Case Report

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Appendiceal actinomycosis mimicking malignant tumor: a rare case report

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Introduction: Actinomycosis is an uncommon bacterial infection caused by *Actinomyces* bacteria that typically progresses slowly and leads to the formation of masses. Although it commonly affects the cervicofacial area, about 20% of cases occur in the abdominopelvic region. Because the disease can be mistaken for a tumour due to its infiltrative mass-like nature on imaging, over 90% of cases are only diagnosed following surgery and histological confirmation. This report describes a case of an appendicular mass, initially suspected to be a malignant tumour, but eventually diagnosed as appendiceal actinomycosis.

Presentation of case: Upon initial presentation, a 53-year-old woman with type II diabetes mellitus and no prior surgical history, displayed abnormal appendiceal uptake during a PET-computed tomography (CT) scan conducted for a suspected spinal tumour. Colonoscopy did not indicate any notable observations, and the patient chose to defer immediate action. Several months later, a CT scan revealed an increased mass-like appearance of the appendix compared to the previous PET-CT scan. After multidisciplinary discussions, a right laparoscopic hemicolectomy was recommended due to suspected malignancy. However, histological staining on microscopy confirmed actinomycosis originating from the appendix.

Discussion: Chronic appendicitis with radiologic features similar to appendiceal carcinoma, or abdominal masses located in the ileocecal area, in patients with or without a previous surgical history should raise suspicion of actinomycosis.

Conclusion: Appendiceal actinomycosis should be considered in the differential diagnosis in the aetiology of chronic appendicitis mimicking appendiceal carcinoma. Awareness and accurate diagnosis of appendiceal actinomycosis can prevent unnecessary extended surgery as was performed in this case.

Keywords: Abdominal actinomycosis, actinomyces, actinomycosis, appendiceal mass, appendiceal neoplasms, case report

Introduction

Actinomycosis is a rare, chronic, and progressive disease that is most frequently brought on by the anaerobic gram-positive bacterium *Actinomyces Israelii* with an estimated prevalence of about one case per 40 – 119 000[1–3]. It is frequently characterized by the development of many abscesses, fistulae, draining sinuses, extensive granulation, and inflammatory pseudotumors. The cervicofacial region accounts for the majority of cases, while 20% of infections also affect the abdominopelvic region and 15% of the thoracic regions[4]. Despite being typical commensals of the gastrointestinal, urogenital, and oral tracts, the *Actinomyces* species can develop harmful properties when necrotic tissue is present[5]. Various factors have been reported to predispose individuals to abdominal disease, including mucosal barrier injuries, abdominal surgery, bowel perforation, trauma, and immunosuppression[6–9].

There are a few cases reported suggesting unusual pathologic presentation of actinomycosis[6,7,10–12]. Since the clinical presentation of abdominal actinomycosis is of nonspecific symptoms and signs, that can mimic other diseases[13], less than 10% of appendiceal actinomycosis cases are diagnosed before surgery due to the need of histological confirmation of the specimen for definitive diagnosis[1,14].

HIGHLIGHTS

- This case report presents the first documented instance of appendiceal actinomycosis in a diabetic patient with no history of abdominal surgery, which mimicked appendiceal carcinoma.
- The patient was treated with a right hemicolectomy for suspicion of appendiceal carcinoma, which was later diagnosed following pathological analysis as appendiceal actinomycosis.
- Accurate diagnosis and awareness of appendiceal actinomycosis can help prevent unnecessary surgery and instead be treated with long-term antibiotic therapy.
- Chronic appendicitis, with radiologic features similar to appendiceal carcinoma, should raise suspicion of actinomycosis.

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Some reports of actinomycosis describe wall thickening and peri appendiceal inflammation, with contrast enhancement, that was misdiagnosed as colon cancer[15,16]. Here, we present the case of an appendicular mass suspected to be a malignant appendiceal tumour due to radiologic imaging, which was eventually diagnosed as appendiceal actinomycosis post-operatively. This case report holds significance as it illustrates the differential diagnosis of an appendiceal mass initially suspected as a malignant tumour. This finding, as described below, may prompt fellow surgeons faced with similar cases to opt for a conservative approach, such as appendectomy, and advance to subsequent steps only after obtaining final pathology reports.

