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Letter to Editor

What can a small bowel obstruction hide: an exceptional combination; Situs inversus, polysplenia, jejunas complex atresia

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Abstract

Introduction: Situs inversus, polysplenia, jejunas complex atresia are uncommon anomalies. Combination in a single patient is even rarer with difficulties in diagnosis and management.

Case repport: A 3-day-old newborn admitted with symptoms of intestinal obstruction. Radiological examinations revealed the presence of an abdominal situs inversus with dilated proximal small intestine handles. At laparotomy, abdominal situs inversus, polysplenia, multiple jejunal atresia with apple peel appearance of the ileum and malrotation were observed.

Conclusion: The combination of situs inversus, polysplenia and complex jejunal atresia is very rare. Preoperative diagnosis of situs inversus is important for proper placement of incisions and better planning of surgical management.

Introduction

Situs inversus is a rare anomaly in which the organs of the body are mirrored from their normal position [1]. It is known to be frequently associated with cardiac and gastrointestinal abnormalities requiring surgical repair. Preoperative diagnosis of the situs inversus is necessary for proper incision placement and improved surgical management. Polysplenia is defined as the presence of more than one spleen. It is associated with the situs inversus in about 20% of cases [2]. The incidence of jejuno-ileal atresia is approximately 1 in 5000 live births [3]. The association of these three conditions is very rare and the authors report that this work was presented according to SCARE criteria [4].

Observation

A 3-day-old girl was referred to us with biliary vomiting and slight abdominal distension since birth. The baby was born at term by C-section to a primiparous 30-year-old mother. The baby weighed 2.79 kg at birth. Prenatal ultrasound at 32 weeks' gestation revealed multiple dilated, fluid-filled intestinal loops. On admission, the baby was dehydrated with a slightly distended abdomen. The rest of the examination was normal. The results of the laboratory tests were within normal limits. X-rays of the abdomen revealed proximal bowel dilatation, and the nasogastric tube was in the right hypochondrium or upper right quadrant of the abdomen (Figure 1). Abdominal ultrasonography revealed a majority of liver on the left side of the abdomen, multiple spleen-shaped structures on the right side with distended intestinal coves. Echocardiography revealed a 5mm ostium secundum type Atrial Septal Defect (ASD) and levocardia.

With the preoperative diagnosis of the abdominal inverus situs confirmed, a classical transverse incision was made in

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the left hypochondrium, and laparotomy confirmed the situs inversus abdominis (Figure 2) with the liver on the left side. The baby had several rats on the right side (Figure 3). The proximal jejunum was massively dilated and there were multiple jejunal atresias (number of 6) (Figure 4). The entire distal small intestine was fed by a single branch of the ileocolic artery and had the appearance of an apple peel. There were several Ladd bands crossing the second part of the duodenum with a narrow-based mesentery. The segment of jejunum containing the atrestic segments was excised and severed from the proximal jejunal termination (Figure 5). Lateral jejuno-ileal anastomosis was performed, and finally the Ladd technique was performed. The residual length of the small intestine was approximately 145 cm, and after the operation the child was transferred to the neonatal intensive care unit. The immediate post-operative period was uneventful. The baby began tube feeding as early as the third post-operative day, but oral feedings were not tolerated until the fourteenth post-operative day. This may be due to hypoperistalsis in the dilated proximal jejunum. Full oral feedings were achieved by 28 days and the baby was discharged. After 18 months of follow-up, the baby is well and asymptomatic.

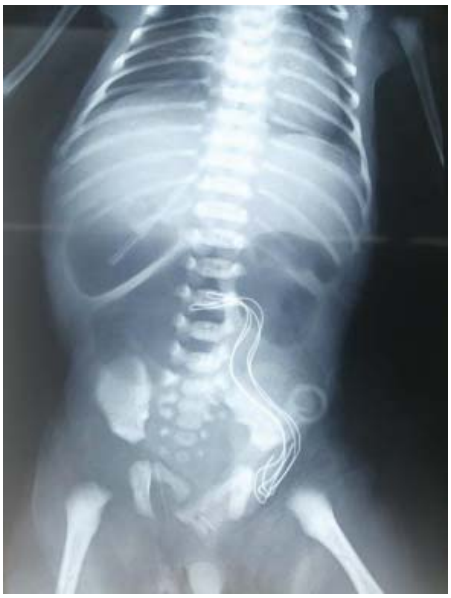

Figure 1: X-ray abdomen showing few dilated bowel loops, paucity of distal gas shad-ows and nasogastric tube in the right upper quadrant of abdomen.

