



Sigmoid Volvulus in An Adolescent Female With Colitis: A Rare Case Report

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ABSTRACT

Background: Sigmoid volvulus is rare among pediatric patients. It predominantly affects adult males and commonly seen in region known as the "volvulus belt" countries. Diagnosis in children is often delayed or missed. **Case Presentation:** A 13-year-old female presented to emergency department with three-days history of abdominal pain, nausea, and inability to defecate without fever or vomiting. Multiple episodes of constipation were noted and patient was menstruating. Physical examination found abdominal distention and decreased bowel sound, suggesting obstruction. Plain abdominal radiograph showed significant colon dilatation and gas-filled loops with absent pelvic gas. Contrast-enhanced abdominal CT revealed large bowel dilatation with "whirl sign" as the diagnostic feature. Colon in-loop result revealed colon redundancy along with "bird-beak sign" at the sigmoid-descending colonic junction, confirming the diagnosis of sigmoid volvulus. Twisted bowel along with colitis was visualized during sigmoidoscopy procedure. Both parents requested discharge two days after procedure as patient's symptoms completely resolved and no further surgical procedures were planned. **Discussion:** Sigmoid volvulus is a potential differential diagnosis in children with symptoms of bowel obstruction. Physicians and radiologists should remain vigilant to avoid morbidity and mortality resulting from delayed and untreated cases. **Conclusion:** It is important to be aware of uncommon causes of abdominal pain in the pediatric age group. Thorough clinical and radiological investigations play a crucial role in preventing treatment delays in volvulus, with histopathologic results aid in determining the underlying cause. Clinicians must "keep an eye out" for uncommon differential diagnosis of abdominal pain, regardless of age.

Keyword: Colitis, Decompression, Pediatric Sigmoid Volvulus, Sigmoid Redundancy, Whirl Sign

ABSTRAK

Latar Belakang: Volvulus sigmoid merupakan kasus yang jarang ditemui pada pasien anak. Kasus ini banyak dijumpai pada pria dewasa dan biasa ditemukan di negara-negara yang dikenal sebagai sabuk volvulus. Diagnosis pada anak seringkali terlambat bahkan terlewatkan. **Presentasi Kasus:** Seorang perempuan berusia 13 tahun datang ke Instalasi Gawat Darurat dengan keluhan nyeri perut, mual, dan tidak dapat buang air besar sejak tiga hari lalu tanpa demam ataupun muntah. Terdapat riwayat konstipasi berulang serta pasien saat ini sedang menstruasi. Pemeriksaan fisik menunjukkan distensi abdomen dan bising usus menurun yang mengarah pada kecurigaan adanya obstruksi saluran cerna. Foto polos abdomen menunjukkan adanya dilatasi kolon dengan *gas-filled-loops* serta tidak terlihat udara pada area pelvis. CT-scan abdomen dengan kontras IV menunjukkan dilatasi kolon signifikan dengan "*whirl sign*" sebagai fitur diagnostik penting. Hasil colon-in-loop menunjukkan adanya redundansi kolon serta "*bird-beak sign*" pada area transisi kolon sigmoid dan kolon desenden, mengonfirmasi diagnosis volvulus sigmoid. Segmen usus yang terpelintir serta kolitis tampak saat prosedur sigmoidoskopi. Kedua orangtua pasien menginginkan pulang karena keluhan sudah hilang ditambah tidak ada rencana operasi lanjutan.



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Diskusi: Volvulus sigmoid merupakan diagnosis diferensial yang potensial pada anak dengan gejala obstruksi saluran cerna. Klinisi dan radiolog harus selalu waspada demi menghindari morbiditas dan mortalitas akibat kasus yang lambat maupun tidak tertangani. **Kesimpulan:** Kasus ini menekankan pentingnya kewaspadaan terhadap suatu penyebab tidak lazim kasus nyeri perut pada anak. Pemeriksaan fisik dan radiologi yang menyeluruh berperan penting dalam mencegah keterlambatan penanganan, serta histopatologi berguna untuk mencari penyebab mendasar terjadinya volvulus sigmoid.

