Indian J Surg (September–October 2009) 71:238–244
DOI: 10.1007/s12262-009-0077-5

REVIEW ARTICLE

Jejunal diverticulae: reports of two cases with review of literature

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Received: 7 May 2009 / Accepted: 21 July 2009
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Abstract

Introduction Jejunal diverticulosis (JD) is a rare disease of elderly people. Majority of diagnosed individuals are asymptomatic and found incidentally. The disease is clinically significant because of associated potential risk of serious complications. Due to the rarity and variable presentation of this clinical entity, diagnosis is often difficult and delayed, resulting in unnecessary morbidity and mortality. Clinical presentations, signs, diagnosis, complications and treatment of JD are discussed through a review of the literature and report of two cases.

Methods A literature review was done for analysis of diagnosis, treatment and complications of JD. Two cases of JD diagnosed and treated in our institution are also presented.

Conclusion JD is a rare disease which has variable presentations and thus poses a challenge to our diagnostic skills. Awareness about complications and presentation of the condition is needed for early detection and avoiding unnecessary mortality.

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Keywords Jejunal diverticulosis · Diverticulitis · Acute abdomen

Introduction

Jejunal diverticulosis (JD) is a rare disease of unknown aetiology which affects elderly people [1]. JD are false and acquired [2, 3] with a prevalence rate of 0.3–2.3% [4–6]. Majority of people with JD are asymptomatic or have minor, non-specific gastrointestinal (GI) symptoms, and found incidentally on imaging studies or exploratory laparotomy done for other reasons [3, 7]. The disease is clinically significant because of associated potential risk of serious complications [8]. Due to the rarity and variable presentation of this clinical entity, diagnosis is often difficult and delayed, resulting in unnecessary morbidity and mortality [4]. Here we discuss the clinical presentations, signs, diagnosis, complications and treatment of JD through a review of the literature and report of two cases diagnosed and treated in our institution.

Case reports

Case 1 A 74-year-old man presented with severe abdominal pain situated around umbilicus associated with vomiting of one day duration. Pain was situated in periumbilical region, continuous and severe in nature, and only slightly increased in severity after meals. There had been three episodes of vomiting and vomitous contained recently eaten food. Raised body temperature and tachycardia was appreciated. He had history of similar pain off and on which had been milder in nature. He had taken treatment on the lines of gastroenteritis. Total leucocyte count was raised

(TLC 19,300/mm^3). An upright abdominal radiograph was normal while ultrasound did not show more than sluggish peristaltic small bowel loops. Patient was initially managed conservatively with intravenous fluids and antibiotics. He improved and on 5th day of admission barium meal examination was done which revealed multiple diverticulae involving proximal jejunum (Fig. 1). His symptoms were attributed to an acute attack of diverticulitis, and antibiotic therapy continued for a week. Patient recovered uneventfully and discharged with advice of regular follow up. He had not suffered another attack for 16 months after discharge.

Case 2 A 65-year-old woman presented with 2-days history of upper abdominal pain associated with upper abdominal fullness, few episodes of vomiting and anorexia. Her past history was notable for intermittent abdominal pain and diarrhoea over previous 4 years. She had been labeled and treated as a case of irritable bowel syndrome during last 4 years by a local practioner. On examination, tenderness was present in epigastric and umbilical regions with fullness. There was no rigidity or rebound tenderness and bowel sounds were present. Leucocyte count showed leucocytosis (TLC 17,000/mm^3) and serum amylase was normal. Chest X-ray was normal and abdominal radiograph showed prominent, dilated small bowel loops in upper abdomen. Ultrasound of abdomen showed dilated fluid filled bowel loops in upper abdomen. A diagnostic laparoscopy was planned, which revealed multiple inter-loop adhesions as a cause of intestinal obstruction, along with multiple jejunal diverticulae involving its proximal 12 inch length of jejunum (Fig. 2). Rest of the bowel was normal. Laparoscopic adhesionolysis with resection of involved segment and jejuno-jejunal anastomosis by laparotomy, was performed. Patient made uneventful recovery.

