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Ileal angiodysplasia presentation as a bowel obstruction: A case report

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ABSTRACT

INTRODUCTION: Angiodysplasia is a common vascular abnormality of the gastrointestinal tract, found in the elderly and most frequently revealed by gastrointestinal bleeding. We report an original case of ileal angiodysplasia in an 83-year-old woman presenting as a bowel obstruction.
CASE PRESENTATION: An 83-year-old woman with a medical history of chronic untreated anemia, presented with cardinal symptoms of bowel obstruction. Computed tomography revealed diffuse ileal wall thickening with multiple zones of stenosis, which were aggravated by an ileal perforation and associated with vascular abnormalities compatible with angiodysplasia. Surgery confirmed the imaging findings. A large resection importing one meter of ileum was performed. The pathology report of the resected specimen revealed ischemic lesions of ileum associated with ileal angiodysplasia. The postoperative period was marked by an acute dehydration in the patient who died 3 weeks after surgery.
DISCUSSION: Angiodysplastic lesions develop with aging due to chronic low-grade intermittent obstruction of submucosal veins. These lesions are the result of increased contractility at the level of muscularis propria, leading to congestion of the capillaries and failure of pre-capillary sphincters, resulting in the formation of small arteriovenous collaterals. The acquired arteriovenous malformation consisting of multiple shunts with rapid blood flow may result in inadequate oxygenation of a segment of the intestine and lead to ischemia and eventually wall thickening, stenosis and even perforation of the small bowel.
CONCLUSION: Angiodysplasia should be kept in the back of one's mind as one of the causes of acute abdomen and bowel obstruction, especially in elderly people suffering from occult gastrointestinal bleeding.

1. Introduction

Angiodysplasia is the most common vascular abnormality of the gastrointestinal tract. The lesions can involve any segment of the intestinal tract, but they are found most frequently in the cecum and ascending colon [1,2]. Angiodysplasia is usually asymptomatic and is often diagnosed in over 60-years patients. However, the lesions may sometimes result in severe bleeding [2,3]. We report here an original case of ileal angiodysplasia revealed by an acute abdomen due to a bowel obstruction in an 83-year-old woman admitted to our academic referral hospital.

2. Case presentation

An 83-year-old woman came walking to our Emergency Department for treatment of acute abdomen due to intestinal obstruction. She presented with a twelve-hour history of abdominal pain and distension with vomiting. Her bowel habit was altered but there is no history of rectal bleeding or melena. She denied any loss of weight or appetite. Her medical history consisted of a chronic and untreated anemia, and she had no past surgical history. Her vital signs upon admission were stable; her abdomen was tender and distended. Bowel sound was sluggish and rectal examination revealed an empty rectum with no palpable mass. Her hernia orifices were free. Her blood investigation results were normal apart from an anemia with a hemoglobin rate of 9.5 g/dl and a C-reactive protein elevation at 25 mg/dl. A supine abdominal x-ray showed dilated loops of small bowel. A clinical diagnosis of small bowel obstruction was then made. A computed tomography imaging was performed, revealing diffuse, circumferential and homogeneous mural thickening of ileum with a "double halo sign" and alternating stenosis and mild dilated zones. Several infarcted small bowel loops with pneumatosis and extraluminal gas bubbles indicating perforation were also found (Fig. 1A). A vascular engorgement was seen in the regional mesentery with a mild amount of free peritoneal fluid. Further findings at computed tomography examination included vascular abnormalities consisting of a dilated and tortuous ileocolic vein with an accumulation of ectasic vessels within the submu-

