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Necrotizing Fasciitis Post-Cesarean Section Leading to Transabdominal Hysterectomy

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Abstract

Necrotizing fasciitis (NF) is a rare but life-threatening disease characterized by rapidly spreading inflammation and subsequent necrosis of the fascial planes and surrounding tissues. Limited literature has described NF as involving an adjacent solid organ beyond fascial planes that has required its removal. We present a case of a 25-year-old white female who underwent a cesarean section and subsequently developed NF involving her uterus and abdominal wall that necessitated a total abdominal hysterectomy, serial surgical debridement of necrotic tissue, and wound vacuum assisted closure (VAC) placement. Her pathology report described her uterus infiltrated by polybacteria, confirming a diagnosis of NF. Despite NF's progressive nature and potential lethality, NF can be challenging to diagnose clinically due to a lack of pathognomonic signs and symptoms. However, early detection of NF with the aid of Laboratory Risk Indicator for Necrotizing Fasciitis score calculation using laboratory values such as white blood cell count, hemoglobin, sodium, glucose, serum creatinine, and C-reactive protein is critical for optimal patient outcomes. A multidisciplinary team approach is vital in treating these patients to debride necrotizing tissue and control the potential sequelae from the infection, particularly for postpartum patients.

Keywords

- necrotizing fasciitis (NF)
- pregnancy
- necrotizing soft tissue infection
- hysterectomy
- postpartum

Necrotizing soft tissue infection, or necrotizing fasciitis (NF), is a rare but life-threatening disease with an overall incidence of 0.24 to 0.4 per 100,000 adults and a mortality rate of about 26.6%.[1] NF is characterized by rapidly spreading inflammation and subsequent necrosis of the fascial planes and surrounding tissues.[2] The etiology of NF typically follows an injury to a minor entry site; however, 45% of the cases reported no identifiable point of entry.[3] Wound cultures at time of admission or at the first surgical debridement are most commonly polymicrobial (58.7%), often including these cultured pathogens: anaerobes, Gram-negative rods, β-hemolytic streptococcal species, coagulase-negative Staphylococcus species, Enterococcus species, and *Staphylococcus aureus*.[4] The early clinical presentation of NF includes primarily erythema, edema, fever, and pain. Late symptoms include bullae, dysesthesia/anesthesia, hard skin upon

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palpitation, crepitation, discoloration, and systemic manifestations,[5] such as pneumonia, urinary tract infections, multiorgan failure,[6,7] sepsis, and death.[8] However, due to a lack of pathognomonic signs and symptoms, the diagnosis of NF poses significant challenges to clinicians. The management of NF begins with establishing a rapid diagnosis, using a combination of clinical intuition, laboratory data using the Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC), and imaging. The LRINEC score is a clinical tool that can be used to distinguish NF from other soft tissue infections early in a patient course that uses laboratory data including total white cell count, hemoglobin, sodium, glucose, serum creatinine, and C-reactive protein. A score of or above 6 points has a positive predictive value of 92% and negative predictive value of 96%[9] for NF. In another systematic review of literature that included 15 studies with 846 patients, the mean LRINEC score was 6.06 for NF patients.[10] Multidisciplinary consultation, surgical debridement, and delayed closing and reconstruction are ultimately required in confirmed cases.[11] The risk factors associated with NF include diabetes mellitus, intravenous drug use, age greater than 50, hypertension, malnutrition, obesity, and immunocompromised states.[12] Because immunocompromised patients with NF may fail to exhibit the typical clinical and laboratory signs of NF, they commonly experience delays in diagnosis and consequential delays in surgical treatment causing higher NF-associated hospital mortalities.[4]

In this case, we present a 25-year-old white female who underwent a cesarean section and subsequently developed NF involving her uterus and abdominal wall that necessitated a total abdominal hysterectomy (TAH), serial surgical debridement of necrotic tissue, and wound VAC placement. After searching three literature databases (PubMed, Embase, Web of Science), we did not find articles or case reports that documented NF spreading beyond fascial planes and leading to an adjacent/nearby organ removal.

Case

A 25-year-old female, gravida 1, para 0, presented for scheduled induction of labor at 39 0/7 weeks' gestation. The patient's pregnancy was complicated by multiple fetal anomalies (heterotaxy, preduodenal portal vein, malrotation), mild polyhydramnios (amniotic fluid index 28.11 cm), gestational diabetes on metformin, chronic hypertension on labetalol, and chronic bipolar disorder. Her group B Streptococcus status at the time of labor was negative. Her labor course was notable for arrest of dilation at 9 cm, and she underwent a primary low transverse cesarean delivery. The patient received perioperative intravenous antibiotics including clindamycin, gentamycin,[13] and azithromycin[14] due to penicillin allergy. During the delivery, a left inferior uterine extension was noted. The hysterotomy and uterine extension were closed using a running locking 0 Monocryl suture on the first layer and an imbricating 0 Monocryl suture on the second layer. The surgery was complicated by a postpartum hemorrhage with an estimated blood loss (EBL) of 1,500 mL secondary to uterine atony. Oxytocin, methylergonovine, carboprost, and misoprostol were administered for uterine atony. A B-lynch suture was placed using #1 chromic suture and several figure-of-eight sutures using 0-Vicryl were required for additional hemostasis. Postoperatively, the patient's hematocrit decreased from 34 to 26%, but she did not require blood transfusion. The patient had no postpartum complications and was discharged on postoperative day (POD) 4 after delivery.