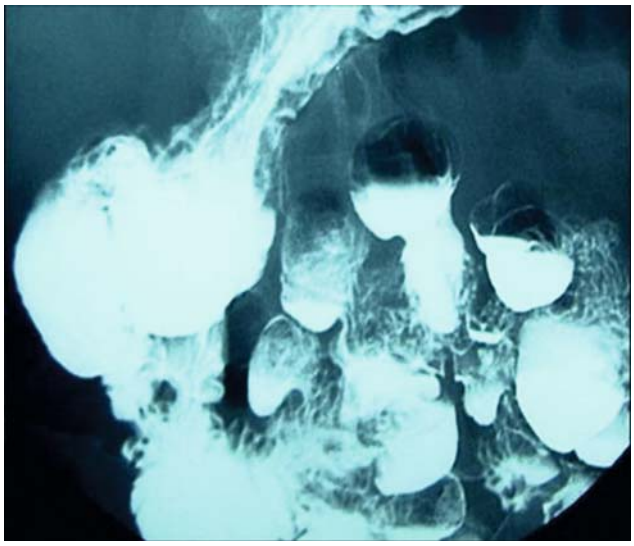

Fig. 1 Barium meal examination of a 74-year-old male showing multiple diverticulae involving proximal jejunum

Discussion

Diverticulae of the intestine may be divided into the true and the false [9]. By definition, the wall of a true diverticulum of intestine is composed of the entire thickness of the intestine; the false diverticulum, actually, on the other hand, represent a herniation of the mucosa and submucosa (Fig. 3) through the muscular coat of the intestine [1, 7, 9]. Therefore, a true diverticulum has all three coats of the gut, while a false one has mucosa and serosa only, with intervening connective tissue [9]. It is also necessary to differentiate between congenital and acquired diverticulae. Congenital diverticulae are true in nature, while acquired ones are mostly false [9]. But, a morphological classification of true and false, is easier and appears to be clinically more applicable. Hence, a true diverticulum can either be congenital or acquired, while a false one is always acquired. JD are false, thus acquired [2, 3].

Jejunal diverticulosis was first described by Sommering in 1794 and later by Sir Astley Cooper in 1807 [2]. It is a rare, acquired disease with a prevalence rate ranging from 0.3–1.3% in autopsy series to 2.3% in imaging studies of general population [4–6]. The actual incidence of reported cases is however lesser [10, 11]. Male-female ratio is 2:1 [12]. Tsiotos et al. analysed 112 clinically diagnosed cases of jejunoileal diverticulosis and found the incidence to be higher in men (58%) than women (42%) [13]. Harris

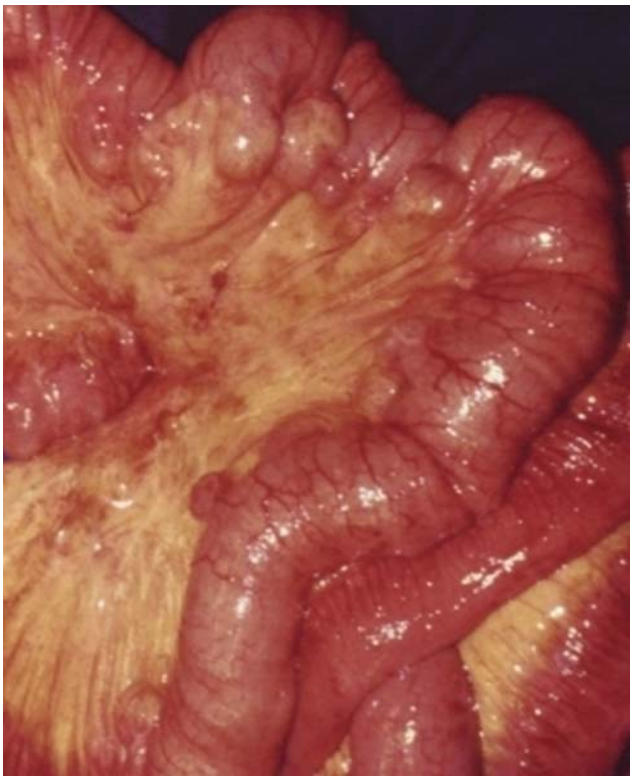

Fig. 2 Laparoscopic view showing multiple jejunal diverticulae involving proximal 12 inch length of jejunum

et al. reported incidence of JD in general population from 0.02–7.17% [14]. JD is a disease of elderly people. Old age favours the development of intestinal diverticulae as the incidence increases with age and over 80% of affected individuals are in 7th decade of life or older [13, 15, 16]. Chendrashekhar and Timberlake, found the average age of presentation to be 62 years and the incidence to be equal in both sexes [8].

Excluding duodenal and Meckel's diverticulum, proximal jejunum is the most common site for diverticulae in small bowel [16]. Edwards noted 75% of these located in the proximal jejunum while only 5% were found in ileum [1]. Predominance of diverticulae in jejunum has been attributed to the larger caliber of vasa recta of jejunum [17, 18] as JD are almost exclusively found on the mesenteric border, near the site of entry of blood vessels into the bowel wall [1, 3, 15, 19, 20].

