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CASE REPORT

Isolated pylorospasm in an infant: A diagnostic and therapeutic conundrum

Dhruv Mahajan,[1,*] Shubhi Gupta[2] and Hari Krishna[3]

[1]*Assistant Professor, Department of Paediatric Surgery, Geetanjali Medical College, and Hospital, Udaipur*
[2]*Assistant Professor, Department of Obstetrics and Gynaecology, Geetanjali Medical College, and Hospital, Udaipur*
[3]*Assistant Professor, Department of Paediatric Surgery, Government Medical College, Goa*

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Abstract

Persistent vomiting in a small infant can be due to delayed gastric emptying which is a significant cause of distress and morbidity. After ruling out common medical causes, evaluation for a surgical cause such as pyloric stenosis is imperative. The diagnosis of pylorospasm is often missed between the medical and surgical realms. We describe a six-week-old infant with persistent nonbilious vomiting who was referred with a suspicion of pyloric stenosis. However, on evaluation clinical findings and imaging were not suggestive of pyloric stenosis. A diagnosis of pylorospasm was made and the patient was managed with anticholinergics without any surgery. The patient had an unremarkable recovery. The diagnosis of pylorospasm needs to be kept in mind in an infant with persistent nonbilious vomiting. It responds satisfactorily with pharmacotherapy and avoids the need for surgery.

Keywords: Pylorospasm, pyloric stenosis, anticholinergic, infant, nonbilious vomiting

*Corresponding author: Dhruv Mahajan
Email: 280691dhruv@gmail.com

Abbreviations

GERD: Gastroesophageal reflux disease
IHPS: Infantile hypertrophic pyloric stenosis
PS: Pylorospasm
UGI: Upper gastrointestinal

Introduction

Repeated episodes of nonbilious vomiting in infants are a common cause of discomfort for the child and parents. Significant vomiting and intolerance of feeds leading to dehydration need careful examination and evaluation. Common causes in infants aged 2-8 weeks include infantile hypertrophic pyloric stenosis (IHPS) and gastroesophageal reflux disease (GERD). Pylorospasm (PS) is often considered as a differential but without any set diagnostic or management criteria. In the absence of any specific symptom, sign, or investigation, it is more of a diagnosis by exclusion [1]. In the recent literature, there have seldom been any reports of this entity.

Pyloric narrowing leading to gastroparesis causes a functional obstruction at the pylorus imitating IHPS and possibly GERD [2]. Clinical examination and imaging can reliably diagnose IHPS. However, no such clinching signs are available for PS. Hence, a high index of suspicion is necessary to avoid any unnecessary surgery or persistent symptoms, as possible PS cases can be managed medically successfully. Here, we describe a six-week-old infant with vomiting managed as PS and discuss the available literature on the same topic.

Case report

A six-week-old first-born male infant born to a thirty-year-old female presented with a history of multiple episodes of nonbilious projectile vomiting for five days. There was no previous comorbidity. The child was born via normal vaginal delivery at full term with appropriate for age weight and had been breastfeeding normally until five days prior. At examination, the patient was well preserved with a weight of 3.6 kilograms with depressed fontanelle and delayed capillary refill time. There was no history of fever, loose stools, inconsolable crying, or any other localizing signs. On abdominal examination, there was no abdominal distension, tenderness, or guarding. No mass or olive was palpable. There was metabolic acidosis (pH 7.2) in blood gas with normal serum electrolytes. Complete blood counts were normal with a total leucocyte count of 10,000/uL. Serum C-Reactive Protein was 2mg/L and serum Procalcitonin was 0.05ng/ml. Along with initial resuscitation with intravenous fluids, ultrasonography was ordered. There were no signs of hypertrophic pyloric stenosis on ultrasonography with a normal pyloric canal wall thickness, length, and diameter (Figure 1). However, a distended stomach was found. A contrast study revealed narrowing and stasis at the pylorus with a distended stomach, which subsequently had a small delayed clearance with a significant hold-up of contrast in the stomach (Figure 2). There was no GERD or malrotation observed in the contrast study. Nasogastric decompression via nasogastric tube was done and the child was kept nil per oral. The nasogastric tube was removed after three days, and small-volume trophic feeds were initiated with anti-GER measures. There was recurrent projectile vomiting and on repeat ultrasound, there was no IHPS.

