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References

1. Katsarelias D, Faisal M, Glavas R, Bartholomä WC. Double papilla of Vater: arare anatomic variant. Endoscopy. 2016;48 Suppl1:E133-4.
2. Rana SS, Bhasin DK. Papilla with separate bile and pancreatic duct orifices. JOP2013;10;14:302-3.
3. Rajnakova A, Tan WT, Goh PM. Double papilla of Vater: a rare anatomic anomaly observed in endoscopic retrograde cholangiopancreatography. SurgLaparoscEndosc. 1998; 8:345-8.
4. Simon MA, Moreira VF, Merono E, Seara JF, del Olmo L. The double papilla of Vater. GastrointestEndosc. 1985;31:353–354
5. Ando H. Embryology of the Biliary Tract. Dig Surg2010;27:87–89
6. Horiguchi S,Kamisawa T. Major Duodenal Papilla and Its Normal Anatomy. Dig Surg2010;27:90–93
7. Kubota K, Fujisawa T, Abe Y, Inamori M, Kirikoshi H, Saito S, et al. Clinical significance of the duodenal papilla in patients with separate orifices for the bile and pancreatic ducts. J Gastroenterol. 2007;42:70-8

Esophageal Actinomycosis, A Rare Complication After Self-Expanding Metallic Stent Deployment

Actinomycosis is a rare, sub-acute to chronic bacterial infection caused by Actinomyces species which are facultative, anaerobic, gram positive bacilli. It is most frequently caused by species Actinomyces israelii. They are mainly present as saprophytes in the soil. In humans, they can occur as commensals of the oral cavity, gastrointestinal tract and vagina. Actinomycosis is preceded by some known insults to the native mucosa. It is characterized by contiguous spread, granulomatous inflammation and formation of multiple abscesses and sinus tracts that may discharge sulphur granules. Though Actinomyces infection can occur in abdomino-pelvic, pulmono-thoracic and cervico-facial areas, oesophageal involvement is an extremely rare occurrence in immunocompetent persons. Here we report a rare case of oesophageal actinomycosis in a 50-year-old lady, following the deployment of a partially covered Self Expanding Metallic Stent (PCSEMS) elsewhere for an indeterminate mid oesophageal stricture. It is noteworthy that this is the first case following oesophageal metallic stenting, reported from India.

Case Report

A 50 year old lady presented with a history of progressive dysphagia to solids and liquids for the past 3 months. Four months ago, she was diagnosed to have an oesophageal stricture at 25 cm, for which a PCSEMS was deployed elsewhere as it was considered malignant.

On physical examination, she was thin built and malnourished. Her blood investigations showed low haemoglobin of 85 g/L and serum albumin of 28 g/L. The oesophagoscopy revealed a proliferative circumferential tissue overgrowth at 20 cm at the proximal end of PCSEMS narrowing the lumen not admitting the endoscope further **(Figure 1)**. Biopsy was taken from the tissue in-growth.

Computed Tomography of the chest revealed an oesophageal metallic stent in the mid and lower third of oesophagus with tissue in-growth into the proximal end of the stent and wall thickening in the mid third of the oesophagus **(Figure 2A & 2B)**. The biopsy and histological examination showed discrete sulphur granules within the granulation tissue in the ulcerated oesophageal tissue consistent with infection by Actinomyces species **(Figure 3A)**.

This patient was treated with a course of intravenous penicillin-G for a month. She had good relief of chest pain and odynophagia with partial alleviation of dysphagia. She was reassessed with upper GI scopy which revealed partial regression of hyperplastic tissue overgrowth. Repeat biopsy from the hyperplastic tissue

revealed only granulation tissue without any evidence of actinomycotic infection or malignancy **(Figure 3B)**. This persistent hyperplastic tissue was subsequently ablated by Argon plasma coagulation.

