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Retrograde pylorogastric intussusception – Case report and review

Efrat Avinadav [a,*], Hagit Poran Feldman [b], Eyal Zifman [c], Yaniv Lakovsky [d], Enrique N. Freud [e]

[a] Department of Pediatric Surgery, Schneider Children's Medical Center of Israel, 14 Kaplan St., Petach Tikva 4920235, Israel
[b] Department of Pediatric B, Schneider Children's Medical Center of Israel, Petach Tikva 4920235, Israel
[c] Institute of Gastroenterology, Nutrition, and Liver Diseases, Schneider Children's Medical Center of Israel, Petach Tikva 4920235, Israel
[d] Department of Radiology, Schneider Children's Medical Center of Israel, Petach Tikva 4920235, Israel
[e] Department of Pediatric Surgery, Schneider Children's Medical Center of Israel, Petach Tikva 4920235, Israel

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ABSTRACT

A case of gastric outlet obstruction in an infant due to retrograde intussusception of the pylorus into the stomach is presented. This anomaly is extremely rare, with almost no reports in the literature. The patient underwent formal Heineke-Mikulicz pyloroplasty with an uneventful recovery and resumed full enteral feeding.

1. Case report

1.1. Patient and history

A full-term female infant perinatally diagnosed with hydrocephalus was born by cesarean section with Apgar score 6 at 1 min and 8 at 5 min. Respiratory difficulties requiring intubation were noted shorty after delivery. A diagnosis of meningomyelocele was made postnatally. The infant was admitted to the neonatal intensive care unit where she underwent surgical repair of the meningomyelocele (day 2), placement of a ventriculoperitoneal shunt (day 10), and surgical gastrostomy for feeding (day 23). After a short period out of hospital, she was readmitted at age 1 month because of poor feeding and apathy.

1.2. Imaging findings

Brain magnetic resonance imaging demonstrated multifocal hemorrhagic lesions, but there were no signs of shunt malfunction. The patient showed poor tolerance for gastrostomy feeding, with bilious drainage from the gastric tube accompanied by bilious and nonbilious vomiting. A small amount of water-soluble contrast was introduced through the gastrostomy, and an X-ray performed a few hours later demonstrated normal passage into the colon and a large, air-filled stomach (Fig. 1). Subsequent abdominal sonography revealed no pathological findings.

During the next 6 days, there was no change in the clinical picture of feeding intolerance and bilious and nonbilious gastrostomy drainage. At that point, a second sonographic examination showed an intussusception in the middle of the abdomen (Fig. 2). Barium enema performed shortly after ruled out ileocolic intussusception (Fig. 3). This was followed by upper fluoroscopy which showed a gastric outlet obstruction as well as a filling defect at the pre-pyloric area (Fig. 4). The finding was confirmed by gastroscopy (Fig. 5).

1.3. Operative course

The patient was prepared for surgery. The abdomen was approached via extension of the upper middle abdominal scar from the previous gastrostomy operation. On exploration, a mass was

* Corresponding author.
E-mail address: EfratAv1@clalit.org.il (E. Avinadav).

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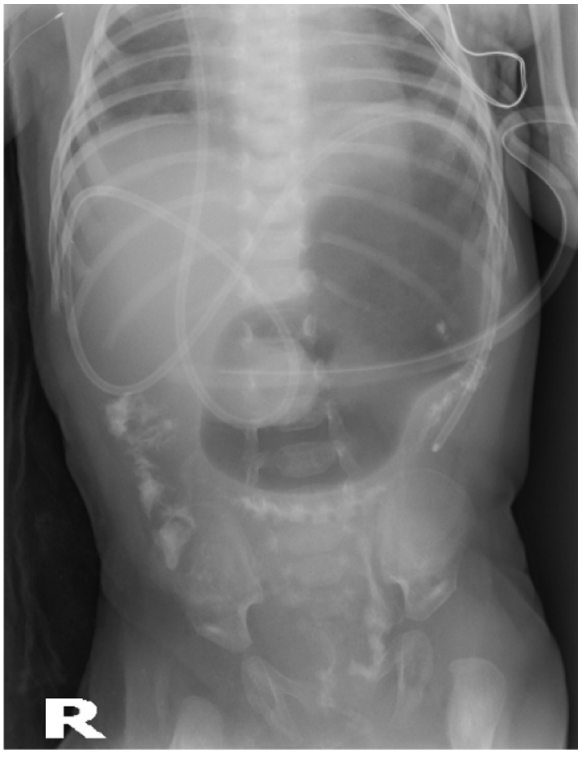

Fig. 1. Abdominal X-ray film demonstrating the gastrostomy tube in a large, air-filled stomach; contrast material had passed into the colon. No intraluminal gas is seen except in the stomach.

identified in the pyloric region and exposed through the surgical incision. Retrograde intussusception of the pylorus into the stomach was noted (Fig. 6).

