CASE REPORT

Carpal synovitis with capitate bone tuberculosis in a child

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SUMMARY

We present a 10-year-old boy with 2-month duration non-traumatic wrist pain and inflammatory signs. Due to elevated inflammatory markers on blood tests, with an increase in radiocarpal and intercarpal joints synovial fluid and no bony lesions, the patient was submitted to wrist arthrocentesis for the suspicion of septic arthritis. The patient did not improve on conventional treatment, however. An MRI showed synovitis around the carpus and a lytic lesion of the capitate bone due to osteomyelitis. A biopsy was able to identify the causative agent as *Mycobacterium tuberculosis*, and the patient was treated with antibiotics. He improved significantly, with no pain and signs of normal capitate bone remodelling on the last radiograph.

BACKGROUND

Tuberculosis (TB) is a major public health problem and although incidence is declining (1.5% from 2014 to 2015), the WHO estimates around 10.4 million new cases every year.[1] In Portugal, the incidence of TB is 20/100.000. However, most cases are seen in large urban centres (with a higher incidence, between 20 and 50/100.000) and are often related to emigrants from sub-Saharan Africa. Of these cases, 28.7% have extrapulmonary infections.[2]

Skeletal TB is an uncommon condition. Although bone and/or joint affection is the third most common extrapulmonary location, it represents only approximately 11% of all cases of extrapulmonary TB.[3]

This condition is usually chronic and commonly affects the spine and weight-bearing articulations, such as the hip or knee,[4] or long bones, as the tibia and femur.[5] Hand and wrist are less frequently involved anatomic areas.[6 7]

Patients with hand and wrist TB present with slow progressing, indistinctive signs that may cause difficulty in diagnosis and delay treatment.[7 8]

This report describes a rare case of extrapulmonary wrist TB, which posed a significant clinical challenge due to its location and indolent presentation. Nevertheless, correctly diagnosing this condition allowed for a specific approach and treatment, with significant improvement in patient's clinical condition.

CASE PRESENTATION

A previously healthy 10-year-old boy presented to the Emergency Department with 2-month duration, non-traumatic, wrist pain. On observation, local inflammatory signs of his left wrist were noted, along with decreased, painful range of motion and a soft, non-tender bulge over the dorsal aspect of the wrist.

Fever, weight loss, sudoresis and cough were denied. He had no other significant clinical findings, such as cervical or axillary lymphadenopathy. There was also no relevant history regarding contact with people infected by TB and his vaccination scheme was up-to-date (including BCG).

INVESTIGATIONS

Although hand and wrist radiographs showed no evidence of acute bony lesions (figure 1A), an increase in radiocarpal and intercarpal joints synovial fluid, mostly around the capitate, was noted on ultrasonography. Leucocyte count was within normal range (9200×10^6/L) whereas inflammatory markers were slighted elevated (C reactive protein 35 mg/L; erythrocyte sedimentation rate 44 mm/hour).

TREATMENT

Due to clinical suspicion of septic arthritis, and since there was no other relevant information that could have made us consider other aetiologies, the patient was submitted to arthrocentesis and started on flucloxacillin (250 mg q6h).

Both blood and synovial fluid cultures were negative.

Despite continuous antibiotic treatment, recurrent joint effusion was noted 6 weeks after arthrocentesis, with a severe increase in joint tension, fistula development and a capitate lytic lesion showing on wrist radiograph (figure 1B). An MRI was immediately performed, identifying extensive synovitis around the carpus, signs of osteomyelitis associated with the lytic lesion of the capitate and tenosynovitis involving flexor digitorum and flexor carpi radialis tendons (figures 2–4).

The patient was scheduled for an arthrotomy followed by a biopsy, sampling for microbiology, aggressive debridement and joint lavage.

Histology from samples collected during surgery exhibited granulomatous caseous necrosis with negative acid-fast bacilli staining. Interferon-Gamma Release Assay (IGRA) was positive and *Mycobacterium tuberculosis*, sensitive to all antibiotics, was identified in tissue cultures. Pulmonary TB and HIV infection were excluded.

