Perforated Isolated Jejunal Diverticula due to Enterolith: A Case Report and Review of Literature

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Abstract

Jejunal diverticula is a rare condition quoted to affect between 0.5% and 7.0% of the population. Usually, it is clinically silent and becomes symptomatic only when complications develop. Perforation of a jejunal diverticulum secondary to enterolith formation leading to generalised or localised peritonitis is extremely rare; even rarer is an isolated perforated jejunal diverticulum. Herein, the authors present a case of perforated isolated jejunal diverticula due to enterolith in a 60-year-old female patient managed by small bowel resection and primary anastomosis.

INTRODUCTION

Small bowel diverticula are sac-like pouchings of the small bowel wall that can occur throughout the small bowel. Small bowel diverticula are most often found in the duodenum, followed by jejunum and ileum. In fact, Akhrass et al.,¹ in their retrospective review of 208 patients with symptomatic small bowel diverticulosis, found that diverticula were located in the duodenum in 79% of cases, in the jejunum or ileum in 18% of cases, and in all three segments in 3% of cases. The incidence of jejunoileal diverticula is variable, reported to occur in 0.5–2.3% of individuals in radiographic series and up to 7% in autopsy studies.² Furthermore, 77% of cases demonstrate multiple, as opposed to solitary, diverticula.³ They are more commonly reported in males, with the highest incidence in the sixth and seventh decades of life.

bowel diverticula Small usuallv are asymptomatic, with а spectrum of presentation ranging from non-specific abdominal pain, dyspepsia, and bloating and reaching presentations with life-threatening complications. In fact, Tsiotos et al.4 analysed 112 cases of jejunoileal diverticulosis where 42% of cases were asymptomatic; in the remaining patients, symptoms of diarrhoea were reported in 58% of patients, chronic abdominal pain in 51%, and bloating in 44% of cases. Furthermore, complication rates as high as 46% for jejunal diverticulosis have been reported and are known to be fatal at times.⁵ Among the complications is perforation with localised or generalised peritonitis. Herein, the authors report a case of perforated isolated jejunal diverticula due to enterolith in a 60-year-old female patient, managed by small bowel resection and primary anastomosis.

CASE REPORT

This is a case of a 60-year-old female patient with previous history of laparoscopic cholecystectomy, presenting with a 2-day history of non-specific epigastric pain associated with decreased food intake. The patient denied having a fever, nausea, vomiting, obstipation, or change in bowel habits. The patient also denied weight loss and personal or family history of malignancy.

Upon arrival to the authors' emergency department, the patient's vitals were stable and a physical examination of the abdomen was within normal limits, with hypoactive bowel sounds and minimal tenderness upon deep palpation of the left upper and left lower quadrants. Laboratory work-up, including complete blood count, inflammatory markers (C-reactive protein), liver function tests, amylase, and lipase, and urine analysis were within normal limits. A CT scan of the abdomen and pelvis with intravenous (IV) contrast was negative for any pertinent signs except for segmental enteritis, with no evidence of bowel suffering and no signs of ischaemia.

The admitted for patient was further management of enteritis by IV hydration and IV antibiotics (ciprofloxacin and metronidazole) for 7 days in total. Two days after the admission, the patient reported acute worsening of her abdominal pain that was associated with one episode of fever of 39 °C and chills. On physical examination of her abdomen, there was diffuse four-quadrant tenderness and rebound tenderness, more so over the left side of the abdomen.

Consequently, another CT scan with IV contrast showed diffusely thickened and enhancing jejunal loops (Figure 1A). A dilated jejunal loop up to 4.5 cm filled with bezoar or enterolith (Figure 1B), significantly increased, and surrounding fat streaking were noted, in addition to free fluid in the pelvis and along the left paracolic gutter that had not been noted on the initial CT scan 2 days previously. In view of worsening pain, the new onset of fever, and the new findings on imaging despite IV antibiotics, the authors opted for a diagnostic laparoscopy.

