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Case report

Miliary tuberculosis presenting as bilateral superficial femoral artery mycotic aneurysm in an immunocompetent patient

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A B S T R A C T

Mycotic tuberculous aneurysm is extremely rare. The aorta is the main affected artery however; iliac and less commonly femoral arteries can also be affected. In 75% of the cases a contagious focus leads to infection through erosion of the vessel wall; on the other hand direct seeding of the blood vessel wall via the vasa vasorum may occur in 25%. In a large number of patients it may be a manifestation of miliary tuberculosis. In this case report we describe an immunocompetent patient who presented with bilateral superficial femoral artery aneurysms, followed by bilateral testicular swellings and inferior mesenteric artery aneurysm, as a presentation of military tuberculosis. Early diagnosis of such patients is essential for initiation of both medical and surgical treatment in order to avoid catastrophic outcome of rupture and bleeding.

1. Introduction

Mycotic aneurysms comprise a small fraction of all aortic aneurysms but have a high mortality if not managed promptly [1]. In a review of more than 22,792 autopsies performed at Boston City Hospital from 1902 to 1951, aortic aneurysms were noted in 338 cases, of which only one case was of tubercular origin [2]. The aneurysms have a predilection for large vessels like the thoracic and abdominal aorta; however, mesenteric, femoral and iliac arteries can also be affected. Femoral arteries are rarely affected, constituting only 0.8% of all aneurysms [3].The usual implicated pathogens are Staphylococcus and Salmonella but *Mycobacterium tuberculosis* (MTB) can be rarely involved [4]. Historically, the first case of aneurysm due to MTB was reported in 1895 by Kamen, while the first attempt to treat it surgically was made by Herndon and colleagues in 1949 [5]. Brockman described a tuberculous mycotic aneurysm of the femoral artery in 1927 in a 14-year-old boy with Potts disease of the spine [6].Recently, there are increasing reports of *M. bovis* mycotic aneurysm following bladder installation of bacilli-Calmette-Guerin (BCG) for the treatment of superficial bladder cancer [7,8]. MTB requires standard antitubercular drugs including pyrazinamide while *M. bovis* contained in the BCG, are inherently resistant to pyrazinamide. Therefore, accurate species identification is essential since the treatment differs according to the causative organism.

In this case report, we describe a rare presentation of mycotic aneurysm secondary to MTB involving bilateral superficial femoral arteries (SFAs) that was treated with endovascular stenting and antitubercular therapy.

2. Case presentation

A 73- year –old retired civil officer presented to the emergency department with bilateral thigh swellings for 8 months. The swelling initially appeared in the left mid-thigh, progressively increasing without associated redness, tenderness or discharge. Two months later he noticed a similar swelling in the right thigh. Additionally, he had a one year history of ® knee swelling and mild pain. He denied history of trauma, fever or any other body swelling. Systemic review was unremarkable apart from anorexia and weight loss of 7Kg.There was a remote history of animal contact but he did not consume raw milk or undercooked meat.

The vital signs were normal. Local examination revealed bilateral mid-thigh swellings. The swellings were pulsatile, oval in shape 6 × 4 cm and 4 × 4cm in the left and right thigh respectively. There was no erythema, discharge or sinuses. All peripheral pulses were bilaterally palpable with no delays. Rest of the systemic examination was normal. Routine investigations including CBC, chemistry and lipid profile were normal. C-reactive protein was 54 mg/dl, erythrocyte sedimentation rate 64mm/hr. CT scan at presentation to our hospital showed fusiform aneurysms involving both SFA measuring 2 cm in the right and 1.7 cm in the left side. The aneurysm dilatation is surrounded by multiloculated collection about 4 by 3cm which was consistent with a haematoma. In addition there was evidence of right knee joint effusion.

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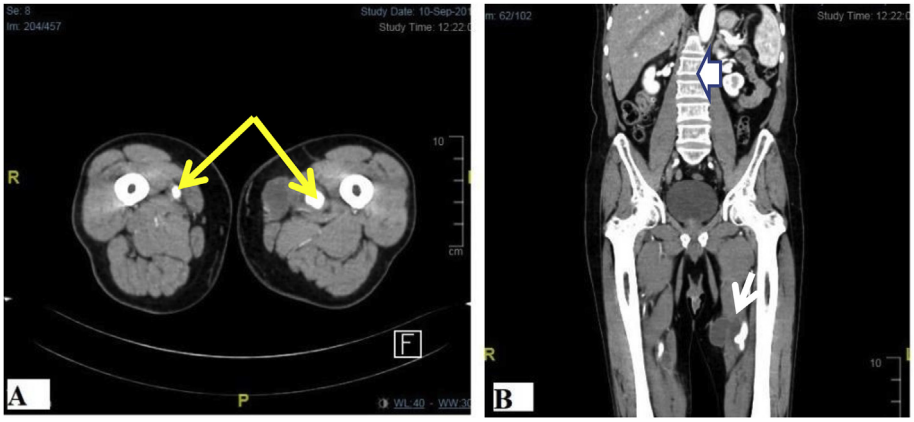

