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ORIGINAL ARTICLE

Fournier's gangrene with massive pulmonary thromboembolism in an infant: case report

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Abstract

Fournier gangrene (GF) is a rare necrotizing fasciitis that affects the perineal and genital regions, most commonly affecting the elderly and immunocompromised men. We report a atypical case of GF with pulmonary thromboembolism (PTE) in a previously healthy infant in the interior of São Paulo, Brazil. Male patient, 1 year and 1 month old, had a history of "red dots" in the genital and inguinal areas and both thighs. Upon evaluation, the patient presented deep bilateral inguinal lesions with necrosis and extensive infraumbilical superficial desquamation. Laboratory investigation revealed a positive blood culture for Pseudomonas aeruginosa. It progressed to cutaneous sepsis, associated with hemodynamic instability and worsening renal function, in addition to overload of the right chambers and PTE. After thrombolysis, the patient progressed with slow and progressive improvement until hemodynamic stability. Considering the rarity and severity of GF in the pediatric population, our case contributes to the benefits of early diagnostic recognition.

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INTRODUCTION

Fournier gangrene (GF) is a rare and fatal form of necrotizing fasciitis that affects the perineal, genital, and perianal regions, causing septic shock and, consequently, multiple organ failure[1,2]. The infection begins in the region adjacent to the portal of entry, leading to obliterative endarteritis, which progresses to cutaneous and subcutaneous vascular necrosis, with localized ischemia and bacterial proliferation[2]. GF is a polymicrobial disease by nature, with commonly cultured pathogens including Staphylococcus spp., Streptococcus spp., Escherichia coli, Pseudomonas spp., and Bacteroides spp., while Candida spp. represent the most common non-bacterial causative pathogens[3]. Studies report varying profiles of microorganism prevalence, emphasizing the importance of local microbial guidelines and antimicrobial resistance[4]. GF is considered rare in the general population and most commonly occurs in elderly men with underlying conditions such as immunosuppressive therapy, alcohol dependence, diabetes mellitus, human immunodeficiency virus disease, chronic steroid use, leukemia, liver disease, malnutrition, low blood pressure, and socioeconomic factors[5]. It is extremely rare in the pediatric population, with only few documented case reports[5-7] in which the disease has been associated with insect bites[8].

Clinical diagnosis of GF is essential. Manifestations include sudden onset of genital or perineal pain and swelling, fever and prostration, which progress to tissue necrosis with purulent secretion, crepitus or fluctuations, and septic shock[4,5]. Early diagnosis and treatment are essential for the management of GF with intensive volume replacement, surgical debridement, and broad-spectrum antibiotics against gram-positive cocci, gram-negative bacilli, and anaerobes. Early surgical exploration with aggressive debridement of devitalized tissue is mandatory, and multiple procedures may be necessary[9]. Pulmonary thromboembolism (PTE) is a complication of GF described in the literature[10]. PTE is a major cause of morbidity and mortality in the general population. Hospital mortality exceeds 30% in patients with massive PTE associated with hemodynamic instability[11], with most deaths occurring during the initial hours of evolution[12]. This study reports a case with a positive outcome of GF with PTE in an infant who was admitted to a hospital in the interior of São Paulo, Brazil.

CASE REPORT

Male patient, 1 year and 1 month old, admitted with a history of "red dots" in the genital and inguinal regions and at some points on the anterior and posterior surface of both thighs, for two days. On the 2nd day of development, the mother sought emergency care and reported that her son had experienced an episode of fever. The patient was diagnosed with chickenpox and administered topical dexamethasone and oral cephalexin. On the 3rd day of progression, the mother noticed a purplish color in the affected areas, continued treatment at home and only sought care the following day, when she observed an increase in the number and severity of lesions. The patient did not experience any other symptoms during this period.

On the 5th day of evolution, the patient was in serious condition and referred to our pediatric ICU service. Upon physical examination, the patient's general condition was normal: hydrated, acyanotic, anicteric, and afebrile. However, signs of toxemia and tearfulness were observed; the patient was taking dexmedetomidine hydrochloride. During perineal evaluation, a violet plaque with irregular edges measuring approximately 15 cm was observed under an area of violet erythematous erosion with irregular scaly edges and precise limits, extending from the pubic region to the right iliac fossa. A white ulcerated plaque (4 cm) with regular edges was present in the right inguinal region, along with millimeter-sized vesicles in the inguinal and scrotal regions (Figure 1A).

