

Case Report

A rare case of fully recovered necrotizing fasciitis

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Abstract: Necrotizing fasciitis (NF) is a severe soft tissue infection caused by bacterial fascia invasion and quick spreading to the muscle and subcutaneous tissues. Treatments of NF should be conducted by extensive debridement and antibiotic therapies. This report presented a 53-year-old woman with diabetes mellitus (DM) and hypothyroid who was referred to our medical center with lower limb pain and significant swelling after mild trauma. The patient was diagnosed with NF due to *E. coli*. She underwent surgical interventions for debridement and long-term antibiotic therapy. The patient recovered successfully without complications or range of motion (ROM) restriction in the hip and knee. It is recommended that immediate actions be taken in NF cases to preserve the remaining tissues and provide better outcomes.

Keywords: Necrotizing fasciitis, trauma, case report

Introduction

Necrotizing fasciitis (NF) is an uncommon but severe soft-tissue infection. This condition is caused by bacterial invasion of the fascia and rapidly spreading to muscles, subcutaneous fat, and overlying skin areas, typically resulting in rapid mortality due to multi-organ failure [1]. NF is exceptionally deadly in 20-25% of patients [2], and an early diagnosis is the cornerstone of prompt therapy and a better outcome [3]. Diagnosis of NF could be challenging because it may occur without a known causative factor or portal of entry for bacteria. Based on data, diagnosis of NF is made by clinical symptoms and with the help of imaging evaluations that could show gas presence in the tissues [4]. Early symptoms of NF are:

- Skin opening.
- Pain in the general area.
- Pain disproportionate to the injury.
- Flu-like symptoms.
- Dehydration.
- Combination of the above symptoms.

Advanced and critical symptoms include swelling in the site of injury and color changes to purple, developing significant, dark marks and blistering with blackish fluid, necrotic wound with a bluish, white, or dark, mottled, flaky appearance, severe hypotension, developing septic shock and loss of consciousness [4, 5]. On the other hand, the lack of specificity of the symptoms at the time of presentation or admission and the rapid progression of septic shock and multi-organ failure (MOF) make it typically exceedingly difficult to make a prompt clinical diagnosis [6].

NF patients frequently present with systemic infection symptoms. As in the present case report, NF must be regarded as a potential issue when screening patients with symptoms [2]. NF is classified into four types: Type I includes 70-80% of cases with polymicrobial or synergistic causes. The responsible microorganisms include Bowel flora-derived, mixed anaerobes and aerobes, *E. coli*, *Pseudomonas* spp., and *Bacteroides* spp. Type II has 20-30% of cases and is often monomicrobial. The responsible microorganisms include skin- or throat-derived bacteria, Group A B-haemolytic streptococcus (GAS), and occasionally streptococcus aureus. Type III is common in Asia and is

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Figure 1. Initial examination of the patients, showing massive limb swelling.

mainly caused by gram-negative bacteria and marine-related organisms such as *Vibrio* spp., *Pasteurella multocida*, *Haemophilus influenza*, *Klebsiella* spp. and *Aeromonas* spp. Type IV is caused by Fungi, including *Candida* spp. and Zygomycetes, in immunocompromised patients [5, 7].

The only treatment option for NF is surgical debridement of the necrotic tissues and broad-spectrum antibiotic therapy [8, 9]. The mortality rate of NF ranges from 11-22% in different studies that could reach 16-33% when associated with streptococcal toxic shock syndrome [10].

This study presented a rare case of fully recovered NF in the left lower extremity. This case recovered successfully without complications or range of motion (ROM) restriction in the hip and knee.

Case presentation

A 53-year-old woman with diabetes mellitus (DM) and hypothyroidism presented with dyspnea and agitation to the emergency ward. This patient had a traumatic accident to the left lower limb two months before admission. After further evaluations, she was discharged from the hospital after further assessment and without any interventions. Afterward, the patient suffered from gradual swelling in the left lower limb (LLL), and she returned to the hospital with limb swelling. She was recommended to undergo magnetic resonance imaging (MRI) for lower limb imaging, but she did not attend the imaging center due to the COVID-19 pandemic. One month after the traumatic accident, the swelling worsened following a falling accident. She underwent MRI imaging of the limb, and during

the imaging procedure, she developed respiratory distress and agitation.

She was referred to the emergency ward of our medical center with these complications. An internal medicine specialist did an initial visit. Massive swelling of the left femur was observed during the initial orthopedic visit. Physical examinations showed a 13/15 Glasgow Coma Scale

(GCS), O₂ saturation of 92%, and body temperature of 38 degrees. In the general examination, massive swelling of the left femur was accompanied by erythema, warmth, fluctuation, creptation, and tenderness. The range of motion (ROM) of the left knee and hip was limited (**Figure 1**). Primary laboratory data are summarized in **Table 1**.

