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Mycobacterium tuberculosis infection of reverse shoulder arthroplasty: a case report

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Tuberculosis (TB) is still 1 of the top 10 causes of death worldwide, and millions of persons continue to become ill each year.[16] Depending on the country, bone and joint infections account for 10% to 15% of the extrapulmonary TB in Europe and the United States and 15% to 20% in low-income countries.[11] Prosthetic joint infections (PJIs) are even more unusual and are only published as case reports or systematic reviews.[14,15] Although medical treatment is the gold standard in pulmonary TB, the treatment of PJIs is unclear: Medical treatment, débridement and irrigation, and 1- or 2-stage prosthetic revision have been reported with good results in selected cases.[7,10,13-15]

This case report describes a reverse shoulder arthroplasty (RSA) infection due to *Mycobacterium tuberculosis* (MTB) treated with 3 consecutive interventions: 2 débridement procedures with partial implant replacement, which failed, followed by a successful 1-stage replacement in combination with anti-TB therapy.

Case report

A 75-year-old woman presented to our center for right shoulder pain that was temporarily improved by an intra-articular corticoid infiltration. This Moroccan patient with a low socioeconomic background had a history of type 2 diabetes, arterial hypertension, and a left rotator cuff tear treated by palliative biceps tenotomy with good clinical results. Human immunodeficiency virus testing showed a negative result, the patient had no known history of active TB, and the findings of preoperative chest radiography were considered normal. Initial magnetic resonance imaging revealed a massive rotator cuff tear with stage 3 retraction of the supraspinatus and infraspinatus tendons.[8] Nonspecific calcified axillary lymph nodes were also described on the examination and seen on a right shoulder radiograph (Fig. 1). An arthroscopic palliative biceps tenotomy was performed.

The patient had temporary pain relief, but a pseudoparalytic shoulder developed without passive range-of-motion limitations. A computed tomography (CT) scan revealed a shoulder arthrosis, calcified axillary formations, and parenchymal sequela images in the right upper lobe (Fig. 2). An RSA associated with a concurrent latissimus dorsi transfer was performed.

Institutional review board approval was not required for this case report.

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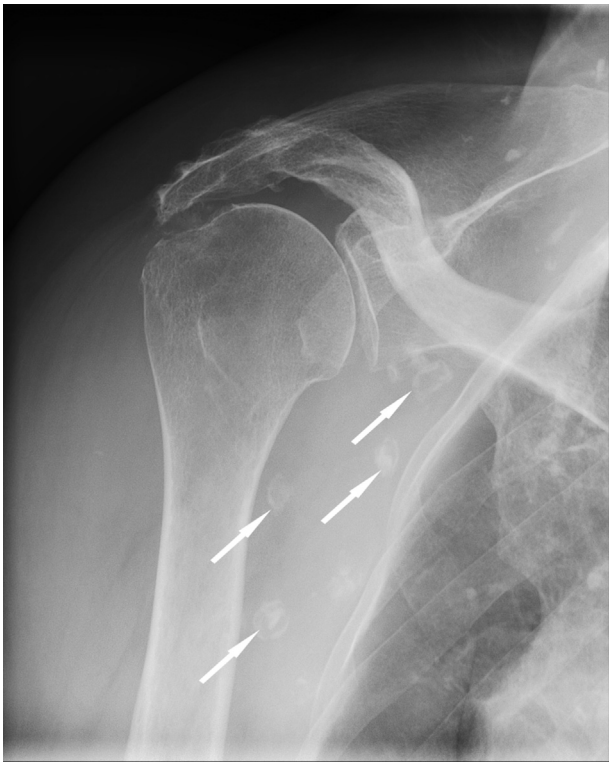

Figure 1 Calcified axillary lymph nodes (*arrows*) on right shoulder radiograph.

Three weeks after the RSA, the patient presented to our center with painful swelling of the right shoulder with low inflammatory marker levels; the C-reactive protein (CRP) level was 11 mg/L (normal level < 6 mg/L). The radiographic control showed an anterior prosthesis dislocation. A subcutaneous purulent collection communicating with the prosthesis was surgically drained and sent to the microbiology laboratory. The uncemented stem was removed without difficulty, and the glenosphere was changed without increasing its size.

