

Pelvic osteomyelitis complicating Crohn's disease

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WCP KWAN, HJ FREEMAN. Pelvic osteomyelitis complicating Crohn's disease. *Can J Gastroenterol* 1993;7(3):293-296. Although rheumatologic complications of Crohn's disease are common, osteomyelitis associated with Crohn's disease rarely is described. In this report, a 29-year-old man with Crohn's disease was seen with ileorectal fistula, pelvic abscess and severe back pain. The abscess and fistula were treated surgically but the patient had persistent fever and pain. Computed tomography scan showed destruction of the sacrum, and bone scan demonstrated increased sacral uptake. Osteomyelitis was suspected and confirmed on bone biopsy. *Streptococcus viridans* was isolated from bone culture. Treatment with parenteral penicillin was successful and follow-up revealed no recurrence of osteomyelitis.

Key Words: Crohn's disease, Fistula, Inflammatory bowel disease, Osteomyelitis, *Streptococcus viridans*

Ostéomyélite pelvienne dans la maladie de Crohn

RÉSUMÉ: Bien que les complications rhumatologiques de la maladie de Crohn soient fréquentes, l'ostéomyélite associée avec cette maladie est rarement décrite. Dans ce rapport, un homme de 29 ans atteint de maladie de Crohn s'est présenté avec une fistule iléo-rectale, un abcès pelvien et une intense douleur au dos. L'abcès et la fistule ont été traités chirurgicalement, mais le patient est demeuré fébrile et souffrant. Une tomographie à densité osseuse a révélé un certain degré de destruction du sacrum et une TDM osseuse a révélé une hyperfixation sacrée accrue. L'ostéomyélite était probablement en cause, ce qui a été confirmé par une biopsie osseuse. *Streptococcus viridans* a été isolé dans la structure osseuse. Le traitement avec de la pénicilline parentérale a été couronné de succès et le suivi n'a révélé aucune récurrence de l'ostéomyélite.

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COMMON MUSCULOSKELETAL complications in patients with Crohn's disease include peripheral arthritis, spondylitis and, less commonly, granulomatous bone or muscle disease as well as periosteal new bone formation with clubbing. Despite the frequent occurrence of inflammatory processes including abscess or fistula formation adjacent to pelvic bones, osteomyelitis has been rarely recorded. The present report describes a patient with Crohn's disease complicated by pelvic sepsis and sacral osteomyelitis, and reviews the previous literature on this apparently unusual complication.

CASE PRESENTATION

A 29-year-old man was admitted to University Hospital in July 1986 for evaluation of fever, weight loss and diarrhea. Crohn's disease had been diagnosed at age 24 with involvement of the ileocecal region and sigmoid colon; improvement resulted from a course of prednisone and sulphasalazine, but within several months he developed fever and increasing abdominal pain. Laparotomy revealed a retroperitoneal abscess that was drained with no intestinal resection done.

The patient remained well until April 1986 when he noted lower ab-



Figure 1) Barium enema showing a lateral view of the pelvis, including the sacrum. A marked increase in the retrorectal space is observed

dominal cramping pain, watery diarrhea with weight loss of 8 kg and onset of constant lower back pain and fever of

39°C. Examination revealed moderate lower abdominal tenderness. Percussion tenderness over the lower lumbar spine and sacrum was also present. A psoas sign was not elicited and he had normal hip joint mobility. No neurological deficit was demonstrated.

A barium enema revealed an increase in retrorectal space with changes of Crohn's disease present in the sigmoid colon, including a pelvic fistulous tract (Figure 1). A small bowel follow-through showed involvement of the terminal ileum, and early filling of the rectum consistent with an ileorectal fistula. Computed tomography (CT) scan of the abdomen showed tethered loops of ileum in the pelvis with an associated inflammatory mass: no clear cut abscess cavity was defined. Radiographs of the lumbosacral spine showed normal sacroiliac and hip joints, but a small bony fragment was seen at the antero-superior aspect of S1 together with spondylolithesis of L5-S1.

