

Management of Intestinal Angiodysplasia and Cataclysmic Hemorrhage

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ABSTRACT

Intestinal angiodysplasia is a common cause for gastrointestinal bleeding. Often, the diagnosis and management of these patients can be challenging. We aim to describe the clinical aspect, imagistic diagnosis and therapeutic options of angiodysplasia. We report the case of a 56 years old woman with multiple severe comorbidities diagnosed with angiodysplasia in an acute setting. The main clinical manifestation was massive gastrointestinal hemorrhage, in the context of disseminated intravascular coagulation. Despite the embolization of the first jejunal artery the hemorrhage continued. The patient underwent right hemicolectomy with ileal resection and intraoperative enteroscopy with good recovery. At one year follow up there were no signs of bleeding. Multiple therapeutic procedures may be necessary in order to control bleeding from angiodysplasia lesions.

Key words: chronic hepatitis HCV, angiodysplasia, gastrointestinal hemorrhage, gastrointestinal bleeding, intraoperative enteroscopy

CASE REPORT

A 56 years old woman was admitted to our clinic for severe fatigue and dizziness, with sudden onset a week before admission. The patient had a history of chronic hepatitis HCV without antiviral treatment, arterial hypertension stage III, high additional risk, type 2 diabetes mellitus insulin dependent on a basal-bolus regimen, autoimmune thyroiditis with hypothyroidism and stage 3 chronic kidney disease (GFR of 43 ml/min/1.73 m²). Her medication included levothyroxine, angiotensin receptor blocker, calcium receptor blocker, alpha receptor blocker, loop diuretic (furosemide), potassium sparing diuretic (spironolactone) and allopurinol. One month before the current admission she presented

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for recurrent episodes of epistaxis and an anemic syndrome. The blood count revealed microcytic hypochromic anemia, remitted after the administration of RBC. On that occasion we performed upper and lower GI endoscopy, revealing no signs of bleeding. Before the current admission she did not report any signs of bleeding. On admission the patient was pale, the BP was 140/90 mmHg, the pulse 50 bpm, the respiratory frequency 20 breaths/minute, the temperature 37 degrees C and the oxygen saturation 96% in room air. She had normal pulmonary and heart sounds, the abdomen was slightly tender in the upper right quadrant, without signs of peritoneal irritation; the remainder of the examination was normal.

The blood tests revealed severe anemia (hemoglobin levels of 5.4 g/dl with hypochromia and microcytosis), normal leukocyte and thrombocyte levels, hyperglycemia (267 mg/dl) with HbA1c 9%, elevated serum creatinine level (2.43 mg/dl, with a GRF of 21 ml/min/1.73 m²), minimal liver cytolysis (ALT, AST two times normal values), elevated fibrinogen level, normal coagulation parameters, increased TSH (72.5 µUI/ml).

The insulin and levothyroxine doses were augmented. On the second day of hospitalization she developed fever (40°C), productive cough, severe dyspnea, with a sudden decrease in O₂ saturation up to 75%, bradycardia (40 bpm), hypotension (SBP 70 mmHg). The desaturation was corrected by non-invasive administration of oxygen (4 l/min) and empiric antibiotherapy was initiated. ECG showed sinus bradycardia without other modifications. Chest

X-ray was normal. Echocardiography showed an ejection fraction of 55%, without left ventricle dyskinesia or signs of pulmonary hypertension. The microscopic examination of the sputum revealed in diplo coci. The patient's status continued to deteriorate; she presented anuria, leukocytosis (38000/mm³), high levels of blood lactate (up to 10 mmol/l) and signs of DIC (decreased thrombocyte and fibrinogen levels, increased coagulation parameters, with and INR of 1.43, positive tests for fibrin degradation products and d-dimers). Also, there were signs of liver failure - cytolysis (ALT, AST ten times normal value, total bilirubin 3 mg/dl). She was transferred to the intensive care unit and hemodialysis by femoral catheter was initiated.

During her stay in the ICU the respiratory symptoms and the fever diminished under treatment. Thrombocytopenia and increased coagulation times persisted. On the 3rd day of ICU she began to present melena and hematochesia- up to 10 stools/day. Her evolution did not improve under administration of PPC, RBC. Upper GI endoscopy was repeated but it did not show any signs of bleeding. Despite the altered coagulation parameters, we decided to perform an intestinal angiography in order to establish the location of the bleeding and try to resolve it by embolization. The approach was via the left femoral artery- the Seldinger technique, with a 5F introducer sheath. Celiac artery catheterization showed no signs of active bleeding. Superior mesenteric artery catheterization revealed active bleeding from the first jejunal artery. This was catheterized selectively with a Cantata 2.5F



Figure 1 - Angiographic aspect of intestinal angiodysplasia showing the vascular tuft and the positioning of the microcoil



Figure 2 - Angiographic aspect after embolization

microcatheter (*fig. 1*) and two Tornado coils, one of 4 cm/3 mm and one of 5cm/2mm were placed (*fig. 2*). Angiographic control proved the success of the procedure. The patient developed left inguinal hematoma and required the placement of a hemostasis device at this level. Despite the apparent success of the procedure, the patient continued to present melena and hematochesia. A colonoscopy was performed revealing a high amount of fresh blood and blood clots in the cecum, with normal aspect of the colon. Abdominal CT scan showed the extravasation and accumulation of i.v. contrast in the cecum, suggesting active bleeding.

The patient was transferred to the General Surgery Department and a right hemicolectomy with ileal resection was performed. Intra operative enteroscopy did not reveal any other lesions beyond the resection limits. The post-operative evolution was favorable, without signs of GI bleeding; the patient resumed normal diuresis and the renal function was stabilized. Hemoglobin levels were corrected by administration of RBC and remained constant. Biopsies taken from the resection piece revealed typical signs of angiodysplasia (*fig. 3 a, b*). She was discharged three weeks after the intervention. At one month and 6 month evaluation she presented constant levels of hemoglobin, around 11 mg/dl (mild anemia due to the chronic kidney disease).

