Delayed presentation of congenital diaphragmatic hernia presenting with intrathoracic gastric volvulus: a case report and review

Hemanta K Nayak, Gaurav Maurya, Neha Kapoor, Premashis Kar

Department of Medicine,
Maulana Azad Medical
College, New Delhi, India

Correspondence to

Dr Hemanta Kumar Nayak,
drhemantanayak@gmail.com

SUMMARY

Acute gastric volvulus associated with congenital diaphragmatic hernia is an unusual surgical emergency and its presentation in adulthood is extremely rare. Gastric volvulus is an uncommon condition which is difficult to diagnose and treat. This case describes a middle aged man presenting with post-traumatic acute abdomen and finally being diagnosed as a case of gastric volvulus due to congenital diaphragmatic hernia which was till now asymptomatic and undiagnosed. Clinical examination and chest x-ray was suggestive of hiatus hernia. Contrast-enhanced CT of thorax and abdomen revealed hiatus hernia and gastric volvulus. A provisional diagnosis of post-traumatic hiatus hernia with associated gastric volvulus was made and was operated on an emergency basis. Surprisingly, the intraoperative findings revealed significant adhesions near the diaphragm suggestive of congenital diaphragmatic hernia. Hence, a diagnosis of gastric volvulus due to congenital diaphragmatic hernia was made which remained asymptomatic and undiagnosed till adulthood. He responded well to operative management and remained asymptomatic afterwards.

BACKGROUND

Congenital diaphragmatic hernia (CDH) is a rare congenital anomaly and typically manifests with respiratory distress occurring immediately after birth or in the first few hours or days of a child's life. It has a high death rate. On rare occasions, CDH can be symptomatic at an older age either as gastrointestinal obstruction or mild respiratory symptoms. In such cases, CDH can be a diagnostic confusion. Here, we described a middle aged man with symptoms of post-traumatic gastric volvulus who was found to have asymptomatic congenital diaphragmatic hernia. The hernia was till now asymptomatic and undiagnosed. This was a very rare manifestation of congenital diaphragmatic hernia.

CASE PRESENTATION

A middle aged north Indian man admitted to medicine emergency with repeated episodes of non-bilious vomiting and epigastric pain of 3 days duration. In addition, he also had postprandial abdominal fullness with dull aching epigastric pain. He had a history of blunt abdominal trauma 3 months back but was asymptomatic at that time. There was no haematemesis, melena, chest pain or dyspnoea. The patient denied any significant history of weight loss, anorexia or fever.

On examination the patient was of average build. He was afebrile and haemodynamically stable. Examination of chest showed decreased air entry in left hemithorax with resonant percussion notes in lower half of left hemithorax with tracheal shift to the right. Abdominal examination was unremarkable except for mild epigastric tenderness.

INVESTIGATIONS

Complete blood count showed normal haemoglobin and normal leucocytic count. Serum amylase and lipase were within normal limit. Chest skiagram showed a defect in left hemidiaphragm with stomach in thoracic cavity. An oesophagogastroduodenoscopy revealed severe oesophagitis with J-shaped stomach and pylorus could not be visualised. Barium swallow revealed an hour glass constriction of body of stomach through defect in the left diaphragm and distal body and pylorus above the left diaphragm in thoracic cavity (figure1). Contrast-enhanced CT of abdomen showed diaphragmatic defect, intrathoracic stomach herniation and gastric volvulus (mesentericoaxial volvulus) (figures 2 and 3).

DIFFERENTIAL DIAGNOSIS

- Post-traumatic diaphragmatic hernia and gastric volvulus
- Pneumothorax

TREATMENT

The patient underwent laparoscopic diaphragmatic hernia repair. Intraoperatively a 4×4 cm size defect in the left diaphragm to the left of the oesophageal hiatus was found and stomach and duodenum were herniated into left thoracic cavity with underlying lung collapse. However, there were fibrous adhesions present along the margin of the defects, which was suggestive of congenital diaphragmatic defect. The defect was closed with 2-0 interrupted ethibond suture.

OUTCOME AND FOLLOW UP

He remained haemodynamically stable in the post-operative period. Ryle's tube was removed on the second postoperative day. Oral sips started on the third postoperative day. He tolerated well and gradually shifted to a normal diet subsequently. Oral Gastrograffin study was normal postoperatively (figures 4 and 5). Postoperative chest skiagram was normal. He was afebrile, ambulatory and accepting orally at the time of discharge and was normal at 6 months follow-up period.

