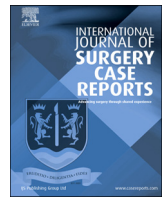


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Posttraumatic pseudoaneurysm of a superficial branch of the ulnar artery: A case report

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ABSTRACT

INTRODUCTION: Hand and Upper limb pseudoaneurysms are uncommon and misdiagnosed. The delayed diagnostic and treatment lead to severe vascular and nerve complications. Many mechanisms are involved like acute injury, chronic micro traumatism of the hand, with specific clinic and para clinic signs.

PRESENTATION OF CASE: The patient was a 30-years old woman, right-handed, admitted at the emergencies for a penetrating hand injury at the junction of the middle third - distal third of the left forearm, palmar surface, with a good radial and ulnar pulse, without sensory or motor deficit. No vessels injuries observed per operatively. A post-operative worsened pain opposite to the scar with a purplish pulsatile swelling appeared after 20 days of complete wound healing. A needle puncture with a red blood contain, motivated an US Doppler revealing a pseudoaneurysm of a superficial artery of the ulnar artery, surgically resected, without complication.

DISCUSSION: Two main mechanisms are involved in upper limb especially hand pseudoaneurysm: penetrating trauma and repeated micro traumatism. Superficial vessels are rarely damaged compare to deep subfascial vessels according to Laplace law. The diagnostic is clinical confirmed with the medical imaging. A pseudoaneurysm is suspected in front of a pulsatile painful tumefaction following a vessel path, with medical imaging in favor. A delayed misdiagnosis lead to a delayed care with severe complications as thrombosis, embolism and vessel. The therapeutic care is mainly surgical.

CONCLUSION: This case reports a delayed diagnostic of posttraumatic pseudoaneurysm of a superficial branch of the ulnar artery, managed with a surgical resection.

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1. Introduction

Upper limb arterial pseudoaneurysm is uncommon, with a non-obvious clinical diagnosis [1]. Subfascial deep arteries are more affected than perifascial and subcutaneous arteries. Pseudoaneurysm is commonly suspected in two clinical situations:

- Direct penetrating or not injury, with a partial, complete or sub-adventitial rupture artery undiagnosed. The false aneurysm results from the hematoma or the partial vessel wall injury. This is a saccular aneurysm [2].
- Repeated micro traumatism leading dysplastic lesion of vessel wall evolving to fusiform aneurysm. This is the main mechanism in the rare Hypothenar Hammer syndrome [3].

The clinical presentation of pseudoaneurysm is a painful and pulsatile mass associated with signs of atypical Raynaud syndrome [3]. The mass can be acute posttraumatic most of the time, or delayed after a complete wound healing [4]. The positive diagnosis is confirmed by medical imaging, either the US Doppler, Magnetic Resonance Angiography MRA, Computed Tomography Angiography CTA or the angiography.

The delayed care leads to severe vascular (thrombosis, rupture, remote emboli) and nerve compression syndrome complications [3,5]. The vascular complications are more important if there is an arterial ulnar pseudoaneurysm thrombosis and a non-existent anastomosis network between the radial artery and the superficial palmar arch. The neurological complications are due to the pseudoaneurysm nerve compression. Robbs and all reported a series of 17 patients presenting a neurological complication following a pseudoaneurysm [6,7].

The upper limb deep arteries are the more affected by pseudoaneurysm (subclavian, axillary, brachial, radial and ulnar arteries), than the superficial arteries which are more vulnerable to penetrating injuries [6,8]. This means that vessels with a small lumen are

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less at risk of pseudoaneurysm according to Laplace's law because they require higher pressures to expand [9]. In addition, these pseudoaneurysms are unlikely to lead to complications with a poor prognosis [8].

Several examples of pseudoaneurysm mechanism are found in the literature. In 1914, Paul Thiebaud described for the first time pseudoaneurysm of the superficial palmar arch, in a series of 24 patients, mainly due to a traumatic injury by glass or non-contending object. Cases with a sperm whale fin etiology on the hand leading to a pseudoaneurysm of the palm of the hand, or chronic irritation of a coachman whose pseudoaneurysm is explained by the repeated impact of the reins and the whip on the palm of his hand, or a locomotive driver with the lever of his machine, or a blacksmith were reported [4]. These presentations were classic at the time but of course scare nowadays.

