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Coexistence of Pyoderma Gangrenosum and Acute Lymphoblastic Leukemia

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Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Background: Pyoderma gangrenosum (PG) is a rare skin disease has been reported in association with acute myeloid leukemia, myeloproliferative disorders, myeloid metaplasia and myeloma is well known, but their association with lymphoid malignant tumors, is extremely rare. (PG) is a serious ulcerating skin disease characterized by the presence of the necrotic ulcer of the skin accompanied by pain.
Case Presentation: We report a rare case of a 25-year- male presented with recurrent fever and general weakness and episodes of typical skin lesion of bullous pyoderma gangrenosum since two months and now associated with pallor and bleeding tendency**.**
Conclusion: Complete remission of blood and bone marrow findings after first induction chemotherapy associated with early regression of all other manifestations.

Keywords: Pyoderma gangrenosum; acute lymphoblastic leukemia; chemotherapy.

1. INTRODUCTION

Pyoderma gangrenosum (PG) described first time in 1930 by Burn-Sting et al. as a rare sterile neutrophilic dermatosis [1]. The causes of PG are unknown in some cases or it may be associated with malignant or systemic diseases. Also there have been association between PG

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and myeloid metaplasia, polycythemia vera rubra and monoclonal gammopathy. We describe PG in association with acute lymphoblastic leukemia in a 25-year-old man, although its association with lymphoid malignancies is rare, who did not respond to extensive antibiotics but responded to steroid-based chemotherapy early and within the first week of induction chemotherapy [2]. PG was not previously reported in Yemen.

2. CASE REPORT

25 year old male presented with recurrent fever and general weakness and episodes of typical skin lesion of bullous pyoderma gangrenosum since two months and now associated with pallor. On physical examination, he was conscious, well oriented, and febrile with marked pallor and bleeding tendency (tarry black stool and red urine) and hepatosplenomegally and no lymphadenopathy. The examination of skin revealed the affected areas include left flank of abdomen and back, left leg with deep ulceration, and associated with virulent exudate at the base of ulceration which not respond to antibiotics and steroids. Cultures of the wounded ulcers showed normal skin microbial flora. He had white blood cell count of 31,000/mm^3, and platelet count of 63,000/mm^3 and a hemoglobin concentration of 5.6 g/dl, Bone marrow aspiration showed hypercellularity with reduction of all cell lines with presence of 95% lymphoblastic cell which was suggestive of ALL and confirmation of acute lymphoblastic leukemia associated with positive B-cell ALL by immunophenotyping expression of CD10 and CD 19.

Pyoderma gangrenosum skin lesions that had previously failed to respond to broad spectrum antibiotics and steroids showed signs of improvement within a week after chemotherapy. In view of the classic morphology of the lesion and patients who did not respond to antibiotics but completely responded to steroid chemotherapy, PG was diagnosed. At the end of first induction chemotherapy, he was in complete remission of her bone marrow revision.

3. DISCUSSION

PG considered one of the rare skin lesions that affects people of all ages. About 4% of the patients are children and infants [3] PG clinical variants include five types: ulcerative / classic, bullous, pustular, perioral, and nutritional [4]. Ulcerative colitis is the most common form, initially with a single small blister or multiple nodules. These lesions became larger and the skin over them began to die off. The typical lesion of a PG ulcer has multiple blind necrotic ulcers with margins, shellac-shaped hematomas, and jagged margins around the edges. The general manifestation of this case is typical ulcerative disease of PG.

