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Case report

Actinomycosis: Case report of an unusual cause of diaphragmatic herniation

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ABSTRACT

Introduction: We present a case of actinomycosis which resulted in a symptomatic diaphragmatic hernia requiring operative repair, an unusual complication not found in the literature. Actinomycosis is a chronic, slowly progressive infection caused by the bacterial genus Actinomyces which characteristically causes necrosis and abscess formation in a myriad of organ systems.

Case presentation: A 5 year old male presented with nonspecific symptoms which were, after a short delay, identified as actinomycosis and treated with appropriate antibiosis. His infection was complicated by development of a diaphragmatic hernia, which subsequently became symptomatic and required surgical repair.

Clinical discussion: While this diaphragmatic hernia is an unusual complication of actinomycosis not previously found in the literature, the patient's delayed diagnosis of the infection is typical. Surgical intervention was warranted for symptoms of the sequelae of the disease, not control of the disease itself.

Conclusion: Laparoscopic repair of the multiple diaphragmatic defects was successful with a intraperitoneal onlay biologic mesh, with resolution of symptoms. It is possible the need for surgical intervention in future cases of actinomycosis could be avoided with higher index of suspicion leading to earlier diagnosis.

1. Introduction

We present a case of thoracoabdominal actinomycosis which resulted in a symptomatic diaphragmatic hernia requiring operative repair, an unusual complication not found in the literature. Actinomyces are commensal flora of the oropharynx, colon, and female genital tract which can cause subacute and chronic infections that can be difficult to diagnose. Infections occur when this non-virulent organism opportunistically invades damaged mucosa, and, often with the help of a companion bacteria in overcoming host defenses, typically results in polymicrobial infection. Once established, the infection stimulates a suppurative and granulomatous inflammatory response which ignores tissue planes and spreads contiguously. Rarely, actinomycosis may also spread hematogenously to other systems. More than half of infections are in the cervicofacial region [1]. Other locations include the female genitourinary tract especially in the setting of intrauterine contraceptive device use, the gastrointestinal tract in the setting of abdominal surgery or perforated viscus, and pulmonary space in the setting of aspiration [2]. The presentation is often nonspecific and varies according to site of infection. In the GI tract, symptoms can include fever, weight loss, abdominal pain and fullness, or change of bowel habits [3]. The diagnostic gold standard is tissue biopsy and anaerobic culture, however, this may be falsely negative in half of cases. Microscopic evaluation of biopsied tissue demonstrating gram-positive filamentous rods or necrosis of yellow sulfur granules helps in preliminary diagnosis [4]. Lab work may show mild leukocytosis with elevated C-reactive protein and erythrocyte sedimentation rate [5]. Imaging may be non-specific, as aggressive infections frequently invade normal anatomic barriers and can be confused with malignancy and other inflammatory diseases with lesions that cross tissue planes [6].

Once actinomycosis is diagnosed, the mainstay of treatment is prolonged high-dose antibiotic therapy. Common courses include parenteral penicillin for 2 to 6 weeks followed by oral therapy for 6 to 12 months until symptomatic and imaging resolution of disease is reached [7]. Surgery is adjunctive therapy, although the diagnosis sometimes is not confirmed until after percutaneous or operative intervention has already been performed [8]. The prognosis is excellent with antibiosis, but worsens if surgery is required to control the disease. Complications

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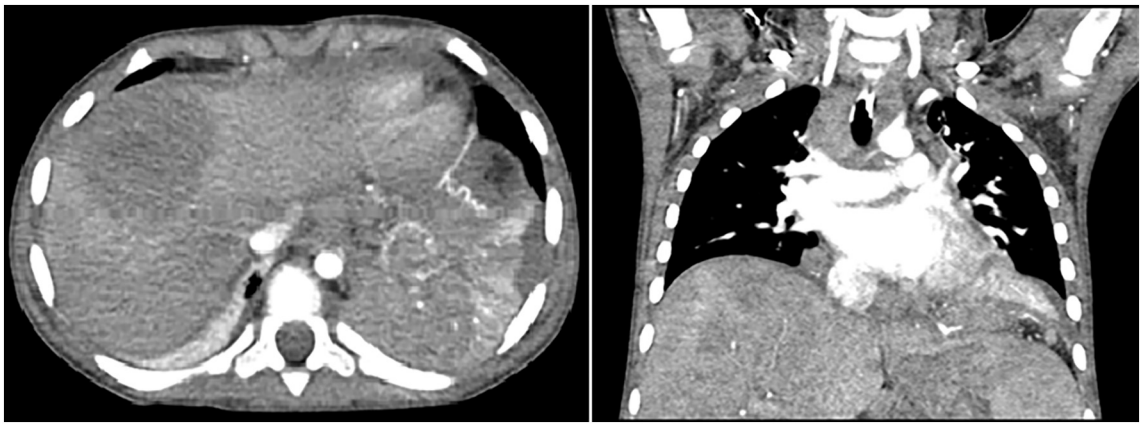

