

Adalimumab Treatment for Hidradenitis Suppurativa Associated with Crohn's Disease

Dominika Diamantova¹, Iva Lomickova², Petra Cetkowska²

¹Department of Dermatology and Venereology, Military Hospital, Olomouc; ²Medical Faculty and Teaching Hospital Pilsen, Department of Dermatology and Venereology, Charles University, Pilsen, Czech Republic

Corresponding author

Dominka Diamantova, MD
Department of Dermatology and Venereology
Military Hospital
Sišilovo nam. 7
Olomouc
Czech Republic
DominikaDiamantova@seznam.cz

SUMMARY Hidradenitis suppurativa (HS) is a chronic, inflammatory disease characterized by recurring abscesses, nodules, and fistulas predominantly in the area of the groin and axillae. The association between HS and Crohn's disease (CD) has already been documented. We report on a case of a patient with CD associated HS, refractory to multiple local and systemic agents. A complete resolution of both diseases was finally achieved after treatment with adalimumab. Our case report supports the co-occurrence of both diseases and suggests that adalimumab approved for CD might also be a safe and effective therapeutic option in the treatment of HS.

KEYWORDS: Crohn's disease; hidradenitis suppurativa; adalimumab

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INTRODUCTION

Crohn's disease (CD) is a sporadic inflammatory disorder of unknown etiology that primarily affects the gastrointestinal tract and often results in discontinuous transmural inflammation. Extraintestinal manifestations are observed in 20-40% of affected patients. Dermatological complications include pyoderma gangrenosum, erythema nodosum, psoriasis, necrotizing cutaneous vasculitis, and epidermolysis bullosa acquisita (1,2).

Hidradenitis suppurativa (HS) is a chronic inflammatory disease of unknown pathogenesis, characterized by recurring painful abscesses, nodules, and fistulas occurring most frequently in the genitofemoral areas and armpits.

The association between HS and CD has been reported, nevertheless the prevalence of their comorbidity is currently unknown (3). The diagnosis of CD generally precedes that of HS by the average of 3.5 years (4-6).

Adalimumab is a fully human monoclonal antibody targeted at tumor necrosis factor α (TNF α). It is approved for the treatment of moderate to severe psoriasis, psoriatic arthritis, rheumatoid arthritis, ankylosing spondylitis, juvenile idiopathic arthritis, and CD. Recently, several case reports and a placebo-controlled study have indicated that adalimumab might also induce resolution in HS (7).

CASE REPORT

A 50-year-old female nonsmoker with a body mass index of 26 was first referred to our department in July 1989 with enlarging ulcers on both shins, diagnosed to be pyoderma gangrenosum. She reported a 6-month history of CD, treated since the onset with high-dose corticosteroids and sulfasalazine due to severe diarrhoeic episodes. Her family history had no bearing on the case. Physical examination revealed necrotic ulcerations on both shins with a characteris-



Figure 1. Genitofemoral region of the patient, with draining fistulas and abscesses before treatment.

tic violaceous undermined border. In addition to oral prednisone for her CD, cyclosporine and consequently azathioprine were administered with a complete resolution of both ulcers in April 1990.

In 1993, the patient developed inflammatory lesions bilaterally in the genitofemoral region with a good temporary response to prednisone, azathioprine, systemic antibiotics, and various types of topical antiseptics.

In December 2010, the disease progressed rapidly with development of painful fistulas, some of which were spontaneously discharging pus, and abscesses, mainly in the left groin (Fig. 1). The patient was clinically diagnosed with HS, Hurley stage II, and an incision of abscesses was performed. The recurrence of inguinal lesions was accompanied by a flare of CD with multiple episodes of diarrhea. In addition, the continuous long-term regimen of prednisone and azathioprine led to a weight gain of 12 kg and elevated blood levels of triglycerides and cholesterol.

Clinical exacerbation resulted in depressive disorder in the patient, so the immunosuppressive treatment was gradually reduced and the patient was included in the adalimumab protocol. In April 2011, adalimumab was started at an initial loading dose of 80 mg, followed by 40 mg every other week, along with oral prednisone 15 mg and azathioprine 25 mg daily. Over the next month, prednisone and azathioprine were tapered and eventually discontinued.

After five weeks of treatment, the groin lesions had almost healed, the pain had improved substantially, and remission of CD was achieved. Adalimumab was well tolerated with no adverse effects. Eight weeks after adalimumab initiation, the fistulas closed completely, leaving scarring in the genitofemoral area. To this date, the patient has not experienced recurrence



Figure 2. Genitofemoral area of the patient, with closed fistulas and no drainage after 8 weeks of treatment.

of either CD or HS (Fig. 2) and is back at her pre-treatment weight.

DISCUSSION

HS is a recurrent inflammatory disease of the apocrine glands that can involve the axillae, groin, submammary folds, perianal region, and genitalia. It is characterized by deep painful nodules, abscesses and draining fistulas that heal with scarring. HS usually develops in young adults and is more frequent in women than man. The etiology is unclear at present, but smoking, obesity, and familial predisposition are considered contributing factors. Current therapy consists of topical or systemic antibiotics, retinoids, and immunosuppressive agents, and only has a temporary or limited effect (4,6). For advanced stages of the disease, ablative surgery is essential (8).

Multiple case reports have been published showing promising responses of HS to off-label treatment with tumor necrosis factor α inhibitors (6). Recently, a placebo-controlled trial with adalimumab showed statistically significant improvements of skin lesions and pain in HS (7).

CD may have various dermatological manifestations, commonly including pyoderma gangrenosum (1,2). An increasing number of articles reports the co-occurrence of CD and HS, and hypothesize the association of the two diseases (3,9).

Interestingly, our patient developed two significant extraintestinal cutaneous complications. Pyoderma gangrenosum occurred shortly after the onset of CD and healed completely with conservative immunosuppressive treatment. Inflammatory lesions in the groins, appearing several years later, were at first thought to also be pyoderma gangrenosum.

However, further development of fistulas and abscesses with purulent draining was consistent with the diagnosis of HS.

The patient was treated with 40 mg adalimumab every other week with successful results, though clinical response rates in HS seem to be higher among those who received adalimumab weekly (7). To date, adalimumab is the only treatment that has been able to control both CD and HS in our patient. Furthermore, it was well tolerated and led to the improvement of her depression and quality of life.

HS is a disabling, difficult-to-manage disease, severely reducing the patient's quality of life. We suggest that TNF α inhibitors might represent a new therapeutic option for patients suffering from severe HS and may be useful in the treatment of CD as a comorbidity. Further studies are needed to better elucidate the efficacy of these drugs for the treatment of HS and to understand the association between CD and HS.

CONCLUSION

This case report demonstrates the likelihood of association between CD and HS the potential efficacy of adalimumab in the treatment and induction of long-term remission in both diseases.

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