Unusual radiological findings of pediatric jejunojejunal intussusception: A case report

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SUMMARY

Intussusception is a common cause of intestinal obstruction in children, especially in those of age <5 years. The typical signs and symptoms of this condition is colicky abdominal pain, bloody mucous stool, and palpated abdominal mass, with a classic target sign finding on abdominal ultrasound. In older children, the symptoms may vary, which necessitates investigation of the cause of intussusception, as it is often caused by a pathologic lead point. We report here the case of a 14-year-old girl with total bowel obstruction, hematochezia, a very dilated reverse C-shaped bowel loop, and intestinal pneumatosis on abdominal X-ray. During laparotomy, we detected jejunojejunal intussusception caused by jejunal polyp. After bowel resection and anastomosis, the patient recovered well and had no other events during follow-up.

INTRODUCTION

Intussusception is a condition in which one segment of the intestine telescope inside of another to cause obstruction, and it most commonly recorded in children aged 6 months to 3 years. Although it is mostly idiopathic, 4% of all cases may be caused by a pathological lead point (PLP) that is usually found in children of age >5 years or in adolescents.[1] Intestinal pneumatosis (IP) is described as an abnormal intramural gas of the digestive tract. The presentation of PI may suggest the occurrence of life-threatening conditions, such as gastrointestinal perforation, ischemic bowel, and bowel necrosis.[2] We report here the case of a 14-year-old girl with bowel obstruction and a very dilated reverse C-shaped bowel loop and IP on abdominal X-ray caused by jejunojejunal intussusception.

CASE REPORT

A 14-year-old girl visited the Emergency Department (ED) of Soetomo General Hospital, Surabaya, Indonesia with intermittent colicky pain for the last 6 days, especially at the epigastric and left abdominal areas. The pain lasted for 5–10 min at each episode. Nausea and vomiting also occurred, eventually progressing to bilious vomiting. The patient did not have any bowel movements for the past 6 days and admitted to enable her to pass gas. At the ED, she had hematochezia with no mucous. Previously, she was admitted to a secondary hospital and then sent to our hospital.

She was weak and somnolent at the time of admission, tachycardic (172 beats per min) with raised body temperature to 38.3°C. The nasogastric tube production was bilious, 300 mL within 12 hours. There was no abdominal distention, bowel contour, or bowel movement. The bowel sound was decreased with muscular guarding. Digital rectal examination revealed collapsed ampulla, no mass, with fresh colored blood on the gloves. Laboratory findings were normal, but her C-Reactive Protein (CRP) level was raised to 27.2 mg/L (normal value: <6 mg/L). Central venous catheter insertion, fluid resuscitation, and broad-spectrum antibiotic administration were accordingly performed. A plain abdominal X-ray showed a reverse C-shaped dilated bowel with massive IP (Fig. 1). Further abdominal CT scan study revealed bowel obstruction due to jejunojejunal intussusception.

We performed exploratory laparotomy and found bloody peritoneal fluid and jejunojejunal intussusception with necrotic intussusception of the bowel, which could only be partially released. The intussusceptum was also necrotic with multiple perforations. We resected 48 cm of the jejunal segment from 10-cm distal to the Treitz ligament and performed end to end jejunojejunal anastomosis. After resecting, the bowel was opened, and we detected a jejunal polyp. We palpated the rest of the bowel and found no other polyp.

Histopathological examination revealed ulcerated jejunum with a wide ischemic and bleeding site, most of which were filled with lymphocytes, histiocytes, erythrocytes extravasation, and infarcted polyp. Multiple trapped air bubbles were found in the submucosa (Fig. 2). The polyp could not be evaluated further because it was probably already necrotic for quite some time, albeit there was no malignant cell.

After surgery, the patient has put nil per month for 2 days due to postoperative paralytic ileus, and oral feeding was started on the 3rd postoperative day. She received 3rd generation of cephalosporin and analgesics. The patient recovered uneventfully after 5 days of recovery time. She recovered well and, at 2 weeks of follow-up, the wound healed nicely. She had no gastrointestinal symptoms at 6 months and 2 years follow-up.

