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CASE REPORT

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Folliculitis decalvans managed with adalimumab: A case report

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Key Clinical Message

Folliculitis decalvans (FD) is a rare neutrophilic cicatricial alopecia of the scalp that manifest with inflammation, pruritus, pustules, and tufted hair. Most dermatologist treat FD with topical or oral antibiotics. We recommend considering treatment with biologics to preserve and stabilize the ongoing inflammatory process in moderate to severe FD.

KEYWORDS

anti-TNF, adalimumab, anti-tumor necrosis factor, dermatology, FD, folliculitis decalvans, TNF, trichology

1 | INTRODUCTION

Folliculitis decalvans (FD) is a primary neutrophilic cicatricial alopecia of the scalp that is challenging entity to manage due to the sparse number of reported cases in literature. To this date, there is no clear pathogenesis that explains FD; nonetheless, alteration in the local immune response and *Staph Aureus* colonization is the primary culprit.[1–3] As it is a progressive inflammatory disease, early age of onset is associated with severe form, but prognosis is undetermined due to lack of studies in the subject. Tufted hair and duration of the disease could predict a worse outcome and a recalcitrant disease.[1] At early stage of the disease, infundibular acneiform dilation is observed along with infra- and peri-follicular neutrophilic infiltration reaching to adventitia of the dermis. At later stages, dermal fibrosis predominates.[1] Herein, we present a case of recalcitrant FD that was managed and stabilized with Adalimumab 40 mg once weekly.

2 | CASE PRESENTATION

A 40-year-old female with history of psoriasis vulgaris presented to the dermatology clinic with a large patch of hair loss over the scalp, associated with pain and itchiness that progressed for years. Dermatological examination showed a central cicatricial alopecia accounting for 40% of the scalp, associated with pustules, crustation, dyspigmentation, tufted hair, fragile skin, and periauricular crustations (Figure 1). Hyperpigmented scaly patches were observed scattered all over the body (Figure 2). Multiple hyperpigmented lichenoid and reticulated patches over buccal mucosa are appreciated (Figure 2). The informed decision was obtaining skin biopsy. Histopathological evaluation showed acanthosis and hyperkeratosis, focal ulcer with acute and chronic inflammation, and abscess formation in the epidermis. The inflammation is seen surrounding the hair shaft with foreign body giant cell reaction with bacterial colonies (Figure 3). Swab culture from the abscess reported heavy growth of isolated *Staphylococcus aureus*. Doxycycline 100 mg twice daily was started for 1 week. In addition, topical betamethasone valerate 0.1%, clindamycin topical solution, and ketoconazole shampoo were all prescribed. Adalimumab (Humira®) has been reported to be utilized as an off-label regimen in recalcitrant FD cases. Thus, Adalimumab (Humira®) was initiated following the hidradenitis suppurativa (HS) prescribing protocol. Serology, basic screening for liver function test, and tuberculosis screening were all within accepted parameters; thus, Adalimumab 80 mg loading dose was administered at week 0, and 40 mg every other week. At follow-up examination after 3 months, stabilization of cicatricial alopecia is appreciated with alleviation of inflammatory pustules (Figure 4). No serious adverse events were reported throughout period of care. At 10-month follow-up, no new foci of scarring alopecia or inflammation have manifested since Adalimumab initiation.

3 | DISCUSSION

As FD is a rarely encountered ailment, there are no treatment guidelines established. A multicenter series of 60 patients proposed a therapeutic protocol for FD management.[1] Their proposed treatment centered around oral antibiotics and topical treatments; however, relapses were more frequent in their reported patients.[1,3,4] Tetracycline were administered for 2–3 months and had 91% response rate in moderate to severe FD.[1] Oral antibiotics are inadequate treatment in FD as the relapse rate with oral antibiotic ranged from 57%–80% in one study.[5]

Another case series, done on 39 male patients, assessed efficacy of isotretinoin. Administration of 0.1–1.02 mg/kg/day (10–90 mg/day) of oral isotretinoin had 82% response rate, with superior outcome demonstrated with 0.4 mg/kg/day prescribed for more than 3 months.