The treatment is surgical most of the time, with ligation, resection and revascularization by direct suture or vascular bypass. The prognostic of this lesion is affected by the delayed diagnosis or misdiagnosis (post-operative seroma, epidermal cyst, and abscess) with unappropriated care (fine needle aspiration, abscess clearance). Therefore, it is important not to ignore the clinical signs of a pseudoaneurysm, despite its rarity, to avoid complications and inappropriate diagnostic and therapeutic care. We report in line with the SCARE guidelines [10], a case of pseudoaneurysm of a superficial branch of the ulnar artery and present a review of the literature on cases of pseudoaneurysm of the upper limb arteries.

2. Case presentation

The present case is a 30-year-old woman, right-handed, consulted at the emergencies of the Nice University Hospital, for a wound at the junction of the middle third - distal third of the left forearm, palmar surface, with a kitchen knife. The patient had any sensitive or motor deficit, with a well-perceived radial and ulnar pulse.

The wound was explored in the operating theatre on the same day, under loco-regional anesthesia and with the humeral tourniquet. Each musculo-tendinous and nervous structures were intact and there were no visible injuries of the vessels. The wound was sutured with separate non-absorbable sutures.

The post-operative period was straightforward. Healing achieved by day 15 with a localized pain in the wound, and any associated functional signs. The pain progressively worsened on day 20 postoperatively, until objective physical examination revealed a subcutaneous inflammatory mass opposite the scar (Fig. 1). This mass was non-pulsatile and suggested the diagnosis of abscess, hematoma or postoperative seroma. Needle aspiration obtained red blood. There was no dysesthesia of the fingers or atypical, unilateral Raynaud's syndrome. A Doppler ultrasound was performed showing a pseudoaneurysm of a cutaneous perforating artery of the ulnar artery which was confirmed by an MR angiography (Figs. 2–4). The lesion was located 2–3 mm subcutaneously in a perifascial plane. Following this diagnosis, the patient had a second surgery under loco-regional anesthesia with humeral tourniquet.

An under skin pinkish colored mass of 3 cm in diameter was found, which was renitent on palpation with thrombotic contents (Fig. 5). This mass was resected and a ligation of the responsible artery was performed with a. The artery at the origin of the pseudoaneurysm was a perforating artery with cutaneous terminal branches, probably due to the initial trauma. The post-operative period was simple with a good healing. At 3 months post-operative, there was no sign of recurrence (Fig. 6). Anatomopathological findings confirmed the diagnosis of arterial pseudoaneurysm.



Fig. 1. Inflammatory tumefaction opposite the scar at 10 days post operative (* tumefaction).

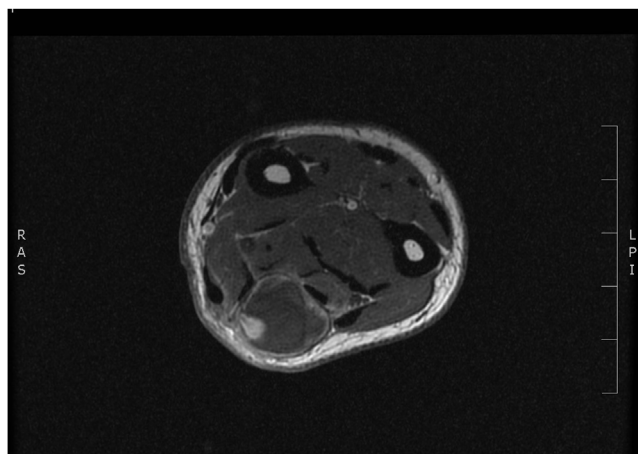


Fig. 2. Axial MRA T1 of the pseudoaneurysm of a superficial branch of the ulnar artery.



Fig. 3. Sagittal MRA T1 of the pseudoaneurysm of a superficial branch of the ulnar artery.

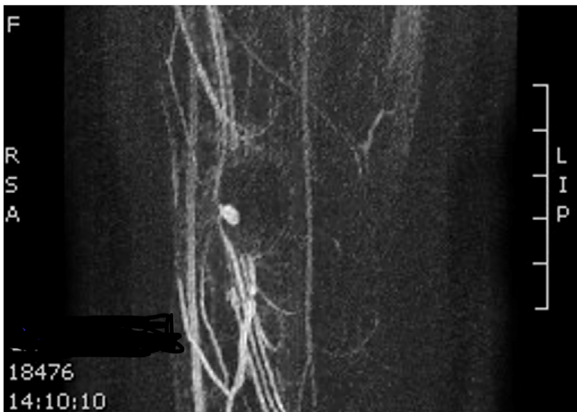


Fig. 4. MRA of the pseudoaneurysm of a superficial branch of the ulnar artery.



Fig. 5. Ulnar pseudoaneurysm with thrombosis.



Fig. 6. Scar at 3 months post operative.

