Gastric duplication cyst

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astric duplication (GD) is uncommon amongst the gastrointestinal tract (GIT) duplication anomalies. None of the theories of their origin can explain the mechanism of these malformations. Typically, they are non-communicating spherical cystic masses along the greater curvature or the posterior aspect of the stomach. GD may present diagnostic as well as therapeutic challenges. As these lesions can be unexpectedly encountered intra-operatively, appropriate surgical management requires that the surgeon be familiar with the anatomy and clinical characteristics of the malformation. In this report, we describe a case of GD who presented in her late childhood with a previously healthy background and who had a good outcome following surgical excision. This is the first such case reported from the Medina region.

Case

A 7-year-old girl was admitted with upper abdominal pain, non-bilious vomiting and an epigastric mass of 5 days duration without any relevant past medical history. She had just started her first year at school, living with her parents in the desert. There was no history of pica or filthy eating habits. The girl was found mild to moderately dehydrated with a visible, globular, non-tender epigastric mass with restricted mobility. Abdominal ultrasonography revealed a cystic mass of 6x4 cm arising from the pylorus of the stomach. She had plain abdominal x-ray, CT and upper gastrointestinal contrast study, all of which pointed to a diagnosis of gastric duplication cyst. During the course of hospital investigation, the mass suddenly disappeared on the 3rd day of admission without any acute abdominal symptoms or signs other than the development of mild tenderness in the epigastrium. At this point, the patient had an esophago-gastroduodenoscopy, which confirmed a residual submucosal lesion at the antrum consistent with duplication. The girl then had a laparotomy, which revealed a cyst originating from the greater curvature at the pyloro-antral area and encircling almost all of the circumference through both the anterior and posterior surfaces. Billroth II gastrectomy with retrocolic Roux-en-Y gastrojejunostomy was done in a bowel loop 40 cm from the duodeno-jejunal flexure. She was discharged from the hospital on the 12th post-operative day and was being followed up regularly in out-patient clinic for 8 months after surgery. She was gaining weight without any relevant symptoms. Histopathological examination of the excised cyst was consistent with GD cyst with areas of ulceration only.

A plain radiograph (Figure 1) showed a round mass surrounded by the air contained in the stomach. The pneuFrom the Department of Pediatric Surgery & Radiology, Maternity & Children's Hospital, Madina Munawara, Saudi Arabia.

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matization of the distal digestive loops appeared normal. A barium meal, 10 minutes (Figure 2) and 1 hour and 30 minutes (Figure 3) after contrast ingestion confirmed the outlet obstruction of the stomach by the previously described mass on plain x-ray. Ultrasound examination (Figure 4) showed the presence of a large round cystic mass with posterior enhancement, adjacent to the lower portion of the greater curvature of the stomach. The solid intra-abdominal organs were normal. Certain ultrasound signs can be helpful to reach a diagnosis and particular importance should be given to the cyst wall. In this case, the wall of the lesion showed 3 layers with "muscular ring sign", which corresponds exactly to a digestive wall.

Discussion

Duplication of the GIT is a rare congenital anomaly seen in 1 in every 4500 autopsies.1 Calder in 17332 first reported this fascinating lesion. Ladd in 1937³ introduced the term "duplication of the alimentary tract" and Gross in 1953⁴ defined their clinical and pathological features. They may occur anywhere in the gut from tongue to anus, attached to the mesenteric border of the bowel, sharing a common blood supply, a smooth muscle coat and GIT epithelia. Ectopic gastric or pancreatic tissues may be found in the lesion. Mostly, they are saccular, while the rest may be tubular or diverticular. The etiology of these malformations has not been well established. Several theories have been proposed, including persistence of foetal gut diverticula, defects in the recanalization of the solid stage of the primitive gut,⁵ partial twining and the Split-Notochard Theory (SNT). Although SNT is the most acceptable one,6 it does not explain duplications not associated with spinal deformities.7



Figure 1.(Plain radiograph) Showing a round mass, partially surrounded by air contained in the stomach with normal pneumatization of digestive loops located below.



