CASE REPORTS

# ANEMIA AND MELENA IN AN INFANT WITH RETROCARDIAC POSTERIOR MEDIASTINAL MASS - CAN IT BE ESOPHAGO-GASTRIC DUPLICATION CYST?

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**Abstract :** Foregut duplication cysts are rare entities seen as mediastinal masses. Gastrointestinal (GI) duplication cysts can occur anywhere along the whole GI tract. We present a 6 month old male infant who presented with severe pallor and Melena. Incidentally done chest X-ray showed an oval retrocardiac posterior mediastinal opacity which was better delineated by CECT of thorax and upper abdomen. Thoracoscopy and mini laparotomy was performed and the mass was diagnosed to be an esophago-gastric intramural duplication cyst with ulceration. Patient recovered well following surgery.

**Keywords:** Anemia, Esophageal duplication cyst, Foregut duplication cyst, Mediastinal mass

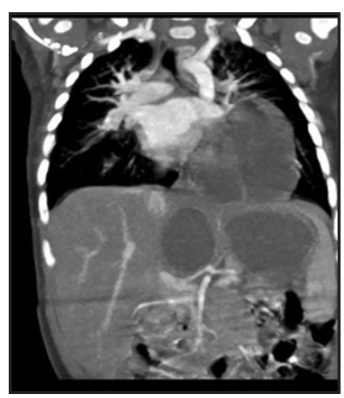
## Introduction

Gastrointestinal (GI) duplication cysts are rare congenital anomalies. They can occur at anywhere along the whole gastrointestinal tract with ileum (over 60%) being the commonest site. (1) The cysts usually have one outer muscular layer and are lined by different kinds of gastrointestinal mucosa at the inner lining. (1) These may be singular or multiple, may or may not communicate with the lumen and may be cystic (80%) or tubular (20%). (1) Both esophageal and gastric duplication cysts are very rare in literature. (2) We present a 6 month old infant who presented to us for evaluation of anemia and was diagnosed as a case of complicated esophago-gastric duplication cyst.

## Case Report

A 6 month old male presented with pallor for one and half months. There was no history of fever or bleeding from any site. On examination, child was pale. Systemic examination was normal. Hemogram showed hemoglobin of 2.7gm% with mean corpuscular volume (MCV) 84.5 fL, mean corpuscular haemoglobin concentration (MCHC) 31%, reticulocyte count 1.6% and peripheral blood smear showed normocytic normochromic anemia. Total leukocyte count was 9,200/cumm with 78% lymphocytic preponderance, platelet count was 3,64,00 cells/cumm. Coagulation profile (prothrombin time 11.3 sec, activated partial thromboplastin time 22.3 sec), high performance liquid chromatography (HPLC) (HbF-21.8%, HbA-77.1%) were normal. Renal and liver functions were normal. We noticed blackish discoloration of the diaper by the stool. The mother, on enquiry, stated that she noticed this colour change of stool but took it as normal. Stool analysis showed 20-25 red blood cells/ high power field. Urine examination was normal. A chest X-ray done incidentally showed an oval shaped retrocardiac opacity over left lower zone. Lateral view of the chest confirmed its presence in posterior mediastinum. Possibility of neurogenic tumour involving gastrointestinal tract was considered. Contrast enhanced computed tomography (CECT) scan of thorax and upper abdomen showed multiloculated thoraco-abdominal cystic lesion compressing left lung and extending from posterior mediastinum to suprapancreatic space with septations measuring 90mm x 38 mm x 46mm in superoinferior, anteroposterior and mediolateral dimensions respectively (Figure 1). No anomalous vascular communication was found. A barium swallow and meal did not reveal any communication between the lesion and gastrointestinal tract. The child was transfused with four aliquots of packed red blood cells to increase the haemoglobin level to 10 gm%. Thoracoscopic surgical exploration with mini-laparotomy revealed a dumb-bell shaped intramural esophago-gastric cyst with ulceration over mucosal aspect. Histopathological study revealed esophago-gastric tissue with mild bronchogenic differentiation at cranial end. Patient recovered well and was discharged without any complication.

**Figure 1: Coronal Computed Tomography scan of thorax and upper abdomen showing thoraco—abdominal multiloculated intramural esophago-gastric duplication cyst extending through esophageal hiatus**



## Discussion

Foregut duplication cysts are extremely rare and constitute around 6-15% of all mediastinal masses in infants and children. (3) There are three types of foregut cysts: bronchogenic, esophageal and enteric, among which esophageal duplication cysts are very rare. (4) The prevalence of esophageal duplication cysts is 0.01%. (2) Right posterior inferior mediastinum is the commonest location and 2/3rd of them involve lower end of esophagus. (2) Esophageal duplication cysts are lined by alimentary epithelium, have double layer of smooth muscles and may be para-oesophageal or intramural in location. (4) On the other hand, gastric duplication cysts make up 4-9% of all intestinal duplication cysts. (5) Usually these are single and without luminal communication. Histologically, gastric duplication cysts have mucosa, connective tissue layer, muscular layer and fibrous capsular layer. (6) They may contain ciliated cells, histiocytes, crystals and protein debris. (6) Around 95% of gastric cysts arise from greater curvature but they can also be found near gastroesophageal junction, cardia and anterior or posterior walls of fundus. (6,7) Our patient had esophago-gastric duplication cyst involving left posterior mediastinum and the greater curvature of

stomach. It had mild bronchogenic differentiation at the cranial end, apart from that was consistent with usual esophageal and gastric histopathological features.

During in-utero life, at 5-8 weeks of gestation, foregut duplication cysts arise as a result of abnormal dorsal (esophageal) or ventral (bronchogenic) budding from the primitive foregut although the exact mechanism still remains an enigma. (7) That is the reason why these are found near the midline structures. Gastrointestinal duplication cysts commonly present before 2 years of age. They present with features like abdominal pain (most common), vomiting, weight loss, respiratory problems, failure to thrive, gastric outlet obstruction or ulcerated mass. (6,7) Gastrointestinal hemorrhage has been reported in rare cases of esophageal cysts having ectopic gastric mucosa. Another rare presentation is malignant transformation of the cysts. (3) Enteric cysts often have vertebral anomalies as a part of split notochord syndrome which are never seen with esophageal duplication cysts. (4) Complications of foregut cysts include infection (most common), peptic ulceration, cardiac arrhythmia, pulmonary artery stenosis and superior vena cava obstruction. (4)

Imaging studies (antenatal screening, conventional or endoscopic ultrasonogram, MRI, CT scans) often find out the cyst but definitive diagnosis is by histopathology examination of biopsy samples. (8,9) Treatment of symptomatic cyst is surgical exploration and excision after antibiotic therapy. For mediastinal cysts, thoracoscopic approach is preferred. Even incidental asymptomatic cysts must be treated due to fear of complications like infection and malignant transformation. (4)

**Conclusion**

In an infant with melena, possibility of surgical causes of gastrointestinal bleeding should be considered.

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**Conflict of Interest:** None

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