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Fournier's gangrene in childhood: a report of 3 infant patients

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Abstract Fournier's gangrene is uncommon in pediatric age group, and little is known about the disease in the newborn period and infancy.Three patients, aged 10 days, 14, and 17 months, with Fournier's gangrene, were treated in our hospital. The predisposing factors were prematurity, a diaper rash, and varicella infection, respectively. Especially, prematurity and diaper rash are rare predisposing factors in the pediatric population; therefore, high index of suspicion, prompt diagnosis, conservative surgery, and multidisciplinary approach are the mainstays of management in children with Fournier's gangrene.

Fournier's gangrene (FG) is an uncommon necrotizing fasciitis of genitalia and perineum that has high mortality and morbidity [1]. Although initially attributed to Baurienne (1764), FG is named after Jean-Alfred Fournier, who, in 1883 described the entity as idiopathic gangrene of sudden onset and rapid development at the level of genitalia, presenting mainly in healthy young males [1-3]. Now, it is well known that this entity is not solely seen in young males in several reports; the age range changed from early infancy up to adulthood [1,4,5]. Fournier's gangrene has been reported in children in the first week of life. It remains relatively uncommon in children, with just 56 cases reported to date in the literature although Legbo et al. [5] concluded that NF was more common in children than adults in their country [6].

In cases originating in the genitalia, the infecting bacteria probably pass through Buck's fascia of the penis and spread along the dartos fascia of the scrotum and penis, Colles' fascia of the perineum, and Scarpa's fascia of the anterior abdominal wall [1,3,6]. The 3 findings characterizing the syndrome are abrupt onset, rapid progression, and absence of a specific etiologic agent. In early stages, involved area is swollen, erythematous, and tender. As infection begins to involve the deep fascia, pain becomes prominent with high fever and systemic toxicity. The swelling and crepitus of the scrotum progresses, and dark purple areas develop resulting in extensive scrotal gangrene [1,3]. Predisposing factors include diabetes mellitus, local trauma, paraphimosis, periurethral extravasation of urine, perirectal or perianal infections, and surgery, such as circumcision or herniorrhaphy [1,3,7]. Fournier's gangrene is a rare infectious entity

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in childhood, which poses diagnostic and therapeutic challenges for the pediatric surgeon. We present rare cases of FG in the present article.

1. Case 1

A premature infant, born in the week 27 of gestation with a birth weight of 980 g, was taken into the newborn intensive care unit because of fetal stress.

The 1-minute and 5-minute Apgar scores were 5 and 8, respectively. Gentamicin, ampicillin, and cefotaxime were administered after blood and urine cultures were taken. Early enteral feeding with breast milk was begun on postnatal day 2. There was no history of rectal body temperature measurement or suppository application.

Perianal hyperemia was detected on postnatal day 10 without any evidence of necrotizing enterocolitis or abdominal distension. The antibiotic regimen was changed to meropenem and teicoplanin. The hyperemic lesion in the anal region showed a rapid progression in 2 days leading to a necrotic tissue, which required surgical debridement (Fig. 1). Cultures of the wound and blood did not yield any specific microorganisms. Subsequently, a protective colostomy was undertaken to prevent wound contamination. Dressings with topical rifampicin and mupirocin were applied twice a day.

The wound developed a healthy tissue in 2 weeks; however, the patient was taken into a dilatation program for anal stenosis because of stricture.

2. Case 2

A 14-month-old male patient was referred to our university clinic because of necrotizing fasciitis in his perianal region and preputial skin.

His history revealed a diarrhoeal episode that appeared 4 days after beginning to take oral antibiotics for an upper respiratory tract infection, diagnosed 10 days before his admission. The perianal region was hyperemic, warm, and tender.

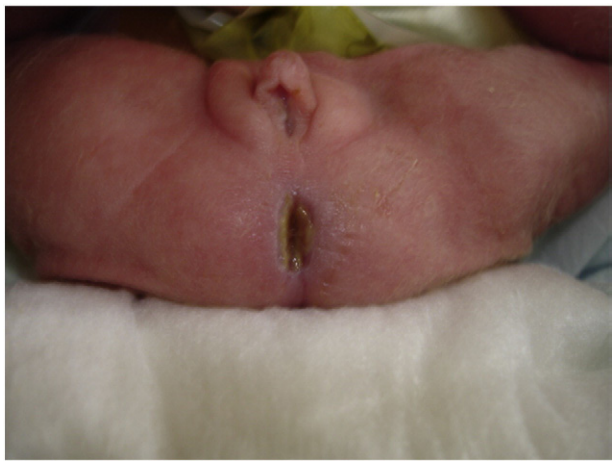

Fig. 1 Healthy granulation tissue of the perianal region after extensive debridement.

