

# Cutaneous Lymphoid Hyperplasia Presenting as Bilateral Post-Auricular Swellings: A Case Report

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**Abstract:** *Objectives:* To report a case of *Lymphocytoma Cutis* in MGM Medical College & Hospital, Aurangabad. *Patients:* 24 yrs. old male patient admitted in surgery ward *Methods:* The information was collected through Clinical examination, Ultrasonography, CT scan, MRI & Histopathological study of the specimen. *Results:* The outcome was *Lymphocytoma Cutis*. *Conclusions:* This case report gives information about the unusual presentation of *Lymphocytoma Cutis*.

**Keywords:** Lymphocytoma Cutis, Cutaneous lymphoid hyperplasia, bilateral post auricular region, Phenytoin, Spiegler–Fendt sarcoid, Lyme disease.

## 1. Introduction

Lymphocytoma cutis or Cutaneous lymphoid hyperplasia refers to a group of benign cutaneous disorders due to an inflammatory response to known or unknown stimuli that results in a lymphomatous-appearing but benign accumulation of inflammatory cells.<sup>(1)</sup>

The patient typically presents with macules or papules over the face with or without any precipitating cause. It is diagnosed by biopsy of the lesion. It usually regresses by removing the offending agent or at times intralesional corticosteroid injections or radiotherapy is used.

## 2. Review of Literature

The term cutaneous lymphoid hyperplasia was coined by Caro and Helwig in 1969.<sup>(2)</sup> It is also known as borreliolymphocytoma, Lymphadenosis benigna cutis, Pseudolymphoma, Pseudolymphoma of Spiegler and Fendt, Sarcoidosis of Spiegler and Fendt, Spiegler–Fendt lymphoid hyperplasia, and Spiegler–Fendt sarcoid.<sup>(3)</sup> Lymphocytoma cutis can be understood as a lymphoreticular and hyperplastic reaction occurring in the dermis.

In most cases there is no identifiable cause but common triggers include ear piercings, tattoos, phenytoin, vaccinations, infections from the herpes simplex virus and zoster, insect bites, and infection by the bacterium *Borrelia burgdorferi* (Lyme disease)

It usually affects Caucasians, females more commonly than males & young adults (<40yrs). Clinically, lesions present as papules and/or nodules that can be shiny. They are not scaly and can be skin-colored, erythematous, brownish or violaceous. Itching can occur. In the literature, it is common to find descriptions of lesions with softened consistency. In the case under discussion, the consistency of the lesions was similar to that of soft tissue tumours.

Various treatment modalities after removing the potential cause include Superpotent topical steroids, Intralesional corticosteroid injections, Cryotherapy, Photodynamic therapy with delta ALA, Topical tacrolimus, Hydroxychloroquine, Surgical excision, Radiotherapy<sup>(4)</sup>.

## 3. Patient Presentation

24 year old male patient came with the chief complaints of spherical swellings over bilateral post auricular region since 6 years. Both the swellings appeared simultaneously & progressively increased in size. They were not associated with any pain or discomfort.

The patient gives a history of a similar swelling behind the left ear 12 years ago which he got excised after 3 years. Three years after the excision both the post auricular swellings appeared & grew painlessly up to their present size when the patient finally came to MGM Hospital.

On examination there were 7x6x6 cms. right sided & 5x4x4 cms. left sided soft non tender spherical pediculated swellings arising from bilateral post auricular regions.

He had a history of itching all over the body since one year with no history of rash for which he took antihistaminics irregularly. The patient is a known case of seizure disorder for which he took Phenytoin Sodium for 6 years & is currently on sodium Valproate.

High frequency USG was s/o bilateral post auricular neoplastic lesion with bilateral cervical lymphadenopathy with necrotic areas in some lymph nodes.

CT Brain (P+C) was s/o ?lipoma/dermoid cyst/sebaceous cyst/nerve sheath tumour/parotid mass/lymphatic malformation. MRI was s/o dermoid cyst.

After taking the opinion of plastic surgeon & oncosurgeon the decision was taken for excision biopsy. On 9/2/2015 under general anaesthesia in-toto excision of both the swellings was done & sent for histopathological examination. Histopath report s/o Lymphocytoma Cutis/Cutaneous Lymphoid Hyperplasia.

## 4. Discussion

Although drug-induced pseudolymphoma remains a rare disorder, more than 100 individual cases have been reported in the literature worldwide.<sup>(5)</sup>

Before advances in immunophenotyping and gene rearrangement analysis, the diagnosis was based on

histologic findings and benign clinical behavior. Clinical follow up for 5 years following initial skin biopsy had been accepted as confirmation of the diagnosis<sup>(6)</sup>

On histologic examination, lesions of CLH may display multiple lymphoid follicles and dense superficial to deep infiltration of mostly mature lymphocytes. Lymphocytes often are admixed with histiocytes and occasional eosinophils and plasma cells. Germinal centers with tingible body macrophages often are apparent. Because these features also may be seen in lymphomas, determination of polyclonality by immunophenotyping with either polymerase chain reaction or other techniques is helpful in the evaluation of CLH.<sup>(7)</sup>

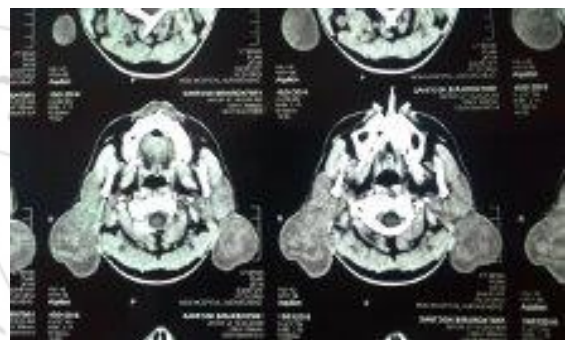
Lesions that interfere with function or are cosmetically undesirable may be surgically removed. No recurrence of excised lesions has been seen after withdrawal of the causative drugs.<sup>(5)</sup> In rare instances, malignant lymphoma has appeared following apparent resolution of phenytoin- and carbamazepine-induced pseudolymphomas. These cases have been termed pseudo-pseudolymphomas.<sup>(8)</sup>

## 5. Conclusion

In this unique case of lymphocytoma cutis the patient presented with bilateral post-auricular swellings. The unusual clinical presentation the uncertain impression even after thorough radiological imaging made this case a diagnostic dilemma. Detailed history taking & histopathological examination lead to the diagnosis of Lymphocytoma Cutis/Cutaneous Lymphoid Hyperplasia, possibly drug induced(Phenytoin). In spite of studies reporting the rare probability of recurrence of Lymphocytoma Cutis after surgical excision, especially after withdrawing the causative agent, the patient is advised follow up for early detection of recurrence or progression to Lymphoma.



Coronal Section of CT Brain(P+C)



Axial View of CT Brain (P+C)



Both specimens after surgical excision of swellings



Patient presenting with Bilateral Post-Auricular Swellings



Pre & post-op images of right post-auricular swelling



Pre & post-op images of left post-auricular swelling

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