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Idiopathic sclerosing mesenteritis in the pediatric patient: A case report[☆]

Mesenteritis esclerosante idiopática en la edad pediátrica: presentación de un caso clínico

Sclerosing mesenteritis makes up part of a spectrum of rare idiopathic conditions characterized by chronic inflammation of the mesentery and fibrosis.¹ It was first described in 1924.² Clinical presentation varies from an asymptomatic abdominal mass to acute systemic syndromes manifested as fever, weight loss, general malaise, bowel obstruction, or intestinal ischemia.

Tomography is a sensitive diagnostic tool, but surgical biopsy is usually necessary for making the diagnosis and ruling out other pathologies. Few patients require surgical treatment, which is generally directed at secondary complications such as bowel obstruction. The epidemiology of sclerosing mesenteritis is unknown. A prevalence of 0.6% has been described from more than 7,000 abdominal computed axial tomography scans.³ The disease is regarded as rare in the pediatric age and only 17 cases have been reported up to the present date.⁴ Even though there are reports on children under 3 years of age, the majority of patients are in the fifth to seventh decades of life.¹

We present the case herein of a 25-month-old male child with a 15-day progression of symptoms that began with abdominal bloating, soft stools, and pain, for which he was hospitalized in order to be tested. An abdominal computerized tomography scan was taken (Fig. 1) that showed important ascites. Paracentesis with fluid analysis was done to look for tuberculosis, but only one positive criterion was found: tuberculin 10 mm (the rest were negative), for which latent tuberculosis was diagnosed.

Given that there were no other paraclinical findings to explain the ascitic process, the patient underwent surgery that revealed abundant clear peritoneal fluid, marked distension of the intestinal segments, and whitish patches on the peritoneum from diaphragmatic recesses. Upper gastrointestinal endoscopy and total video colonoscopy were performed that showed grade III esophagitis, antral gastritis,

mild duodenitis, and nodular colitis. Biopsies demonstrated micro-abscesses at the distal duodenal level and special stains were negative.

Two weeks after surgery and with no clear diagnosis, the patient's symptoms of abdominal bloating and pain recurred. A control abdominal ultrasound was done that showed evidence again of free fluid in the cavity. A second surgery was decided upon and diagnostic laparoscopy was performed in which samples of the entire thickness of the intestine were taken; this had not been possible in the first surgery because the distension of the segments had not allowed for adequate visualization. Surgery revealed critical stricture of the ileum with severe hardening in almost its entirety. Samples were taken for histopathology and the macroscopic results were: the serous membrane of the distal ileum presented with thickened fibrous areas like a «breastplate», compressing it.

Microscopically: the mucosa, submucosa, and muscle were normal; the serous membrane had a thickened fibrous band that entirely covered the distal ileum resulting in complete stricture and compression. Diagnosis in the distal ileum was idiopathic sclerosing mesenteritis (Figs. 2 and 3).

Systemic steroid therapy was begun, along with isoniazid for 9 months for treating the latent tuberculosis infection. Due to the severity of the clinical symptoms, cyclophosphamide was added. Once treatment was started, the patient progressed adequately, he tolerated oral intake, there were no febrile peaks, and the ascites and distension were resolved.

Given the atypical nature of the clinical symptoms and the immediate improvement, it was decided to continue the steroids for 6 complete weeks, without prolonging cyclophosphamide administration.

It has been suggested in the literature that sclerosing mesenteritis is a rare entity in children and adolescents due to the lower quantity of mesenteric fat,⁵ and even though the precise etiology is still unexplained, mechanisms including previous abdominal trauma or surgery, autoimmunity, infection, and ischemia have been proposed.⁶ A reasonable hypothesis is that this subgroup of patients has abnormal responses in the cicatrization and repair of connective tissue. This inflammatory and fibrotic process can affect the integrity of the gastrointestinal lumen and the mesenteric vessels through a mass effect, causing a variety of gastrointestinal and systemic manifestations that include abdominal pain, nausea and vomiting, diarrhea, weight loss, and fever. Therefore a high degree of suspicion is necessary for making the diagnosis.²

