Necrotizing fasciitis due to late-onset group B streptococcal bacteremia in a 2-month-old girl

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Necrotizing fasciitis (NF) is a severe soft tissue infection, characterized by rapid and fulminant progression. Thus, early suspicion, and prompt medical and aggressive surgical management are important for the clinical outcomes of NF. Despite the rarity of NF in infants and group B streptococcus as its cause, a 2-month-old preterm girl presented with NF involving the suprapubic, inguinal, and lower lateral abdominal regions caused by group B streptococcus. The girl recovered after early medical and surgical management.

Key words: Fasciitis, Necrotizing; Infant; Neonatal Sepsis; Soft Tissue Infections; Streptococcus agalactiae

Introduction

Necrotizing fasciitis (NF) is the most severe form of soft tissue infection, characterized by rapidly spreading inflammation and subsequent necrosis of the superficial fascia and subcutaneous tissue[1]. The annual incidence of NF is 0.08 cases per 100,000 children[2]. Despite the rarity, it has mortality rates of 59.1% in neonates and 25% in children[3,4]. While the common pathogens of NF in adults include group A streptococcus (GAS), Enterobacteriaceae, and non-group A streptococci[5], in pediatric NF, GAS is the most common pathogen, followed by *Staphylococcus epidermidis*, *Pseudomonas aeruginosa*, *Escherichia coli*, and others[2,6]. *Staphylococcus aureus*, GAS, and group B streptococcus (GBS) usually cause NF in neonates and infants[1,6,7].
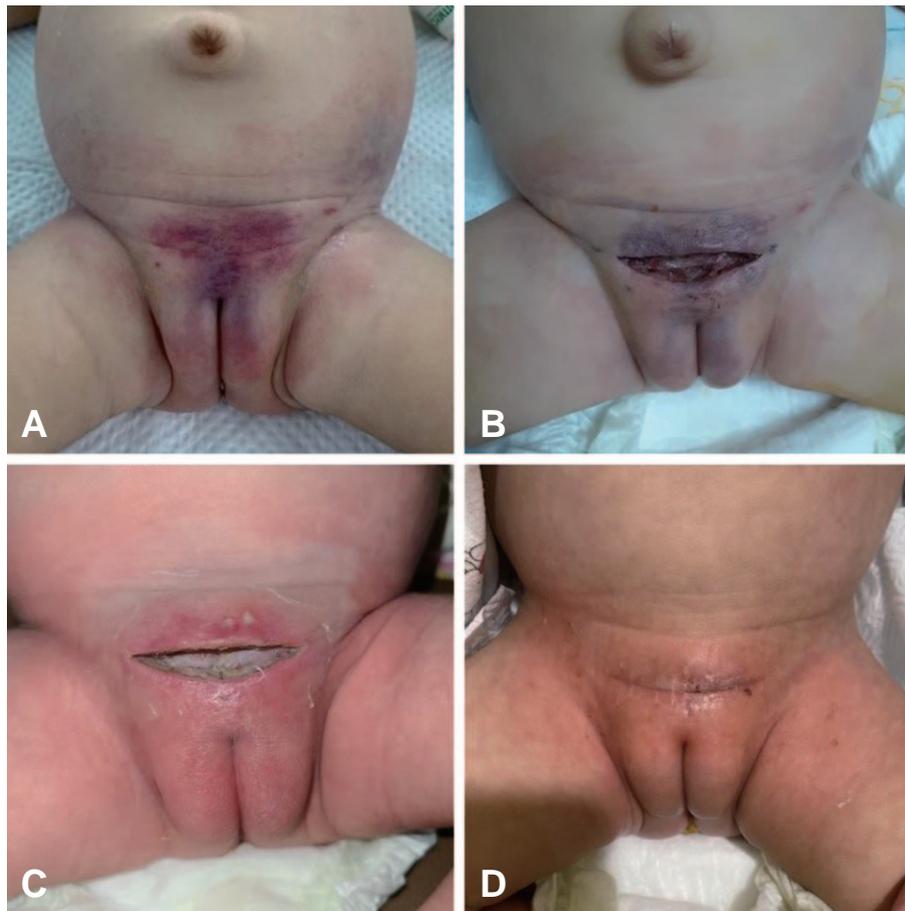
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Here we describe an infant with GBS NF who was treated with early medical and surgical management and successfully recovered without skin complications. This study was approved by the institutional review board of the Keimyung University Dongsan Medical Center with a waiver for informed consent (IRB no. 2022-08-053).

