Rare Presentation of Perforated Rectal Cancer with Right Buttock Necrotising Fasciitis

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1. Abstract
Necrotizing Fasciitis (NF) is a life-threatening infection that affects soft tissues and deep fascia. On rare occasions, perforated rectal cancer can be presented with NF, requiring urgent recognition and management to reduce mortality and optimise clinical outcomes. We present a 56-year-old female who had been referred by a General Practitioner (GP) to the emergency department due to ongoing right buttock pain. Computed Tomography (CT) abdomen and pelvis revealed a large pelvic mass involving the rectum, sigmoid colon, and uterus/vagina. Multiple gas locules were noted in the right ischiorectal fossa. The patient was taken to the operating theatre and underwent flexible sigmoidoscopy, and drainage/debridement of the right ischiorectal abscess. A large fungating circumferential near-obstructing mass was found in the sigmoid colon and rectum (5cm from the anal verge). Endoscopic biopsies were taken. Laparoscopic diverting proximal sigmoid colon loop colostomy was formed and the right ischiorectal fossa was debrided. The ischiorectal fossa was debrided and packed with betadine soaked gauze which was changed daily. The histopathology of the recto-sigmoid mass has returned as adenocarcinoma. The patient was discussed in the multidisciplinary meeting and treated with palliative chemoradiotherapy. Extraperitoneal causes of necrotising fasciitis should be sought in patients who present with buttock subcutaneous emphysema to reduce mortality and improve clinical outcomes.

2. Introduction
Colorectal cancer is the fourth most common cancer and the second most common cancer death in Australia in 2022 [1]. It is estimated about 15,000 patients are diagnosed with colorectal cancer and over 5,300 deaths annually in Australia [1]. The common presentations of colorectal cancer include altered bowel habits, weight loss, blood in stool, and night sweats [1]. On rare occasions, colorectal cancer can be complicated by perforation or obstruction, which requires emergency surgery [2,3]. Most patients with perforated colorectal cancers present with peritonitis and intra-abdominal sepsis [2-4]. In rare instances, perforation of rectal cancer may present with necrotising fasciitis [2]. Necrotizing fasciitis is a devastating soft tissue condition characterized by rapidly advancing infection just above the deep fascia by gas forming bacteria, leading to ischemia and necrosis of the skin, and the fat layer above [2,5]. Common causes of NF include complicated bacterial infections, blunt trauma, or even minor wounds [2,5]. Causative bacterial organisms may be monomicrobial (Streptococcus pyogenes) or polymicrobial [5].

This case report presents a rare case of a patient who presented with right gluteal necrotising fasciitis due to perforation of an undiagnosed rectal cancer.

3. Case Report
Mrs RP is a 59-year-old female who was referred by GP to the emergency department with right buttock gluteal pain and symptomatic anaemia. Her past medical history included lower segment caesarean section. Her biochemical panel revealed haemoglobin 60 (g/L; normal range 115-160), white cell count 29.2 (x10⁹/L; normal range 4.0-11.0), platelet 555 (x10⁹/L; normal range 140-400), c-reactive protein 157 (mg/L; normal range <5.0), blood culture positive with Escherichia coli (E.coli), and carcinoembryonic antigen 8.2 (ug/L; normal range <3.5). CT abdomen/pelvis scan revealed a large pelvic mass involving the rectum, sigmoid colon, and uterus/vagina. Multiple gas locules were identified at the right side of the perianal region as well as the right buttock (Figures 1 and 2). A nodular appearance to the distal extent of the medial limb of the right adrenal gland approximately measures 14x10mm.
concerning of the metastatic mass (Figure 2).

The patient was febrile with temperature of 39.1°C, heart rate of 99, the abdomen was soft with guarding/rebound tenderness over the suprapubic and lower iliac fossa, right buttock was indurated and tender on palpation. Her Digital Rectum Examination (DRE) exam showed a palpable rectal mass at 4-5cm from the anal verge with hard induration and faecal incontinence.

