Infected pseudoaneurysm of the superficial femoral artery in a patient with *Salmonella enteritidis* bacteremia

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Mycotic aneurysms, defined as irreversible dilation of an artery due to destruction of the vessel wall by infection, are rare but are associated with a high risk of rupture if not treated promptly. The case of a healthy 52-year-old smoker who presented with pyrexia, rigors, night sweats and severe right leg pain with swelling is presented. He was diagnosed with a superficial femoral artery mycotic aneurysm, with *Salmonella enteritidis* as the causative agent. He was treated with high-dose antibiotics, local debridement and autologous reconstruction. A high index of suspicion is needed to make the correct diagnosis in these cases. Prompt surgical intervention and antimicrobial therapy are the cornerstones of treatment to reduce the associated high morbidity and mortality.

Key Words: Aneurysm bacteriology; Femoral; Mycotic; Pseudoaneurysm; Salmonella; SFA

Mycotic aneurysms are defined as a localized, irreversible dilation of an artery due to destruction of the vessel wall by infection, which can arise following an infection of a previously healthy artery wall or through secondary infection of a pre-existing aneurysm. The name mycotic aneurysm was coined by Osler (1) to describe aneurysms associated with bacterial endocarditis.

Infected aneurysms are a serious clinical condition that are associated with significant morbidity and mortality. Early diagnosis and treatment of infected pseudoaneurysms are essential to prevent rupture or distal embolization. Treatment consists of antibiotic therapy combined with aggressive surgical debridement of the infected tissue and vascular reconstruction, as needed. Endovascular therapies may have a role in the treatment of a ruptured infected aneurysm, particularly for the treatment of patients at prohibitive risk for open surgery. The current case is presented to illustrate the pathogenesis, microbiology, clinical manifestations, diagnosis and treatment of infected aneurysms.

CASE PRESENTATION

A 52-year-old male smoker with no medical history presented with pyrexia, rigors, night sweats and severe right leg pain with swelling. Physical examination revealed an indurated swelling on the medial aspect of his right thigh. Investigations demonstrated a white blood cell count of $18 \times 10^9/L$, a normochromic, normocytic anemia of 94 g/L, an elevated erythrocyte sedimentation rate of 125 mm/h and a C-reactive protein level of 278 mg/L. A computed tomography (CT) scan revealed a 3.3 cm right superficial femoral artery (SFA) pseudoaneurysm with no evidence of leak and deep vein thrombosis (Figure 1). Radiological appearances suggested the aneurysm was mycotic in origin. There was no history of intravenous (IV) drug use, recent arterial catheterization or immunosuppression. Two weeks earlier, he had experienced an episode of gastrointestinal upset, which was self-limiting. *Salmonella enteritidis* sensitive to IV ceftriaxone was grown in blood cultures. The aneurysm was treated successfully with in situ reconstruction using autogenous superficial femoral vein from the contralateral limb and wide tissue debridement. The excised aneurysm tissue was sent for culture and *S enteritidis* was

Un pseudoanévrisme infecté de l'artère fémorale superficielle chez un patient ayant une bactériémie à *Salmonella enteritidis*

Les anévrismes mycosiques, définis comme la dilatation irréversible d'une artère causée par une destruction de la paroi découlant de l'infection, sont rares mais s'associent à un risque élevé de rupture s'ils ne sont pas traités rapidement. Les auteurs exposent le cas d'un fumeur en santé de 52 ans qui a consulté à cause d'une pyrexie, de frissons, de sueurs nocturnes et de graves douleurs à la jambe droite accompagnées d'un œdème. Il a reçu un diagnostic d'anévrisme mycosique de l'artère fémorale superficielle causée par une *Salmonella enteritidis*. Il a reçu de fortes doses d'antibiotiques, un débridement local et une reconstruction autologue. Il faut un indice de présomption élevé pour poser le bon diagnostic. Une intervention chirurgicale et une thérapie antimicrobienne amorcées rapidement sont la pierre angulaire du traitement pour réduire la morbidité et la mortalité élevées qui s'associent à ce problème.

Figure 1) A computed tomography angiogram axial image illustrating the right superficial femoral artery pseudoaneurysm measuring 3.3 cm. The left leg is shown for comparison

isolated. The patient was treated for six weeks with high-dose IV antibiotics and remained well at follow-up after two years.

DISCUSSION

Mycotic aneurysm, defined as a localized, irreversible arterial dilation due to destruction of the vessel by infection, is a rare entity comprising only 0.9% of total aneurysms (2,3). A primary mycotic aneurysm arises following infection of a previously normal arterial wall, whereas infection of a pre-existing aneurysm is defined as a secondary mycotic aneurysm (2). There are several proposed mechanisms for infection of an arterial wall: septic emboli to the vasa vasorum, bacteremic seeding of the arterial wall, trauma causing direct bacterial inoculation and contiguous infective focus extending to the arterial wall (2).

In a series of 180 patients with mycotic aneurysms, the femoral artery was the most common location (38%), followed by the abdominal aorta (31%), superior mesenteric (8%), brachial (7%), iliac (6%) and carotid arteries (5%) (4). The etiology of mycotic aneurysms has been changing, from endocarditis being the most common cause before the antibiotic era, to arterial trauma in the postantibiotic era (4). This is believed to be due to increased intravascular drug use and

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catheters for intravascular monitoring. Femoral mycotic aneurysms, in particular, are associated with increasing percutaneous arterial access procedures and IV drug use (5). However, neither of these risk factors were present in the current case and there was no known history of immunosuppression, malignancy or bacterial endocarditis.

