

TUBULAR COLONIC DUPLICATION WITH ANORECTAL MALFORMATION RECTO-VESTIBULAR FISTULA: A CASE REPORT

Muhammad Noor Fitriansyah¹, Ken Sanden^{2*}, Dadik Agus Soenjoto³

^{1,2} Universitas Mulawarman, Indonesia

³ Abdoel Wahab Sjahranie Regional General Hospital, Samarinda, Indonesia

Email: sandenken39@gmail.com*

Abstract

Anorectal malformations associated with colonic duplications are a rare condition. Double terminations of tubular colonic duplication with Anorectal Malformation are even rarer. Colonic duplication may have different presentations according to its location and size and incidence of less than 15% of all gastrointestinal duplications. These cases also illustrate the diagnostic challenge and variable presentation of colonic duplications in female patients with recto-vestibular fistula. This report aims to present and analyse the clinical presentation, diagnostic approach, and management of anorectal malformations associated with colonic duplications highlighting the challenges and treatment strategies for such a rare case. Case Presentation: We present the case of an 11-month-old female referred from a network hospital with a history of stool leakage from genital, status post diverting colostomy at age of 4 month and was referred to our center for further evaluation and treatment. During the procedure of diverting colostomy, colonic duplication was identified at the colostomy site. Further radiographic evaluation and exploratory laparotomy with complete resection of the colonic duplication was performed. Conclusion: Colonic duplication is a rare congenital disorder, with an even rarer association with anorectal abnormalities. This condition presents with diverse clinical manifestations, necessitating a thorough preoperative differential diagnosis. Surgical intervention remains the treatment of choice, with complete resection being the preferred approach for cystic, diverticular, and short tubular duplications. However, total colon duplications may require alternative management strategies.

Keywords: Colonic duplication, Anorectal malformations, Pediatric Surgery

Introduction

Duplications of the alimentary tract is a rare and diverse congenital disorder that can be found anywhere from the mouth to the anus. These unusual congenital anomalies present both diagnostic and therapeutic challenges to surgeons. Because these lesions occur so infrequently, they are often not suspected until encountered intraoperatively (Duess et al., 2022). Due to the complicated anatomy with common blood supply shared between the duplication and the associated native bowel, and a possibility of associated anomalies in the urogenital system, an appropriate management requires familiarity with the anatomy and clinical characteristics of each symptom (Piplani et al., 2022).

An 11-month-old female was brought by her parents to the outpatient installation of the pediatric surgery clinic of the Abdoel Wahab Sjahranie Regional General Hospital

with a referral letter from another network hospital for further examination and treatment related to her branching intestine condition. Based on information provided by the parents, the patient had a history of passage stool per vaginal area and stool per normally sited anus since her first day of life. Additionally, the patient's parents reported frequent difficulty with defecation and a history of irregular bowel movements (Isa et al., 2021).

Initially, the parents consulted with general practitioner who subsequently referred them to a pediatric surgeon at previous hospital. The patient was diagnosed with congenital anorectal malformation, specifically a recto-vestibular fistula, and underwent a diversion colostomy procedure at 4 months of age. During the procedure, it was discovered that the patient's colon was duplicated. After 6 months of routine follow-up, during which the patient remained stable and the surgical wound had healed properly, she was referred to Abdoel Wahab Sjahranie Hospital for further management. For additional information the patient is the 3rd child of the mother's 3rd pregnancy with history of regular antenatal check-ups. Mother's labour was assisted by a local midwife, the delivery was spontaneous vaginal at 39 weeks gestation. The patient was born with a birth weight of 3100 grams and a body length of 51 centimeters.



Figure 1. Proximal and Distal Stom

Upon arrival at Abdoel Wahab Sjahranie Hospital, physical examination of the patient's abdomen showed that the proximal and distal stomas were in viable condition with no signs of inflammation, the abdomen did not appear distended, tender on palpation and bowel sounds were normal. On examination of the genitourinary organs, a fistula was found in the vestibular area without any other comorbidities. The anal opening was in a normal position with good tone. Routine blood examination revealed a decreased Haemoglobin level (9.6 g/dL) with normal leukocyte (13,070 /uL) and chemical blood test shown a normal blood sugar level, serum electrolytes, coagulation factors and renal function. A chest X-ray, renal ultrasound and echocardiography were performed at the referring hospital to evaluate for potential urinary or cardiac anomalies. The results showed no evidence of abnormalities, and no additional comorbidities were identified.



