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Case Report

True pylorus duplication: a case report

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ABSTRACT

True pylorus duplication cysts are particularly rare, the literature describes only a small number of cases. They account for nearly 2.2% of all alimentary tract duplications. Most of the gastric duplications presents at an age younger than one year with one-third diagnosed in the neonatal period. However, they are not limited to that period. We report a case of a true pyloric duplication cyst in a 3-year-old girl who had been experiencing frequent vomiting with occasional abdominal pain for about 15 days prior to her presentation. The patient had an abdominal ultrasound that showed an epigastric cystic mass. CT of the abdomen demonstrated a 3.8×3.8 cm mass with a claw sign proximal to the first part duodenum. The diagnosis of pyloric duplication cyst was confirmed during the operation and with the histopathological examination following the resection. The patient is doing well with an appropriate weight gain at the follow-up appointments. The patient is doing well with an appropriate weight gain at the follow-up appointments. In summary, this is an an unusual case of gastric outlet obstruction that presented outside the usual age period caused by a pyloric duplication cyst. Imaging studies including US and CT are useful aids in the diagnoses of gastrointestinal duplication cysts.

Keywords: Pyloric duplication cyst, Gastrointestinal duplication, Resection, Pyloroplasty, Case report

INTRODUCTION

The term “intestinal duplication” was promoted by Ladd in 1937.[1] gastrointestinal tract duplications are scarce and may occur in 1 out of 4500 births.[2] True pylorus duplications are even immensely rare, only few cases has been described in the literature. They account for approximately 2.2% of all alimentary tract duplications with a female predominance.[3,4]

Duplication of the pylorus clinically displays a myriad of signs and symptoms. This includes, but not confined to, abdominal mass, pain, and gastric outlet obstruction. The precise diagnosis is rarely made before surgery despite the imaging studies. In the end the principal of treatment is complete resection.

CASE REPORT

A 3-year-old girl was brought to our hospital by her mother with frequent vomiting of undigested food for the last 15 days. She also had occasional abdominal pain that was relieved by vomiting. By the time of presentation, the vomiting happened only at the end of the day. Prior to her presentation, she was hospitalized for two days for hydration and observation then was discharged. However, she relapsed again so her mother took her to another hospital where CT abdomen and pelvis was done, and an intra-abdominal cyst was found.

Her past medical and surgical histories were unremarkable. Her prenatal and birth history were also unremarkable. Clinical examination revealed an epigastric fullness with no tenderness.

CBC, renal profile, LFT, α-feto-protein, ß-hcg, CA 125, and carcinoembryonic antigen were ordered and were all unremarkable. US abdomen showed a cystic mass containing multiple debris at the epigastric area with a claw sign to the first part of the duodenum measuring 3.8×3.8 cm (Figure 1) CT abdomen was done and showed a cystic mass closely related to the duodenum measuring 4×4×5 cm. It was exerting a mass-effect to the stomach and duodenum which caused upstream dilatation of the stomach (Figure 2).

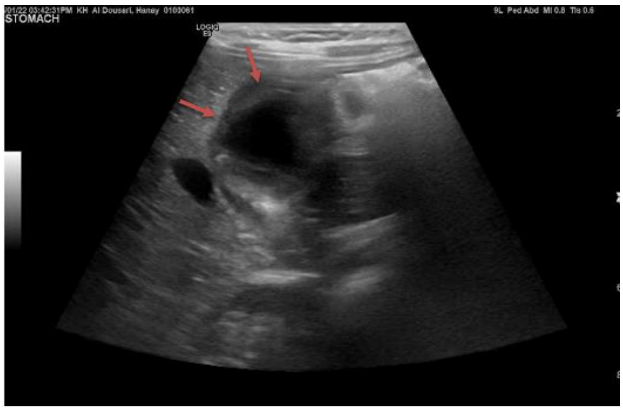

Figure 1: Ultrasonography is showing a 3.8×3.8 cm cyst with claw sign to the first part of the duodenum.