The patient represented to the hospital on POD 7 after delivery with severe abdominal/incisional pain, erythema at incision site, body aches, chills, and subjective fevers. Vital signs revealed blood pressure of 121/71 mm Hg, heart rate of 128 beats/minute, oxygen saturation of 99%, respiratory rate of 20, and a temperature of 103.2°F. Physical examination was notable for an ill-appearing, flushed woman in mild distress. Lungs were clear to auscultation bilaterally. Cardiac exam revealed tachycardia with regular rhythm. Abdominal exam was notable for erythema extending from the mons pubis to the level of the umbilicus with associated edema. There was no overt drainage noted from the incision or crepitus in the abdominal wall. A 3×4 cm ecchymotic area was present at midpoint of Pfannenstiel suture.

Labs were significant for: white blood count $21.6 \times 10(3)$/mcL, hematocrit 24%, sodium 140 mmol/L, creatinine 0.76 mg/dL, and glucose of 63 mg/dL. The patient's calculated LRINEC score was 5. An emergent computed tomography (CT) of the abdomen and pelvis was ordered and demonstrated extensive subcutaneous fat stranding, skin thickening, and multiloculated fluid and gas within the anterior abdominal wall near the incision. Necrotizing infection of the uterus was diagnosed when moderate foci of gas was seen within the endometrium extending into the body of the uterus. Findings were highly concerning for necrotizing infection. The general surgery (GS) service was immediately consulted.

The patient was taken to the operating room (OR) by the gynecology team for an exploratory laparotomy and GS joined for intraoperative support and assistance with debridement and source control. Abdominal examination in the OR noted extensive expansion of the previously noted abdominal wall erythema with the presence of newly formed bullae over the central portion of the Pfannenstiel incision. The necrotic tissue also increased in size to 4×4 cm.

Intraoperatively, the prior Pfannenstiel incision was reopened. Purulent fluid was noted immediately beneath the skin along with necrotic subcutaneous tissue extending across the anterior abdominal wall. Additionally, defects in fascia were also noted. Upon inspection, the uterus was also found to have purulent discharge from the hysterotomy. GS debrided $15 \times 8 \times 6$ cm from the anterior abdominal wall. The gynecologic team then proceeded with hysterectomy and bilateral salpingectomy. The EBL was 1,500 mL, and the patient had a hematocrit of 18%. Three units of packed red blood cells were transfused intraoperatively. The abdomen was closed with an advanced open abdomen dressing, and the patient remained intubated and sedated. She was transferred to the surgical intensive care unit with plans for additional debridement surgery.

The tissue culture was notable for *Enterococcus faecalis*, *Streptococcus anginosus*, and *Prevotella bivia*. The patient's pathology report described a post-TAH post-gravid uterus

with focal area of ischemic necrosis infiltrated by many bacteria (Gram-positive cocci and Gram-negative rods). There was evidence of endocervical acute inflammation and acute salpingitis. The abdominal biopsies showed fibrofatty tissue with marked acute inflammation with many bacterial colonies (Gram-positive cocci and Gram-negative rods) and focal ischemic necrosis.

The patient returned to the OR 3 days later. Other than a necrotic portion of the rectus muscles requiring further debridement, the tissue appeared otherwise healthy and viable. A 16×11 cm fascial defect was repaired with a biologic mesh sutured to the fascial edges using running #1 polydioxanone. The vaginal cuff was examined under anesthesia and was unremarkable with prior sutures intact. During the postoperative period, the patient had a pre-renal acute kidney injury secondary to hypovolemia/sepsis and likely exacerbated by nephrotoxic medications.

On POD 5 following exploratory laparotomy and hysterectomy, intravenous antibiotics were transitioned to oral antibiotics and a wound VAC was placed. The patient was discharged on POD 9 to inpatient rehab. At her postpartum visit 3 weeks later, the patient continued to use a walker to ambulate.

Discussion

NF is a rare but rapidly progressive and often lethal bacterial infection that primarily causes necrosis of the subcutaneous tissue, fascia, and/or muscle.[15,16] In our patient, the endometrium and the body of the uterus were found to be infiltrated with polymicrobial bacteria, later confirmed with pathology, due to NF. However, an extensive review of the literature has found limited cases of NF affecting organ systems beneath subcutaneous soft tissue or facial planes. There was also a paucity of literature of NF involving the uterus as a complication of a cesarean section resulting in the need for a TAH.

