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Case Report

CASE REPORT

An unusual cause of recurrent pediatric vomiting (an extraluminal pyloric duplication cystic): A case report

Leen Jamel Doya[1,*,†], Naya Talal Hassan[2], Fareeda Wasfy Bijow[1], Hanin Ahmed Mansour[1], Sawsan Ahmad[1], Fedaa Nooh[1] and Ali Ibrahim[3]

[1]Department of Pediatrics, Tishreen University Hospital, Latakia, Syria, [2]Faculty of Medicine, Tishreen University, Latakia, Syria, [3]Department of Pediatrics, Professor of Gastroenterology and Hepatology, Tishreen University Hospital, Latakia, Syria

*Correspondence address. Department of Pediatrics, Tishreen University Hospital, Latakia, Syria. Tel: 96399285698; E-mail: dr.leen.doya@gmail.com

Abstract

Vomiting is a common symptom of a multitude of diseases in children. It is usually part of benign illness and can occur at any age. Recurrent vomiting can be a symptom of life-threatening medical or surgical emergencies. It can be rarely caused by an extraluminal pyloric duplication cyst. Early recognition is essential for preventing delays in management and potential complications. Here we report a case of an extraluminal pyloric duplication presenting as progressive gastric outlet obstruction cyst in a 14-month-old Syrian boy. The diagnosis was made through abdominal ultrasound, gastrointestinal endoscopy and abdominal computed tomography scan.

INTRODUCTION

Vomiting is a common complaint in a multitude of disorders, ranging from gastrointestinal etiologies, central nervous system disease, pulmonary problems, renal disease, endocrine/metabolic disorders, and drugs (either as side effects or in overdosages) [1]. Gastric Duplication Cysts (GDC) in particular are rare causes of pediatrics vomiting. It is accounting for 4% of gastrointestinal tract duplications or roughly 17 per 1 000 000 births [2]. Cystic duplication of the pylorus is scarcely reported, only 2.2% are of gastric origin.

To our knowledge, no more than 50 cases of pyloric duplication have been reported in the literature, most of them were extraluminal masses [3].

CASE REPORT

A 14-month-old Syrian boy was referred to our clinic for recurrent incoercible, non-bilious vomiting after feeding, 4-6 episodes a day, with abdominal pain and constipation for 1 month. He was born full-term, normal pregnancy without any complications after birth. His birth weight was 4 kg. The parents deny any history of medical ingestion, toxin exposure, trauma, or falls. In his medical history, the boy was diagnosed with patent duct arteriosus (PDA) that surgical ligation at the age of 12 months.

There was no family history of genetic disorders. On physical examination, his body weight was 11 kg (50.79%; z score −1.5 SD), the length was 82 cm (87.49%; z score −2.5 SD), he was vitally stable and generally well. The abdomen was soft with no masses or organomegaly. The patient underwent multiple investigations: Complete blood count (CBC), Blood gas test, Amylase, lipase, renal and liver function, electrolytes, glucose, and thyroid-stimulating hormone were normal. Urinalysis, culture, and sensitivity were within normal limits. Echocardiography was normal.

An ultrasound (US) of the abdomen revealed a thick-walled, round cystic mass approximately measuring 3.5×3.5 cm which was adherent to the posterior wall of the pylorus, and

†Leen Jamel Doya, http://orcid.org/0000-0003-0814-5135
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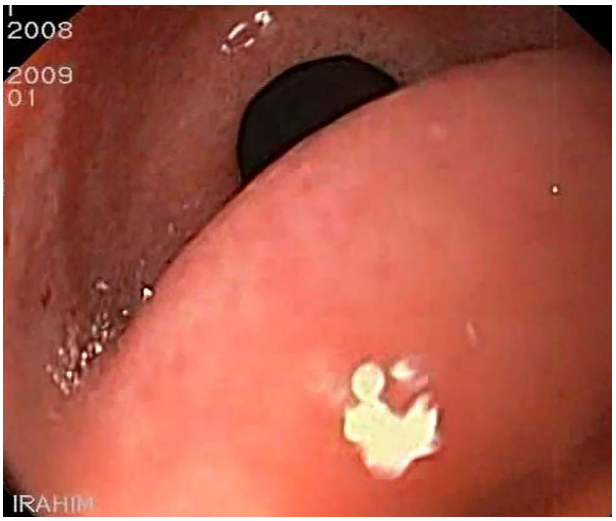

Figure 1: Esophagogastroduodenoscopy (EGD) showed narrowing in the outlet of the stomach.

compressed the pyloric channel, resulting in partial gastric outlet obstruction. An esophagogastroduodenoscopy (EGD) was performed which showed a narrowing in the outlet of the stomach (Fig. 1). An abdominal computed tomography (CT) scan showed cystic mass compressing the pyloric channel (Fig. 2a and 2b). Based on the clinical presentation and radiologic findings, a pyloric duplication cyst was suspected.

An open laparotomy was undertaken for assessment and cyst removal. At laparotomy, a cystic mass was identified inferior and lateral to the pyloric channel which appeared to compress it, resulting in gastric outlet obstruction. The cyst had no communication with the pyloric channel but it shared a muscular wall with the antimesenteric border of the pylorus.

