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Case Report

Non-invasive treatment of the sigmoid volvulus. A pediatric case report☆,☆☆

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ABSTRACT

Sigmoid volvulus occurs when the sigmoid colic loop gets wrapped around its own mesocolon. While this condition is categorized as an extremely rare emergency in the pediatric population, diagnosis is often difficult due to the fact that its aspecific manifestations or sneaky symptoms are similar to other medical conditions. The available treatment options remain controversial up to this day, and the non-operative approach is more preferred in treating hemodynamic stable patients.

This paper examines the case study of a 13-year-old girl suffering from sigmoid volvulus, who was treated with water-soluble contrast enema, in order to determine whether this method is efficient and effective in successfully treating the pathology of this condition.

Introduction

In the pediatric population, the sigmoid volvulus is an extremely rare emergency. It results in abdominal pain that commonly affects adults more than children. Diagnosing this condition is generally difficult as there are only a few data in the literature that are currently available [1-3].

Salas et al reported 63 cases of sigmoid volvulus in children with a median age of 7 years old and a male predominance [4]. After which, Smith et al examined other 48 cases of sigmoid volvulus in children with a median age of 8 years [5,6].

Clinical manifestations of volvulus can include acute abdominal pain with nausea, vomiting, and abdominal distention, or nonspecific and chronic symptoms such as recurrent abdominal pain, constipation, or gastroenteritis [1,4].

An early diagnosis and treatment are essential to avoid complications related to untreated volvulus as it can lead to ischemia, necrosis, perforation, septic shock, and in more serious cases, death [1].

The available treatments for pediatric sigmoid volvulus remain controversial and dependent on the hemodynamic stability of the patient. If stable, a non-operative approach along with barium enema or sigmoidoscopy should be utilized. In

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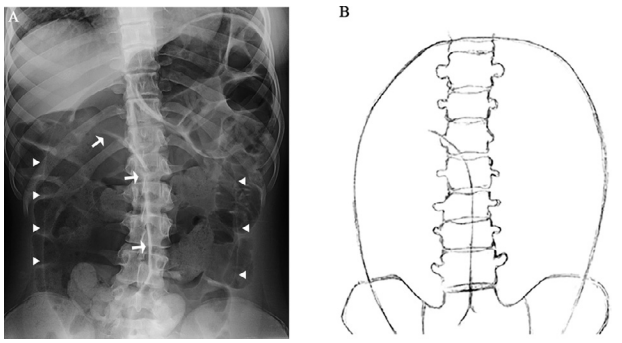

Fig. 1 – (A) Plain radiograph showing the coffee bean sign suggestive for sigmoid volvulus. This is formed by grossly-dilated. (B) Graphic representation of coffee bean sign.

unsuccessful cases or hemodynamic instability, surgical intervention is needed to remove the sigmoid loop and prevent other possible recurrences [4,5,7,9].

We present a clinical case of a 13-year-old girl with a sigmoid volvulus diagnosed and resolved with water-soluble contrast enema. The aim is to describe how this method could be useful not only for the diagnosis but also for the definitive and successful treatment of this pathology.

Case presentation

A 13-year-old girl was admitted to our institution because she was suffering from constipation, worsening abdomen pain, and inappetence for 4 days.

The patient's medical history was unremarkable, with no previous abdominal surgeries, except for a long-standing history of constipation.

The clinical examinations revealed a greatly distended, non-tender, and painless abdomen, without muscle guarding or rebounding tenderness. All her vital signs including body temperature were normal and stable. Based on her history of long constipation, suspecting a fecaloma colonic obstruction, a proctoscopy was performed, and that excluded this diagnosis, showing an empty rectum.

An abdominal plain film revealed severe distention of the entire colon, particularly the sigmoid colon (maximum diameter 12 cm). (Fig. 1) In addition, the findings also indicated great distention in the small bowel loops with consistent stepladder air-fluid levels, suggesting a mechanical intestinal obstruction.

