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Case report

A case report of Actinomyces-associated vulvar necrotizing soft tissue infection: Aspects of multidisciplinary care

Bruce Lee *, Peter Mroz

Rochester General Hospital, Department of OBGYN, 1425 Portland Ave., Rochester, NY, 14621, United States of America

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ABSTRACT

Introduction and importance: Necrotizing soft-tissue infections are life-threatening infections with significant morbidity and mortality.

Case presentation: A 36-year-old female gravida 1 para 1001 with a history of intravenous drug use and homelessness presented to the emergency department for a three-day history of labial pain, swelling, and fevers. A contrast tomography scan revealed necrotizing soft tissue infection of the mons and vulva with subcutaneous gas. Empiric broad-spectrum antibiotics were initiated. She underwent an emergent two-staged surgical debridement and delayed primary closure after 1-month in the hospital. She was discharged to complete a 9 to 12 month course of antibiotics. Multidisciplinary team was central to patient care.

Clinical discussion: Necrotizing soft tissue infections are a diverse group of infections affecting the subcutaneous tissue, muscle, and fascia. Actinomyces is a rare cause and require long-term antibiotics. Early recognition and emergent surgical debridement has been shown to reduce morbidity and mortality. A multidisciplinary approach is critical for proper wound care and healing, pain management, and long-term follow up.

Conclusions: Actinomyces-associated vulvar NSTIs are life-threatening infections requiring early surgical debridement, long-term antibiotics, and multidisciplinary approach.

1. Introduction

Necrotizing soft-tissue infections (NSTIs) are a diverse group of rapidly-advancing and potentially life-threatening infections of the subcutaneous layer, muscle, and fascial tissues. According to a National Inpatient Sample (NIS) from 1998 to 2010, the estimated annual incidence ranges from 3800 to 5800 admissions [1]. The combined obesity and opioid epidemic are direct risk factors in developing NSTIs. Gynecologists will undoubtedly encounter more vulvar NSTIs.

Current mortality rates range from 6 % to 33 %; however they have decreased overtime [2]. National registries such as the National Surgical Quality Improvement Program (NSQIP) quote a 12 % mortality rate [3]. The mainstay of treatment remains the same: early recognition, aggressive intravenous (IV) antibiotic treatment, and most importantly—early surgical debridement. With improved mortality rates, prognostic discussions now revolve around significant morbidity with repeat debridements, medical complications and prolonged hospitalization stays [1]. Given the complexities and severity of disease course, a multidisciplinary approach is essential for prompt treatment and a safe hospital discharge.

We present a significant case of *Actinomyces*-associated vulvar NSTI in a morbidly obese woman with a history of intravenous drug use (IVDU) at a local community hospital. This case report follows guidelines from the SCARE Criteria [4].

2. Case

A 36-year-old female gravida 1 para 1001 presented to the emergency department (ED) for a three-day history of labial pain, swelling, and fevers. Her past medical history is significant for hypertension, body mass index (BMI) 56 kg/m^2, polysubstance abuse (intravenous drug use, opioid abuse, and tobacco), asthma, bipolar disorder, obsessive-compulsive disorder, chronic pain syndrome, and homelessness. Her surgical history is remarkable for a prior cesarean section.

Her vital signs were: body temperature 38.4 °C, blood pressure 135/69, heart rate 121 beats per minute (bpm), respiratory rate 18. Physical exam was remarkable for a large right labial and mons abscess with a small opened wound with drainage (Fig. 1: A).

* Corresponding author.
E-mail addresses: Bruce7ee920@gmail.com (B. Lee), Peter.Mroz@rochesterregional.org (P. Mroz).

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Admission labs were remarkable for mild leukocytosis 16.5 (normal, 4–11 × 10^3/μL), lactic acid 0.6 mmol/L (normal, 0.4–2.0 mmol/L), and hemoglobin A1C 5.6 (normal, 4.2–5.6 %). Aerobic cultures grew *Streptococcus viridians*, *Staphylococcus aureus* and *Enterococcus faecalis*. Anaerobic cultures grew numerous *Actinomyces* species (4+) and *Corynebacterium* species (2+). CT abdomen and pelvis showed moderate soft-tissue inflammatory stranding and subcutaneous gas overlying the pubis and extending into the right labia majora concerning for a necrotizing infection (Fig. 2).

In the ED, she was started on standard sepsis protocol with aggressive IV fluids and empiric antibiotics (vancomycin, piperacillin-tazobactam, and clindamycin). The patient desired to leave against medical advice and our psychiatric assignment officer (PAO) determined that the patient did not have capacity primarily due to lack of insight into her medical condition. In addition, four-point restraints were placed due to combative behavior and security and local police department were involved.

