Case Report

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Intraluminal pyloric duplication cyst- a rare cause of non-bilious vomiting in a neonate: A case report

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KEYWORDS

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ABSTRACT

Background: Duplications of the alimentary tract are rare congenital malformations, out of which, true pyloric duplications constitute only 2.2%. They present with non-bilious vomiting and mimic hypertrophic pyloric stenosis (HPS). Pyloric duplications that are intraluminal are not separately visible at laparotomy, making their diagnosis difficult.

Case Presentation: Our case is a neonate with an intraluminal pyloric duplication cyst who presented with recurrent vomiting. The radiological evaluation suggested a duplication cyst medial to the second part of the duodenum towards the stomach's lesser curvature with features of gastric outlet obstruction. Intraoperatively, a cystic mass of 1 x 2 cm intraluminally was found on opening the pylorus which was excised and pyloroplasty was done. Postoperatively the baby was discharged in a stable condition.

Conclusion: Intraluminal pyloric duplication cysts are rare and the clinical presentation mimics HPS. They should be considered as a differential diagnosis in a neonate presenting with features of gastric outlet obstruction.

INTRODUCTION

Duplications of the alimentary tract along the pylorus are rare congenital malformations, with the intraluminal variants being even rarer. The clinical presentation mimics that of hypertrophic pyloric stenosis (HPS). They are diagnosed intraoperatively as they are not commonly considered during imaging. [1] Complete excision of the cyst along with the mucosal lining is the recommended surgical treatment. [2] In this report, we share the case of a neonate presenting with features of gastric outlet obstruction due to a pyloric duplication cyst that was intraluminal.

CASE REPORT

A term, male baby with a birth weight of 2.8kg, with no antenatally diagnosed congenital anomalies, was born by normal delivery to a gravida 2 mother. The neonate passed meconium on day 1 of life. From 3rd day of life, the child developed non-bilious vomiting after feeds, which was non-projectile, however, the feeding was normal. The child was brought to our center on day 9 of life with persistent non-bilious vomiting.

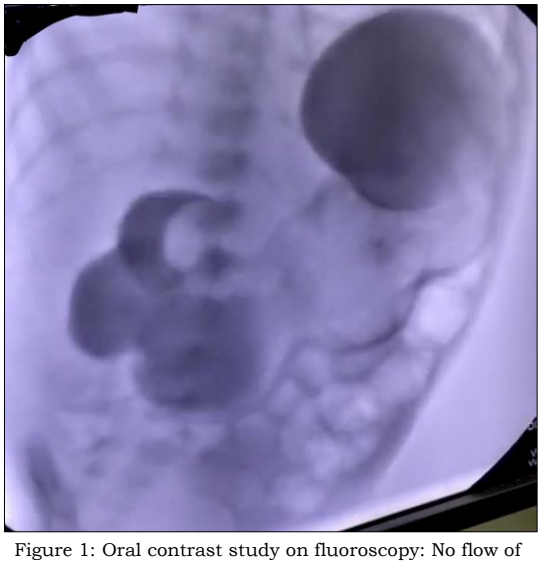

Figure 1: Oral contrast study on fluoroscopy: No flow of contrast distal to the second part of the duodenum.

On arrival, the child weighed 2.6kg, heart rate was 124 per minute, respiratory rate was 40 per minute and there was mild dehydration. Oxygen saturation was 98% on ambient air. The abdomen was not distended, there was no visible peristalsis, no mass palpable per abdomen, and bowel sounds were normal.

On the X-ray abdomen, the stomach appeared distended and there were a few air shadows in the small bowel. On Ultrasonography (USG) of the abdomen, there was a cystic lesion with bowel signature measuring 12.1mm x 20.6mm x 22.5mm, inferomedial to gall bladder suggestive of enteric duplication cyst. Venous blood gas analysis revealed mild alkalosis with a pH of 7.46. Serum sodium was 133mEq/L and serum potassium was 3.5mEq/L. On oral contrast study, there was no flow of contrast distal to the second part of the duodenum (Fig. 1).

A clinico-radiological diagnosis of duodenal duplication cyst was considered and the baby was taken up for surgery. Intra-operative findings included a bulky pylorus with a thickened wall on the side of the lesser curvature and a normal wall on the greater curvature. No other lesion was seen separately (Fig. 2).

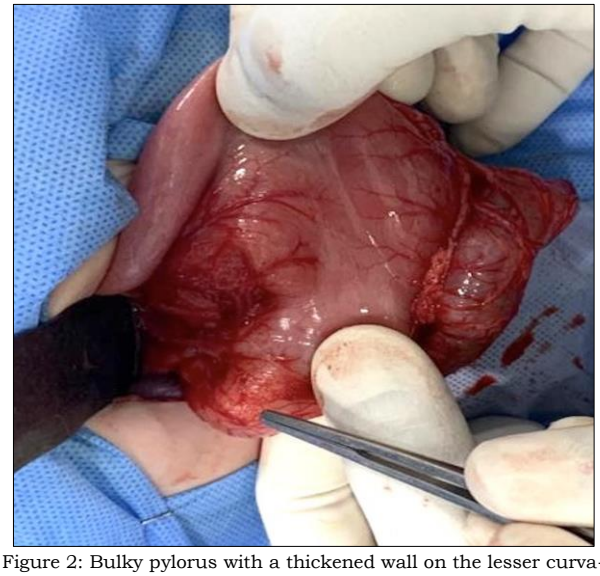

Figure 2: Bulky pylorus with a thickened wall on the lesser curvature of the pylorus.

