Concomitant alopecia areata and hypertrichosis after infliximab therapy: rara avis

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ABSTRACT

We present a 37 year-old man with HLA-B27 positive ankylosing spondylitis for the last 3 years. Interestingly, he developed both alopecia areata and hypertrichosis simultaneously following infliximab treatment. Reporting this interesting patient of ours -to our best notice for the first time in the literature- we call attention of clinicians to the contradiestinctive effects of anti-TNF-α agents on hair growth cycle.

Keywords: Ankylosing spondylitis; TNF-α; Infliximab; Alopecia; Hypertrichosis

INTRODUCTION

The role of TNF-α in the pathophysiology of autoimmunity, and more specifically of alopecia areata appears complex. On the one hand, TNF-α has been considered an important cytokine involved in the hair loss, the more so because it inhibits hair growth in vitro and TNF-α producing cells can be found in the mononuclear infiltrate surrounding the hair follicle¹. On the other hand, TNF-α appears to protect from hair loss since the blockade of TNF-α mediated effects by monoclonal antibodies resulted in the worsening of alopecia areata with infliximab².

In this report, we present a patient who developed alopecia areata and hypertrichosis simultaneously following infliximab treatment – to our best notice for the first time in the literature. Our report suggests that anti-TNF agents can be causally related to the development of alopecia areata concurrently with hypertrichosis in the same patient.

CASE REPORT

A 37 year-old man with HLA-B27 positive ankylosing spondylitis of 3 years duration, who had been treated with different non-steroidal anti-inflammatory drugs and sulfasalazine but who had only partial improvement, was seen in our center. Due to the persistence of bilateral sacroiliac pain, and loss of lumbar range of motion, biological therapy with infliximab was started at an intravenous dose of 5 mg/Kg and thereafter his symptoms (daytime and night-time sacroiliac pain) started to improve. On the other hand, after the second infusion, he suffered both nonscarring, patchy hair loss, typical of alopecia areata in the abdominal region and the front of both thighs (Figure 1), and hypertrichosis in the forearms and hands (Figure 2) simultaneously. Complete blood count, liver/renal function tests were normal; antinuclear and anti-dsDNA antibodies were negative. On further questioning, he denied personal or family history of hair loss, atopy and hypertrichosis. Accordingly, the patient’s findings were attributed to infliximab infusion. Therapy with infliximab was discontinued and the patient evolved favourably, with recovery of abdominal and thigh hair in 2 weeks and no further alopecia, also recovery of hypertrichosis in 5 weeks.

DISCUSSION

Although anti-TNF-α agents are expected to treat autoimmune-related processes similar to ankylosing spondylitis, these drugs have been associated with the development of other autoimmune diseases such as alopecia areata³. It is an autoimmune inflammatory disea-

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...se, in which T-lym phocytes play a central pathogene -
tic role. Although the exact pathophysiology of alope-
cia areata remains unknown, it is postulated that CD4+ 
and CD8+ T cells reactive to hair bulb autoantigens in-
duce the autoimmune process leading to non-scarring 
hair loss.

Recent fundamental studies have shown that TNF-α 
promotes both the expansion and suppressive func-
tions of CD4+ regulatory T cells which play an essen-
tial role in maintaining immune tolerance and pre-
venting autoimmunity. Therefore, it is possible that 
anti-TNF-α induced autoimmunity is mediated by the 
inhibition of the suppressive functions of regulatory T 
cells.

Philpott et al. showed that IL-1α, IL-1β and TNF-α 
were potent inhibitors of hair follicle growth in vitro. 
On the other hand, a few cases of patients develo-
ping AA after biological therapy have been reported. 
Our case suffered both nonscarring, patchy hair loss, 
typical of alopecia areata in the abdominal region and 
the front of both thighs, and hypertrichosis in the fo-
rearm simultaneously after the treatment of infliximab. 
We believe that in our patient, the eventual scenario 
might be due to complex mechanisms pertaining to 
TNF-α effects on hair growth cycle.

CONCLUSION

To summarize, presenting this interesting patient of 
ours in whom alopecia areata and hypertrichosis en-
sued concurrently after the treatment of anti-TNF-α 
therapy, we draw attention of physicians towards this 
rare eventuality that can occur due to contradi

FIGURE 1. Alopescia areata in the different regions of the body. 
(A) Abdominal region; (B) Right thigh; (C) Left thigh

FIGURE 2. Hypertrichosis in forearms and hands

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