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Jejunal atresia associated with jejunal diverticulosis

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ABSTRACT

Jejunoilial atresia is a frequent cause of bowel obstruction in neonates, with prevalence around 0.7 per 10000 livebirths. It is believed to be related to an intrauterine vascular accident that commonly presents as isolated pathology. Although, other associated anomalies have been reported on rare occasions, the association between the atresia and jejunal diverticulosis in newborn has never been described in literature. Furthermore, jenunal diverticulosis is rare below 40 and never been reported in children. Here in, we share the first reported case of this association and the first case of jejunal diverticulosis in this age category, the challenge in its management and provide review on literature.

1. Introduction

Jejunoileal atresia (JIA) is a frequent cause of neonatal intestinal obstruction with a prevalence of 0.7 per 10000 livebirths [1]. It is universally accepted that the etiology of this anomaly is linked to a late intrauterine mesenteric vascular accident due to conditions like intussusception, malrotation, internal hernia, segmental volvulus, intestinal perforation, or thromboembolism [2,3]. Furthermore, maternal ingestion of drugs such as acetaminophen, ephedrine, or cocaine during pregnancy have been related to occurrence of JIA [4]. Although, this anomaly is usually isolated, it may also be associated with other anomalies in other body systems with the GIT anomaly being the most frequent. JIA has also been described in association with abdominal wall defects and meconium ileus [5]. However, JIA associated with jejunal diverticulosis is never described in the English literature. Furthermore, Intestinal diverticulosis, the presence of multiple mucosal outpouchings at weak points in the bowel wall, has never been reported in children [6–8]. Here, we describe the first case of JIA associated with multiple diverticula in a newborn and difficulty in decision making when the pathology faced intra-operatively. We also include a literature review.

2. Case report

A preterm baby boy aged 34 weeks with a low birth weight of 1.935 kg was delivered by normal spontaneous vaginal delivery from a 30-year-old mother. The Apgar scores were 1, 4 and 7 at 0.5 and 10 min, respectively. The antenatal ultrasound showed dilated bowel loops and polyhydramnios. He was in respiratory distress at birth so he was intubated and shifted to neonatal intensive care and put on low setting parameters. His vitals were maintained within the normal ranges for age. The oro-gastric tube drained 60 ml of greenish aspirate. The abdomen was moderately distended, and the anus was patent and in its normal position. Abdominal X-ray showed features of low neonatal intestinal obstruction (Fig. 1). Contrast enema confirmed the diagnosis of jejunal atresia and patency of the colon. The echocardiogram was normal and abdominal ultrasound revealed right single kidney. The baby was kept fasting and resuscitated with intravenous fluids, and antibiotics (ampicillin and gentamycin) were started. The baby underwent surgery on the 3rd day, and there were one jejunal atresia type 3 and other multiple atresias (type 1 distally). Another rare finding was the associated diverticulosis in a long segment of the small bowel (Fig. 2).

After intraoperative assessment and calculation of the predicted remaining bowel, we decided to resect the atretic segment, the proximal dilated jejunum, and the segment of the bowel containing the diverticula. Thus, about 85 cm was removed followed by bowel anastomosis. Histopathology examination confirmed the presence of diverticulosis (Figs. 3 and 4). Histopathological examination of 15 serial tissue sections of each formalin fixed paraffin embedded jejunum tissue blocks stained with H&E supported the diagnosis of true and pseudo jejunal diverticulosis. The majority of the diverticulae had a muscularis propria all-around and can therefore be considered as a true diverticula due to congenital malformation (Fig. 3). Few pseudodiverticula with defect in the muscularis propria also detected (Fig. 4).

Postoperatively, the boy was extubated on the 2nd postoperative day. His nutrition was maintained by total parenteral nutrition. The bowel movement was recovered after 25 days, and he passed stool, but

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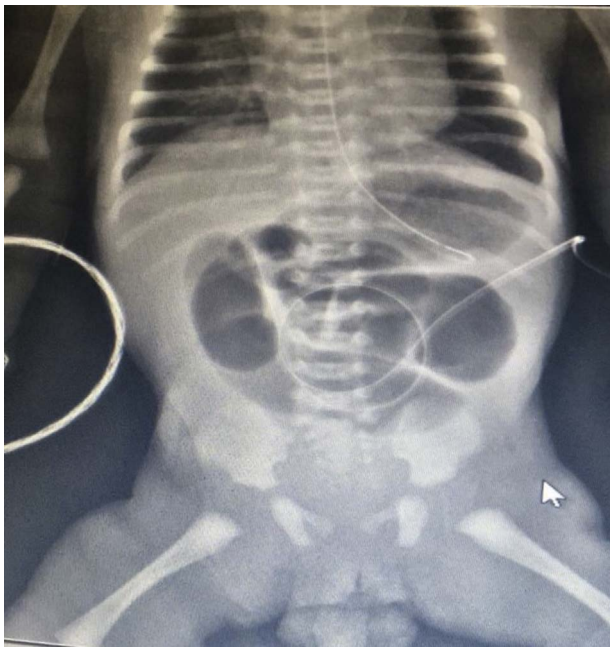

Fig. 1. Abdominal x-ray shows the gas distribution pattern of low intestinal obstruction.

the gastric residual remained high for another 15 days before declining to a minimal level. Oral feeding with elemental formula was started gradually after that over 2 weeks, and he was discharged after that in good condition.

