

## A prenatally diagnosed gastric duplication cyst connecting the pancreatic duct and paraspinal region

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### ABSTRACT

The gastrointestinal duplication of stomach is uncommon congenital anomalies of gastrointestinal tract. Herein a rare case of gastric duplication with newborn is being reported. A 14-day-old term female baby was admitted to our clinic with intermittent bilious vomiting and abdominal distention. In evaluation, abdominal cystic mass was detected in the left upper quadrant. The cyst which was attached to both the pancreatic duct and spinal cord was removed totally by laparotomy. A histologic examination showed gastric duplication cyst. The postoperative course was uneventful. There were no any complications at the 8-month follow-up period. This rare entity should be included in the differential diagnosis of cystic masses of the gastrointestinal tract and treated surgically by complete resection due to the risk of malignant transformation and other complications. This is the first case report that the gastric duplication cyst has a connection with both the pancreatic duct and spinal cord.

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### Introduction

Alimentary tract duplications comprise a wide range of mass lesions throughout the gastrointestinal tract. Gastric duplications (GD) are often cystic and asymptomatic, and can reach very large sizes. They can have a wide range clinical presentations due to mass effect and pressure on the surrounding structures ranging from abdominal pain to perforation [1]. Although there are cases of GDs connecting with the pancreatic duct in the literature, to the best of our knowledge a GD communicating both with pancreas and spinal cord have not been reported up to date.

Herein, we present a case of a newborn with a prenatally detected GD cyst.

### Case Presentation

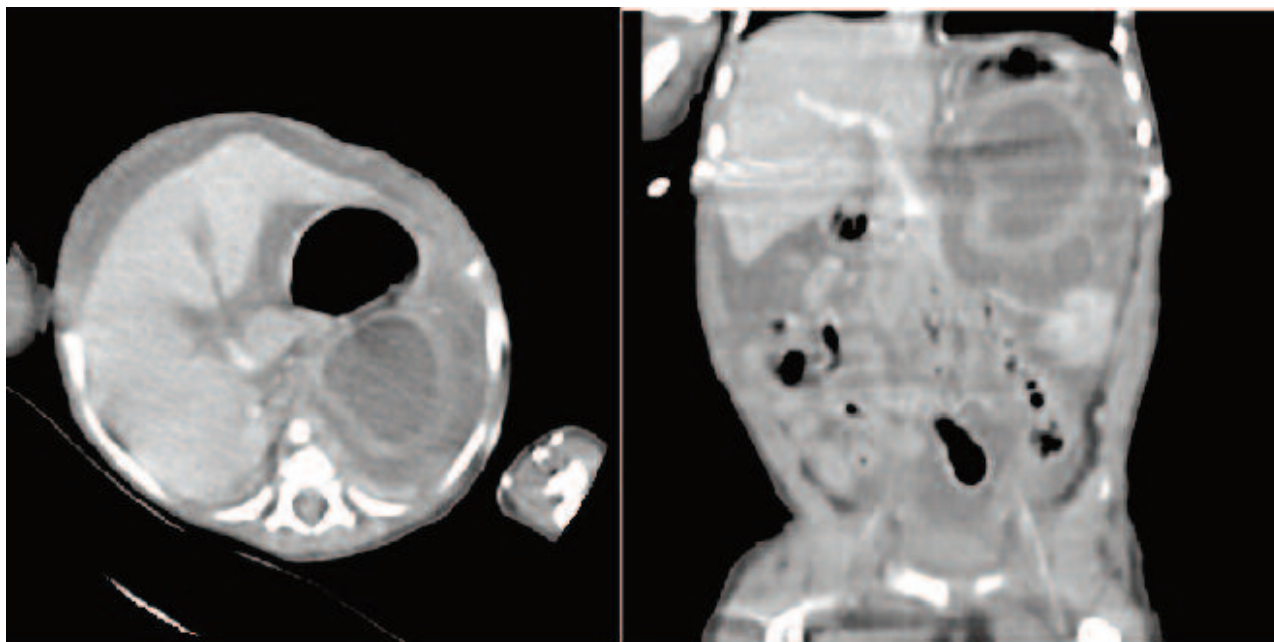
A 14-day-old term female infant presented to the emergency department with intermittent vomiting and abdominal distention. In her history, it was demonstrated that she was diagnosed with a homogenous cystic mass (3 × 4 × 4 cm) on the upper