Ultrasound examination showed a gas collection in the anterior compartment of the left femur. The X-ray of the left femur demonstrates gas accumulation at this site (**Figure 2**).

The diagnosis of NF was ascertained because the patient had the following symptoms: pain disproportionate to the injury, flu-like symptoms, swelling in the site of injury, and the presence of gas in the mentioned site, according to imaging evaluations.

Treatment

Two hours after the first orthopedic visit, the patient was transferred to the operation room (OR), and the anterior and lateral compartments of the left femur opened with one incision. The gas was expelled rapidly with a foul odor. Purulent discharge was seen and sent for culture. Irrigation and debridement were done with normal saline serum. The wound was opened, and the dressing was done.

Outcome and follow-up

After the first surgery, the general conditions of the patient improved. Incision and drainage (I and D) were done several times, and necrotic tissues were removed from the site of infection (**Figure 3A**). A defect was seen in the anterior of the left femur that required a flap for closing. The result of the tissue culture test was *E. coli*.

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Table 1. Results of the primary laboratory tests

Variable	Result
White blood cell (WBC)	15000 cells per cubic millimeter (cmm) with NEUT dominant
Hemoglobin (Hb)	8.5 g/dl
Platelet	275000 mL
Erthrocyte sedimentation rate (ESR)	132 millimeters per hour
C-reactive protein (CRP)	96 mg/L
Prothrombin Time (PT)	17.5
partial thromboplastin time (PTT)	22
International Normalized Ratio (INR)	1.5
Blood sugar (BS)	400 mg/dL
Na	135 mEq/L
K	4.5 mEq/L
Blood urea nitrogen (BUN)	43 mg/dL
Creatinin (Cr)	2.03 mg/dL



Figure 2. The X-ray of the left femur demonstrating gas accumulation.

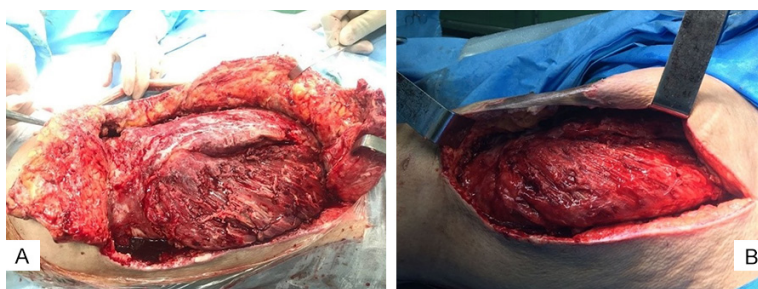


Figure 3. A. Necrotic tissue was removed with a skin defect, B. After several times of incision and drainage, the wound was cleaned.

Based on the antibiogram and consultation with infectious disease specialists, treatments with vancomycin and meropenem were initiated. When the wound was cleaned (**Figure 3B**), the wound was closed without any flap

after two rounds of surgeries (**Figure 4**).

After treatments with several I and D surgeries and long-term intravenous antibiotic therapy, the patient recovered without skin flaps, and the entire hip and knee ROM was preserved. She was discharged in good general condition. The patient was visited three months after discharge. She had no complaints, and the lower limb physical examinations were normal. No pathologic findings were observed. It should be noted that informed consent on disclosing the data was taken from the patient.

Discussion

Origin of soft tissue infection

In most cases, microorganisms that cause NF infection gain access to the fascia through a break in the skin (generally due to trauma), creating a continuous interface between the surface and the subcutaneous tissue. Indeed, the etiology of NF has been linked to a variety of traumas, including varicella skin lesions [11], surgery, blunt or penetrating trauma, colonic perforation [12], tattoos [13], nee-

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Figure 4. First round (A) and final wound closure (B).

dle stick injuries [14, 15], intestinal hernia [13], insect bites [16], burns, stingray injuries, toothpick injuries, childbirth [17], and/or rectal mucosal injuries associated with temperature measurements [18]. NF can also develop after trauma and can occur without evidence of skin damage (e.g., bruises) or trauma, particularly in pre-existing conditions [19], such as diabetes mellitus [20], older age [21], rheumatoid arthritis [22, 23] alcoholic liver cirrhosis, peripheral vascular disease, chronic renal failure [19], malignancy [24], liver disease [25], malnutrition and/or myelotoxic chemotherapy. In situations like these, it was hypothesized that the bacteria responsible for the NF infection would first colonize an extracutaneous part of the body, then transit through the blood to a location of dermal inflammation (such as a contusion or hematoma), and finally cause the NF infection.

