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Case Report
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Gastrointestinal Fistula with Thigh Extension in a Palliative Care Patient with Intractable Pain: A Case Report

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Abstract

Spontaneous enterocutaneous fistulae occur as a complication in cancer patients, especially following irradiation, chemotherapy or cytoreductive operations. They are associated with worse prognosis, higher costs of treatment, and impaired quality of life. Proper recognition of the problem and fast implementation of selective therapy including water-electrolyte resuscitation, infection control, and nutritional support, control of output volume, proper wound care, and ultimately surgical treatment in selected patients may result in better symptom relief and improved quality of life of patients. We describe a patient with advanced rectal cancer and acute pain in the lateral aspect of thigh caused by the presumptive presence of an enterocutaneous fistula, in which therapy with antibiotics and surgical incision enabled rapid pain relief and comfort of dying.

Introduction

Gastrointestinal fistula is a pathological communication between the gastrointestinal tract and hollow viscera (internal fistula) or the skin (external fistula; enterocutaneous fistula). Although most gastrointestinal fistulae occur as a complication of surgery, especially for malignancy, 15% to 25% may occur spontaneously in patients with inflammatory bowel disease, following irradiation, ischemia, abdominal trauma, and in subjects with cancer [1]. They are associated with high morbidity and mortality rates, prolonged hospital stay, and increased costs of treatment [2]. The presence of a gastrointestinal fistula deteriorates patient's quality of life, as it is associated with pain, psychological distress, restrictions in daily life, dependence and social isolation [3].

We report a case of gastrointestinal fistula extending along the muscle planes to thigh in a terminally ill patient with rectal cancer, presenting with severe pain in the thigh.

Case Presentation

SD, a 44-year old man with a history of rectal cancer, was admitted to Palliative Care Ward of the University Hospital of Lord's Transfiguration because of pain in the lateral aspect of right thigh. The pain started abruptly one week before admission, as a consequence of a fall. The character of pain was somatic, and its intensity increased while moving or touching the limb. The maximum intensity of pain in Numerical Rating Scale (NRS) 0-10 was 9. The patient was bed-ridden since the moment of the fall. However, the home care staff had ruled out a fracture or other serious injury.

According to the family, the patient had slightly elevated body temperature, and his general condition had been rapidly deteriorating since the fall.

Seven months before admission the patient was diagnosed with a rectal cancer T4N2, infiltrating mesorectal fat tissue, right ischiorectal fossa, levator muscle of anus, sacral plexus and piriform muscle attachment to sacral bone, and obturating the rectum in the MRI imaging. After an exploratory laparotomy, a colostomy was performed followed by radiotherapy (5 Gy x 5 Gy). Control Magnetic Resonance Imaging (MRI), carried out three months after the radiotherapy and one month before the admission to Palliative Care Ward, showed decrease in the tumor size (from 75 mm x 75 mm to 53 mm x 60 mm x 68 mm) (Figure 1). The degree of infiltration of the surrounding tissues was comparable to the previous assessment. Moreover, two fistulas to the right and left gluteal muscles were revealed (100 mm x 20 mm to right buttock and 52 mm x 13 mm to

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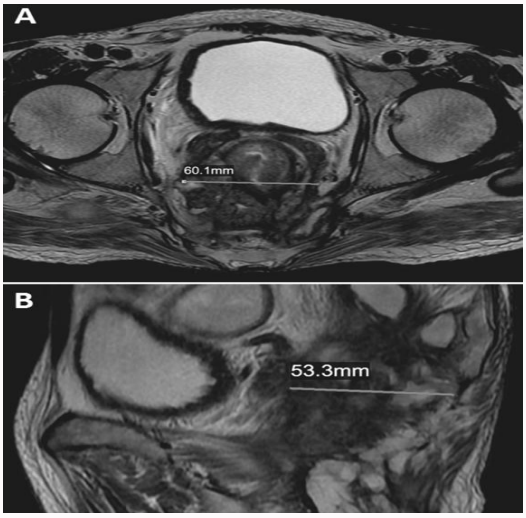

Figure 1: T2-weighted fast spin-echo magnetic resonance images. A) Axial image, B) Sagittal image. Tumor infiltrating mesorectal fat tissue, right ischiorectal fossa, levator muscle of anus, sacral plexus and piriform muscle attachment to sacral bone, and obturating the rectum.