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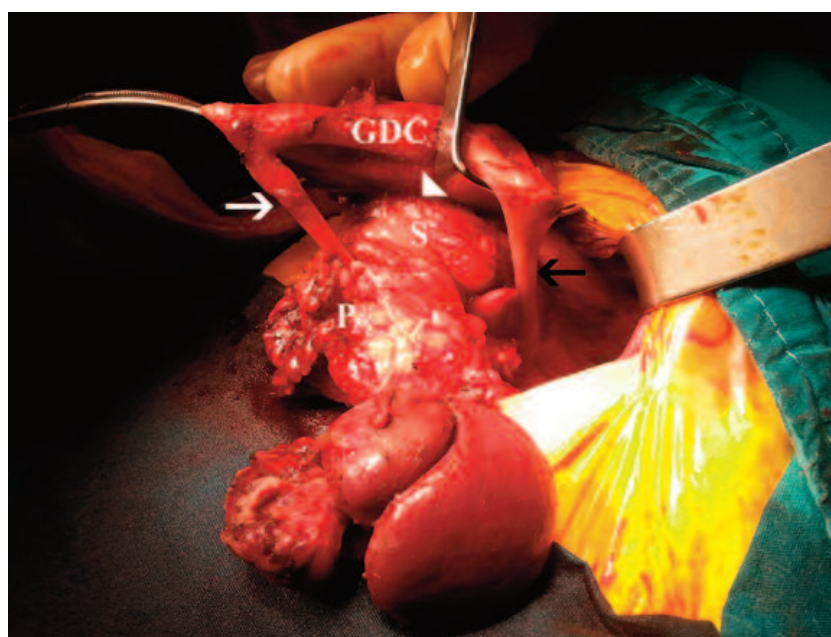
**Figure 1.** CT demonstrates an intraabdominal cyst on upper left quadrant.

left area in the abdomen. A general physical examination revealed a temperature of 38° C, pulse rate of 110 per minute and respiratory rate of 35 per minute. The general condition of the infant was poor and she was intubated and a nasogastric tube was inserted. The drainage fluid was bile stained with an amount of 50 mL per day. On abdominal examination, there was a marked abdominal distention without rebound tenderness and the bowel sounds were normo-active.

