



Fournier's gangrene in an infant: Is primary surgical repair a better alternative for management? A case report

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Abstract Fournier's gangrene is an uncommon form of necrotizing fasciitis in children. It needs prompt diagnosis and treatment to avoid the high morbidity and mortality. In the present study we are reporting a 3 months old infant with Fournier's gangrene predisposed by diaper rash due to poor hygiene. The infant was treated by debridement and primary repair with a good outcome.

Keywords Fournier's gangrene; necrotizing fasciitis; infant; diaper rash; primary repair.

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INTRODUCTION

Fournier's gangrene (FG) is defined as an infective necrotizing fasciitis, which affects perianal and genital regions, leading to severe skin necrosis and thrombosis of subcutaneous vasculature [1,2]. It is a rare, but life-threatening process with high

morbidity and mortality [1,2]. It produces extensive cellulitis with severe involvement of the subcutaneous tissue, fascia, and muscles or both, resulting in extensive necrosis [1,3]. FG, initially described as a disease of young males, is now known to affect both sexes and all ages [4]. It represents a very rare clinical entity in childhood, and the majority of cases have been described in infants [5]. The incidence of necrotizing fasciitis of various anatomic localizations, including the perineal, has

been reported to be 0.08/100.000 in children in comparison to 0.4/100.000 in adults [5]. In the present study, we are reporting a 3 months old infant with Fournier's gangrene managed successfully.

CASE REPORT

A 3 months old infant presented with fever, diaper rash in the groin with redness of the anterior abdominal wall and scrotal wall, irritability, dehydration, lethargy with poor feeding. Fluid resuscitation was done, blood investigations showed TLC of $18000/\text{mm}^3$, hemoglobin of 11 gm/dl, platelet count of $170000/\text{mm}^3$, serum urea of 38 mg/dL, serum creatinine of 0.8 mg/dL, serum sodium of 137 mEq/L, and serum potassium of 4.7 mEq/L. Ultrasonography showed normal intra-abdominal findings, edema in the lower anterior abdominal wall, normal blood flow to the testes. Wound swab culture showed *Escherichia coli* but the blood culture did not grow any organism. He was started on piperacilin with tazobactam, amikacin and metronidazole. The erythema of the anterior abdominal wall and groin rapidly progressed to a gangrenous patch in the scrotal wall within 2 days (Fig. 1). The erythema and cellulitis of the anterior abdominal wall subsided on antibiotics with development of line of demarcation of gangrenous patch in the scrotal wall (Fig. 2).



Fig. 1. Early stage of Fournier's gangrene showing scrotal wall cellulitis and necrosis along with abdominal wall erythema.



Fig. 2. Fournier's gangrene showing gangrenous scrotal wall.

Surgical debridement was done under general anesthesia, till the healthy margin was reached (Fig. 3) and wound was repaired primarily mobilizing the lateral edges covering the testes which were exposed after the debridement (Fig. 4).



Fig. 3. Intra-operative photo after excising the gangrenous scrotal wall, note the presence of necrotic tissue which needs to be excised till the healthy margin.



Fig. 4. Primary repair after debridement.

Foley catheter was indwelled for a period of 5 days to avoid soiling of the wound. The wound was healed except for minor gaps of about 1 cm and 0.5 cm (Fig. 5).



Fig. 5. Minor gapping of scrotal wound.

Patient was discharged on the 7th post operative day after a total of 14 days in the hospital. Histopathology of debrided tissues showed necrotic changes in the fascia with acute inflammatory cellular infiltration and thrombosis of the nutrient arterioles. In the follow up, the gaps have healed only on local antibiotic ointment.

DISCUSSION

Even though, Baurienne in 1764 and Avicenna in 1877 had described the same disease earlier, this clinical entity is credited to the Parisian venerologist Jean-Alfred Fournier, who described it as a fulminant gangrene of the penis and scrotum in young men [1,6,7].

FG was defined as an idiopathic entity, but vast majority of cases have a source of infection, as either perineal or genital skin

infections [4]. The source of infection may be urogenital (45%), anorectal (33%), or cutaneous (21%) [8]. Although it is not possible to grow microorganisms in every patient, *Escherichia coli*, *Bacterioides*, *Streptococcus*, *Peptostreptococcus*, and *Clostridium spp* are frequently identified as causative microorganisms in this often polymicrobial infection [1,2,3]. The polymicrobial nature is needed to create the synergy of enzyme production that promotes rapid multiplication and spread of the infection [9]. The suppurative bacterial infection results in microthrombosis of the small subcutaneous vessels leading to the development of gangrene of the overlying skin [1].

