ANNALS OF GASTROENTEROLOGY 2000, 13(2):142-144

Case report

Complete gastric duplication herniated in the right hemithorax: imaging appearance

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SUMMARY

This is the first report of a complete gastric duplication cyst that herniated through the esophageal hiatus into the right hemithorax. A study of x-ray chest films, abdominal ultrasonography, computed tomography of the duplication cyst and a brief review of the recent literature are presented.

Key words: gastric duplication cyst, infant, imaging

INTRODUCTION

Duplication cyst (DC) of the alimentary system is an unusual congenital abnormality that may be found anywhere between the mouth and the anus.[1] Commonly, it is encountered in the ileum and esophagus. Gastric duplication cyst represents only the 4-5% of all gastrointestinal duplications.[2] It may vary widely in size and shape.[3] Complete gastric duplication is rare and few cases have been reported.

Histologically, gastric DC has a well developed smooth muscle wall and is lined by gastric mucosa. The etiology of the duplication cyst of the alimentary tract has not been well established. Etiologic theories have postulated that the duplication cyst is a consequence of: a) persistence of fetal gut diverticula,[3] b) recanalization and fusion of longitudinal folds,[3,4] c) intrauterine vascular occlusion.[4,5]

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CASE REPORT

A 5-month-old male infant was referred with dyspnea and cough. On physical examination, a mobile mass in the left upper quadrant of the abdomen was palpable. Routine blood tests were normal.

The anteroposterior and the lateral chest x-ray film demonstrated the presence of a huge soft-tissue mass in the middle and posterior lower mediastinum (Fig. 1).

An abdominal sonogram revealed a large cystic mass located in the left upper abdomen adjacent to the great-

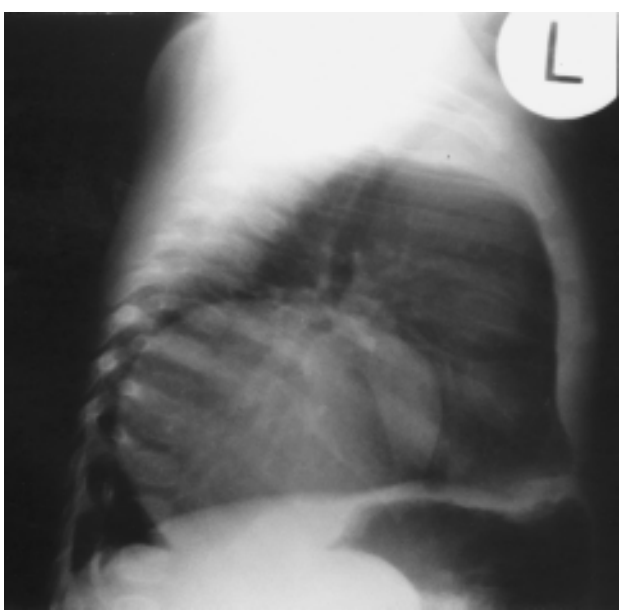

Figure 1. Lateral chest radiograph. A large posterior and medial mediastinal mass is shown obliterating the right diaphragm and ascending to the level of the sixth thoracic vertebra

er curvature of the stomach displacing it anteriorly. A double layer wall was partially identified consisting of an inner echogenic ring and an outer hypoechogenic. The mass appeared to ascend into the left posterior mediastinum through the diaphragm (Fig. 2). An abdominal enhanced CT scan confirmed the presence of the cystic abdominal mass along the greater curvature of the stomach extending anteriorly to the pancreas (Fig. 3). The cystic mass ascended into the left hemithorax through the hiatus and extended in the right hemithorax, to the level of the sixth thoracic vertebra. An incomplete diaphragm in the mediastinal part of the mass was identified amplifying the suspicion of an inflammatory process. Although scintigraphy by Tc-99m pertenchnetate is the method of choice for the diagnosis of alimentary duplication, it was not performed, due to the fact that it was not available. Barium enema was also not performed as the preoperative diagnosis was that of the esophageal and gastric duplication cyst. In addition, we considered that it was preferable not to expose the infant once again to ionizing radiation, as the operation was mandatory.

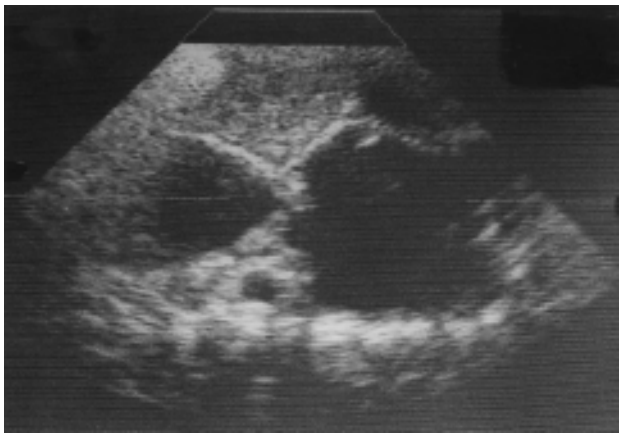

Figure 2. Sonogram, longitudinal image. A large abdominal cystic mass is demonstrated posteriorly to the left hepatic lobe. The mass communicates with another cystic intrathoracic lesion through the diaphragm which appeared as an echogenic line

