Original Article

Annular Pancreas as a Rare Cause of Neonatal Duodenal Obstruction: A 6-year Review

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Abstract

Background: Congenital duodenal obstruction is one of the most common causes of neonatal intestinal obstruction that occurs 1 in 2500–10,000 live births. Owing to various causes, annular pancreas causing it is very rare. We present our 6-year experience by analysing clinical spectrum, management and outcome in neonates with annular pancreas causing duodenal obstruction at our hospital. **Materials and Methods:** We have reviewed all available hospital records of admitted neonates with duodenal obstruction from March 2016 to February 2022. Annular pancreas causing duodenal obstruction was sorted out from it and analysed all collected data according to age, gender, time of presentation, clinical presentations, associated anomalies, management, outcomes and follow-up period of 1 year. We have excluded malrotation of the gut associated with congenital diaphragmatic hernia and abdominal wall defects. **Results:** A total of 116 neonates with duodenal obstruction were admitted from March 2016 to February 2022. Fourteen cases were annular pancreas. Out of 14 cases of Annular Pancreas causing Duodenal obstruction, 9 cases were Male, 5 cases were Female (M: F =1.8:1), Mean gestational age at presentation was 36 +/- 1 week, and 4 neonates were preterm. The mean birth weight was 2124 ± 700 g. Most common presenting symptoms were vomiting in all 14 cases, bilious in 12 and nonbilious in 2 cases, upper abdominal distention in 11 (78.5%), dehydration in 6 (42.8%), hyperbilirubinemia seen in 4 (28.57%) cases. Associated anomalies were found in 5 cases, malrotation of the gut in 3, Down's syndrome in 2 and congenital heart disease (CHD) along with Down's syndrome in 1 case. The diagnosis was made with plain abdominal X-ray with a characteristic double-bubble sign in all 14 cases, but the diagnosis of annular pancreas was confirmed during laparotomy. Doppler Ultrasonography of abdomen and pelvis detect malrotation of gut in 3 out of 14 cases of Annular Pancreas causing Duodenal obstruction. All cases were managed surgically by open laparotomy with diamond-shaped duodenoduodenostomy, bypassing the obstructed duodenum by pancreatic tissue without dividing it. In 3 cases with associated malrotation of gut, were corrected with Ladds' procedure along with duodeno-duodenostomy. The average post-operative hospital stay was 12 ± 2 days. Two neonates died (mortality rate was 14%), one due to CHD and the other due to sepsis. All the cases were followed up for 1 year; during follow-up period, 2 babies were admitted again for bilious vomiting and dehydration. Both were improved with conservative management. **Conclusion:** Congenital duodenal obstruction is one of the most common anomalies of neonates. Annular pancreas, though rare, is an important cause of duodenal obstruction. Billious vomiting and characteristic double bubble sign in plain X-ray abdomen was key for its diagnosis, but confirmed during laparotomy. Diamond-shaped duodenoduodenostomy was the best surgical option. Mortality in neonates with duodenal obstruction caused by annular pancreas depends on associated anomalies and delayed presentation.

Keywords: Annular pancreas, duodenal obstruction, neonates

Introduction

Congenital duodenal obstruction is one of the most common causes of neonatal intestinal obstruction, accounting for almost 50% of cases.[1] The incidence of duodenal obstruction is 1 in 2500–10,000 live births.[1,2] Duodenal obstruction is classified as intrinsic, extrinsic or both, and it may be complete or partial.[3,4] Common causes are duodenal atresia, malrotation of the gut and rarely annular pancreas [Table 1].

Annular pancreas occurs 1 in 20,000 live births. Annular pancreas is a rare condition, in which the second part of the duodenum is surrounded by a ring of pancreatic tissue continuous with the head of the pancreas. This portion of the

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pancreas can constrict the duodenum and block or impair the flow of food distally. It is due to abnormal rotation of the ventral pancreatic bud in its course around the posterior aspect of the duodenum. The duodenum is encircled and might be obstructed by normal pancreatic tissue.[5] Three theories explain the development of annular pancreas, and it can result from the growth of a bifid ventral pancreatic bud around the duodenum, where the parts of the bifid ventral bud fuse with the dorsal bud, forming a pancreatic ring. It can also result if the ventral pancreatic bud fails to fully rotate, so it remains on the right or if the dorsal bud rotates in the wrong direction, such that the duodenum is surrounded by pancreatic tissue.

The treatment of the annular pancreas is surgical. Its aim is the relief of the duodenal obstruction. For this, various procedures are being used.

1. Division of a portion of the annular pancreas: This approach has many disadvantages. It increases the risk of pancreatitis, pancreatic or duodenal fistula.[6,7] Direct division of the annular pancreas should be avoided because this does not address the underlying intraluminal duodenal obstruction, and there is a significant risk of injury to the accessory pancreatic duct[8]
2. Gastrojejunostomy or duodenojejunostomy with Roux-en Y loop.[7,9] They are associated with peptic ulcer, and the risk of anastomotic ulcer suggests the need for a procedure like a vagotomy, which reduces acid secretion by the stomach.[7,10] These complications lead to the reluctance of many paediatric surgeons to perform these procedures
3. Duodenoduodenostomy. Kimura *et al*.[11] stated that diamond-shaped duodenoduodenostomy provided early functioning and wide anastomosis with the avoidance of a blind loop.