Samples cultured in the usual medium remained sterile. An unfavorable clinical evolution at 2 weeks led to a new CT scan, showing 2 fistulized collections of 5 × 3 × 3 cm and a superior dislocation of the prosthesis; the CRP level was 8.5 mg/L. Irrigation and débridement with new bacteriologic samples associated with an increase in the glenosphere's size and of the polyethylene were performed. The usual cultures remained sterile but were positive for a susceptible strain of MTB in Löwenstein-Jensen and Coletsos media. A combination of rifampin, isoniazid, and pyrazinamide was administered orally for 2 months, followed by a rifampin-isoniazid combination for 7 months.

Two months later, the patient came back to the emergency department for a persistent fistula; the CRP level was 11 mg/L. An antibiotic-free period of 1 month preceding a revision with new microbiological sampling of the prosthesis was decided after a multidisciplinary discussion. The humeral stem and the glenosphere were changed, but the metaglene and its screw were left in place. A rifampin-resistant strain of *Staphylococcus epidermidis* considered a contaminant was found in the samples. Cultures remained positive for MTB, leading us to continue the anti-TB treatment.

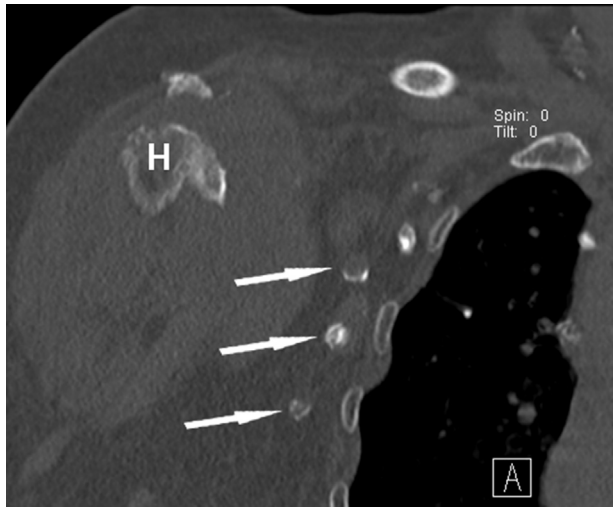

Figure 2 Coronal view of right shoulder computed tomography scan, showing calcified axillary formations (*arrows*). *H*, humerus; *A*, right lung.

Three months after the last surgical intervention, the patient had shoulder pain with an inflammatory scar and an elevation in the CRP level to 50 mg/L. Joint aspiration was performed, which was negative on culture including for MTB, the treatment of which was conducted for 9 months as scheduled initially.

Six months after the end of the anti-TB treatment, the patient had increasing pain with a noninflammatory scar and a normal CRP level, at 4 mg/L; the radiographic control found a new anterior prosthetic dislocation. After a new CT scan, a complete change of the implants was performed (Fig. 3). The new bacteriologic samples were positive (2 of 5) for *Cutibacterium acnes,* treated with a combination of clindamycin and doxycycline for 4 months, and were negative for MTB. One year after the last surgical procedure, the patient had no pain with a noninflammatory scar and had a good range of motion with elevation of 90° with no inflammatory markers.

Discussion

Prosthetic shoulder infections remain difficult to diagnose despite microbiological advances in this field.[12] The most frequently isolated pathogen is *C acnes*,[9] a slowly developing germ with poor clinical manifestations.[1]

Although prosthetic loosening always needs a replacement surgical procedure, implant revision is still debated for all types of germs and joints, especially in MBT infections. The result depends on the involved joint: Treatment with débridement, antibiotics, and implant