Discussion

Actinomyces are facultative anaerobic, gram-positive bacilli that colonize oral and GI tract as well as the vaginal mucosa.[1] Although these organisms have a low degree of pathogenicity, mucosal disruption can result in chronic granulomatous and suppurative infection that can involve almost any organ system.[2] Over time, the bacteria can cross natural anatomic boundaries, resulting in multiple abscesses, draining sinus tracts, and fistula formation.[3]

Esophageal actinomycosis has been described in both immunocompetent and immunocompromised patients. Dysphagia and odynophagia are the most common presenting symptoms.[4] Endoscopy may reveal multiple discrete round ulcers. The primary diagnostic tools remain Gram stain and histology which may show typical microscopic findings that include necrosis with yellowish sulphur granules and filamentous Gram-positive fungus-like pathogens.[2] Definitive diagnosis with positive cultures requires 5 to 7 days but is seen in only a minority of cases.[5] Barium oesophagogram may demonstrate deep ulcers with fistulas and multiple sinus tracts. CT may demonstrate a thickened oesophageal wall in early stages with or without sinus tracts.

The thoracic and abdominal forms of actinomycosis is treated with a 4-6 week course of intravenous penicillin - G, 3-4 million units every four hours, followed by oral pencillin or amoxicillin for 6-12 months. Prolonged (6 to 12 months) high doses of penicillin G or amoxicillin is needed to facilitate the drug penetration in abscesses and infected tissues. Tetracycline is the preferred antibiotic for individuals with penicillin allergy, although minocycline, erythromycin, clindamycin, ceftriaxone, and imipenem are available alternative treatments.

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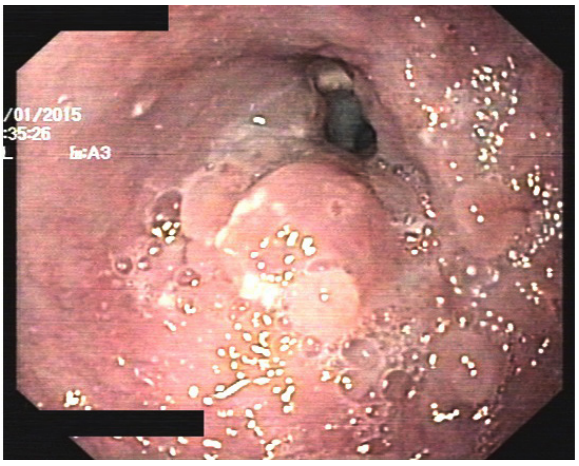

Figure 1: Oesophagoscopy revealing edematous oesophageal mucosa with tissue overgrowth at 20 cm narrowing the lumen eccentrically, obscuring the PCSEMS.

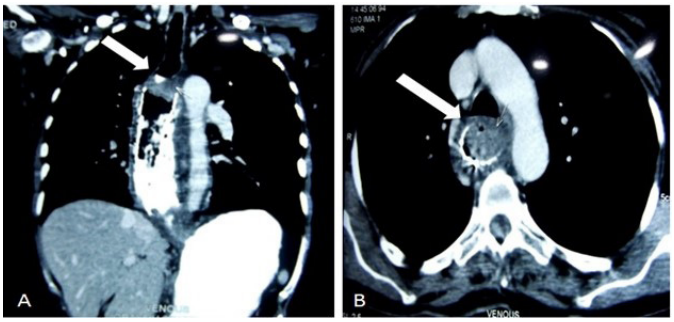

Figure (2A & 2B): Computed Tomography of the chest revealing oesophageal metallic stent in the mid and lower third of oesophagus with tissue in-growth into the proximal end of the stent and wall thickening in the mid third of the oesophagus.