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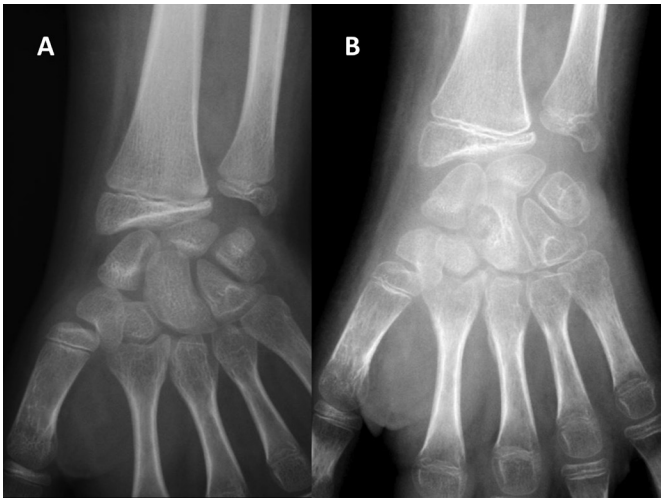

Figure 1 Anteroposterior wrist radiography. (A) Patient's wrist on first observation. (B) Patient's wrist 6 weeks later, with capitate bone destruction.

The patient was started on daily isoniazid (300 mg), rifampicin (600 mg), pyrazinamide (2000 mg) and ethambutol (1200 mg), for 2 months, followed by 10 months of daily isoniazid (300 mg) and rifampicin (600 mg). No restriction to wrist movement was advised, but the patient's arm was placed on an arm sling for 3 weeks.

OUTCOME AND FOLLOW-UP

At a 9-month follow-up, the patient regained full wrist range of motion and presented no pain. Currently, with an 18-month follow-up, there is no evidence of infection and wrist radiograph shows signs of capitate bone remodelling (figure 5).

DISCUSSION

This case depicts a successful diagnosis and treatment of capitate bone TB, associated with an aggressive infectious synovitis, both extremely rare conditions that represent a diagnostic challenge, especially in children.

Musculoskeletal TB is an uncommon condition,[3] usually occurring in long bones, such as the tibia or femur.[5] Infection

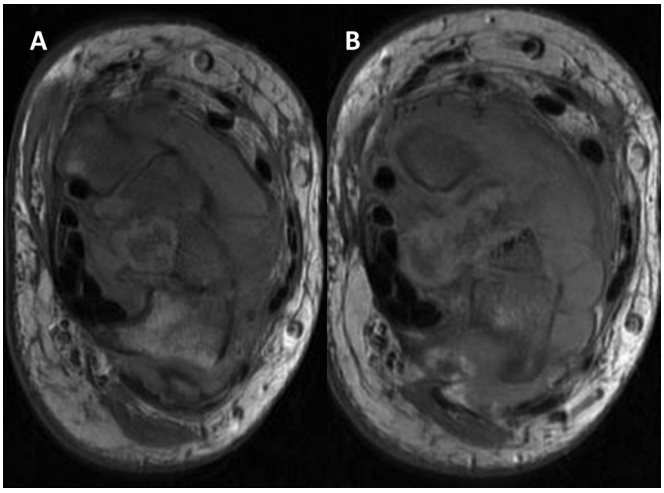

Figure 2 Wrist MRI in the transverse plane, showing a lytic lesion of the entire capitate bone (A,proximal segment; B, distal segment), with associated inflammatory signs extending to the flexor tendons.

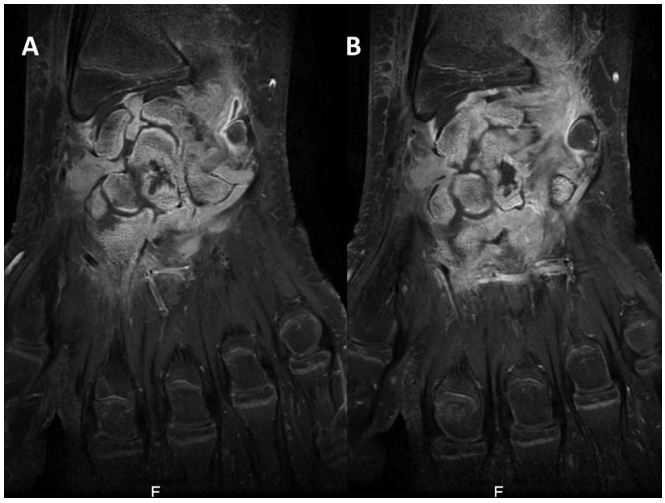

Figure 3 Wrist MRI in the coronal plane, showing the extent of the capitate bone lytic lesion (A, dorsal segment; B, volar segment).