Case

A 2-month-old girl visited the emergency department with 1-day fever and irritability. The infant was alert but ill-looking with initial vital signs as follows: blood pressure, 80/50 mmHg; heart rate, 164 beats/min; respiratory rate, 32 breaths/min; temperature, 39.4°C; and oxyhemoglobin saturation, 99%. Four hours before the visit, she suddenly developed swelling, redness, firmness, and tenderness with petechiae in the suprapubic area. Approximately 2 hours after the visit, the skin lesion had rapidly spread to the both inguinal, lower lateral abdomen, and upper thighs with bluish discoloration and ecchymosis, without bulla, crepitus or foul odor (Fig. 1). She became lethargic with mild pallor, and had otherwise normal findings of physical examination.

Fig. 1. The skin lesion before operation (**A**), on day 2 (postoperative day 1) (**B**), on day 6 (**C**), and on day 16 (**D**).
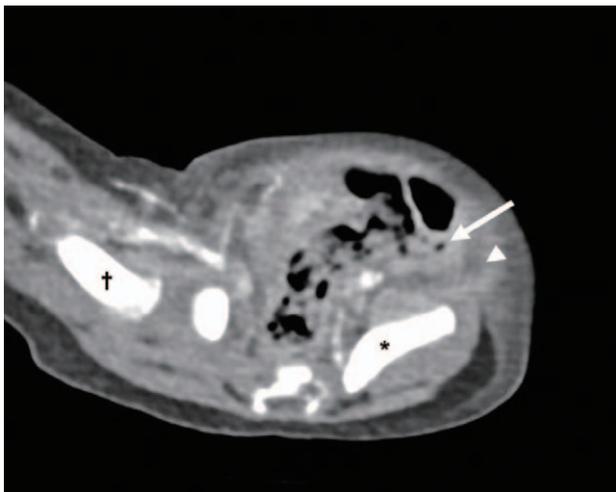
Fig. 2. Computed tomography scan showing a tiny free air collection in the left pelvic cavity (arrow). Of note, diffuse soft tissue edema and necrosis (arrowhead) are shown from the pelvic cavity to both inguinal regions.

* Left ilium.

† right femur.

The infant was born through emergency cesarean section due to maternal preterm labor with 18 hours of premature rupture of membranes (PROM), at the postconceptional age of 30 4/7 weeks, with the birth weight of 1,580 g. She did not undergo skin trauma or invasive procedures, such as endotracheal intubation or umbilical venous catheterization, in the neonatal intensive care unit (ICU). Her mother's laboratory tests showed the white blood cell (WBC) count of 20,830/μL with an absolute neutrophil count of 16,570/μL, and C−reactive protein concentration of 0.4 mg/L. Despite the absence of fever, the mother was treated with prophylactic antibiotics because of prolonged PROM. After the delivery, pathology of the mother's placenta showed chorioamnionitis, with GBS and *Enterococcus faecalis* colonization of the maternal cervix.

Abdominal ultrasonography and computed tomog−

raphy showed diffuse soft tissue edema with tiny movable air foci from the left pelvic cavity to both inguinal regions and an edematous lower lateral abdominal wall along both inguinal ligaments (Fig. 2). These findings were consistent with NF. Findings of inflammatory markers were as follows: WBCs, 1,760/μL (absolute neutrophil counts, 720/μL); C-reactive protein, 11.0 mg/L; and procalcitonin, 47.1 ng/mL (reference value, 0.02-0.5 ng/mL). Cerebrospinal fluid (CSF) study showed the yellowish clear fluid with a WBC count of < 5 cells/mm^3, protein concentration of 100.0 mg/dL, and glucose concentration of 50 mg/dL.

She was treated with vancomycin (60 mg/kg/day) and piperacillin-tazobactam (320 mg/kg/day) with intravenous immunoglobulin (0.5 g/kg/day), and underwent emergency surgical debridement under general anesthesia on day 1. The surgical finding with the timing of surgery showed the grayish subcutaneous tissue with a small amount of serosanguineous discharge, consistent with NF. However, biopsy and culture of the excised tissue were not performed.

Blood and CSF cultures were positive for GBS, which was sensitive to benzathine penicillin. At this time, we changed the antibiotic to ampicillin-sulbactam (300 mg/kg/day) and cefotaxime (200 mg/kg/day) due to the unavailability of aqueous crystalline penicillin G in Korea and of tissue culture results.

The infant underwent intensive medical management with mechanical ventilation and twice-daily wound dressing with or without debridement in the ICU. In the subsequent days after the intensive care, the wound improved and was closed on day 9. The follow-up culture isolated no bacteria, and she was discharged healthy on day 22 after completion of 3-week antibiotic therapy.