Patients with mycotic aneurysms of the SFA may present with a pulsatile groin mass, signs of localized infection in the groin, hemorrhage or thrombosis (5). In a study of 57 infected femoral pseudoaneurysms occurring in IV drug users, 83% of patients presented with pain (6). Edema and erythema were present in 80% and 78% of the patients, respectively, whereas fever was reported in only 30% of the patients. Our patient presented with severe leg pain, medial thigh swelling and fever. Other features, as reported in a series of 52 upper and lower extremity mycotic aneurysms in IV drug users, include audible bruit (50%), leukocytosis (70%) and hemoglobin levels <120 g/L (56%) (7). In two different series, blood cultures were reported positive in 46% and 61% of patients with mycotic aneurysms and organisms were isolated from the aneurysm or surrounding tissue in 73% and 84% of the patients, respectively (7,8). In our patient, leukocytosis of $18 \times 10^9/L$ and a hemoglobin level of 94 g/L were present, in addition to positive blood and tissue cultures for *S. enteritidis*.

Diagnosis can be confirmed by ultrasound, CT scan, angiography or magnetic resonance angiography. Air within the aneurysm, local inflammation, contained rupture or saccular or lobulated aneurysms are some radiological findings evident on tests (9). The diagnosis in the present case was confirmed with CT that demonstrated a 3.3 cm SFA pseudoaneurysm.

The bacteriology of mycotic aneurysms has been undergoing a constant change. Initially enterococcus, streptococcus and pneumococcus were responsible for the majority of mycotic aneurysms secondary to endocarditis in the preantibiotic era (4). Brown et al (4) reported a change in etiology to *Staphylococcus aureus* (28%) and *Salmonella* (15%) as the use of broad-spectrum antibiotics, immunosuppression and arterial puncture from vascular procedures and IV drug use has increased. Recently, a series of 26 patients by Brossier et al (8) showed the majority of mycotic infections were related to atypical bacteria, including *Campylobacter fetus*, *Listeria monocytogenes*, *Mycobacterium tuberculosis*, *Coxiella burnetii* and *Aspergillus* species. This trend may be attributed to improved diagnostic tests in detecting microorganisms and increasing numbers of patients with immunosuppression related to cancer, immunosuppressive therapy or long-term steroid therapy (8).

Salmonella are flagellated Gram-negative, facultatively anaerobic, nonspore-forming bacilli of the family *Enterobacteriaceae*. They can be broadly categorized into two forms, typhoidal and nontyphoidal. The two typhoidal strains *Salmonella typhi* and *Salmonella paratyphi* are known to cause enteric fever characterized by fever and abdominal pain. Nontyphoidal *Salmonella* such as *S. enteritidis*, on the other hand, cause self-limited acute gastroenteritis and cardiovascular complications (10,11). The likely etiology for the mycotic aneurysm in the present case was *S. enteritidis*-induced acute gastroenteritis two weeks before his presentation, which was then complicated by *S. enteritidis* bacteremia and subsequent endovascular infection.

The pathogenesis of *Salmonella* causing endovascular infection involves three main steps. First, the bacteria use fimbriae to attach to intestinal epithelial cell membranes. Once phagocytosed in the submucosal space by lymphoid tissue, *Salmonella* can be disseminated via the hematogenous route or lymphatic system. The final step is progression from bacteremia to endovascular infection, the mechanism of which is still unclear (12,13). Nontyphoidal *Salmonella* gastroenteritis is known to cause bacteremia in up to 8% of patients, with immunodeficiency the major risk factor (10,13). Of patients with *Salmonella* bacteremia, 10% to 25% older than 50 years of age develop endovascular infections (14,15). Persistent or high-grade bacteremia in a patient with a history of atherosclerotic disease increases the risk of endovascular infection (10). Because our patient was a known smoker, he likely had atherosclerotic disease that facilitated endovascular infection from *Salmonella* bacteremia and subsequent development of the mycotic aneurysm.

Surgical treatment of femoral mycotic aneurysms is controversial, with studies supporting ligation with debridement alone, ligation with revascularization and routine revascularization (16). The use of stent-grafts is less invasive but the outcome is unproven in the setting of infected aneurysms. The patient in the present case was treated successfully with in situ reconstruction using autogenous superficial femoral vein from the contralateral limb and wide tissue debridement. The use of bactericidal antibiotics together with early surgical intervention and long-term suppressive antibiotic therapy has led to improved survival and decreased amputation rates (5,7,17). High-dose bactericidal therapy should be maintained for at least six weeks and longer if inflammatory biomarkers such as C-reactive protein level, erythrocyte sedimentation rate and white blood cell count do not subside (17,18). Our patient was treated with six weeks of high-dose intravenous antibiotics and remained well at follow-up at two years.

An infected femoral aneurysm is a challenging clinical entity that should be diagnosed and managed promptly because it is associated with high mortality and morbidity, including limb loss. Our clinical experience in the present case highlights that a high index of suspicion should be present whenever an inflammatory mass appears near a large artery even in the absence of previous arterial trauma, IV drug use or immunosuppression, so that timely management is commenced.

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