CASE REPORT

# Unusual Recto-colonic Tubular Duplication in a Female Neonate

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## ABSTRACT

Complete tubular colonic duplication is exceedingly rare. A second ectopic opening in the perineum other than a normally cited anus could be an unusual presentation. We report an unusual case of recto-colonic duplication in a 16-day-old girl who presented with fecal discharge from a vestibular opening in addition to a normally situated anus. The diagnosis of total recto-colonic tubular duplication associated with a rectovestibular fistula and a normal anus was confirmed by barium enema and computed tomography scan with double contrast. At operation, we performed a long transanal incision of the common septum to create an anastomosis between the normal and duplicated colon with complete submucosal excision of the rectovestibular fistula. The patient was doing well at 3-year follow-up examination.

**Key words:** Gastrointestinal duplication; Anorectal malformation; Colonic duplication

## INTRODUCTION

Gastrointestinal duplications are a rare condition in children [1,2]. Rarest of them are the tubular duplications that affect several segments of the colon or the entire colon. A clinical diagnosis of colonic duplication is rarely made, possibly due to unfamiliarity with the syndrome and the non-specificity of clinical signs. Colonic duplication revealed by ectopic opening in the perineum is exceedingly rare, posing a problem in the differential diagnosis with anorectal malformations.

We report an unusual case of total recto-colonic tubular duplication associated with a normal anal opening and a fistulous opening in the vestibule in a female neonate. We present it for its rarity, unusual presentation and discuss pertinent review of literature.

## CASE REPORT

A 16-day-old girl, product of full-term normal vaginal delivery and birth weight of 3.4 kg, was brought to our department by her parents with the complaint of fecal discharge from a small opening in the vestibule since birth. She had regular bowel movements through a normally situated anus. There was no history of constipation, abdominal pain, or urinary problem. On physical examination, there was no palpable abdominal mass, the anal opening and the external genitalia were normal, and an opening in the vestibule just behind the vaginal opening discharging feces. There were no other obvious associated anomalies.

Computed tomography scan with double contrast showed a tubular recto-colonic duplication with proximal communication at the left colic angle level (Figure 1). It revealed also associated anomalies: A left ectopic kidney and spina bifida. There was no gastric heterotopia revealed on digestive scintigraphy.

Examination under anesthesia revealed the presence of a narrow fistula opening in the vestibule that would allow only size 8 Foley's catheter. On digital rectal examination, a long submucosal fistulous tract was palpable. A simultaneous contrast study through normal anus and the fistula opening was done. It showed a fistulous tract that extended right up to the sigmoid and apparently communicating with it. An endoscopic examination performed from

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fistula opening, confirmed the presence of the tubular duplication of colon.

Saline enemas were given the night before and on the morning of operation. Under general anesthesia, in the lithotomy position, the vestibular orifice was exposed. The fistulous opening was dissected submucosally from vestibular orifice to the anterior rectum wall and fistula was resected close to it.

A transanal distal communication was made by creation of a communicating window by dividing the mucosal septum between the two sigmoid colons and rectums as low down as possible.

The child was kept without oral intake for 2 days and received antibiotics intravenously for 48 h. The perineum was kept clean by frequent local cleansing. She made an uneventful post-operative recovery and was started on diet after 48 h. She continues to do well with no constipation or bowel-related complaints at 3-year follow-up.

## DISCUSSION

Colonic duplications account only for 4–18% of all gastrointestinal duplications [1,2]. Majority of them (60–80%) may be asymptomatic and remain undiagnosed for years or diagnosed later incidentally. If symptomatic, they manifest by obstruction, bleeding, constipation, and perforation. Few cases revealed by malignancy are also reported [3].

Both lumina may be unobstructed and function normally as two perineal ani or terminate distally blindly as imperforate anus of one or both lumina. In some cases, the ventral colon may end as a rectourinary, rectovaginal, or vestibular fistula [4]. Our case was revealed by ectopic opening of the duplicated colon which is one of the rarest presentations. During the search of English language literature, we came across only five cases of total colon duplication with a normal anus and a vulvar or vestibular fistula [4-8].

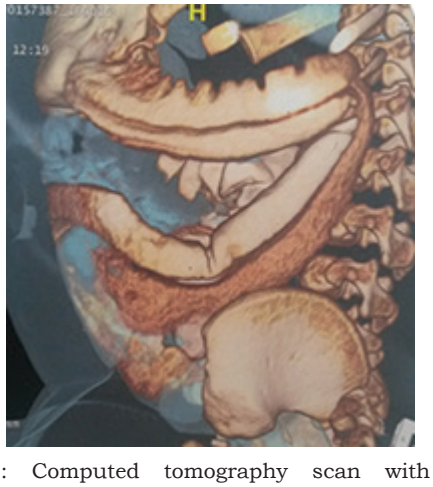


Figure 1: Computed tomography scan with double opacification showed a tubular recto-colonic duplication with proximal communication at left colic angle level

Yucesan et al. [5], in 1986 described a case of complete tubular colonic duplication (CTCD). The patient was a 1-year-old girl who had defecation problems for 3 months and a rectal bleeding after every defecation. On physical examination, in addition to the anus, a vestibular opening was also noticed. There were no associated malformations. The second case of a 3-month-old girl presented with complaints of passing feces through two openings in the perineum was described by Kaur et al. in 2004 [4]. The third case of newborn girl with a total colonic duplication was presented by Sarpel et al. in 2005 [6] with one colon ending in the rectum and the other emptying into a vestibular fistula. There was also a 7-cm diverticulum arising off the primary cecum. The two colons shared a common wall, but there was no communication. Karkera et al. in 2015 [7] described the fourth case of a 2-month-old female with a Y-shaped tubular colonic duplication which presented with a rectovestibular fistula and a normal anus. Kothari et al. in 2015 [8] described a case of a 4 ½-year-old girl with total colon duplication with a normally situated anus, vestibular fistula, associated with double bladder, and urethra.

The key to successful management of these rare cases is recognizing the entity with the aid of radiologic imaging studies. Yousefzadeh et al. reported that opacification of two colons is the diagnostic sign of colonic duplication, with simultaneous evaluation of both colons being facilitated using contrast media of two different densities [9]. However, for duplications with communication to the normal bowel, the barium enema may be more suitable to accurately delineate the anatomy of the duplicated segment.

Duplications should be treated for local control of symptoms including obstruction, bleeding, and possible septic sequelae and to prevent the risk of malignant change.

The treatment of choice consists of resection of the duplicated bowel with an extension of 2 cm in the normal bowel. Although malignant changes have been reported in adults [10], colorectal duplications are essentially benign lesions and radical surgical excision is not required. Furthermore, total resection of a long tubular duplication represents a real challenge, due to the common blood supply at the two organs. Hence, for patients with long segment involvement, a blood supply not amenable to resection and with no gastric heterotopia, conservative management is appropriate. Various surgical techniques have also been described. A long colostomy and incision of the common septum to cre-

ate an anastomosis between the normal and duplicated colon have been successfully used, but in such a surgical approach, the division of the lowest part of the septum may be difficult. A small blind pouch of rectum may be left, which could result in fecal impaction, constipation, or a recurrent fistula [4,5]. Yucesan et al. [5] in 1986 dissected the fistula tract from below and created a large anastomosis between the duplication and the normal colon to eliminate any blind end. However, they only joined the distal portion of the two lumens. Kaur et al. [4] created a communicating window of 8 cm between the two sigmoid and the rectum as low as possible with excision of fistula from below. Recurrence of rectovaginal fistula was noted in their patient. Sarpel et al. [6] resected the diverticulum, and the duplicated terminal ileum, and created an end-to-end ileoileostomy between the proximal ileum and the remaining terminal ileum. The shared colonic wall was divided proximally and distally with several applications of a stapling device, uniting the lumens at these points. The distal duplicated rectum was excluded from the fecal stream by transecting with a stapler, leaving a blind pouch to the fistula. In the case presented by Karkera et al. [7] a high sigmoid loop colostomy was done which resulted in four stomal lumens. Later, an endorectal mucosal resection of the duplicated distal segment till the colostomy site was done, with division of the septum of the proximal segment. Karkera et al. in 2015 [7] made a distal end to side anastomosis of the duplicated segment to the native colon ensuring emptying of the segment, eliminating blind end and complete submucosal excision of the fistulous connection preserving integrity of vagina and normal pelvic musculature.

In our case, there was no gastric heterotopia in digestive scintigraphy. Hence, there was no risk of malignant change or digestive tract bleeding. The conservative management was appropriate, by dividing the distal mucosal septum between the two sigmoid colons and rectums as low down as possible and a submucosal excision of the rectovestibular fistula. We also found that the endorectal approach manages the problem of the blind end without compromise of the continence mechanism and creates a balance between the rates of successful surgical management of CTCD and the pain, disability, and functional implications due to abdominoperineal approach. The result was excellent in our case.

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