# Colon bleeding from both arteriovenous malformation and inflammatory bowel disease. Case report

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SUMMARY: Colon bleeding from both arteriovenous malformation and inflammatory bowel disease. Case report.

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Introduction. Although endoscopy and angiography have changed the management of lower gastrointestinal bleeding and the majority of patients respond to conservative treatment 10-20% of cases have no recognizable site of hemorrhage. About 10-30% of all patients will require operative intervation. A very rare case of massive lower gastrointestinal bleeding in a young patient who was found to suffer from two causes of gastrointestinal hemorrhage in the same time is reported. The patient had to undergo surgery for the control of bleeding.

Case report. A 23 years old male Greek patient presented to the emergency department of our hospital because of three episodes of hematochezia during the last 10 hours. He was admitted to the surgical department for monitoring of his condition. In the next 10 hours the hematochezia continued and the patient although being transfused with three units of packed red blood cells, started to become unstable with his vital signs affected, having also a syncoptic episode. Emergent colonoscopy could not recognize the site of hemorrhage or any other pathology in the colon, but revealed an intestinal lumen full of blood from the anus to the cecum. It was decided that the patient should undergo operation to stop bleeding. An extensive right hemicolectomy was performed. After that the patient remained stable and showed no signs of hemorrage. The histopathological examination of the specimen showed an arteriovenous malformation but also lesions of the mucosa compatible with early inflammatory bowel disease.

Conclusions. In young patients with massive lower gastrointestinal bleeding of unknown origin, extensive right hemicolectomy provides a good and safe therapeutic choice that will control hemorrhage in most cases with the advantage of lower mortality and morbidity rates compared to subtotal colectomy. Close monitoring of the patient postoperatively is essential. RIASSUNTO: Trattamento chirurgico di emorragie massive del colon di origine sconosciuta. Caso clinico

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Introduzione. L'endoscopia e l'angiografia hanno cambiato il trattamento delle emorragie gastrointestinali inferiori e la maggioranza dei pazienti risponde bene al trattamento conservativo, ma circa il 10-30% dovrà sotroporsi ad un intervento chirurgico. Nel 10-20% dei casi il sito dell' emorragia non sarà riconosciuto. Presentiamo un raro caso di emorragia massiva gastrointestinale inferiore in un paziente giovane che nello stesso tempo soffriva di due cause responsabili di emorragia gastrointestinale. Il paziente veniva sottoposto a laparotomia per controllare l'emorragia.

Caso clinico. Paziente di 23 anni, si presenta al reparto di chirurgia d'urgenza con tre episodi di ematohezia nelle ultime dieci ore. Nelle successive ore la perdita di sangue continua e il paziente diventa emodinamicamente instabile anche se trasfuso. Un esame colonscopico d'urgenza veniva effetuato senza poter riconoscere il sito di emorragia; c'era però la presenza di sangue in tutto il lume intestinale dal retto al cieco. Il paziente veniva sottoposto a laparotomia, con emicolectomia destra allargata. Dopo l'intervento il paziente era stabile senza ulteriori segni di emorragia. L'esame anatomopatologico del pezzo operatorio evidenziava malformazioni arterovenose ma anche lesioni della mucosa intestinale compatibili con malattia infiammatoria cronica intestinale.

Conclusioni. In pazienti giovani con emorragie massive intestinali inferiori, la emicolectomia destra potrebbe essere una scelta di terapia abbastanza sicura con il vantaggio di minore morbidità e mortalità rispetto alla colectomia subtotale.

KEY WORDS: Colon bleeding - Arteriovenosus malformation - Inflammatory bowel disease. Emorragia del colon - Malformazioni arterovenose - Malattia inflammatoria cronica intestinale.

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## Introduction

Acute lower gastrointestinal (GI) hemorrhage accounts 20% of all cases of GI bleeding. In the last four decades there has been great progress in diagnostic methods for locating the precise bleeding point, with colonoscopy and mesenteric angiography being the most important of these methods (3). Despite the improvement in diagnostic imaging and procedures, a 10-20% of patients with lower GI bleeding have no demonstrable bleeding source (3). Arteriovenous malformation (AVM) of gastrointestinal track is one of the significant causes of lower gastrointestinal bleeding, along with diverticulosis, neoplasms, inflammatoty bowel disease (IBD), anorectal disorders etc. In young patients IBD, AVM, Meckel's diverticulum and polyposis are the most common causes of such symptoms (1,3). It is very rare that two of these conditions co-exist in the same patient. No matter what is the cause of lower gastrointestinal bleeding about 80% of patients will stop bleeding spontaneously (2,10). Those who don't or those who rebleed are at particularly high risk to suffer an inhospital complication or require surgical control of their hemorrhage. We present a very rare case of a young patient with hematochezia who was found to suffer from two causes of lower GI bleeding in the same time. Specifically the histopathological examination of the specimen showed an AVM and also early IBD in a young patient with massive lower GI bleeding. The patient had to undergo extensive right hemicolectomy in order to stop bleeding. Right hemicolectomy with close monitoring of the patient is a different therapeutic approach to treat young patients with massive lower GI bleeding of unknown origin.