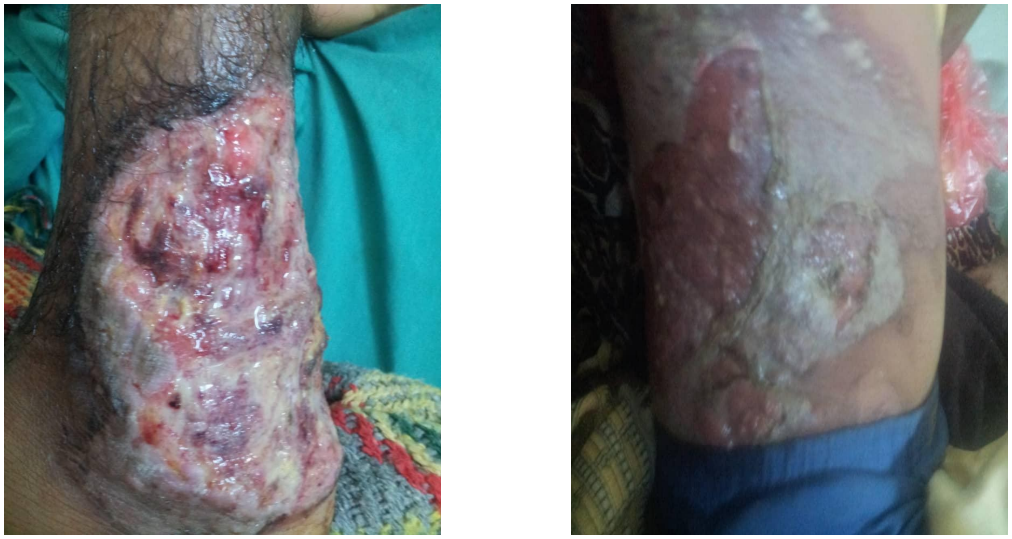

Fig. 1. Before treatment; deep ulceration of left leg and left flank of abdomen and back with virulent exudate at the ulcer base

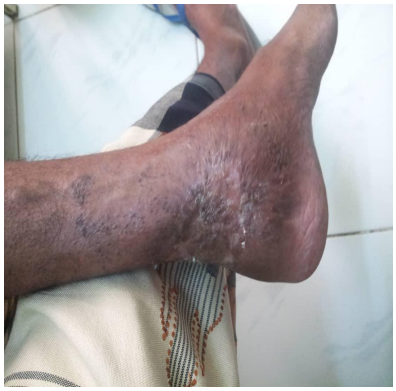
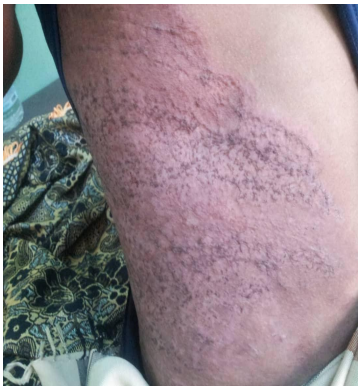

Fig. 2. After treatment; healing of deep ulceration of left leg and left flank of abdomen and back

The causes and pathogenesis are not well understood, but are believed to be due to an underlying immune regulatory disorder. Diagnosis is often delayed and only considered after other processes that can lead to skin ulceration have been ruled out and only supported by histopathology.

There are different ulcerative cutaneous lesions interfere with the differential diagnosis of PG which include malignant disease, insect bites, infectious diseases, arterial and venous insufficiency. Maldonado et al reported the association of acute leukemia with PG in 1968 [5]. Acute myeloid leukemia being the most common type of malignant diseases associated with PG [6] Acute lymphoblastic leukemia is rarely associated with PG but this case reported this rare relationship.

It is an important diagnostic basis for advanced clinical features and is usually only supported in histopathology. The typical changes of PG is massive neutrophilic infiltrates without vasculitis and granuloma formation [7]. PG is a skin lesion that is caused by neutropenia but can be a symptom of neutropenia, such as autoimmune neutropenia and type 1 leukocyte adhesion defect. A clinical criterion for PG has been suggested [8,9]. Our patient meets these criteria.

In adult the prevalence of lesions, often affecting the lower extremities, although PG in children is more common in the head and face [10]. Most patients with PG have multiple lesions, like our patient, although the exact etiology and pathogenesis are not understood. The association with leukemia may occur more frequently than previously thought, and leukemia monitoring should be maintained in atypical cases of PG to allow early diagnosis and appropriate treatment.

4. CONCLUSION

Acute lymphoblastic leukemia can be preceded by pyoderma gangrenosum as an early manifestation in rare cases. This case emphasizes the task of the PG diagnosis, and emphasizes the critical clinical feature to help diagnose, and the clinical effects of this state delay or misdiagnosis of this condition. The role of blood investigations and bone marrow aspirations are very helpful for early diagnosis of acute leukemia associated with pyoderma gangrenosum. Complete remission of bone marrow after first induction chemotherapy in the first 2 weeks associated with early improvement of skin lesions.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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