Following an unsuccessful attempt to reduce the intussusception, Heineke-Mikulicz pyloroplasty was performed. The pylorus was found to be thickened, and a biopsy sample was sent for analysis. Histologic study revealed a thickened muscularis propria of 3 mm, normal neural plexuses, and early mucosal ischemic changes (Fig. 7).

The ventriculoperitoneal shunt was exteriorized on neurosurgical consult due to contamination of the peritoneal cavity during the procedure. A feeding tube was passed through the pyloroplasty to the jejunum.

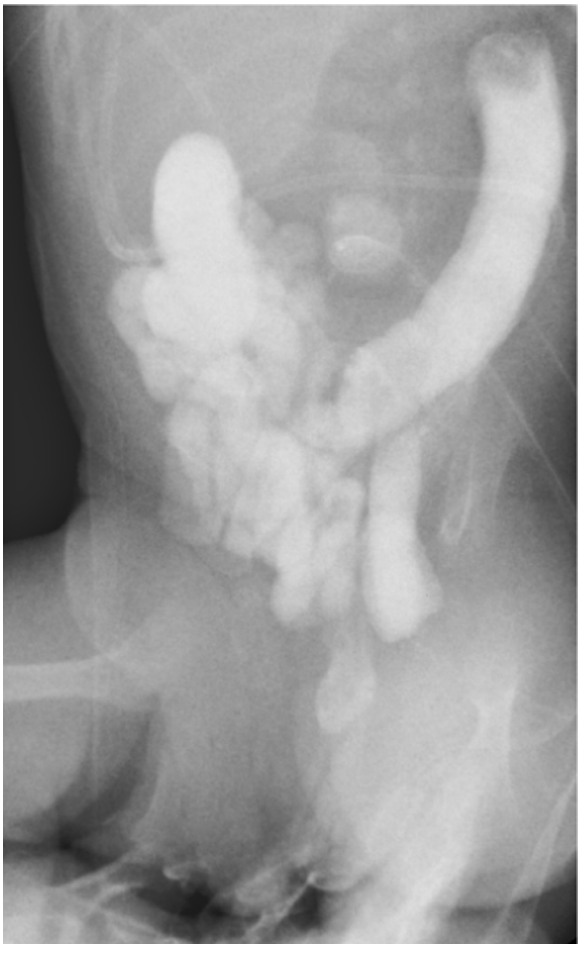

Fig. 3. Barium enema performed following the sonographic findings in Fig. 2.

1.4. Follow-up

The postoperative course was uneventful. Jejunal feeding was started on postoperative day 1 with good tolerance. One week after surgery, contrast study through the gastrostomy demonstrated good passage and no leakage in the pyloroplasty (Fig. 8). The feeding tube was removed, and full gastrostomy feeding was initiated. A few days later, the shunt was replaced and repositioned in the peritoneal cavity.

