Case Report

An Unusual Surgical Cause of Pyloric Stenosis in an 8-Month-Old Infant

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Abstract

Infantile hypertrophic pyloric stenosis (IHPS) is the most common surgical cause of nonbilious, projectile vomiting in children. Its incidence is estimated to be 2–5/1000 live births and has been noted to be quite rare in African-Americans, Asians, and Indians. It is more common in infants under 6 months of age and is quite rare in older infants. While IHPS is invariably intrinsic in nature, extrinsic pyloric stenosis is very rare. Children who present with nonbilious, projectile vomiting after the age of 6 months should, therefore, be thoroughly investigated for causes other than IHPS. This is a case report of an 8-month-old child who had a band of tissue constricting the pylorus which mimicked IHPS.

Keywords: Hypertrophic, infantile, pyloric stenosis, surgical cause, unusual

Introduction

Infantile hypertrophic pyloric stenosis (IHPS) is the most common surgical cause of non-bilious, projectile vomiting in children. It is estimated to occur at a rate of 2–5/1000 live births. It is rare in African-Americans, Asians, and Indians. It is more common in infants under 6 months of age than it is in older ones [1].

Although childhood pyloric stenosis is usually due to intrinsic causes (of which IHPS forms the bulk)[2,3], extrinsic obstruction of the pylorus has been reported to simulate IHPS.[2,4] We report an 8-month-old child who had a band of tissue constricting the pylorus, and mimicking IHPS.

Case Report

An 8-month-old boy, human immunodeficiency virus (HIV) exposed but HIV deoxyribonucleic acid polymerase chain reaction negative at 6 weeks, presented to Princess Mar ina Hospital (the main public referral hospital in Gaborone, Botswana) with a 3 weeks' history of vomiting after feeds. Vomiting was projectile, nonbilious, and nonbloody. It was not associated with diarrhea, but there was a 1-month history of intermittent episodes of passing small amounts of very hard stools every three or more days. There was no history of fever, shortness of breath, or cough.

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He was born at term by uncomplicated spontaneous vaginal delivery with a birth weight of 3355 g and Apgar scores 9, 10, and 10 at 1, 5, and 10 min, respectively. He was not dysmorphic. His mother's Venereal Disease Research Laboratory result was negative at antenatal booking, and he had never received antibiotics.

His mother was HIV infected, and hence, he received single-dose nevirapine after birth followed by zidovudine for 4 weeks. Postnatally, he was a well baby and was discharged from hospital on day 2 of life. He was exclusively formula fed. His child vaccine card showed suboptimal growth between −2 and −3 Z-scores on the World Health Organization weight-for-age growth chart. At 7 months, he had a bout of acute gastroenteritis followed by weight loss, and he then plotted below −3 Z-scores for weight-for-age. This weight decline coincided with developmental regression in language and gross motor milestones. At 6 months, he was reported to have been attempting single words and sitting without support, but from 7-month onward, he could no longer sit or speak any words. His immunizations were up-to-date.

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On physical examination, he was emaciated and irritable. He had no pallor, jaundice, or lymphadenopathy. He had no dysmorphic features or any signs of respiratory distress. His vital signs were axillary temperature 36.5° C, respiratory rate 40/min, and pulse rate 148/min. His anthropometric measurements revealed weight 3.7 kg, length 59 cm, head circumference 39 cm, weight for age −3 Z-score, weight for length −3 Z-score, and length for age −3 Z-score.

He had a soft, grossly distended abdomen. There were no distended veins, scars, or hepatosplenomegaly. Bowel sounds were present but reduced. There was no palpable mass in the epigastrium. Repeated abdominal examination after feeding revealed a mass in the left upper quadrant that appeared to move to the right in a peristaltic fashion. It measured about 4 cm in diameter approximately. Examination of other systems was unremarkable.

Arterial blood gas showed a metabolic alkalosis with PH 7.49, pCO2 37 mmHg, pO2 89 mmHg, standard bicarbonate 28 mmol/L, and base excess +4.6. Serum urea and electrolytes were essentially normal with sodium 135 mmol/L, potassium 4.5 mmol/L, chloride 94.4 mmol/L, creatinine 20 mmol/L, and urea 4.6 mmol/L. The full blood count was normal with white blood cells 6.9 × 10^3/mL, hemoglobin 11.3 g/dL, mean cell volume 80 fL, and platelets 218 × 10^3/mL.

