

Congenital diverticular disease of jejunum presenting as obstruction in adulthood: A case report from Ladakh

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Received - 29 December 2019

Initial Review - 14 January 2020

Accepted - 24 January 2020

ABSTRACT

Of the many causes of small bowel obstruction, volvulus of the midgut is seen mainly in paediatric age group only, along with malrotation of the gut. However, the volvulus of the small gut in an adult is rare. Volvulus of the midgut in adulthood could be due to many reasons like long mesentery, adhesive bands, internal herniation, endometrioses, etc. But volvulus occurring due to diverticulosis of small intestine is a rare occurrence. The diverticulosis in most of the adult patient are acquired (pulsion diverticuli) with a diverticular wall having only two layers of the intestine. Congenital diverticulosis with the diverticuli bearing all the layers of the intestine leading to the volvulus of the midgut makes it still rarer. We present the case of midgut volvulus arising due to congenital true jejunal diverticulosis in a 50-years-old adult along with a brief review of the literature. In our case report, volvulus occurred in an adult due to congenital diverticulosis of jejunum making it a difficult preoperative diagnosis and a rare case reporting.

Keywords: *Jejunal diverticulosis, Ladakh, Midgut volvulus.*

Small bowel obstruction arising due to midgut volvulus is a rare entity in an adult [1] and the occurrence of midgut volvulus arising due to multiple jejunal diverticulosis is very rare. Jejunal diverticulosis may remain asymptomatic unless complicated [1]. The incidence of jejunal diverticulosis is 0.3-1.3% in autopsy series and 2.3% in radiographical series [2]. The prevalence of the condition increases with age and peak during the sixth and seventh decade. Jejunal diverticulosis is of two types congenital (true) and acquired (false or pulsion). Most of the diverticuli reported so far in an adult are acquired with only two layers of bowel wall [3]. Congenital (true) jejunal diverticulum if present manifest either in childhood or even in adulthood due to complications secondary to it like bleeding, perforation, and obstruction from midgut volvulus [3,4,5,6].

Jejunal diverticuli causes volvulus when multiple diverticuli overloaded with fluid function as a pivot causing volvulus of the affected segment of the gut [5]. The importance of this condition lies in the fact that because of its rare occurrence, it is a not a common differential diagnosis for upper abdominal symptoms like postprandial fullness, pain, bloating, nausea, and dyspepsia, nor is it picked by usual diagnostic modalities like ultrasound and the upper gastrointestinal endoscopy. In most of the cases, the condition is picked up only on laparotomy when it presents itself as an acute abdomen due obstruction, perforation, haemorrhage, and diverticulitis. We present the case of midgut volvulus arising due to congenital true jejunal diverticulosis in an adult along with a brief review of the literature.

CASE REPORT

We report the case of a 50-years-old labourer who presented to the accident and emergency department of our hospital with complaints of sudden onset, severe colicky abdominal pain, centred around umbilicus associated with nausea and a repeated episode of vomiting. The vomitus contained ingested food particles initially and later contained bilious fluid. The pain was non radiating, non shifting and had no change in intensity with a change of posture. The pain started while he was straining to defecate and was not relieved by taking the usual proton pump inhibitor and the antispasmodic which he had been abusing for the past several years. Slowly over a period of about two hours, the pain which was intermittent initially had become continuous and he had a bloated sensation with an inability to pass flatus.

There was a history of recurrent pain abdomen in the past for which he used to take medication from a local practitioner but never visited the hospital before this episode. Retrospectively, he told about similar abdominal pain in his elder brother, but because of the lack of facility in his village, his brother succumbed to his illness. Since in our case the diverticulum on histopathological examination was true diverticulum, congenital nature of the diverticulum cannot be ruled out as his brother also died after a similar episode of pain and distension.

On arrival in the casualty, the patient was conscious, oriented and was rolling in pain. His vitals on admission were pulse 110/minute, blood pressure 140/100 and SPO₂ 86%. The examination

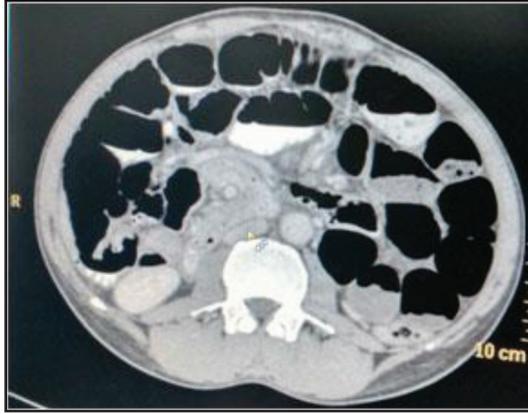


Figure 1: CECT film showing positive Whirl sign.

of the respiratory and cardiovascular system was unremarkable. Per abdominal examination showed that on inspection all quadrants of the abdomen were moving well with respiration, there was epigastric fullness, but no definite lump was noted. On palpation, there was generalised guarding and tenderness but there was no rebound tenderness. On auscultation, the bowel sounds were exaggerated. The digital rectal examination was unremarkable.

