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Case Report

Surgery Section

Laparoscopic Pyloroplasty for Jodhpur Disease

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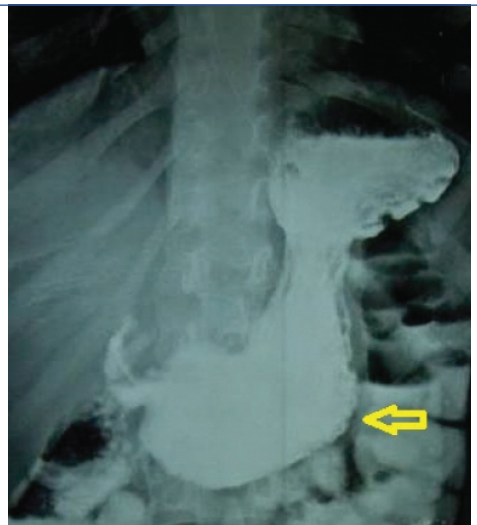
ABSTRACT

Jodhpur disease is the name given to acquire Gastric Outlet Obstruction (GOO) of idiopathic origin. Persistent non bilious vomiting, pain and occasional distension of upper abdomen are the usual clinical presentation. This condition responds well to Henieke-Mikulicz' pyloroplasty; however, laparoscopic management of Jodhpur disease is yet to become a widely accepted modality of treatment. Herein, we report a case of a three-year-old male child, diagnosed with Jodhpur disease who was managed successfully with three port laparoscopic Henieke-Mikulicz' pyloroplasty.

Keywords: Gastric outlet obstruction, Paediatrics, Vomiting

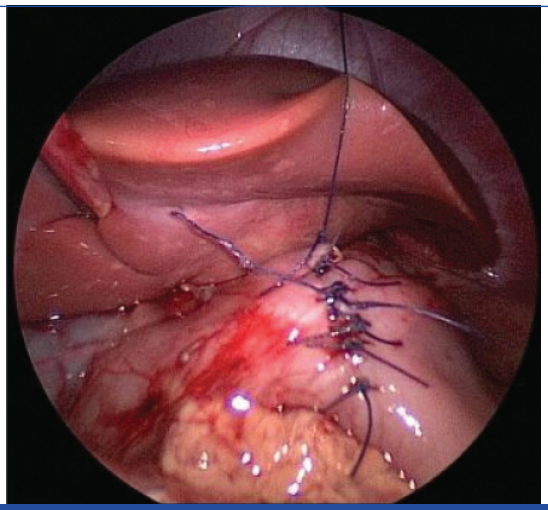
CASE REPORT

A three-year-old male child was brought to paediatric surgery outpatient department with history of postprandial upper abdominal distension and recurrent non bilious postprandial vomiting for last two months. There was a history of significant weight loss. There was no episode of gastrointestinal bleeding and history of caustic ingestion. During this period, the patient was treated with prokinetics and antacids without any improvement of symptoms as narrated by the parents. On examination, the patient was dehydrated and emaciated. There was epigastric distension with visible peristalsis. Patient was admitted and resuscitated. Ultrasonography of abdomen did not reveal any abnormal findings. Upper GI contrast study was done which demonstrated dilated stomach with delayed passage of dye through the pylorus [Table/Fig-1]. Upper GI endoscopy revealed a dilated stomach and narrow pylorus not allowing passage endoscope into the duodenum. There was no membrane and ulceration in the antropyloric region.



[Table/Fig-1]: Upper GI contrast study demonstrating dilated stomach.

Patient underwent diagnostic laparoscopy with a 30° angled telescope through a 5-mm optic port at the umbilicus. Stomach was found to be dilated but there was no evidence of any pyloric hypertrophy or extrinsic compression of the pylorus. Decision to perform Henieke-Mikulicz' pyloroplasty was taken. Two other 3 mm working ports were inserted along the right and left midclavicular line above the level of umbilicus. Longitudinal pyloromyotomy was done extending from the duodenal edge to the anrtum with monopolar hook diathermy. Mucosa was opened. The longitudinal pyloroplasty incision was then closed transversely with 3-0 Polyglactin 910 suture. After tying the first upper corner knot the suture was brought out through the anterior abdominal wall and was kept in traction which helped us to alleviate the need of liver retraction and made pyloroplasty incision closure easy [Table/Fig-2]. Single layer closure was accomplished with intermittent full thickness bites followed by intracorporeal knotting. Air insufflation was done through nasogastric tube to ascertain the integrity of suture line. Ports were removed under vision. Umbilical port incision was closed with 3-0 Polyglactin 910. Nasogastric tube was removed on fourth postoperative day and oral liquid diet was started on the next day. The patient was discharged after one week. He was revaluated at outpatient clinic after 15 days and was found to be healthy and gained one kg weight. Presently after six months, the patient is fine and attained the growth indices as per his age.



[Table/Fig-2]: Laparoscopic pyloroplasty near completion with uppermost corner suture suspended to abdominal wall.

