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ABOUT COVER

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CASE REPORT

Fournier gangrene in an infant, complicated with severe sepsis and liver dysfunction: A case report

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Abstract

BACKGROUND

Fournier gangrene is a rare, life-threatening infection characterized by necrotizing fasciitis in the perineal, genital and/or lower abdominal regions. Despite its rarity, the unfavorable prognosis associated with this disease is dependent on the timing of medical care.

CASE SUMMARY

A 3-month-old boy was admitted to our pediatric intensive care unit in critical condition after a 5-day history of fever and scrotal erythema with breaching skin lesions and swelling. Despite ambulatory antibiotic treatment, the child's clinical condition deteriorated. At the time of admission, the child had necrotizing scrotal fasciitis that had spread to the abdomen. Following reanimation, the surgeon decided on an immediate intervention to rule out testicular torsion and to debride the affected area. Despite optimal antibiotic and supportive therapy, the patient developed severe sepsis with liver dysfunction, making treatment more challenging.

CONCLUSION

Recognizing Fournier gangrene, prompt referral to pediatric surgery, and appropriate antibiotic coverage are critical for avoiding sepsis and multiorgan dysfunction.

Key Words: Fournier gangrene; Infant; Early diagnosis; Sepsis; Liver dysfunction; Case report

Core Tip: This is a case report of an infant with Fournier gangrene. Despite its rarity, Fournier gangrene remains a disease with severe complications and high mortality. The prognosis is influenced by the timing of medical treatment. Despite receiving optimal antibiotic and supportive care after hospital admission, the patient developed sepsis with severe liver damage, making treatment more challenging. The prognosis to sepsis and liver dysfunction was influenced by delayed diagnosis. Through this case report we highlight the importance of early recognition and high clinical suspicion by all doctors (pediatricians, dermatologist, surgeons, *etc.*), to make an accurate diagnosis.

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INTRODUCTION

Fournier gangrene is a severe and aggressive form of infective necrotizing fasciitis that affects the perineum and genitalia as a result of polymicrobial infection[1-4]. Despite being named Fournier (Jean-Alfred Fournier) after he published a series of five cases, the condition was first described as an idiopathic, rapidly progressive necrotic process of the soft tissues leading to genital organ gangrene in 1764 by a physician named Baurienne[5]. Fournier gangrene in children is rare, with an annual incidence of 0.8 per million patients, and little is known about the disease in this age group[2,4-10]. A review of the literature revealed only 56 pediatric cases, with 66% of those involving infants under the age of 3 mo[5,7]. Despite its rarity, Fournier gangrene was and continues to be a disease with severe complications and high mortality. The prognosis for this disease is dependent on the timing of medical care. Because early manifestations are frequently subtle, through this case report we would like to emphasize the importance of early recognition and high clinical suspicion by all doctors in order to make an accurate diagnosis.

CASE PRESENTATION

Chief complaints

A 3-month-old boy was admitted to our pediatric intensive care unit in critical condition, intoxicated and drowsy, after a 5-day history of fever and progressively increasing scrotal swelling with scrotal zone discoloration.

History of present illness

He initially displayed skin breaches in the scrotum and right inguinal area (Figure 1A). The child was examined 3 days before hospitalization and began antibiotic treatment (oral cefaclor and topical fusidic acid). Despite outpatient treatment, the child's clinical condition worsened (Figure 1B).

History of past illness

His medical history revealed that he was born at term, appropriate for gestational age, and weighed 3500 g. The child was in good general condition 5 d before admission. There was no history of surgical intervention, perineum or lower abdomen injury, catheterization, insect bite, or other predisposing conditions.

Personal and family history

His parents are in good health.

Physical examination

The scrotal area appeared edematous at admission, with a dark to black color that extended towards the dexter inguinal area and the abdomen, with some ulcerative areas (Figure 1C).

Laboratory examinations

The initial laboratory results revealed leukocytosis (white blood cell count of 23200/mm^3) with 78% neutrophils, high C-reactive protein (9.95 mg/dL), blood urea nitrogen 80 mg/dL; creatinine 0.72 mg/dL, high aspartate transaminase (AST) levels of 563 IU/L, high alanine transaminase (ALT) levels of 187I U/L, total bilirubin of 1.39 mg/dL, low total protein of 3.5 g/dL, prothrombin time of 21 s, activated partial thromboplastin time of 47.7 s, and fibrinogen level of 302 mg/dL.