The patient was resuscitated and put on intravenous fluid and intravenous antibiotics. A battery of investigation was ordered which included complete blood count, bleeding time clotting time, renal function test, liver function test, and viral markers. X-ray abdomen showed multiple dilated small bowel loops with air-fluid levels seen predominantly in the left upper abdomen with no evidence of pneumoperitoneum. Ultrasound abdomen showed multiple dilated small bowels loop with increased peristalsis and free fluid in the lower abdomen. Contrast-enhanced computerised tomography (CECT) done after oral contrast showed multiple dilated small bowel loops (jejunal loops) predominantly in the upper abdomen with air-fluid levels (Fig. 1). Free-fluid was seen in the pelvis.

Relationship of the superior mesenteric vein and the superior mesenteric artery was reversed (superior mesenteric vein was seen on the left side and superior mesenteric artery on the right) along



Figure 3: Preoperative photograph showing multiple diverticuli from the mesentery side of Jejunum.

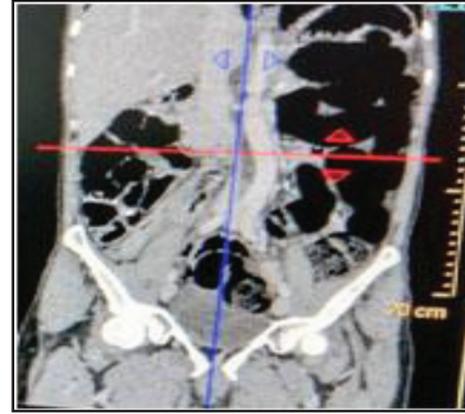


Figure 2: Rotation of the small gut to the left side around its mesentery.

with midgut volvulus (Fig. 1&2). Few round to oval-shaped air/ fluid/ contrast filled structures also seen adjacent to the proximal jejunum loop with thin imperceptible wall and absent fold, valvulae conniventes were seen which was suggestive of diverticulae.

The patient was subjected to laparotomy in view of clinical and radiological findings. There was minimal serosanguinous fluid in the pelvis. The proximal jejunum had rotated by about 500 degrees towards the left. The jejunum on derotation showed the presence of about 10 multiple small and large diverticuli arising from the mesenteric border of the gut, the smallest measuring 4 x 3cm and the largest was as big 6 x 7cms (Fig. 3). The diverticuli were located at about 40 cm from the duodenojejunal junction. The diverticulum bearing segment was as long as -40 cm. The rest of the gut was healthy. The affected segment was resected and end to end anastomosis was carried out.

Microscopically, the histopathological examination of multiple sections examined from the diverticula shows all layers of the intestine including the small intestinal mucosa, submucosa, muscularis propria and serosa consistent with true diverticulum. Lamina show moderate lymphoblastic inflammatory infiltrate admixed with few eosinophils. No ulceration, perforation and atypical cells were seen (Fig. 4).

The postoperative period was uneventful and the patient was discharged on 9th postoperative day. The patient was followed

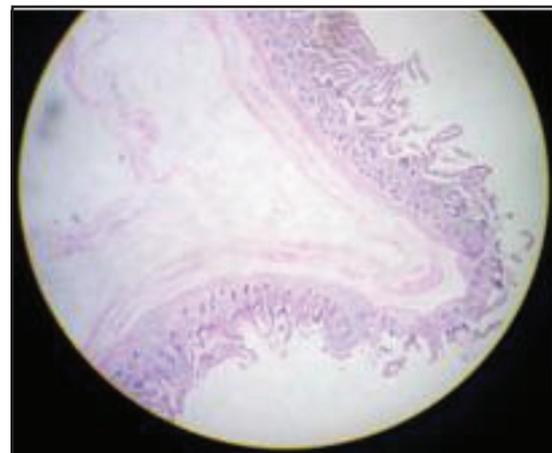


Figure 4: Histopathological examination of the resected specimen show diverticuli has all layers of the intestinal wall proving the congenital nature of the diverticuli.

up for three months at regular intervals during which he was symptom-free. Thereafter, he was lost to follow-up as he went back to his native place.

DISCUSSION

Volvulus refers to the rotation of the gut around its mesentery. Volvulus occur frequently in the large intestine (sigmoid and caecum) but volvulus of the small intestine is a rare entity [1]. Small gut volvulus is seen mainly in infants with malrotation of the gut. Midgut volvulus in an adult is a rare phenomenon and its occurrence secondary to jejunal diverticulosis is very rare. Rarer still is our case report of true (congenital) multiple diverticuli presenting in adulthood as midgut volvulus. To the best of our knowledge, till now only three cases of true jejunal diverticulum presenting with complication have been reported two cases in children and one familial case of jejunal diverticulosis [7-9].