Erect and supine abdominal X-rays showed a distended gastric bubble with dilated loops of bowel, and a granular pattern suggestive of fecal impaction in the colon. Abdominal ultrasound scan showed a gaseous abdomen with feces in the colon. No other abdominal masses were appreciated.

A barium swallow and meal showed the unrestricted passage of contrast in the esophagus but delayed and restricted passage through the pylorus into the duodenum, suggestive of a partial but significant gastric outlet obstruction [Figures 1 and 2]. We thus entertained the diagnosis of infantile hypertrophic pyloric stenosis (IHPS).

The child underwent a laparotomy. There was a soft band of tissue crossing from the porta hepatis to the transverse colon which was constricting the pylorus. The pylorus was so severely stenosed that a size 8 feeding tube could not pass through the pyloric sphincter. The band of tissue was so tight that the gastric antrum was partly incorporated into the inferior surface of the left lobe of the liver [Figures 3 and 4]. The pylorus and gastric antrum were freed after ligating the constricting band of tissue.

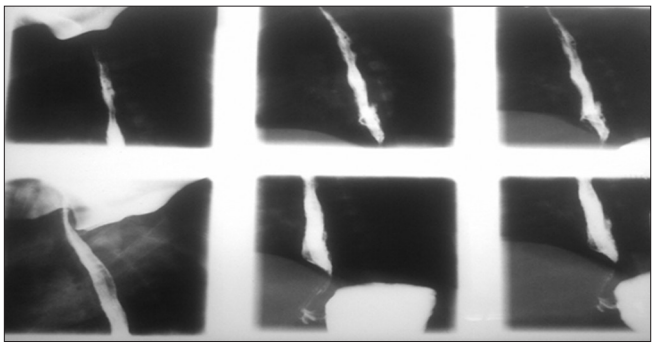

Figure 1: Barium swallow. Note the unlimited flow of barium through the esophagus into the stomach

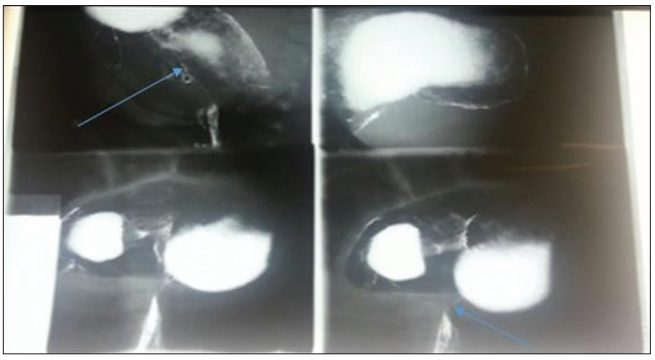

Figure 2: Barium meal. Note the "string sign" (arrows) indicating restricted gastric emptying

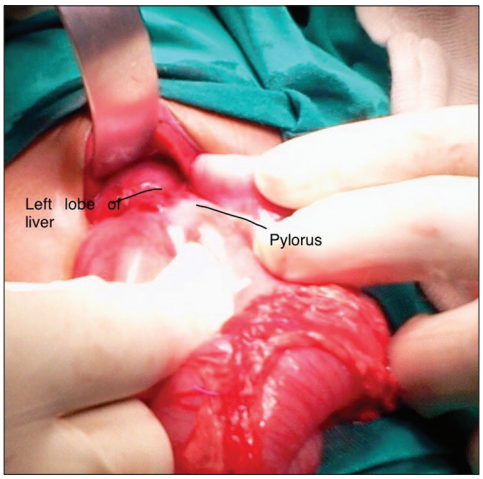

Figure 3: Intraoperative image. Note the firm attachment of the gastric pylorus to the liver

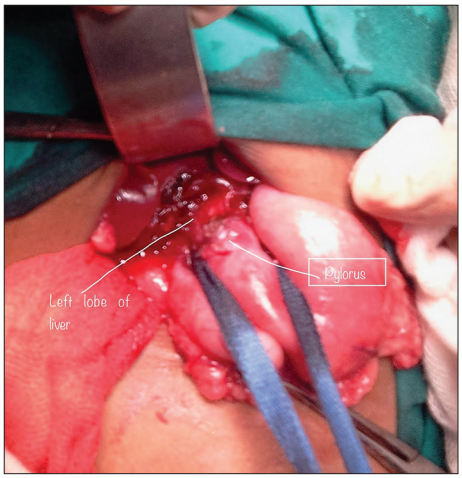

Figure 4: Intraoperative image showing the gastric antrum incorporated into the inferior surface of the left lobe of the liver

Postoperatively, his recovery was uneventful. He was later successfully followed up in our nutritional rehabilitation unit until the malnutrition resolved.