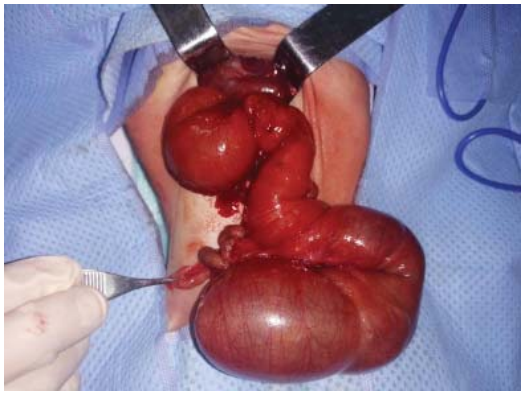

Figure 2: Laparotomy showing liver and duodenal C loop on the left side.

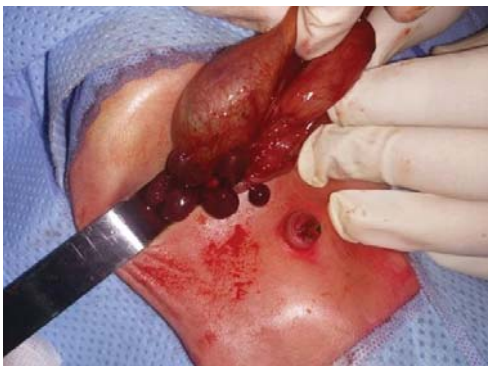

Figure 3: Laparotomy showing ultiple spleens on the right side.

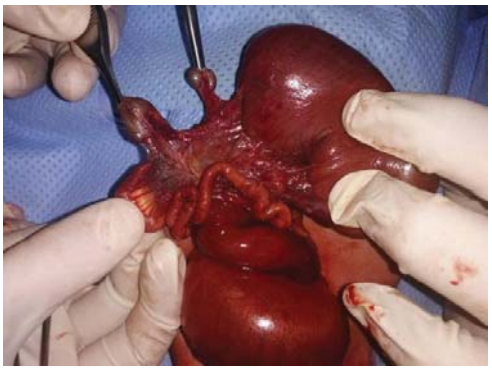

Figure 4: Intra-operative photograph showing massively dilated proximal jejunumwith multiple atresias.

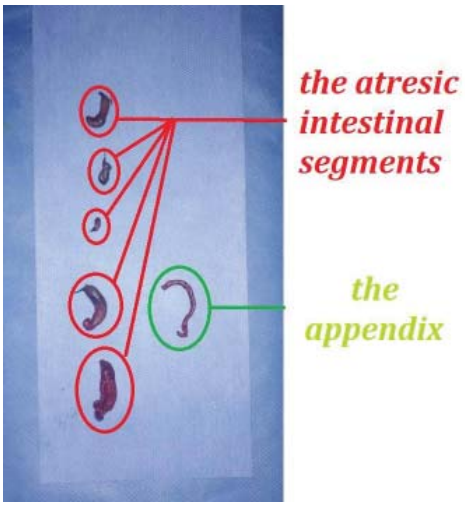

Figure 5: Picture of the atresic intestinal segments and appendix.

Discussion

The normal anatomy of the internal organs is known as the situs solitus. The mirror transposition of the situs solitus is known as the situs inversus in which the solid organs are reflected. It may be total or partial. Fonkalsrud et al, in their study of patients with inverus situs, showed that 15 patients with inverus situs had major abdominal abnormalities requiring surgery and 8 of them had duodenal or jejunal atresia [5]. The preoperative diagnosis of situs inversus is important in planning surgical incision and abdominal procedures. polysplenia refers

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to the presence of two or more spleens. The location and number of spleens are variable; their number may range from 2 to 16. The common presentation is vague abdominal pain, nausea and vomiting. Polysplenia refers to its association with various abnormalities of the organs of the abdomen and chest. It is often recognized in childhood, although approximately 10% of cases are present in late adulthood. Lee, et al. recently reported a 12.1% rate of complex jejuno-intestinal atresia (type IIIb appl-peel atresia and type IV multiple intestinal atresia) in all patients with intestinal atresia, and these patients experienced greater morbidity or mortality [6]. None of these patients had a combination of type IIIb and type IV atresia [6]. Federici, et al. [7] and Rich, et al. [8], have recently described the association of complex jejunal atresias, as well as malrotation and polysplenia in a patient with an inverus situs is very rare. There have been only clinical cases in the literature. Rasool et al. reported a syndrome of polysplenia associated with type I jejunal atresia in a 2-day-old girl [2]. Abdur Rehman, et al. reported a case of dextrogas-tria, reverse rotation of the midgut and intestinal atresia in a neonate [9]. However, in their case, the spleen was absent. Peetsold et al.reported a girl with jejunal atresia appl-peel with situs inversus operated in the neonatal period [10]. Ruben, et al. reported one case of jejunal atresia with situs inversus without malrotation or splenic malformation [11]. The embryology of the situs inversus has not been well established. However, the rotation abnormality may lead to a vascular accident resulting in multiple atresias and the appearance of appl-peel, as in our case.

Conclusion

The association of situs inversus, polysplenia and complex jejunal atresia is very rare. Preoperative diagnosis of the situs inversus is important for proper incision placement and surgical planning.

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