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Fournier's Gangrene after Radiation Therapy for Rectal Carcinoma: A Rare Case Description and Review of Literature

Aman Kumar Vaid[1], Jagroop Panchal[2], Rajiv Atri[3], Rakesh Dhankhar[4], Ashok Chauhan[5]

[1]Senior Resident, Department of Radiation Oncology, Regional Cancer Centre, Pt. B.D. Sharma PGIMS, Rohtak

[2]Junior Resident, Department of Radiation Oncology, Regional Cancer Centre, Pt. B.D. Sharma PGIMS, Rohtak

[3,4]Professor, Department of Radiation Oncology, Regional Cancer Centre, Pt. B.D. Sharma PGIMS, Rohtak

[5]Professor and head, Department of Radiation Oncology, Regional Cancer Centre, Pt. B.D. Sharma PGIMS, Rohtak

ABSTRACT

Background: Fournier's Gangrene (FG) is a rapidly progressive infective necrotizing fasciitis primarily affecting the genital region[1]. It is predominantly associated with various urological, gastrointestinal, and retroperitoneal diseases, some of which may involve malignancies of these regions[2]. The occurrence of Fournier's gangrene following radiation therapy for an underlying carcinoma is exceedingly rare, and therefore, only sporadic cases have been documented in the literature. Treating Fournier's gangrene in this specific context presents increased complexity compared to cases stemming from other causes, and it is associated with elevated mortality rates[3]. Consequently, a heightened level of vigilance is necessary in the management of such cases. In this report, we present the case of a 71-year-old male patient diagnosed with rectal carcinoma who developed Fournier's Gangrene subsequent to radiotherapy, and the treatment approach was tailored accordingly.

Case Description: A 71-year-old male presented to our department with a chief complaint of persistent rectal bleeding for a duration of one year. Further assessment, including a contrastenhanced computed tomography (CECT) scan of the entire abdomen and pelvis, revealed a 5.4x4.7x4.5 cm lesion located in the ano-rectal region. Due to the patient's compromised overall health status, a decision was made to administer palliative external beam radiotherapy (EBRT) to the pelvic region. The treatment plan consisted of a total radiation dose of 20 Gy delivered over five fractions spanning five consecutive days (20 Gy/5 fraction/5 days), employing anteriorposterior (AP) and posterior-anterior (PA) fields. Shortly after the completion of radiotherapy, the patient developed severe bilateral swelling of the scrotum, which subsequent evaluation confirmed to be Fournier's gangrene. Prompt surgical intervention in the form of incision and drainage, in conjunction with intravenous administration of appropriate antibiotics, was performed, yielding positive a therapeutic outcome. Currently, the patient is being maintained on a low-dose oral metronomic chemotherapy regimen with tablet Capecitabine at a dose of 500 mg twice daily for a duration of two weeks, followed by a one-week treatment-free interval. The patient has demonstrated favorable response and tolerability to the prescribed treatment regimen, given the presence of residual disease.

Conclusion: We report an extremely rare case of development of Fournier's Gangrene following Radiotherapy in a case of Carcinoma Rectum which can be associated with very high mortality if not diagnosed early and treated properly. Hence, prompt diagnosis and aggressive management of this rare complication is necessary for patient's favorable outcome.

KEYWORDS: Fournier's Gangrene, Carcinoma rectum, Radiotherapy

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INTRODUCTION

Fournier's Gangrene (FG) is characterized as an infective necrotizing fasciitis primarily affecting the perineal, genital, or perianal regions, leading to thrombosis of subcutaneous blood vessels and subsequent gangrene of the overlying skin as a result of infectious processes[1]. The condition is accompanied by pronounced systemic intoxication and can lead to multiple organ failure. The condition commonly arises from various urological, gastrointestinal, and retroperitoneal diseases, with or without malignancy involvement[2].Initially considered idiopathic, Fournier's Gangrene (FG) is now found to have an identifiable cause in as many as 95% of cases. The infection can result from anorectal, urogenital, traumatic, or dermatologic pathology, with a potential inoculation of both anaerobic and aerobic bacteria [4, 5]. In the majority of cases, the infectious agent is polymicrobial (62%), with Streptococcus spp. (31%), Bacteroides spp. (27%), and E. coli (24%) being the most prevalent isolates [6]. The development of FG as a secondary manifestation of cancer is an infrequent occurrence. Radiation-induced Fournier's Gangrene in cancer patients undergoing pelvic radiotherapy is an exceptionally rare event and hence has only been documented in the literature sporadically[1,2]. The mortality rate associated with Fournier's Gangrene is remarkably high, underscoring the critical importance of early diagnosis and aggressive management for favorable patient outcomes[3]. The largest case series conducted on FG has revealed a significant mortality risk, reaching 20.8% if not managed proactively[4]. Treatment of FG entails extensive debridement of necrotic areas, along with the administration of broad-spectrum intravenous antibiotics and supportive intravenous fluid therapy. In cases where FG develops as a consequence of rectal cancer, the causative rectal tumor should be surgically excised, followed by diligent supportive care. Treatment for Fournier's gangrene associated with radiotherapy poses greater complexity compared to other instances of FG due to the wound healing complications that arise as a side effect of external beam radiotherapy (EBRT) and hence should be dealt with aggressively.