### **Case report**

A 23 years old male Greek patient presents to the emergency department of our hospital complaining about three episodes of hematochezia during the last ten hours. He has no other symptoms and he claims that this is the first episode.

From his medical history he has no chronic disease and takes no medication.

Eight months before he had been hospitalized to the pneumonology department because of an episode of hemoptysis that was considered to have happened due to a respiratory infection. The patient was in good condition, hemodinamically stable with good vital signs (blood pressure:128/86 mm Hg, heart rate: 78 beats per minute). His temperature was 36.8 C. From the clinical examination his abdominal wall was painless during palpation and he had normal bowel sounds. Inspection of oropharynx and nasopharynx reveals no blood. Inspection of the anus shows no signs of hemorrhoids or other anorectal disease. Digital rectal examination shows bright red colored stools. A complete blood count, a chemistry panel and a coagulation test were performed. All laboratory tests were normal except from a slight decrease on the hematocrite that was found to be 39,5% with a hemoglobin level of 13,1. In the emergency department during initial evaluation the patient had one more episode of hematochezia, the fourth from the beginning of his symptoms. Because of the ongoing bleeding it was decided that he needed hospitalization monitoring. A nasogastric tube was placed. Nasogastric aspiration showed bilious fluids and no signs of upper gastrointestinal bleeding.

A urinary catheter was also placed to measure the urine output and the patient was transferred to the surgical department. Intravenous fluids were started.

Crystalloid solutions were given with an infusion rate of 120 ml/h. Also 40 mg of omeprazole were given every 12 hours. Vital signs and urine output were measured hourly.

In the next few hours the patients condition evolved. Three more episodes of hematochezia occurred. The patients vital signs begun to alter. After 4 hours he was able to maintain normal blood pressure but his heart rate was elevated up to 110 bpm. During that time a second blood count was taken and a hematocrit of 33,2 with a hemoglobin of 13,1 was found. 10 hours later a third blood count was taken and a transfusion of three units of packed red blood cells was started.

During that time the patients blood pressure begun to drop (88/58 mm Hg) and his heart rate was even more elevated (123bpm). His urine output was also decreased (25ml per hour). The patient was pale and he had a syncoptic episode due to hypovolemic shock that was the result of the ongoing bleeding. From the blood count a hematocrit of 24,5% was found with a hemoglobin of 8,1. This represented a fall of 15% in the patients hematocrit during the last 10 hours. An emergent colonoscopy that was performed found the lumen of large intestine full of blood from the rectum to the ileocecal valve. The site of bleeding was not possible to be determined. An emergent gastroscopy that was performed showed no remarkable findings. It was decided that the patient should undergo surgery because of the ongoing hemorrage and his hemodinanic instability. After colonoscopy he was immediately transferred to the operation room. During operation the findings were similar to those of colonoscopy. The large intestine was full of blood but the site of bleeding could not be recognized. The small intestine was normal. An extensive right hemicolectomy was decided and performed. The patient had to receive 3 blood units during operation to maintain hematocrit and hemoglobin values. After the operation the patient was stable and showed no signs of rebleeding. The 8th postoperative day he was released from the hospital in good condition. Reexamination of the patient after 20 days showed noremarkable findings and no signs of rebleeding. The histopathologic examination of the specimen showed an AVM but also signs of early IBD. AVM was thought to be the most probable cause of the patients symptoms.

## Discussion

Lower GI hemorrhage is defined as an abnormal intraluminal blood loss from a source distal to Treitz ligament (3). Lower GI bleeding accounts about 20% of all GI bleeding with the small intestine being the site of bleeding in about 1% of cases and almost all the remaining cases located in the colon (2). It is classified in three groups according to the amount of bleeding, occult, moderate and massive bleeding. Massive lower GI bleeding is characterized by passage of a large volume of red or maroon blood through the rectum, hemodynamic instability and shock, initial decrease in hematocrit level of 6g/dl, transfusion of at least 2 units packed red blood cells, bleeding that continues for 3 days or significant rebleeding in 1 week (3). Usually patients with GI bleeding present to the hospital after observing the passage of blood from the GI tract as hematemesis, melena or hematochezia. Melena is a black, tarry stool resulting from the

degradation of blood by enteric bacteria. It can be a sign of upper GI bleeding, but also it can arise from small intestine or right colon. Hematochezia is the passage of red bright blood from the rectum and it is usually indicative of a lower GI source although massive upper GI hemorrhage can also cause hematochezia. Acute lower GI bleeding presented with hematochezia in the majority of cases (55,5%) with the next most frequent presentations being maroon stools (16,7%) and melena (11%) (3).

