Multifocal necrotizing fasciitis following Hirshsprung's disease surgery away from the surgical wound site

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Necrotizing fasciitis (NF) is a life-threatening infection with rapidly progressive necrosis. Escherichia coli is rarely reported as causative agent of type 2 NF. NF typically arises in a single area usually secondary to penetrating injury. NF was only reported as a postoperative complication of Hirshsprung's disease in one report, where the causative agent was Pseudomonas aeruginosa. We here present a case of synchronous multifocal NF in a 7-month Hirshsprung's disease patient after abdominal Soave procedure. The patient presented with constipation and had a colosotomy undergone at the splenic flexure through a left transverse supraumbilical incision during neonatal period. At the age of 7 months and after preoperative preparation, abdominal Soave procedure was performed through a Pfannestiel incision. Few days following operation he had fever, tachycardia, and pain. Local examination showed red edematous areas at both flanks away from the wounds. Shortly afterwards skin became dark, tense, and started to slough. At this stage the clinical diagnosis of NF was raised and surgical

debridement was done. Blood and tissue cultures were positive for *E. coli.* Five days later the patient had another debridement due to necrotic wound edges. Vacuum assisted closure therapy with Aquacel Ag dressings was fitted and changed every 48 h. Eighteen days later split thickness skin-graft was carried out. The patient was discharged home; 10 days later he is doing fine on followup. *Ann Pediatr Surg* 11:159–161 © 2015 Annals of Pediatric Surgery.

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Introduction

Necrotizing fasciitis (NF) is a life-threatening infection, characterized by rapidly progressing necrosis involving the subcutaneous tissue and fascia [1]. NF can be classified into type I and type II on the basis of the causative agent [2]. Type I infections represent 70% of

cases and are polymicrobial, including gram-positive cocci, gram-negative rods, and anaerobes. Type 2 infections are monomicrobial, with group A *Streptococci* spp. and *Staphylococcus aureus* being the primary pathogens reported [3]. *Escherichia coli* is rarely reported as a causative agent of type 2 NF [2]. NF typically arises in

Fig. 1



Patient on fourth postoperative day. Note the lesions on the flanks separated from the surgical wounds by healthy skin.

a single area usually secondary to a penetrating injury [4]. Multifocal NF in which there is more than one separate area of necrosis is much less common [4]. NF is rarely reported in infants [5].

Hirshsprung's disease (HSD) is a common cause of neonatal intestinal obstruction requiring operative intervention during infancy [6]. NF was not reported as a postoperative complication of HSD, except in one report in which the causative agent was *Pseudomonas aeruginosa* [7]. Herein, we present a case of synchronous multifocal NF occurring in a 7-month-old HSD patient after undergoing abdominal Soave procedure.

Case presentation

The patient was presented to us during the neonatal period with constipation and was diagnosed after rectal suction biopsy. At the age of 17 days abdominal exploration was carried out through a left transverse supraumbilical incision. Multiple biopsies were taken, and a colosotomy was performed at the splenic flexure. The patient had no associated comorbidities. At the age of 7 months he was admitted for definitive surgery. Following 2 days of clear fluids, followed by 1 day of nil per mouth and rectal washouts through the anus and colostomy, antibiotics

Fig. 2



Patient in the operation room during the debridement. Note lack of bleeding.



(a) VAC device. (b, c) Patient 6 months after surgery. VAC, vacuum assisted closure.

(cefuroxime and metronidazole) were administered intravenously. Abdominal Soave procedure was performed through a muscle splitting Pfannestiel incision. The proximal end of the stoma was pulled through the anus and the anastomosis was performed. The procedure went smoothly, but during the first few postoperative days the patient had spikes of fever, tachycardia, and pain; however, his white blood cell count was normal. Local abdominal examination showed red edematous areas at both flanks away from the wounds. Local signs rapidly deteriorated, and skin became dark, tense, and started to slough (Fig. 1). The patient was septic with multiorgan system failure. At this stage the clinical diagnosis of NF was raised. The patient was intubated and put on inotrops. Antibiotics were changed to intravenous gentamycin, meropenem, and vancomycin. The patient was taken to the operation room and surgical debridement was performed (Fig. 2). Blood and tissue cultures were positive for extended spectrum B-lactamase-producing E. coli. Histopathology revealed acute and chronic fasciitis with ischemic changes. Five days later the patient had another debridement due to necrotic wound edges. Vacuum assisted closure (VAC) therapy with Aquacel Ag dressings (ConvaTec Bristol-Myers Squibb, New York, USA) was carried out to provide a negative wound pressure of 50 mmHg. It was changed

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Fig. 3

every 48–72 h depending upon need. There was a marvelous improvement, and healthy granulation tissue appeared. Eighteen days later and after three negative wound cultures were obtained, split thickness skin graft was performed. The patient was discharged home 10 days after grafting. He is on regular follow-up in the outpatient clinic and doing fine (Fig. 3).