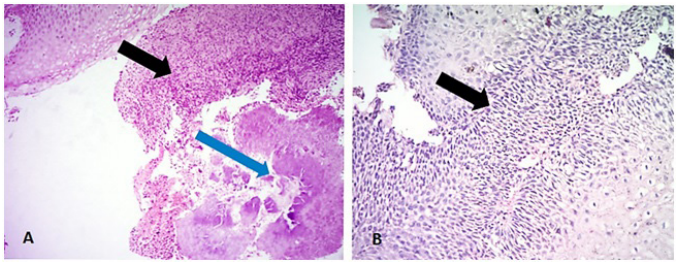

Figure 3(A): Histological examination of a Hematoxylin and Eosin (H&E) stained biopsy specimen (Before treatment) showing discrete sulphur granule (Blue arrow) within the granulation tissue (Black arrow) of an ulcerated oesophageal mucosa, consistent with infection by Actinomyces species (magnification×200). (B): Biopsy specimen after penicillin therapy showing chronic inflammation with acanthosis, basal hyperplasia and dysplasia with no evidence of Actinomyces (magnification×400).

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References

1. Chou F, Cheng K, Chiang I. Oesophageal actinomycosis. Advances in Therapy. 2006;23(4):623-626.
2. Kosseifi S, Dittus K, Nassour D, Shaikh M, Young M. Actinomycosis Esophagitis in a Patient with Persistent Dysphagia. Southern Medical Journal. 2005;98(6):662-664.
3. Lee SA, Palmer GW, Cooney EL. Oesophageal actinomycosis in a patient with AIDS. Yale J Biol Med. 2001;74(6):383-9.
4. Abdalla J, Myers J, Moorman J. Actinomycotic infection of the ooesophagus. Journal of Infection. 2005;51(2):E39-E43.
5. Arora A, Nord J, Olofinlade J, Javors B. Oesophageal Actinomycosis: A Case Report and Review of the Literature. Dysphagia. 2003;18(1):27-31.

Iron Deficiency Anemia Masking Adenomyoma of the Jejunum

Intestinal adenomyomas, also known as myoepithelialhamartomas, are benign non-neoplastic tumor-like lesions characterized by glandular structures lined by cuboidal or tall columnar epithelium and surrounded by smooth muscle bundles. Reported cases of adenomyoma of the gastrointestinal tract have been largely confined to the stomach and duodenum, with lesions distal to the duodenum occurring less frequently. Here we report a case of adenomyoma of the jejunum that presented with anemia, along with a review of the literature.

Case Report

A 62 year old woman with history of heartburn and cholecystectomy presented for bronchial thermoplasty for treatment of asthma. She was incidentally found to have iron deficiency anemia. Anemia was evaluated with upper and lower endoscopy, and findings were negative for suspicious pathology or source of bleeding. Capsule video endoscopy revealed a pedunculated 10 mm polyp in the distal jejunum/proximal ileum. Endoscopic removal of the polyp was completed by double balloon enteroscopy. Grossly, the lesion was polypoid shaped measuring 0.9x0.8x0.6 cm **(Figure 1)**. Microscopically, it was a well-circumscribed submucosal lesion composed of variably sized glandular structures surrounded by smooth muscle bundles. The glandular structures showed pyloric morphology and were lined by cuboidal or columnar epithelium with interspersed goblet cells and occasional Paneth cells **(Figure 2)**. Immune-stains were performed with appropriate controls. The lesional epithelial component was strongly and diffusely positive for CK7 and CK19 and weakly positive for CDX2, but negative for CK20 **(Figure 3)**. The smooth muscle component was positive for desmin and alpha-SMA. Ki-67/MIB-1, highlighting rare lymphocytes in the lesion and basal cells of the small intestinal mucosal epithelium **(Figure 4)**. All pathologic features supported a final diagnosis of adenomyoma. The patient's postoperative course was complicated by transient abdominal pain and bloating after the procedure. Abdominal X-ray was negative for free intra-abdominal air or perforation. Her pain resolved overnight and she was discharged the following morning. Upon follow-up one year later, she continues to do well, and the anemia has resolved.

Discussion

Myoepithelialhamartoma of the gastrointestinal tract was first described by Clarkein 1940.[5] While the incidence of small intestine adenomyoma is relatively rare, the actual incidence is unclear because very few cases