G Chir Vol. 33 - n. 11/12 - pp. 383-386
November-December 2012

clinical practice

Arterial entrapment syndrome in the cubital fossa: a rare cause of acute stress-related arterial thrombosis in a patient with brachial artery duplication

F. DE SANTIS[1], G. MARTINI[1], N. DECAMINADA[2], G. MANI[1]

SUMMARY: **Arterial entrapment syndrome in the cubital fossa: a rare cause of acute stress-related arterial thrombosis in a patient with brachial artery duplication.**

F. DE SANTIS, G. MARTINI, N. DECAMINADA, G. MANI

Arterial entrapment syndrome (AES) at elbow level is very rare and to our knowledge no case of AES by lacertus fibrosus in the cubital fossa in presence of brachial artery duplication has been described to date. We describe a rare case of acute arterial thrombosis of one of two brachial arteries highlighted in the cubital fossa which developed after strenuous right elbow flexor muscle activity and hyper-extensions presumably related to AES by lacertus fibrosus at elbow level.

A 43-year-old right-handed woman, experienced paleness, coldness and numbness of the right hand, after 8 consecutive hours of gardening. As she worked, her ipsilateral flexor elbow muscles remained in prolonged and inappropriate tension. Clinical examination evidenced the absence of radial artery pulse in the wrist and mild hypothermia in the second and third finger. During surgical exploration two anastomosed brachial arteries were detected in the cubital fossa under the lacertus fibrosus. The lateral superficial brachial artery was occluded. Intraoperative arteriography evidenced brachial artery duplication at the third superior of the arm and normal vascular pattern at the forearm level. In cases of unexplained atypical intermittent upper extremity claudication or acute ischemic symptoms an AES should always be ruled out, particularly when symptoms are exacerbated by strenuous upper extremity activity or when upper limb muscular hypertrophy is evident. In these cases a thorough dynamic clinical and instrumental examination is mandatory to confirm a diagnosis of AES and to avoid possible future ischemic complications.

RIASSUNTO: **La sindrome da entrapment arterioso a livello della fossa cubitale: un raro caso di trombosi arteriosa acuta da stress in un paziente con "duplicazione" dell'arteria brachiale.**

F. DE SANTIS, G. MARTINI, N. DECAMINADA, G. MANI

La sindrome da entrapment arterioso (SEA) a livello della fossa cubitale è molto rara e, a nostra conoscenza, nessun caso di SEA da parte del lacerto fibroso in presenza di "duplicazione" dell'arteria brachiale è stato fino ad oggi descritto. Si riporta un raro caso di trombosi arteriosa acuta di una di due arterie brachiali presenti a livello della fossa cubitale, sviluppatasi dopo intensa attività muscolare (con gomito in flessione e in iperestensione) presumibilmente legata a SEA da parte del lacerto fibroso.

Donna di 43 anni destrimane riferiva, dopo 8 ore consecutive di giardinaggio, pallore, sensazione di freddo e intorpidimento della mano destra. Durante l'attività lavorativa, i muscoli flessori del gomito erano rimasti in prolungata ed inappropriata tensione. L'esame clinico mostrava assenza di polso radiale e ipotermia del secondo e terzo dito. Durante l'esplorazione chirurgica venivano rinvenute, al di sotto del lacerto fibroso, due arterie brachiali anastomizzate l'una all'altra nella fossa cubitale. L'arteria brachiale laterale, più superficiale, era occlusa. L'arteriografia intraoperatoria confermava la "duplicazione" dell'arteria brachiale al terzo superiore del braccio con un normale pattern vascolare a livello dell'avambraccio. In casi di claudicazione intermittente atipica degli arti superiori o sintomi di ischemia acuta dovrebbe sempre essere sempre esclusa una SEA, soprattutto quando i sintomi sono aggravati dalla intensa attività degli arti superiori o quando è evidente una ipertrofia muscolare dell'arto superiore stesso. In questi casi è indispensabile eseguire un'accurata valutazione "dinamica" clinica e strumentale al fine di confermare la diagnosi di SEA ed evitare possibili complicanze ischemiche.

