Case Report

First Necrotizing Fasciitis Caused by Haemophilus influenza Serotype a

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Abstract

Necrotizing fasciitis (NF) is an infrequently encountered skin infection that has high morbidity and mortality, even with prompt medical and surgical intervention. We describe the case of a 67-year-old male presenting with significant NF in his left lower extremity, despite aggressive surgical intervention, and included multiple surgical debridements, ACell Matrix, split-thickness, and negative wound VAC therapy. Ultimately, this patient required a below the knee amputation. This is the first documented case of Haemophilus influenza type a causing NF.

Keywords

necrotizing fasciitis, necrotizing soft tissue, Haemophilus influenza serotype a, Haemophilus influenza

Case Report

A 67-year-old male presented to the Beaumont Dearborn emergency department with the complaint of worsening left leg pain for 6 days. He was seen 2 days prior to this hospital visit at another facility after cutting his leg on his bed frame. He was discharged with Bactrim and Norco for presumed cellulitis. His medical history was significant for insulin-dependent type 2 diabetes mellitus, hypertension, and arthritis.

There was muscle weakness and loss of motion and sensation in his left leg. Extensive bullae and erythema were found along the anterior and posterior leg, from the medial malleolus to proximal thigh (Figure 1). His vital signs were within normal limits: blood pressure 114/56, temperature 97.6, and pulse 77. Laboratory values were significant for hyponatremia (Na: 127), hypochloremia (Cl: 84), hypokalemia (2.7), hyperglycemia (Glu: 218), acute renal failure (blood urea nitrogen: 120; creatinine: 5.53; glomerular filtration rate: 10), and leukocytosis with bands (white blood cells: 23,000; bands: 8.7%).

In the emergency department, the patient was given vancomycin, clindamycin, and Zosyn. He was then brought to the operating room for emergent excisional debridement and placement of Quinton catheter for dialysis. Surgical findings included extensive necrosis of the skin, subcutaneous tissues, fascia, and underlying muscles. These findings extended from the medial thigh to the medial malleolus. Surgical debridement yielded 60 x 25 cm of necrotic tissue. A pulse lavage system was used to irrigate the wound and sterile dressing was applied. The patient was brought to the intensive care unit for further management, dialysis, and the plan to return for further debridement.

Patient was brought back to the operating room, hospital days 1 and 2, with continued debridement of necrotic tissue (Figure 2). Wound cultures returned showing growth of Haemophilus influenza serotype a that was sensitive to ceftriaxone and resistant to Bactrim. His antibiotic therapy was

Figure 1. Left lower extremity on presentation; extensive necrotic tissues with multiple bullae observed from medial malleolus to proximal thigh.

Received July 25, 2017. Accepted August 28, 2017.

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changed to ceftriaxone monotherapy. Blood cultures were negative for growth.

On hospital day 7, there was an application of a skin substitute to promote wound healing. After debridement of residual necrotic tissue, portions of ACell MatriStem was applied to the 30 x 100 cm area of exposed wounds and tendons. Sterile dressing was applied.

On hospital day 17, wound exploration was performed. Extensive necrotic and desiccated tissue was found and debrided. Wet to dry dressing was applied.

On hospital day 22, the patient had another irrigation and debridement with the intention of a split-thickness skin graft. Approximately 720 cm² of skin was harvested from the right calf and thigh and applied to the left leg.

On hospital day 27, the patient returned for another wound exploration. The wound was deemed unsuitable for further skin grafting, so approximately 2000 cm² of VAC negative pressure dressing was applied over the exposed wound areas.

On hospital day 30, additional debridement and replacement of the VAC negative pressure wound took place.

On hospital day 37, VAC dressing replacement and wound exploration was performed. Intraoperatively, the left knee joint was found open with synovial fluid leakage with rupture of the quadriceps and adductor tendons. Orthopedics was consulted. They agreed that a complete limb salvage due to the presence of infected tissue in the proximal thigh. Additionally, a complete hip disarticulation has a greater hindrance on mobility and an increase in mortality. It was our goal to stop the spread of infection while attempting to preserve the patient’s quality of life. A BKA or AKA would have been an acceptable compromise between these goals.