CASE DESCRIPTION

A 71-year-old male presented to our department with a chief complaint of persistent bleeding per rectal for a duration of one year. A comprehensive medical history and general physical examination were conducted, which included a digital rectal examination that revealed no apparent growth or lesion in the anorectal region. Subsequent evaluation involved a contrast-enhanced magnetic resonance imaging (CEMRI) scan of the entire abdomen and pelvis, which revealed a rectal lesion measuring 5.4x4.7x4.5 cm, located 7 cm away from the anal verge. The invading edge of the lesion spanned from the 7 o'clock to 4 o'clock position. The distal edge of the tumor extended into the rectal region, with a maximum longitudinal extent of 5.4 cm. The tumor exhibited infiltration of the peritoneal reflection and mesorectal fascia, with peritoneal spread involving the left seminal vesicle. Several enlarged lymph nodes in the presacral region were observed, with the largest measuring 1.7x1.8 cm. A colonoscopic biopsy confirmed the presence of Moderately Differentiated Adenocarcinoma of the Rectum. Based on the CEMRI findings, the clinical TNM stage was determined as T4N1M0, classifying the patient into prognostic group stage IIIb. Considering the patient's compromised overall health status, a decision was made to administer palliative radiotherapy (RT) to the pelvic region. The treatment plan consisted of a total radiation dose of 20 Gy delivered over five fractions spanning five consecutive days, utilizing anteriorposterior (AP) and posterior-anterior (PA) fields. The patient was advised to return for follow-up after one month to assess the tumor response to radiotherapy. However, the patient presented two days after completing radiotherapy with severe bilateral scrotal swelling measuring approximately 20x20 cm. A surgical consultation was taken, leading to a diagnosis of Fournier's Gangrene (FG). The patient underwent incision and drainage (I&D) of the gangrenous wound and received aggressive antibacterial therapy, involving intravenous administration of antibiotics (Imipenem and Amikacin) and supportive fluid therapy. The patient exhibited a favorable response to the treatment and experienced rapid recovery. Subsequently, the patient was placed on a low-dose oral metronomic chemotherapy regimen. This entailed the administration of Capecitabine tablets in three-week cycles, with a dosage of 500 mg twice daily for a duration of two weeks, followed by a one-week treatment-free interval. Currently, the patient's condition is stable, and regular followup visits are being conducted in accordance with medical advice.

DISCUSSION

Fournier's gangrene (FG) is an uncommon yet critical condition characterized by necrotizing fasciitis primarily affecting the perineal or genital regions. The condition dates back to 1764 when Baurienne initially documented it, but it was later named after Jean-Alfred Fournier, a prominent French dermatologist and venereologist who presented five cases in 1883. Fournier's clinical series highlighted the rapid progression of gangrene involving the penis and scrotum in previously healthy young men. In modern terminology, the term encompasses a polymicrobial necrotizing soft tissue infection that involves the superficial and deep fascial planes of the genital, perineal, or perianal areas. [2]. Bacteremia is recognized as the initial trigger in the pathogenesis of fascial necrosis, initiating a cytokine cascade that results in endothelial damage. This, in turn, activates the coagulation cascade via thromboplastin, leading to fibrinolysis inhibition and the formation of disseminated microthrombosis in the vessels supplying the fascia. Concurrently, endothelial damage leads to extravasation of plasma, tissue swelling, leukocyte infiltration, all culminating in ischemic necrosis of the fascia [2].