The patient was treated with total parenteral nutrition and antibiotics, including gentamicin and metronidazole. Laparotomy, revealed a large pelvic inflammatory mass involving the distal

ileum, cecum and rectosigmoid. Fistulous tracts were demonstrated between the ileum and rectum. Ileocecal resection and reanastomosis was done with a sigmoid resection and colostomy.

Postoperatively, the patient's condition improved and the antibiotics were discontinued. His abdominal pain resolved and the colostomy functioned normally. However, he continued to have low grade fever and complained of increasing back pain with radiation down his right leg and mild urinary hesitancy. Multiple blood cultures were negative. Repeat neurological assessment revealed no abnormality. CT scan of his spine now showed multiple bony fragments anterior to the first sacral segment, with irregular resorption of the body of S1. These changes were consistent with sacral osteomyelitis (Figure 2). A bone scan demonstrated increased uptake in the upper sacrum and CT-guided needle biopsy of the sacrum showing histological changes of osteomyelitis but cultures from the biopsy material revealed no organism. A myelogram showed no evidence of obstruction or epidural defect.

The patient was treated with penicillin G and gentamicin. In an effort to obtain a specific bacteriologic diagnosis, an open biopsy of the sacrum was done. Tissues from the sacrum yielded a heavy growth of *Streptococcus viridans* and the histology demonstrated areas of osteonecrosis along with acute and chronic inflammation consistent with osteomyelitis. No granuloma was seen and an acid fast stain was negative. As *Strep viridans* is an unusual cause of pelvic bone osteomyelitis, the patient had further blood cultures and underwent an echocardiogram to exclude endocarditis (these studies were negative).

The patient received six weeks of parenteral penicillin G therapy with good clinical response. There was resolution of fever and back pain, and his erythrocyte sedimentation rate decreased from 50 to 12 mm/h. A follow-up bone scan near the conclusion of treatment showed only slight uptake in the sacrum with no appreciable change in plain radiographs. The patient was discharged in October 1986 and his colostomy was closed in November 1986.

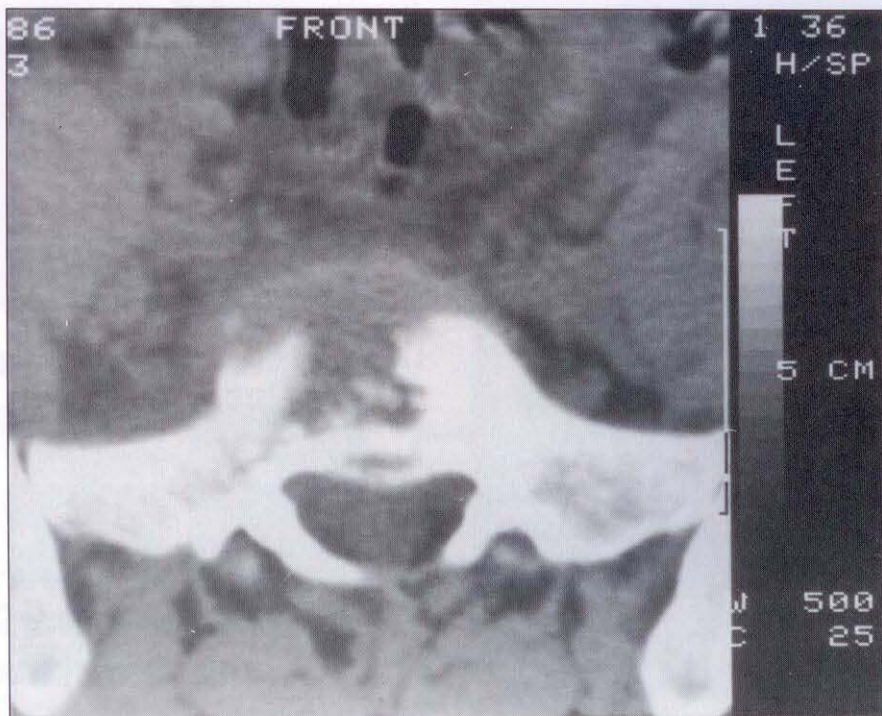


Figure 2) Computed tomography scan view of S1 showing a lytic lesion associated with sacral osteomyelitis. Subsequent biopsies confirmed the presence of osteomyelitis and cultures revealed