With the exposure of the knee joint and tendons, we attempted to salvage the limb with ACell matrix. However, this was inhibited by desiccation and continuing tissue necrosis. Split-thickness skin grafting was then attempted. Unfortunately, poor vascularization over the areas of exposed joints and tendons likely caused the graft to fail. Subsequently, negative pressure wound VAC was placed with the hope to facilitate neovascularization and further wound healing. Wound VAC therapy, in this case, was found to be much more successful than previous methods. However, we were still unable to preserve the areas over the exposed joints and tendons. The rupture of the quadriceps and adductor tendons coupled with open knee joint and synovial fluid leakage ultimately led to no other choices but amputation.

Another remarkable feature of this case is the distinctive organism identified from the wound culture. NF is commonly caused by group A streptococci or mixed facultative/anaerobic bacteria. Haemophilus influenzae is a gram-negative coccobacillus and a common cause of childhood upper respiratory tract infections. H influenzae has also been known to cause meningitis, bacteremia, and pneumonia. H influenzae are either encapsulated (typable) or unencapsulated (non-typable). Six serotypes (a-f) are known and each has capsules made from various polysaccharides. This capsule allows H influenzae to resist phagocytosis and complement mediated lysis. Before the advent of Hib vaccination, it was the most common cause of bacterial meningitis, epiglottitis, and pneumonia in children. With the advent of Hib vaccinations, the rates of invasive Hib infections have been almost eliminated. Unencapsulated H influenzae stains still cause upper respiratory tract infections.

Discussion

In this case, our patient presented with extensive necrotizing fasciitis (NF) of his left lower extremity that resulted in an AKA, despite aggressive interventions to salvage the limb. The decision for amputation was initially considered, but it would have required a complete hip disarticulation due to the presence of infected tissue in the proximal thigh. Additionally, a complete hip disarticulation has a greater hindrance on mobility and an increase in mortality. It was our goal to stop the spread of infection while attempting to preserve the patient’s quality of life. A BKA or AKA would have been an acceptable compromise between these goals.

With the exposure of the knee joint and tendons, we attempted to salvage the limb with ACell matrix. However, this was inhibited by desiccation and continuing tissue necrosis. Split-thickness skin grafting was then attempted. Unfortunately, poor vascularization over the areas of exposed joints and tendons likely caused the graft to fail. Subsequently, negative pressure wound VAC was placed with the hope to facilitate neovascularization and further wound healing. Wound VAC therapy, in this case, was found to be much more successful than previous methods. However, we were still unable to preserve the areas over the exposed joints and tendons. The rupture of the quadriceps and adductor tendons coupled with open knee joint and synovial fluid leakage ultimately led to no other choices but amputation.

Another remarkable feature of this case is the distinctive organism identified from the wound culture. NF is commonly caused by group A streptococci or mixed facultative/anaerobic bacteria. Our patient is a unique case where NF is caused by H influenzae serotype a.

Haemophilus influenzae is a gram-negative coccobacillus and a common cause of childhood upper respiratory tract infections. H influenzae has also been known to cause meningitis, bacteremia, and pneumonia. H influenza are either encapsulated (typable) or unencapsulated (non-typable). Six serotypes (a-f) are known and each has capsules made from various polysaccharides. This capsule allows H influenzae to resist phagocytosis and complement mediated lysis. Before the advent of Hib vaccination, it was the most common cause of bacterial meningitis, epiglottitis, and pneumonia in children. With the advent of Hib vaccinations, the rates of invasive Hib infections have been almost eliminated. Unencapsulated H influenzae stains still cause upper respiratory tract infections.
in both children and adults, but cannot cause disseminated diseases with their lack of polysaccharide capsules.2

In a population study of 91 cases of invasive disease due to \textit{H influenza}, serotype a was the most common serotype of \textit{H influenza}. In children under the age of 5, the average incidence of Hia invasive diseases was 0.8/100 000 with the average incidence of Hib invasive diseases being 0.11/100 100 child-years. This case did not report any cases of soft-tissue infections due to \textit{H influenza}.4