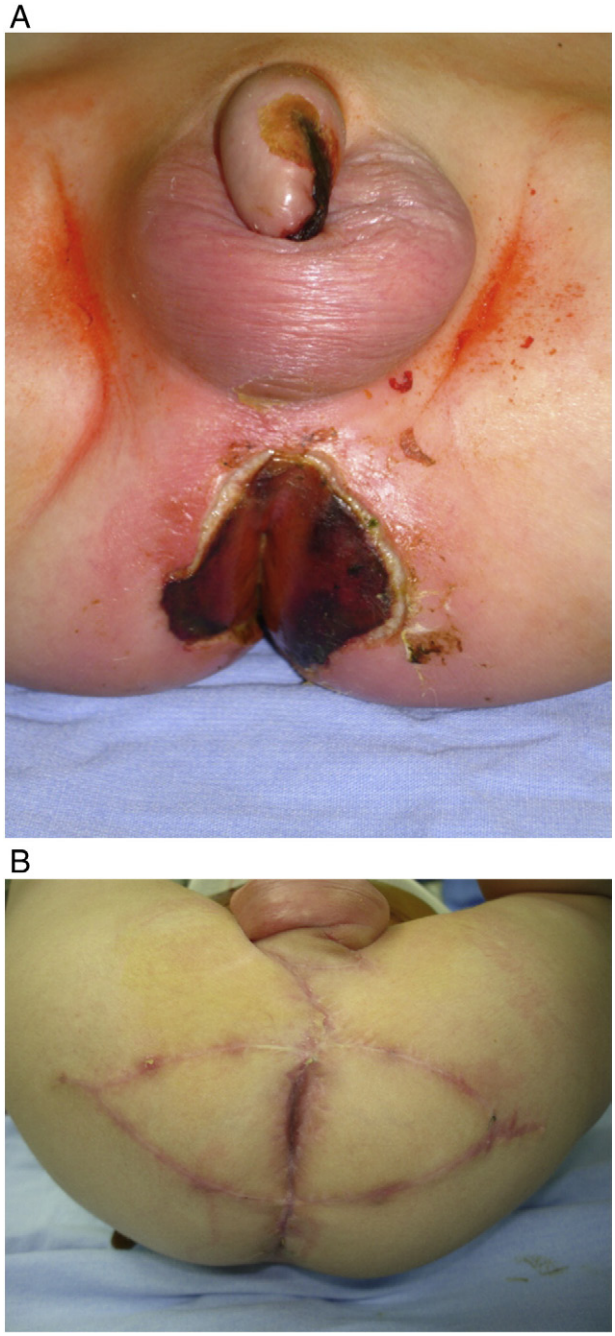

Fig. 2 A, Necrosis of perianal area and penis. B, Final appearance of the perianal region after reconstructive surgery.

Within 2 days of the onset of hyperemia, the color of the affected area became darker and the temperature rose to 40°C. The patient appeared toxic with generalized edema, tachycardia, hypotension, and other signs of sepsis. Edema and increase in local temperature became prominent, and cellulitis was observed around a circular necrotic lesion of 10 cm diameter (Fig. 2a). In addition to diffuse edema and tenderness on the penis, there was a necrotic area with 1 × 3

cm dimensions on the dorsal surface of the penis extending distally to the proximal portion of the preputium.

Laboratory tests showed anemia and neutropenia.

Ornidazole and meropenem were started. Protective colostomy was performed after debridement of the necrotic tissue described above. Because the tissue culture grew *Pseudomonas aeruginosa* and *Enterococcus faecium,* antimicrobial therapy was changed to teicoplanin + meropenem + gentamicin according to antibiotic sensitivities. After removal of the infected tissue, the wound was thoroughly irrigated and dressed with closed vacuum-assisted (VAC) device [8]. Subsequently, the patient underwent suction-assisted device foam dressing changes every 48 hours for 2 weeks. After the VAC device was dislodged, the tissue defect was closed with bilateral v-y advancement flaps together with an anoplasty, performed on the edges of the wound with the addition of circumcision (Fig. 2b). There were not any postoperative complications. The loop colostomy was closed 2 months later.

3. Case 3

A 17-month-old female patient was evaluated for hyperemia and tenderness that had developed in her right groin and perineum after a varicella infection. The patient was admitted to surgical ward with the diagnosis of soft tissue infection and was administered intravenous ampicillin and clindamycin. A rapid local tissue necrosis with high fever developed in the affected area. The patient had a white blood cell count of 28,800/μL and a hemoglobin concentration of 11 g/dL. Blood and urine cultures did not reveal any bacterial growth. On day 7 of her admission, the necrotic tissue was removed surgically to the level of well-vascularized tissue. Daily dressings were applied after this debridement. The tissue defect healed by itself with granulation tissue. The resultant defect was allowed to heal by secondary intention without any further reconstruction.