The duplication was resected as well as the common wall and a pyloroplasty was performed (Fig. 3).

Histologic examination revealed gastric mucosa with a smooth muscle coat, which was consistent with a pyloric duplication cyst. No aberrant tissue was identified. The patient was discharged 7 days after surgery, with no postoperative complications. The patient was asymptomatic 4 years later.

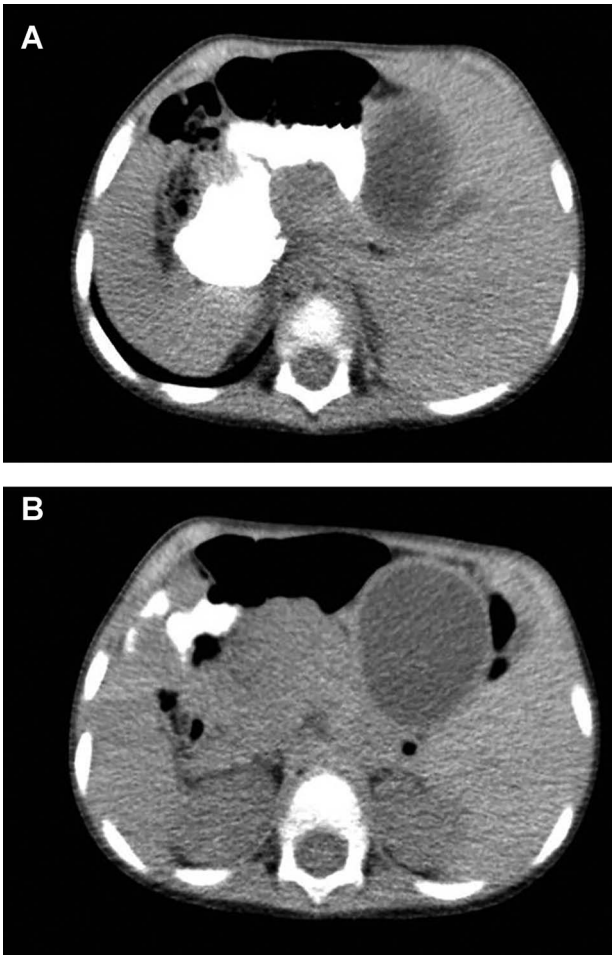

Figure 2: CT showed cystic mass compressing the pyloric channel.

Figure 3: A cystic mass measuring ~35 × 35 mm.

DISCUSSION

Vomiting after feeding in a 1-year-old patient is frequently seen in gastroenteritis (GE), but GE is usually associated with diarrhea, and recurrent vomiting is not usual. The physical examination and laboratory were normal. That can be excluded the systemic infections, metabolic and endocrine causes. There was no medical history. A surgical condition such as a gastric duplication cyst usually presents at younger than one year of age, it is more prevalent in females. Only one-third of the duplications are present in the neonatal period [4].

GDC may be associated with various other congenital anomalies like; alimentary tract duplications, genitourinary abnormalities, and an esophageal diverticulum in about 50% of cases, vertebral and spinal cord abnormalities (16–26%) [5].

In our case, the patient had a corrected cardiac malformation associated with an extraluminal pyloric duplication cyst which was the first case described in the literature.

Symptoms of pylorus duplication are non-specific that vary according to the size, site and the type of fluid secreted by their mucosa. Symptoms usually range from a nonobstructing lesion to an obstructing mass, even mimicking infantile hypertrophied pyloric stenosis [6]. Our patient is male and has vomiting with good weight gain at 14-months-old.

The first report on the duplication cyst was by Calder in 1733. However, it was not until 1937 when William E. Ladd

introduced the term duplication of the gastrointestinal tract [7]. Cystic Duplication of the pylorus has an embryonic origin, it probably develops in the sixth week of the embryonic period but the etiology has not been elucidated [8].

GDC must be continuity of the cyst wall with the stomach and shared in blood supply. It should be no interruption between the smooth muscle layer of the doubling area and the normal intestine. The cyst should have an epithelial lining of the gastrointestinal tract [6]. Diagnostic confirmation often requires complementary imaging tests such as US, CT and/or magnetic resonance imaging (MRI) [8]. The US is the most common modality used to image duplication cysts. GDC characterizes in US as a 'double-wall sign,' delineating the echogenic inner mucosa from the hypoechoic outer rim of muscle [9]. In the current case, US only made a diagnosis and showed an anechoic cystic mass with a double-wall sign.

Treatment of gastric duplication is complete surgical resection due to the risk of malignant transformation. Surgical resection can be performed by open and laparoscopic techniques [10].

In summary, this was an unusual case of gastric outlet obstruction caused by an extraluminal pyloric duplication cyst in a boy with cardiac malformation. This case demonstrates the effectiveness of using the US to diagnose gastrointestinal duplication cysts.

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CONFLICT OF INTEREST STATEMENT

None declared.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This case report did not require review by the Ethics Committee Tishreen University Hospital, Latakia, Syria.

FUNDING

None.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient's parents for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor.

GUARANTOR

Leen Jamel Doya, Department of Pediatrics, Tishreen University Hospital, Latakia, Syria.

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