CASE REPORT

Abdominopelvic actinomycosis mimicking disseminated peritoneal carcinomatosis

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SUMMARY

We present a case of a 38-year-old woman who presented with symptoms suggestive of intra-abdominal or pelvic malignancy: marked weight loss, abdominal pain, altered bowel habit, anorexia and fatigue. The findings of multiple peritoneal deposits, adnexal and presacral masses on CT imaging and appearances on diagnostic laparotomy also suggested malignancy. However, the histological analysis was inconsistent with malignancy and revealed an infection with *Actinomyces israelii*. The patient started a course of intravenous antibiotics and complete resolution is expected. An intrauterine contraceptive device was identified as the likely source of the infection.

BACKGROUND

This case highlights the importance of considering alternative diagnoses masquerading as intra-abdominal or pelvic malignancy, particularly actinomycosis in association with an intrauterine contraceptive device (IUCD). We discuss the diagnosis as well as the treatment of abdominopelvic actinomycosis and highlight similar case presentations. The impact on the patient of this diagnostic dilemma is illustrated in the patient's account.

CASE PRESENTATION

This 38-year-old woman presented with a 3 month history of altered bowel habit, loose stools alternating with constipation, postprandial abdominal pain, anorexia, 15 kg weight loss and in the 2 weeks prior to admission, nausea, vomiting and night sweats.

During this period, she had been investigated for possible Crohn's disease on an outpatient basis and, 1 month previously, had normal findings on an oesophagogastroduodenoscopy and a colonoscopy. She also had a normal ultrasound of her abdomen and pelvis at this time. At the time of admission, an outpatient magnetic resonance enterography was pending.

She had previously been healthy with only a history of mild asthma, for which she took budesonide inhalers. She took no other regular medication, was a non-smoker and had minimal alcohol consumption. She had an IUCD inserted 4 years previously. Approximately 10 years ago, she had travelled to West Africa as part of her job as a hydrogeologist, but had not travelled abroad since. Her paternal grandmother and great-grandmother had died of colorectal cancer.

On examination, the patient was cachectic, pale and dehydrated, with a palpable mass in her left iliac fossa and mild hepatomegaly.

INVESTIGATIONS

Blood tests showed raised inflammatory markers and a microcytic anaemia (white cell count $13.1 \times 10^9/L$, C reactive protein 120 mg/L and haemoglobin 7.2 g/dL, mean cell volume 74 fL).

A CT scan of her abdomen and pelvis was performed, showing large, complex, cystic/solid masses in the right adnexa and presacral areas, and smaller masses in the left flank, left iliac fossa and anterior pelvis with tethering of small bowel loops in the pelvis and several low attenuation lesions in the liver (figure 1). Findings from a subsequent staging CT chest were normal.

DIFFERENTIAL DIAGNOSIS

At this stage, the most likely differential diagnosis was considered to be disseminated malignancy, with the right ovary as the most likely primary site. However, the normal ultrasound performed 6 weeks earlier was somewhat inconsistent with this. Ultrasound has a high sensitivity for diagnosing ovarian tumours, estimated to be 89.5% in a recent large multicentre study.¹ Tumour markers were also largely normal, with only a slightly elevated CA-125 (83 IU/mL). Primary gastrointestinal malignancy or Crohn's-associated inflammatory deposits were considered to be less likely in the

Figure 1 Sagittal CT scan of the patient's abdomen and pelvis demonstrates numerous pelvic masses (arrows). This appearance, combined with the presence of hepatomegaly, raised the concern of an ovarian malignancy with peritoneal spread.

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context of her normal colonoscopy findings. Infection, especially tuberculosis given this patient's previous travels, was also considered. Unexpectedly, tumour markers were largely normal, with only a slightly elevated CA-125 of 83 IU/mL.

An attempted ultrasound-guided percutaneous biopsy of peritoneal deposits was unsuccessful owing to overlying loops of bowel. A diagnostic laparoscopy for tissue biopsy was therefore performed (figure 2). Again, intraoperative laparoscopic appearances were considered to be suggestive of malignancy. While awaiting the tissue biopsy results, the suspicion of malignancy was discussed with the patient and she was referred to, and seen by, the oncology team.

OUTCOME AND FOLLOW-UP

However, to the medical team's and the patient's relief, the histological analysis of the tissue biopsy, in conjunction with microbiological analysis, revealed colonies of *Actinomyces israelii* bacteria surrounded by acute suppurative exudate and fibrosing chronic inflammation (figure 3). The presence of the IUCD was recalled and the IUCD was immediately removed. The patient was started on a 2-week course of intravenous benzylpenicillin, with four further weeks of intravenous ceftriaxone and 4 weeks of amoxicillin oral follow-on therapy planned. Complete resolution of infection is expected, and will be assessed by repeat cross-sectional imaging. Interestingly, the culture of the IUCD and the ascitic fluid obtained intraoperatively were both negative for infection. However, actinomyces-like organisms had been noted on a previous smear test from the patient in 2012.