TABLE 1
Crohn's disease and osteomyelitis

Year (reference)	Age (gender)	Location of Crohn's disease	Location of osteomyelitis	Organism(s)
1969 (3)	51 (male)	Ileum, cecum, ileocutaneous fistula	Right ileum	Not stated
	28 (male)	Ileum, presacral abscess	L5, sacrum	β -hemolytic, streptococcus
1971 (4)	17 (male)	Ileum, colon, right psoas abscess	Right ileum, right femoral head	<i>Bacteroides</i> , peptostreptococcus, <i>Clostridium perfringens</i> , <i>Escherichia coli</i> , group D streptococcus
1973 (5)	20 (female)	Small bowel, colon, perirectal abscess	Sacrum	α -hemolytic streptococcus
1973 (6)	12 (male)	Left colon, retroperitoneal abscess	Left ileum	Not stated
	16 (male)	Ileum, paracecal abscess, enterocutaneous fistula	Right ileum	Not stated
	17 (male)	Ileum, cecum, rectum, enterovesical fistula, intra-abdominal abscess	Right ileum	Not stated
1984 (7)	20 (female)	Ileum, colon, right psoas abscess, intra-abdominal abscess	Sacrum	Not stated
1987 (8)	17 (male)	Terminal ileum, presacral abscess	Sacrum	<i>E coli</i>
Current case	29 (male)	Ileum, cecum, sigmoid sacrum, ileorectal fistula	Sacrum	<i>Streptococcus viridans</i>

Subsequent follow-up to December 1991 has been uneventful with no further relapse of his Crohn's disease or recurrent osteomyelitis.

DISCUSSION

Although many inflammatory disorders of the intestinal tract develop close or adjacent to the bony pelvis, pyogenic osteomyelitis involving the pelvic bone distinctly is uncommon. In a review of 616 cases of osteomyelitis, only 5% involved the pelvic bony structures (1). Usually, the infection is spread by contiguous extension from soft tissue foci and, less commonly, from intra-abdominal or pelvic abscesses. The infection usually is localized to the ileum since it is the largest pelvic bone and has an abundant blood supply.

Pelvic osteomyelitis appears to be an unusual complication of Crohn's disease despite the frequent presence of an associated chronic inflammatory pelvic mass, abscess and/or fistulous tracts. This contrasts the frequency of other musculoskeletal disorders experienced by patients with inflammatory bowel disease which can be as high as 40 to 50% (2). The precise incidence of pelvic osteomyelitis in Crohn's disease is unknown; however, based on the clinical descriptions of this associated complication in the literature, it seems to be rare. The first two cases of pelvic

osteomyelitis in Crohn's disease were described in 1969 by Goldstein et al (3). Since that time, only 10 additional cases have been reported (Table 1).

The majority of reported cases involved the right ileum. This undoubtedly is related to the fact that the coadjacent terminal ileum and cecum are the most common sites of Crohn's disease. In most reported patients, an adjacent abscess or fistula was present, suggesting that infection resulted from seeding to contiguous bone. The only exception was a case with osteomyelitis of the left femur and Crohn's disease; the patient described also had an *E coli* septicemia and was being treated for Crohn's disease with corticosteroids and immunosuppressive agents (4). The current patient had osteomyelitis involving the sacrum, likely as a sequela of the pelvic inflammatory mass with an ileorectal fistula: associated presacral infection and abscess formation became evident. In all three previously described cases with sacral osteomyelitis in the setting of Crohn's disease, presacral and perirectal abscesses were also present. In almost all instances the diagnosis of Crohn's disease was made before osteomyelitis was discovered – this is not surprising since osteomyelitis appears to occur exclusively in the setting of complicated Crohn's disease. However, Schwartz et

al (5) described a case of sacral osteomyelitis as the initial presentation with Crohn's disease found only at surgery despite extensive radiographic preoperative evaluation.

