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

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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.

KEY WORDS: Arterial entrapment syndrome - Brachial artery anomalies - Stress-related acute arterial thrombosis - Surgery.

Arterial 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...
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 intramuscular 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 eco-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 musculotendinous 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-
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Scler or tendon anomalies. Ten years later Basset 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 fibrosus 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 muscolotendinous 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 aponeurosis (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-related 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 hyper-extension 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 musculo-tendinous or nervous anomalies, or other causes of cardiac or peripheral thrombo-em-
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.

References