Case report

Ulnar infected pseudoaneurysm in the course of extended-spectrum beta-lactamase *Escherichia coli* septicemia

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ABSTRACT

We describe the first case of proximal ulnar pseudoaneurysm due to extended-spectrum beta-lactamase-producing *Escherichia coli*. We performed an open ligation of the ulnar artery and partial excision of the aneurysmal sac. This article discusses the etiology and surgical management of ulnar-infected pseudoaneurysms according to the literature data.

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Case report

A 66-year old bedridden man presented with a one-day history of painful, swollen, and red left forearm (Fig. 1). Known to have hypertension and hypercholesterolemia, he has an interventricular communication with a pulmonary artery stenosis. Five years ago, the patient underwent an aortic valve replacement with a biological valve following a *Streptococcus equinus* endocarditis complicated with cerebral embolization, epilepsy, and transient swallowing disorders. Since he was immobilized because of sequellar right hemiplegia, he is prophylactically anticoagulated daily. On examination, he was pyrexial (39.2 °C). An indurated mass was palpated in his proximal forearm. The hand perfusion was good. Palpitation was clearly observed on the radial artery, but not on the ulnar artery. The patient had sensory deficits in his left hand. The blood analysis showed an elevated inflammatory response (435 mg/L) and hyperleukocytosis (20.000/mm³). An enhanced computed tomography revealed an ulnar pseudoaneurysm (Fig. 2). An emergency transesophageal echocardiography was performed and did not reveal any signs of endocarditis. He received a combination of vancomycin and ceftriaxone and was surgically treated. After a skin incision on the palmar face of his forearm, we carefully dissected the muscle and

individualized the humeral artery at the level of its radial and ulnar branches division. We performed a distalia dissection of the fine ulnar artery and the entire aneurysmal circumference. The ulnar artery was safely ligated. The aneurysm was flattened and dark pus was eliminated. Internal thrombus and pseudo-aneurysmal shell samples were collected for bacteriological and anatomopathological testing. Wide debridement of the infected tissue was performed. Lastly, we performed a raphia of the pseudo-aneurysmal shell (Fig. 3). The hand maintained good vascularization because of the radial artery. Regarding the microbiology exams, extended-spectrum beta-lactamase (ESBL) producing *Escherichia coli* were identified from blood culture, per-operative samples, and urinalysis. Therefore, we changed to meropenem 1 g three times a day for a total of fifty days. Repeat cultures were drawn and no persistent bacteremia was detected. PET/CT ruled out the diagnosis of infective endocarditis but sternal osteitis was suspected. Anatomopathological analysis revealed an inflammatory granulation tissue dotted with calcifications. The patient made a good recovery and was discharged.

Discussion

Our patient developed an infected ulnar pseudoaneurysm during a course of ESBL *Escherichia coli* septicemia. Ulnar pseudoaneurysms are extremely rare and mostly induced by a penetrating injury to the vessel, either by trauma or iatrogenically [1,2]. It results from arterial wall disruptions with hematoma into the surrounding tissues and

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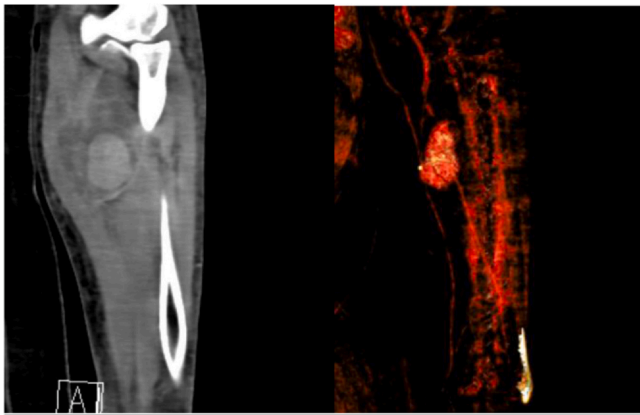
Fig. 1. Initial clinical presentation.

Fig. 2. Pseudoaneurysm described on the CT-scanner of the left forearm.

containment by a reactive fibrous capsule, which was clearly observed by our anatomopathological analysis [3]. Our first hypothesis is that the patient developed a bacteremia from a urinary tract infection promoted by an indwelling catheter. In fact, we found the same bacteria in the urinalysis and in the per-operative samples. Furthermore, the patient reports he had suffered from a severe acute cystitis with hematuria two weeks before his admission, which was treated for two weeks by ciprofloxacin. The bacteria probably seeded into a damaged intima by atherosclerosis or into a previous pseudoaneurysm, which led to an infected pseudoaneurysm. Essentially, it is not excluded that a previous ulnar pseudoaneurysm was already present and promoted the infection. For example, a previous invasive monitoring could injure the vessel and contribute to its infection during the sepsis. Our patient suffered recurrent urinary tract infections while on a long-term catheter with the use of fluoroquinolones, which are risk factors to develop ESBL-producing