KEY WORDS: Arterial entrapment syndrome - Brachial artery anomalies - Stress-related acute arterial thrombosis - Surgery.
Sindrome da "entrapment" arterioso - Anomalie arteria brachiale - Trombosi arteriosa acuta da stress - Chirurgia.

Introduction

The majority of reported cases of entrapment syndrome in the forearm and elbow involve neurological structures, while arterial entrapment syndrome (AES) is rare (1-3). The overall prevalence of forearm arteries entrapment has been estimated as low as 1% (4), while AES

Bressanone Hospital, Bressanone, Italy
[1] Department of Vascular Surgery
[2] Department of Radiology

in the cubital fossa has been only sporadically described (1-3, 5); to our knowledge no case of acute local stress-related arterial thrombosis due to AES in presence of brachial artery duplication at elbow level has been described to date.

We report a case of acute segmental thrombosis of one of two brachial arteries highlighted in the cubital fossa, which developed after strenuous upper extremity activity and hyper-extensions, apparently related to prolonged arterial entrapment, stretching and compression by lacertus fibrosus (biceps brachii aponeurosis) at elbow level. The possible etiopathogenetic mechanisms of this singular case of acute AES in the cubital fossa are extensively discussed.

Case report

A 43-year-old right-handed woman, after 8 consecutive hours of gardening, experienced paleness, coldness and numbness of the right hand. As she worked, her ipsilateral flexor elbow muscles remained in prolonged and inappropriate tension with frequent elbow and upper extremity hyper-extensions. The symptoms disappeared in about six hours. Clinical examination six weeks later evidenced the absence of radial artery pulse in the wrist and mild hypothermia in the second and third finger. CW Doppler showed monophasic flow in the radial artery. No other preoperative investigations were performed and arterial thrombo-embolectomy was recommended. During surgical exploration two anastomosed brachial arteries in the cubital fossa were detected under the lacertus fibrosus. The lateral superficial brachial artery was occluded for about 2,5 cm, while the medial, smaller brachial artery was patent. The radial artery originated from the superficial brachial artery; while the ulnar artery from the medial, smaller brachial artery (Fig. 1). Thromboembolectomy resulted difficult because the thrombus are firmly attached to the vessel wall. Intraoperative arteriography showed brachial artery duplication at the third superior of the arm (Fig. 2) and normal radial, ulnar and intraosseous vascular pattern at the forearm level (Fig. 3). A postoperative MR angiography control showed patency of both brachial arteries at arm and elbow level and clearly evidenced the anatomical proximity between the distal insertion site of the biceps brachii aponeurosis and the anomalous superficial brachial artery (Fig. 4). No other vascular or musculotendinous anomalies were observed. The postoperative course was uneventful and echo-color-Doppler evaluation of the contra-lateral upper extremity evidenced normal vascular tree anatomy. Ecocardiography and Holter electrocardiography excluded both cardiac arrhythmias and other possible cardiac embolic sources. A thorough hematologic screening did not evidence significant thrombo-embolism risk factors. The patient was discharged under anticoagulation therapy and at 12 months no arterial thrombosis recurrence was observed.

Discussion

The first case of brachial artery entrapment syndrome at elbow level was reported in a muscular thirty-nine year old man in 1977 (6). After this first report only isolated cases of AES in the cubital fossa have been described. Both upper limb muscle and lacertus fibrosus hypertrophy (1, 2, 6) and anatomical musculotendineous anomalies (3-5, 7) have been considered capable of promoting vascular compression and entrapment in the forearm and cubital fossa. In 1984 Biemans (1) described eight cases of brachial artery entrapment syndrome as a result of muscular hypertrophy in absence of congenital mu-

Fig. 1 - Intraoperative findings. Two anastomosed (thin arrow) brachial arteries in the middle of the cubital fossa. Lateral SBA is located in proximity of the residual fibers of the "lacertus fibrous" which is partially retracted deep into the surgical wound. At this level, acute arterial thrombosis developed and the thromboembolectomy was performed (large arrow). Medially, the BA is clearly evidenced. The RA originates from the SBA and UA from the BA. SBA, superficial brachial artery; BA, brachial artery; RA, radial artery; UA, ulnar artery.