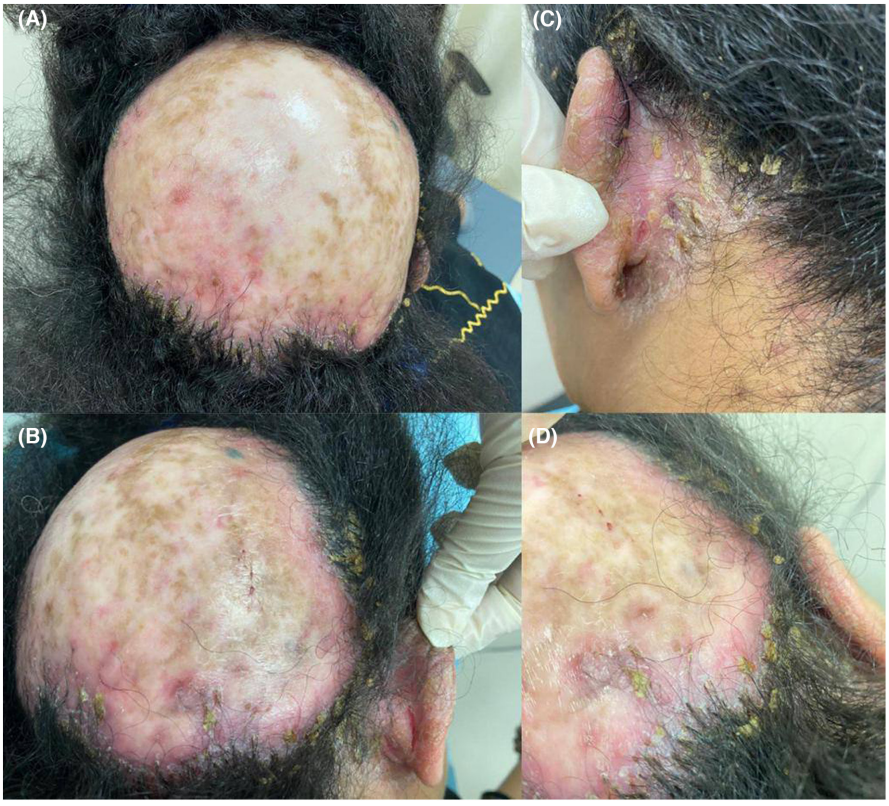

FIGURE 1 (A) Erythema, patches of hypo- and hyper-pigmentation, multiple tufted hair around periphery of the scalp over the 6 o'clock position. (B) Erythema with scales and crusts formation around left ear. (C) Cicatricial alopecia of the scalp with close-up to the ongoing inflammation, pustules, crust formation, and severe erythema around periphery of the scalp. (D) Close-up to the intense erythema, pustules, and crusts around scalp periphery, denoting to an active FD.

