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Case Report

SPONTANEOUS HEMOPERITONEUM AND ISOLATED GANGRENE OF SPLENIC FLEXURE AND DESCENDING COLON IN A PATIENT OF RHEUMATOID ARTHRITIS: AN EXTREMELY RARE EVENT

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ABSTRACT

Spontaneous hemoperitoneum is the presence of intraperitoneal hemorrhage from a nontraumatic and noniatrogenic cause and there are only a few reported cases of in rheumatoid arthritis-associated gastrointestinal vasculitis. We present an extremely rare case of spontaneus hemoperitoneum with isolated gangrene of splenic flexure and descending colon due to non traumatic left colic arterial haemorrhage along with pneumoperitoneum in a patient of rheumatoid arthritis. An emergency laparotomy with resection of the gangrenous colon and transverse terminal colostomy was performed after preoperative resuscitation. We propose that in our case the cause of isolated gangrene could be due to the conglomeration of factors such as occlusion of small and medium-sized vessels of mesentric circulation particularly the involvement of collateral channels like Arch of Riolan and marginal artery of Drummond due to rheumatoid arthritis-associated gastrointestinal vasculitis compounded by splanchnic hypoperfusion and systemic hypotension as a result of left colic arterial bleed. All these factors led to ischemic injury at "watershed" areas of mesenteric circulation. Although rare, rheumatoid vasculitis can cause spontaneous hemoperitoneum and gangrenous changes in the bowel. The presence of gastrointestinal (GI) symptoms in a patient of rheumatoid arthritis should be investigated thoroughly.

Key Words: *Spontaneus Hemoperitoneum, Isolated Gangrene, Splenic Flexure, Descending Colon, Rheumatoid Arthritis, Rheumatoid Vasculitis*

INTRODUCTION

Spontaneous hemoperitoneum is defined as the presence of intraperitoneal hemorrhage from a nontraumatic and noniatrogenic cause (Furlan *et al.,* 2009). This condition is associated with high mortality if not managed appropriately (Skourasa *et al.,* 2011). Though rare spontaneous hemoperitoneum has been earlier reported in patients with gastrointestinal (GI) vasculitis. Upper gastrointestinal (UGI) bleeding and hemorrhagic shock in rheumatoid arthritis-associated gastrointestinal vasculitis is very rare (Parker and Chattopadhyay, 2007; Burt *et al.*, 1983; Achkar *et al.,* 1995; Pagnoux *et al.,* 2005; Babian *et al.,* 1998; Jayawardena *et al.,* 2001). Our aim is to report an extremely rare case of spontaneous hemoperitoneum with isolated gangrene of splenic flexure and descending colon due to non traumatic left colic arterial haemorrhage with pneumoperitoneum in a patient of rheumatoid arthritis.

CASES

A 28-year-old Indian male patient was admitted to the Emergency department of NIMS University Medical College, Shobha Nagar, Jaipur complaining of generalized severe pain abdomen, abdominal distension and absolute constipation for the last 4 days.

The pain was acute in onset, non-radiating, not related to meals and initially situated in the left half of the abdomen and latter became generalized. It was associated with anorexia, nausea, few attacks of vomiting.

There was no evidence of bleeding from any site. There was history of prolonged low grade fever, vague abdominal pain and weight loss. There was no history of urinary symptoms. The patient was a known case of rheumatoid arthritis for the last 6-7 years and was on steroids (prednisolone 10 mg daily for the last 6 years). He had episodes of morning stiffness, but, there was no apparent orthopaedic deformity. He had no history of surgeries or allergy, and had no special habits.

On physical examination the patient was febrile, grossly anemic and there was severe muscle wasting. Pulse 124/min and blood pressure was 92/64 mm Hg. There was no evidence of heart failure. There was generalized abdominal distension and tenderness. There was a vague lump abdomen in left half of the abdomen. Abdominal guarding and rigidity was present. Rectal examination revealed an empty rectum with dark colored blood in the rectum.

