

A rare case of mycobacterial pseudoaneurysm of the superficial femoral artery

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Abstract. – OBJECTIVE: Extrapulmonary localization of tuberculosis accounts for about 15-20% of cases. Several cases of *Mycobacterium tuberculosis* with vascular involvement have been described, but only few cases for limb vessels.

CASE REPORT: We report the case of a 33-year-old man from Gambia with a symptomatic pseudoaneurysm of the right superficial femoral artery. Total body positron emission tomography/computed tomography with [18F]FDG revealed an active infection. The patient underwent vascular reconstruction with a straight reversed vein graft. Molecular testing for *Mycobacterium tuberculosis* was non-diagnostic. Cultures of the pseudoaneurysm wall and thrombus removed during surgery grew *Mycobacterium tuberculosis*.

CONCLUSIONS: The diagnosis of vascular tuberculosis infection due to *Mycobacterium tuberculosis* is a challenge. Epidemiology remains the primary criterion for maintaining a high index of suspicion.

Key Words:

Mycobacterium tuberculosis, Femoral pseudoaneurysm, Mycotic aneurysm.

Introduction

Extrapulmonary localization of tuberculosis (TB) accounts for about 15-20% of cases^{1,2}. Medical and surgical management of extrapulmonary TB is a challenge for clinicians and surgeons. Vascular TB is the rarest clinical manifestation of extrapulmonary TB, usually involving the descending thoracic aorta³. There are only a few cases in visceral or peripheral arteries, such as the mesenteric, iliac, and femoral arteries⁴.

Mycobacterium tuberculosis (MTB) is not usually involved in the development of mycotic aneurysms. The first case of MTB-related aneu-

rysm was reported in 1895 by Kamen⁵, while the first attempt at surgical treatment of an MTB-related aneurysm was performed by Herndon et al⁶ in 1949. Since then, several cases of MTB with vascular involvement have been described⁷⁻¹³. The lower aortic segments are rarely involved, as is the case for visceral and lower/upper limb vessels^{14,15}.

We report the case of a young man with an MTB-related superficial femoral artery pseudoaneurysm.

Case Report

A 33-year-old man from Gambia was admitted to the Emergency Department with a symptomatic pulsatile mass in the middle part of the right thigh. The patient denied a history of trauma, fever, or other symptoms. He reported swelling of the right leg during walking. Peripheral pulses were palpable.

A computed tomography angiography of the abdomen and lower limbs revealed a 6-cm partially thrombosed pseudoaneurysm of the right superficial femoral artery. All lower limb arteries were patent.

Laboratory examinations revealed hemoglobin of 13 g/dL, a total white blood cell count of $6.02 \times 10^3/\mu\text{L}$, procalcitonin of 0.10 ng/mL, C-reactive protein of 2.90 mg/L, erythrocyte sedimentation rate of 120 mm/h, and serum creatinine of 0.75 mg/dL. No clinical signs of infection were detected; however, the chest X-ray showed micronodules along the fissures and parenchyma. These findings were confirmed by chest computed tomography angiography that revealed malignant involvement of the right pleura and lymphadenopathy of the mediastinum and mesentery. There was a high suspicion of mesothelioma or TB infection; therefore, the patient underwent a

