CASE REPORT

Acute visceral obstruction

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oliburdall@doctors.net.uk**SUMMARY**

Gastric volvulus is a rare cause for acute visceral obstruction, with a high mortality rate that rises with delay in definitive treatment. A 33-year-old woman with a rare chromosomal mutation (46,XX,del(6)(q25.1q25.3)) presented with evidence of acute visceral obstruction. Diagnostic difficulties delayed treatment and she clinically deteriorated. Urgent CT imaging revealed acute mesentero-axial gastric volvulus. At laparotomy, global failure of colonic and splenic fixation was found. The viscera were de-rotated, the stomach salvaged and gastropexy and colopexy were performed. This is the first report of gastric volvulus secondary to congenital absence of colonic and splenic ligamentous attachments occurring in a patient over 30 years of age. This case is interesting not only due to unique pathology, but also highlights that general surgeons must be aware of the possibility of unusual causes for intestinal obstruction in patients with recognised genetic abnormalities, even in adult cases, to avoid harmful diagnostic delay.

BACKGROUND

Gastric volvulus is a rare cause for acute visceral obstruction. It describes the abnormal rotation of the stomach of more than 180° along the transverse (mesentero-axial (MA)) or longitudinal (organo-axial (OA)) axis. The diagnosis is frequently delayed due to unusual presentations, with potentially catastrophic consequences; it carries a mortality rate of 30–50%.¹

Here we present a case report of a 33-year-old woman with a rare chromosomal mutation who was found to have acute MA gastric volvulus associated with complete absence of splenic and colonic fixation. This is the first report of gastric volvulus secondary to congenital absence of such ligamentous attachments occurring in an individual over the age of 30 years.

CASE PRESENTATION

A 33-year-old woman was admitted with acute abdominal distension and generalised abdominal pain. Her symptoms had begun 3 days previously with vomiting, followed by persistent non-productive retching. She had not opened her bowels or passed flatus for 3 days.

This woman was known to have a rare chromosomal mutation—an interstitial deletion on the long arm of chromosome 6, with the karyotype 46,XX,del(6)(q25.1q25.3). She had the following features associated with this chromosomal abnormality: short stature, craniostenosis mild pulmonary stenosis, parachute mitral valve, mild learning disability. Despite these problems, the patient had no

previous surgical history and was independent with daily activities.

She was tachycardic and tachypnoeic on admission. She had a grossly distended tympanic abdomen which was tender throughout, but not peritonitic. Per rectum examination demonstrated some hard stool in her rectum, without obstruction or other masses.

INVESTIGATIONS

Initial radiography showed a poorly visualised large hypodense mass, suggestive of the 'coffee bean' sign, occupying most of the mid-abdomen, thought to be in keeping with sigmoid volvulus. Decompression was attempted with rigid sigmoidoscopy. This procedure was hindered by large amounts of impacted stool in the rectum and thus phosphate enemas were given and the procedure reattempted once these had an effect. This again failed to decompress her abdomen. Blood tests were only received at this point, which showed a raised white cell count and C reactive protein, acute kidney injury and an amylase level >1000 U/L. Her initial arterial blood gas demonstrated a mild metabolic alkalosis and an elevated lactate (2.2 mmol/L).

An urgent diagnostic CT was requested due to her clinical deterioration, which concluded that she had a massively distended stomach (see figure 1), with MA gastric volvulus (demonstrated in figures 2 and 3). The jejunum-ileum was compressed into the right para-colic gutter (see figure 4). The aorta was compressed at T12 with associated left renal and hepatic hypoperfusion/segmental infarction (see figure 4). There was also compression of the inferior vena cava, renal veins and left common iliac artery. The spleen was also noted to be positioned in the right-iliac fossa (see figure 2).

DIFFERENTIAL DIAGNOSIS

Prior to the CT scan, the differential diagnosis included sigmoid volvulus or pancreatitis with an associated intestinal ileus.

TREATMENT

A nasogastric (NG) tube was placed for decompression, with 2800 mL initial output. She was resuscitated with intravenous fluids and taken to theatre for an urgent laparotomy. The findings were as follows:

1. MA gastric volvulus, stomach grossly distended.
2. Ischaemic (but viable) gastric fundus.
3. Global failure of colonic fixation to the retroperitoneum resulting in a mobile caecum, hepatic and splenic flexure. Absence of the gastrocolic ligament.

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Figure 1 Sagittal view of CT abdomen shows a grossly distended stomach which has rotated in the mesentero-axial plane, moving the antrum (A) above the fundus (B).

4. Intestinal rotation with the appendix positioned in right upper quadrant.
5. Absence of splenic ligamentous attachments, including the gastrosplenic ligament.
6. Acute pancreatitis.

The gastric volvulus was reduced, and the spleen and colon returned to their anatomical positions. The ischaemic segment

Figure 2 Sagittal view of CT abdomen shows the oesophagus (C) entering the stomach relatively low down, beneath the bulge of the antrum (D). This also demonstrates the spleen (E) in the right iliac fossa.

Figure 3 Sagittal view of CT abdomen shows the gastric antrum emptying into the pylorus (F) above the fundus (G).

of stomach was plicated. Gastropexy to the left abdominal wall was performed followed by fixation of the ascending colon and caecum to the right lower quadrant and fixation of the sigmoid colon to the left lower quadrant. A feeding jejunostomy was inserted.

OUTCOME AND FOLLOW-UP

Postoperatively this patient was transferred to the intensive care unit where she was managed supportively, with NG-free

Figure 4 Sagittal view of CT abdomen shows ischaemia of the left kidney (H) and compression of the small bowel into the right para-colic gutter (I).