Most pediatric cases of FG have been reported in previously healthy children, who may appear well in the early course of the disease and, thus, may escape diagnosis [10,11]. The predisposing factors as mentioned in the literature are diarrhea, use of NSAIDs, post varicella infection, cutaneous HPV lesions, diaper rash, prematurity, circumcision, omphalitis, strangulated hernia, testicular torsion, trauma, insect bites, burns, periurethral and anorectal diseases/infections, needle puncture of the hydrocele, systemic infections, foley catheterization, immune-compromised states, and hematologic

malignancies [2,3,5,8,11-18]. In the present case the predisposing factor was possibly diaper rash due to poor hygiene.

Most of pediatric FG usually presents below the age of 3 months [4,11]. Patients may present with fever, malaise, and nonspecific symptoms of infection without any complaints from the perineal area, although typically in the early stages the clinical hallmark of FG is severe local pain, disproportionate with physical findings which is abrupt onset, rapidly spreading edema and hyperemia of perineal skin [1,8,10]. The infection commonly starts as a cellulitis adjacent to the portal of entry, commonly in the perineum or perianal region. The patient also has pronounced systemic signs; usually out of proportion to the local extent of the disease. As the subcutaneous inflammation worsens, necrotic patches start appearing over the overlying skin and progress to extensive necrosis [1]. In the present case, the erythema progressed to gangrene in a span of 2 days.

Unless aggressively treated, the patient can rapidly progress to sepsis with multiple organ failure. The spread of infection is along the facial planes and is usually limited by the attachment of the Colles' fascia in the perineum. Infection can spread to involve the scrotum and penis, and can spread up the

anterior abdominal wall as in the present case [1]. Since the main predisposing factors of the FG are due to infections in the perineal and genital region, maintaining good hygiene and proper aseptic and antiseptic measures with judicious use of antibiotics during genital and perineal surgeries will help to prevent most cases. The main components of aggressive management of FG should be started as soon as possible, which includes 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 debrided aggressively, for necrosis may progress rapidly in hours. Antimicrobial treatment with broad-spectrum antibiotic combinations should be initiated before surgery, to be tailored according to culture report [1].

The goal of surgical debridement is to remove all non-viable tissue, halt progression of infection and alleviate systemic toxicity [4]. Attah recommended excision so radical as to permit primary closure of the resulting wound [19]. But it did not gain popularity because of the infiltrating nature of necrotizing fasciitis; multiple surgical debridements are paramount to remove any nidus of remaining infection [4]. Multiple

debridements are the rule rather than the exception, an average of 2 (1 to 5) debridement steps have been reported [20]. But many authors have described FG in children to be more benign with a more favorable outcome compare to adults [11,21]. Adeyokunnu reported successful medical treatment of the gangrenous area in 6 infants, and the children survived with no appreciable scrotal skin loss or adhesions [17]. Adams et al. [11] analyzed the data of 55 previously reported cases of pediatric FG, they have been successfully managed with a more conservative surgical approach and have had a significantly lower mortality rate than adult cases.

These findings of previous studies along with well defined line of demarcation of scrotal gangrene prompted us to decide on the debridement and primary repair of the defect. In our study, the early initiation of broad spectrum antibiotic along with the more benign course of infantile Fournier's gangrene allowed us to delay the debridement till the healthy demarcation of the gangrenous scrotal wall favoring debridement with primary repair. The wound was debrided along with primary repair without significant complication except for minor wound gapping, which shorten the hospital stay significantly. In the extensive search of literature, wound debridement with

primary repair of the defect has not been attempted in children. Most of the publications showed from single to multiple debridements with secondary repair after healthy granulation tissue appeared, secondary healing by granulation, reconstruction with flaps etc. [1,4].

Once the wound is clean with healthy granulation tissue after the debridement, one can either perform reconstructive surgery or allow closure by secondary intention [1,4]. The potential benefits of VAC devices on healing process has been described by many authors [4,15]. Urinary and fecal diversion may be needed to prevent wound contamination [1,15,20]. Although their efficacy is still debated, heparin, hyperbaric oxygen, and honey have been suggested as adjuncts to treatment [1,4].

The exact figures of mortality 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 [8,13,20] against the adult mortality of 3 to 45 per cent [4].

Conclusion

Even though fulminant, Fournier's gangrene in infants can be treated in a more conservative approach with early initiation of antibiotics and meticulous wound debridement to decrease the high morbidity and mortality. Debridement with primary repair is a good option for treatment of the infantile Fournier's gangrene with significant shortening of recovery period.

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