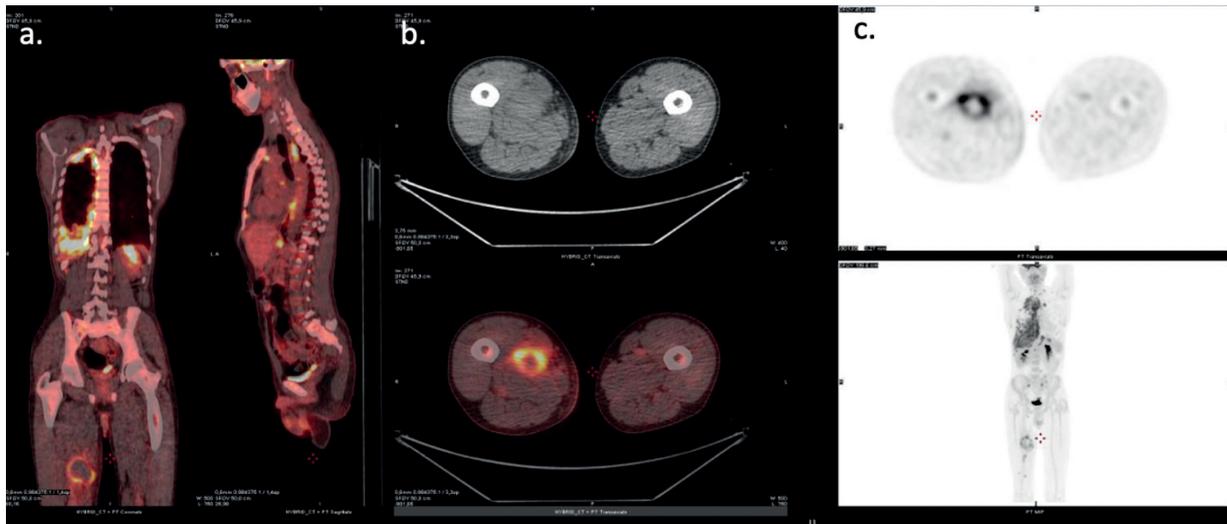


Figure 1. **a**, Superimposed [18F]FDG PET/ CT images in the coronal, sagittal and transaxial view shows large uptake in pleural effusions (SUV max 13), in lung micronodules (SUV max 6.4), in chest lymphadenopathy (SUV max 9.5), cervical and dorsal vertebrae, sacrum and posterior iliac crest (SUV max 10.7). **b**, Superimposed [18F]FDG PET/CT images in the transaxial shows intense radiopharmaceutical uptake at the femoral pseudoaneurysm (SUV max 10) and at the surrounding tissues (SUV max 7.5). **c**, [18F]FDG PET/CT images shows the same intense radiopharmaceutical uptake at the femoral pseudoaneurysm and in the other body regions.

total body positron emission tomography/computed tomography (PET/CT) with [18F]FDG. Intense radiopharmaceutical uptake was detected at the right femoral pseudoaneurysm (SUV max 10) and the surrounding tissues (SUV max 7.5). Moreover, similar uptake was found in a pleural effusion (SUV max 13), lung micronodules (SUV max 6.4), chest lymphadenopathy (SUV max 9.5), cervical and dorsal vertebrae, and the sacrum and posterior iliac crest (SUV max 10.7) (Figure 1).

The patient underwent surgery under general anesthesia with a vascular reconstruction of the right superficial femoral artery. The great saphenous vein was used to perform a straight reversed vein graft with end-to-end anastomoses (Figure 2). Doppler ultrasound was performed at the end of the procedure, and peripheral pulses on the right limb were palpable.

The pseudoaneurysm was sent for histological examination (Figure 3). Signs of erosion of the