Fig. 1. Computed tomography (CT) at initial diagnosis, showing (left) dominant hepatic abscess and splenic phlegmon, and (right) hepatic and splenic lesions with left lower lobe infiltrate.

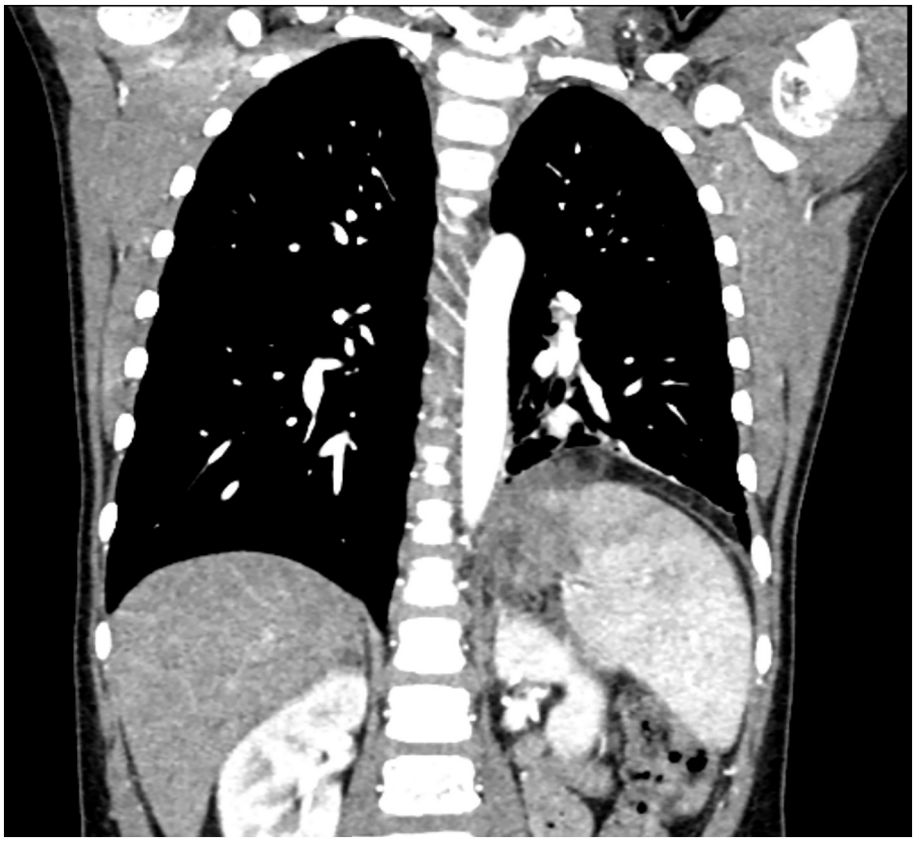

Fig. 2. CT angiogram one year after initial diagnosis showing elevated left hemidiaphragm and gastric herniation.

include abscess development and spread of infection. This case has been reported in line with the SCARE 2020 criteria [9].