JD can range from a few millimetre to up to 10 cm in size [20]. Diverticulae of jejunum tend to be larger in size than more distally situated diverticulae of small bowel [6]. Finding of multiple diverticulae as compared to single diverticulum is commoner [6, 14]. Fischer described a case of 85-years-old female with more than 400 diverticulae, mostly situated in jejunum [9]. Associated diverticulae in other parts of gastrointestinal tract and even urinary bladder is another part of the disease. Simultaneous diverticular disease can occur in, colon in 35–75% cases of JD, duodenum in (15–42%), oesophagus (2%), stomach in (2%) and urinary bladder (12%) [6, 9, 16, 21, 22].

The aetiology of JD is still not clear [1, 4]. Anatomic wall defect seems not to be the only factor [4]. Most SBD are thought to be pulsion lesions [16]. Weakening of the bowel wall when coupled with increase in pressure inside the bowel lumen leads to development of diverticulae known as 'Pulsion diverticulae' [4, 15]. Bowel wall weakening can be due to, dysfunction of smooth muscles or myenteric plexus in bowel resulting in jejunoileal dyskinesia or defect in the intestinal muscle coat [3]. Microscopic examinations of JD specimens have shown that fibrosis along with degeneration or decrease in number of normal muscle cells, or, de-

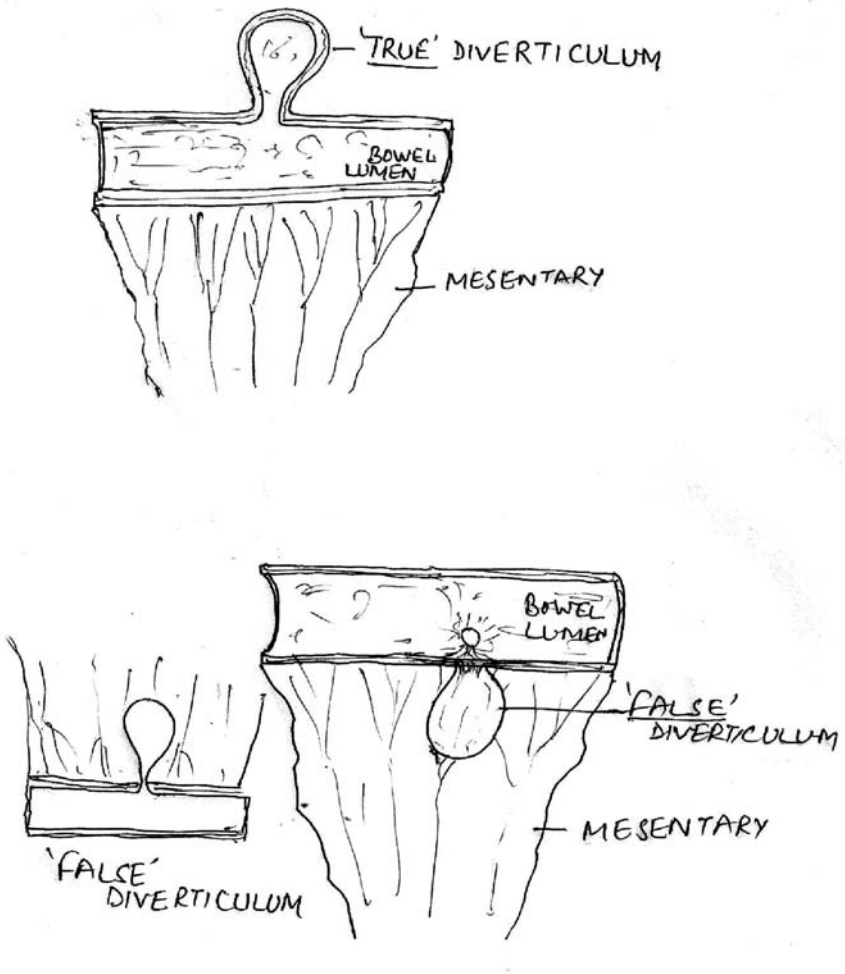

Fig. 3 Line diagram clearly differentiating true and false jejunal diverticulae. True diverticulum (upper diagram) with all layers of the bowel, while false one (lower diagram) with only mucosal pouting from in between the other layers of the bowel, and usually situated on mesenteric border

generation of neurons occurring in intestinal wall underlies these dysfunctions. Also to add, these histological findings are consistent with GI motility disorders like progressive systemic sclerosis, visceral myopathy and visceral neuropathies [7]. In fact, JD can be seen in elderly people with peristaltic disorders, such as progressive systemic sclerosis, leading to intestinal obstruction which in turn causes retention of faeces or gas in distal bowel, producing increased intraluminal pressure [4, 7]. Increased intraluminal pressure acts at the weakest points in the muscle layers of jejunum i.e. the site of entry of blood vessels on mesenteric border, and leads to herniation of mucosa [1, 3, 7, 15, 19, 20]. Association of these lesions with connective tissue disorders such as systemic lupus erythematosis, amyloidosis, Sjögren syndrome and GI scleroderma have also been recorded [13, 15, 19, 23–25]. Tsiotos et al. also found an association with Raynaud's phenomenon [13].