The surgical management of the annular pancreas is still developing and under revision. We herein describe our 6 years' experience of incidence and management of annular pancreas causing duodenal obstruction in neonates.

Materials and Methods

It is a retrospective study conducted at SVPPGIP, SCB Medical College, Cuttack, Odisha. We had analysed all available hospital records of admitted neonates from March 2016 to February 2022. We have analysed all collected data according to age, gender, time of presentation, clinical presentations, associated anomalies, management and outcomes. All cases were followed up for 1 year. We have excluded malrotation of the gut associated with congenital diaphragmatic hernia and abdominal wall defects.

All the neonates referred from peripheral hospitals and other medical colleges were admitted. All neonates stabilised haemodynamically with IV fluid and electrolytes and clinically examined. Clinically, neonates with duodenal obstruction usually presented with bilious vomiting and epigastric fullness. The diagnosis was confirmed with plain X-ray of the abdomen erect posture with a characteristic double-bubble sign [Figure 1]. Doppler ultrasonography (USG) of the abdomen and pelvis was done to rule out other associated anomalies, the position of the superior mesenteric artery and superior mesenteric vein to diagnose malrotation of the gut. After diagnosis, all neonates were subjected to surgical management. During laparotomy, annular pancreas causing duodenal obstruction was confirmed and grouped in our study [Figure 2].

Results

The study was conducted at SVPPGIP, SCB Medical College and Hospital, Cuttack, Odisha, from March 2016 to February 2022. The total number of patients admitted during this time was 7635, of which neonates with intestinal obstruction were 1803. After confirmation of diagnosis, neonates with duodenal obstruction were 116 (6.4%) and most common cause of it being Duodenal atresia in 57 (49.13%), malrotation of gut in 45 (38.79%) and Annular pancreas are less common cause in 14 (12.06%) cases [Table 1].

Out of 14 cases of Annular Pancreas causing Duodenal obstruction, 9 cases were Male, 5 cases were Female (M: F

Table 1: Causes of neonatal duodenal obstruction

Causes	Number of patients (%)
Duodenal atresia	57 (49.13)
Malrotation of the gut	45 (38.79)
Annular pancreas	14 (12.06)

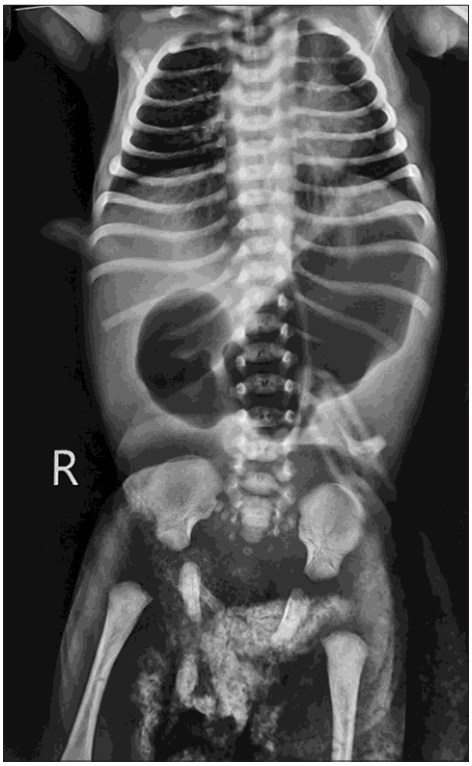


Figure 1: Plain X-ray of the abdomen showing characteristic double bubble sign

=1.8:1), Mean gestational age at presentation was 36 +/- 1 week, and 4 neonates were preterm. The mean birth weight was 2124 ± 700 g. Most common presenting symptoms were vomiting in all 14 cases, bilious in 12 and nonbilious in 2 cases, upper abdominal distention in 11 (78.5%), dehydration in 6 (42.8%), hyperbilirubinemia seen in 4 (28.57%) cases. Associated anomalies were found in 5 cases, malrotation of the gut in 3, Down's syndrome in 2 and congenital heart disease (CHD) along with Down's syndrome in one case.

The diagnosis was made with plain X-ray of the abdomen with a characteristic double-bubble sign in all 14 cases [Figure 1], but the diagnosis of annular pancreas was confirmed during laparotomy [Figure 2]. USG of the abdomen and pelvis done in all cases with features of distended stomach and proximal duodenum, in 3 out of 14 cases, there is alteration of SMA and SMV position suggestive of malrotation of the gut.

The management of annular pancreas causing duodenal obstruction was always surgical. Diamond shaped duodeno-duodenostomy as stated by Kimura *et al*.[11] by passing the obstructed duodenum by pancreatic tissue without dividing it is the best and commonly used procedure were done in all cases [Figure 3]. In 3 cases with associated malrotation of the gut, Ladd's procedure was done before duodenoduodenostomy.

