CASE REPORT

UDC: 616.379-008.64-052:617.584-089.873-001.41-08
DOI: https://doi.org/10.2298/VSP220315074P

Above-knee amputation due to necrotizing fasciitis caused by a gas-producing strain of *Escherichia coli* and negative pressure therapy assisted closure of a large open wound

Natkolena amputacija zbog nekrotizujućeg fasciitisa izazvanog sojem *Escherichia coli* koji produkuje gas i zatvaranje velike otvorene rane uz pomoć terapije negativnim pritiskom

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Abstract

Introduction. Nonclostridial gas-forming soft tissue infections (NGSTI) are rare, rapid progressive infections characterized by high mortality and high amputation rates. Surgical debridement is crucial in therapy, and it results in complex wounds that need to be closed in order to prevent secondary morbidity. **Case report.** Herein we present a case of NGSTI in a 68-year-old diabetic patient with acute thrombosis of popliteal artery aneurysm and radiological signs of gas in his right leg and the urinary bladder wall. The infection was caused by a gas-forming strain of *Escherichia coli.* In the early stage of the disease, the patient was treated with antibiotics and femoral amputation. A vacuum-assisted closure (VAC) treatment was applied to close the amputation wound. Administered VAC therapy resulted in primary wound closure without complications 17 days after surgery. **Conclusion.** Negative pressure increases the clearance of fluid and infection from the wound but also increases wound contraction and approximation of skin flaps. To avoid extensive reconstructive surgery, VAC therapy can be a good adjunctive treatment for closing large open wounds in patients with NGSTI.

Key words:
amputation, surgical; diabetes mellitus; escherichia coli; necrotizing fasciitis; wound closure techniques.

Apstrakt

Uvod. Neklostridijalne infekcije mekih tkiva sa pisustvom gasa (NIMTPG) su retke, rapidno progresivne infekcije, koje se karakterišu visokim mortalitetom i visokom stopom amputacija. U terapiji je ključan hirurški debridman, posle koga ostaju kompleksne rane, koje je potrebno zatvoriti da bi se sprečio sekundarni morbiditet. **Prikaz bolesnika.** Prikazan je bolesnik sa NIMTPG, dijabetičar, star 68 godina, sa akutnom trombozom aneurizme poplitealne arterije i radiološkim znacima gasa u desnoj nozi i zidu mokraćne bešike. Infekcija je bila izazvana sojem *Escherichia coli* koji produkuje gas. Bolesnik je u u ranoj fazi lečen antibioticima i amputiran mu je femur. Za zatvaranje amputacione rane primenjeno je vakumom-asistirano zatvaranje (VAZ). Primenjena terapija je dovela do primarnog zatvaranja rane bez komplikacija, 17 dana posle operacije. **Zaključak.** Negativan pritisak povećava klirens tečnosti i infekcije iz rane, povećava i pritisak u rani, kao i približavanje kožnih „flapova“. Terapija VAZ može biti dobar pomoćni tretman za zatvaranje velikih rana kod bolesnika sa NIMTPG u cilju izbegavanja velikih rekonstruktivnih zahvata.

Ključne reči:
amputacija, hirurška; dijabetes melitus; escherichia coli; fasciitis, nekrotizujući; rana, zatvaranje, tehnike.

Introduction

Nonclostridial gas-forming soft tissue infections (NGSTI) are characterized by fulminate widespread necrotizing changes of any soft tissue compartment, production of gas, and systemic toxicity. The disease has an aggressively invasive course, frequent loss of extremities, and mortality as high as 42.9–64.5% [1, 2]. Incidence of severe necrotizing soft tissue infections caused by all known microorganisms has been estimated between 0.4 and 0.53 cases *per* 100,000 people, and only a handful of them are characterized as gas-forming [1, 3, 4]. It is a relatively rare entity and can appear after trauma, surgical intervention, minor injury, and sometimes even spontaneously [3].

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The infection is usually caused by gram-positive cocci, gram-negative rods, or a combination of microorganisms. Of all affected patients, it is most common in those with diabetes mellitus (DM). Most often, it has a short incubation time with sudden onset of pain, development of crepitus, and soft tissue induration; discoloration may also be present. Plain X-rays identify gas in deep tissues, and computed tomography (CT) imaging or magnetic resonance imaging may assess the spreading of infection within muscles. Signs of intoxication can develop rapidly, and many patients present with septic shock at the time of their admission [3, 5].

Treatment consists of prompt initiation of antimicrobial therapy, which consists of broad-spectrum antibiotics until a causative agent is identified, intensive care support, and wide surgical debridement. Delay of surgical treatment is associated with higher morbidity and mortality. Usually, amputation is necessary to control infection, whereas functional limb salvage is rare [5]. After such wide surgical debridement, large open wounds persist. The prolonged open wound period induces wound skin marginal contraction and inversion. To close such wounds, complex surgical interventions are needed and delay in coverage may result in infection and secondary morbidity (suboptimal stump function, deformation, infection of skin graft donor site, necrosis of free flap).