2. Patient information

A 5-year-old male presented with one day of right-sided abdominal pain. His history was significant for six months of intermittent diarrhea, fevers, and night sweats. He was evaluated three months prior for these symptoms, at which time he tested positive for rotavirus and was found to have an anemia of hemoglobin 7.0 g/dL, for which he was prescribed but did not take iron supplementation. On his subsequent presentation, his physical examination revealed tenderness of the right abdomen worse in the lower quadrant as well as weight loss of 1 pound from the year prior. He had no contributory family, drug, or psychosocial history. Laboratory findings included a microcytic anemia with hemoglobin of 8.5 g/dL, a leukocytosis of 17,900 WBC/mm3 with 72% neutrophils, and a C-reactive protein of 130 mg/L. Ultrasound of the right lower quadrant showed free pelvic fluid with a non-visualized appendix, and computed tomography (CT) of the abdomen found hepatosplenomegaly with multiple liver and splenic rim-enhancing lesions, heterogeneous decreased attenuation of the superior spleen, and a left lower lobe consolidation (Fig. 1). He was diagnosed with a left lower lobe pneumonia and hepatic and splenic abscesses. He was treated with empiric ceftriaxone and metronidazole, and image-guided drainage of the largest hepatic collection. His symptoms and clinical picture improved and he was discharged. His final antibiotic regimen, upon return of his

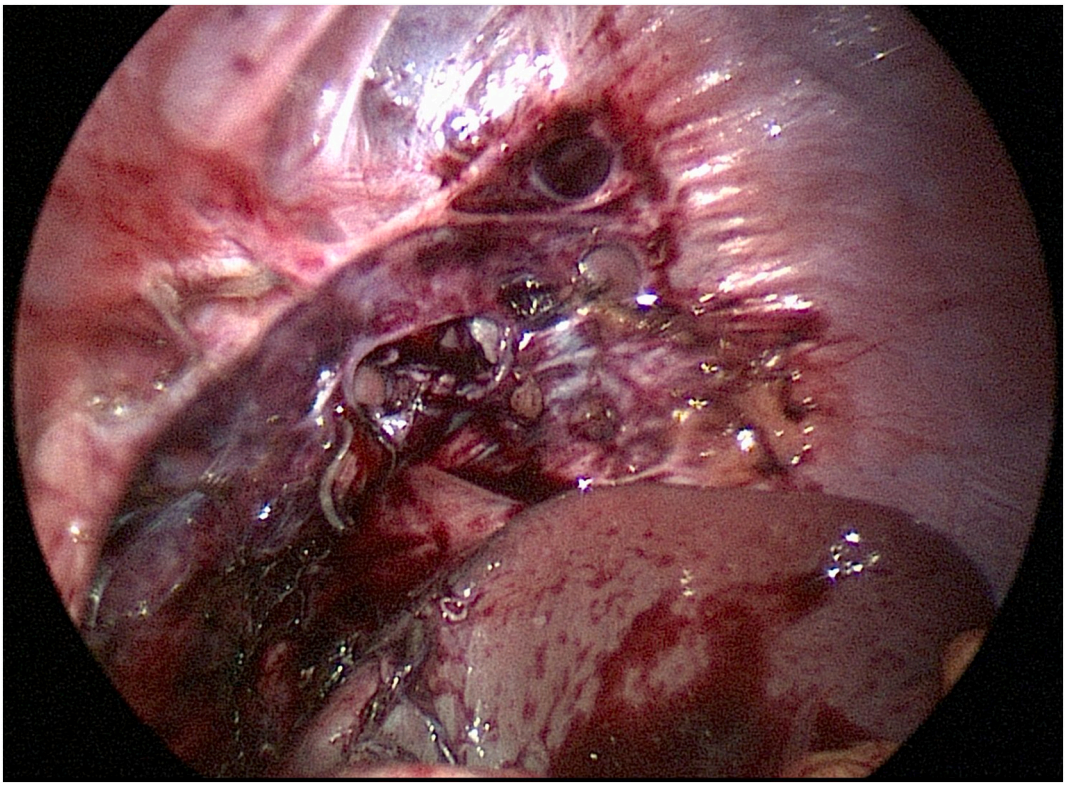

Fig. 3. Left hemidiaphragm with multiple defects and general thinning and fibrosis.

