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*Case Report*

# Abdomino-pelvic actinomycosis of urachal remnant mimicking lower abdominal tumour

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**Abdominal-pelvic actinomycosis constitutes three percent (3%) of all human actinomycotic infections. It is usually insidious, and is one of the great imitators in clinical practice, particularly when it occurs in abdominal cavity and one is struck by the frequency with which actinomycotic lesions are diagnosed to be diverticulitis, abscesses, inflammatory bowel disease or even a neoplasm. We report a case of urachal actinomycosis that presented with a progressively enlarging infraumbilical mass associated with infraumbilical discharge. Ultrasound and CT scan examinations of the abdomen and pelvis revealed an heterogenous mass extending through and through intraperitoneal to extraperitoneal planes with bowel loops adherent to this mass on its intra-peritoneal side. Gas filled spaces were also apparent within the mass. A provisional differential diagnosis of an inflammatory bowel disease, tumour of abdominal wall, chronic infections such as tuberculosis or actinomycosis was made. All the involved bowels with partial cystectomy and involved abdominal wall were resected. The pathologic examination revealed actinomycosis. The patient was treated with penicillin and no recurrence was noted on postoperative follow-up. The case is therefore presented to raise the awareness of this rare condition, to be included in the differential diagnosis of an abdominal or abdomino-pelvic mass.**

**Keywords:** Actinomycosis, Abdomen, Pelvis, Tumour.

## INTRODUCTION

Actinomycosis is a subacute-to-chronic bacterial infection caused by filamentous, gram-positive, non–acid-fast, anaerobic-to-microaerophilic bacteria called Actinomyces israelli.

Actinomyces israelii are normal inhabitants in the oral cavity and upper intestinal tract of humans. The most common clinical forms of actinomycosis are cervicofacial followed by abdominal actinomycosis; (Cintron et al., 1996) but it has been found recently in the female genital tract.

It is characterized by contiguous spread, suppurative and granulomatous inflammation, and formation of multiple abscesses and sinus tracts that may discharge sulfur granules (Sumer et al., 2004).

Risk factors for abdomino-pelvic actinomycosis include history of recent or remote bowel surgery such as perforated acute appendicitis (Erdal et al., 2008), perforated colonic diverticulitis penetrating trauma to the abdomen or ingestion of foreign bodies like chicken or fish bones, patent urachal remnants, use of intrauterine

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contraceptive devices (IUCD) (Arend et al., 1998; Soria-Aledo et al., 2004; O'Connor et al., 1989) in females during which actinomycetes are introduced into the deep tissues.

In the case of IUCD use, pelvic actinomycosis is postulated most commonly to ascend from the uterus and the IUCD must have been in place for an average length of about eight (8) years or more.

Involvement of any abdominal organ, including the abdominal wall occurs by direct spread, with eventual formation of draining sinuses. The involvement of the urinary tract is rare and primary actinomycosis of urinary bladder is still rarer (Kawahara et al., 1998; Matsumura et al., 2003; Dhamborvorn et al., 2001; Bianchini et al., 2012).

Actinomycosis is considered the most misdiagnosed disease (Gning et al., 2011; Thompson et al., 1982; Pusiol et al., 2011) and the rarity of the disease in general and its non-specific clinical, biochemical, and radiological features are the causes for the frequently mistaking the condition as neoplasm.

The diagnosis remains difficult clinically unless there are multiple right lower abdominal sinuses.

They have a variety of clinical manifestations and can even mimic a malignancy (Simsek et al., 2011) and often a surgical intervention with resection is needed before a definitive diagnosis is made.

Delay in diagnosis makes the course worse and leads to abdominal septic complications.

We report this case of a 35 year old female with an abdomino-pelvic pseudo-tumoral fistulous mass and irritative lower urinary symptoms for about two months (2) associated with loss of appetite and weight.

The possible differential diagnosis included a chronic inflammatory mass, tumour of abdominal wall, chronic infections such as tuberculosis or actinomycosis. The disease was eventually diagnosed histologically from the resected specimen after exploratory laparotomy.

We therefore recommend that this disease entity should be included in the differential diagnosis of any lower abdomino-pelvic tumorous mass with or without fistulous presentations and better still with history of some of the risk factors associated with abdominal actinomycosis enumerated above.

## CASE REPORT

A 35 year old female was referred from the Urologist with two- month (2) history of lower abdominal pain, deep yellow to orange urine associated with frequency and burning.