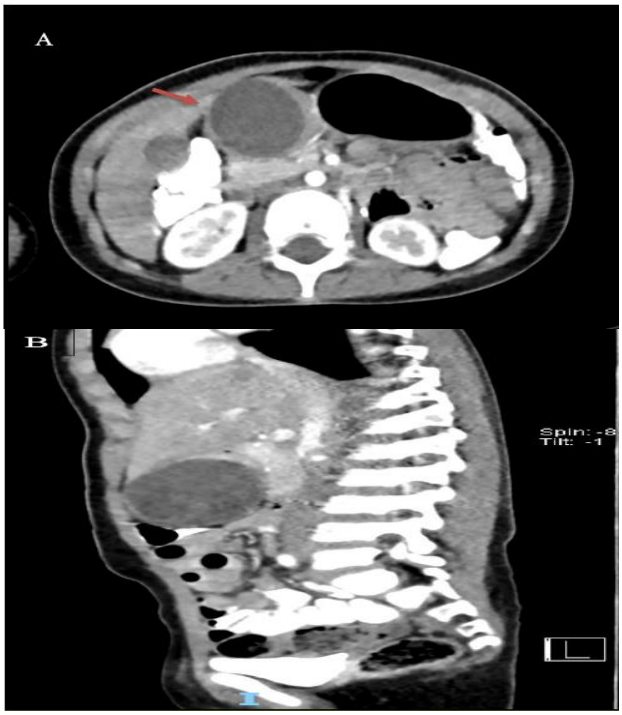

Figure 2 (A and B): CT showed cystic mass compressing duodenum and stomach. Transverse palne and Saggital plane.

Based on the results of the investigations, the patient was scheduled for laparoscopic cyst resection. Three five-millimeter ports were used at the supra-umbilical, right iliac fossa and left iliac fossa. At the laparoscopy, a cystic mass was found adherent to the posterior and inferior aspect of the pylorus and was stretching the first part of the duodenum (Figure 3). During mobilization of the cyst, a communication between the cyst and the pylorus was discovered of two centimeters in diameter.

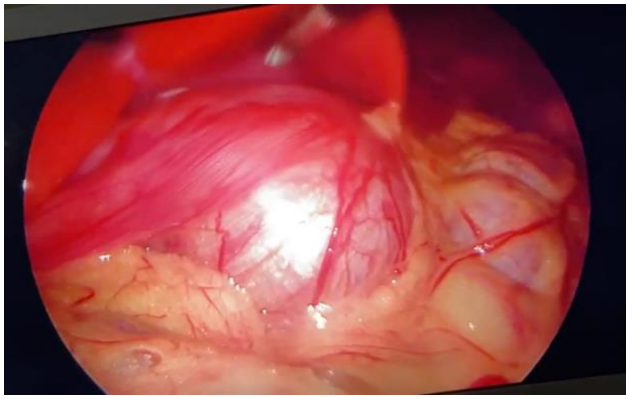

Figure 3: The cyst was found at the anterior-inferior aspect of the pylorus.

Due to the uncertainty of the diagnosis and the presence of the communication, conversion to laparotomy was decided. The Supra-umbilical port incision was utilized. The communication between the duplication cyst and the pyloric channel was identified (Figure 4). The cyst was enucleated and excised en bloc from the pyloric wall.

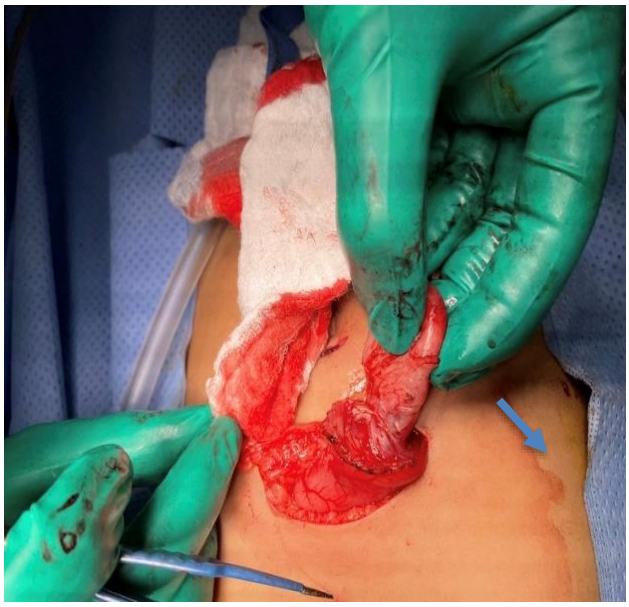

Figure 4: Site of communication between the cyst and the pyloric channel.

The site of the communication was closed transversely starting with the mucosa and then the shared muscular wall with the cyst was closed in two layers restoring the

pyloric anatomy and hence minimizing the post-operative complications (Figure 5).

