

Ulcerated Meckel’s diverticulum

Divertículo de Meckel que aparenta una ileítis de Crohn

Sr. Director:

Meckel’s diverticulum (MD) is a true intestinal diverticulum containing all layers of the small intestine, resulting from the failure of the vitelline duct to obliterate. MD occurs so infrequently that most articles have reported either small series or isolated cases.¹ Crohn’s disease (CD) is a disorder of uncertain etiology characterized by ulceration and transmural inflammation of the gastrointestinal tract. We present a case in which acid-secreting MD mimic Crohn’s ileitis.

Case report

A 21-year-old male presented a 4-day history of gastrointestinal bleeding (passage of bloody stool). He had no significant previous medical history, was on no regular medication, had no allergies known and had no family history of gastrointestinal pathology. He denied abdominal pain, episodes of diarrhea or use of non-steroidal anti-inflammatory drugs.

Physical examination revealed paleness of the skin and mucosae and sinus tachycardia with normal blood pressure. Rectal examination revealed red blood in stools.

Laboratory data showed severe anemia with hemoglobin level of 3.8 g/dl. Platelet count, coagulation parameters and inflammatory markers were normal. Nasogastric tube insertion showed a clear aspirate. He required transfusion of 5 units of red blood cells and hemoglobin level raised to 7.7 g/dl.

An oesophagogastroduodenoscopy (OGD) was performed, but it was negative. A colonoscopy was also performed and revealed about ten ulcers (<20 mm) in the terminal ileum, with red blood in the ileum coming proximally, and digested blood in the colon. Biopsies of the ileum and colon were undertaken.

Although hemodynamically stable, he kept bleeding intermittently and had to receive 3 more units of red cells. He underwent a small bowel capsule endoscopy, although this procedure was incomplete, fresh blood was seen in the distal ileum, without identification of any lesion. At this time, a single-balloon enteroscopy (SBE) by anal route was

proposed to clarify these findings. It revealed some ulcers in the distal ileum, one of those serpiginous (Fig. 1), with apparent hematin pigmentation and no fresh blood was seen in the lumen. The preliminary diagnosis of regional enteritis (Crohn’s disease) was assumed.

However, the patient presented a massive recurrent bleeding within 24 h and was decided to perform another SBE. It revealed large amounts of fresh blood not allowing the procedure. The patient became hypovolemic and was submitted to an intraoperative enteroscopy.

A large Meckel’s diverticulum was recognized 80 cm from the ileocaecal valve and intraoperative enteroscopy (Fig. 2a) identified a peri-diverticulum ulcer with visible vessel (Fig. 2b), (Anexo-video). An enterectomy with resection of the diverticulum was carried out (Fig. 2c). Pathology confirmed the presence of a Meckel’s diverticulum with focal ectopic gastric mucosa.

The patient did well postoperatively, with no recurrent episodes of bleeding, and was discharged seven days after surgery.

Biopsies at first colonoscopy revealed non-specific ileitis. He has since remained asymptomatic.

Discussion

Congenital gastrointestinal malformations make up to 6% of all congenital anomalies and of these MD is the most common.² MD was described in detail by Hohann Friedrich Meckel in 1808, and bears his name.³

Anatomically MD is a true intestinal diverticulum containing all layers of the small intestine, resulting from the failure of the vitelline duct to obliterate in the first trimester of gestation, in the antimesenteric border of the ileum.² MD is usually asymptomatic, only about 2–4% develop a complication over the course of their life. The most common complications are hemorrhage, small bowel obstruction, and diverticulitis, which occur more frequently in children rather than in adults. Sixty percent of those who become symptomatic are under 10 years old.⁴

Hemorrhage is reported in the majority of symptomatic cases of MD in children (over 50% of cases) but is unusual in adult patients (only 11.8%).⁵ As we show in our case report, bleeding is usually painless and can be massive and fast, presenting fresh red blood in stool. Furthermore, the patient can develop hypovolemic shock.

Heterotopic tissue is present in approximately 50% of resected diverticula, with gastric mucosa being the most

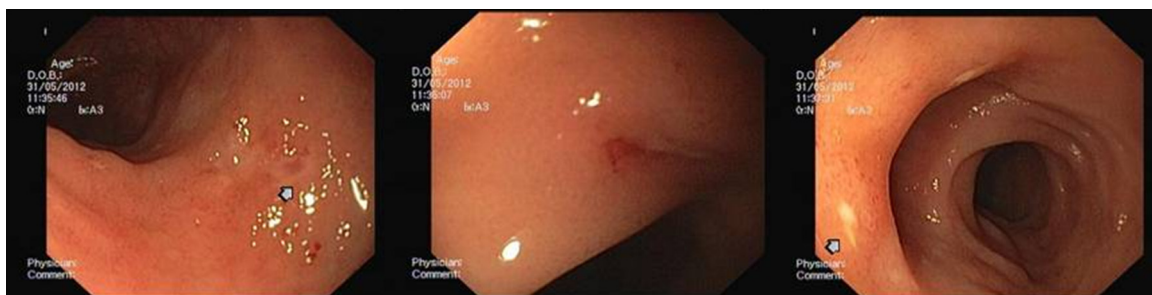


Figure 1 (see arrows) Ulcers in SBE (single-balloon enteroscopy).