Current literature has identified risk factors for NF. These include diabetes mellitus, hypertension, cirrhosis, obesity, and immune suppressive states like pregnancy, malignancy, chronic disease, malnutrition, advanced age (> 50), and/or use of nonsteroidal anti-inflammatory drugs (NSAIDs) in early postoperative period.[17] Our patient had several risk factors associated with NF prior to her presentation, including gestational diabetes mellitus, anemia due to chronic disease, immunosuppressive state due to pregnancy, recent surgery (cesarean section), and the recent NSAID use in early postoperative period. Because NSAIDs could mask signs and symptoms of NF,[18] some clinicians believe that NSAID use in early postoperative period is associated with higher rates of *Streptococcus pyogenes* infection.[19] However, new data suggests that NSAIDs may also accelerate disease progression and limit antibiotic efficacy in established group A Streptococcal soft tissue infections.[18]

Due to its progressive nature and potential lethality, NF should be promptly diagnosed. A delay in diagnosis has been linked to higher mortality rates. Physical exam is the most important clinical tool for the diagnosis of NF. However, the paucity of specific cutaneous or clinical signs makes early recognition difficult for the disease. Some external clinical signs associated with NF include foul-smelling lesions with serosanguineous discharge.[5] Signs of sepsis such as fever, hypotension, tachycardia, erythema, crepitus, and/or purulent drainage particularly in the groin, perineum, or recent incisions should call for a suspicion for NF. In cases that may be ambiguous, the LRINEC score can be calculated to compute variable sensitivity and risks of developing NF.

The LRINEC score is a clinical tool that can be used to distinguish NF from other soft tissue infections early in a patient course, given that early operative debridement is a major determinant of outcomes in NF. Total white cell count, hemoglobin, sodium, glucose, serum creatinine, and C-reactive protein are the lab values that contribute to the score. However, there are limitations of the LRINEC score. First, the LRINEC score does not accurately predict 100% of NF cases as Tarricone et al found that the LRINEC score has a specificity of 83.17% and a sensitivity of 49.39%.[20] In our patient, her score was 5, which puts her at a low risk for an NF-predicted diagnosis. Second, the LRINEC score is calculated based on laboratory value, which can sometimes be time consuming and logistically challenging. Given the limitation, if there is a high suspicion for NF through clinical history and physical exam, it is pertinent to proceed to operative debridement without calculating a LRINEC score. The diagnosis of NF can be confirmed by the presence of gas in tissues on a CT scan without contrast.

The distribution of causal pathogens in NF can vary widely in literature reports, partially related to the geographical and clinical context of the cases. Nevertheless, polymicrobial cases account for approximately 50% of the cases, often consisting of a combination of Enterobacteriaceae and anaerobic bacteria.[21] On the other hand, B-hemolytic *Streptococci* and *S. aureus* are frequently associated with monomicrobial NF.[21] Water exposure associated pathogens such as *Vibrio vulnificus* and *Aeromonas hydrophila* have been regarded as rare causes of monomicrobial NFs, and literature has suggested that these pathogens predominantly affect subjects with preexisting chronic liver conditions[15] and who reside in certain countries such as Taiwan.[22] Due to the various potential causative pathogens, treatment with an empirical broad-spectrum antibiotic therapy consisting of third-generation cephalosporin, penicillin, and metronidazole[23] is recommended. Moreover, other literature has suggested adding penicillin for treatment of *Streptococci* and particularly when *Clostridia* is suspected.[24] As an alternative, clindamycin and chloramphenicol can be substituted empirically to facilitate coverage of Gram-positive cocci and anaerobes until culture results return.[24]

The management of NF often requires a multidisciplinary approach to provide the best care for the patient. In this case, the GS and obstetrics and gynecology teams collaborated early upon the patient's arrival to the hospital. Since NF can affect soft tissue in various body parts, muscle, or bone, the care of the patient can involve different specialties ranging from orthopaedics, GS, otolaryngology, plastics, to obstetrics and gynecology.

The treatment for NF is operative since the disease is progressive. The operative findings in NF include grayish necrotic fascia, a decrease/lack of resistance of normally adherent

superficial fascia to blunt dissection, lack of bleeding fascial tissue, and the presence of foul-smelling "dishwater" pus.[5] The goal of the debridement is to remove all tissue that is not viable. The excised tissue is sent to pathology and histology to confirm the causative agents so that bacteria-specific antibiotics can be utilized rather than continued broad-spectrum antibiotics, with its inherent risk of selection of resistant bacteria and organ dysfunction (renal or *Clostridium difficile* colitis).

After operative debridement, our patient's abdomen was temporarily closed with a negative pressure wound device with plans to return to the OR for final closure. In our patient, a biologic mesh and a vacuum dressing were also placed to allow for development of granulation tissue. The vacuum-assisted closure system has proved to be helpful in wound management with its combined benefits of continuous cleansing of the wound and the formation of granulation tissue.[25]

Other postoperative considerations that should be highlighted in NF patients include pain management and psychosocial support postoperation. It has been reported that patients who survived NF have a lower self-reported quality of life for multiple domains when compared to multiple reference populations, particularly regarding physical functioning.[26] This underlines the importance of providing patients, particularly those who are postpartum, with enough comprehensive information during admission to ensure more realistic expectations during their recovery. Furthermore, involving other providers of the care teams like physiotherapists and social workers during admission can be helpful to streamline patient care during admission and after discharge. These considerations are important to ensure optimal and holistic care of the postpartum patient and to minimize residual complaints after NF.

Conflict of Interest

None declared.

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