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JEJUNAL DUPLICATION IN A 10-YEAR-OLD BOY PRESENTING WITH GENERALIZED OEDEMA AND ABDOMINAL DISTENSION

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ABSTRACT

Background: Gastrointestinal tract duplications are rare congenital anomalies, with the jejunum being the second most common site after the ileum. These duplications can vary in size, location, and presence of ectopic mucosa, significantly impacting clinical presentation and often requiring surgical intervention.

Case presentation: A 10-year-old Thai boy with haemoglobin (Hb) E trait and iron deficiency anaemia presented with significant oedema and abdominal distention. Laboratory investigations revealed isolated hypoalbuminemia. Imaging studies, including computed tomography (CT) scan, demonstrated a large, loculated, rim-enhancing air-fluid collection adjacent to the jejunum, along with mild dilatation and wall thickening of the affected jejunal loop. The patient underwent exploratory laparotomy, which included jejunal resection and jejunojejunal end-to-end anastomosis.

Conclusion: Jejunal duplications are uncommon in children and can present diagnostic challenges, potentially leading to serious complications. This case underscores the importance of considering jejunal duplication in paediatric patients with unexplained abdominal distention and protein-losing enteropathy. Early recognition and timely surgical management are crucial to prevent severe complications and improve patient outcomes.

KEYWORDS

Jejunal duplication, pediatric surgery, hypoalbuminemia

LEARNING POINTS

- Jejunal duplications are rare congenital malformations, which can cause a range of symptoms, including bilious vomiting, abdominal pain, and obstruction.
- Our patient presented with iron deficiency anaemia, generalized oedema, including swelling of both legs, scrotum, and abdominal distension.
- The diagnosis was made by computed tomography scan.

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INTRODUCTION

Alimentary tract duplications are extremely rare congenital malformations, occurring in approximately 1 in 25,000 live births[1,2]. These duplication anomalies can develop anywhere along the gastrointestinal tract, from the oesophagus to the colon[2-5]. They are generally classified as either cystic or tubular, with the cystic form being the most prevalent [6,7]. Despite their structural resemblance to the gastrointestinal tract, duplications may contain ectopic mucosa, such as gastric or pancreatic tissue, which may potentially cause bleeding or ulceration[5]. While alimentary tract duplications in children are usually benign, their clinical manifestations range from incidental findings to severe complications, including abdominal pain, distension, nausea, vomiting, and abdominal mass formation[6-8]. More severe but less common presentations include intussusception, bowel obstruction, malabsorption, and protein-losing enteropathy. The symptoms primarily depend on the size, location, and presence of ectopic mucosa. Reports indicate that 75% of duplications are located within the abdominal cavity[8]. Although ileal duplications are the most prevalent 53%[3,7-9], jejunal duplications present unique diagnostic issues challenges due to their nonspecific symptoms, making early identification difficult.

This case report presents a rare instance of tubular jejunal duplication in a paediatric patient, emphasizing the diagnostic challenges, the role of imaging, and the successful surgical management that led to symptom resolution.

CASE DESCRIPTION

A 10-year-old Thai boy with a history of haemoglobin (Hb) E trait and iron deficiency anaemia presented with generalized oedema, including swelling of both legs, scrotum, and abdominal distension. Two years prior, he experienced a similar episode of isolated hypoalbuminemia (albumin 1.5 g/dl, stool alpha-1 antitrypsin 18.59 mg/dl), which resolved with albumin transfusion and anthelmintic therapy. Upon admission to Naresuan University Hospital, his weight and height were 24.2 kg and 124 cm, respectively. Physical examination revealed abdominal distension with a positive fluid thrill but no peripheral oedema. Laboratory tests showed transaminitis (aspartate aminotransferase 71 IU/l, alanine aminotransferase 77 IU/l), and urinalysis revealed trace protein without significant abnormalities in the protein-creatinine ratio. Imaging studies, including abdominal X-ray and contrast-enhanced computed tomography (CT) scan, revealed a large, loculated, rim-enhancing air-fluid collection in the mid-abdomen, suspected to be connected to the jejunum. Mild dilatation and wall thickening of the jejunal loop were also observed. An upper gastrointestinal (GI) series with small bowel follow-through confirmed the presence of a large, contrast-filled cystic structure occupying most of the abdomen and obscuring the small bowel, with delayed small bowel transit time *(Fig. 1)*. Esophagogastroduodenoscopy revealed mild reactive changes in the stomach but no evidence of primary lymphangiectasia. Due to severe abdominal distension, colonoscopy was deferred. Given the progressive distension, the patient underwent an exploratory laparotomy, which confirmed a jejunal duplication. The affected segment was resected, followed by jejunojejunal anastomosis *(Fig. 2)*. Histopathological examination of the resected segment confirmed the diagnosis of jejunal duplication, with benign mucosa, focal ulceration, and granulation tissue *(Fig. 3)*.

