the literature and all of them are secondary to a known etiology. This is the first report of a an adult patient who was free of any medical treatment with primary spontaneous brachial artery pseudoaneurysm which was mis-diagnosed for 8 years and treated surgically without any complications.

Address: Azmy M. Hadidy MD, FRCR, Department of Diagnostic Radiology, Faculty of Medicine, Jordan University Hospital, the University of Jordan, 11942 Amman, Jordan, e-mails: ahadidy@ju.edu.Jo, amshadidy@yahoo.com
DOI: 10.1016/j.crvasa.2014.04.007

Introduction

Pseudo-aneurysm (PSA) is a pulsatile hematoma due to bleeding into the surrounding soft tissue with fibrous encapsulation and persistent communication between the disrupted vessel and the hematoma, but without normal arterial wall components. Non-healing of the arterial wall disruption will cause the blood to flow back and forth between the two spaces during the cardiac cycle [1]. The overall incidence of PSA after trans-femoral catheterization was reported between 1.1–14% depending on type of intervention, period of manual compression and patient age and gender [2,3]. PSA of the upper extremity accounted for 27% of all the PSA recorded in the Vietnam vascular registry [1]. In neonates, PSA occurs at a rate of about 0.05 after diagnostic catheterization and up to 1.2 after more complex procedures [4]. The arm is an extremely rare site with only a few cases of PSA of brachial artery reported [5]. In literature search there was an underlying cause or risk factor for all cases but this is the first case of spontaneous primary PSA of brachial artery in adult patient [5–7].

Case report

A 41-year-old male patient who is working as a long-distance lorry driver for 22 years and who had never seen a doctor in his life presented to primary healthcare center complaining from painless small swelling on the medial side of his Rt. arm 8 years ago and was diagnosed as muscular herniation and was advised to ignore it, in the last 3 years the lesion slowly progressed in size so the patient was transferred to our orthopedic outpatient clinic. The mass did not cause any impairment in the functional capacity of the right upper limb, the patient reported no pain, numbness, tingling sensation, wasting or color changes in the forearm and hand, there was no history of trauma to the arm, instrumentation (multiple venopuncture, arteriography, dialysis, intervention, drug abuse) or any surgery in the affected area. The patient was not diabetic or hypertensive and had no previous or family history of similar swelling. Examination showed a healthy looking man with no evidence of anemia having an ovoid swelling 4×4 cm on the medial aspect of the right arm with no visible pulsation, scar marking, skin pigmentation or prominent veins and the color of the skin overlying the swelling was the same as that of the surrounding skin. Palpation revealed 4×4 cm non tender, pulsatile, expansile, non-fluctuant mass having same temperature as that of the surrounding skin, which was compressible, non reducible, not blanching on pressure, not attached to the overlying skin or underlying muscle or bone, no skin necrosis or ulceration. Axillary and supraclavicular lymph nodes were not palpable bilaterally. Distal neurovascular status was intact. On auscultation no bruit was audible over the swelling, the heart examination revealed no added heart sound or murmurs to suggest vascular heart disease. The remainder of systemic examination was unremarkable. Laboratory investigations including rheumatology evaluation were normal. Grayscale ultrasound showed a pulsating juxta-arterial heterogeneous mass with hypo-echoic eccentric area (Fig. 1A) compressing the brachial artery. Color Doppler ultrasound showed swirling color flow (yin yang sign) in the mass (Fig. 1B) separated from the brachial artery with color flow within the tract between the artery and the mass consistent with PSA neck while spectral Doppler analysis showed the characteristic to and fro flow pattern within the track confirming the diagnosis of PSA. Magnetic resonance imaging (MRI) showed 4×4 cm mass of heterogeneous signal anteromedially with a peripheral thrombus anteriorly and hypo-intense eccentric area (Fig. 2A), which showed enhancement after I.V contrast injection (Fig. 2B) with phase misregistration artifact (Fig. 2C). While MRI with I.V. contrast (Fig. 3) showed the brachial artery displaced posteromedially by the mass which showed marked central enhancement (contrast extravasations into the mass), also it appeared in continuity with the brachial artery and a diagnosis of PSA was established.

Treatment options were fully discussed and explained to the patient; taking into consideration delayed diagnosis (8 years) of PSA, location and size of PSA, patient's age and career as full-time long-distance lorry driver, and the need to eliminate chances of stent complications (mainly fracture and occlusion) in this young physically active patient with upper limb dependent career. Accordingly the patient decision was to proceed for surgical repair option.

