

An Isolated Tubular Intestinal Loop in a Neonate with Type II Intestinal Atresia

Soon-Ok Choi, M.D., Ph.D., Eunyoung Jung, M.D., and Woo-Hyun Park, M.D., Ph.D.

Department of Pediatric Surgery, Keimyung University Dongsan Medical Center, Daegu, Korea

An isolated tubular intestinal loop (ITIL) means an anatomical or vascular communication with rest of the bowel loop and may provide an insight into the pathogenesis of intestinal atresia. We experienced a case of an ITIL identified in omentum of a 4-day-old neonate with type-II intestinal atresia. To our knowledge, this association has never been reported in the English literature. Omental wrapping of the incompletely resorbed ischemic bowel segment may explain this phenomenon in a case of congenital intestinal atresia.

Key Words : Isolated intestinal loop, Intestinal atresia, Neonate

Intrauterine vascular accident is a well-known etiology of jejuno-ileal atresia.¹⁻³ Vascular occlusion of the fetal mesenteric vessels results in irreversible changes with disintegration of the ischemic bowel, which remains as the atretic segment. However, the in-depth mechanism of the remnant loop of bowel remains unclear. Herein, we present an isolated tubular intestinal loop (ITIL) in a neonate with type II intestinal atresia, which was anatomically separate from the main intestinal continuity and was embedded in omentum. This case may enhance the understanding of ischemic insult as a cause of jejunoileal atresia and support the suitability of omentum as a potential source of blood supply for ischemic intestine.

Case Report

A 3 day-old male was transferred to our hospital because of abdominal distention and multiple episodes of bilious vomiting after birth. The pregnancy was uncomplicated and no abnormality was detected on

prenatal ultrasound examination. He was delivered by spontaneous vaginal delivery at 38⁺² weeks of gestation with a birth weight of 3,020 g. The delivery was uneventful but the neonate began to have bilious vomiting, upper abdominal distention, and had passed white meconium. He had never been fed after birth. On admission to our neonatal intensive care unit, he was active and slightly distended abdomen on examination. The body weight was 2,850 g at the time of transfer. A plain abdominal radiograph showed a significantly dilated bowel loop with air-fluid levels with an absence of air in lower abdomen. A contrast enema with gastrografen showed microcolon with an influx of contrast to the terminal ileum and jejunal atresia was diagnosed. The continuity operation was performed on the 4th day of life. Intraoperative findings were suggestive of type I jejunal atresia (Fig. 1). The atresia was located approximately 110 cm distal from the duodenojejunal junction. The omentum had adhered to this atretic segment, and a fibrous band to the mesentery was noted. In addition, we found a 3 cm-long tubular structure located in the omentum, and this was suspicious of an intestinal loop (Fig. 2). A double barrel ileostomy was performed due to great disparity of sizes (>6 times) between the proximal and distal

접수 : 2013년 5월 14일, 수정 : 2013년 8월 31일

승인 : 2013년 9월 2일

주관책임자 : 최순옥, 700-712 대구광역시 중구 달성로 56번지

계명대학교 의과대학 동산병원 소아청소년과학교실

전화 : 053)250-7322, 전송 : 053)250-7322

E-mail : choi1635@dsmc.or.kr

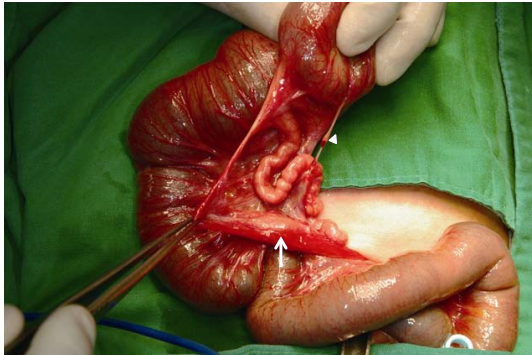


Fig. 1. Operative findings of intestinal atresia showing type I atresia and a tubular structure (indicated by arrow) embedded in omentum, and a cord-like band (indicated by arrowhead).

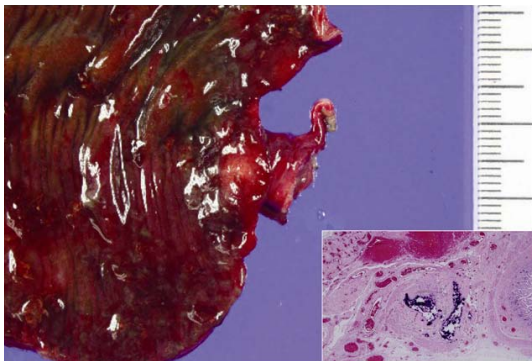


Fig. 2. A longitudinal section of the atretic portion showed atresia with interruption of bowel continuity. A microscopic findings of the atretic part shows loss of continuity with separation by fibrous septum (inside box).

ends of the bowel. Pathologic examination revealed type II small intestinal atresia with separation of the 2 blind ends by fibrous tissue (Fig. 2). The microscopic findings of the cross section of the isolated tubular structure showed a degenerated intestine with four distinct tissue layers – mucosa, submucosa, muscle, and serosa (Fig. 3).