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Figure 1 Barium swallow was suggestive of hour glass constriction of body of stomach through defect in the left diaphragm and distal body and pylorus above the left diaphragm in thoracic cavity.

DISCUSSION

CDH is a rare congenital anomaly and manifested as respiratory distress immediately after birth or in the first few hours or days of a child's life with a very high death rate. On rare occasions, CDH can be symptomatic for the first time in the elderly either as gastrointestinal obstruction or with mild respiratory symptoms. In such cases, CDH be a diagnostic confusion.

Congenital diaphragmatic hernias with late onset symptoms in adulthood are exceedingly rare with few cases reported in the literature.¹⁻³ They can occur through an anterior parasternal foramen (Morgagni) or through a posterolateral, mainly left-sided, defect Bochdalek hernia (BH) representing persistence of the pleuroperitoneal canal. The overall prevalence of asymptomatic BH in adults is 6%.⁴ From all patients with a congenital BH only 5% will be diagnosed in childhood or adulthood.⁵ Adult BHs can present in two ways. They can give rise to vague, mainly gastrointestinal (abdominal pain, nausea and vomiting and constipation) or respiratory (chest pain, dyspnoea and wheezing) symptoms, followed by severe attacks and episodes of incarceration with serious consequences.⁶⁻⁹ On the contrary,

Figure 2 Contrast-enhanced CT of abdomen showed diaphragmatic defect, intrathoracic stomach herniation and gastric volvulus (mesentericoaxial volvulus).

Figure 3 Contrast-enhanced CT of abdomen showed diaphragmatic defect, intrathoracic stomach herniation and gastric volvulus (mesentericoaxial volvulus).

these symptoms can be intermittent, as herniated viscera can spontaneously reduce causing symptom regression. In such cases, radiological investigations demonstrate reduction of the hernia with symptom resolution.¹ Others may present with serious complications of strangulated herniated viscera, especially when the diagnosis has been missed or treatment delayed.⁹ There have been reports of BH presenting with sudden death from intrathoracic complications.¹⁰ Gastric volvulus is one of the rare but recognised complications of BH.⁸ Presentation with severe symptoms has been reported in 46% of cases and the mortality in these has been high (32%) because of visceral strangulation.¹¹ Diagnosis can be made with chest skiagram during an attack, especially when hollow viscera herniates though large defects.⁶ CT scan can detect small asymptomatic BH and a definitive diagnosis can be achieved with barium or gastrograffin meal and swallow.^{8, 12}

This patient developed acute abdomen after 1 month of abdominal blunt trauma. Chest skiagram was suggestive of intrathoracic herniation and the CT was that of gastric volvulus. The decision for immediate surgery was appropriate and no time was wasted with further investigations. The outcome was good and the hernia was repaired before strangulation took

Figure 4 Postoperative oral Gastrograffin study was found to be normal study.

Figure 5 Postoperative oral Gastrograffin study was found to be normal study.

place. However, intraoperative findings were suggestive of congenital diaphragmatic hernia. Hence, it was evident that the patient had a congenital diaphragmatic hernia, gastric volvulus of the herniated stomach leading to its acute presentation.

Delay in diagnosis and treatment of GV will lead to a poor outcome. Death rate up to 80% has been reported in non-operated patients with GV. To reduce intragastric pressure and prevent gastric necrosis, a nasogastric tube should be promptly inserted. The principles of the surgical procedure include de-rotation of the volvulus, reduction of the herniated stomach, closure of the associated diaphragmatic defect and fixation of the stomach to the anterior abdominal wall.^{13 14}

In conclusion, acute gastric volvulus associated with congenital diaphragmatic hernia is an unusual surgical emergency and its presentation in adulthood is extremely rare. However, prompt diagnosis and early surgical intervention leads to a good outcome.

Learning points

- Congenital diaphragmatic hernia presenting with post-traumatic gastric volvulus in the elderly is a very rare manifestation.
- Early diagnosis and prompt surgical intervention were required for good outcome.
- No time should be wasted before surgery once a diagnosis of gastric volvulus is made because it is highly prone to incarceration.

Competing interests None.

Patient consent Obtained.

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