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The Importance of Hyperbaric Oxygen Therapy in Patients with Fournier Syndrome

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1. Abstract

Fournier syndrome is a synergistic polymicrobial infection that mainly affects the perianal, perineal and genital regions. It is a relatively rare disease with high mortality and an estimated overall incidence of 1.6/100,000 men. Diagnosis is clinical and an early detection is crucial to improve patient outcomes. Treatment is based on three pillars: initial patient stabilization, broad-spectrum antibiotic therapy and extensive surgical debridement of the entire necrotic area. Currently, hyperbaric oxygen therapy has been used as an adjunctive treatment. This study aimed to report the case of a patient with Fournier syndrome who underwent the three-pillar treatment and also hyperbaric oxygenation with a satisfactory result.

2. Keywords: Fournier syndrome; Anti-bacterial agents; Debridement; Hyperbaric oxygenation

3. Introduction

Fournier syndrome, also known as Fournier gangrene, is a polymicrobial infection caused by aerobic and anaerobic bacteria, of synergistic character, mainly affecting the perianal, perineal and genital regions [1,2]. This infectious disease process can be potentiated and spread to other regions of the body if the treatment is ineffective. Retroperitoneum, abdominal wall, upper and lower limbs are some regions likely to be affected by this condition [3,4]. The syndrome was first reported by Baurienne in 1764 and described in detail by Jean Alfred Fournier, a French urologist, in papers published in 1863 and 1864 [1,5]. It is a relatively rare disease, with high mortality rates and an estimated overall incidence of 1.6/100,000 men [2,6,7]. It is predominant in males, with more than 90% of cases. Recent reports have indicated epidemiological changes of the syndrome, shifting from a younger population to individuals between the fifth and sixth decade of life [8].

The manifestation of this disease is insidious and the patients present with pain (100%), erythema (100%), fever (72%), crackling sound when touching the affected area caused by the gas (62%), shock (40%) and change in the level of consciousness (32%). It may be associated with some risk factors such as diabetes mellitus, hypertension, obesity, smoking, alcoholism and immunosuppressive conditions [1,3,9].

The diagnosis is clinical and the main laboratory finding is leukocytosis. Early detection is of paramount importance to improve patient outcomes and the treatment is based on three pillars: initial stabilization of the patient, broad-spectrum antibiotic therapy and extensive surgical debridement of the entire necrotic area [2,8]. Hyperbaric oxygen therapy (HBOT) has been currently used as an adjunctive treatment. The exposure to increased atmospheric pressure in

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the chamber causes toxicity to anaerobic bacteria due to the release of peroxide and superoxide radicals, leading to decreased microbial proliferation. In addition, during hyperoxygenation, oxygen-related functions such as polymorphonuclear bacterial phagocytosis, fibroblast proliferation and osteogenesis increase, while the areas of fluid resorption in the interstitial secondary to vasoconstriction and of hypoxia decrease. Moreover, hyperbaric oxygen stimulates angiogenesis and fibroblastic activity, thus increasing collagen deposition, resulting in better wound healing [10-12].

The prognosis is based on the Fournier Gangrene Severity Index (FGSI), a numerical score that comprises nine parameters in two categories: vital signs - temperature, heart rate, respiratory rate; and laboratory tests - serum sodium, potassium, creatinine and bicarbonate levels, hematocrit and leukocyte count. The individual values are summed to reach the FGSI score. A score of nine corresponds to a survival probability of 78%, while a score greater than nine is associated with a 75% probability of death [8,10].

The case of a 64-year-old male patient complaining of abscess in the perianal region and scrotal pouch, diagnosed with Fournier syndrome, is here reported.

4. Case Presentation

A 64-year-old male, married, was admitted to the outpatient clinic of our service complaining of abscess in the perianal region as well as in the scrotal pouch, with large amounts of purulent discharge, associated with asthenia and fever (not measured). The patient affirmed that he had been a chronic alcoholic for 42 years and had a 50-pack-year history of smoking. He denied any comorbidities, but reported having already treated two abscesses in the anal region, ten and four years before, exclusively with antibiotic therapy.

At admission, antibiotic therapy with cefepime and clindamycin was initiated and the patient was hospitalized for surgery. The physical examination evidenced that the scrotal pouch had edema, accompanied by phlogistic signs and a necrotic circular area of approximately 6 cm in diameter in the right hemiscrotum. Additionally, the perianal region was hardened, with edema, hyperemia and a fistula in the left inferolateral quadrant, about 3 cm from the anal edge.

Figure 1: Perineal injury observed at admission to the emergency medical service.

The patient was referred to the operating room to perform the debridement of the perineal necrotizing fasciitis, with extensive involvement of the anterior and posterior perineum, as well as the terminal rectum, requiring transversectomy.

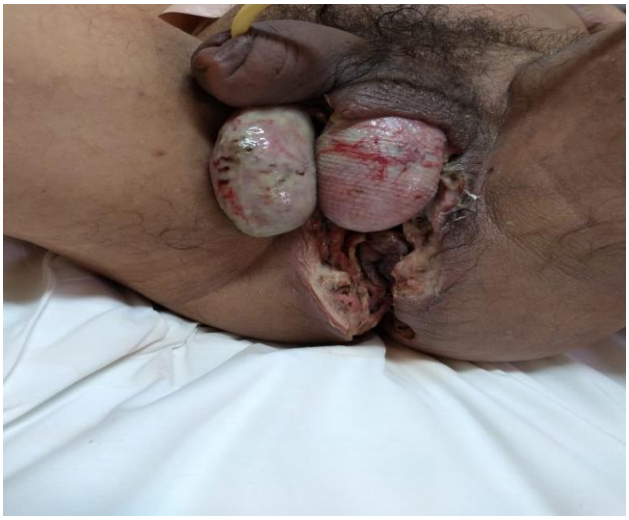

Figure 2: Postoperative surgical debridement.

