International Scholars Journals

International Journal of Public Health and Epidemiology ISSN 2326-7291 Vol. 5 (2), pp. 244-245, February, 2016. Available online at www.internationalscholarsjournals.org © International Scholars Journals

Case Report

Management of severe and refractory HLA-B27 negative heel enthesitis using adalimumab

Faisal Salman

Department of Rheumatology, King AbdulAziz University Hospital, Jeddah, Saudi Arabia.

Accepted 15 November, 2015

Enthesitis is a common manifestation of HLA-B27 positive spondyloarthritis. Here, we report the successful use of adalimumab for the treatment of an unusual case of isolated heel enthesitis, in an HLA-B27 negative patient without additional clinical symptoms of undifferentiated spondyloarthritis. These findings contribute to the growing body of evidence that supports the beneficial use of anti-tumor necrosis factor-alpha therapies for patients with enthesitis.

Key words: Adalimumab, enthesitis, spondyloarthritis, magnetic resonance imaging.

INTRODUCTION

Patients with enthesitis-related arthritis display a similar clinical presentation to those meeting traditional definitions of juvenile spondyloarthropathy (Michelsson, 2005; Flatø, 2006). Although no guideline regarding their treatment has been provided, several reports recently propose the use of tumor necrosis factor-alpha (TNFα) antagonists to treat juvenile-onset Human Leukocyte Antigen (HLA)-B27-associated with severe and refractory heel enthesitis (Olivieri, 2006). We present here a case of successful adalimumab-based therapy for an unusual occurrence of isolated heel enthesitis in an HLA-B27 negative patient without clinical symptoms of undifferentiated SpA.

CASE REPORT

A 28-year-old male patient of Arab descent presenting with an 18-month history of heel pain in the left foot, which was more severe during the first steps after rising and worsened by sport activities, was referred to our Department of Rheumatology. Overall, the discomfort led to serious disability for the patient, who had no known previous history of medical illness or traumatic injury. In addition, he did not display any typical SpA features, nor did he have a family history of spondyloarthropathy. Furthermore, there was no other joint involvement, history of similar episodes, inflammatory back pain, personal/family history of psoriasis, previous gastrointestinal/genitourinary symptoms, or evidence of inflammatory bowel disease. Moreover, due to the severity of the symptoms, the patient had been previously treated with celecoxib (200 mg orally, twice-daily for two months), arcoxia (120 mg orally, daily for four months), two local steroid injections in the left foot's plantar fascia, and sulfasalazine (up to 3 g daily for five months), all resulting in little or no improvement.

Upon further clinical examination, we identified soft tissue swelling at the insertion of the left Achilles' tendon, with tenderness of the plantar fascia. Also, the patient did not display restricted spinal mobility, psoriatic skin lesions, nail dystrophy, or pain/swelling of the peripheral joints or entheseal sites. We found that the Maastricht Ankylosing Spondylitis Enthesitis Score (MASES) was 1, and the Visual Analogue Scale (VAS) pain level was 10 (Sieper, 2009). Laboratory tests revealed that the erythrocyte sedimentation rate and C-reactive protein levels were normal. Also, rheumatoid factor and HLA-B27 were negative. MRI of the left foot showed increased signal intensity within Achilles's tendon and the plantar fascia, while MRI of the right foot was unremarkable.

Due to the severity of the clinical findings, and because the patient did not improve following full doses of two separate NSAIDS, local steroid injection or sulfasalazine,

*Corresponding author. E-mail: abdulaziz_faisal@yahoo.com

this study started a therapy with a TNFα antagonist (adalimumab, 40 mg, every other week) and Methotrexate (10 mg orally, on weekly basis) plus folic acid (5 mg orally, on weekly basis). Adalimumab treatment of our patient led to a rapid and marked improvement of signs and symptoms after the second injection, with significant improvement in the MASES score and the VAS pain level (scores of 1 and 6, respectively). At three months, the MASES score was 0, and the VAS pain level was 4. Following six months of continuous therapy, the MASES and VAS scores were both 0. A new MRI of the left heel performed nine months after treatment onset with adalimumab revealed evidence of plantar fasciitis. The Achilles' tendon was intact and no retrocalcaneal bursitisorerosive changes in the calcaneus could be seen. Therefore, significant improvement was observed clinically, as well as radiologically.

