A Rare Case of Lichen Planus Follicularis Tumidus Involving Bilateral Retroauricular Areas

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Abstract
Lichen planus follicularis tumidus (LPFT) is an extremely rare variant of lichen planus characterized by white to yellow milia-like cysts and comedones on a violaceous to hyperpigmented plaque most commonly involving retroauricular area. Clinically, it resembles milia en plaque. It is usually asymptomatic, more common in middle-aged females. Histopathologically, it has features of lichen planopilaris along with follicular cysts in dermis surrounded by lichenoid infiltrate. We are reporting a case of LPFT in a 62-year-old male patient involving bilateral retroauricular areas due to the rarity of this condition.

Key Words: Lichen planopilaris, lichen planus follicularis tumidus, milia en plaque

Introduction
Lichen planus follicularis tumidus (LPFT) is an extremely rare variant of lichen planus. In 1977, Belaich et al. first described three patients with a new variant of lichen planus with retroauricular involvement and called LPFT. It is clinically characterized by white to yellow milia-like cysts and comedones on a violaceous to hyperpigmented plaque. The most common presentation is unilateral plaque in retroauricular area but rarely is bilateral. It is usually asymptomatic but may be mildly pruritic. Histopathologically, LPFT is similar to lichen planopilaris and additionally is characterized by follicles and cysts in dermis surrounded by lymphohistiocytic infiltrate. Here, we report a case of LPFT involving bilateral retroauricular areas.

Case Report
A 62-year-old male patient presented to our outpatient department with asymptomatic violaceous lesion in the right retroauricular area for 6 months. He noticed similar smaller size lesion on the left retroauricular area for 1 month. Physical examination revealed hyperpigmented tumid plaque of size 1–4 cm studded with whitish to yellowish milia-like cysts and open comedones. Examination of hair, nail, and oral mucosa revealed no abnormality.

The patient denied any history of trauma, bullous lesion, use of any topical treatment or hearing aid. The patient was a known case of hypertension and diabetes since 10 years and was on treatment. Based on the clinical examination, the differential diagnosis of milia en plaque, LPFT, and nevus comedonicus was considered. Histopathological examination of biopsy taken from retroauricular lesion revealed epidermis showing focal hypergranulosis and basal cell layer vacuolization, dense infiltration with lymphocytes and melanophages in dermis. Moreover, at places, epidermis showed follicular plugging, dermis showed keratin-filled cysts and dense lymphocytic infiltration around cystic spaces. Routine laboratory investigations were within normal limits. Serology for hepatitis B and C virus was negative, and thyroid function test was within normal limit. Based on the clinical and histopathological examination, we reached a final diagnosis of lichen planus follicularis tumidus involving bilateral retroauricular areas. We put him on oral prednisolone 0.5 mg/kg body weight with slow tapering. The patient showed mild improvement after 4 weeks of treatment in the form of decrease in tumidity of lesion.

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Discussion

Lichen planus is an inflammatory disorder of unknown etiology with a broad spectrum of cutaneous and oral involvement. Belaich et al. described the first case of LPFT as a very rare variant of lichen planus in 1977 involving retroauricular area. A case of bilateral retroauricular LPFT associated with oral lichen planus was described by Grupper et al. Baptista et al., in 1980 described a case of LPFT involving the cheek. Jiménez-Gallo et al. in 2013 described most severe form of LPFT involving face extensively. LPFT is more common in middle-aged females and clinically manifest as tumid violaceous plaques associated with multiple cysts and comedones. The most common location is retroauricular area, others sites being bilateral retroauricular area, cheeks, neck, ears, nose, and scalp. Our patient had tumid hyperpigmented plaque studded with multiple milia-like cysts and comedones, in the bilateral retroauricular areas.

LPFT has been reported in association with diseases, such as hepatitis B, hepatitis C, and autoimmune thyroiditis. It may be associated with other forms of lichen planus as well. In our case, no such association was noticed. Histopathologically, LPFT has been described as a variant of lichen planopilaris. In our case, histopathological examination revealed lichenoid infiltrate around follicles and cysts in dermis along with classical findings of lichen planus. Clinically, considering the classical location and morphology, the closest differential diagnosis kept was milia en plaque. Milia en plaque has milia on an erythematous base; comedones are usually absent. Whereas in our case, retroauricular plaque was studded with milia-like cysts and multiple comedones. Furthermore, on histopathology, milia en plaque cysts are not associated with lichenoid reaction, which was a consistent finding in our case. Nevus comedonicus consists of closely arranged dilated follicular openings with a keratinous plug with a...
predilection for face and neck area. It may present at birth or later in life approximately at the age of 10 years. LPFT is a chronic relapsing disease, and it is very resistant to treatment.[2] Although treatment in this disorder is disappointing, topical and systemic steroids, topical tretinoin, isotretinoin,[3] and excision have been tried. We are reporting this case because of its rarity. In addition, to the best of our knowledge, very few cases of LPFT are reported in literature in male patients.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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