Chapter

Fournier's Gangrene of the Shoulder Girdle

Gyoguevara Patriota, Luiz Marcelo Bastos Leite, Nivaldo Cardozo Filho, Paulo Santoro Belangero and Benno Ejnisman

Abstract

Fournier's gangrene is uncommon, a high-mortality infection that affects the subcutaneous tissue with rapidly progressive necrosis. Reports on cases involving the shoulder girdle are more rare. Similar to the presentation on other regions of the human body, fundamental is early diagnosis and surgical intervention.

Keywords: Fournier's gangrene, necrotizing fasciitis, shoulder girdle infection, acute necrotizing, shoulder

1. Introduction and epidemiology

Fournier's gangrene is known by a variety of other names, necrotizing fasciitis (NF) such as hospital gangrene and hemolytic streptococcal gangrene, among others [1]. Necrotizing fasciitis (NF) is a rare and serious infection, characterized by extensive and rapidly progressive necrosis. Around 500–1500 cases/year are reported in the United States [2] but do not exist studies with profile in Brazil. It has an estimated greater involvement of males (3:1) at a mean age of 50 years. The predominant region of the body is in the perineal area. The mean mortality rate is high (32.2%), and if untreated, it can reach 100%.

Fournier's gangrene is a death-threatening infection caused by aerobic and/or anaerobic microorganisms. It affects the fascia and subcutaneous tissue with microcirculation thrombosis and rapidly progressive necrosis of the skin in the affected region (evolution reaches 2–3 cm/h) [3–6].

Reports on cases involving the shoulder girdle are uncommon. There are only eight cases of Fournier's gangrene (on the shoulder girdle) described in the literature, beginning after surgical procedures in the shoulder (arthroscopy or osteosynthesis) [7–9], or intra-articular infiltration in the shoulder (with corticosteroids) [10], or after closed trauma [11–13], or even without any trauma reported [14]. The description of two cases of necrotizing fasciitis after closed trauma, without the presence of injuries or immune depression conditions, intrigues and alerts us to the possibility of occurrence in any person. Fundamental is the early diagnosis and intervention in early surgery.

2. Etiology

The acute necrotizing inflammatory process initially affects the deep subcutaneous tissue and the fascia. The most superficial tissues and the skin are affected secondarily, due to vascular injury, thrombosis, and ischemia—resulting from the action of pro-inflammatory cytokines, proteinases, and endothelin. The destruction of subcutaneous nerves occurs in advanced stages. Initially described as a disease of unknown cause, it is now known that an underlying pathological process can be found in most cases of Fournier's gangrene; nonetheless, in a significant number of patients, the cause cannot be determined [15–17]. Therefore, a careful investigation can indicate the point of entry, which is located primarily in the digestive tract, in cutaneous affections, in the urogenital tract, or in cutaneous affections [18]. Eke et al. [16], in 2000, published a series of 1726 cases published cutaneous conditions accounted for 24% of the cases. The agents most associated with NF are group A beta-hemolytic *Streptococcus* and *Staphylococcus aureus*. However, other pathogens have already been linked to this disease, namely, *Clostridium perfringens*, *Peptostreptococcus*, Enterobacteriaceae, Proteus, *Pseudomonas*, and *Klebsiella* [16].

The most commonly observed comorbidity is diabetes mellitus, with a prevalence of 40–60%. Other common comorbidities include hepatic cirrhosis, immunodeficiencies, heart failure, systemic lupus erythematosus, obesity, alcoholism, hypertension, Addison's disease, and peripheral vascular disease. However, NF can present in healthy individuals, without comorbidities [19].

3. Classification

There are two classifications for necrotizing fasciitis. The US Food and Drug Administration (FDA) classifies NF according to its microbiological characteristics: type I, aerobic/anaerobic polymicrobial pattern (streptococci, staphylococci, enterococci, *Bacteroides*); type II, only one agent (*S. aureus* or more commonly group A beta-hemolytic *Streptococcus*) and less aggressive lesions, accounting for 10–15% of the cases; and type III, gastric myonecrosis and necrotizing fasciitis caused by *Clostridium perfringens* (less than 5% of the cases) [20].

