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Sphacelation with auto-anastomosis of the intestine: a rare outcome of intussusception in a child—a case report

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Abstract

Background: Intussusception is the telescoping of a proximal segment of the bowel into a distal segment. It can be idiopathic or pathological. Children commonly present with colicky abdominal pain, vomiting, a palpable abdominal mass, and bloody stools. Our case describes the unusual presentation of bowel sphacelation with auto-anastomosis in a child with intussusception and its clinical progression.

Case presentation: A 3-year-old boy with underlying stage IV rhabdomyosarcoma of the left orbit presented with high-grade fever and diarrhea for 1 day. He was treated for neutropenic sepsis in view of low absolute neutrophil count and recent history of chemotherapy. During his admission, he developed abdominal distension, high bilious aspirates, and diarrhea with bloody stools. Abdominal X-ray showed dilated bowel loops. Impression was septic ileus with coagulopathy. He was treated with blood transfusion and bowel rest. On the 6th day of illness, he passed out a tubular structure per rectum which was confirmed to be a segment of gangrenous bowel by histopathological examination. A diagnosis of intussusception with bowel sphacelation was made. He was treated conservatively, and his obstruction was resolved. He was discharged well with no abdominal symptoms during follow-up.

Conclusion: Intussusception is a common cause of small bowel obstruction in children. A high index of suspicion of intussusception should be maintained in children presenting with vomiting and bloody stools complemented by ultrasound to avoid missing this diagnosis. Sphacelation of the intussuscepted bowel with auto-anastomosis is a rare presentation of intussusception with a favorable outcome.

Keywords: Intussusception, Gangrene, Sphacelation, Bowel auto-anastomosis, Case report

Background

Intussusception is the telescoping of a proximal segment of the bowel into a distal segment. It is a common cause of bowel obstruction in children 5 months to 3 years old. The etiology maybe idiopathic or pathological. Idiopathic intussusception accounts for 90% of cases and is believed to be caused by enlarged Peyer's patches [1]. Pathological intussusception occurs in 2–12% of cases in the presence of a surgical lead point. The probability of a pathological lead point increases with age, and the success of non-surgical reduction is low [2].

The classical presentations of intussusception are colicky abdominal pain, vomiting, red currant jelly stools, and palpable abdominal mass [3]. Prognosis is excellent with early detection and treatment with radiological reduction. However, if undiagnosed, intussusception may lead to mortality in 2–5 days. In rare instances, sphacelation with auto-anastomosis of the bowels occurs with a resolution of symptoms [4].

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Case presentation

A 3-year-old boy presented with fever, reduced oral intake, and diarrhea for 1 day. He has underlying stage IV rhabdomyosarcoma of the left orbit with lung metastasis. He underwent his 4th cycle of chemotherapy a week prior to the onset of symptoms. He was febrile with a temperature of 38.8 °C and respiratory rate of 20, lethargic, and dehydrated. The lungs were clear, and the abdomen was soft, non-tender, and not distended. His blood investigations showed Hb 8.4 g/dL, TWC 0.17 × 10^3/μL, ANC 0.06, platelet 6 × 10^3/μL, CRP >156 mg/L, urea 4.1 mmol/L, sodium 126 mmol/L, potassium 3.9 mmol/L, and chloride 89 mmol/L. Blood culture grew *Escherichia coli* (ESBL producer). He was treated for neutropenic sepsis and was started on intravenous fluids maintenance and replacement, meropenem, and granulocyte colony-stimulating factor. Packed cells and platelets were also transfused.

His general condition improved after initiation of treatment; however, on day 2 of admission, he developed abdominal distension, vomiting, and diarrhea with watery mucous-like stools. There was no abdominal pain. He was able to sit up comfortably in bed. The abdomen was distended, soft, and non-tender with no guarding. Abdominal X-ray showed dilated small bowel loops (Fig. 1). An impression of septic ileus was made, and he was treated conservatively with bowel rest and nasogastric tube insertion with free flow and four hourly aspiration.

On the 4th day of admission, he started to pass out bloody stool. Gastric aspirates were coffee ground. Despite daily platelet transfusions, he remained thrombocytopenic with a platelet of 2 × 10^3/μL. The coagulation profile was prolonged with INR 1.82, PT 20.9 s, and APTT 38.4 s. He was treated for coagulopathy secondary to sepsis. Coagulopathy was corrected with transfusion of platelets and fresh frozen plasma.

On day 6 of admission, he passed out a tubular structure per rectum which resembled a segment of necrotic small bowel in his diapers (Fig. 2). His abdomen remained soft and distended with no signs of peritonitis. Ultrasound abdomen showed bowel wall thickening with poor peristalsis. A diagnosis of intussusception with bowel sphacelation with auto-anastomosis was made. He was observed for signs of peritonitis and was continued on bowel rest for a week. TPN was initiated. Abdominal distension gradually resolved, bowel opening was brownish colored stools, and he was able to tolerate orally. Repeated abdominal X-ray showed no dilated bowels with normal bowel gas distribution (Fig. 3). He was discharged home after resolution of sepsis and was well at 6 months follow-up. Histopathological examination showed an acute-type extensive full-thickness (transmural) ischemic necrosis of the bowel wall (Fig. 4).

