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CASE REPORT

Fournier's Gangrene with Growth of *Actinomyces europaeus*: A Case Report

Shurong Zhang · Yunkai Xie · Yanqiu Wang · Guoyu Jin · Rongtao Cui · Yonghui Zou

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ABSTRACT

Fournier's gangrene (FG) is a rare infectious disease with rapid disease progression and a high mortality rate. We report a case of a 61-year-old female with type 2 diabetes who developed FG caused by *Actinomyces europaeus*. *A. europaeus* is associated with abscesses, decubitus ulcers, and purulent urethritis. Although *A. europaeus* rarely causes FG as the main causative pathogen, we should still be alert to this pathogenic microorganism. To our knowledge, this is the first case report of FG caused by *A. europaeus* mono-infection, and it adds to the evidence that *A. europaeus* has the potential to cause necrotizing fasciitis.

Keywords: Fournier's gangrene; Necrotizing fasciitis; *Actinomyces europaeus*

Key Summary Points

Fournier's gangrene is a rare infectious disease with a high mortality rate.

This is the first case report of FG caused by *Actinomyces europaeus* mono-infection.

Actinomyces europaeus has the potential to cause necrotizing fasciitis.

S. Zhang · Y. Zou (✉)
Department of Gynecological Oncology, Shandong Provincial Hospital, Shandong University, Jinan 250000, China
e-mail: 18769735015@163.com

Y. Xie
Department of Gynecological Oncology, Beijing Obstetrics and Gynecology Hospital, Capital Medical University, Beijing Maternal and Child Health Care Hospital, Beijing, China

Y. Wang
Department of Operating Room, Shandong Provincial Hospital, Jinan, China

G. Jin
Department of Gynecological Oncology, Shandong University of Traditional Chinese Medicine, Jinan, China

R. Cui
Department of Burns and Plastic Surgery, Shandong Provincial Hospital, Shandong University, Jinan, China

INTRODUCTION

Fournier's gangrene (FG) is a rapidly worsening, necrotizing infection of the soft tissues and fascia of the perineum and genital region. During FG, the superficial fascia and subcutaneous tissues cause necrosis, resulting in sepsis and even death [1]. Its common pathogens includes Group A *Streptococcus*, *Bacteroides fragilis*,

Staphylococcus aureus, *Clostridium* species, *Pseudomonas aeruginosa*, *Enterobacteriaceae*, and others [1]. In extreme cases, FG can be caused by *Actinomyces*. *Actinomycetes* is a common genus of opportunistic pathogens found in the oral cavity, gastrointestinal tract, and genitourinary tract [2].

Actinomyces europaeus is one of the subspecies of *Actinomyces*. It was first isolated in humans in 1997 [2]. In 2019, the first case report on necrotizing fasciitis caused by *A. europaeus* and *Actinotignum schaalii* was published [3]. To our knowledge, this is the first case report of FG caused by *A. europaeus* mono-infection. This study was conducted following the 1964 Declaration of Helsinki and its subsequent amendments. Informed consent was obtained from the patient for being included in this case report.

CASE REPORT

A 61-year-old female with a history of type 2 diabetes mellitus and hypertension presented with a soft, painless, 4 cm mass in the perineum without obvious inducement, on March 7 2022. The patient had a history of diabetes for 6 years and was taking metformin irregularly. She did not take hypoglycemic drugs nor monitor her blood glucose recently. However, on March 9, the lesions had spread to the left labia majora and mons pubis. The pain had affected her sleep and motion; she reported no fever or difficulty in urinating. She was managed at the local community hospital with infusion therapy (drug unknown) for painful swollen perineum, but she had no obvious improvement following the infusion therapy, and her symptoms further worsened with increased pain and enlarged lumps. The patient was then advised to present to our gynecological department by her primary care physician on March 15. On the day of presentation, she developed fever and cough with no known aggravating factors.