Fig. 1. A. CT angiogram (axial section) both lower limbs showing bilateral fusiform aneurysms involving SFA surrounded by multiloculated fluids (yellow arrows). B. Coronal section both lower limbs showing SFA aneurysms and surrounding fluid collection (white arrow).

The rest of the CT scan of chest, abdomen and pelvis were reported as normal (Fig. 1).

The differentials diagnosis included atherosclerosis, vasculitis, infective endocarditis, mycotic aneurysms secondary to tuberculosis, brucellosis, and syphilis. The autoimmune workup for vasculitis was all negative. Blood culture was sterile including prolonged incubation for brucella. HIV, brucella and syphilis serology were also negative. Both *trans*-thoracic (TTE) and transoesophageal (TEE) echocardiography were normal.

He was planned for surgical correction of the aneurysm, debridement and to obtain tissue for histopathology, microbiology and molecular testing. Unfortunately, the patient rejected surgery but agreed for conservative management. Bilateral stenting of the superficial femoral arteries was undertaken and placement of a flexible, self-expanding endoluminal endoprosthesis with heparin bioactive surface was carried out with preservation of peripheral pulses. The stents consisted of an expanded polytetrafluoroethylene (ePTFE) lining with an external nitinol (titanium). Post operatively the patient remained stable however he started complaining of (L) scrotal swelling and pain. Ultrasound scrotum revealed bilateral epididymitis and two left testicular fluid collections (Fig. 2). Fluid aspirate gram stain and culture did not grow bacterial pathogen. He was discharged home on oral ciprofloxacin.

6 weeks later, he presented with worsening thigh swellings. A repeat CT angiogram showed development of a new pseudo aneurysm at the proximal segment of inferior mesenteric artery. A new enlargement of necrotic aorto-caval lymph nodes was also noted. Multiple collections were observed in the left testis (Fig. 3A and B). There was increase in the size of the multiloculated collection surrounding the femoral aneurysms of both the thighs however no leakage from the stent was identified(Fig. 4). In addition a whole body PET scan confirmed an enlarged (L) adrenal with a hypermetabolic activity.

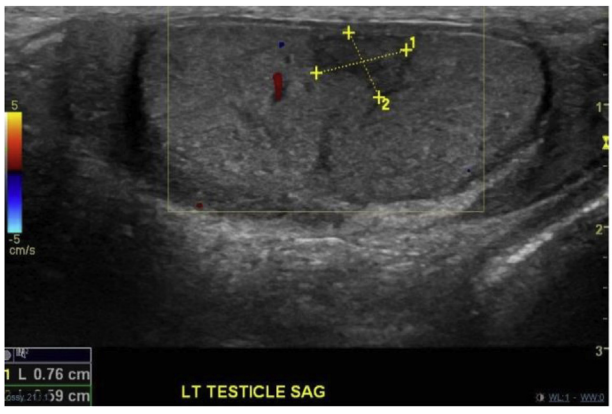

Fig. 2. US of the testis showing (L) testicular abscess measuring 2.5 × 1.6cm.

A reddish brown fluid was aspirated from the femoral perianeurysmal collection. PCR for MTB was reported as positive; fortunately, the gene for rifampicin resistance was not detected. CT Chest showed innumerable miliary nodules particularly in the upper lobes (Fig. 5B). In retrospect the lower CT chest cuts of the initial admission CT angiogram showed more extensive miliary shadows than the current one (Fig. 5A). He was started on Isoniazid 300mg, rifampicin 600mg, ethambutol 1200mg and pyrazinamide 1.5 gm once daily. Eventually cultures of sputum, right and (L) thigh drainage grew fully sensitive MTB.

On follow up after 2 weeks, the patient complained of pain and further increase in the size of femoral swellings. At this point he was prescribed anti-inflammatory agent and advised to continue on his antitubercular medications and attend the infectious disease clinic.

Unfortunately, approximately 6 weeks later he developed a massive gastrointestinal bleeding from which he could not be resuscitated and died.

