Facial lichen planopilaris: a novel treatment using oral isotretinoin: a case series study

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Abstract

This case series study was performed on 19 patients with facial papule The patients were treated with oral isotretinoin 20 mg for 6 months. Oral isotertinoin was an effective and safe treatment in patients with facial LPP. There were some patients with only facial involvement without FFA or scalp LPP

Case series

Title:

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Farahnaz Fatemi: Case presentation, data collection,

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Clinical key message:

Despite the little information about the facial papules due to LPP, we have many cases with facial skin roughness in which histological study have showed LPP. Additionally, in those patients treating for FFA or scalp LPP there were no improvement in facial papules.

Keywords: lichen planopilaris, frontal fibrosing alopecia, facial papules, isotertinoin,

Introduction

Lichen planopilaris (LPP) is the most common scaring alopecia characterized by lymphocytic infiltration around hair follicles. Although it is considered an autoimmune disease, its exact mechanism of pathogenesis is still unknown (1). Frontal fibrosing alopecia (FFA) is a distinctive form of primary lymphocytic cicatricial alopecia, considering as a variant of LPP (2). FFA has markedly increased over the last years and considered "a growing epidemic" disease (3, 4). It mostly affects postmenopausal women, but it is also described in premenopausal women and men (3). Association with autoimmune diseases has also been reported (e.g. hypothyroidism) (5,6). Eventually, the number of cases with FFA has increased in recent years as a result of rising incidence (7). It has been considered as a variant of LPP that involves scalp hairs in frontotemporal hairlines, eyebrows and eyelashes. Involvement of facial vellus hairs presented as skin-colored follicular papules due to lichenoid perifollicular inflammation was first described by Donati et al. in 2011 (8). Beside it may be accompanied with body vellus hair involvement suggests that FFA has more pathological expansion (9-11). Facial vellus hair involvement is reported as a clinical feature of FFA in the majority of studies, although there are some rare reports of facial LPP in the absence of scalp disease (12).

Herein we present 14 patients with facial papules who have scalp involvement (FFA or LPP) and 5 patients with isolated facial LPP in the absence of other sites of disease activity and evaluate response to treatment with isotretinoine.

Cases' presentation

This case series study was performed on 19 patients with facial papule referred to Alzahra hospital and clinics affiliated with Isfahan University of Medical Sciences in Isfahan-Iran during 2018-2019. Patients who met the inclusion criteria were those with clinical feature of facial papules whose diagnosis was proved histopathologically. To quantify pre-treatment and post-treatment response to isotretinoin, we use Global Improvement Scale Assessments (GISA)

Nineteen patients were diagnosed with facial LPP enrolled to the study. Except for 2 males all patients in this series were females (41.17% postmenopausal and 58.82% premenopausal). The average patients age was from 32 to 68 years with mean age ± SD of 49.36 ± 11.61. FFA was found in 9 (52.9%) patients who were all females, whereas classic form of LPP was evident in 5 (29.4%) patients (2 males and 3 females). Five (29.4%) patients who were all females presented only with facial LPP without other sites involvement.

Concerning the presenting signs and symptoms, 9 (47.36%) patients had been referred with chief complaints facial skin roughness. Interestingly some of these patients were misdiagnosed and undergo laser resurfacing (2 cases) and needle radiofrequency (1 case) for their lesions. Following these procedures exacerbation of facial lesions, suggesting Koebner's phenomenon were observed. In 10 out of 19 cases, facial papule was recognized following scalp involvement.

Two male and three female patients with LPP of scalp were undergone treatment with systemic drugs (cyclosporine, hydroxychloroquine and or MTX, prednisolone, and phototherapy) for LPP of scalp, but in none of them facial papules respond to these treatments.

We observed different clinical patterns of facial papules according to age and sex of the patients. All patients who were older than 50 years (postmenopausal women) have subtle clinical expression and had previously been given a diagnosis of LPP or FFA. On the other hand facial lesions were better observed in younger patient and many of them present with such lesions. Two male patients presented with severe papular eruption over the face. In both of them, physical examination revealed LPP of the scalp.

The shape of lesions and pattern of their distribution varied between males and females and in female patients; also There was difference between patients under 50 years of age and those over 50 years of age (Fig.1 and Table.1).

In 11 of 19 cases (64.7%) eyebrows eyebrows were affected – partially in 9 patients (47.36%) and totally in 2 patients (10.52%) – whereas eyelash loss were present in 2 patients (10.52%).

The skin biopsy of facial papules showed lymphocytic infiltration around the vellus hairs accompanied by vacuolar degeneration of basal epithelium of these hairs and replacement of vellus hairs by fine fibrous tracts.

