Pediatr Urol Case Rep 2019; 6(5):114-118 DOI: 10.14534/j-pucr.2019554537

PEDIATRIC UROLOGY CASE REPORTS
ISSN 2148-2969
http://www.pediatricurologycasereports.com

Fournier's gangrene with urethral involvement in a four-year-old child with neuropathic bladder: A rare case report

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ABSTRACT

A four-year-old boy presenting with neuropathic bladder and hydrocephalus developed Fournier's gangrene of the scrotum and perineum. At first, he was in septic shock and intubated. After debridement and grafting, he was taken to the normal service room from the intensive care unit. He was discharged on the 29th postoperative day. We performed bilateral orchiectomy due to testicular involvement and also repair and flap application to urethra due to its involvement. We think that our case is interesting because Fournier's gangrene is already rare in children and our case also contains atypical involvement sites such as testes and urethra.

Key Words: Fournier's gangrene, pediatric, testicular and urethral involvement, orchiectomy.

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Received 2019-05-26, Revisions 2019-08-07
Accepted 2019-08-16
Publication Date 2019-09-01

Introduction

Fournier's gangrene (FG) was first described by venereologist Alfred Fournier in 1883 [1]. It is more common in adulthood and can be fatal if it is not intervened early [2,3]. The disease is usually seen in immunosuppressive patients with diabetes, alcoholism, HIV infection and malignancy. FG spreads rapidly from the perineum and genitals to the wide area by progressing in the fascial layer [4,5]. FG is very rare in children. Testicular involvement is rare because the blood supply to the testis is independent of the affected area [6]. However, our patient underwent bilateral orchiectomy due to testicular involvement. It has been reported that the recovery is faster and mortality is lower with the intervention in pediatric patients [7]. Urethral involvement in FG is very rare and has been reported in only two cases so far [8,9]. In our case, the urethra was involved and repaired before the graft procedure. We report the early results of debridement after FG in a four-year-old child who was operated for meningomyelocele after birth and has a neuropathic bladder.

Case report

A four-year-old patient with operated meningomyelocele and hydrocephalus was

admitted to the emergency department with complaints of fever, vomiting, diarrhea, necrosis in the scrotum and lower abdomen. The patient's blood pressure was very low, circulation was distorted and superficial spontaneous breathing was observed. He was intubated and admitted to the intensive care unit upon deterioration of his general status. The patient whose blood pressure could not be measured was loaded 1 time with saline from 20 cc / kg. Dopamine and Adrenaline infusion was started in the patient with low blood pressure. Laboratory tests showed a white blood cell count of 26.800/mm^3 (normal, 6000–12,500/mm^3) and a C-reactive protein of 113 mg/l (normal, < 6 mg/l). Combination of vancomycin, meropenem, amikacin, ambisome, clindamycin, and metronidazole was given in antibiotherapy. Patient underwent surgery after supportive treatment. Lower abdomen, genital area and perineum were completely involved (Fig. 1).

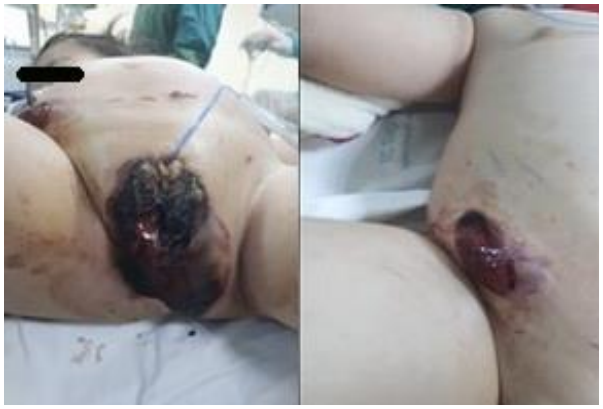

Fig. 1. In the first examination, all scrotum and perineum was involved and there was spontaneous opening in the lower abdomen.