Diverticulosis is the most common cause of lower GI bleeding followed by colitis, neoplasms and angiodysplasia (2). In a retrospective review of medical records from approximately 1100 patients with acute lower GI bleeding all of whom were admitted to the surgical service of a single urban emergency hospital, it was determined that the most common etiologies for bleeding in these patients were diverticular disease (33,5%), hemorrhoids (22,5%) and carcinoma (12,7%) (3,4). An other retrospective study taking account of 4410 patients found that among patients with lower GI bleeding the most common causes of bleeding were diverticulosis (60%), IBD (13%), anorectal disorders (11%). Although some publications have reported AVM as a common cause of lower GI bleeding the true incidence seems to be low (3%) (3,5). Lower GI bleeding is much more common in the elderly. Studies show an average age of 60 years of such patients (2,6). Other studies found a 200-fold increase to the frequency of lower GI bleeding from the 3rd to the 9th decade of life (2,7). The common causes of lower GI bleeding in children and young adults differ from those found in older ages. Meckel diverticulum, intussuspeeption, polyposis syndromes, AVM and IBD are the most common causes in the age group from 1 to 30 years old (1,3).

Arteriovenous malformation (AVM) of gastrointestinal tract is one of the causes of lower gastrointestinal bleeding. It can happen without any specific medical history or family history. It is rare in people under 50 years old. AVM is also known as angiodysplasia, vascular ectasia, angioma and represents a degenerative lesion that increases in proportion of aging. It results from intermittent low grade obstruction of submucosal veins since it penetrates the muscular layers of the colon and cause AVM. Histologically it is noted as a hypertrophy of submucosal layer, infiltration of inflammatory cell and irregularly thickened vessel (8). AVM tends to cause slow but repeated episodes of bleeding. Therefore patients with angiodysplasia present with anemia and syncopal episodes. Infrequently angiodysplasias can cause abrupt loss of large quantities of blood. It is reported that 80% of angiodysplasia was investigated in the distal ileum, ascending colon, hepatic flexure and especially in cecum (1,8,9). Actively bleeding lesions can be treated with colonoscopic electrocoagulation. Incident discovered lesions should be left alone (3).

Massive hemorrhage due to IBD is rare. Ulcerative colitis cause bloody diarrhea in most cases. In up to 50% of patients with ulcerative colitis, mild to moderate lower GI bleeding occurs, and approximately 4% of patients with ulcerative colitis have massive hemorrhage. Lower GI bleeding in patients with Crohn's disease is not as common as in patients with ulcerative colitis. 1-2% of patients with Crohn's disease may experience massive bleeding (3).

Fortunately 75-80% of all cases of acute lower GI bleeding stop spontaneously (1,2,10). In these cases conservative treatment with intravenous fluids infusion, careful monitoring of the patient, nasogatric and urinary catheters placement are enough. Blood products should be given if indicated to maintain hematocrit above 24%, platelets above 50000 and prothrombine time under 15 seconds (1). In the remaining 20-25% of cases of acute lower GI bleeding conservative measures are not enough and surgery is necessary to control hemorrhage. The most common indications for surgical control of the bleeding are, bleeding that requires >1500ml/24 hours of packed red blood cells to maintain blood volume, ongoing bleeding that continues over 48 hours and rebleeding in a patient with lower GI bleeding that was temporarily controlled (1,2).

If surgery is necessary a more limited resection is possible if the site of bleeding is known. Segmental colectomy has been shown under these circumstances to be associated with a lower mortality than subtotal colectomy, and the incidence of recurrent hemorrhage after segmental resection is low (10,14). To determine the site of bleeding several diagnostic methods are used. Colonoscopy can be both diagnostic and therapeutic using thermal modalities or sclerosing agents (3). Selective mesenteric angiography permits the identification of vascular abnormalities and the precise bleeding point. Since its discovery selective mesenteric angiography remains the criterion standard in precise localization of the bleeding. Superselective visceral arteriography for infusion of vasoconstrictors and superselective embolization of the mesenteric vessels as an alternative technique to treat massive lower GI bleeding can also be used with the danger of ischemic colitis (11-13). Vasopressin infusion also causes vasoconstriction and can arrest bleeding. The recurrence rate can be as high as 71%. Therefore vasopressin infusion is used to temporize the acute event and to stabilize patients before surgery (3,10).