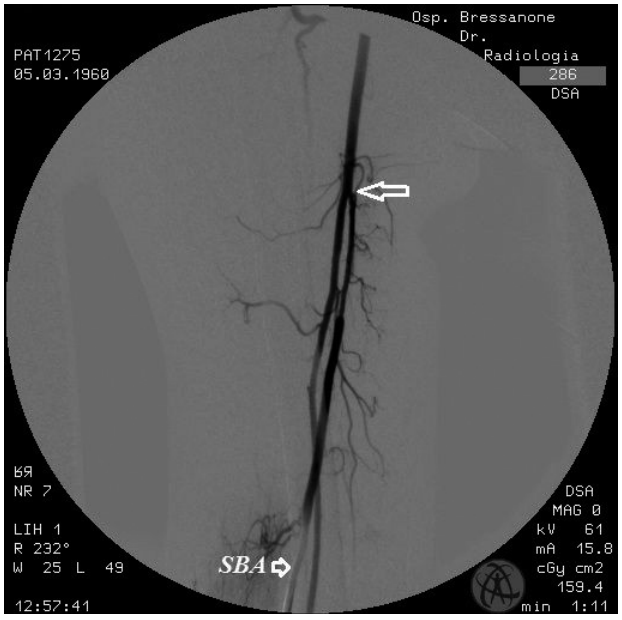

Fig. 2 - Intraoperative arteriography. Brachial artery duplication (arrow) at the third superior of the right arm.

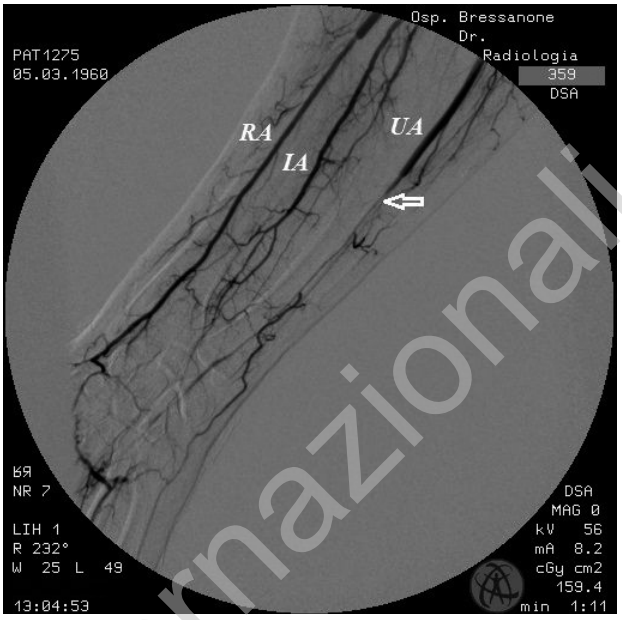

Fig. 3 - Intraoperative arteriography. Normal radial, ulnar and intraosseous vascular pattern at the forearm level; a Fogarty-related UA spasm is evident (arrow). RA, radial artery; UA, ulnar artery; IA, intraosseous artery.

scle or tendon anomalies. Ten years later Bassett et al. (2) reported five similar cases of intermittent brachial artery compression by lacertus fibrosus, exacerbated by strenuous upper extremity activity. In all these cases, releasing biceps brachii aponeurosis restored normal pulses (1,2). Recently Chemla et al. (4) reported two cases of forearm arteries entrapment followed by recurrent angioaccess thrombosis due to arterial compression at elbow level from the flexor digitorum fibrosous arcade in one case (8) and an anomalous fibrous band arising from the pronator teres muscle in the other (9).