This case report has been reported in line with the SCARE Criteria[17].

Case presentation

A 53-year-old female presented with high metabolic appendiceal uptake on PET-computed tomography (CT) with [F18]-2-fluoro2-deoxy-d-glucose (FDG) (Fig. 1), which was performed due to suspicion of a spinal tumour. The patient's medical history included type II diabetes mellitus and essential hypertension, which were being treated with medication at an outpatient clinic. Past social history (history of tobacco, alcohol, and drug use) were negative, and all review of systems, such as weight loss, weakness, and fever, were also negative. Additionally, the patient had no previous surgical history, and physical examination and laboratory results were normal. Colonoscopy revealed no remarkable findings. The patient opted to delay further action at the time.

Subsequent to the PET-CT scan conducted several months prior, a follow-up CT scan was administered. This decision was made to assess potential growth of the mass, its relation to the caecum, and the presence of any new lymphadenopathy. The CT demonstrated focal thickening of the appendix with a width of about 18 mm, and a relatively high density. A fine striped infiltration of fat and lymph nodes measuring about 8 mm in diameter near the appendix was also noted (Fig. 2). From these findings, an appendiceal tumour was highly suspected. Following a multidisciplinary discussion with the gastroenterology and general surgery departments, an elective laparoscopic right hemicolectomy was performed.

Surgical findings revealed an intraluminal growing small mass with no signs of macroscopic serosal invasion. Gross pathology revealed an ill-defined mass lesion originating from the appendix. Upon microscopy, the appendiceal wall was positive for *Actinomyces* on periodic acid-schiff (PAS) stain (Fig. 3A, B) and on hematoxylin and eosin (HE) stain (Fig. 3C), with no evidence of malignancy of the appendix. The ileum, colon, and pericolic lymph nodes, along with the pericolic fatty tissue, were all unremarkable.

One month long antibiotic regimen of intravenous Penicillin G treatment was decided as adjuvant therapy.

Discussion

Actinomycosis confined to the appendix represents a distinctive and uncommon manifestation of abdominopelvic actinomycosis, with the underlying pathogenesis not completely understood. In many cases, the presentation of the patient combined with the imaging studies can raise the possibility of appendicitis.