Benign gynecology and general surgery teams were consulted. A two-attending consent by the benign gynecology attending and emergency room physician was obtained for emergent surgical debridement 17 h after initial evaluation. Our approach was a skin-sparing debridement in two stages. Exam in the operating room (OR) showed a 15 × 21 × 9 cm vulvar and mons NSTI and was successfully debrided until there was healthy well-vascularized tissue of the wound bed (Fig. 1: B). The fascia was not involved and care was taken to avoid major underlying vasculature and clitoris. She was taken back to the OR on post-op day (POD) #1 for the planned second-stage debridement (Fig. 1: C, D). Skin-sparing debridement has been shown to be non-inferior to en-bloc debridement

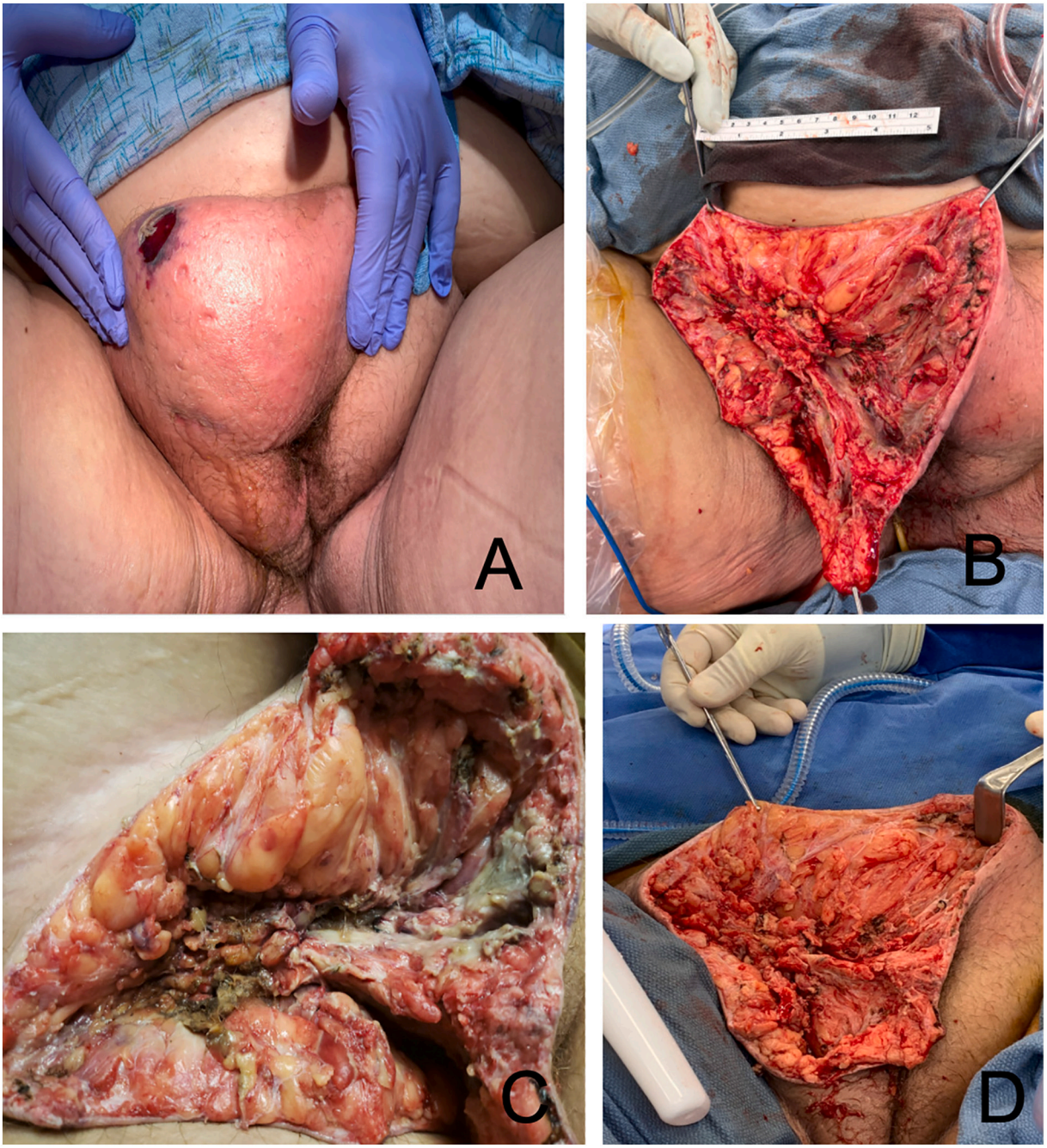

Fig. 1. A) Infected mons pubis and vulvar with a dark circular opening wound. B) First surgical vulvar debridement. C) Vuvlar wound prior to second debridement. D) Second surgical vulvar debridement.