In the operating theatre, the patient was given general anaesthesia, a urinary Foley catheter was inserted, and the patient was placed in the modified lithotomy position. An infraumbilical incision, an open technique

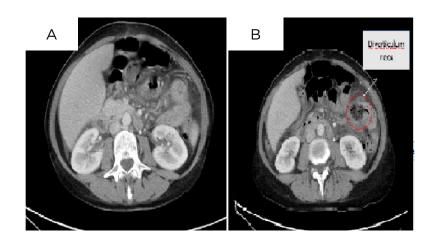


Figure 1: A) Diffuse jejunal wall thickening; and B) highlighting an enterolith or bezoar.

to access the abdominal cavity, insufflation, and the insertion of two 5 mm trocars were performed under direct vision in the suprapubic and left lower quadrant area. Sharp lysis of adhesions was done up until reaching the area of inflammation, whereby it was dissected using blunt dissection. A large jejunal diverticula on the mesenteric border measuring approximately 7 cm in diameter was identified, located 40 cm distal to the ligament of Treitz. Further blunt dissection of the diverticula from the jejunal mesentery identified an enterolith (Figure 2A). Furthermore, a jejunal perforation measuring approximately 3 cm was identified (Figure 2B). This was followed by a left paramedian 5 cm incision and placement of Alexis retractor. A small bowel segment was exteriorised and segmental resection with primary side-to-side anastomosis performed, followed by closure of the mesenteric defect (Figure 2C). Running of the small bowel from the ileocaecal valve until reaching the ligament of Treitz, the authors did not identify any other diverticula and no colonic diverticula identified intra-operatively. were Final

histopathology studies confirmed the diagnosis of perforated false diverticulum with signs of necrosis and gangrene at the edges of perforation (Figure 2D). The patient's postoperative course was smooth and the patient was discharged home on Day 4 post-operation.

DISCUSSION

Small bowel diverticulosis was first reported by Sommering in 1794.6 Its aetiology is thought to be related to a combination of intestinal dyskinesia and abnormal peristalsis, causing high segmental intraluminal pressures.7 In fact, the current hypothesis focuses on abnormalities in the smooth muscle or myenteric plexus. Having said this, microscopic evaluation of jejunal specimens with diverticulosis has shown that there are three different abnormalities: fibrosis and decreased numbers of normal muscle cells, fibrosis and degenerated smooth muscle cells, and neuronal and axonal degeneration.⁸ The presence of any of the above-mentioned abnormalities will consequently lead to distorted smooth muscle contractions of the affected small

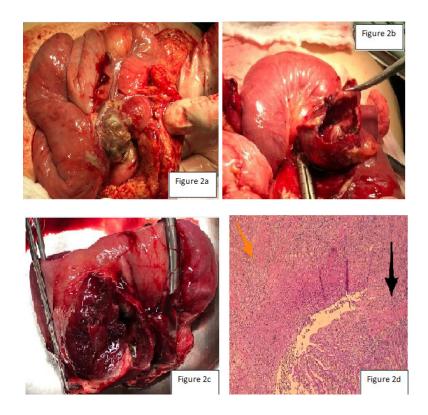


Figure 2: A) An enterolith, seen intra-operatively; B) diverticular perforation of the small bowel; C) the resected small bowel; and D) the transition from diverticular mucosa to bowel rupture, inflammation, and gangrenous formation. The black arrow represents normal mucosa while the orange arrow shows the rupture.

bowel, generating areas of increased intraluminal pressure, which in turn will lead to herniation of the bowel mucosa and submucosa through the weakest point in the bowel wall, the mesenteric border at the site of entry of the vasa recta, leading to the formation of small bowel diverticula.

Small bowel diverticula are usually clinically silent and discovered incidentally or once they become complicated. However, they may have a wide spectrum of presentation, ranging from nonspecific abdominal pain, dyspepsia, and bloating and reaching presentations with life-threatening complications. Life-threatening complications include gastrointestinal bleeding, massive volvulus, diverticulitis, and perforation with localised or generalised peritonitis. Furthermore, jejunal diverticulosis complicated by enteroliths is a rare entity. The enterolith, formed within small bowel diverticula, can be either de novo or secondary to a piece of undigested food. Those that form *de novo* are comprised of cholic acid, the end-product of bile salt metabolism, as their primary constituent. It is theorised that cholic acid enteroliths form within small bowel diverticula due to an acidic pH shift within the diverticula.9 Enterolith in jejunal diverticula can result in enterolith ileus or lead to perforation if it becomes impacted. The synergistic effect of intestinal dyskinesia with the abnormal peristalsis leads to the enterolith being stagnated due to this abnormal transit. This may eventually lead to perforation due to pressure necrosis or acute necrotising inflammation.^{10,11} Presentation varies widely depending on the severity of perforation. For instance, patients where the

