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Chilaiditi's syndrome associated with colonic volvulus and intestinal malrotation—A rare case

A. Williams, R. Cox, B. Palaniappan, A. Woodward *

Royal Glamorgan Hospital, Llantrisant, Rhondda Cynon Taf, Wales, CF72 8XR, United Kingdom

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ABSTRACT

INTRODUCTION: Chilaiditi's syndrome (symptomatic hepatodiaphragmatic interposition of the colon) is an exceptionally rare cause of bowel obstruction and may present difficulty in diagnosis and management. This is the first reported case of colonic volvulus occurring in Chilaiditi's syndrome in association with intestinal malrotation and this case study describes its successful management.
PRESENTATION OF CASE: An 18 year old male presented as an emergency with vague abdominal pain and a past history of gastroschisis repair with intestinal malrotation. CT scanning showed a closed loop obstruction due to a volvulus of the colon herniating under the falciform ligament. The patient was successfully treated by surgical reduction of the hernia, anatomical correction of the malrotation and caecopexy with a tube caecostomy. At six month follow up the patient was well and asymptomatic.
DISCUSSION: In nine of the previously reported cases of Chilaiditi's syndrome with colonic volvulus, treatment was by partial colonic resection of which a third underwent stoma formation. One patient died as a consequence of anastomotic leak following primary anastomosis. We therefore suggest an alternative approach to management.
CONCLUSION: Chilaiditi's syndrome with colonic volvulus in association with intestinal malrotation has not previously been described. As there is no consensus in the literature as to how to manage such a case we suggest that reduction of the volvulus, anatomical correction of the malrotation and fixation of the caecum by tube caecostomy results in a successful outcome. This approach avoids the need for colonic resection and possible stoma formation.

1. Introduction

In 1910, Chilaiditi described the first cases of hepatodiaphragmatic interposition of the colon.[1] This was considered an incidental radiographic finding and is known as Chilaiditi's sign. However, this rare radiographic finding may be symptomatic and is then more correctly known as Chilaiditi's syndrome. It is exceptionally rare for this condition to cause bowel obstruction or require surgical correction. Furthermore, colonic volvulus causing bowel obstruction in an adult with Chilaiditi's syndrome has been reported only eleven times in the world literature.[2–12] We describe the twelfth such case and believe this is the first reported case associated with adult malrotation.

2. Case report

A previously fit and well 18-year-old man presented as an emergency with severe central colicky abdominal pain and vomiting. The severe pain settled quickly but he continued to have intermittent discomfort and vomiting. He had previously undergone repair of gastroschisis as a neonate and he reported that he had malrotation of his intestines. On clinical examination, his abdomen was mildly tender without guarding or rigidity and his full blood count, electrolytes and amylase level were normal. An erect chest radiograph revealed the presence of gas under the right hemidiaphragm suggesting the possibility of Chilaiditi's syndrome. Subsequent CT scanning demonstrated volvulus of the colon corresponding to the splenic flexure overlying the liver but no evidence of ischaemia (Figs. 1 and 2). There was inversion of the superior mesenteric artery and vein (Fig. 3) with the caecum lying in the left iliac fossa (Fig. 4). There was volvulus of the colon corresponding to the splenic flexure. Laparotomy was undertaken and the patient was found to have extensive adhesions from his previous gastroschisis repair. Having freed some of the upper abdominal adhesions it became apparent that the splenic flexure which had undergone volvulus in

* Corresponding author. Tel.: +44 01443 443541; fax: +44 01443 443224.
E-mail addresses: alan.woodward2@wales.nhs.uk, alanwoodward1@gmail.com, alanwoodward@prontomail.com (A. Woodward).

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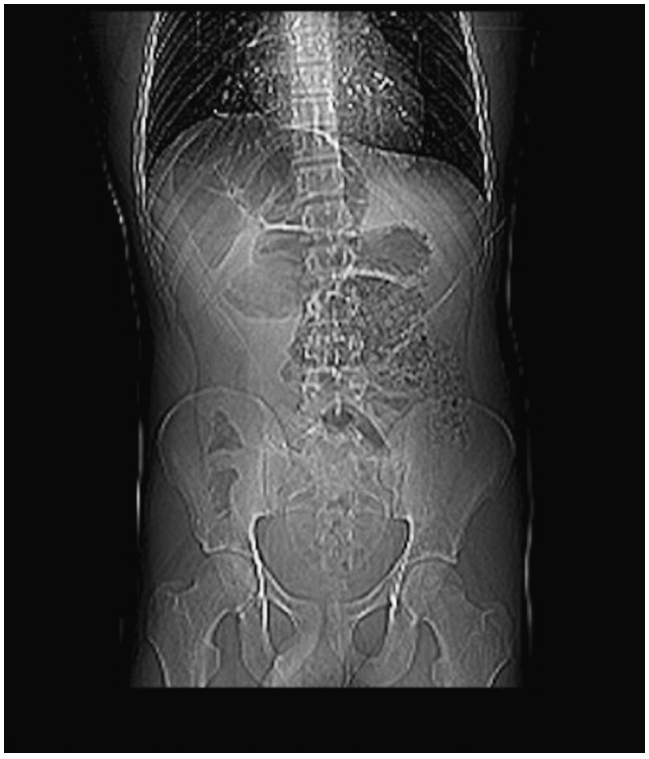