To facilitate and accelerate the closure of open wounds, negative pressure therapy [vacuum-assisted closure (VAC) technique] is described in literature [6, 7]. Positive effects of the VAC technique on open wounds are control of tissue edema by removing tissue fluid, preventing bacterial and fungal colonization, tissue angioneogenesis, and enhancing granulation tissue growth. Although VAC therapy can improve the condition of a wound, it cannot close it completely; therefore, other operations are required for definitive wound closure [6, 8, 9].

Case report

A 68-year-old male with a previous history of DM presented to the Emergency Department with a history of pain in his right lower limb for the past five days. The patient did not have any history of preceding trauma. The pain was dull, aching, with high intensity, and non-radiating. He was not aware of his diabetes status.

On physical examination, he was afebrile, with a pulse of 100 bpm, his blood pressure was 180/100 mmHg, and he had dyspnea. Locally, his right leg was generally swollen and tender. He had local areas of erythema on the medial aspect of his right thigh and the ventral side of his knee. In the pretibial area, skin discoloration was present with crepitations, but also with areas of fluctuation. The popliteal, posterior tibial, and *dorsalis pedis* pulsations were absent. The foot was cold, but there were no ulcers or gangrene signs over the foot.

Laboratory analysis showed red blood cells count of 3.46×10^{12}/L [reference range (RR) 4.44–5.6 × 10^{12}/L], white blood cells count (WBC) of 15.14×10^9/L (RR 3.91–10.0 × 10^9/L), granulocytes count of 12.95×10^9/L (RR 1.8–6.9 × 10^9/L), platelet count of 241×10^9/L (RR 166–308 × 10^9/L), hemoglobin level of 84 g/L (RR 135–169 g/L), and random blood glucose level of 16.9 mmol/L (RR 3.9–6.1 mmol/L). His blood urea was 11.1 mmol/L (RR 3.0–9.2 mmol/L), creatinin 102 µmol/L (RR 62–115 µmol/L), creatinine kinase 8.445 U/L (RR 30–200 U/L), aspartate-aminotransferase 460 U/L (RR 5–34 U/L), alanine-aminotransferase 254 U/L (RR 0–55 U/L), albumin level was 24 g/dL (RR 32–46 g/L), D-dimer 3.348 ng/mL (normal values < 198 ng/mL), sodium level 132 mmol/L (RR 136–146 mmol/L), and potassium level was 3.7 mmol/L (RR 3.5–5.1 mmol/L). Of inflammatory markers, he had C-reactive protein (CRP) of 303.3 mg/L (RR 0.0–5.0 mg/L), procalcitonin of 1.97 ng/mL (normal values < 0.05 ng/mL), and fibrinogen of 8.25 g/dL (RR 2.10–4.0 g/dL).

CT scan of his right lower extremity showed the presence of gas inclusions in the region of his plantar fascia, muscles of the calf, popliteal *fossa*, and medial aspect of his thigh up to his hip. There were also gas inclusions in the muscular layer of the urinary bladder wall. The scan also showed a thrombosed popliteal aneurysm diameter of 60 mm, with no flow distally (Figure 1).

Because of extensive infection and muscle necrosis with inclusions of gas and ischemia, the patient underwent

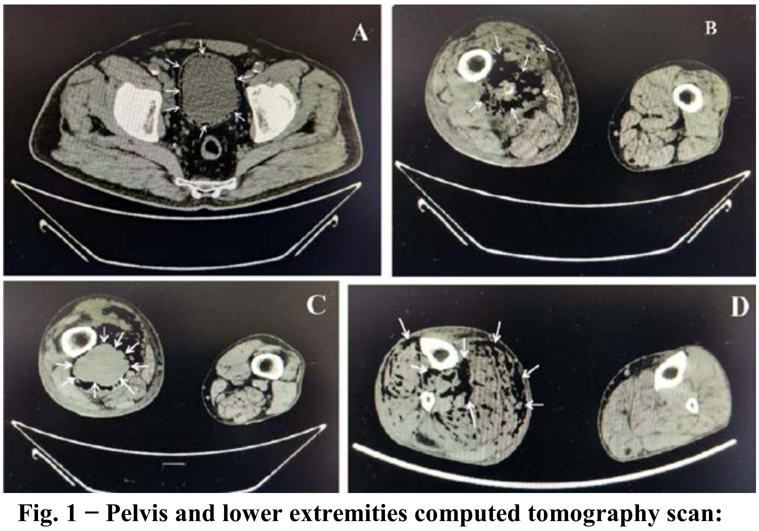

Fig. 1 − Pelvis and lower extremities computed tomography scan: A) Gas inclusions in urinary bladder wall (arrows); B) Gas inclusions in muscles of the right thigh; C) Right popliteal artery aneurysm (arrows) with gas inclusion around it; D) Gas inclusions in muscles of the right calf (arrows).