Féres et al. [21] proposed an anatomic classification. This classification considers two relevant criteria: extension of the necrosis area and correlated it with mortality; these authors defined four groups (increasing mortality), in which group I presented a 12.5% rate, while the mortality rate in group IV was 68.75% (**Table 1**).

Groups	Description	Mortality (%)
Group I	Necrosis of the anterior perineum, scrotum, and penis or vulva	12.5
Group II	Group I + posterior perineum, perianal region up to 7 cm in diameter, rectum, and perirectal fat	34
Group III	Group II + sacral region, gluteal, inguinal region, and necrosis of the penis	37
Group IV	Group III + abdominal wall, suprapubic region, flank, thoracic wall, axillary region, and retroperitoneum	68.75

Table 1.
Anatomical classification of the necrosis area and correlation with mortality in Fournier gangrene. Féres et al. [21].

4. Diagnosis

The diagnosis is eminently clinical and corroborated by surgical findings, which include low adherence of the subcutaneous tissue, observed to the surgical manipulation, absence of bleeding, and liquefaction of the subcutaneous fat. Due to its severity and speed of evolution, Fournier's gangrene is an emergency. The clinical diagnosis must be suspected of the classic triad, pain, edema, and erythema with fever/tachycardia as soon as possible so that early treatment can be initiated. Necrotizing fasciitis could evolve rapidly with necrotic tissues and hemorrhagic blisters [19]. Patients may present laboratory abnormalities such as elevated serum urea (more than 18 mg/dl), serum creatinine (more than 1.2 mg/dl), leukocytosis (>20,000 WBC/mm^3), and CPK (more than 600 μ/l) [22].

Furthermore, imaging exams may also be used, such as radiographs (subcutaneous gas formation—low sensitivity and specificity), ultrasonography (has little practical use), computed tomography (lesion extension and gas formation), and magnetic resonance imaging (more accurate but more costly) [23]. CT provides additional information, such as asymmetric thickening of the fascia and changes in subcutaneous fat, as well as the presence of gas and abscesses. MRI, on the other hand, is considered superior to other imaging methods. It has high sensitivity and allows to define the area of necrosis of the fascia and to schedule the surgical procedure. The absence of changes in the deep fascia practically excludes in the diagnosis. It is worth mentioning that the critical condition of the patient often makes transportation impossible to perform the exam, limiting its use.

Culturing the debrided tissue is important in order to guide antibiotic therapy [19]. Biopsy of the fascia is considered the gold standard for diagnosis and should be performed in all patients during debridement, even in those whose macroscopic appearance is normal.

5. Case report

A 42-year-old female patient [12] who had previously been a victim of a motorcycle accident was attended to at a hospital unit in the interior of the state, diagnosed with a fracture of the middle third of the clavicle (Allman's group I) with deviation (>2 cm).

The physical examination showed no neurovascular deficit, no imminence of bone exposure at the fracture site, or apparent deformity, but she presented with right shoulder abrasions (posteriorly). Initially she was medicated with analgesics but has not received orientation about the use of a sling or necessity of follow-up with a specialist.

After the trauma (2 weeks), she presented with fever, local hyperemia, pain, and fever, requiring hospitalization (city of origin). She evolved local abscess (right clavicle region) and followed by spontaneous drainage of a purulent secretion (through a small orifice). Seventeen days after trauma, the patient underwent abscess drainage with 0.9% saline solution (in the ward) but no debridement (cultures/swab wasn't collected) (**Figure 1**). Results of laboratory exams are as follows: white blood cells (WBC), 2000/mm^3; erythrocyte sedimentation rate (ESR), 25 mm/h; and C-reactive protein (CRP), 11 mm/dl. At this stage, intravenous antibiotic therapy was initiated (with clindamycin 600 mg 8/8 h, metronidazole 500 mg 8/8 h, and ceftriaxone 1 g 12/12 h) and was transferred to a referral hospital in orthopedics surgery in the city of Salvador-BA (Brazil).

