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CASE REPORT

Jejunal diverticulosis: A rare cause of massive bleeding

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Abstract

INTRODUCTION: Jejunal diverticulosis is a rare diagnosis that occurs mainly in old age, more often in men than in women. It is usually an incidental diagnosis of unclear aethtiology. In some cases, visceral myopathy can also be the cause. It is most often manifested by abdominal pain and bleeding. Bleeding from the small intestinal diverticula represents only 0.6-5% of all small intestinal bleeding.
CASE REPORT: The authors describe the case of a 66-year-old man with massive gastrointestinal bleeding who did not respond to conservative hemostyptic treatment. Following negative gastrofibroscopic and colonoscopic examinations, an angioCT examination was indicated, which revealed a source of bleeding in the jejunal diverticula. The patient was indicated for surgical treatment. The extent of bleeding was determined by perioperative enteroscopy and subsequently, the affected jejunal segment was segmentally resected with a primary anastomosis.
CONCLUSION: Bleeding from the jejunal diverticula is a very rare diagnosis, which poses challenges in the diagnostic process in particular. Capsule enteroscopy plays an important role in the diagnosis, as well as CT angiography and scintigraphy in the event of massive bleeding. In addition to conservative treatment, the embolization of a bleeding vessel may subsequently be used in therapy. In indicated cases, surgical resection treatment is also possible.

INTRODUCTION

Diverticulosis is a typical diagnosis for the colon, the incidence of which increases with age. Diverticula in the jejunum and ileum are a rare, usually incidental diagnosis. Their incidence represents 0.9-1% of all gastrointestinal diverticula (Romera-Barba *et al.* 2017). In contrast X-ray examinations, they were described in 0.5-2.3% of small bowel imaging examinations for other causes and in 0.06-4.5% of autopsy findings (Patel *et al.* 2008). Jejunoileal diverticula are more common in men, while duodenal diverticula are the same in both sexes. Most cases of duodenal diverticula are observed in patients > 50 years of age, while jejunoileal ones occur in patients aged 60-70 years (Akhrass *et al.* 1997). The incidence of small intestinal diverticula at a younger

age is typical of congenital syndromes *(Ehlers-Danlos, Marphan)* and visceral myopathy (McLean *et al.* 1985).

Small intestinal diverticula occur most often in the jejunum (80%), less frequently in the ileum (15%), or throughout the small intestine (5%). They are usually multiple and range in size from just a few millimetres to 10 cm in diameter. The small intestinal diverticula do not have a muscle layer and are usually located on the mesenteric margin. Meckel's diverticulum, on the contrary, is located on the antimesenteric margin (Choi *et al.* 2013; Mansoori *et al.* 2016).

The clinical manifestations of jejunoileal diverticula are very insignificant and non-specific. They include chronic abdominal pain, dyspepsia, nausea, vomiting, borborygmi and altered intestinal habits (Alves *et al.* 2018; Ceuppens *et al.* 2018). However, they are much more often manifested by complications such as diverticulitis, massive bleeding, intestinal obstruction, perforation, and the multiplication of anaerobic bacteria in a stagnant content (Kouraklis *et al.* 2002; Alves *et al.* 2018; Rangan *et al.* 2020).

Small bowel bleeding is a rare pathology that accounts for 5-10% of gastrointestinal bleeding, with only 0.6% to 5% of cases due to the presence of small bowel diverticula (Cellier 2008; Blake-Siemsen *et al.* 2017). Bleeding from small intestinal diverticula is more typical of duodenal diverticula, where it manifests itself as hematemesis and melena. Bleeding from jejunoileal diverticula is most often manifested by hematochezia (Rangan *et al.* 2020).

MATERIAL AND METHODS

On December 23, 2018, a 66-year-old patient with chest pain was brought to the Emergency Department at Martin University Hospital.

Due to a suspicion of STEMI, he was given 200 mg of acetylsalicylic acid (ASA), 180 mg of ticagrelor and 5,000 units of unfractionated heparin (UFH) during transport to the hospital. Only insignificant changes not typical of the acute coronary syndrome were present in the initial ECG; laboratory values of troponin T and CK-MB were in physiological intervals. The patient was admitted to the coronary unit and indicated for an acute coronary angiography, which did not confirm the presence of the acute coronary syndrome.

In a laboratory examination following the coronary angiography, the patient had severe anaemia. When being questioned about his medical history, the patient mentioned the presence of a bloody stool two days earlier. During the rectal examination, the patient had enterorrhagic stool. The patient with bleeding from the GIT of unclear aetiology, with severe anaemia, was subsequently transferred to the ICU of the surgical clinic.

