



## Bryan Murphy<sup>1\*</sup>, Helen Hunter<sup>1</sup> and Jennifer Somerville<sup>2</sup>

<sup>1</sup>Department of Dermatology, Belfast City Hospital, Lisburn Road, Belfast, Northern Ireland

<sup>2</sup>Department of Pathology, Belfast City Hospital, Lisburn Road, Belfast, Northern Ireland

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**\*Corresponding author:** Bryan Murphy, Department of Dermatology, Belfast City Hospital, Lisburn Road, Belfast, Northern Ireland, E-mail: bryan.murphy@hscni.net

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## Case Report

# A Case of Jessner's Lymphocytic Infiltrate and Cutaneous B-Cell Follicular Lymphoma: Coexistence or Transformation?

## Abstract

Jessner's lymphocytic infiltrate of the skin is a rare, benign cutaneous disorder. Difficulty arises in differentiating it from cutaneous lymphoma on histology, thus requiring immunophenotyping, however the literature lacks reports demonstrating malignant transformation or dual pathology.

We present a case of Jessner's lymphocytic infiltrate who later presented with areas of cutaneous B-cell follicular lymphoma.

## Abbreviations

JLI: Jessner's lymphocytic infiltrate

## Introduction

Jessner's lymphocytic infiltrate of the skin is a rare disorder of unknown aetiology consisting of erythematous, typically asymptomatic papules commonly found on the head and neck. It is characterised by a sleeve-like perivascular lymphocytic infiltrate within the dermis and is considered a benign entity showing some response to topical steroid and antimalarials. Discussion around its relationship with cutaneous lymphoma has stemmed from the difficulty in differentiating the two histologically. Our review of the literature however found only two cases of Jessner's where presence of malignant cells was suggested on retrospective analysis of specimens but they lacked immunostaining [1]. We present a case of Jessner's lymphocytic infiltrate who later presented with areas of cutaneous B-cell follicular lymphoma.

## Case Report

A 35 year old male presented with juicy, erythematous papules affecting both cheeks and pre-auricular areas which were exacerbated by sunlight and cleared completely within winter months. He had a history of lichen planus and nickel allergy but was otherwise well. He had no significant family history and was a smoker. The lesions were not typical of lichen planus so punch biopsy showed dermal perivascular lymphocytic infiltrate with peri-appendigeal lymphocyte

inflammation confirming Jessner's lymphocytic infiltrate of the skin (JLI). He was treated unsuccessfully with elocin but responded well to sunscreen and short courses of dermivate. Four years following diagnosis flares became more frequent so the patient commenced hydroxychloroquine 200mg twice daily.

Two months into treatment the patient incidentally highlighted a firm 15x10mm nodule in the left supraclavicular area which had slowly enlarged over a year. This area had never previously been affected by JLI to our knowledge in this patient but clinically the lesion was atypical for this. He felt otherwise well and his facial lesions were clearing. He denied night sweats and attributed 1/2 stone weight loss to dieting. Initial biopsy was non-diagnostic so complete excision ensued. This demonstrated cutaneous B-cell follicular lymphoma with nodules of predominantly B lymphocytes. Immunostaining of follicle centres was negative for BCL2, PCR confirmed a monoclonal population and t(14;18) chromosomal translocation was positive. Renal and liver function, full blood picture and bone profile were normal. Serum lactate dehydrogenase was 174 iu/L and C - reactive protein 3mg/L. CT PET scan showed no FDG uptake and bone marrow was trephine negative for lymphoma infiltration. Hydroxychloroquine was stopped and haematology multidisciplinary meeting concluded an involved excision peripheral margin so the patient received adjuvant radiotherapy.

Five months following lymphoma diagnosis the patient presented with a winter flare of JLI so hydroxychloroquine

was restarted at 200mg once daily. Clearance resulted but four months later he reported a new, raised 12x15mm raised, erythematous area on his right shoulder. He denied any B-symptoms. Excision confirmed cutaneous B-cell follicular lymphoma. Haematology multidisciplinary input advised no radiotherapy was indicated as it would not affect the risk of recurrence. Hydroxychloroquine was reintroduced six months later once daily. The patient has since been clear of JLI with infrequent flares controlled with short courses of dermatone and hydroxychloroquine.

## Discussion

Jessner's lymphocytic infiltration of the skin (JLI) is known to follow a chronic, benign, relapsing-remitting course with some response to topical steroid and antimalarials. Our review of the literature found no confirmed cases of JLI progressing to malignant lymphoma at different sites. Lange Wantzin et al [1], discuss two cases where sequential biopsies of relapsing lesions raised suspicion of malignancy but only on a retrospective analysis of JLI specimens. These cases signify the difficulty in differentiating benign and malignant diagnosis in JLI histology however they lack immunophenotyping. On the contrary however Ploysangam et al [2], state confusion with lymphoma is unlikely to occur with Jessner's because atypical lymphocytes and lymphoid follicles are not present.

Malignant transformation of other benign lymphocytic disorders such as cutaneous lymphoid hyperplasia and B-cell pseudolymphoma have been reported [3-6]. These reports propose: continued antigenic stimuli such as previous trauma or tattoos; more generalised or aggressive non-malignant disease; and identification of clonal B-cell populations in pseudolymphoma samples as markers for potential of malignant transformation. Considering our patient; he was otherwise well and had no obvious antigenic triggers. Following diagnosis of cutaneous B-cell follicular lymphoma; for which specimens demonstrated no plasma B-cells or sclerosed dermal stroma to suggest an Immunoglobulin G4-related pseudotumour [7]; the patient's original JLI specimens were re-examined and the typical histological findings of JLI were confirmed with no suspicion of lymphoma. There were no follicular structures and on immunostaining the infiltrate was composed mainly of T cells. There is no single criterion for differentiating benign or reactive B and T-cell conditions from lymphoma, however Ploysangam et al [2], suggest histological features that favour benign B-cell infiltrates over cutaneous B-cell lymphoma include: acanthosis; a top-heavy infiltrate favouring the papillary dermis; a mixed rather than monomorphous infiltrate; presence of germinal centres and tingible bodies; vascular proliferation; and preserved adnexal structures. Concerning benign over malignant T-cell infiltrates they propose: lack of epidermotropism; prominent spongiosis; benign appearing lymphocytes; and normal CD7 expression are favourable features.

Finally, hydroxychloroquine's role in the evolution of lymphoma might also be questioned as the second lymphoma nodule arose almost a year following first dose. On the contrary our patient states that the first lymphoma nodule was present for over ten months before starting treatment and considering he is now three years following excision and on continuous treatment without relapse would make hydroxychloroquine's culpability less likely. The literature also negates this theory; hydroxychloroquine has been identified as having anticancer properties [8,9] and potentially as a therapeutic agent in B-cell malignancies [10].

## Conclusion

We report a case of JLI and cutaneous B cell lymphoma and question a transformative process but cannot exclude the possibility of two concurrent, separate diseases. Given the diagnostic difficulty in differentiating benign from malignant, one might never be absolutely certain of the absence of clonal B-cells in initial specimens thus supportive immunophenotyping is required. This case signifies the importance of close follow-up of JLI patients and early biopsy of new, atypical lesions.

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