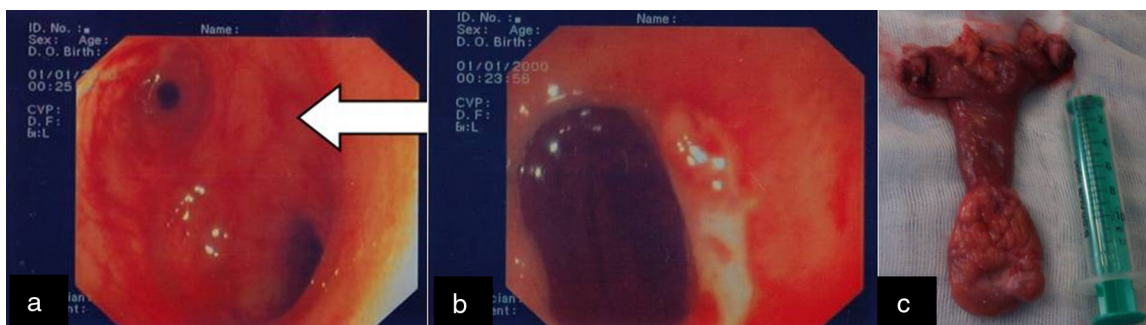


Figure 2 (a) Intraoperative enteroscopy – Meckel's diverticulum; (b) intraoperative enteroscopy – ulcerated Meckel's diverticulum with visible vessel; (c) Meckel's diverticulum – surgical specimen.

common tissue type found (23–50% of cases). The main mechanism of bleeding is the acid secretion from heterotopic gastric mucosae, which leads to ulceration of adjacent ileal mucosa in 62–100% of cases. However ulceration can be seen in cases without gastric heterotopic tissue, and may be explained by either ischemia and/or trauma.⁵ In our case, the pathology showed gastric mucosa in the MD and our patient presented ulceration of ileal mucosa adjacent to the diverticulum with visible vessel resulting in massive bleeding culminating in hypovolemic shock, being this presentation unusual in this age group.

The traditional diagnostic test of choice for MD has been scintigraphy using ⁹⁹Tc^m pertechnetate with an accuracy of 90% in children. However, in adults the accuracy is only 46%.⁴

In our case report the presence of multiple ulcers, some of those serpiginous, in the terminal ileum and the fact that single-balloon enteroscopy did not detect MD, conducted to a false hypothesis of inflammatory bowel disease (IBD). Inaccurate diagnoses with single-balloon enteroscopy may result from a small opening of the diverticulum and/or the absence of active bleeding during the procedure or massive active bleeding that influences the endoscopic view.⁶

Meckel's ileitis is an uncommon manifestation of MD.⁷ Levesque et al.⁸ reported in 2011 a case of a 60-year-old man who presented brisk obscure gastrointestinal bleeding and ileal ulcerations on capsule endoscopy, but no pathologic evidence of IBD and who was diagnosed with CD. After an extensive revision of the case a laparoscopy was performed and MD was resected, with resolution of the symptoms. The authors suggest that "Meckel's ileitis" is an alternative diagnosis to CD in patients with gastrointestinal bleeding, ulcerations in capsule endoscopy but no pathologic evidence of CD.⁸ As far as we know few cases^{8–10} have been reported, in adults, in which ulcerated MD mimic Crohn's ileitis. Preoperative detection of symptomatic MD requires a high index suspicion, and arrangement of the diagnostic tests.¹

The treatment of symptomatic MD is surgical resection. In cases where an inflammatory or ischemic process involves the adjacent ileum, intestinal resection with the diverticulum and anastomosis may be necessary.² In our case the presence of ileal ulcerations, led to an enterectomy with diverticulectomy, without complications.

In conclusion, the commonest cause of ulceration in the terminal ileum is IBD, however as we demonstrated the rare condition of "Meckel's ileitis" can be misdiagnosed as CD in adults. In cases of isolated small bowel ulceration

alternative etiologies to CD should be carefully investigated since IBD treatments carry significant costs and side effects for patients.⁸ As this case report shows despite being the most prevalent congenital anomaly of the gastrointestinal tract, MD continues to be a diagnostic challenge. Therefore, it is important for clinicians to keep this entity in mind when looking at cases of gastrointestinal bleeding even in adults.