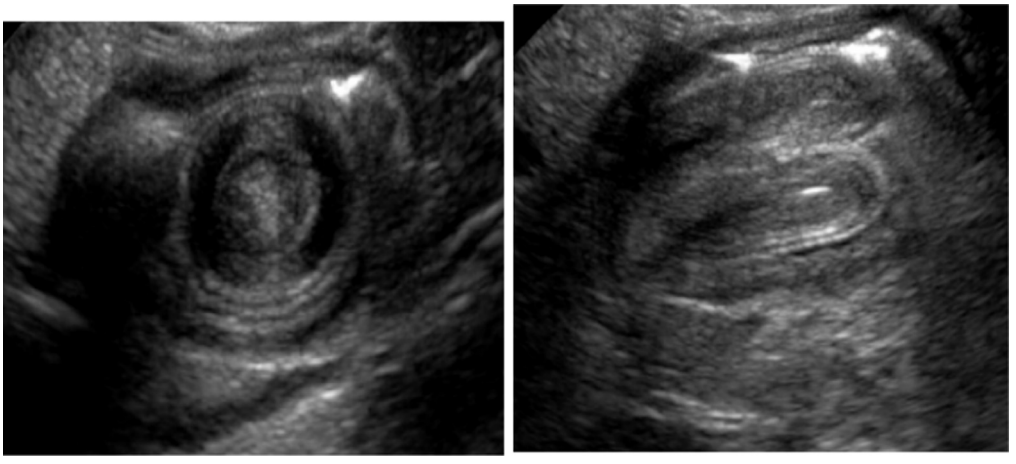

Fig. 2. Sonographic picture consistent with intussusception.

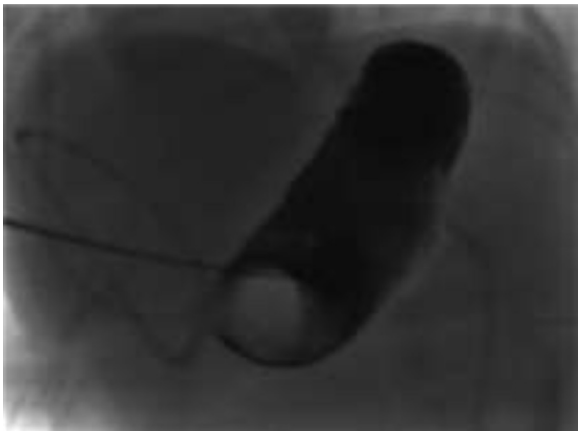

Fig. 4. Filling defect in the pre-pyloric area of the stomach.

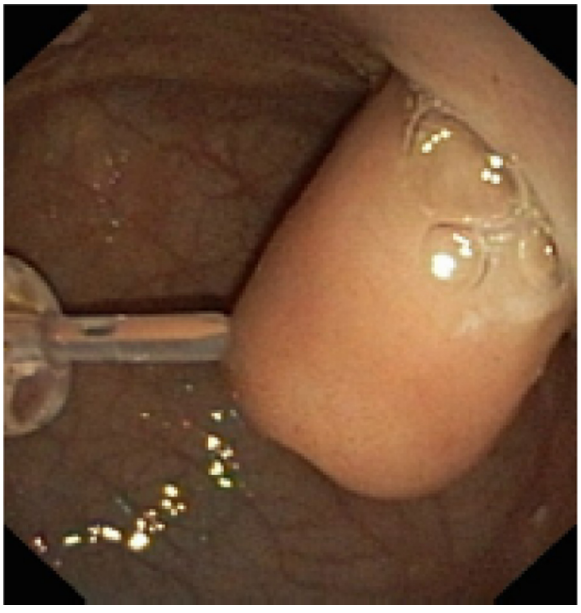

Fig. 5. Gastroscopy demonstrating a bulge in the pyloric area. Gastrostomy tube is present.

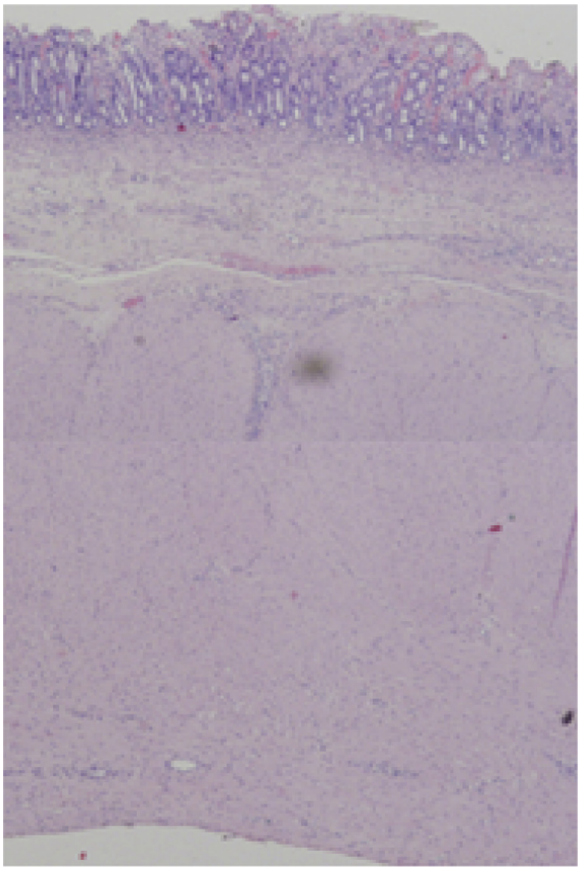

Fig. 7. Histological appearance.

