

Paediatric jejunal diverticulum masquerading as severe constipation with intestinal obstruction: A rare primary care encounter

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SUMMARY

Paediatric jejunal diverticulum is rare. It is characterised by the presence of sac-like outpouchings of the small bowel wall. Although mostly asymptomatic, it is called diverticular disease when it becomes symptomatic and causes gastrointestinal (GI) bleeding, abdominal pain, and bowel obstruction. We describe a young boy who initially presented to primary care with the complaints of chronic colicky abdominal pain and constipation refractory to medical therapy. As his abdominal distension worsened and radiological findings remained inconclusive, he was referred to a tertiary centre for an emergency laparotomy. A segment of jejunal narrowing was found adjacent to a diverticulum, causing partial bowel obstruction. After segmental jejunal resection, he made an uneventful recovery and was discharged well. Histopathology confirmed a true diverticulum. This case highlights the need for clinicians, especially in primary care settings, to hold a high index of suspicion to consider the diagnosis of small bowel diverticular disease when patients of the paediatric age group presented with constipation with atypical features.

INTRODUCTION

Jejunal diverticulum (JD) is a rare entity. Historically, it has a reported incidence of 2% in small bowel contrast studies and 0.7% in autopsies.^{1,2} Diverticulum can be classified as true and false. In the paediatric population, JDs are sometimes true diverticula that are congenital and involve the herniation of all layers of the gastrointestinal (GI) tract, forming a sac-like outpouching over the small bowel.¹ Contrastingly, a false diverticulum is acquired and more common among adults, in which a weakness in the muscularis propria layer allows the protrusion of mucosa and submucosa outward through the small bowel wall.^{1,2} JDs are usually an incidental finding during surgery or radiological investigation done to assess other pathologies.¹⁻³

JDs are mostly asymptomatic.^{2,3} However, when symptomatic, JDs can present with a wide spectrum of clinical features, from minor complaints like colicky abdominal pain and constipation, to life-threatening complications including diverticulitis, perforation and bowel

obstruction.¹⁻⁶ These potential sequelae can be fatal and often require urgent surgical intervention.³⁻⁶

JDs are exceedingly rare and tend to affect the elderly more.¹⁻⁵ Only a handful of paediatric JDs have been described worldwide.^{3,6} Due to its exceeding rarity, the pathophysiology of paediatric JDs is not thoroughly understood.^{2,4} This presents a diagnostic challenge, especially to primary care providers in resource-limited centres without radiological or surgical capacity. Therefore, treating clinicians must recognise alarming clinical features and consider early referral to tertiary centres for escalation of care in a timely manner. This case illustrates a rare encounter of a child with JD, who initially presented to primary care with constipation that was refractory to medical treatment, followed by intestinal obstruction and eventually required surgical resection.

CASE PRESENTATION

A 3-year-old boy was brought to primary care by his mother with complaints of colicky abdominal pain for 4 weeks. It was associated with worsening abdominal distension, constipation, and reduced flatus. Otherwise, the child remained well with no vomiting or other clinical features suggestive of infection or malignancy. He experienced similar symptoms two months ago, which resolved spontaneously within two days. He had previously had multiple visits to the local clinics and emergency department with similar complaints, but was discharged with laxatives. Otherwise, the child was born full term with no known antenatal complications or medical illness. Serial abdominal examination demonstrated a mildly distended abdomen but was otherwise unremarkable. He was discharged with syrup lactulose and glycerine enema.

The patient's symptoms worsened despite medical therapy. Besides the persistent constipation, the child developed postprandial vomiting with poor oral intake and worsening abdominal distension. He exhibited severe distress during episodes of abdominal colic, which was subsequently relieved after bowel opening. Otherwise, he was haemodynamically stable with good hydration status. Repeated abdominal

