

# Massive calcaneal enthesopathy in a non-healing leg ulcer: A case report

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## ABSTRACT

Enthesopathy at the superior or inferior surface of a calcaneus may be seen in normal individuals having degenerative osteoarthritis. This condition is also known to occur in patients with rheumatoid arthritis, seronegative spondyloarthropathy, trauma, as well as inflammatory and metabolic diseases. Enthesopathy may sometimes be the first manifestation of a variety of rheumatic diseases. In this report, we present a case of massive enthesopathy of the superior and inferior surface of the calcaneus giving rise to an 'axe effect'.

**Key words:** arthritis, rheumatoid; calcaneus; rheumatic diseases

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## INTRODUCTION

An enthesis is a site of insertion of a tendon, ligament, or articular capsule into the bone. An alteration at these sites is termed an enthesopathy.<sup>1</sup> Wright and Moll<sup>2</sup> first used the term 'seronegative spondyloarthropathy' to describe conditions that involved the sacroiliac joints, vertebral column, and similar extra-articular manifestations. The involvement of cartilagenous joints and enthesopathy is known to occur in rheumatoid arthritic patients, but appears less frequently than in seronegative spondyloarthropathic patients.<sup>3</sup> Posterior calcaneal spurs rarely occur in Reiter's syndrome compared with rheumatoid arthritis, psoriasis, and ankylosing spondylitis.<sup>4</sup> Enthesophytes in degenerative disease are well defined compared with the fluffy, poorly marginated calcaneal outgrowth as seen in seronegative spondyloarthropathy.<sup>1</sup>

## CASE REPORT

An 87-year-old man presented with a 40-year history of a single non-healing ulcer over the anterolateral aspect of the lower one third of the right leg and right ankle. Radiographs of the spine and sacroiliac joints showed flowing calcification and ossification along the anterolateral aspect of more than 4 contiguous vertebrae. The absence of apophyseal and sacroiliac joint bone ankylosis and the absence of radiographic changes related to degenerative disc diseases were consistent with diffuse idiopathic skeletal hyperostosis. The radiograph of the right ankle showed massive enthesopathy of the superior and inferior surfaces of the calcaneus mimicking the shape of an axe (Fig. 1). The radiograph also showed some periosteal reaction in the tibia caused by osteomyelitis. The radiograph of the left ankle showed typical insertional enthesopathy of the calcaneus (Fig. 2). Culture from the wound revealed a heavy growth of *Escherichia coli* and *Enterobacter*. The serum titre results were negative for rheumatoid factor and human leukocyte antigen B27. The test for C-reactive protein was positive and the erythrocyte sedimentation rate was 60 mm/h. All other routine blood parameters were within normal limits.

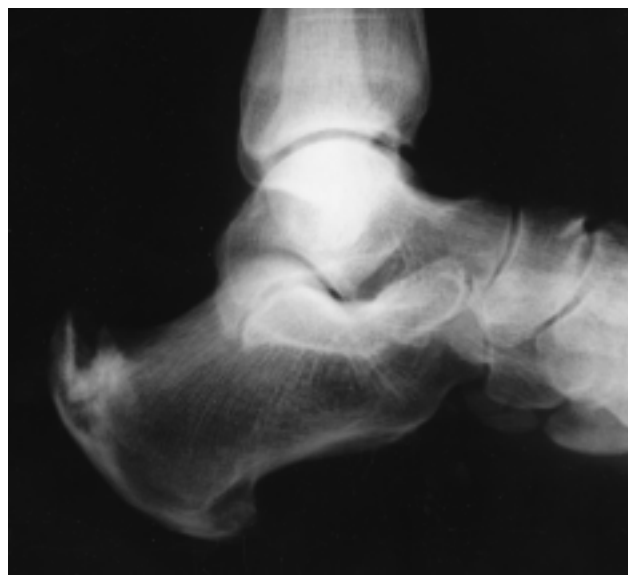
## DISCUSSION

Well-defined calcaneal enthesophytes are seen in as many as 25% of the population, according to studies from the US.<sup>1</sup> In addition to the enthesis occurring in or around the synovial joints, cartilaginous joints, and syndesmoses; abnormalities may appear at the calcaneus, patella, humeral tuberosities, olecranon process of ulna, iliac-crest, ischial tuberosity, and femoral trochanters.<sup>1</sup> Although rheumatoid arthritis and seronegative spondyloarthropathy share many radiological and pathological features, fundamental differences exist between their distribution and the osteoarticular lesions.<sup>5</sup> In seronegative spondyloarthropathy, the absence of osteoporosis, the presence of bony proliferation, and intra-articular bony ankylosis with enthesopathy help to differentiate it from rheumatoid arthritis.<sup>6</sup>

Diffuse idiopathic skeletal hyperostosis is accompanied by alterations in both the spinal and extraspinal locations, which are manifestations of an enthesopathy. Diagnostic criteria of diffuse idiopathic skeletal hyperostosis include flowing calcification and ossification along the anterolateral aspect of at least 4 contiguous vertebrae, the absence of radiographic changes relating to the degenerative disc disease, and



**Figure 1** Lateral radiograph of the right ankle showing massive enthesopathy of the superior and inferior surfaces of the calcaneus with periosteitis of tibia.



**Figure 2** Lateral radiograph of the left ankle showing the typical finding of insertional enthesopathy of the superior and inferior surfaces of the calcaneus.

the absence of apophyseal and sacroiliac joint bone ankylosis. Changes at the superior and inferior surfaces of the calcaneus, patella, tarsal bones, and olecranon are frequently seen.<sup>1</sup>

The pathology in enthesopathy is an inflammatory process that results in the replacement of the enthesis by soft connective tissue containing plasma cells and lymphocytes, which tend to spread along the undamaged part of the ligament. The surrounding bone shows evidence of erosion. Ultimately, the

ligament and bone are repaired by woven bone, which is then remodelled into the lamellar bone.<sup>1</sup>

Infectious agents have long been implicated as trigger factors for spondyloarthropathies. Human leukocyte antigen B27 is now acknowledged to play an important role, although its action is yet to be elucidated and the relationship is still not completely understood. Most of the organisms implicated are gram-negative bacteria. They can penetrate and survive within mammalian cells and can persist for prolonged periods in the synovium and monocytes. Living microorganisms have not been recovered from articular tissue, although microbial antigens have been identified using polymerase chain reaction techniques.<sup>7</sup>

In this patient, the long-standing history of infections in the vicinity of the right leg and ankle, and the presence of gram-negative pathogens in the wound could explain the massive enthesitis of the superior and inferior surface of the calcaneus.

## CONCLUSION

Several disorders cause enthesopathy and differentiation among them is often difficult. Therefore, understanding the morbid anatomy of enthesopathy is important because it may be the first manifestation of a variety of rheumatic diseases.

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