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Case Study
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A Rare Entity for Spondyloarthritis: Extremititis

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Case Study

46 year-old male admitted for pain in his second foot finger and foot dorsum for two days. He was diagnosed as non radiographic axial spondyloarthritis five years ago with chronic inflammatory back pain and a diagnostic sacroiliac MRI. He was under sulfasalazine and diclofenac sodium treatment up to 3 years and has been taking no medication for two years because he had no complaints. He firstly feels pain on the dorsal side of his second foot finger and consequently it spread along the foot dorsum with the redness and pain. At the time of assessment he had no back pain and any other articular complaint. His second finger was wholly swollen and 2nd metatarsal region and half distal of dorsum of his food was swollen, red and painful revealing dactylitis of the second finger and metatarsophalangeal arthritis and metatarsitis. His blood tests revealed high acute phase results with no other specific changes. The differential diagnoses were Spondyloarthritis and Gouty Arthritis. Naproxen sodium 1000 mg/d and sulfasalazine treatments were started. But on the consequent day, the pain and swelling spread into his right ankle revealing additive ankle arthritis. This Picture brought the possibility of infection to the fore. Amoxicillin and ciprofloxacin was started. On the second day of antibiotics and the 4th day of the complaints the pain spread through his whole right leg up to distal board of the knee and leg was swollen and sensitive. This picture forced to consider the possibility of the deep vein thrombosis but the Doppler ultrasound of right lower extremity was completely normal. So the MRI of the right lower extremity was planned. MRI just revealed a subcutaneous edema and elevated fluid intensity of the fascias between the muscles (Figure 1). Antibiotics were continued on and a 250 mg pulse steroid treatment was started. After the first dose of pulse steroid, with no clinical improvement, a consequent knee involvement was added (Figure 2). Knee was also hyperalgesic and swollen and the physical treatment revealed a little ballotman. So the intra-articular steroid injection was administered for the local and systemic benefits, but articular fluid seemed completely normal with its color and optimal viscosity. So the required knee ultrasound revealed no knee effusion. But the periarticular tendon and soft tissue inflammation was reported.

Discussion

As a brief reconsideration, this phenomenon was firstly a dactylitis-like or gouty-like beginning that progressed to whole extremity inflammation in the consequent 5 days and the spread of the inflammation was completely due to anatomical proximity. The clinical improvement was not

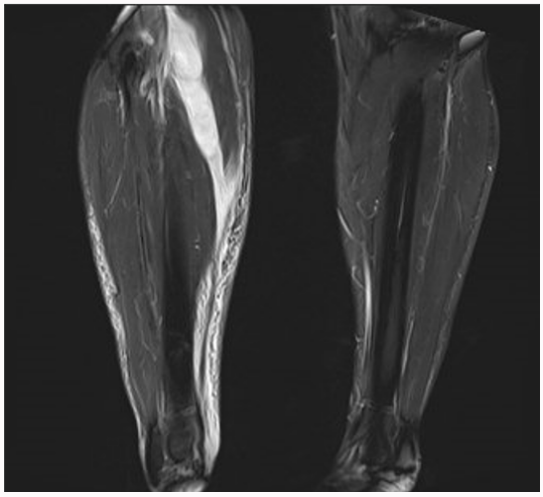

Figure 1: Lower extremity MRI, T2 sequence.

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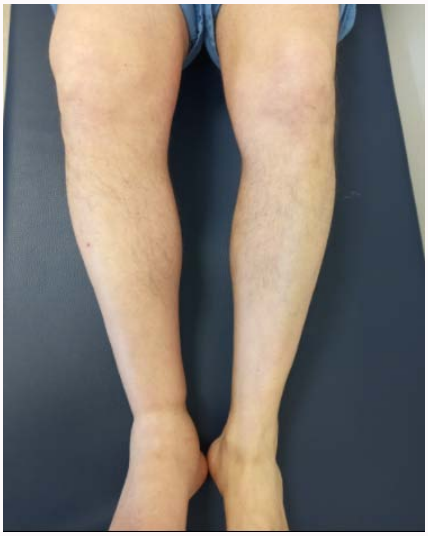

Figure 2: After the first dose of pulse steroid.

possible with the conventional therapies and the pulse steroid doses. So biological treatment was planned as the emergent requirement because the working position of his business was under risk due to his lack of performance. Adalimumab was administered after a negative QuantiFERON testing and after the second dose in the first month of medication, he completely healed.

From the final point of view, it is seen more clearly that the clinical decision of arthritis due to physical assessment was a misdiagnose and it was mostly periarthritis from the first clinical picture of the dactylitis like swelling to the last clinical feature as the knee periarthritis. As a valuable experience, a single dose of anakinra was administered to patient due to his confirmation on the day swelling spread up to the distal border of knee, and after just one dose of 100 mg SC Anakinra, swelling of the leg got improved but it showed no benefit to the knee, ankle, and foot dorsum and finger periarthritis. So it shows some similarity with the fasciitis of the FMF patients which is known as well responding to single dose Anakinra. As an academic requirement, MEFV gene was analyzed and resulted as M694V heterozygous. Fasciitis in autoimmune or auto inflammatory diseases have been reported in several cases. It is mostly seen in patients with Tumor Necrosis Factor Receptor-Associated Periodic Syndrome (TRAPS) [1,2]. In addition, patients with psoriatic arthritis, polyarteritis nodosa, eosinophilic fasciitis and inflammatory myopathy with abundant macrophages may have fasciitis [3,4]. Also there is an entity as "Protracted Febrile Myalgia Syndrome" with the Familial Mediterranean Fever. PFMS is no known to well respond to single doses of Anakinra [5]. Periarticular inflammation and bone marrow edema is critical foot the confirmation of Spondyloarthropathies [6]. As it is proven, the progress of the current case is thought to have similarity about the partial response to IL-1 blockage. The three major points of this case may be summarized as; 1. A newly entity which is not named literally may be described as extremititis. 2. The pattern of fasciitis is responsive to IL-1 blockage and the periarthritis of the joints do not. 3. Anti TNF treatment can alleviate all the features of the pathologic progress. Weak point of the report is the lack of knowledge about the HLA B27 result but it is clear that the diagnostic process did not require to reveal HLA B27 profile.

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