Fournier's disease is characterized by pronounced symptoms of intoxication. Local manifestations include ulceration in the balanus, prepuce, penile skin, or scrotum. Within a few hours, hyperemia of the genitalia intensifies, accompanied by tissue necrosis. Painful and difficult urination is observed. The disease typically runs its course over a duration of 5 to 8 days [2].

FG, in colorectal cancer patients receiving radiotherapy is an extremely rare but life-threatening complication that has been described only thrice in literature[9]

The clinical symptomatology involves widespread necrosis of the skin, subcutaneous tissue, and muscles, often resulting in the development of sepsis and multi-organ failure, ultimately leading to death. The scrotum is the most frequent initial site of Fournier's gangrene (FG) resulting from rectal cancer. In patients presenting with anorectal pain, rectal bleeding, tenesmus, alterations in bowel habits, and unintentional weight loss, the development of scrotal edema should prompt suspicion of rectal cancer. The septic state manifests with the swift onset of severe toxemia, resulting in pyrexia with or without hypothermia, tachycardia, hypotension, and oliguria. Sepsis can develop within a few hours, rapidly progressing to organ failure and death. The clinical presentation is fairly consistent regardless of the etiologies, including carcinomas of the pelvis[7]. Early diagnosis may be hindered by various factors, such as patient obesity and the patient's reluctance to report genital pain complaints.

The essential pillars of FG management lie in early diagnosis and aggressive treatment, as the condition is associated with considerable morbidity and mortality rates. Despite receiving appropriate treatment, the mortality rate remains alarmingly high, ranging from 20% to 35% [1]. In one of the largest single institute study by Yilmalzar et al, the mortality rate was found to be 20.8% even after early diagnosis and aggressive treatment[8].

Numerous factors contribute to the predisposition for FG, including trauma, diabetes mellitus, intravenous drug abuse, peripheral vascular disease, hypertension, malnutrition, smoking, obesity, immunocompromised status, renal insufficiency, malignancy, and spinal cord injury. In the case of cancer, which represents an immunocompromised state, Fournier Gangrene can develop. Moreover, the utilization of large radiation portals in pelvic cancer treatment increases the susceptibility to FG due to associated skin reactions that may become infected, leading to subsequent necrosis and gangrene. [3] There are three distinct pathways of development in this context: (1) the manifestation of trauma in the perineal region, (2) the progression along the muscular membrane lining the infected urinary tract, and (3) the infection affecting the perianal region and retroperitoneum [4]. In this case, the patient was in very good overall condition without comorbidities. He did not take any medication, smoke, or consume alcohol, and did not report any trauma. Risk factors in our patient included immunosuppressed state related to cancer, and the high dose per fraction radiotherapy (20 Gy/5 Fractions corresponding to 4 Gy/fraction) which was given via large pelvic portals which may also have contributed to the severe skin reactions which got infected and developed gangrene. In order to effectively address this condition, it is recommended to promptly initiate treatment involving broad-spectrum antibiotics that provide coverage against staphylococci, streptococci, enterobacteriaceae, and anaerobic bacteria. Nevertheless, the cornerstone of treatment continues to be aggressive surgical intervention, which entails the comprehensive removal of all necrotic tissue until viable, actively bleeding tissues are encountered [6]. In many cases, repeated surgical debridements may be necessary to adequately eliminate all affected tissue. While instances of rectal cancer perforation subsequent to radiation therapy have been well-documented, occurrences of FG following radiation therapy for rectal cancer treatment are exceedingly rare. The condition poses a significant threat to life and is associated with a high mortality rate. Therefore, it is crucial to promptly diagnose and initiate treatment, encompassing debridement and administration of appropriate antibiotics. In the context of medical intervention, it is imperative to maintain a heightened awareness of the possibility of Fournier's gangrene and commence treatment expeditiously.

CONCLUSION

We present an exceptionally uncommon case of Fournier's Gangrene that emerged following radiotherapy in a patient with carcinoma rectum at our institute. This disease can be linked to a significantly elevated mortality rate if not promptly diagnosed and managed appropriately. Therefore, urgent and assertive intervention is imperative to ensure a favorable patient outcome in this rare complication.

Fig 1: CECT scan pelvis showing mass lesion in the rectum

Fig 2: Severe scrotal swelling developed after completion of radiation therapy

Figure 3: Resolving FG post Incision and Drainage

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