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Submission date: 19-May-2023 08:57AM (UTC+0700)
Submission ID: 2096696047
File name: Case_report_penile_gangren.pdf (522.56K)
Word count: 1756
Character count: 9717

Bali Medical Journal (*Bali MedJ)* 2021, Volume 10, Number 1: 108-110
P-ISSN.2089-1180, E-ISSN: 2302-2914

Bali Medical Journal
Published by Bali Medical Journal

Penile gangrene as a priapism sequele due to Chronic Myeloid Leukemia (CML): the first report in Indonesia

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Received: 2020-12-09
Accepted: 2021-03-16
Published: 2021-04-01

ABSTRACT

Background: Priapism is defined as a prolonged erection of the penis lasting over 6 hours in the absence of sexual stimulation. Priapism is divided into two types, Low-Flow (ischemic priapism) and High-Flow (trauma). This case study aims to evaluate penile gangrene as a priapism sequele due to Chronic Myeloid Leukemia (CML) as the first report in Indonesia.
Case Presentation: A 45-years old male patient admitted at Muara Teweh hospital before referred to Ulin Hospital presented with Fournier gangrene with unconfirmed leukemia as comorbidities used in this case report. In his medical history, "Snake Maneuver Shunting" has been carried out in this patient as an indication for priapism lasting more than 24 hours.
Conclusion: Hyperleukocytosis is thought to be the cause of priapism in patients with leukemia. Leukostasis is the most common medical emergency seen on CML patients in blast crisis.

Keywords: Priapism, CML, Fournier Gangrene, Penile Gangrene.
Cite This Article: Yulizar, D.R., Maulani, E.P., Prasetya, H., Sutapa, H., Rahman, E.Y. 2021. Penile gangrene as a priapism sequele due to Chronic Myeloid Leukemia (CML): the first report in Indonesia. *Bali Medical Journal* 10(1): 108-110. DOI: 10.15562/bmj.v10i1.2125

INTRODUCTION

Priapism is defined as a prolonged erection of the penis lasting over 6 hours in the absence of sexual stimulation.[1] It is a potentially painful condition in which the erect penis does not return to its flaccid state, despite the absence of both physical and psychological stimulation.[1] It is a medical emergency and for its function to return, early treatment is essential. Priapism is divided into two types, Low-Flow (ischemic priapism) and High-Flow (trauma). Ischemic priapism is a surgical emergency that requires urgent intervention.[1,2]

Priapism etiology ranges from idiopathic, pharmacological, hematologic disorder (sickle cell disease, leukemias, penile metastases, etc.) or particular neurological disorder.[3] The sequele of priapism, if not treated appropriately, can either become permanent erectile dysfunction or, rarely, penile gangrene.[4] Penile gangrene associated with priapism has been reported in patients with sickle cell disease, urethral cancer, thrombotic thrombocytopenic purpura, bladder carcinoma.[3-5]

Based on those mentioned above, this case study aims to present a chronic myeloid leukemia patient presented with ischemic priapism, penile gangrene, and Fournier's gangrene.

CASE REPORT

A 45 years old male patient admitted at Muara Teweh hospital before referred to Ulin Hospital presented with Fournier gangrene with unconfirmed leukemia as comorbidities. In his medical history, "Snake Maneuver Shunting" has been carried out in this patient as an indication of priapism that lasts more than 24 hours. Two days after the treatment, Fournier gangrene occurred, and the patient was immediately referred to Ulin Hospital. The patient presented with a complaint of the darkened penis shaft and both sides of the scrotum accompanied by pain (Figure 1). A shunt connecting the right and left corpus cavernosum is attached to the patient's glans penis. Clinically, the patient showed signs of sepsis and based on a laboratory test, and his leukocytes are 241,500 mg/dL. A debridement-necrotomy that exposes scrotum to tunica vaginalis and penis shaft to tunica dartos has been done (Figure 2).

After degloving the penile shaft, the glans appeared to be blackened, and

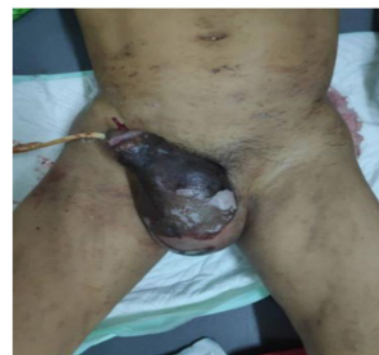

Figure 1. Anterior Aspect Clinical Picture in Emergency Room (ER)

