Actinomycosis: an unusual cause of unresectable rectal cancer

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ABSTRACT

We present a very unusual case of a woman with an intrauterine contraceptive device (IUCD) who developed pelvic actinomycosis during long course chemoradiotherapy for rectal cancer, which presented a significant diagnostic challenge and eventually rendered the cancer unresectable. IUCDs are often implicated in the development of pelvic actinomycosis but there is no current evidence or guideline to suggest that they should be removed prior to oncological treatment. Owing to the devastating consequences of this combination of disease, we suggest that it may be prudent to remove IUCDs in this setting.

KEYWORDS

Rectal cancer – Actinomycosis

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A 47-year-old woman presented with rectal bleeding and urgency, and was diagnosed with a low rectal adenocarcinoma. Staging with magnetic resonance imaging and computed tomography (CT) suggested a T3N2M0 cancer with probable extramural vascular invasion and the tumour extending to the mesorectal fascia on the right (Fig 1). An intrauterine contraceptive device (IUCD) was noted. The patient underwent long course chemoradiotherapy and was restaged three months later. Around this time she began to feel unwell and feverish, with increasing abdominal pain. Her repeat imaging showed some response in the tumour but new abnormalities in the right adnexa with tethering of the rectosigmoid, which was thought to represent an inflammatory process (Fig 2). Omental infiltration was also noted. Although the pattern was unusual for disease progression, this was clearly high on the list of differential diagnoses and, initially, a CT guided omental biopsy was planned.

Prior to this taking place, the patient presented urgently with a huge abdominal wall abscess in her right iliac fossa and abnormal induration of the left upper abdominal wall (Fig 3). The abscess was incised under anaesthesia and over a litre of pus drained. Bacterial culture yielded mixed anaerobes but no specific organisms. At the same time, operation biopsies were taken from the thickened tissue in the left abdominal wall, which was thought to represent possible metastatic disease. Histological analysis revealed fibrosis and active chronic inflammation only.

A month later, further CT showed increased volume of the intra-abdominal soft tissue abnormalities, with disease

in the small bowel mesentery. Again, it was not possible from a radiological perspective to firmly diagnose either metastatic or non-malignant disease and there was concern regarding CT guided biopsy through an infected abdominal wall.

Figure 1 Computed tomography prior to neoadjuvant therapy

Figure 2 Computed tomography for restaging following long course chemoradiotherapy

Figure 3 Computed tomography two months after Figure 2

Without any less invasive means of gaining a definitive diagnosis, a decision was made to perform a laparotomy with a view to proceeding to an anterior resection if this proved to be possible. At operation, the pelvis was ‘frozen’ with what appeared to be an advanced tumour that was partially obstructing the ileum. There also seemed to be disseminated serosal/peritoneal deposits of tumour. Given the extent of the disease and that there were no dissectable tissue planes in the pelvis, an ileotransverse colon bypass was performed and biopsies were taken.

Histology showed no neoplasm, however, but rather fibrosis, inflammation and *Actinomyces*-like organisms. What had appeared to be disseminated malignancy was likely to be abdominopelvic actinomycosis. The patient’s IUCD was removed as the likely source of infection and she was started on a six-month course of penicillin. Unfortunately, owing to both the nature of actinomycosis and issues with compliance, she continued to have active infection with recurrent relapse of abdominal wall abscesses. Her malignant disease eventually progressed: she developed bilateral ureteric obstruction and distant metastases, and died just over two years after her original cancer diagnosis.

Discussion

Actinomycosis is a chronic granulomatous disease caused by an anaerobic, Gram-positive organism normally found in the mouth, gut and genitourinary tract. It is thought to cause infection in instances where there is a break in the mucosal continuity. It is a particularly difficult organism to detect because of the indolent nature of the infection. Sulphur granules, either micro or macroscopic, are strongly suggestive but not pathognomic of actinomycosis infection.¹ Other common findings include anaemia, leucocytosis and

elevated inflammatory markers (erythrocyte sedimentation rate and C-reactive protein).^{2,5}

Actinomycosis is not considered to be opportunistic in the sense that it does not appear to have a predilection for those who are immunocompromised (eg those with human immunodeficiency virus, diabetes and some malignancies) although it has been described in such patients.² It is generally considered to be an infection of low virulence and to affect the immunocompetent.¹

The association of *Actinomyces* species with IUCDs has long been established.^{3,4} Although there is some debate regarding whether or not they should be removed in the setting of oncological treatment for pelvic cancers,⁵ there is no evidence that they cause harm in this setting.

Manifestations of abdominopelvic actinomycosis vary widely. Differential diagnoses include Crohn’s disease, diverticular disease, malignancy (including of the rectum) and isolated cases of abdominal wall infection. The difficulties in diagnosis often lead to extensive and unnecessary de-bulking operations under the presumption that the pathology is malignant and not infectious.

It is generally suggested that patients with actinomycosis infection be treated with high dose, long course antibiotics to prevent relapse. Long-term therapy is recommended because of the granulomatous, filamentous nature of the infection and the resultant poor penetration of treatment. It is accepted that extensive surgery should be withheld for the rare cases when the abnormal tissue is unresponsive to antibiotic treatment.

Although there are multiple cases reported of abdominopelvic actinomycosis mimicking malignancy, our case appears to be unique in that the infection coexisted with rectal cancer, developing during its treatment and leading to irresectability. We do not know whether the patient’s rectum may have become resectable had her compliance with

antibiotic therapy been better. Indeed, it is possible that progressive actinomycosis as well as advancing malignancy played a significant role in her demise.

Conclusions

The learning points from this case are that conventional imaging and findings at laparotomy struggled to differentiate carcinoma from actinomycotic lesions. The role of chemoradiotherapy in the development of actinomycosis is less apparent although it may be prudent to suggest removal of an IUCD prior to embarking on neoadjuvant therapy for rectal cancer. What seems clear is that this is a rare but potentially fatal combination of disease and a hitherto undescribed cause of unresectable rectal cancer.

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