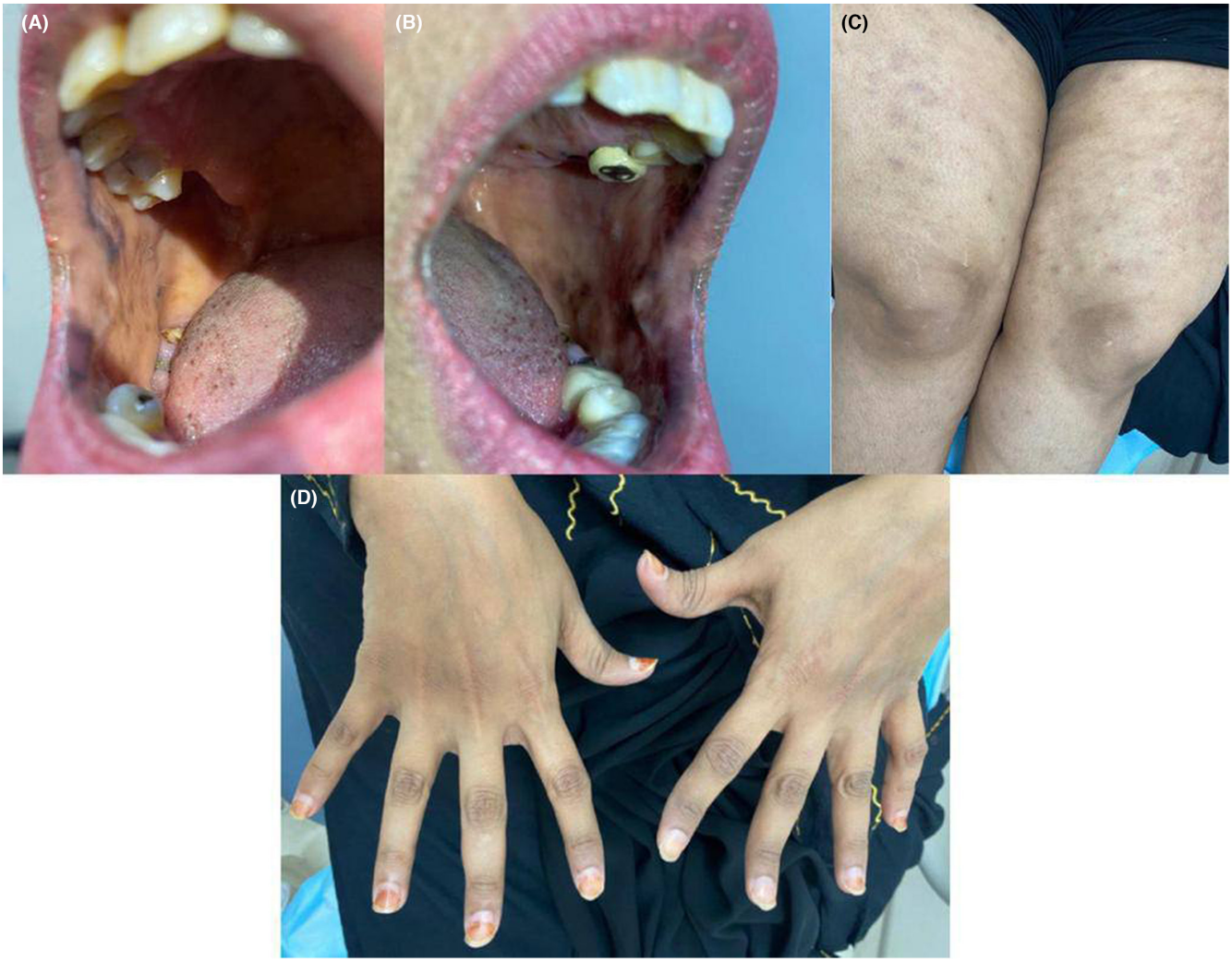

FIGURE 2 (A, B) Hyperpigmented lichenoid reticulated patches over buccal mucosa. (C) Multiple Hyperpigmented patches scattered around bilateral thighs. (D) No nails changes.

Relapse rate was 34%.[6] It is proposed that isotretinoin mechanism of action in FD is inhibiting neutrophil migration in the inflamed skin by decreasing the toll-like receptor that mediates immune response against gram-positive bacteria.[5]

Collier et al[7] have reported a 26-year-old male with recalcitrant FD who was successfully controlled with systemic photodynamic therapy (PDT) using ultraviolet light (100–140 J/cm^2) with porfimer sodium 1 mg/kg as monotherapy. Our patient had marked FD accounting for 40% surface area; Collier et al[7] reported a young male patient with localized FD involvement. Their patient's presentation and youth made it preferable to utilize PDT 1 mg/kg to avoid radiotherapy. Their patient had solely improved with PDT, and all topical and oral antibiotics have been stopped 5 months prior to treatment. At our institution, PDT and systemic photosensitizers are unavailable.

Lin et al[8] have also reported the utilization of photodynamic therapy with topical aminolevulinic acid in managing a recalcitrant FD in an adult 63-year-old male. Photodynamic therapy demonstrated an immune-modulatory and antibacterial action, improved scar healing, anti-inflammatory properties, and can target fibroblasts, keratinocytes, sebaceous glands, and hair follicles.[8,9] Lin et al[8] reported FD stabilizing after 4 cycles of PDT at 2-week intervals. Their patient had received systemic antibiotics and isotretinoin for FD without improvement. And due to his liver damage, their approach is to utilize PDT as a sole treatment regimen. Due to the delayed and severe presentation of our patient and presence multiple hair tufts, our decision is not to delay the introduction of Adalimumab by a trial of antibiotics. As multiple studies have shown greater relapse rate and no improvement.

Another case series of 4 patients treated with textile PDT, 12 and 37 J/cm^2, have demonstrated an excellent tolerance and great outcomes.[10] One patient from the previous report, a 24-year-old male, has been prescribed

