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Case Report

Pathology Section

Case of Ray Fungus Presenting with Abdominal Distension

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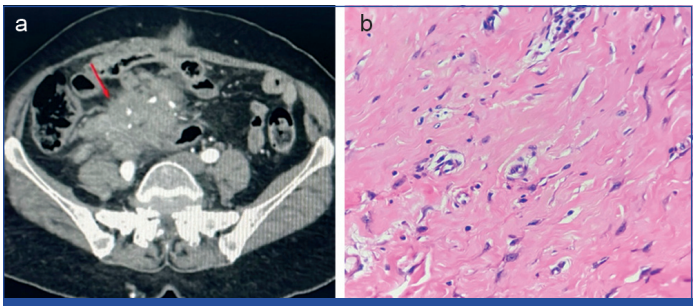
ABSTRACT

Actinomycosis is a rare infection caused by filamentous, non acid fast staining and gram positive bacteria. The diagnosis is rarely made preoperatively, and only histopathological examination can confirm it, as the condition presents with varied clinical manifestations, often resembling a malignant lesion on radiographic images. Hereby, the authors present a case of a 67-year-old female, who was admitted for evaluation of an abdominal mass. Radiologically, the mass resembled a desmoplastic fibroma, and clinically, it was suspected to be the same. The mass was excised and sent for histopathological examination. Grossly, two specimens were received. The first consisted of adherent bowel loops, including the ileum, appendix, caecum and descending colon, with the mass arising from mesentery involving the bowel, measuring 11.5x9.5x6 cm. The second specimen was a partial cystectomy specimen, with the abdominal wall mass infiltrating the bladder anteroposteriorly, measuring 11x10x8 cm. Microscopic examination revealed extensive areas of inflammation with actinomycotic organisms exhibiting the Splendore-Hoeppli phenomenon, along with numerous multinucleated giant cells and acute on chronic inflammatory infiltrate. Surrounding areas showed dense collagen bundles. The prognosis after complete surgical resection is good, along with the administration of broad spectrum intravenous antibiotics.

Keywords: Abdomen, Broad spectrum antibiotics, Disseminated actinomycosis

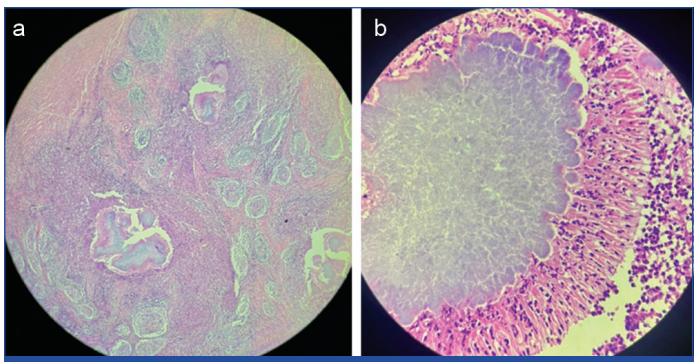
CASE REPORT

A 67-year-old female, known case of systemic hypertension for the past two years and on B blockers, presented with complaints of altered bowel habits with vague abdominal pain on and off for the past one month. The pain was presented as a vague discomfort in the whole abdomen radiating to other sites. Physical examination was apparently normal, with mild abdominal distension and no tenderness. Laboratory investigations revealed an elevated total leucocyte count of 17830 cells/microliter, predominantly neutrophils, ruling out the possibility of an abscess or infection. Patient has a history of angioplasty performed one year ago and a hysterectomy performed ten years ago. Radiological imaging studies were advised as it was difficult to establish a provisional diagnosis based on the history and clinical findings. Imaging studies suggested a soft tissue attenuating lesion in the mesentery and right rectus abdominis muscle, favouring desmoplastic fibroma [Table/Fig-1a]. Clinically, the lesion was thought to be a desmoid tumour. An attempted core needle biopsy of the mass showed predominantly areas of fibrosis on Hematoxylin and Eosin (H&E) [Table/Fig-1b].



[Table/Fig-1]: a) Ill-defined irregular soft-tissue lesion seen in the midline to the mesentery towards right; b) Core biopsy showing only areas of fibrosis (H&E; 200X).

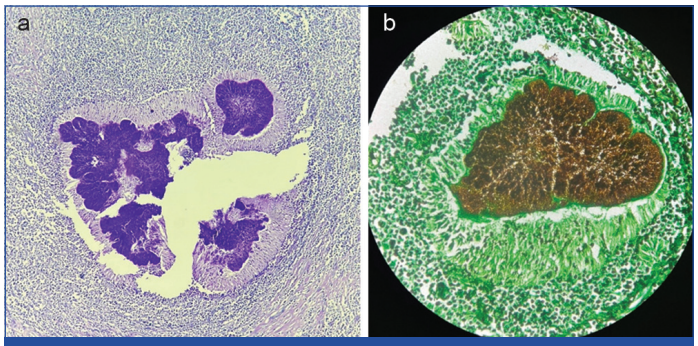
Since, a definite diagnosis could not be made, complete excision of the lesion was advised. An exploratory laparotomy was performed, and the tumour was completely resected and sent for histopathological examination. Grossly, two specimens were received: one was the mass arising from the mesentery involving the bowel, and the other was the abdominal wall mass infiltrating the bladder. Adherent bowel loops composed of the ileum, appendix, ascending colon, and caecum with an infiltrative ill-defined lesion involving the root of the mesentery were seen, measuring 11.5×9.5×6 cm. The lesion appeared grey-white and nodular with focal grey-brown areas. The appendix showed the presence of a faecolith. The serosal aspect of the ileum showed multiple grey-red to grey-black irregular nodules, with the largest nodule measuring 3×2.5×1.1 cm. The mucosal aspect of the small intestine was predominantly unremarkable, with focal grey-black areas. The partial cystectomy specimen with the abdominal wall tumour measured 11×10×8 cm, and the anteroposterior aspect showed an ill-defined infiltrative lesion measuring 9.5×7×6 cm. It was inferiorly attached to the rectus abdominis muscle. On serial sectioning of the bladder and abdominal mass, a few ill-defined grey-brown areas measuring 2×1.5×1 cm were noted. The rest of the bladder mucosa appeared unremarkable. Microscopy revealed extensive areas of inflammation, actinomycotic species showing splendore-hoeppli phenomenon with multinucleated giant cells, and dense acute on chronic inflammatory granulation tissue [Table/Fig-2]. Also seen were thick-walled blood vessels and lymphoid follicles with germinal center formation. The small intestinal mucosa mainly showed areas of haemorrhage. The



[Table/Fig-2]: a) Section from lesion bit showing actinomycotic species and lymphoid follicles with germinal centers (H&E stain; 10X); b) Actinomycotic species showing splendore-hoeppli phenomenon (H&E stain; 10X).

appendix showed features of acute appendicitis. The proximal and distal margins that were submitted were free of tumour.

The bladder mucosa revealed areas of ulceration and extensive infiltration by the actinomycotic species, admixed with numerous foreign body-type giant cells and haemosiderin-laden macrophages. The surrounding areas showed dense collagen bundles with areas of chronic inflammation. Special stains such as Periodic acid-Schiff (PAS) stain and Grocott–Gömöri's Methenamine Silver (GMS) stain were performed, highlighting the presence of actinomycosis [Table/Fig-3]. With the above findings, a diagnosis of disseminated abdominal actinomycosis was confirmed. The patient was then started on intravenous ampicillin for six weeks and was subsequently discharged with close follow-up. Although the patient initially showed signs of improvement, later developed intolerance to the antibiotic regimen and developed diarrhoea, which was managed. On follow-up after three months, the patient was hospitalised due to dyselectrolytemia and severe vitamin D deficiency.



[Table/Fig-3]: a) Positive Periodic acid-Schiff (PAS) stain for actinomycotic species (10X); b) Positive Grocott–Gömöri's Methenamine Silver (GMS) stain for actinomycotic species (40X).