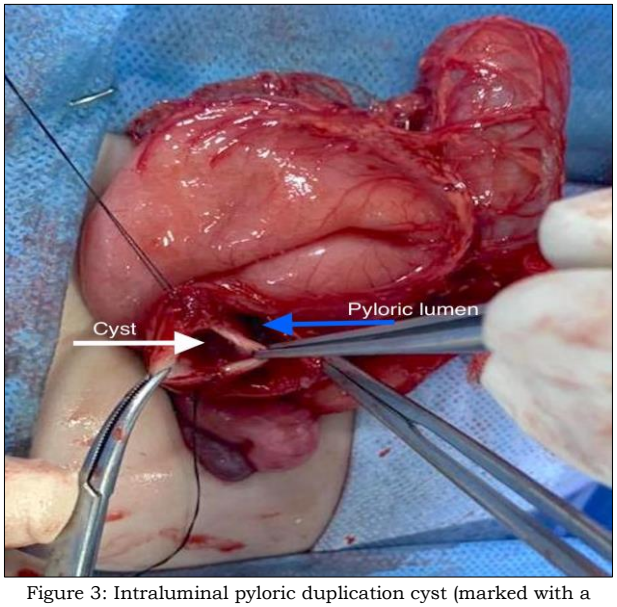

Figure 3: Intraluminal pyloric duplication cyst (marked with a white arrow) sharing a muscular wall with the pylorus (marked with a blue arrow) on the lesser curvature side.

On incising the anterior surface of the pylorus longitudinally, a cystic mass of 1 x 2 cm was seen adjacent to the pylorus towards the lesser curvature compressing the pylorus, resulting in gastric outlet obstruction (Fig. 3). The duplication cyst was found to share a wall with the pylorus however was not communicating with the pyloric channel. The cyst was excised, a Heineke-Mikulicz pyloroplasty was done and a feeding jejunostomy was made to initiate early feeding.

The baby was started on jejunostomy feeds on the 3rd postoperative day, which was progressively increased, and on postoperative day 13 was started on breastfeeds. The feeding jejunostomy tube was removed prior to the discharge of the child. Histopathology confirmed it a pyloric duplication cyst. The child remained asymptomatic during postoperative follow-ups. The last follow-up was at six months after surgery and the child has been gaining weight and has normal developmental milestones.

DISCUSSION

Our patient was a term-born male child who presented with non-projectile, non-bilious vomiting after feeds from the third day of life. With this history, the differential diagnosis considered was any of the causes of non-bilious vomiting in a term neonate like gastroesophageal reflux, metabolic disorders, HPS, antral web, pyloric atresia, or antropyloric duplication cysts. X-ray abdomen with air shadows in the small bowel ruled out the possibility of pyloric atresia. Venous blood gas with mild alkalosis was not characteristic of HPS. USG abdomen in correlation with an upper GI contrast study suggested a duodenal duplication cyst, for which the patient was taken up for surgery. Intraoperatively, an intraluminal duplication cyst sharing a common wall with the pylorus was identified which was resected. As the pyloric channel along with the mucosa was opened longitudinally in the incision, a Heineke-Mikulicz pyloroplasty was done to avoid any possibility of narrowing of the pylorus in the postoperative period. To establish feeding early, feeding jejunostomy was made.

Clinical identification of the exact cause of gastric outlet obstruction in a neonate is difficult, however radiological investigations usually indicate the most likely etiology, but they are also not always accurate, as has been in the present case.

Gastric outlet obstruction is rarely caused by gastrointestinal tract duplications. Gastrointestinal duplications have an incidence of 1: 4500. [3] They are spherical or tubular cystic structures arising anywhere along the gastrointestinal tract, lined with alimentary tract epithelium and with smooth muscle in its wall. [3] Less than 4% of all duplications involve the stomach, and most (85%) do not communicate with the gastrointestinal lumen. [4] Pyloric duplica-

tion cysts are uncommon lesions of which most are extraluminal. Intraluminal pyloric duplication cysts are extremely rare [5] and hence the diagnosis is almost always never made before surgery. [6]

