International Pathology Day 2024 #IPD2024

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Case Report: A Rare Case of Oesophageal Actinomycosis with Underlying Poorly Differentiated Adenocarcinoma

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Background

Actinomycosis is a rare, chronic infection caused by Gram-positive, anaerobic, filamentous Actinomyces bacteria, typically found in the oral cavity, urogenital tract, and gastrointestinal tract. Although *Actinomyces spp*. have low pathogenicity in healthy individuals, mucosal disruption and immunosuppression can lead to infection. Infection can be granulomatous and suppurative, and in some cases aggressive and infiltrative. It is known to mimic appendicitis, Crohn's disease and invasive malignancy. Oesophageal actinomycosis is extremely rare, with only around 24 cases reported in the U.S., often presenting as erosions or ulcers on endoscopy. These infections generally arise following trauma, infection, or instrumentation, but can also occur without clear predisposing factors. In this case, a rare actinomycotic infection of the lower oesophagus was discovered in a young, otherwise healthy patient who was being investigated for suspected malignancy.

Case Presentation

A 37-year-old male patient presented to the emergency department with a 4-week history of epigastric pain and dysphagia, worsening over the past 3 days, along with nausea and an inability to tolerate food. The patient reported a 10 kg weight loss over the last 2 months. He had no past medical history and was not on any regular medications. The patient was admitted under the medical team for imaging and an oesophago-gastro-duodenoscopy (OGD) with biopsy.

CT and OGD Findings

CT chest abdomen and pelvis showed circumferential thickening of the oesophagus at the GOJ with adjacent nodularity in the surrounding mediastinal fat. There were bulky lymph nodes around the lesser curvature of the stomach and coeliac axis. Hypoattenuated lesions within the liver were suggestive of hepatic metastasis.
First OGD identified a deep necrotising ulcer in the lower third of the oesophagus (40cm from the incisors) extending to the gastro-oesaphageal junction (GOJ). The stomach and duodenum appeared normal. Biopsies were taken.
Second OGD and biopsy was undertaken 3 weeks later, showing a fungating and invasive mass covering three quarters of the circumference of the oesophagus (Figure 2).

Histopathological Assessment

The oesophageal biopsy (Figure 1 A-C) shows small fragments of inflamed, superficial squamous epithelium, along with abundant basophilic filamentous aggregates of bacteria, consistent with *Actinomyces spp*. Actinomyces was confirmed by positive Gram and Periodic Acid-Schiff (PAS) stains. The typical causative agent in humans is *Actinomyces israelii*, but this is not distinguishable by histology.

This case was discussed at the multidisciplinary team meeting. Actinomyces can mimic invasive carcinoma, however, given the liver lesions and bulky nodes, an underlying malignancy needed to be excluded. A decision was made to to treat with intravenous benzylpenicillin and re-biopsy in 2-4 weeks. Human immunodeficiency virus and other immunodeficiency screens were negative.

The second oesophageal biopsy showed sheets of pleomorphic malignant cells, diagnostic for poorly differentiated adenocarcinoma, in addition to ongoing actinomyces infection (Figure 3). The patient is awaiting definitive treatment.

Conclusion

This case illustrates the diagnostic complexity of dual pathologies whereby actinomyces is known to mimic and co-exist with invasive malignancies. The rarity of this case, combined with its atypical presentation, underscores the need for heightened clinical suspicion and thorough evaluation in similar cases. Timely diagnosis and appropriate management for both the infection and the underlying malignancy are crucial for patient outcomes. This case contributes valuable knowledge to the limited body of literature on esophageal actinomycosis.

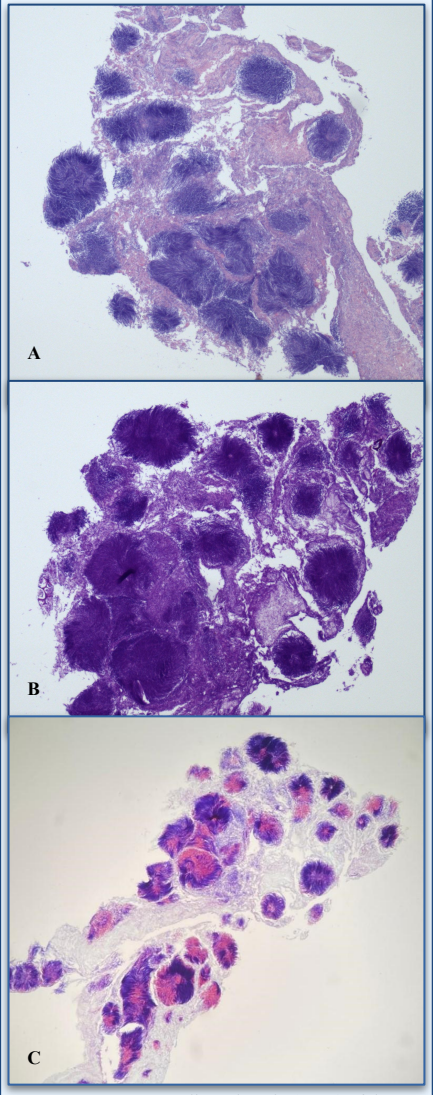

Figure 1 A: Haematoxylin and Eosin (H&E) of the first biopsy showing basophilic filamentous aggregates of bacteria. **B:** Periodic Acid-Schiff (PAS) stain showing positive staining of the organisms. **C:** Gram stain highlighting the bacterial elements

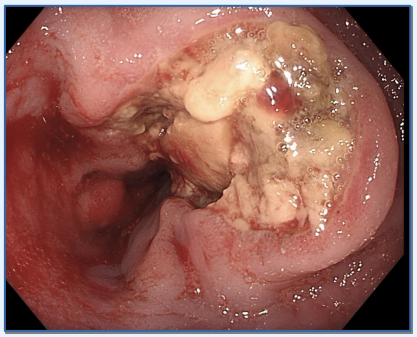

Figure 2: Esophagogastroduodenoscopy shows a deep necrotising ulcer in the lower third of the oesophagus, extending to the gastroesophageal junction.

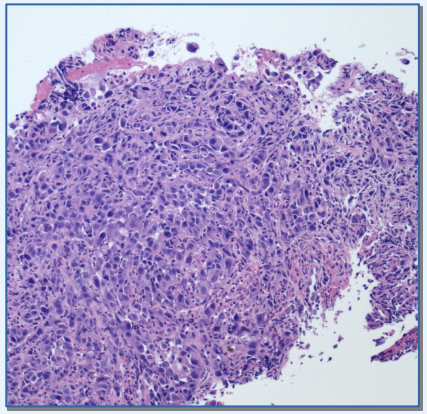

Figure 3: Second oesophageal biopsy (H&E) showing a poorly differentiated adenocarcinoma. The cells are arranged in sheets with significant pleomorphism. Poorly formed bacterial colonies were also present on this biopsy (not in this image).

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