

in diameter.³ In 85% of cases, hemangiomas are clinically asymptomatic and are incidentally detected in imaging. Diffuse hepatic hemangiomatosis usually occurs in neonates and is associated with Rendu-Osler-Weber disease or skeletal hemangiomatosis.^{4,5} Surgical resection is the most commonly used treatment modality. A stable hemangioma has a rupture rate of 1% to 4% and after rupture, a fatality rate of 60% to 75%. The natural history is variable, and they can regress spontaneously, undergo thrombosis, scarring (called as sclerosed hemangioma) or calcification. Close follow-up without intervention is the best way of management of asymptomatic cases. The available treatment options include radiotherapy, interferon therapy, transcatheter arterial embolization (TAE), radiofrequency ablation, arterial ligation, corticosteroid therapy and liver transplantation. Spontaneous or traumatic rupture, intra tumoral hemorrhage, consumption coagulopathy (Kasabach-Merritt Syndrome) and a rapid growth rate are absolute surgical indications. Surgery remains the only consistently effective curative treatment for giant haemangiomas. An everlasting debate exists between enucleation and hepatic resection, both having equal efficacy and a decision is usually made on the size of tumor. However, Kayan et al⁶ reported that the combination of preoperative percutaneous trans-catheterarterial embolization and surgery is a safe procedure that eliminates the risk of intraoperative bleeding. In unresectable lesions, when there is multiple bilobar involvement, and in symptomatic cases with hepatic hilum involvement, liver transplantation is the most appropriate approach. Indications for liver transplantation in cases of hepatic hemangioma include acute and chronic hepatic failure, such as occurrence of Kasabach-Merritt syndrome, giant hepatic hemangioma affecting the normal liver tissue eventually causing liver dysfunction, and a life-threatening giant hepatic hemangioma that cannot be resected.⁷ Stapler Hepatectomy mode of transection is fast with less bleeding from parenchyma and easy for left lobe resection.

M MANISEGARAN
ANUROOP THOTA

NRI Academy of Medical Sciences, Andhra Pradesh.

*Correspondence: Anuroop Thota
Email: anuroopt@gmail.com*

References

1. Weimann A, Ringe B, Klempnauer J et al. Benign liver tumors: differential diagnosis and indications for surgery. *World J Surg.* 1997;**21**:983-990; discussion 990-991.
2. Ishak KG, Rabin L. Benign tumors of the liver. *Med Clin North Am.* 1975;**59**:995-1013.
3. Adam YG, Huvos AG, Fortner JG. Giant hemangiomas of the liver. *Ann Surg.* 1970;**172**:239-245.
4. Gutierrez RM, Spjut HJ. Skeletal angiomas: report of three cases and review of the literature. *Clin Orthop Relat Res.* 1972;**85**:82-97.
5. Haitjema T, Westermann CJ, Overtom TT et al. Hereditary hemorrhagic telangiectasia (Osler-Weber-Rendu disease): new insights in pathogenesis, complications, and treatment. *Arch Intern Med.* 1996;**156**:714-719.
6. Kayan M, Cetin M, Aktaş AR et al. Pre-operative arterial embolization of symptomatic giant hemangioma of the liver. *Prague Med Rep.* 2012;**113**:166-171.
7. Lin Zhong, Tong-Yi Men, Gao-di Yang et al. Case report: living donor liver transplantation for giant hepatic hemangioma using a right lobectomy without the middle hepatic vein. *World Journal of Surgical Oncology.* 2014;**12**:83.

Spontaneous perforation of an ileal J pouch

Perforation of the ileal pouch is a rare complication of an ileal pouch-anal anastomosis with only a few cases reported till date.

Case Report

A 52-year-old woman who had undergone a 3-stage restorative total proctocolectomy for refractory severe ulcerative colitis one year ago, presented to the emergency with complaints of severe pain in the abdomen, abdominal distension and obstipation. She was apparently well till 5 days ago when these symptoms started and gradually

progressed. She also developed intermittent fever for the past 3 days. She had not passed urine for some hours before presentation. On examination, she was irritable and dehydrated. Her pulse rate was 124/minute and blood pressure was 86/60 mmHg. Her abdomen was distended and there was generalized tenderness along with rebound tenderness and absent bowel sounds. A per rectal examination did not reveal any abnormality. A chest radiograph including both domes of diaphragm was obtained which showed free air under the diaphragm.

Her past surgical history revealed that a subtotal colectomy was done in January 2010 as the first stage for acute severe colitis unresponsive to a 5-day course of intravenous steroids. Subsequently, a completion proctectomy and ileal pouch–anal anastomosis using a J pouch with a covering loop ileostomy was done in October 2010. Finally, the loop ileostomy was closed in June 2012. She made an uneventful recovery after each of the procedures. The pouch function was good with a stool frequency of five to six times per day and no incontinence.