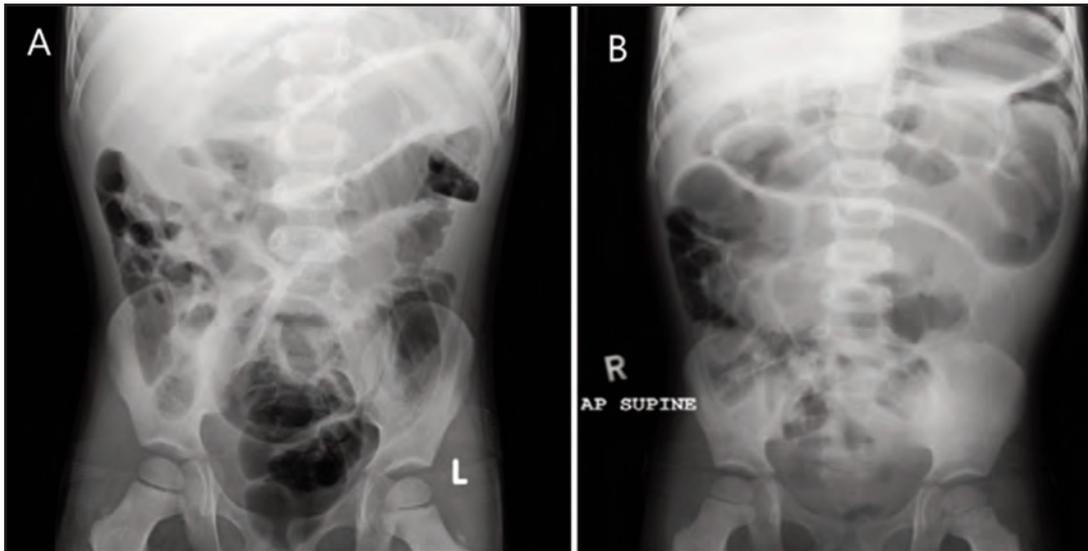


Fig. 1: Images of 2 abdominal plain films as the patient's abdominal pain worsens. Image (A) is the abdominal X-ray done in a local health clinic to show dilated bowels with faecal matters and image (B) was done before surgery to show dilated small bowels.

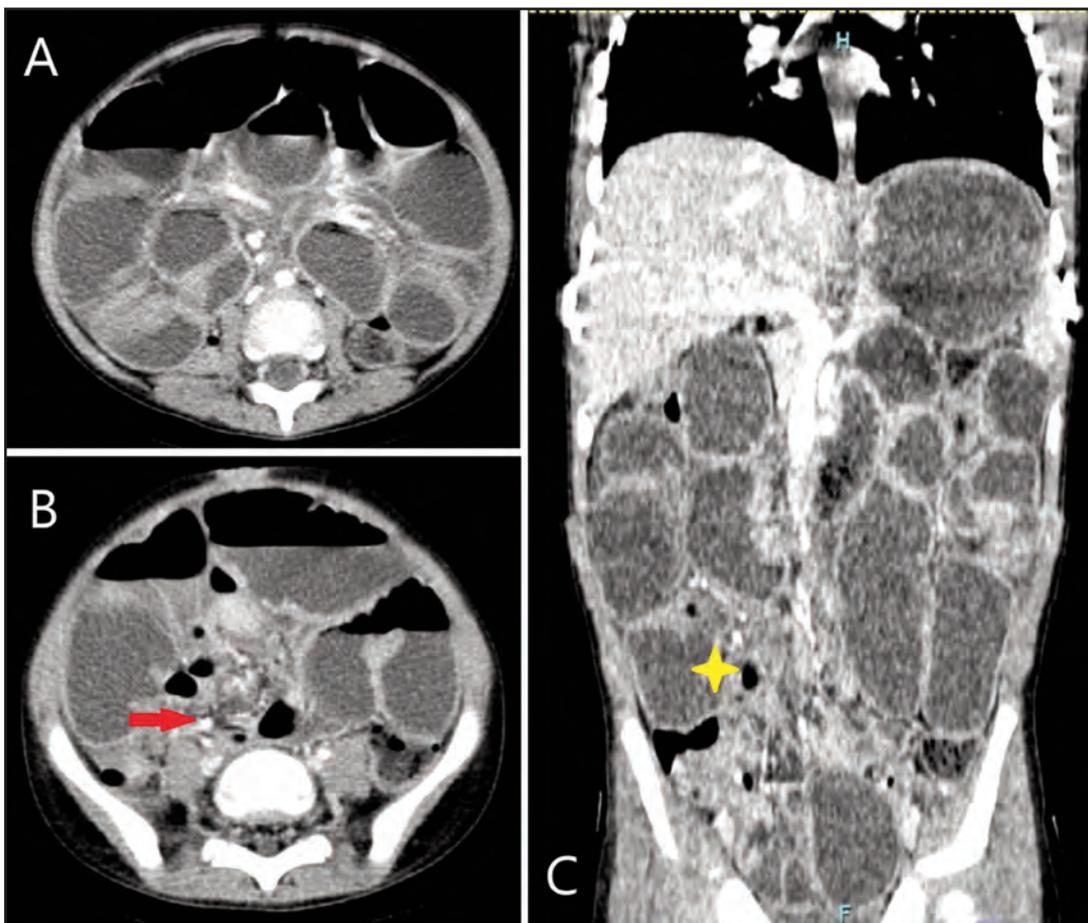


Fig. 2: Images of pre-operative contrast CT of abdomen and pelvis of the child. Images (A) and (B) are the axial CT view, showing dilated small bowels. In image (B), the transition point of the small bowel is seen at the right side of the pelvis, with surrounding mesenteric lymphadenopathy as shown with the red arrow. The anatomical landmark of the transition zone was further confirmed, as shown with a yellow star, in the coronal CT plane in image (C).