enterolith has been sitting within the diverticulum for an extended period of time, resulting in progressive erosion into the bowel wall and consequently bowel wall perforation, usually present with a localised abscess without causing the patient to become acutely ill. This is either due to the perforation into the mesentery, hence a contained perforation, or due to the walling off by the adjoining small bowel mesentery so that only localised peritonitis will occur. On the contrary, patients with acute perforation present in an acutely ill situation with gross contamination of the abdomen and generalised peritonitis. For patients with localised peritonitis and clinically stable patients, non-surgical management by IV antibiotics and CT-guided aspiration of collections may be appropriate for some patients. On the other hand, for patients with generalised peritonitis or haemodynamic instability, resection of the affected segment should be done.

Reviewing the English literature regarding perforated isolated jejunal diverticula due to enterolith has revealed only few reported cases. In fact, in the authors' review of the medical literature, they identified a total of 23 cases of perforated jejunal diverticula (Table 1), of which five patients had isolated jejunal diverticula; their patient is the sixth reported in literature. The mean age of presentation was 71 years, with a male to female ratio of 2:1. Abdominal pain was the most frequent initial presentation.

All reported cases were managed surgically by laparotomy. The authors' case was the first to

	Author/	Age	Clinical	Radiologic	Surgical	Intra-operative	Multiple/	Treatment
	year	(years)/sex	presentation	finding	approach	finding	isolated	
							jejunal	
							diverticula	
1	Cegla et	65/F	Localised	CT: free fluid in the	Laparotomy	Mesenteric	Multiple	Segmental
	al., ¹² 2007		peritonism	abdominal cavity;		abscess;		resection
			in left iliac	free air in the		perforated		with primary
			fossa	retroperitoneum;		diverticula		anastomosis
				small bowel				
				perforation				
				detected				

Table 1: A review of medical	literature where 23 case	s of perforated jejunal	diverticula were identified
Table I. A review of medical	interature where 25 case	s of perforated jejunal	diverticula were identified.

Table 1 continued

			1	1	1		1	,
2	Kassahun	85/M	Fever,	CT: normal findings	Laparoscopy	Jejunal	Multiple	Resection of the
	et al.,13		diffuse		converted to	diverticulitis		entire diverticula-
	2007		abdominal		open	and abscesses		bearing segment
			pain, and			in the		of jejunum
			abdominal			mesentery/		with primary
			distension			sigmoid		anastomosis
						diverticulitis		and Hartmann's
								procedure
3	Lempinen	78/M	4 days of	US: No free air; no	Laparotomy	Extensive	Multiple	Jejunal resection
	et al.,³		right upper	fluid collections		jejunal		with primary
	2009		abdominal			diverticulosis;		anastomosis
			pain and			adjacent		
			chills			mesenteric		
						abscess		
4	Lempinen	75/F	1 day of	AXR: no free air	Laparotomy	Single jejunal	Isolated	Jejunal resection
·	et al., ³		abdominal			diverticula		with primary
	2009		pain (RLQ)			with adjacent		anastomosis and
	2005					mesenteric		appendectomy
						abscess		appendectoring
						abscess		
5	Lempinen	83/F	Increasing	CT: free air	Laparotomy	Perforated	Multiple	Segmental
	et al., ³		abdominal			diverticula		resection
	2009		pain and			and faecal		with primary
			signs of			peritonitis		anastomosis/lysis
			peritonitis				ļ	of adhesions
6	Lempinen	77/M	4 days of	CT: 5x10 cm solid	Laparotomy	Large,	Isolated	Jejunal resection
	et al., ³		abdominal	tumour		bleeding		with primary
	2009		pain and			jejunal		anastomosis
			melaena			diverticulum		
						impacted with		
						haematoma		
7	Lempinen	75/M	Acute	CT: Normal	Laparotomy	Perforated	Isolated	Jejunal resection
	et al.,³		abdominal			jejunal		with primary
	2009		pain and			diverticula,		anastomosis
			fever			30 cm from		
						ligament of		
						Treitz		
8	Lempinen	78/M	1 week of	CT: intra-abdominal	Laparotomy	Intra-	Multiple	Segmental
	et al., ³		abdominal	abscess		abdominal		resection
	2009		pain, nausea,			abscess		with primary
			vomiting,	US: thickened, slow				anastomosis
			1	peristaltic colonic				anastoniosis
			and fever;	bowel loops				
			increased					
			intensity of					
			abdominal					
			pain					
9	Lempinen	72/M	1 day of	NA	Laparotomy	Adhesions	Multiple	Adhesiolysis
	et al., ³		abdominal					
	2009	1	pain and	1				
	2005		1				1	