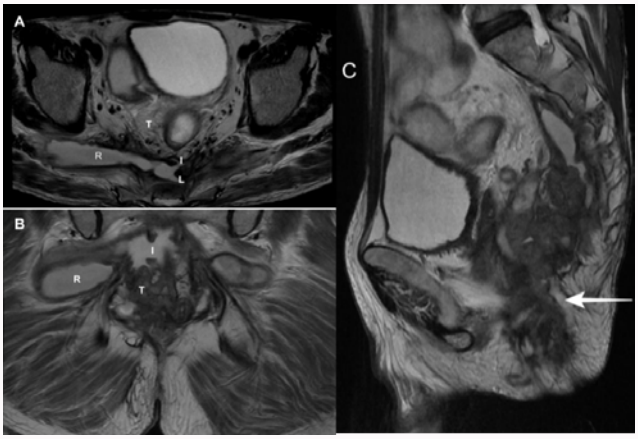

Figure 2: T2-weighted fast spin-echo magnetic resonance images. A) Axial image, B) coronal image, C) sagittal image. Fistulas to the right (R) and left (L) gluteal muscle. Internal opening of fistula's canal (I). Tumor mass (T). Arrow indicates the origin of fistula to the right gluteal muscles.

left buttock) (Figure 2).

Patient's comorbidities included cachexia, anaemia requiring several blood transfusions within previous seven months, and cigarette smoking. His daily treatment included 30 mg of morphine given in repeated Sub Cutaneous (SC) injections, 75 mcg/hr fentanyl patch, 20 mg of metoclopramide SC, and 750 mg of intravenous (IV) metronidazole.

On admission to the Palliative Care Ward, the patient was drowsy. His Karnofsky Performance Scale score was 30. On physical examination, crepitations were found in the right groin, and a fistula in the right buttock, near the anus, was revealed. The right thigh was swollen, with a cylindrical, slightly tender prominence along the anterolateral aspect, suggestive of an abscess. A 6-cm tumour, fluctuating and severely painful, was found at lateral aspect of the right knee. The skin over the prominence and tumor was slightly warm, but normal in color. Similar changes were found along the anteromedial side of the left thigh. Patient's body temperature was normal (36.2°C).

Figure 3: An incision of the tumor near the right knee resulted in discharge of feces-smelling material.

His heart rate was 104 bpm, and his systolic/diastolic arterial pressure was 82/55 mmHg. The first, tentative diagnosis was lymphoedema complicated with anaerobes infection, cellulitis, and subcutaneous emphysema.

Symptom treatment was continued with transdermal fentanyl patch 75 mcg/hour, continuous IV morphine infusion in increasing doses (from 40 mg to 80 mg per day) and metamizole 3,0 IV per day. Empirical therapy with IV amoxicillin/clavulanic acid 1.0 g b.i.d. and IV metronidazole 500 mg t.i.d. was started, and IV fluid therapy and low molecular weight heparin in prophylactic dose were given. On the second day of hospitalization, the pain control was much better (NRS 5-6). The patient was still drowsy, on physical examination tachycardia (121 bpm) and low blood pressure was found (94/62 mmHg). The tumor near the right knee was better formed. An incision of the tumor was performed with discharge of 500 mL of feces-smelling material (Figure 3), and fascial space drainage was placed. A continuous discharge was observed. The material was taken for bacteriological culture. The pain control was better (NRS 2-3), and patient's general condition improved. The character of discharge, together with the results of MRI performed before admission, suggested the presence of enterocutaneous fistula. The patient died (in good comfort) two days later, which precluded further diagnostics. Bacteriological culture revealed Escherichia coli, Proteus mirabilis and Proteus vulgaris, which indirectly confirmed the diagnosis of enterocutaneous fistula.