Discussion

NF is a rare and severe infection with a poor outcome. NF more commonly affects the trunk and extremities [8]. Type 2 infections are monomicrobial and associated with multiorgan failure. Group A *Streptococci* spp. and *S. aureus* are the primary pathogens reported [9]. The predisposing factors in type 2 are burns, trauma, and surgery [3]. In our case, *E. coli* was isolated in wound and blood cultures.

The diagnosis of NF is essentially made on clinical findings and needs a high index of suspicion. NF may be misdiagnosed as cellulitis, especially in children [2]. Severe wound pain not correlating with skin findings, septic shock, and multiorgan failure are usually associated clues that suggest NF, as in our case. Early diagnosis, broad-spectrum antibiotics including gram-positive, gram-negative aerobes and anaerobes, and widespread debridement of devitalized tissues are the cornerstones in the management of this potentially fatal disease [10]. Some authors also advocate the use of adjunctive therapies such as hyperbaric oxygen and intravenous immunoglobin [11]. VAC is a relatively new technique that consists of the application of a sterile, open-cell foam sponge onto the wound, in combination with a negative external pressure. Although VAC was well described in adults, it is still rarely used in children [12]. VAC improves granulation tissue formation, reduces the surface area of the wound, and reduces bacterial counts by enhancing microcirculation [13]. VAC device removes excess exudates and reduces local edema [14]. VAC therapy requires less number of dressing changes compared

with the traditional dressings. It increases patient comfort and decreases pain.

Acknowledgements Conflicts of interest

There are no conflicts of interest.

References

- 1 Wilson B. Necrotizing fasciitis. Am Surg 1952; 18:416-431.
- 2 Swartz MN, Pasternack MS. Cellulitis and subcutaneous tissue infections. In: Mandell GL, Bennett JE, Dolin R, editors. *Principles and practice of infectious diseases*, 6th ed. New York: Churchill Livingstone; 2005. pp. 1172–1194.
- 3 Sarani B, Strong M, Pascual J, Schwab CW. Necrotizing fasciitis: current concepts and review of the literature. J Am Coll Surg 2009; 208:279–288.
- 4 El-Khani U, Nehme J, Darwish A, Jamnadas-Khoda B, Scerri G, Heppell S, Bennett N. Multifocal necrotising fasciitis: an overlooked entity? *J Plast Reconstr Aesthet Surg* 2012; 65:501–512.
- 5 Hsieh WS, Yang PH, Chao HC, Lai JY. Neonatal necrotizing fasciitis: a report of three cases and review of the literature. *Pediatrics* 1999; **103**:e53.
- 6 Wang G, Sun XY, Wei MF, Weng YZ. Heart-shaped anastomosis for Hirschsprung's disease: operative technique and long-term follow-up. *World J Gastroenterol* 2005; 11:296–298.
- 7 Karakus SC, Kilincaslan H, Koku N, Parmaksiz ME. Necrotizing fasciitis following Soave procedure in Hirschsprung disease. *Eur J Pediatr Surg* 2014; 24:190–192.
- 8 Darmstadt G. Subcutaneous tissue infections and abscesses. In: Long S, Pickering L, Prober C, editors. *Principles and practice of pediatric infectious diseases*, 2nd ed. New York: Churchill Livingstone; 2003. pp. 449–457.
- 9 Abass K, Saad H, Abd-Elsayed AA. Necrotizing fasciitis with toxic shock syndrome in a child: a case report and review of literature. *Cases J* 2008; 1:228.
- 10 Sakata S, Das Gupta R, Leditschke JF, Kimble RM. Extensive necrotising fasciitis in a 4-day-old neonate: a successful outcome from modern dressings, intensive care and early surgical intervention. *Pediatr Surg Int* 2009; 25:117–119.
- 11 Young MH, Aronoff DM, Engleberg NC. Necrotizing fasciitis: pathogenesis and treatment. Expert Rev Anti Infect Ther 2005; 3:279–294.
- 12 Bütter A, Emran M, Al-Jazaeri A, Ouimet A. Vacuum-assisted closure for wound management in the pediatric population. *J Pediatr Surg* 2006; 41:940–942.
- 13 Mouës CM, van den Bemd GJ, Heule F, Hovius SE. Comparing conventional gauze therapy to vacuum-assisted closure wound therapy: a prospective randomised trial. J Plast Reconstr Aesthet Surg 2007; 60:672–681.
- 14 Schaffzin DM, Douglas JM, Stahl TJ, Smith LE. Vacuum-assisted closure of complex perineal wounds. *Dis Colon Rectum* 2004; 47:1745–1748.