Case report

Abdominopelvic actinomycosis: An unexpected diagnosis in an elderly female with a destructive-appearing soft tissue mass

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\textbf{A B S T R A C T}

We present the case of an elderly female who underwent a workup for acute blood loss anemia that incidentally led to the discovery of abdominopelvic actinomycosis. While esophagogastroduodenoscopy and colonoscopy were unremarkable, CT abdomen/pelvis displayed a soft tissue mass in the left sacral ala and presacral area that appeared suspicious for malignancy. MRI pelvis revealed a presacral abscess, and an IR-guided biopsy cemented the diagnosis. This case exemplifies how actinomycosis can mimic the presentation of cancer. Risk factors included a history of ischemic colitis and lumbar laminectomy, as mucosal tissue compromise and orthopedic hardware can be niduses for infection.

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1. Introduction

Actinomycosis constitutes a rare invasive infectious disease caused by gram positive aerobic or anaerobic filamentous bacilli which are usually found in the oropharynx, gut, and female genitourinary system.\textsuperscript{[1]} Actinomyces carries a low potential for virulence, so processes compromising the integrity of mucosal surfaces such as surgery, trauma, and foreign body insertion can give rise to infection.\textsuperscript{[2]} Infection is insidious in onset, indolent in course, and presents with nonspecific symptoms like abdominal pain, fever, weight loss, and diarrhea.\textsuperscript{[1,2]} This clinical picture can mislead clinicians to suspect other infectious, inflammatory, or oncological differentials, including tuberculosis, inflammatory bowel disease, diverticulitis, nocardiosis, and solid tumors.\textsuperscript{[1,2]} The natural history of actinomycosis is marked by a progressive, tumor-like pattern of tissue infiltration.\textsuperscript{[1,2]} Invasive procedures are warranted to obtain tissue for histological stain or culture for diagnostic confirmation.\textsuperscript{[1,2]} We describe an elderly female with a previous lumbar laminectomy and ischemic colitis found to have abdominopelvic actinomycosis despite imaging concerning for malignancy. Figs. 1 to 3.

1.1. Case report

An 86 year old female with a history of diabetes, hypertension, hyperlipidemia, chronic kidney disease, lumbar laminectomy, and ischemic colitis presented with a fall in the setting of generalized weakness and melena. Physical exam was remarkable for pallor. Her white blood cell count was 12,100/\textmu L and hemoglobin was 7.5 g/dL. A gastrointestinal workup sought an etiology for iron deficiency anemia secondary to blood loss superimposed on anemia of chronic disease. Esophagogastroduodenoscopy and colonoscopy ruled out an active bleed. Sigmoid and descending colon diverticulosis was present. CT abdomen and pelvis did not reveal an intra-abdominal or retroperitoneal bleed. Incidentally, the patient was found to have a soft tissue mass in the left sacral ala and presacral region.

MRI pelvis demonstrated infiltrative areas in the bilateral sacral ala and a multifocal rim-enhancing presacral fluid collection. The presacral abscess was drained and the sacral mass was biopsied. Neurosurgery was consulted given the spinal surgery history, and MRI spine ruled out epidural abscesses.

An IR-guided biopsy solidified a diagnosis of actinomycosis. Cytology revealed gram positive filamentous organisms with yellow sulfur granules, along with dense fibrous tissue infiltrated by neutrophils and some lymphocytes. Additionally, the wound culture grew \textit{E coli}, \textit{Bacteroides fragilis}, and \textit{Parvimonas micra}. Urine culture grew \textit{E coli}. The patient was discharged on a 6-week course of piperacillin-tazobactam and trimethoprim-sulfamethoxazole. She followed up with infectious disease to transition to an oral regimen.

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2. Discussion

Actinomycetes represent a diverse group of aerobic and anaerobic bacteria united by a common structural feature, filamentous branching structures that fragment into bacillary or coccoid forms [3]. Forty-seven species of Actinomyces exist, 25 of which have the capacity to induce infection among humans [4,5]. Actinomycosis can be localized to any of the following anatomical regions: cervicofacial, abdominopelvic, and thoracic [3,4]. When the integrity of a vulnerable mucosal barrier is compromised, Actinomyces can spread contiguously in a manner that ignores anatomical planes and leads to infection [2,6]. Actinomycosis is often referred to as “the great masquerader” in the clinical setting as it can mimic other disease states given its propensity to impact a wide variety of organ systems [5,7]. In fact, the chronic progression of symptoms, ability to form masses, and pattern of infiltration renders actinomycosis an excellent mimic of malignancy [3,7–9].