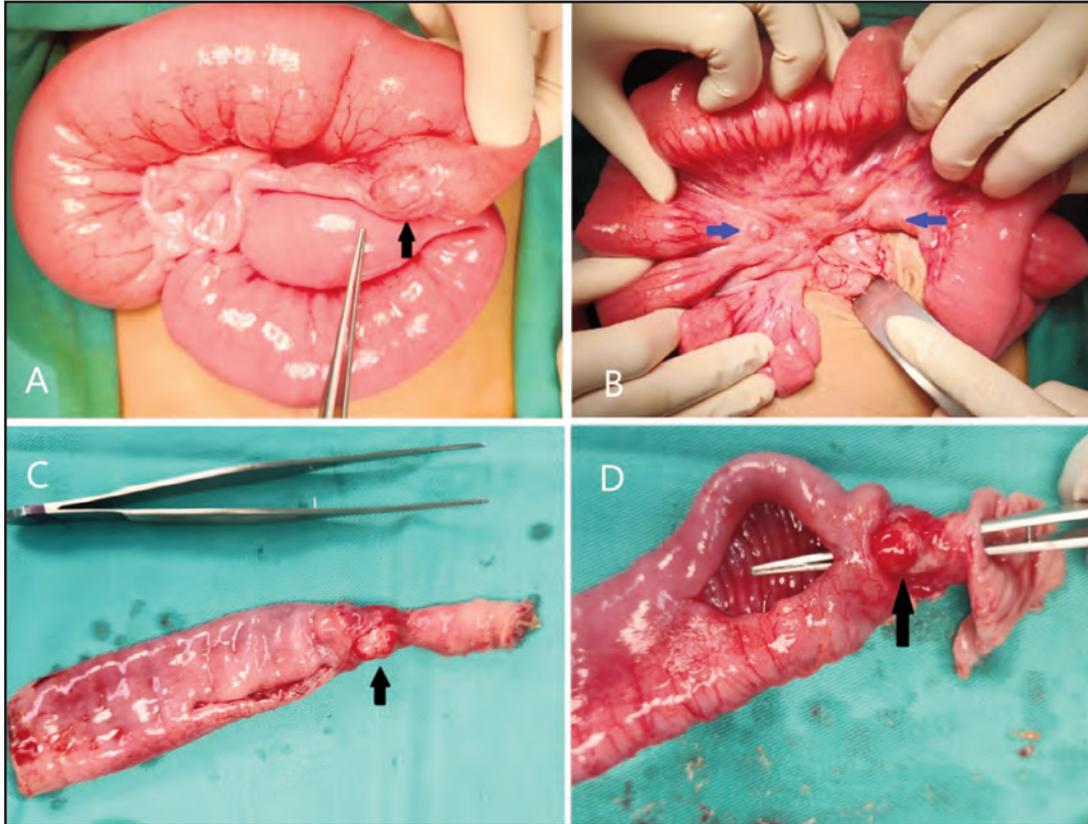


Fig. 3: The intraoperative images of a jejunal mass. Image (A) shows the mass of size 1.0cmx1.5cm as marked with a black arrow, at the antimesenteric border of the jejunum with a collapsed small bowel segment distal to it. Enlarged mesenteric lymph nodes can be seen as marked with blue arrows in image (B). Images (C) and (D) show the specimen of segmental jejunal resection, including the mass. It appears round, contracted, causing circumferential stenosis of the bowel adjacent to it, causing partial obstruction. The calibre of the jejunal lumen was so narrowed that it allowed only the passage of the tips of the forceps, as shown in image (D).

assessment did not find evidence of peritonism. Bowel sounds were normal. Plain abdominal radiograph showed dilated bowels with impacted stool. Due to multiple visits and constipation despite medical therapy, the diagnosis of bowel obstruction secondary to faecaloma was made, and the child was referred to a tertiary centre for specialised care.