To find the infection's most likely source, it is critical to inspect the body (including inner organs) without focusing only on soft tissues. Indeed, imagine that trauma caused by human activity is the main reason for the illness (e.g., vehicle accidents, physical assaults, etc.). This means that the person or entity being sued may be held legally responsible for the decedent's NF-related mortality (e.g., manslaughter or accidental death). Even with a thorough search, a pathologist should be mindful that it would not always be possible to locate the infection's origin using solid forensic evidence.

The microorganisms causing the infection

As widely reported in the literature, NF can be classified into different types based on the ante-and/or postmortem cultures (**Table 1**) [6]. The autopsy should be done immediately after the exitus to prevent the invasion of tissues

by saprophytic bacterial flora. The forensic pathologist should review any clinical microbiology data and collect peripheral blood and necrotizing tissues during the autopsy to properly identify the microorganism(s) involved. Identifying the microorganisms involved in the necrosis can be crucial for determining the prognosis of the disease and evaluating the correctness of the medi-

cal interventions (e.g., antibiotic therapy, and surgical debridement). Type II NFs show a rapid clinical development to septic shock and death despite any medical/surgical treatment or intensive support, especially when group A β -hemolytic streptococcus (GAS) is present. In some other articles, two different types of NF have been described. Type 1 is usually caused by multiple organisms (70-90%, streptococcal species, staphylococcal species, enterococcal species, Enterobacteriaceae, Bacteroides species), and type 2 typically involves group A streptococci (10%) [8-10]. Some researchers have included a type 3 that includes uncommon methicillin-resistant *Staphylococcus aureus* or *Vibrio* species as the causal agents. Some patients with GAS showed a fulminant development to MOF and died from septic shock with MOF in less than 12 hours [25].

Clinical evaluation and staging

NF is typically subdivided into three clinical phases: a) During the first 24 hours (i.e., the early phase, development of the infection to NF), pain out of scale to the injury or skin infection is typically noted. Possible supplementary findings include erythema and/or hyperpigmentation of the skin. b) During the subsequent 2-4 days (i.e., the intermediate phase), clinical toxicity symptoms such as vomiting, dizziness, weakness, nausea, fever, disorientation, diarrhea, malaise, and dehydration may manifest. The painful area exhibits "hard signs" of NF, including edema, purplish rash, or blisters with "dish-wash" hemorrhagic or purulent fluid, gas buildup under the skin and local emphysema. The discomfort increases and is out of proportion to the skin lesions. c) After 4-6 days, septic shock and numerous organ dysfunction syndromes (late phase) manifest,

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followed by hypotension, tachycardia, cardiac shock, an increase in white blood cell count, weakness, acidosis, mental status abnormalities, and coagulopathy [3].

According to a typical clinical course, inflexible signs and symptoms of NF should appear many days before multi-organ failure, raising the possibility of NF. However, the infection spreads fast and systemically in the acute and fulminant types, with early indications of severe septic shock. In each of the seven instances that were discussed, NF manifested as a fulminant course with harsh characteristics that only appeared in septic shock, followed by a swift death that occurred from sepsis within 24 hours. Therefore, while clinically staging NF in death cases of NF in an ex-post situation, the forensic pathologist must exercise extreme caution. Radiological imaging can help in NF evaluation even though it is a clinical diagnosis, especially concerning the development of consequences and the evolution of the disease [3]. Ultrasound, which shows fascial thickening and fluid accumulation, is most useful in pediatric patients [26, 27].

Plain radiographs are only marginally useful since soft tissue thickening and opacity are symptoms that can resemble cellulitis. Additionally, soft tissue gas, regarded as a diagnostic feature, is infrequently seen [26]. Even though they are intermittent, soft tissue air and fluid in the deep fascia can be easily detected using computed tomography (CT) [27, 28]. Additionally, CT can detect fascial thickness and enhancement, lymphadenopathy, and serious consequences such as vascular rupture [26]. Magnetic resonance imaging (MRI) is particularly suitable for diagnosing NF due to its high soft-tissue contrast. Indeed, high signal strength in the fluid-sensitive segments in the deep fascia, particularly the deep intermuscular fascia, is an observable diagnostic feature [27, 29].

The treatment of NF is based on prompt surgical debridement of the affected tissue and wide-spectrum antibiotic use [26]. As described above, the use of antibiotics depends on the type of responsible microorganisms. Ampicillin or ampicillin-sulbactam combined with metronidazole or clindamycin for anaerobic coverage is the most effective [25, 26].

