



A Rare Case of Colonic Triplication with Associated Imperforate Anus in a Newborn Male

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European J Pediatr Surg Rep 2022;10:e118–e121.

Abstract Keywords

- ▶ anorectal malformation
- ▶ colonic triplication
- ▶ imperforate anus
- ▶ rectovesical fistula
- ▶ colonic duplication

We present a case of a newborn male with imperforate anus who was found to have colonic triplication with a high rectovesical fistula. The case is presented with a focus on surgical strategies for the management of this rare malformation.

New Insights and the Importance for the Pediatric Surgeon

Colonic triplication with associated imperforate anus and rectovesical fistula is a rare congenital malformation that requires a complex surgical approach. In addition to correction of the imperforate anus and repair of any fistula, one must plan for the possibility of bowel/bladder incontinence by keeping the appendix for future use and creating a single colonic lumen for mechanical antegrade flushes

Introduction

Alimentary tract duplications are rare congenital lesions that can occur anywhere from the mouth to the anus. Large bowel duplications account for only a small percentage (~10%) of all alimentary tract duplications.¹ Triplication of the colon is even rarer, with only a handful of cases described in the literature. Colorectal tubular duplications may be associated with rectogenital or rectourinary fistulae, duplication of internal or external genitalia, or vertebral anomalies.¹ The surgical treatment of these complex hindgut duplications can be very challenging. Here we present a case of a newborn male with colonic triplication and imperforate anus with a high rectovesical fistula.

Case Presentation

A newborn male, born full-term, weighing 4kg, with no relevant prenatal history, was found to have an anorectal malformation (ARM) in the delivery room. He was taken to the operating room at birth to create a colostomy with mucus fistula. Upon division of the colon, he was noted to have three separate proximal colonic lumens and two distal colonic lumens. The ostomy and mucus fistulas were matured (▶**Fig. 1**) and the patient was referred to our center for further management.

Upon presentation to our center at 5 months of age, an exam under anesthesia, cystoscopy, and contrast studies of the colon were performed. There were three easily

received
November 15, 2021
accepted after revision
April 22, 2022

DOI <https://doi.org/10.1055/s-0042-1750318>.
ISSN 2194-7619.

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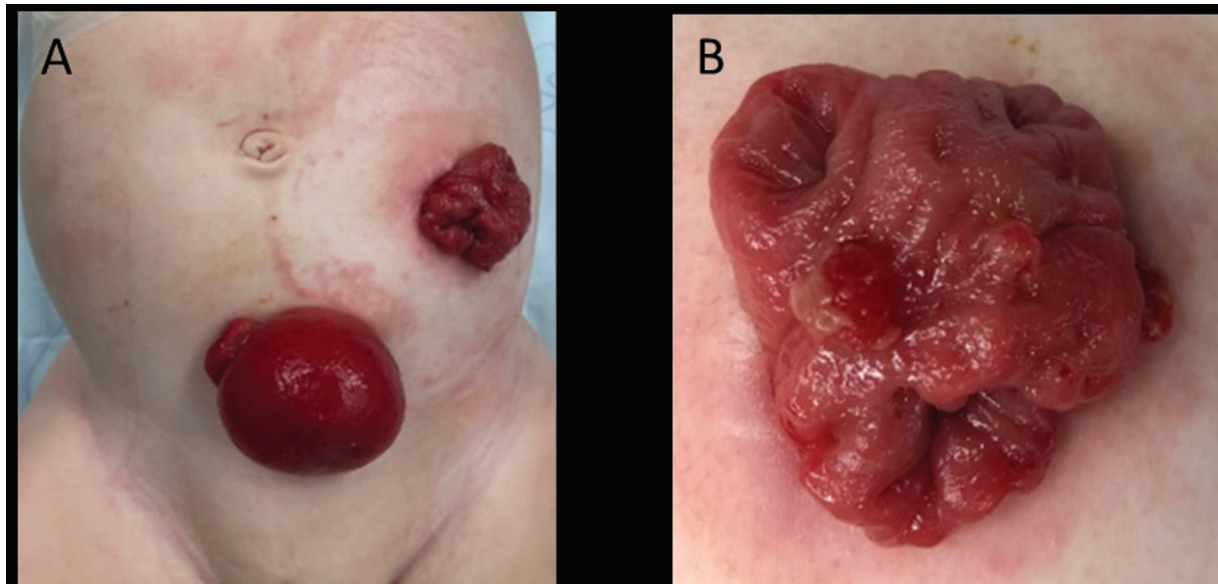


Fig. 1 Colostomy and prolapse mucus fistula (A). Three distinct lumens of the colostomy (B).