Fig. 2 – Postamputation wound on the first (left) and fourth (right) postoperative day.

right above-knee amputation with an incision on the medial aspect of the proximal third of the right thigh. Intravenous penicillin G combined with clindamycin was administered for two days until the necrotic tissue and urine culture showed the growth of *Escherichia coli (E. coli)*. According to the antibiogram, the patient was on piperacillin-tazobactam therapy. Furthermore, tissue swabs and samples were taken for testing on anaerobic agents, but the results were negative. On the day when antibiotic therapy was changed, inflammatory marker values were as follows: CRP 194.6 mg/L and procalcitonin 1.05 ng/mL.

In response to intensive therapy, the patient showed a significant improvement postoperatively. On the fifth day after admission, a control CT scan was made, which showed no signs of gas in the urinary bladder wall. On the eighth day, the wound swab was sterile (Figure 2). After ten days of thorough wound debridement and irrigation, the patient underwent two cycles of VAC therapy for wound closure. VAC system was placed over the patient in analgosedation. The wound volume had decreased prominently, and a retraction of muscles had been achieved. We were able to close the wound without tension 17 days after the treatment without infection. The patient was discharged without complications 22 days after admission. The patient was followed up regularly at the outpatient department and without complications.

Discussion

NGSTI is quite a rare, life-threatening condition, and diagnosis is based on clinical findings, radiological examinations, and microbiological investigations [3]. Most of these patients have underlying diseases such as DM or peripheral atherosclerotic vascular diseases [8–10]. However, to the authors' knowledge, there are very few reports on patients with such conditions and concurrent peripheral artery aneurysms or acute ischemia [11]. Likewise, in the available literature, it is rare for *E. coli* to produce gas and severe deep soft tissue infection [12, 13].

In the presented case, multiple tissue sample cultures showed the growth of *E. coli* as the monomicrobial causative agent. Gas inclusions were present in the lower limb and the muscle layer of the urinary bladder wall. A possible explanation for gas in the urinary bladder wall is that this was the primary site of infection with a facultative anaerobic strain of *E. coli*, which was in the surrounding of glucosuria [14], and that it was secondarily transferred to a skin defect on the right lower limb. Moreover, due to a thrombosed popliteal artery, limb ischemia further contributed to bacterial growth.

The most important in therapy are surgical intervention, antibiotic therapy, and intensive care support [3, 15]. Surgical intervention consists of surgical debridement of all necrotic tissue. In many cases, NGSTI surgical treatment of involved extremities consists of amputation and/or fasciotomy. Amputation might be beneficial in cases where compromised neurovascular supply exists and when attempts to salvage the involved limb may lead to metabolic overload and secondary organ failure [5, 16]. In this infective setting, an open stump wound is necessary for wound debridement and infection clearance. However, this open surface results in exposing muscles and soft tissues with wound marginal skin contraction and inversion, thus aggravating the approximation of wound skin flaps. This condition diminishes the possibility of later wound closure.

In this case, there were clinical and radiological signs of widespread infection and, simultaneously, signs of acute critical limb-threatening ischemia because of a thrombosed popliteal artery aneurysm. Since then, reconstructive vascular surgery treatment was associated with extremely high risk in this concurrent infective and ischemic settings. It has been decided that the above-knee amputation be done as a life-saving procedure without prior fasciotomy.

Lee et al. [6] showed that the restoration of the tissue pressure provided by fascia and skin is the key to large open wound closure. Following the same principle, we decided to apply the VAC system on the postamputation stump. We achieved it by fixating a sponge on the muscle fascia layer to achieve the negative pressure on muscles while preserving marginal skin and subcutaneous tissue. The VAC therapy suction pressure was set at 100 mmHg to increase tissue pressure and wound fluid removal while maximizing wound contraction and microvascular blood flow [6, 17]. Centripetal compression effect enhanced approximation of skin flaps.

In our patient, there was no skin necrosis, and the administration of negative pressure therapy facilitated primary wound closure. For successful contouring of wound surface area, an appropriate wound preparation was vital for successful closure. Negative pressure was acting twofold; it was involved in wound bed preparation and infection clearance.

Hyperbaric oxygenation as an adjunct treatment [3, 15, 18] has been discussed, but it has been decided not to be used because of clinical and radiological signs of good response to previous therapeutic modalities and in order to accelerate closure of stump wounds by administering negative pressure therapy.

Conclusion

We presented a rare case of necrotizing fasciitis with gas inclusions caused by a gas-producing strain of *E. coli* with concurrent thrombosed popliteal aneurysm and acute limb ischemia, which was treated with above-knee amputation and modulated negative pressure therapy for closure of the large open wound. Negative pressure therapy decreases the open wound area and minimizes the necessity for secondary operation after treatment of NGSTI.

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Received on March 15, 2022
Revised on June 16, 2022
Accepted on July 5, 2022
Online First July 2022