Case Report

An unusual, delayed, solitary manifestation of a penile lesion in chronic lymphocytic leukemia

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ABSTRACT

Chronic lymphocytic leukemia (CLL) is the most common type of leukemia in the adult western population. It is characterized by the proliferation of mature but dysfunctional lymphocytes, primarily $CD5^+$ B cells. It primarily affects the reticuloendothelial system in the majority of the cases, but can rarely manifest as extranodal and extramedullary lesions. One of the rare presentations is genitourinary cutaneous infiltration, and only a handful of cases of secondary metastases to the genitourinary skin, have been reported in the literature. The current report describes a patient with solitary lesion of CLL in the penis, manifesting almost two decades after the complete treatment of CLL.

INTRODUCTION

Chronic lymphocytic leukemia (CLL) is the most common type of leukemia affecting the adult population in the developed countries. It is characterized by the proliferation of mature but dysfunctional CD5+, CD19+, CD20+, CD23+, and CD10 – clonal lymphocytes. The disease primarily affects the peripheral blood, bone marrow, lymph nodes, and spleen in the majority of the cases. Extramedullary and extranodal involvement can rarely be seen in these patients at the time of diagnosis.[1] Leukemia cutis, an uncommon presentation of CLL, is characterized by any type of cutaneous infiltration by the leukemic cells, including the infiltration of the genitourinary skin.[2] Penile manifestation of CLL is rarely reported, and globally, only 30 such cases have been reported to date,[3] with the first being reported in 1953.[1] In this report, we describe a patient with solitary lesion of CLL in the penile skin, presenting several years after complete treatment and remission of CLL.

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CASE REPORT

A 63-year-old male patient presented with lower urinary tract symptoms and was diagnosed to have benign prostatic enlargement. He also complained of itching sensation on the penis and had a past history of CLL stage IIA non-Hodgkin's lymphoma, for which he had received treatment and was in remission for the last 17 years. On physical examination, a 1-cm subcutaneous lesion was palpable at the level of the coronal sulcus [Figure 1], and was treated as an abscess with topical and oral antibiotics, but showed no improvement.

After 4 weeks, the lesion was excised, and the histopathological examination showed angiofollicular lymphoid hyperplasia with eosinophilia and was positive for CD20, CD3, and Ki67, indicating the predominance of B cell component with heavy inflammation and abscess formation. Hence, the patient was referred to the hematology department and was started on acalabrutinib 100 mg twice a day. The patient's follow-up visits, both in the department of urology and hematology, were unremarkable. On the subsequent urology visit after 24 weeks, the patient

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complained of a painful swelling on the dorsal aspect of the coronal sulcus, which was excised. The histopathology and immune-histochemistry studies revealed an atypical lymphoproliferative lesion suggestive of small lymphocytic lymphoma without transformation [Figure 2], for which the hematologist's advised to continue acalabrutinib 100 mg twice daily.

At 32 weeks follow up, the patient had persistent lesion which was re-excised and the histopathology results demonstrated diffuse deep atypical lymphoproliferative infiltrates consistent with low-grade small cell lymphoma, which was strongly positive for CD20, CD5, and bcl-2, moderately positive for CD23, and weak to moderately positive for CD43 and bcl-6. Hematology review was requested and the patient was started and maintained on acalabrutinib and venetoclax with subsequent follow-ups demonstrating complete remission of the penile lesion, without an evidence of any acute symptoms or tumor lysis syndrome.

DISCUSSION

CLL is an indolent cancer diagnosed by the presence of persistently elevated (for >3 months) monoclonal B-lymphocytes ($\geq 5 \times 10^9$/L) in the peripheral blood and the expression of dysfunctional CD5+, CD10–, CD19+, CD20+, and CD23+ clonal lymphocytes on immuno-phenotyping. These patients can rarely present with clinical features of extramedullary or extranodal involvement.[1] Up to 15% of the cases with myeloid disorders and 4%–20% of the patients with CLL may present with cutaneous involvement.[2] Although the genitourinary system is an uncommon site of involvement in CLL, leukemic infiltration should be suspected in a previously diagnosed/treated case of CLL who presents with urological symptoms.[1]

In the most of the previously reported cases, the penis was involved by secondary metastasis rather than by a delayed primary lesion as in our case. Metastatic lesions commonly spread to the penis via the venous, arterial, or the lymphatic routes, as well as by direct extension or iatrogenic implantation.[4] Primary cancers metastasizing to the penis include prostate, bladder, colon, and kidneys, however, penile involvement by CLL is extremely rare.[5] The penile metastasis clinically presents as either penile nodules, skin lesions, or as malignant priapism, but presentation as a solitary penile lesion is rare.[6] Some investigators propose that retrograde lymphatic spread could explain the infiltration of the foreskin and superficial fascia.[7] Diagnosis can be made by a thorough physical examination followed by imaging studies including computed tomography and magnetic resonance imaging.[8] Currently, the diagnosis of metastatic or primary penile lesion depends on the pathological examination of the tissue specimen.[8]

As per the literature review, secondary metastasis was the most common cause of genitourinary involvement in the majority of the cases of CLL, contrary to the current case, who presented with a penile lesion almost two decades after the complete remission of CLL. This lead us to conclude that the penile lesion in our patient was a primary delayed penile CLL.

The treatment of choice for the majority of cases with secondary penile lymphoma is systemic chemotherapy, owing to the good functional as well as cosmetic results.[6] The index case also received acalabrutinib and venetoclax with good response and the penile lesion resolved completely. Radical surgery should be reserved for cases who do not respond to systemic therapy.[3,6] Similarly, partial or total glans resurfacing can be performed for noninvasive penile lesions, in patients with relapse after topical chemotherapy.[9] Other therapeutic options include laser ablation, photodynamic therapy, and Moh's micrographic surgery.[6]

Consent

Informed consent was taken from the patient, can be provided upon request.

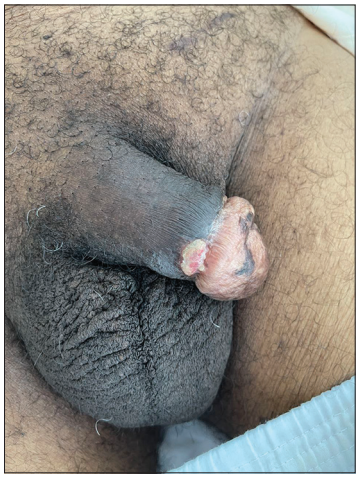

Figure 1: A 1 cm subcutaneous lesion noted at the level of the coronal sulcus

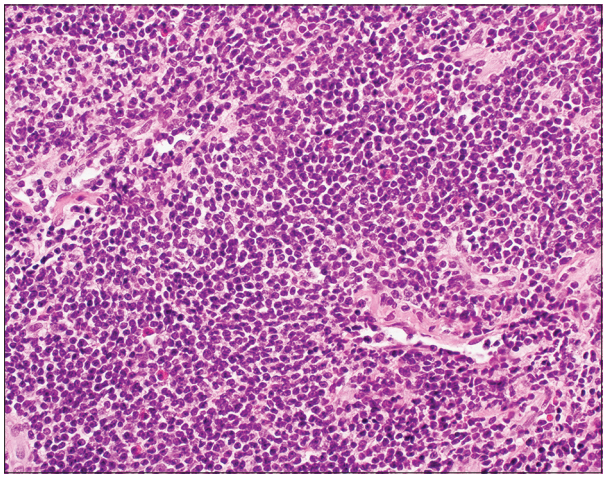

Figure 2: Atypical diffuse lymphoproliferative lesion

Declaration of patient consent

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

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