Figure 2. Distal Lopography in Left Lateral Projection

A barium enema examination through distal colostomy (Lopography) revealed that the distal end of the contrast medium was located in the middle third of the rectum, approximately 2.4 centimeters from the anal dimple marker. Anterior leakage of contrast was observed, suggesting the presence of a rectovaginal fistula without any communication between the native colon and the duplicated part. Based on these findings, patient was diagnosed with Tubular Colon Duplication with Anorectal Malformation and the patient was scheduled for surgical intervention by the pediatric surgical team.

Laparotomy exploration colonic resection and posterior sagittal anorectoplasty (PSARP) procedure were performed on August 12, 2024. Intraoperatively, two Foley catheter (16Fr and 8Fr) were placed in the fistula and the rectum to help identify the duplicated colon and the native colon respectively. Colonic duplication was identified with the proximal end on the distal stoma and the distal end on the vestibule. Colonic resection was performed throughout the colonic duplication with approximately 12 centimeters in length, along with fistula excision and then followed by the PSARP procedure (Wu et al., 2018). At the end of the procedure, the diversion colostomy is retained with the mucus fistula now consisting only of the native colon (AbouZeid et al., 2019).

Postoperative the patient was admitted to the Pediatric Intensive Care Unit (PICU) for monitoring and management. On the third postoperative day, wound infection was noted, necessitating a wound debridement procedure and culture samples were obtained to guide targeted antibiotic therapy. The patient was finally discharged after treatment for 17 days postoperatively.



Figure 3. Preoperative: catheter was advanced through the mucus fistula and observed exiting from the vestibule and the other from the anus.

Method Research

This study employs a normative juridical research method, focusing on a systematic analysis of legal norms governing construction contract dispute resolution. The normative-juridical approach is chosen for its ability to examine the coherence between regulations and their effectiveness in practical implementation. This methodology is reinforced by a conceptual approach to explore legal doctrines relevant to dispute resolution in the construction sector.

The research data consists of primary, secondary, and tertiary legal materials collected through a comprehensive literature review. Primary legal materials include hierarchical regulations ranging from the 1945 Constitution to technical regulations related to construction services, such as Law No. 2 of 2017 on Construction Services, Government Regulation No. 22 of 2020, and regulations governing public procurement. Secondary legal materials comprise accredited academic journals, legal textbooks, previous research findings, and documentation of dispute resolution practices. Meanwhile, tertiary legal materials, such as legal dictionaries and encyclopedias, are used to enhance conceptual understanding.

The data analysis process follows three main stages. First, a comprehensive inventory of regulations governing construction dispute resolution is conducted, including an analysis of regulatory hierarchy and harmonization. Second, a content analysis of secondary legal materials is performed to identify patterns, trends, and issues in the implementation of regulations. Third, findings are synthesized to formulate applicable and contextually relevant recommendations for optimizing dispute resolution mechanisms in Indonesia's construction industry.

A qualitative juridical analysis technique is applied, utilizing systematic and teleological interpretation methods to understand the intent and objectives of legislators and the socio-legal context of regulation implementation. The validity of research findings is strengthened through data triangulation and peer review by construction law experts. This approach enables the study to generate not only a comprehensive theoretical analysis but also practical insights into improving construction dispute resolution mechanisms.

Result and Discussion

Congenital colonic duplication is a rare anomaly of the intestinal tract, with the complete tubular form being exceptionally uncommon. These duplications exhibit significant variability in size and morphology, presenting as either cystic or tubular structures. The estimated incidence, derived from fetal and neonatal autopsy data, ranges from 1 in 4,500 to 1 in 5,000. Among gastrointestinal duplications, colonic duplications account for only 6–14% of cases, making them particularly rare.^{1,2} Colonic duplication typically presenting with symptoms such as constipation, vomiting, volvulus, perforation, and most commonly bowel obstruction. From a morphological perspective, cystic duplications along the colon are the most common anomalies, followed by tubular duplications. Typically, tubular colonic duplication presenting as either a double-barrel or Y-shaped structure (Bahmad et al., 2021).