Despite the use of intravenous antibiotics, she evolved with toxemia, sepsis (heart rate (HR), 110 bpm; respiration frequency (RF), 26 ripm; temperature, 38.5°C), and an extensive lesion of the right hemithorax and base of the neck (with necrosis) and clavicle bone exposure but no neurovascular alterations (**Figure 2**). Their exams evolved worse (21,000 WBC/mm^3; ESR, 44 mm/h; CRP, 20 mm/dl, creatinine, 1.3 mg/dl; urea, 48 mg/dl; and CPK, 900 u/l), and the magnetic resonance imaging (MRI) of the thorax evidenced inflammatory process in the anterior region of the thorax (but no involving deep tissue layers), typical of Fournier's gangrene. Because of this clinical condition, she was admitted in the intensive care unit (ICU).

After clinical stabilization, the patient needed surgical debridement (with collected culture material), with clavicle preservation and modified antibiotic therapy (changed to vancomycin 1 g 12/12 h and meropenem 1 g 8/8 h).

The Fournier's gangrene continued with an increase in the necrotic area and of osteolysis in the clavicle exposure area. The orthopedics surgeons decided to perform a total clavicle resection with debridement (**Figure 3**). Two days after,

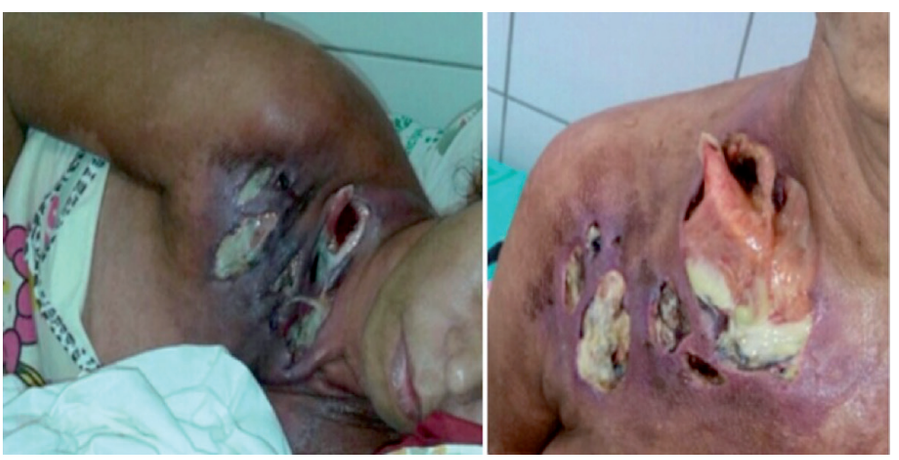

Figure 1.
Initial Fournier's gangrene of the shoulder girdle.

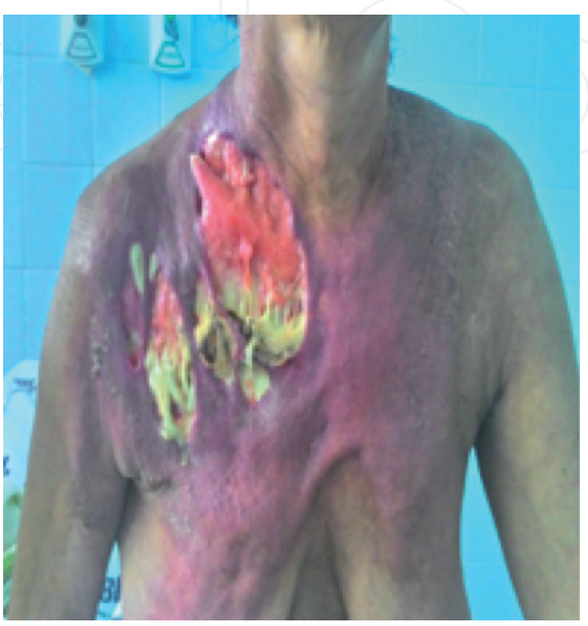

Figure 2.
Evolution of Fournier's gangrene of the shoulder girdle.

