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Case Report

Uncommon Locations of Gas Gangrene Treated Successfully With Surgical Debridement and the Vacuum-Assisted Closure Device

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Gas gangrene is a life-threatening condition implying necrosis of dermis and hypodermis, along with necrosis of the superficial muscular aponeurosis. Fournier s gangrene is a subtype of the disease affecting the perineal and genital area. The aim of this study is to analyze the clinical presentation, diagnosis, medical, and surgical treatment of three cases of gas gangrene affecting uncommon locations in the human body, treated with extensive surgical debridement followed by the vacuum assisted closure method in two of these cases. Three cases of gas gangrene affecting uncommon locations treated surgically in our Department are presented. In one case the perineal and scrotal region was infected with invasion of the lateral abdominal wall and the peritoneal cavity. In the second case the axillary regions were infected bilaterally and in the third case the left axillary and subscapular regions were infected after a left arm disarticulation. All cases were treated successfully with successive surgical debridement and/or the vacuum-assisted closure method. Gas gangrene is a curable disease if diagnosed early and treated effectively with successive surgical wound cleaning and debridement. The vacuum assisted closure method can be helpful in promoting wound healing.

Key words: Gas gangrene – Fournier's gangrene – Debridement – Vacuum-assisted closure method

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Fournier's gangrene is a synergistic polymicrobial necrotizing fasciitis of the scrotum and perineum that can possibly spread to the lateral abdominal wall. Occasionally the infection runs a fulminant course with a gradual destruction of the fascia approaching 2 to 3 cm/h[1] leading to extensive soft tissue necrosis and often sepsis.

The first description of the disease has been attributed to Avicenna (1025).[2] In the 18th century, Baurienne reported a pediatric case,[3] but the first documented clinical case was reported by Jean Alfred Fournier in 1883, a French venereologist, who gave the disease its eponymous name.[4] In his 4-case report, he described the condition as a fulminant gangrene of the scrotum and the perineal region with a sudden onset, rapid extension and excessive morbidity, in the absence of any predetermined cause.

Up to present, more than 1500 cases have been reported in the literature under various terms: necrotizing fasciitis, gas gangrene, necrotizing cellulitis, infectious gangrene of the scrotum and the perineum, clostridial cellulitis, and synergistic infection of the perineum.[1,5] Also the age pattern has changed: Fournier described a young, otherwise healthy man, whereas today, the majority of cases occur in elderly or immunocompromised hosts.[6]

Occasionally necrotizing fasciitis (NF) may start as a local infection below the subcutaneous tissue and the fascial planes in other parts of the body. Thus it may primarily affect the upper or lower extremities or the axillary regions. The pathophysiologic mechanisms are in all cases identical: both anaerobes and aerobes act synergistically via the action of enzymes such, as collagenases, hyaluronidases, to invade and destroy fascial planes. This process rapidly leads to tissue hypoxia, vascular thrombosis, and irreversible necrosis.

In this report we present 3 cases of NF in uncommon locations in the human body treated successfully with repeat surgical debridement followed by the vacuum-assisted closure (VAC) technique[7] in 2 of the cases for improving wound healing.

Case 1

A 42-year-old male patient was admitted to the Emergency Department with a history of high fever (40°C) since 1 week before, and 2 inflamed ulcers with necrotic material in the gluteal regions bilaterally, with excessive edema in the scrotal area and the inguinal regions. The patient had undergone incision and drainage of 2 bilateral perianal abscesses 4 days before. At the Operating Department, 2 longitudinal incisions were done in the gluteal regions bilaterally uncovering extensive necrosis of the subcutaneous tissue and the aponeuroses spreading to the scrotal and the inguinal areas. After surgery, the patient's general condition improved and the fever subsided.

