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CASE REPORT

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# Pyloric duplication in a neonate: a rare entity

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**Abstract** Duplications of the alimentary tract are rare anomalies that have been reported to occur all along the gastrointestinal tract. Of the various alimentary tract duplications, pyloric duplications are extremely rare. We report the case of a 3-day-old neonate who was antenatally diagnosed as having a cystic mass in the abdomen and who presented with vomiting on the 2nd day of life. At operation, a duplication cyst of the pylorus was removed successfully and a pyloroantrectomy performed.

**Keywords** Duplication cyst · Pylorus · Neonate

## Introduction

Alimentary tract duplications are uncommon congenital malformations. Of these, only 3.8% are of gastric origin [1]. Only a third present in the neonatal period, with the most common presentation being an abdominal mass [2, 3]. Most of the patients present with symptoms of gastric outlet obstruction [4–6]. We report a neonate with pyloric duplication who was diagnosed antenatally as having a cystic mass in the abdomen.

## Case report

A 3-day-old girl was referred with a mass in the abdomen. She was born at term, weighed 2.5 kg, and was delivered by emergency cesarean section for fetal distress. The mother was 29 years old, para 3. There were no perinatal complications. An antenatal ultrasound done at 36 weeks of gestation showed a 3.6×3.7×4.2-cm intrabdominal cyst. Beginning the 2nd day of life, the infant had nonbilious vomiting, the frequency of which increased and necessitated hospital admission. On examination of her abdomen, an ill-defined, firm 5 cm×5 cm mass was noted in the right hypochondriac and epigastric regions extending to the right lumbar region. The mass had restricted mobility and could not be pushed down toward the pelvis. There was no organomegaly. Hematological and biochemical examinations were normal. Blood gas examination was suggestive of metabolic alkalosis. Blood and urine cultures were done, which showed no growth. Plain x-ray of the abdomen showed a soft tissue mass in the upper abdomen with no bowel distention. Chest and spine x-rays were normal. Ultrasound showed a thin-walled 5.4 cm×5.9 cm×4.7 cm simple cyst anterior and inferior to the body of the stomach. The rest of the viscera were normal on scan. Two-dimensional echosonography was also normal.

After adequate resuscitation, an exploratory laparotomy was undertaken, during which we found a 6 cm×5 cm cystic mass arising from the anterior and inferior surface of the pylorus, covering the anterior surface of the gastric antrum. The antrum "appeared" to be entering the cyst on the medial side, with the duodenum emerging on the right lateral side (Fig. 1). The cyst contained clear yellow fluid and had a common wall with the pylorus and a part of the gastric antrum. Excision of the cyst with sparing of the pyloric channel was attempted but was not possible. A pyloroantrectomy was performed, excising the cyst with a part of the gastric antrum. After a free flow of bile from the duodenal end was confirmed, it was anastomosed end-to-end with the gastric antrum over a transanastomotic Silastic tube placed in the jejunum. A nasogastric tube was placed in the stomach. The rest of the bowel and viscera were normal.

The child had an uneventful postoperative recovery. There were no organisms on culture of the cyst fluid. On

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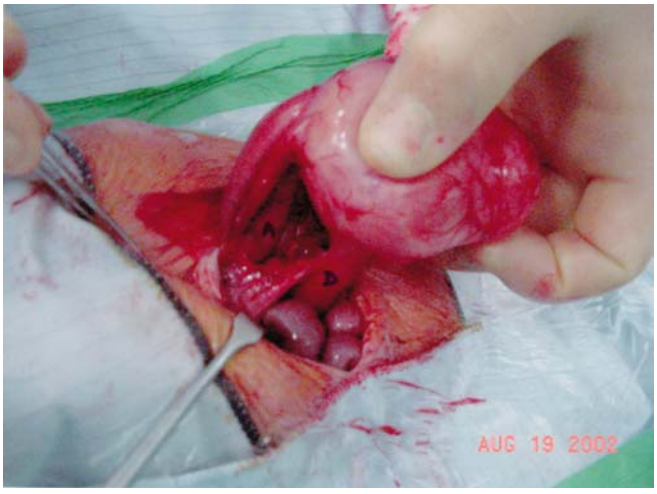


**Fig. 1** Pyloric duplication cyst with gastric antrum (*A*) and duodenum (*D*)

the 2nd postoperative day, feeding was begun through the transanastomotic tube and was gradually increased. Bottle feeds were started on the 8th postoperative day. Early evidence of dumping syndrome, such as irritability, pallor, sweating, abdominal distension, or watery diarrhea, were not observed even after bolus feeding was started. Histology of the cyst showed gastric and small bowel mucosa with congestion, erosions, and hemorrhage present on both sides, along with duplication of the muscular layers. No aberrant tissue was seen. Twenty months after the procedure, the child is feeding and growing normally with no signs of dumping syndrome.

## Discussion

Though these lesions were first reported by Calder in 1733, Ladd in 1937 introduced the term ''duplications'' and described the characteristic features of the lesion, which are the following [7]:

1. Presence of a well-developed coat of smooth muscle
2. Presence of epithelial lining resembling some part of the alimentary tract
3. Intimate attachment to at least one point of the alimentary tract

Enteric duplications can occur anywhere along the gastrointestinal tract. They are most commonly found in the ileum and rarely along the pyloric canal [8]. In 1957, Ramsay [9] reported the first case of a juxtapyloric cyst. Theories of embryologic origin, including incomplete twinning, phylogenetic reversal, persistent embryonic diverticula, dysvacuolation, tracheobronchial foregut duplication theory, and split notochord theory have been proposed to explain duplications.

Duplications of the stomach are frequently located along the greater curvature, and most have no communication with the stomach. The presenting symptoms may be nonbilious vomiting, a palpable mass, weight loss, or/and failure to thrive [3, 10]. Older children may also present with abdominal pain, hematemesis, or even perforation and peritonitis [11, 12]. Associated anomalies can occur in up to 50% of cases, including other alimentary tract duplications and vertebral defects. Pyloroduodenal duplication cysts are an extremely rare congenital anomaly that present with symptoms of gastric outlet obstruction and a palpable abdominal mass. Pancreatic heterotopias have also been known to occur with pyloric duplications [13].

Kremer [10] suggests that the possibility of gastric duplication should be kept in mind especially for female infants presenting with abdominal mass, pain, vomiting, anemia, and an x-ray showing an indentation along the greater curvature and depression of the transverse colon. Tihanski et al. [8] reported the first case of pyloric duplication cyst accurately diagnosed before surgery by an upper gastrointestinal barium study. Diagnostic studies such as ultrasound, upper gastrointestinal barium study, endoscopy, computed tomography, and even intravenous cholangiography with spiral computed tomography (IVC-SCT) [14, 15] are useful in the preoperative diagnosis, but a correct preoperative diagnosis is seldom possible because the symptoms are so varied and the entity so rare [8].

The surgical treatment involves total excision of the cyst. In the case of incomplete excision, procedures including stripping the residual mucosal lining, patching the raw area with omentum or colon [4, 16], or leaving the seromuscular gap [2] have been described. In the present case, it was not possible to dissect the common wall between the duplication and the gastric wall. As a result, the pyloric duplication with a wedge of the gastric antrum had to be excised, resulting in a limited pyloroantrectomy [17].

Though rare in infancy, dumping syndrome is a known complication following gastric surgery [18]. Despite pyloroantrectomy, our patient has shown no signs suggestive of dumping syndrome following bolus feeds, either in the postoperative period or at follow-up.

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