Laboratory tests were carried out, with abnormalities described below:

- Blood count: hemoglobin, 8.9 g/dL; hematocrit, 26.6% (37–47%); leukocytes, 25,050/mm; rods, 4%; segmented, 66%; eosinophils, 0%; basophils, 0%; typical lymphocytes, 17%; monocytes, 13%; and platelets, 147,000/mm³
- Arterial lactate: 8 mg/dL, electrolytes within normal limits, absence of any alterations in renal and hepatic functions, C-reactive protein, 127.47 mg/L
- Blood culture: *P. aeruginosa*

The patient progressed to cutaneous sepsis, was intubated, and started on vasoactive drugs. Due to extensive lesions, necrotic area, and hemodynamic instability presented by the patient, vancomycin 60 mg/kg + meropenem 120 mg/kg in combination with clindamycin 40 mg/kg/day was initiated. The lesions progressed significantly (Figure 1B), with slight response to the proposed antibiotic therapy, requiring follow-up with pediatric surgery to assess the best time to approach. Over time, the patient developed hemodynamic stability, allowing a reduction in the dosage of vasoactive drugs.

On the 8th day of evolution, the patient presented a significant worsening condition, with a drop in blood pressure and oxygen saturation, along with worsening renal function and oliguria, requiring re-initiation of vasoactive drugs. On the 9th day, the patient underwent echocardiography, which revealed indirect signs of pulmonary arterial hypertension, significant tricuspid reflux, dilation of the right chambers, moderate systolic dysfunction and inverted D-shape of the right ventricle. An electrocardiogram with an s1q3t3 pattern was also performed, which led to the diagnosis of PTE. Due to the patient's excessive hemodynamic instability, alteplase was administered at a dose of 0.18 mg/kg and heparin was started using a continuous pump. The patient showed slow and progressive improvement in the kidney function and did not require dialysis. On the 11th day of evolution, dosages of