In order to determine the underlying cause behind the mechanical intestinal obstruction, a Computed Tomography (CT) was performed. This confirmed the presence of marked gaseous distention of the colon, especially sigmoid, as well as revealed a swirling appearance of the mesosigmoid colon. (Fig. 2) Furthermore, the results showed that there were no bowel walls thickening nor free fluid in the abdomen.

Based on these radiological findings (compatibles with an intestinal obstruction induced by a sigmoid volvulus) and

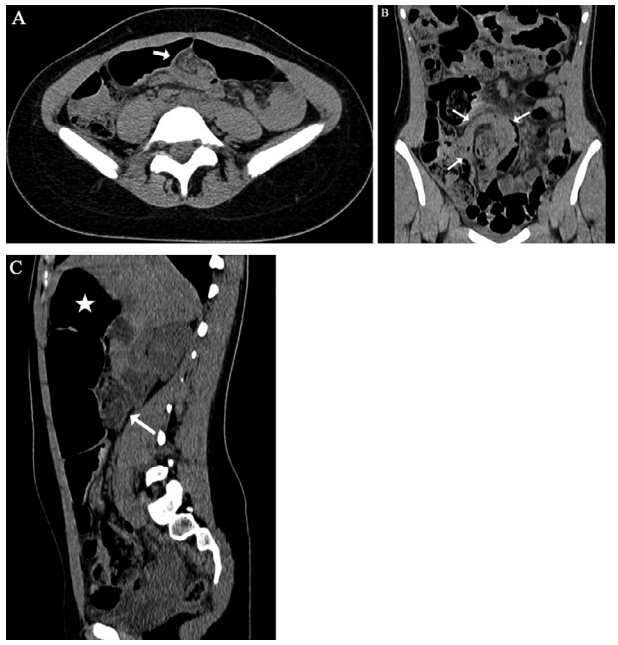

Fig. 2 – (A) CT axial view shows the whirlpool sign (arrow), representing twisted mesentery. (B) CT coronal view shows the whirlpool sign (arrows), representing twisted mesentery. (C) CT sagittal view show dilated sigmoid colon (star) that extends cephalad to the transverse colon (arrow), northern

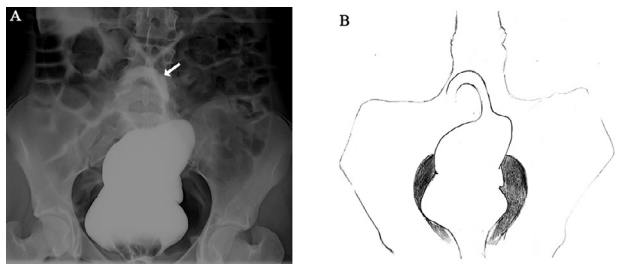

Fig. 3 – (A) Gastrografin enema showing a markedly narrowing of the sigmoid colon with a classic "birds beak" (arrow) appearance at the level of the twist. (B) Graphic representation.

worsening of abdominal pain, a water-soluble fluoroscopy-guided contrast enema was performed to confirm the diagnosis of the sigmoid volvulus (Figs. 3-4-5) and to offer her a potential non-invasive treatment.

After rectum catheterization, 300 mL Gastrografin and 600 mL of 0.9% saline solution were infused through the catheter, until the opacification of the point of torsion of descending colon. The enema was successful in the first attempt, achieving the complete detorsion of the sigmoid colon and the immediate relief of symptoms.

Four days later, an abdominal ultrasound was performed, which showed a significant reduction of intestinal distention of the colon and sigmoid colon (maximum diameter 2 cm).