DISCUSSION

Intussusceptions are a common cause of acute abdomen in

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Table I: Conditions associated with pneumatosis intestinalis

Pulmonary Asthma COPD Emphysema Bronchitis Cystic Fibrosis	**Drug Induced** Corticosteroids Chemotherapy agents Lactulose Sorbitol Chloral hydrate	**Organ transplantation** Bone marrow e.g. for leukemia Kidney, Lung, Liver Graft versus host
Gastrointestinal IBD Diverticulitis Colitis Necrotizing enterocolitis Enteritis Toxic Megacolon Appendicitis Intestinal Obstruction Bowel stenosis Adynamic Ileus Malignancy Peptic Ulcer Celiac sprue	**Infectious** HIV and AIDS Virus (CMV, rota-, adeno-, varicella- zoster virus) Candida albicans Mycobacterium tuberculosis	**Autoimmune and systemic** Lupus variants Polymyositis Dermatomyositis Polyarteritis nodosa Scleroderma Sacroidosis
Vascular Mesenteric vascular disease Intestinal infarction and ischemia	**Iatrogenic** Blunt abdominal trauma Endoscopy Postsurgical intestinal anastomosis Jejunoileal bypass Barium enema Enteric tube placement PEEP ventilation	**Idiopathic (primary)**

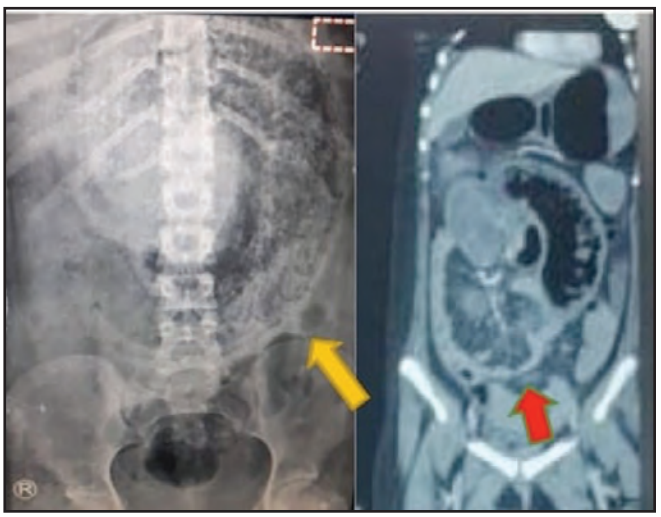

Fig. 1: (left) abdominal X-ray showed reverse C shaped dilated (yellow arrow) bowel and multiple IP ;(right) abdominal CT scan revealed jejunojejunal intussusception (red arrow) with dilated proximal duodenojejunum.

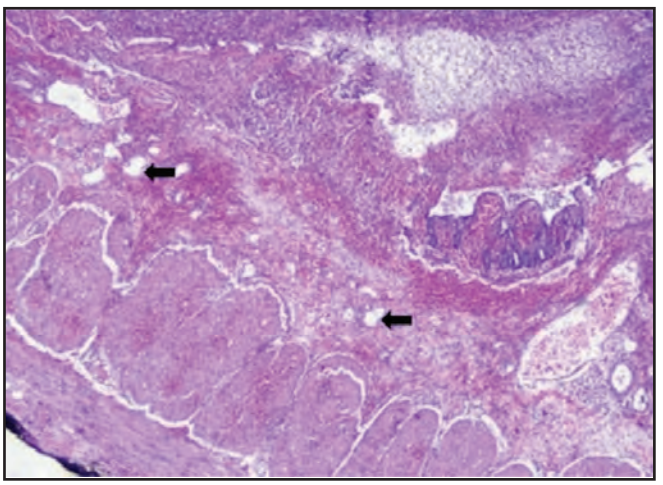

Fig. 2: Air bubbles (black arrow) trapped in the submucosal area (HE staining).

children. In our case, the patient presented with acute abdominal pain, a sign of bowel obstruction, and rectal bleeding, which should have raised a high index of suspicion for a life-threatening condition due to bowel strangulation. After rapid resuscitation, subsequent acute abdomen imaging studies were performed without delaying the definitive surgical treatment.

The abdominal X-ray revealed large reverse C-shaped bowel consistent with the dilated third portion of the duodenum, duodenojejunal junction, and proximal jejunum. The fixed retroperitoneal located distal duodenum, traction from the Treitz ligament, and narrow proximal jejunal mesentery made this reverse C-appearance possible. At the end of the reverse C-shaped bowel, we detected soft tissue mass opacity indicating intussuscepted bowel. There were also multiple linear and cystic-shaped lucency on the wall of proximal dilated part of the duodenum and jejunum, and the also in the walls of the jejunal intussusception, which was consistent with IP.