Imaging examinations

Ultrasound was performed to rule out a deep soft tissue infection and a torsion testis. The presence of liquid with corpuscular content was confirmed by ultrasound, as was thickening of the scrotal walls, but without a clear assessment of blood flow in the appropriate testis.

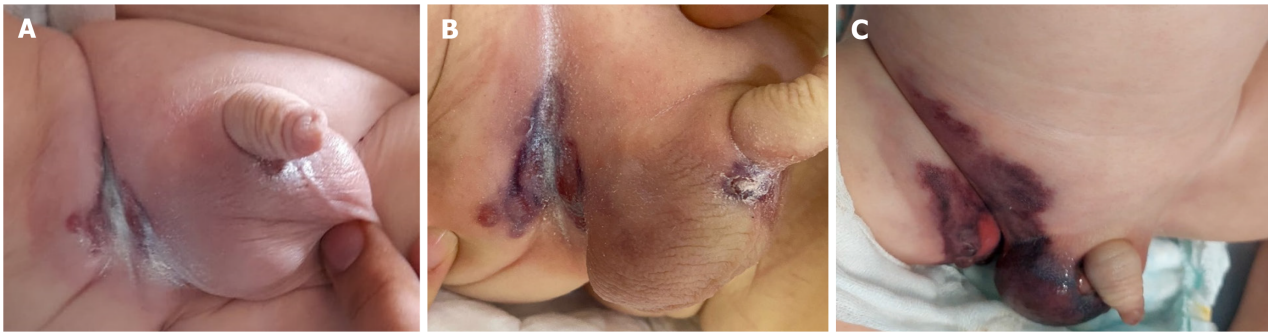

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Figure 1 Evolution of scrotal necrosis from the day 2 of history to day 1 of hospital admission. A: Skin breaches in the scrotum and right inguinal area; B: The child's clinical condition worsened; C: The scrotal area appeared edematous at admission, with a dark to black color that extended towards the dexter inguinal area and the abdomen, with some ulcerative areas.

MULTIDISCIPLINARY EXPERT CONSULTATION

After vigorous resuscitation, the surgeon decided on an immediate intervention to rule out testicular torsion and to debride the affected region.

FINAL DIAGNOSIS

The intervention clarified the child's diagnosis of Fournier's gangrene.

TREATMENT

In addition to other supportive measures, the baby was given intravenous fluids and broad-spectrum antibiotics that covered both aerobic and anaerobic bacteria.

OUTCOME AND FOLLOW-UP

Despite receiving optimal antibiotic and supportive therapy, the patient developed sepsis with severe liver injury that led to liver failure, making treatment more challenging. AST, ALT and bilirubin levels peaked on the second day of admission (ALT: 651 IU/L, AST: 2993 IU/L, total bilirubin: 3.9 mg/dL, prothrombin time: 6 s). The liver parameters improved 4 d after admission and returned to normal levels after 10 d. The results of blood and urine cultures were both negative. The immunological parameters were within the normal range. In the postoperative period, antibiotics and other supportive treatments, as well as regular dressing, were continued. The child was discharged from the hospital in a good clinical condition.

DISCUSSION

Fournier gangrene is a type of necrotizing fasciitis that affects the external genital organs as well as the perianal region. It is characterized by thrombosis of the feeding arteries, which leads to gangrene of the skin and subcutaneous tissues, as well as severe intoxication and multiple organ failure. The necrotizing process is frequently caused by an infection in the anorectum, urogenital tract, or genital skin[1,17,18]. Omphalitis, strangulated hernia, prematurity, diaper rash, varicella infection, circumcision, and perineal abscesses have all been reported as etiological factors in the pediatric age group[1,8]. Trauma, minor skin breaches (as in our case), insect bites, surgeries or invasive procedures in the perineal region, urethral instrumentation, burns, and systemic infections are some of the other causes in children[1,5].

Despite its rarity, the unfavorable prognosis is dependent on the timing of medical care. The treatment delay is accompanied by a high lethality rate, reaching 90%, due to the development of septic shock and its associated complications[13]. Fournier gangrene progresses more quickly in children than in older people as their immune systems are immature and their skin barrier is more fragile. As a result, the situation is more likely to lead to septic shock, dysfunction of many organs, and a poor prognosis[12].

The liver plays an important role in defensive responses to scavenge bacteria and produce inflammatory mediators during sepsis. However, the liver can also be a target of a dysregulated inflammatory response. Although liver dysfunction is not the most common type of organ injury seen in septic patients, when it progresses to liver failure, it

becomes a serious complication. Based on the new consensus for sepsis definition, the clinical situation, and the additional investigation, sepsis appears to have been complicated by significant liver injury in our case[14-16].

