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Case Report

Gastric Outlet Obstruction Secondary to Gastric Actinomycosis: A Case Report and Literature Review

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Abstract **Actinomycosis is a rare chronic infection caused by the bacteria of the Actinomyces species. Gastric actinomycosis is an extremely rare disease. The usual presentations are low-grade fever, epigastric pain, weight loss, and upper gastrointestinal hemorrhage. To date, about 21 cases have been reported in the literature. It is difficult to diagnose preoperatively. This chronic infection has a propensity to mimic malignancy. An accurate diagnosis is always obtained in a histological examination of the resected specimen. We report here a 39-year-old woman with primary gastric actinomycosis presenting with gastric outlet obstruction. Because of its rarity, gastric actinomycosis is an entity overlooked by most surgeons. Reporting of such case may help increase the awareness of this important and curable disease. To our knowledge, this is the first reported case of gastric outlet obstruction secondary to gastric actinomycosis in the literature. The literature on gastric and abdominal actinomycosis were also reviewed.**

Keywords: **actinomycosis, gastric outlet obstruction**

INTRODUCTION

Actinomycosis is a rare chronic suppurative and granulomatous inflammation caused by an anaerobic, filamentous, Gram-positive bacteria of Actinomyces species.[1-6] Human actinomycosis was first described by Israel in 1878[7] and it was first diagnosed in a live patient by Ponficke in 1879.[8] The four main clinical forms of actinomycosis include cervicofacial (31%-65%), abdomino/pelvic (20%-36%), thoracic (15%-30%) and cerebral form.[1,9-11] The clinical presentation of abdominal actinomycosis is variable depending on the site of involvement. The appendix and ileocecal region are the most commonly involved sites (65%),[3,5,9,10,12] but infection of any intra-abdominal organs is possible. Primary gastric actinomycosis is a rare infection. As a result of the low gastric pH, the organisms may be killed or growth is inhibited.[5,13,14] Until 2007, only 21 cases of primary gastric actinomycosis have been reported in the literature.[3,5,13-22]

Herein, we report a case of primary gastric actinomycosis presented with gastric outlet obstruction and the literature were also reviewed. To our

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knowledge, this is the first reported case of gastric outlet obstruction secondary to gastric actinomycosis in the literature.

Case Report

A 39-year-old woman presented with a history of non-bilious vomiting for two weeks. She had mild epigastric pain without fever for about one month. She had no previous history of peptic ulcer disease, abdominal surgery, and intrauterine device (IUD) use. Abdominal examination revealed a positive finding of splashing sound without palpable mass. Laboratory investigations showed only mild leukocytosis (15,000/mm^3). Plain abdominal X-ray showed a fluid-filled dilated stomach. Gastroscopy revealed a circumferential submucosal mass at prepyloric antrum, and the scope could not pass through the pyloric canal (Figure 1). CT scan showed a circumferential mass (5-cm size) at prepyloric antrum causing gastric outlet obstruction with dilatation of the stomach. An operation was performed with the presumptive diagnosis of gastric outlet obstruction due to gastric malignancy (possible gastric lymphoma). The operative findings revealed a circumferential mass at prepyloric antrum. No distant metastasis nor peritoneal dissemination was noted. D2 subtotal gastrectomy with Billroth II anastomosis was performed. Pathological examination showed a focal markedly chronic suppurative inflammation with clumping of bacteria morphologically consistent with Actinomyces (Figure 2). All lymph nodes were negative for malignancy (27 nodes). The post-operative course was uneventful. Following surgery, she was treated with oral penicillin for two months. She remained well six months after surgery.

