An unusual clinical state: true ulnar artery aneurysm in a five-year old girl

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Abstract

We report the case of a 5-year old girl, who presented with a true ulnar artery aneurysm, and the aetiology, clinical presentation and treatment of the disease.

Keywords: True aneurysm • Children • Ulnar artery

INTRODUCTION

Ulnar artery aneurysm may develop for traumatic, atherosclerotic and infectious reasons. Distal ulnar artery aneurysms, although uncommon, have been well described in adults, as a clinical finding, as a part of the hypothenar hammer syndrome. True aneurysms have a sac, which is formed by dilatation of the whole arterial wall and contain elements of internal elastic or muscular fibres [1, 2]. True congenital aneurysms of the ulnar artery, however, are rare. The youngest patient reported in literature with a true ulnar artery aneurysm is a 1-year old male infant [3]. Pseudoaneurysms result from arterial wall disruptions with haematoma into the surrounding tissues and containment by a reactive fibrous capsule [4].

To our knowledge, our case is the youngest patient with a true ulnar artery aneurysm reported in the literature. She was treated by restoring normal anatomy.

CASE

A 5-year old, right-hand dominant, girl presented with a pulsatile mass located in the volar aspect of the right wrist. The mass was incidentally discovered by her mother 2 months before the referral. Upon history-taking from patient’s mother, we learned that the mass had enlarged during the referral. There was no history of trauma, previous cannulation or vascular or connective tissue disease, and her medical history was unremarkable.

On physical examination, the patient appeared comfortable and without any dysmorphic features. The right-hand examination showed a 2 × 2 cm, clearly pulsatile, compressible mass in the volar aspect of the wrist. There was no thrill over the mass. The radial and ulnar arteries were palpable at the wrist, with a normal Allen test result. The Allen test was performed by asking the patient to clench her fist for 1 min while both radial and ulnar arteries were compressed. The radial artery was then released and the time of the return of the pallor in the hypothenar and palmar area was recorded. There was a normal Allen test in the hand with refill of the palmar vasculature in less than 5 s. No signs of finger ischaemia were observed.

Blood examination, including erythrocyte sedimentation rate, and C-reactive protein, showed normal values. The test result for antinuclear antibody was negative. Duplex imaging revealed a saccular ulnar artery aneurysm, which did not contain thrombus. The aneurysm was 2 cm in diameter and 2 cm in length.

The surgical approach was a complete extraction of the aneurysm under general anaesthesia. The proximal and distal parts of the ulnar artery were exposed by vertical incision over the lower part of forearm, and skeletonized (Fig. 1). The aneurysm sac was excised, and this revealed transaction of the ulnar artery. Proximal and distal vascular control was achieved by vascular bulldog clamps after heparinizing both ends of the transected artery. The ulnar artery was repaired by end-to-end anastomosis, using 7/0 polydioxanone (PDS) with interrupted sutures, and the flow was restored on release of the bulldog vascular clamps (Fig. 2). The clamping time of the ulnar artery was approximately 20 min.

Pathological analysis of the aneurysm sac demonstrated a true aneurysm. No evidence of vasculitis was observed.

The patient’s postoperative course was uneventful, and she was discharged home on the second postoperative day. The follow-up period was 3 months. Duplex study on the 10th postoperative day and at the 3rd month showed the patency of the ulnar artery.

DISCUSSION

The diagnostic and treatment algorithms for ulnar artery aneurysm are not well established because the disease is very
rare. A positive Allen test suggests occlusion, stenosis or incomplete development of the superficial palmar arch or distal ulnar artery. In one series, however, the Allen test was negative in 14% of patients with hypothenar hammer syndrome [5].

Although conventional angiography is known to be the most sensitive radiological method for the diagnosis and detection of aneurysms of arterial origin [4], we used duplex scanning, as further radiological examinations were not readily available at our hospital.

Arterial duplex scanning for the diagnosis of ulnar artery aneurysm has many advantages: the tests are not invasive and do not require general anaesthesia in infants. They can delineate and localize the ulnar artery aneurysm with accuracy and they can identify the presence of mural thrombus. They are of great help in the assessment of the adequacy of hand perfusion perioperatively and therefore help in the decision of whether to ligate or to reconstruct the aneurysm [6]. The disadvantage of the arterial duplex scanning is its operator-dependence.

In patients with symptoms of ischaemia or a positive Allen test, we think that further radiological examinations like conventional arteriography, magnetic resonance image angiography and multi-detector computed tomography angiography are necessary. We believe that, for assessing a risk stratification of surgery in children with ulnar artery aneurysms, an interdisciplinary approach involving both a vascular surgeon and a radiologist is needed during the preoperative evaluation.

In our case, general anaesthesia was applied during the surgical procedure. Local anaesthesia might have been problematic from the surgical comfort point of view, because the patient was an infant, and the regional anaesthesia like peripheral nerve blockage was not appropriate due to the lack of technical support in our surgery room.

Surgical options for ulnar artery aneurysms include: arterial ligation (assuming an intact radial/palmar arch), resection of the thrombosed arterial segment or aneurysm with end-to-end anastomosis, and resection and vascular reconstruction with vein or artery graft.

The surgical options for ulnar artery aneurysms depend on the presence of adequate perfusion in the hand after the aneurysm is excluded from the hand circulation. Simple resection is the surgical option if the hand is adequately perfused and the radial artery is intact; however, if the hand perfusion is inadequate, ulnar artery reconstruction is mandatory [7].

We think that the normal anatomy should be restored as far as possible, especially in infants, considering the potential damage of a radial artery in the future.

Conflict of interest: none declared.

REFERENCES