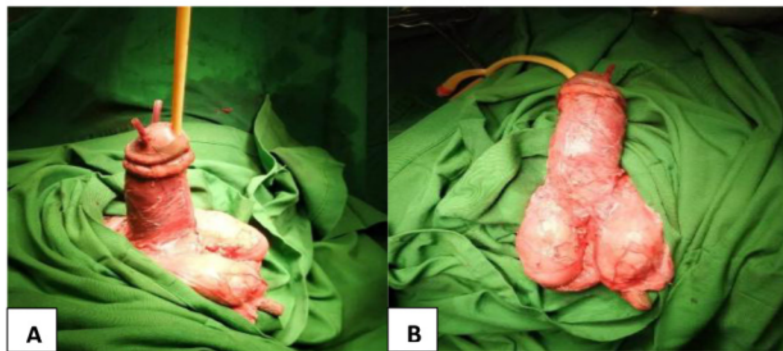

Figure 2. Clinical pictures first debridement on A) Lateral Side and B) Dorsal Side

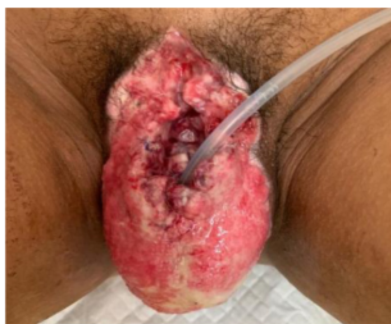

Figure 3. Clinical Picture After Partial Penectomy

the vitality of the penile tissue show a reddish picture, so we decided to keep observing him. In 3 days of evaluation, we obtained a picture of the blackened penis shaft and did a USG examination. The USG results show splenomegaly. BCR-ABL test showed "Detected" as a result; therefore, we confirmed the diagnosis of CML. The internal medicine department was consulted and decided to give hydroxyurea therapy for 10 days. As for surgical treatment, we chose to do partial penectomy, but when the patient was given the information to do a perineostomy, he refused and wanted to maintain the upper urinary area (Figure 3). He was discharged 9 days later after penectomy in a stable condition and a skin flap had been planned.

DISCUSSION

Priapism is defined as involuntary, painful, and prolonged erection of the penis lasting more than 6 hours unrelated to sexual stimulation and unrelieved by ejaculation.[1,2] Ischemic priapism accounts for >95% of cases of priapism.[5] Underlying hematological causes such as sickle cell disease, thalassemia, leukemia, G6PD deficiency, fat emboli associated with hyperalimentation, and rarely multiple myeloma need to be evaluated in cases of ischemic priapism.[6] Penile gangrene is a rare sequel of priapism. In our case, priapism with penile gangrene was the initial presentation.[5-7]

Prompt recognition and appropriate treatment of an episode of priapism in patients with CML are critical. This is the result of prolonged or repeated episodes of priapism can result from ischemia and fibrosis of the corpus cavernosa of the penis, potentially leading to impaired sexual function and impotence.[8] The goal of management for stuttering priapism is the prevention of future episodes. A practical approach to the diagnosis and management of priapism in patients with CML will be presented here. Priapism is an unusual and rarely presentation for CML.[9-11]

Hyperleukocytosis is thought to be the cause of priapism in patients with leukemia.[12] Three different mechanisms have been described such as: 1) venous congestion of the corpora cavernosa resulting from mechanical pressure on the abdominal veins by the splenomegaly; 2) sludging of leukemic cells in the corpora cavernosa and the dorsal veins of the penis; and 3) infiltration of the sacral nerves and central nervous system with leukemic cells.[13] In our case, significant leukocytosis with hepatosplenomegaly supports the first mechanism in the pathogenesis. Leukostasis is a medical emergency most commonly seen in patients with CML in blast crisis.[7,10,11]

In this case, it was uncertain whether the patient had received less invasive treatment of priapism before received shunting therapy from a surgeon or not. In addition, priapism had already occurred for more than 24 hours since the incident because the health facilities were quite far from the patient's area. Therefore, it worsened the penis condition besides of patient's CML. This supported by the development of penile gangrene due to prolonged erection, resulting in ischemia of the blood vessels. Furthermore, it developed into Fournier gangrene during the treatment process due to infection in the Colles fascia. It spread to the fascia buck for several days due to the patient weak immune system.

CONCLUSION

In this case report, we conclude there is a correlation between CML, based on our clinical and laboratory findings, with priapism. This is also followed by the occurrence of Fournier gangrene in this patient.

CONFLICT OF INTEREST

This statement is to certify that all Authors have seen and approved the manuscript being submitted. We warrant that the article is the Author's original work. We warrant that the article has not received prior publication and is not under consideration for publication elsewhere. On behalf of all Co-Authors, the corresponding Author shall bear full responsibility for the submission. This case report has not been submitted for publication, nor has it been published in whole or in part elsewhere. We attest that all authors listed on the title page have contributed significantly to the work, have read the manuscript, attest to the validity and legitimacy of the data and its interpretation, and agree to its submission.

FUNDING

No funding sources.

AUTHOR CONTRIBUTIONS

All authors equally contribute to the study from the conceptualization, formal

analysis and investigation, writing original draft preparation, writing-review and editing, resources, and supervision of manuscript.

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