

CASE REPORT**A Rare Case of Crohn's Disease Presenting as a Chronic Gluteal Abscess with Underlying Sacral Osteomyelitis**Reice Brown,¹ Aria Rezaie,¹ Philip Napolitan, DO²¹Kansas City University, Kansas City, MO, United States²Department of Medicine, North Kansas City Hospital, North Kansas City, MO, United States

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ABSTRACT

This case report discusses the atypical presentation of Crohn's Disease (CD) in a male in his early twenties. The patient presented to the Emergency Department with a chronic left gluteal abscess that failed to resolve despite previous interventions. Over the course of several months and multiple hospital admissions, the patient was seen by various consulting services in a workup that revealed sacral osteomyelitis and an associated pelvic abscess. During an outpatient follow-up, the patient was again admitted to the hospital for the suspicion that irritable bowel disease was the root cause of the infection. The patient was subsequently diagnosed with CD and underwent an ileocectomy. Patients with perianal Crohn's Disease (pCD) are often met with a delayed diagnosis, which creates a deferred treatment plan and negatively

impacts patient health. This case report serves as a learning tool and reminder for clinicians to consider CD in patients presenting with isolated perianal disease.

INTRODUCTION

Crohn's Disease is a form of irritable bowel disease that continues to grow in incidence in the United States.¹ The effects of untreated CD can have grave consequences for a patient's health, so it is important for patients to be diagnosed in a timely manner. It is uncommon for CD to initially present as a severe disease, such as pCD, but it does occur. These presentations tend to have a poorer prognosis for patient health.² It is imperative for healthcare providers to recognize symptoms of perianal disease as a possible initial presentation of CD.

The presentation of CD as sacral osteomyelitis is an exceedingly rare

complication of the disease process, let alone the initial presentation. A review of the literature revealed minimal case reports of CD presenting as sacral osteomyelitis and no analyses relating the two processes as far as we could tell. This rare presentation of CD as sacral osteomyelitis with associated abscess and fistula serves as an essential educational tool for healthcare providers in recognizing insidious irritable bowel disease in patients.

CASE PRESENTATION

A male patient in his early twenties presented to the Emergency Department for sustained drainage from an abscess in the left upper buttock. He reported that he had previously undergone an incision and drainage procedure by his primary care provider and the wound culture grew *Group B Streptococcus* at that time. The patient had been started on amoxicillin and was still taking antibiotics at the time of Emergency Department presentation. He only admitted to fatigue and occasional heartburn. The patient had no significant medical or surgical history. His medications included amoxicillin and ibuprofen as needed. The patient reported he consumed 1-2 glasses of wine per month and denied tobacco or illicit substance abuse. His family history was significant for type 2 diabetes mellitus and hypertension in both biological parents.

Examination revealed vital signs within normal limits. His weight was 132.2 pounds. There was a left upper buttock abscess that was 8 cm from the midline with a small fistulous opening. A small amount of purulent fluid came out with palpation. The

opening extended inferiorly at least 3 cm with probing. There was no evidence of pilonidal disease, midline pits, or masses. The patient subsequently underwent a surgical incision and debridement for a large chronic abscess of the left upper outer buttock. Operational findings were significant for a 10x8 cm cavity with a large amount of chronic granulation tissue. The cavity appeared to go below the fascia of the buttocks and could be probed along the bone. No purulent drainage was expressed from this area. A complete blood count revealed microcytic anemia with hemoglobin of 11.8 g/dL, so the patient was started on oral iron supplementation at discharge. The patient followed up with wound care for vacuum-assisted wound closure (wound VAC) and further management.

Over the course of the next two months, the patient's wound failed to heal and generated two deep sinus tracts. The failed treatment plan included a wound VAC, a 10-day course of amoxicillin-clavulanate, and repeated debridement. At the two-month mark, the patient began to experience low-grade fevers at home and felt poor overall. The patient stated he was experiencing some alternating constipation and fecal urgency along with mild dyspnea on exertion. He also reported neurologic pain down the back of his left leg for several days. The physical exam was only remarkable for the left gluteal area with copious amounts of purulent drainage and suspected cavities over the left sacroiliac joint and posterior superior iliac spine area. At this time a computerized tomography (CT) scan was ordered, and it was

recommended the patient be admitted to the hospital.

