CASE REPORT

Congenital diaphragmatic hernia coexisting with acute intra-thoracic gastric volvulus – a case report

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Abstract

Gastric volvulus, a rare condition in children, is the twisting of the whole or part of the stomach to at least 180º about an axis that causes foregut obstruction with a life-threatening presentation and fatality if prompt diagnosis and intervention are not done. Association of gastric volvulus with congenital diaphragmatic hernia is a rare occurrence because of the temporal differences in their occurrence. Gastric volvulus is a disease of adults, while congenital diaphragmatic hernia is a disease of newborns. We report a case of congenital diaphragmatic hernia coexisting with intra-thoracic gastric volvulus in a 9-year-old girl who had a successful prompt diagnosis and surgical intervention. This report is aimed at showing that coexisting congenital diaphragmatic hernia and gastric volvulus, though a rare phenomenon, occurs in children.

Background

The word volvulus is derived from the Latin word *Volvore* which means to roll or twist. Gastric volvulus means twisting of the whole or part of the stomach to at least 180º about an axis that causes foregut obstruction.[1] When the twisting of the stomach is less than 180º it is termed partial gastric volvulus or gastric torsion.[1] Gastric volvulus in children was first described by Olfmann in 1899.[2] It remains a rare condition in children that usually presents before the age of 1 year with equal sex distribution.[3] The presentation can be acute, intermittent, or chronic. The acute presentation is a life-threatening condition that requires prompt diagnosis and treatment.

Singleton[4] proposed a classification of gastric volvulus based on their axis of rotation; Organo-axial volvulus is characterized by rotation of the stomach along its long axis while in mesenteric-axial volvulus, the rotation is around the short axis of the stomach from the lesser to greater curvature. Whereas the Mesenteric-axial volvulus is commoner in children, the Organo-axial volvulus is commonly associated with diaphragmatic defect. Gastric volvulus can also be classified into intra-abdominal or intra-thoracic gastric volvulus based on their location, and based on their aetiology, it is classified into primary or secondary gastric volvulus. Primary gastric volvulus is caused by distortion of the normal attachment of the stomach. Secondary gastric volvulus is due to abnormality of the structures adjacent to the stomach such as diaphragmatic defect and wandering spleen.

Congenital diaphragmatic hernia is a congenital disorder associated with abnormal growth affecting 3.6:10,000 newborns.[5] It occurs due to incomplete closure of the pleuro-peritoneal channel during foetal development.[6] It commonly presents on the left side in 88% of cases with difficulty in breathing in the neonatal period because of the peritoneal viscera protrusion into the pleural cavity in the chest.[6] Late presentation is rare and is seen in 5-30% of all cases of congenital diaphragmatic hernia.[7,8]

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This report is a rare case of late presentation of a congenital diaphragmatic hernia coexisting with acute intra-thoracic gastric volvulus in a 9-year-old child.

Case report

H.A.M. is a 9-year-old girl who presented to the emergency paediatric unit of our hospital with a two-day history of difficulty in breathing, dysphagia, non-bilious vomiting, and epigastric pain. The vomitus was characterised by initial gastric content followed by non-productive retching. The difficulty in breathing and dysphagia were associated with failed nasogastric tube insertion.

There was a previous history of recurrent left hypochondrial discomfort with associated early satiety which was managed symptomatically with an over-the-counter drug purchased from a nearby patent medicine store. There was no history of recurrent vomiting, or haematemesis before the current presentation. The patient was however noticed to be small for her age compared to her peers and siblings.

On physical examination, the patient was dyspnoeic, not pale, not cyanosed, and not febrile. The child was underweight, height was <95 percentile (stunted) and the respiratory system examination revealed tachypnoea with a respiratory rate of 38c/min, normal SPO2, and absent breath sound was noted in the left hemithorax. Other systemic examinations were normal.

The chest radiograph (Fig. 1) revealed non-visualization of the outlined left hemidiaphragm and upward displacement of the gastric fundal gas into the left mid and lower zones with a resultant shift of the mediastinum to the right hemithorax. There is crowding of the left broncho-vascular markings. The abdominal radiograph (Fig. 2) also demonstrated an absence of fundal gas in the abdomen. The barium meal (Figs. 3-4) confirmed the intra-thoracic location of the stomach. The stomach was also found to be rotated along its long axis with the greater curvature lying superior to the lesser curvature. The gastric fundus is rotated postero-inferiorly while the gastric antrum is rotated antero-superiorly.