DISCUSSION

Jejunal duplication is a rare congenital malformation of the gastrointestinal tract that can present with a range

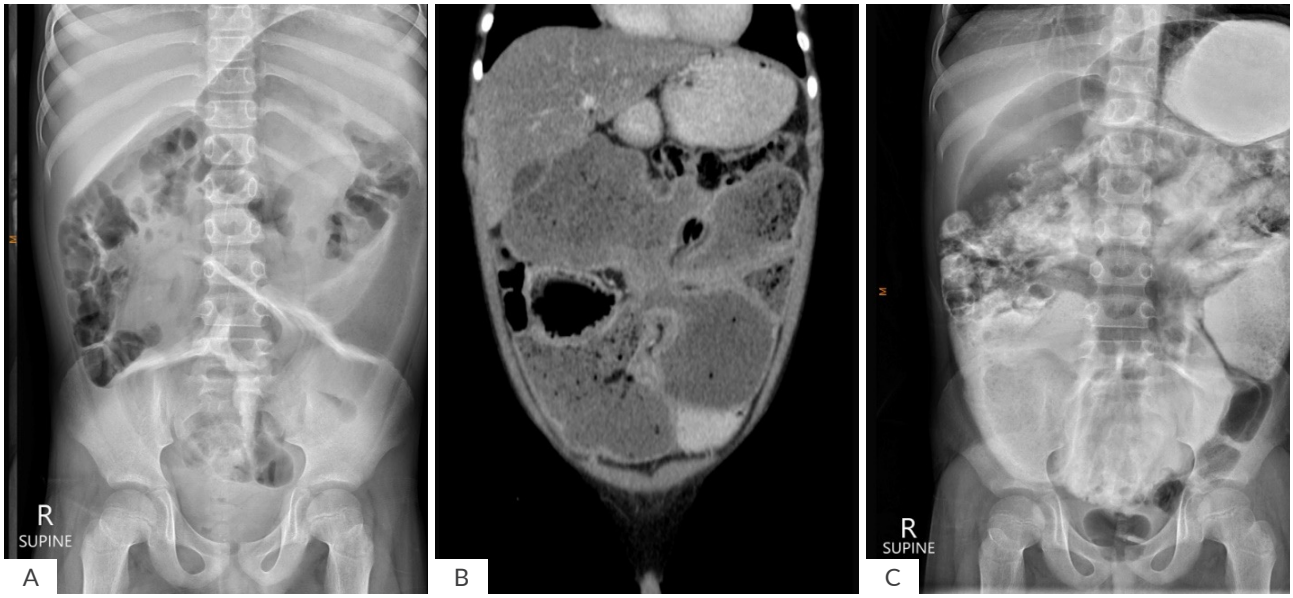


Figure 1. Abdominal radiology findings. A) Abdominal X-ray showing small bowel dilatation; (B) Computed tomography scan of the abdomen revealing a large, loculated, rim-enhancing air-fluid collection with a suspected connection to the jejunum at the mid-abdomen, along with mild dilatation and wall thickening of the jejunal loop; (C) Upper gastrointestinal study with follow-through demonstrating a contrast-filled, large loculated cystic structure occupying the entire abdomen, obscuring the small bowel, and causing delayed small bowel transit time.