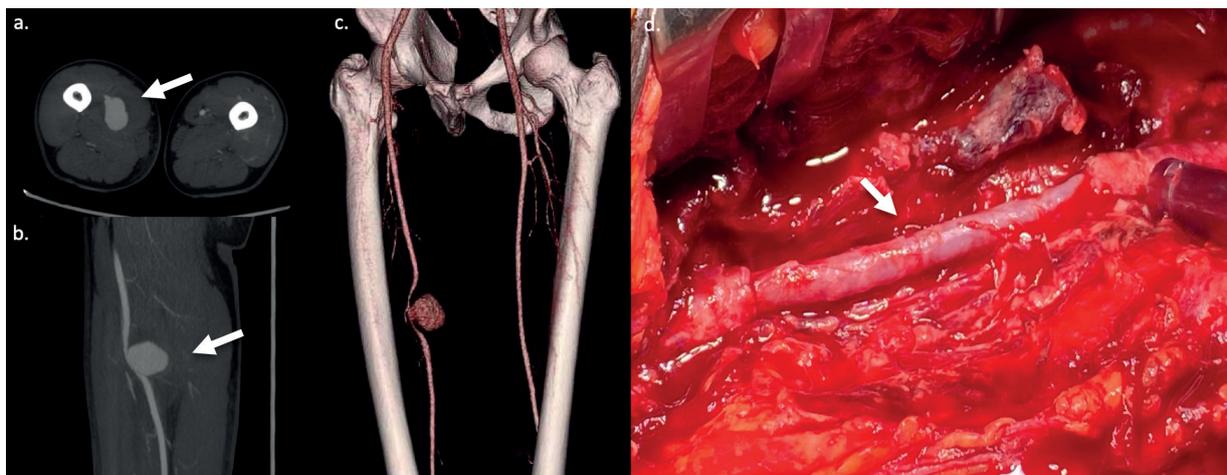


Figure 2. **a-b**, CT images in the transaxial and sagittal view shows the large femoral pseudoaneurysm (thin arrow). **c**, 3D image showing the same right femoral pseudoaneurysm. **d**, intraoperative end-to-end anastomosis between the great saphenous vein (GSV) graft (thin arrow points GSV) and superficial femoral artery.

intima with lymphomonocytic infiltrates were found. The media had several atypical microcalcifications.

The day after the procedure, bronchoscopy and bronchoalveolar washing was performed. The amount of lavage was only sufficient for a direct microscopic examination, which did not reveal atypical cells or bacterial microorganisms. The sputum's direct microscopic examination was negative, as was molecular testing for MTB.

Fine needle aspiration of the pleural lesions was performed. The histological study revealed morphological aspects of chronic giant cell necrotizing inflammation. Molecular testing for MTB revealed an uncertain outcome (GeneXpert MTB/RIF is standardized for respiratory samples only).

Screening for syphilis and HIV were negative, while the Widal-Wright reaction was positive for the O antigen of *Salmonella typhi*. Subsequent blood and stool cultures were negative. An interferon-gamma release assay for MTB complex was positive.

After eleven days, the patient was discharged with the suspected diagnosis of disseminated tuberculosis and was prescribed oral anti-TB therapy with isoniazid (300 mg daily), rifampicin (600 mg daily), ethambutol (1,200 mg daily), and levofloxacin (750 mg daily). The choice of quinolone was necessary due to a temporary lack of availability of pyrazinamide.

During the diagnostic process at the outpatient infectious disease unit, cultures of the pseudoaneurysm wall and thrombus collected during surgery grew MTB, confirming active MTB infection. Susceptibility testing revealed resistance to ethambutol.

Magnetic resonance imaging (MRI) of the brain revealed two lesions in the temporal lobes (maximum diameter 4 mm) suggestive of tuberculomas. Spinal cord MRI confirmed the presence of focal abnormalities of several vertebrae as a sign of spondylodiscitis, consistent with the findings from the PET/CT with [18F]FDG.

Anti-TB treatment was continued with three drugs (isoniazid, rifampin, and pyrazinamide because of ethambutol resistance detection and pyrazinamide availability) for two months (intensive phase), then with two drugs (isoniazid and rifampin) for the continuation phase extended to 10 months, for a total duration of treatment of 12 months. Corticosteroids were added and tapered over eight weeks for central nervous system involvement on MRI and concomitant pericardial effusion detection on echocardiography. A one-

