Case Report

Asymptomatic Perforated Gastric Duplication Clogged by Omentum, Anorectal Malformation and Agenesis of the Corpus Callosum: A Rare Combination

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Abstract

Gastric duplication cysts are uncommon congenital anomaly and its association with other malformations is rarely reported. Many theories exist for the development of these lesions. This case report describes coincidental detection of perforated gastric duplication clogged by the omentum associated with anorectal malformation and agenesis of the corpus callosum.

Keywords: Anorectal, congenital, duplication, gastric

Introduction

Digestive tract duplications represent 0.1%–0.3% of malformations of the child. More rarely, gastric duplication represents 4%–9% of these duplications.[1-4] Its etiopathogenesis remains unclear. Gastric duplication is often isolated. Only few published cases reported associated malformations. This case report describes a unique combination and unusual circumstance of discovery of a perforated gastric duplication.

Case Report

A 3-month-old female infant born in term, without antenatal diagnosis of congenital malformation and with vaginal delivery, was followed from birth for vulvar anus.

The clinical examination showed hypo-reactive baby, without dysmorphic facies, no limb anomalies, normal auscultation with soft depressible abdomen and permeable vulvar anus.

Through an assessment of associated malformations, abdominal ultrasound revealed a cystic mass, in the left upper quadrant, evoking a digestive duplication depending on small bowel, 4-cm long axis [Figure 1]. Cardiac and renal ultrasounds

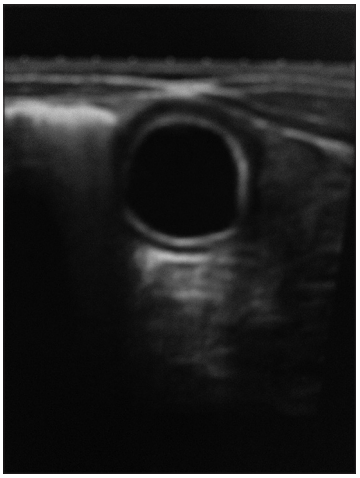

Figure 1: Abdominal ultrasound revealed a cystic mass suggestive of digestive tract duplication

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Received: 18-01-2018 **Revised:** 26-02-2018 **Accepted:** 13-05-2018 **Available Online:** 16-09-2020

Access this article online

Quick Response Code:

Website: www.afrjpaedsurg.org

DOI: 10.4103/ajps.AJPS_4_18

How to cite this article: Belhassen S, Ammar S, Zouaoui A, Abdellatif N,. Asymptomatic perforated gastric duplication clogged by omentum, anorectal malformation and agenesis of the corpus callosum: A rare combination. Afr J Paediatr Surg 2019;16:46-8.

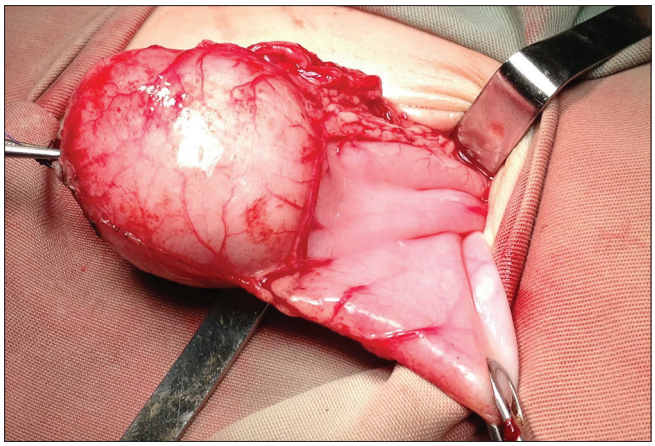

Figure 2: Spherical non-communicating gastric duplication located in the greater curvature of the stomach with a small perforation clogged by a flange of omentum

were normal, but a slight hydrocephalus was noted. Cerebral magnetic resonance imaging showed agenesis of the corpus callosum with hydrocephalus.

We decide to treat digestive tract duplication. Intraoperatively, it was a spherical non-communicating gastric duplication located in the great curvature of the stomach with a small perforation clogged by a flange of omentum [Figure 2]. The mass was completely excised from the greater curvature and a flange of the gastric muscular was sutured with 6/0. Post-prandial discomfort syndrome (PDS) histological examination confirmed a gastric duplication with fundic mucosa.