The pathogenesis of gastrointestinal duplications remains unclear and is believed to involve multifactorial mechanisms, including molecular genetic factors. Alimentary tract duplications present a significant diagnostic challenge due to their variable clinical and anatomical manifestations, some cases are even diagnosed after the presentation of complications or found intraoperatively for other malformation conditions. Same as the case that has been presented, the patient had a mild symptom of difficulty in defecation and irregular bowel movements (Ahmed et al., 2021). However, colonic duplication was incidentally discovered during surgery performed for an anorectal malformation.

Accurate characterization of colonic duplication often requires selective radiologic imaging studies to determine the precise anatomy. Common diagnostic modalities include ultrasonography, barium swallow, computed tomography (CT), and magnetic resonance imaging (MRI). In pediatric populations, MRI is particularly advantageous as it can not only delineate the anatomy of the duplication but also identify associated spinal anomalies. Barium enema studies may provide a more detailed assessment of colonic duplications, enabling precise anatomical mapping. Comprehensive preoperative planning, guided by these imaging techniques, is essential to optimize surgical outcomes.^{6,7}

Definitive management of anorectal malformation with colonic duplication necessitates disconnection of the fistula associated with the colon or the duplicated bowel. This is typically achieved through the usual posterior sagittal reconstruction anoplasty (PSARP).⁸ The management of gastrointestinal duplications itself varies based on the type, location, size, and presence of associated anomalies. Total excision is considered the optimal treatment, particularly for short-segmented or cystic duplications to prevent future complications including the potential for malignant transformation.² However, for some cases with long-segmented or total tubular colonic duplication, complete resection of the duplicated colon may not be feasible due to shared vascular supply. Various surgical approaches have been described for the management of long-segment colonic duplication. One of the methods involves fenestrate or connecting the distal portion of the duplicated colon to the normal colon using either a hand-sewn side-to-side anastomosis or a linear stapler. The anastomosis must be performed distally enough to prevent the duplication from collecting stool or forming an obstructive mass that could lead to compressive symptoms such as constipation. Alternatively, the distal portion of the duplication can be resected, which is an ideal approach when the rectum is normal and the duplication is anatomically distinct. In cases where the colonic duplication is connected to the urinary tract or vagina, separation of the duplication may be performed, followed by an endorectal mucosectomy of the distal segment like the soave procedure.

Mucosectomy and rectal transection of the fistula under peritoneal reflection can be performed transabdominally and a posterior sagittal approach can be used to excise the fistula; this prevents vaginal discharge of mucus from the residual rectal pouch as was done in our case. The muscle cuff is subsequently closed, and an end-to-side anastomosis is created to connect the remaining distal end of the duplicated colon to the normal colon. However, the separation of the shared wall in rectal duplication and the excision of associated fistulas present significant challenges, often necessitating complex pelvic dissection that risks compromising surrounding healthy tissues and may disrupt the continence mechanism of the normally located anus.^{6,9,10}

In cases where duplication of the hindgut is found unexpectedly at the initial laparotomy for stoma formation in anorectal malformations, sigmoidostomy has been shown to be well tolerated as it does not interfere with subsequent reconstruction and facilitates radiologic evaluation of complex anatomy.¹¹

Generally, patients with isolated cystic or tubular duplications of the stomach, small intestine and colon have excellent outcomes, some studies reporting a low mortality or morbidity around 4-8% after surgical management. Postoperative complications have been rarely reported in the literature, with a few authors documenting cases requiring reoperation for incomplete division of the distal rectal septum and death resulting from persistent sepsis. Patients presenting with associated anomalies or complication such as total gastrointestinal tract obstruction, bleeding, perforation and mesenteric vascular involvement often have a more complicated and less favourable prognosis. When treated prior to the onset of complications, total tubular colon duplication generally carries a favourable prognosis, highlighting the critical importance of early diagnosis.^{5,6,9}

Conclusion

We report an 11-month-old girl with an extremely rare association of colonic duplication with a recto-vestibular fistula with mild symptom of difficulty in defecation and irregular bowel movements. The diagnosis was made during prior colostomy surgery, initially aimed at managing the anorectal malformation. Proper examination and radiologic evaluation, even before colostomy exploration through examination, fistulogram, or contrast enema, will help delineate the anatomy in such cases, especially in obstructed cases. Colostomy at initial laparotomy is useful for further radiographic evaluation and future reconstruction. Colostomy should be made proximal to the duplication, if possible, to determine the best surgical option at a later stage, reduce the number of operations, and make the procedure technically easier. Colonic resection to remove the duplicated colonic segment is the main option in the management of a duplicated colon. Options to manage a duplicated colon that cannot be resected include cutting the septum to turn it into a single colonic duct. Limited dissection of the submucosal plane preserves the duplicated orthotopic rectal muscle wall to ensure normal continence in the future.