Under general anesthesia surgical resection of the PSA and interposition of brachial vein graft was performed. There was no pre- or post-operative neurological defi-

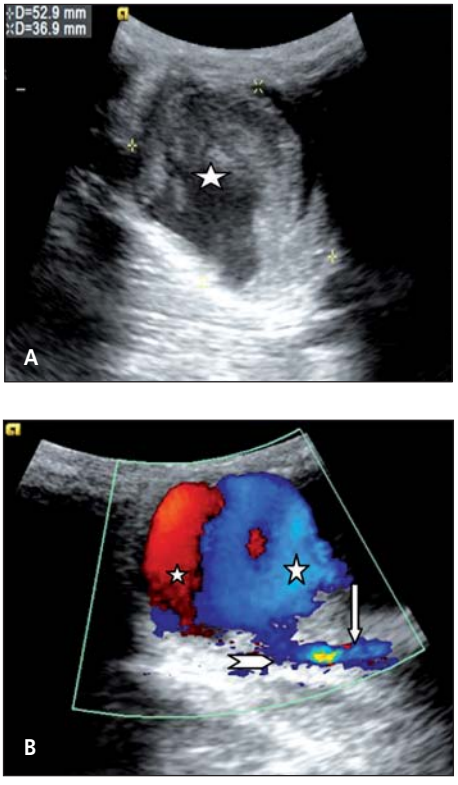

Fig. 1 – (A) Ultrasound showing a pulsating juxta-arterial heterogeneous mass with hypo-echoic eccentric area (star), (B) color Doppler ultrasound showing (stars) swirling color flow (yin, yang sign) in the mass (PSA) separated from the brachial artery (arrow) with color flow within the tract (chevron) between the artery and the mass.

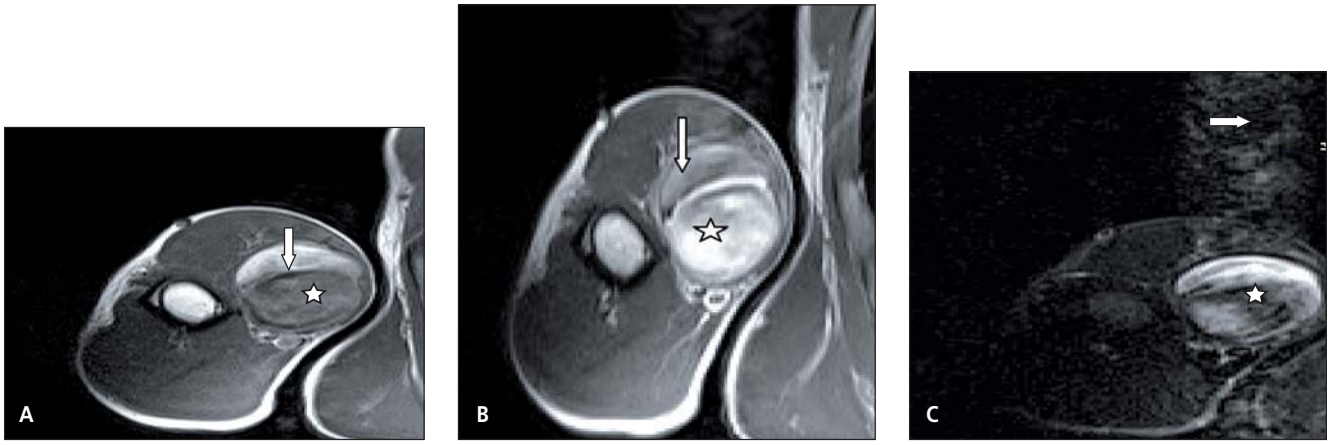

Fig. 2 – (A) Axial T1WI showing the brachial artery PSA with the thrombosed part anteriorly (arrow) and the patent part posteriorly (star), (B) axial post contrast injection T1WI showing the enhancement in the patent part of the PSA (star), while the thrombosed part (arrow) is seen anteriorly. (C) Axial T2WI with fat saturation showing the misregistration artifact (arrow) due to pulsation in the right brachial artery PSA (star).

