International Journal of Current Advanced Research
ISSN: O: 2319-6475, ISSN: P: 2319-6505, Impact Factor: 6.614
Available Online at www.journalijcar.org
Volume 10; Issue 05 (B); May 2021; Page No.24393-24398
DOI: http://dx.doi.org/10.24327/ijcar.2021.24398.4838

Case Report

HYPERBARIC OXYGEN THERAPY WITH EARLY REPOSITION OF TESTIS IN FOURNIER'S GANGRENE: CASE REPORT AND BRIEF REVIEW

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A R T I C L E I N F O

Article History:
Received 14th February, 2021
Received in revised form 29th March, 2021
Accepted 05th April, 2021
Published online 28th May, 2021

Key words:

Fournier's Gangrene (FG), Hyperbaric Oxygen Therapy (HBOT),

A B S T R A C T

Background*:* Founrier's Gangrene (FG), an infective necrotizing fasciitis affecting crotum and perineal region, requires extensive debridement, usually it takes a very long duration for wound healing and scrotal reconstruction and reposition of testes. We are presenting a case report with review of literature, where we used HBOT in immediate postoperative period in an operated case of FG which promoted wound healing and allowed us early reposition of testes and reduced overall duration of hospital stay

About The Case A 46-yr-old male driver was admitted with FG and deranged coagulation profile. On admission his surgical debridement was done. As vacuum dressing was not possible due anatomical reasons like size, site and extent of the wound; daily Hyperbaric Oxygen Therapy (HBOT) was done for five days. During this period healing of wound, slough disappearance and development of granulation tissue was monitored. Wound became healthy but swab taken from floor of ulcer grown resistant pseudomonas and klebsiella which required tow weeks of antibiotic treatment. His testicular reposition and scrotal reconstruction was done on 35th day of primary surgery and he was discharged on 48th day of admission in the hospital. To our opinion HBOT has helped to do early reposition in this case. We have done search of articles on FG using Google, Pubmed and Medline data search. We are presenting report of our case and review of literature.

Conclusions*:* HBOT in immediate postoperative period promots wound healing and can be used do early reposition of testes. It can reduce overall duration of hospital stay. HBOT has role in treatment of Fornier's gangrene. Further multicentric systematic studies are required to analyse exact role of HBOT in FG.

INTRODUCTION

Fournier's Gangrene (FG) is a form of infective necrotizing fasciitis affecting scrotum and perineal region. It requires extensive debridement; usually it takes a very long duration for wound healing and scrotal reconstruction with appropriate reposition of testes. Hyperbaric oxygen therapy (HBOT) promotes wound healing and can be useful to cut short the duration required for healing and definitive procedure. We have used 5 sessions of HBOT in immediate postoperative period in an operated case of FG to promote wound healing and so we could do early reposition of testes. We are presenting a report of same case along with review of literature.

CASE REPORT

A 46 year old male, a driver by occupation, admitted with complaints of swelling and blackish discoloration of scrotum associated with pain and foul smelling discharge since 12 days. Patient also had pain in lower abdomen and perineum since 3 days. On further enquiry patient was apparently normal two weeks ago and initially he had only itching over the scrotum. Subsequently, he developed swelling in the scrotum which gradually progressed over the following 12 days to the present condition as shown in image 1.

On further enquiry patient is not a known case of diabetes, hypertension or any immunocompromised status. On general examination he was slightly breathless, his pulse rate was 100, blood pressure was 90/70. On local examination his scrotum was swollen, there was blackish discoloration, necrotic skin patch, reddening of rest of scrotal skin, loss of rugosity and a foul smelling serous discharge was coming from scrotal skin. On palpation crepitus and tenderness with local rise of temperature was elicited. Signs of inflammation were extending to both the groins above and to the perineum below. He was clinically diagnosed as a case of fournier's gangrene and investigated.

