CASE REPORT

Actinomycosis presenting as an anterior abdominal mass after laparoscopic cholecystectomy

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Accepted 9 May 2017

SUMMARY

Abdominal actinomycosis is a rare disease caused by different anaerobic *Actinomyces* species. We report the case of a 55-year-old woman who presented with a painless, slow growing, left upper abdominal mass that developed after a laparoscopic cholecystectomy. A CT scan and MRI of the abdomen revealed a desmoid tumour of the left rectus abdominis muscle. Surgical excision was performed with an uneventful postoperative course. The histological analysis of the specimen was inconsistent with a desmoid tumour and revealed an infection of *Actinomyces israelii* in the anterior abdominal wall that was confirmed with a microbiology culture. The surgical treatment was followed by a course of penicillin antibiotic therapy for 6 months. This treatment resulted in full recovery with no further complications. Although it is rare, the patient's history of laparoscopic cholecystectomy was identified as the likely source of infection.

BACKGROUND

This case was an unusual presentation of abdominal wall actinomycosis (AMC) masquerading as a soft tissue tumour of the abdominal wall.

Although abdominal AMC is a rare, chronic, granulomatous infection, it should always be considered in the differential diagnosis for patients presenting with an abdominal mass who have a history of surgical or invasive procedures.

Diagnosis of abdominal AMC is often difficult: the diagnosis is made preoperatively in less than 10% of cases. Radiological imaging is often suggestive of neoplasm. Definitive diagnosis is based on histochemical and microscopic examination of tissue specimens.

Abdominal AMC is an extremely rare infection to occur after cholecystectomy. At present, only eight cases have been reported in the existing literature.^{1–7} This case is the first case to be reported as an abdominal wall mass after laparoscopic cholecystectomy related to bile spillage, without identification of other previous predisposing risk factors.

CASE PRESENTATION

A 55-year-old woman presented to the general surgery department with a painless, slow-growing abdominal mass in the left hypochondrium. She also had intermittent mild abdominal pain, anorexia and fatigue. Repeat history revealed that this swelling had developed after a laparoscopic cholecystectomy operation in another hospital

1 year ago. This operation was planned electively after the patient was diagnosed with symptomatic cholelithiasis. Postoperatively, she had bile leak and was managed conservatively with antibiotics for 7 days (Augmentin and metronidazole).

The patient's medical history included well-regulated, type 2 diabetes mellitus. There was no history of intrauterine contraceptive devices (IUCDs) use or previous bowel perforation. She was a non-smoker, married and had three sons. Her family history was unremarkable and showed no malignancy.

On examination, the patient was generally looking well, with a palpable mass in her left hypochondriac area. This mass was well defined, with an irregular surface, and approximately 3.5×4×5 cm in size. Located near to this swelling was a small scar from the previous laparoscopic operation (figure 1).

During this period, the patient was investigated for possible soft tissue tumours of the abdominal wall as an outpatient.

INVESTIGATIONS

Laboratory investigations showed raised inflammatory markers (C reactive protein 110 mg/L and erythrocyte sedimentation rate (ESR) 40 mm/hour).

A CT scan of the abdomen and pelvis revealed that the left rectus abdominis muscle was swollen and contained a well-defined, irregular soft tissue mass lesion, measuring approximately 3.7×5×6 cm in size. This lesion had an intra-abdominal extension indenting the underlying small bowel loops with loss of fat planes at some points in between and surrounding fat stranding (figure 2A,B). This discovery of a left rectus abdominis muscle soft tissue mass lesion was suggestive of a desmoid tumour or rectus sheath haematoma.

MRI of the abdomen was also performed and showed irregular enhancement of the left rectus sheath muscle, extending deep to include the adjacent part of the omentum and measuring approximately 12×10×6 cm in maximum dimension (figure 3). This appearance was in-keeping with a desmoid tumour of the left rectus sheath with direct invasion of the omentum rather than a rectus muscle haematoma.

DIFFERENTIAL DIAGNOSIS

At this stage, based on the patient's medical history and radiological investigations, the most likely diagnosis was a desmoid tumour of the left rectus abdominis muscle, with the other possibility being a left rectus muscle haematoma.

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To cite: Alhumoud Z, Salem A. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-220357

Figure 1 Well-defined abdominal mass, with an irregular surface, and approximately 3.5×4×5 cm in size. Located near to this swelling was a small scar from the previous laparoscopic operation.

TREATMENT

Excision of the left anterior abdominal wall mass was conducted with help from a plastic surgery team for closure of the wound. The gross appearance of the mass was suggestive of a desmoid tumour of the anterior abdominal wall. Histological examination revealed the appearance of an exuberant inflammatory reaction

Figure 2 (A,B) CT scan of abdomen (A, coronal section; B, sagittal section) showing the left rectus abdominis muscle containing a well-defined irregular soft tissue mass lesion measuring about 3.7×5×6 cm in size intra-abdominal extension indenting the underlying small bowel loops.

Figure 3 MRI of abdomen. showing irregular enhancement of the left rectus sheath muscle, extending deep to involve the adjacent part of the omentum and measuring about 12×10×6 cm.

with abscess and extensive fibrous scarring of surrounding tissue. There were colonies of filamentous Gram-positive bacteria, forming sulfur granules, surrounded by eosinophilic material (Splendore-Hoeppli phenomenon). These findings were consistent with abdominal wall AMC with secondary abscess formation (figure 4).⁸

Once the diagnosis was confirmed by a pathologist, the patient was started on intravenous penicillin G for 2 weeks. Treatment with Augmentin—1 g, two times a day—was continued, with a planned treatment timeline of 6 months.