Lab investigations revealed Hb 6.9 gm%, total leucocyte count of 23.5 thousand/ml, sodium 139 mmol/L, potassium 4.5 mmol/L and bilirubin 1.15 mg%, SGOT 118 U/L, SGPT 158 U/L, ALP 390 U/L. Abdominal x-ray revealed gas under the diaphragm (Figure 1). On ultrasonography (USG) a large collection with internal echoes was noted in pelvis and an organized hypoechoic area was noted in left lumbar region. Free fluid was also seen in perihepatic, perisplenic and Morrisons pouch.

Patient was prepared for emergency laparotomy and 4 units of blood were arranged. Abdomen was opened by a midline incision. There was hemoperitoneum with 1.5 litres of blood present in the peritoneal cavity (pelvis), which was aspirated. The abdomen was explored. The source of hemoperitoneum was from Left colic artery, which was ruptured and bleeding. It was controlled with an artery forceps and ligated. There were multiple hematomas present in the mesentery with otherwise normal small bowel loops (Figure 2). The omentum was wrapping the splenic flexure and descending colon which were found to be gangrenous after separating it from omentum (Figure 2). There was full thickness gangrene, starting from the splenic flexure to descending colon, just proximal to the beginning of sigmoid colon. Whole of the gangrenous descending colon was removed. Liver, spleen and other viscera were normal. Peritoneal toileting was done. In view of the poor clinical condition of the patient a transverse terminal colostomy was done along with closure of the distal end (sigmoid colon). Abdomen was closed with tension sutures. The resected bowel was sent for histopathological examination. Postoperatively patient recovered well, except for minor wound infection which was treated with regular dressings. Patient was discharged on 25th postoperative day in a stable condition and is under follow up.

DISCUSSION

Rheumatoid vasculitis (RV) is a systemic necrotizing vasculitis affecting small and medium-sized vessels (Parker and Chattopadhyay, 2007). It is an extra-articular manifestation of rheumatoid arthritis and is a well-known complication of long-standing seropositive rheumatoid arthritis (Parker and Chattopadhyay, 2007). It is present in approximately 25.0% of all patients with rheumatoid arthritis, whereas less than 1% of them developed clinical signs of vasculitis (Burt *et al.*, 1983). GI involvement is rare and among these patients, 10%-38% will have gastrointestinal vasculitis and this may be the first manifestation (Achkar *et al.*, 1995).

Other well known causes of gastrointestinal vasculitis are polyarteritis nodosa, microscopic polyangiitis, Wegener granulomatosis, Churg-Strauss syndrome, mycotic and bacterial infections, trauma, pancreatitis (Skourasa *et al.*, 2011; Pagnoux *et al.*, 2005).

The common gastrointestinal manifestations seen in gastrointestinal vasculitis are abdominal pain, nausea or vomiting, diarrhea, hematochezia or melena, hematemesis, gastroduodenal ulcerations including esophageal and colorectal (Pagnoux *et al.*, 2005). Peritonitis, bowel perforations, bowel ischemia/infarction, intestinal occlusion, acute appendicitis, cholecystitis, and acute pancreatitis and abdominal visceral aneurysm, aneurysmal rupture with syncope and hemorrhage are reported complications in gastrointestinal vasculitis. Mortality is high in cases with surgical abdomen (Skourasa *et al.*, 2011; Achkar *et al.*, 1995; Pagnoux *et al.*, 2005; Babian *et al.*, 1998; Jayawardena *et al.*, 2001).