His sonography of abdomen and pelvis revealed hepatomegaly. Sonography of scrotum revealed extensive bilateral scrotal wall edema with significant surrounding fat stranding. There were multiple hypoechoic areas involving

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inferior aspects of bilateral scrotal walls with multiple interspersed hyperechoic regions suggestive of air foci with appearance of dirty shadowing within. Subcutaneous oedema was extending in bilateral inguinal region and perineal region. There was mild right sided hydrocoele. His laboratory investigations revealed anaemia with features of sepsis. (Hb-10 g/dl, TLC-9100, Neutrophils-80%, platelet count-60000, prothrombin time 22.5 sec, INR 1.58)

Clinical Images: 1 to 4

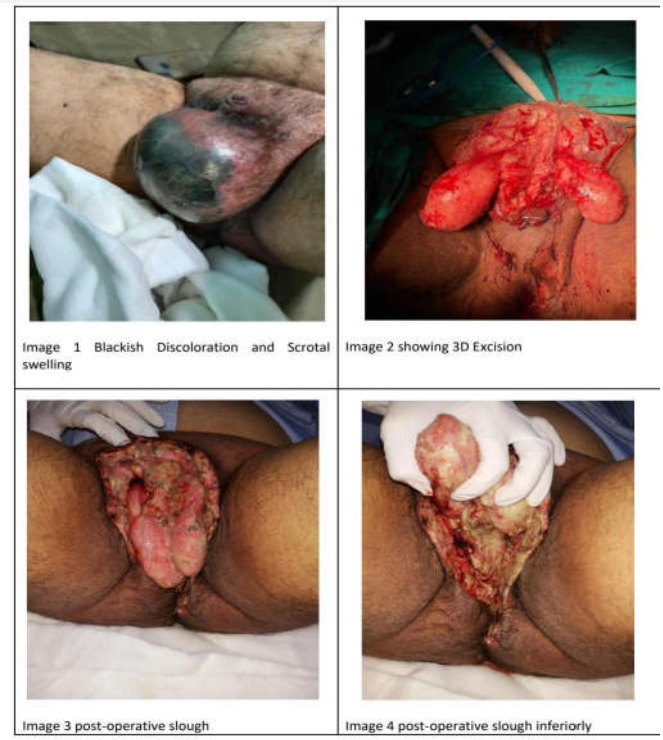

Image 1 Blackish Discoloration and Scrotal swelling

Image 2 showing 3D Excision

Image 3 post-operative slough

Image 4 post-operative slough inferiorly

Considering clinical findings and investigation, he was kept in ICU and resuscitated with transfusion of random donor platelets, fresh frozen plasma, higher antibiotics and IV Fluids. On stabilization he was taken for debridement under general anaesthesia.

Clinical Images: 5 To 8

Image 5 patient placed in the HBOT Chamber

Image 6 disappearance of slough

Image 7 showing wound Contracting

Image 8 showing reposition of Testis

As shown in image 2, three-dimensional wide excision of gangrenous scrotal skin was done up to the verge of anal sphincter. Testicles were exposed, mild reactionary hydrocoele of right testis is noted and tunica vaginalis is not opened. Postoperatively he was treated with antibiotics, blood products and other supportive care. On check dressing slough and discharge were noted as in images 3 and 4. Regular dressing and mechanical debridement of slough followed by application of desloughing agent was done.

Vacuum dressing was not possible due anatomical reasons like the site and extent of the wound. Hence we decided to try HBOT which is normally used in chronic non healing wounds and ulcer. Patient was sent to a facility providing HBOT for 5 consecutive sessions, on daily basis. In each session the patient was kept in HBOT chamber as shown in image 5 for a duration of 60 minutes and healing process of the wound was monitored closely. With HBOT, all slough disappeared (image 6), there was appearance of healthy granulation and wound started contracting (image 7). With HBOT, his entire wound became healthy, however culture swab taken from wound grown klebsiella, pseudomonas and escherichia coli which required treatment with antibiotics gentamicin, linezolid for 2 weeks.

Clinical Images: 9 and 10

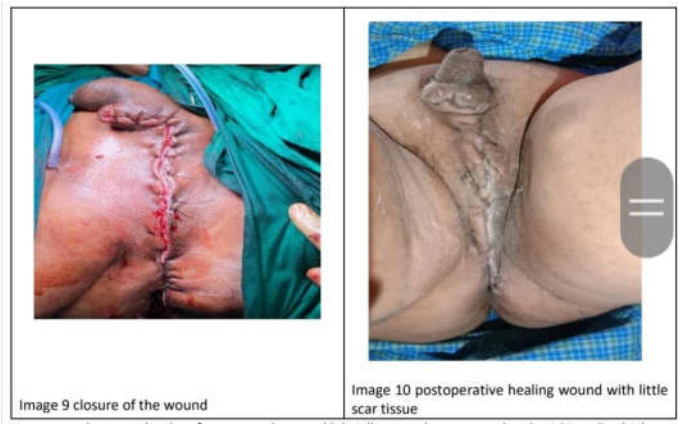

Image 9 closure of the wound

Image 10 postoperative healing wound with little scar tissue

Five weeks following initial debridement surgery, the wound was healthy but bigger in size and both the testicles were exposed to outside environment with hydrocoele on right side. We decided to do fenestration procedure for right hydrocoele with repositioning of right testis in thigh (image 8) and to cover left testis with skin flaps (image 9). The procedure was carried out under spinal anaesthesia. Postoperatively he had flap oedema which was managed with local magnesium sulphate with glycerine dressing. His wound healed well (image 10), and he was discharged on postoperative day 48 and post testes reposition day 13. To our opinion, timely use of hyperbaric oxygen therapy has helped in this case for early healing and early repositioning of testis.