Conflict of interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.gastrohep.2013.07.011>.

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Stanozolol-induced bland cholestasis

Colestasis canalicular inducida por estanozolol

Sr. Director:

It is difficult to estimate the prevalence of toxic hepatitis, especially by anabolic-steroids, because frequently they are taken without medical prescription. The use of anabolic-steroids by sportsmen¹ and teenagers has dramatically increased, raising the question about their adverse effects, especially hepatotoxicity. The hepatotoxic effects include cholestasis,² hepatocellular carcinoma,³ nodular regenerative hyperplasia and variceal bleeding, secondary to portal hypertension presumably due to nodular regenerative hyperplasia.⁴ Stanozolol is a 17 α -alkyl anabolic-androgenic steroid, which is used in therapeutic doses for some medical indications such as hereditary angioedema⁵ or aplastic anemia,⁶ but its use is extended among sportsmen and body-builders. The effect of this drug is dose-dependent, although it is influenced by individual susceptibility and the presence of other toxic habits, such as alcohol abuse.⁷

We report the case of a 37-year-old European Caucasian man, who was admitted to our hospital after developing acute severe jaundice and itching, but without fever or chill. Furthermore, he reported passing dark urine simultaneously. He did not have problems such as abdominal pain, nausea or vomiting. There was no history of pre-existing liver disease. Furthermore, he denied unsafe sexual practices, drug abuse or other toxic habits (except smoking twenty cigarettes a day). However, he ingested a protein-enriched diet for two years to increase the muscle mass. The patient did not take other medications.

On admission, physical examination revealed jaundice. The biochemical test showed serum levels of bilirubin of 19.16 mg/dL (normally <1) (direct fraction 15.84 mg/dL), with aspartate aminotransferase (AST) 45 U/L (normally 5–37), alanine aminotransferase (ALT) 58 U/L (normally 5–41), alkaline phosphatase (AP) 152 U/L (normally 40–129) and gamma-glutamyl-transpeptidase (GGT) 19 U/L (normally 10–66). Other biochemical parameters such as creatinine, C reactive protein, sodium and potassium remained normal. Hemoglobin, leucocytes, platelet count and prothrombine time were normal as well. The presence of viral infection (hepatitis A, hepatitis B, hepatitis C, cytomegalovirus, Epstein–Barr virus and HIV) and autoantibodies (including anti-mitochondrial antibody, anti-smooth muscle

antibody, liver kidney microsomal type 1 antibody and antinuclear antibodies) was excluded. An ultrasound scan of the abdomen was performed, showing a normal volume of the liver and no evidence of biliary dilation. During admission, he admitted to have self-administered high doses of stanozolol (Winstrol®) by injections (intramuscularly, three times a week) for three weeks prior to the onset of symptoms. After that, we thought of a toxic hepatitis so we used the CIOMS scale resulting in 9 points, supporting our impression. Thus, we decided not to perform a liver biopsy. Accordingly, during admission the patient was provided with supportive medical treatment and showed a good progress.

Eight weeks after discontinuation of stanozolol, biochemical tests gradually improved, itching disappeared and he was completely asymptomatic. Finally, in three months, all tests were normal. Therefore, clinical signs and laboratory findings improved substantially in following weeks after discontinuation of stanozolol.

Discussion

Our patient developed severe cholestatic jaundice with a slight elevation of liver enzymes and itching, after self-administration of stanozolol injections. The patient mentioned that he went to the gym and ingested a protein-enriched diet to increase the muscular strength but, at the beginning, he did not recognize to take steroids, which delayed the diagnosis. The temporal relationship between the administration and the appearance of symptoms, and the return to normal values after drug withdrawal, clearly suggest the association. CIOMS scale is validated to find out the relationship between drugs and toxic hepatitis, being highly probable values over 8 points⁸ (our patient scored 9 points). CIOMS scale has the following elements: type of liver injury, time of onset of the event, time from drug intake, until reaction onset, time from drug withdrawal until reaction onset, risk factors, and course of reaction. Finally, we considered that the liver biopsy was not necessary due to three reasons: (a) the temporal relationship; (b) the CIOMS scale score; and (c) exclusion of other causes.

Bland cholestasis is almost always associated with the use of 17 α -alkyl anabolic-androgenic steroids. The course of illness is marked by an insidious onset of itching followed by dark urine and jaundice, with minimal serum enzyme elevations or evidence of hepatocellular necrosis (ALT levels are usually <200 U/L, AP <230 U/L). Typically, bland cholestasis shows a slow recovery (usually, more than 4 weeks).