Similar to this case, actinomycosis commonly occurs in the context of a polymicrobial infection [3–5,8]. Tissue culture and pathology ultimately facilitate the diagnosis [1,5,8,10]. Importantly, multiple factors can prevent isolation of Actinomyces from a culprit infection site, namely prior antimicrobials, inhibition of growth by concomitant bacteria, and insufficient incubation time [5,8]. Sub-optimal conditions for culture growth can interfere with the diagnosis, and appropriate options for successful growth are chocolate blood agar media at 37 °C, brain heart infusion broth, Brucella blood agar with hemin and vitamin K, phenylethyl alcohol, and mupirocin-metronidazole blood agar [5]. While one or more of the above factors prevented Actinomyces growth on culture in this case, biopsy was pathognomonic for actinomycosis. Granulomatous tissue surrounded acute inflammatory lesions stemming from the infection and sulfur granules were observed [3,4,9,10]. Methods in the field of microbiological taxonomy, such as implementation of comparative 16S ribosomal RNA sequencing, can be utilized to solidify the diagnosis [1,5].

Abdominal actinomycosis infection can arise in a variety of clinical settings, such as abdominal surgery, ruptured appendicitis, inflammatory bowel conditions, perforated colonic diverticulitis, and tuboovarian abscess [3,7,8]. A prior hospitalization for ischemic colitis was the inciting event for our patient’s infection. Abdominal actinomycosis involving the appendix, cecum, or colon can manifest years following the predisposing gastrointestinal mucosal disruption [4,5,8]. Furthermore, mucosal atrophy associated with aging can increase the risk of infection as a result of reduced mucosal resistance [8]. This patient had diverticulosis, which can occasionally raise the chance of acquiring actinomycosis by weakening the epithelium to establish a nidus for infection.

Cases of pelvic actinomycosis are more frequently observed in patients who have undergone placement of an intrauterine device in the past [3]. However, invasive disease can also follow pelvic reconstructive surgery [7]. At the time of diagnosis, patients may experience vaginal bleeding, fevers, pelvic pain, and weight loss [3]. Ovarian tumors are often suspected in this clinical setting [3]. Since our patient never had an intrauterine device or gynecological surgery, the suspected portal of entry was gastrointestinal. Similar to abdominal cases, pelvic actinomycosis can take years to identify due to the gradual progression of the disease process and nonspecific symptom presentation [7–9].

The patient’s history of lumbar laminectomy rendered her susceptible to hardware infection. Since Actinomyces have the capacity to form a biofilm on hardware, removal of hardware was advised. Actinomyces can spread to the central nervous system via hematogenous dissemination and cause significant morbidity or mortality via the development of meningitis, epidural abscess, and subdural empyema [5].
Appropriate antimicrobial therapy is essential. Actinomycosis typically responds to treatment with penicillin G or amoxicillin, thus these agents are frequently referred to as the antimicrobials of choice [5,8,9]. Macrolides as well as clindamycin can be reasonable alternative treatment options [5]. Broad spectrum antimicrobials such as piperacillin-tazobactam and certain carbapenems have been successfully utilized as well [5]. Our patient’s 6-week course of piperacillin-tazobactam and trimethoprim-sulfamethoxazole led to clinical improvement even before transition to oral antimicrobials.

While CT abdomen and pelvis led us to suspect malignancy, biopsy directed the team to the diagnosis [1,2,4]. Capsule endoscopy was recommended to complete the anemia workup, yet the patient did not follow up with gastroenterology.

The diagnosis required careful analysis of risk factors, radiographic findings, and pathology results [8–10]. Ischemic colitis and diverticular disease made the colonic epithelium more vulnerable to Actinomyces invasion [9]. Hardware removal was recommended to prevent the spread of infection to the central nervous system, however the patient opted for lifelong antimicrobials.

3. Conclusion

While abdominopelvic actinomycosis is a rare clinical entity, it is an important differential to consider in the presence of a process that compromises a mucosal barrier, an infiltrating mass that resembles a tumor, and a range of non-specific gastrointestinal symptoms [1,6]. In our patient, the mass that prompted additional investigations was an incidental finding on a workup for acute blood loss anemia. Actinomycosis may take years to manifest following the initial gastrointestinal mucosal insult. Bacterial translocation from ischemic colitis one year prior was believed to be the primary source of infection for our patient [2]. Since the patient underwent lumbar laminectomy 10 years ago, and we are unable to rule out that hardware contributed to actinomycosis, hardware removal was recommended. Her refusal to pursue hardware removal made it reasonable for her to remain on lifelong suppressive antimicrobial therapy. In uncomplicated cases, a 6-month course of antimicrobials represents the mainstay of treatment, and high-dose penicillin G is often the medication of choice [1,5,8,9]. In complicated cases with suboptimal response to antimicrobials or those demonstrating widespread tissue necrosis, drainage or operative management is warranted [2].

CRediT authorship contribution statement

Dr. Elise Hyser was involved in the conceptualization and writing of the manuscript. Dr. Drashti Antala was involved in writing the manuscript as well. Dr. Harvey Friedman and Dr. Jonathan Stake were contributors to the manuscript through the process of revising it, and thus had a supervisory role in the creation of this work. Dr. Elise Hyser, Dr. Drashti Antala, and Dr. Jonathan Stake were all directly involved in caring for the patient.

Conflict of interest

We have no conflicts of interest to disclose.

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