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Case Report

# Recurrent Hypertrophic Pyloric Stenosis in a Male Child: A Case Rarity

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## Abstract

Infantile hypertrophic pyloric stenosis (IHPS) is the most frequent surgical condition in children. Incomplete pyloromyotomy is an infrequent complication, but recurrent pyloric stenosis (RPS) is extremely rare. We report here a case of late-onset recurrence of IHPS in a 7-year-old male child, after successful pyloromyotomy. He presented with abdominal pain, recurrent episodes of non-bilious vomiting and upper abdominal fullness after taking meals that relieved on vomiting. These symptoms started around 4 months after successful pyloromyotomy done at 4 weeks of age. Child underwent series of radiological investigations including upper GI endoscopy that confirmed gastric outlet obstruction with thickened pylorus. Multiple management options are proposed but we preferred 'heineke mikulicz pyloroplasty' in our case.

## Introduction

Hypertrophic pyloric stenosis (HPS) is a common cause of gastric outlet obstruction (GOO) in neonates, with an incidence of 1.5 to 3 per 1000 live births. Onset is seen usually between 2 to 8 weeks of life, with classic presentation of feeding intolerance with projectile, non-bilious emesis. Diagnostic work up usually reveals a hypokalemic, hypochloremic metabolic alkalosis. Ultrasound or fluoroscopic upper gastrointestinal series can be used to facilitate diagnosis when the clinical picture and physical exam are equivocal. The etiology of IHPS is not properly understood. Various authors have suggested its congenital or acquired nature [1]. Ramstedt's pyloromyotomy is the current standard of care, with high curative rate and minimal post-operative morbidity. As incomplete pyloromyotomy is a well-documented post-operative complication, there are very few cases of true recurrent PS.

Multiple options are proposed for the management of recurrent PS including pneumatic dilatation or botulinum toxin injection as well as operative management in form of pyloroplasty, Billroth I, Billroth II procedures [2-4]. Here, we present a case of isolated recurrent PS in 7 years old male child following successful management with routine pyloromyotomy.

## Case Report

A 7-year-old male child presented with abdominal pain, recurrent episodes of non-bilious vomiting and upper abdominal fullness after taking meals that relieved on vomiting. Parents also gave history of vomiting of partially digested food material eaten few hours earlier. These symptoms started around 4 months after the child had undergone successful pyloromyotomy at 4 weeks of age. At the time of first presentation during neonatal period, symptoms of repeated episodes of vomiting and upper abdominal fullness immediately after feeding were recognised by parents and child underwent open Ramstedt pyloromyotomy. At home, he tolerated full feeds with no vomiting and gained weight. Child was symptoms free until about 4 months postoperatively. Initially the symptoms were not severe and child had bloating, early satiety, foul-smelling regurgitations and over the following year, child experienced few episodes of infrequent non-bilious vomiting. As per his mother child was gaining weight at good pace until when the frequency of vomitings gradually increased over 4-5 years. Child later developed abdominal pain and recurrent non bilious vomitings after about 20 to 30 minutes of a feed.

During follow-up child was being treated for gastro-esophageal reflux with persistent symptoms. Later, child presented to our emergency. On examination, he had signs of dehydration, with pulse rate of 119/minute, low volume and B.P was 92/48 mmHg. Blood investigations revealed Hb 9.3g/dl, TLC 7250/dl, Platelet count 3.59L/$mm^3$, HCT 41%, protein 4.8g/dl, serum albumin 2.3g/dl, blood urea 21 mg/dl, serum creatinine 0.9mg/dl, $Na^+$ 129meq/L, $K^+$ 3.1meq/L & $Cl^-$ 95meq/L. Venous blood gas (VBG) analysis showed pH 7.47, $pCo_2$ 34, $pO_2$ 59, $Hco_3$ 29. Weight of the child at presentation was less than 3rd Centile on growth chart. Child was managed with fluid resuscitation, and blood investigation revealed correction of dehydration as manifested by normalisation of hematocrit to 36% and VBG pH 7.36, pCo2 31, pO2 66, Hco3 24. Child underwent series of radiological investigations that included USG abdomen, which showed dilated stomach and contrast enhanced CT scan with oral contrast confirmed grossly dilated stomach extending inferiorly to the level of iliac fossa and displaced the bowel loops posteriorly. On successive sections there was circumferential thickening of distal stomach. All these features pointed towards GOO because of PS (Figure 1).