Discussion

IHPS is the most common cause of nonbilious projectile vomiting in infants although a similar presentation has been reported with gastroenteritis, gastroesophageal reflux disease, hiatal hernia, adrenal insufficiency, and inborn errors of metabolism.

In IHPS, there is hyperplasia of the smooth muscle fibers in the antropyloric portion of the stomach resulting in thickening and narrowing of the pyloric canal, with subsequent obstruction to gastric emptying. This condition is typically diagnosed between the 3rd and 8th weeks of life and the majority of cases occur in children under 6 months of age. Of note, males are more affected than females, and there may be preponderance toward firstborn males.[1]

The initial presentation is usually nonbilious, projectile vomiting. Vomiting characteristically occurs at 2–8 weeks of life, often noticed about 10–30 min after feeding.[5] The affected infant is usually very hungry after a vomiting episode, and this forms a vicious cycle of eating, vomiting, and wasting. Due to the wasting, the infant tends to have active visible peristaltic waves due to a thin abdominal wall. Constipation may also be noted.

A palpable olive-shaped mass is pathognomonic for IHPS on physical examination. Although it is not so easy to elicit, it has a positive predictive value of 85%–100%. It may be argued that the accuracy of abdominal examination is dependent on the experience of the examiner, the calmness of the infant and the presence of gastric distention.[1]

Metabolic alkalosis and hypochloremia are common findings in IHPS and have been shown to be independent predictors of IHPS. These metabolic derangements must be corrected before pyloromyotomy, the definitive corrective surgery for IHPS, is done.

Other investigations that can aid in the diagnosis of IHPS are abdominal sonography, barium studies and abdominal X-ray. On sonography, there is often a thickened prepyloric antrum bridging the duodenal bulb and variable degrees of gastric distension. Studies have demonstrated that a sonographic diagnosis of IHPS can be made reliably if the pylorus is elongated to more than 15 mm or thickened to more than 3 mm, with a high degree of accuracy, suggesting that bedside sonography is an acceptable rule-in test.[6] Barium swallow/meal is another useful test for diagnosing IHPS. Characteristically, IHPS is diagnosed when the pyloric canal is outlined by a restricted string of contrast material coursing through the mucosal interstices (string sign) or by several linear tracts of contrast material separated by the intervening mucosa (double-track sign) suggesting that there is a filling defect. Its sensitivity has been shown to be around 95% and is more affordable than endoscopy,[1] which has been proposed as the most expeditious and accurate method for diagnosing IHPS. However, it is very expensive and is therefore rarely used.

The clinical presentation of our patient was identical to that of IHPS. For instance, he is a boy, had a history of postprandial projectile nonbilious vomiting and intermittent constipation with marked wasting, had a hypochloremic metabolic alkalosis and had visible peristaltic waves per abdomen although no abdominal mass was visualized on abdominal ultrasound. Furthermore, his abdominal X-ray and barium studies were very typical. However, the late onset of symptoms and signs from the age of 7 months made IHPS less likely although late IHPS has been reported in two boys aged 8 years and 14 years.[5,7]

Most causes of pyloric stenosis are, by far, intrinsic with IHPS comprising the majority of the cases. Developmental abnormalities of the pyloroduodenal canal in the form of duplication cysts causing intrinsic pyloric stenosis have also been reported to mimic IHPS in children.[2]

Idiopathic cases of intrinsic pyloric stenosis, that is, cases without any muscular hypertrophy (Jodhpur disease) have also been reported.[3]

Extrinsic obstruction of the pylorus mimicking IHPS is very rare in children. Postoperative extrinsic pyloric stenosis resulting from surgical adhesions binding the pyloroduodenal region to the under surface of the liver are possible.[2] Extrinsic obstruction of the duodenum and the pylorus by a gallstone in the presence of a cholecystoduodenal fistula (Bouveret's syndrome) has been described in adults.[4] This is the first time that a band of tissue crossing from the porta hepatis to the transverse colon has been reported to cause extrinsic pyloric obstruction and mimic IHPS.

We recommend that there should be a high index of suspicion for causes other than IHPS in all children who present with nonbilious, projectile vomiting after the age of 6 months.

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Conflicts of interest

There are no conflicts of interest.

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