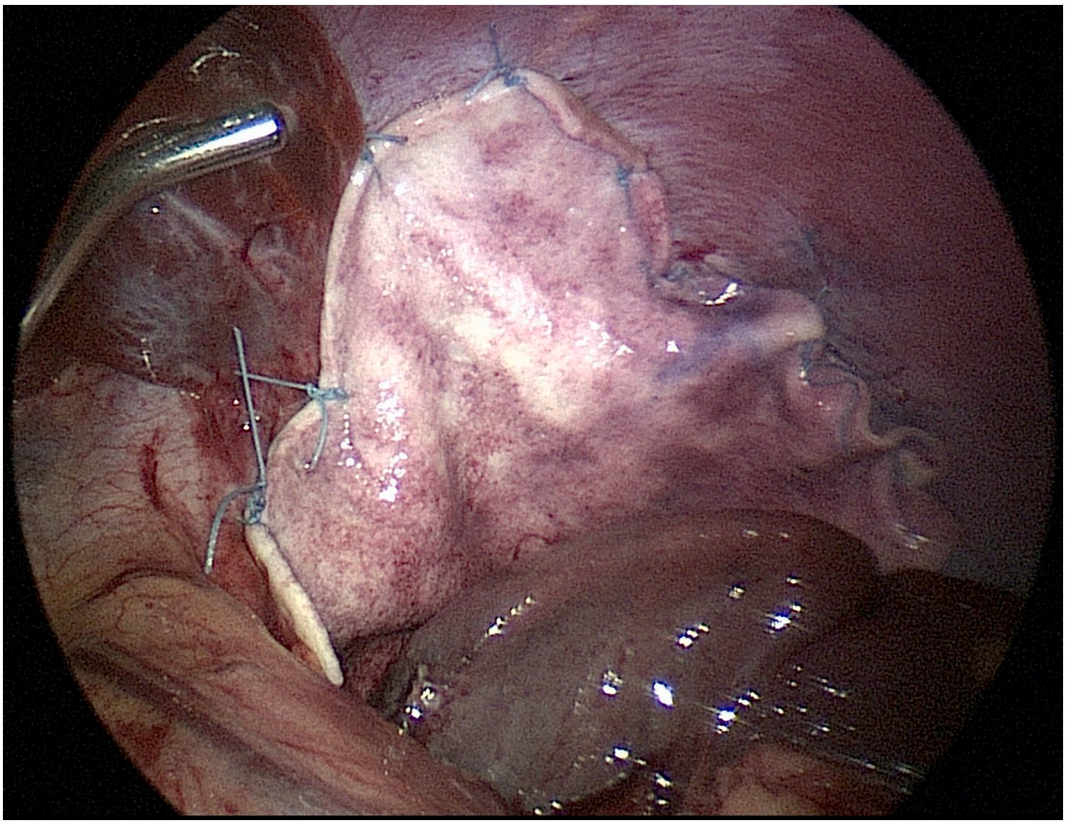

Fig. 4. Alloderm intraperitoneal mesh repair of left diaphragmatic defects.