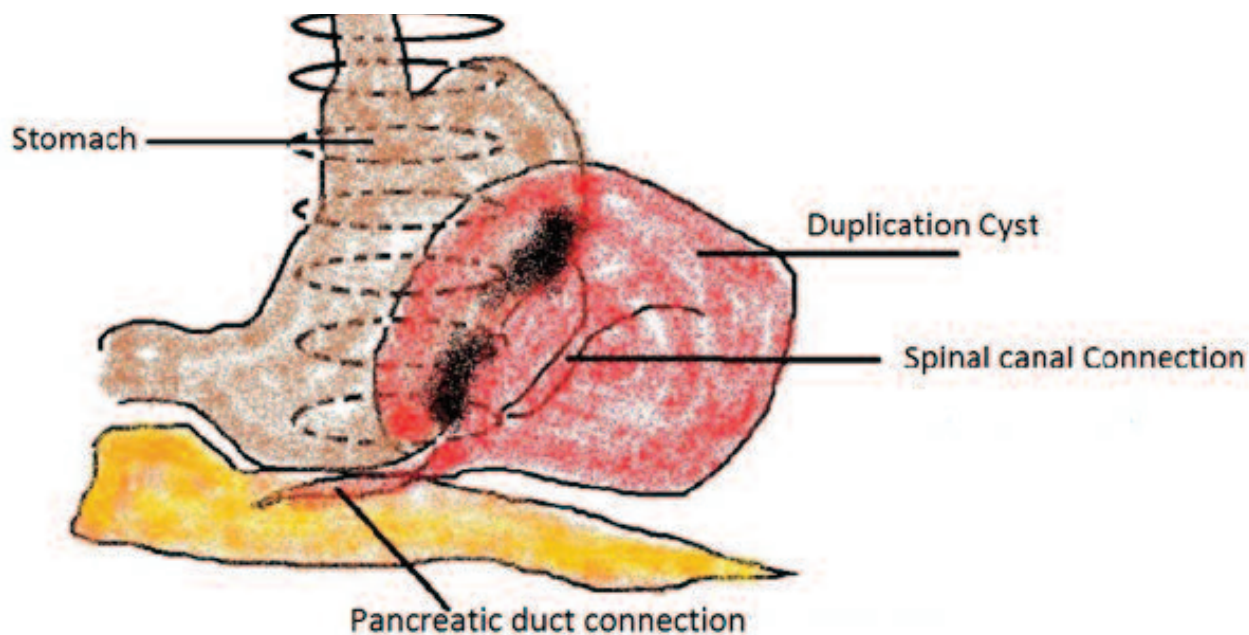
On follow-up, the plain abdominal radiographs

were normal and ultrasonography (US) of the abdomen demonstrated 7 × 8 × 6 cm homogenous cystic mass with millimetric echogenities arising from the upper pole of the left adrenal gland extending into the paraumbilical area lateralizing the left kidney and spleen.

The abdominal tomography (CT) showed a big encapsulated cystic lesion which was located inferior-posterior of great curvature of stomach (Figure 1). It was extending from the left upper suprarenal side to the para-umbilical area without a dilated stomach and



**Figure 2.** The intraoperative photograph shows the gastric duplication cyst (GDC) which has two connections with both the pancreas (P, white arrow) and the spinal cord (black arrow). Cyst also attached to the stomach (S, white triangle).



**Figure 3.** The illustrated image shows common wall with the stomach at two points and connections with pancreatic duct and spinal canal.

pancreatic ducts. An urgent exploratory laparotomy was planned after preparations.

On laparotomy, a cystic mass of about  $6 \times 10$  cm was noticed, and it was considered to be a GD cyst. The cyst was firmly adhered to the spleen and attached to the posterior wall of the stomach. The cyst was shown to share a common wall with the stomach at two points. GD cyst had also a connection with both the spinal canal and the pancreatic duct. The view of the operation and illustrated image are showed (Figures 2 and 3). About 100 ml of dark brown fluid was aspirated. Both connections were ligated and transected. The cyst was dissected from spleen's and stomach's adhesion and mucosectomy of the two points was performed.

Postoperative course was uneventful. She was discharged on postoperative fifth day. In the eight months of the follow - up period, the patient was healthy.