BRIEF REVIEW OF LITERATURE

FG is a fulminant form of infective necrotising fascitis of the perineal, genital, or perianal regions, which commonly affects men, but can also occur in women and children [1]. The Parisian Venerologist Jean -Alfred Fournier, in 1883, described it as a fulminant gangrene of the penis and scrotum in a young man [2]. Baurienne, in 1764 and Avicenna in 1877 had described the same disease earlier [3]. Many terms have been used to describe this clinical condition which includes idiopathic gangrene of the scrotum, periurethralphlegmon,

streptococcal scrotal gangrene, phagedena, and synergistic necrotising cellulitis [4,5].

Initially, FG was defined as an idiopathic entity, but with diligent search, the source of infection in the vast majority of cases is either perineal and genital skin infections. Anorectal or urogenital or perineal trauma, pelvic and perineal injury or pelvic interventions are other causes of FG. The most common foci include the gastrointestinal tract (30%–50%), followed by the genitourinary tract (20%–40%), and cutaneous injuries (20%) [6]

Various co-morbid systemic disorders are seen associated with FG; the commonest being diabetes mellitus and alcohol misuse. Diabetes mellitus is reported to be present in 20%–70% of patients [7] with FG and chronic alcoholism in 25%–50% patients [8]. In FG, cultures from the wounds commonly show poly microbial infections by aerobes and anaerobes. It includes organisms like coliforms, klebsiella, streptococci, staphylococci, clostridia, bacteroids, and corynbacteria which could be because of the commensal nature of these organisms in the perineal region. On an average, at least three organisms are cultured from each diagnosed patient [9]. E-coli has been reported to be the commonest organism isolated from the wound, anaerobes are rarely reported. Candida albicans and lactobacillus gasseri has also been reported in some cases [10 - 12].

Pathophysiology and Clinical Presentation

The suppurative bacterial infection, synergistic activity of aerobes and anaerobes lead to the production of various exotoxins and enzymes like collagenase, heparinase, hyaluronidase, streptokinase, and streptodornase, which aid in tissue destruction and spread of infection. The platelet aggregation and complement fixation induced by the aerobes and the heparinase and collagenase produced by the anaerobes lead to results in microthrombosis of the small subcutaneous vessels leading to the development of dermal necrosis and gangrene of the overlying skin. The impaired defense mechanisms like phagocytic activity, loss in necrotic tissue in the host help the infection to proceed unchecked at alarming speed along the facial planes.

A vast heterogeneity in clinical presentation has been described ranging from insidious onset and slow progression to rapid onset and fulminant course, the latter being the more common presentation. In contrast with initial description by Fournier, the disease tends to present more in elderly men, and also has been reported in women and children [13]. The infection commonly starts as a cellulitis adjacent to the portal of entry, depending on the source of infection, commonly in the perineum or perianal region. The spread of infection is along the facial planes and is usually limited by the attachment of the colles' fascia in the perineum. Infection can spread to involve the scrotum, penis and can spread up the anterior abdominal wall, up to the clavicle [14]. The testes are usually spared as their blood supply originate intra-abdominally. When testis are involved, it suggests retroperitoneal origin or spread of infection. FG originating from Urogenital infections travel posteriorly along the bucks and dartos fascia to involve the colles' fascia, but are limited from the anal margin by the attachment of the colles' fascia to the perineal body. In contrast anorectal sources of infection usually start perianaly and this variation in initial clinical presentation can serve as a guide to localizing the foci of infection.

The local signs and symptoms are usually dramatic with significant pain and swelling. The patient also has pronounced systemic signs; usually out of proportion to the local extent of the disease. Crepitus of the inflamed tissues is a common feature because of the presence of gas forming organisms. As the subcutaneous inflammation worsens, necrotic patches start appearing over the overlying skin and progress to extensive necrosis [15]. Unless aggressively treated, the patient can rapidly progress to sepsis with multiple organ failure which is a common cause of death in these patients [16].

