



Fournier's gangrene in a two-month-old infant

Muhammad Asykar Palinrungi^{a, *}, Sulmiati^b, Sachraswaty Rachman Laidding^c,
Farid Nur Mantu^b, Elanda Putri Madyaningtias^d, Robert Christeven^d, Muhammad Faruk^d

^a Division of Urology, Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia

^b Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia

^c Division of Plastic and Reconstructive Surgery, Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia

^d Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia

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ABSTRACT

We present a case of Fournier gangrene (FG) in a two-month-old male infant which admitted to the hospital with chief complaining of progressive scrotal swelling for two weeks. Scrotal ultrasonography showed a free fluid on right hemiscrotal and normal testicles size. Debridement was conducted to remove devitalized tissue. Forty days after the surgery, the patient remained well without any complication.

1. Introduction

FG, a rare and potentially life-threatening disease, is a progressive gangrenous process of the genitalia, perineum and perianal regions [1,2]. FG has an abrupt onset with the progressive gangrenous formation which can lead to gangrenous genitalia destruction. Most of the reported FG cases are caused by polymicrobial infection, which includes gram-negative, gram-positive, aerobic and anaerobic bacteria [2,3]. Establishing the diagnosis of FG is challenging because of surgical emergency as a progression from genitalia to perineum to the abdominal wall may occur in a short time (often within hours). The exclusion of FG should be a priority during every consultation for acute scrotal swelling [2].

FG is more common in adults with peak incidence occurs between 20 and 50 years of age. Even though FG can occur in all age groups, it is infrequently found in pediatric age groups [4]. Literature review only found 56 reported cases of FG in children which 66% of them have been found in the first three months of life [5]. In this report, we present our case of a two-month-old infant with FG who was referred to our hospital.

2. Case presentation

A two-month-old infant was hospitalized with a chief complaint of progressively increasing scrotal swelling for two weeks and discoloration of the scrotal skin for the past seven days before hospitalization.

The baby had been in a fever for five days before hospitalization. The baby was born full-term from a 33-year-old P3A0 mother by cesarean section due to premature rupture of the amniotic membrane. The baby's birth weight was 2600 g. There was no history of any type of invasive intervention, indwelling catheters, localized trauma, surgical procedure, insect bite, steroids use, prolonged hospitalization, and history of human immunodeficiency virus from mother, or the other predisposing conditions.

On physical examination, the baby was irritable, well-nourished, he had heart rate of 142 beats/minute, respiratory rate of 42 times/minute, and body temperature of 100.4 °F. On external genitalia, scrotal skin was found to be grayish on left hemiscrotal and blackish-brown on right hemiscrotal, indurated with sharp and clear demarcation with the normal skin (Fig. 1). The penis was erythema, purulent, ulcerated, with necrotic tissue on the dorsal of the penis. The skin of suprapubic, inguinal, umbilical region and bilateral medial thigh was involved and peeled. Laboratory investigation showed a leukocyte count of 16.200/μl with neutrophil of 66.6%, hemoglobin 10.6 gr/dl, platelet count of 261.000/μl, blood urea 11 mg/dl, creatinine 0.28 mg/dl, and electrolyte investigation result was within normal limit. Abdominal X-Ray showed soft tissue density of the scrotal region (Fig. 2). Scrotal ultrasonography showed a free fluid on right hemiscrotal and normal testicles size (Fig. 3).

* Corresponding author. Division of Urology, Department of Surgery, Faculty of Medicine, Hasanuddin University, Perintis Kemerdekaan KM 11, Makassar, 90245, Indonesia.

E-mail address: apalinrungi@yahoo.com, apalinrungi74@gmail.com (M.A. Palinrungi).