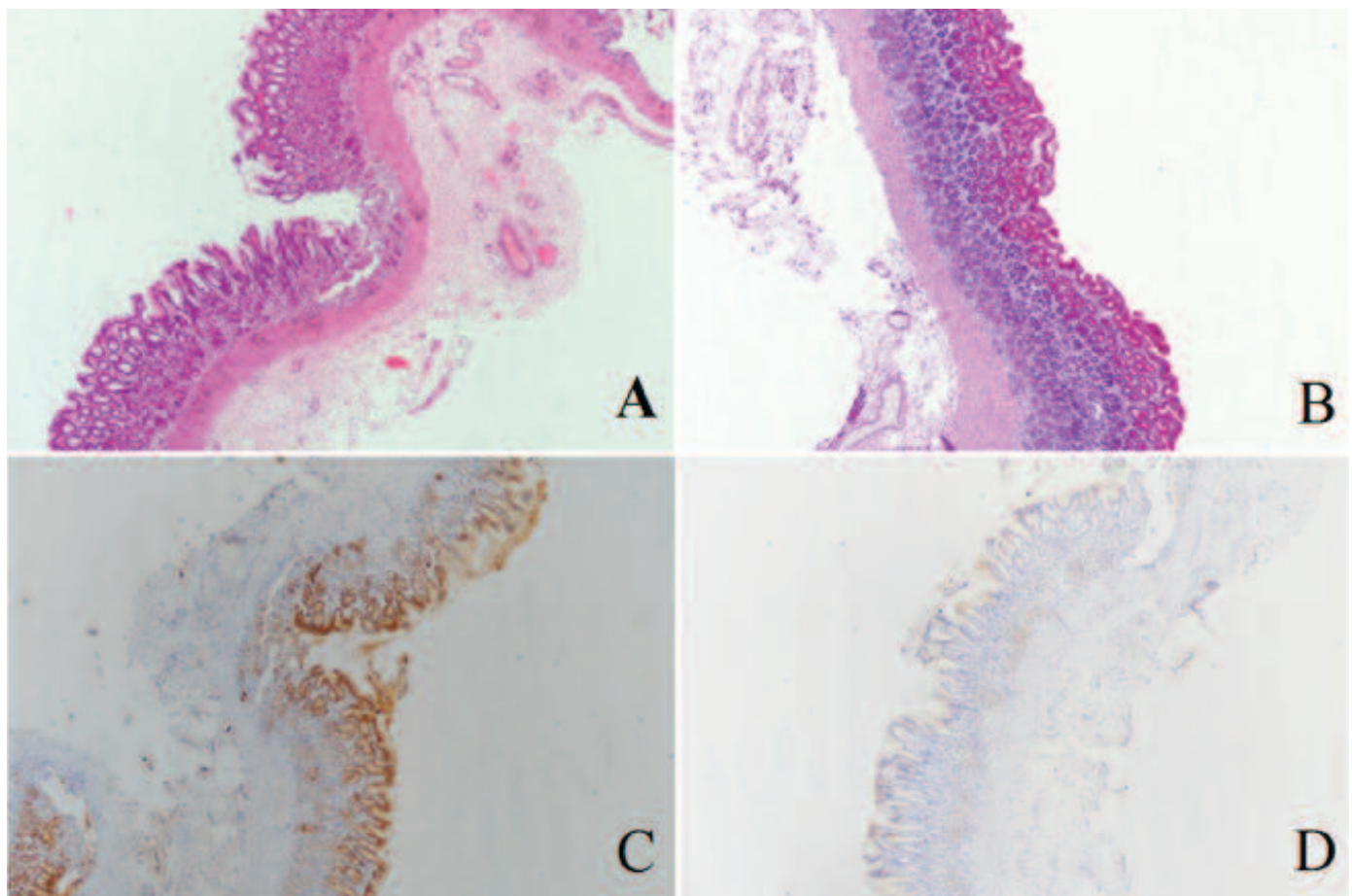
Histopathologically, on the examination by light microscopy, the sections stained with hematoxylin-eosin revealed the layers of gastric wall that is mucosa, sub mucosa, muscularispropria and serosa (Figure 4a). Periodic acid – Schiff-alien blue staining showed us that, the most of the mucosal cells were gastric cell and the some of them were cells containing apical intestinal mucus (Figure 4b). The most mucosal cells had positivity of CK7 immunohistochemical staining (Figure 4c), but some cells were positive with CK20 (Figure 4d).

## Discussion

The duplication cysts can be occurring along the whole alimentary tract [2]. Although the most involved tract is ileum (30-35%) the GD represent only 4 to 5 % of all duplications [3]. GD, most of which are cystic duplication (80%) and have not communicated with lumen of the stomach. The remainder structures are tubular duplication which associated with gastric lumen [4]. In general, patients typically present at neonatal period with symptoms of gastric outlet obstruction and palpable abdominal mass which may be seen on antenatal ultrasonography [1, 3]. In determining of GD cyst, US and CT are an effective diagnostic modality.