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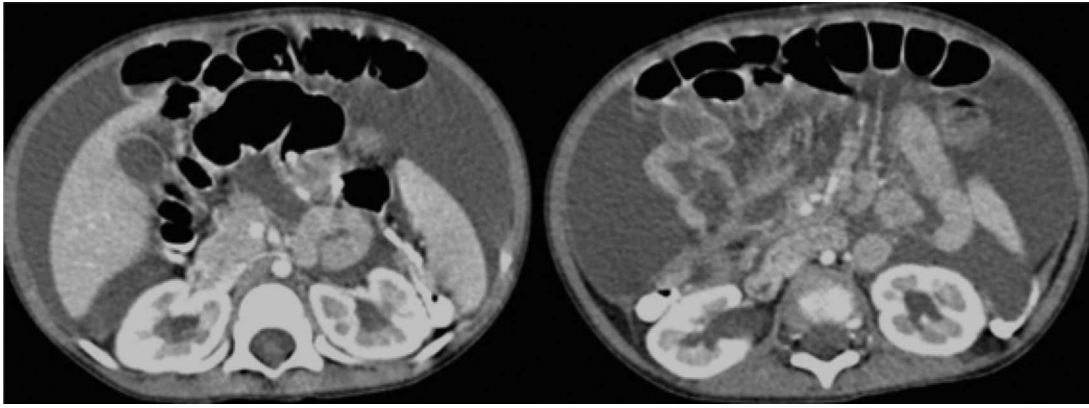


Figure 1 Abdominal tomography scan: A great quantity of ascites of undetermined cause; the liver, pancreas, and kidneys have a normal aspect and size; there is no primary tumor or metastasis and no adenomegaly or masses. The vascular structures have normal caliber and there are no lesions in the digestive tract.

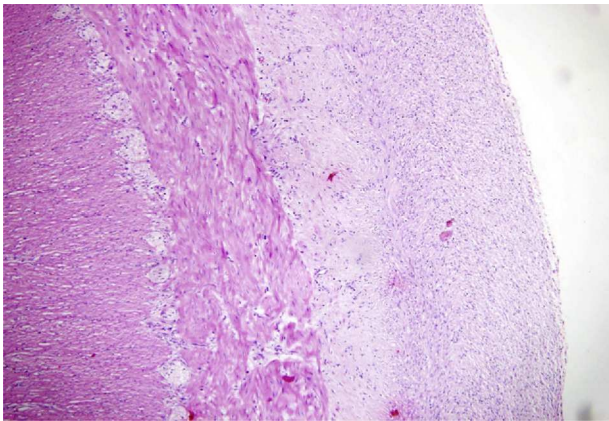


Figure 2 Histopathology, hematoxylin and eosin stain: serous membrane of the intestine with a fibrous band obliterating the lumen.

Diagnosis is established through histologic evaluation; plain x-ray is non-specific. Ultrasound imaging can aid in better visualizing the mesenteric fat. Computerized tomography scan is the most sensitive image for detection.

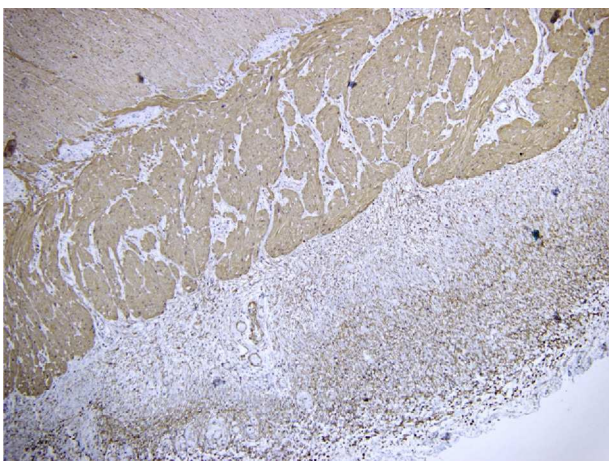


Figure 3 Histopathology, immunohistochemistry: a more evident fibrous band is observed that is strictured and completely compresses the lumen of the distal ileum.

Calcifications are present in approximately 20% of the lesions as a result of fat necrosis or vascular obstruction.^{7,8}

Pathologic confirmation should be obtained in all cases, as was the case with our patient. It was the basis for establishing the definitive diagnosis.

There is no standard therapy and treatment should be individualized. In the large majority of cases, complete surgical resection is impossible due to the associated vascular involvement and disease extension.