3. Discussion

Upper limb arterial pseudoaneurysm are rare, leading to severe complications. In all ages, these lesions can be seen [8]. It can be clinically suspected before a penetrating or not injuries, painful, pulsatile inflammatory swelling along the arterial path [4]. The pain is usually slow and progressive, but may appear abruptly and be uncomfortable. If the two main arteries of the forearm, radial and ulnar, are compressed, there will be suppression of pulse accompanied with the mass collapse. The notion of direct trauma associated with the appearance of a pulsatile and troublesome mass in the following days may suggest the diagnosis.

Unlike a real aneurysm, a pseudoaneurysm does not affect all layers of the arterial wall. Indeed, a true aneurysm is a damage of the arterial wall, which causes a weakness of the wall with dilatation [9]. A pseudoaneurysm corresponds to the formation of a capsule with a false lumen following a penetrating trauma of the arterial wall. The tear in the wall will constitute an extra-arterial hematoma, organized with a fibrous capsule around the damaged part [5,7]. It is in communication with the lumen of a perforated artery forming the pseudoaneurysm. These are saccular aneurysms. The shape depends on the anatomical arrangement

and the resistance of the neighboring parts. Vessels with a small lumen (< 1 mm) barely leading to aneurysm formation according to Laplace law. Indeed, their formation requires higher pressures to expand. This explains why the occurrence of pseudoaneurysms in small superficial or muscular vessels is rare.

The diagnosis must be evoked in front of the classic triad: palpable, pulsatile and painful mass. The swelling occurs on the initial scar on the path of the artery. It is unique, isolated, well limited, of variable circumscribed volume, renitent and elastic. This mass is reducible and depressible, slightly mobile. Sometimes, for old trauma, the skin can be thinned, red or purplish, preceding ulceration. This aspect is an indication of an imminent rupture [4]. The beats of the mass are isochronous with the pulse, regular, rhythmic and disappear completely when the humeral artery is compressed. The invasion of the aneurysmal sac by blood clots helps prevent the beats from being noticeable. There may be an expansion syndrome, which corresponds to dilatation and retraction. The auscultation found an intermittent isochronous murmur at the systole of the heart.

The peripheral vessels of the upper limbs are vulnerable for the formation of pseudoaneurysms. There are several cases in the literature. These anatomical regions are very exposed to wounds, and vascular injuries of the superficial arteries can be encountered even if in most cases thrombosis of the vessel involved is the rule. The etiology of these pseudoaneurysms is often the result of simple or complex trauma: penetrating wounds, gunshot wounds, stab wounds, and iatrogenic arterial injury (by either osteosynthesis material, cardiac catheterization, or arterial). Puncture wounds (drug addiction), infections, and fractures can also often be involved.

The first case described in the literature was in 1914 in Paul Thiébauld's Thesis. In this work, the author reported cases of pseudo traumatic aneurysms of the superficial palmar arch. The determining causes were the alteration of the arterial layers and the risk of rupture of continuity by a penetrating wound (prickly, sharp instruments or firearms). Similarly, cases have been reported following a facial trauma, or iatrogenic pseudoaneurysm of the superficial temporal artery [11]. It is surprising, given the high incidence of forearm and hand injuries, that pseudoaneurysms are less frequent. This may be explained by the fact that all wounds are now explored for noble organ damage and that systematic repair of the vascular lesions encountered is carried out. The interval between trauma and the formation of the pseudoaneurysm is variable. From a few days to several months, most of the time after the wound has completely healed. The first sign is the appearance of pulsatile swelling opposite the traumatized area.

The clinical presentation can be accompanied with vascular signs such as a sensation of cold fingers, discoloration and trophic disorders, but also complications such as nerve compressions in the upper limb [6,7,12]. Several cases of compressions secondary to a pseudoaneurysm have been reported, the most common being compression of the ulnar nerve in the Guyon Canal. Hunt et al. [7] described this compression as early as 1908. It may be responsible for motor damage with a positive Wartenberg sign, atrophy of the interosseous muscles and ulnar claw, which is resolved after rapid surgical management. Compression of the median nerve in the carpal tunnel and compression of the ulnar nerve in the Guyon canal have been reported following acute rupture of a pseudoaneurysm [13]. These complications can result in significant sequelae such as motor paralysis or sensory neuropathy. The morbidity created by the compression of a nerve caused by a false aneurysm is considerable [6].

Beyond complications due to compression of the surrounding structures, the evolution of these aneurysms can lead to rupture or thromboembolic complications with risk of ischemia or hemorrhagic complications [13]. For this reason, it is important to look

the clinical signs in order to make an early diagnosis and treatment [14].