Brain magnetic resonance imaging before discharge showed no abnormal signal intensity. There were no neurologic sequelae after discharge. She obtained normal findings of immunologic battery of tests, including next-generation sequencing panel, lymphocyte subset, immunoglobulins (G, A, M, E, and D), C3, C4, and CH50.

Discussion

We reported a rare case of infant NF caused by GBS bacteremia, despite the rarity of GBS as a cause of NF[1]. In infants, GBS infection can be categorized into early- (< 7 days of age) and late-onset diseases (7-90 days)[7]. Early-onset disease tends to present as respiratory distress, sepsis or pneumonia while late-onset disease presents as bacteremia or meningitis[7]. Although skin and soft tissue infection is a rare manifestation of GBS infection, primary soft tissue infection may be a sign of GBS bacteremia[8,9]. Although the case patient's corrected age of 7 days was borderline of early- and late-onset diseases, her bacteremia without respiratory distress indicated the latter disease. We speculate that the GBS NF stemmed from bacteremia due to the following reasons. First, she did not undergo skin trauma or invasive procedures during previous hospitalization, which is a prerequisite for diagnosis of secondary NF. Second, the skin lesion worsened after the occurrence of fever, an essential symptom of bacteremia. As our case was positive for GBS in both blood and CSF, about 13%-25% of GBS bacteremia may have meningitis due to the same pathogen[7]. Hence, culture should be performed using both blood and CSF, as well as aspiration from the affected regions, to make microbiological diagnosis.

The most common site of neonatal NF is the abdominal wall or back[1,4]. This feature contrasts with the fact that NF has a predilection for the extremity in adults[6]. This discrepancy may be due to differences in pathogens and risk factors of the patient group. Common risk factors for pediatric NF include omphalitis, balanitis, placement of electrodes and catheters for monitoring, postoperative complications, necrotizing enterocolitis, bullous impetigo, maternal mastitis, septicemia, prematurity, and immunodeficiency[1,7,10]. Risk factors for neonatal GBS sepsis include prematurity, immun-

odeficiency, prolonged PROM, prior antibiotic use, African-American race, previous infant GBS infection, maternal GBS colonization, and maternal age younger than 20 years[7,11,12]. Among these risk factors, the case patient had prematurity with low birth weight, maternal history of chorioamnionitis with PROM, and maternal cervical GBS colonization.

Early suspicion and aggressive treatment are important for the outcome of NF. It is sometimes difficult to differentiate NF from other soft tissue infections because the initial presentations of NF are similar to those seen in cellulitis with nonspecific features of sepsis accompanying localized skin and soft tissue erythema[7]. NF should be suspected in infants with rapidly progressive erythema with crepitus or signs of systemic toxicity, such as fever, tachycardia, hypotension, and anxiety or lethargy[6,13]. Ultrasonography, computed tomography or magnetic resonance imaging can aid in the early diagnosis of NF[1]. The most useful imaging finding is the presence of gas in the soft tissues and fluid collections, as well as edema and thickening of the fascia or abscess formation[1,13]. Definitive diagnosis of NF is usually made surgically by the presence of the dusky gray subcutaneous fat and the fascia with scanty serosanguineous discharge[1].

Mainstays of treatment are empirical antibiotic therapy covering gram-positive, gram-negative, and anaerobic bacteria pending culture results, along with early surgical debridement[1,3]. Conventionally, the antibiotic regimens include high-dose penicillin or cephalosporin, aminoglycoside, and clindamycin or metronidazole[3,4]. In a case review of pediatric NF, 2 antibiotics were frequently used with combinations of aminoglycoside or third-generation cephalosporin (cefoperazone sodium) plus clindamycin[2]. However,

vancomycin or linezolid is currently recommended due to concerns over possible methicillin-resistant *S. aureus* infection. Also in adult NF, vancomycin or linezolid plus piperacillin-tazobactam is recommended as empirical antibiotics[14].

Infants with NF need multi-systemic support in ICUs. Nutrition is equally important in the healing process, and parenteral support is needed[10]. It has yet to be evaluated regarding hyperbaric oxygen, corticosteroids, intravenous immunoglobulin or granulocyte colony-stimulating factor[1].

To summarize, we reported a case of infant NF caused by GBS bacteremia, a rare pathogen-induced rare disease. NF is a rare but rapidly progressing and fatal soft tissue infection. If there is suspicion of NF, it is essential to implement aggressive antibiotic therapy and surgical management.

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Conflicts of interest

No potential conflicts of interest relevant to this article were reported.

Funding sources

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