

Case Report

A case of CD20 positive peripheral T-cell lymphoma: not otherwise specified masquerading as botryomycosis

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ABSTRACT

Peripheral T-cell lymphoma (PTCL), a type of non-Hodgkin's lymphoma, is an uncommon malignancy frequently involving the skin either as primary or secondary manifestation of the disease. Lymphomas may occasionally masquerade as infectious diseases especially when it is not associated with significant lymphadenopathy. Botryomycosis is a chronic granulomatous suppurative bacterial infection, predominantly caused by *Staphylococcus aureus*, which is seen on trauma prone sites. Herein we present a case of a 33 years old male who presented to us with multiple rapidly growing friable masses on the chest wall of six weeks duration which clinically pointed towards botryomycosis. Lack of response to treatment with intravenous antibiotics and absence of grains and granuloma in the biopsy specimen made us further proceed for immune-histochemical evaluation which confirmed a rare type of CD20 positive PTCL. In conclusion, our case alerts physician to maintain a high index of suspicion for unusual entities in cases they encounter, cases with unusual presentation or atypical response to treatment.

Keywords: Non-Hodgkins lymphoma, CD20, Bacterial pseudomycosis, Immunohistochemistry, T-cell lymphoma

INTRODUCTION

Botryomycosis is a chronic granulomatous suppurative bacterial infection with varying skin lesions. They are usually located on areas exposed to trauma (head, arms, legs) and genital regions. The skin lesion is characterized by multiple fistulae draining purulent secretion and grains that are PAS positive. The most frequent etiological agent is *Staphylococcus aureus* (40%), followed by *Pseudomonas species* (20%).¹ Its treatment consists of antibiotic therapy and if required surgical debridement.

Peripheral T-cell lymphomas account for approximately 25% of all NHLs.² Immunohistochemistry is used in identifying the cell types and thereby classifying lymphomas.

CD20, is a pan-B cell marker, and its presence on benign and neoplastic lymphocytes is generally considered specific for B-lineage. However, few recent studies have indicated that peripheral T cell lymphomas can rarely express CD20 and hence can sometimes lead to misdiagnosis and failure of usual treatment regimens. Very few cases of CD20 positive T-cell lymphomas have been reported in the literature so far. There are various case reports on various mimickers of PTCL but none till date mimicking botryomycosis.

CASE REPORT

A 33 year old male presented with a six-week history of multiple dark, raised masses of varying sizes on the anterior chest wall, with fatigability, weight loss and anorexia of the same duration. He stated that this lesion

had been 'growing fast' in the last two weeks and also recalled a history of a fall six weeks earlier, following which he developed redness and swelling of the chest wall. In addition there was a history of multiple topical applications including many traditional medications and herbs. He also gave a history of multiple sand like particles and purulent discharge from the lesions and fever. Clinical examination showed multiple well defined hyper-pigmented friable masses on the anterior chest wall which showed multiple openings on the surface, purulent discharge from the sides of the lesions and surrounding skin showing tenderness, erythema and edema (Figure 1). A few enlarged, 1×1 cm tender, mobile and firm axillary lymph nodes were present.



Figure 1: Multiple well defined masses on anterior chest wall with purulent discharge and surrounding erythematous skin.

Investigations revealed Hb 7.9 g/dl, total count of 23000 cells/mm³, platelets of 90000 cells/mm³ and ESR 150 mm/hr. Chest X ray was normal. Gram stain from the discharge showed occasional gram negative cocci. No fungal elements or AFB were noted. Culture demonstrated staphylococci. Mantoux and fungal culture were negative. Routine investigations were not suggestive of a specific diagnosis. USG of chest wall showed multiple focal areas of subcutaneous fat thickening with increased echogenicity, fluid collection and mild increase in vascularity in colour Doppler with normal underlying lung parenchyma suggesting chest wall cellulitis. CT thorax demonstrated minimal bilateral pleural effusion.

A history of trauma, discharge of grain like material and clinical presentation of the lesion gave us a high clinical suspicion of botryomycosis. The other differentials considered were deep fungal infections (mycetoma, chromoblastomycosis, blastomycosis), cutaneous tuberculosis (nodular TBVC), cutaneous metastases and lymphoma.

The patient was treated with multiple intravenous antibiotics including amoxicillin, clavulanic acid,

metronidazole and amikacin but showed no signs of improvement and deteriorated drastically. Histopathology showed an ulcerated stratified squamous epithelium with pseudoepitheliomatous hyperplasia and edema. The dermis showed a dense inflammatory infiltrate composed of predominantly lymphocytes extending to the subcutaneous tissue and with focal areas of necrosis (Figure 2). No granulomas or fungal elements were noted.