On admission, her vitals were stable, with a temperature of 37 °C, blood pressure of 112/70 mmHg, respiratory rate of 21 breaths per minute, heart rate of 96 beats per minute, and her random blood glucose level was 23.4 mmol/L. Physical examination showed there was extensive erythema in the left groin, the upper one-third of the inner side of the left thigh, the pubis, and the left labia majora with tenderness, edema, and skin necrosis (Fig. 1A). She underwent baseline laboratory investigations including routine bloods, pus and blood culture, and biochemical liver function tests (Table 1). After 3 days of bacterial culture, on March 20, bacterial culture of the pus detected *Actinomyces europaeus* (Fig. 1C), and blood cultures were negative. After a discussion with the dermatologist, the patient was initially diagnosed as FG according to her clinical manifestations on March 17. After the clinical diagnosis of FG was made, the patient underwent multiple staged debridement and graft skin closure surgery in the lithotomy position (Fig. 1B). On March 18, during the first operation, we found that the necrotic subcutaneous soft tissue extended from the lower abdomen to the anus, and the necrotizing fasciitis involved the perineum extending to both pelvic floor muscles and left hip joint. The necrotic tissue was excised and intraoperative wound cultures grew *A. europaeus*. The postoperative period remained uneventful and she showed improvement in her general condition.

During hospitalization, her antibiotic treatment plan was adjusted several times. On March 15, she was empirically given cefoperazone sulbactam and levo-ornidazole. On March 17, after the clinical diagnosis of FG was made, the antibiotic treatment plan was not changed. On March 18, after the first debridement, cefoperazone sulbactam 1.5 g twice daily was changed to 3 g three times daily. After the second debridement, the cefoperazone sulbactam/levo-ornidazole combination was changed to piperacillin sodium and tazobactam sodium 4.5 g three times daily.

DISCUSSION

FG is a rare but fatal disease, with extremely high mortality. The annual incidence is 1.6 cases in 100,000 and the average mortality is 7.3% [4]. Recognized predisposing factors include advanced age, lowered immune function, diabetes mellitus, and decubitus ulcer [1].

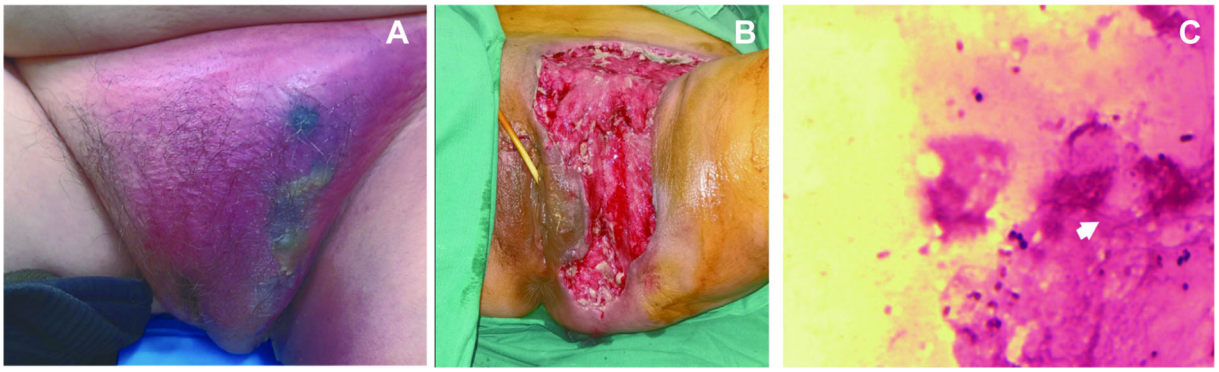

Fig. 1 **A**, Representative images of gangrenous perineum; **B** postsurgical debridement; **C**, bacterial culture of the pus detected the growth of *A. europaeus*

Table 1 Initial laboratory workup on admission

Test	Results	Reference range
White blood cell count	21.73	3.5–9.5 × 10^9/L
Neutrophils count	19.51	1.8–6.3 × 10^9/L
C-reactive protein	234.00	0–10 mg/L
D-dimer	2.71	0–0.5 mg/L (FEU)
Fibrinogen level	8.19	1.75–4.35 g/L
Human serum amyloid A	> 320.00	1–10 mg/L
Bilirubin direct	8.93	0–4 μmol/L
Aspartate aminotransferase	18	13–35 U/L
Alanine aminotransferase	20	7–40 U/L
Bilirubin total	28.99	0–21 μmol/L
Sodium	130.90	137–147 mmol/L
Creatinine	91.16	40–105 μmol/L
HbA1c	11.2	4–6%