Keyword: Dekompresi, Kolitis, Redundansi Sigmoid, Sigmoid Volvulus Pediatri, Whirl Sign

1. Introduction

Sigmoid volvulus accounts for 3-54% of cases of intestinal obstruction worldwide. It predominantly affects adult males and is frequently observed in region known as the "volvulus belt", encompassing countries such as Africa, India, the Middle East, South America, and Turkey.^[1] Currently there are no data regarding its prevalence in Indonesia or other Southeast Asia countries. A long, redundant sigmoid colon with an elongated mesocolon is identified as the primary predisposing factor. Other predisposing factors include a high-fiber diet, chronic constipation, Hirschsprung's disease, and chronic use of laxatives.^{[1][2][3]} Its occurrence among pediatric patients is rare, and diagnosis of sigmoid volvulus in children is often delayed and frequently missed.^[1] Late diagnosis and treatment may lead to complications such as colon ischemia, hemorrhagic infarction, and even life-threatening situations.^[4] In this report, we present a rare case of sigmoid volvulus in a 13-year-old adolescent female.

2. Case Presentation

A 13-years-old Indonesian female patient presented at the hospital's ED with persistent, worsening abdominal pain and nausea for three days. Distended abdomen was observed and she had been unable to pass stool for three days but could still pass flatus. There was no history of fever, vomiting, and bloody "red currant jelly" stools. Multiple previous episodes of constipation had occurred. Family history, routine drug consumption, changes in dietary pattern, and drug allergies were all denied. Patient was currently menstruating.

Physical examination showed stable vital signs with mild abdominal distention and decreased bowel sounds. No rebound tenderness or muscle guarding on palpation was found in all four abdominal quadrants. Digital rectal examination revealed normal sphincter tone with no palpable fecal mass, no collapsed rectum, or blood findings. Routine laboratory examination showed normal results. Initial fluid and intravenous drugs were administered. Supine abdominal plain radiograph showed significant colon dilatation and gas-filled loops with ground-glass appearance in the pelvic area (Figure 1a). The left lateral decubitus view showed multiple air-fluid levels consistent with large bowel obstruction and no sign of free air (Figure. 1b).

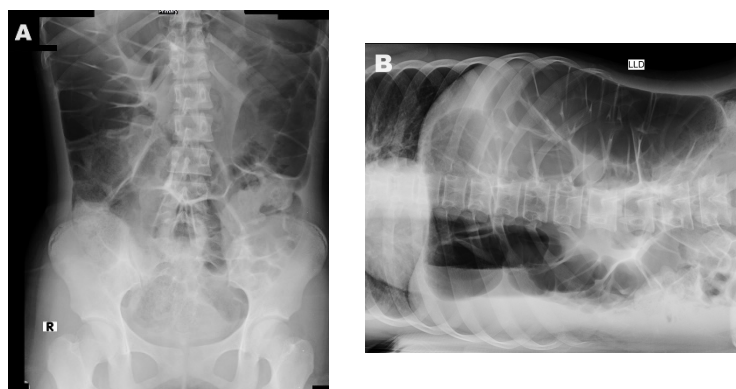


Figure 1. (a) Supine AP photo depicted a dilated colon with a ground-glass appearance in the pelvic area on the day of admission; (b) Multiple air fluid levels spotted on LLD photo

Further investigation with abdominal IV contrast CT identified "whirl sign" appearance causing large bowel obstruction with marked dilatation (Figure 2), highly suggestive to sigmoid volvulus. The small bowel appeared normal in caliber. There were no signs of free air in the abdominal cavity indicating

pneumoperitoneum, nor was there free fluid in the abdominal cavity. Additionally, there were no masses in the pelvis or abdominal cavity and no lymph node enlargement.

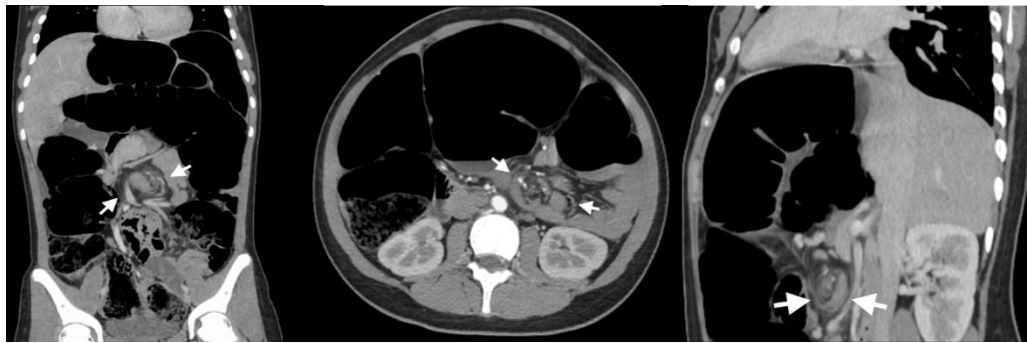


Figure 2. (a) Frontal plane; (b) transverse plane and; (c) sagittal plane. Abdominal CT scan images revealed colonic dilatation along with whirlpool sign (blue arrows) as a diagnostic finding, marking transitional point from the normal to the twisted part of the colon.

