IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 17, Issue 2 Ver. 13 February. (2018), PP 65-66
www.iosrjournals.org

Jodhpur disease (Primary acquired gastric outlet obstruction in infancy and childhood): A rare case

Tayal N[1], Kadian YS[2],Ahari P[3], Kadian A[4]
[1, 3](Department of General Surgery, PGIMS Rohtak)
[2](Department of Paediatric Surgery, PGIMS Rohtak)

Abstract: *Jodhpur disease (Primary acquired gastric outlet obstruction in infancy and childhood) is a rare clinical entity causing symptoms of gastric outlet obstruction. This disease was first described in 1997 in Jodhpur, India. We present a case report of a 2-year old male child presented to us with recurrent non-bilious vomiting and upper abdominal distension after meals. Patient was operated and hypertrophic mucosa and sub-mucosa were found for which Hienke-Mikuliczpyloroplasty was done. Postoperative period was uneventful and patient was discharged under satisfactory conditions. Jodhpur disease is a rare entity with presentation as gastric outlet obstruction. It is a curable disease and management is essentially surgical and Heineke-Mikuliczpyloroplasty has excellent results.*

Keywords: *Jodhpur Disease, Primary acquired gastric outlet obstruction in infancy and childhood, Hienke-Mikuliczpyloroplasty*

Date of Submission: 14-02-2018 Date of acceptance: 28-02-2018

I. Introduction

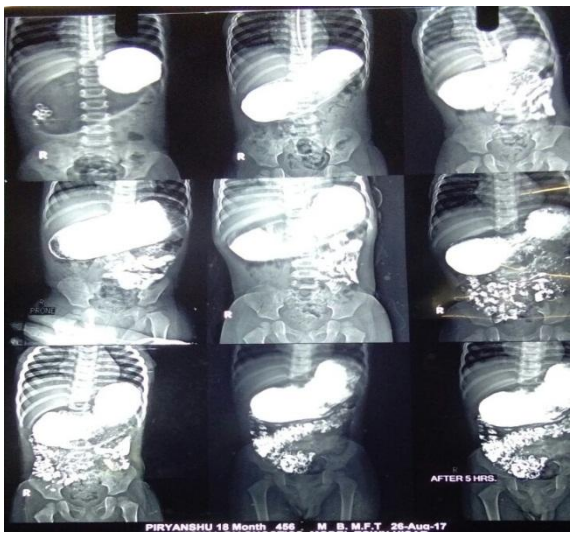
Gastric outlet obstruction is a common cause of recurrent projectile non-bilious vomiting in children. Most common cause is infantile hypertrophic pyloric stenosis. Jodhpur disease is another rare clinical entity causing symptoms of gastric outlet obstruction. This disease was first described in 1997 in Jodhpur, India. A very few cases have been reported since then.

II. Case report

A 2-year old male child presented in outpatient department with recurrent vomiting and failure to gain weight. The child was admitted and further worked-up for the underlying cause. The patient had history of vomiting for past six months which was characteristically non bilious. The patient fed less and failed to gain weight in past six months. He had history of abdominal distension following meals in upper abdomen.

The child was born at 39+2 weeks by a normal vaginal delivery with no delay in crying and no immediate complication. He was vaccinated in time for BCG, DPT, Hepatitis B and Measles. The milestones of growth were normal. There was no history of recurrent vomiting in past. There was no history of jaundice, pneumonia, breath holding spells, seizures, previous surgery or any contact with tuberculosis in family.

The blood investigations revealed hemoglobin 10g/dL, bleeding time – 1’45’’, clotting time – 3’45’’, blood urea –24mg/dL, random blood sugar level – 69mg/dL, serum sodium – 140mEq/dLand serum potassium – 3.5mEq/dL. His barium meal study showed grossly distended stomach with poor passage of barium distally and delayed emptying (Fig. 1). A diagnosis of gastric outlet obstruction was made and a decision was made to operate the patient.A transverse incision was made over right subcostal region extending beyond the midline. Pylorus was identified and opened in longitudinal manner. Hypertrophic mucosa and sub-mucosa were almost completely obliterating the lumen in pyloric region(Fig. 2). Excess of mucosa and sub-mucosa were excised and sent for histopathological examination. Pyloric muscle was normal.Hienke-Mikuliczpyloroplastywas done using vicryl 3-0. The abdomen was closed in layers. The HPE of the specimen sent showed mucosal and sub-mucosal congestion.



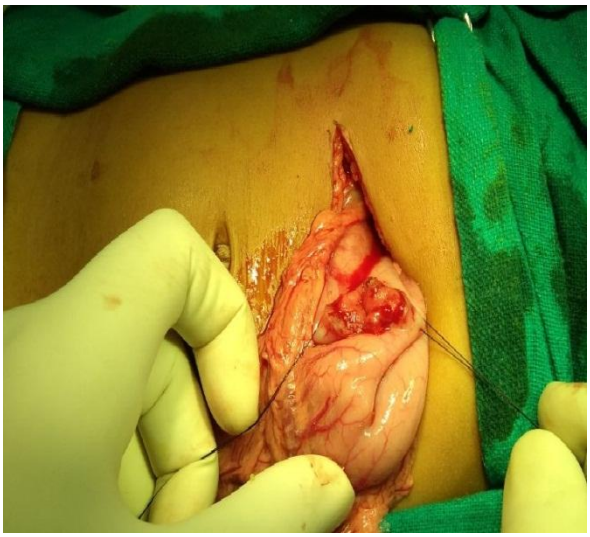


fig. 1Showing grossly distended stomach in barium study of the patient **fig. 2 showing hypertrophied mucosa and submucosa**

In postoperative period, vitals of patient were normal. The patient remained stable and passed stool and flatus on 3rd postoperative day when the nasogastric tube was taken out and he was allowed sips of clear fluid orally. The oral intake of patient was increased gradually to semisolids and solids as he tolerated it. He was discharged on 6th postoperative day under satisfactory condition.

III. Discussion

Gastric outlet obstruction in infancy and childhood was first described in India in 1997 in Jodhpur and was nicknamed 'Jodhpur disease' [1]. Recently it has been renamed as "primary acquired GOO in infancy and childhood". It is a rare entity with incidence of 1: 100,000 and predilection for males. Mean age of presentation is 2.9 years (range 1 month – 6 years) [2]. In this condition, pyloricmuscle remains absolutely normal.The exact etiology is still unknown. Most of the cases have been reported from northern India (Jodhpur), so dietary factors may be contributing to this condition. Various hypotheses have been put forward. One states that lack of nitric oxide synthase in neurons leads to failure to relax pyloric smooth muscle causing neuromuscular incoordination and later GOO, while another claims that the cause is an abnormality of the interstitial cells of Cajal that act as electrical pacemakers of GI tract smooth muscles [3]. Management is essentially surgical with "Heineke-Mikuliczpyloroplasty" as the procedure of choice with excellent outcome [2]. Pneumatic dilatation by balloon inflation via upper endoscopy has also been tried [4].

IV. Conclusion

Jodhpur disease is a rare entity with presentation as gastric outlet obstruction. A high degree of clinical suspicion should be kept in mind in preschool children presenting with recurrent non-bilious vomiting.It is a curable disease and management is essentially surgical and Heineke-Mikuliczpyloroplasty has excellent results.

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Tayal N "Jodhpur disease (Primary acquired gastric outlet obstruction in infancy and childhood): A rare case.". "IOSR Journal of Dental and Medical Sciences (IOSR-JDMS), Volume 17, Issue 2 (2018), PP 65-66.