Our patient had high risk factors for developing FG, including older age and having poorly controlled diabetes mellitus. FG is characterized by polymicrobial infection. However, our case is different. Bacterial smears of wound pus from our patients found Gram-positive cocci and Gram-negative bacilli, wound cultures showed *A. europaeus*, but blood cultures were negative. Thus, the microbiologists explained that the results of smears were normal flora, and only *A. europaeus* were cultured in an anaerobic environment, and that blood cultures in FG patients are usually negative. Therefore, we concluded that this is a special case of FG caused by *A. europaeus* mono-infection.

To our knowledge, this is the first case of *A. europaeus* as the primary organism causing FG. There are only two reports of *A. europaeus* causing necrotizing fasciitis, one of which was abdominal wall necrotizing fasciitis and the other was *A. europaeus* along with *Actinotignum schaali* causing necrotizing fasciitis [3, 5]. This case report reinforces the link between *A. europaeus* and necrotizing fasciitis. When *A. european* are cultured in specimens, we should be highly alert to necrotizing fasciitis and adjust antibiotics in a timely manner.

Early diagnosis, early use of effective antibiotics, and urgent surgical debridement are important for proper treatment of this disease. Wound cultures of FG often reveal polymicrobial infection of both aerobes and obligate anaerobes, but rarely show *Actinomyces* species. Generally, FG patients receive broad-spectrum antibiotics to cover as many bacterial species as possible. Then, antibiotics should be tailored according to Gram staining and cultures. Classical triple therapy involves generation cephalosporins or aminoglycosides, plus penicillin and metronidazole. Metronidazole and penicillin-based antibiotics are the most commonly used antimicrobials in the past two decades [4].

However, sometimes FG can be caused by atypical pathogens, such as *Actinomyces*. Arshan [6] reported a case of FG caused by *Streptococcus anginosus*, *Actinomyces turicensis*, and *Peptoniphilus harei*. Tongchun [7] reported that FG can be caused by *Actinomyces turicensis*, and Sección [8] reported that *Actinomyces funkei*, *Fusobacterium gonidiaformans*, and *Clostridium hathewayi* caused FG. These cases suggest that atypical pathogens, especially *Actinomyces*, should be considered as potential pathogens of FG. Therefore, in our case, *A. europaeus* particularly may be a suspicious organism causing necrotizing fasciitis. The results of Gram staining and cultures are significant for doctors to effectively manage these patients. However, cultivation of *Actinomyces* is quite difficult, and for this genus, metronidazole generally has poor activity, but β-lactam antimicrobial agents have good activity [9]. Steininger et al. recommended using β-lactam antimicrobial agents to treat *Actinomyces* [9].

CONCLUSIONS

Here we report a rare case of FG induced by *Actinomyces europaeus*, which was effectively treated by early aggressive debridements, broad-spectrum antibiotics, and skin transplantation. FG mainly caused by *A. europaeus* is seldom reported, so it is very difficult to correctly identify. It ought to be regarded as a suspected causative agent to avoid missed diagnosis.

Declarations

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Author Contributions Shurong Zhang and Yunkai Xie assisted in drafting and revising the manuscript. Yonghui Zou and Rongtao Cui performed the surgery together. Yunkai Xie and Guoyu Jin searched all the cases and made the analysis. All authors read and approved the manuscript for publication.

Disclosures Shurong Zhang, Yunkai Xie, Yanqiu Wang, Guoyu Jin, Rongtao Cui and Yonghui Zou have nothing to disclose.

Compliance with Ethics Guidelines Compliance with Ethics Guideline. This study was conducted following the 1964 Declaration of Helsinki and its subsequent amendments. Informed consent was obtained from the patient for being included in this case report.

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