There was also loss of appetite and weight.

In her past medical history was the fact that she had caesarean section carried out seven (7) years earlier.

Clinical examination revealed a slightly ill-looking anicteric lady; temperature 37.8$^{o}$C; BP 110/80mmHg; Pulse 78/min; respiratory rate (RR) 30/min.

Abdominal examination revealed an angry looking tender oedematous, lymphangitic palpable well circumscribed mass about 12 by 15 cm over the supra-pubic region and through its center was an old transverse healed caesarean section scar carried out seven years earlier.

The mass had limited movement along its transverse axis but none on its longitudinal axis.

The whole area of the mass was red with features of an abscess that was about to rupture.

Routine complete blood count revealed hematocrit value was 37%; WBC count was 12,200 /mm3 and ESR of 84 mm/h. Biochemical investigation was unremarkable

Both ultrasound and CT scan examinations of the abdomen and pelvis revealed heterogenous mass extending through and through intraperitoneal to extraperitoneal planes, with bowel loops adherent to this mass on its intra-peritoneal side. Gas filled spaces were apparent within the mass (see figures 1 and 2 below).

A provisional differential diagnosis of an inflammatory bowel disease, tumour of abdominal wall, chronic infections such as tuberculosis or actinomycosis was made.

Patient was then transferred from the urological service to general surgery and was placed on intravenous fluids, parenteral combination antibiotics, and metronidazole. Swab was taken for culture and sensitivity.

An exploratory laparotomy was later carried out which revealed an inflammatory firm mass at the anterior bladder wall connected to another mass at the anterior abdominal wall.

All the involved structures (caecum, appendix, loops of small bowel, sigmoid colon, and greater omentum) were carefully dissected and separated with success. Excision of the mass with the involved anterior abdominal wall plus partial cystectomy since the bladder dome was involved were carried out. (see figure 3 below).

The resected specimen was sent for histopathology and the result came back as actinomycosis (see figure 4 below).

It must be stressed that the actinomycotic infection had sinus and abscess formation right from the dome of the bladder to the skin outside.

Abdomen was then closed after adequate haemostasis; a haemovac drain was left in-situ and an indwelling Foley's catheter left in the urinary bladder for about two (2) weeks.

The patient was subsequently treated post operatively with penicillin.

Therapy continued parenterally for about fourteen (14) days and she remained well and was later discharged home on oral penicillin therapy for the next two (2) months.

Post-operative period was uneventful. The initial swab taken pre-operatively was negative for any microbes.