Figure 2. (Barium meal) 10 minutes after contrast ingestion showing the obstruction of the gastric outlet by the lesion.





Figure 3. (Barium meal) 1 hour and 30 min after contrast ingestion showing same appearance with no passage of contrast in the duodenum.

Figure 4. (Ultrasound) Longitudinal section of the lesion showing a round well-defined cystic mass with posterior enhancement. The wall of the lesion is thick and shows 3 layers with "muscular rim sign".



Figure 5. Enhanced Abdominal CT Scan: Showing a rounded well-defined intraperitoneal cystic mass with thick wall,adjacent to the antropyloric region, displacing the stomach to the left & superior mesenteric vessels to the right.

GIT duplication is most common in the ileum near the ileo-caecal valve and uncommon in the stomach.^{8,9} There are only 38 cases of GD in the English language literature up to 1961¹⁰ but in recent years, more cases have been reported. In an extensive review, a 9% incidence for GD has been recorded.¹¹ Three cases of complete duplication extending from the oesophagus to the duodenum have been reported.¹²

GD is usually a non-communicating cyst located in the greater curvature and occasionally in the pyloric end producing symptoms of gastric outlet obstruction, as in this case. Symptoms may vary according to the site and size of the cyst. Manifestations may range from a non-obstructing lesion to an obstructing mass, even mimicking infantile hypertrophied pyloric stenosis.¹³⁻¹⁵

Cysts in other parts of the stomach may present as an abdominal mass. A complicating cyst may present with peptic ulceration, unexplained GIT haemorrhage,¹⁶⁻¹⁸ perforation and peritonitis, which could be a misleading sequelae. Cases of GD communicating with the pancreas have been reported as a cause of recurrent pancreatitis.¹⁹ Malignant changes in the gastric mucosa have been mentioned.²⁰

Duplications are more prevalent in males, one-third are diagnosed during the neonatal period and two-thirds during the first year of life ^{21,22} and 85% cases are detected by the age of two years.¹⁶ Cases have been reported in early childhood to adulthood.^{4,16} More than one duplication in one patient in some series has also been reported.²¹

A small cyst may not be detectable on clinical evaluation while ultrasonography(USG)²³⁻²⁶ will show a hypoechoic cystic mass with a mucosal lining and a muscular ring (muscular ring sign) to establish the diagnosis, as in our case. Although USG is the most useful diagnostic modality, an upper GIT contrast study, CT, and MRI, may be helpful. CT will show the relationship to the surrounding structures and the organ of origin. Nowadays, antenatal diagnosis is possible upon detection of a cyst with peristaltic activity in the right upper quadrant of the foetal abdomen. Another dilemma in diagnosing GD may happen secondary to collapse of the cyst due to communication with the GIT, which may open or close spontaneously²⁶ as was observed in our patient during her preoperative course of management.

Management of GD is surgical excision. The options may be dictated by the site and size of the anomaly, ranging from partial gastrectomy (as inour case) to stripping of the mucosa of the cyst from the stomach wall and then patching the raw area with transverse colon if the defect is large,²⁷ or leaving it as a seromuscular defect as in the case of Ramstedt's pyloromyotomy.²⁸ In our case, because of the large size and pyloroantral location with involvement of most of the circumference of the involved part, we performed a Billroth 2 partial gastrectomy with a 40 cm Roux-en-Y jejunal loop. Nowadays, laparoscopic surgical treatment is also a viable option in selected cases.

In children with cystic intra-abdominal mass, GIT duplication should be one of the differential diagnoses. Sudden collapse of the cyst due to communication with the GIT may be encountered during investigation and this should not delay the confirmation of the diagnosis. Awareness of this may reduce suffering and avoid any delay in definitive surgical treatment. During ultrasonographic evaluation, emphasis should be given to analyzing the wall of the cyst, which we consider an important step towards the diagnosis. Once GD is diagnosed, appropriate surgical measures should be undertaken to avoid complications. Anatomicopathologic analysis of the resected segment allows confirmation of the diagnosis.

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