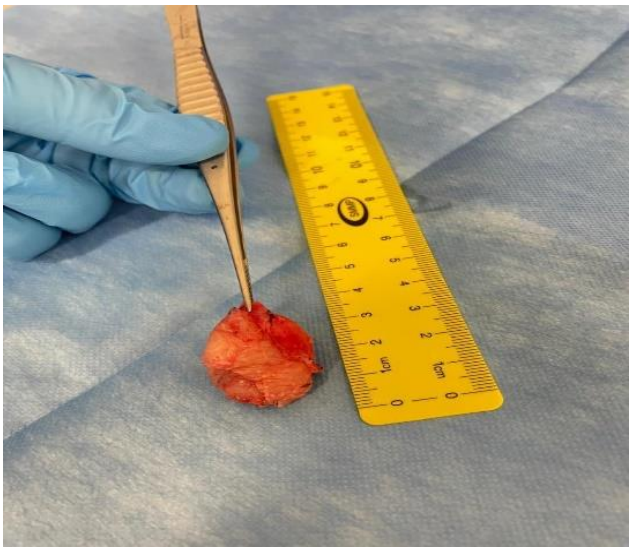

Figure 5: Pyloric cystic mass measuring ~30×30 mm.

After the procedure, patient had an uneventful recovery and started feeding on the second day. Histological evaluation of the resected specimen revealed the presence of a cyst containing fibromuscular wall with inflamed and ulcerated pyloric type mucosa. This confirmed the diagnosis of pyloric duplication cyst. The patient was asymptomatic during the 12-month follow-up period.

DISCUSSION

The phrase duplication of the alimentary tract was first coined by William E. Ladd in 1937. He described three common findings: a well-developed smooth muscle coat, epithelial lining, and attachment to the alimentary tract.[1] However, Ramsay reported the first pyloric duplication cyst discovered in an 8-day-old girl in 1957.[5]

Enteric duplications may arise anywhere in the gastrointestinal tract, with an incidence of 1 in 4500 births.[2] Among them, gastric duplication cysts are behind 4% of the gastrointestinal tract duplications or roughly 17 per 1 000 000 births. Cystic duplication of the pylorus is scarcely reported, and they only accounts for 2.2% of all alimentary tract duplications cysts.[3] The female to male ratio is 2:1.[4]

To our knowledge, no more than 34 cases of true pyloric duplication have been reported in the literature and most of them were extraluminal and non-communicating. The present study describes the first true pyloric duplication cyst in Saudi Arabia.

Symptoms usually range from a non-obstructing lesion to an obstructing mass, even mimicking infantile hypertrophied pyloric stenosis. Common clinical manifestations include an abdominal mass and non bilious vomiting which are almost present in all the cases in the literature.[17] Vomiting is a shared symptom of a multitude of diseases in children. It is usually part of benign illnesses but can also be a symptom of lethal surgical emergencies. Gastric duplication cysts in particular are rare causes of pediatrics vomiting.

The etiology of cystic duplication of the pylorus has not been explicated. Several theories have been suggested to clarify their development. These include the persistence of embryonic diverticulae throughout alimentary tract development, abortive attempts at twinning, prenatal vascular accidents, and recanalization and merging of embryological longitudinal folds.[6-10]

Associated anomalies are described in approximately 20% of duplication cases. This includes genitourinary malformations, spinal malformations, intestinal atresia and malrotation.[11]

Gastric duplication cyst are mostly diagnosed in the neonatal period, but may present anywhere from 1 day of age up to 5 years of age and is thus not limited to the neonatal period alone.[6,12] Our patient presented at 3 years of life. Such a late presentation is uncommon and was encountered in only five cases in the literature with ages between 2 and 5 years old.[12]

The most common image modality used in these cases is ultrasonography. Other diagnostic studies, including upper gastrointestinal contrast study, gastrointestinal endoscopy, and CT abdomen, are used in establishing the preoperative diagnosis. Because of the nonspecific symptoms and rarity, the correct preoperative diagnosis is seldom possible.[17]

Surgery is the undisputed mainstay of treatment. Surgical options should be individualized for each case and varies from simple excision to pyloroantrectomy. In all surgical procedures it is essential to excise all of the cyst lining. Marsupialization, however, must be avoided because carcinoma has been found in an adult gastric duplication.[16]

CONCLUSION

Duplications involving the pylorus are particularly rare. This was an atypical case of gastric outlet obstruction with late presentation caused by a pyloric duplication cyst. Imaging studies including US and CT are useful to diagnose gastrointestinal duplication cysts.

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