Jejunal diverticulosis is a rare condition rarely diagnosed unless complicated [3-5]. It has an incidence rate of 0.3 to 4.5% in autopsy series and 0.5%-2.3% in radiographical series [2]. Prevalence of the disease increases with the age with peaking in the sixth and seventh decades. The rarity of this condition has been reported by Nataranjan K et al. According to them till the year 2015, only seven cases of jejunal diverticulosis has been reported so far in India [1].

The condition was described by Sommering in 1794 and later followed up by Astley Cooper in 1809. Jejunal diverticuli has been classified into true and false diverticuli. True jejunal diverticuli are mostly congenital in nature and consist of all layers of intestine while false diverticuli are herniation of mucosa and submucosa through the muscularis layer of the intestine. False diverticuli are also called pulsion diverticuli [6,7]. True/congenital jejunal diverticuli in some cases have shown to have familial predisposition [7]. These patients have multiple diverticuli over multiple sites including the esophagus and large intestine.

While false diverticuli has been attributed to abnormalities in peristalsis, intestinal dyskinesia due to motor dysfunction of smooth muscle or myenteric plexuses in the small bowel leading to high segmental intraluminal pressure and penetration of mucosa and submucosa through the weak mesenteric sites where the blood vessels penetrate the wall [7]. It is commonly seen in an individual with visceral neuropathy and visceral myopathy like Ehlers Danlos syndrome, Systemic lupus erythematosus, Scleroderma [8]. Congenital diverticuli and pulsion diverticuli could be solitary and are multiple, in our case the diverticuli in spite of being true were multiple arising from the mesenteric border contrary to reported cases of true diverticuli arising from antimesenteric border [7-10].

The majority of the jejunal diverticuli are incidental findings with many of them being diagnosed incidentally during barium studies/enteroclysis done for some other cause. Some of them are diagnosed during laparotomy/laparoscopy done for unrelated causes. These patients have clinical symptoms that are easily

misdiagnosed as dyspepsia/ irritable bowel syndrome. Patients with chronic post-prandial pain, nausea, vomiting, weight loss, alternating constipation, and diarrhoea, etc. should be actively evaluated for jejunoileal diverticuli if no other cause is found during the investigation. If done, so we will be able to pick up the cases of jejunoileal diverticuli before the patient developing complications.

Complication requiring surgical intervention is reported in 8- 30% of patients with jejunal diverticuli [11]. Complications include Volvulus, mechanical intestinal obstruction due to enterolith, intussusceptions, adhesive band formation from the previous episodes of diverticulitis, peritonitis due to perforation, gastrointestinal bleeding, etc [1-3,9-11]. Report of malignancy arising in the diverticuli is also there [12,13].

Volvulus, a common complication of jejunal diverticulosis which occurs when the fluid-filled involved heavier segment act as a pivot around which the gut rotates. In many cases, adhesive band arising from previous diverticulitis facilitate this rotation. Lobo et al [5,14] reported dynamic intestinal obstruction as the most frequent complication of jejunal diverticulosis which needs surgical intervention.

Most of the jejuna diverticuli even when asymptomatic can be diagnosed by barium studies that show cork-screw appearance. The enteroclysis is considered to be the best diagnostic modality for jejunal diverticuli. However, when complicated, CT is the investigation of choice and the Whirl sign on abdominal CT is highly suggestive of midgut volvulus [15]. Whirl sign refers to whirling or spiralling appearance of the mesenteric vessel which may accompany the intestinal loop and the feeding vessels [16]. Onangiographic studies, the barber pole sign is a significant finding of midgut volvulus.

Midgut volvulus presents as acute intestinal obstruction with a pain abdomen associated with nausea and a repeated episode of vomiting and constipation. In initial stages, the abdominal sign is minimal and if neglected, the patient presents with distension and peritonitis.

No established criteria have been described for the management of jejunal diverticuli. Asymptomatic cases which are picked up incidentally on routine contrast studies or during laparotomy for some unrelated condition do not require any treatment and kept on regular follow-up. Multiple jejunal diverticulitis spanning over the short segment of bowel and single solitary diverticuli are managed by resection and end to end anastomosis. Simple diverticulectomy is not recommended because it has been linked with postoperative leakage and sepsis [17]. If jejunal diverticuli are multiple and scattered over the large segment, resection would entail short gut syndrome. In such cases, resection should be limited to the symptomatic segment [17].

CONCLUSION

Jejunal diverticulosis should be suspected in all cases of abdominal pain so that, they are picked up early in the course of the disease so that we can reduce the mortality and reduce the morbidity.

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Funding: None; Conflict of Interest: None Stated.

How to cite this article: Deskit P, Badwal M, Dolma Y. Congenital diverticular disease of jejunum presenting as obstruction in adulthood: A case report from ladakh. *Indian J Case Reports.* 2020;6(1):41-44.

Doi: 10.32677/IJCR.2020.v06.i01.014