

Case Report

A Chinese man with chronic recalcitrant hidradenitis suppurativa successfully treated with infliximab

以因福利美成功治癒華人男性患者慢性頑固性化膿性汗腺炎

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A 31-year-old Chinese male suffered from recalcitrant hidradenitis suppurativa for seven years causing disfiguring scars over the face and intertriginous areas, particularly the axillae and groins. Multiple medical treatments and surgical operation were tried but in vain. Infliximab infusion led to significant improvement. To our best knowledge, this is the first Chinese patient with hidradenitis suppurativa treated with infliximab in Hong Kong.

31歲中國男性患有化膿性汗腺炎七年，面上、腋窩、腹股溝都長滿了疤痕。多種藥物治療和外科手術都沒有成效。當轉用生物製劑因福利美（infliximab）後，結果病情有明顯的改善。就我們所知，這是香港首名化膿性汗腺炎病人使用生物製劑因福利美（infliximab）。

Keywords: Ankylosing spondylitis, hidradenitis suppurativa, infliximab

關鍵詞：強直性脊椎炎，化膿性汗腺炎，因福利美

Introduction

Hidradenitis suppurativa (HS) is a chronic inflammatory dermatosis of unknown aetiology. It affects mainly the apocrine gland bearing areas,

especially the axillae, groins and perineal regions. HS is believed to be due to follicular occlusion and together with acne conglobata, dissecting cellulitis of the scalp and pilonidal sinus, is known as the follicular occlusion tetrad.¹ It is characterised by recurrent formation of multiple abscesses and sinuses causing dystrophic scarring. Its chronicity and poor response to traditional medical therapy are debilitating to both the patient and physician. Radical surgical excision with healing by secondary intention remains the mainstay of therapy but carries the risk of additional scarring. Scarring may lead to contractures and loss of limb mobility.² Recently, promising results have been reported with the use of biologics in many inflammatory

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diseases such as rheumatoid arthritis, ankylosing spondylitis, Crohn's disease, psoriasis and psoriatic arthropathy. We report a 31-year-old Chinese male with recalcitrant HS and HLA B27+ve ankylosing spondylitis unresponsive to multiple systemic treatments and surgeries treated with infliximab. To the best of our knowledge, this is the first Chinese patient with HS treated with infliximab in Hong Kong.

Case report

A 31-year-old Chinese male was first seen in February 2003 because of severe acne over the face and upper trunk. He also complained of multiple sinuses with discharge over the intertriginous areas of axillae and groins for two years. The scalp was unaffected. Physical examination revealed multiple papules and pustules over bilateral cheeks, chin and upper trunk. Multiple large sinuses with yellowish discharge from bilateral axillae were found (Figure 1). Furthermore, extensive scarring over face, upper trunk, axillae and groins were present. The clinical diagnoses were hidradenitis suppurativa and acne conglobata.

He responded poorly to multiple systemic and topical antibiotics, including doxycycline, minocycline, metronidazole, amoxicillin/clavulanate, clindamycin, ofloxacin and erythromycin. Intralesional injection of triamcinolone into recalcitrant lesions also failed to control the disease. Wide excision of both axillae was performed but only resulted in increased scarring. Subsequent treatment with oral isotretinoin resulted in further deterioration and had to be discontinued. The patient also suffered from HLA B27+ve ankylosing spondylitis that was treated with sulphasalazine and non-steroidal anti-inflammatory drugs by rheumatologist.

A skin biopsy from the left axilla revealed active chronic inflammation with scarring compatible with chronic HS. In view of the refractory disease in our patient, treatment with intravenous infliximab infusion was given. Baseline investigations included complete blood picture, liver and renal function tests, and erythrocyte sedimentation rate (ESR). He had no symptoms and signs suggesting active pulmonary tuberculosis infection and both chest x-ray and Mantoux test were unremarkable.

Infliximab 5 mg/kg (total 300 mg) was infused intravenously at 0, 2 and 6 weeks. The infusion was initiated at 10 ml/hr and then increased to 20 ml/hr, 40 ml/hr and 80 ml/hr every 15 minutes to avoid fluctuation in blood pressure. Afterwards, the infusion rate was increased to 150 ml/hr for next 30 minutes and finally 250 ml/hr until the end of therapy.

Significant improvement was noted after three courses of infliximab, beginning after the second dose of infusion and becoming clinically obvious around week 4. Clinically, the sinuses started to heal with significant reduction of discharge (Figure 2). The inflammatory markers demonstrated an even earlier improvement, observable immediately after the first dose of infliximab infusion. The ESR dropped from a baseline of 102 mm/hr to 35 mm/hr after the first dose (week 0) and further down to 20 mm/hr after the second infusion (week 2) and 6 mm/hr after completing the third dose (week 6). C-reactive protein (CRP) also showed similar improvement from a baseline of 48.4 mg/L (normal <5 mg/L) to normal (4.7 mg/L) after the first dose (week 0) and <3.2 mg/L after the second infusion (week 2). Reactive thrombocytosis at baseline ($543 \times 10^9/L$; normal range $145-370 \times 10^9/L$) believed to be due to the inflammatory effects of HS was also corrected after infliximab infusion, falling



Figure 1. A large sinus over left axilla (before infliximab treatment).