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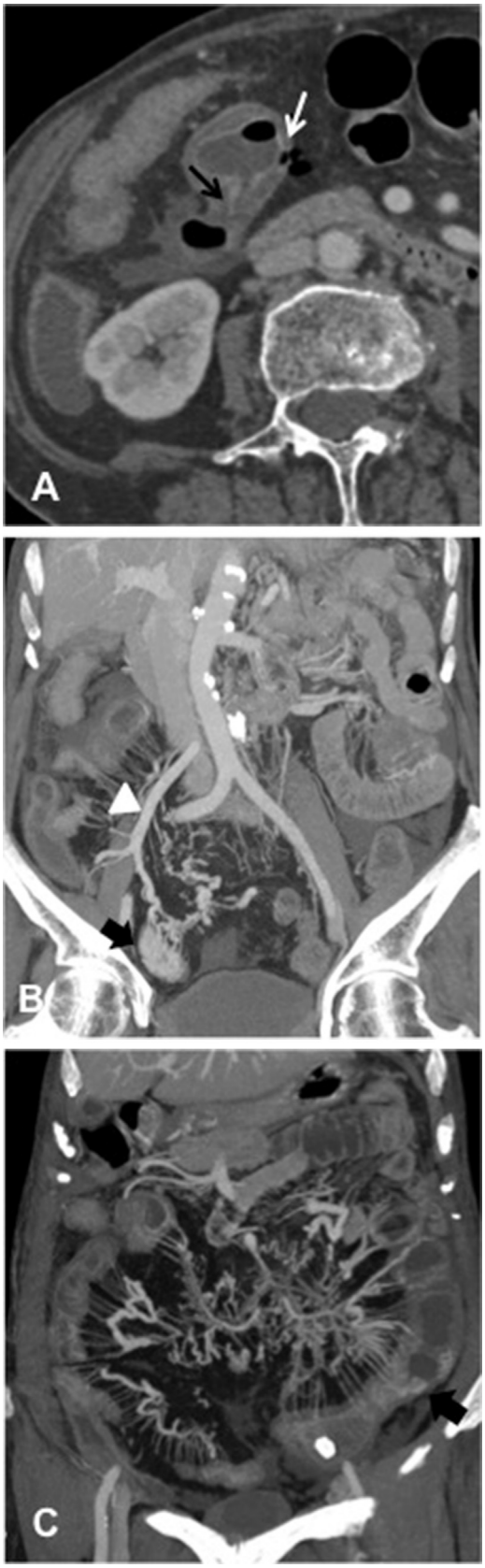

Fig. 1. Enhanced abdominal computed tomography in axial and coronal plans showing: (A) circumferential wall thickness of ileum with a target appearance due to submucosal edema and an ileal perforation (white arrow) upstream of a wall stenosis (black arrow); (B, C) dilated ileocolic vein (white arrow) with accumulation of ectasic veins within the submucosa of right and left ileal loops (black arrows).

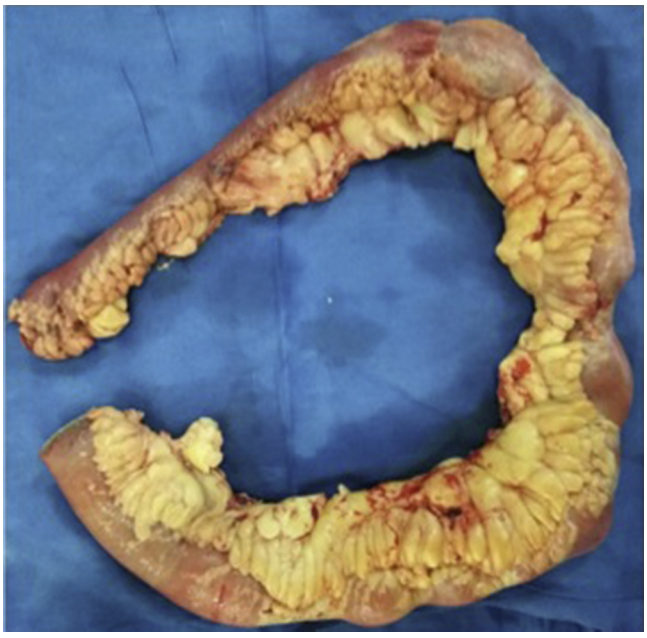

Fig. 2. Ileal resected specimen showing alternating zones of stenosis and mild dilatation. The vascular abnormalities cannot be seen because of their submucosal localization.