A diagnosis of bowel perforation causing peritonitis, possibly secondary to intestinal obstruction due to adhesions, internal herniation or a band was made. She was taken up for an emergency exploratory laparotomy after initial resuscitation and optimization of electrolytes.

Intraoperatively, there was 500 ml of faeculent fluid in the peritoneal cavity. There were dense adhesions; however, there was no internal herniation or band. There was a 5 mm perforation with unhealthy margins in the proximal limb of the J pouch (**Figure 1**). The margins of the perforation were freshened and it was closed with interrupted 4-0 polydioxane sutures. A thorough peritoneal lavage was done and a diverting loop ileostomy was fashioned.

No definite cause for the perforation could be identified. A Widal test and Cytomegalovirus PP65 antigen test done postoperatively were negative. The histopathology of perforation margins revealed mucosal ulceration with acute as well as chronic inflammation. A formal pouch examination had not been done previously but she did not give history of any symptoms suggestive of pouchitis.

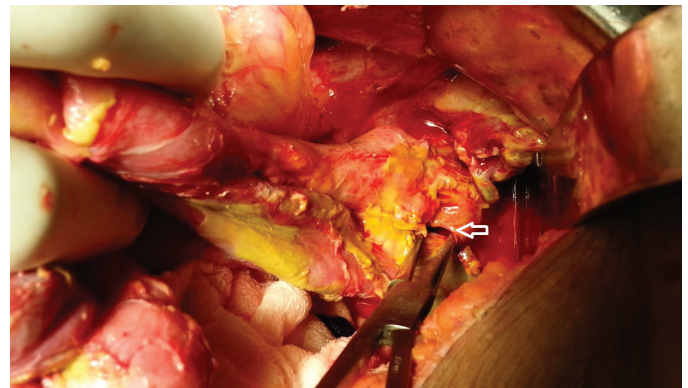


Figure 1: Perforation in ileal J pouch (arrow).

Discussion

All the previous reported cases of pouch perforation had underlying risk factors for perforation like trauma,¹ pregnancy,² volvulus³ or twisting of a long blind end.⁴ In another case, there was a combination of a long blind end and pouchitis that resulted in perforation.⁵ However, our patient had none of these risk factors. The pouch was not dilated and there was no distended blind limb. Moreover, the perforation was located in the ascending limb and not in the blind limb. The biopsy from the margin of the perforation revealed mucosal ulceration with features of acute as well as chronic inflammation. However, there was no previous history of any symptoms of pouchitis like increased stool frequency, per rectal bleeding or fever. In view of the negative aetiological workup and non-specific operative and histopathology findings, a diagnosis of idiopathic spontaneous pouch perforation was made.

In India, ileal perforations are most commonly caused by infections like *Salmonella typhi* and tubercular mycobacteria. However, no definite cause is identified in a large proportion of patients and the histopathology shows only non-specific inflammatory changes.⁶ Since the ileal pouch is constructed from the terminal ileum, it is likely to be susceptible to all the diseases that affect the small bowel. Thus, in the absence of any other aetiological clues, we presume that the ileal pouch perforation was secondary to non-specific ileal inflammation.

To the best of our knowledge, this is the first report of a spontaneous perforation in an ileal J pouch. The treatment for non-specific ileal perforation is either

primary repair or exteriorizing the perforated ileum as a loop ileostomy. The only option available in pouch perforation is primary repair with or without a proximal stoma. We chose to create a proximal stoma as the patient had presented late with generalized peritonitis and haemodynamic instability requiring inotropic support and the margins of the perforation were unhealthy.

RAJESH PANWAR
ARIF ALI KHAN
SUJOY PAL
NIHAR RANJAN DASH
VINEET AHUJA

All India Institute of Medical Sciences, New Delhi.

Correspondence: Rajesh Panwar
Email: rajeshpanwar81@gmail.com

References

1. Hsu TC. Traumatic perforation of ileal pouch. Report of a case. *Dis Colon Rectum*. 1989;**32**(1):64-6.
2. Aouthmany A, Horattas MC. Ileal pouch perforation in pregnancy: report of a case and review of the literature. *Dis Colon Rectum*. 2004;**47**(2):243-5.
3. Arima K, Watanabe M, Iwatsuki M, et al. Volvulus of an ileal pouch-rectal anastomosis after subtotal colectomy for ulcerative colitis: Report of a case. *Surg Today*. 2014;**44**(12):2382-4.
4. Pezim ME, Taylor BA, Davis CJ, Beart RW Jr. Perforation of terminal ileal appendage of J-pelvic ileal reservoir. *Dis Colon Rectum*. 1987;**30**(3):161-3.
5. Takahashi K, Funayama Y, Fukushima K et al. Ileal J-pouch perforation at the blind end: report of a case. *Surg Today*. 2009;**39**(12):1080-2.
6. Mittal S, Singh H, Munghate A, Singh G et al. A comparative study between the outcome of primary repair versus loop ileostomy in ileal perforation. *Surg Res Pract*. 2014;2014:729018. doi: 10.1155/2014/729018.