Diagnosis and Scoring

Even though the diagnosis of FG is primarily clinical, imaging modalities may be useful in those cases where the presentation is atypical or when there is concern regarding the true extent of the disease. Plain radiography may show air within the tissues. Ultrasonography is useful to differentiate intrascrotal abnormality and can also show thickened and swollen scrotal wall, with gas within [17]. Computed tomography and magnetic resonance imaging are useful in select cases to diagnose or rule out retroperitoneal or intraabdominal disease process [18]

Laor *et al* developed a scoring system (fournier's gangrene severity index), to quantify the severity of infection, using common vital signs and laboratory data. This score helps to prognosticate the illness and helps to predict the mortality. With a score of over 9, they found a 75% probability of death while a score of less than 9 was associated with 78% probability of survival [19]. Chawla *et al* used this scoring system in their series of 19 patients and found that fournier's gangrene severity index was useful in predicting survival but not length of hospital stay [20].

Treatment

FG warrants an aggressive multi-modal approach, which includes haemodynamic stabilization, broad spectrum antibiotics, and surgical debridement. It must be highlighted however, that early surgical debridement is the primary component of treatment and if delayed will have a negative impact on the prognosis [21]. All non-viable and necrotic tissue must be excised, until well perfused viable tissue is re ached. the full extent of the disease may not be apparent from the areas of cutaneous involvement, which is usually less than the subcutaneous disease. Care must be taken not to accidentally open up deeper facial planes, which were not initially involved. Urinary or faecal diversion may be necessary depending upon the foci of origin of the disease. Multiple surgical debridement is the rule rather than the exception, with an average of 3.5 procedures required per patient. Even though testes are classically spared in the process of FG, orchidectomy, for non-viable testis, is eventually required in up to 21% patients.

Antibiotic therapy should be broad spectrum to empirically cover all possible organisms. The usual combination includes penicillin for the streptococcal species, third generation cephalosporin, with or without an aminoglycoside, for the gram negative organisms, plus metronidazole for the anaerobes [22]. Some topical agents like Dakins solution (sodium hypochlorite), hydrogen peroxide, or unprocessed honey has been tried to aid in the separation of the slough and accelerate granulation tissue [23]. If the initial tissue stain using potassium hydroxide shows the presence of a fungus or if

grown in the culture, then addition of Amphotercin-B is necessary.

After initial aggressive surgical debridement it is important to do desloughing regularly so that wound start healing. Various measures are available for the promotion of wound healing. With vacuum assisted closure system dressing, there seems to be some improvement with minimising skin defects and speeding tissue healing. It simply works by exposing a wound to subatmospheric pressure for an extended period to promote debridement and healing. Hyperbaric oxygen is widely believed to be an effective adjunctive therapy in the treatment of FG. In our case we have given five sessions of HBOT to our patient in the early post operative period and we found it useful as the entire slough disappeared with the appearance of healthy granulation tissue in 5 sessions of HBOT.

Hyperbaric Oxygen Therapy

HBOT is breathing 100% oxygen under increased atmospheric pressure. It was introduced as treatment modality in 16th century. A British clergyman, Henshaw, built a structure called domicilium which can be pressurized or unpressurized with air using bellows; that was used to treat several diseases [24]. French surgeon Fontaine built a pressurized, mobile operating room in 1879 [25].

When a patient is given 100% oxygen under pressure, hemoglobin is saturated, but the blood can be hyperoxygenated by dissolving oxygen within the plasma. Normal wound healing proceeds through stages of hemostasis, removal of infectious agents, resolution of the inflammatory response, reestablishment of a connective tissue matrix, angiogenesis, and resurfacing [26,27]. Benefits of HBOT include neutralisation of anaerobic organisms, improvement in neutrophil function, increased fibroblast proliferation, and angiogenesis [28]. HBOT can be administered in patients via two basic chambers: Type A, multiplace (for multiple patients at the same time); and Type B, monoplace (for one patient at a time).

Multiple clinical studies suggest that HBOT is efficacious in the treatment of necrotizing soft tissue infections. These include case series, retrospective and prospective studies, and non-randomized clinical trials. They suggest significant reductions in mortality and morbidity. Initial HBOT is aggressively performed at least twice per day in coordination with surgical debridement. Typically, a treatment pressure ranging from 2.0-2.5 ATA is adequate. However, in the specific case of clostridial myonecrosis, 3 ATA is often used to ensure adequate tissue oxygen tensions to stop alpha toxin production. for the same reason, HBOT should be initiated as quickly as possible in this circumstance and performed 3 times in the first 24 h if at all feasible.