REFERENCES

- AbouZeid, A. A., Mohammad, S. A., Ibrahim, S. E., Fagelnor, A., & Zaki, A. (2019). Late diagnosis of complete colonic and rectal duplication in a girl with an anorectal malformation. *European Journal of Pediatric Surgery Reports*, 7(01), e47–e50.
- Ahmed, S., Saifuddin, M. S., Ng, C. Y., Salwati, N. F., Hayati, F., & Ismail, S. (2021). Congenital colorectal tubular duplication in an infant: A tale of radiological diagnostic challenges. *Radiology Case Reports*, 16(11), 3289–3294.
- Aworanti, O., Twomey, E., & Awadalla, S. (2014). Terminal ileum and total colonic duplication associated with a rectovestibular fistula in a child. *Irish Medical Journal*, 107(8), 241–242.
- Bahmad, H. F., Alvarado, L. E. R., Muddasani, K. P., & Medina, A. M. (2021). Tubular colonic duplication in an adult patient with long-standing history of constipation and tenesmus. *Autopsy and Case Reports*, 11, e2021260.
- Craigie, R. J., Abbaraju, J. S., Ba'ath, M. E., Turnock, R. R., & Baillie, C. T. (2006). Anorectal malformation with tubular hindgut duplication. *Journal of Pediatric Surgery*, 41(6), e31–e34. <https://doi.org/10.1016/j.jpedsurg.2006.03.045>
- Duess, J. W., Zimmermann, P., Hirsch, F. W., Graefe, D., Lacher, M., & Gosemann, J.-H. (2022). Complete Colonic Duplication and Perineal Fistula: Transanal Mucosectomy of the Ectopic Rectum. *European Journal of Pediatric Surgery Reports*, 10(01), e102–e106. <https://doi.org/10.1055/s-0042-1750028>
- Jellali, M. A., Mekki, M., Saad, J., Zrig, A., Elanes, I., Mnari, W., et al. (2012). Perinatally discovered complete tubular colonic duplication associated with anal atresia. *Journal of Pediatric Surgery*, 47(e19–e23). <https://doi.org/10.1016/j.jpedsurg.2012.03.045>
- Isa, M. M., Thayeb, A., Meildi, M., & Hutagalung, M. B. Z. (2021). Complete tubular duplication of colon presenting as rectovestibular fistula: A case report. *Bali Medical Journal*, 10(2), 526–528. <https://doi.org/10.15562/bmj.v10i2.2386>
- Ildstad, S. T., Tollerud, D. J., Weiss, R. G., Ryan, D. P., McGowan, M. A., & Martin, L. W. (1988). Duplications of the alimentary tract: Clinical characteristics, preferred treatment, and associated malformations. *Annals of Surgery*, 208(2), 184–189. From the Department of Pediatric Surgery, Children's Hospital Medical Center, Cincinnati, Ohio.
- Kaur, N., Nagpal, K., Sodhi, P., & Minocha, V. R. (2004). Hindgut duplication: Case report and literature review. *Pediatric Surgery International*, 20(8), 640–642. <https://doi.org/10.1007/s00383-004-1248-x>
- Piplani, R., Acharya, S. K., & Bagga, D. (2022). Recto-vestibular fistula with colonic duplication: report of three cases and review of literature. *Annals of Pediatric Surgery*, 18(1).
- Platt KD, Wu AJ, Nunez Lopez O, Wang KS, Bowen-Jallow KA. Anorectal malformation with colonic duplication and dual rectovaginal fistulae. *Int J Surg Res*. 2016;S1:002:6–9.
- Wu, X., Xu, X., Zheng, C., & Li, B. (2018). Tubular colonic duplication in an adult: case report and brief literature review. *Journal of International Medical Research*, 46(7), 2970–2975.
- Ziegler MM, Azizkhan RG, Allmen D, Weber TR. Operative pediatric surgery. 2nd ed. Cincinnati, Ohio; McGraw-Hill; 2014:651–663

Copyright holder:

Muhammad Noor Fitriansyah, Ken Sanden, Dadik Agus Soenjoto (2025)

First publication right:

Syntax Literate: Jurnal Ilmiah Indonesia

This article is licensed under:

