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Necrotizing fasciitis following routine genitourinary surgery in healthy infants

Harry H. Lee, BA¹, Ted Lee², Richard S Lee², Joseph G. Borer²

¹Georgetown University School of Medicine, 3900 Reservoir Rd NW, Washington, DC 20007

²Boston Children's Hospital, Department of Urology, 300 Longwood Avenue, Boston, MA 02115

Abstract

Morbidity and mortality associated with pediatric necrotizing fasciitis are strongly dependent on early diagnosis and timely intervention. Yet, the lack of early cutaneous findings and nonspecific symptoms may result in initial delayed diagnosis or misdiagnosis. Infants may be particularly at risk of missed or delayed diagnosis due to inherent barriers in communication and rarity of the condition, especially among healthy patients. We describe two cases of necrotizing fasciitis following routine genitourinary surgery in healthy infants.

Keywords

pediatric urology; necrotizing fasciitis

Introduction

Necrotizing fasciitis (NF) is characterized by soft tissue necrosis secondary to rapid bacterial spread.[1, 2] Symptoms may include systemic signs of toxicity as well as more localized symptoms, including skin erythema and blistering, tense edema, pain disproportionate to exam, and crepitus on palpation.[3] NF in the genital area is also referred to as Fournier's gangrene.

Without early diagnosis and intervention, NF is associated with substantial mortality rates in the pediatric population. However, timely diagnosis may be complicated by lack of early cutaneous findings and nonspecific symptoms.[2] Infants are particularly at risk of delayed diagnosis due to inherent barriers in communication and rarity of the condition, especially among healthy patients.[4]

Corresponding Author: Ted Lee, Department of Urology, 300 Longwood Avenue, Boston, MA 02115, ted.lee@childrens.harvard.edu, 617.355.6000.

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Herein, we present two cases of NF following routine genitourinary surgery in healthy infants. Low level of suspicion and nonspecific presentation may have resulted in initial misdiagnosis in one patient.

Case Presentations

Patient 1

Post-operative day 5 following left inguinal hernia repair and distal hypospadias repair via meatal advancement and glanuloplasty, an 8-month-old male presented with significant edema and erythema surrounding the groin incision site. Given concern for incarcerated left inguinal hernia, the patient was taken emergently for inguinal exploration. Exploration was extremely challenging due to significant erythema, hyperemia, and induration within the prior surgical site. What was thought to be incarcerated omentum was reduced. The hernia was revised prior to closure and the patient received intravenous (IV) cefazolin post-operatively.

The following morning, exam revealed rapidly spreading erythema, tenderness, and edema of the inguinal, scrotal, and perineal areas. The patient exhibited systemic toxicity, including a fever of 39.6°C, tachycardia, and leukocytosis (18,200 WBC/ μ l). Crepitus was palpable surrounding the incision site. The patient was taken to the operating room emergently for re-exploration, primarily to assess for possible necrosis and obtain tissue sample for diagnosis and culture. A small area of necrotic tissue was noted near the inguinal incision, and surgical debridement of the inguinal and scrotal regions was performed. A full-thickness supra-fascial (1 $\times$ 3cm biopsy; skin and fat, to level of external oblique fascia) tissue biopsy was taken in addition to tissue culture studies. Bilateral scrotal full thickness tissue samples were also taken.

Histopathology revealed tissue necrosis involving fascial layers and tissue cultures positive for *Peptostreptococcus*. Infectious disease expertise recommended against wound closure. Planned re-examination and dressing change in the operating room the next day revealed significant improvements (Figure 1). With moist-dry saline gauze dressing changes, the patient's post-debridement wounds healed by secondary intention. The patient was discharged in stable condition following an elongated course of antibiotics.

Patient 2

Post-operative day 8 following penoscrotal hypospadias repair via tubularized incised plate approach and ventral dorsal plication for correction of chordee, an 8-month-old male presented with pale skin, tense edema, and erythema of the left hemiscrotum and ventral penile shaft extending to the left lower abdomen. The patient was seen the day prior for urethral stent removal, at which time exam was consistent with expected post-operative swelling and healing. Notably, the patient was voiding with a straight stream following stent removal.