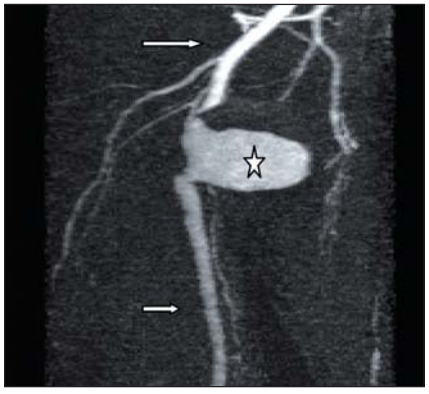

Fig. 3 – MRI with intravenous contrast injection of the right brachial artery (arrows) showing extravasation of contrast in the PSA (star).

cit. The patient was discharged and followed up for 18 months without symptoms or signs of recurrence or complication. Patient gave consent to the publication of this report.

Discussion

The brachial artery is wholly superficial and covered anteriorly only by the skin and superficial and deep fascia. The median nerve enters the arm at first lateral to the brachial artery near the origin of coracobrachialis, it crosses in front (rarely behind) of the brachial artery [8]. But brachial artery PSA is extremely rare with only a few cases reported in the literature and all of them are secondary to a known etiology [5]. After brachial artery arteriotomy PSA occurs in less than 0.04 of cases, but it is possible to be increased and became more common with the use of percutaneous access to the brachial artery [9]. Risk factors for PSA include osteochondroma, bone fracture, joint dislocation, orthopedic procedures with implants, arterial catheterization, antiplatelet agents, peri-procedural anticoagulation, hemophilia, penetrating trauma (stab) and missile injuries (gunshot, nail gun, bullets, weed wacker) and blunt trauma [10]. Other PSA causes include infection, inflammation (vasculitis, pancreatitis), collagen disorders (Marfans and Ehlers-Danlos syndrome), erosion from malignancy, intravenous drug abuse, failure of an anastomosis after arterial reconstruction or repairs, and arterial graft degeneration [7,11], also congenital arterial defect and trauma play a role in the pathogenesis of upper extremity PSA [12]. PSA at the site of hemodialysis fistula commonly forms within the draining venous segment of the fistula over time (cosmetically undesirable) but seldom interferes with the fistula function, but PSA of native arteries have unpredictable natural history which may include expansion, rupture, artery thrombosis, or spontaneous resolution [13]. Differential diagnosis of brachial artery PSA includes pulsating tumors (sarcomas, osteoblastomaes), arteriovenous malformation, lymphadenopathy, lipomas, abscesses and hematoma [10]; but color Doppler ultrasound has characteristic appearances and a sensitivity and specificity of 100% in differentiating PSA from peripheral hematoma and that is what makes it the imaging method of choice allowing non-invasive evaluation of such masses [14,15]. Delayed presentation of brachial artery PSA has been reported in adults in the literature with prominent retained hardware but not in pediatric population [6]. Complications associated with PSA include infection, compartment syndrome, thromboembolic complication in hands and fingers and cutaneous and osseous erosion [1]. But immediate and long-term morbidity include Volkmann ischemia contracture [6]. The potential sequale of missed diagnosis of PSA includes rupture of the aneurysm with hemorrhage, nerve compression with possible permanent neuropraxia [11].

Treatment choices include primary surgical repair and interposition vein grafting especially with arterial defect more than 1 cm. Brachial artery ligation which in children is often associated with compensation through collaterals circulation [6]. But care must be taken not to injure the nerves and veins adjacent to PSA or scar tissue during surgical procedures [12], which is recommended in patients with neurovascular deficit [6]. Some of these operations may result in serious complications such as limb amputation, brachial plexus involvement and flexion contractures [16]. Other alternative approaches include

ultrasound-guided compression obliteration, percutaneous injection of thrombin into the PSA to produce instant thrombosis and endovascular procedures which include exclusion of PSA by covered stent and embolization with detachable ballons and coils [9,13,15,16].

Treatment delay causes hemorrhage, venous edema, cutaneous erosion, and adjacent neurological structure compression [12]. Although the diagnosis of our patient was delayed for 8 years and treated surgically, no immediate or late complications were seen.

Conclusion

This is the first case of spontaneous primary brachial artery PSA in an adult patient who was mis-diagnosed for 8 years, but without any sequel or post-operative complications. Endovascular procedure is another alternative but surgical intervention was the treatment of choice for this case study.

Conflict of interest

There are no known conflicts of interest associated with this publication.

Funding body

There is no financial support for the conduct of the research or preparation of the article.

Ethical statement

We admit that the research was done according to ethical standards.

Informed consent

We admit that the patients agreed to participate in the research.

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