Patient was later transferred to an inpatient room in the evening and a bedside decompression was attempted with the insertion of rectal tube along with bowel lavage. To evaluate further, the patient underwent a colon-in-loop procedure. Water soluble contrast was injected and entered the rectum and sigmoid thoroughly but then slowed. A narrowing area appeared between the sigmoid and the distal descending colonic junction, shown as “bird-beak sign”, resulting from volvulus with partial low-lying obstructive ileus. There was no sign of hollow-organ perforation. Redundant colon was also observed, with no visible transitional zone as seen on Hirschprung disease (Figure 3).

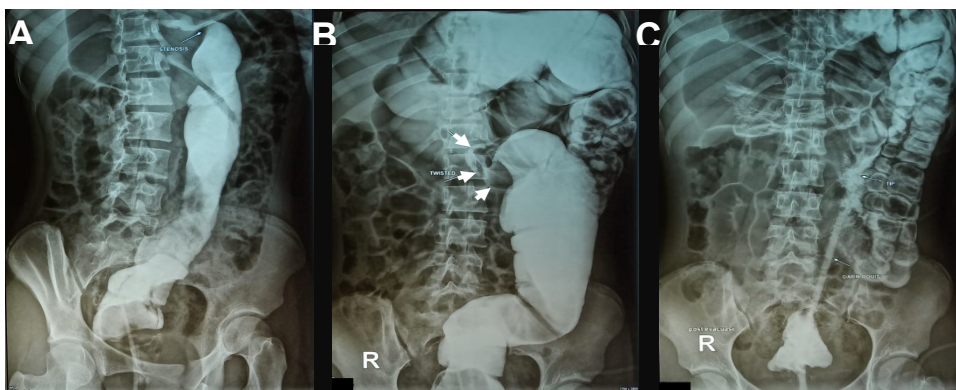


Figure 3. Colon-in-loop: (a) Contrast entering rectum and part of sigmoid colon; (b) “bird-beak sign” (white arrows); (c) colon redundancy.

Diagnostic and decompression sigmoidoscopy was performed in the operating room the next day. During procedure, “pinwheeling” torsion indicating volvulus was visualized, along with erythematous and edematous mucosa (Figure 4). A Biopsy was done in conjunction. The bowel was successfully decompressed, and a follow-up abdominal X-ray showed no colon distention (Figure 5). Patient was able to pass stool as well as abdominal distention was reduced. Both parents requested discharge two days after procedure as patient’s symptoms completely resolved and no further surgical procedures were planned. Parents was advised to return for further evaluation after the histopathology results were available.



Figure 4. “Pinwheeling” torsion visualized on sigmoidoscopy, indicating twisted of bowel lumen along with colitis



Figure 5. Supine AP photo post-sigmoidoscopy decompression, revealing normal colon caliber and gas distribution.

3. Discussion

Volvulus derived from the Latin word "volvere", meaning twist. The sigmoid colon is the most common twisted part in adults, while the small intestine and stomach are more common in children.^[3] Sigmoid volvulus is a rare condition in children, with approximately 100 cases reported since 1940 to date.^[6] ^[2]^[5] Males outnumber females (3:1) with a median age of 7 years old.^[1] The latest similar cases have been reported in male adolescents by Totadri, Vetri, & Sainath and in female adolescents by Kiyaka et al. and Damkjaer et al.^[5]^[6]^[7]

In this rare case, a 13-year-old female presented with acute, worsening abdominal pain, abdominal distention, and inability to pass stool. Multiple previous instances of constipation were identified as a risk factor for her. Upon examination, mild abdominal distention with decreased bowel sounds was observed. The absence of fever, rebound tenderness, and muscle guarding indicated that peritonitis had not occurred. Digital rectal examination revealed normal sphincter tone with no collapsed rectum and no palpable stool. Comprehensive history taking and thorough physical examination are essential to develop clinical suspicion of bowel obstruction in children and to avoid associated mortality. Without careful consideration, abdominal pain in this patient could be misdiagnosed as merely dysmenorrhea, considering that patient was menstruating at that time. All findings were consistent with a study conducted by Colinet et al., which showed that the most frequent initial symptoms in children were abdominal pain in all cases (100%), followed by abdominal distention (84.6%), and nausea and/or vomiting (53.8%), with constipation present in 23% of cases.^[8] Clinical examination revealed abdominal tenderness (100%) and absence of stool in the rectum (46%). Differential diagnosis includes Hirschsprung's disease, cecal volvulus, pseudo-obstruction, malignant tumors and other colonic diseases such as toxic megacolon, paralytic ileus, giant colonic diverticulum, and ileosigmoid knotting.^[1]