Four days after surgery the patient's general condition deteriorated, presenting with temperature elevation, abdominal distension, and respiratory distress. An abdominal and pelvic computed tomography revealed edema in the muscular and aponeurotic planes in the lateral abdominal wall bilaterally, intestinal paresis, and peritoneal fluid collection in the pelvis. The patient was transferred to the operating theater where abdominal exploration was done, revealing peritonitis due to spread of the inflammatory process through the lateral abdominal wall to the peritoneal cavity. Two oblique incisions were done in the lateral abdominal wall bilaterally followed by placement of drainage tubes. Saline infusion and curettage of the abdominal cavity followed. A prophylactic straightforward loop ileostomy was done to avoid continuous spillage of the gluteal wounds at defecation, since it was technically easier to perform than a sigmoidostomy. The patient was transferred to the Intensive Care Unit where he remained intubated in severe septic condition for almost 2 months. Worsening general condition and high temperature spikes urged to continuous bring backs to the operating theater. At surgery cleansing and irrigation of the peritoneal cavity and the wounds of the perineal region and the lateral abdominal wall was performed. A vacuum-assisted closure (VAC) device was placed at the abdominal wound, above the viscera, under suction to absorb the pus and necrotic material from the wounds (Fig. 1). This reduced the patient's need for the regular every-other-day bring-backs in the operating room and gradually improved the patient's general condition. The patient was extubated after 1 month. The device remained in the patient's abdomen for almost 3 weeks and after a total 70-day staying in the ICU the patient was transferred to the surgical ward. The wounds gradually healed and the patient was discharged in good condition 95 days after admission. One and a half years later the patient was admitted again for reconstruction of his huge abdominal hernia. A large mesh was placed intraperitoneally and the patient recovered soon after. At present, 3 years after

Fig. 1 In the first patient a vacuum-assisted closure device was placed in the open abdominal cavity to absorb the peritoneal fluids.

the first operation, he is in excellent physical condition and able to work.

Case 2

A 70-year-old diabetic (type II) patient was admitted to the Emergency Department because of 2-month history of dermatitis. The patient was under corticosteroid treatment for the last month without clinical improvement. His past medical history included hypertension under treatment.

He was originally hospitalized at the Dermatology Department and treated with high doses of corticosteroids and levocetirizine dihydrochloride, hydroxyzine, NSAIDs, and fluconazole for his foot dermatophytosis, but his clinical condition deteriorated.

At the 15th day of hospitalization the patient became febrile and presented erythema, calor, circumscribed swelling, and excessive tenderness in the left forearm and to a lesser extent to the right. Laboratory tests revealed elevated C-reactive protein and procalcitonin level, and also elevated lactic acid levels. Blood and urine cultures were obtained and piperacillin-tazobactam and vancomycin was administered after consultation by an infectious disease specialist.

An ultrasound was also performed and no sign of air in the soft tissue was observed. In addition no sign of thrombosis or ischemia of the upper extremity was confirmed.

The next 48 hours the infection expanded to the left elbow with excessive swelling and tenderness. The patient was transferred to the Surgical Department. Vancomycin was replaced with linezolid due to elevated creatinine levels. CT revealed the presence of air in soft tissues (left deltoid and subscapularis muscle) and between pectoralis minor and pectoralis major muscles, along with inflammation of the dermal and subcutaneous areas locally. Antibiotic treatment converted to meropenem, daptomycin, and clindamycin due to clinical severity and isolation of methicillin-resistant staphylococcus aureus from blood culture.

The patient underwent immediate surgical intervention. An aggressive debridement of the posterior venter of the left biceps brachii muscle was done. Extensive fasciotomy and necrosectomy was performed at the areas of axillary cavity, posterior to the left acromion, as well as in a third cavity toward the left elbow. Pus, aspiration fluid, and tissues were sent for culture. All opened cavities were copiously irrigated with hydrogen peroxide, saline, povidone iodine. Three Penrose drains were placed and the wound was covered with elastic dressings. After the initial debridement the wounds were carefully monitored and dressing changes were done twice daily. In the course of the next 10 days the wound stabilized and fresh granulation tissue formed but the patient presented swelling, erythema, exquisite tenderness in the right elbow, and a new episode of fever with elevated CRP levels. A sequential CT scan revealed deterioration of previous findings regarding the right arm with presence of air in the soft tissues, especially posterior of the triceps muscle. The patient was surgically treated with extended fissions of necrotized tissues, fasciotomy, and debridement of the right arm (Fig. 2). Furthermore, the incision of the left arm was extended to the left axilla. All pus, aspiration fluid, and tissues were sent for culture. All wounds were copiously irrigated with hydrogen peroxide, saline, povidone iodine, and elastic dressings. The patient made an encouraging recovery during the following days. The wounds improved within the following 2 weeks and the patient was discharged at 3 weeks after surgery in good condition.