Lu *et al*[12] also reported the case of an 8-month-old girl with Fournier gangrene caused by *Escherichia coli*, which was complicated by septic shock and septic associated encephalopathy[12]. The majority of pediatric cases of Fournier gangrene have been reported in previously healthy children, who may appear well in the early stages of the disease and thus escape diagnosis, as in our case[3].

Radiography, ultrasound, computed tomography, and magnetic resonance imaging (MRI) are all used to diagnose Fournier gangrene. An X-ray of the region may show gas in the soft tissues, which is an absolute indication of surgical intervention. However, the diagnosis is usually clinical[5].

In soft tissues, ultrasound can detect fluid or gas. Gas in the scrotal wall is the sonographic hallmark of Fournier gangrene. Scrotal wall edema is possible. Blood flow and testicular status can also be determined using ultrasound. Contrary to most urological diseases, Fournier gangrene does not affect the testes. This is due to their supply's independence from the blood supply of the scrotum and penis[13]. Even in our case, we were able to keep the tests. The use of MRI in the treatment of Fournier gangrene is not well documented in the literature. MRI should not be used to postpone surgery if the diagnosis is highly suspect.

The treatment for Fournier gangrene is based on an emergency surgical intervention combined with antibacterial therapy. Surgery is required for a definitive diagnosis and the removal of necrotic tissue. A delay to the surgical intervention is the most significant modifiable risk factor associated with mortality. Once Fournier gangrene diagnosis has been made, all necrotic tissues must be removed. A systemic pediatric review determined that early aggressive surgical treatment was the best option[17]. A study confirmed the opinion that early surgical intervention reduces mortality in a large retrospective review of 379 patients. Those who received earlier intervention had a lower fatality rate (odds ratio = 0.38) than those who received intervention 3 d or later[5].

In approximately 95% of cases, the cause of Fournier gangrene can be identified. Necrotizing fasciitis is classified into two types based on the organisms that cause it. Type 1 bacteria are typically polymicrobial, containing both aerobic and anaerobic bacteria. Type 2 is monomicrobial and is frequently caused by *Streptococcus* group A alone or in combination with *Staphylococcus aureus*[18]. Zundel *et al*[17] with their systematic review, aimed to better characterize the pediatric Fournier gangrene. In their study, 53 pediatric patients were included. Monomicrobial necrotizing fasciitis (type 2) was far more common than polymicrobial fasciitis (type 1). *Pseudomonas aeruginosa* was frequently isolated alongside streptococci and staphylococci[17]. In our case, we were unable to determine the etiology.

Considering the polymicrobial (aerobic and anaerobic) causative microflora, the drugs of choice for antibacterial therapy of Fournier gangrene are second or third generation cephalosporins combined with antibiotics from the nitroimidazole, fluoroquinolone and aminoglycoside groups. Antibiotics from the carbapenem class are used in the complex processes of antibacterial therapy in severe cases of the disease[13]. Streptococci and staphylococci are more common in children than Gram-negative and anaerobic bacteria, which predominate in adults[1,17,19]. As a result, multiple antibiotics are recommended, including one specifically targeted toward *S. aureus*[12,20]. This aggressive treatment modality was effective in our patient.

According to various authors, mortality ranged from 3% to 45% due to severe sepsis, coagulopathy and renal failure[1,17,20,21]. Necrotizing fasciitis in neonates is associated with a nearly 50% mortality rate[10]. The prognosis of this disease is influenced by age, the presence of infectious agents, extent of the disease, and underlying medical conditions. A multicenter retrospective study found that older age, diabetes, anemia, sepsis, a delay in initial treatment, and a Fournier Gangrene Severity Index score of 9 are important predictors of disease severity[12,22]. The mainstays of management in children with Fournier gangrene are a high index of suspicion and prompt diagnosis, as well as surgical debridement and a multidisciplinary approach[12,20,21].

CONCLUSION

Recognizing Fournier gangrene, referring the children for a pediatric surgery as soon as possible, and providing the appropriate antibiotic coverage are crucial for avoiding sepsis and multiorgan dysfunction and improving prognosis.

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FOOTNOTES

Author contributions: Bakalli I conceptualized, supervised data collection, drafted the initial manuscript, and corrected the final English version; Heta S and Celaj E contributed to the acquisition of data; Kola E revised the manuscript; and all authors have read and approved the manuscript critically.

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