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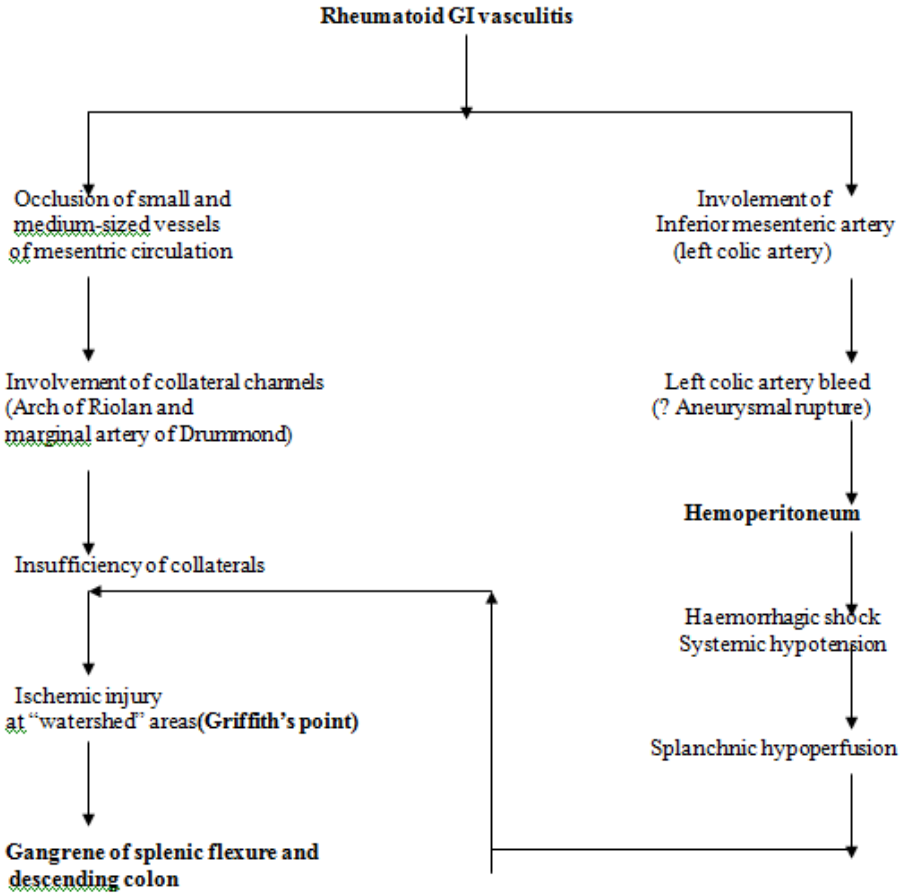

Flow Chart: Conglomeration of factors leading to isolated gangrene of descending colon and splenic flexure (Griffith's Point) and spontaneous hemoperitoneum

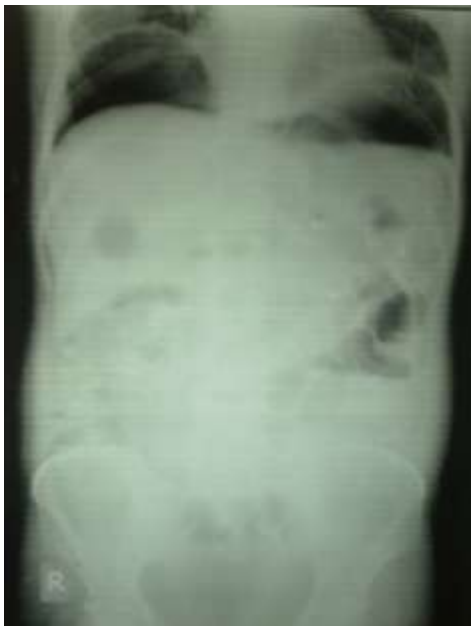

Figure 1: Radiograph showing pneumoperitoneum with gas under both the domes of diaphragm

