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A rare case of recurrent hypertrophic pyloric stenosis☆

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ABSTRACT

Hypertrophic pyloric stenosis (HPS) is perhaps the most commonly encountered gastrointestinal disease process in the pediatric surgical patient. While the etiology remains unclear, progression of the disease is well described and uniformly results in non-bilious emesis, weight loss and metabolic derangement. Current management of HPS is the Ramstedt procedure; complications are minimal and the procedure is curative. True recurrence of HPS is rare, and usually mistaken for an incomplete myotomy. We present here a case of recurrent pyloric stenosis after complete myotomy.

We present here a case of true, recurrent pyloric stenosis. This is relatively rare and can usually be attributed to an incomplete myotomy at the time of initial surgical intervention. In reviewing this patients history we believe that a complete myotomy was performed for pyloric stenosis. Subsequent to this, our patient had a recurrence of his disease requiring additional surgical management.

Hypertrophic pyloric stenosis (HPS) is the most commonly encountered surgical disease among pediatric patients. Incidence of HPS varies with geography, but can be estimated at 2–3 in every 1000 live births [1]. Onset is typically between two and eight weeks of life. The classic presentation includes feeding intolerance with projectile, non-bilious emesis [1]. Diagnostic work up usually reveals a hypokalemic, hypochloremic metabolic alkalosis [2]. Ultrasound or fluoroscopic upper gastrointestinal series can be used to facilitate diagnosis when the clinical picture and physical exam are equivocal.

The current standard of care for management of HPS is a surgical pyloromyotomy. Initially described by Ramstedt in 1912 [3], this procedure is associated with a high curative rate and minimal post-operative morbidity. Of note, while incomplete pyloromyotomy is a well documented post-operative complication, there are very few cases of true recurrent pyloric stenosis. A recent, 30 year review of all HPS cases at a children's hospital identified only two cases of recurrent pyloric stenosis [4].

1. Case report

Our patient is an African American, male infant born at thirty-nine weeks via C-section, who initially presented at two weeks of life with a two day history of non-bilious, non-bloody emesis after each feed. The patient was evaluated at an outside institution where an abdominal ultrasound demonstrated a 4.7 mm thick pyloric wall with a 20 mm channel length. Weight at time of admission was 3.4 kg. Following transfer the patient was resuscitated with intravenous fluids and taken to the operating room where a laparoscopic pyloromyotomy was performed without complication. Complete repair was achieved after incising the serosa from the proximal pyloroduodenal junction to the gastric fibers and down to the duodenal mucosa. The muscle was spread, subsequently exposing the mucosa. Both sides of the incised muscle rocked independently [5]. The postoperative course was relatively uncomplicated; the patient tolerated Pedialyte, then formula per hospital protocol and was discharged on post-operative day one.

Five weeks after discharge from our institution the patient returned with a one week history of worsening emesis; non-bilious, non-bloody and projectile in nature. During this time the patient was seen by his primary care doctor who initiated Zantac for presumed reflux disease without success. The patient did not return to surgery clinic for a postoperative evaluation. It is important to note that per the patient's mother the child had gained weight (from 3.4 kg to approximately 4.5 kg) between the first pyloromyotomy and readmission. At the time of readmission the patient weighed

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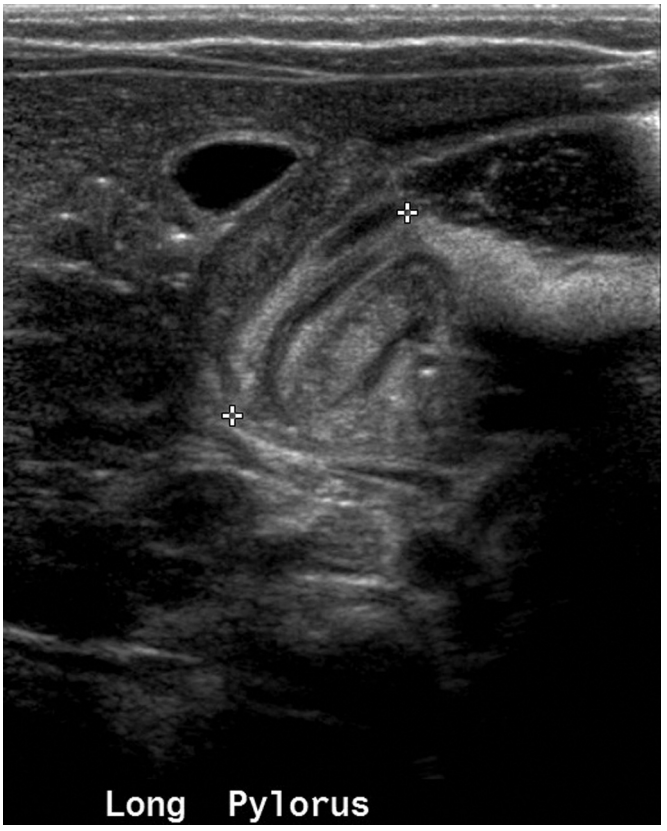

Fig. 1. Pylorus channel – 22 mm.