Figure 2. The sinus at left axilla healed after three doses of infliximab.

to 492 at week 1 and then to the normal value of 272 after the second infliximab infusion (week 2).

Furthermore, better symptomatic control led to improvement in quality of life. Prior to treatment with infliximab, our patient's daily activities and social activities were limited by low back pain and knee pain secondary to HLA B27 positive ankylosing spondylitis and embarrassment due to malodour and soaking of clothes from the axillary discharge. After infusion of three doses of infliximab, his joint pain decreased dramatically. The axillary discharge almost cleared leading to improvement in morale.

Our patient did not have any major side effects during infusion and the three doses of infliximab were completed uneventfully. A small staphylococcal abscess occurred over his occipital scalp approximately 2 weeks after the third dose of infliximab, which subsided after simple drainage and a course of oral ampicillin and cloxacillin. A repeat chest X-ray after completing three doses of infliximab infusion did not reveal any changes or evidence of pulmonary tuberculosis.

Discussion

Hidradenitis suppurativa is a recalcitrant inflammatory dermatosis that often causes frustration to patients and physicians. The results of various medical therapies including antibiotics, oral retinoids, dapsone, steroid and immunosuppressive have been disappointing.² The recent use of biologics for treatment of HS has demonstrated promising results. In 2003, Sullivan et al reported five patients with HS who had significant decrease in both patient's self-reported disease activity scores and physician-observed clinical improvement after the treatment with infliximab.³ Lebwohl and Sapadin reported the use of infliximab to treat HS with nearly complete re-epithelialisation of lesions and reduction of pain at around five weeks after the third dose.⁴ Adams et al also treated a patient with 3-year history of HS with the same regimen of infliximab (5 mg/kg at 0, 2, 6 weeks) who had complete resolution of pain, purulence, discharge, malodour and with greatly reduced inflammation.⁵ Rosi et al reported a 30-year-old woman with HS and Crohn's disease treated

with infliximab 5 mg/kg at 0, 2, 6 weeks demonstrating significant improvement just only two weeks after the first dose.⁶ In 2006, Cusack and Buckley used etanercept 25 mg subcutaneously twice weekly for six patients with severe HS who responded with a marked reduction in self-reported disease activity and Dermatology Life Quality Index (DLQI).⁷ Furthermore, Moul and Korman treated a patient with a twenty year history of HS with adalimumab subcutaneously (40 mg every other week) with significant improvement.⁸

Infliximab is a chimeric monoclonal antibody composed of a murine antigen-binding region joined to the human IgG1 constant region. It has a molecular weight of 149,100 Da with binding specificity for human TNF- α .⁹ Each molecule of infliximab can bind two molecules of TNF- α and binds to both monomers and trimers of TNF- α thus blocking all receptor-binding sites. This stable binding complex formed by infliximab and TNF- α prevents binding of TNF- α with its receptor sites,⁹ and leads to lysis of TNF- α producing cells. Subsequently, it induces apoptosis of these activated lymphocytes. The final reduction in TNF- α producing cells may contribute to its clinical anti-inflammatory effects.

Infusion reactions like fever, dyspnoea, urticarial rash, arthralgias, myalgia and fluctuation in blood pressure have been reported in around 16% of patients.⁹ Another concern in using infliximab is infections such as reactivation of tuberculosis. This is particularly relevant in Hong Kong where tuberculosis is endemic. Since TNF- α plays a role in the host defence, blocking its activity may predispose the patients to higher risk of infections. Our patient did not have any evidence of tuberculosis infection after infliximab infusion and no adverse reaction was reported during infusion. His only adverse effect probably due to infliximab

infusion was the small skin abscess at the occipital scalp that subsided after simple drainage and a course of antibiotics. In a report by Fardet et al, three patients with HS treated with infliximab were found to have abdominal pain caused by colon cancer, multifocal motor neuropathy with conduction block and severe allergic reaction respectively.¹⁰ A lupus-like syndrome has been reported as a rare adverse effect of infliximab. One patient developed infliximab induced lupus-like reaction with arthralgia, diffuse alopecia and positive anti-nuclear antibody (ANA) after eleven months of infliximab infusion for HS. The clinical symptoms and serological markers improved after stopping the drug.¹¹ This lupus-like syndrome was also seen in patients with rheumatoid arthritis using infliximab. Despite the fact that ANA was positive up to 62% of infliximab treated patients when compared with 27% in the control group, the development of lupus-like syndrome was rare.¹²

In summary, we reported a 31-year-old Chinese man suffering from a seven year history of recalcitrant hidradenitis suppurativa and HLA B27 positive ankylosing spondylitis who showed a significant improvement after three doses of infliximab infusion without major adverse reactions.

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