A rare coexistence of two gastric outlet obstructive lesions: infantile hypertrophic pyloric stenosis and organoaxial gastric volvulus

Pelin Oğuzkurt[1], M. Emin Şenocak[1], Akgün Hiçsönmez[1]

[1]*Department of Pediatric Surgery, Hacettepe University Faculty of Medicine, Ankara, Turkey*

SUMMARY: Oğuzkurt P, Şenocak ME, Hiçsönmez A. A rare coexistence of two gastric outlet obstructive lesions: infantile hypertrophic pyloric stenosis and organoaxial gastric volvulus. Turk J Pediatr 2000; 42: 87-89.

Infantile pyloric stenosis is one of the most common conditions requiring surgery during the first few weeks of life. The association of infantile pyloric stenosis with gastric volvulus in an extremely uncommon occurrence. A 10-month-old male infant operated for infantile pyloric stenosis at two months of age is presented. His current problem was recurrent pulmonary infections and he was diagnosed to have organoaxial gastric volvulus and gastroesophageal reflux. The common features of presentation, radiological findings, surgical procedures and possible mechanisms of gastric volvulus associated with infantile pyloric stenosis are discussed.

Key words: gastric volvulus, infantile hypertrophic pyloric stenosis, gastric outlet obstructions.

The association of infantile pyloric stenosis (IPS) with other gastrointestinal tract anomalies is rarely encountered[1,2]. The uncommon association of IPS and gastric volvulus is not throughly mentioned in the literature. Gastric volvulus (GV), mesenteroaxial or organoaxial, is a rare condition in infancy and childhood[3,4]. It is known that conditions such as mental retardation with aerophagia, gastric outlet obstruction and tracheo-esophageal fistula, which result in gastric distention, may predispose to gastric volvulus[4,5]. It may occasionally be associated with diaphragmatic hernia or eventration, congenital bands and disorders causing gastric distension, and deficient gastric attachments as well as asplenia, or it may be idiopathic[6-9]. The association of two obstructive upper gastrointestinal disorders and late presentation of organoaxial volvulus after relief of infantile pyloric stenosis are discussed in this report.

Case Report

A 10-month-old male infant was hospitalized with symptoms of fever, cough, grunting respirations, and intercostal and subcostal retractions. He had had several attacks of pneumonia over the previous four months with clinical pictures milder than with the current attack. His past history revealed an operation for IPS at the age of two months. During Ramstedt's pyloromyotomy, duodenal perforation was noticed and was primarily sutured. The postoperative period was uneventful and he had been discharged on the 8th postoperative day. He was in good health until the time of his first severe pneumonia attack at six months of age. Chest examination revealed fine rales in both lungs. The physical examination of the abdomen revealed a supraumbilical right transverse incision and epigastric distention. Routine laboratory tests including complete blood count, urinalysis and blood chemistry were within normal limits. The sweat chloride test and quantitative immunoglobulins A, M, G were normal. The posteroanterior chest X-ray revealed interstitial pneumonia and right paracardiac infiltration with a distended stomach with double superimposed air-fluid level (Fig. 1). While the technetium-99m sulphur colloid scan revealed

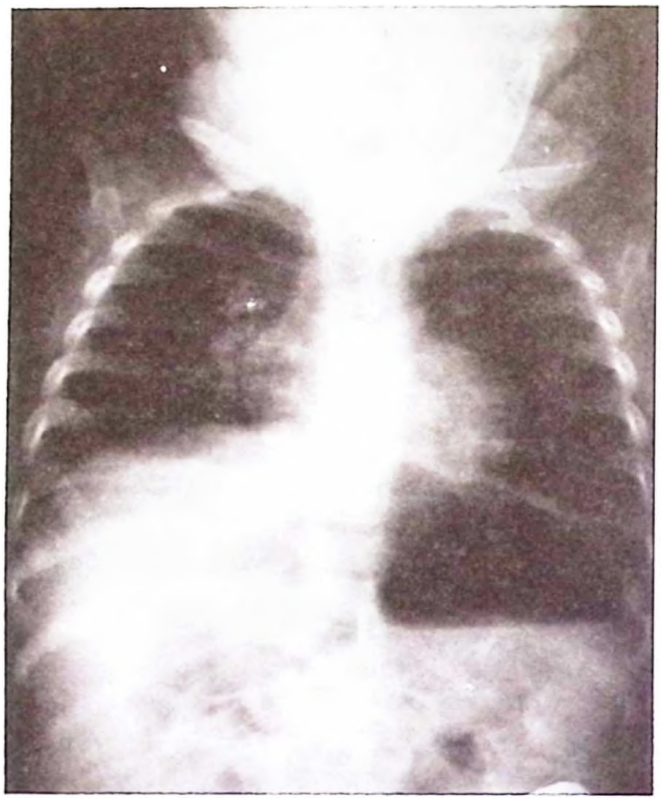

Fig. 1. Configuration of the distended stomach and double superimposed air-fluid levels suggestive of upside down stomach on plain erect X-ray consistent with organoaxial gastric volvulus.