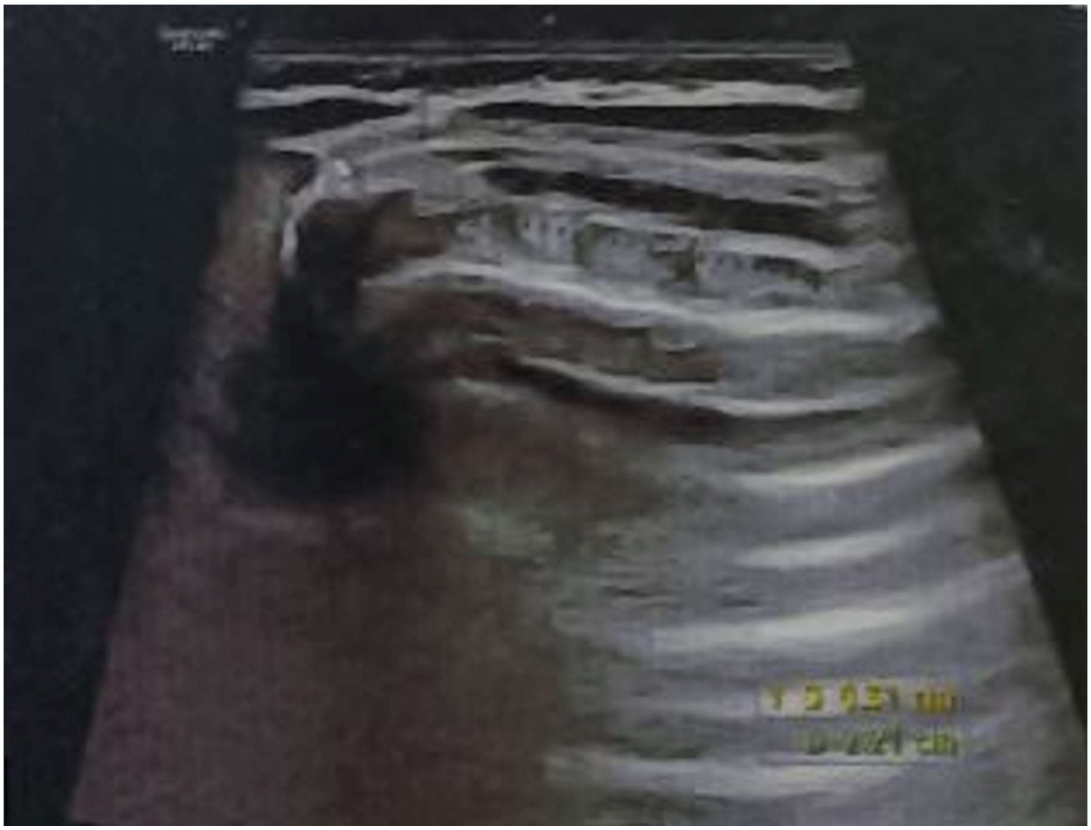

Figure 1. Ultrasound depicting normal pyloric parameters: normal wall thickness, normal diameter, and canal length, not suggestive of pyloric stenosis. Passage of gastric contents and opening of the pylorus were observed while monitoring via ultrasound over a span of 10 minutes.