Clinical features of the condition are not constant. Majority of people with JD are asymptomatic or have minor, non-specific GI symptoms, and found incidentally on imaging studies or exploratory laparotomy done for other reasons [3, 7, 12, 26]. Tsiotos et al. retrospectively analysed 112 diagnosed cases of jejunoileal diverticulosis, and of these, 42% were asymptomatic [13]. If symptomatic, the most common symptomatology includes minor non-specific GI symptoms in the form of chronic vague abdominal pain, nausea and vomiting, alternating diarrhoea and constipation, weight loss, anaemia and steatorrhoea. A characteristic feature of this condition is the intermittent nature and, variable periodicity and severity of symptoms extending over many years. Intermittently occurring vague postprandial pain over epigastric or umbilical region with bloating sensation is usually the earliest symptoms [22]. These symptoms reflect functional pseudo-obstruction [12, 13]. Ten to 20% present with acute abdomen due to development of complications such as diverticulitis, fistula formation, GI haemorrhage, perforation and obstruction [3, 4, 6, 12, 13, 18, 26–29]. Haris et al. demonstrated complications in 10% of cases while rest were asymptomatic [14]. Kouraklis et al. [30] showed the incidence of different presentations to be as follows: abdominal pain 64%, chronic obstruction 10–25%, GI bleeding 15%, malabsorption 3.5–12% and perforation 2%. Among two cases presented in this article, patient in the first case presented with acute jejunal diverticulitis, while in second case JD was a coincidental finding on diagnostic laparoscopy. Therefore, presentation of JD is vague and diverse, and varies from a coincidental symptomless finding on imaging studies to acute emergencies; the fact that can lead to delay in diagnosis. Also, most of the common imaging studies give atypical appearance without key diagnostic features, which may not correlate with clinical symptoms. It is recommended that surgeons should be aware of this condition and its various ways of presentation, so that early clinical recognition could be made.

Laboratory findings in JD tend to be non-specific, but elevated WBC counts indicate acute diverticulitis or perforation. Plain abdominal imaging is frequently the first investigation done, but alone is not diagnostic in most cases [31, 32]. However, in symptomatic patients plain abdominal X-ray film can show distension of jejunal loops and air-fluid levels inside diverticulae [4]. Noble in 1971 described a triad consisting of obscure abdominal pain, anaemia and dilated small bowel loops on plain abdominal X-ray as highly suggestive of JD [33]. USG can show JD as multiple perintestinal hypoechoic structures; but is neither diagnostic nor preffered [34]. Upper GI radiological contrast studies using various forms of barium, like enteroclysis, are needed for specific diagnosis which will clearly show the presence of multiple diverticulae [4]. But these are contraindicated if acute diverticulitis or perforation is suspected. CT will give definitive diagnosis in such situations, and must be considered early [3]. It is the investigation of choice [3], and has been proven to be superior to barium studies for demonstrating acute inflammatory changes, and the mural, serosal, and mesenteric stranding and extent of disease itself or its complications [35]. CT also helps to rule out other conditions with which JD and its complications are frequently confused [36]. Capsule endoscopy and double-balloon enteroscpy are newer modalities of benefit in the diagnosis [37]. However, their use in emergency situations is limited [38]. Mesenteric angiography and RBC scan are very helpful in detecting the site of haemorrhage in case of haemorrhage from JD.

Complications of JD These often present a diagnostic dilemma and clinician must have a high index of suspicion, so that an operative intervention if needed can be done as early as possible. Chances of developing complications increase with increase in number of diverticulae [8].

