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# Short bowel syndrome as a result of sigmoid volvulus in an 8-year-old child. The first reported case worldwide: A case report

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## ABSTRACT

**INTRODUCTION:** We present a case of a male child 8 years old presenting with a sigmoid volvulus causing ischemia of most of bowel loops resulting in a short bowel syndrome. It is the first reported case worldwide.

**PRESENTATION:** A male child presented with a picture of intestinal obstruction. After complete laboratory and radiological investigation, laparotomy was done revealing a sigmoid volvulus compressing most of the small bowel loops with gangrenous sigmoid colon for which sigmoidectomy with end colostomy, resection of gangrenous small bowel loops and primary anastomosis of the remaining healthy part.

**DISCUSSION:** There are few reported cases describing sigmoid volvulus in this age group. However, none of them resulted in short bowel syndrome. The median age was 7 years with a higher ratio in males than females (3.5:1).

**CONCLUSION:** Sigmoid volvulus is not a common problem in children and adolescents, and is rarely considered as a cause of intestinal obstruction and it was never reported as a cause of short bowel syndrome. Early diagnosis and prompt treatment confer an excellent prognosis.

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## 1. Introduction

Sigmoid volvulus is not commonly considered a diagnosis in children and adolescents [1]. There are few reported cases described sigmoid volvulus in this age group. However, none of them resulted in short bowel syndrome. Sigmoid volvulus is more commonly recorded in the Middle East, Africa, the Indian subcontinent, Turkey, and South America [2]. It is a potentially life-threatening problem in children. A high index of suspicion is needed to reach a true diagnosis and prevent morbidity and mortality. We report an uncommon case of a male child presenting with a sigmoid volvulus causing ischemia of most of the small bowel, requiring a massive resection and resulting in short bowel syndrome (SBS). The patient was managed via total parenteral nutrition feeding for four months until he was able to tolerate oral feeding.

## 2. Case presentation

We report an uncommon case of a male child presenting with a sigmoid volvulus causing ischemia of most of the small bowel, requiring a massive resection and resulting in short bowel syndrome (SBS). The patient was managed via total parenteral

nutrition feeding for four months until he was able to tolerate oral feeding.

Our case is an 8-years-old boy referred to our hospital with 4 days' history of diffuse abdominal pain, bilious vomiting, and absolute constipation. No previous episodes of constipation mentioned in the past history. He was examined at our emergency department and was found to have a blood pressure 100/60 mmHg, pulse rate of 110 beats per minute and temperature of 38.7c. Abdominal examination revealed a severely distended abdomen and tenderness all over the abdomen. Bowel sounds were very sluggish and a per rectal examination revealed an empty rectum. A plain abdominal film showed only multiple air-fluid levels, but not conclusive of a sigmoid volvulus (Fig. 1) and so we did multi-slice computed tomography (C.T) of abdomen and pelvis that confirmed the diagnosis of a sigmoid volvulus (whirl sign of volvulus, dilated bowel loops, multiple air-fluid levels) and the presence of pneumointestinalis (Figs. 2 and 3).

Preoperative laboratory investigations were as follows; Hb (15gm/dl), WBCs (18000), serum sodium (137 meq/l), serum potassium (4.3 meq/l), blood urea (35 mg/l), serum creatinine (1 mg/dl). He was admitted to our pediatric surgical unit and a nasogastric tube was inserted, intravenous fluids and broad spectrum antibiotics were given and prepared for urgent abdominal exploration. A preoperative written consent was secured. As this was considered an emergency, his bowel was not prepared, he underwent laparotomy (via a midline incision) with the finding of the sigmoid

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**Fig. 1.** Plain x ray showing multiple air-fluid levels (jejunal and ileal).



Fig. 2. CT of the abdomen showing multiple air fluid levels most of them are jejunal loops and pneumatosis intestinalis (marked by arrows).

