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Pyloric Atresia in a Neonate: Harbinger of clinical suspicion; A Case Report

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ABSTRACT

Pyloric atresia is an extremely rare congenital anomaly that typically presents in the neonatal period with symptoms of non-bilious vomiting, feed intolerance and upper abdominal distension. X ray abdomen and pelvis in a classical case shows a single gas bubble/single large stomach shadow in the abdomen. Differential diagnosis include supraampullary duodenal atresia, antral web and malrotation with midgut volvulus. Due to its rare occurrence, diagnosis requires a high index of suspicion. Isolated Pyloric atresia has a relatively good prognosis whereas Pyloric atresia associated with other congenital conditions such as epidermolysis bullosa (most commonly associated), multiple atresia syndrome and aplasia cutis congenita carries a more dire prognosis. Prompt diagnosis and surgical intervention are critical to prevent severe complications. Here we describe a three-day-old female neonate, who presented to us with history of feed intolerance and non bilious vomiting since birth. Upon evaluation the neonate was suspected to have pyloric atresia,. The patient underwent a pyloroplasty for membranous atresia (Type 1 pyloric atresia) and thereafter had an excellent postoperative recovery period. This Case report illustrates the clinical presentation, diagnostic findings, surgical management and reviews the literature available with respect to pyloric atresia, with the aim of highlighting the importance of early recognition and appropriate surgical treatment of one of the rare types of gastrointestinal atresia.

Keywords: *pyloric, atresia, pyloroplasty, congenital, rare*

1. INTRODUCTION

Pyloric atresia (PA) is one of the rarest types of atresia noted in neonates and occurs in 1 in 100,000 live births, accounting for 1 percent of all bowel atresias.[1,2] It is characterized by a complete or partial obstruction at the level of the pylorus, leading to symptoms such as non-bilious vomiting and upper abdominal distension in the neonatal period.[3-5]

Because of the rarity of PA, there is limited research and case studies available for review. PA can present in isolation or be associated with other anomalies, such as epidermolysis bullosa, aplasia congenita cutis, sensorineural deafness and multicystic dysplastic kidney[6-8,9,10]. Isolated PA has a good prognosis. In reviewing available literature, the clinical presentation of PA is consistent across reports. Non-bilious vomiting typically starts within the first 24-48 hours of life, and abdominal X-rays usually show a large gastric bubble with no distal bowel gas. The key to comprehensive management is timely diagnosis with early surgical intervention. The specific surgical management depends on the type of atresia identified intraoperatively. Here, we highlight a case of isolated pyloric atresia, and elaborate the management protocol followed to achieve a successful outcome.

2. CASE REPORT

A three day old female, pre-term, low birth weight neonate, was brought to Casualty with a history of non-bilious vomiting, intolerance of feeds and upper abdominal distension since birth. Antenatal and maternal history was not significant. On examination, baby had a distended upper abdomen with visible gastric peristalsis. Nasogastric tube was inserted which showed gastric aspirate only. Routine hematological and biochemical tests were done followed by an X ray abdomen and pelvis erect which revealed a large gastric bubble with complete absence of distal bowel gas (Figure 1). Given the radiographic findings and clinical presentation, pyloric atresia was highly suspected.

After resuscitation, surgical exploration was done, which showed hugely distended stomach, with hard and prominent pylorus. On attempting to pass nasogastric tube beyond the pylorus, it could not be negotiated, confirming the diagnosis of pyloric atresia. A longitudinal incision was made along the pylorus, just proximal to the site of visible obstruction. Pyloric membrane was identified within (mucosal membrane) causing obstruction- suggestive of Type 1 pyloric atresia. Membrane was divided and mucosal repair done for establishing continuity (Figure 2). A Heineke Mikulicz pyloroplasty was done and transanastomotic umbilical vein catheter was placed via naso-jejunal route to aid in enteral feeding. An orogastric tube was also placed to drain gastric secretions postoperatively. The neonate was transferred to the NICU for postoperative care. Enteral feeding was initiated on the third postoperative day via transanastomotic stent and incremented daily. Transanastomotic feeding tube was removed on POD (postoperative day) 10 and thereafter orogastric feeding was initiated followed by direct breast feeds. Abdominal drain was removed on POD 8. Serial abdominal X-rays demonstrated the passage of air into the small and large intestine confirming the resolution of the obstruction (Figure 3). An Upper GI dye study done 6 weeks postoperatively, showed passage of dye smoothly from the stomach into the small bowel loops with mild Gastroesophageal reflux (Figure 4).