Cystography was carried out post-operatively and

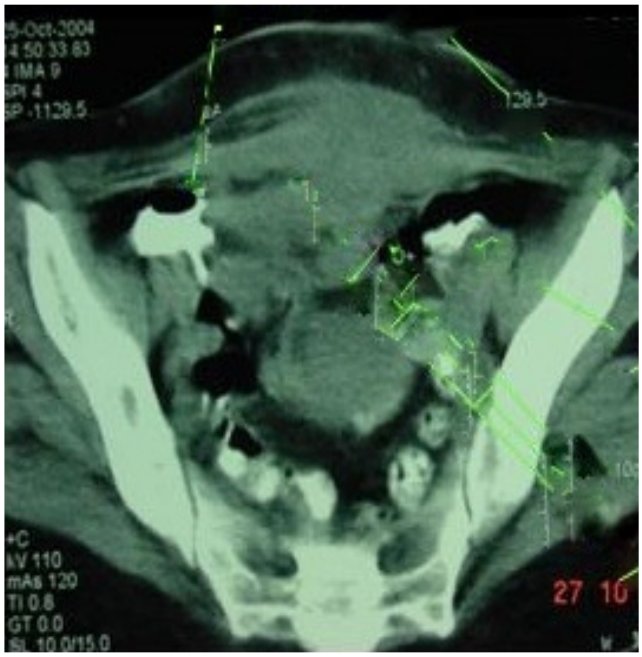

**Figure 1**

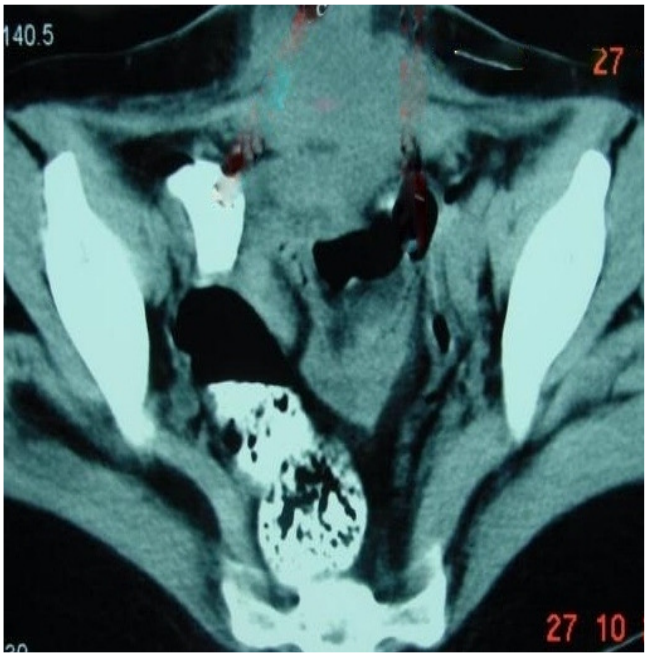

**Figure 2**

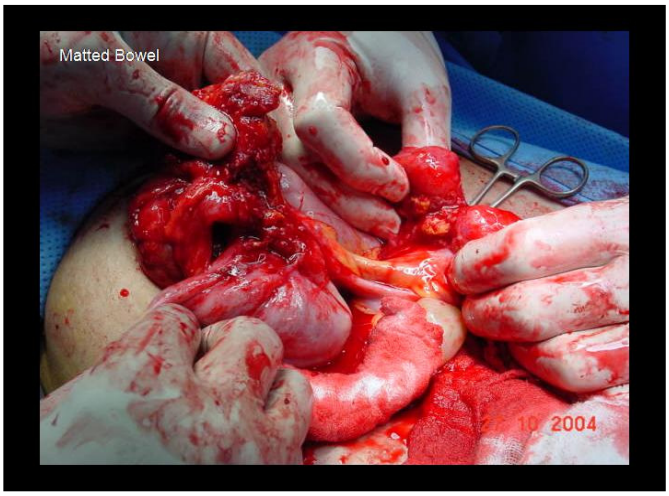


**Figure 3**

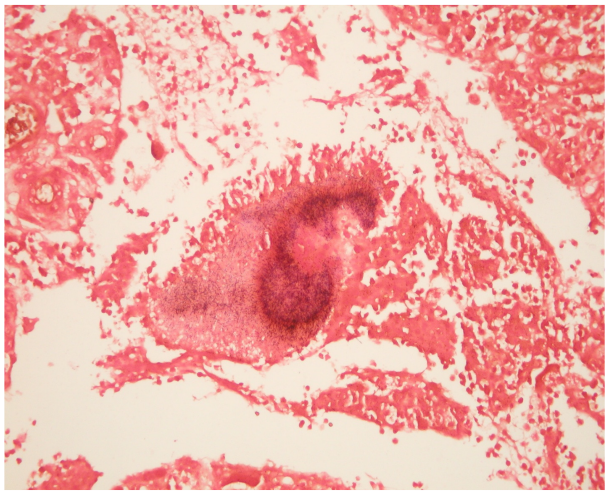

**Figure 4.** Actinomycosis gram stain

found to be clear.

Follow-up post operative ultrasound (U/S) and CT scan of abdomen done did not reveal any residual or recurrent lesion.

## DISCUSSION AND CONCLUSION

Between 1948 and 1998 Yeguez et al had investigated 505 cases (Yegüez et al., 2000) with abdominal actinomycosis, although the first identification of actinomycosis was made nearly a hundred years ago; however the factors that initiate the infection are still not well known.

Patients with immune deficiency, previous abdominal surgery, penetrating abdominal trauma, inserted IUCD for a period not less than eight years, (Arend et al., 1998; O'Connor et al., 1989) and oropharygeal surgery are predisposing conditions in the development of abdomino-pelvic actinomycosis.

The prolonged use of IUCDs associated to pelvic actinomycosis was not given in this case, however predisposing factors are not always present in many cases.

Non specific nature of presentation such as abdominal pain, occasional fever, anorexia, weight loss, change of bowel habit and abdominal mass can easily mislead clinicians to an incorrect diagnosis.