Fig. 1. CT scannergram showing caecum lying in the left iliac fossa with volvulus of the colon in the right subdiaphragmatic space.

the right subdiaphragmatic space was trapped by the taut inferior aspect of the falciform ligament. The ligament had to be divided to free the herniated colon and only then was it possible to untwist the volvulus. During the dissection, the colon became very distended and it was unclear as to the exact lie of the intestines. Therefore, the remainder of the abdominal adhesions were freed until the colon was fully mobilized and the anatomy became clear. The small intestines were confirmed to be on the right side of the abdomen with the caecum in the left iliac fossa. Having freed all

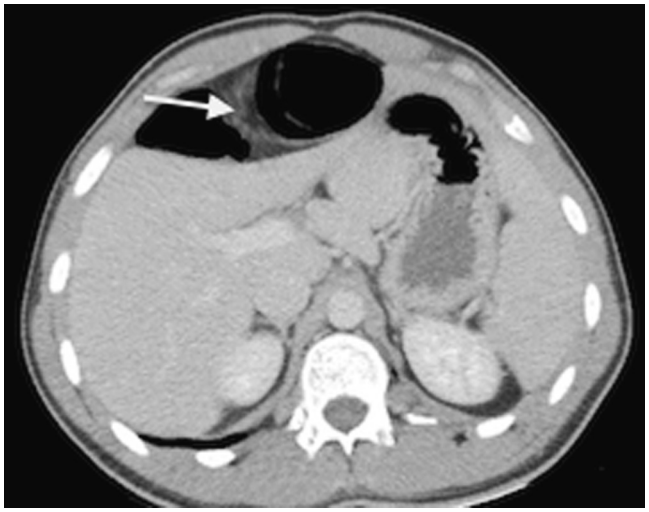

Fig. 2. CT scan showing volvulus of the colon with closed loop obstruction compressing the left lobe of the liver.

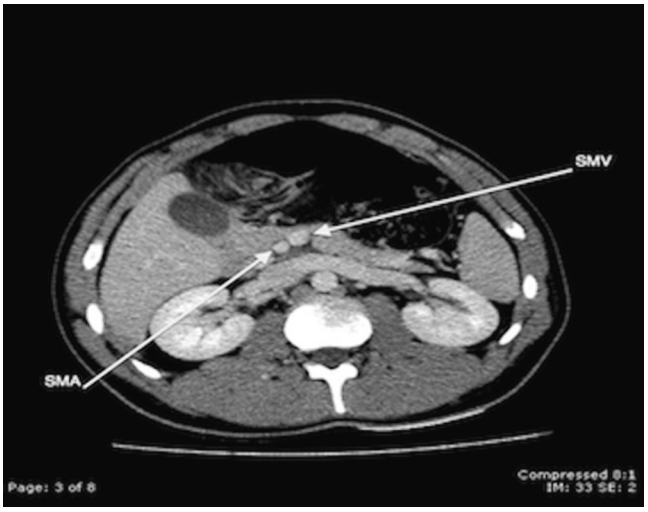

Fig. 3. CT scan showing inversion of the superior mesenteric artery (SMA) and the superior mesenteric vein (SMV).

adhesions the caecum was placed in the right iliac fossa restoring normal colonic anatomy. Due to massive colonic distention it was not possible to return the colon to the abdominal cavity in order to close the abdominal wall. Therefore, an appendicectomy was performed with a tube caecostomy placed via the appendix stump to decompress the colon. The caecostomy tube was a size 24 Foley catheter and was secured with 20 ml of saline in the balloon and a pursestring suture of 3/0 polydioxanone. Successful colonic decompression resulted, fixing the caecum in the correct position and enabling abdominal closure. Postoperatively, he developed an ileus for a few days and a CT scan was undertaken to confirm that there was no evidence of intestinal ischaemia or abscess around the caecostomy tube. He subsequently made an uneventful recovery and the caecostomy tube was removed after ten days. He was reviewed