DISCUSSION

Angiodysplasia is a common vascular malformation of the GI tract (1). It accounts for 3% to 6% of all lower GI bleeding and 2% to 5% of acute upper GI bleeding. One recent study found a prevalence of asymptomatic angiosyplasia in 9,8% of patients undergoing upper endoscopy (2). However, angiodysplasia is the most frequent cause of bleeding from the jejunum an ileum.

Intestinal angiodysplasia can be associated to chronic kidney disease (1). It has also been described in association with aortic stenosis, chronic obstructive pulmonary disease, venous thromembolism and heart failure (3). In patients with chronic kidney disease, intestinal angiodysplasia appears more frequently in patients with advanced age, arterial hypertension, overall high comorbidities and dialysis (4). Our patient had a history of chronic kidney disease and arterial hypertension, which may have contributed to the development of the lesions. However, the most important aspect and the particularity of this case lies within the acute and life threatening situation in which the angiodysplasia was manifested. It is debatable whether or not the disease would have remained asymptomatic had it not been for the combination of metabolic disorders (uncontrolled diabetes mellitus leading to high susceptibility for infections and poor prognosis, severe hypothyroidism leading to cardiac dysfunction) and sepsis leading to DIC and the entire panel of coagulation disorders that accompany it.

Endoscopic procedures are effective in an acute setting in order to control the hemorrhage but do not prevent long term bleeding, possibly due to lesions located elsewhere (5). Small bowel lesions of angiodysplasia can be difficult to find and treat. Lesions can be identified by capsule endoscopy, CT enterography or CT angiography, but these investigations have no therapeutic potential. Furthermore, in young patients with small bowel bleeding, videocapsule endoscopy may not accurately diagnose bleeding lesions, especially if they present active hemorrhage (6,7). Risk factors for rebleeding after a negative videocapsule exam include overt bleeding and use of non-steroidal anti-inflammatories (8). A recent retrospective study found that isolated gastrointestinal angiodysplasias of the small bowel have been outcomes and lower

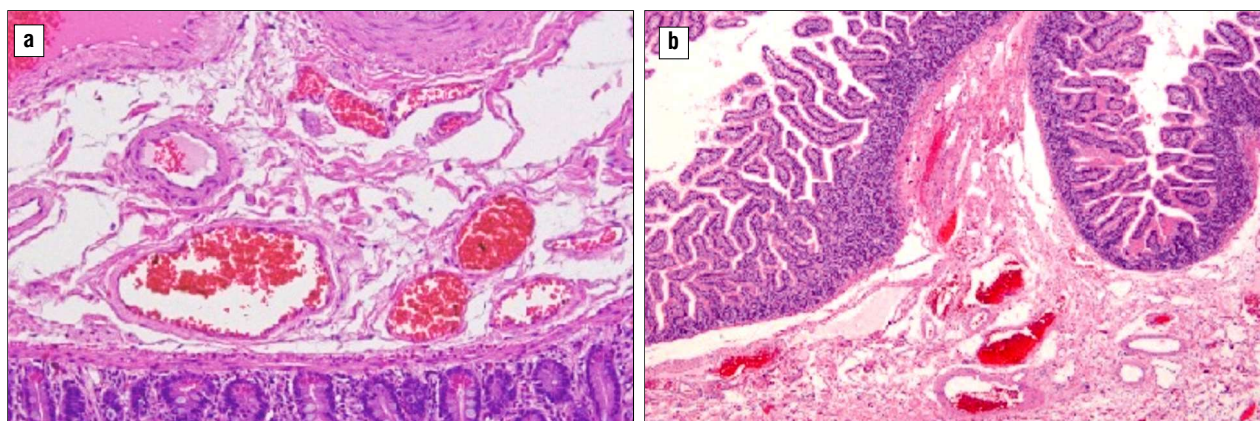


Figure 3 - (a) Optic microscopy (20x) aspect of colonic angiodysplasia: large distorted blood vessel in the submucosa. (b) Optic microscopy (4x) aspect of small bowel angiodysplasia: dilated tortuous vein in the submucosa

rebleeding rates than patients who associate lesions in other parts of the digestive tract (9). On the other hand, efforts are made to provide severity scores in patients with endoscopic diagnosis, taking into account the number, location and probability of bleeding (10).

Several hemostatic methods can be performed, depending on the localization of the bleeding: endoscopy and argon plasma coagulation, hemocliping or band ligation, or selective embolization by angiography (11). Intestinal angiography has both diagnostic and treatment advantages. It requires no special preparations and it can accurately detect bleedings as low as 0.5ml/min (12). Surgical resection of the affected bowel is indicated as a rescue option, when the lesions are too extensive or the angiographic treatment has failed (13). Future therapies for angiodysplasia may be based on angiogenesis inhibition, as studies have shown altered expressions of specific cytokines and chemical mediators (TNF- α , Ang-1, Ang-2) in intestinal bleedings (14).

CONCLUSION

Angiographic approach to angiodysplasia is a specific and sensitive means of diagnosis and treatment. Although there may be occasional complications, embolization is relatively safe and efficient in the management of intestinal angiodysplasia. Surgical resection of the affected segments of the GI tract is an important step in the treatment of angiodysplasia. However, due to the multiple localizations of the lesions, it can be ineffective if not associated with other interventions.

Conflict of interest

All author declare that they have no conflict of interest.

Ethical statement

Institutional ethical clearance was obtained prior to the study.

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