Among complication of JD, acute diverticulitis is quite uncommon with frequency of 2.3% in known cases of JD [30], but has mortality rate as high as 24% [6, 8]. It is thought to develop secondary to obstruction of intestinal lumen which predisposes to bacterial stasis and localised inflammation. It has a non-specific presentation which is similar to other acute abdominal conditions like perforated peptic ulcer, acute appendicitis, pancreatitis, colonic diverticulitis and acute cholecystitis [26, 39]. Diverticulitis may, further complicate the situation by mechanical obstruction, perforation of bowel with peritonitis or fistulisation [39]. Diagnosis of jejunal diverticulitis is rarely made preoperatively, because of non-specific presentation that is similar to that of other acute abdominal conditions [26, 39]. Plain upright abdominal X-ray is not specific for jejunal diverticulitis. It can show intestinal obstruction and hollow viscous perforation. Upper GI radiological contrast studies using various forms of barium are needed for specific diagnosis which will clearly show the presence of multiple diverticulae [4, 40].

Acute intestinal obstruction in a case of JD is usually caused by adhesions and/or inflammatory stenosis caused due to repeated attacks of diverticulitis. Other possible

causes include pressure on the bowel by JD, intussusception, volvulus or enteroliths developed within diverticulae [3, 4, 6, 41].

Perforation of jejunal diverticulae is uncommon [42]. It usually occurs into the mesentery of involved part of jejunum leading to a contained perforation and development of a mesenteric abscess [43]. Perforation and haemorrhage are the result of progressive ulceration and erosion of mucosa, because of necrotising inflammatory reaction in case of acute diverticulitis [3, 4, 6]. Other common causative factors for perforation of JD are blunt trauma and foreign body imopaction [42].

Very very rarely malignancy may arise in a jejunal diverticulum. Although JD do not have a muscle coat, there may be a thin muscle coat layer which can give rise to a neoplasm. Ulrick et al. [44] reported a leiomyosarcoma arising from a jejunal diverticulum. A unique case of malignant fibrous histiocytoma arising from solitary jejunal diverticulum which had lead to perforation has also been described [45].

Management

No established criteria for the management of JD with or without complications are available. Management depends on patients symptoms. Asymptomatic patients diagnosed incidentally on routine contrast studies or at laparotomy for some other cause, do not require any treatment and can be kept on follow up and observed. Surgical treatment is not needed unless refractory symptoms or complications occur [8, 12, 41].

Conservative therapy is appropriate for symptomatic JD, in cases of chronic abdominal pain [21, 46]. It includes treatment of acute and chronic diverticulitis with chronic and intermittent antibiotic therapy which may or may not be followed by surgery [12].

The management of uncomplicated JD found incidentally on diagnostic laparoscopy in cases of undiagnosed abdominal pain, is again controversial. Literature has offered very little on this aspect of the topic. Authors suggest that chronic undiagnosed abdominal pain in such cases can be left alone if JD is uncomplicated, as the patients are usually elderly with some comorbidity [13, 29]. If adhesions are there, these should be lysed. If the symptoms are severe in a patient presenting for first time or if frequently recurring chronic symptoms make the patient to seek medical advice frequently, then the decision can be taken on the basis of laparoscopy findings. Multiple JD spanning over a short length of bowel and large diverticulae should be handled by resection anastomosis of involved segment, which should also be considered if no other reason is seen on laparoscopy other than JD, to blame for patients symptoms.

Surgical management is needed in approximately 8.5% of all patients of JD [47], and is frequently required for acutely symptomatic patients with complicated JD [28]. Resection of the involved segment with primary end-to-end anastomosis is mostly recommended in this setting [4, 19, 48–50]. Simple diverticulectomy is not recommended because it has been linked to postoperative leakage, sepsis and death [26]. Resection should be limited only to the involved segment or to the segment with largest diverticulum in case multiple diverticulae are scattered over a long segment of bowel [51]. Localisation of small JD which are frequently hidden in the mesentery, may be aided by insufflation of air into jejunum [50]. Laparoscopic approach is also being used for exploration and resection anastomosis [12, 50].

JD complicated by enteroliths with obstruction, are initially managed conservatively with decompression, rehydration and correction of electrolyte imbalances. Sugical treatment needs to be planned if conservative therapy fails. In most cases it consists of milking of enteroliths distally, crushing, or enterotomy and stone extraction [50, 52–54]. When these measures are not possible and/or there is associated bowel perforation or multiple diverticulae are present over limited part of bowel, resection and anastomosis is indicated [52–54].

Conclusion

JD is a rare, usually benign and clinically silent but potentially serious condition of old age. Rarity of this condition, usually silent nature, and non-specific presentation in symptomatic cases may pose a diagnostic challenge leading to missed or late diagnosis. Clinical awareness and high index of suspicion for early detection is the key to decrease the potentially possible complications and thus mortality. Elderly patients, with chronic vague abdominal symptoms, or recurrent symptoms need to be evaluated further.

Conflict of interest The authors do not have any disclosable interest

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