Table 1 continued.

10	Lempinen	59/M	Chronic	Gastro/colon	Laparotomy	Turbid fluid	Multiple	Segmental
	et al., ³		symptoms of	normal		in abdomen/		resection
	2009		abdominal	CT: normal		large		with primary
			pain,			impacted		anastomosis
			occasional	Enteroclysis: jejunal		small bowel		
			fever	diverticulosis		stone		
						(enterolith) in		
						mid-jejunum		
11	Chugay et	79/M	Abdominal	CT: extensive	Laparotomy	Turbid fluid	Multiple	Segmental
	al.,10 2010		pain,	inflammation of		in abdomen/		resection
			nausea, and	a small bowel		large		with primary
			vomiting	segment on		impacted		anastomosis
				left; free air;		small bowel		
				pneumatosis of		stone		
				mid-jejunum		(enterolith) in		
						mid-jejunum		
12	Chugay et	89/F	Abdominal	CT: pan-colic	Laparotomy	Multiple	Multiple	Segmental
	al.,10 2010		pain and	diverticulosis/		diverticula		resection
			constipation;	dilated small bowel		and dilated		with primary
			SBO	loops		SBO loops;		anastomosis
						impacted		
						stone (3 cm		
						enterolith) in		
						distal jejunum		
13	Butler et	82/F	Generalised	AXR: multiple	Laparotomy	2 pinhole	Multiple	Primary repair
	al., ¹⁴ 2010	02/1	abdominal	dilated small bowel	Laparotorny	jejunal		of the 2 sites
	01., 2010		pain and	loops CT: thickening		perforations		of perforation
			vomiting	of duodenum/		associated		with abdominal
			Vorniting	dilatation of		with faecal		washout
				proximal jejunum;		contamination		Washout
				multiple diverticula		Contarnination		
				with air surrounding				
14	Akbari et	74/M	2 days of	AXR: prominent but	Laparotomy	Multiple jejunal	Multiple	Segmental
14	al., ¹⁵ 2013	74/11		not dilated bowel	Laparotorny	diverticula,		resection
	dl., ¹⁰ 2015		constipation, anorexia,	loops		one of which		
			Í Í	loops				with primary
			fever, and			perforated		anastomosis
			left-sided			(40 cm distal		
			abdominal			to ligament of		
			pain			Treitz)		
15	Webster et	54/M	Diarrhoea,	CXR: air under the	Laparotomy	Generalised	Isolated	Segmental
	al., ¹⁶ 2014		vomiting,	diaphragm		purulent		resection
			and			peritonitis due		with primary
			abdominal			to perforation		anastomosis
			pain			jejunal		
						diverticula,		
						containing		
						impacted		
		1	I	1		faecalith		
						laccaliti		
						20 cm from		

Table 1 continued.