of the wrist is a rare occurrence, comprising only up to 1% of all musculoskeletal TB infections.[6] The infection begins either in the synovial membrane, progressing into the whole carpus, as well as flexor and extensor tendons,[9] or in bone metaphysis/diaphysis, with later spread into adjacent structures, such as the joint.[10]

The typical clinical presentation may feature mild pain, tenderness, soft tissue oedema, sinus and pathological fractures.[5 7 10 11] Indeed, Prakash and Mehtani[12] reported a review on 44 cases of hand and wrist TBs, where most patients presented with pain and swelling, which did not preclude using the hand, moderate limitation in range of motion and sinus formation as one of the presenting symptoms. These symptoms started, in average, 7.6 weeks before patients sought medical advice.[12]

Despite describing four cases of capitate bone TB,[12] this study did not give any more specific information about patient or infection characteristics, as well as conducted treatment or results.

Similarly, Kotwall and Khan[11] also presented 32 cases of wrist and hand TB, 2 of which with capitate bone infection reported: both patients were men, with 9 and 19 years of age, respectively, and treated conservatively. No recurrence was identified during follow-up (21 and 31 months, respectively) but, regrettably, not

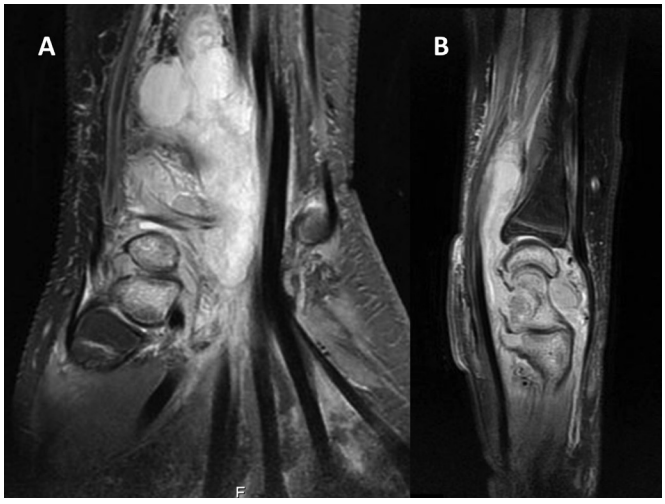

Figure 4 Wrist MRI, (A) coronal plane and (B) sagittal plane, showing extensive flexor tenosynovitis on the palmar aspect of the wrist.

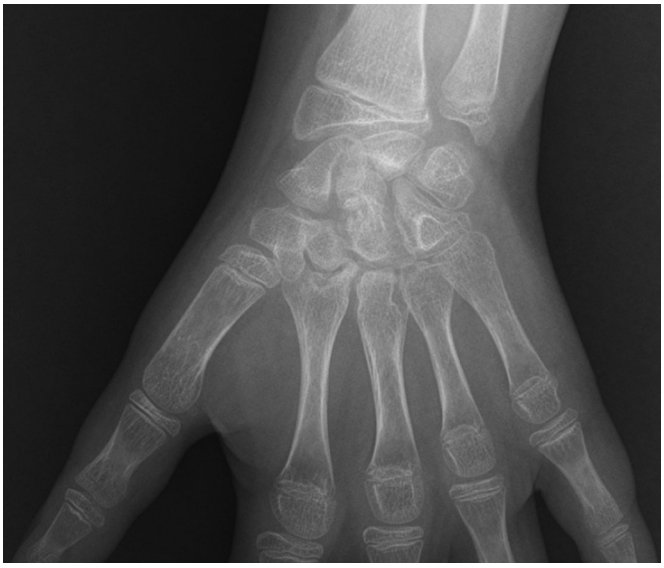

Figure 5 Anteroposterior wrist radiography at the final follow-up, showing capitate bone remodelling.

enough information was provided to perform a full comparison to our case.