Escherichia coli infection. This strain is associated with an increased risk of endovascular infection in extra-aortic sites [4]. Moreover, a link appears to exist between urological processes and infected aneurysms, arising by dissemination of organisms originating from urinary tract infections with overspill to the blood, facilitated by instrumentation and indwelling catheters [5,6]. Few cases described urosepsis as the origin of infective pseudoaneurysm, including reports of pseudoaneurysms due to ESBL *Escherichia coli* infection [4,7,8].

The second infected aneurysm etiology is sternal osteitis, suspected on PET-CT. Our patient underwent cardiac surgery five years earlier but sternal wound hypermetabolism is detected until six months after the surgery [9]. This may be explained by hypercaptation five years later as chronic mediastinitis with sternal osteomyelitis, which are rarely described post cardiac surgery [10,11].

The literature data concerning extremity pseudoaneurysms that developed in the context of chronic sternal infection after cardiac surgery are nonexistent to this day. However, cases of aortic pseudoaneurysm are described by extension of the sternal wound infection and not by embolism or through bacteremia [7,12].

The development of an infected aneurysm in the context of osteomyelitis can be explained by septic embolism or seeding of a preexisting aneurysm through bacteremia.

Two cases describe infected aneurysms that occurred in the course of osteomyelitis. One reports a right subclavian mycotic aneurysm in the setting of chronic clavicular osteomyelitis by septic emboli [13]. The other describes an iliac mycotic aneurysm formation as a result of tibia osteomyelitis [14]. However, aneurysm due to septic emboli are true and not false aneurysm, as in our case.

On the other hand, bacteremia or severe sepsis caused by sternal osteomyelitis post-cardiac surgery as the origin are described [15]. In this context, the pseudoaneurysm must be due to the seeding of preexisting pseudoaneurysm.

Regarding the therapy, there are no randomized trials to guide the management of infected aneurysms. The standard treatment of most infected aneurysms is antibiotic therapy combined with surgical debridement with or without revascularization, depending upon the affected vascular bed and status of distal perfusion [16]. A small retrospective review of patients treated for peripheral infected aneurysms showed that resection or ligation of peripheral infected aneurysms without revascularization was well tolerated. Extensive reconstruction or extra-anatomic procedures are avoided in these infected fields, without the need for revascularization [17]. Most reported cases of ulnar infected aneurysm have been managed surgically. Endovascular techniques are emerging as a treatment alternative [18]. Effective alternatives to surgical intervention include ultrasound-guided compression repair or ultrasound-guided thrombin injection; however, their usefulness in treating ulnar artery pseudoaneurysm is unknown [19]. Non-treatment or delayed
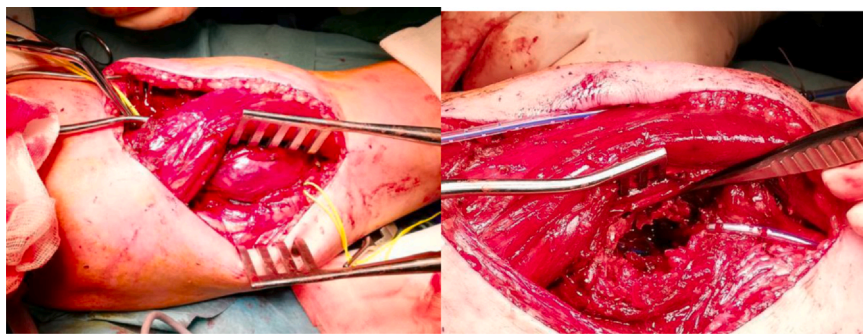

Fig. 3. The fibrous infected capsule were debrided.

treatment of infected pseudoaneurysms often leads to fulminant sepsis, spontaneous arterial rupture, and death [20]. Early diagnosis is important since, without medical and, often, surgical management, catastrophic hemorrhage or uncontrolled sepsis may occur.

Conclusion

We reported the first case of infected ulnar artery pseudoaneurysm due to ESBL *Escherichia coli*. Septicemia in patients with atherosclerosis can lead to pseudoaneurysm, which can be difficult to identify and have serious consequences. The treatment by simple ligation of the ulnar aneurysm is well tolerated.

Ethical approval

This case report is exempt from ethnical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request.

CRediT authorship contribution statement

Scaillet Jessika: Research, Writing. Tessely Heloise: Writing. Cherifi Soraya: Critical revision.

Declaration of competing interest

The authors report no declarations of interest.

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