4. Discussion

Necrotizing fasciitis of genitalia and perineum, also known as *FG* is a rare, but life-threatening process [1-5]. Nowadays, *FG* is defined as an infective necrotizing fasciitis, which affects perianal and genital regions, leading to severe skin necrosis and thrombosis of subcutaneous vasculature. Necrotizing fasciitis in children is a rare disease that produces extensive cellulitis with severe involvement of the subcutaneous tissue, fascia, and muscle or both, resulting in extensive necrosis [1,3,6,7].

The source of infection may be urogenital (45%), anorectal (33%), or cutaneous (21%) [9]. Most of the affected adult patients have an underlying systemic disorder such as alcoholism, diabetes mellitus, malnutrition, immunosuppressive therapy, or low socioeconomic status [1,3,5,6]. In children, predisposing factors include trauma, insect bites, circumcision, burns, periurethral and anorectal diseases, systemic infections, immunocompromised states, and hematologic malignancies [2,7,9]. Interestingly, in case 1, we could not identify any underlying causes. The situation may have been triggered by the immature immune system of the preterm infant. The predisposing factors in the other patients were perianal infection caused by diaper rash and cutaneous varicella infection.

The diagnosis of perineal necrotizing fasciitis is primarily based on clinical evidence. Early on, FG can mimic many other conditions such as cellulitis, erysipelas, viral illness, and others [3,4,7,9]. Clinical findings that point to FG include rapidly spreading edema and hyperemia of perineal skin, numbness of the overlying skin, and severe pain, out of proportion to skin findings [1,3,9]. Additional laboratory tests and histopathologic studies can aid in the diagnosis of FG [3,5,6].

Gram staining of the infected tissue and blood cultures may be helpful in identifying the causative organism and in choosing convenient antibiotics. Although it is not possible to grow microorganisms in every patient, *Escherichia coli*, *Bacteriodes*, *Streptococcus*, *Peptostreptococcus*, and *Clostridium spp* are frequently identified as causative microorganisms in this often polymicrobial infection [1-3,7]. The most commonly isolated organism is *E. coli* in adults and streptococci in children [9]. Only in case 2 a bacterial growth (*Klebsiella*) has been detected in wound culture, whereas no bacteria were identified in the other 2 patients.

The main components of the management of FG, which, in our opinion, should be started as soon as possible, are fluid resuscitation, hemodynamic support, broad-spectrum antibiotics, surgical debridement, and supportive care. As soon as the patient's condition has stabilized, devitalized tissue should be excised aggressively, for necrosis may progress rapidly in hours. Antimicrobial treatment with broad-spectrum antibiotic combinations should be initiated before surgery, and either changed or continued according to results of tissue culture [1,3,5].

Fournier's gangrene is a life-threatening disease in adults, the mortality rates ranging between 11% and 45%. Although the exact figures are not known for children because of limited number of cases, general opinion is that mortality rates varies from 9% to as high as 30% in infants younger than 3 months [6,9,10]. For this reason, although the goal of surgical approach is to remove all devitalized tissue, one can proceed more conservatively in children because of the relatively benign course of the disease in this age group [2,6,9,10]. Wound care after surgical debridement includes frequent wound irrigation (every 6-8 hours) with saline and dressing changes and application of an antibacterial ointment. Once the wound is clean and granulation tissue develops, one can either perform reconstructive surgery or allow closure by

secondary intention [1,3,6,11]. Allowing these wounds to heal by secondary intention may prolong hospital stay. Silberstein et al. [8] reported benefits of VAC devices on healing process.

Urinary and fecal diversion may be needed to prevent wound contamination or to treat an underlying condition that may have caused the infection [1,5,6]. A suprapubic cystostomy is required when there is gross urinary extravasation or periurethral inflammation. To avoid urinary contamination, urinary catheterization is preferred to cystostomy, which involves a less safe route [3,12]. Colostomy has been suggested for FG, particularly for the involvement of anorectal area and sphincter and for patients with a high risk of fecal contamination to allow a better wound healing [3,12]. Nevertheless, colostomy should be performed only in selected cases, as it is not a procedure free of complications. The incidence of colostomy-related complications ranges from 28% to 74% [13]. Colostomy prolapse, bleeding, obstruction, wound infection, revision, and skin excoriation are the major complications [13]. No colostomy-related complications have developed in our 2 patients. We preferred colostomy in 2 patients; because fecal contamination was unavoidable because of the very young age of the patients.

Although their efficacy and practicableness is debated, heparin, hyperbaric oxygen, and honey have been suggested as adjuncts to treatment [1,3,8]. We did not use any of these treatments in our patients except for the one who required a flap reconstructive surgery for wound closure.

In conclusion, the key to a successful outcome in FG includes a high index of suspicion, a multidisciplinary approach, prompt diagnosis, and surgical intervention, which may be more conservative in children because of the lower mortality rate than in adults.

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