A review of the patient's clinical history revealed that swelling started after a laparoscopic cholecystectomy performed 1 year prior, which confirmed the diagnosis of secondary abdominal wall AMC as a rare complication of previous laparoscopic surgery.

OUTCOME AND FOLLOW-UP

During follow-up—3 and 6 months after discharge from the hospital—the patient had no more complaints. The treatment

Figure 4 Characteristic of actinomycosis. Colonies of actinomyces (sulfur granules) surrounded by purulent exudate (H&E).

with surgical intervention and antibiotics resulted in full recovery with no further complications. Blood tests showed normalisation of ESR and C reactive proteins.

After 6 months of therapy with Augmentin—1 g, two times a day—therapy was discontinued.

DISCUSSION

AMC is a subacute to chronic bacterial infection characterised by contiguous spread, suppurative and granulomatous inflammation. This clinical infection is caused by *Actinomyces israelii*, which was discovered in 1878. It is an anaerobic, Gram-positive, filamentous bacterium that was previously misdiagnosed as fungus.⁹

A. israelii colonise the respiratory, gastrointestinal and genitourinary tracts. The bacterium has low virulence potential and only causes disease when the normal mucosal barrier is broken. It leads to multiple abscess formation, fistula or mass lesions.¹⁰

The most common infection site is the cervicofacial area, which accounts for 40%–50% of cases. Fifteen per cent of AMC occurs in the thorax, and 20% occurs in the abdomen and pelvis.¹⁰ Pelvic and abdominal AMC is increasing in frequency.¹¹ The most common sites of abdominal AMC are the appendix and the ileocaecal region. The intestines, stomach, liver, pancreas, anorectal region, pelvis or abdominal wall may also be involved. A patient with abdominal AMC usually presents with non-specific symptoms including abdominal pain, nausea, vomiting, fever, weight loss and palpable mass.¹⁰ This clinical presentation increases the consideration of malignancy.

Predisposing factors for abdominal AMC include recent abdominal surgeries or trauma, endoscopic procedures, the presence of intra-abdominal malignancy and hollow viscous perforation. In women of childbearing age, prolonged use of IUCDs has been suggested as the primary colonisation site of AMC in pelvic involvement.¹² Any age group can be affected by AMC, but it is rare at ages younger than 10 years.¹⁰ Male patients are affected more commonly than female patients, with a reported ratio of 3:1. The reported annual incidence of AMC is 1/300 000.¹⁰

Abdominal AMC is an extremely rare infection after cholecystectomy procedures, and only eight cases have been reported in the literature to date.^{1–7} Isolated involvement of the abdominal wall after cholecystectomy surgery was reported in one case. However, prolonged use of IUCDs was reported in this case, which has led them to be associated with colonisation of the abdomen with actinomyces species in addition to spilled gallstones. To our knowledge, our case is the first case to be reported as an abdominal wall mass after laparoscopic cholecystectomy related to bile spillage without identification of other previous predisposing risk factors.

Preoperative diagnosis of AMC is difficult to perform and has been reported in only 10% of all cases. Laboratory and radiological investigations are inadequate to differentiate between abdominal AMC and other inflammatory or neoplastic process.¹² However, CT scans can show the site and content of the lesions, in addition to their relation to adjacent tissue before any procedure.

Surgical intervention is required for a diagnostic biopsy or total excision. Definitive diagnosis is based on microscopic examination and histopathology of the surgical specimens, which demonstrate the characteristic sulfur granules and filament aggregates in 50% of cases. However, sulfur granules are not pathognomonic for this disease as other organisms such as nocardia and streptomyces may aggregate the granules resembling the actinomyces infections.^{10 11}

Histopathology is not the only reliable way to diagnose AMC. Microbiology cultures can be positive in about 50% of cases. However, the anaerobic characteristic and slow growth of these actinomyces species mean the samples will take up to 2 weeks to develop.¹¹

Following the confirmation of diagnosis of abdominal AMC, a systemic application of a high dose of intravenous penicillin (penicillin G 10–20 units daily) is the initial therapy of choice. In case of penicillin allergy, tetracycline, erythromycin and clindamycin are acceptable alternatives. After intravenous treatment for 4–6 weeks, oral penicillin (2–4 g/day) or amoxicillin should be administered for 6–12 months.^{10–12}

Abdominal AMC represents a diagnostic challenge for every physician, considering the clinical picture is highly non-specific and laboratory investigations are only indicative of an inflammatory process. However, actinomyces should always be part of the differential diagnosis of patients who present with previous abdominal surgery or trauma, the presence of intra-abdominal malignancy, viscous perforation and prolonged use of IUCDs. A CT scan of the affected area is advisable and the final diagnosis could be determined by the combination of histopathology of the surgical specimen and actinomyces cultures.

In 90% of all cases, the combination of surgical intervention and antibiotics results in complete recovery.

Learning points

- ▶ Abdominal actinomycosis (AMC) should always be considered as part of the differential diagnosis of patients presenting with an abdominal mass who have a history of surgical or invasive procedures.
- ▶ Radiological investigations can identify the mass from abdominal AMC, but they cannot differentiate between the infection and malignancy.
- ▶ Abdominal AMC can occur secondary to spilled gallstones after laparoscopic cholecystectomy.

Contributors ZA was involved in case scenario, literature review and manuscript preparation; AS was involved in management of the case, informed consent and final revision.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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