The patients were provided with an adequate explanation about the project and informed consent was obtained. All patients referred to the laboratory for initial blood tests including CBC, BUN, Cr, LFT and beta-HCG for female patients, ., patients received 20 mg of isotretinoin orally daily for 6 months. During treatment course, follow-up appointment was arranged after 1, 2, 4 and 6 months of treatment. Comparison of the change in lesions and the 4-point grading scale score (table. . . .) from base line was performed, based on photography.

After 6 months of treatment, before and after photographs were scored by a blind dermatologist. Response to treatment was dramatic in 2 males and significantly varied with female patients and in a way; the lesions were significantly reduced after 6 month of treatment. 10 patients (58.8%) were satisfied with the treatment and 3 cases (17%) had satisfactory results. The papular lesions were clearly reduced, and the skin was smoother especially in male patients (Fig.2), but this treatment did not affect the FFA and LPP of scalp.

Improvement with oral isotretinoin was apparent in all patients within 6 months. Many papular lesions disappeared rapidly and remained smooth skin. At the last visit, 63.15% of the patients stated that they were satisfied with the results of the of the treatment. Interestingly, scalp disease not affected by such treatment.

The scoring system was used to evaluate the treatment results by a dermatologist was as: 0 = no response, 1 = mild to moderate response, 2 = good response and 4 = a very good response.

The response to treatment in patients was evaluated by a blinded dermatologist and according to her opinion, 42.1% of patients had a good response and 26.3% had a very good response (Table.2).

Discussion and conclusion

LPP is a scaring alopecia presents as patchy or diffuse hair loss perifollicular erythema, perifollicular scaling and follicular keratosis. It can be subdivided into 3 variants, including classic LPP FFA and Graham-Little-Piccardi-Lasseur syndrome (1).

FFA was first described in 1994 by Kossard as a progressive loss of frontotemporal hairlines in postmenopausal women (13). Although premenopausal women or even men might be affected, We found a high rate of premenopausal women affected by FFA (36.8%). In our series, we had 2 men (10.5%) that they presented more severe facial papules than women. Recently, involvement of facial vellus hairs presenting as peculiar facial papules have been reported in association with FFA. On the other hand the disease might extend beyond the frontotemporal hair line and affect peripheral body hair and lead to loss of eyebrows and eyelashes. Thus, FFA has been recognized as a generalized than localized process.

FFA may be associated with clinical or serologic evidence of autoimmunity e.g. hypothyroidism. In concordance with previous reports, we found a high prevalence of hypothyroidism. Among our patients there were 3 cases (15.7%) of hypothyroidism and 3 cases (15.7%) of vitamin D deficiency.

Facial vellus hair involvement present as noninflammatory facial papules has been described in association with FFA (8). The clinical picture includes follicular micropapules randomly distributed over the facial skin, readily more visible over temporal and cheek regions. At the scalp biopsy, an inflammatory lymphocytic infiltrate around the upper portion of hair follicle can been observed with common findings of perifolliculr lamellar fibrosis and fibrosis of follicular tract (15), so that it is considered as a variant of LPP.

In our experience these skin changes are more prominent in male patients due to terminal nature of facial hair follicles. In the other hand, aged patients may have subtle clinical expression because of coexisting wrinkles and solar elastosis. Premenopausal women present with more evident papules even before scalp or eyebrows alopecia.

In parallel our observation, Maele et al. have described facial LPP in the absence of FFA in premenopausal women (12). In current study we found facial LPP in 9 women (47.3% of our case) in the absence of scalp involvement whom 7 cases (41.17%) were premenopausal. Because of these findings this question comes to mind that whether these facial papules are in the clinical spectrum of FFA or a variant of LPP that can occur as a single presentation without involvement of other body sites. More studies and longer follow up studies are need to recognize the nature of these lesions.

In some studies, histological evaluation of these papules revealed elastic fiber involvement accompanied with preserved large sebaceous glands (16).

Retinoids including isotretinoin are known to have several actions include anti-inflammatory effects and induction of apoptosis in sebaceous glands. (17). There are some reports of successful treatment of FFA with topical retinoids (18). Also there is a report of successful treatment of facial papules in FFA with oral isotretinoin (16). In addition, we have never observed in our practice FFA or LPP patients using other treatments (for example hydroxychloroquine, cyclosporine, methotrexate and prednisolone) have improved their facial lesions.

These observations, prompted us to conduct this study to evaluate the therapeutic effects of oral isotretinoin as a first-line treatment of facial LPP. The results were promising, showed the facial papules dramatically disappeared (42.1%good and 26.3% very good response). It seems valuable to consider the use of oral isotretinoin for treatment of facial papules with LPP nature. But more studies with greater number patients are necessary to better evaluate the efficacy of oral isotretinoin optimum dosage and treatment duration and comparison between topical and systemic retinoids and long term fallow up of the patients and determining the incidence of recurrence after stopping the treatment.. On the other hand, isotretinoin-induced sebaceous gland atrophy is not permanent, so long-term follow-up is necessary to confirm our observations

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