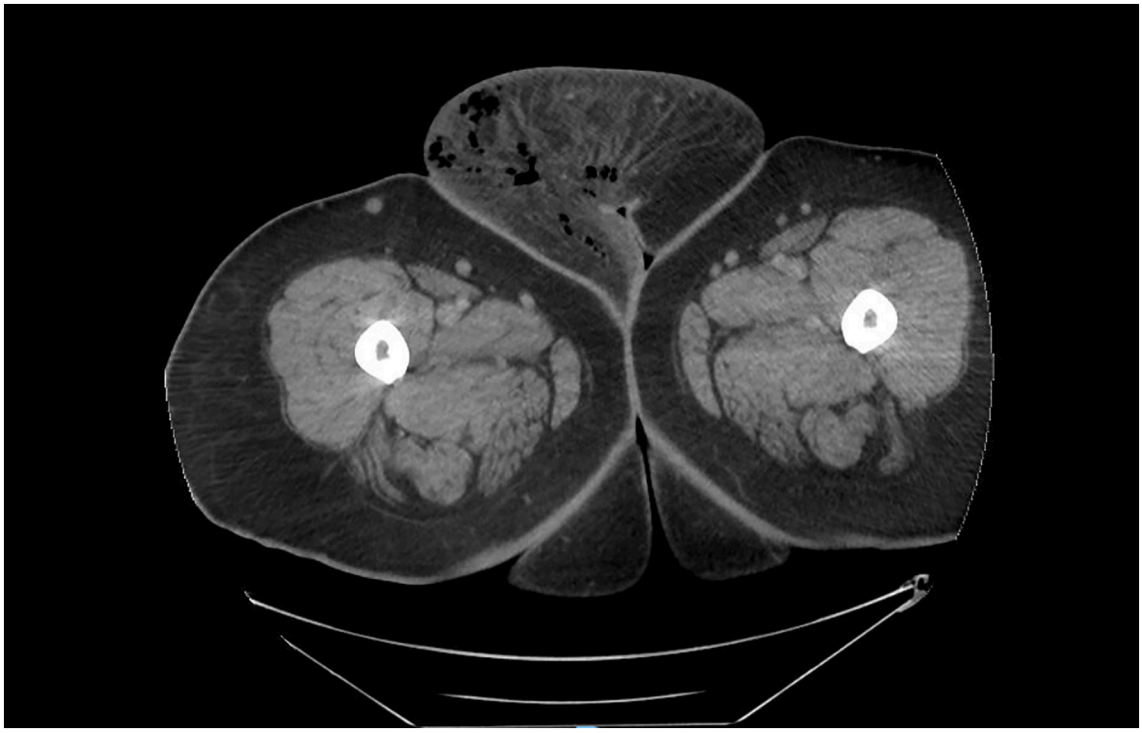

Fig. 2. CT abdomen and pelvis showing soft tissue inflammatory stranding and subcutaneous gas of the pubis and right vulva concerning for a necrotizing infection.

for NSTI [5].

Post-operatively, her wound dressings were changed daily and by POD#10 a Veraflow™ wound vacuum was placed every 2–3 days. Plastic surgery was consulted to perform a delayed-primary closure and mons pubis panniculectomy after one month (Fig. 4). A #19 Blake drain was placed and removed after 7 days. The infectious disease team transitioned her broad-spectrum antibiotics to Augmentin twice daily to complete a 9 to 12 month course due to growth of *Actinomyces* (4+) and *E. faecalis*.

Due to her severe agitation, maladaptive personality, and emotional dysregulation the primary team faced several challenges. Early in her post-operative period, additional four-point restraints were utilized due to safety concerns. IV access was commonly lost. In addition, several attempts to leave from the hospital were made. Wound care was extremely challenging as we suspected intentional wound disruptions which led to vacuum changes up to 2-3× per day. Frequent changes led to pain exacerbations initially requiring IV narcotics.

Psychiatry and palliative care team were essential in managing severe agitation and chronic pain syndrome. This was first managed by a combination of haloperidol and lorazepam. Atypical antipsychotics such as olanzapine and quetiapine proved to be efficacious. Adjuncts such as scheduled trazadone and hydroxyzine were useful to phase out any medications that triggered a reward for acting out. A multimodal pain approach was used with scheduled acetaminophen and non-steroidal anti-inflammatory agents, gabapentin, and oral narcotics as needed. IV opiates were discontinued and then avoided. A suboxone regimen was initiated inpatient with a plan to connect her to an outpatient chemical dependency program.