Due to the severity of the exam at presentation on post-operative day 8, and concern for NF, infectious disease expertise was sought and empiric broad-spectrum IV antibiotics were started. Upon admission, the patient exhibited low-grade fevers, mild tachycardia, and

leukocytosis (15,960 WBC/ μ l). Despite antibiotics, skin erythema, swelling and induration rapidly progressed to the lower abdomen and scrotum within the first 8 hours following onset of symptoms (Figure 2). A lower abdominal ultrasound showed severe cellulitis and focal areas of phlegmon, but subcutaneous air was not visualized. Given the rapidly worsening exam findings, the decision for urgent exploration and tissue biopsy in the operating room was reached following discussion with our infectious disease and pathology colleagues.

Our infectious disease colleagues emphasized the importance of a full thickness, 1 $\times$ 3cm tissue biopsy during surgical exploration. Intraoperatively, incisions were made per recommended tissue biopsy size over the left inguinal region. Skin and subcutaneous tissue was obtained down to the level of the external oblique fascia. Fascia appeared intact and grossly unaffected at the external oblique fascial plane. The wound was irrigated with antimicrobial solution. A deep tissue biopsy was obtained and sent for histopathologic evaluation and anaerobic and aerobic culture. The wound was subsequently packed with Iodoform gauze. Suprapubic cystostomy tube was decided against due to risk of potentially promoting sub-fascial spread. A urethral catheter was carefully placed in order to prevent any potential contamination from urine or urine leakage per repair.

Histopathology displayed subcutaneous necrosis with preservation of external oblique fascial planes. Tissue cultures grew extended-spectrum beta-lactamase-positive *E. coli*, which included resistance to trimethoprim-sulfamethoxazole, which was originally prescribed as prophylaxis, but sensitive to piperacillin-tazobactam, which was given IV intraoperatively. Post-operatively, patient's symptoms improved, and he was discharged after 14 days of tailored IV antibiotics. The wound healed successfully by secondary intention.

Discussion

Early diagnosis and subsequent treatment are critical for improved outcomes following NF. However, nonspecific symptoms and lack of early cutaneous findings leads to challenges in timely diagnosis. These challenges are exacerbated in healthy children due to the rarity of the condition.[1]

Diagnosis of NF requires a high index of suspicion and the incorporation of various diagnostic tools such as laboratory findings, imaging studies, and pathology. In comparing necrotizing and non-necrotizing soft-tissue infections, studies have found the presence of elevated white blood cell count, elevated C-reactive protein, creatinine, and glucose, and decreased hemoglobin and sodium levels to be associated with NF.[5, 6] Diagnostic studies such as plain x-rays, ultrasound, CT scans, and MRI scans, can help identify subcutaneous gas, a non-sensitive but very specific finding.[1] Although acquiring bedside biopsies to facilitate early diagnosis have also been recommended in the past [1, 3], this may delay diagnosis and surgical intervention.[1] Additionally, a bedside biopsy would be inadequate with regard to the size of the tissue biopsy needed to meet the standard recommended by our infectious disease colleagues for our two patients. It also would not permit wound debridement if potentially needed. With regards to microbiology, past studies have found that there are no bacterial species that are diagnostic for NF.[1, 7] The procurement of tissue

for histopathology and culture, at biopsy, was critical for the management of our second patient, who was infected with a highly resistant organism. Without this knowledge and subsequent adjustment of antibiotic treatment, the clinical course of this patient may have stagnated.

Proper management entails a multifaceted approach that incorporates surgical debridement, antimicrobial therapy, and physiologic support.[1] We emphasize the importance of full thickness tissue biopsy and cultures, which substantially aided in our management of both patients by allowing us to tailor antimicrobial medications postoperatively.

Conclusion

Necrotizing fasciitis may be easily misdiagnosed due to nonspecific early symptoms and rarity among children. However, NF should remain in the list of differential diagnoses during postoperative evaluation given its significant morbidity and lethal potential if diagnosis is missed or delayed. Upon suspicion of NF, timely surgical exploration should be performed, in addition to supportive care and administration of broad-spectrum empiric antibiotics. Deep tissue biopsy and wound and tissue cultures are essential components of the surgical exploration that must not be forgotten. The mortality and morbidity associated with these infections can be decreased with clinical awareness, early diagnosis, and adequate and urgent surgical biopsy and debridement.

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Figure 1. One day following surgical debridement and deep tissue biopsies for *patient 1*. Blue markings outline the extent of swelling and edema prior to treatment.

Figure 2.
Preoperative exam for patient 2