Table 1: Literature on cases of neonatal pyloric duplication

S No	Author	Age/ Sex	Clinical presentation	Diagnosis	Management	Histology	Outcome
1	Ramsay [7] (1957)	8d/F	Projectile vomiting, visible peristalsis, mass	Discovered intraoperatively	Local excision antrum closed transversely	Pyloric mucosa	Vomiting for 2 days post-op, then recovered
2	Abrami & Dennison [10] (1961)	7d/F	Vomiting, no mass	UGI	Posterior gastrojejunostomy	-	Died
3	Grosfeld et al [11] (1970)	3d/M	Non-bilious vomiting. No mass	Discovered intraoperatively	Cyst excision	Pyloric, small bowel mucosa, ectopic pancreatic tissue	Recovered
4	Anas & Miller [12] (1971)	12d/M	Mass, later vomiting	UGI	Cyst mucosal stripping	-	Recovered
5	Bower et al [13] (1978)	3 weeks	Vomiting, mass	-	Cyst excision	-	Recovered
6	Keramidas et al [14] (1980)	20 d/F	Intermittent vomiting since day 3 of life, mass	UGI	Cyst excision	Pyloric mucosa	Recovered
7	Birenbaum et al [15] (1982)	2d/F	Vomiting	UGI & EGD	Cyst excision and Heinecke Miculicz pyloroplasty	Gastric and duodenal mucosa	Recovered
8	Bommen & Singh [16] (1984)	5d/M	Preterm, Hyperbilirubinemia, vomiting, mass	UGI & USG	Cyst excision	Gastric mucosa	Recovered
9	Goyert et al. [17] (1991)	Perinatal/F	Antenatal abdominal cyst of unknown etiology. Asymptomatic	USG	Cyst excision	Antral and pyloric mucosa	Recovered
10	Murty et al [18] (1992)	Newborn/M	Mass, Melena on day 2	UGI & USG	Cyst excision	-	Recovered
11	MP Patel et al [19] (1997)	3d/M	Vomiting, mass	USG	Pyloroantrectomy	Gastric mucosa	Pseudomonas meningitis on POD 10 and died
12	Shah et al. [20] (2005)	3d/F	Vomiting, mass	USG	Pyloroantrectomy	Gastric, small bowel mucosa	Recovered
13	Saad et al [4] (2005)	Newborn	Mass	USG	Lap assisted resection	Gastric mucosa	Recovered
14	CK Sinha [21] (2007)	9d/F	Non-bilious vomiting, mass	USG	Cyst excision and cauterizing remnant mucosa	Pyloric mucosa	Recovered
15	VD Upadhyaya [22] (2009)	21d/M	Non-bilious vomiting, mass	USG, UGI	Cyst excision and pyloro- duodenal anastomosis	Duodenal lining	Uneventful
16	Anthony Chin [23] (2010)	11d/F	Non-bilious vomiting, mass	USG, UGI	Cyst excision	Gastric epithelium and accessory pancreatic tissue	Recovered
17	Trainavicius et al [1] (2013)	2d/F	Vomiting, mass	Antenatal USG & CT	Cyst excision	Duplication cyst pylorus	Recovered
18	Cristina Marginean [24] (2014)	7d/M	Vomiting, dehydration	Antenatal USG	Initial cyst aspiration then cyst excision and cauterizing remnant mucosa	Pyloric duplication and ectopic pancreatic tissue	Symptomatic 20 days after 1st surgery then underwent second surgery, subsequently recovered
19	KD Lee [25] (2017)	3d/F	Non-bilious vomiting, dehydration, mass	USG, UGI, MRCP	Partial cyst excision and mucosectomy	Gastric mucosa	Recovered

Gordon Ramsay, in the year 1957, reported the first case of neonatal pyloric duplication. [7] Subsequently, literature on pyloric duplication in neonates has been limited to case reports. Symptoms of enteric duplication cysts include recurrent abdominal pain, vomiting from intestinal obstruction, or hematochezia resulting from ulceration of ectopic gastric mucosa.

Cysts near the pylorus, as in our case, are known to present in the neonatal period with persistent non-bilious vomiting simulating HPS. [8] Pyloric duplications in neonates usually present with vomiting and a palpable mass per abdomen. Complete resection is considered the mainstay of treatment. [1] The surgeries performed vary from simple excision to pyloroantrectomy. [2] Marsupialization is to be avoided because carcinoma has been reported in an adult gastric duplication. [9] Drainage procedures are required only when extensive resection would otherwise be needed because of large cyst size or proximity to the common duct etc. Procedures such as cystgastrostomy, cystenterostomy Roux-en-Y, and a "window" procedure have been done in various situations. [2]

The demography, presentation, diagnosis, surgical management, histology, and outcome of cases of neonatal pyloric duplication that have been mentioned in the literature has been tabulated in Table1. [1,4,10-25]

In the present case, the child was taken up for surgery with suspicion of a duodenal duplication cyst, however, an intraluminal pyloric duplication cyst was discovered and excised. The histopathology of the excised cyst showed Brunner's gland in submucosa and the muscularis layer was present suggestive of a duplication cyst.

In conclusion, pyloric duplication cyst is a rare congenital malformation that mimics HPS. It may be considered as a differential in neonates with non-bilious vomiting and surgical management is the mainstay of treatment.

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