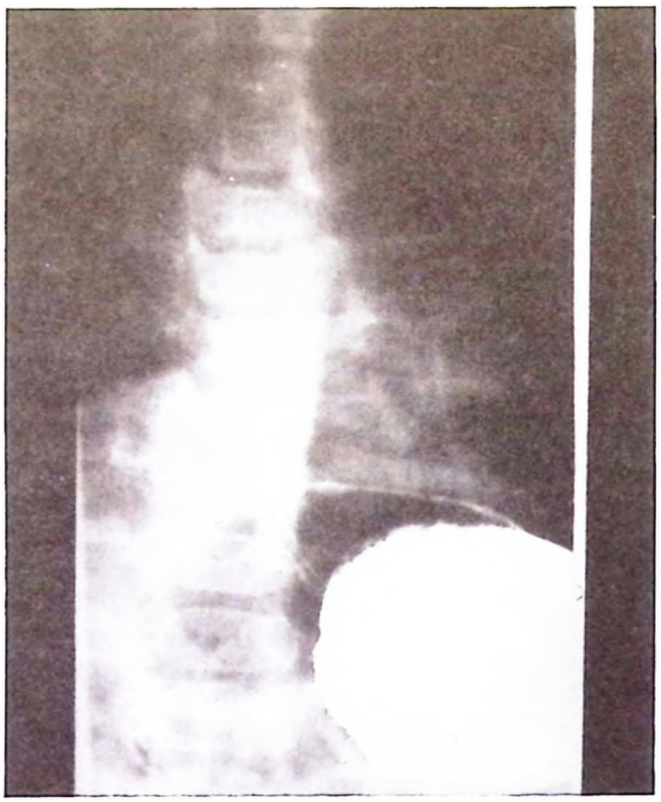

Fig. 2. Upper gastrointestinal series confirms the presence of organoaxial gastric volvulus associated with gastroesophageal reflux.

grade 2 gastroesophageal reflux, the barium swallow revealed both gastroesophageal reflux and organoaxial gastric volvulus (Fig. 2). After 15 days of antibiotic treatment, he underwent surgery. The greater curvature of the stomach was free and was located below the diaphragm under the left lobe of the liver. No adhesive bands due to the previous operation were identified. The greater curvature of the stomach was reduced and anterior gastropexy was performed with 3/0 nonabsorbable interrupted sutures. The postoperative period was uneventful and the patient was discharged on the 7th postoperative day. The barium swallow, performed 15 days after the operation, was normal. During six months follow-up, the patient did not have symptoms of gastroesophageal reflux disease and had no respiratory infections.

Discussion

Infantile pyloric stenosis, one of the most common conditions requiring abdominal surgery during infancy, may be rarely associated with other alimentary tract anomalies[1,10]. In large series, the malformations mentioned are esophageal atresia, malrotation, Meckel's diverticulum, diaphragmatic hernia, Hirschsprung's disease and exomphalos[1]. The occurrence of GV has not been previously mentioned among the associated alimentary tract anomalies in IPS.

Gastric volvulus, which is defined as an abnormal degree of rotation of one part of the stomach around another, is a rather uncommon condition in children[11]. The stomach is securely held in place by gastrophrenic ligaments and esophageal hiatus, the retroperitoneal fixation of the duodenum, the short gastric vessels and the gastrocolic ligament[12]. For a volvulus to occur, these ligaments must be absent or stretched[4]. It is reported that gastric distention may also predispose to GV[3,4]. Although ulceration or tumors causing gastric outlet obstruction may result in gastric volvulus in adults[4], in children GV may occur in mentally retarded and aerophagic patients[5], as well as in patients with tracheoesophageal fistula (TEF),

both of which result in gastric distention[4]. According to the clinical course, GV is classified as acute or chronic. It has been reported that no identifiable anomaly has been detected in 33.3 percent of the cases with chronic gastric volvulus[7].