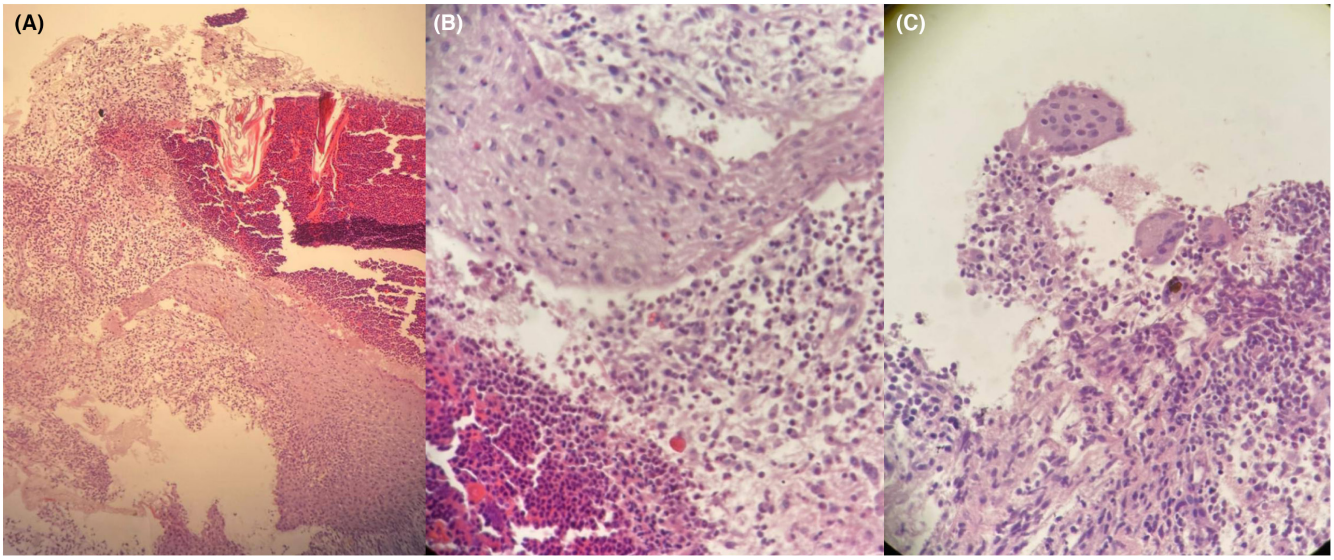

FIGURE 3 (A) There are dense perifollicular neutrophilic infiltrates with dense dermal infiltrate composed of lymphocytes, plasma cells, and abundant neutrophils, Hematoxylin and Eosin (H&E) stain, 100x. (B) Perifollicular collections of neutrophils (H&E), 400x. (C) The extruded hair fiber has stimulated granulomatous reaction, (H&E), 400x.