During the admission, the child passed out only a small amount of stool despite regular sodium phosphate enema and rectal washout. Moreover, he had worsening colicky abdominal pain and distension. Abdominal ultrasound (USG) showed dilated bowels but was inconclusive. Contrast computed tomography (CT) showed dilated small bowels with a transition zone at the right iliac fossa, surrounded by adjacent mesenteric lymphadenopathies. No normal appendix was visualised. Large bowels were normal with no faecaloma. No bowel diverticulum was noted on CT. At this point, the mother also recalled that the child had an episode of blunt injury to his abdomen one month ago. Considering the atypical clinical trajectory and radiological suspicion, the differential diagnoses at this point were small bowel obstruction secondary to possible delayed bowel injury, perforated appendicitis, or a duplication cyst. A decision was made for emergent laparotomy. Intraoperatively, an oval-shaped mass with the size of 1cm x 1.5cm was seen at the segment of distal jejunum, midway between the

duodenojejunal (DJ) flexure and the ileocecal valve (ICV), at the antimesenteric border. It appeared contracted, causing circumferential narrowing of the adjacent jejunum and serving as a transition zone of the obstruction (Figure 3). The small bowel proximal to the constricting point was grossly dilated and distally collapsed. There was surrounding mesenteric lymphadenopathy. Otherwise, the bowels appeared viable. No adhesion, tumour or Meckel's diverticulum was identified. The diseased segment was resected, and primary anastomosis of the jejunum was performed.

The histopathological assessment of the diseased jejunal segment revealed the outpouching of all the layers through the bowel wall, which is consistent with the presence of a true diverticulum. There was no ectopic tissue seen within its lumen. No features suggestive of chronic inflammation or perforation were noted.

Post-operatively, the child made a swift recovery. He tolerated oral feeding without abdominal pain and distension and was discharged from the ward within 1 week. There was no evidence of wound complications or intra-abdominal collection upon 1-month follow-up. His constipation resolved and he was able to have bowel motion daily.

DISCUSSION

Paediatric jejunal diverticulum (JD) is rare and is characterised by the presence of sac-like protrusions or pouches (diverticula) in the jejunum in children.^{1,4,6} It can be acquired or congenital. The congenital diverticula are true diverticulum, which is composed of all layers of the intestinal wall, distinguishing them from the false diverticulum. The formation of congenital diverticulum stems from developmental anomalies during foetal growth.^{1,3,4}

The most important barriers to diagnosing this case at primary care are the non-specific clinical features combined with the rarity of paediatric JD. Notably, constipation is a ubiquitous complaint among the paediatric population. It would be difficult to attribute constipation, a common paediatric ailment, to the sequelae of a JD in a primary care setting. Constipation in children is commonly seen with a reported prevalence of varying degrees.⁸ The prevalence of constipation or functional constipation in children varies according to different research, ranging from 1.1% to 31.4%.^{8,9} Additionally, children with constipation also often present with abdominal colic which is typically non-specific and poorly localised.⁹ Additionally, frequently changing history from caretakers can be misleading for clinicians to detect alarming features of this patient.

Besides that, the aetiology of paediatric JD is also not well understood.^{1,3,6} Moreover, due to the non-specific features of diverticular disease, it presents a diagnostic challenge to primary care providers with a reported prevalence of diagnosis of 0.1% of primary care consultations.⁷ Certain genetic syndromes such as Marfan's syndrome, cystic fibrosis and other genetic disorders, may predispose children of younger age to develop diverticular disease. So, in such a group of children presenting with non-specific abdominal symptoms, primary care doctors should consider JD as a differential diagnosis, despite its rare prevalence.

Notably, the current management algorithm of diverticular disease is largely based on the understanding of the pathophysiology of colonic disease in adults.⁵ This is because it is more common in adults, especially among the elderly. The adult diverticular disease is acquired and caused by the formation of false diverticula in the large bowel.⁵ Due to increased intraluminal pressure and abnormal intestinal motility, a weakness or defect in the muscularis propria occurs, allowing the herniation of mucosa and submucosa outward through the bowel wall.⁵ The stretch of the layers contributes to the structural weakness of the bowel wall and chronic thinning of the surrounding vessel wall, making it prone to complications like perforation and bleeding.⁵

However, such pathophysiology does not apply in diagnosing and managing paediatric JDs. Many cases remain asymptomatic and are discovered incidentally during imaging or surgical procedures for unrelated conditions. When symptomatic, they tend to present with non-specific features like vomiting, bloating, altered bowel habits and abdominal colic. It can mimic other GI disorders like constipation, perforated appendicitis, and Crohn's disease. However, paediatric JDs can also cause potentially fatal complications like GI bleeding, perforation and obstruction,

which warrants urgent surgical intervention.⁶ Some paediatric JDs can also present with diverticulitis, sepsis and peritonitis.^{3,6}