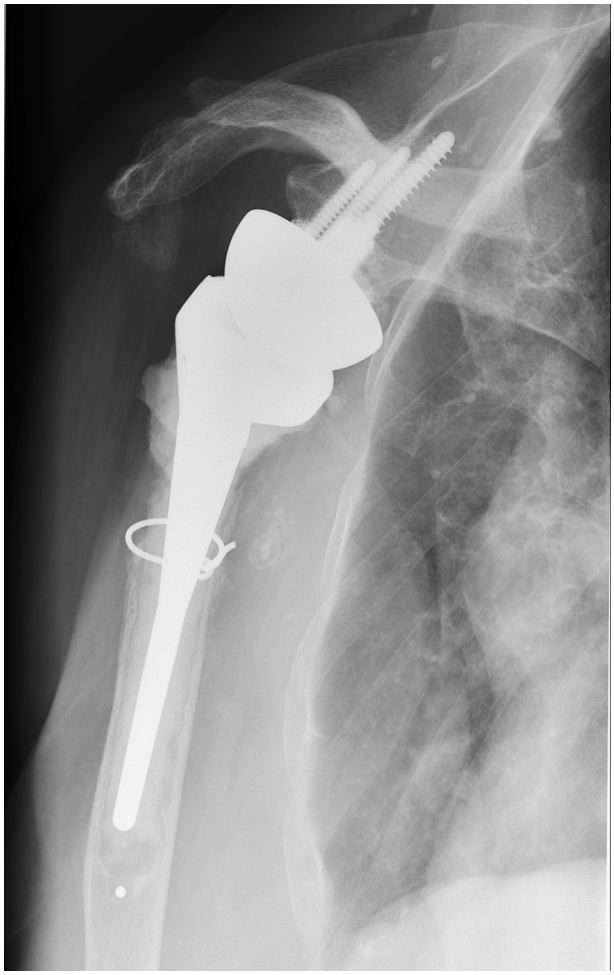

Figure 3 Reverse shoulder arthroplasty at last follow-up on radiograph of right shoulder.

retention (DAIR) for total knee arthroplasty gives poorer results than that for other joints. If it seems established that implant retention gives better results when the infection is being taken care of within 3 weeks after the prosthesis implantation, decision making is more complex beyond that.[7]

DAIR appears to be successful for MBT infections, with some authors having even achieved clinical healing without surgery with only an adapted and extended anti-TB treatment for 9 to 12 months.[6,10,13] The apparent success of this medical approach may be attributed to a lower adherence of MBT to the biofilm unlike for *S epidermidis*, as well as the high efficiency of the anti-TB treatment containing rifampin.[4] In our case, however, DAIR failed despite the medical and surgical management and the full susceptibility of the MTB strain to the anti-TB agents.

Functional failures in the management of MBT infections are linked to the frequent diagnostic delays because standard bacteriologic cultures cannot identify MBT. During such delays, patients can undergo multiple iterative surgical procedures compromising the subsequent function of the shoulder as they are not followed by adequate antibiotic treatment and thus the infection is not being treating.[2,5] We performed 3 surgical procedures before obtaining the diagnosis of MBT infection. At the last surgical procedure, a *C acnes* infection was identified, probably as a result of the previous surgical interventions.[17] Keeping in mind the diagnosis of MBT infection could have spared us multiple interventions as we retrospectively analyzed the calcified lymph nodes as TB scars. Multiple operations may have reactivated the TB.

It is necessary to ask for an extended culture with a specific search for mycobacteria, direct analysis with a Ziehl-Neelsen stain, and extended cultures in Löwenstein-Jensen and Coletsos media. As these tests are not performed routinely, intraoperative joint samples usually show negative findings, which delays the optimal management of the infection. These analyses should be routinely requested in cases of a high suspicion of PJI with negative intraoperative sample findings in patients considered at high risk. Multidisciplinary consultation involving orthopedic surgeons, infectiologists, and microbiologists may help diagnose this type of infection and treat it in the most efficient way.[3]

Conclusion

This case report shows the diagnostic difficulties encountered when dealing with extrapulmonary TB. To ask for the correct sample bacteriologic analysis, we have to keep in mind TB infection, especially in patients with low socioeconomic backgrounds or from low-income countries. In our patient, the failure of the DAIR approach required the replacement of the RSA to finally achieve a cure.

Disclaimer

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