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(CASE REPORT)

Penile cyanosis revealing polycythemia vera: A case report

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Abstract

Polycythemia vera is a chronic myeloproliferative neoplasm characterized by excessive erythrocyte production, resulting in increased blood viscosity, impaired microcirculation, and a heightened risk of thrombotic events. It may present with a wide spectrum of cutaneous manifestations related to hyperviscosity and microvascular dysfunction, including erythromelalgia, livedo, pruritus, acral ischemia, and, more rarely, penile cyanosis.

We report the case of a 61-year-old male with no significant medical history, presenting with penile cyanosis associated with a penile blister, preceded by a febrile syndrome and acute urinary retention. Laboratory investigations incidentally revealed polycythemia (hemoglobin 18.5 g/dL), acute renal failure, and myocarditis on transthoracic echocardiography. The patient underwent therapeutic phlebotomy followed by curative anticoagulation with low-molecular-weight heparin and antiplatelet therapy, resulting in favorable clinical evolution, with regression of penile cyanosis and partial healing of post-blister erosions.

Keywords: Penile Cyanosis; Polycythemia Vera; Hyperviscosity; Cutaneous Manifestations; Rare

1. Introduction

Polycythemia Vera (PV) is an acquired myeloproliferative syndrome (MPS) characterized by an elevation of the absolute red blood cell mass due to uncontrolled production, often associated with the overproduction of leukocytes and platelets [1]. In this report, we describe a rare presentation of PV in which penile cyanosis was the initial clinical manifestation, leading to the incidental diagnosis of Polycythemia vera. Such genital ischemic presentations are exceptionally uncommon in the literature, highlighting the atypical and misleading nature of the initial clinical picture.

2. Case report

A 61-year-old male with no prior medical history presented to the urological emergency department with penile cyanosis and blistering of the shaft evolving over four days. The onset was preceded by chills, febrile sensations, and acute urinary retention, for which symptomatic treatment was administered without improvement.

Initial laboratory investigations revealed significant polycythemia (hemoglobin: 18.5 g/dL), acute renal failure, and echocardiographic evidence of myocarditis. The patient denied any medication intake before symptom onset, had no history of similar episodes, and did not report palmar-plantar erythema or warmth. The clinical course was accompanied by fever and asthenia.

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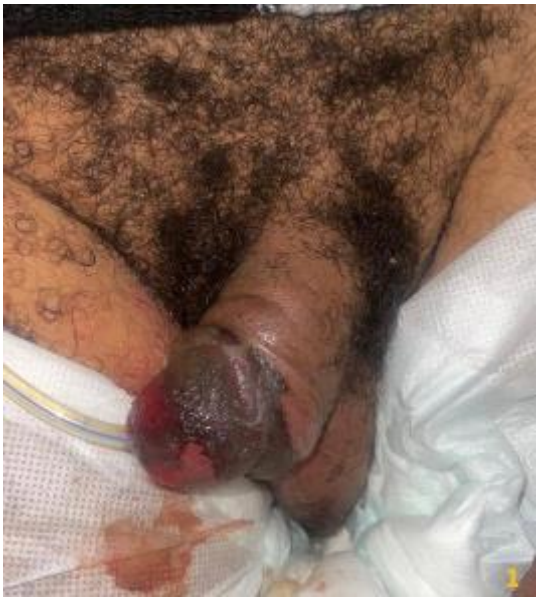