Discussion

The earliest descriptions of intussusception could be found in the Hippocratic Writings [5]. Bonetus in his volume *Sepulchretum* (1679) complied the autopsy findings of ileus in which descriptions of intussusception were made by Colombus (1494 to 1557), Barbette (1629–1557), and Morgagni (1682–1771) [6]. John Hunter (1728–1793) can be credited for the first detail description of the pathological features of intussusception. During this period of Renaissance, the term ileus was used to describe midgut volvulus, intussusception,

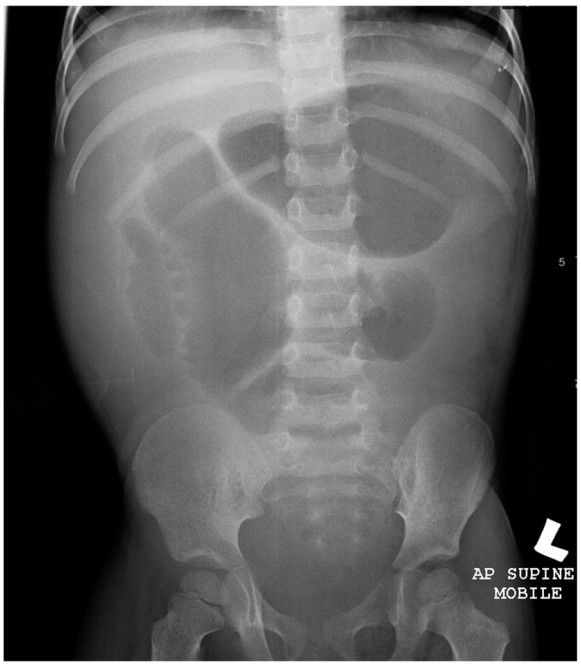

Fig. 1 Abdominal X-ray which showed dilated small bowel loops

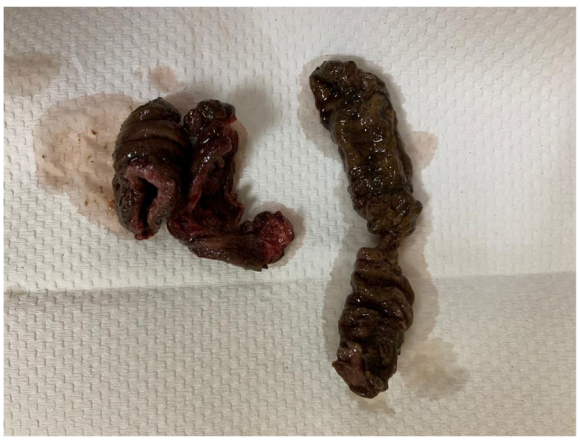

Fig. 2 Tubular structure passed out on day 6 of illness which resembled necrotic small bowel

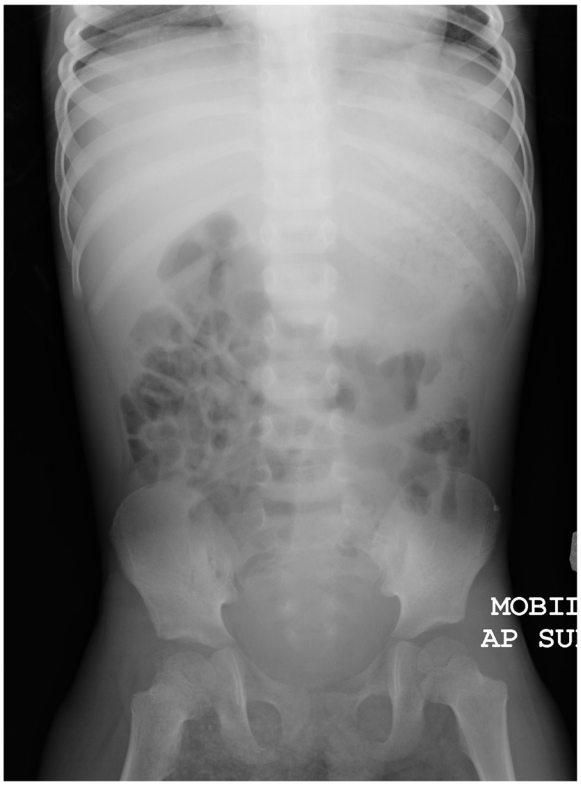

Fig. 3 Repeated abdominal X-ray which showed normal bowel gas distribution with no dilated bowel loops

and incarcerated hernias due to their similar clinical presentation of abdominal pain, obstipation, and fecal vomiting [7]. The outcome of intussusception was almost always fatal [8]. There were only a few historical reports of recovery after sphacelation with auto-anastomosis of the bowel [9].

