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ORIGINAL STUDY

Chronic Mycobacterium kansasii tenosynovitis in an immunocompromised host : case report

Laura Lemmens, Nathalie Van Beek, Luk Corluy, Stefaan Verfaillie

From the AZ Herentals, Belgium

Mycobacteriuim kansasii is a nontuberculous atypical mycobacterium which typically causes respiratory infections. Localized extrapulmonary diseases, such as tenosynovitis or arthritis are rarely seen in the immunocompetent population *(1)*. We present a case of an immunocompromised 55-year-old man with a chronic Mycobacteriuim kansasii tenosynovitis of the hand.

Keywords : mycobacterium ; infection hand ; immuno-compromised.

CASE REPORT

A 55-year-old man with a history of psoriatic arthritis presents to the hand clinic with pain and swelling on the dorsum of the left hand since 5 weeks. He is a technician in the chemical industry and builds swimming pools in his free time. For his psoriatic arthritis he takes methotrexate and certolizumab. A couple of months ago he went for a canoeing trip and from that moment on he noticed a minimal skin lesion on his hand. A clinical examination revealed profound swelling of the dorsum of the fifth metacarpophalangeal (MCP) joint and, as mentioned before, a minimal skin lesion.

Initially normal laboratory results were found with a C-reactive protein (CRP) of 5 mg/dL, a sedimentation rate of 1 mm/h and white blood cells count of 6.7x10*9/L. An ultrasound demonstrated subcutaneous edema without underlying abscess. The patient received a local corticosteroid injection (40mg) with initial good clinical improvement. Two months later he came back to the clinic with recurrent swelling. An MRI scan showed no signs of synovitis at the fifth metacarpophalangeal joint (MCP) joint and no erosive lesions. Severe subcutaneous edema with impression of a small abscess appeared, mainly at the fourth and fifth MCP joints. No thickening of the extensor tendons was seen, but more synovial swelling around the extensor tendon of the fifth row was visible on MRI. A surgical debridement was performed and biopsies were taken. Immediately after the surgical debridement, clarithromycin was given because of the high suspicion of a mycobacterial infection. The reason for this suspicion was that profound tenosynovitis was seen clinically and on the MRI, and the patient was immunocompromised. A few days after the debridement, pain, swelling and redness recurred. CRP at that moment was found to be slightly raised (13 mg/dL). Amoxicillin/

■ Laura Lemmens,
■ Nathalie Van Beek,
■ Luk Corluy,
■ Stefaan Verfaillie
AZ Herentals, Belgium
Correspondence : Laura Lemmens, Orthopaedic Surgeon, Resident, AZ Herentals, Nederrij 133, 2200 Herentals
E-mail : Laura.lemmens@azherentals.be

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clavulanic acid 3g twice daily was first given intravenously and afterwards orally 875mg three times a day. Due to clarythromycin intolerance and no clinical improvement after the first debridement, a new surgical debridement was performed. Postoperatively levofloxacin (500mg 1x/d) and doxycycline (200mg 2x/d) was given. After rheumatologic advice, the immunosuppressiva (certoluzimab) were stopped. Three weeks after the second debridement the culture was found to be positive for Mycobacterium kansasii and the antibacterial therapy was switched to a fourfold therapy, consisting of clarithromycin 500mg twice daily, and once a day isoniazid 300mg and rifampicin 600mg, and ethambutol 400mg four times a day for at least two months. Pathology was suggestive of necrotizing granulomatous tenosynovitis.

Two months after starting the fourfold therapy with antimycobacterial drugs, the culture results suggested resistance to ethambutol. As such, ethambutol was discontinued and only isoniazid, rifampicin and clarythromycin were continued. Because there was no clinical improvement after 2 months of therapy, a new debridement was performed. After 6 months of triple therapy of tuberculostatic drugs, there was an improvement of pain and swelling. Clarythromycin was stopped and only rifadin and isoniazid were continued for the next six months. A follow-up 3 months after discontinuing his therapy has shown no recurrence of the infection so far. In addition, the patient oddly did not have any recurrence of psoriatic arthritis after stopping the immunosuppressiva.