In the rectal examination, it was observed that the anal sphincter and rectum intact. The stool consistency was normal and therefore we did not need colostomy opening. First, all necrotic tissues in the scrotum and perineum were excised until healthy subcutaneous tissue was observed. Necrosis was observed in the testes and so bilateral orchiectomy was performed (Fig. 2). It was observed that the mid-urethra was opened and the urethral catheter was seen (Fig. 3). We decided to repair it just before the grafting. Due to the observation that necrosis spreads from both inguinal canals and spread to the lower abdomen, inguinal region was entered on both sides and excision was performed until muscle tissues were observed.

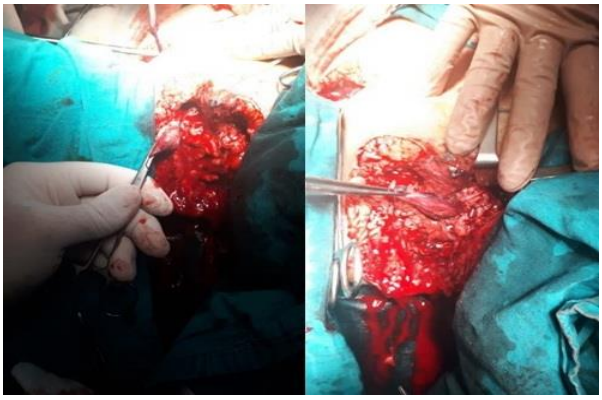

Fig. 2. Testicular involvement and bilateral orchiectomy.

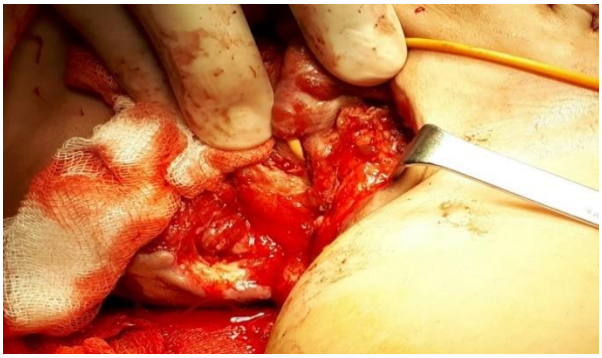

Fig. 3. Urethral involvement.

All necrosis was cleared (Fig. 4a). After surgery, daily dressings were performed. C reactive protein (CRP) and procalcitonin levels were significantly reduced. The tissue culture confirmed Proteus Mirabilis and his antibiotics were continued for 14 days. On the 7th day of hospitalization, inotropic support was stopped by decreasing. On the 8th day of hospitalization, the patient extubated himself.

On the 12th day of his hospitalization, the patient was started on oral feeding and the oxygen supply given by the nasal cannula was discontinued. On the 14th postoperative day the wound was completely cleared and the patient was operated again (Fig. 4b).

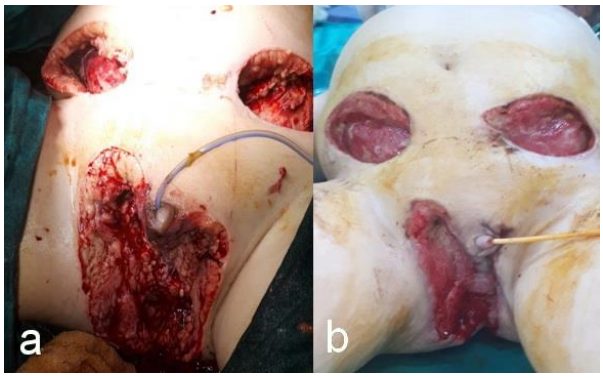

Fig. 4. a) After the first debridement, muscle fibers visible. **b)** Before Grafting, all necrotic tissues were cleaned.

The urethral region 7/0 PDS was sutured after all remaining residual necrotic areas were cleared and was closed with flap from surrounding granulated tissues. Wide skin grafts were removed from the thigh front with two parts. It was sutured to the region of the skin defects of the scrotum and perineum (Fig. 5a, b). The open areas in the abdomen were closed primary.