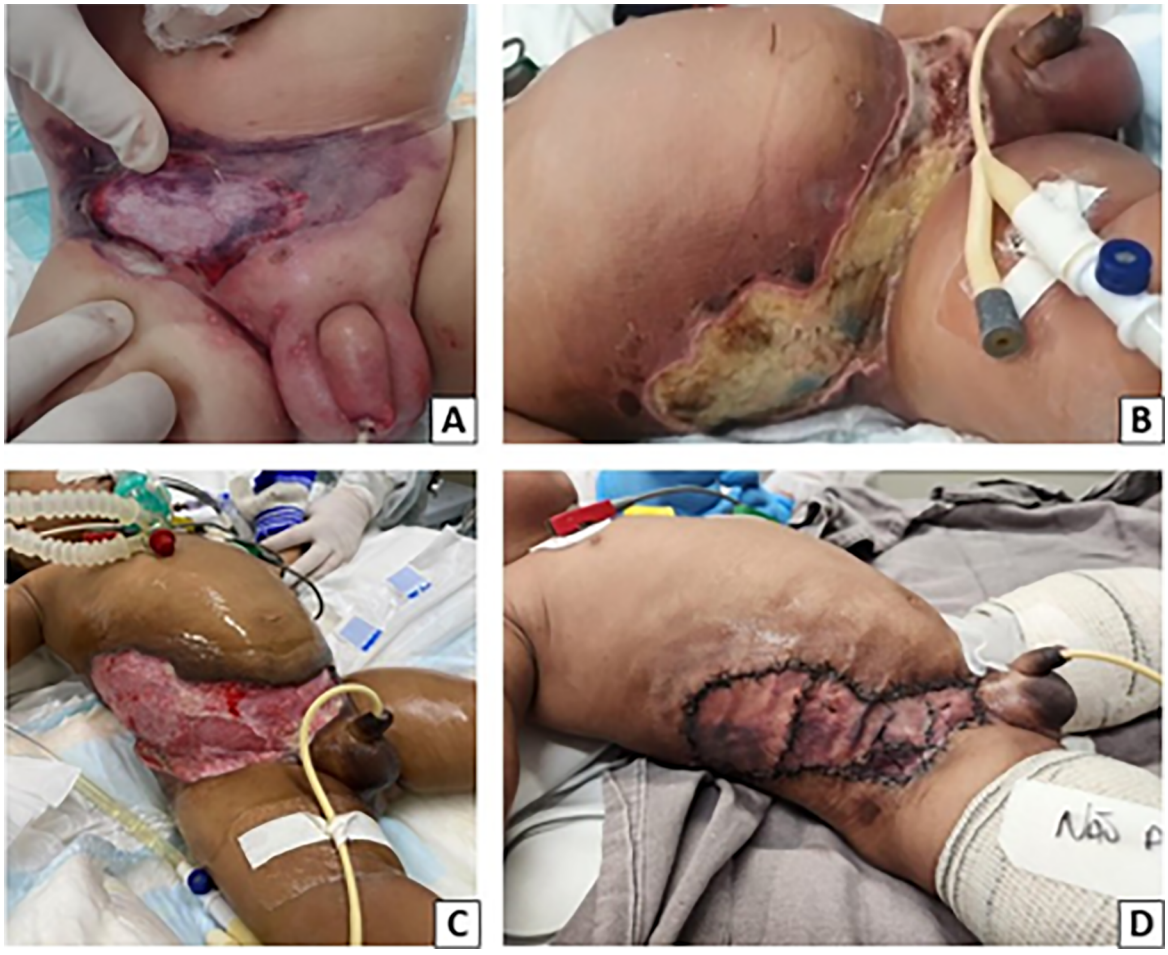


Figure 1. A. Violet plaque (15 cm) with irregular edges under erythematous erosion in the pubic region, extending to the right iliac fossa. White ulcerated plaque (4 cm) with regular edges in the right inguinal region and millimeter-sized vesicles in the inguinal and scrotal regions. B. Ulcerated plaque (35 cm) with irregular edges, exposing subcutaneous tissue and muscular fascia from the right flank to the pubis. C. Erythematous ulcerated plaque (45 cm) with active bleeding, irregular edges, and precise limits from the right flank to the pubis. D. Synthesis.

vasoactive drugs were reduced; there was a significant improvement in diuresis, progressing with hemodynamic stability.

On the 25th day of evolution, debridement of the injury to the abdominal wall and root of the thighs was performed. On the 27th day, a new debridement was performed by the pediatric surgery team using a vacuum dressing. On the 34th day, debridement of the abdominal and perineal regions was performed (Figure 1C).

In the synthesis of the left flank abdominal region, primary suture was suggested, and in the right flank abdominal region, a skin graft was performed from the vastus anterior of the thigh (Figure 1D).

After clinical evolution with good graft adhesion and extubation, the patient was discharged on the 55th day of the condition. The patient was followed up with pediatric surgery on an outpatient basis.

DISCUSSION

Here, we present a rare case of GF in an infant[5-7]. The case is noteworthy for affecting an age group that is not commonly impacted, and for presenting in a patient without prior comorbidities[5]. The causative microorganism was identified as *Pseudomonas* spp., one of the most frequently described etiological agents in the literature[3-4]. The patient presented a significant progression of the disease despite being on early antibiotic therapy, requiring surgical intervention using several approaches, and a subsequent need for skin grafting, and good prognosis. The patient also developed a massive PTE, which is a rare outcome in pediatric patients[13]. Due to the low incidence of this condition in this age group, a study conducted by Silva et al. (2023)[14] highlighted the challenges in identifying and diagnosing PTE, particularly due to the lack of validated tests

for pediatric patients, unlike the well-established methods available for adults. This limitation directly impacts the clinical progression of the condition. It is estimated that 10% of patients with symptomatic PTE die within 1 hour of the onset of the condition[15]. Patients who survive acutely, generally present with nonspecific symptoms; therefore, the diagnosis of PTE is often delayed or completely ignored[15]. Finally, despite the current technical and scientific knowledge regarding PTE in the pediatric population, in the present case, at the onset of the PTE, the patient was placed on aggressive hemodynamic support with access to advanced diagnostic and therapeutic resources. Within less than 24 hours, the administration of alteplase was initiated, resulting in a favorable clinical response.

CONCLUSION

Considering the rarity and severity of GF in the pediatric population, this case report contributes to the benefits of early diagnostic recognition. In our experience, early antibiotic therapy and surgical approach are essential for favorable outcomes.

ETHICAL APPROVAL AND INFORMED CONSENT

The project was approved by the Research Ethics Committee of Anhembi Morumbi University under approval number 6.691.807. The legal guardian of the child was informed about the objectives and all procedures involved in the research and signed the informed consent form (ICF), authorizing the use of medical records, examination results, and their publication in a non-identifiable manner.

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