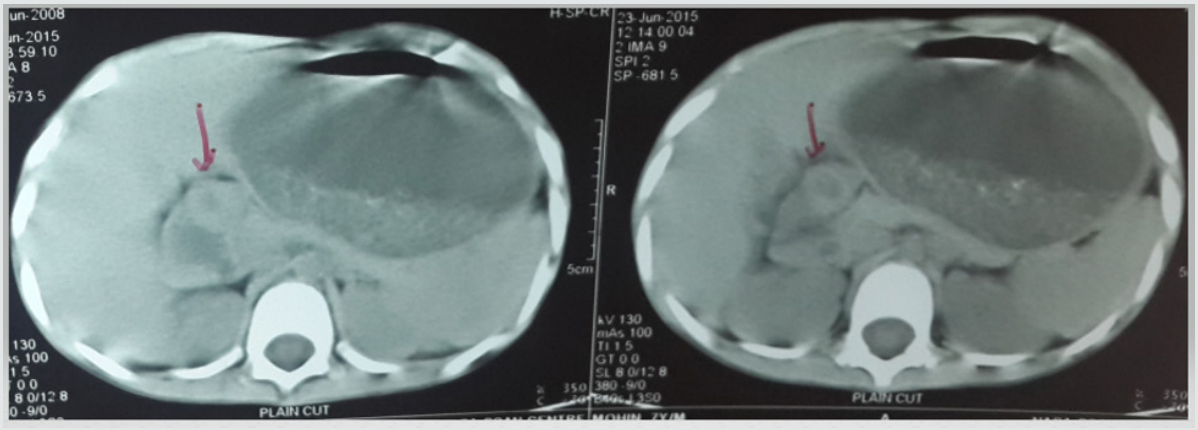


**Figure 1.**

Upper GI endoscopy showed multiple ulcerations with friability at the lower end of esophagus. Stomach was dilated with food residue and normal fundus, body and antral regions but pyloric opening couldn't be negotiated. After workup child was planned for surgery. Intra-operative findings were dilated stomach and thickened pylorus. We performed 'heineke mikulicz pyloroplasty' involving transversely closed longitudinal incision across the pylorus. The nasogastric tube was removed on 3rd post-operative day (POD) and child was allowed liquids on 4th POD with soft diet on 5th POD (Figure 2).