2. Discussion

This case posed a clinical challenge because of the unusual diagnosis of retrograde intussusception of the pylorus.

Gastric outlet obstruction in infancy may result from congenital or acquired causes [1–5]. The most common cause is infantile hypertrophic pyloric stenosis, which has an incidence of 1.5–3 per

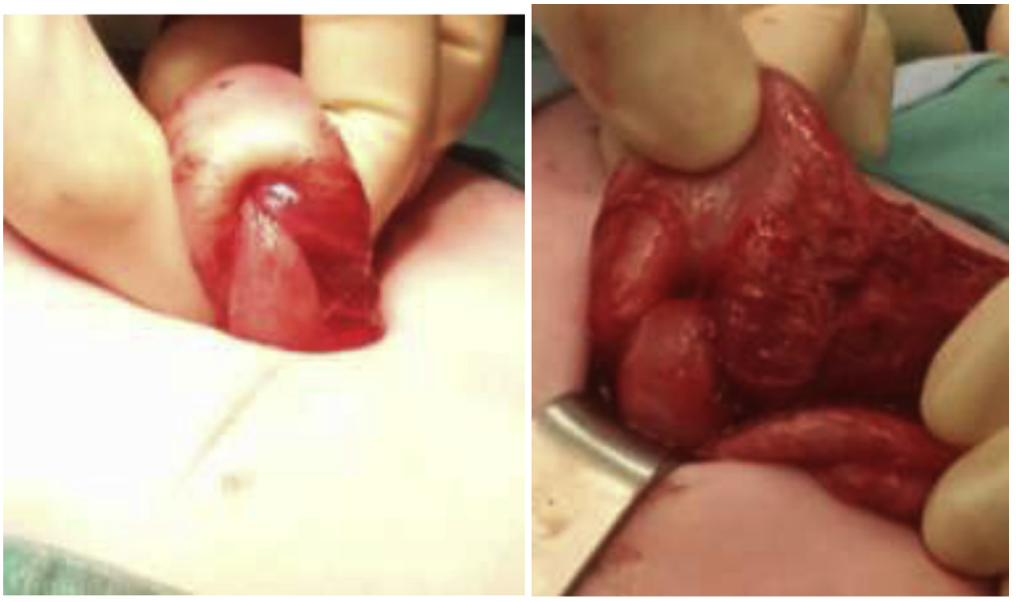

Fig. 6. Intraoperative findings.

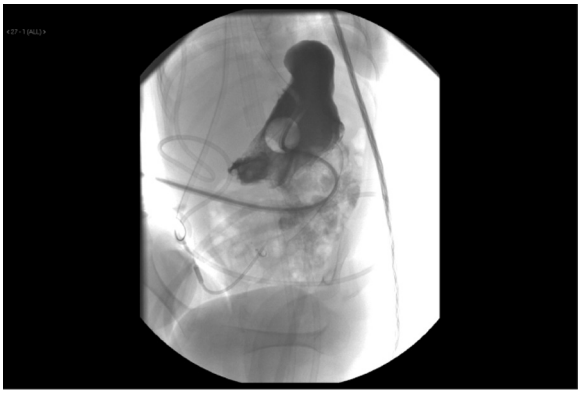

Fig. 8. Contrast study one week postoperatively.

1000 live births. Our extensive search of the English medical literature failed to yield any previous reports of retrograde intussusception of the pylorus in humans. The condition has been described in dogs [6–8], including puppies [9], in which it was either identified postmortem or successfully treated by surgery or endoscopy.

The operative and histologic findings in our case supported the diagnosis of hypertrophic pyloric stenosis. Therefore, the pyloric thickening was probably an important pathogenetic factor, preventing spontaneous reduction of the intussusception.

3. Conclusion

A possible cause of the retrograde intussusception itself could have been retrograde peristalsis associated with the vomiting induced by the intracranial hemorrhages. Another possibility is an iatrogenic mechanism involving post-pyloric migration and manipulation of the gastrostomy tube, although the tube was not documented in the post-pyloric position on X-ray examination.

Conflicts of interest

None.

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