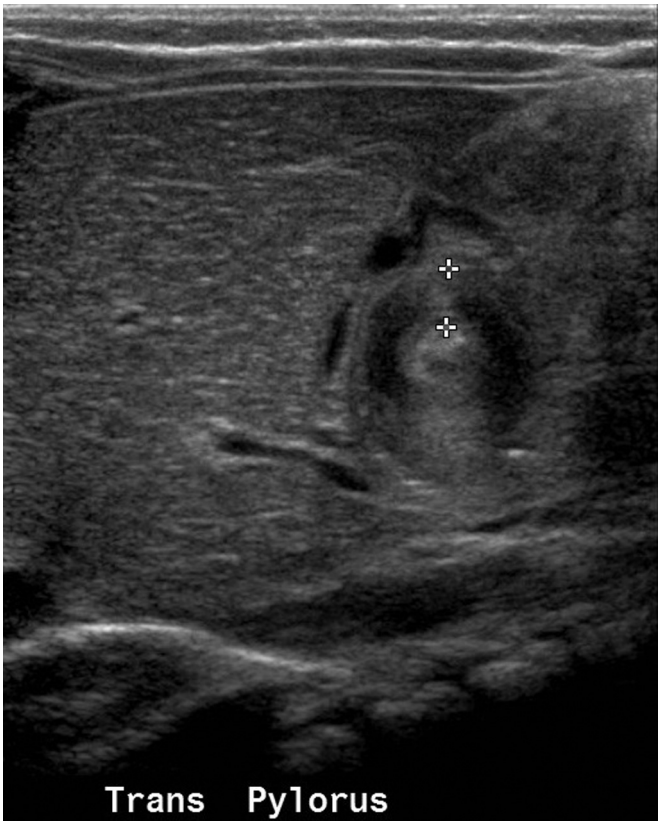

Fig. 2. Pylorus wall thickness – 4.6 mm.

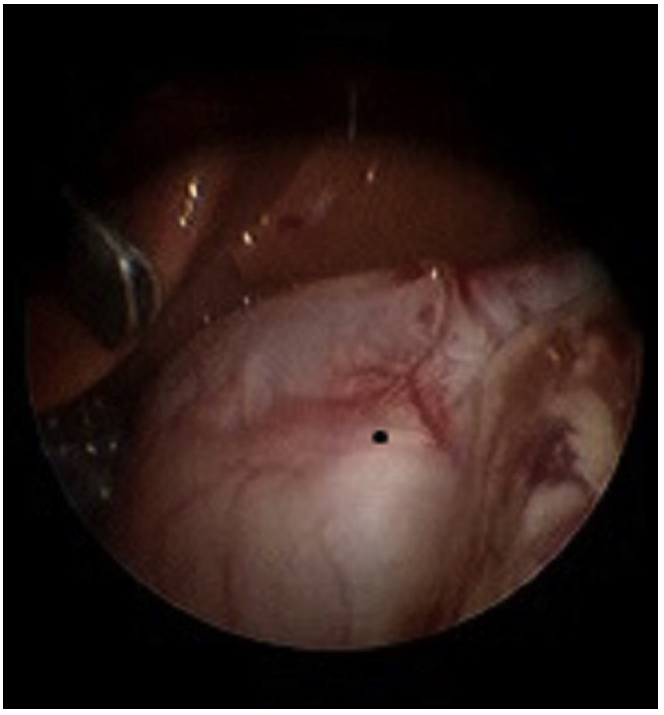

Fig. 3. Well healed anterior scar from prior pyloromyotomy.

4.3 kg. Additionally, the patient had ceased to have episodes of emesis with feeds for an entire month following his operation. This element of the history suggests that this is a true recurrence as opposed to an incomplete pyloromyotomy.