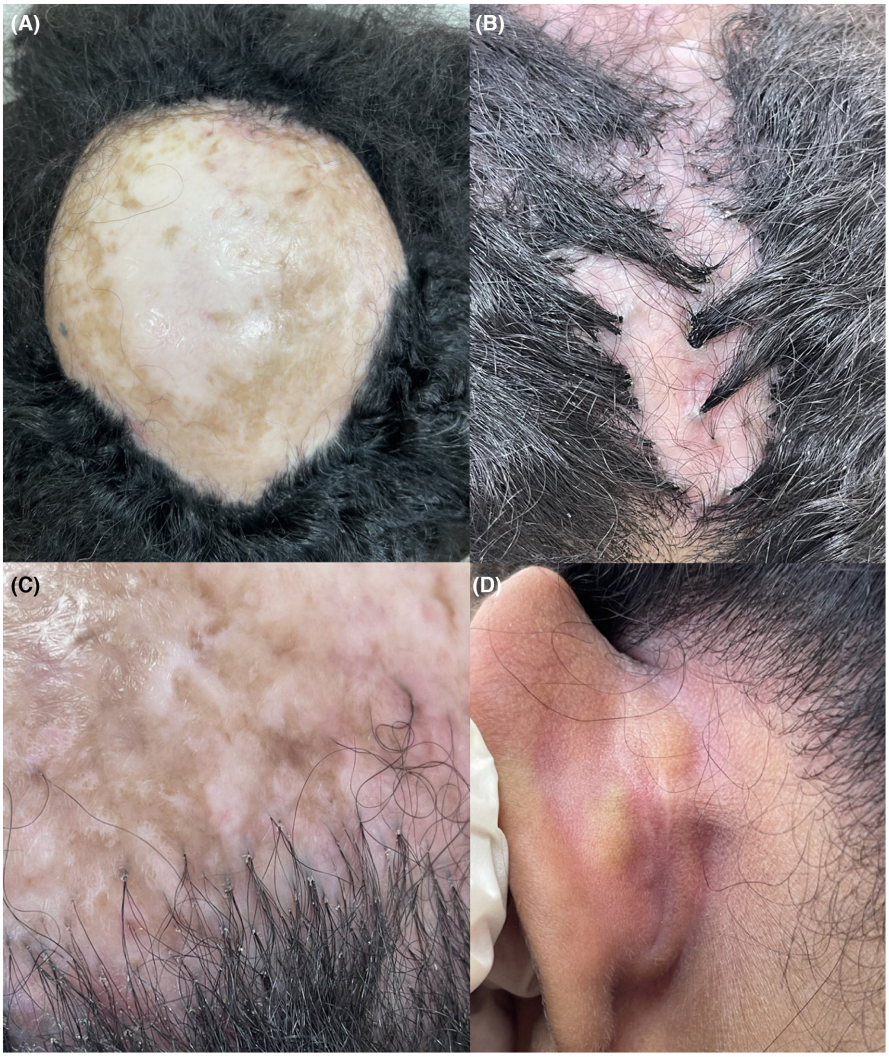

FIGURE 4 (A) Crown of the scalp showing alleviation of the inflammation 3 months after Adalimumab initiation. (B, C) Tufting of hair is apparent. No erythema, pustules, crusting, or suppuration is noted. (D) Close up behind the ear demonstrating improvement of the crusting and inflammation.

Adalimumab and undergone 37 J/cm^2 textile PDT sessions. It is believed that combining Adalimumab, anti-tumor necrosis factor (TNF)-α, and PDT had synergistic effect in managing recalcitrant FD.[10]

Alhameedy et al[11] have utilized Adalimumab in a 54-year-old female with recalcitrant FD who achieved remission after 3 months, with one relapse after drug unavailability. Their patient had undergone topical, systemic (acitretin and isotretinoin), and intralesional injections without any clinical improvement.[11] Our patient was started with Adalimumab and showed improvement after 1 month and remission after 3 months.

Adalimumab has been utilized in managing recalcitrant FD in four cases from literature, three females (aged 50, 58, and 68 years) and one male (aged 23 years).[12,13] As there are no treatment guidelines in FD and all therapeutic interventions are recommendations, the initiation of Adalimumab was on off-label basis and following the HS dosing.[11–13]

Adalimumab (anti-TNF-α) is a human recombinant IgG1 monoclonal antibodies with specificity for human TNF.[11] It has an anti-inflammatory property and is utilized for a variety of dermatological and autoimmune diseases. Off-label use of Adalimumab had been reported in multiple neutrophilic dermatosis, including pyoderma gangrenosum and dissecting cellulitis.[14,15] Due to fear of preserving the remaining hair follicles and decreasing inflammation, our team initiated Adalimumab subcutaneous injections following HS prescribing protocol.[10–13]

Iorizzo et al[16] have reported a case series of 23 patient with biopsy-proven FD, 21 of them were successfully managed with Adalimumab as disease progression and inflammatory process have halted. Two subjects have failed to show any clinical response. Their patient is still undergoing Adalimumab to prevent recurrence; nonetheless, recurrence can be managed by restarting Adalimumab as Alhameedy et al[11] have reported. Their Adalimumab administration was 160 mg at week 0, 80 mg at week 2, and 80 mg every other week.[16] However, we followed the HS prescribing protocol 80 mg at week 0 and 40 mg every other week. Our patient continued to clinical improving and no new foci of scarring alopecia or inflammation have presented since Adalimumab initiation.

We advocate for further reports that demonstrate the outcome of recalcitrant FD patients who have been treated with innovative therapeutic interventions, such as Adalimumab. Antibiotics are utilized in effort to alleviate the ongoing inflammatory process. Nonetheless, it is associated with increased resistance, and higher chances of relapse after completing the regimen. We recommend starting Adalimumab in moderate to severe FD to preserve the remaining hair follicles, halt the ongoing inflammatory process, and prevent any new foci of scarring.

The patient in our report have presented late with severe active FD. Proper laboratory evaluation and histological correlation to confirm the diagnosis of FD have been made in efforts to initiate Adalimumab to halt the ongoing inflammatory process, stabilize the scarring alopecia, and preserve the remaining hair follicles.

AUTHOR CONTRIBUTIONS

Adel Alsantali: Conceptualization; supervision; writing – review and editing. **Razan Baghdadi:** Investigation; visualization; writing – review and editing. **Yara Alghamdi:** Conceptualization; writing – original draft; writing – review and editing. **Elham Bin Abbas:** Data curation; software. **Rafah Ghazi:** Investigation; software.

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The patient had agreed to participate and to publish the findings of this research.

DATA AVAILABILITY STATEMENT

Data available on request due to privacy/ethical restrictions.

CONSENT

Written informed consent has been obtained from the patient to publish this report in accordance with the journal's consent policy.

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