Figure 5 Diagram showing organo-axial (OA) and mesentero-axial (MA) volvulus. In OA volvulus the stomach rotates around its longitudinal axis, running from the fundus to the pylorus. In MA volvulus the stomach rotates around its transverse axis, a line from the middle of the lesser curve to the middle of the greater curve, causing the pylorus to lie over the fundus.

drainage and 4-hourly aspirates. She recovered well and was discharged to the ward 3 days postoperatively.

DISCUSSION

The normal anatomy of the stomach involves four ligamentous attachments which fix the stomach in its normal anatomical position in the left upper quadrant: the gastrosplenic, gastrohepatic, gastrophrenic and gastrocolic ligaments. Despite these attachments it has been established that the stomach can twist and untwist up to 180° during normal gastric function.² It is only found to volve when twisted more than 180°, thus defining gastric volvulus.³ This requires absence/abnormal laxity of the ligaments, and is a greater risk with a fluid-filled stomach.³ The aetiology can therefore be logically divided into primary, the absence or disruption of ligamentous attachments, or secondary, disorders which predispose to either lax ligaments or gastric distension. The latter may include any defect of the diaphragm allowing gastric herniation and thus stretching of the ligamentous attachments, such as congenital diaphragmatic hernia, para-oesophageal hernia, or traumatic diaphragmatic hernia, or any condition predisposing to gastric distension such as malrotation of the intestine or strictures from gastric ulceration. Gastric volvulus has a peak incidence in the fifth decade, due to the prevalence of para-oesophageal hernia in the adult population.⁴

In 60% of cases of gastric volvulus the rotation occurs along the OA plane, where the stomach will rotate along the axis running from the fundus to the pylorus (longitudinal axis), as seen in para-oesophageal hernia (see figure 5).⁵ In these cases strangulation is more likely to occur due to obstruction at the gastro-oesophageal junction, pylorus or both.¹ In 30% of cases the stomach rotates along its transverse axis, taking the pylorus over the fundus (see figure 5).⁵ This is known as MA volvulus and requires significant laxity or absence of the ligamentous attachments in order to occur. MA volvulus therefore more often has a congenital cause. The remaining 10% of cases are those which remain unclassified, or where the stomach twists in both directions and often presents with chronic, intermittent abdominal pain.¹

This case demonstrates the difficulty in the diagnosis of acute gastric volvulus. Borchardt⁶ described a triad of presenting features comprising epigastric pain, persistent non-productive retching and inability to pass an NG tube;

however, many atypical presentations have been described. In this case, the stomach was so large that her pain and distension were no longer confined to the upper abdomen and the abdominal radiograph showed a distended viscus in the central abdomen, which is not classical of gastric volvulus. Acute gastric volvulus has a mortality of 30–50% most commonly due to strangulation, perforation and hypovolaemic shock, which have a heightened risk with delay in the diagnosis and definitive treatment.¹ Patients presenting as an emergency may have a history of vague intermittent abdominal pain as the stomach twists and untwists and thus an opportunity for elective correction is missed.

Controversy exists in the management options of gastric volvulus. Correction can be made endoscopically to decompress the volvulus; however, this will not correct the underlying cause, thus predisposing the individual to a recurrence. Ultimately these patients require surgical correction and reconstruction of the normal anatomical attachments. Laparoscopic options exist, however, in this case, given the degree of gastric distension and the patient's critical condition, open surgery was necessary.

In this case we found that the colon and spleen were not attached to the peritoneum and hence these organs were freely mobile. At laparotomy, the small bowel was entirely in the right para-colic gutter, the appendix lay in the right upper quadrant and spleen in the right lower quadrant. These operative findings are similar to those of wandering spleen and intestinal malrotation. After extensive literature search the authors found only one case of MA volvulus secondary to acquired wandering spleen in an adult; however, there were no other reported cases of a patient with global absence of colonic and splenic ligamentous attachments causing gastric volvulus in an adult over the age of 30 years.⁷ This unique finding is consistent with a large study looking at cases of intestinal obstruction secondary to congenital abnormalities, which showed a significant tail-off in numbers over the age of 12 years.⁸ It is of interest that this was her first presentation given her mobile viscera.

In conclusion, this case is not only interesting due to its unique pathology, but also demonstrates difficulty in the diagnosis of this condition, which carries a high complication and mortality rate that rises with delay in definitive treatment.

Learning points

In this case, there was a significant element of diagnostic uncertainty which resulted in a delay in definitive management. As such the following points should be highlighted:

- ▶ An awareness that gastric volvulus does not commonly fit with the usual triad of symptoms, and should therefore be considered among the differential diagnosis of all patients who present with acute obstructive symptoms, may have hastened the diagnosis.
- ▶ In the absence of a clear diagnosis and in the presence of significant symptoms, there should be a low threshold for definitive investigations such as CT imaging. In this case there were a number of hours before a CT scan was requested while the rigid sigmoidoscopy was repeated.
- ▶ General surgeons must be aware of the possibility of unusual causes for intestinal obstruction in patients with recognised genetic abnormalities, even in adult cases. This association should therefore be considered in the differential diagnosis of adults with chromosomal abnormalities who present with acute visceral obstruction to avoid harmful diagnostic delay.

for the conception and design of the report, literature search, initial draft and revision of the article, and had overall control over the publication of the paper. She is the guarantor for the paper. OCB was involved with the management of the patient after the initial presentation. He was also involved with the literature search, drafting the discussion and revising the content of the report, and reviewing the final version. SK was responsible for the initial idea to publish the report, and was involved with the operation and the long-term management of the case. He was involved with the planning and critical appraisal of the report and reviewed the final draft before submission for publication.

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Contributor SEM was involved with this patient when she initially presented to the emergency department and was present during her operation. She was responsible

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