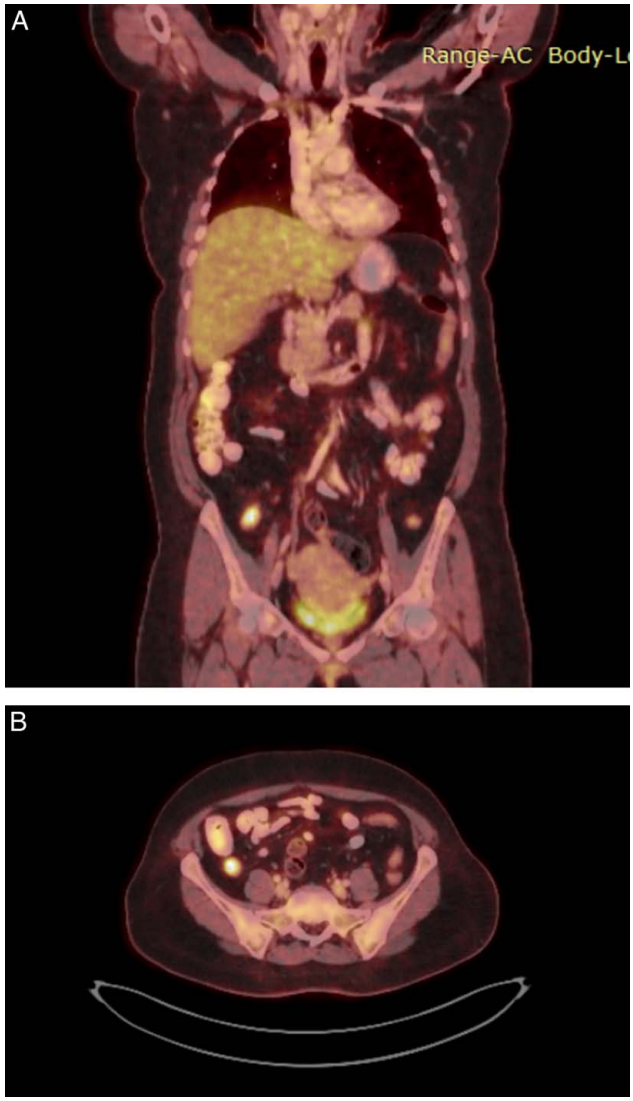

Figure 1. (A) High metabolic appendiceal uptake on coronal abdominal PET-computed tomography (CT) with [F18]-2-fluoro2-deoxy-d-glucose (FDG). (B) High metabolic appendiceal uptake on cross sectional abdominal PET-CT with FDG.

Consequently, patients commonly undergo appendectomy, and the definitive diagnosis is established through histopathological examination[13,18,19].

However, appendiceal actinomycosis mimicking a malignant tumour of the appendix is an even more uncommon incident, with a review of the literature describing one other case report[11]. In our case described above, the working diagnosis in this patient leaned towards a malignant appendiceal tumour. This inclination was rooted in the tumour-like appearance of the appendix, the heightened FDG uptake, and the concurrent enlargement of an adjacent lymph node, as observed in the CT scan. When comparing the similarities between the two diagnoses, appendiceal actinomycosis like malignant appendiceal carcinoma can also present with dense contract enhancement[1,5,15]. However, contrary to the literature, in which actinomycosis is generally not known to disseminate through the lymphatic system, our CT scan revealed signs of localized lymphadenopathy. This observation

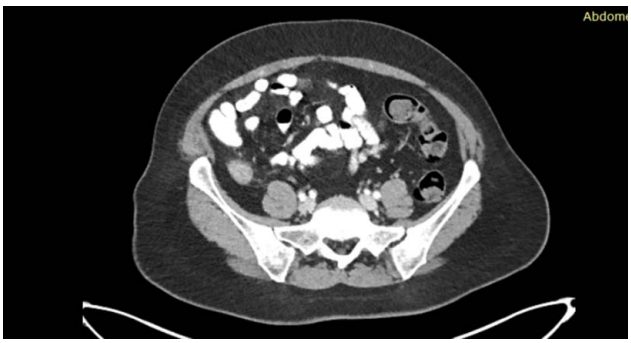

Figure 2. Follow-up cross sectional computed tomography of the abdomen showing fine striped infiltration of fat and lymph nodes measuring about 8 mm in diameter near the appendix.

led us to dismiss the possibility of actinomycosis and consider malignant appendiceal carcinoma as the more likely diagnosis[4,16]. This prompted our choice to proceed with an initial laparoscopic right hemicolectomy. However, retrospectively, it was feasible to circumvent a right hemicolectomy by conducting an appendectomy and awaiting the final histological results.

As we review this case collectively, we want to bring to attention the gravity of appendiceal actinomycosis to fellow clinicians. Although rare, the diagnosis should be added to the differential in patients presenting with or without abdominal symptoms, evidence of enhanced contrast on CT scan, and with or without regional lymphadenopathy. Furthermore, differentiating between the two diagnoses intraoperatively via histology can help prevent an incorrect diagnosis and the need for unnecessary surgeries.

Upon diagnosing a digestive tract infection with actinomycosis prior to surgery, extended antimicrobial treatment becomes imperative for favourable outcomes. Surgery is only warranted in complicated cases, such as in the development of fistulas in patients[1]. Nevertheless, to our knowledge, there are no established guidelines regarding administrating penicillin following removal of an affected organ with actinomycosis, as in the case of appendiceal actinomycosis. In our case, after a comprehensive multidisciplinary discussion, a decision was made to administer adjuvant antibiotic therapy as we could not completely rule out the potential for bacterial spillage during the surgery.