DISCUSSION

Gastric Outlet Obstruction (GOO) during infancy and childhood can be due to rare congenital causes like aplasia, atresia, diaphragms and webs in the pylorus or anrtum or it can be due to common condition like idiopathic hypertrophic pyloric stenosis. GOO is mostly acquired secondary to chemical injury or scarring due to acid peptic disease [1]. There is another group of GOO without any obvious extrinsic, intrinsic or anatomical aetiology that was classified by Sharma KK et al., as Jodhpur disease [2]. Sharma KK et al., reported five cases of GOO between the age ranges of three months to six years [3]. They did not find any

aetiological factor for the disease. They propose that some form of neuromuscular inco-ordination due to unspecified agent may have caused the permanent changes in the pylorus. All diagnosed patients responded well to Henieke-Mikulicz' pyloroplasty. Sharma KK et al., modified the earlier classification for GOO and grouped these patients into a new disease entity called as acquired primary GOO [3]. In a subsequent re-modification in 2008 Sharma KK et al., coined the term Jodhpur disease for acquired GOO as abnormally high number of cases including the first case was reported by his centre at Jodhpur [2].

The usual presentation of Jodhpur disease is persistent non bilious vomiting. There can be associated pain and occasional distension of upper abdomen. Diagnosis is based on clinical history and examination supported by some radiological investigations. Ultrasound of abdomen shows dilated stomach with normal sized pyloric canal. Upper GI contrast study demonstrates dilated stomach with delayed passage of dye through the pylorus. Upper GI endoscopy shows narrowed pyloric canal. Though various hypotheses have been proposed, the exact aetiology of this condition is still not known. Histopathology examination of the pyloric area shows a normal number of ganglion cells without any inflammatory, fibro proliferative or neoplastic cells. We classified our case as Jodhpur disease as there was no extrinsic cause of pyloric obstruction, pylorus was not hypertrophied, and endoscopy showed narrowed pylorus with failure to negotiate the scope through the pylorus. Management of this condition is essentially surgical [2-4] though some cases have been reported with endoscopic management [5,6] of the condition. We preferred pyloroplasty as it is the most widely accepted option for the surgical treatment of GOO. Endoscopic management of primary acquired GOO is reported in a few cases but it is yet to be a widely accepted method of treatment for Jodhpur disease [5,6]. Pyloroplasty is also a treatment option for failed endoscopic management of primary acquired GOO [7]. Laparoscopy provides benefits of minimal access surgery to the treatment of Jodhpur disease with same outcome as that of open surgery. Jiménez-Méndez MG et al., were the first to report laparoscopic management of Jodhpur disease by Henieke-Mikulicz' pyloroplasty in Spanish literature [8]. They used four ports for the procedure with one port dedicated for liver retraction while we had completed the procedure with only three ports. From the technical standpoint, utilising the uppermost corner suture of pyloroplasty and suspending it from the anterior abdominal wall helps in vertical alignment of the incision for comfortable intracorporeal suturing without a need for liver retraction as in our case. The index case is the youngest reported patient to have undergone laparoscopic pyloroplasty for successful management of Jodhpur disease in English literature.

CONCLUSION

Henieke-Mikulicz' pyloroplasty continues to remain the treatment of choice for Jodhpur disease. Laparoscopic approach is as safe and effective as open surgery and should be the preferred approach for the management of this disease.

REFERENCES

[1] Feng J, Gu W, Li M. Rare causes of gastric outlet obstruction in children. Pediatr Surg Int. 2005;21:635-40.
[2] Sharma KK, Ranka P, Goyal P, Dabi DR. Gastric outlet obstruction in children: An overview with report of Jodhpur disease and Sharma's classification. J Pediatr Surg. 2008;43(10):1891-97.
[3] Sharma KK, Agrawal P, Toshniwal H. Acquired gastric outlet obstruction during infancy and childhood: a report of five unusual cases. J Pediatr Surg. 1997;32(6):928-30.
[4] Aggarwal M, Sood V, Kumar A, Saurabh K. Jodhpur disease revisited: a rare cause of severe protein energy malnutrition. Ann Gastroenterol. 2012;25(3):262-64.
[5] Lin JY, Lee ZF, Yen YC, Chang YT. Pneumatic dilation in treatment of late-onset primary gastric outlet obstruction in childhood. J Pediatr Surg. 2007;42(2):e1-4.
[6] Karnsakul W, Cannon ML, Gillespie S, Vaughan R. Idiopathic non-hypertrophic pyloric stenosis in an infant successfully treated via endoscopic approach. World J Gastrointest Endosc. 2010;2(12):413-16.
[7] Ceccanti S, Mele E, Frediani S, Di Nardo G, Roggini M, Cozzi DA. Laparoscopic pyloroplasty for idiopathic non-hypertrophic pyloric stenosis in a child. J Pediatr Surg. 2012;47(10):1955-58.
[8] Jiménez-Méndez MG, Carmona-Barba R. Jodhpur disease resolved by laparoscopic pyloroplasty. Acta Med Costarric. 2015;57(1):38-40.

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