

Volvulus of the sigmoid colon in a child

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ABSTRACT

Sigmoid volvulus although a common cause of large bowel obstruction in the elderly, is considered rare in the pediatric age group. We report a case of sigmoid volvulus in a 10-year-old child with mental retardation and myopathy. The various predisposing factors for sigmoid volvulus in children are discussed, and the literature on the subject is also reviewed.

Keywords: Sigmoid volvulus, children.

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Sigmoid volvulus is a common cause of large bowel obstruction in adults, especially those older than 60 years of age where an acquired redundancy of the colon as a result of constipation is an important predisposing factor.¹⁻⁴ However, this is not the case in the pediatric age group where sigmoid volvulus is rare, and in a large number of reported cases, a definite predisposing factor is present.⁵⁻⁷ This is a case report of sigmoid volvulus in a 10-year-old male child with mental retardation and myopathy, treated at Tawam Hospital, Al Ain, Abu Dhabi, United Arab Emirates, outlining aspects of diagnosis and management. The various predisposing factors for sigmoid volvulus in children are discussed, and the literature on the subject is also reviewed.

Case Report. A 10-year-old male child was admitted to the hospital with abdominal pain, abdominal distension, vomiting and fever of 18 hours duration. He was a product of a full term spontaneous vaginal delivery with a birth weight of 2.7 kg. At the age of 40 days he was admitted to the hospital due to irreducible right inguinal hernia and was also found to have a left inguinal hernia, for

which he had bilateral inguinal herniotomies. He also had a small umbilical hernia which closed spontaneously. On follow up, he was noticed to have delayed development, muscle weakness and mental retardation. His chromosomal analysis was normal and the computerized tomography (CT) scan of the brain showed slight ventricular dilatation. He was investigated for myopathy but the father refused further investigations including a muscle biopsy. At the age of 6 years he was referred to the Pediatric Surgery Clinic due to constipation. He was found to have a loaded colon with a large fecaloma in the rectum. He was advised admission to the hospital but the father refused. He was treated with enemas and home toilet training. He had barium enema which showed a distended, elongated rectosigmoid and the rectum was loaded with fecal material. Eighteen hours prior to his presentation to the hospital he started to complain of sudden onset, generalized abdominal pain, vomited 3 times and his abdomen was distended. He submitted one small watery stool. On examination, he was found to be mentally retarded with limb muscle weakness. He was pale and his temperature was 38.2°C. The abdomen was tense, distended and with generalized tenderness.

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The bowel sounds were hypoactive. Per rectal examination showed an empty rectum with a small amount of normal color stool. He was resuscitated and prepared for possible emergency laparotomy. Plain abdominal x-ray showed extensive dilation of the sigmoid and probably also the transverse colon with a big air fluid level. There was no air in the rectum. He also had Gastrografin enema (**Figure 1**). The rectum filled up with contrast, but in the junction of the upper rectum and sigmoid there was a beak-shaped narrowing pathognomonic of sigmoid volvulus. An attempt was made to derotate the volvulus, but this was not successful. The patient underwent an emergency laparotomy. He was found to have a sigmoid volvulus. The sigmoid colon was ischemic and the mesentery twisted 360° (**Figure 2**). After derotation of the volvulus, the sigmoid colon was still ischemic, so it was resected and the 2 ends were brought out as double stoma. Post operatively, the patient did well and 8 weeks later the colostomy was closed. Histology of the resected colon showed necrotic colon with viable resection margins.

Discussion. Sigmoid volvulus is a common cause of intestinal obstruction in adults, but the incidence worldwide is variable. It is particularly common in Eastern Europe, Scandinavia, India and Africa, while it is rare in Britain, Western Europe and North America.^{1-4,7} Chronic constipation resulting from irregular bowel function or secondary to drugs as in those with mental retardation and