Case 3

A 63-year-old patient, who had undergone a closed strain reduction of the left shoulder, presented to our Emergency Department with ischemia of the left arm, as a result of axillary artery injury and oligemic shock. The patient was diagnosed with partial tear of axillary artery, thrombectomy with a Fogarty

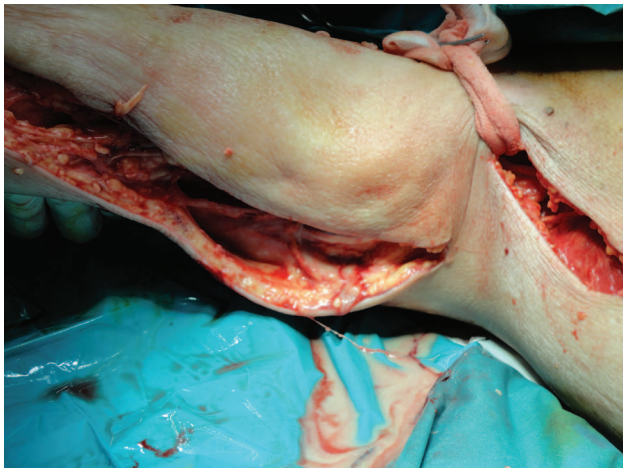

Fig. 2 At the second operation the second patient underwent extended fissions of the necrotic tissues, fasciotomy, and debridement of the right arm.

catheter was done followed with reconstruction of the artery with a venous graft from the saphenous vein. The patient's condition deteriorated; computed tomographic scan (CT) was done immediately and the findings were: extensive left axillary hematoma, which extended to major and minor pectoralis, left latissimus dorsi, and left pronator muscle edema; obstruction of the ipsilateral axillary artery and left lower lobe atelectasis.

The patient was hospitalized because he had signs of hypoxia, metabolic acidosis, rhabdomyolysis, and oliguria. In addition, the patient was hemodynamically unstable and, a couple of days later, the patient had a new episode of shock. Although he was under antimicrobial therapy, and his hematoma had been drained, the ischemia of the left arm and the associated septic syndrome were persistent, necessitating amputation of the left upper limb.

The patient remained septic because of the infected stump of his left arm, and a few days later he underwent disarticulation of the left shoulder, followed by surgical debridement due to abscess formation in the surgical wound (Fig. 3). Blood cultures were positive to multiresistant strain of *Klebsiella pneumoniae* and *Escherichia coli*. The patient's condition during the next days improved, with good communication after interruption of sedation, hemodynamic stabilization, recovery of renal function, but 20 days later he had a new septic episode and fall of hematocrit. A new CT chest / abdomen showed: Collection at the surgical field and colitis, which was confirmed afterward with

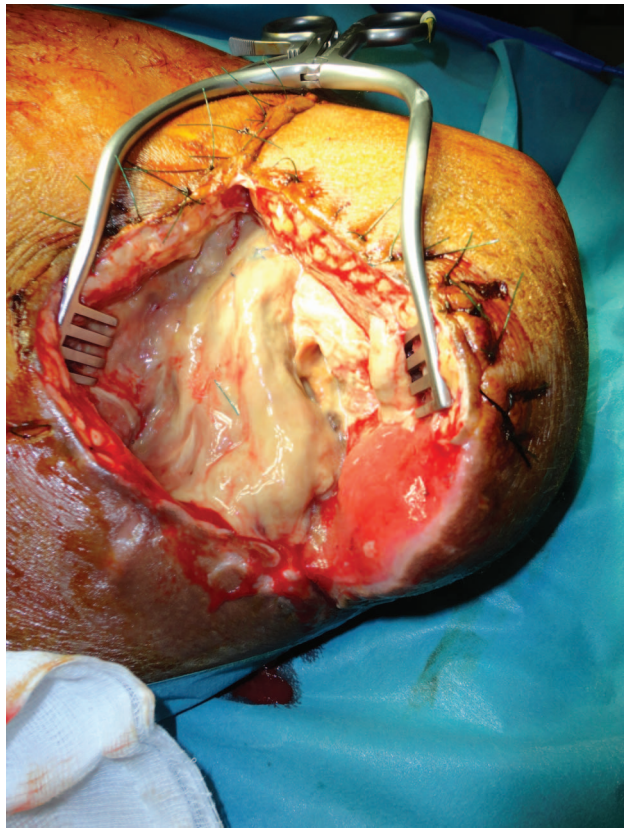

Fig. 3 The third patient underwent surgical debridement of the stump of the left arm.