In our opinion, the above reported case of stress-related acute arterial thrombosis in the cubital fossa is the consequence of prolonged anomalous superficial brachial artery entrapment and compression by Biceps brachii aponeurosis at elbow level. In this regard several reports have proven the critical role of Biceps brachii muscolotendineous insertions in both nervous and *AES* development in the cubital fossa. In 1991 Spinner et al. described a case of median nerve entrapment due to accessory bicipital brachii aponeourosis (10) while, more recently, Mahato reported a case of bilateral median nerves and brachial arteries entrapment due to an additional biceps brachii muscle origin (5). Finally, in 2009 Clark et al. reported a case of critical brachial artery compression from the bicipital aponeurosis after swelling in a supracondylar fracture (11).

Moreover, it has been proven that posture of the upper extremities may also promote *AES* in the cubital fossa, specifically in particular anatomical and local stress-realated conditions (3,4,9). In the above reported case arterial thrombosis occurred after strenuous upper extremity activity with the elbow flexor muscles in excessive and protracted tension and frequent elbow and upper extremity hyper-extensions. In this regard, Tahila et al. described a case of upper extremity ischemia occurring in a 25-year-old woman during upper limb hyperextension in presence of anomalous insertion of the pronator teres muscle (3).

In the reported case brachial artery variation may also have had an important role in acute arterial thrombosis development. Brachial artery anomalies are frequently described in anatomical studies (12,13) and not infrequently encountered in clinical practice (14). In this case a brachial artery duplication in the third superior of the arm with vessels anastomosis in the cubital fossa was detected. This anatomical variation, clearly described in a morphological study performed in 1995 on 150 routine anatomical dissections of the upper limbs, is related to the total or partial persistence of the superficial arterial segments of the upper extremities (13). As documented in the postoperative Angio-MR control the anatomical location of the anomalous superficial brachial artery (adjacent to biceps brachii aponeurosis), in our opinion could have significantly facilitated arterial entrapment and compression by lacertus fibrosus in the cubital fossa (Fig. 4).

Finally, it should be underlined that in this case no evidence of muscolo-tendineous or nervous anomalies, or other causes of cardiac or peripheral thrombo-em-

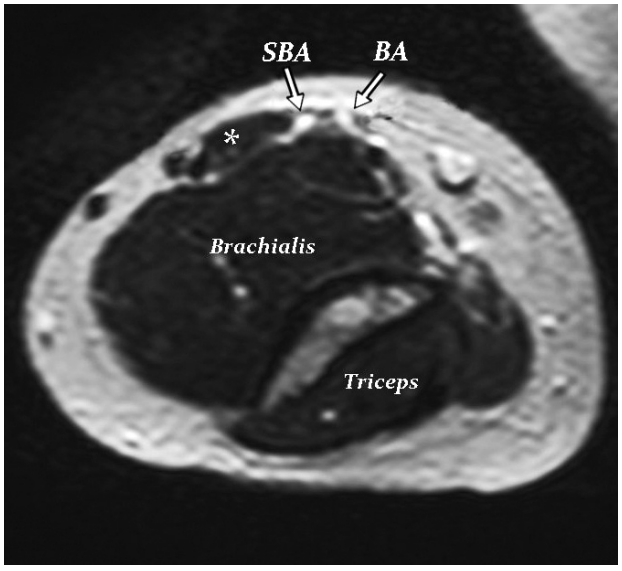

Fig. 4 - Postoperative angio-MR control (transverse imaging at elbow level). Both SBA and smaller median BA are patent. Note the anatomical proximity between the distal insertion of the biceps brachii muscle (asterisk) and the anomalous SBA. SBA, superficial brachial artery; BA, brachial artery.

bolisms were detected. Acute arterial thrombosis was therefore most probably the direct consequence of a positional and local stress-related *AES*-mechanism.