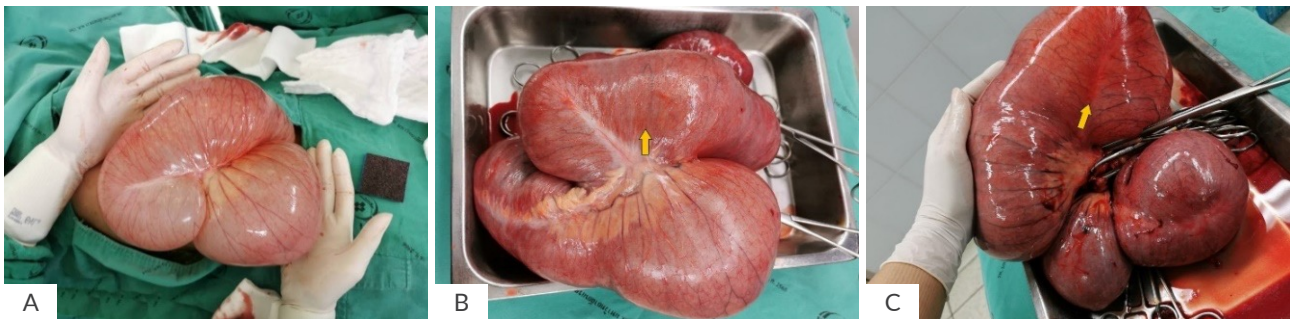


Figure 2. Gross findings of the dissected jejunal duplication segment. A) Markedly dilated loops of the small intestine sharing a common wall and mesentery at the jejunum, leading to an intraoperative diagnosis of tubular jejunal duplication; B) The native bowel is bifurcated into the tubular duplication (arrow) and the native intestine. C) Small bowel duplications that share a common wall (arrow) with the native intestine, arising from the mesenteric side and sharing a common blood supply.

of symptoms[10], depending on its size, location, and the presence of ectopic mucosa. It is typically diagnosed in infancy or early childhood[5,7,9], with symptoms varying from asymptomatic to various manifestations, including abdominal pain, abdominal mass, nausea, vomiting[5], and more rarely, bowel obstruction[1], haematochezia, malnutrition, and protein-losing enteropathy. Jejunal duplications should be considered in the differential diagnosis of paediatric patients, particularly those under 2 years of age[1,7], who present with abdominal pain, distension, vomiting, or failure to thrive[5]. This case highlights the importance of timely recognition and surgical intervention in symptomatic cases. Jejunal duplications can be classified into two types: cystic and tubular [6], with tubular forms being more likely to cause symptoms. One study reported that 9% of alimentary tract duplications involve the jejunum, and all cases were of the cystic type[10]. In our case, the patient had a tubular jejunal duplication, leading to abdominal distension, vomiting, and malnutrition due to its effect on intestinal motility and absorption. In contrast, cystic duplications may be incidentally detected during imaging and often remain asymptomatic unless they become large enough to exert pressure on surrounding structures or become infected. The pathophysiology of jejunal duplications is believed to result from abnormal embryonic development between the 4th and 8th weeks of gestation[2,7,9]. Proposed mechanisms include incomplete recanalization of the primitive gut tube and failure of gut splitting during development, leading to the formation of a duplicated segment[3,9]. These duplications can communicate with the native intestine, sharing a common muscular layer and blood supply[2,8]. Other theories, such as the abortive twinning theory and the intrauterine vascular accident theory, have been proposed but remain controversial[2,9]. The presence of ectopic tissue, such as gastric mucosa, may contribute to complications like ulceration or bleeding, further complicating the clinical presentation.

Liaqat et al.[4] conducted a retrospective study of 8 ileal duplication cases, all of which involved tubular or cystic types. Most cases presented with intestinal obstruction, but none exhibited symptoms of abdominal distension or protein-losing enteropathy. Mucosal ulceration within the duplicated segment may contribute to protein-losing enteropathy[7]. In our patient, tubular jejunal duplication was suspected based on imaging studies. Plain radiographs were nonspecific, and abdominal ultrasound findings were inconclusive, likely due to overlying bowel gas and a low index of suspicion[3]. Contrast-enhanced CT scan and an upper gastrointestinal series revealed a large, contrast-filled cystic structure in the abdomen, leading to delayed small bowel transit time.

