

Ileocolic Atresia with Appendicular Duplication: A Case Report

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Abstract

INTRODUCTION: Intestinal atresias are rare entities, the incidence of ileocolic atresia is 1 in 5000 to 10,000 live births. The incidence of appendiceal duplication is 0.004% to 0.009% and is associated with other anomalies of the digestive and genitourinary tract. Less than 100 cases of Appendiceal Duplication have been reported since 1902. **PRESENTATION OF CASE:** Two-day-old male newborn born by caesarean section due to fetal distress, was referred for abdominal distension due to necrotizing enterocolitis, sepsis and probable syphilis. The physical examination found abdominal distension, the X-ray of the abdomen showed pneumoperitoneum, the associated lab work indicated leukocytosis, severe thrombocytopenia, high potassium. During surgery, a greenish and purulent fluid was found, dilated distal ileum with necrosis and a blind pouch: subseral cecal appendix with coprolite inside, collapsed colon, suppurated cecum with necrotized blind pouch with fibrin, the dilated distal ileum was resected with cecal appendix. The cecum and blind pouch are resected; an ileostomy and colostomy are performed with mucous fistula. The patient had infection by Klebsiella and pseudomonas, was discharged at one month of life and was transferred to a more specialized hospital for severe dehydration, the intestinal tract was restored without major complication. **DISCUSSION:** Ileocolic atresia are rare entities, appendix duplication also, which may be an incidental finding during surgery. It is important to have the surgery as soon as possible to avoid complications. **CONCLUSION:** This is an extremely rare pathology, the surgery should be performed immediately after diagnosis or suspicion of obstruction to prevent further complications.

Keywords

Ileocolic Atresia, Double Appendix

1. Introduction

Intestinal atresias are rare entities; ileocolic atresia has been described in very few cases [1]. The incidence is 1 in 5000 to 10,000 live births, with colonic atresia being the least frequent [1]. Appendiceal duplication has an estimated incidence between 0.004% and 0.009% [2]-[4], most anomalies of the appendix have been observed in adults [4], double appendices are usually incidental findings during surgical procedures [4], Picoli (1892) reported the first case of appendix duplex in a female patient who had associated anomalies of duplication of the entire large bowel, two uteri with two vaginae, ectopia vesicae and exomphalos [4]. The Wallbridge-Waugh classification divides them into three groups according to the location of the base of the appendix [5], for this case, it is type B2 [4] [5]. Appendiceal duplication is associated with other anomalies of the digestive and genitourinary systems [6]. Fewer than 100 cases of appendiceal duplication have been reported in world literature since 1902 [6]. The following is a case of a newborn with type IIIA ileocolic atresia and appendiceal duplication type B2.

2. Case Report

One-day-old male newborn born by caesarean section due to fetal distress was referred from level II hospital for abdominal distension due to necrotizing enterocolitis suspicion, sepsis and probable syphilis. The patient is admitted to the neonatal intensive care unit, the physical examination found abdominal distension with an absence of bowel sounds. The X-ray of the abdomen showed pneumoperitoneum, the associated lab work (CBC) indicated leukocytosis, severe thrombocytopenia, potassium in 8 meq/L and respiratory distress. The on-call surgical team decided to wait until the patient was stabilized. A day after, the patient remained in serious condition, for the past two days; his potassium level dropped to 7 mEq/L in 24 hours, and the patient was at risk of death. A medical board meeting was held with neonatology and anesthesiology, and despite the patient's condition, surgery was decided upon. During surgery a greenish and purulent fluid was found, dilated distal ileum with necrosis and a blind pouch: subseral cecal appendix with coprolite inside (**Figure 1**), collapsed colon, suppurated cecum with necrotized blind pouch with fibrin (**Figure 2**), the dilated distal ileum was resected with cecal appendix. The cecum and blind pouch are resected (**Figure 3** and **Figure 4**). An ileostomy and colostomy are performed with mucous fistula. The patient received broad spectrum antibiotics, enteral nutrition, had infection by *Klebsiella* and *Pseudomonas*, was discharged at one month of life, two weeks later, patient returned to the emergency room for severe dehydration, was transferred to a more specialized hospital, because the hospital doesn't have pediatric unit care, after stabilizing and colon irrigations, the intestinal tract was restored without major complication, oral nutrition was resumed three days after surgery and patient was discharged later without complications. The appendicular duplication was confirmed by histopathology.