At the ICU, the patient had another enterorrhagia with a volume of 800 ml. The patient received hemostyptic therapy (etamsylate, 4-aminomethylbenzoic acid), vitamin K, proton pump inhibitors by continuous infusion, and 2TU of red blood cells. Due to the recent administration of acetylsalicylic acid, the patient was given a platelet concentrate and an urgent gastrofibroscopic examination was performed, which did not find any source of bleeding in the upper gastrointestinal tract. A colonoscopy examination revealed enterorrhea without finding a source; the patient had diverticulosis without bleeding in the entire colon. Despite treatment, the bleeding was ongoing (the patient had already received 5 TU of red blood cells), the anaemia worsened (control haemoglobin decreased to 60g/l), and therefore urgent an angioCT examination was indicated. A contrast leak was present in the jejunal loop on CT examination (Fig. 1).

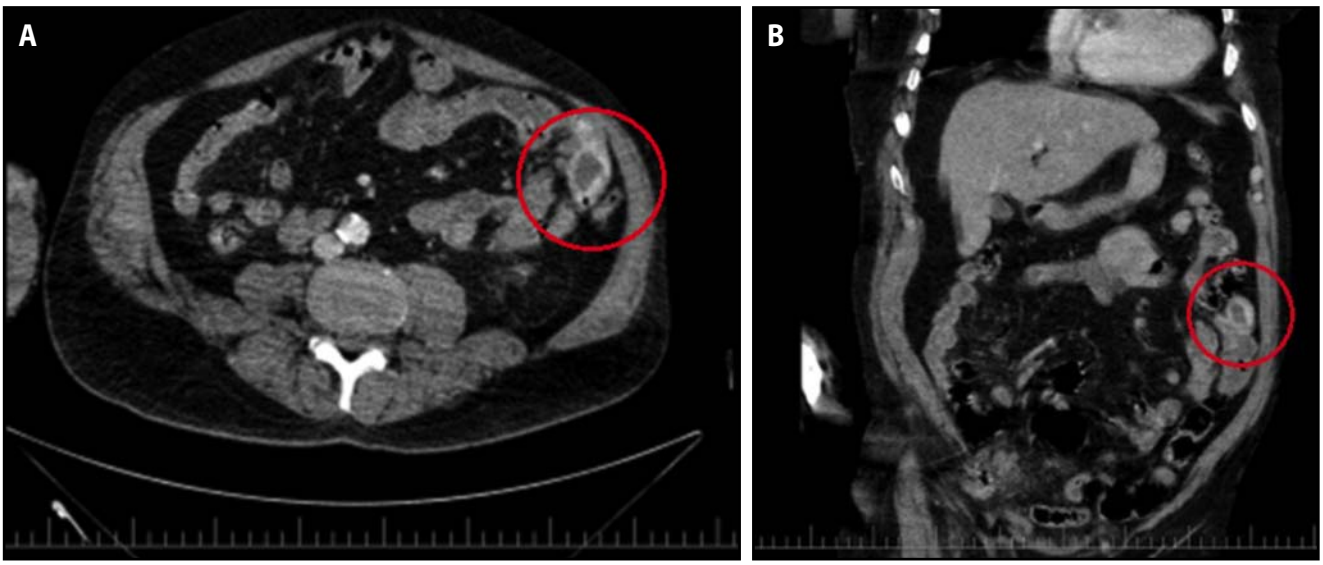

Fig. 1. CT angiography, leak of the contrast from the jejunal region in the red circle:
a) axial projection,
b) sagittal projection