neuropsychiatric disorders is considered the main predisposing factor in adults. Other predisposing factors include high fibre diet, pregnancy, pelvic tumors or cysts, and adhesion secondary to previous abdominal surgery.^{1,2,8} In the pediatric age group, sigmoid volvulus is rare. Over a period of 37 years from 1955 to 1992, Mellor and Drake⁹ treated only 14 cases of colonic volvulus in children between 3 months and 15 years of age, 10 of them had sigmoid volvulus. Smith et al⁹ in a review of the literature collected only 48 cases of sigmoid volvulus in children. Khanna et al⁶ from India, an area known to have a high incidence of sigmoid volvulus in adults, treated only 6 cases of sigmoid volvulus in children over a period of 10 years. Whereas sigmoid volvulus in adults is considered to be rare in North America, most pediatric cases however have been reported in the United States of America (USA).^{5,9,10} This point to different predisposing factors in children, and whereas constipation is reported in 33%-55% of children with sigmoid volvulus, it is a sequela and not a predisposing factor for volvulus, and other factors play a role.^{5,8} These include: (1) Congenital anomalous fixation of the colon.⁶ The sigmoid colon in these patients is usually long and redundant and the mesocolon is long and narrow at its parietal attachments. Our patient prior to the attack had barium enema which showed a distended, elongated sigmoid colon. (2) Hirschsprung's disease. Sarioglu et al¹¹ reported 2 cases and reviewed 8 other cases from the literature of Hirschsprung's disease with colonic volvulus, 8 of them had sigmoid volvulus



Figure 1 - Gastrografin enema showing the classic bird's beak, sign of sigmoid volvulus.



Figure 2 - Intra-operative photograph showing sigmoid volvulus with dilated, necrotic sigmoid colon.

and interestingly all were males. These patients usually have short segment Hirschsprung's disease and if the mesosigmoid is freely mobile, the dilated ganglionic sigmoid colon twist forming a sigmoid volvulus. (3) Segmental dilatation of the colon, which is an abnormally dilated intestinal segment located between normally functioning proximal and distal bowel segments. Ravansse et al¹² reported the first 2 cases of sigmoid volvulus as a complication of segmental dilatation of the colon. (4) imperforated anus,¹³ (5) absent mesocolon,¹⁴ (6) Prune-belly syndrome,⁵ (7) Ivemark's syndrome,¹⁵ (8) mental retardation,^{5,16} (9) malrotation,⁵ and (10) myotonic dystrophy.^{5,16} Our patient had a compound problem of mental retardation and myopathy and these 2 factors, as well as the already present congenitally long and redundant sigmoid colon, contributed to the development of sigmoid volvulus.

In children, the common presenting symptoms of sigmoid volvulus are abdominal pain 66%, vomiting 31% and constipation 10%, while the main signs are abdominal distension in 69% patients, tenderness in 41% and a palpable mass in 10%.^{5,17} The pre-operative diagnosis of sigmoid volvulus in children is more difficult than adults. A plain abdominal x-ray is suggestive of sigmoid volvulus in 29% of cases, while barium enema is diagnostic in 61% of cases.⁵ Plain abdominal x-ray frequently shows distended loops of bowel with marked distension of the large bowel as seen in our patient. Although, the classic bird's beak deformity seen on contrast enema is pathognomonic for volvulus, Mellor and Drake⁹ described a twisted appearance to be more common. Barium enema is valuable not only in terms of diagnosis, but it may also relieve the obstruction by reducing the volvulus. However, this may require more than one enema. Mellor and Drake⁹ were successful using barium enema to reduce 11 out of 14 children with colonic volvulus. The definitive treatment of sigmoid volvulus is surgical resection of the sigmoid colon and primary anastomosis. However, this is not always possible in the emergency setting, where a Hartman's procedure is more appropriate. In the absence of peritonitis or signs of bowel ischemia, an attempt at non-operative reduction can be made. This should be followed by bowel preparation and elective resection with end to end anastomosis. Simple derotation alone is not sufficient as this is associated with a high rate of recurrence.⁵ A variety of operative procedures to treat

sigmoid volvulus have been tried in adults including mesosigmoidoplasty, sigmoidopexy, mesocoloplasty, and extraperitonealization.^{3,18-20} Although, some of these have been occasionally used in children,⁶ they are not considered the treatment of choice.

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