The pathophysiology of sphacelation was first described by Treves (1899). Invagination of the intussusceptum together with its bowel mesentery leads to compression and kinking of the vessels. Early in the process, lymphatic drainage is obstructed. As edema develops and pressure builds up within the intussusceptum, venous congestion occurs followed by arterial impairment and infraction of the intussuscepted bowel. Treves demonstrated that gangrene was maximum at the area of greatest constriction which is at the neck of the intussusception. Perforation commonly occurs with peritonitis. However, if the ischemic process is gradual, it allows for adhesions to form at the neck between the serosal layer of the intussusceptum and the intussuscepien forming an auto-anastomosis. Other complications that may occur after sphacelation are stricture or perforation at the line of separation, bleeding, obstruction caused by the gangrenous bowel, and recurrence of intussusception [10].

Many significant advancements were made over the past century on the pathological understanding, diagnostic methods, and treatment of intussusception. Harald Hirschsprung was an early adopter of hydrostatic pressure reduction of intussusception and had reported 107 cases with a mortality of only 35% in 1905 [11]. In 1913, Lehmann was credited for the first contrast enema demonstrating an intussusception [12]. Robert Gross was a strong advocate of surgical reduction. In 1953, he showed how mortality had declined from 60 to 0% over 40 years in 702 cases [13]. The adoption of pneumatic reduction was stimulated by a study of 6396 cases in China which reported a higher success rate compared to hydrostatic reduction [14]. In 1977, the introduction of ultrasonography led to the description of the target sign and pseudo kidney sign in intussusception [15]. Ultrasound is now widely used as the main diagnostic and

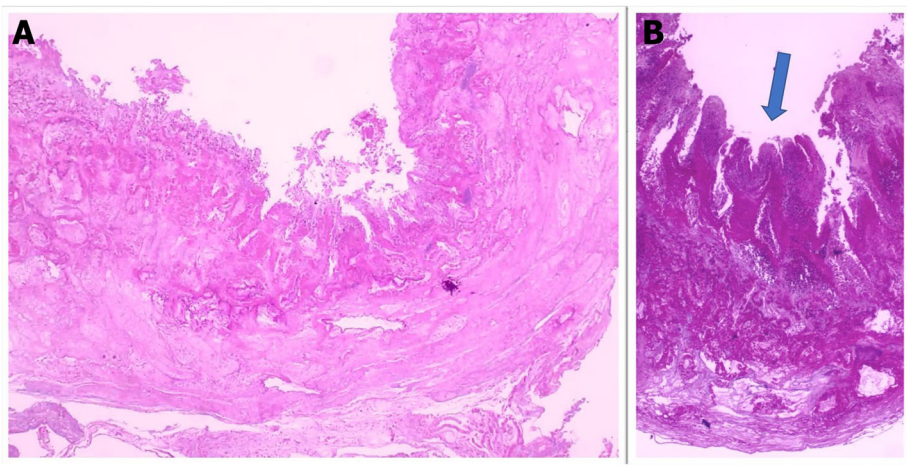

Fig. 4 Microscopic images showing acute-type extensive full-thickness (transmural) ischemic necrosis of the bowel wall. No viable area was observed on histology. Note the ghost-like outline of the previous small bowel villi (arrow). H&E stain. **A** ×20 and **B** ×40 magnification

therapeutic method of reduction because it avoids exposure to radiation, it is minimally invasive, and it is repeatable with high sensitivity and specificity [16]. Today, the outcomes of intussusception are excellent if diagnosed and treated early.

Sphacelation of an intussuscepted bowel is extremely rare and unheard of in today's practice. The last reported case of autoamputation of an intussuscepted bowel was in 1984 in an adult patient [17]. Our present case demonstrates a unique presentation of intussusception. Contrary to the fact that the majority of intussusception present in well-nourished children, frequently after a viral illness [12], our patient presented ill with neutropenic sepsis after recent chemotherapy. The classical symptom of colicky intermittent abdominal pain was absent, and other nonspecific signs of distension and gastrointestinal bleeding were thought to be attributed to his primary condition. In retrospect, supportive management led to the improvement of his general condition which allowed time for the process of sphacelation and auto-anastomosis to occur. Considering his generally ill condition (sepsis, immunocompromised, coagulopathy), operative intervention would have a high risk of morbidity or mortality. This child was very fortunate as he recovered from this ordeal without any untoward sequel.

Conclusion

Intussusception is a common cause of small bowel obstruction in children. A high index of suspicion of intussusception should be maintained in children presenting with vomiting and bloody stools complemented by ultrasound to avoid missing this diagnosis. Sphacelation of the intussuscepted bowel with auto-anastomosis is a rare presentation of intussusception with a favorable outcome.

Abbreviations

ESBL: Extended spectrum beta-lactamase; INR: International normalized ratio; PT: Prothrombin time; APTT: Activated partial thromboplastin time; TPN: Total parenteral nutrition; H&E: Hematoxylin and eosin

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Authors' contributions

SKT: conception, design of work, acquisition, analysis of the data, and preparation of the manuscript. JAB: acquisition and analysis of the data and drafting. PP: acquisition, analysis of the data, and critical review. CWT: acquisition and analysis of the data and critical review. JH: analysis of the data and critical review. MAN: design of the work, analysis of the data and critical review, and final approval of the manuscript. All authors have read and approved the final manuscript.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written consent had been obtained from the parent of the study participant.

Competing interests

The authors declare that they have no competing interests.

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