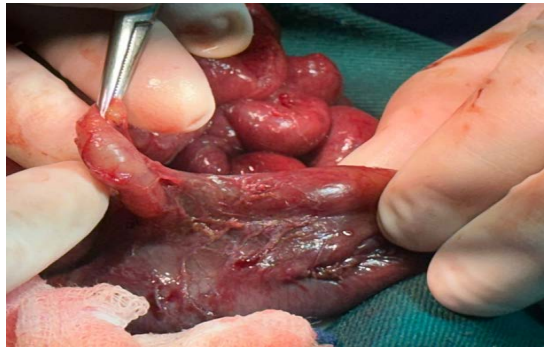


Figure 1. Perforated terminal ileum with cecal appendix.

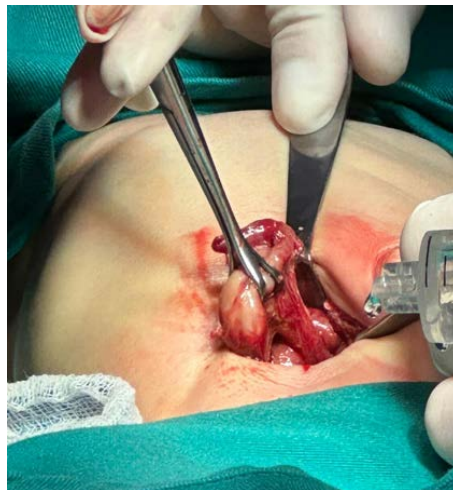


Figure 2. Intestine with cecal appendix and blind sack.

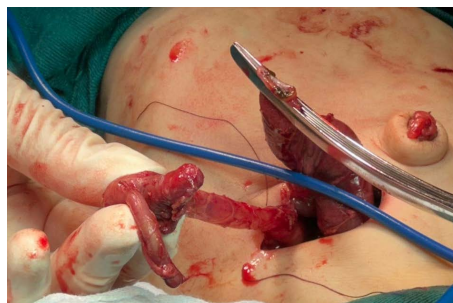


Figure 3. Ileocolic atresia.



Figure 4. Distal ileum and cecum with appendix and blind sack.

3. Discussion

Ileocolic atresias are the rarest of the gastrointestinal tract, and may or may not be associated with the absence of the ileocecal valve [1] [7] [8]. Mesenteric vascular occlusion is a cause for distal jejunal and ileal atresia [7] [9]. Some authors report intrauterine intestinal intussusception as a possible cause of ileocolic atresia [7] [8]. The importance of ileocecal atresia lies in the presence or absence of the ileocecal valve (ICV).

Appendiceal duplication is an even rarer condition, which may be an incidental finding during surgery. The embryogenesis of the appendix is well known, however, the pathogenesis of duplication is not well documented although some theories have been put forward including the split notochord theory, median septum formation, normal regression of embryonic diverticula and the partial twinning procedure [4] [6] [10] [11], environmental factors such as trauma or hypoxia during early fetal development also play an important role [6] [11]. According to the Cave-Wallbridge classification (**Table 1**) [4]-[6], in our case, it would be type B2. Surgical resection of both appendages is indicated, even if only one is inflamed, because it could develop abnormalities in the future [11]. In our case, we decided to resect the IVC because a perforation was found at that level, an ileostomy and colostomy were performed as a mucosal fistula, for future ileocolic anastomosis as described in other studies [9], although other authors describe the creation of a new IVC [10]. It is therefore important to search for other anomalies in the same sitting or afterwards where possible [6], in our case, no other malformation was found. It is important to stabilize the patient in the first few hours and make the surgery as soon as possible to avoid complications and death. The prenatal diagnosis of intestinal atresia helps to make an early surgery and avoid complications, signs such as polyhydramnios on ultrasound, could help to refer the pregnant woman to a more complex hospital to expand the study; appendiceal duplication is an incidental finding.

Table 1. Cave-Wallbridge classification.

Type A	Single caecum and incomplete duplication
Type B1	Symmetric duplication at both sides of the iliocaecal valve
Type B2	Duplication, one normal and the other at a different localization
Type C	Duplication occurring with Caecum duplication

4. Conclusion

The ileocolic atresia and appendiceal duplication is an extremely rare pathology, prenatal diagnostic of atresia prevent complication, it is important to stabilize the patient in the first few hours and perform the surgery immediately after diagnosis or suspicion of obstruction to prevent further complications such as sepsis even death, and also to provide irrigation in the distal segment.

Conflicts of Interest

The authors declare that they have no conflict of interest. Ethical approval for this study was exempted by our institution, and the parents written informed consent.

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