The significant clinical feature that suggested osteomyelitis in the present patient as well as in all reported cases to date, was severe and persistent pain in the affected area. This can sometimes be overlooked because the patient is frequently chronically ill with other complications of Crohn's disease, and the pain in the abdomen or affected areas associated with of either an abscess or fistula is expected. Furthermore, sacroileitis and spondylitis complicating chronic inflammatory bowel disease can certainly result in disabling and sometimes severe back pain. The plain radiographs of the lumbosacral spine in the current patient revealed progressive changes resulting in further evaluation with CT and bone scans that strongly suggested sacral osteomyelitis. Subsequent definition of the typical pathological features combined with microbiological studies unequivocally demonstrated its presence. It is well known that typical radiographic osseous changes of osteomyelitis may not appear for days or even weeks (as illustrated in the patient described here). Bone scans may offer improved sensitivity approaching 100%, and

other imaging modalities, such as CT scan, may aid in detection of sequestra (6) and help delineate anatomical alterations. At the time the patient was evaluated, magnetic resonance imaging was not available in the authors' hospital. Recent literature has demonstrated that this imaging modality is indispensable to evaluate osteomyelitis – not only is its specificity reported to be superior but it is useful in defining the extent of the inflammatory process and can distinguish osteomyelitis from cellulitis (7).

Neurological complications of pelvic sepsis in Crohn's disease were considered in the current patient, especially with the development of radiating pain to his right thigh and urinary hesitancy. However, no objective neurological abnormality was evident and his myelogram was normal. Although no permanent neurological deficit was seen, the development of pelvic sepsis, particularly if sacral osteomyelitis is documented, may result in further extension of the inflammatory process into the spinal canal.

Indeed, there are a number of prior descriptions of Crohn's disease with fistula formation extending to the spinal canal causing serious complications, such as spinal epidural abscess. Aitken (8) first reported a case of epidural abscess in a 36-year-old man with Crohn's disease of the terminal ileum and a pelvic inflammatory mass resulting in paraplegia. Sacher (9) described a case of spinal epidural abscess from L2 to S4 in an 11-year-old boy with Crohn's disease of the distal colon and right psoas abscess. Hershkowitz (10) reported a 19-year-old man with epidural and subdural spinal empyema originating from rectal fistula.

It is anticipated that the organisms involved in pelvic osteomyelitis originate from the bowel flora; however, documentation of the offending pathogen often appears to be difficult and may be complicated by the frequent use of antibiotics to treat septic complications in these patients. Culture from draining sinuses, adjacent abscesses or infected cavities may be misleading. A

bone biopsy may offer the best opportunity to document the bacteria involved. In a comprehensive review of osteomyelitis, the frequency of positive cultures in relation to the source of specimens was 60% for bone aspirate and 65% for bone pus obtained at surgery (11). There was a heavy growth of a single organism, *Strep viridans*, from the bone biopsy and the present patient was treated successfully with penicillin.

In summary, despite the many intra-abdominal complications that occur in patients with Crohn's disease, osteomyelitis is rare and invariably develops in the presence of an intra-pelvic abscess or fistula with contiguous seeding of infection. Diagnosis may be made with appropriate radiographic imaging and bone scan methods along with microbiological studies. Heightened suspicion in the patient with persistent back or bone pain, however, is most important as this likely will enhance detection and limit potential serious morbidity due to osteomyelitis and associated neurological complications.

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