Gastric volvulus may cause nonspecific symptoms such as epigastric distention and episodic vomiting or retching, or chronic aspiration symptoms due to gastroesophageal reflux which was successfully treated with conservative measures without any need for fundoplication[11]. Localized distention of stomach and horizontal position with a single air fluid level or double superimposed air-fluid levels may be seen[5]. It may even be missed in barium swallow showing the lower position of the esophagogastric junction and distortion of the antrum and duodenum[5].

In our clinic, 116 patients were diagnosed as IPS between December 1989 and December 1996. In this series, the associated alimentary tract anomalies were H-type TEF in one case, Hirschsprung's disease in one case and duodenal web in the second part of the duodenum in another case. The only operative complication was duodenal perforation which was recognized during the operation and primarily sutured. The postoperative complications in this series were two cases of eventration.

Organoaxial gastric volvulus was not recognized as an associated malformation or postoperative complication in our series nor in the large series in the literature[1]. In the presented case, organoaxial gastric volvulus was probably present when the patient was operated for IPS. An upper gastrointestinal series was not performed preoperatively because of the greater curvature of the stomach during the operation. Our patient presented with the complications of chronic GV such as gastroesophageal reflux and pneumonia at 10 months of age, but no responsible anomaly was identified.

Hypertrophic pylorus in association with gastric volvulus was reported in a newborn who was found to have a distended stomach and hypertrophied pylorus at laparotomy for organoaxial gastric volvulus; both gastropexy and pyloroplasty were performed[5]. Therefore, IPS causing gastric distention may be a predisposing factor for the occurrence of gastric volvulus. Khemani et al.[13] reported organoaxial gastric volvulus following vagotomy and pyloroplasty for chronic gastric outlet obstruction, and attributed the pathology to the laxity of the gastric suspensory ligaments and postoperative adhesions. This may explain the late presentation of organoaxial gastric volvulus after gastric decompression due to the relief of gastric outlet obstruction[13].

The presented case represents and idiopathic form of GV with chronic symptoms which manifest at a later age probably after subclinic attacks of mild vomiting or gastroesophageal reflux disease. It is therefore suggested that the presented case represents a coexistence of two obstructive upper gastrointestinal pathologies.

REFERENCES

1. Ahmed S. Infantile hypertrophic pyloric stenosis associated with major anomalies of the alimentary tract. J Pediatr Surg 1970; 5: 660-666.
2. Zenn MR, Redo SF. Hypertrophic pyloric stenosis in the newborn. J Pediatr Surg 1993; 28: 1577-1578.
3. El-Gohary MA, Etiaby A. Gastric volvulus infants and children. Pediatr Surg Int 1994; 9: 486-488.
4. Ziprkowski MN, Teele RL. Gastric volvulus in childhood. AJR 1979; 132: 921-925.
5. Cameron AE, Howard ER. Gastric volvulus in childhood. J Pediatr Surg 1987; 22: 944-947.
6. Aoyoma K, Tateishi K. Gastric volvulus in three children with asplenic syndrome. J Pediatr Surg 1986; 21: 307-310.
7. Dudgeon DL. Lesions of the stomach. In: Ashcraft KW, Holder TM (eds). Pediatric Surgery (2nd ed). Philadelphia: W.B. Saunders Company; 1993: 295-297.
8. Rowe MI, O'Neill JA Jr, Grosfeld JL. *Essentials of Pediatric Surgery*. St Louis: Mosby Company; 1995: 504-505.
9. Şenocak ME, Büyükpamukçu N, Hiçsönmez A. Chronic gastric volvulus in children: a 10 year experience. Z Kinderchir 1990; 45: 159-163.
10. Bell MJ. Infantile pyloric stenosis: experience with 305 cases at Louisville Children's Hospital. Surgery 1968; 64: 983-989.
11. Samuel B, Burge DM, Griffiths DM. Gastric volvulus and associated gastro-oesophageal reflux. Arch Dis Child 1995; 73: 462-464.
12. Scherer LR. Peptic ulcer and other conditions of the stomach. In: O'Neill JA, Rowe MI, Grosfeld JL, Fonkalsrud EW, Coran AG (eds). Pediatric Surgery (5th ed). St Louis: Mosby; 1998: 1127-1128.
13. Khemani R, Kantheria C, Shah P. Organoaxial volvulus of the stomach following corrective surgery for gastric outlet obstruction. Indian J Gastroenterol 1992; 11: 144.