The patient then presented to the hospital as a direct admission. His temperature was 102.8 degrees Fahrenheit along with a heart rate in the low 100s beats per minute on admission; other vitals were within normal limits. CT scan of the abdomen and pelvis with contrast revealed no definitive CT evidence for osteomyelitis. It did however reveal a probable subcutaneous abscess measuring 6 cm along the left posterior paraspinal musculature with air pockets that extended towards the gluteus maximus. It also showed diffuse bowel wall thickening involving the distal/terminal ileum measuring 10 cm, and angulated bowel in the sigmoid colon and proximal rectum with associated bowel wall thickening in the area adjacent to the soft tissue density. There was no definitive fistula identified on CT. The patient was started on empiric vancomycin, cefepime, and metronidazole. Labs were significant for elevated C-reactive protein at 13.6 and erythrocyte sedimentation rate at 64. The iron panel was consistent for anemia of chronic disease. The following day, a magnetic resonance image of the pelvis with and without contrast revealed extensive inflammation surrounding the sacrum and also involving the posterior aspect of the gluteal muscles, as well as the piriformis muscles. It also showed multiple areas of gas and fluid along with bone marrow edema of the sacrum and the first lumbar segment. These findings were strongly suspicious for osteomyelitis. The wound culture grew positive for *Escherichia coli*, *Bacteroides*, *Clostridium*, and coagulase-

negative *Staphylococcus*. The methicillin-resistant *Staphylococcus aureus* swab was negative. Empiric antibiotics were changed to daptomycin and ertapenem. A peripherally inserted central catheter line was placed for a 6-week course of intravenous antibiotics upon discharge.

Two days following discharge, the patient followed up with wound care. The physician had a suspicion that the patient's fistula could be caused by underlying irritable bowel disease due to the nature of the wound and terminal ileal findings on prior CT. The patient was again directly admitted to the hospital for surgery and gastroenterology consultation. Surgery was consulted, and a laparotomy was planned with a possible ileal colectomy, as there appeared to be a suspicious area on CT indicating the presence of a fistulous connection between the small bowel and the phlegmonous presacral process which connected to the skin. Gastroenterology recommended a biological agent once the infection subsided. Daptomycin and ertapenem were continued inpatient. Laparotomy revealed a single loop of thickened rubbery distal ileum welded to the presacral fascia. An ileocecectomy with subsequent ileocolic anastomosis was performed. The pathology report of the terminal ileum and cecum revealed marked transmural acute on chronic inflammation with architectural distortion that confirmed the diagnosis of CD. The patient was medically stable and discharged home with follow up appointments with gastroenterology and infectious disease in the outpatient setting.

DISCUSSION

A severe phenotype of CD, known as perianal Crohn's Disease (pCD), is characterized as severe fistulizing lesions with or without abscesses. This form of CD is correlated with poorer health outcomes and increased risk for hospitalizations, surgical resections, and the need for immunosuppressive therapy.² In the Crohn's population, 10-30% of patients develop abdominal or pelvic abscesses. Most of these incidences occur in the postoperative setting, and it is less common for this manifestation to be the presenting symptom.³ However, there are instances where extraintestinal and perianal manifestations of CD present before typical luminal symptoms.^{1,2} One study revealed that 17.2% of patients with CD had symptoms of pCD before an official diagnosis of CD was able to be made.⁴ A meta-analysis identified that approximately 4% of patients with CD initially present with pCD without any luminal symptoms.² While pCD is a rare presentation, it is necessary for clinicians to be aware that perianal disease can be due to underlying CD.

The pathogenesis of pCD starts with an initial insult to the tissues that triggers a granulomatous response with local infiltration pockets of macrophages, epithelioid cells, and multinucleated giant cells.⁵ This can lead to an increase in transforming growth factor- β , tumor necrosis factor (TNF), and interleukin 13, which triggers an epithelial-to-mesenchymal transformation in the tissues. With this transformation, there is an increase in matrix metalloproteinases that cause tissue

remodeling leading to fistula formation.⁶ Abscess formation occurs during an active disease process because the altered barrier function increases permeability in the intestinal wall and allows an invasive bacterial infection to occur. The immune dysfunction secondary to malnutrition in active disease also permits bacterial infection.⁷ The rare occurrence of sacral osteomyelitis in this process is possible due to the direct extension of the fistula into the bone, or erosion of the bone in a contiguous abscess.³

In regards to pCD, the duration of fistula presence is a poor prognostic factor.⁸ It is estimated that 20% of patients had strictures or penetrating disease at the time of CD diagnosis, which suggests that there is clinically silent inflammation occurring before diagnosis. A study in China revealed that 17.5% of patients with CD had asymptomatic perianal fistulas. Those with CD who are diagnosed early in the disease process and receive proper treatment have decreased risk of fistulas, abscesses, hospitalization, and surgical intervention. These patients also have better long-term prognoses.⁹

Early recognition of CD and efficient management is the key to successful treatment. The early use of anti-TNF therapy, along with surgical intervention, can minimize the complications of pCD.¹⁰ Delayed diagnosis of CD is a common issue and is associated with poorer health outcomes.⁹ A study showed that the median duration of time from the presentation of pCD in a patient with a new diagnosis to anti-TNF therapy was 365 days. Even with optimal interservice cooperation, the healing

rate for Crohn's fistulae is reported to be less than 30%. Delays in care often lead to substantially decreased quality of life for patients and have a significant influence on the effectiveness of treatment.⁸ The presentation of isolated perianal disease should increase the suspicion of CD among clinicians. Increased clinical suspicion for CD and more efficient healthcare management can lead to better treatment efficacy for patients with pCD.

Notes

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