3. Discussion

Bilateral SFA aneurysm secondary to tuberculosis is an extremely rare complication of mycobacterial tuberculosis. Zhang et al., in 2014 reported 32 aneurysm cases secondary to tuberculosis encompassing the whole arterial system, variably reported from 1993 to 2013. Out of these, only 2 cases involved the femoral vessels [9]. Fortunately rupture of tubercular mycotic aneurysm into the digestive tract is very rare.

Tuberculosis spreads directly by eroding through the vascular wall due to contiguous focus in over 75% of all mycotic aneurysm cases. Cases of tuberculosis spread to paraarterial lymph nodes, lungs, pericardium, or spine (Pott's disease) have been previously reported [10]. Less commonly, haematogenous infection directly from the blood stream or through the vasa vasorum, with bacteria invading the tunica and adventitia of the blood vessels may occur. The subsequent vascular wall thinning will lead to the formation of pseudoaneurysm typical of mycobacterial mycotic aneurysms. Occasionally, infection follows a lymphatic spread. The association of mycotic aneurysm and disseminated tuberculosis is well recognized [11]. In our patient, the most probable mode of spread was haematogenous, as multiple sites of infection, including the lungs, leg, abdomen, testes, and the knee were involved. Additionally, a review of the initial CT images obtained at the time of first presentation showed miliary infiltration. Intriguingly, the infiltrations were shown to have regressed on subsequent CT scan, which we assume was an effect of ciprofloxacin prescribed for epididymo-orchitis.

The mainstay treatment for mycotic aneurysm includes a combination of prolonged antimycobacterial drugs and surgical intervention. Either medical or surgical treatment alone is not sufficient. Eighty-

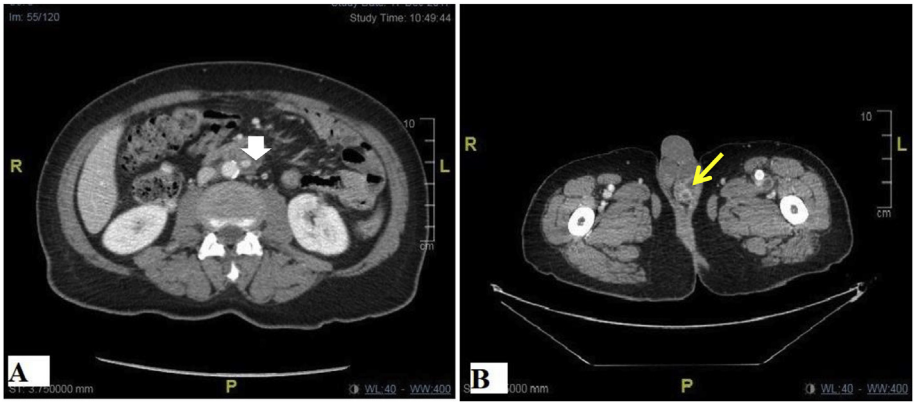

Fig. 3. A. CT scan of the abdomen showing aneurysm of the inferior mesenteric artery (white arrow head). B. CT scan of the lower limbs confirming collection of the left testis (yellow arrow).

seven percent of the patients who were provided antitubercular drugs in addition to surgical intervention survived. On the other hand, there were no survivors among patients who were recommended only one form of treatment or no treatment at all [12]. Remarkably, the mortality in patients with associated military TB can be significantly higher (22.2%) when compared to the rest of the patients (5.2%). On the other hand even elderly patients can have a favourable outcome when a thorough approach for an early diagnosis and treatment is initiated [13].

Both endovascular replacement and open surgery are used as surgical interventions. Classic open surgery is preferred since it eliminates the necrotic tissue and improves response to medical treatment. It is plausible that active anti-tubercular drugs may have decreased access to the aneurysm, and may not penetrate the necrotic tissue and thrombus [12]. However, open repair is invasive and associated with a higher mortality compared to endovascular aneurysm repair (EVAR). Intravascular stenting is less invasive and avoids the complications of surgery. In one review, 42% of patients were managed by intravascular repair and stenting, with a favourable outcome. Additionally, adjunctive drainage of collections and soaking of stents with rifampicin could play a role in stented patients [14].Overall, it is not yet clear which modality of intervention is appropriate in patients with tuberculous aneurysm. The rarity of the condition may not allow for a randomised controlled trial to reach a conclusion, and patients will continue to have to be managed based on individual characteristics. In general, it is suggested that EVAR is used as a bridge to definite surgical intervention or as palliative treatment in debilitated patients with poor surgical risk [15]. Furthermore, Guillermo et al. has mentioned in their article that dacron stents soaked with rifampicin inhibits the growth of common organisms causing this condition, and it is worth considering this option in cases where open repair of aneurysms is performed [11].