Our patient had an ultrasound at our institution (Figs. 1 and 2) which showed a 4.6 mm thick wall with a 22 mm channel, his metabolic profile was consistent with a hypochloremic, hypokalemic metabolic alkalosis. The patient was taken back to the operating room where laparoscopy revealed an enlarged pylorus with a well healed anterior scar (Fig. 3). The pylorus was rotated caudally

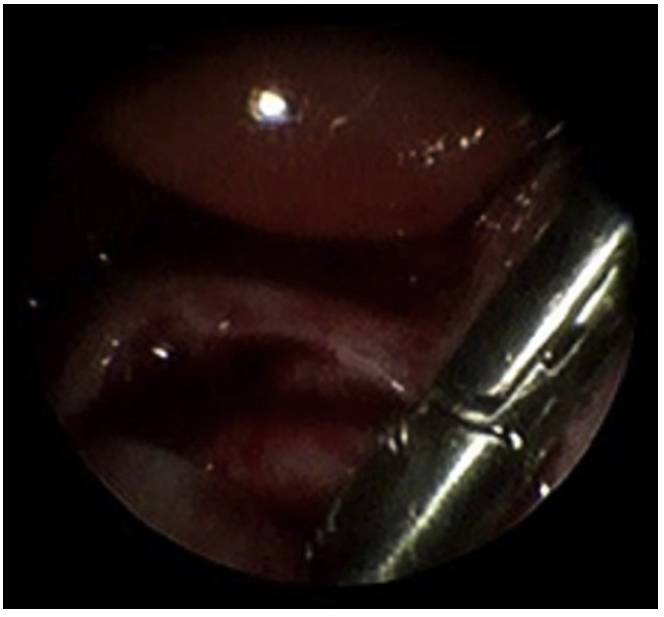

Fig. 4. Posterior pyloromyotomy with bulging mucosa through divided muscle fibers.

and a pyloromyotomy was made through the muscle in a location separate from the well healed anterior scar. Again, both sides of the muscle were rocked independently after spreading the incised tissue and mucosa was exposed (Fig. 4). The patient tolerated the procedure well and was discharged on post-operative day one tolerating 60 mL of formula every 3 h without emesis.

2. Discussion

Pyloromyotomy is the current standard of care for managing HPS. Initially described by Ramstedt [3], the procedure has evolved over the last century with changes in surgical technology. However, in skilled hands the outcome is nearly uniform, resolution of symptoms with little morbidity to the patient. Complication rates, both intraoperative and postoperative, are low and rarely require operative intervention. The most common operative complications include mucosal perforation, wound infection, wound dehiscence, incisional hernia, and persistent emesis. A retrospective study by Kim et al. [6] described no significant difference in the occurrence of these complications when comparing laparoscopic, circumumbilical and right upper quadrant approaches. Complication rates of up to 8% have been reported previously [7]. One series of 901 patients reported a 4% intraoperative and 6% postoperative complication rate. There were three cases of incomplete pyloromyotomy, but no recurrences [8]. True recurrence of HPS is quite rare, defined as complete resolution of symptoms with subsequent weight gain followed by representation with sonographic and/or operative evidence of re-stenosis. This should not be confused with an incomplete myotomy. The presentation of an incomplete myotomy is usually within the immediate post op period and warrants evaluation if the patient has post feed emesis greater than five days post op [8].

A literature search performed by the authors revealed only four documented cases of true recurrent HPS [4,9,10], all of which required surgical or endoscopic intervention. Interestingly, there is some controversy surrounding the diagnosis of recurrent HPS. There is literature to suggest that infants who need a second pyloromyotomy may not have simply a recurrence, but instead suffer from progression of disease [10]. The implication is that some cases of HPS are operated on early in the natural history of the disease process and eventually require a second intervention. Going beyond this, a paper by Rollins et al. [11], challenged whether or not HPS is truly a congenital disease. 1400 infants were evaluated with ultrasound at birth, none of these infants met sonographic criteria for HPS. Nine of the patients evaluated at birth went on to develop HPS requiring operative intervention. Additionally, the most recent Updated National Birth Prevalence Estimates for Selected Birth Defects in the United States does not include HPS, as it occurs after the first day of life [12].

3. Conclusion

In conclusion, this case demonstrates the rare event of a true recurrent pyloric stenosis. It suggests that even after a complete myotomy the process of hypertrophic pyloric stenosis is a dynamic one, and recurrence can in fact happen.

Conflict of interest

The authors have no financial or personal relationships to disclose.

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