\textit{H influenza} serotype f has also caused invasive infections. Of the 91 patients in a study by Urwin et al, invasive Hif infections carried a 30% mortality rate. Seventy-two percent of these patients had underlying comorbidities, such as diabetes or chronic obstructive pulmonary disease. Eighty-two percent of these 91 cases involved the upper respiratory tract. As before, no soft-tissue infections were reported in this case.5

There have only been 13 documented cases of NF caused by \textit{H influenza} serotypes b, f, and e.6,7 This is the first documented case of \textit{H influenza} type a causing NF, so it is unclear why one serotype was found over another. Most of the patients in previous cases of \textit{H influenza} NF had underlying comorbidities such as immunosuppression, malignancy, and diabetes.6,7 In the study by Azar et al, the patient had a documented case of hypocomplementemia along with immunosuppressants for auto-immune diseases. Complement and immunoglobulin opsonization play a major role in fighting encapsulated bacteria. In particular, deficiency of C3 significantly increases the risk of \textit{H influenza} infections. Interestingly, C5-9 deficiency is associated with increased \textit{Neisseria} infections, not \textit{H influenza}.8,9 Azar et al and Winkelstein and Moxon are the only \textit{H influenza} NF cases that have documented hypocomplementemia, so its role is unclear in this disease process. Immunoglobulin deficiency is known to a wide variety of infections, including \textit{H influenza}. But again, it is unknown if there is a high incidence of \textit{H influenza} NF in patients with immunoglobulin deficiency.7

**Declaration of Conflicting Interests**

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

**Funding**

The author(s) received no financial support for the research, authorship, and/or publication of this article.

**Ethics Approval**

Our institution does not require ethical approval for reporting individual cases or case series.

**Informed Consent**

Verbal informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

**References**

1. Espandar R, Sibdari SY, Rafiee E, Yazdanian S. Necrotizing fasciitis of the extremities: a prospective study. \textit{Strategies Trauma Limb Reconstruct.} 2011;6:121-125. doi:10.1007/s11751-011-0116.
2. Kasper DL, Harrison TR. \textit{Harrison’s Principles of Internal Medicine}. New York, NY: McGraw-Hill; 2005.
3. Agrawal A, Murphy TF. \textit{Haemophilus influenzae} infections in the \textit{H. influenzae} type b conjugate vaccine era. \textit{J Clin Microbiol.} 2011;49:3728-3732.
4. Bender JM, Cox CM, Mottte S, et al. Invasive \textit{Haemophilus influenzae} disease in Utah children: an 11-year population-based study in the era of conjugate vaccine. \textit{Clin Infect Dis.} 2010;50:e41-e46.
5. Urwin G, Krohn JA, Robinson KD, Wenger JD, Farley MM. Invasive disease due to \textit{Haemophilus influenzae} serotype f: clinical and epidemiologic characteristics in the \textit{H. influenzae} serotype b vaccine era. \textit{Clin Infect Dis.} 1996;22:1069-1076.
6. Arnold CJ, Garrigues G, St Gene JW 3rd, Sexton DJ. Necrotizing fasciitis caused by \textit{Haemophilus influenzae} serotype f. \textit{J Clin Microbiol.} 2014;52:3471-3474.
7. Azar MM, Folk D, Henderson T, Mehr S, Vining E, Banach DB. Cervicofacial necrotizing fasciitis caused by \textit{Haemophilus influenzae} serotype e in a patient with Sjogren’s syndrome, systemic lupus erythematosus and hypocomplementaemia. \textit{JMM Case Rep.} 2015. doi:10.1099/jmmcr.0.000040.
8. Tedesco F. Inherited complement deficiencies and bacterial infections. \textit{Vaccine}. 2008;26(suppl 8):I3-I8.
9. Winkelstein JA, Moxon ER. The role of complement in the host’s defense against \textit{Haemophilus influenzae}. \textit{J Infect Dis.} 1992;165(suppl 1):S62-S65.