The patient was taken to the operating theatre and underwent flexible sigmoidoscopy, and drainage/debridement of the right ischiorectal abscess. A large fungating circumferential near-obstructing mass was found in the sigmoid colon and rectum (5cm from the anal verge). Endoscopic biopsies were taken. Laparoscopic diverting proximal sigmoid colon loop colostomy was formed and the right ischiorectal fossa was debrided. The ischiorectal fossa was debrided and packed with betadine-soaked gauze which was changed daily. The histopathology of the recto-sigmoid mass has returned as invasive adenocarcinoma. Lymphovascular invasion and perineural infiltration are not seen.

Intra-operatively large circumferential fungating low rectal cancer was identified at 4-5 cm from the anal verge. The tumor was near obstructing with a pinpoint lumen. On the bimanual exam, the tumor had invaded into the left posterior wall of the vagina. Crepitus was palpable over the right ischiorectal fossa with the entire right ischiorectal fossa destroyed by necrotic tumor and abscess. An extensive debridement was performed down to the gluteus maximus and up to the levator ani (pelvic floor). A diverting laparoscopic colostomy was formed.

A staging CT scan of the chest, abdomen and pelvis showed no distant metastases.

Magnetic Resonance Imaging (MRI) of the abdomen and pelvis showed a 12.8 x 9 cm fungating rectal cancer up to the sacral promontory (Figure 3). The tumor was invading beyond the fascia propria to involve the lateral pelvic side wall, sacrum as well as pelvic organs anteriorly including uterus and vagina (Figures 4 and 5). Histopathology from the biopsy of the ischiorectal fossa shown a moderately differentiated adenocarcinoma. Bacterial culture of the right ischiorectal abscess confirmed polymicrobial infection with gas-forming organisms (E.coli, P.aeruginosa).

In the multidisciplinary meeting, it was decided that the tumor is inoperable, with the patient being offered palliative chemotherapy and radiotherapy. The patient was discharged on day fourteen after the initial surgery.

**Figure 1:** CT Abdominal/pelvis portal venous axial image to demonstrated multiple gas locules in the right perianal region extending to the right buttock soft tissues.

**Figure 2:** CT Abdominal/Pelvis Portal venous coronal image to demonstrated 3mm wall off collection in the right side of the lower pelvis/perianal region approximately 70 mm in craniocaudal length and 52mm x 22mm maximum axial dimensions.
Figure 3: Magnetic resonance image scan pelvis T2 TSE sagittal demonstrating large cavitating fungating mass extending to the upper, middle and lower third rectum. Craniocaudal length is measured at 12.8cm and anterior posterior diameter is measured at 9.01cm T2 hyperintense foci in the lesion.

Figure 4: Magnetic resonance imaging scan pelvis T2 TSE Axial demonstrating lesion extends to and infiltrates the mesorectal fascia particularly on the right side anteriorly and posteriorly.

Figure 5: Magnetic resonance imaging scan pelvis T2 TSE Axial demonstrating fungating mass in the rectum infiltrating to posterior myometrium suggested by the same signal intensity.

4. Discussion

NF is a life-threatening soft tissue infection caused by either monomicrobial or polymicrobial bacteria [5]. Polymicrobial NF is usually prevalent in immunosuppressed patients, and monomicrobial NF is more common after blunt trauma, complicated infections, and minor wounds [5]. In other articles, NF secondary to colorectal cancer perforation have been reported to have an association in patients who are taking steroids, chemotherapy, and radiotherapy which are risk factors for polymicrobial NF. Polymicrobial NF is reported to be more common than monomicrobial and is mainly caused by a combination of aerobic and anaerobic bacteria [5]. Monomicrobial NF is more uncommon, and usually affects healthy individuals with a history of trauma, and or minor infection or injury [5]. In patients with NF, typical clinical presentations include sepsis, toxic appearance, edematous, skin discoloration of the affected area, erythema, subcutaneous emphysema, and crepitus on palpation due to subcutaneous emphysema [5].
Subcutaneous emphysema is a gas accumulation in the subcutaneous tissue plane, and it can be due to various causes including trauma, recent surgery, and necrotizing fasciitis, which can be a surgical emergency [2]. The first case of subcutaneous emphysema caused by a perforated hollow viscus was reported in 1853 [3]. Subcutaneous emphysema may be caused by either intraperitoneal or extraperitoneal [3]. In rare cases, non-traumatic subcutaneous emphysema in the lower extremities can be a sign of perforation of intestinal rupture [4,6]. Subcutaneous emphysema of the leg has been previously described in cases of perforated appendicitis, ileum, diverticulitis, and perforated colorectal cancer [3,4]. The aetiology of the formation of subcutaneous emphysema in intestinal rupture can be contributed by either gas from the perforated organ or from secondary gas forming bacterial infection [3]. Subcutaneous emphysema of the lower limb is thought to be that the free air can track down inferiorly through the pelvic floor into the subcutaneous tissues of the buttock and thighs [3].