LRINEC's potential for clinical staging and early detection of NF

Several scoring systems based on a combination of clinical data and blood tests have been presented to anticipate the clinical diagnosis of NF. The Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score, which has recently been updated to the modified LRINEC and now includes considering clinical characteristics, is the one that is utilized the most.

The LRINEC is a dependable scoring system that uses data from six commonly ordered laboratory tests, including total white cell count per cubic millimeter, hemoglobin, glucose, C-reactive protein, creatinine, and sodium. It is utilized to differentiate NF from other severe soft tissue infections at an early stage [30]. Different cut-offs were used to stratify the patients into risk categories for NF. These cut-offs corresponded to a different probability of developing NF and several statistical metrics such as specificity, sensitivity, negative and positive predictive values, and likelihood ratios. The accuracy of the LRINEC in making NF diagnostic forecasts has been subjected to extensive research and analysis in published works. There are also cases of people diagnosed with NF even though they had a standard LRINEC score [31]. When patients undergo surgery within 12 hours of the onset of symptoms, the mortality rate is approximately 6%; however, it can reach up to 30% after 24 hours [11].

Conclusion

Treatments of vast NF should be conducted as soon as possible with invasive surgical debridement and antibiotic therapies based on cultures. This report highlights the importance of immediate surgical interventions to preserve the patient's tissues and full ROM.

Disclosure of conflict of interest

None.

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References

- [1] Cai Y, Cai Y, Shi W, Feng Q and Zhu L. Necrotizing fasciitis of the breast: a review of the literature. *Surg Infect (Larchmt)* 2021; 22: 363-373.
- [2] Sanchez IM, Lowenstein S, Johnson KA, Babik J, Haag C, Keller JJ, Ortega-Loayza AG, Cohen J, McCalmont TH, Demer AM, Mansh MD, Hylwa SA, Liu J and Shinkai K. Clinical features of neutrophilic dermatosis variants resembling necrotizing fasciitis. *JAMA Dermatol* 2019; 155: 79-84.
- [3] Roje Z, Roje Ž, Matić D, Librenjak D, Dokuzović S and Varvodić N. Necrotizing fasciitis: literature review of contemporary strategies for diagnosing and management with three case reports: torso, abdominal wall, upper and lower limbs. *World J Emerg Surg* 2011; 6: 46.
- [4] Gunaratne DA, Tseros EA, Hasan Z, Kudpaje AS, Suruliraj A, Smith MC, Riffat F and Palme CE. Cervical necrotizing fasciitis: systematic review and analysis of 1235 reported cases from the literature. *Head Neck* 2018; 40: 2094-2102.
- [5] Chen LL, Fasolka B and Treacy C. Necrotizing fasciitis: a comprehensive review. *Nursing* 2020; 50: 34-40.
- [6] Morgan M. Diagnosis and management of necrotising fasciitis: a multiparametric approach. *J Hosp Infect* 2010; 75: 249-257.
- [7] Huang TY, Tsai YH, Kuo LT, Hsu WH, Hsiao CT, Hung CH, Huang WY, Wu HR, Chuang HJ, Li YY and Peng KT. Different types of bullae of limbs with necrotizing fasciitis predict different outcome: a prospective study. *Infection* 2021; 49: 135-144.
- [8] Dhawan G, Kapoor R, Dhamija A, Singh R, Monga B and Calabrese EJ. Necrotizing fasciitis: low-dose radiotherapy as a potential adjunct treatment. *Dose Response* 2019; 17: 1559325819871757.
- [9] Leiblein M, Marzi I, Sander A, Barker J, Ebert F and Frank J. Necrotizing fasciitis: treatment concepts and clinical results. *Eur J Trauma Emerg Surg* 2018; 44: 279-290.
- [10] Al-Qurayshi Z, Nichols RL, Killackey MT and Kandil E. Mortality risk in necrotizing fasciitis: national prevalence, trend, and burden. *Surg Infect* 2020; 21: 840-852.
- [11] Hidalgo-Carballal A and Suárez-Mier MP. Sudden unexpected death in a child with varicella caused by necrotizing fasciitis and streptococcal toxic shock syndrome. *Am J Forensic Med Pathol* 2006; 27: 93-96.
- [12] Reisman JS, Weinberg A, Ponte C and Kradin R. Monomicrobial pseudomonas necrotizing fasciitis: a case of infection by two strains and a review of 37 cases in the literature. *Scand J Infect Dis* 2012; 44: 216-221.
- [13] Hourmozdi JJ, Hawley DA, Hadi CM, Tahir B and Seupaul RA. Streptococcal necrotizing myositis: a case report and clinical review. *J Emerg Med* 2014; 46: 436-442.
- [14] Finn SP, Leen E, English L and O'Briain DS. Autopsy findings in an outbreak of severe systemic illness in heroin users following injection site inflammation: an effect of clostridium novyi exotoxin? *Arch Pathol Lab Med* 2003; 127: 1465-1470.
- [15] Dunbar NM and Harruff RC. Necrotizing fasciitis: manifestations, microbiology and connection with black tar heroin. *J Forensic Sci* 2007; 52: 920-923.
- [16] Rellinger EJ, Craig BT, Craig-Owens LD, Pacheco MC, Chung DH and Danko ME. Clostridium sordellii necrotizing omphalitis: a case report and literature review. *J Pediatr Surg Case Rep* 2016; 10: 35-38.
- [17] Elliott D, Kufera JA and Myers RA. The microbiology of necrotizing soft tissue infections. *Am J Surg* 2000; 179: 361-366.
- [18] Christensen R, Pysner T and Christensen S. Case report: perianal necrotizing fasciitis in a near-term neonate. *J Perinatol* 2007; 27: 390-391.
- [19] Tajiri T, Tate G, Miura K, Masuda S, Ohike N, Kunimura T, Mitsuya T and Morohoshi T. Sudden death caused by fulminant bacterial infection: background and pathogenesis of Japanese adult cases. *Intern Med* 2008; 47: 1499-1504.
- [20] Cope TE, Cope W and Beaumont DM. A case of necrotising fasciitis caused by serratia marsescens: extreme age as functional immunosuppression? *Age Ageing* 2013; 42: 266-268.
- [21] Wilson MP and Schneur AB. A case of necrotizing fasciitis with a LRINEC score of zero: clinical suspicion should trump scoring systems. *J Emerg Med* 2013; 44: 928-931.
- [22] Sugimoto T, Nakazawa J, Yomoda M, Hirata K, Soumura M, Kawasaki T, Uzu T, Nishio Y, Sugihara H and Eguchi Y. Necrotizing fasciitis resulting from Escherichia coli in a patient with chronic rheumatoid arthritis. *APLAR J Rheumatol* 2007; 10: 57-59.
- [23] Iwata K, Arinuma Y, Nakayama H, Nogi S, Futami H, Takaoka H, Hashimoto A, Shimada K, Komiya A, Matsui T, Saito I and Tohma S. An autopsy case of necrotizing fasciitis with rapidly progressive purpura caused by hemolytic streptococcal infection in a patient with rheumatoid arthritis. *Mod Rheumatol* 2011; 21: 669-672.
- [24] Muldrew KL, Miller RR, Kressin M, Tang YW and Stratton C. Necrotizing fasciitis from Vibrio vulnificus in a patient with undiagnosed hepa-

