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Pathological Co-localization of Lichen Planopilaris and Folliculitis Decalvans with Clinical Manifestations of LPP only

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ABSTRACT

Cicatricial alopecias are categorized based on their immune cell infiltration with Lichenplanopilaris (LPP) predominantly showing lymphocytic infiltration and folliculitis decalvans (FD) presenting with neutrophilic infiltration. Case series have been reported in previous literature introducing a concomitant or sequential occurrence of both diseases known as folliculitis decalvans lichen planopilaris spectrum (FDLPPS). In all these studies, concomitant cases presented with both clinical and histopathological features of FD and LPP, while sequential cases showed a precedence of FD with over time transition to LPP both clinically and histologically. This raised a discussion over the importance of the correct diagnosis and treatment in cicatricial alopecias, particularly in cases who's clinical and trichoscopic signs can't be fully attributed to one disease only, or cases of FD that prove unresponsive to treatment over time. We aim to present a rare case in this spectrum, that doesn't fall into any of the categories reported previously. Our patient manifested clinical signs of LPP only while showing histopathological features of LPP and FD simultaneously. It seems that this case is showing a precedence of clinical LPP over FD unlike other literature, while presenting with a concomitant occurrence of histopathological signs. Based on these findings, it was decided by the authors to treat the patient for both types of cicatricial alopecias simultaneously.

Keywords

Cicatricial alopecia, Folliculitis decalvans, Lichen planopilaris, Neutrophilic infiltration, Lymphocytic infiltration.

Introduction

Primary cicatricial alopecias (PCA) are primarily categorized into two different groups based on their cellular infiltration upon histopathological evaluation: Lichen planopilaris (LPP) and Folliculitis Decalvans (FD) presenting with lymphocytic and neutrophilic infiltration of the hair follicles respectively [1]. However, in clinical settings we often encounter cases of scarring alopecias with presentations that cannot be fully attributed to one type of PCA only or have been primarily diagnosed as either one of the two PCAs typically FD, but prove unresponsive to treatment. Thus, the term FDLPPPS also known as the folliculitis decalvans/lichen planopilaris phenotypic spectrum was introduced to define a third category of cicatricial alopecias in which neutrophilic and lymphocytic infiltration occur either concomitantly which typically involves clinical and pathological criteria of both diseases such as pustules, crusts and tufted hair follicles (FD) as well as perifollicular erythema and scaling, or occurring sequentially presenting with manifestations of one disease at first and transitioning to features of the other over time [2,3]. The latter is usually seen as FD preceding LPP [4,5]. In this study, we aim to present a case of FDLPPS presenting with clinical and trichoscopic features of LPP only, but with a concomitant neutrophilic and lymphocytic infiltration of hair follicles upon pathological evaluation, which indicates the consideration of simultaneous anti-inflammatory and anti-microbial treatment in order to achieve the desired clinical response.

Case Report

We report the case of a 35-year-old female with a negative past medical history who visited our office with the chief complaint of gradual hair loss over the past several years as well as a burning sensation associated with crusting and occasional pain and itching of the scalp. She had no history of previous medication use for her condition. Upon inspection, multiple patches of hair loss along with a general thinning of hair could be seen. An overall fibrotic appearance of the scalp was also noticed. Trichoscopic examination revealed anisotrichosis and focal atrichia as well as more specific signs such as honeycomb pigmentation, white dot, pig tails, fibrotic bands and arborizing blood vessels accompanied by perifollicular scaling and erythema, indicating LPP. Biopsy of the scalp was performed for a definitive diagnosis. Two samples, horizontal and vertical, were taken and sent for pathologic evaluation. Microscopic examination revealed hyperorthokeratosis, focal hypergranulosis and mild acanthosis in the epidermis. Moderate perifollicular and perivascular infiltration of the lymphocytes along with scarring at the isthmus level of hair follicles were also noted, which shows that the disease is at a moderate level of activity. Additionally, neutrophilic infiltration was seen at the infundibulum and isthmus of the follicles, suggestive of Folliculitis decalvans (FD). Bacterial culture was positive for S. Aureus. The pathology report concluded the diagnosis of Lichenplanopilaris (LPP) associated with deep neutrophilic folliculitis/perifolliculitis, even though the patient reported no symptom indicating folliculitis/perifolliculitis and our examination for the latter was negative. Due to the patient's reluctance to undergo treatment with oral medications such as corticosteroids, therapy was based on a twice daily application of topical combination solutions with 5% minoxidil, clobetasol, bimatoprost, zinc sulfate, dimethicone and caffeine. 1% clindamycin solution was added to the topical combination therapy to target neutrophilic involvement of the follicular and perifollicular area. Oral antibiotic therapy with Minocycline 50mg twice daily for 30 days was initiated to prevent any signs or symptoms of folliculitis decalvans from manifesting, seeing as the neutrophilic involvement of the follicles was reported to be deep. Two intramuscular injections of triamcinolone were also administered to systematically combat the inflammation of the scalp. Follow up was done after 2 months, in which the patients reported complete relief of the itching and burning of the scalp along with a significant decrease in active hair loss.

