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Gastric antrum hypertrophy causing outlet obstruction in an infant with congenital diaphragmatic hernia

Ahmad Saad[a,*], **Barton Kenney**[b], **Robert Touloukian**[a]

[a]*Section of Pediatric Surgery, Yale University School of Medicine and Yale New Haven Children's Hospital, New Haven, CT 06520, USA*
[b]*Department of Laboratory Medicine, Yale University School of Medicine, New Haven, CT 06520, USA*

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Abstract Congenital diaphragmatic hernia (CDH) is associated with multiple congenital anomalies affecting several organ systems, including the gastrointestinal system. Pyloric stenosis and bands are known and previously reported etiologies of gastric outlet obstruction in infants with CDH. We report the first case of gastric antrum hypertrophy causing gastric outlet obstruction in an infant with CDH.

Major anomalies have been associated with congenital diaphragmatic hernia (CDH), in approximately 39% of liveborn cases [1], rising to approximately 95% in those with intrauterine death [2]. The most commonly encountered anomalies are cardiac, renal, central nervous system, and gastrointestinal [3]. Both major and minor gastrointestinal anomalies have been reported in association with 14% of cases of CDH. The most common gastrointestinal associations are intestinal fixation anomalies and gastroesophageal reflux. Several cases of hypertrophic pyloric stenosis have been reported [4,5], with an incidence of 1.2% in a recently published database of outcomes with CDH [6].

Feeding intolerance, secondary to ileus and gastroesophageal reflux, is often experienced postrepair; but more rarely encountered conditions may be overlooked. We report such a patient with delayed diagnosis of gastric antrum stenosis causing outlet obstruction.

1. Case

A 3.2-kg male infant was born after 39 weeks of gestation, by cesarean section, to a 33-year-old gravida 1 mother. Diagnosis of left CDH was known by prenatal ultrasound at 21 weeks of gestation with the stomach and small bowel seen in the left hemithorax. Gestation was complicated by diabetes and polyhydramnios.

The infant was immediately intubated; an orogastric suction tube was placed for decompression. Chest x-ray at birth showed evidence of left diaphragmatic hernia and mediastinal shift to the right (Fig. 1). The infant maintained oxygen saturation of 95% on room air but gradually became tachypneic with a respiratory rate of 40 to 50 breaths per minute. Echocardiogram and renal ultrasound studies were normal.

On the fourth day of life, the infant was taken to the operating room for open abdominal repair of CDH. The stomach, spleen, loops of small intestine, left kidney, and adrenal were found in the left hemithorax. The posterior leaf of the diaphragm was absent. The abdominal viscera were reduced back to the abdominal cavity, the diaphragmatic

* Corresponding author. Tel.: +1 203 785 7890; fax: +1 203 737 5209.
E-mail addresses: ahmad.saad@yale.edu (A. Saad), barton.kenney@yale.edu (B. Kenney), robert.touloukian@yale.edu (R. Touloukian).

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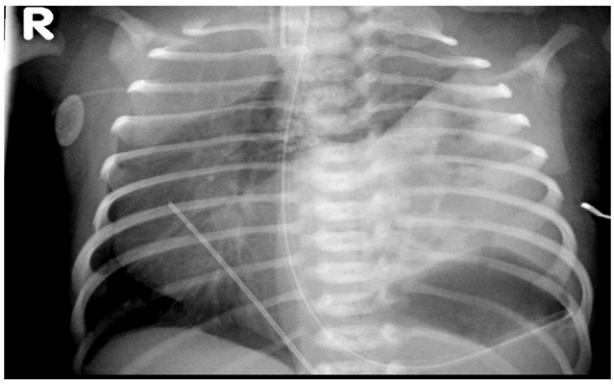

Fig. 1 Initial chest x-ray showing bowel gas pattern in the left hemithorax with mediastenal shift to the right consistent with CDH.

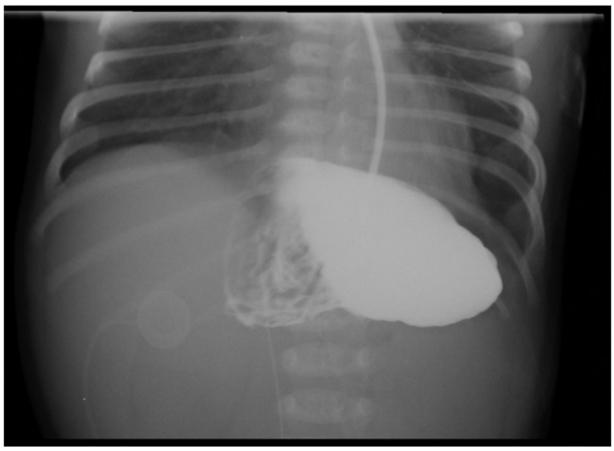

Fig. 2 Upper gastrointestinal study at 21 days of life shows no progression of the contrast beyond the stomach.

defect was closed using 4.0 silk sutures approximating the anterior portion of the diaphragm to the posterior ribs, and a left chest tube was placed. At this time, the bowel was noted to be air filled and distended, requiring ventral hernia repair.