cosa of the distal ileum (Fig. 1B and C). The diagnosis of ileal angiodysplasia associated with ileal ischemic lesions was then made. An exploratory laparotomy was subsequently performed on the patient. The results showed a diffuse ileal wall thickness with segmental and multifocal ileal wall stenosis zones alternating with dilated zones (Fig. 2). An ileal perforation was found 50 centimeters far from the ileocecal junction. No evidence of a malignant mass was found. A large ileal resection importing one meter of ileum and a double ileostomy were then performed. The resected ileum was examined histologically. Ischemic changes of ileum were found (Fig. 3A). They consisted of multiple mucosal ulcerations covered by pseudo-membranes with segmental wall thickness and stenosis. The serosa was hyperemic, marked by petechial hemorrhage, edema with congestive capillaries and inflammatory cell infiltration. The inflammatory cell population was mainly composed of lymphocytes, plasma cells and neutrophil leucocytes. The mucosa between ulcers revealed chronic regenerative changes with cuboidal epithelial cells and inhomogeneous distorted crypts. No malignant cells were found. Further findings on histologic examination included vascular lesions characterized by the presence of dilated and tortuous vascular veins and veinules in the submucosa of the ileum (Fig. 3B). These findings were compatible with ileal angiodyslasia associated with ischemic lesions of ileum. After surgery, the patient was taken to the intensive care unit and she was closely monitored. She was put on broad spectrum antibiotics and intravenous fluid replacement. The patient remained stable during ten days after surgery. Then, her postoperative course was marked by an acute dehydration. The physical examination showed low blood pressure, rapid pulse rate and low urine output. Laboratory tests showed increasing serum urea, sodium and potassium which reached 6.5 mmol/L. Unfortunately, the patient died 3 weeks after surgery.

3. Discussion

Angiodysplasia is the most common vascular abnormality of the gastrointestinal tract, found mainly in patients older than sixty years without gender predilection [1,2]. The lesions are frequently multiple and can involve any segment of the GI tract; but they are

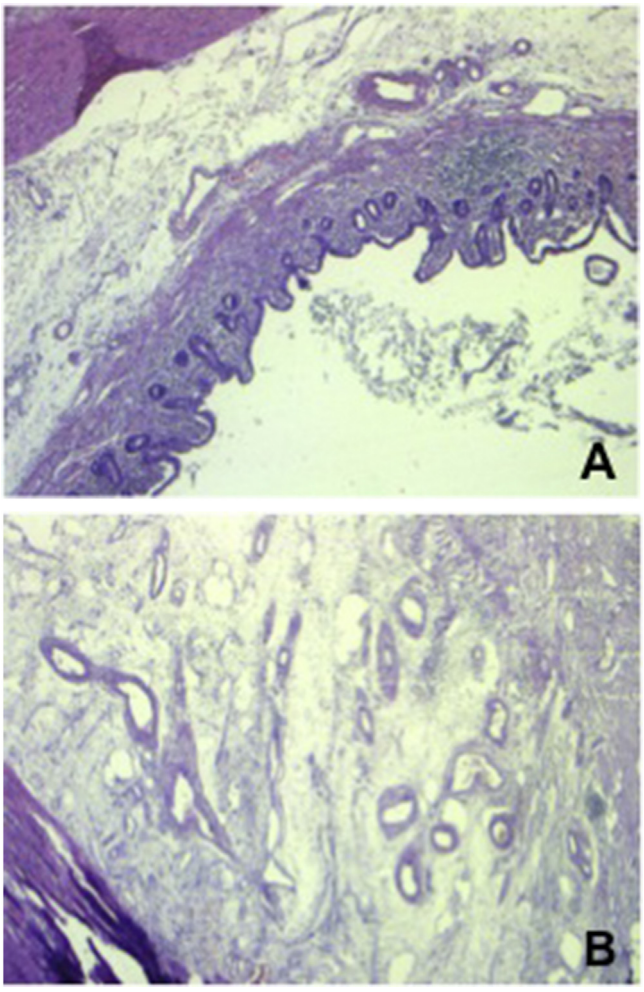

Fig. 3. Microscopic analysis of ileal resected specimen showing ishemic lesions of ileal mucosa (A) (HE, original magnification ×25) and cluster of dilated veins and veinules in ileal submucosa (B) (HE, original magnification ×25).