16	Kavanagh	63/M	DLO main	CT: contained	Lanavatanav	Perforated	Multiple	Commontal
16	÷	63/14	RLQ pain		Laparotomy		Multiple	Segmental
	et al., ⁷ 2014			perforation with		jejunal		resection
				central calcification		diverticula		with primary
				within diverticula				anastomosis
17	Chaudhery	84/F	Abdominal	CT: Locules of free	Laparotomy	4-quadrant	Isolated	Segmental
	et al.,17 2014		pain and	gas; picture of SBO		purulent		resection
			vomiting			peritonitis;		with primary
						micro-		anastomosis
						abscesses;		
						isolated		
						perforated		
						jejunal		
						diverticula due		
						to enterolith		
						(12x6 cm)		
18	Baksi et	55/F	Diffuse	AXR: free air under	Laparotomy	Isolated jejunal	Isolated	Segmental
	al.,18 2014	00/1	abdominal	the diaphragm	Laparotoriny	diverticula	15010100	resection
	ul., 2014		pain,			50 cm from		with primary
			constipation,			DJ flexure,		anastomosis
			and fever			with 1 cm		anastomosis
			and rever					
						perforation		
19	Hubbard et	Unknown/M	Abdominal	CT: large, calcified	Laparotomy	Multiple jejunal	Multiple	Segmental
	al., ¹⁹ 2015		pain	mass within the		diverticula;		resection
				lumen of the small		perforated		with primary
				bowel; enterolith		diverticula		anastomosis
				within the lumen		with a 4x5 cm		
				of the small bowel;		enterolith		
				mesenteric twist				
20	Natarajan	56/M	Abdominal	CT: multiple small-	Laparotomy	Purulent	Multiple	Segmental
	et al., ²⁰		pain,	bowel diverticula;		exudate with		resection
	2015		vomiting,	air under the		perforated		with primary
			and low-	diaphragm		jejunal		anastomosis
			grade fever			diverticula		
21	Sehgal et	82/M	Abdominal	CT: hollow viscus	Laparotomy	Multiple	Multiple	Segmental
	al., ²¹ 2016		pain, nausea,	perforation;		diverticula;		resection
			and low-	pneumoperitoneum		single		with primary
			grade fever			perforated		anastomosis
						jejunal		
						diverticula		
22	Gunta and	50/M	Abdominal	N/A	Laparotomic		Multiple	Sogmental
	Gupta and	50/14			Laparotomy	Multiple	multiple	Segmental
	Kumar, ²²		pain,			diverticula;		resection
	2017		nausea, and			sealed		with primary
			fever			perforated		anastomosis
						diverticula		

Table 1 continued.

23	Alvez	74/F	Abdominal	CXR: free air under	Laparotomy	Multiple	Multiple	Segmental
	Martins et		pain,	the diaphragm		diverticula;		resection
	al., ²³ 2018		nausea, and			perforated		with primary
			vomiting			jejunal		anastomosis
						diverticula,		
						20 cm from		
						ligament of		
						Treitz		
24	Authors'	60/F	Abdominal	CT: thickening and	Laparoscopy	Isolated	Isolated	Segmental
	case		pain, fever,	enhancing jejunal		perforated		resection
			and chills	loops; enterolith		jejunal		with primary
				within the jejunum		diverticula due		anastomosis
				with fat streaking		to enterolith,		
						40 cm distal		
						to ligament of		
						Treitz		

AXR: abdominal x-ray; CXR: chest radiograph; DJ: duodenojejunal; F: female; M: male; N/A: not applicable; RLQ: right lower quadrant; SBO: small bowel obstruction; US: ultrasound.

utilise the laparoscopic approach and hence minimally invasive surgery in the treatment of such a rare entity. Due to its rarity, diagnosis is usually delayed. The authors' case report highlights the importance of maintaining a high clinical suspicion of a perforated small bowel diverticulum in any patient presenting with an acute abdomen with localised signs of peritonitis. Furthermore, although non-operative management has been shown to be beneficial in stable patients, surgical exploration with segmental resection and primary anastomosis remains the mainstay of management. The authors believe that the acute onset of pain in absence of gross contamination of the abdomen was due to ischaemia, caused by the stagnated enterolith. Furthermore, the authors opted for laparoscopic approach as the patient was haemodynamically stable and this decreased the post-operative morbidity.

CONCLUSION

Jejunal diverticula is a rare entity, and their perforation is a challenge to the unaware. A delay in the diagnosis can be fatal, especially as this disease is more common in the elderly population with multiple comorbidities. Early surgical intervention when indicated is the key for successful treatment. While there is emerging evidence for the role of conservative management, the concern for progression to free perforation, especially in the setting of large, perforated diverticula, makes segmental resection the preferred intervention. Surgical approach, being laparoscopic or open, depends on the surgeon's expertise and available resources.

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