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- titis and cirrhosis. *J Clin Microbiol* 2007; 45: 1058-1062.
- [25] Abella BS, Kuchinic P, Hiraoka T and Howes DS. Atraumatic clostridial myonecrosis: case report and literature review. *J Emerg Med* 2003; 24: 401-405.
- [26] Fugitt JB, Puckett ML, Quigley MM and Kerr SM. Necrotizing fasciitis. *Radiographics* 2004; 24: 1472-1476.
- [27] Malghem J, Lecouvet FE, Omoumi P, Maldague BE and Berg BCV. Necrotizing fasciitis: contribution and limitations of diagnostic imaging. *Joint Bone Spine* 2013; 80: 146-154.
- [28] Chaudhry AA, Baker KS, Gould ES and Gupta R. Necrotizing fasciitis and its mimics: what radiologists need to know. *AJR Am J Roentgenol* 2015; 204: 128-139.
- [29] Hayeri MR, Ziai P, Shehata ML, Teytelboym OM and Huang BK. Soft-tissue infections and their imaging mimics: from cellulitis to necrotizing fasciitis. *Radiographics* 2016; 36: 1888-1910.
- [30] Wong CH, Khin LW, Heng KS, Tan KC and Low CO. The LRINEC (Laboratory Risk Indicator for Necrotizing Fasciitis) score: a tool for distinguishing necrotizing fasciitis from other soft tissue infections. *Crit Care Med* 2004; 32: 1535-1541.
- [31] Matsubara K, Mikamo H, Numa M, Yamamoto G, Kusano H and Takamine Y. Three fatal cases of invasive serotype VI group B streptococcal infection. *J Infect* 2006; 53: e139-e142.