A number of pharmaceutical agents have been used in an attempt to stabilize the disease. Glucocorticoids, azathioprine, methotrexate, colchicine, cyclophosphamide, and/or radiotherapy have been proposed as therapeutic options.^{1,9,10} The patients with a greater inflammatory component, with fever, weight loss, and general malaise appear to be the most receptive to glucocorticoids, alone or in combination. In the case of our patient, the use of the steroid and cyclophosphamide had favorable results in disease improvement and modulation.

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Conflict of interest

The authors declare that there is no conflict of interest.

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Capsule endoscope aspiration: A common complication?[☆]

Aspiración de cápsula endoscópica: ¿complicación frecuente?

Obscure gastrointestinal bleeding (OGIB) is defined as occult bleeding or it manifests as bleeding of unknown origin after negative endoscopy and colonoscopy. OGIB can present as occult bleeding characterized by anemia, iron deficiency and/or fecal occult blood, or it can manifest as hematochezia or melena, with no evidence of the bleeding site through conventional studies. Approximately 5% of gastrointestinal bleeding occurs between the Treitz ligament and the ileocecal valve.¹ Diagnosis and treatment of patients with OGIB are often long and difficult processes.² Capsule endoscopy (CE) has been shown to be a cornerstone in evaluating the patient with OGIB. The benefits of CE include the fact that it is a painless, invasive method that is widely accepted by the patient for studying the small bowel (SB).¹ All the patient has to do is swallow the capsule endoscope. Despite its being a minimally invasive method, several complications must be taken into account, such as retention of the capsule endoscope (0.7%), impaction in the cricopharyngeus or diverticula, and aspiration into the respiratory tract.³⁻⁸ Only a few cases of capsule endoscope aspiration have been reported worldwide⁶⁻⁸ and none in Mexico. Therefore, we believe it is of interest to report the present case, so that the physicians carrying out this type of study are aware of this possible complication.

A 78-year-old man had no past history of chronic degenerative diseases and no complaints of dysphagia or swallowing disorders. He was hospitalized 3 years prior due to upper gastrointestinal bleeding from a gastric ulcer secondary to NSAID ingestion for which he received satisfactory combined endoscopic treatment. He continued with occasional NSAID ingestion. One year ago the patient began to present

with postprandial fullness, a weight loss of 14 kg during that year, and fecal occult blood on 2 occasions. Panendoscopy revealed a hiatal hernia and chronic gastropathy of the body and antrum. Colonoscopy showed uncomplicated diverticular disease of the descending colon and the sigmoid colon. The patient was referred for CE as part of the approach to OGIB. A bowel transit time test was done on the patient before he was given the capsule endoscope to swallow, and it was normal. There were no complications in relation to the preparation for CE.

The patient was programmed for the CE study with the modified PillCam COLON 1 technique for studying the SB. The patient had some difficulty in swallowing the capsule endoscope, but he was able to do so. Immediately after swallowing it he had the sensation of a foreign body in his throat and a coughing spell of 2 min. He did not complain of dyspnea or any other symptom and physical examination showed no cyanosis. He drank water again and had the sensation of having swallowed the capsule. However, one minute later his cough was exacerbated and he expelled the capsule into his mouth. He was given water, he swallowed the capsule with no incident, and the study was completed with no apparent complication. The images were downloaded and when the study was examined, it was seen that the capsule had been aspirated, taking photographs of the respiratory tract (figs. 1 and 2); the capsule was then expelled into the mouth and swallowed again, evaluating the usual trajectory in its entirety. The capsule was passed 8 hours later and the cause of the bleeding was identified.

CE has currently become the method of choice for studying the patient with OGIB. It has been approved in the United States by the Food and Drug Administration for the study of these patients, and it is also indicated in the study of Crohn's disease and polyps, among others.⁹

The present case showed the aspiration of a capsule endoscope and the successful and non-invasive recovery of the patient. The first reported case of aspiration required surgical extraction,⁸ in another published case extraction was carried out through rigid bronchoscopy, using a foreign body forceps and basket,⁷ and in another, the patient did not require any invasive recovery procedure,⁶ as with our case. Our patient did not present with the classic symptoms of acute aspiration or any symptom of respiratory distress.

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