Our case with antenatal diagnosis of intra-abdominal cystic lesion presented with postnatal gastric outlet obstruction. Based on US and CT scan, we didn't make the preoperative diagnosis of a duplication cyst. Although it is difficult to diagnose GD cyst preoperatively, imaging studies have provided some evidence informative. It is stated that the characteristic “double wall sign,” delineating the acrogenic inner mucosa from the hypo echoic outer rim of muscle can be used to identify on US [5]. Additionally, a contrast-enhanced CT scan can also be demonstrated GD as a thick-walled cystic lesion with enhancement of the inner lining [4].

Many varied theories have been claimed regarding the origin of GD cyst, like recanalization defect of alimentary tract, adhesions between endoderm and



**Figure 4.** Histopathological appearance; (a) Gastric duplication wall comprised of all layers; mucosa, submucosa, muscularis propria and serosa (Hematoxylin and Eosin, X40), (b) Mucosa was predominantly of gastric type (pink colored) with some cells containing apical intestinal mucus (blue colored) (Periodic acid-Schiff-alcian blue staining, X40), (c) CK7 positive mucosal epithelium (CK7 immunohistochemical staining, X40) and (d) CK20 positive mucosal areas (CK20 immunohistochemical staining, X40).

neuroectoderm, theory of split notochord and abortive attempts of twinning [1-3]. Most commonly accepted theory is “split notochord syndrome” to explain the development of this congenital anomaly. In this theory, persistence of ectoendodermal connection that has developed at an early embryonic stage causes diverticular elongation of the gut and leads to the formation of duplication cysts [6]. In previously reported gastro duodenal duplications with communication to the pancreatic ducts, it has been proposed that the dorsal traction onto the portion of the foregut that gives rise to the dorsal pancreatic bud might represent the initial trigger for formation of the cyst [6]. Indeed, as previously reported, connections between GD cyst and pancreas well described. To our knowledge, the present case is the first case report of a connection with both the pancreas and spinal cord as well. It also is a valuable case to prove split notochord theory.

The management of GD is essentially surgical. In GD cyst, the treatment procedure is resection without

injury to the lumen of the stomach. No communicated GD cysts are classically treated by complete excision of cyst and resection of common wall between stomach and cyst with mucosal stripping. In the communicating GD cyst drainage and marsupialization of the cyst have been suggested. Gastrostomy or even a feeding jejunostomy may be helpful according to patient position [1, 4]. Furthermore, leaving the residual tissue of cyst wall is given potential risk for inflammatory complications and malignant transformation [2, 4, 7]. In presented case we performed totally cystic excision with mucosal stripping at two points in common wall.

In asymptomatic or untreated patients, such as infection, gastrointestinal bleeding, ulceration or perforation may be occur because of ectopic gastric mucosa, also may lead to pancreatitis and mimic a pancreatic pseudo cyst due to ectopic pancreatic tissue [4]. In addition, duplication cysts have the potential risk for neoplastic transformation especially adenocarcinomas which has been reported [4, 8, 9].

In histopathological evaluation, most of GD cyst contains gastric mucosa with smooth muscles. In addition, up to 10% gastric duplications may contain ectopic pancreatic tissue [2, 4, 7]. In our case cyst surface was covered with gastric mucosa, and smooth muscles in the wall.

## Conclusion

This rare entity should be included in the differential diagnosis of cystic masses of the gastrointestinal tract. US and CT facilitates the diagnosis. This case emphasizes that the GD cyst should be treated surgically by complete resection due to the risk of malignant transformation and other complications. According to the literature and with the results of the present report, this is the first report to demonstrate a communication between both the pancreas and spinal cord, and it would help to explain the development of GD cyst by the split notochord theory.

### *Informed consent*

Written informed consent was obtained from the patient for the publication of this case report.

### *Conflict of interest*

The authors declared that there are no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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