The procedure lasted approximately 5 hours, under spinal and general anesthesia and was performed without complications.

After the surgery, the patient was transferred to the intensive care unit, where he remained for two days.

4.1. Laboratory exams showed

leukocyte count 21.3×109/L, C-reactive protein 16.20 mL/L and glycemia 258 mg/dL. On the third day of the postoperative period, he was transferred to a ward, but immediately after, a surgical re-approach was needed due to the presence of devitalized tissue.

Figure 3: Surgical re-approach.

The secretion culture performed turned out to be positive for Gram-positive cocci. Therefore, we associated 2 g cefepime every 8 h, 1 g vancomycin every 12 h and 500 mg metronidazole every 8 h with the previously adopted antibiotic therapy, for 14 days. During hospitalization, the patient was also diagnosed with type II diabetes mellitus, requiring the correction of glycemia with insulin therapy. At this point, based on the microbiological culture results, the multiple antibiotic therapy, the need of insulin therapy, the recurrence of abscesses in the anal region and several surgical debridement procedures in this region, we decided the patient would benefit from HBOT. This treatment was administered in a monoplace chamber (ECOBAR 800, Sistema Hiperbárico Monopaciente, Ecotec Equipamentos e Sistemas, Mogi das Cruzes, SP, Brazil) at 100% oxygen, 2.4 Absolute Atmosphere (ATA) [1 ATA = 760 mmHg = 101.3 KPa) and a PO_2 of 2,280 mmHg. Each HBOT session lasted 90 minutes and 30 sessions were required to achieve a satisfactory result.

By the 20th HBOT session, however, the patient already had well-established granulation tissue in the perianal region and in the scrotal pouch.

Immediately upon completion of 30 HBOT sessions,

Figure 4: Scrotal pouch after the 20th session of hyperbaric oxygen therapy.

the patient was scheduled for plastic surgery in order to reconstruct the injured area.

5. Discussion

Fournier syndrome was initially described as an idiopathic disease, rapidly progressing to fulminant gangrene. To date, mortality rate remains high, around 20% - 40%, increasing by up to 80% in elderly and diabetic patients [8,10,13]. It is mainly caused by aerobic and anaerobic microorganisms, which act together so that it quickly evolves into progressive necrotizing fasciitis, affecting principally the perineum and the genital region. After the infection is established, it develops to cutaneous and subcutaneous vascular thrombosis, resulting in necrosis of the skin at the affected site [1,8].

Most often, this syndrome is caused by an anorectal infection of genital origin, commonly leading to perirectal abscesses, especially when diagnosis is delayed. Moreover, it may cause urinary tract lesions such as renal abscess, urethral calculus and urethral stenosis. In a study that included 32 patients, an initial lesion with abscess (genital/perianal) was observed in 27 (84.3%) individuals, of which 17 were of genital origin (penile and/or scrotum), 9 of perianal origin and 1 in the inguinal region. Other causes involved were urethral stenosis in 4 patients (12.5%) and local trauma in 1 patient (3.2%) [14].

Although the diagnosis of Fournier syndrome is mostly clinical, complementary imaging can be used to assist in the determination of the extension of the infection

[10]. The treatment should start immediately, since this condition is aggressive and considered a surgical emergency [4,14]. The major objective of the surgical treatment is to extensively remove the devitalized tissue aiming to interrupt the progression of the infectious process.

In general, multiple surgical procedures (three to four) are required to control the infectious process. Moreover, protective colostomy is indicated in cases that would otherwise result in fecal infection. A cystostomy can be performed or a urinary catheter can be passed to avoid extravasation of urine on the surgical wound [1,15].

Considering the severity of the lesion, a broad-spectrum antibiotic regimen may be instituted. It should be administered parenterally and at doses high enough to achieve an effective concentration in the infected tissues. The therapeutic regimen should cover Gram-negative aerobic, anaerobic and streptococci and the most recommended is a combination of benzathine penicillin or amoxicillin against streptococci, metronidazole or clindamycin against anaerobic microorganisms and third generation cephalosporins or aminoglycosides against Gram-negative bacteria. As soon as drug treatment has been instituted, surgical debridement should not be delayed [1,8].

HBOT is also recommended as adjuvant therapy for patients presenting with Fournier syndrome, inasmuch as it displays antibacterial effect on anaerobic species and reduces endothelin activity. This method helps decrease the extent of necrosis, facilitating the healing process and accelerating the recovery of the patient after the surgical procedure. On average, 10 to 15 2-hour daily sessions should be performed, depending on the evolution and extent of the lesion [11,12,16].

Reparative surgery is indicated whenever the patient shows no more signs of necrotic tissue or infection and granulation tissue is present to ensure adequate tissue viability. Certainly, reconstruction methods play an important role to minimize deformities and restore patients' body image and self-esteem [14].

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