The increase in size of the femoral aneurysms noted in this case is noteworthy. We believe it is due to an immune paradoxical response. Enlargement of a pre-existing tubercular lesion or the development of new lesion in patients who are compliant to treatment of a fully sensitive mycobacteria fulfils the criteria for a paradoxical response to medications. It is conceivable that a similar reaction could have led to the formation of a new intraabdominal aneurysm that in turn led to massive gastrointestinal bleeding, as a terminal event in our patient. Post-mortem examination could not be performed to confirm the diagnosis, since our patient was also on non-steroidal anti-inflammatory drugs, which could have led to massive hematemesis. Rupture of tubercular mycotic aneurysm into the digestive tract is very rare, as mentioned before, with only a few cases having been reported previously. A paper reported the case of a 60-year-old male patient who died suddenly following a massive gastrointestinal bleeding; a ruptured aortic aneurysm was detected on autopsy [16]. The risk of rupture is not dependant on the size of the aneurysm and may occur with unpredictable speed [17]. This emphasizes the importance of surgical intervention irrespective of the size of the aneurysm.

This case report highlights the difficulty of reaching an early diagnosis and prompt treatment in cases of mycotic tuberculous aneurysms. The condition of patients receiving antitubercular treatment that paradoxically worsen or develop new lesions may not indicate failure of treatment. Major bleeding could occur irrespective of the size of the aneurysm; therefore, early surgical intervention is required to improve the prognosis.

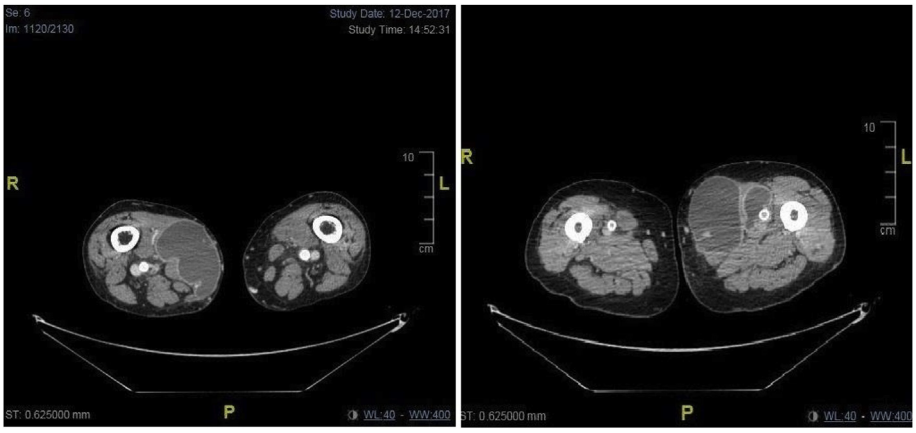

Fig. 4. CT scan angiography of the lower limbs showing increasing SFA aneurysms with bilateral collections.

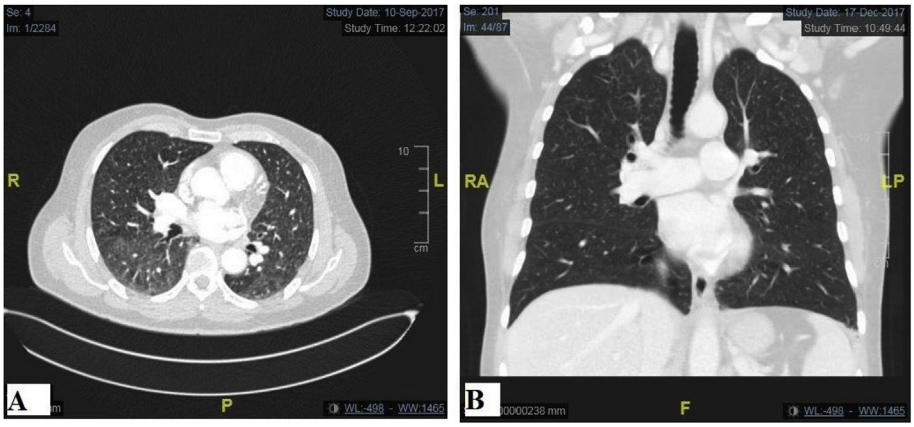

Fig. 5. A. CT scan angiogram on initial admission showing innumerable miliary nodules. B. CT scan of the chest 3 month following initial admission showing similar shadowing but less marked than first CT scan.

Conflicts of interest

No conflict of interest.

Funding

None.

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