DISCUSSION

Atypical mycobacterial infections are frequently diagnosed using the genetic identification technique *(2)*. About 50% of the diagnosed atypical mycobacterial infections are due to M. marinum, with the second most common infecting organism being M. kansasii and the third M. avium *(3)*. Hobbies and occupation related to water and the sea, aquariums, and soil are potential risk factors for a M. kansasii infection, together with immunosuppressiva and trauma of the skin *(4)*. In our case the patient had two risk factors for a mycobacterial infection. One is his immunocompromised status because of the psoriatic arthritis, and the other risk factor is that he builds swimming pools and thus has a much higher exposure to contaminated water.

M. kansasii usually causes a granulomatous lung infection, but tendinitis, synovitis, arthritis and carpal tunnel syndrome can also occur *(5)*. In our case the patient suffered from an isolated tenosynovitis and had a negative X-ray of the chest.

This case demonstrates, coupled with the majority of cases described by other authors previously, the diagnostic problem that atypical mycobacterial infections can present. Previously reported studies show that in nearly all cases at least a year would elapse before a correct diagnosis was made and often steroid or non-steroid anti-inflammatory drugs were prescribed without any significant effect before a biopsy was considered as an alternative treatment. One of the main reasons for this delay is that the clinical disease pattern is often dormant and thus difficult to diagnose. A clinical presentation with chronic swelling of the involved area not responding to conventional treatments, an immunocompromised patient and exposure to contaminated water, are the keystones of the diagnosis. The laboratory results such as CRP and sedimentation rate are often normal in previously described cases. If they are increased, it will be in a much smaller order than it is with gram-positive soft tissue infections *(6)*. Another possible reason for the delay in diagnosis could be found in the biopsy itself. It is mandatory that tissue is submitted for microbiological investigation as well as histology, however as the organisms may be scarce or absent from tissue sections, an accurate identification is highly dependent on the specific characteristics of the performed culture and biochemistry.

In our patient case there was resistance for ethambutol in one culture and in another culture it was shown to be sensitive. The reason for these differing susceptibility results of ethambutol between the two reference labs is unclear. The cultures were performed by 2 different laboratory technicians, and this could be a possible explanation. In most cases, the antibiotic regime involves a combination of isoniazid, rifampicin, ethambutol and erythromycin. The variability in resistance

of M. kansasii and the other organisms makes it essential to know the drug sensitivities before or after starting treatment. Varying time points for the administration of tuberculostatic drugs have been reported including just after surgical debridement, after susceptibility for drugs has been proven, and many more. Although drug administration just after debridement is often recommended, there is so far no consensus *(7-9)*. The optimal duration of drug therapy is also uncertain, but a minimum of 3-6 months is usually recommended, guided by clinical impression. An extension for 3-4 months after a complete resolution of clinical signs has previously been advised by Gunther and Levy *(10)*. For this reason, as we followed the guidelines, our patient received another 6 months of rifadin and isoniazid after clinical improvement. The decision of drug use was made in a multidisciplinary approach together with an orthopedic surgeon, a rheumatologist and an infectiologist.

Although tenosynovectomy alone cannot remove all infected tissue, it will decrease the overall diseased tissue load, and this may give antimycobacterial therapy a better chance of eradicating the residual infection *(11)*. As such, we performed multiple debridements every time there was no clinical improvement after a period of antimycobacterial drugs. This helps to reduce the lesion, but can also shorten the duration of drug therapy and decrease the side-effects *(12)*.

CONCLUSION

Our immunocompromised patient with chronic Mycobacterium kansasii tenosynovitis of the hand was successfully treated with multiple surgical debridements and 14 months of antimycobacterial therapy. We recommend careful monitoring of antimicrobial therapy by an infectious disease specialist to minimize the complications and rate of recurrence.

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