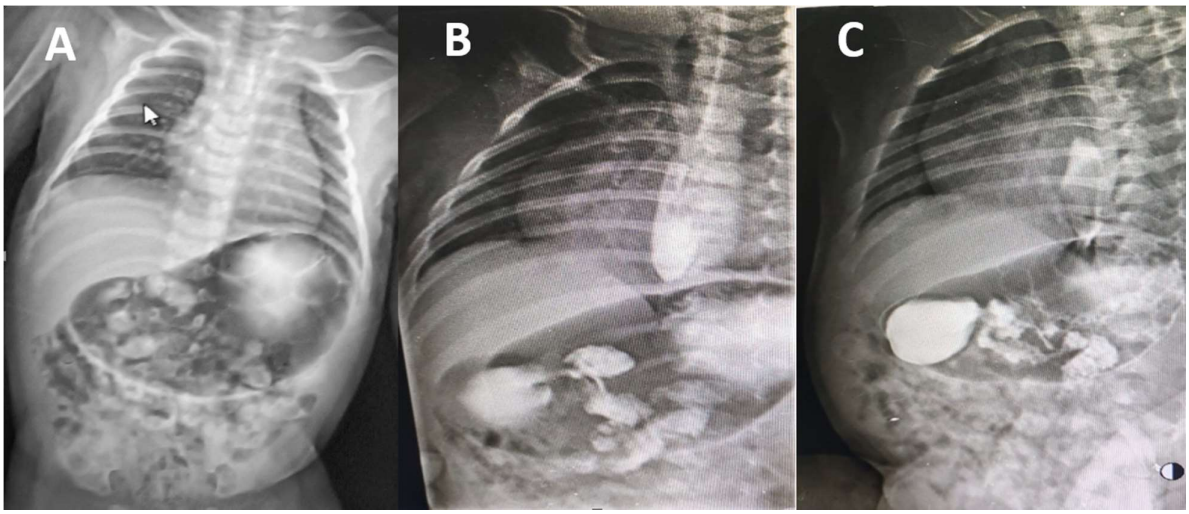

Figure 2. Upper gastrointestinal contrast study showing (**A**) a distended stomach with (**B**) hold up of contrast at the pylorus. However, a slow distal contrast run-off is observed after 40 minutes (**C**).

Isolated anti-GER measures were unable to alleviate the symptoms. All the history, investigations, and treatments were reviewed and a suspicion of pylorospasm as the cause was considered. Gastroesophageal reflux was ruled out based on history and even then anti-reflux measures were started after admission. However, there was no response. Gastroenteritis and Drug-induced gastroparesis were ruled out based on feeding history. The baby was fed only breastfed and no top feed was given. Hygiene-related issues were ruled out after evaluating the mother's feeding habits. The stool microscopy and culture were also negative. There was mild dehydration, which was corrected on admission. However, the symptoms persisted even then. Sepsis was ruled out clinically and laboratory tests as CRP and PCT were negative. Intestinal obstruction and malrotation of the gut were ruled out on plain abdominal radiographs and ultrasonograms, respectively. There were no signs, symptoms, or any other indication suggestive of a raised intracranial pressure.

Since the symptoms were intractable and persistent even on conservative management and reflux measures. The parents were duly explained about the differential diagnosis of pylorospasm. The parents were also explained that hyoscine is not the traditional management and has been rarely used in children, so the side effect profile is not well known in children. The patient was kept in the PICU for 48 hours after starting the medication, without any cost to the patient.

Given the suspicion of PS, the child was started on antispasmodic therapy in the form of hyoscine butylbromide at 0.2 mg twice daily. The patient was kept nil per oral for two days and then small-volume feeds (expressed breast milk) were started initially at the rate of 5ml every 3 hourly. The feeds were gradually increased and breastfeeding was started on the third day after initiating medication. By the morning of the fifth day, the baby was accepting breastfeeding ad-lib. The child tolerated feeds without any vomiting. The child was discharged on the fifth day and the dose was decreased to 0.2mg once daily. Repeat ultrasonography after two weeks, showed normal pyloric wall thickness, length, and diameter. Following this, 0.2mg alternate day was given for one week and then the medication was stopped completely. The child had no recurrence of symptoms and is thriving well at follow-up performed at six months of age with adequate weight gain.