If no bleeding site can be recognized in a patient with ongoing bleeding then a blind subtotal colectomy is advocated by most authors (3,10,15). This happens because blind limited resections had a high recurrence rate of 30-60% (16). On the other hand morbidity and mor-

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tality of urgent subtotal colectomy are high. Bender et al collate the series of lower GI bleeding treated with total colectomy, published between 1972 and 1989 and found a mortality rate of 22,6% (16,17). In our case taking account the young age of our patient and the fact that AVM was the most probable diagnosis we chose to perform an extensive right hemicolectomy and put him under close observation and monitoring postoperatively for the possibility to do a more extensive operation if he rebleed.

Fortunately the hemorrhage was controlled and the patient remained stable after surgery.

# References

- 1. Papadimitriou I. Current general surgery. Parisianos editions' Athens 2001:1109-1121.
- Mulholand MW, Lillemoe KD, et al. Greenfield's surgery. Scientific principles and practice. Lippincott, Williams and Wilkins' 5<sup>th</sup> editions 2010.
- Burt Cajir, Elizabeth Cirincione, Lower gastrointestinal bleeding, Surgical Treatment. e Medicine>specialities>General surgery>Colorectal Oct 2009.
- Gayer C, Chino A, Lucas C, Tokioka S, Yamasaki T, Edelman DA, Sugawa C. Acute lower gastrointestinal bleeding in 1,112 patients admitted to an urban emergency medical center. Surgery 2009;146(4):600-6.
- Vernava AM, Longo WE, Virgo KS. A nationwide study of the incidence and etiology of lower gastrointestinal bleeding. Surg Res Commun. 1996;18:113-120.
- Peura DA, Lanza FL, Gostout CJ, Foutch PG. The American College of Gastroenterology Bleeding Registry: preliminary findings. Am J Gastroenterol 1997;92(6):924-8.
- 7. Longstreth GF. Epidemiology and outcome of patients hospitalized with acute lower gastrointestinal hemorrhage: a population-based study. Am J Gastrenterol 1997;92(3):419-24.
- Choi HK, Park CI, Shin JA, Moon JT, Lee SJ, Park CI, Park H. Hematochezia with colonic polypoid angiodysplasia in a young female patient. Gut Liver 2008 Sep;2(2):126-9.
- Cavett CM, Selby JH, Hamilton JL, Wiliamson JW. Arteriovenous malformation in chronic gastrointestinal bleeding. Ann

## Conclusions

In young patients with massive lower GI bleeding of unknown origin extensive right hemicolectomy provides a good and safe therapeutic choice that will control hemorrhage in most cases with the advantage of lower mortality and morbidity rates compared tosubtotal colectomy. Close monitoring of the patient postoperatively is essential. If one decides to perform a right hemicolectomy in such cases he should take into account the risk that a second operation to do a more extensive colectomy may be necessary if the patient rebleeds.

#### surg 1977;185(1):116-21.

- 10. Roberts P, Thomas PA. Massive large-bowel haemorrhage. Ann R Coll Surg Engl 1981; 63(2):108-10.
- Rösch J, Dotter CT, Rose RW. Selective arterial infusion of vasoconstrictors in acute gastrointestinal bleeding. Radiology 1971;99(1):27-36.
- 12. Athanasoulis CA, Baum S, Rösch J, Waltman AC, Ring EJ, Smith JC Jr, Sugarbaker E, Wood W. Mesenteric arterial infusin of vaso pressin for hemorrhage from colonic diverticulosis. Am J Surg 1975;129(2):212-6.
- 13. Allison DJ. Therapeutic embolization. Br J Hosp Med 1978;20(6):707-15.
- 14. Welch CE, Athanasoulis CA, Galdabini JJ. Hemorrhage from the large bowel with special reference to angiodysplasia and diverticular disease. World J Surg 1978;2(1):78-83.
- Drapanas T, Pennigton DG, Kappelman M, Lindsey ES. Emergency subtotal colectomy: prefered approach to management of massively bleeding diverticular disease. Ann Surg 1973; 177(5):519-26.
- Simoens CH, Mendes da Costa P. Surgical management of recurrent colon haemorrhage of unknown origin. Acta Chir Belg 2003;103(5):481-4.
- Bender JS, Wiencek R, Bouwman DL. Morbidity and mortality following total abdominal colectomy for massive lower gastrointestinal bleeding. Am Surg 1991;57(8):536-40.