Our patient presented with fever, and right buttock infection, raising concern for NF. CT abdominal and pelvis showed multiple gas locules in the right side of the perianal region extending to the right buttock which is suggestive of subcutaneous emphysema. With no recovery of intravenous antibiotics use with rapid progression of infection, clinical diagnosis of necrotising fasciitis is confirmed. The risk factors of necrotising fasciitis include diabetes, obesity, alcoholism, intravenous drug use, and peripheral artery [4], which our patient had none of them.

Perforated colorectal cancer normally presents with peritonism, and pneumoperitoneum in imaging [4], however, it is essential to remember that it can be present with necrotising fasciitis of gluteal/buttock region, and lower extremities with radiological evidence of subcutaneous emphysema [4].

Amongst different diagnoses, perforated colorectal cancer needs to be considered for prompt treatment. Perforated colorectal cancer has a mortality rate of 30 to 40% and in the case of subcutaneous emphysema developing NF, can go higher up to 50 to 75% even with appropriate management [7]. There are only a few case reports on colorectal cancer perforation presenting with subcutaneous emphysema, and a few other case studies with NF in perforated colorectal cancer suggest a high mortality rate of over 60% [2,3,6]. As a tumour progresses, it can perforate, which can give rise to abscesses in adjacent tissues [3,4]. Gas-producing bacteria such as Escherichia coli, Group A beta-hemolytic streptococcus or a clostridium species from an abscess can produce gas that will dissect through soft tissues [8].

In our case, the patient was operated on day 2 post-admission by a colorectal surgeon as the patient was responding to appropriate resuscitation initially. The histology from the debrided tissue had unfortunately shown the involvement of cancer and the tissue culture grew Escherichia coli and Pseudomonas aeruginosa. She recovered well and was discharged on day fourteen with medical oncology follow-up for palliative chemotherapy.

Early recognition of necrotising fasciitis and aggressive surgical intervention are cornerstones of treatment to reduce patient mortality and morbidity [4]. Treatment for perforated rectal cancer is damage control resection of the perforated area with cancer and primary anastomosis or diversion stoma formation for stabilisation [4]. Delay in early recognition can lead to further tissue necrosis and higher mortality [2]. Areas of necrotic tissues need to be debrided, and swab cultures and biopsies should be done for targeted antibiotic therapies with possible secondary washout/relook to check the viability of the tissue [2].

This is a compelling case to discuss given its unusual presentation of a rectal cancer perforation with no background risk factors of NF. The patient did not have peritonism, nor was she in shock on presentation, the main notable clinical sign the patient had was buttock subcutaneous emphysema with tenderness. This case report reiterates the possibility of NF in patients with colorectal cancer perforation and the importance of early recognition and prompt management of NF and perforated colorectal cancer. The early management of NF includes antibiotic therapy and aggressive debridement surgery, and the management of perforated colorectal cancer is resection of the affected area with primary anastomosis or temporary stoma formation [2]. In conclusion, clinical suspicion of extraperitoneal cause should be sought in patients who present with lower limb and buttock subcutaneous emphysema [2,8].

5. Conclusion
NF is a rare presentation of perforated rectal cancer and subcutaneous emphysema is one of the clinical findings. This case and the images demonstrated that subcutaneous emphysema of the buttock should raise the concern of NF and the extraperitoneal cause should be sought promptly to decrease mortality and improve clinical outcomes.

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