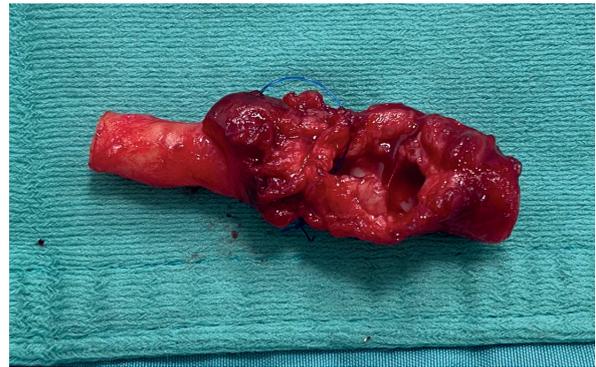


Figure 3. Pseudoaneurysm removed from superficial femoral artery and collected for histological examination.

month follow-up Doppler ultrasound revealed complete patency of the vein graft without restenosis.

One-year follow-up (including chest CT, PET/CT, and brain and spinal cord MRI) showed almost complete resolution of the baseline alterations with only persistence of some lung and pleural lymph nodes and central nervous system calcifications consistent with disease resolution. Laboratory exams revealed complete and stable normalization of parameters of inflammation. The patient is currently asymptomatic and in good health.

The patient gave his consent to the publication of this report. Institutional Review Board approval was waived due to the descriptive nature of the study.

Discussion

MTB-related aneurysms often develop in the thoracic aorta and innominate arteries because of contiguous spreading from pulmonary infection sites^{16,17}. Less frequently, MTB bacilli spread hematogenously in extrathoracic arteries, such as the abdominal aorta and the iliac and peripheral arteries. In these cases, additional active sites of infection are frequently present. When a TB aneurysm is diagnosed, there is usually a source of active disease elsewhere, most commonly in the respiratory tract¹⁰. Mycotic aneurysms of the femoral arteries were first described in 1927 by Brockman¹⁸ and (more recently) in immunocompromised¹⁷ and immunocompetent patients⁸, confirming that individual immunological status is only a concomitant factor in the development of MTB-related aneurysms.

Routine mycobacterial culture of surgical specimens is recommended only for spinal bone samples suspicious for osteomyelitis²⁰. In our case, the decision to include MTB screening of surgical specimens was based on epidemiological criteria. The patient came from Gambia (Western Africa), an area with a high incidence of TB, as estimated by the 2020 World Health Organization TB report.

Amplifying mycobacterial nucleic acids could help detecting MTB due to its rapidity and high specificity. Nevertheless, in our case, this examination was negative, revealing its low sensitivity²¹ and confirming the culture of intraoperative specimens as the diagnostic gold standard despite the long growth time of MTB (i.e., 4 to 8 weeks).

Mechanisms of vascular involvement during mycobacteremia remain unclear. A previous endothelial injury could be a possible pathophysiological mechanism. Indeed, our patient showed a positive Widal-Wright reaction, and it is recognized that *Salmonella typhi* bacteremia could result in mycotic aneurysms. Nevertheless, we cannot sustain the hypothesis that the possible previous bacteremia could have led the patient to develop TB arteritis.

Although many laboratory tests could help clinicians to determine the causes of infection, the management of tubercular pseudoaneurysms is strictly surgical. Nevertheless, medical treatment is mandatory during follow-up.

Conclusions

The diagnosis of vascular TB infection due to MTB is a challenge. Epidemiology remains the primary criterion for maintaining a high index of suspicion. Strict collaboration between infectious disease specialists and vascular surgeons with surgical treatment and antituberculous therapy was mandatory to manage our case successfully.

Authors' Contributions

Raffaella Berchiolli: writing, final revision, final approval. Giulia Bertagna: writing, final revision, final approval. Paola Erba: writing, final revision, final approval. Claudio Caroselli: writing, final revision, final approval. Silvia Fabiani: writing, final revision, final approval. Lorenzo Roberto Suardi: writing, final revision, final approval. Marco Falcone: final revision, final approval. Mauro Ferrari: final revision, final approval. Nicola Troisi: writing, final revision, final approval. All authors read and approved the final version of the manuscript.

Conflicts of Interest

The authors declare no conflicts of interest.

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