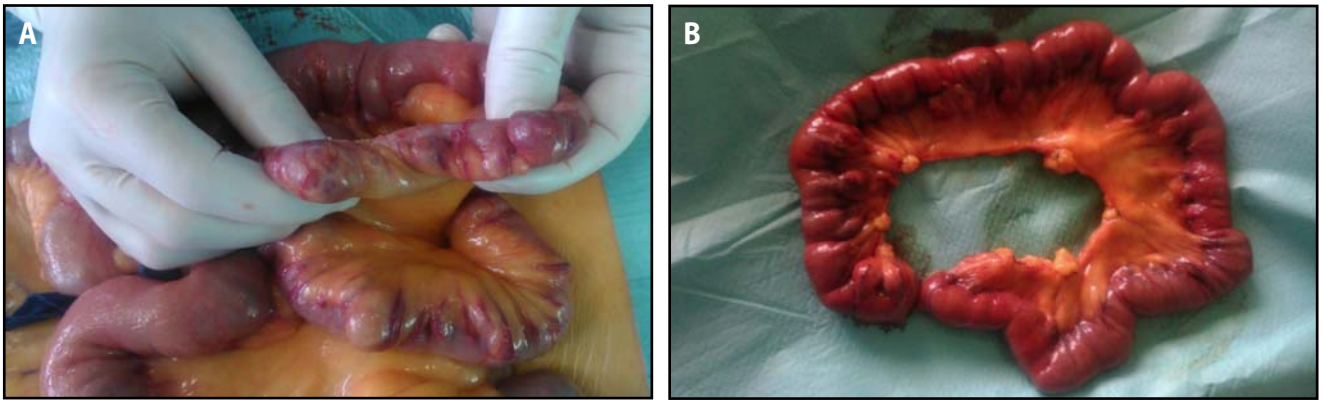

Fig. 2. Jejunal diverticula:
a) perioperative finding,
b) resection of 70 cm of the jejunum

Since conservative therapy did not stop the bleeding, urgent surgical treatment was indicated. During surgical revision, we found diverticulosis of the entire colon and multiple Graser diverticula of the jejunum on the mesenteric side, starting around 40 cm behind the Treitz ligament and continuing for a total length of 70 cm (Fig. 2A). Some diverticula were up to 3 cm in diameter. The part of the intestine orally above the diverticula was intraluminally without the presence of blood. We indicated intraoperative enteroscopy and the segmental resection of the jejunum along a total length of 70 cm in the range of the presented bleeding diverticula (Fig. 2B). After the transection of the intestine with a linear cutter, we restored intestinal continuity with a latero-lateral anastomosis of the small intestine.

RESULTS

The operation was performed without complications. The abdominal drain was removed on the first postoperative day. From the second postoperative day, the patient began to drink fluids, followed by a slurry diet on the fourth postoperative day and a solid diet on the fifth, which the patient tolerated well. On the sixth day, the patient was discharged home in good condition. The haemoglobin at discharge was 112 g/l.

The histological examination confirmed multiple right small intestinal diverticula with predominantly completely absent small intestinal muscle. The wall of the diverticula consists only of the mucosa and connective tissue of the submucosal tissue; in the vicinity of most diverticula, there are only residues of significantly fibrotised smooth muscle (Fig. 3).

DISCUSSION

Small bowel diverticulosis is most frequently an acquired disease, but cases of the familial occurrence of massive jejunal diverticulosis have also been published in academic literature (Andersen *et al.* 1988; Koch *et al.* 2007). The involvement of both layers of muscularis propria is typical of rare familial visceral

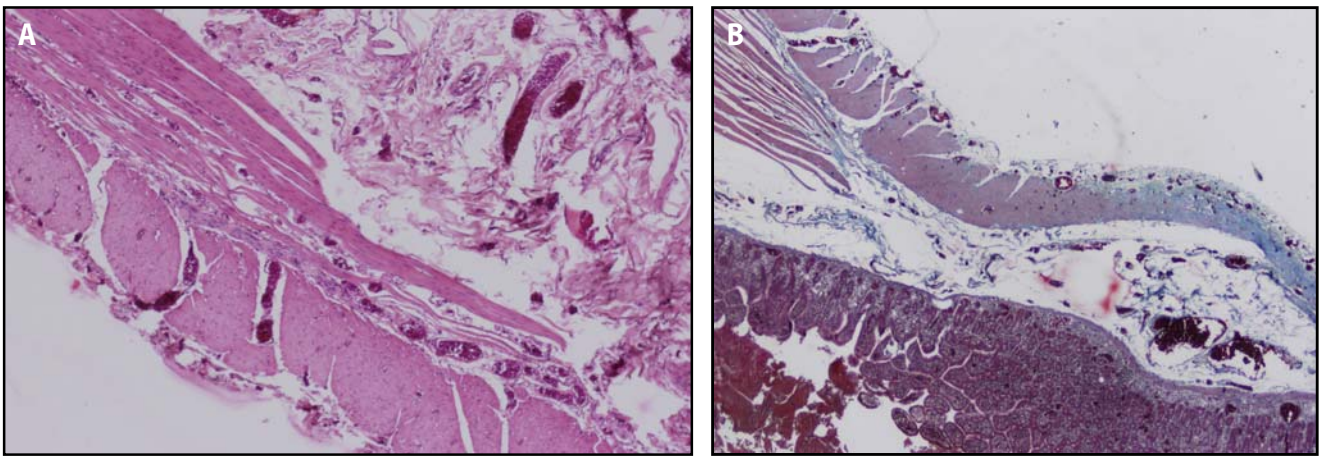

Fig. 3. Focal reduction to complete absence of the circular layer of small intestinal muscle in the diverticulum region:
a) hematoxylin-eosin staining,
b) trichrome staining

myopathy with autosomal recessive transmission. In addition to small intestinal diverticula, the dilatation of the stomach and small intestine dominates the clinical picture and the patient experiences severe abdominal pain (Anuras *et al.* 1983; Alstead et al 1988).