Multiple multidisciplinary meetings occurred to determine safe and appropriate discharge planning. This included conversations with a local women's shelter, engaging with outpatient chemical dependency, and obtaining appropriate follow up with gynecology. Unfortunately, due to behavioral issues with prior police officers, the patient was transferred to jail. She was loss to follow-up with gynecology.

3. Discussion

NSTIs are a diverse group of infections affecting the subcutaneous tissue, muscle, and fascia. They carry significant morbidity and mortality and require a multidisciplinary approach. Our patient had significant risk factors including class III obesity, IVDU, chronic homelessness, and poor mental health status. It is important for this patient to recognize the life-threatening nature of this disease process and to have a PAO involved.

A case series examined 14 cases of vulvar NSTI and found increasing rates attributed to a rise in obesity, hypertension, and diabetes [6]. This case series also emphasized the importance of early surgical debridement and a multidisciplinary approach to include aggressive intensive care unit (ICU) care and consultations with infectious disease, plastic surgery, and wound care management [6].

The goal of surgery is the complete debridement of all infected and necrotic tissue [7]. A general rule of surgical debridement is to perform surgery within 24 h, however more recent literature suggests within 12 h (and potentially within 6 h) improves morbidity and mortality [2]. Early surgical debridement is not only associated with decreased mortality but also reduce risk of septic shock, number of surgical debridements, and hospital length stay [8,9]. NSTIs often require a two-stage debridement as it is difficult to see all infected tissue grossly, and to return to the OR with a planned second stage debridement to salvage tissue and ensure a complete debridement [5].

For this patient, primary surgical debridement occurred after 17 h with benign gynecology and general surgery team due to PAO assessment.

Given the stability of our patient and lack of ICU beds during the Coronavirus pandemic, she was managed on the general floor. We elected to use a Veraflow™ wound vacuum device. This is a negative pressure wound therapy with instillation and dwell time (NPWTi-d). The basic principles include a facilitated removal of microorganisms, dilution of inflammatory and cytotoxic molecules, wound hydration, and promotion of angiogenesis with intermittent suction [10]. Despite a multitude of wound vacuum changes, there was significant

improvement over the following 4 weeks (Fig. 3: A, B, C, D). Proper wound care is essential as large complex wounds are a significant source of bacteria and driver for nosocomial infections. *Re*-admission rate are estimated at 30 % within 90 days [11].

The Infectious Diseases Society of America (IDSA) recommends empiric treatment of polymicrobial infection against aerobes, anaerobes, and MRSA for NSTIs [12]. Antibiotic duration continues until debridement is no longer necessary, significant clinical improvement, or absent of fevers for 48–72 h [12]. The *Actinomyces* genus colonizes the oropharynx, gastrointestinal tract, and vagina and is found in polymicrobial flora [13].

They can commonly form a chronic suppurative infection leading to abscess formation mimicking pelvic malignancies [14]. *Actinomyces* also do not produce beta-lactams, and thus not useful in combination with beta-lactam inhibitors such as clavulanic acid, except in cases where co-pathogens such as *Enterobacteriaceae* are present [13]. Thus, infectious disease team recommended a 9 to 12 month course.

Lastly, the role of psychiatry and palliative care team to assess

Fig. 3. A) Vulvar wound 1-week post-op. B) Vulvar wound 2 weeks post-op. C) Vulvar wound 3-weeks post-op. D) Vulvar wound 4-weeks post-op.

Fig. 4. Vulvar wound after delayed primary closure by plastic surgery with #19 Blake drain placement.

mental capacity and to develop treatments for acute agitation and chronic pain syndrome were essential. This was challenging given patient's non-compliance and prior substance use history.

Numerous multidisciplinary meetings were completed to develop a safe discharge plan. Despite our efforts, the patient was unexpectedly discharged to jail due to prior conflicts with police officers. This case also highlights the incongruent and non-constructive relationship between the medical and criminal justice system as institutionalized patients can be lost to follow-up. The patient did not show up for outpatient gynecologic care after discharge.

4. Conclusion

Actinomyces-associated vulvar NSTIs are life-threatening infections requiring early surgical debridement and long-term antibiotics to reduce morbidity and mortality. A multidisciplinary team is necessary to address the medical, surgical, and biopsychosocial aspects of patient care and improve outcomes.

Funding

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Ethical approval

N/A.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Bruce Lee performed the initial surgical debridement under the guidance of general surgery and gynecology attendings. He obtained

patient consent and participated in all aspects her post-operative care. He reviewed the patient's chart, performed a literature review, and wrote the manuscript. Peter Mroz obtained surgical consent, supervised the initial surgery, and reviewed the case report and manuscript.

Registration of research studies

N/A.

Guarantor

Bruce Lee.

Declaration of competing interest

The authors report no conflicts of interest.

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