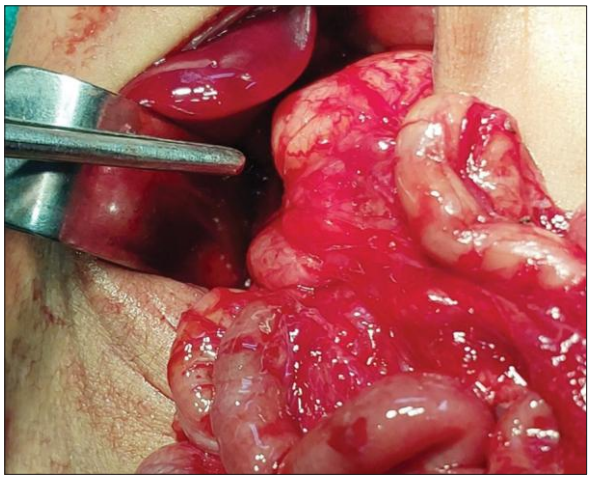


Figure 2: Annular Pancreas causing duodenal obstruction



Figure 3: Duodeno-duodenostomy, bypassing the obstructed duodenum by pancreatic tissue

All the cases were managed postoperatively in the neonatal intensive care unit. Oral feeding was started when the neonates passed stool. The average period to start oral feed was 6 days. After tolerating full feed, they were discharged from the hospital. The average post-operative hospital stay was 12 ± 2 days. During the post-operative period, 2 neonates died (mortality rate was 14%), one due to CHD and the other due to sepsis. All the cases were followed up for 1 year; during the follow-up period, 2 babies were admitted again for bilious vomiting and dehydration. Both improved with conservative management.

Discussion

In our study, 14 cases of annular pancreas causing duodenal obstruction were studied. The diagnosis of duodenal obstruction was made with clinical and radiological studies, but the differentiation of duodenal obstruction caused by annular pancreas was difficult and usually confirmed during laparotomy. Male neonates were affected more than females (1.8:1), which was comparable to study by Almoamin *et al*.[8] where M: F 2:1. Mean age of referral to our centre was 4.8 days (48hrs-12days), whereas Almoamin *et al*.[8] noted it was 3.6 days (24hrs-6days), Li *et al*.[12] showed it was 4.2 days (24hrs-13 days). In our study Mean Birth weight was 2124+/-700 gm, whereas in other study as noted by Yigiter *et al*.[13] it was 2285.2+/- 675 gm, cause of Low birth weight in Annular Pancreas causing Duodenal obstruction could be due to failure of Amniotic fluid absorption in the G I Tract in addition to pancreatic insulin secretion impairment.[14] Prematurity was noted in 4 neonates (28.5%), whereas Almoamin *et al*.[8] noted in one out of six neonates (16.7%), and Jimenez *et al*.[15] study it was 4 out of 16 neonates (25%). Association of Malrotation of gut with Duodenal obstruction due to Annular pancreas in our study were three (21.43%), whereas study by Almoamin *et al*.[8] it was 16.7%. Sencan *et al*.[16] showed it was 42.8%. Down's syndrome association in our study was in one case (7.1%), whereas Almoamin *et al*.[8] study it was 16.7%. In our study, duodenal obstruction caused by annular pancreas was complete in all cases which was comparable to a study by Ali Almoamin *et al*.[8] which was 100%. Vomiting was bilious in 14 cases (85.7%), nonbilious in 2 cases (14.2%), whereas Yigiter *et al*.[13] noted bilious in 59%, nonbilious in 41% and Jimenez *et al*.[15] showed bilious in 94%, nonbilious in 4% neonates. Moderate Jaundice (Indirect hyperbilirubinemia) were noted in 4 (28.5%) neonates and needed phototherapy. The diagnosis was done by plain X-ray abdomen with a characteristic double-bubble sign in all neonates (100%) and confirmed during laparotomy and comparable with a study by Sencan *et al*.[16] who reported the presence of a characteristic double-bubble sign in all (100%) neonates.

All neonates were treated with diamond-shaped duodenoduodenostomy with excellent result [Figure 3]. Kimura

et al.[11] stated that diamond-shaped duodenoduodenostomy provided wide anastomosis leading to early functioning and results. In our study, the average post-operative hospital stay was 12 ± 2 days whereas in other study by Ali Almoamin *et al*.[8] it was 10 ± 2 days. Survival rate in our study was 85.7% whereas study by Almoamin *et al*.[8] it was 100%. The prognosis of patients with duodenal obstruction caused by annular pancreas depends on associated anomalies and delayed presentation.

Conclusion

Congenital duodenal obstruction is one of the most common anomalies of neonates. Annular pancreas, though rare, is an important cause of duodenal obstruction. Billious vomiting and characteristic double bubble sign in plain X-ray abdomen was key for its diagnosis, but confirmed during laparotomy. Diamond-shaped duodenoduodenostomy was the best surgical option. Mortality in neonates with duodenal obstruction caused by annular pancreas depends on associated anomalies and delayed presentation.

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Conflicts of interest

There are no conflicts of interest.

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