Discussion

Intestinal atresia is the most common congenital small bowel anomaly and is a major cause of intestinal obstruction in neonates.⁴ The exact prevalence is not known, it is reported to be around 2.9 cases in the

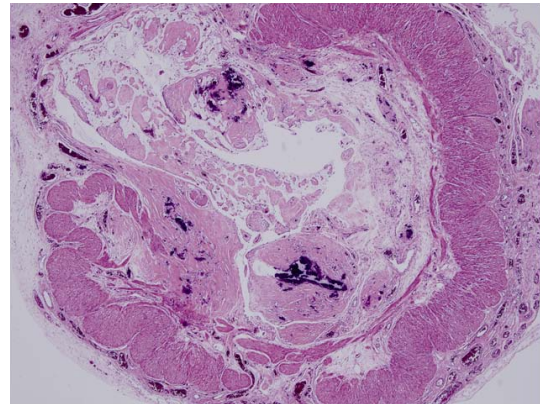


Fig. 3. Cross section of the tubular structure shows degenerated intestine with 4 layers with luminal obstruction (H&E stain).

United States and 4.5 cases in Far East Asians per 10,000 live births in 2004.⁴ Intestinal atresia is believed to be caused by an intrauterine vascular insult with interruption of local blood supply and by subsequent atrophy and involution of the intestinal segment involved.^{1-3,5,6} Various events could induce intrauterine vascular insults such as volvulus, internal hernia, intussusception, and infarction.⁴ Theoretically, a loop of intestine might become isolated, and this loop could undergo resorption if its own blood supply is poor.⁶ However, this feature has not been reported so far. In this case, we believe that the isolated tubular intestinal loop (ITIL) represented an incomplete degradation of the ischemic intestinal loop, which was partially sustained by omentum.

An ITIL lacks an anatomical or vascular communication with the remainder of bowel loop. The exact mechanism of ITIL is unclear, however, ITIL is considered to be a bowel segment, which has not fully involuted after the ischemic event. It is not a well-known fact that such findings of less degrees of vascular impairment in the intestine. Intestinal infarction could be remained by fibrosis, stricture and obstruction at the involved segment. Interruption of the mesenteric blood flow creates a V-shaped region of

ischemia in bowel wall and its mesentery. If the omentum grabs the bowel loop, the bowel loop will be taken away from the main bowel and embedded in the omentum. In case of simultaneous adjoining the two ischemic ends of main bowel and its mesentery, type II intestinal atresia could be formed. The intestine will preserve its outer shape even though the lumen is not patent just like our case. In an animal model, omentum had been found to promote angiogenesis when fixed to an ischemic bowel segment.⁷ In our case, the omentum may be the reason why the isolated bowel loop had not undergone complete resorption. The gross operative findings suggested mucosal atresia with an intact bowel and mesentery (Type I atresia). However, 2-3 mm fibrous septum was occluding the lumen, between the two atretic ends even the preservation of the bowel wall in continuity with an intact mesentery. Hence, the final diagnosis of this case was type II atresia in pathology.

In conclusion, we have presented a rare case of ITIL in a neonate with type II intestinal atresia. The isolated loop was embedded in the omentum and had his-

tological features suggestive of continued blood supply from the omentum. This case of omental ITIL could explain the in-depth mechanism of intestinal atresia.

References

- 1) Louw JH, Barnard CN. Congenital intestinal atresia; observations on its origin. *Lancet* 1955;269:1065-7.
- 2) Nixon HH, Tawes R. Etiology and treatment of small intestinal atresia: analysis of a series of 127 jejunoileal atresias and comparison with 62 duodenal atresias. *Surgery* 1971;69:41-51.
- 3) Dalla Vecchia LK, Grosfeld JL, West KW, Rescorla FJ, Scherer LR, Engum SA. Intestinal atresia and stenosis: a 25-year experience with 277 cases. *Arch Surg* 1998;133:490-6; discussion 496-7.
- 4) Frischer J, Azizkhan R. Jejunoileal Atresia and Stenosis, in *Pediatric Surgery*. 7th ed. ed. Edited by Coran AG, Philadelphia, PA: Elsevier/Saunders; 2012. p1059-71.
- 5) Abrams JS. Experimental intestinal atresia. *Surgery* 1968; 64:185-91.
- 6) Dickinson SJ. Origin of intestinal atresia of newborn. *JAMA* 1964;190:119-21.
- 7) Shoshany G, Cohen E, Mordohovich D, Hayari L, Har-Shai Y, Bar-Maor JA. Creation of the isolated bowel segment in animals by omentoenteropexy. *J Pediatr Surg* 1994;29:1344-7.

= 국 문 초 록 =

제2형 소장 무공증 신생아에서 동반된 분리된 장고리

계명대학교 동산의료원 소아외과

최순옥·정은영·박우현

정상 장과 분리되어 존재하는 관상 장고리는 정상적인 장과 해부학적 혹은 혈액공급이 분리되어 있는 퇴화성 장조직으로 소장 무공증의 발생과정에서 관찰할 수 있는 소견이다. 저자들은 제2형 소장 무공증으로 진단된 4일된 남아의 수술 중 정상 장과 분리되어 존재하는 관상 장고리를 발견하였기에 보고하는 바이다. 저자들이 아는 바로 이러한 소견은 현재까지의 문헌에서는 아직까지 보고되지 않았다. 자궁 내에서 허혈된 소장의 일부가 불완전하게 흡수되면서 대망에 싸여있는 소견은 선천성 발생학적으로 소장 무공증이 형성되는 과정을 제안해 줄 수 있을 것으로 생각된다.

중심 단어 : 분리된 장, 소장 무공증, 신생아