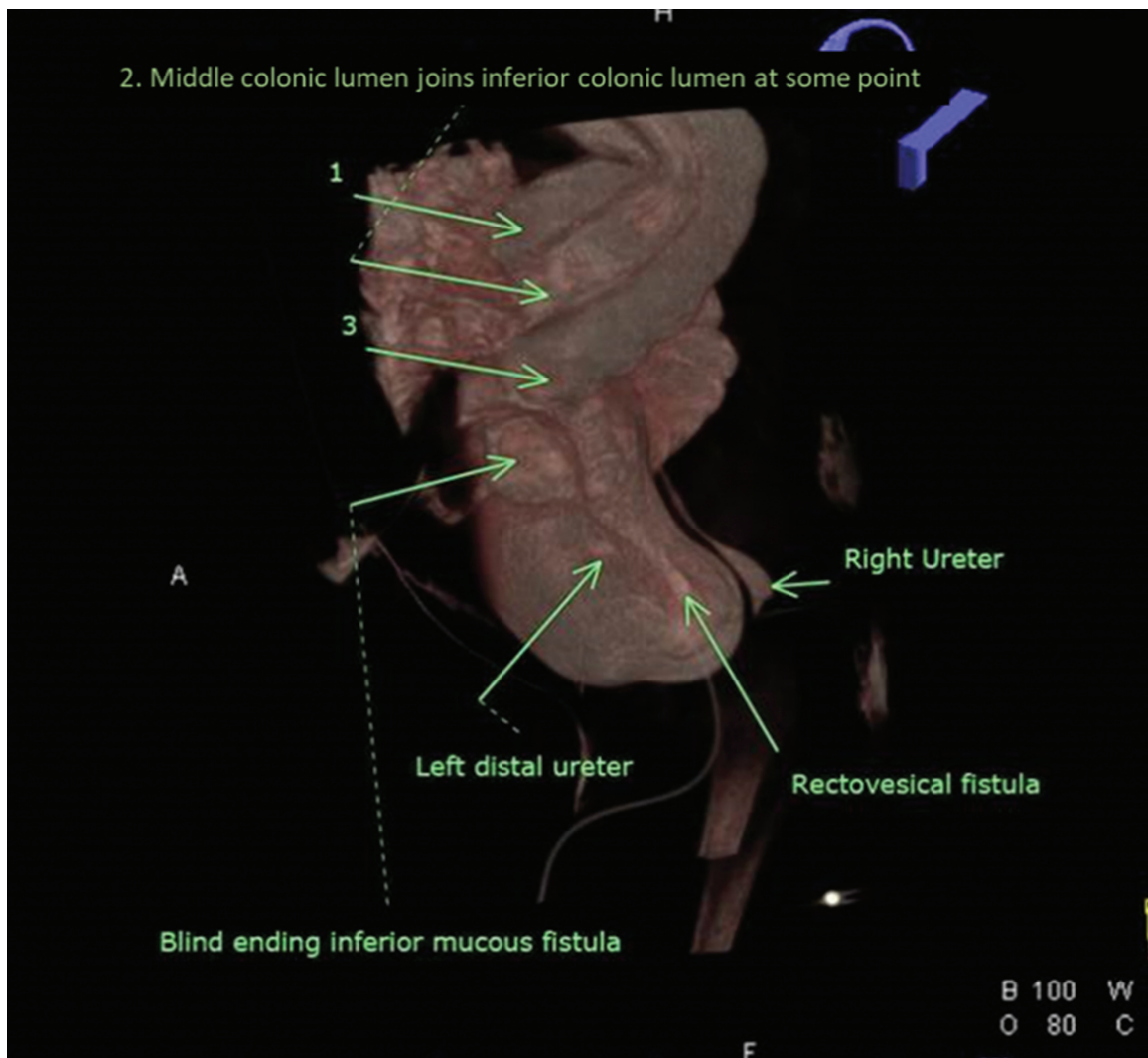


Fig. 2 High-resolution three-dimensional imaging showing three colonic lumens of proximal colon, distal colon with one blind ending lumen, and one fistulizing to the bladder.

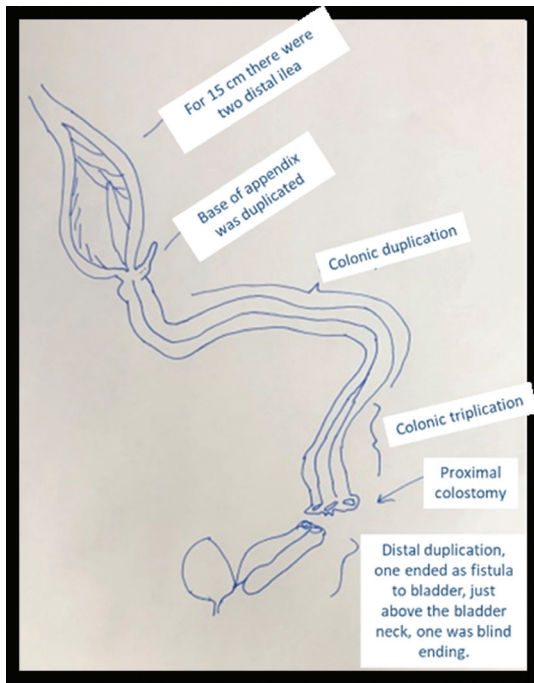


Fig. 3 Diagram of the operative findings.

identifiable colonic lumens on the end colostomy. The mucus fistula was prolapsed and only two lumens were able to be identified. There was no anus identified, but there was a good gluteal cleft. On cystoscopy, a colovesical fistula was seen just distal to the bladder neck, in close proximity to the left ureteral orifice. High-resolution and three-dimensional contrast imaging demonstrated two lumens of the distal colon with one ending as a fistula to the bladder and the other blind ending (→Fig. 2). Proximally there were three lumens in the descending colon, which merged to form two lumens around the splenic flexure. The colonic duplication continued all the way to the cecum (→Fig. 3).

The patient was taken to the operating room where an exploratory laparotomy was performed. The anal sphincters were marked at the start of the case using an electrical stimulator. An ileal duplication and fused appendiceal duplication were also identified intraoperatively (→Fig. 4). A stapler was used to create a single lumen in the ascending and descending colons (→Fig. 4). A posterior sagittal anorectoplasty (PSARP) was performed, the distal rectum connected to the bladder neck was separated, and then the two distal lumens were made into one with stapler and pulled through for the anoplasty. The colostomy was closed, and the thinner of the two distal ileums was resected. A diverting

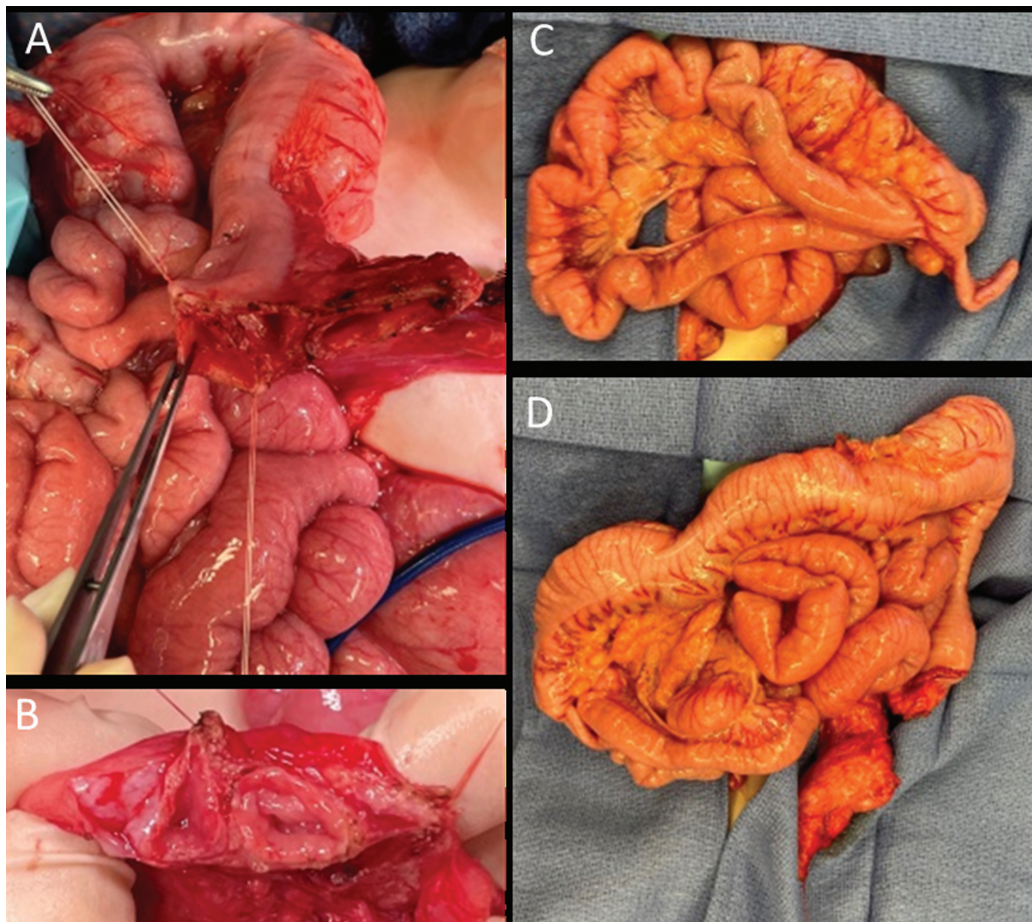


Fig. 4 Intraoperative findings. Three colonic lumens of colostomy (A). Two colonic lumens of mucus fistula (B). Ileal, cecal, and appendiceal duplication (C). Entire colon with normal outer appearance despite multiple internal lumens (D).

ileostomy was created to protect the repair and was subsequently reversed 6 weeks later. The patient is currently clean with Malone flushes and is working toward control of the flushes as a bridge to voluntary bowel movements.

Discussion

Triplication of the colon is extremely rare, with only a few cases reported in the literature.²⁻⁵ These complex patients create a unique surgical challenge for which there is no defined approach. In a patient such as this, diversion is the most appropriate initial step. The anatomy should be well delineated and a comprehensive surgical plan created prior to undertaking major reconstruction.

At reconstruction, creating a single distal rectum with no communication with the urinary tract is vital to success. Any remnant of the original fistula that remains can lead to urinary tract infections, kidney damage, and even affect continence. It is also important to use a muscle stimulator to premark the anal sphincters prior to dissection to place the rectum within the center of the muscle complex.

Another important consideration in the surgical approach to these patients should always include the possibility that they may need management of fecal or urinary incontinence in the future. Using the ARM continence index⁶ as a guide, we anticipated that this patient has moderate potential for bowel control because they have a high rectovesical fistula, a normal spine, and an intermediate sacral ratio. The ARM index is a nonvalidated way to guide conversations between clinician and parents of an ARM child, as it appears that prognosis for bowel control is dependent on the type of malformation, and the quality of the sacrum and spine. To account for this, we left the duplicated appendix untouched and created a single lumen channel at the start and end of the

colon so that if mechanical flushes are needed, antegrade flushes would achieve emptying of the entire colon at the same time. While we did leave a short area of duplication in the transverse colon, the two lumens were equal in size and communicating, so we do not anticipate any issues with the antegrade flushes he will likely need in the future.

Conclusion

Colonic triplication with associated imperforate anus and rectovesical fistula is a rare congenital malformation that requires a complex surgical approach. In addition to correction of the imperforate anus and repair of any fistula, one must plan for the future possibility of bowel/bladder incontinence by keeping the appendix for future use and creating a single colonic lumen for mechanical antegrade flushes.

Conflict of Interest

None declared.

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