Localized Follicular Mucinosis in a Child Treated Successfully with Pimecrolimus and Targeted Phototherapy

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Abstract
Follicular mucinosis (FM)/alopecia mucinosa is not commonly seen in children. There are a limited number of case reports, and its prognosis and long-term outcome are unpredictable. We describe a case of FM in a 3-year-old child, which was confirmed on histopathology. The lesion resolved with topical pimecrolimus cream and targeted phototherapy with an excimer lamp and has not recurred.

Introduction

Follicular mucinosis (FM) is rarely seen in children. Topical corticosteroids, antimalarial drugs, minocycline, bexarotene, and UV phototherapy are few of the available therapies. We treated our patient effectively with topical pimecrolimus cream and targeted phototherapy with an excimer lamp. A solitary lesion of FM/alopecia mucinosa in children rarely evolves into FM. Nevertheless, a long-term follow-up is highly essential.
Case Report

A healthy 3-year-old boy presented with a 3-month history of an asymptomatic, well-circumscribed indurated rounded plaque (4 cm in diameter), located on his glabellar forehead, with follicular prominence and mild scales at places (Fig. 1a). Desonide cream was used for 3 weeks before, which was ineffective. The lesion was gradually but slowly evolving with loss of hair affecting the medial part of left eyebrow. Skin biopsy showed (Fig. 2a, b) patchy lichenoid and dense perifollicular lymphocytic infiltrate, with clustering of lymphocytes in the outer root sheath of the hair follicles (folliculotropism). Follicular epithelium showed moderate reticular epithelial degeneration with intercellular stands of bluish stained mucin confirmed with Alcian blue stain (Fig. 2). Epidermotropism or atypia was not seen; hence, immunohistochemistry and T-cell receptor gene rearrangement studies were not done.

Although spontaneous remission is known in the pediatric age group, various therapies have been tried including topical and intralesional corticosteroids, retinoids including bexarotene, minocycline, antimalarial drugs, UVA and NBUVB phototherapy, calcineurin inhibitors, etc. All the reported therapies are anecdotal. The condition itself being rare, controlled studies are not available. Although this condition is known to be self-limiting, keeping in mind even the least possibility of transformation into MF, we treated this child with a nonaggressive therapy, application of pimecrolimus 0.1% cream once a day combined with excimer light therapy (Exciplex: 308 nm: 100–180 mJ) once in a week with eye shields for protection. The infiltrated plaque started thinning out gradually and resolved completely (Fig. 1b) over 6 months (25 sessions of excimer light therapy). As of now, there is no recurrence on following up for more than a year.

Fig. 1. a Well-circumscribed infiltrated plaque on the glabella with partial alopecia of the left eyebrow. b 6 months after treatment with pimecrolimus and excimer lamp phototherapy.

Fig. 2. a H&E 10 X: Patchy perivascular and dense perifollicular infiltrate of lymphocytes with folliculotropism. b 40 X: Moderate reticular follicular epithelial degeneration with intercellular stands of bluish staining mucin. c Alcian blue 40 X: mucinous degeneration and mucin deposits.
Discussion

FM is not commonly seen in children. Precise diagnostic and prognostic criteria for FM in children are still undetermined [1]. FM getting transformed into mycosis fungoides or Hodgkin's lymphoma has been reported but again is not so common, especially in children having a solitary lesion over the head and neck region [2].

FM is a variant of mycosis fungoides with localized disease and excellent prognosis. Thus, if FM and CTCL are in the same spectrum of diseases, the same treatment could be applied for both [3]. There are studies that show a significant improvement in FM with PUVA therapy [4, 5].

In our case, Desonide cream was used for 3 weeks earlier did not have any effect. Considering the age and the location of the lesion (glabella), we thought of treating with topical pimecrolimus cream and targeted phototherapy at 308 nm. The lesion resolved completely in 6 months. We are regularly following up the case every 3 months, and there is no recurrence even after a year. Both calcineurin inhibitors and excimer light are known to induce T-cell apoptosis [6–8].

This safe and effective combination therapy of FM, a rare disorder in children, is hitherto unreported to the best of our knowledge. More number of studies will help to establish our findings.

Statement of Ethics

The paper is exempt from the Ethical Committee Approval as it is a retrospective case report. Written informed consent was obtained from the parent/legal guardian of the patient for the publication of the details of their medical case and any accompanying images. Ethical approval was not required for this study in accordance with local/national guidelines.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Sharad Mutalik has made a substantial contribution to conception and designing of the manuscript. He gave guidelines not only on the manuscript preparation but also for designing the manuscript and reviewing it later. A major contribution was of intellectual content and literature search. Balkrishna Nikam is a dermatopathologist, whose guidance and expertise helped in interpretation of histopathological changes and to reach the diagnosis of the case, and who contributed to manuscript editing and image editing and reviewing. Contribution was instrumental in the manuscript preparation. Tejaswini Salunke is actively involved in drafting the manuscript, in literature search, and in revising the manuscript for intellectual content and contributed to concept visualization and design. All three authors have given a final approval to the manuscript to be published and are accountable for all aspects of the work.
Data Availability Statement

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

References

1. Alikhan A, Griffin J, Nguyen N, Davis DM, Gibson LE. Pediatric follicular mucinosis: presentation, histopathology, molecular genetics, treatment, and outcomes over an 11-year period at the Mayo Clinic. Pediatr Dermatol. 2013 Mar–Apr;30(2):192–8.
2. Hooper KHBR, Brown JA. Idiopathic follicular mucinosis or mycosis fungoides? Classification and diagnostic challenges. Cutis. 2015;95:E9–14.
3. Fernández-Guarino M, Castaño AH, Carrillo R, Jaén P. Primary follicular mucinosis: excellent response to treatment with photodynamic therapy. J Eur Acad Dermatol Venereol. 2008 Mar;22(3):393–4.
4. Borgia F, Giuffrida R, Lentini M, Palazzo R, Cannavò SP. Follicular mucinosis with diffuse scalp alopecia treated with narrow-band UVB phototherapy: the role of trichoscopy in monitoring therapeutic outcomes. G Ital Dermatol Venereol. 2016 Apr;151(2):212–5.
5. Kenicer KJA, Lakshmipathi T. Follicular mucinosis treated with PUVA. Br J Dermatol. 1982;107(Suppl 22):48–9.
6. Kluk J, Krassilnik N, McBride SR. Follicular mucinosis treated with topical 0.1% tacrolimus ointment. Clin Exp Dermatol. 2014 Mar;39(2):227–8.
7. Gorpelioglu C, Sarifakioglu E, Bayrak R. A case of follicular mucinoses treated successfully with pimecrolimus. Clin Exp Dermatol. 2009;34(1):86–7.
8. Al-Mutairi N. 308-nm excimer laser for the treatment of alopecia areata in children. Pediatr Dermatol. 2009;26(5):547–50.