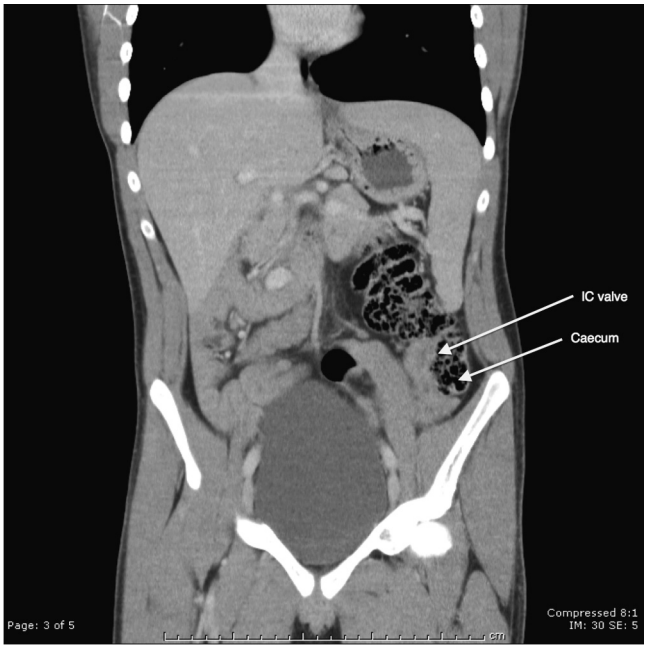

Fig. 4. CT scan showing ileocaecal valve and caecum on the left side and small bowel on the right.

at six weeks and six months postoperatively and was found to be asymptomatic on both occasions.

3. Discussion

This is only the twelfth reported case of Chilaiditi's syndrome with colonic volvulus and is the first reported case with intestinal malrotation. Two of these cases were reported in foreign languages[3,10] and one case was in a child.[8]

The initial presentation was nonspecific and it was not immediately apparent that he would require surgery. CT scanning confirmed the presence of a closed loop obstruction due to splenic flexure volvulus beneath the falciform ligament along with the presence of intestinal malrotation. In the absence of colonic volvulus, Chilaiditi's syndrome may be successfully treated by conservative means including intravenous fluid hydration.[14] However, the presence of volvulus necessitated laparotomy during which the herniated colon was found to be trapped beneath the falciform ligament. Falciform ligament hernias have previously been described and may contain small or large bowel though usually through a defect in the ligament.[13] This case was unusual in that the hepatodiaphragmatic interposition contained splenic flexure[15] as opposed to transverse colon[2,3,5,7–12] or sigmoid colon.[4] This was a consequence of the intestines rotating only 90 deg during foetal development placing the caecum in the left iliac fossa to the left of the small intestine enabling the splenic flexure to gain access to the right subdiaphragmatic space. This degree of rotation is termed nonrotation of the intestines as opposed to incomplete rotation and is technically a misnomer.

Having freed the volvulus, the distended colon could not be returned to the peritoneal cavity. A decision was taken to free all adhesions, identify the anatomy fully and undertake anatomical correction of the malrotation. This enabled decompression of the colon via a caecostomy tube in the right iliac fossa. In children, anatomical correction of malrotation is generally not advised but has been described in adults.[15] After full colonic mobilization it was a simple matter to place the caecum in the right iliac fossa restoring the normal colonic configuration. Furthermore, this manoeuvre ensured that the colon would be prevented from recurrent herniation into the subdiaphragmatic space.

In nine of the previously reported cases of Chilaiditi's syndrome with colonic volvulus, treatment was by partial colonic resection[2–7,10–12] Three of these patients underwent stoma formation[2,4,5] and there was one mortality from anastomotic leak following primary anastomosis.[7] In this case, we judged that in the absence of ischaemia, colonic resection would have been excessive and unnecessary.

4. Conclusion

In summary, we describe a rare and challenging case of colonic volvulus in an adult with Chilaiditi's syndrome. Extensive adhesions from a previous gastroschisis repair and the presence of malrotation added to the complexity of the case. There can be no consensus from the literature as to how to manage such a rare problem. However, we suggest that reduction of the volvulus, anatomical correction of the malrotation and fixation of the caecum by tube caecostomy is an acceptable alternative to colonic resection in the absence of intestinal ischaemia.

Conflict of interest

The authors report no conflict of interest.

Funding

None.

Consent

Written informed consent was obtained from the patient 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.

Authors' contributions

Anwen Williams: Assisting surgeon and subsequently writing of the case and reviewing the literature. Rebecca Cox: Reviewing the literature and obtaining relevant images. Balan Palaniappan: Additional radiology and revising the manuscript. Alan Woodward: Lead surgeon and editing the paper.

Key learning points

- CT is essential in diagnosis and management of our case.
- Colonic resection ± stoma formation is unnecessary in absence of ischaemia.
- Anatomical correction of the malrotation is acceptable in adults.

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