Radiological investigation is crucial for confirmation and ruling out differential diagnosis. This patient initially underwent abdominal plain radiography, followed by IV-contrast abdominal CT-scan. These tests are often ordered if there is suspicion of bowel obstruction. Although less expensive and readily available in daily practice, abdominal plain radiography is less useful in distinguishing volvulus from other large bowel obstructions. In the pediatric population, the "coffee-bean" sign is seen in just 16% of cases.^[1]^[9]^[10] IV-contrast abdominal CT remains the gold standard in localizing the site of obstruction, although a study by Tannouri et al. successfully diagnosed only 55.6% of pediatric cases compared to adult volvulus (sensitivity 93%; specificity 96%). The "whirl sign" on CT-scan represents tension on the tightly twisted mesocolon by the afferent and efferent limbs of the dilated colon.^[10] Barium enema/ colon-in-loop examination increases sensitivity in pediatric patients (71-82%).^[4] A Long and redundant colon was an interesting finding in this patient. Redundant sigmoid colon, with its narrowed mesenteric attachment to posterior abdominal wall allows close approximation of two limbs of sigmoid colon, becoming main predisposing factor in sigmoid volvulus.^[1] ^[8] ^[11] Another finding was the demonstration of "bird-beak sign", which is common and characteristic of sigmoid volvulus.^[12] Rectal tube insertion was performed bedside in attempt to decompress the colon while waiting for definitive treatment.

It is crucial to determine whether this patient presented with isolated case or if there was another associated disease. In particular, Hirschsprung disease must be ruled out, as 17% of pediatric sigmoid volvulus cases are associated with it and it acts as one of predisposing factor.^[1]^[7] However, in this case we didn't find any feature

indicative of Hirschprung disease besides chronic constipation: There was no history of delayed meconium passage (>48h) after birth, no vomiting and no collapsed rectum with explosive stools on digital rectal examination. Transitional zone was not visualized during colon-in-loop procedure. Possibility of Hirschprung-associated Enterocolitis (HAEC) due to neglected Hirschprung disease can also be excluded as patient didn't exhibit fever, vomiting or bloody diarrhea.

In this presented case, a sigmoidoscopic procedure was performed to decompress the twisted colon. This aligns with some studies, state that sigmoidoscopic decompression should be the first-line treatment for sigmoid volvulus in hemodynamically stable children with no signs of bowel perforation, as in adults. It is safe and not associated with any complications. Additionally, it allows for visualization and assessment of colonic mucosa viability. While the success rate of sigmoidoscopic decompression in adults ranges from 50% to 100%, data regarding pediatric cases remain scarce.^{[3][13][14][15]} Parolini et al. reported 69% success rate in managing 56 cases, while Colinet et al. observed a success rate of 92.3% in 13 cases.^{[8][18]} This procedure should be followed by elective bowel resection, which was not performed in this patient due to surgery's assessment. Some studies reported children who were readmitted with the same complaints after initial decompression procedure without being followed by surgery. Recurrence rates in children were observed from 50% to 100%, similar to those observed in adults with shorter recurrence times varying from 2 days to 4 months after the initial decompression, compared to 5-year median in adults.^{[8][9][12][15][16][17]}

Colitis visualized during sigmoidoscopy suggests an inflammatory process, which may lead to predisposition of volvulus. Inflammation may lead to hyperperistaltic and one proposed theory suggested that abnormal fixation of an inflamed colon to surrounding viscera might predispose to bowel twisting.^[2] Histopathology examination is important and considered the gold standard for further confirming the cause of colitis. Unfortunately, post-decompression images were not obtained and histopathology result has not been followed up yet. Physicians and radiologists should remain vigilant about sigmoid volvulus as a potential differential diagnosis to avoid morbidity and mortality resulting from delayed and untreated cases, especially in the pediatric population.

4. Conclusion

It is essential to "keep an eye out" for uncommon differential diagnosis of abdominal pain, regardless of age. Early diagnosis and prompt treatment result in better prognosis, prevent treatment delays and complications such as bowel ischemia, perforation, shock and death. Histopathologic results aid in determining the underlying cause.

5. Conflict of Interest Statement

The authors have no conflict of interests to declare.

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