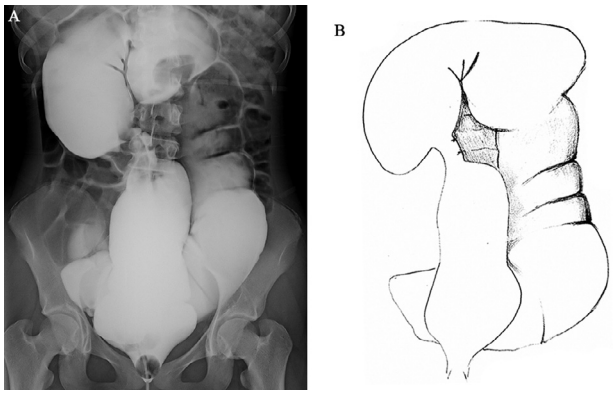

Fig. 4 – (A) Reduction of narrowing during progressive administration of the contrast medium. (B) Graphic representation.

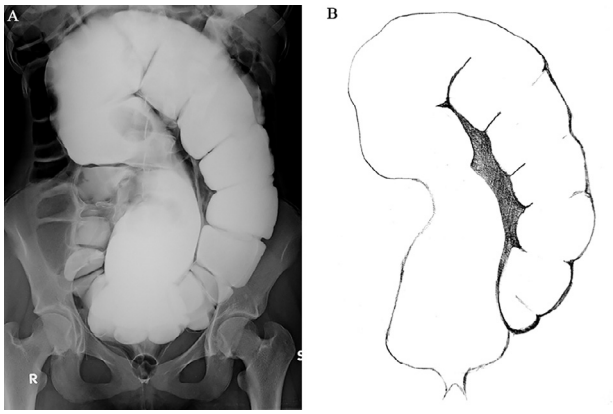

Fig. 5 – (A) Detorsion of the sigmoid volvulus. (B) Graphic representation.

Neither free fluid nor imaging findings of inflammatory disease were present.

After 6 days from the treatment, based on the improvement of her general clinical conditions, the imaging findings and, the resolution ad not-relapsed of symptoms, the patient was discharged home. After 2 months, follow-up radiographs confirmed the resolution of the sigmoid volvulus with only minimal distention of the sigmoid colon.

Thus far, the patient had no relapses and no symptoms related to intestinal occlusion. She is only treated for chronic constipation with Macrogol 0.5 mg/kg as prescribed by the pediatric surgeon.

Discussion

Sigmoid volvulus is an extremely rare cause of abdominal pain in children, which occurs when the sigmoid colonic is wrapped around its own elongated fixed mesocolon.

When the degree of torsion exceeds 180°, an intestinal occlusion can occur. On the other hand, when it exceeds 360°, a reduction of blood flow can determine intestinal hypoperfusion leading to critical bowel complications such as ischemia or infarction [8,10].

Although volvulus is a rare occurrence in the pediatric population, the differential diagnosis of acute or chronic abdominal pain should be taken into account.

An association between sigmoid volvulus and congenital anomalies including malrotation, internal hernia or Hirschsprung disease, omphalomesenteric abnormalities, anal stenosis, chronic constipation have been demonstrated [7,9,11-15].

Abdominal radiographs represent the first approach in making a diagnosis, showing various indirect signs that are suggestive of sigmoid volvulus, such as the coffee bean and northern exposure signs [9,16,17].

The coffee bean (also known as the kidney bean sign) is obtained by the sigmoid colon distention by gas, which forms an area of hyperlucency that is similar to the shape of a coffee bean [16]. Conversely, the northern exposure sign is the result of the vertical dislocation in the upper part of the abdomen of the dilated sigmoid colon, crossing the transverse colon [17].

Other common findings in radiographs are: the marked ahaustral dilated colon, the absence of rectal gas, multiple air-fluid levels in intestinal loops, and other signs of mechanical intestinal obstruction [1,4,6,18].

Although the sensitivity of plain radiographs to make a diagnosis of sigmoid volvulus is high, the specificity remains low (20%), and most commonly in children than in adults the plain films are often non-diagnostic and aspecific [1,4]. Smith et al reported the presence of the coffee bean in only 8 out of 28 pediatric patients, and most recently, Atamanalp et al, documented this particular sign in 12 out of 19 children with sigmoid volvulus (63% of patients) [5,8].

