Adalimumab in severe hidradenitis suppurativa – a case report of a novel experience

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Abstract

Hidradenitis suppurativa (HS) is a chronic inflammatory disabling disorder which affects apocrine gland bearing skin, characterized by discharging sinuses, nodules and abscesses that relentlessly progress and frequently cause scarring, contractures and immobility. Exact pathogenesis of the HS remains incompletely understood. Current theories suggest follicular obstruction and rupture leading to subsequent inflammation. Therapeutic options are often unsatisfactory in severe cases. However, biologics have shown promising results recently and tumor necrosis factor - α (TNF-α) inhibitors have been reported as effective therapy in severe cases. Herein we report a case of severe recalcitrant HS which improved with adalimumab.

Keywords: hidradenitis suppurativa, biologics, TNF-α inhibitor, adalimumab

Introduction

HS is a complex, chronic and disfiguring disease characterized by inflamed nodules, sinus tracts and abscesses leading to scarring. It primarily involves apocrine gland bearing skin with a predilection for the intertriginous areas. Hence, it is also called acne inversa and is part of the follicular occlusion tetrad. Prevalence of HS is estimated at 4.1% with a female predominance. HS typically present in post pubertal individuals with the symptoms of discomfort and/ or itching associated with tender papules or deep-seated nodules which can lead to abscesses and sinus tract formation. Individual lesions may resolve completely but often recur in groups. Lesions heal with fibrosis and dermal contractures.

The pathogenesis of HS is not fully understood, but it is likely that a multitude of factors play a role. Follicular occlusion, follicular rupture and an associated immune response appear to be important events in the development of HS. Genetic susceptibility, mechanical stresses on the skin, obesity, smoking and hormonal factors are also associated with the pathogenesis of HS. The contribution of the immune system in the HS has become apparent by the favorable response of HS to TNF-α inhibitors.

Despite the availability of a variety of treatment options including topical and systemic antibiotics, retinoids and surgery, severe HS often ends up with frequent relapses and severe morbidity. In this era of biologics, TNF-α inhibitors have drawn interest as effective therapy in severe cases.

Although infliximab, another TNF-α inhibitor and a chimeric monoclonal antibody, has been tried in Sri Lanka previously, patients who showed response initially developed loss of response over time. This secondary loss of response to infliximab even with the optimal dosage may occur due to antibody formation of the drug due to chimeric nature of the infliximab.

We present a case of a young man with severe recalcitrant HS which previously responded poorly to conventional antibiotics, surgery and acitretin whom we treated with adalimumab with marked clinical improvement.

Case report

A 23-year-old Srilankan young man with a 6-years history of HS presented with multiple recurrent painful discharging sinuses and nodules involving both axillae and groins. Prior treatment for his HS included multiple courses of oral antibiotics for more than 4 years, frequent surgical drainage of abscesses and acitretin 30 mg per day for last 12 months. These therapies yielded very little improvement and the patient continued to have severe pain and severe disease. He has had no notable medical histories apart from one episode of acute glomerulonephritis following infected HS lesions. He was distressed and depressed due to this prolonged disability.

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Physical examination revealed an averagely built male with BMI (Body mass index) of 24 with multiple tender purulent discharging sinuses, nodules and cysts involving both axillae and groins with scarring and nodulocystic acne with scars involving face. Dissecting folliculitis of the scalp and pilonidal sinus were not present. His Hurley stage was iii, with a sartorius score of 19. Dermatology life quality index (DLQI) was 13 which indicating severe disease activity.

He was commenced on subcutaneous adalimumab as per guideline. 160mg on day 1 followed by 80mg on day 15 and 40mg on day 29. 40 mg of subcutaneous adalimumab was continued weekly.

Therapeutic improvement started to appear from the second dose with significant clinical, sartorius score and DLQI response. Lesions were completely healed with scarring after 6th dose of adalimumab.

Discussion

HS often troubles both patient and dermatologist due to its chronic nature. Severe cases are often difficult to treat and needs combination therapy.

The initial pathological event in HS is a follicular hyperkeratosis with plugging and dilation of the hair follicle that causes inflammation. This persistent inflammation appears to be the main mechanism leading to HS chronicity, progression, and irreversible structural damage. A key therapeutic target may therefore be the control of the underlying inflammatory process before significant and irreversible structural damage occurs. Tumor necrosis factor α is a potent and central ubiquitous proinflammatory cytokine. Adalimumab is a fully human monoclonal IgG1 antibody specific for TNF-α that is administered subcutaneously.

Our patient has had a severe case of HS, for which numerous aggressive therapies have failed. Adalimumab elicited a rapid and sustained clinical improvement after second injection. After 1 month of commencement of adalimumab all sinus discharges and cyst had subsided with healing of ulcers.

Figure *1a & 1b axillae lesions before starting adalimumab, 2a & 2b before 2nd dose of adalimumab, 3a & 3b before 3rd dose of adalimumab, 4a & 4b completely healed lesions.*

Adalimumab is the only internationally registered biologic for HS and should be considered in severe cases when conventional therapies appear insufficient, especially in young people.

Although it has high safety profile and easy to administer compared to infliximab, adalimumab is not without additional risks. It has a black box warning recommending evaluation and treatment of latent tuberculosis. Proper pre evaluation before selecting the patients to be done to avoid unnecessary treatment complications.

In addition to the potential side effects, the introduction of adalimumab in the management of HS raises questions regarding the cost-efficacy of treatment especially in a developing country like Sri Lanka. Every week therapy roughly cost about 75000 rupees. But a study done in Greece showed that treatment with anti-TNFs does achieve a significant containment of HS exacerbations as well as significant cost-savings compared to the expenditure for the hospitalization, out-patient treatments and surgery for severe HS.

Given its strong anti-inflammatory properties, along with its convenient subcutaneous dosing regimen, adalimumab represents a therapeutic option for patients with severe HS and is to be considered as a next line option once the conventional therapies fail to show the response in severe HS.

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