colonoscopy. Colitis was treated successfully with interruption of enteral feeding. The patient underwent another surgical debridement 2 days later and was transferred back to the ICU. *Candida non albicans* was isolated in wound cultures.

During the following days, the patient showed progressive improvement, undergoing bench wound cleansing; the VAC device was used to improve wound healing. He underwent another CT 6 days later, which showed improvement at the surgical field, and the tracheostomy tube was occluded. The VAC device remained for 2 weeks. He completed the antimicrobial and antifungal treatment (last figure: gentamicin, tigecycline, liposomal amphotericin B). The patient was instructed to undergo daily physical therapy. Two months after admission he was discharged home in acceptable condition.

Discussion

NF, a potentially life-threatening condition, implies necrosis of dermis and hypodermis, along with

necrosis of superficial muscular aponeurosis.[8] Fournier's gangrene is a subtype of NF that affects the perineal and genital area, which can possibly spread to the lateral abdominal wall. The incidence of NF in United States is estimated at 500 to 1000 cases annually,[9] and its prevalence globally has been reported to be 0.40 cases per 100,000 population.[10] NF is more common in middle-aged and elderly patients,[11] and the male-to-female ratio of NF is 3:1. The median mortality rate of NF is 32.2%,[12] but it may be even higher, up to 76%.[13] Mortality rate of Fournier's gangrene in particular is much lower (median mortality rate 16%).[14] Generally without treatment the mortality rate approaches 100%. According to Anaya *et al*[15] infection of the extremities are the most common site of NF (57.8%), followed by the abdomen and the perineum. However, NF of the upper limbs is rare, compared to that of the lower limbs,[16] and NF in the axillary region is extremely rare.[17]

There are numerous preexisting conditions associated with an increased risk of NF. Diabetes mellitus is the most common predisposing factor, as over half of patients with NF, have diabetes.[16,18,19] However, in Fournier's gangrene the presence of sepsis is thought to be the only significant independent risk factor for mortality.[20,21] Other common comorbidities are chronic alcohol disease,[18] immunosuppression, (especially in patients with inflammatory bowel disease),[21] age,[1] and malignancy, particularly leukemia.[22,23]

The natural history of NF begins with the microbial invasion of the subcutaneous tissue and the skin, which can occur either directly through an external trauma or a surgical wound, or through bacterial invasion from a perforated viscus.[11,24] The infection develops from the hypodermis or the superficial fascia,[25] due to the synergistic activity of the different microorganisms producing various toxins. These toxins can cause prolonged vasoconstriction in the dermal capillary network that may lead to microvascular thrombosis of the skin and the subcutis perforators, as well as macrovascular thrombosis of the digital arteries, in case of infection of the upper limb. As a result, tissue ischemia may occur, and the infection disseminates causing cutaneous necrosis.[25]

An early diagnosis of NF is the major challenge for clinicians, as with instant surgical treatment the mortality is reduced to 38%.[26] Moreover, a delay in diagnosis can be fatal, because it has been associated with higher mortality rates, more extensive surgery, and higher rates of amputation, especially in case of upper limb infection.[22,27] There are 4 vital sign abnormalities strongly suggesting NF. These are fever (44%), tachycardia (59%), hypotension (21%), and tachypnea (26%).[28] The typical early clinical picture includes erythema, swelling, tenderness to palpation, and local warmth. As the infection progresses, skin ischemia with blisters and bullae may appear leading to tissue necrosis, skin anesthesia, and crepitus.[29–31] Pain is usually proportionless to lesion severity.[22,23,26] Gradually the pain becomes more severe, especially when it affects the abdominal wall, although it still remains disproportionate to the clinical picture.[32] If diagnosis is delayed, the patient may develop septic shock. When the infection is located in the upper limbs the general condition of the patients is better.[20] Patients with an accompanying disease will have a worse clinical status, because of additional symptoms of these diseases.[11,33]