Because the goals of HBOT for wound healing include cellular proliferation and angiogenesis, HBOT is generally performed daily for a minimum of 30 treatments. Treatment is generally at 2 to 2.4 ATA for a total of 90 minutes of 100% oxygen breathing time. Based on the response to therapy, extended courses of therapy may be indicated.

HBOT should be used in conjunction with a complete wound healing care plan. As with all chronic wounds, other underlying host factors like large vessel disease, glycemic control, nutrition, infection, presence of necrotic tissue, offloading must be simultaneously addressed. Emi Latham *et al* reviewed all the literature related to mechanism of action and clinical application of HBOT and he found that HBOT has been successfully used in treatment of variety of conditions. HBOT has been used in chronic non healing ulcers in diabetic patients, necrotising soft tissue infections, crush injuries, compartment syndrome, compromised grafts and flaps, thermal burns, carbon monoxide poisoning and central retinal artery occlusion [29].

The cornerstones of therapy in FG are wide surgical debridement and aggressive antibiotic therapy. HBOT is used adjunctively with these measures, as it offers several mechanisms of action to control the infection and reduce tissue loss. In 2020, Laila Schneidewind et al. conducted a systematic review of HBOT in treatment of FG where literature search for primary studies yielded 79 results of which 13 studies were considered, which included a total of 376 patients with FG, of whom 202 (53.7%) received HBOT [30-42]. Final conclusion of Laila Schneidewind et al. was that, despite the risk of bias, HBOT has the potential as an adjunct in FG treatment, but it is challenging to carry out further studies or even randomized control studies due to the rareness of this disease, restricted availability of HBOT and the complex character of FG [43].

Post Operative Testicular Reposition

Various workers have used different techniques to provide skin cover to exposed testis in FG. Various described options included free skin grafts, axial groin flaps, myocutaneous flaps and auto transplantation of testes. Split thickness skin graft seems to be the treatment of choice in treating perineal and scrotal skin defects. Parkash *et al* reported their series of treatment of 43 cases, in three cases the gangrene had spread beyond the scrotum and involved penile skin; split-skin grafts were used to cover it. In all the other cases, cover was provided with scrotal skin remnants at the edge of the lesion and on the penis with the inner layer of the prepuce, which had remained intact [44]. Black PC *et al* reported their series of meshed unexpanded split-thickness skin grafting for skin defects. They treated nine patients with penile skin loss with meshed split thickness skin grafts to the penis [45]. In our case, as exposed area was big fenestration procedure for right hydrocoele with repositioning of right testis in thigh was done. We could cover left testis with skin flaps raised from remnant scrotal as well as groin skin.

Outcome

Early surgical debridement of necrotic tissues, appropriate antibiotics, Post operative wound management and appropriate skin closure with reposition of testis are the important components in the treatment of FG. Despite advanced management mortality in describe literature is still high and ranges between 20%–30% [46]. Very few authors have done study of post operative time required to do testicular reposition and overall hospital stay duration in FG. Mark Ferretti *et al* described study of 20 patients of FG in which HBOT was used in 4 cases and overall hospital stay was 22 days in this group of patients. [47]. Oussama Baraket *et al* described comparison of duration of wound healing in 23 patients of FG treated with or without HBOT, mean duration of healing was 15 days in HBOT group verses 24 days in Non HBOT group [48]. In our case wound was improved after HBOT and was ready for closure on 20th day but because of organisms grown in culture we have to give antibiotics for two weeks still over all hospital stay was 48 days from the day of admission. To our opinion

HBOT promoted early wound healing and thus helped to decrease duration of hospital stay.

CONCLUSION

HBOT in immediate postoperative period promots wound healing and can be used do early reposition of testes. It can reduce overall duration of hospital stay. HBOT has role in treatment of Fornier's gangrene. Further multicentric systematic studies are required to analyse exact role of HBOT in FG.

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How to cite this article:

Kailash Jawade *et al* (2021) 'Hyperbaric Oxygen Therapy With Early Reposition of Testis In Fournier's Gangrene: Case Report And Brief Review', *International Journal of Current Advanced Research,* 10(05), pp. 24393-24398.
DOI: http://dx.doi.org/10.24327/ijcar.2021.24398.4838

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