3. DISCUSSION

Pyloric atresia is a rare congenital anomaly[1-3,8-10]. It can present in isolation or associated with congenital anomalies, like epidermolysis bullosa, aplasia congenita cutis, sensorineural deafness and multicystic dysplastic kidney[5-7]. The clinical presentation typically includes non-bilious vomiting, upper abdominal distension, and occasionally failure of passage of meconium. This clinical presentation may be a conundrum due to overlap with similar complaints in gastro-oesophageal reflux. Other differential diagnosis to be considered inludes causes of high intestinal obstruction like proximal (supraampullary) duodenal atresia which also typically presents with non bilious vomiting[4]. X ray abdomen and pelvis showing a single gastric shadow/air bubble with no gas distally, as seen in our case, gives a strong suspicion of PA[1,5-7,8,9]. Though the condition is well read theoretically, the number of cases seen clinically remains a fraction. The delay in diagnosis could be attributed to the rarity, and this contributes to the mortality. This case study highlights the feasibility of simple investigative modalities for a timely diagnosis.

The diagnosis is often confirmed by imaging studies, with abdominal X-ray being the first-line and sometimes, only required investigation.

Pyloric atresia is classified into three main types based on the anatomical findings[6-9]:

1. Type A (Membranous Atresia):- Most common type, characterized by a thin or thick membrane obstructing the pyloric canal. Surgical approach is a pyloroplasty.

2. Type B (Cordlike Atresia):- Fibrous cord replaces a segment of the pylorus. Surgical treatment involves excising the fibrous cord and performing an anastomosis between the stomach and duodenum.

3. Type C (Complete Atresia):- Most severe form, with complete separation of stomach and duodenum with no continuity of lumen. It requires a more complex surgical procedure, such as a gastroduodenostomy or Roux-en-Y gastrojejunostomy.[1]

Parelkar et al[5] advocated that all neonates with pyloric atresia should be evaluated for components of other anomalies, particularly to rule out HMIA (hereditary multiple intestinal atresia). This requires a thorough bowel walk through during laparotomy. While other physical congenital anomalies, particularly epidermolysis bullosa, can be evaluated on clinical examination of the neonate, genitourinary anomalies must be evaluated for radiologically. The evaluation should not preclude timely surgical interventions, and should ideally be completed prior to discharge of the patient, in order to prognosticate and follow up.

In a study by Gupta in 2013[6], all patients with pyloric atresia were diagnosed with a preoperative X ray abdomen and pelvis showing a single, large air bubble representing the dilated stomach with no gas distally. This classical sign should raise strong suspicions of pyloric atresia, as was in our case. Though an Upper Gastrointestinal contrast study is recommended to confirm failure of passage of contrast beyond the pylorus, the same was not followed in our patient as no significant alteration in management would be required, with findings subsequently confirmed intraoperatively without surgical delay.

Intraoperatively, it is imperative to locate the site of obstruction precisely, especially in those with a memabrane, to avoid missing a windsock diaphragm. A nasojejunal catheter or a feeding tube should be passed distally via orogastric/nasogastric route and in some instances even via a small gastrostomy. This aids in identifying a windsock deformity if present or another diaphragm.

In a review of the available minimum literature, it is evident that all neonates present classically with similar symptoms with radiological finding of single gastric bubble[1,5-10]. This should strongly prompt resuscitation and early management, lest dyselectrolemia and metabolic disturbances develop. Surgical correction is the definitive treatment for pyloric atresia. In cases of membranous atresia, as seen in our patient, pyloroplasty is an effective procedure to restore continuity and function of the gastrointestinal tract. Other surgical options include gastroduodenostomy or pyloric resection with end-to-end anastomosis in more complex cases. Placing a transanastomotic tube aids in early enteral nutrition. The most common postoperative complications following surgery include delayed gastric emptying and gastroesophageal reflux, which usually respond well to medical treatment and proper feeding practices.

4. CONCLUSION:

We can conclude from this case that Pyloric atresia, though rare, should be considered in neonates presenting with non-bilious vomiting and upper abdominal distension. Prompt diagnosis and surgical intervention are crucial for a favorable outcome. This case underscores the importance of early recognition using simple radiologial investigations and appropriate management of pyloric atresia depending on the type, to ensure normal feeding and good postoperative outcome in neonates. Furthur prospective studies would be required over a longer duration so as to assess in depth the outcome comparatively between patients with Isolated pyloric atresia and Pyloric atresia associated with other congenital anomalies in our centre.

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