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SCIENTIFIC LETTER

Esophageal actinomycosis simulating circumferential neoplasia

Actinomicosis esófagica simulando una neoplasia circunferencial

Esophageal actinomycosis is an extremely rare condition, with very few cases published in the literature. The vast majority manifest as ulcers or erosions in the esophageal mucosa mimicking an esophageal tumour.[1]

We present the case of a 76-year-old woman with a history of antireflux surgery by Nissen fundoplication in 2014. She attended our clinic in September 2021 with a 15-day history of progressive dysphagia for solids and liquids, with no associated constitutional symptoms. Lab tests showed only elevation of C-reactive protein up to 50, with no other findings. Her physical examination was unremarkable.

An upper gastrointestinal endoscopy was requested and she was started on a puréed diet until investigations could be completed. Four days later, the patient went to Accident and Emergency because of oral intolerance. Upper gastrointestinal endoscopy showed some difficulty in passage at the level of the lower esophageal sphincter. In retroversion, it was possible to see the sutures from the earlier fundoplication with a slight foreign body reaction; the area was biopsied with results suggesting non-specific inflammation. There were no signs of lesion in the esophageal mucosa.

The patient was admitted to the hospital ward for investigation. A barium swallow was performed, demonstrating an image compatible with achalasia-pseudoachalasia. Tests were completed with a chest/abdomen computed tomography scan, which revealed circumferential thickening of the distal middle third of the esophagus with lymphadenopathy, suggesting malignancy as the top possibility. An endoscopic ultrasound was then performed in which it was not possible to access the stomach. Esophageal thickening was seen which included all layers, in addition to peri-esophageal fat and lymphadenopathy, raising suspicions of malignancy. The area of thickening was punctured, but the samples were insufficient. As we needed a histological sample, the endoscopic ultrasound was repeated. This time a 4 cm segment was described with normal stenotic mucosa with a 1 cm thickening with hypoechoic fraying towards mediastinal fat, with the muscularis propria appearing to be normal. There were several benign-looking enlarged lymph nodes in the vicinity. On this occasion, the endoscopist interpreted the image as thickening with inflammatory, benign characteristics. A biopsy was taken from the thickened area for cytology. The results came back negative for malignancy, with the finding of sulphur granules suggestive of the presence of actinomyces.

With this diagnosis, the immunosuppression study was completed with serology and detailed medical history but without any findings. The patient was started on treatment with intravenous penicillin G for three weeks. After that period, the patient made good progress, being able to ingest solids, and her C-reactive protein levels returned to normal. As the initial suspicions were highly compatible with malignancy, endoscopic ultrasound was repeated after the three weeks of intravenous antibiotic therapy. This time, it was possible to access the stomach with the ultrasound-guided endoscope, verifying a decrease in the thickening and a reduction in the size of the lymphadenopathy. The patient completed treatment with a six-month course of oral amoxicillin.

Actinomyces is a Gram-positive, facultatively anaerobic bacillus. It is a commensal microorganism of the flora of the mouth and gastrointestinal tract. They are slow-growing bacteria which can cause purulent abscesses in infected tissues. Esophageal involvement is usually preceded by a disruption of the mucosa which allows the entry of the bacteria. This disruption can occur after surgical or endoscopic mucosal trauma, even years after the event.[1]

Clinically it tends to present as odynophagia and dysphagia. It is slightly more common in males than in females. It has a higher prevalence in immunosuppressed patients, but cases have been reported in immunocompetent patients.[2] Endoscopically, esophagitis, ulcers and esophageal mass have been described.

This was a particularly interesting case. She was the first reported case of esophageal actinomycosis presenting with circumferential esophageal thickening without mucosal lesions. And ultimately, she was an immunocompetent patient, significantly reducing the degree of suspicion for the diagnosis.

Diagnosis is challenging, as the histological identification of the bacterium is only possible in very few patients. Microscopic examination is essential and typically shows necrosis and sulphur granules. Treatment consists of administering intravenous penicillin for two to four weeks, followed by oral amoxicillin for approximately six months.[3]

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Migratory intestinal stenosis by primary intestinal amyloidosis. A case report

Estenosis intestinal migratoria por amiloidosis intestinal primaria. A propósito de un caso

Amyloidosis is a disease characterized by the deposit of amyloid protein material in the organs causing their dysfunction. Different types of amyloidosis have been described according to the aetiology. These include primary amyloidosis (AL) and secondary amyloidosis (AA), among others. It can also be classified according to its location: systemic or localized amyloidosis. Localized amyloidosis most frequently affects the respiratory and lower urinary tract and subcutaneous tissue.[1] Isolated intestinal involvement is infrequent. In fact, in a meta-analysis published by Alsheri et al., there were only 14 cases identified that were published between 1960 and 2019.[2] This report presents the case of a patient with AL localized in the intestine with an atypical presentation based on migratory stenosis that responded to oral corticosteroid treatment.

A 72-year-old man with no toxic habits and no medical history was admitted because of 3 days of evolution of abdominal pain, vomiting and constipation. The patient had suffered 4 similar episodes in the previous two months.

Blood analysis showed an increase of acute phase reactants (15,000 leukocytes and 8 mg/dl C-reactive-protein). Abdominal computed tomography (CT) revealed a progressive calibre change in the right iliac fossa without identifying the cause and three segmental thickenings in the jejunum (Fig. 1a). An upper intestinal enteroscopy was performed, visualizing a stricture in the jejunum with prestenotic dilatation and two ulcerations with fibrin. Biopsies were taken (Fig. 1b). 72 h after the procedure, he presented clinical worsening so a CT-scan was repeated. Jejunal thickenings in different locations to those observed in the previous scans were seen (Fig. 1c). While waiting for the results of the biopsies, a systemic steroid treatment was started under inflammatory bowel disease suspicion. The patient evolved favourably, presenting both clinical and radiological resolution.

The histological study showed nodular deposits of amorphous eosinophilic hyaline material in the submucosa, Congo red positive and permanganate resistant, compatible with primary intestinal amyloidosis (Fig. 1d). The study was completed with bone marrow aspirate, a subcutaneous fat biopsy, a cardiac resonance and a urine study, ruling out systemic involvement.

The digestive tract is involved in up to 60% of AL cases. However, the isolated involvement of this organ is exceptional. The clinical presentation depends on the area, abdominal pain and intestinal obstruction in the small intestine (commonly in jejunum) and rectal bleeding when involving the colon (commonly in sigma).[2]

Localized AL has a low risk of progressing to a systemic disease and therefore has a better prognosis than systemic AL. Localized AL is often managed with local endoscopic and surgical treatment. A systemic treatment is usually avoided due to unfavourable risk/benefit.[3] In our case, local treatment was ruled out due to the location of the lesions and their migratory nature. After the clinical response to the steroid therapy, a progressive withdrawal of treatment was attempted. However, the patient presented recurrence of the suboclusive symptoms when the dose of steroids was reduced below 10 mg. It was decided that the treatment would be maintained chronically at the lowest effective dose.

An extensive review of literature found no cases of intestinal localized AL treated with steroids. However, in systemic AL, corticosteroids are used extensively with other chemotherapy agents such as bortezomib.[4]

In conclusion, intestinal localized AL is a rare entity. Systemic steroid therapy could be helpful in those cases where local treatment cannot be performed. Studies to confirm the hypothesis that our case suggests are needed.