Abdominal actinomycosis accounts for 20% of all cases of causes of acute to chronic lower abdominal pain. Since malignancy and acute abdomen present similar clinical pictures (Stringer and Cameron, 1987; Alam et al., 2001; Tamer et al., 2006; Cirafici et al., 2002; Wagenlehner et al., 2003; Sung et al., 2011) to abdominal actinomycosis, usually a pre-operative definitive diagnosis is not made in most cases. Accurate diagnosis is largely made with an examination of the specimen removed during surgery.

Actinomycosis is an indolent and slowly progressive chronic granulomatous infection. It is characterized by the development of indurated swellings, mainly in the connective tissue, suppuration and discharge of sulphur granules.

Actinomyces species exist in the normal flora of the gastrointestinal system and female genitourinary tracts, and rarely the disease involves the genitorurinary system, usually by the haematogenous route from a primary site of infection. Primary actinomycosis in urachal remnant is documented rarely in the English literature; and now we are reporting another case of this rare disease.

The most common etiologic organism associated clinically with actinomycosis is *Actinomyces* Israeli (Wagenlehner et al., 2003). However other species like Actinomyces gerencseriae, Actinomyces turicensis, Actinomyces radingae, Actinomyces europaeus could also be responsible.

Almost all actinomycotic lesions contain so-called companion bacteria the most important of these bacteria include actinobacillus actinomycet emcomitans, peptostreptococcus, bacteroides, staphylococcus, and streptococcus species, depending on the location of actinomycotic lesions. These companion bacteria appear to magnify the low pathogenic potential of actinomycetes but none were present in our case after we received the culture report.

A patent urachus may also increase a person's susceptibility to urinary tract infections in some cases. Since Actinomyces species have low virulence, they cause disease only when the normal mucosal barrier is broken leading to abscess formation, fistula or mass lesions.

There are no specific radiological signs of gastrointestinal actinomycosis. Ultrasonography, Computerized abdominal scan (CT), magnetic resonance imaging (MRI) and angiography may fail to distinguish between actinomycosis and carcinoma (Lee et al., 2001; Böhm et al., 2006; Brook, 2008).

CT scanning irrespective of the anatomic area of involvement usually reveals an infiltrative mass with focal areas of decreased attenuation that enhance with contrast. This infiltrative mass tends to invade surrounding tissues.

However, when patients present with superficial lesions with pus discharging sinuses that contains sulfur granules or accessible to biopsy, diagnosis can be established pre-operatively.

In a suspected mass, a preoperative fine CT scan or ultrasound-guided fine-needle aspiration and/or biopsy and anaerobic culture can establish the diagnosis. This was not attempted in our case because of the risk of bowel perforation and possible development of peritonitis.

Increased awareness of this entity will help in raising the index of suspicion and possibility of preoperative diagnosis in suitable cases.

In most cases of actinomycosis, antimicrobial therapy is the only treatment required, although surgery can be adjunctive in selected cases. Penicillin G is the drug of choice for treating infections caused by actinomycetes.

With the precise diagnosis established, and no penicillin allergy exists; the antibiotic regimen will be penicillin G, 10 million IU/day parenterally for two (2) weeks, followed by oral therapy of 2 g/day for two (2) months (Atad et al., 1999). It has a high capability of penetrating abscesses, sinuses and dense fibrotic structures.

Other antibiotics that could be used include tetracycline, erythromycin, doxycycline, clindamycin, imipenem, ceftriaxone and ciprofloxacin; they are all suitable alternatives if there is penicillin allergy.

Attempt to cure actinomycosis, especially those with extensive disease, involves antimicrobial therapy combined with surgical treatment such as incision and drainage of abscesses, excision of sinus tracts and recalcitrant fibrotic lesions, decompression of closed-space infections, and interventions aimed at relieving obstructions provided a pre-operative diagnosis is made (Hamid et al., 2000; Michihiro et al., 2010).

It is important to be aware of this uncommon, yet significant, presentation of a common infectious disease which is curable with antibiotics in order to avoid misdiagnosis and over treatment as a case of malignancy (Wong et al., 2011; Ferrari et al., 2000; Al-Kadhi et al., 2007).

Actinomycosis is a fully curable disease with good prognosis. A prolonged course of antibiotics is usually required for cure.

In conclusion, the diagnosis of abdominal actinomycosis is difficult due to its rarity and specific presentation.

It should however be considered in the differential diagnosis of an abdomino-pelvic mass with or without fistulous presentations and better still in patients with history of some of the risk factors earlier enumerated.

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