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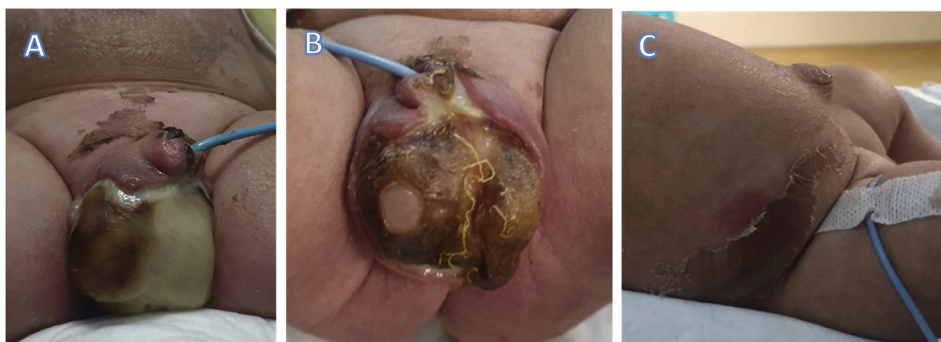


Fig. 1. A. Showing scrotal swelling and discoloration of scrotal skin. The penis and surrounding skin were also involved. B. Showing the scrotal skin were worsened on the second day of hospitalization. C. Showing the surrounding skin were worsened on the second day of hospitalization.



Fig. 2. Showing soft tissue density of scrotal region (arrow).

The patient was resuscitated with intravenous fluids. Cefotaxime and metronidazole, the broad-spectrum antibiotics which covered both aerobic and anaerobic organisms, were administered in the first hours of hospital admission. On the second day of hospitalization, the scrotal swelling was worsening with blackish-brown discoloration of scrotal skin was widened (Fig. 1b and c). Surgical debridement was undertaken under general anesthesia. All devitalized and necrotic tissues were excised, thus exposing the unaffected testicles (Fig. 4a). A swab of pus was obtained and sent to the microbiology laboratory for bacterial culture. The penis and the surrounding skin lesions were also debrided in the area of active bleeding. The wound was irrigated repeatedly with normal saline and povidone-iodine; then packed with antibiotic tulle dressing and dry sterile gauze. This dressing procedure was continued in the postoperative period. Microbiology culture from pus swab showed the growth of *Pseudomonas Aeruginosa* and antibiotics were continued according to the sensitivity report. After several days of postoperative treatment, the surgical wound was improved (Fig. 4b) and the baby was discharged from the hospital on the 7th postoperative day. The patient's parents had been educated regarding the maintenance of proper hygiene and wound care. The baby has remained well, and the surgical wound has improved at follow-up in an out-patient clinic. On the 40th post-operative day, the baby remained healthy and the surgical wound was healed perfectly with minimal scarring (Fig. 4c).

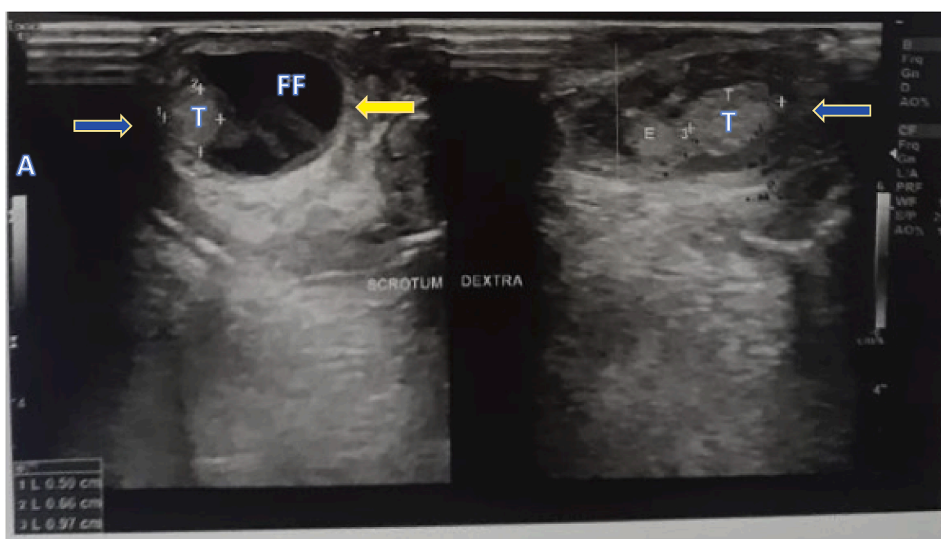


Fig. 3. Free fluid (FF) on right hemiscrotal (yellow arrow) and normal testicle (T) (blue arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