Discussion

We searched EBSCO database with 'gastrointestinal fistula', 'enterocutaneous fistula', and 'thigh as keywords. We found only one previously reported case similar to our patient [4], and three further cases, which were markedly different [5-7]. Saldua et al. [4] described a 64-year old patient with a history of postsurgical radiation for rectal cancer seven years earlier, who presented with subcutaneous emphysema of the thigh in the presence of urinary sepsis. Although diagnosis of necrotizing fasciitis was first suspected, abdominal Computed Tomography (CT) revealed an enterocutaneuos fistula to be the source of emphysema. The patient was successfully treated with four subsequent surgical debridements and wound vacuum placement. Similar to our patient, he was afebrile on admission, with tachycardia and low blood pressure on physical examination, and his patient complaint was pain in the thigh and deterioration of functional status.

Gastrointestinal fistulae in patients with cancer are often iatrogenic, with operative procedures and radiation being the most

important risk factors [8,9]. Of note, both our patient and the one described by Saldua et al. [4] were treated with radiotherapy for their rectal cancer. The risk of a fistula formation is also increased in cancer patients receiving bevacizumab or following cytoreductive surgery and hyper thermic intra peritoneal chemotherapy [8].

Management of gastrointestinal fistulae includes water-electrolyte resuscitation, infection control, and nutritional support, control of output volume, proper wound care, and ultimately surgical treatment [9]. From 20% to 30% of enterocutaneous fistulae close spontaneously (without surgical operation) within 2-3 months, but the prognosis is worse in patients with cancer and/or following radiation [9]. In patients with advanced gastrointestinal cancer, spontaneous closure of a fistula is not probable, and surgical treatment is usually impossible because of local advancement of the disease, and poor general status. In such patients, management should be focused on symptom relief.

As the predominant symptom in our patient with advanced rectal cancer was pain, we intensified systemic treatment with opioids, which resulted in moderate improvement in pain control. Local signs indicated anaerobes infection, so empirical therapy with amoxicillin/clavulanic acid and metronidazole was started. In subjects with *enterocutaneous fistulas*, antibiotics active against large bowel flora, including *Bacteroides spp*, *Enterobacteriaceae*, *Enterococcus spp*, and Proteus spp limits the extent of inflammatory infiltration and decreases the risk of life-threatening sepsis. In such patients, mono therapy with ticarcillin/clavulanic acid, tigecycline, or ertapenem should be started. Alternatively, metronidazole may be used in combination with cefazolin, cefuroxime, ceftriaxone, or cefotaxime [10,11].

In patients with better prognosis, further measures, including parenteral and/or enteral nutrition and negative pressure wound therapies could be implemented [8,9]. In high-output fistulas therapies with anti-diarrheal or octreotide has also been used with success [9], including terminally ill patients [12]. In our patient, the applied management combining empirical antibiotic therapy and surgical debridement was implemented too late to improve his life, but it improved quality of life in his last days and comfort of dying.

Spontaneous enterocutaneous fistulas may occur as a complication of advanced gastrointestinal cancer. The possibility of a gastrointestinal fistula should be taken into account in patients with gastrointestinal cancer, presenting with otherwise unexplained or exacerbated pain, especially in subjects with fever, following radiotherapy, cytoreductive surgery, hyper thermic intra peritoneal chemotherapy, or treatment with bevacizumab. General and local symptoms and signs of infection may be vague or even absent. A poor general condition of patients usually precludes confirming the diagnosis with CT or MRI imaging. Although the therapeutic experience is very limited, proper recognition of the problem and fast implementation of selective therapy together with symptomatic treatment may result in better symptom relief and improved quality of life of patients.

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