Discussion

Scarring or cicatricial alopecias are primarily divided into two main diseases known as lichen planopilaris (LPP) and folliculitis decalvans (FD) [6]. These two are distinguished by their immune cell infiltrates in the perifollicular areas with the former showing lymphocytic infiltrates and the latter showing neutrophilic infiltrates [7-9]. Although clinical and trichoscopic features can sometimes be mutual between the two, there are manifestations that are primarily attributed to either of the two. Lichen planopilaris is commonly suspected by perifollicular erythema, hyperkeratosis and scaling while folliculitis decalvans presents with pustules, yellow crusts and polytrichia. The folliculitis decalvans lichen planopilaris spectrum (FDLPPS) was introduced as part of an effort to effectively diagnose and treat cases with combined or mixed features [6]. This is either defined as a concomitant occurrence, where patients manifest clinical and histological criteria of both diseases at once, or as sequential occurrence also known as a biphasic occurrence. This is due to the fact that in such cases, folliculitis decalvans usually precedes lichen planopilaris and is hypothesized to be due to the dysbiosis causes by FD, that patients develop features of LPP over time [10-12]. Treatment is usually based on primary presentation. Patients who present with mixed features or in our case only show an overlap in pathologic evaluation, should be treated for both types of alopecias [1]. However, cases that are initially diagnosed with FD, don't respond fully to treatment in later stages of the therapy course due to lichenoid changes developing over time. In these cases, topical corticosteroids as a first line treatment for LPP, seem to be the best option after definite diagnosis of FDLPPS [2,4]. Previous literature have reported multiple occurrences of FDLPPS. Yip et al. presented a series of 13 cases, 6 of which were initially diagnosed with FD and showed LPP like changes over time in a biphasic manner while the other 7 cases present with clinical and in some cases histopathological features of FD and LPP concomitantly [2]. Egger et al. reported a 7-patient case series of FDLPPS with concomitant clinical and histologic findings of FD and LPP [1]. Ramos et al. have reported the concomitant manifestation of FD and LPP both clinically and histopathologically in the first two pediatric cases [3]. Melian-Olivera et al. studied a larger group of 31 patients diagnosed with FDLPPS and have stated that most of the cases were the result of a biphasic nature of FD progressing to LPP over time after multiple periods of antimicrobial treatment [5]. Thus, concomitant features of FD and LPP seem to occur both in clinical and histological aspects and sequential occurrence is typically seen as FD preceding LPP [7]. However, our case of FDLPPS presents concomitant histologic findings of FD and LPP while clinically, LPP seems to be preceding FD.

Conclusion

According to our investigations, the simultaneous presence of histopathological criteria of lichen planopilaris and folliculitis decalvans while clinically manifesting only one disease, has been rarely reported in previous literature. It is a very uncommon case, given the fact that the hair follicles are targeted by both lymphocytic and neutrophilic infiltration, however presenting as one disease clinically, with LPP being the preceding clinical presentation unlike previous reports. This raises the importance of future studies, evaluating the possibility of LPP preceding FD in sequential cases of FDLPPS and potential pathways to explain this.

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