Conclusion

Appendiceal actinomycosis is a rare occurrence. It can manifest in various conditions: either in a subacute or chronic presentation, resembling appendicitis, or even as an incidental discovery, as seen in our case. While there are some typical, but not specific, characteristics that can allude to a diagnosis of actinomycosis, it notably exhibits increased FDG uptake when a PET-CT is performed. Although an initial knee-jerk reaction of an appendiceal mass with FDG uptake may represent a malignant tumour, it is crucial to bear in mind that alternative diagnoses, including actinomycosis, should be considered. We believe that if surgically feasible, prioritizing an appendectomy and awaiting final pathology results before deciding on further intervention would be preferable. Given that a definitive diagnosis is only confirmed postoperatively by pathological analysis, clinicians should be aware of this rare diagnosis in order to deliver appropriate treatment and mitigate the possibility of unnecessary extensive surgery.

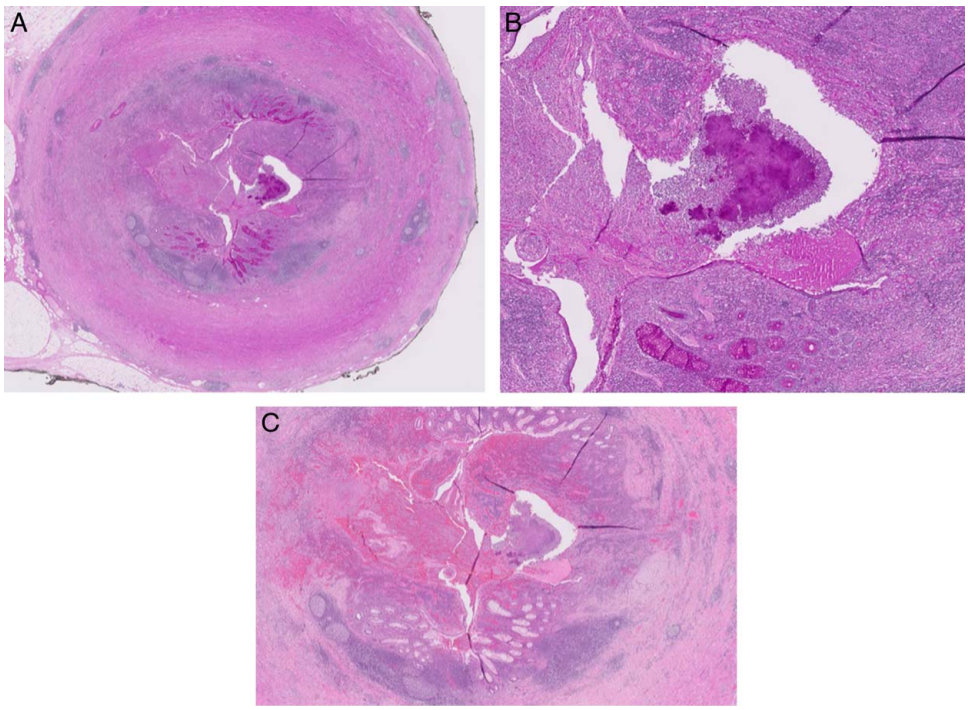

Figure 3. (A and B) Microscopic images of the appendiceal wall positive for *Actinomyces* on periodic acid-schiff stain. (C) Microscopic image of the appendiceal wall positive for *Actinomyces* on hematoxylin and eosin stain.

Ethical approval

This is a case report, and not a research study, therefore ethical approval is not mandatory.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal upon request.

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Author contribution

N.K.-H. and S.Z.: study concept; literature review; design; writing the paper; submission. E.D.: data collection, analysis and interpretation. SA.: study concept and design; data collection and analysis.

Conflicts of interest disclosure

The authors declare that they do not have any commercial or associative interest that represents a conflict of interest in connection with the work submitted.

Research registration unique identifying number (UIN)

This manuscript is a case report and not a human study; therefore does not need to be registered.

Guarantor

Shmuel Avital and Nathan Khabyeh-Hasbani accept full responsibility for the work and the conduct of the study, had access to the data, and controlled the decision to publish.

Data availability statement

This submission is a case report and therefore does not include any results derived from research data.

Provenance and peer review

This paper was not invited, commissioned, nor externally peer-reviewed.

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