DISCUSSION

Here, we report the use of adalimumab to treat an unusual case of isolated heel enthesitis in an HLA-B27 negative patient without additional clinical symptoms of undifferentiated spondyloarthritis. Because this case proved so difficult to manage, we attempted anti-TNFα therapy as recommended by the ASAS and the European League Against Rheumatism (EULAR) for the management of ankylosing spondylitis in patients with symptomatic enthesitis after failure of appropriate local treatments (Zochling, 2006).

Additionally, Italian guidelines for the appropriate use of anti-TNFα therapy for psoriatic arthritis suggested the use of TNFα antagonists in cases of peripheral enthesitis resistant to conventional treatments (Salvarani, 2006).

Upon review of the literature for similar cases of isolated heel enthesitis, we identified reports of HLA-B27 positive enthesitis that responded to anti-TNFα treatment (Marzo-Ortega, 2001; McGonagle, 2003). Additionally, in cases where one anti-TNFα failed to control the disease, benefit was achieved following treatment with a second TNFα antagonist (Olivieri, 2007). Consistent with the findings of this study, a similar case of adalimumab-treated HLA-B27 negative heel enthesitis documented using MRI was also identified (Mancarella, 2010). In the previous report, the enthesitis was also severe and resistant, failing to respond to three months of full doses of diclofenac and ibuprofen, physical therapy, and multiple steroid injections prior to the successful use of adalimumab.

The findings of this study contribute to the growing body of evidence that supports the use of anti-TNFα therapies for the treatment of patients with enthesitis. In particular, we believe that these biologic therapies can be beneficial for HLA-B27 negative patients with isolated heel enthesitis in the absence of other signs of undifferentiated SpA. Future studies of patients will be needed to confirm these findings.

REFERENCES

Flatø B, Hoffmann-Vold AM, Reiff A, Førre Ø, Lien G, Vinje O (2006). Long-term outcome and prognostic factors in enthesitis-related arthritis: a case-control study. Arthritis Rheumatol., 54: 3573-3582.

Mancarella L, Battaglia M, Addimanda O, Pelotti P, Galletti S, Meliconi R (2010). Successful adalimumab treatment of HLA B27 negative heel enthesitis documented with MRI and US. Clin. Exp. Rheumatol., 28: 443-444.

Marzo-Ortega H, McGonagle D, O'Connor P, Emery P (2001). Efficacy of etanercept in the treatment of the entheseal pathology in resistant spondylarthropathy: a clinical and magnetic resonance imaging study. Arthritis Rheumatol., 44: 2112-2117.

McGonagle D, Marzo-Ortega H, Benjamin M, Emery P (2003). Report on the Second international Enthesitis Workshop. Arthritis Rheumatol., 48: 896-905.

Michelsson O, Konttinen YT, Paavolainen P, Santavirta S (2005). Plantar heel pain and its 3-mode 4-stage treatment. Mod Rheumatol., 15: 307-314.

Olivieri I, Scarano E, Gigliotti P, Giasi V, Padula A (2006). Successful treatment of juvenile-onset HLA-B27-associated severe and refractory heel enthesitis with adalimumab documented by magnetic resonance imaging. Rheumatology (Oxford), 45: 1315-1317.

Olivieri I, Scarano E, Padula A, D'angelo S, Cantini F (2007). Switching tumor necrosis factor alpha inhibitors in HLA-B27-associated severe heel enthesitis. Arthritis Rheumatol., 57: 1572-1574.

Salvarani C, Olivieri I, Pipitone N, Cantini F, Marchesoni A, Punzi L (2006). Recommendations of the Italian Society for Rheumatology for the use of biologic (TNF-alpha blocking) agents in the treatment of psoriatic arthritis. Clin. Exp. Rheumatol., 24: 70-78.

Sieper J, Rudwaleit M, Baraliakos X, Brandt J, Braun J, Burgos-Vargas R (2009). The Assessment of SpondyloArthritis international Society (ASAS) handbook: a guide to assess spondyloarthritis. Ann Rheum Dis., 68(Suppl 2): ii1-44.

Zochling J, van der Heijde D, Burgos-Vargas R, Collantes E, Davis JC, Dijkmans B (2006). ASAS/EULAR recommendations for the management of ankylosing spondylitis. Ann Rheum Dis., 65: 442-452.