Another important barrier of diagnosis in this case is also the lack of advanced radiological assessment modality in primary care. Generally, many diagnoses of paediatric diseases are often made clinically with minimal radiological assessment to reduce the unnecessary exposure to radiation risk for children. However, the diagnosis of paediatric JD often involves a combination of clinical suspicion supported by radiological assessment. This includes imaging techniques like CT scans which carries radiation risk, GI contrast study that may induce contrast allergy and intraoperative bowel examination which is invasive.^{1,2} Moreover, very few JDs can be diagnosed with CT due to the difficulty in delineating it from the bowel loops.² While GI contrast study and advanced endoscopic techniques like video capsule endoscopy can be useful, it would be inappropriate and unsafe in this patient due to the development of bowel obstruction. Most importantly, all these are not available in resource-limited settings or in local clinics.

Management of paediatric JDs depends on the severity of the condition and the presence of complications.^{1,4,6} Surgical resection is not needed if they are asymptomatic and an incidental finding. However, in the development of complications, surgical intervention is necessary. The most common surgical approach is segmental resection of the affected portion of the small bowel, followed by primary anastomosis to restore intestinal continuity. Laparoscopic surgery is increasingly favoured due to its minimally invasive nature and faster recovery times. While the minimally invasive technique is favourable, it is more suitable in elective cases of paediatric JDs. It was not performed in this patient because the laparoscopic port insertion and manoeuvre may injure the already dilated bowels. They also take up a larger space within the intraabdominal cavity, which can reduce the viewability of laparoscopic surgery. Overall, long-term follow-up is recommended to monitor of complications after surgery.

In this case, the initial differential diagnosis of a duplication cyst was nullified after histopathological confirmation of the presence of a true diverticulum. The possibility of Meckel's diverticulum was briefly considered. Meckel's diverticulum is a true diverticulum that is a remnant of an incompletely obliterated vitelline duct and has a completely different aetiology than a true JD.¹⁰ Notably, this diverticulum was found in the jejunum, which is not typical of Meckel's diverticulum, which is located two feet from the ICV.¹⁰ It has no ectopic mucosa within. It is also round, short and contracted, distinguishing it from the typical appearance of a Meckel's diverticulum that is often two inches long.¹⁰

Some debates remain that histopathology surprisingly showed no evidence of chronic inflammation. Intraoperative specimens showed a contracted diverticulum, causing the luminal stenosis of the jejunal segment and causing obstruction. This is likely the sequelae of chronic inflammation of the JD, causing contracture and scarring of the surrounding tissue. We postulate that even as the chronic

inflammation over the JD has subsided, over time, the healing process has caused the gradual and ongoing contracture and lumen stenosis. This gives rise to the recurrent and insidious onset to the child's constipation and abdominal distension.

In summary, paediatric JD is a rare but potentially significant condition that requires a high index of suspicion for diagnosis. Its nonspecific symptoms and potential for severe complications underscore the importance of prompt and accurate diagnostic evaluation. Advances in imaging and surgical techniques have improved outcomes for affected children, but further research is needed to better understand the condition's pathogenesis and optimal management strategies. Early recognition and appropriate intervention are critical in ensuring favourable outcomes and minimising morbidity in this rare paediatric disorder.

Recognition of alarming features

The alarming features of abdominal distension should prompt primary care physicians to consider differential diagnoses, beyond functional constipation. As illustrated in the case above, there are instances in which the attending health care professionals may have missed the alarming features of constipation during the initial clinical evaluation, leading to the child recurrently failing pharmacological treatment and multiple visits with no clear diagnosis. Health care professionals, especially those at primary care settings are the patients' first contact to the healthcare system. They must be aware and capable of identifying alarming features based on thorough history taking and physical examination. This would help to improve patients' prognosis by prompting an early referral to the tertiary centre for escalation of therapy and further investigation.

CONCLUSION

We have encountered a rare case of paediatric jejunal diverticulum which presented initially as constipation and progressed to develop intestinal obstruction. Although constipation is commonly seen in children, recognition of alarming clinical signs and symptoms based on thorough history taking and physical examination in primary care settings is crucial, to ensure early and appropriate referral to tertiary centres is made in a timely manner.

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DECLARATIONS

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