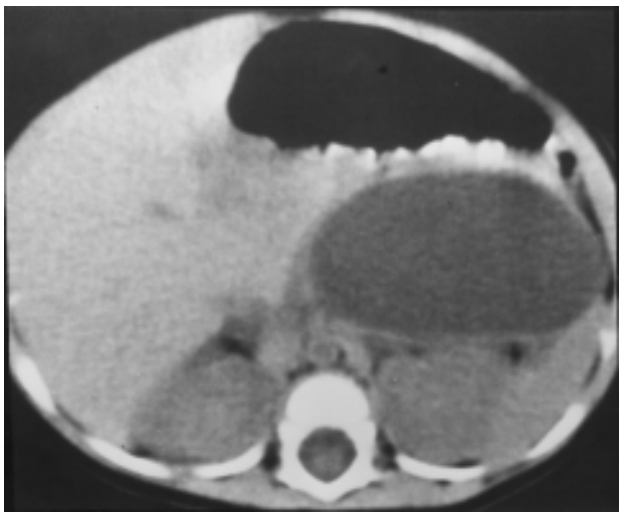

Figure 3. Computed tomography after intravenous administration of contrast medium. An abdominal cystic mass is demonstrated adjacent to the greater curvature of the stomach

Through a thoracoabdominal incision, a huge cystic mass loosely attached to the greater curvature of the stomach was completely excised. The mass did not share a common muscle layer with the stomach and did not communicate with it. Histologic examination revealed the presence of a viscous yellow fluid within the cyst. The cyst was lined with gastric mucosa and had a smooth muscle wall. Scattered superficial ulcerations, especially in the intrathoracic part of the cystic mass, were noted.

Exploration revealed no other duplication cysts. The patient had an uneventful recovery and follow-up five weeks later revealed no abnormality.

DISCUSSION

Gastric duplication cyst is a rare abnormality of the alimentary system, usually asymptomatic discovered by an imaging method that is required for unrelated reasons. When symptomatic, it is usually detected during infancy, especially when it involves the thorax and the upper gastrointestinal tract.[1] Symptoms and signs are often non specific and include vomiting, nausea and a mobile palpable epigastric mass. Abdominal pain is also another relatively common symptom.

Gastric duplication is often associated with other duplications or atresias of the alimentary tract or other congenital anomalies, such as vertebral malformations,[3] aberrant pancreas[6] and pancreatic duct.[6,7] Gastric duplication cysts may infrequently be multiple[8,9] or may coexist with other duplications arising from the alimentary system.[10]

Occasionally, its clinical presentation is secondary to a complication, such as partial gastric obstruction,[11] perforation,[9] due to gastric ulceration or fistulas formation with the pancreas.[12] Preoperative diagnosis is not easy when the cyst is small.[13]

Gastric DC usually arises from the greater curvature.[2] It may be tubular or saccular in its shape.[1,3] Tubular du-

plication usually communicates with the gastric lumen while saccular does not.[3] A common blood supply and a common muscular coat with the alimentary tract is found in most cases.[13] Occasionally, it may be found in the submucosal or subserosal layer.[3] Large gastric DCs are usually associated with esophageal duplications.[10]

Gastric duplication cyst, as any duplication cyst of the alimentary system, can occur as a focal lesion or involve the entire stomach.[14] A duplication cyst of the entire stomach is developed only in exceptional cases.[15] The last 17 years only one case of complete gastric duplication cyst in association with complete esophageal duplication cyst has been reported.[14]

In our case, a duplication cyst of the entire stomach, extending into the mediastinum, caused respiratory distress in a 5-month-old infant. Although the sonographic appearance of the duplication cyst has been considered characteristic, due to the visualization of its double wall consisting of an echogenic ring (mucosa) and an outer hypoechogenic (muscle layer),[13,16,17] our diagnosis could not be based on this sonographic appearance. The double wall of the cyst appeared only in some areas probably due to the large size of the cyst. A communication between the cystic component of the mediastinum and the cystic part of the mass in the abdomen was revealed through the diaphragm which was visualized as an echogenic line.

A computed tomography examination was required, even though it has been postulated that it may mislead in the case of DC, especially when hemorrhage or infection occurs.[17] The main reason of the performance of CT was to detect the accurate relation of the cyst with the mediastinal structures (especially vessels).

The treatment of a symptomatic non communicating DC is total surgical excision or excision of the shared wall between the stomach and the duplication cyst, converting the non communicating cyst in a communicating one.

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