Certain radiologic examinations must be done to establish diagnosis. Ultrasonography may provide useful information about the nature and extend of the infection, especially when diagnosis is unclear.[34] The most significant finding in ultrasonography is the hyperechoic foci with reverberation artifact and dirty shadowing in the site of infection,[35] representing the subcutaneous gas. However computed tomography (CT) and magnetic resonance imaging (MRI) are more sensitive and specific and therefore are the main diagnostic tools. A CT scan can show the extent of tissue infection, fascial swelling, inflammation, and gas formation. The same finding can also be found in a MRI scan, but it is not often used, because of its high cost.

Broad spectrum antibiotic coverage is always required but has a limited value because tissue ischemia does not allow proper delivery of antibiotics to the target issue.[36] Early and aggressive drainage and debridement are the mainstays of treatment. Surgical debridement should be done always as soon as the diagnosis is established with the clinical and the CT findings. An incision over the infection site is made, under local anesthesia, to allow washing with hydrogen peroxide and saline solution. At the same time, subcutaneous drainages (Penrose) are inserted. Necrotic skin should be excised, while the noninfected skin remains unattached.[37,38] Surgical debridement must be repeated every 24 to 48 hours, depending on the patient's clinical status.

Fournier's gangrene requires special consideration, as the infection can spread into the scrotum, inguinal region, and lower abdominal wall. In some

cases, as in our first case, it is necessary to perform a diverting colostomy, or orchiectomy. Skin incision in the lateral abdominal wall must be performed longitudinally along the muscle-fascial layers, until healthy fascia is encountered.[18] Postoperative wound management on the lateral abdominal wall consists of successive dressing changes every 24 hours to 48 hours, until the wound is clean. After wound stabilization, abdominal wall and soft tissue defect reconstruction may be required.[39]

NF of the axilla also requires special consideration. In that case, a delay in surgical debridement will be proved highly lethal, even more than an infection in a common site.[40] The axillary region is rich in blood vessels and lymphatics, and, as a result, the infection can be spread rapidly to distant sites. Another consideration in surgical treatment of axillary NF is the need to avoid axillary contractures, by covering the exposed neurovascular network.[17] Again, the surgical reconstruction of the wound is essential for successful wound closure.

Special attention should be paid to infection of the upper extremities. The extent of debridement is very important. Additional fasciotomies of all fasciocutaneous bridges should be performed in cases with widespread infection.[36] The incision proceeds in a longitudinal manner, until healthy fascia is founded. After examining tissue's viability, vigilant hemostasis should be performed.[36] There are certain criteria for amputation of the upper limb,[41] and these are concurrent disease with high anaesthetic risk (such as diabetic neuropathy), extensive soft tissue necrosis with involvement of underlying muscles, presence of shock, concurrent vascular insufficiency, and fulminant infection. The patient in Case 3 had met the criteria of amputation. Postoperative wound management is the same as the other cases, but in the treatment of the upper limb there are some extra points to mention. After initial surgery, the wound must be carefully examined under general anaesthesia every 24 hours, to assess the tissue viability and the need of amputation.[36] Consecutive debridement must be performed, because the necrotizing infection of upper limb is rarely eradicated after a single debridement.[36]

Lately, many surgeons worldwide have started using the vacuum-assisted closure therapy, for earlier and effective wound closure. The VAC therapy consists of a polyurethane sponge, which is placed over the wound (its size is adjusted to wound size). This sponge, which also has a suction tube, is usually covered with a second sterile, adherent, occlusive dressing. Evacuation is applied to the sponge using a portable pump. The dressing must be changed every 24 to 72 hours. The VAC therapy is beneficial for wound management; reduction of the wound area and the formation of new granulation tissue are the most prominent benefits of this therapy.[7,42] Other benefits, such as effective wound cleaning by continuous sucking of the exudate, make VAC a promising adjuvant therapy for wound closure.

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