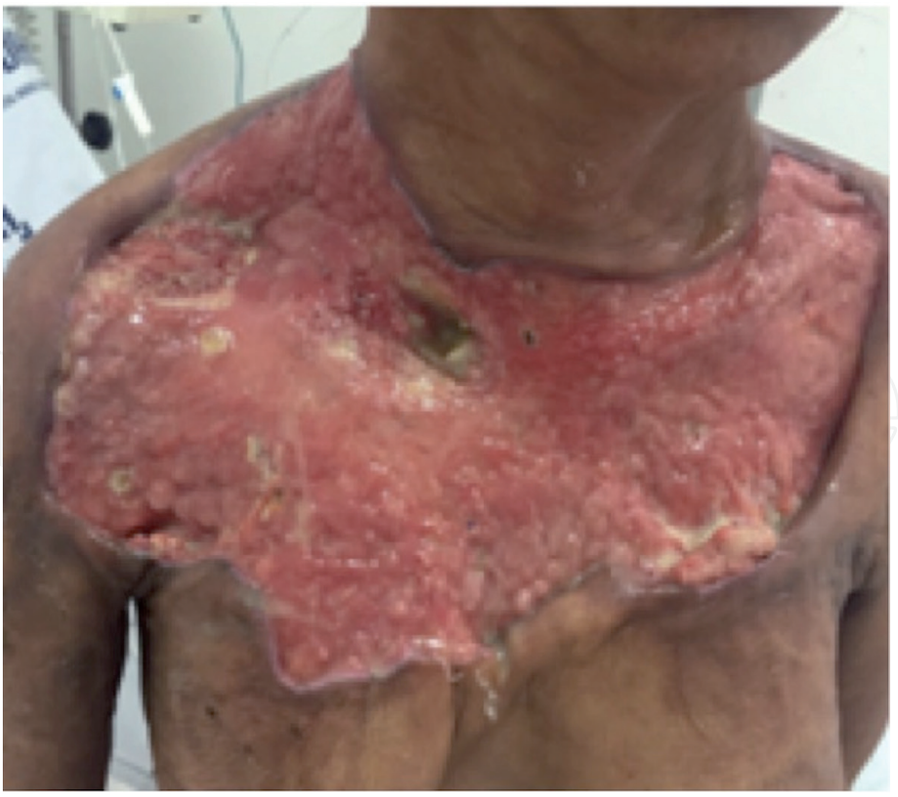

Figure 3.
Lesion before grafting.