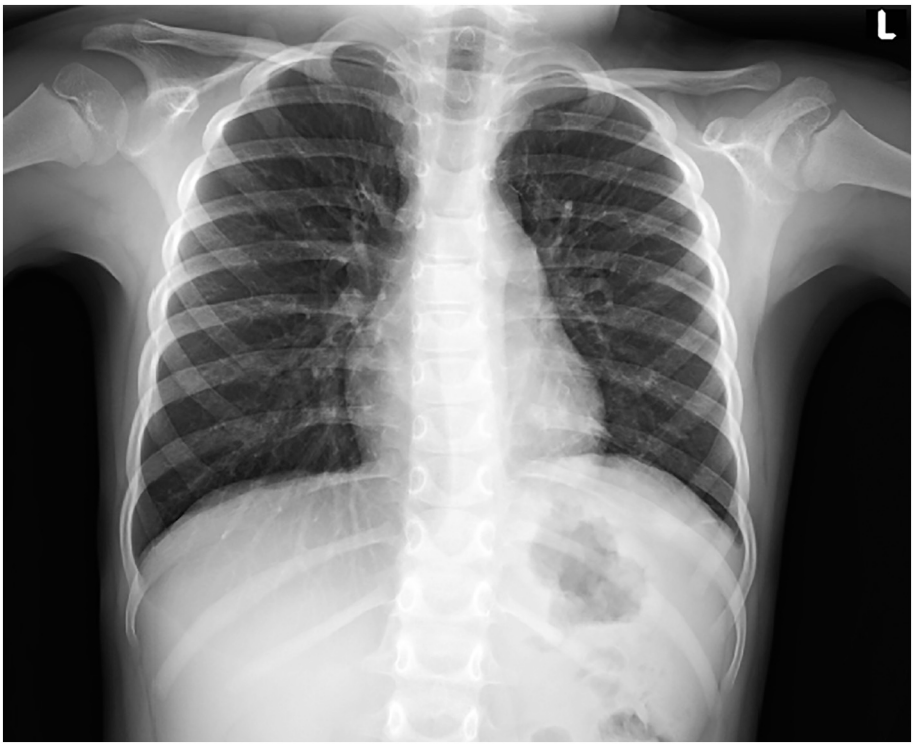

Fig. 5. Chest radiograph obtained three months postoperatively with resolution of left basilar opacity.

hepatic drain cultures (polymicrobial growth including Actinomyces odontolyticus, Capnocytophaga species, Fusobacterium species, *Eikenella corrodens*, and Aggregatibacter actinomycetecomitans), was guided by consultation with Infectious Disease and was consistent with typical therapy for actinomycosis: a six-week course of ceftriaxone and metronidazole followed by ten months of oral amoxicillin monotherapy.

A follow up chest X-ray one year after his drainage procedure showed persistent left lower lobe opacity. A CT angiogram-chest performed for further evaluation revealed elevation of the left diaphragm, with thinning at its mid-portion with at least one complete defect seen, with herniation of intraabdominal fat, a portion of the gastric fundus, and possibly the splenic dome (Fig. 2). The patient was referred to our surgery clinic. In clinic, he reported mild post-prandial pain with heavy meals, and had a normal physical exam. A fluoroscopy study showed elevation of the left diaphragm with decreased motion that was not paradoxical. He was taken to the operating room and underwent diagnostic laparoscopy, performed by the attending surgeon who was trained in Pediatric Surgery and assisted by a Pediatric Surgery fellow and General Surgery resident. The patient's left hemidiaphragm was indeed significantly elevated and was fibrotic and rigid. Reduction of herniated fat and multiple splenules revealed four small separate diaphragmatic defects (Fig. 3). We patched the defects with an Alloderm (Allergan; Dublin, Ireland) intraperitoneal mesh (Fig. 4). There were no deviations from the initial mamagement plan and the patient was discharged with standard postoperative laparoscopy instructions. At one and three month follow up appointments he was doing well with normal activity and no respiratory or gastrointestinal symptoms. A chest radiograph obtained 3 months post-operatively showed resolution of the left basilar opacity (Fig. 5).

3. Discussion

In this case, the patient had nonspecific gastrointestinal symptoms for six months and had received medical evaluation three months prior to his eventual diagnosis of Actinomycosis. It is most likely his primary infection was pulmonary with direct and hematogenous spread to the spleen and liver, given his left lobe involvement and absence of risk factors for a gastrointestinal source. Percutaneous drainage and antibiosis were sufficient in this case for controlling the infection. Unfortunately, the characteristic breakdown of tissue planes in this patient led to a complication for which we have not found precedent: an elevated, fibrotic hemidiaphragm with multiple diaphragmatic hernias, and symptomatic herniation of abdominal contents through the diaphragmatic defects. For this complication, surgery was indicated, even though the initial infection had already resolved. We were unable to perform a primary tissue repair after reduction of the hernias due to fibrosis and thinning of the diaphragm. Therefore, we chose a resorbable biologic implant over a synthetic one in the setting of a previously infected surgical bed with a causative organism known to recur up to years after the initial infection. It is possible this surgery could have been avoided had a higher index of suspicion at the patient's first encounter with the health system led to an earlier diagnosis.

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Consent

Written informed consent was obtained from the patient's parent 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 on request.

Ethical approval

This case report was reviewed and determined exempt from specific ethical approval by the Drexel University Office of Research and Innovation Institutional Review Board, as it is not human subjects research as defined by DHHS or FDA regulations. Exemption number 2104008458.

Author contributions

Hurwich: writing the paper, data collection, data interpretation.
Pennell: editing the paper, data interpretation.
Prasad: study concept, data interpretation, editing the paper.

Research registration

Not applicable.

Guarantor

Dr. Rajeev Prasad.

Provenance and peer review

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Declaration of competing interest

None declared.

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