3. Discussion

Atresia is a continual cause of intestinal obstruction in neonates. In a retrospective review of 277 neonates, Vecchia et al. [9], found that the level of obstruction was duodenal in 49.8% (138 cases), jejunoileal in 46.6% (128 cases), and colonic in 7.58% (21 cases). The atresias were multiple in 3.6% (10 cases). Considering JIA, the jejunum and the ileum are evenly affected in some reports [10,11]. The proximal jejunum is affected in 31% of cases, the distal jejunum in 20%, the proximal ileum in 13%, and the distal ileum in 36%. Multiple atresia are recorded in 6–20% of cases [12]. However, In another retrospective review for jejunoileal atresias at a huge children referral hospital in the Netherlands in the period between 1974 and 2004, Stollman et al. [13] found 114 infants with this anomaly: 62% of these atresia were in the jejunum, 30% in the ileum, and 8% in both. Heij et al. reviewed 21 neonates with jejunal atresia and 24 with ileal atresia. They found that jejunal atresia is usually multiple, more common in those with low birth weight and often associated with other anomalies [14]. Among the 114 neonates studied by Stollman et al. [13], they discovered other associated GI anomalies in 24% of patients, genitourinary malformations in 9%, cystic fibrosis in 9%, neurologic anomalies in 6%, and congenital heart disease in 4%. Other rare associations reported by other investigators were pyloric atresia and pylorocholedochal fistula [15]. Reviewing the English literature for jejunoileal atresia associated with jejunal diverticulosis or jejunal diverticulosis alone using the popular search engines; PubMed and Medline revealed no previous reports. There were a few reports describing the occurrence of isolated jejunal diverticulum in pediatric age group. These were in a child, presenting with small bowel obstruction [16], one caused diverticulitis and volvulus [16], another presented as perforated congenital jejunal diverticulum [17], and one was acquired diverticulum after resection and anastomosis for jejuno-ileal atresia [18].

In general, Jejunal diverticulosis is uncommon and rare in individuals under age 40. The reported incidence in adult literature was 0.02% to 7.1% [19–21], and based on enteroclysis studies, the small bowel diverticular disease incidence was 2.3% [20]. Simultaneous occurrence of jejunal diverticulosis and diverticulosis elsewhere in the GIT was described in adult by Rankin and Palder. They found in patients with jejunal diverticulosis, synchronous colonic diverticulosis in 30%–61% and synchronous duodenal diverticula in 22% [21,22]. The cause for the small bowel diverticular disease is not yet known; however, it is likely due to increased intraluminal pressure causing mucosal herniation along the mesenteric side of the intestine [23]. Marfan's syndrome, cystic fibrosis, and other genetic disorders can predispose these people to diverticular disease at a younger age [24–26]. It is usually asymptomatic but it may cause abdominal pain, and potentially life-threatening complications like perforation, diverticulitis, and bleeding [27].

Managing and making decision in such difficult and complex clinical case are not always easy especially when there is no previous reported cases, so the decision are usually depend on the personal knowledge and experience.

The surgical management for jejunal diverticulosis based on adult literatures is resection of the affected part. In this case, the decision was a challenge. Leaving the segment bearing the diverticulosis means to keep the child at long-life risk of complications that carry high morbidity and mortality while resection of such segment remove this potential. The final decision was resection of bowel segment bearing diverticulosis in addition to the atretic area and dilated jejunal segment proximal to the atresia was performed to avoid life-threatening complications, after calculating the adequacy of the remaining segment to maintain the bowel function. This decision may be not accepted by others and the argument of removing a long segment of bowel that contains the diverticula just because of the potential complication may result in short bowel. This case open the discussion among experts regarding what is the optimal management for case like this, if we face the situation again.

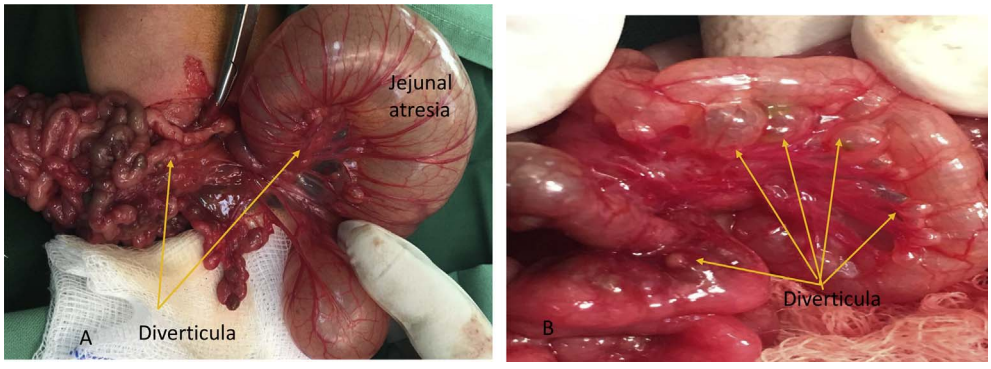

Fig. 2. Shows the intraoperative findings; A) The atresia and the diverticula even in atretic segment. B) diverticula are more clear after injection of normal saline in the segment distal to the atresia. The arrows point to the diverticula.