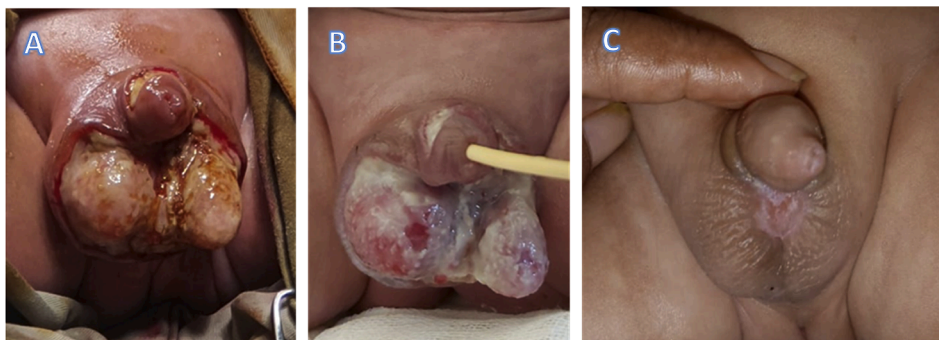


Fig. 4. Showing the surgical wound A. After debridement, B. 4th day postoperatively; C. The wound was healed perfectly with minimal scarring on the 40th postoperatively day.

3. Discussion

Idiopathic scrotal gangrene is an uncommon disease, but it has a fearful effect. FG is a vascular disaster of genitalia caused by infective origin [6]. Although originally described as idiopathic genitalia gangrenous disease, FG has an identifiable etiology and predisposing factors in approximately 95% of cases [3,7]. Mortality of FG as reported by different literature ranged from 3% to 45% and it was due to severe sepsis, coagulopathy, and renal failure [3]. In infants, FG is associated with almost 50% mortality [6]. The necrotizing process commonly originates from an infection in the urogenital tract, anorectal region, and/or skin of the genitalia.

Any condition which leads to a decrease of cellular immunity may predispose a child to the development of FG. Preterm and low-birth-weight infants with impaired immune status and those with poor local hygiene appear to have increased risk of this rare disease [3]. In children, some predisposing factors are urethral instrumentation/in-dwelling catheterization, strangulated inguinal hernia, circumcision, omphalitis, insect bites, trauma, poor genitalia hygiene, peri-rectal abscess, systemic infection, and burn injury [3,8]. In our case, the predisposing factors were poor local hygiene status and immaturity of the infant immune system.

Wound cultures from patients with FG reveal that it is a polymicrobial infection with an average of four isolates per case. *Escherichia coli* is the commonest aerobe, and *Bacteroides* species the commonest anaerobe. Other common micro-flora include *Staphylococcus*, *Enterococcus*, *Proteus*, *Streptococcus*, *Pseudomonas*, *Klebsiella*, and *Clostridium* [7]. The bacteria act synergistically to produce enzymes that invade the fascial planes and lead to vascular thrombosis with subsequent gangrene of the overlying skin [8]. The first type of microorganism might produce enzymes needed to cause coagulation and thrombosis of nutrient vessels which cause a decrease in local blood supply [7] resulting in ischemia and tissue hypoxia that allow facultative anaerobic growth and micro-aerophilic microorganisms. The resultant tissue hypoxia due to ischemia allows the growth of facultative anaerobes and micro-aerophilic microorganisms [3]. These latter microorganisms produce enzymes (e.g., *Lecithinase*, *Collagenase*), which lead to digestion of fascial plane barriers, thus causing the rapid extension of the infection. Necrosis and digestion of the fascial are hallmarks of FG, and it can spread from the fascial layer of the genitalia throughout the perineum, along the trunk, occasionally into the thighs [3].

The pus swab in our case revealed only *Pseudomonas Aeruginosa*, but the existence of the other microorganism could not be excluded because the only poly-microbial infection could cause extensive and rapid progressing necrosis. The negative growth of the other microorganisms in culture medium could be caused by improper sample swab handling and the absence of specific nutrients in medium culture.