Discussion

An infant with repeated episodes of persistent nonbilious vomiting requires urgent attention. Since smaller infants have poor reserves and are prone to dehydration leading to electrolyte disturbances, this can be deadly. With the advent and wide-scale acceptance of ultrasonography, the diagnosis of hypertrophic pyloric stenosis as a cause is readily picked up and treated. However, in the absence of sonographic findings, the remaining cases are attributed to either GERD or anecdotal. PS is an uncommonly described entity in patients with similar symptoms. The patients with PS are unlikely to improve with therapy directed at GERD and may worsen if not treated promptly.

However, the exact etiology or incidence of PS in infants is unknown. One case reported pylorospasm in a neonate due to an underlying subhepatic abscess due to *staphylococcus aureus* [3]. PS is a commonly suspected, seldom found, and

rarely proven entity in infants. There is no concrete literature available on the incidence or correlation with pyloric stenosis. Repeated episodes of non-bilious vomiting in a 4–6-week-old infant could be due to IHPS, GERD, PS, antral webs, duplication cyst, gastritis, gastroenteritis, raised intracranial pressure, or metabolic disorders among other causes. For an infant with typical examination findings of olive with corroborative evidence in ultrasonography and/or UGI contrast study, the diagnosis of IHPS can be made conclusively. However, in the absence of findings of IHPS or reflux on imaging, the diagnosis of PS could be considered. The double-track sign seen in ultrasound can be seen in both IHPS and PS and lacks specificity [1,4]. An ultrasonographic examination that suggests IHPS for a part of the study but changes during the study itself could be suggestive of PS [5]. In the future, investigations like antroduodenal manometry or ENDOFLIP could help to diagnose this entity. But at present, the lack of availability and expertise limits its use in general clinical practice.

Another school of thought is that PS as such could be a precursor in the natural history of IHPS. Sustained contraction of the pylorus could lead to hypertrophy of the pyloric muscle leading to IHPS over time. Wesley et al. followed up on ten patients with PS and found that all ten patients developed IHPS requiring surgery after 2 to 46 days (mean 13 days) [6]. Considering this, it is paramount to follow-up these patients to screen for resolution of symptoms. In case of persistence of symptoms, repeat imaging to rule out IHPS is warranted.

A plethora of therapies have been described in the literature for the management of PS. Thick cereal feeding was described initially, but at an age of 2-8 weeks, when these patients are most present; it is not a suitable therapy, especially for those on exclusive breastfeeding. Decreasing the volume of individual feeds and increasing the number of feeds has also been described to cause alleviation of PS [7]. A therapy of frequent stomach wash along with an injection of atropine before feeding with a mixture of albumin milk and the dextrin-maltose combination has been tried in earlier times. Drugs such as banthine bromide, procaine amide, and rociverine have also been used successfully [8]. Anticholinergic therapy in the form of intravenous atropine has been documented to have a success rate in the management of PS [9]. Oral anticholinergics that function as antispasmodics along with watchful observation are therapeutic. A strict follow-up is essential along with repeat evaluation if symptoms recur. Since it is a single case, so it is difficult to conclude that the resolution of symptoms with treatment has a temporal resolution or is serendipitous. Further research is necessary for a similar cohort of patients regarding diagnostic and management guidelines.

Conclusions

PS should be suspected in a small infant with recurrent nonbilious vomiting not attributable to any other cause. Medical treatment should be started when in doubt, for resolution of the distressing symptoms. It is a treatable cause of repeated vomiting in an infant with a good outcome, so a high index of suspicion should be maintained.

Statements and Declarations

Ethics approval

Ethical approval is not applicable.

Consent for publication

Written informed consent was obtained from the patient's parents for the publication of this case report and accompanying images.

Availability of data and material

Not applicable

Competing interests

The authors declare that they have no competing interests.

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