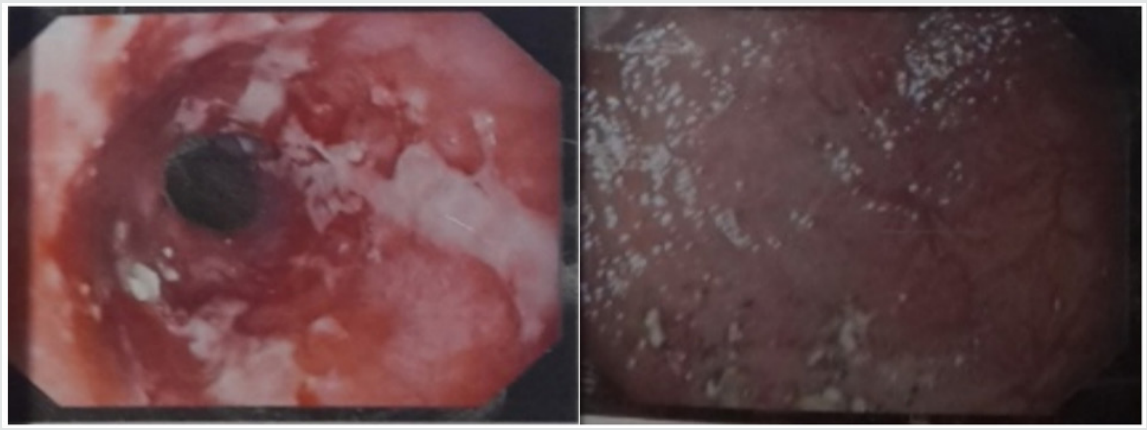

**Figure 2.**

## Outcome and follow-up

He was discharged on POD-7 on soft diet and tab Lanzol 15mg DT for 2 weeks. At his 2-week post-operative visit, he was asymptomatic and tolerated feedings without bloating or emesis. The histopathological examination was consistent with HPS. He remained completely asymptomatic at 1-year follow-up.

## Discussion

Ramstedt's pyloromyotomy (PM) remains the gold standard treatment for IHPS [5]. Emesis is a frequent postoperative complaint after pyloromyotomy and it occurs in 36–90%of cases [4]. Usually vomiting stops spontaneously after few days. Incomplete pyloromyotomy is suspected when vomiting persists and lasts of

more than 5 days. There is a criterion to differentiate recurrent PS from incomplete pyloromyotomy which includes [6];

a. The patient must have resolution of symptoms for duration of at least three weeks postoperatively.

b. The patient must demonstrate weight gain.

c. Evidence of restenosis must be confirmed with imaging or on exploration (all these conditions are consistent with our case).

Huang et al. [7] showed that the immediate postoperative resolution of emesis after a successful pyloromyotomy is from an increase in the diameter of the intramuscular pyloric canal rather than an immediate regression in the thickness or length of the pyloric muscle. In incomplete pyloromyotomy pyloric canal diameter will not be seen to increase in diameter on USG who presented with repeated vomiting in immediate post-operative period. True recurrence of HPS is quite rare with only a few case reports in literature.

The etiology of recurrent PS, as IHPS itself, is unclear. It appears that pathological evolution of HPS as well as recurrent PS as a true surgical entity is similar and separate from incomplete PM. It has been suggested that the process which drives hypertrophy of the pylorus is in still in evolution when initial pyloromyotomy was performed which after initial symptom free interval progressed to true recurrent PS [3,8]. In any case of recurrent PS, initial management is conservative with bowel rest and nasogastric decompression with trial periods ranging from 7 to 21 days. The medical management using atropine, botulinum toxin injection and surgical management including pyloroplasty, Billroth I, Billroth II procedures, and pneumatic balloon dilatation has been proposed [2-4]. We did 'heineke mikulicz pyloroplasty' in our case because of its ease and simplicity with preservation of normal anatomical tract and less complications.

A close differential diagnosis of Jodhpur disease (JD) needs mention. It presents as primary acquired gastric outlet obstruction in infancy and childhood, with very similar presentation to our case. Jodhpur disease has predilection for male sex [9]. There are certain differentiating features between these two entities [10]. A) Ultrasonography shows normal pyloric canal with no pyloric muscle hypertrophy in Jodhpur disease (JD) but shows narrow pyloric canal with pyloric muscle hypertrophy in HPS. B) UGIE in JD shows no intra-luminal pathology with normal gastric mucosal rugosities in a large-sized stomach while in HPS Antral folds hypertrophy with dilated stomach. C) On histopathological examination (HPE), Jodhpur disease shows normal cellular pattern of all coats without inflammatory and fibro-proliferative nature [9]. HPE was consistent with HPS in our case. It showed marked congestion of pyloric mucosa. Muscularis mucosa and muscularis propria showed hypertrophy and hyperplasia of muscle bundles along with haphazardly directed muscle bundles with interspersed fibro-collagenous tissue.

## Conclusion

True Recurrent PS is a rare condition with unclear etiology. There may be an increasing incidence due to early diagnosis with modern investigations. Utilizing the evidences & criteria available for diagnosis, we believe pyloroplasty seems to be the most effective and safest intervention for recurrent PS.

## Conflicting Interest

Nil

## Acknowledgement

Patient was managed at Maulana Azad Medical college, Lok Nayak Hospital.

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