The post-operative course was uneventful with refeeding at day 3 postoperatively. Mean follow-up was 1 year. The patient was operated 3 months later for the vulvar anus with a good post-operative result. However, she still has neurological problems due to the callosum agenesis.

Discussion

Gastric duplication can be tubular or cystic.[2] The cystic type has no communication with gastric lumen.[2] Gastric duplications are usually diagnosed in a young age due to their mass effect.[4] A wide range of symptoms and signs have been reported and vary from asymptomatic to complicated or non-specific presentations. The most frequents are vomiting, abdominal pain, abdominal distension and weight loss. In our case, this gastric cyst was incidentally diagnosed during an assessment of associated malformations with a low anorectal malformation.

Prenatal diagnosis is difficult posing the problem of differential diagnosis with any intra-abdominal cystic such as ovarian cyst, renal cyst, choledochal cyst and mesenteric cyst.[2] After birth, multiple imaging modalities exist to evaluate duplication cysts including plain radiographs, upper gastrointestinal contrast studies, ultrasound, computed tomography and technetium-99m. The sensitivity of ultrasound to identify bowel wall signals facilitates the diagnosis and is considered the imaging modality of choice for evaluation of duplication cysts.[5]

Multiple theories have arisen to explain the occurrence of enteric duplications, but no single theory accounts for all the known variants.

The embryological origin of these anomalies is still controversial.[4,6,7] Many theories exist for the development of these lesions including a persistent embryological diverticulum, aberrant recanalization of the alimentary tract. The theory of split notochord is postulated to describe many associated anomalies involving the spine, gastrointestinal tract and skin. The theory of environmental factors is supported by data that suggest other anomalies, particularly intestinal atresias, and may be induced by intrauterine vascular accidents.[3]

Duplication of the stomach is usually single. It can sometimes be associated with a second duplication such as oesophageal[1] or pancreatic duplication.[6] Rare malformative associations with gastric duplication are described in the literature (mature teratoma with vertebral anomalies,[8] omental pseudocyst,[3] diaphragmatic eventration and accessory pancreatic).[2,4] Only one case of anorectal malformation associated with spinal lipoma and transdiaphragmatic duplicated gastric cyst is published in the literature.[4]

Anorectal malformations are often associated with other malformations, essentially high forms, which are in order of frequency urogenital, vertebral, gastrointestinal (atresia, oesophageal fistula and omphalocele), cardiovascular, central nervous system and limb malformations.[5] They may be part of syndromic associations (vertebral defects, anal atresia, cardiac defects, tracheo-oesophageal fistula, renal anomalies and limb abnormalities or Currarino syndrome) or chromosomal abnormalities (cat eye syndrome). In our case, it was a low anorectal malformation. Association of anorectal malformation and agenesis of the corpus callosum evoke Opitz syndrome, but there were no characteristic dysmorphic facies. A complementary genetic study was asked for our patient.

The association of gastric duplication, anorectal malformation and agenesis of the corpus callosum can probably been explained by the theory of split notochord.

Gastric duplication can be complicated such as digestive tract bleeding,[9] fistula and ulceration,[10] intra-pleural perforation,[2] intra-peritoneal perforation and malignant transformation.[11] In our case, gastric duplication was perforated by acid fluid secreted by the fundic mucosa, but fortunately, it was clogged by the omentum. There could be mass effect on adjacent normal stomach causing symptoms such as easy satiety, vomiting and when it ruptures an acid peritonitis.

The treatment of gastric duplication is complete surgical resection. Surgical alternatives are a total excision of the duplication by enucleation or a total excision with resection of adjacent segment and end-to-end anastomosis. Other modalities were described such as endoscopic or surgical drainage but exposing to the risk of recurrence. Successful management by laparoscopic approaches had been reported.[4]

In our case, we made an enucleation, but in a little part, there were adhesions, gastric muscular was sutured.

Gastric duplication is a rare congenital malformation. It still represents a challenge for pre-operative diagnosis. The treatment is surgical even when it is not symptomatic.

Conclusion

We should have a high index of suspicion for gastric duplication cyst in children with epigastric masses and always exclude associated malformations. Complete surgical resection of the duplication is the best treatment because the risk of complication, especially malignant transformation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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