If there is high clinical and radiographic suspicion, a contrast enema or an abdominal contrast-enhanced CT should be performed [11].

To identify a sigmoid volvulus, CT examination has higher sensitivity and specificity (93% and 96% respectively) than plain radiographs. It is characterized by a diagnostic accuracy of almost 100% [1,6], identifying some specific findings, including the mesocolon whirlpool sign and the sigmoid "bird-beak" sign [9]. Moreover, it can be useful to valuing coexisting disorders or excluding complications (as pneumoperitoneum, or signs of bowel wall ischemia or hemorrhagic infarction). Thanks to Multiplanar reconstruction (MPR) of CT scan, is possible to assess the exact level of the colon obstruction, as well [1,6,19].

Currently, the treatment for pediatric sigmoid volvulus is still up for debate as it poses risks and complications. In stable patients with sigmoid volvulus, non-surgical reduction, such as enema examination, should be considered the treatment of choice, in order to reduce the high complication rate associated with emergency surgery [1,11,14].

In 2016 ACR–SPR PRACTICE provided specific indications for the performance of pediatric enema examinations. Based on these guidelines, patients with complications related to volvulus as perforation, ischemic colon, toxic megacolon, hypovolemic shock, peritonitis, or other potentially unstable clinical conditions, cannot undergo to enema [20].

In most patients, no specific bowel preparation is needed. Enema can be performed with a pneumatic or hydrostatic

technique. The first is preferred in the case of pneumoperitoneum or peritonitis, which represents absolute contraindications to hydrostatic contrast enema with barium. In this case, a water-soluble contrast that is less irritating to the peritoneum can be a viable alternative to reduce the risk of peritonitis [1,4].

The contrast enema is identified through the appearance of the "twisted taper" or "bird-beak" structure of the colon [4]. Contrast enema has diagnostic and therapeutic advantages, which can be beneficial in the differential diagnosis (identifying other possible bowel obstruction causes) and in the non-invasive treatment of volvulus, although not always successful at the first attempt [1,4]. In the uncomplicated stable patients, contrast enema should be considered as the initial approach, in the prospect of definitive surgery management [2,11].

Mellor et al reported that the sigmoid volvulus de-rotation without resection is associated with a 31% of recurrence rate [18]. This data was substantiated by Smith's examination, which indicated a recurrence rate of 35% [5]. However, comparing contrast enema to other non-operative treatments such as sigmoidoscopy and rectal-tube reduction, Salas et al reported a success rate of 77% vs 47% of the rectal tube, in absence of complications [4]. Mellor et al described a 79% success rate using the same technique, suggesting that contrast enema should be preferred over endoscopic detorsion, since children have thinner-walled colon than adults, making it more susceptible to perforation [6,18,21].

Over the years, other studies confirmed a similar contrast enema success rate (67% in Arnold and Nance and 69.2% in Oren) [22,23].

Conclusions

To the best of our knowledge, this is the first reported case in the literature of sigmoid volvulus detorsion by water-soluble enema (Gastrografin and 0.9% saline solution), an efficient and safe preliminary treatment of sigmoid volvulus in children.

In conclusion, we suggest that the water-soluble contrast enema fluoroscopy-guided intervention for the reduction of sigmoid volvulus should be used as an immediate non-operative treatment for this condition.

Patient Consent

Patient privacy was maintained.

Consent has been obtained for the radiological procedures performed.

The images are anonymised from which the individual cannot be identified.

These do not contain any identifying marks and are not accompanied by text that might identify the individual concerned.

Authors Contribution

RT, CV, SMM, AS and IS conceptualized the work and developed a strategy of research. RT, SMM and AS collected the evidence from the literature and screened upon CV and IS's suggestions. The authors reviewed the manuscript and approved the final version. All the authors contributed in writing.

Declaration of Competing Interest

RT, CV, SMM, AS, IS have no potential conflicts of interest to disclose.

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