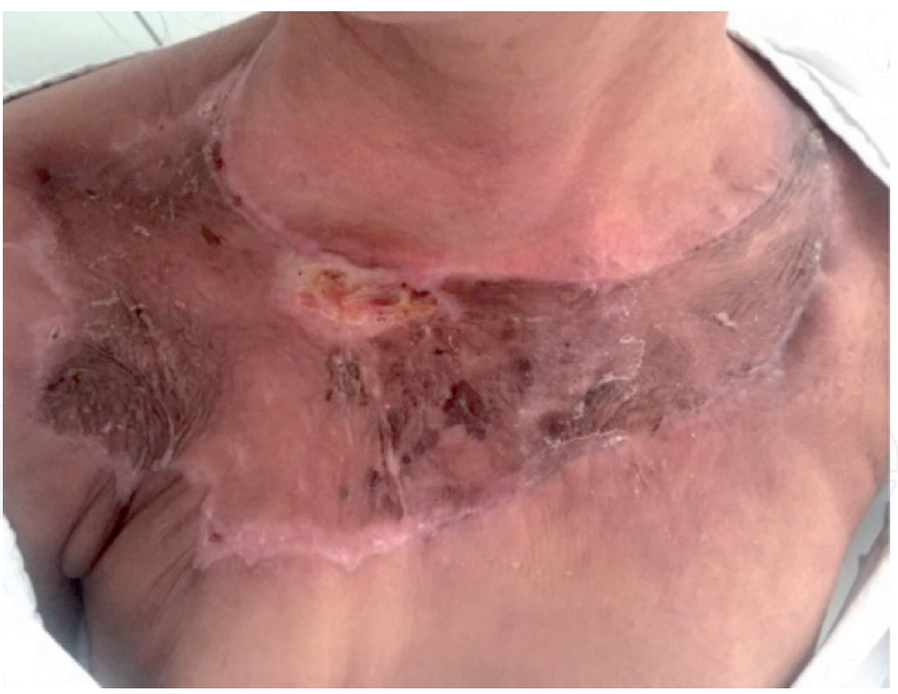

Figure 4.
Three months after grafting.

she evolved with a reduction of the WBC/inflammatory markers and presented an important clinical improvement.

The borders of the lesion ceased to evolve with necrosis and purulent secretion (granulation tissue started). The bone and soft tissue culture results were negative (maybe due to previous use of antibiotics).

Twenty days after the clavicle resection, with normal laboratory tests, a skin graft was performed by the plastic's surgeon. The patient was discharged from

Figure 5.
Six months after the procedure, the patient presented excellent functional results and a completely healed wound.

the hospital after evolving without new signs of infection. The wound presented complete healing of the graft after 60 days **(Figure 4)**.

At the last outpatient visit (after 6 months of trauma), the patient presented excellent upper limb function (33 points on the UCLA score and 93 points on the constant score) **(Figure 5)**.

6. Treatment

Once the diagnosis is established, treatment must be instituted immediately and consists of volume replacement; ample surgical debridement, with removal of all necrotic material, including the fascia; and the use of broad-spectrum antibiotics.

Although didactic, the classification of NF, in types I–IV, has little practical utility and should not be decisive in the choice of antimicrobials. The polymicrobial form is responsible in 80% of cases, which justifies the initial broad-spectrum empirical antibiotic therapy, formed by the association of clindamycin, with aminoglycoside or ciprofloxacin, used in the reported cases. Recently, the American Society of Infectious Diseases indicated the combination of ampicillin-sulbactam, clindamycin, and ciprofloxacin as the scheme of choice for community-acquired infections. In cases of nosocomial infection, the association of carbapenems with anaerobicides is indicated, according to the profile sensitivity of the most prevalent bacteria in the institution [10].

Mallikarjuna et al. [24] described the treatment of Fournier's gangrene consists of drainage, radical debridement of the necrotic tissues, and antibiotic therapy for approximately 4 to 6 weeks (initially with ampicillin/sulbactam or ampicillin combined with clindamycin or metronidazole and de-escalation guided by culture results), plus good hemodynamic stabilization. Hyperbaric oxygen therapy (OH) and the use of immunoglobulins are adjuvant and remain controversial; at the same time, further studies are needed before they can be recommended [24, 25]. The use of a vacuum drain dressing has shown to be beneficial in the follow-up after debridement; this dressing should be changed every 24–72 h [26]. Tetanus prophylaxis should be performed; however, randomized controlled trials are still required to prove the efficacy of the use of immunoglobulins as a neutralizer of *Streptococcus* toxins [27]. After the absence of infectious sign and clinical stabilization, reconstructive surgery would could be performed with grafting (if necessary) [28].

Conflict of interest

The authors do not have any conflicts of interest to declare.

Author details

Gyoguevara Patriota[1*], Luiz Marcelo Bastos Leite[1], Nivaldo Cardozo Filho[1], Paulo Santoro Belangero[2] and Benno Ejnisman[2]

1 Cardiopulmonar Hospital – Salvador-BA, Salvador, Brazil

2 Sports Medicine Division, Orthopaedics Department, Federal University of São Paulo/UNIFESP, São Paulo, Brazil

*Address all correspondence to: ombroecotovelo@drpatriota.com.br

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