US Doppler and MRA [2,15] confirm the diagnosis. Ultrasonography characterizes the pseudoaneurysm (size, site, shape).

Saccular formation arises directly from the adjacent artery, and will be diagnosed by 3 signs on ultrasound [16]:

- the neck communicating with the pseudoaneurysm.
- The Yin-Yang [5] indicates bidirectional flow, due to the swirling of blood but in the pseudo aneurysmal cavity.
- To and Fro (back and forth) which corresponds to the reversal of flow in the neck due to the change in pressure gradients during diastole.

Ultrasound should be the first medical imaging in the evaluation of a possible pseudoaneurysm with a specificity of 97% and sensitivity of 94% [16]. MR angiography is useful as a complement to ultrasound [2] and allows for choice and planning of therapy by visualizing normal anatomy and its variants. It asserts the vascular nature of the lesion, the site, the quality of the downstream vascular bed and the supply routes.

These superficial pseudo aneurysmal lesions may be clinically confused most often with postoperative abscesses [14], postoperative hematomas and seroma for which there is no expansion or reducibility syndrome. Other differential diagnoses of these lesions are benign or malignant soft tissue tumors [17] (fibroids, lipomas, giant cell tumors of the synovial sheaths, sarcomas), chondromas and osteomas, synovial cysts for which there is no change in volume if the humeral artery or the forearm arteries are compressed.

Different therapeutic attitudes have been proposed more than 100 years ago [4]: elevation of the limb, refrigeration, flexion, direct or indirect compression, injection of coagulant liquids, electrolysis, Galvano puncture, introduction of foreign bodies or ligature. Today, the treatment of pseudoaneurysms consists of a multitude of options, depending on the characteristics of the lesion, its location and associated lesions [4]: surgical, endovascular, or ultrasound guided.

The main treatment proposed is surgical. Surgical treatment immediately initiated avoid the complications. It consists of resection of the lesion allowing decompression if necessary [18]. This treatment may be associated or not with revascularization depending on the presence of adequate perfusion of the limb and hand [19]. Suture removal and venous bypass to restore vascular anatomy are the techniques used to carry out revascularization [17]. If the hand is sufficiently perfused, simple resection without revascularization is possible, especially if the aneurysm is thrombosed. It is necessary to perform an Allen test as part of the preoperative workup. Surgical treatment with revascularization seems to be the best approach [20] and has a high success rate with minimal morbidity. Nevertheless, simple ligature is possible depending on the location of the aneurysm. Treatment should be early to relieve pain and avoid potential complications such as thrombosis, rupture, or embolism.

Some authors propose endovascular procedures such as angioplasty with stent or stent grafting, or embolization by interventional radiography [5,17]. These alternative options remain controversial [19]. In 2004, Komorowska-Timek et al. reported the successful use of ultrasound-guided thrombin injection in the treatment of a false radial artery aneurysm at 3 cm and another false ulnar artery aneurysm at 2.5 cm resulting from arterial trauma during a failed catheterization [3,5,21]. Others offer direct compression ultrasound-guided treatment [22]. The latter appears to have a low risk and can be attempted. Ultrasound-guided thrombin injection is effective for uncomplicated post-catheterization pseudoaneurysms of the femoral artery with a success rate of 90% [23]. However, experience with thrombin injection is limited and embolization can result in fatal ischemia [24]. The authors favor

surgical treatment. This treatment is still the subject of debate, but surgical management seems to us to be the most appropriate [19,25]. Abstention from therapy also seems possible, with close monitoring if the aneurysm is asymptomatic or small [3]. As soon as there is a painful mass with neurological signs, surgical management is no longer questionable.

4. Conclusion

In case of an arterial wound, one must always think about the risk of aneurysm. The diagnosis of a pseudoaneurysm must be evoked early and should not be ignored, despite its rarity, in front of any pulsatile and painful mass or other evocative signs following a trauma, most often penetrating, because it can lead to complications (thrombosis, emboli, compression of adjacent structures such as nerves and neighboring vessels) with important consequences. Diagnosis allows for early, rapid and appropriate management. Revascularization or surgical ligation are the recommended treatment.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

The ethical committee of the hospital gave the agreement to report this case.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

- Alexandra Maertens: Surgery, writing and proofreading.
- Marie Anne Poumellec: surgery and proofreading.
- Olivier Camuzard: surgery and proofreading.
- Frederica Jessie Tchoungui Ritz: writing and proofreading.
- Thierry Balaguer: writing and proofreading.

All authors have revised and agreed to the submission of this manuscript.

Registration of research studies

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