

Lymphadenosis Benigna Cutis or Cutaneous Lymphoid Hyperplasia: A Rare Case Report

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ABSTRACT

Lymphadenosis benigna cutis or cutaneous lymphoid hyperplasia or lymphocytoma cutis or pseudolymphoma is classified as one of the inflammatory disorder in which accumulation of lymphocytes on skin resemble, clinically and histologically as, cutaneous lymphomas. It manifests as asymptomatic, indolent, nodular lesions of different sizes varying between 2 and 5 cm, usually solitary, mainly on exposed areas of the body like face and neck. The presence of polymorphous cell infiltrates comprising of T and B lymphocytes, plasma cells, eosinophils, histiocytes and dendritic cells along with lack of atypical lymphocytes after incisional biopsy support diagnosis of pseudolymphoma. We report a 25 year old man who presented with bilateral postauricular swellings. The diagnosis was made as lymphocytoma cutis histologically and confirmed by immunohistochemistry. We report this case due to its distinct presentation and rarity of site and unusual size.

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Introduction

Lymphocytoma cutis or lymphadenosis benigna cutis are localized or disseminated cutaneous proliferations of lymphocytes that are considered benign or reactive in nature. These are localized to head and neck region, and are seen as violaceous tumor ranging upto 4cm in diameter or as groupings pink small papules.^[1]The presence of polymorphous cell infiltrates comprising of T and B lymphocytes, plasma cells, eosinophils, histiocytes and dendritic cells along with lack of atypical lymphocytes after excisional biopsy supports diagnosis of pseudolymphoma.^[2] Final diagnosis is confirmed by Immunohistochemistry. Herein, we report a case of lymphadenosis benigna cutis due to its rarity and unusual size.

Case Report

25 year old male presented with bilateral post auricular swellings (fig. 1) since 12 years. Left sided swelling was seen from 12 years, it was excised six years back, since then it has recurred. Right sided swelling appeared since 6 years, gradually increased to present size. Onset was insidious and symptomless. The swelling was non tender and caused no discomfort for the patient. The patient came for cosmetic reasons. Left sided swelling measured 6x5x3 cm while right sided swelling measured 4x3x1 cm. The surface was smooth and no regional lymphadenopathy noted. There was no past history of drug intake, insect bite, tattooing, scabies, vaccination, acupuncture or photosensitivity. CT scan gave the differential diagnosis of

lipoma, dermoid cyst, sebaceous cyst, nerve sheath tumor, keloid or parotid tumor. Both the swellings were excised and send for pathological examination.

Gross: Received two skin covered masses of sizes 5x4x3cm and 4x3x2 cm. Cut section was grayish white. (fig 2)

Microscopy: Multiple sections studied showed skin lining, underneath dermis showed mild increase in sebaceous glands and hair follicles along with scant perivascular and periadnexal lymphoid infiltrate. The deeper dermis showed infiltration by lymphocytes and histiocytes. There were lymphoid follicles with germinal centres along with admixture of inflammatory cells. (fig 3 & 4) The case was diagnosed histopathologically as cutaneous lymphoid hyperplasia or lymphocytoma cutis. Immunohistochemistry showed positivity for T cell marker CD3 as well as B cell marker CD20, suggesting polyclonal nature, thereby differentiating from cutaneous lymphomas. (fig 5 & 6).

Discussion

The term *cutaneous lymphoid hyperplasia (CLH)* was coined by Caro and Helwig^[2] in 1969. The disease also has been called lymphadenosis benigna cutis, Spiegler-Fendt pseudolymphoma, lymphocytoma cutis, and cutaneous lymphoplasia.^[3] Cutaneous lymphadenoma is an uncommon benign epithelial neoplasm with a prominent lymphocytic infiltrate with pilosebaceous and an eccrine origin which clinically resembles CLH.^[4]



Fig.1: Image showing bilateral postauricular swellings.



Fig 2 Figures showing two skin covered swellings, on cut surface grayish white in colour

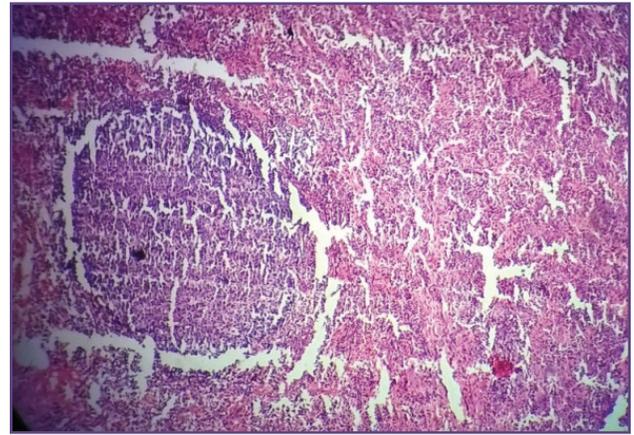


Fig 3 Section showing lymphoid follicles in dermis with inflammatory infiltrate in background (H & E, 10X)