Jejunal diverticulosis can also be associated with several diseases such as scleroderma, celiac disease, Fabry disease, Cronkhite-Canada syndrome (Zuber-Jerger *et al.* 2008) or systemic connective tissue disorder Coffin-Lowry syndrome (Machin *et al.* 1987). The pathogenesis of these lesions is multifactorial. The site of herniation appears to occur at the points where blood vessels penetrate the mesentery into the bowel wall (Longo *et al.* 1992). Also, abnormal contractions of the intestinal wall muscle (Maull *et al.* 1981), neuronal and axonal degeneration and neuronal intranuclear inclusions, findings consistent with a visceral neuropathy can play a role in pathogenesis (Krishnamurthy *et al.* 1983; Martínez-García *et al.* 2001).

Clinical manifestations of small intestinal diverticula are usually absent. However, if they do occur, the most common are abdominal pain (49%) and bleeding (29%) (Chiu *et al.* 2000). Rare but serious complications of diverticula include diverticulitis, perforation, volvulus, bacterial overgrowth, intussusception, jejunocolic fistula, and massive bleeding (Vilallonga *et al.* 2012).

In the case of bleeding from the small intestine, finding the source of the bleeding is the most problematic issue. Following a negative gastrofibroscopy and colonoscopy, the small intestine is the presumed source of bleeding. For chronic slow bleeding, capsule enteroscopy is the method of choice, with a diagnostic yield ranging from 59.4 to 66.9% (Katsinelos *et al.* 2014). However, the disadvantage is the length of the examination, since the passage of the capsule through the digestive tract takes several hours, which is unacceptable in the event of acute bleeding with a repeated need for blood transfusions. Capsule retention in the diverticulum may also occur with the need for surgery (Committee *et al.* 2013). In acute bleeding with hemodynamic instability, the method of choice is imaging. Current options for small bowel imaging in patients with small bowel bleeding include a multiphase CT scan (CT angiography (CTA) and enterography (CTE)), MR enterography (MRE), 99mTc radioisotope bleeding scans, mesenteric angiogram, and Meckel's scan. Multiphase CT scanning is the recommended first line imaging to evaluate detected small bowel bleeding (ASGE Standards of Practice Committee *et al.* 2017). CTA is most sensitive when patients are hemodynamically unstable or have a transfusion requirement of 5 or more units of blood (Abbas *et al.* 2005). 99mTc-bleeding scan and angiography can detect haemorrhage when the bleeding rate is in the range of 0.1 mL / min and 0.5 to 1.0 mL / min respectively (Mantas *et al.* 2011).

In the event of the failure of conservative and interventional radiology treatment, surgical treatment is indicated as the last option. This may include intraoperative enteroscopy or resection. The yield of intraoperative enteroscopy is 58-88% (Kuo *et al.* 2019) and the rate of bleeding recurrence after such treatment can range from 13 to 52% (Voron *et al.* 2017). Both laparoscopic (Ertem *et al.* 2010) and open procedures can be used in surgical treatment. The most common type of operation is the segmental resection of the bleeding section of the small intestine with a primary anastomosis (Vilallonga *et al.* 2012; Mazahreh *et al.* 2019). During resection, only the necessary part of the small intestine needs to be removed to prevent short bowel syndrome.

CONCLUSION

Jejunal diverticulosis is a rare finding in the small intestine and is usually asymptomatic. Abdominal pain and bleeding are its most common manifestations. Capsule enteroscopy can be used in the diagnosis of jejunal diverticula haemorrhage, with CT angiography or scintigraphy being the first choice for massive haemorrhage. A conservative approach is initially applied in therapy. If it fails and massive bleeding continues, it is possible to stop the bleeding by embolizing the bleeding vessel. In the case of bleeding from multiple sources or the unavailability of an interventional radiology method, surgical treatment with a laparoscopic or open approach is indicated. The operation involves a segmental resection of the bleeding section with the preservation of as large a section of the small intestine as possible to prevent short bowel syndrome.

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