Figure 1 A well-demarcated cyanosis localized to the glans penis

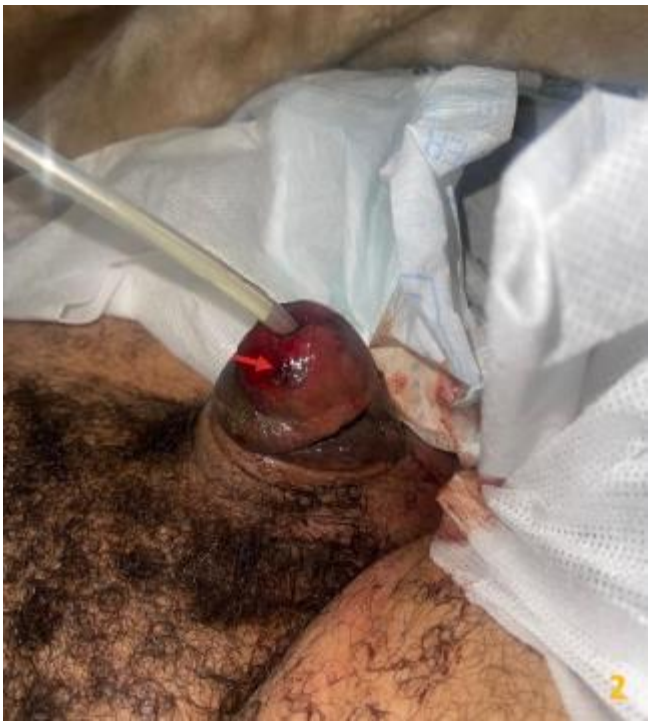

Figure 2 Presence of a well-defined erosion with a clean surface, partially covered by a haemorrhagic crust (red arrow), involving the glans in the peri-meatal region

Dermatological examination revealed a well-demarcated cyanosis localized to the glans penis (Figure 1), with overlying erosions following blister rupture (Figure 2). The erosions displayed clean surfaces with geographic borders, were hyperalgesic in some areas, but showed no necrosis or sensory deficits. There were no signs of palmar-plantar erythema or local heat.

The patient underwent urgent phlebotomy to reduce hematocrit levels, followed by cytoreductive therapy (AML protocol) combined with curative-dose low molecular weight heparin (Hibor) and antiplatelet therapy. Local wound

care included saline-soaked compresses, topical fusidic acid, and a healing cream applied twice daily, along with gauze packing.

The clinical course was favorable, with complete regression of penile cyanosis and partial healing of the post-blister erosions.

3. Discussion

PV can occur at any age, with a predominance between 50 and 70 years. Symptoms are often insidious at the beginning, including headaches, dizziness, vertigo, tinnitus, and visual disturbances [1].

PV may be complicated by arterial and venous thrombotic events, including cerebrovascular accidents, myocardial infarction, deep vein thrombosis, pulmonary embolism, and splanchnic vein thrombosis. Hemorrhagic manifestations may also occur. Advanced disease can evolve into post-polycythemia myelofibrosis, myelodysplastic syndrome, or acute leukemia [2].

Symptoms are mainly related to blood hyperviscosity, leading to impaired microcirculation due to increased circulating blood elements. Dermatologic manifestations of Polycythemia vera include progressive cutaneomucosal erythrosis, particularly involving the face, palms, and nail beds. Aquagenic pruritus is a common and characteristic symptom associated with the underlying myeloproliferative neoplasm. In our patient, penile cyanosis was observed [3,4].

Diabetes mellitus, vascular calcification in end-stage renal disease, thromboembolic disorders, hypercoagulable states secondary to neoplastic diseases, trauma, and infections represent the main etiologies of penile ischemia and distress. In our case, a thrombotic tendency secondary to Polycythemia vera was considered the most likely mechanism responsible for penile cyanosis [5].

The treatment of Polycythemia vera (PV) is based on correcting coagulation disorders, notably through phlebotomy to improve blood circulation by reducing hematocrit (Hct) to 45%. Hydroxyurea remains the standard treatment for elderly patients. In the absence of contraindications, low-dose aspirin reduces the risk of thrombosis. Treatment with interferon-alpha may be offered to pregnant women. Anagrelide can also be used to reduce platelet counts [6]. According to the 5th edition of the World Health Organization (WHO) Classification of Haematolymphoid Tumors (2022), Polycythemia vera is classified as a BCR ::ABL1-negative myeloproliferative neoplasm [7].

4. Conclusion

Polycythemia vera is a myelodysplastic syndrome characterized by a polymorphic clinical presentation with systemic involvement. Dermatological manifestations are mainly represented by erythromelalgia; however, atypical signs such as penile cyanosis, which was the only revealing symptom in our patient, can also occur.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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