most commonly found in the cecum and ascending colon, while 15% of them are located in the small bowel [4,5]. Angiodysplasia has varied clinical expressions. It is usually asymptomatic, but may sometimes be a source of significant morbidity from bleeding [1]. Small bowel angiodysplasia accounts for 30–40% of cases of GI bleeding of obscure origin [3]. In the current report, our patient, with a medical history of chronic anemia, presented with an acute abdomen due to a bowel obstruction. To the best of our knowledge, this report describes the first case of an elderly person whose bowel obstruction was the presenting symptom of angiodysplasia and not intestinal bleeding. Only two other reports have described similar presentations in children including ileal and cecal stenosis [6], and intestinal perforation [7], both presenting in infancy. Even the cause of angiodysplasia is unknown, the disease is probably acquired, resulting from a degenerative process of previously healthy vessels, associated with aging and do not occur in association with other vascular malformations [1,8]. Angiodysplasia has been purported to occur with higher frequency in patients with renal failure, Von Willebrand's disease, aortic stenosis, cardiovascular and pulmonary disease [1,8,9].

In the case of our patient, CT angiography has showed vascular abnormalities, compatible with ileal angiodysplasia. Besides, CT and laparotomy have also shown ileal wall thickness with alternating dilated and stenosis zones, aggravated by ileal perforation and explaining the clinical presentation of bowel obstruction and acute abdomen. Computed tomographic angiography (CTA) has become a sensitive, specific and minimally invasive tool for the diagnosis of angiodysplasia, particularly in the ileum because of the limitations of the endoscopy [2]. The other advantage of CT is related to its capability to accurately evaluate additional extra-luminal abnormalities such as intestinal parietal thickening. In patients with angiodysplasia, intraoperative exploration usually produces little information because of the mucosal and submucoal localization of the lesions [10]. Surgery is curative. However, it is only carried out on symptomatic patients usually suffering from acute, severe or chronic bleeding not controlled by other alternatives [1,10]. Histopathological examination of the resected specimen provided definite evidence of angiodysplastic lesions which were associated with histologic features of ileal ischemia.

Unlike malignant diseases such as lymphoma or inflammatory ones such as Crohn's disease, angiodysplasia is not considered as a common cause of diffuse thickened ileal wall or a common cause of small bowel ischemia, stenosis and perforation [11]. Understanding the pathogenesis of angiodysplasia can help to explain such unusual findings. The etiology and mechanism for the development of angiodysplasia are not fully understood. The most widely quoted hypothesis suggests that angiodysplastic lesions develop with aging due to chronic low-grade intermittent obstruction of submucosal veins as a result of increased contractility at the level of muscularis propria. This leads to congestion of the capillaries and failure of pre-capillary sphincters, resulting in the formation of small arteriovenous collaterals composed of a dilated arteriolar-capillary-venular unit [1,8]. This is consistent with the computed tomographic angiography appearance, namely that of a normal arterial phase, the appearance of a "vascular tuft" in the capillary phase and early filling and late emptying thereafter of large veins [8]. The acquired arteriovenous malformation consisting of multiple shunts with rapid blood flow may result in inadequate oxygenation of a segment of intestine, explaining in our case, chronic ischemia, wall thickness, stenosis and perforation of the small bowel.

4. Conclusion

Rare cases similar to the one we investigated should highlight the importance, for practitioners, of becoming more knowledgeable about unusual clinical, radiological and operative angiodysplastic features. Clinicians need to have suspicion of angiodysplasia in their diagnostic workup when dealing with an acute abdomen due to a bowel obstruction, especially in elderly patients suffering from occult gastrointestinal bleeding.

Conflicts of interest

No conflicts of interest are to be stated.

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Ethical approval

This work does not require a deliberation by the ethics committee.

Consent

Written informed consent was obtained from the hospital for publication of this case report and any accompanying images. Every effort has been made to protect the patient's identity and there is no

reason to believe that our patient, if she had been still alive, would have objected to publication.

Authors' contributions

Ghdes O is the first author of this article; Gaja A and Mnif N performed radiological diagnosis and contributed to preparing the figures; Jarraya H performed clinical treatment including surgical operation and Blel A performed histologic diagnosis. Mnif N performed critical revision of the manuscript for important intellectual content. All the authors have read and approved the final manuscript.

Guarantors

Ons Ghdes and Ali Gaja accept full responsibility for the elaboration of this work and the control of the decision to publish it.

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The study complies with the SCARE Statement: Consensus-based surgical case report guidelines [12].

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