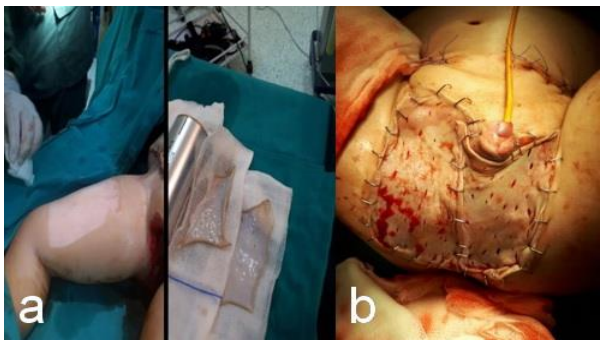

Fig. 5. **a)** Two large pieces graft from thigh front. **b)** Grafting and primary suturization.

On the 6th day of the graft application, it was observed that the graft was alive and abdominal wounds were also healed (Fig. 6 a, b).

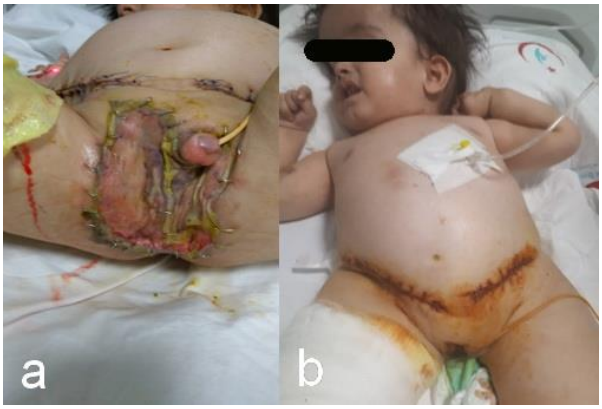

Fig. 6. a) On the 6th day of the graft application, the graft was alive. **b)** Abdominal wounds were healed and the child was extubated.

Discussion

Fournier's gangrene is an extremely rare condition in pediatric patients and may be mortal if not treated early. FG begins with focal skin infection and spreads along the facial plane, which results in inflammation, ischemia and necrosis. In adulthood, FG usually occurs due to underlying predisposing conditions such as diabetes mellitus, alcohol dependence, immunosuppressive therapy, long-term steroid therapy, malnutrition and HIV. However, local traumas, insect bites, burns, systemic infections, previous genital surgery and paraphimosis may be the causative agents in children [1-4]. The sources of FG-causing infections are usually urogenital (45%), anorectal (33%) or cutaneous (21%) [4]. Our case was a patient with myelomeningocele and bed dependent. In the history of patient, firstly, the erythema of the scrotum started and then progressed rapidly to other areas. It was a FG that developed after a possible epididymoorchitis. In additionally, our biggest disadvantage in evaluating this patient was that we did not know his past. The

development of scrotal and perineal gangrene in a bed-dependent child was most likely secondary to a urinary tract infection. However, this was the first time the patient presented to us and we do not know the history. From the information received from the family, we learned that scrotal erythema developed a few days ago and was given standard antibiotic treatment, but quickly went to necrosis. The family lived in a village very far from the city center and therefore, may have been delayed to admit. Multiple infectious agents such as coliforms, klebsiella, streptococci, staphylococci, clostridia, bacteroids, and corynbacteria may be responsible for the etiology [4-8], but only Proteus Mirabilis produced in the debridement tissue samples of our case.

Fournier's gangrene is a surgical emergency that requires rapid recognition and multimodal treatment, including resuscitation, broad-spectrum antibiotics and early surgical debridement. All necrotic tissues should be excised, as applied in our case [1-8]. In addition, testicular involvement is rare due to the the blood supply to testis is different, but we performed bilateral orchiectomy due to the involvement and pathological evaluation was in favor of necrosis. In endocrinological evaluation, it was stated that there was no need for early hormone replacement and it could be started in prepubertal period. The urethral involvement is very rare due to the protection of the Buck fascia [9,10]. However, in our case, the mid-urethra had involvement exist and was sutured before the graft and covered with flap.

Conclusion

We think that our case is interesting because FG which is already rare in children and our case also contains atypical involvement sites such as testes and urethra. In addition, as in the current case, Fournier's gangrene has high mortality and morbidity in children. Therefore, early diagnosis and surgical debridement are crucial for the success of treatment.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: All photos were taken with parental consent.

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