DISCUSSION

Actinomycosis is a rare infection, constituting approximately 15.9% of cases [1]. The organism responsible is a gram-positive, filamentous, non acid-fast, anaerobic bacterium [1]. Actinomyces israelii is the most common type affecting humans. The infection typically occurs in middle-aged individuals and is more common in males [2]. It is also more prevalent among low socio economic groups. Due to its variable clinical presentation, the diagnosis of actinomycosis is rarely made before surgery and is often not confirmed until the chronic phase. A detailed clinical history plays a vital role in aiding the diagnosis. Isolating the organism in culture is a time consuming process that can yield false negative results. Actinomyces is usually present as a normal commensal in the oral cavity and large intestine.

It is divided into three clinical forms: cervicofacial, thoracic and abdominopelvic types. The abdominopelvic type usually develops after a localised inflammatory process or, more commonly in women, with Intrauterine Devices (IUD) usage. Infection of the oral cavity is due to poor oral hygiene. Thoracic infections may occur in patients with an alcohol disorder, and pulmonary infections can occur as a complication of aspiration. They acquire pathogenicity by invading with the help of companion bacteria through breached or necrotic tissue [3,4].

It spreads by direct extension across tissue planes, resulting in the formation of abundant granulation tissue, abscesses and sinuses. It may involve multiple sulphur containing abscesses. These sulphur granules are responsible for intense fibrosis of the tissues. However, detecting sulphur granules of the actinomyces organism is very difficult, with only 26% of cultures being positive [5]. Haematogenous spread to organs like the liver and lungs is documented, which can produce nodules that, on radiographic images, can be mistaken for malignancy [6].

The appendix, colon and caecum are most affected by actinomyces infection [7]. The presentation is similar to irritable bowel disease, tuberculosis and malignancy. When the surrounding structures are involved, it contributes to the insidious onset and delays in diagnosis, mimicking a tumour. Serological tests do not help in making the diagnosis. Imaging findings are non specific for the infections but rather help in assessing the degree of soft tissue involvement. The infection is treatable with intravenous antibiotics, with surgery being used as an adjunctive treatment. The duration of therapy is usually 6-12 months, but can be shortened if surgical resection is done, depending on the type of resection [8]. The curative rate is currently very high due to the availability of a wide range of antibiotics and the absence of resistance by actinomycotic species to these antibiotics. Hence, recurrence is very rare. The outcomes are poorer for immunocompromised individuals. Leaving the infection untreated is life-threatening for the patient, leading to systemic spread and sepsis. The response to treatment can be monitored with imaging [9-11]. Therefore, a well-integrated team approach is essential to provide optimal care to patients infected with actinomycosis.

The tabulated cases show various sites affected in the abdomen by actinomycosis, which also revealed a good response to surgical resection, since, it is usually mass forming, followed by intravenous long-term, broad spectrum antibiotics and close follow-up of the patients [Table/Fig-4] [12-15].

Name of the author and year of the study	Age (years)	Sex	Affected site	Treatment	Outcome
García-Zúñiga B and Jiménez-Pastrana MT, 2016 [12]	41	Male	Distal ileum	Resection and ileostomy by Hartmann's procedure, followed by intravenous penicillin.	Fully recovered. Follow-up done.
Eskarous H et al., 2021 [13]	88	Female	Sigmoid	Exploratory laparotomy with loop colostomy, followed by long term antimicrobial use.	Fully recovered.
Pamathy G et al., 2021 [14]	40	Female	Descending colon	Exploratory laparotomy with extended left hemicolectomy, followed by oral penicillin for 6-12 weeks.	Recovered and on follow-up.
Tarzi M et al., 2021 [15]	59	Male	Abdominal wall	Parenteral crystalline penicillin 24 million units/ day for 1 month, followed by oral penicillin V for 6 months.	Fully recovered.
Present case, 2023	67	Female	Abdomen	Given intravenous ampicillin.	Discharged with penicillin and is on follow-up.

[Table/Fig-4]: List of actinomycosis in abdomen sites from previous studies [12-15].

CONCLUSION(S)

The present case is presented due to the radiological assumption of a soft tissue tumour and clinical suspicion of the same, with a possibility of malignancy, as well. This emphasises the significance of histopathological evaluation of the lesion. The condition responds well to broad spectrum antibiotics, which is reassuring for the patient and treating physician. Light microscopy, along with fungal stains, helps in eliminating any confusion regarding the diagnosis of the condition.

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