The clinical manifestation of affected overlying skin may be erythematous, edematous, cyanotic, bronzed, indurated, blistered, and

gangrenous. A feculent odor may be present secondary to infection with anaerobic microorganism. Crepitus may be present, but its absence does not exclude the gas-forming microorganisms. The imaging finding of soft-tissue air may be present before clinical crepitus is identified. However, the absence of subcutaneous air in the scrotum or perineum does not exclude the diagnosis of FG [9]. Systemic symptoms (e.g. fever, tachycardia, and hypotension) may be present. It is a necessity to establish the diagnosis of FG as soon as possible because the progression of the gangrenous process from genitalia to perineum to the abdominal wall may occur extremely rapidly (often within hours) [2]. In our case, we also encountered rapid progressing of scrotal swelling and tissue necrosis.

Treatment of FG involves a combination of broad-spectrum antibiotics and extensive surgical debridement to margins of healthy bleeding tissue [2,7]. Some patients may often require a second operation after 24–48 hours to exclude further disease progression. The testicles and other structures within the tunica vaginalis can almost always be spared, although a loss of tissue in the abdominal wall may be extensive due to bacterial spread along fascial planes [2].

In our case, we had administered broad-spectrum antibiotics, such as the combination of cefotaxime and metronidazole from the first hours of hospital admission and surgical debridement had been performed until reaching healthy bleeding tissue. The testicles were spared from the infection. Having proper wound care, the post debridement wound was healed perfectly, only leaving minimal scarring.

Since this patient had poor hygiene and no obvious inciting factors were found, we have concluded that poor hygiene can predispose FG in an infant. Proper health education to the parents regarding good hygiene can probably prevent FG in infants.

4. Conclusion

FG of the scrotum is an extremely rare case in the infant but has a fearful effect which can lead to death due to sepsis. It should be considered as a differential diagnosis in the clinical management of the acute scrotum. The FG is a surgical emergency as a progression from genitalia to perineum to the abdominal wall may occur extremely rapidly. The exclusion of FG, should be a priority during every consultation for soft tissue infection of the genitalia Proper health education to the parents regarding good hygiene can probably prevent FG in infants.

Consent

Written informed consent was obtained from his parent patient for publication in this case report.

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Declaration of competing interest

No potential conflict of interest relevant to this article was reported.

References

- [1] Ameh EA, Dauda MM, Sabiu L, Mshelbwala PM, Mbibu HN, Nmadu PT. Fournier's gangrene in neonates and infants. *Eur J Pediatr Surg* 2004 Dec;14(6):418–21.
- [2] Link RE, Theodore R. Fournier gangrene (necrotizing fasciitis of the perineum). In: Partin AW, Peters CA, Kavoussi LR, Roger R, Dmochowski AJW, editors. *Campbell-walsh urology*. Twelfth. Philadelphia: Elsevier; 2020. p. 211–7.
- [3] Eke N, Fournier's Gangrene EJ. In: *gangrene - current concepts and management options*. InTech; 2011.
- [4] Rouzrokh M, Tavassoli A, Mirshemirani A. Fournier's gangrene in children: report on 7 cases and review of literature. *Iran J Pediatr* 2014 Oct;24(5):660–1. 2014/07/20.
- [5] Adams JR, Mata JA, Venable DZ, Culkun DJ, Bocchini JA. Fourniers gangrene in children. *Urology* 1990 May;35(5):439–41.
- [6] Idweini Y. Necrotizing scrotum fasciitis (Fournier's Gangrene) in an Infant: a case report and review of the literature. *Clin Res Urol* 2018;1(1):1–3.
- [7] Dey S, Bhutia KL, Baruah AK, Kharga B, Mohanta PK, Singh VK. Neonatal Fournier's gangrene. *Arch Iran Med* 2010 Jul;13(4):360–2.
- [8] Singh A, Gupta A, Pardeshi R, Garg D. Neonatal Fournier's gangrene. *J Clin Neonatol* 2018 Jul 1;7(3):174–6.
- [9] Levenson RB, Singh AK, Novelline RA. Fournier gangrene: role of imaging. *Radiographics* 2008;28(2):519–28.