Computerized Tomography (CT) imaging is a common examination modality performed to detect IP as well as other signs of digestive pathology. The absence of bowel enhancement is a specific finding associated with ischemia that occurs during the late stage. IP with the associated absence of bowel enhancement, bowel wall thickening, mesenteric fat stranding, and ascites are prominent findings that indicate surgical situation.[2] In this case, the abdominal CT scan confirmed the X-ray findings, showed dilatation of the stomach, duodenum, and proximal jejunum. The intussuscepted bowel mass was also clearly depicted at the distal portion of the reverse C-shaped bowel. The heterogenous hypodensity at the proximal and medial portion of the C-shaped bowel may constitute the overlapping intraluminal gas, bowel wall edema, and multiple IP.

A massive intramural gas detected in this patient was considered as a rare finding in intussusception. It may appear in the intramural layer in a cystic, bubbly, curvilinear, or linear shape. In general, IP is classified into 2 types, primary and secondary IP3. Primary IP, found in approximately 15% of all IP cases, is a benign condition that is usually asymptomatic and caused by respiratory diseases, systemic diseases, after organ transplants, pathological process, endoscopic procedures, immunological imbalance, mucosal disruption, and other intraabdominal pathology. The management of primary IP depends on the cause and, not every case requires surgical management.[1,4] On the other hand, secondary IP is described as an air collection, forming a linear pattern that reflects on a pathological condition. Pear, in 1998, divided the cause of IP into 4 big groups: bowel necrosis, mucosal disruption, increased mucosal permeability, and pulmonary disease; while St. Peter divided IP based on pathology that reflected the origin of the gas: intraluminal gastrointestinal gas, bacterial gas, and pulmonary gas.[5] Various conditions associated with IP are described in Table I.

In our case, the IP was secondary, probably due to bowel mucosal disruption, caused by both leaked intraluminal gastrointestinal gas and gas-forming bacteria due to bacterial translocation after a long period of bowel obstruction that leads to distention and ischemia.

Intussusception in an older child is usually caused by PLP. It is different from the idiopathic intussusceptions that usually occur at ileocolon, the predilection of intussusceptions with PLP are at the caecocolic (2.5%) and jejunojejunal (2.5%).[1] In this patient, we detected a jejunojejunal intussusception caused by a jejunal polyp. Although rare, the jejunal polyp is often recorded as an intussusception caused in various ages, arising only from an obstruction to atresia.[6–8]

Some past studies have reported solitary jejunal polyp in association with Peutz Jegher syndrome, but, in this case, we detected no signs of this syndrome such as dark skin freckling at the mouth, eye, nostril, or anus, and other polyps along the gastrointestinal tract.[6,7] Other causes included juvenile polyp, adenocarcinomatous polyp, and inflammatory fibrous polyp.[9,10] In this patient, due to an old ischemic cell, the pathologist only described an infarcted polyp, but could not differentiate the type of the polyp.

Treatment for IP is mostly performed conservatively unless signs of peritonitis or strangulation are detected. Cause of IP that can be managed medically: autoimmune disease, vascular disease, or drug-induced IP. In case of doubt, surgery decision making should depend on several points; concomitant critical CT findings (mesenteric ischemia, bowel obstruction, bowel perforation, portal venous gas), critical laboratory findings (Leukocyte/inflammatory marker increase, lactic acid increase, acidosis), conspicuous physical examination (muscle guarding, peritonitis, and bowel sounds), and past medical history and its medications. The decision to perform surgery should be tailored to the clinical conditions of patients and supported by meticulous examination.[5]

The ideal follow-up should be based on the probable diagnosis of the polyp, but the limitation, in our case, is that the histopathology of the polyp was inconclusive due to old infarction and bleeding. As it might have been an isolated juvenile polyp, we considered it as a benign polyp that did not necessarily need further evaluation.

CONCLUSION

IP can be a sign of life-threatening conditions in children. Although it is most commonly seen in neonates with necrotizing enterocolitis, other pathology such as intussusception, gastrointestinal perforation, bowel ischemia, and necrosis must be considered seriously in older children.

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