These findings underscore the critical role of advanced imaging techniques in diagnosing alimentary tract duplications, as they help identify key features, such as air-fluid levels, bowel wall thickening, and loculated cystic masses[8]. Exploratory laparotomy confirmed the diagnosis of tubular jejunal duplication. Surgical resection of the duplicated segment resulted in complete resolution of the patient's symptoms. Surgical management remains the cornerstone of treatment for symptomatic jejunal

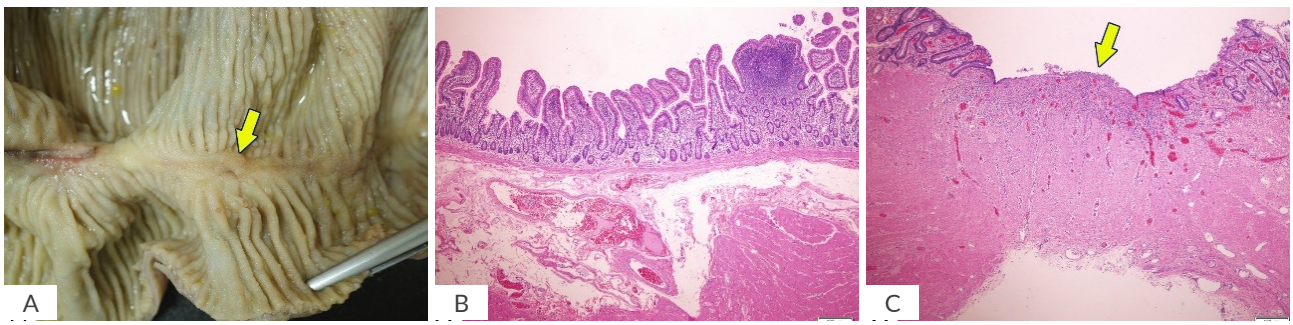


Figure 3. Pathologic findings of the resected intestinal segment. A) Dilated intestinal wall with ulceration (arrow); B) Histopathology showing a normal small intestinal wall (H&E 10×); C) Area of ulceration and granulation tissue (arrow) (H&E 10×).

duplications[1,7]. The primary goal is complete resection of the duplicated segment while preserving healthy bowel tissue. In our case, a jejunojejunal anastomosis was performed following resection. The postoperative course was uneventful, and the patient showed significant improvement in both his abdominal symptoms and nutritional status. Surgical approaches typically involve either end-to-end or side-to-side anastomosis, depending on the location and size of the duplication. Long-term follow-up is essential to monitor potential complications, including bowel obstruction, adhesions, and malabsorption[4]. Associated anomalies have been reported in 16-76% of cases and may lead to complications such as bleeding, erosion, or malignant degeneration[10]. Although malignant transformation is rare, it remains a potential long-term risk[9]. A critical aspect of managing jejunal duplications, particularly in the postoperative period, is addressing potential nutritional deficiencies. The duplicated segment may interfere with nutrient absorption, leading to hypoalbuminemia, as observed in our patient. Following surgical resection, the patient's nutritional status improved significantly. Paediatric patients with gastrointestinal anomalies require close nutritional monitoring, including a high-protein diet, enteral nutrition, or oral protein supplements as necessary. Regular follow-up of growth parameters and serum albumin levels is recommended to ensure proper recovery and development. Complications following surgery for jejunal duplication are rare but may include bowel obstruction, anastomotic leaks, infections, and malabsorption. While the patient in this case did not experience significant postoperative complications, the risk of intestinal adhesions remains a concern, particularly in paediatric patients. Regular follow-up visits, imaging studies, and nutritional assessments are essential for the early detection of potential complications.

This case highlights the importance of considering jejunal duplication in the differential diagnosis of paediatric patients presenting with unexplained abdominal distension, proteinuria, or failure to thrive. Early recognition through imaging studies, followed by appropriate surgical intervention, can prevent long-term complications and improve patient outcomes. Although jejunal duplications are rare, they should not be overlooked, as timely diagnosis and treatment are essential for optimal management and resolution of symptoms.

CONCLUSION

This case of jejunal duplication in a young child highlights the importance of including considering rare gastrointestinal anomalies, such as jejunal duplications, in the differential diagnosis of paediatric patients presenting with unexplained abdominal distension, oedema, and protein-losing enteropathy. Imaging modalities, including contrast-enhanced CT scan and upper GI series, are crucial for identifying these anomalies. Early detection and surgical excision of symptomatic duplications can result in significant clinical improvement and prevent serious complications. Furthermore, long-term follow-up is necessary to monitor potential postoperative complications and ensure optimal patient outcomes.

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