Therefore, a literature review on published cases of capitate bone TB returns only two fully depicted cases, one of them in a child.[13 14]

All cases share some characteristics. All three cases report to a male patient, either still a child or a young adult (10, 12 and 23 years old), with wrist pain and swelling for at least 2 months prior to seeking medical advice. Also, the occurrence of sinus in the affected wrist was seen in all cases. This is in line with most reported reviews on hand and wrist TB.[5 10]

Coincidentally, all patients presented with infection of their left wrist, although this might have no clinical relevance.

There were no respiratory complaints or other relevant history that could make the treating physicians suspect about a possible pulmonary TB infection. Since extrapulmonary TB is only associated with active pulmonary disease in about one-third of cases,[6 10] this finding comes as no surprise.

Whenever osteolytic lesions are found in radiological examinations, one must consider other diagnoses besides infection, such as Langerhans cell histiocytosis, osteosarcoma/Ewing sarcoma, chronic recurrent multifocal osteomyelitis or osteoid osteoma. When these lesions occur in the hand, both enchondroma and osteosarcoma should be investigated.[15]

However, at an early stage, radiological findings are either unspecific or may not even exist, as any anomalous findings on image examinations will vary in accordance to the extent of the infection.[9] Nevertheless, MRI may present significant changes, which may allow for an early diagnosis of capitate bone infection and even synovitis and involvement of flexor/extensor tendons, as depicted in some cases found in the literature.[11–14] Retrospective evaluation of our case made us consider that requesting a MRI could have given more information and allow for an earlier diagnosis of TB infection. We now rely more on MRI for chronic, painful, inflammatory joint conditions in children.

Treatment of osseous TB usually includes isoniazid and rifampicin for 9 to 12 months combined with pyrazinamide and ethambutol for the first 2 months.[6] Surgical procedures in wrist TB are controversial, with some authors advocating that it is unnecessary in most patients.[6 13]

Patient's perspective

'I believe the worst thing was not knowing the reason for the pain my child was experiencing. We were informed through the entire process, and we were able to see the doctors' struggle to reach a diagnosis. Fortunately, when the cause for the infection was known, the treatment was given and my boy recovered fully. He is now able to play sports again!'

Learning points

- Maintaining a high suspicion level in all atypical cases of chronic wrist pain is essential in order to achieve a prompt diagnosis and adequate treatment of wrist tuberculosis (TB).
- Patients with capitate bone TB will typically present with chronic wrist pain and swelling, as well as sinus formation in the affected wrist.
- MRI presents as the best examination to diagnose early changes caused by TB, as radiographical findings are unspecific or may not even exist on an early stage.
- Due consideration should be given to performing a biopsy of osteolytic lesions of the hand, in order to rule out osteosarcoma, as well as test for bacterial and mycobacterial infections.
- Conservative treatment should be considered the primary treatment option.

One study evaluated risk factors for surgery in patients initially treated with antituberculous drugs. Time range between initiation of symptoms and adequate antituberculous treatment was the most important risk factor encountered.[12] Likewise, elevated erythrocyte sedimentation rate, skin ulceration, multiple lesions or presence of sinus were also risk factors for surgical intervention.[12] As for capitate bone TB, at least one surgical procedure, with or without debridement, was performed in each case.

All patients recovered normal range of motion, with sinus closure and radiographical signs of bone formation on lytic lesions of the capitate. There were also no signs of recurrence on any case at final follow-up evaluation.[13 14]

Most authors also highlight the importance of histological changes, with epithelioid granulomatous reaction and central caseous necrosis, together with mycobacterial cultures and nucleic acid amplification tests for TB diagnosis.[12–14]

Being a rare and misleading infection, one can easily understand why evaluation of this disease is difficult, as clinical manifestations may either mimic other wrist pathologies or be too unspecific to allow for a simple and direct diagnosis. Nevertheless, physicians must still consider TB in all cases of chronic wrist pain that does not respond well to usual treatment options.

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