On postoperative day 4, the abdomen was primarily closed.

During his hospital course, the infant was noted to have increased orogastric tube output. On the 18th day of life, radiographic studies showed absent gas and contrast in the bowel distal to the stomach (Fig. 2). A Ladd procedure was performed for obstruction with duodenal bands extending to the pylorus. At that time, air could be expressed from the pylorus into the small bowel.

On the fifth week of life, the infant continued to have high orogastric tube output with intolerance of tube feeds. Upper gastrointestinal study continued to show contrast in the stomach without further progression into the small intestine. At reexploration, a proximal gastrostomy was performed, and a stenotic area was identified in the gastric antrum. The pylorus and small intestine were normal distal to the stenosis. A proximal antrum gastrectomy with gastrogastric anastomosis was performed. Postoperatively, the infant tolerated tube feeds. Upper gastrointestinal contrast study 4 weeks after partial gastrectomy showed a rapid emptying of contrast from stomach into intestine. The infant was discharged home at 2 1/2 months of age on full feeds.

2. Pathology

Histologic examination of the resection specimen revealed prominent hypertrophic changes within the gastric

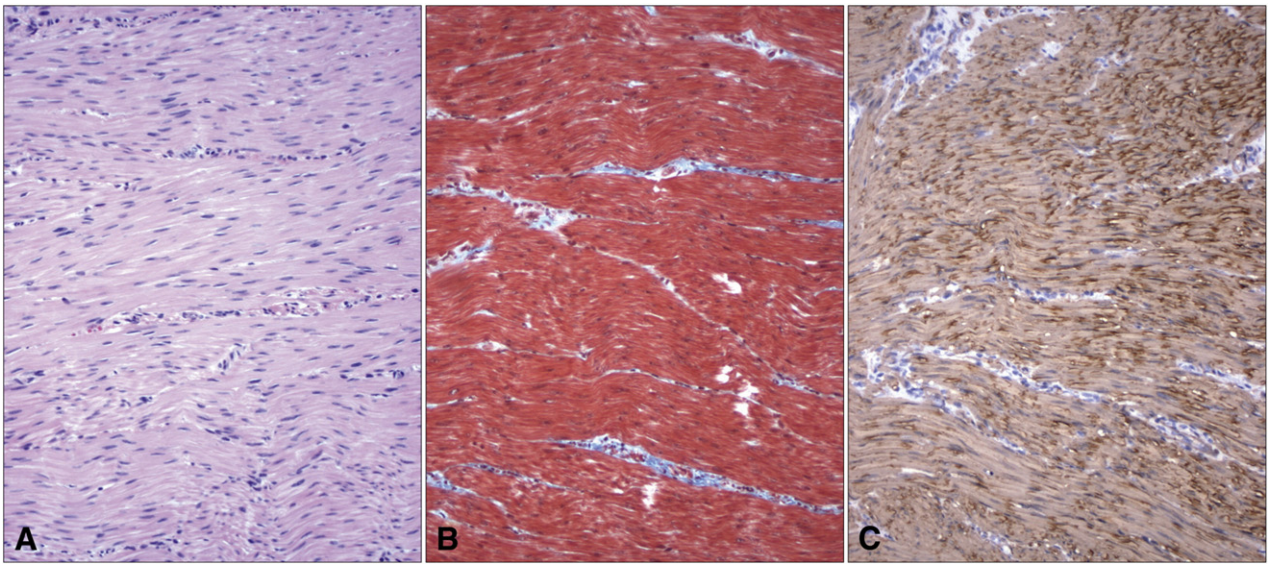

Fig. 3 A, Muscular hypertrophy, with abundant, haphazardly arranged muscle fibers (hematoxylin and eosin [H&E], original magnification ×10). B, Red histochemical staining with trichrome, confirming muscle differentiation and revealing no significant component of fibrosis (Klatskin trichrome, original magnification ×10). C, Positive immunostaining for smooth muscle actin, again confirming muscle differentiation (smooth muscle actin immunostain, original magnification ×10).