Figure 2: H and E stain showing ulcerated epidermis, pseudo-epitheliomatous hyperplasia and dense infiltrate (40X magnification).

The clinical features, the doubtful histopathology with dense inflammation involving the deep dermis and the lack of response to antibiotics made us doubt the diagnosis and to request immune-histochemical (IHC) studies. The IHC report demonstrated diffuse CD3+, CD20 positive aggregates, CD8+/CD4-, Ki67-90-95% positive favouring peripheral T-cell lymphoma. The patient was referred to higher centres for further management. Over a period of two weeks, the patient developed breathlessness and other constitutional symptoms which could be due to tumour lysis syndrome.

DISCUSSION

Peripheral T-cell lymphoma is a diverse group of aggressive lymphomas that develop from mature-stage white blood cells called T-cells and natural killer (NK) cells. NHL affects two particular types of white blood cells i.e., B-cells and T-cells. PTCL has a poor prognosis with average duration of survival of 6 months.³

T-cell lymphomas comprise a heterogeneous group of neoplasms that are highly diverse, which shows positive expression of T-cell antigens (CD2, CD3, CD5, and CD7) and negative expression of B-cell antigens (CD19, CD20, CD79 α , and PAX5). While there are cases of B-cell lymphoma with significant expression of T-cell antigens the opposite is rarely found. Because of its rarity, little is known about this subtype of disease, its treatment and prognosis.

Peripheral T-cell non-Hodgkin lymphoma (PTCL)- not otherwise specified (NOS), is the most common PTCL subtype, accounting for at least 25% of PTCL.⁴ They are mature T-cell lymphomas that do not correspond to any of the defined T-cell entities, according to the WHO classification. It mostly affects adult patients, with a median age at presentation of 60 years and male predominance. Advanced stage at presentation is common, with almost two-thirds of patients presenting with an intermediate to high International prognostic index (IPI).

According to World Health Organization-European Organization for Research and Treatment of Cancer classification, PTCL-NOS are categorized under primary cutaneous lymphomas with aggressive clinical behaviour. Little attention has been given to the highly variable skin manifestations of the disease in the literature. In a study by Tolkachjov, out of 30 cases of PTCL-NOS, 47% had skin-only disease and 50% had concurrent skin and systemic disease at presentation.⁵ A case series by Wallett et al highlighted the cutaneous manifestations of PTCL-NOS which included urticarial papules and plaques, maculopapular rash, necrotic ulcers, nodules and indurated plaques.³

Histologically, diffuse, nodular or band like infiltrate of atypical lymphocytes occur in the dermis. Medium to large sized pleomorphic or immunoblast like T-cells are present in variable numbers and epidermotropism is generally mild or absent.⁶ Immunophenotype analysis is an important diagnostic method for PTCL-NOS. The most common immunophenotype was CD4+/CD8-.³ In a study done by Bekkenk et al, they found that a favorable outcome was noticed in cases with CD3+CD4+CD8-phenotype and with a clinical presentation of localized skin lesions.⁷ Our patient had CD3 diffusely positive, CD8+/CD4-. The Ki67 in our patient was 90-95%. Ki67 is a nuclear antigen expressed by dividing cells and a rate >80% heralds a worse prognosis.

CD20 has been used to identify the subtypes of malignant lymphoma as a B-cell marker. However, studies showed that peripheral T-cell lymphomas rarely express CD20 antigen with very few cases reported in the literature so far.⁸⁻¹⁰ CD20 positive T-cell lymphoma is a rare condition that is associated with the co-expressions of CD20 and T-cell markers, such as, CD3, CD5, or UCHL-1. The majority of cases of CD20 positive T cell lymphoma have been reported as immature peripheral T-cell lymphoma, not otherwise specified. The importance of this being a possibility of misdiagnosis and a high chance of failure of usual treatment regimens. Some studies have evaluated the possible role of rituximab in this variant of PTCL.

Here the patient had PTCL with chest wall cellulitis which could have led to the purulent discharge. It is possible that the patient mistook the friable mass for grains discharging from the lesion. History of trauma prior to onset of the lesions, discharge of grains with

purulent discharge, culture showing *Staphylococcus aureus* and absence of significant lymphadenopathy made us suspect botryomycosis. Immunohistochemical analysis proved this to be an incorrect diagnosis. He also had multiple factors suggesting a poor prognosis-younger age of onset, secondary bacterial infection, sudden increase in size, multiple lesions and immunophenotypically a high Ki67 rate.

As it is an aggressive tumour with poor prognosis, immediate therapy is warranted and hence a slight delay in diagnosis may do great harm.

CONCLUSION

A rare case of CD20 positive peripheral T-cell lymphoma, which can masquerade multiple other conditions and highlights the importance of maintaining a high index of suspicion in cases with atypical presentations and responses to treatment.

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