CASE REPORT

Hypertrophic pyloric stenosis in an older child: a rare presentation with successful standard surgical management

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SUMMARY

Infantile hypertrophic pyloric stenosis is a disease of neonatal period usually manifest between the third and fourth weeks of life. Metabolic alkalosis and paradoxical aciduria are two common sequel of this entity. We report an unusual case of an 8-year-old boy who presented with recurrent, long-standing episodes of non-bilious vomiting and poor weight gain without any other metabolic derangement.

BACKGROUND

Gastric outlet obstruction (GOO) in older children is a broad spectrum condition that prevents the passage of gastric contents into the duodenum. Persistent non-bilious vomiting results in dehydration and electrolyte imbalances. The correct diagnosis of GOO can be achieved by clinical presentation with appropriate imaging, leading to optimal and timely patient management. Pylorus in these cases is usually normal. Our case was different from this entity because there were no electrolyte imbalance, no visible peristalsis and presence of thickened pylorus.

CASE PRESENTATION

An 8-year-old boy was admitted to the paediatric medicine department with long-standing multiple episodes of non-bilious vomiting along with poor weight gain and stunted growth for last 2 years. Working diagnosis at the time of admission was reflux gastritis.

INVESTIGATIONS

The child underwent multiple investigations including arterial blood gas analysis, haematological, biochemical, urine, stool examination, X-ray and ultrasound of the abdomen. On upper gastrointestinal endoscopy, it was found that the pylorus was completely occluded and scope could not be negotiated across the pylorus. Our department was consulted to assess this case to rule out any surgical pathology. A contrast-enhanced CT of the abdomen was performed to rule out any mass lesion causing extrinsic compression of the pylorus. No mass lesion was found but there was concentric hypertrophy of the pylorus with hardly any lumen (figure 1). Contrast swallow was also performed, which showed dilated stomach with no contrast going across the pylorus even for 2 h delayed film (figure 2). On clinical examination, the child was

Figure 1 Contrast-enhanced CT showing dilated stomach and concentric hypertrophy of pylorus.

otherwise normal; abdomen was soft non-tender, no visible peristalsis. On the basis of available investigation and clinical feature, a diagnosis of GOO was made and laparotomy was planned.

DIFFERENTIAL DIAGNOSIS

- ▶ Gastric volvulus
- ▶ Gastric antral web
- ▶ Gastric duplication cyst
- ▶ Gastric polyps
- ▶ Neoplasm
- ▶ Crohn disease
- ▶ Malrotation and midgut volvulus
- ▶ Jodhpur disease

Figure 2 Contrast swallow showing dilated stomach with no contrast seen distally.

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Figure 3 Intraoperative finding of hypertrophic pyloric stenosis.

TREATMENT

Intraoperatively the pylorus was found to be of normal outline, but thickened for about 3 cm in length (figure 3). On opening the pylorus the mucosal lining was normal. The wall thickness of the pylorus was about 5 mm. The patency of lumen was confirmed proximally and distally. Heinke-Miculicz pyloroplasty (HMP) was performed with a covering gastrostomy for drainage of the stomach and a feeding jejunostomy for starting feeds early. A full thickness biopsy was also taken from the wall of the pylorus and sent for histopathological examination.

OUTCOME AND FOLLOW-UP

Postoperative period was uneventful and the child recovered well. A contrast swallow was performed on postoperative day 7 which showed good pyloric canal with free flow of contrast into duodenum. Oral feeds were started on day-7 and gastrostomy and feeding jejunostomy tubes were kept clamped and removed after 3 weeks. The biopsy report was hypertrophied and irregularly arranged muscle fibres in the pylorus. The child presently is in regular follow-up and is accepting food normally and there is no vomiting or regurgitation.

DISCUSSION

Infantile hypertrophic pyloric stenosis (IHPS) is a disorder of the neonatal period described with the other causes of GOO. The reported incidence of GOO is 1.5–3/1000 live-births.¹ However, GOO without IHPS is a rare entity with incidence of 1:100 000 live-births.² Pathologically, GOO has been classified as congenital or acquired. Congenital causes include duodenal atresia, duodenal web, annular pancreas, pyloric web, pyloric atresia, malrotation, etc, while acquired group include peptic ulcer disease, neoplasia and acid ingestion.^{3–10} There is another entity, which is known as acquired obstruction during infancy and childhood that is Jodhpur disease first reported in 1997 by Sharma *et al.*¹¹ Various other authors have reported similar cases from all over the globe.^{12–16} Our case was different from this entity because there were no electrolyte imbalance, no visible peristalsis and presence of thickened pylorus. Histologically the pylorus was found to be hypertrophied a finding not described for Jodhpur disease until now. Our approach was different from that described for Jodhpur disease. Instead of doing only HMP, we did HMP along with gastrostomy and feeding jejunostomy. Reason for doing feeding

jejunostomy was to start early feed and gastrostomy to provide external drainage for gastric content just to preserve the precarious anastomosis. Regarding the final diagnosis we can conclude that this case might be the modified part of the spectrum of diseases called as acquired GOO during infancy and childhood, also called as Jodhpur disease. We recommend that the spectrum of Jodhpur disease should be widened to encompass cases like this in the near future.

Learning points

- ▶ Surgical causes should be kept in mind while working up a child with recurrent vomiting and malnutrition.
- ▶ Hypertrophic pyloric stenosis in older children is very rare but can be managed successfully by standard surgical procedure.
- ▶ For starting of early enteral feeds to overcome malnutrition, feeding jejunostomy should be performed along with the definitive surgical procedure.

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Competing interests None.

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