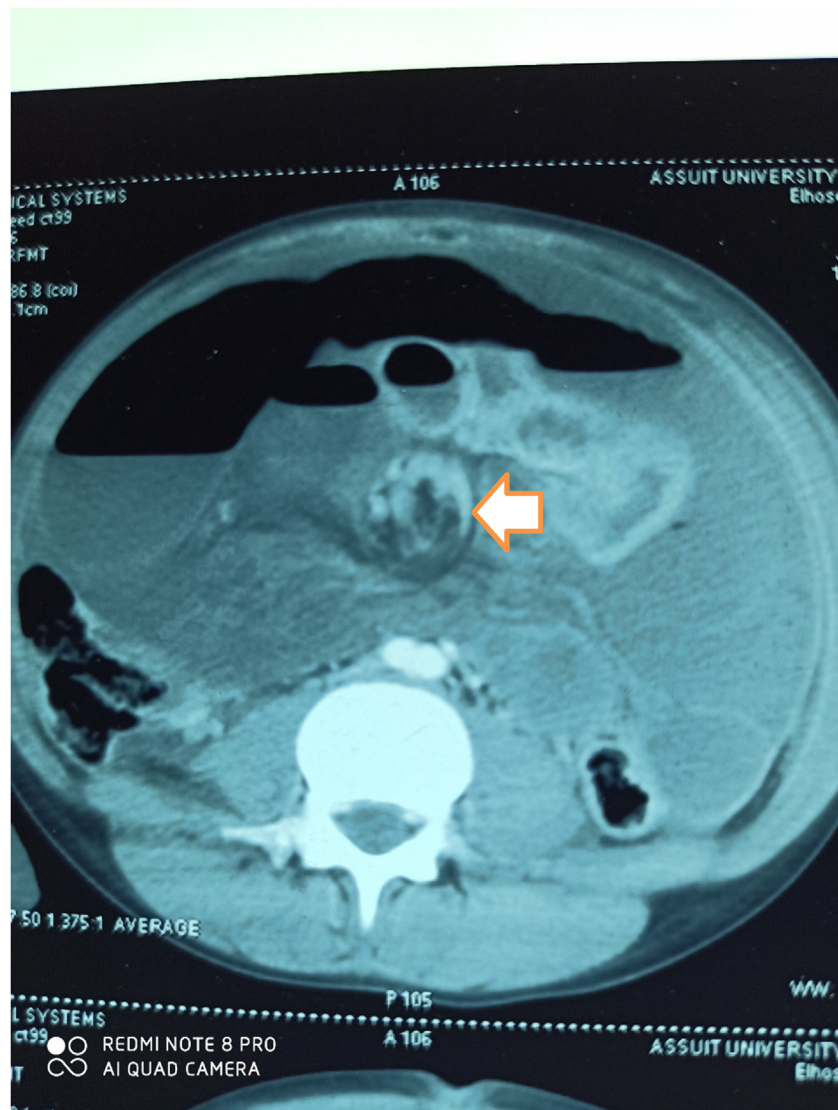


Fig. 3. CT of the abdomen showing whirl sign suggestive of sigmoid volvulus (marked by an arrow).

occupying most of the abdominal cavity, the sigmoid was delivered from right side and hugely dilated with a 360° twist (Fig. 4).

The sigmoid colon was crossing to the opposite side compressing the mesentery of the small intestine (Fig. 5).

There were traces of intra-peritoneal collection (serous in nature). There was gangrene of the sigmoid colon and causing compression of the mesentery of the small bowel causing gangrene of most of the small bowel, we used warm saline and good oxygenation in an attempt to salvage the small intestine but with no benefit, loops were blackish in color and covered with patches of white slough. Only about 100 cm of ileum were healthy (Figs. 6 and 7).

We performed a sigmoidectomy with end colostomy (Hartman's) and primary anastomosis of the healthy part of small intestine. This massive resection of most of the small intestine resulted in a short bowel syndrome. As regarding postoperative treatment, the patient was on TPN and broad spectrum antibiotics against gram positive, gram negative and anaerobe bacteria. The postoperative period was uneventful with no postoperative complications and no surgical site infection.

On Follow up, he is free of symptoms 1 year after operation.

As regarding the resected sigmoid and small bowel, the pathology report showed that both resection margins had a preserved

architecture with no features of ischemia. The mucosal lining from the discolored areas mild chronic inflammation. The lamina propria has congested blood vessels with a hypertrophic muscular coat. There were visible ganglion cells and hypertrophic nerve trunks in the muscularis propria.

This case was reported in line with the SCARE 2020 criteria [3].

### 3. Discussion

Sigmoid volvulus occurs when a redundant loop rotates around its elongated mesentery. Obstruction of the intestinal lumen and compromise of its blood supply occur when the degree of twist exceeds 180 and 360°, respectively [4]. The median age was 7 years with a higher ratio in males than females (3.5:1) [5].

Among children several predisposing factors have been identified. Hirschsprung's disease has been implicated in both transverse colonic as well as sigmoid volvulus. This was excluded in our patient by the detection of abundant ganglion cells shown in the biopsy specimen. Chronic constipation is another predisposing factor for sigmoid volvulus, but our patient and his family denied any history of constipation. So, the causative factor in this reported case will remain idiopathic.