Other laboratory investigations such as

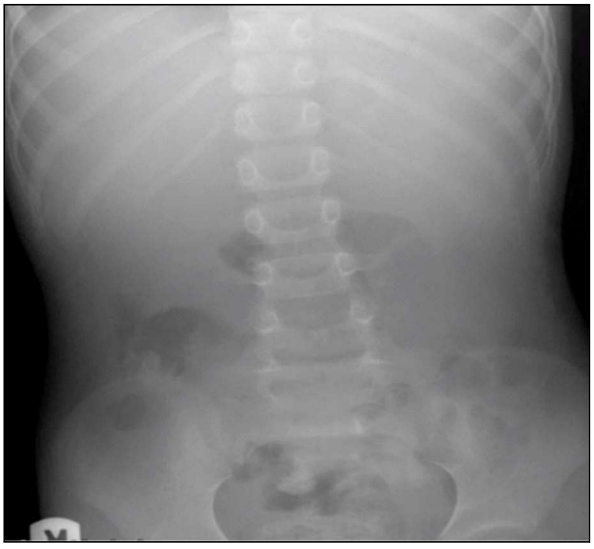

Figure 1: PA chest radiograph revealed non-visualisation of the outline left hemi diaphragm. There is a thick wall lucency occupying the left mid and lower zones which continue into the left hypochondrium reflecting air within the displaced stomach.

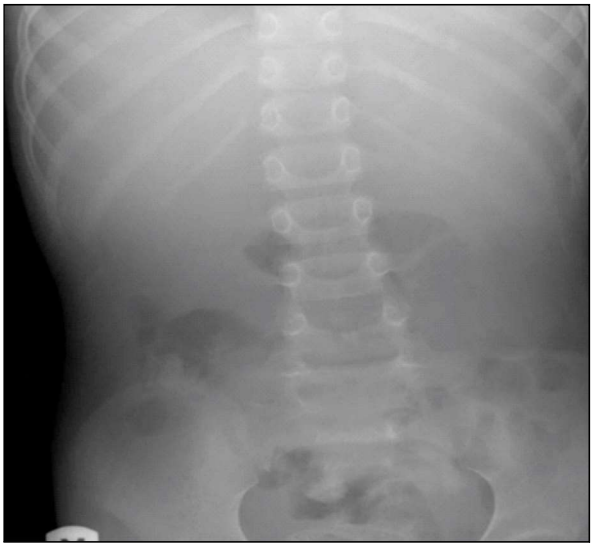

Figure 2: Abdominal radiograph showing the absence of fundal gas in the abdomen.

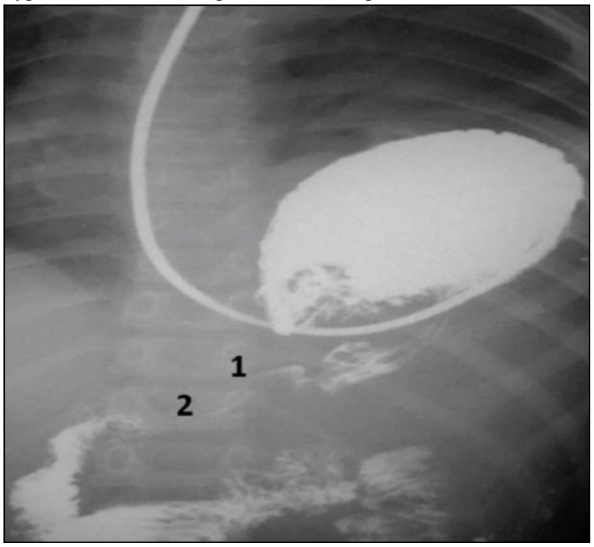

Figure 3: AP spot film of the barium meal confirming the intra-thoracic location of the stomach. The first (1) and the proximal portion (2) of the second part of the duodenum were seen dragged superiorly and across the midline.