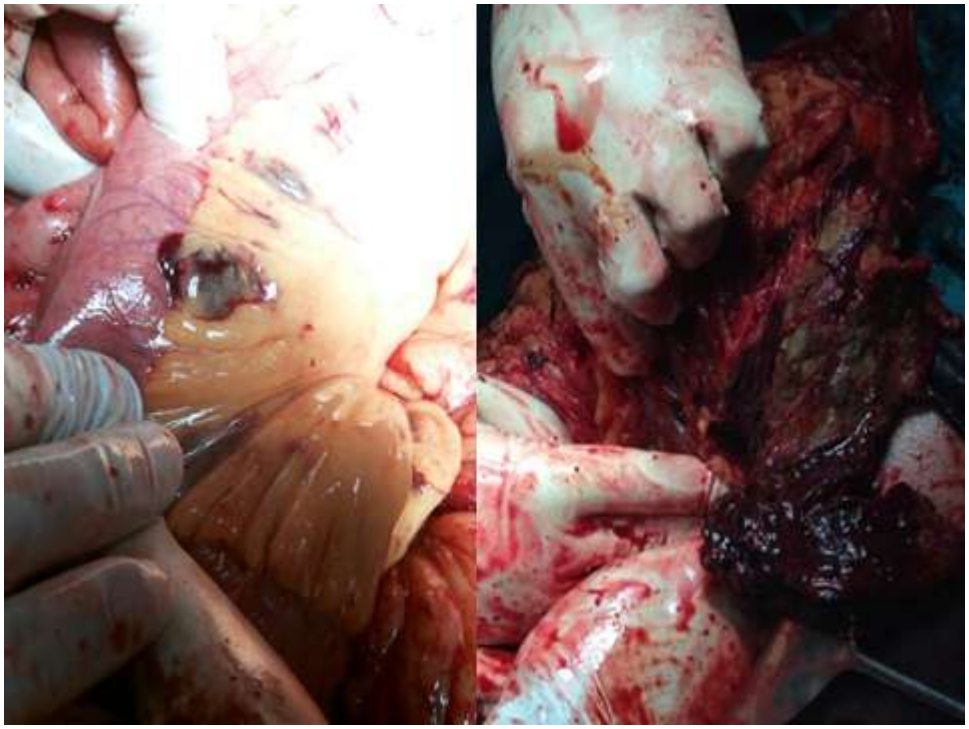

Figure 2: Intraoperative photographs showing multiple hematomas in the small bowel mesentery with otherwise normal bowel loops in the left photograph, gangrenous descending colon being separated from the omentum (which was wrapping the gangrenous colon) in the right photograph

Hemoperitoneum is very rare event in a patient of rheumatoid vasculitis and its simultaneous occurrence with isolated gangrene of splenic flexure and descending colon in is extremely rare (Skourasa *et al.,* 2011; Achkar *et al.*, 1995; Pagnoux *et al.*, 2005; Babian *et al.*, 1998; Jayawardena *et al.*, 2001). The source of bleeding in gastrointestinal vasculitis could be left gastric artery (Yahata *et al.*, 2009), superior mesenteric artery (Choo and Yen, 2013), ileo-colic artery (Cheung *et al.,* 2010), middle colic artery (Skourasa *et al.,* 2011), but in our case, the cause was ruptured left colic artery.

Pneumoperitoneum without bowel perforation seen in our patient was possibly due to transmigration of gas forming bacteria from the gangrenous bowel.

The arterial circulation to the gut can develop extensive collaterals to supply areas of relative ischaemia in the mesenteric circulation, as often happens with chronic mesenteric ischaemia (Menon *et al.,* 2005). Well recognised main collateral channels between the SMA and Inferior Mesenteric Artery (IMA) occur in the region of the splenic flexure (Griffith's point) at both the base and border of the mesentery, through the arch of Riolan (an ascending branch of the left colic artery anastomosing with the branches of SMA) and the marginal artery of Drummond, respectively (Menon *et al.*, 2005). Another important collateral circulation is between internal iliac artery and the left colic artery via the superior haemorrhoidal arteries (Menon *et al.*, 2005).

However, these collaterals are insufficient when an acute insult occurs in mesenteric circulation and during systemic hypotension. This is because mesenteric vasoconstriction is a normal physiological response to shock mediated largely by the renin angiotensin axis. Ischemic injury most often occurs at "watershed" areas of mesenteric circulation, i.e. splenic flexure (Griffith's point) where the collateral arteries are small and narrow (Menon *et al.*, 2005). In our case the cause of isolated gangrene of descending colon and splenic flexure (Griffith's point) and spontaneous hemoperitoneum could be due to the conglomeration of factors as explained in the flow chart.

The treatment of spontaneous hemoperitoneum revolves around aggressive patient resuscitation followed by surgical management which includes emergency laparotomy, and ligation of the source of bleeding. Bowel resection is indicated if the intestinal blood supply has been compromised and if there is evidence of gangrene (Skourasa *et al.,* 2011).

Conclusion

Although rare, rheumatoid arthritis-associated gastrointestinal vasculitis can cause spontaneous hemoperitoneum and gangrenous changes in the bowel. The presence of GI symptoms in a patient of rheumatoid arthritis should be investigated thoroughly.

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