Conclusions

In conclusion arterial entrapment by lacertus fibrosus in the cubital fossa may promote impairment of arterial perfusion and rarely, acute arterial thrombosis, especially in particular anatomical and local activity-related conditions. In cases of unexplained atypical intermittent upper extremity claudication or acute ischemic symptoms an *AES* should always be ruled out, particularly when symptoms are exacerbated by strenuous upper extremity activity or when upper limb muscular hypertrophy is evident. In cases of suspected *AES* patients should always be warned to avoid excessive and inappropriate upper limb movements or hyper-extensions. In these cases a careful dynamic clinical and instrumental examination is mandatory to confirm the *AES* diagnosis and to avoid possible future ischemic complications.

Congresses: None

Funding: None

Conflicts of interest: None

Acknowledgements: The authors wish to thank Ms Elena Harwood for the linguistic revision of this paper.

References

1. Biemans RG. The Popeye syndrome - brachial artery entrapment as a result of muscular hypertrophy. Neth J Surg 1984;36,4:103-6.
2. Bassett FH, Spinner RJ, Schroeter TA. Brachial artery compression by the lacertus fibrosus. Clin Orthop Relat Res 1994;370:110-6.
3. Talha H, Enon B, Chevalier JM, L´Hoste P, Pillet J. Brachial artery entrapment: Compression by the supracondylar process. Ann Vasc Surg 1987;1,4:479-82.
4. Chemla ES, Raynaud A, Mongredien B, Combes MA, D´Attellis N, Cardon CF, Julia PL, Toussaint JF, Fabiani JN. Forearm arteries entrapment syndrome: A rare cause of recurrent angioaccess thrombosis. J Vasc Surg 2001;34:743-47.
5. Mahato NK. Entrapment of the median nerve and brachial arteries in the lower arms bilaterally and additional origin of biceps brachii muscle. Case Report. Int J Morph 2010;28,4:1241-44.
6. Biermans RG. Brachial artery entrapment syndrome. Intermittent arterial compression as a result of muscular hypertrophy. J Cardiovasc Surg 1977;4:367-71.
7. Gunther SF, Di Pasquale D, Martin R. Struthers' ligament and associated median nerve variations in a cadaveric specimen. Yale J Biol Med 1993;66:203-208.
8. Shimizu K, Iwasaki R, Hoshikawa H, Yamamuro T. Entrapment neuropathy of the palmar cutaneous branch of the median nerve by the fascia of flexor digitorum superficialis. J Hand Surg 1988;13:581-3.
9. Stal M, Hagert CG, Moritz U. Upper extremity nerve involvement in Swedish female machine milkers. Am J Ind Med 1998;33:551-9.
10. Spinner RJ, Carmichael SW, Spinner M. Partial median nerve entrapment in the distal arm because of an accessory bicipital aponeurosis. J Hand Surg (Am) 1991;16:236-44.
11. Clark D, Astle L, Fergal M, James L. The bicipital aponeurosis may be involved in the anatomical etiology of arterial compromise after swelling in supracondylar fracture. J Orth Trauma 2009;23,10:731-33.
12. Patnaik VVG et al. Branching patterns of brachial artery - A morphological study. J Anat Soc India 2002;5,2:176-86.
13. Rodriguez-Baeza A, Nebot J, Ferreira B, Reina F, Perez J, Sanudo JS, Roig M. An anatomical study and ontogenic explanation of 23 cases with variations in the main pattern of the human brachio-anterobrachial arteries. J Anat 1995;187:473-9.
14. De Santis F, Schiavone M, Chaves Brait CM, De Santis P, V Di Cintio. Management of complex post-traumatic injury of the upper extremity in the presence of early brachial artery branching. Eur J Vasc Endovas Surg 2011;41,4:576, EJVES extra 2011; 21:e17-e20.