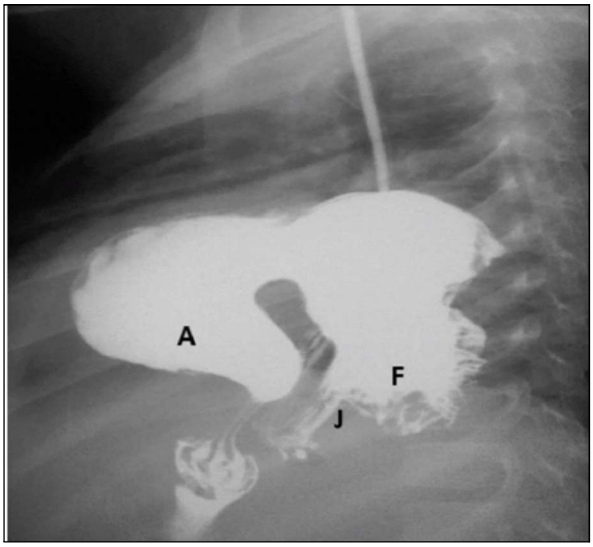

Figure 4: Lateral spot film of the barium meal showing the intra-thoracic location of the stomach. The stomach is rotated along its long axis with its greater curvature lying superior to the lesser curvature. The gastric fundus (F) is rotated posterior-inferiorly while the gastric antrum (A) is rotated anterior-superiorly. The gastroesophageal junction is labelled (J).

electrolytes urea and creatinine, full blood count, and urinalyses were within normal limits. Diagnosis of left diaphragmatic hernia was made. The patient was transferred to the paediatric surgery unit where an emergency laparotomy was performed. Intraoperatively the patient had repair of the diaphragmatic defect and anterior gastropexy after reduction of hernia content. The postoperative period was unremarkable; the patient had antibiotics and intravenous fluid post-op and was discharged home six days after surgery.

Discussion

Congenital diaphragmatic hernia is a defect that results from incomplete closure of the pleuro-peritoneal canal. The defect allows intrabdominal organs including the stomach to herniate into the thoracic cavity. The diaphragmatic defect is usually associated with agenesis or elongation of the gastro-phrenic ligament which will predispose the stomach to rotates along its mesentery.[9,10] More so, the diaphragmatic defect also increases the space under the diaphragm which also predisposes the stomach to an increased risk of gastric volvulus like the current case.[11]

Diaphragmatic hernia typically presents at birth as a congenital condition, affecting newborns with a delayed presentation being a rare occurrence. In a seven-year review of thirty-three children with diaphragmatic hernias in Egypt, Elhalaby *et al*[12] reported that fifteen had delayed presentation with ages ranging from two months to 14 years. The report also noted that the late-presenting diaphragmatic hernia was frequently left-sided accounting for about 60% of the cases presenting outside the neonatal age group. The highlighted case report was a delayed presentation of a diaphragmatic hernia in a 9-year-old girl which was also on the left side.

The patient in this case presented with epigastric pain and vomiting with an initial gastric content followed by non-productive retching. These are classical presentations of gastric volvulus.[7] The index patient also presented with difficulty in breathing and dysphagia associated with failed nasogastric tube insertion. Although the inability to pass a nasogastric tube is a common feature in adults, this occurrence is rare in children.[12] These symptoms may have resulted from reduced volume of the thoracic cavity and the compression of the lungs and oesophagus due to the herniated stomach.

Although CT and MRI have been suggested for correct diagnosis, plain radiographs, which is the usual first-line imaging modality and barium meal study are diagnostic for both diaphragmatic hernia and organo-axial gastric volvulus.[8] The index case was diagnosed based on the classical features demonstrated on barium meal and chest and abdominal radiographs. These features include diaphragmatic defect, herniation of the stomach into the thoracic cavity, organo-axial gastric malrotation seen as an inverted stomach with the greater curvature above and medial to the lesser curvature, posterior-inferior displacement of the gastric fundus, and anterosuperior displacement of gastric antrum on barium meal.

The chest and abdominal radiographic features of diaphragmatic hernia fulfilled by the index case include the displacement of the gastric fundal gas into the left mid and lower zones with resultant shift of the mediastinum to the contralateral side, crowding of the left broncho-vascular markings and absence of fundal gas in the abdomen.

The treatment of choice for patients with diaphragmatic hernia is diaphragmatic defect repair and anterior gastropexy via open or laparoscopic technique. The index patient had an open diaphragmatic defect repair with anterior gastropexy. The postoperative period was uneventful, and the patient was discharged home 6 days after surgery.

Conclusion

Diaphragmatic hernia with coexisting gastric volvulus is a rare phenomenon in children. Plain radiographs and barium meal are diagnostic, and prompt surgical intervention reduces associated mortality.

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