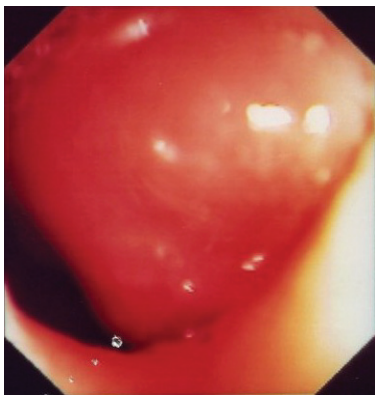

Figure 1 Submucosal mass at prepyrolic antrum

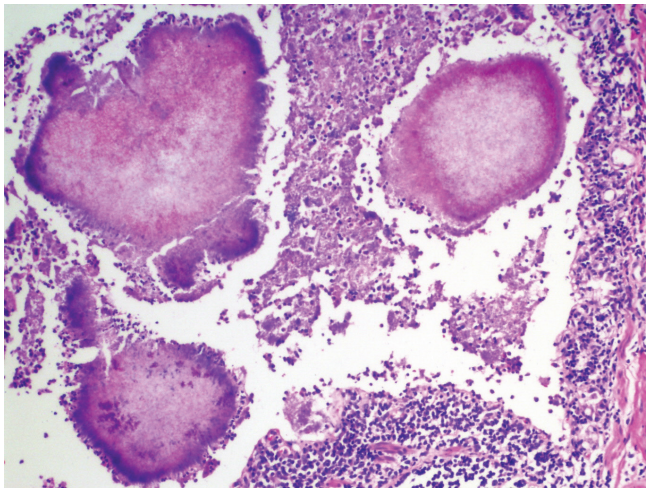

Figure 2 Actinomyces

Discussion

Actinomycosis in human is most commonly caused by *Actinomyces israelii.*[1-2,6,10,23-26] They are normal commensal inhabitants of the human GI tract and female genital tract.[1,2,6,9,23,25,27] This bacteria does not invade intact mucous membrane.[1,5,11,23] This opportunistic organisms can become pathogenic in the presence of damaged mucosal barrier.[5-6,23] Factors that precipitate abdominal actinomycosis include previous abdominal surgery, bowel perforation, penetrating abdominal trauma, and foreign bodies (IUD). Berardi in 1979 first reported the association between this commensal organism *(Actinomyces israelii)* and clinical abdominal actinomycosis.[6] He postulated that the pathogenesis of this condition was due to a disruption in mucosal integrity.[6] In most cases, it has been impossible to trace the mechanism by which Actinomyces had reached the gastric wall. Our patient did not have any identifiable predisposing factors. There is no documented person-to-person transmission of the disease.[1]

The usual presenting clinical manifestations of gastric actinomycosis are low-grade fever, epigastric pain, weight loss, and upper GI bleeding.[1,5,14,16] The duration of symptoms ranged from one month to several years.[5,13,14,16] Our patient developed symptoms of gastric outlet obstruction for two weeks. There had no previously reported case of this manifestation from gastric actinomycosis from the literature review. The pathogen produces a characteristic granulomatous inflammation, followed by extensive reactive fibrosis.[25]

Hematogenous and lymphatic disseminations are uncommon as in our case. The term "woody induration" has been used to describe this lesion. Abdominal actinomycosis is one of the greatest challenge for diagnosis. Actinomycosis was once described as "the most misdiagnosed disease".[29] There is no specific radiological evidence or endoscopic appearance of the disease. CT findings usually demonstrated an infiltrative lesion with diffuse gastric wall thickening. The appearance suggested adenocarcinoma or lymphoma of the stomach[4,9] as in our case. Similar to radiologic studies, the endoscopic findings of gastric actinomycosis may simulate gastric neoplasm and may include submucosal tumor-like or infiltrative lesion. Because of the submucosal localization of the inflammatory process, endoscopic biopsy specimens usually revealed nonspecific inflammatory change.[5,18,22] The diagnosis is almost always ascertained after surgery and histopathological examination of the resected specimen. Only one case is reported by Lee et al in 2004,[5] in which the diagnosis of gastric actinomycosis was made on microscopic evaluation of endoscopic biopsy specimen. Culturing is negative in most cases of actinomycosis (>76%).[24,25] In our case, as no material had been sent for culture, because during operation, actinomycosis was not suspected, and all the resected tissue had been formalin-fixed and paraffin-embedded, we had no chance to perform microbiological studies. However, the histopathological examination was typical of actinomycosis. As in our case and a review of the literature, the rarity of the disease and its non-specific clinical, radiological, and endoscopic findings are the causes for frequently mistakening this condition as neoplasm.[26] This may lead to unnecessary radical surgical procedure with high morbidity and even mortality rates. In our case, D2 subtotal gastrectomy was performed without morbidity and mortality. In case of gastric actinomycosis, an extensive surgical procedure or oncologic resection should be avoided, only limited surgical resection is effective.

Uncomplicated actinomycosis can be medically treated by antibiotics[10,30]. A prolonged treatment course is required because of the poor penetration of antibiotics into the fibrotic tissue. Actinomyces species are susceptible to penicillin, but the duration of treatment varies from several weeks to months to achieve permanent recovery[1]. The recommended antibiotic after surgery is oral penicillin 2-4 gm/day for 3-12 months.[4,10,23-25] However, in case Actinomycosis can be completely removed surgically, a shorter period of antibiotic therapy could be effective.[24] Despite the good response to penicillin therapy, cure is rarely achieved without operation to eradicate the inflammatory process.[10,23,31] Only four cases treated exclusively with antibiotics have been cured of the disease[15]. Surgery is valuable as a therapeutic adjunct and relieve the symptoms and remove infected fibrotic tissue.

Conclusions

We report the first case of gastric outlet obstructions secondary to gastric actinomycosis. An awareness of this extremely rare condition is the key to make the diagnosis. Maintaining a high index of suspicion for this condition will prevent unnecessary radical surgery for presumed gastric malignancies. Even though gastric actinomycosis is an extremely condition, it should be included in the differential diagnosis of the cause of gastric outlet obstruction.

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