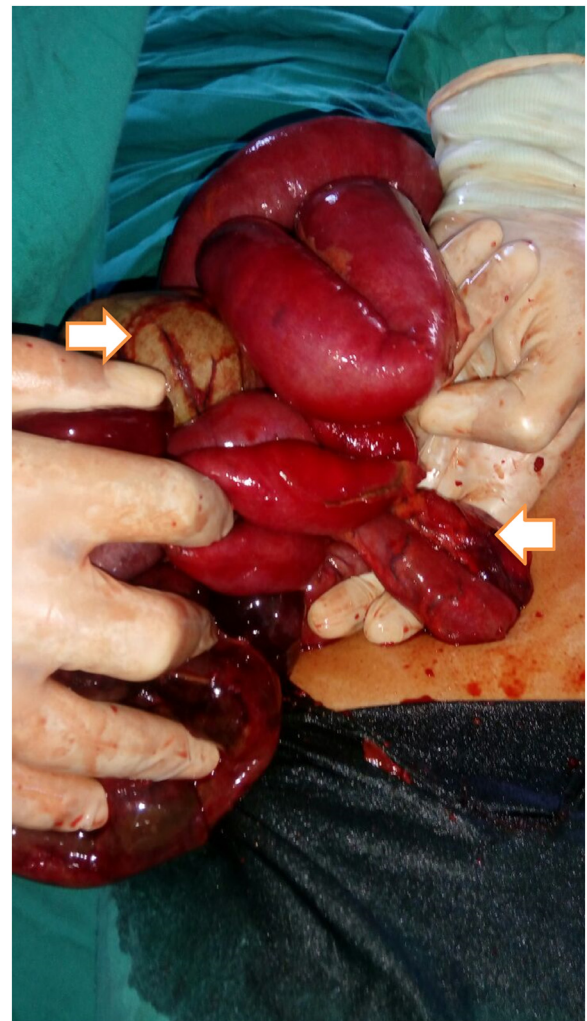
**Fig. 4.** The sigmoid was delivered from right side (marked by an arrow) and hugely dilated with a 360° twist.

If untreated, it can cause hemorrhagic infarction, perforation, septic shock, and death [6]. In this patient, it resulted in gangrene of the sigmoid colon and the majority of small bowel. Sigmoid volvulus has two different presentations (acute and recurrent). In our patient, it was acute presentation, no history of recurrent attacks. The most frequent manifestations are abdominal pain, abdominal distention, and vomiting [7]. All these symptoms were found in our patient.

The diagnosis of sigmoid volvulus depends on a detailed history, physical examination, and careful interpretation of plain abdominal films. The sensitivity of the whirl sign for sigmoid volvulus in pediatrics is only 16–29% [7]. A plain abdominal X-ray is suggestive of sigmoid volvulus in approximately 29% of patients [8]. In our case, it showed only multiple air fluid levels.

The typical bird's beak sign seen on contrast enema study is specific for volvulus. However, Mellor and Drake found a twisted appearance to be more frequent [9]. This twist was very obvious in our case. Reduction by barium enema succeeded in 77% (10 of 13) of the cases [7]. Treatment for sigmoid volvulus is still controversial in pediatrics. In stable patients, non-operative reduction of the volvulus using barium enema or sigmoidoscopy may be tried first [10]. When there are no evidence of peritonitis and an endoscopy unit is available, endoscopic decompression and detorsion should be the first step of treatment aiming to relieve symptoms and to prepare the patient for the surgical intervention [11]. But, our patient was presenting late with diffuse peritonitis, so he was not suitable for those previous measures and was prepared for urgent exploration.

The definitive treatment is sigmoidectomy, either with primary anastomosis or colostomy. Our patient was managed by sigmoidectomy, with end colostomy (Hartman) and primary anastomosis of the remaining healthy small bowel. Recurrence is common in cases managed by detorsion without resection (operative 25%, non-operative 35%) [7]. Excellent prognosis of sigmoid volvulus is reported, provided it is diagnosed and treated promptly by the



**Fig. 5.** Showing sigmoid colon (marked by arrows) crossing the opposite side compressing mesentery of small intestine.

definitive measures [5]. The resection of most of the small intestine in our case resulted in a short bowel syndrome that was managed via total parenteral nutrition feeding until the patient was able to tolerate orally on the standard nutrition support for short bowel syndrome.

#### 4. Conclusions

Sigmoid volvulus is not a common problem in children and adolescents, and is rarely considered as a cause of intestinal obstruction and it was never reported as a cause of short bowel syndrome. Hence, pediatric surgeons should maintain a high index of suspicion, in order not to miss them, as any delay in instituting treatment has a devastating effect on morbidity as well as mortality. Early diagnosis and prompt treatment confer an excellent prognosis

#### Declaration of Competing Interest

No conflict of interest.

#### Sources of funding

No sources of funding.



Fig. 6. Showing gangrene of the sigmoid colon and most of the small bowel.



Fig. 7. Showing the resected gangrenous part of small intestine.

**Ethical approval**

The case report is exempt from ethical approval in my institution.

**Consent**

Written informed consent was obtained from the patient’s parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Author contribution**

Hussein Ibrahim: main author of the paper and wrote the manuscript.

Ahmed Maher: literature review and revised the manuscript.

Tarek Abdelazeem Sabra: supervising and editing.

**Registration of research studies**

Not Applicable.

**Guarantor**

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**Provenance and peer review**

Not commissioned, externally peer-reviewed.

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