DISCUSSION

We presented a case in which pseudotumours and pelvic masses resulting from actinomycosis were initially suspected to be malignancy. Actinomycosis is a rare, chronic, granulomatous infection caused by *Actinomyces* species and may affect the orocervicofacial, thoracic or abdominopelvic regions. The most important human pathogen among the genus *Actinomyces*, and the causative agent in this case, is *A israelii*, a filamentous, Gram-positive, anaerobic, commensal bacterium that normally colonises the oral cavity, lower gastrointestinal and genitourinary tracts.² *A israelii* can produce opportunistic infections; any body site can be infected provided that a break in the mucous membrane exists and, although rare, disseminated spread of the infection can occur. However, infection is endogenous as there is no person-to-person spread.³

Figure 2 Right ovarian actinomycosis deposit.

Figure 3 Histological analysis of the peritoneal biopsy demonstrated acute fibrosing inflammation in association with actinomycetes colonies. These bacterial colonies are composed of filamentous organisms and are surrounded by an acute suppurative exudate. There was no evidence of dysplasia or malignancy within the specimen (H&E, ×200 magnification).

A israelii becomes saprophytic in low oxygen levels, allowing penetration of the mucosa. The resulting host inflammatory response can lead to the formation of slow growing, multiloculated abscesses and pseudotumours. These become symptomatic once they become sufficient in size to obstruct or fistulate with nearby structures, discharging pus. Abdominopelvic actinomycosis is rare, but most commonly seen in association with an IUCD or after surgery to the region, for example appendicectomy.³ *A israelii* can affect 1.65–11.6% of IUD users and infection is more common in women who have had an IUD for more than 4 years.⁴ Notably, actinomyces-like organisms were seen on our patient's cervical smear test in 2012, making her IUCD a highly likely source.

As was the case in this patient, actinomycosis classically presents with non-specific symptoms and is therefore difficult to diagnose on clinical grounds. When examined microscopically, the pus discharged from fistulae contains clusters of bacteria, producing the classical macroscopic appearance of yellow 'sulfur granules': rounded basophilic masses with eosinophilic terminal clubs on staining with H&E. This 'sulfur granule' appearance is not unique to *Actinomyces* species, but can be differentiated from other bacterial infections with a similar appearance by the observation of filamentous branching bacteria at the periphery on Gram, Gomori methanamine-silver and Giemsa stains. Alternatively, direct identification can be achieved using a species-specific antibody. Gram stains are usually more sensitive than culture, which can be insensitive in up to 76% of cases.^{2 3} In addition, actinomycetes are slow growing bacteria and can take up to 3 weeks to culture on selective media.

Prolonged courses of antibiotic therapy are the cornerstone of treatment for actinomycosis. In vitro studies show sensitivity of actinomycetes to a wide range of antimicrobials, although they are resistant to cephalexin and metronidazole. Penicillin is a common first line agent for the treatment of actinomycosis and the development of penicillin-resistance is low.⁵ Antibiotics are typically given for at least 3 months, although complete resolution has been reported with shorter courses.⁶ Surgical excision of necrotic tissue and drainage of abscesses may also be beneficial, particularly in the context of refractoriness to medical therapy.⁷ Resolution of infection should be confirmed by repeat CT imaging.

Several previous case reports highlight the fact that actinomycosis can mimic malignancy.² Failure to consider a diagnosis of actinomycosis can lead to unnecessary surgery and delay in appropriate treatment. Indeed, it is estimated that fewer than 10% of cases are diagnosed preoperatively.² Delays in diagnosing actinomycosis can also lead to unnecessary distress for the patient and their family.

Patient's perspective

I accumulated symptoms over the first few weeks, including tiredness, abdominal pain, loss of appetite, and diarrhoea. Blood tests revealed inflammation and I was told that it was likely that I had inflammatory bowel disease. In agony and despair, I was admitted to hospital. A CT scan showed masses and we were gently told that this was not Crohn's disease but might be cancer, infection, tuberculosis, or inflammatory disease. This was a shock after believing for so long that I had Crohn's.

I was told that a biopsy should be diagnostic and waited several days for a laparoscopy: days of nil by mouth were ironic for someone admitted to hospital with malnutrition. I was so relieved at having had the laparoscopy and being able to eat again, that I was fairly calm at the news that it looked like ovarian cancer. I already understood that this was possible, now it was highly likely. However, my husband was devastated and started planning for the worst, leaving me to try and absorb his anguish.

When the biopsy results came back I was almost numb to the news – an infection! I was amazed that simple antibiotics would make me well again. Of course, I felt hugely relieved, but the whole process of illness, waiting and uncertainty, has left me feeling fragile and vulnerable.

Learning points

- ▶ Actinomycosis is an important differential diagnosis in suspected abdominal or pelvic malignancy, particularly in the context of an IUCD.
- ▶ Clinical symptoms, radiological findings and macroscopic appearance are non-specific and diagnosis relies on histological assessment.
- ▶ Direct tissue biopsy is often required due to the prolonged incubation time and relative insensitivity of microbiological culture.
- ▶ Actinomycosis is treatable with prolonged doses of intravenous antibiotics, plus surgery for more extensive disease.

Competing interests None.

Patient consent Obtained.

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