

## Case Report

# Severe Recalcitrant Acrodermatitis Continua of Hallopeau with Complete Response to Infliximab

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## Abstract

Acrodermatitis continua of Hallopeau (ACH) is considered a rare variant of pustular psoriasis characterized by chronic and recurrent pustular eruptions of the distal fingers and toes as well as atrophic skin changes, onychodystrophy and osteolysis. ACH is traditionally very recalcitrant to topical and systemic anti-psoriatic agents. We report on a 44 year-old female with severe debilitating ACH who was successfully treated with infliximab for 11 years with complete clearance.

## ABBREVIATIONS

ACH: Acrodermatitis Continua of Hallopeau

## INTRODUCTION

Acrodermatitis continua of Hallopeau (ACH) is considered a rare variant of pustular psoriasis characterized by chronic and recurrent acropustular eruption of the distal fingers and toes as well as atrophic skin changes, onychodystrophy and osteolysis [1]. The trigger for eruption is yet to be deciphered, but on occasion has been reported to develop in the context of both plaque and pustular psoriasis as an initial presentation or subsequent manifestation [2]. ACH is an exceptionally challenging condition proved to be a challenging condition to treat. Being invariably recalcitrant to all topical and systemic anti-psoriatic agents, it has recently been reported to respond to tumor necrosis factor-alpha inhibitors as well as other immune modulators. There is limited guidance for treatment of ACH, which emphasizes the importance of case reports to direct specific treatment of this debilitating disease.

## CASE PRESENTATION

We present a 44 year-old female who was referred to our specialty psoriasis clinic with an 18-month history of disabling Acrodermatitis Continua of Hallopeau (ACH). When first seen, she presented with her hands totally enwrapped in towels. Her husband had recently divorced her as a result of her skin ailment. Her condition started with a single area of her right thumb and progressed to severe pustular eruption with confluent lakes of pus oozing from all ten fingertips (Figure 1). There were minor remnants of nail on her fourth and fifth digits with associated

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- Acropustular psoriasis
- Dermatitis repens
- Acrodermatitis perstans
- Anti-tumor necrosis factor-alpha

multiple periungual discrete pustules and erythema. There was also an unusual greenish discoloration of the areas of pustulation that suggested pseudomonas bacterial involvement. Imaging revealed osteolysis of multiple fingers. The patient's past medical and family history were noncontributory. Her condition had been recalcitrant to all topical and systemic agents with the exception of a short-term partial response to high dose cyclosporine (7 mg/kg/day). Previous therapies included cyclosporine, PUVA soaks, methotrexate, acitretin, 6-thioguanine, azathioprine, cladribine, and thalidomide. The patient initiated immediately on infliximab 5mg/kg with infusions at weeks 0, 2, and 6 with maintenance infusions every 8 weeks. She also was treated with topical chloramphenicol for the pseudomonal involvement. Four months after initiation of treatment, she presented with a dramatic response. At her one-year follow up, her hands were totally clear



**Figure 1** Initial evaluation - severe acropustular eruption of all ten fingers.



**Figure 2** One year subsequent to maintenance infliximab treatment. Severe ACH resolved with significant nail re growth noted.

with significant nail re growth noted (Figure 2). The patient had regained significant functionality of her hands despite psoriatic arthritic changes and has remained clear for 11 years of follow up.

## DISCUSSION

Acrodermatitis continua of Hallopeau (ACH) were first described in 1888 by Radcliffe-Crocker [1]. It has since remained an uncommon clinical condition reported infrequently in the literature. Patients with active ACH experience significant impact on their quality of life due to reduced functionality of their hands and/or feet. Patients often struggle to maintain employment and carry out activities of daily living resulting in significant disability. While the condition is rare, it is imperative that specific guidelines for therapy are recommended to prevent further debilitating impact of the disease.

The increasing knowledge of the pathophysiology of psoriasis and the availability of multiple immune modulating therapies has encouraged dermatologists to experiment with the drug class for the treatment of ACH. As a result, there are a handful of case reports describing treatment with both TNF-alpha inhibitors and ustekinumab [1,3]. There are six reported cases of infliximab used for the treatment of ACH [4-9]. Challenges described with infliximab infusion treatments include rising antibody titers in two patients with resulting relapse of the ACH [5,7-9].

We submit this case as further long-term evidence to support the initial use of infliximab for patients with recalcitrant

ACH. Double blind, placebo controlled trials would be logical in assessing the efficacy and safety of biologic therapy in this patient population despite the limited number of cases of ACH. While we recognize the challenges of this type of study for such a rare condition, the debilitating nature of the disease certainly deserves more concrete treatment guidelines.

## CONFLICT OF INTEREST

Dr. Menter has been a consultant, researcher and/or invited lecturer for: Abbott Laboratories, Allergen, Amgen, Inc., Celgene, Eli Lilly, Galderma, Janssen, LEO Pharma, Novartis, Novo-Nordisk, Pfizer, Stiefel, SyntrixBiosystems, Wyeth. Dr. Menter has no stock ownership or Board Membership on any pharmaceutical or Biotechnology company. There are no other conflicts of interest to report.

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