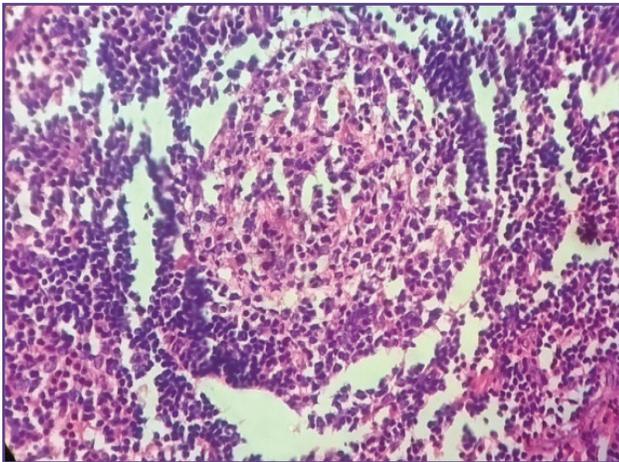


Fig. 4: Section showing lymphoid follicles with germinal centres & lymphocytes, histocytes (H&E-40X)

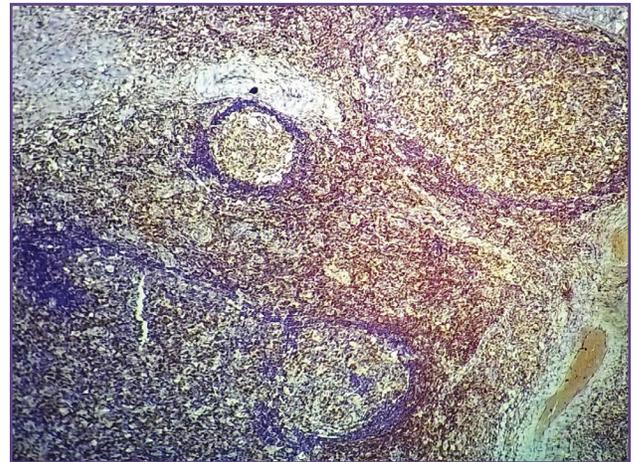


Fig. 5: CD3 positivity -10X (Tcell marker)

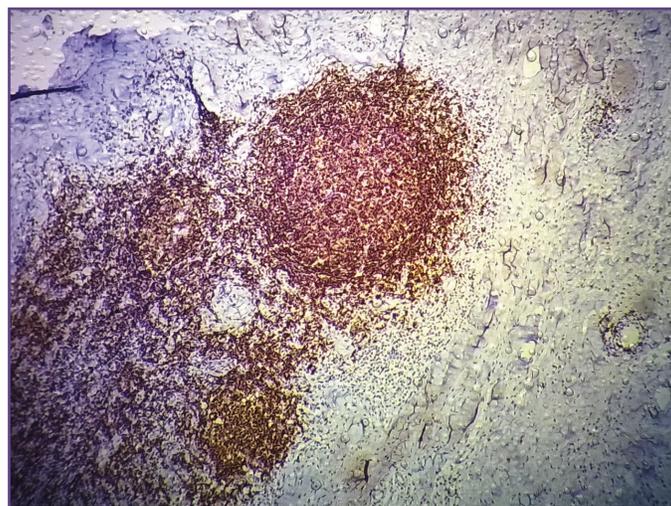


Fig. 6: CD20 positivity -10X (B cell marker)

Although the pathogenesis of CLH remains unknown and most cases are idiopathic, certain drugs and long-term antigenic stimulation are implicated in many cases.^[5] Other causes include arthropod bite, borrelia infection, as a postzoster phenomenon^[6]; in HIV infected individuals^[7] or after tattoos, exposure to gold and nickel, vaccinations, and taking drugs such as antihistaminics and phenytoin.^[8,9] The natural history of localized lesions is initial enlargement, sometimes followed by involution; disseminated lesions tend to be more persistent. Evolution to lymphoma over a period of years has been reported, but some of these cases may have been lymphomas from the beginning that were undetectable from the beginning with older diagnostic methods.^[1]

A female preponderance is seen. It is more common in patients under 40 years of age.^[10] Microscopy shows nodular or diffuse infiltrate more in superficial dermis (top heavy). Adnexal structures are spared, though sometimes show distortion or hyperplastic hair follicles with mild inflammation. The hallmark of pseudolymphomas is formation of lymphoid follicles containing mixed population of lymphoid cells. Flow cytometry shows polyclonal lymphocytic population. Differential diagnosis include: cutaneous lymphoma, sarcoidosis, erythematous lupus, angiolymphoid hyperplasia, lumina eruption and rosacea.^[11,12]

Histopathologically cutaneous, follicular, centre cell lymphoma is characterized by diffuse growth pattern with proliferation of neoplastic centrocytes and centroblasts.^[13] Follicle composition in follicular lymphoma is neoplastic, monomorphous and clonal, whereas in cutaneous lymphoid hyperplasia it is benign, polymorphous and polyclonal.^[14] Other IHC markers which can be applied to these cases can be CD19+, CD79a+, Ki 67 / MIB-1 antibody and BCL-6 expression.

Our patient had an unusually large lesion seen as bilateral post auricular swelling. The patient was suffering for more than 6 years without any malignant transformation.

In most of the cases, the lesion subsides spontaneously. Response to various treatment modalities are doubtful.^[15] In our case, the patient underwent excision of swellings. Further follow up of this patient unfortunately could not be maintained.

Conclusion

Differentiation of pseudolymphomas from lymphomas is difficult on histopathology alone. Immunohistochemistry is very helpful for final diagnosis. Pseudolymphomas can be cured in most of the cases, Proper follow up of the cases is needed to look for malignant transformation.

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Competing Interests

None declared

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