

Ahmad Khaleghnejad Tabari (MD)^{*1}
Alireza Mirshemirani (MD)¹
Nasibeh Khaleghnejad Tabari (MD)¹

1- Pediatric Surgery Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

* **Correspondence:**
Ahmad Khaleghnejad Tabari, Pediatric Surgery Research Center, Mofid Children's Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

E-mail: khalegh@ams.ac.ir
Tel: 0098 21 22908181
Fax: 0098 21 22908181

Received: 29 Nov 2011
Revised: 13 Jan 2012
Accepted: 6 Feb 2012

Complete colonic duplication in children

Abstract

Background: Complete colonic duplication is a very rare congenital anomaly that may have different presentations according to its location and size. Complete colonic duplication can occur in 15% of gastrointestinal duplication. We report two cases of complete colonic duplications, and their characteristics.

Case Presentation: We present two patients with complete colonic duplication with different types and presentations. Case 1: A 2-year old boy presented to the clinic with abdominal protrusion, difficulty to defecate, chronic constipation and mucosal prolaps covered bulging (rectocele) since he was 6 months old. The patient had palpable pelvic mass with doughy consistency. Rectal exam confirmed perirectal mass with soft consistency. The patient underwent a surgical operation that had total tubular colorectal duplication with one blind end and was treated with simple fenestration of distal end, and was discharged without complication. After two years follow up, he had normal defecation and good weight gain. Case 2: A 2-day old infant was referred with imperforate anus and complete duplication of recto-sigmoid colon, diphallus, double bladder, and hypospadiasis. After clinical and paraclinical investigations, he underwent operations in several stages in different periods, and was discharged without complications. After four years follow up, he led a normal life.

Conclusion: The patients with complete duplication have to be examined carefully because of the high incidence of other systemic anomalies. Treatment includes simple resection of distal common wall, fenestration, and repair other associated anomalies.

Keywords: Complete duplication, Colon, Surgical Treatment, Children.

Caspian J Intern Med 2012; 3(2): 436-439

Duplication of the alimentary tract is an uncommon occurrence in pediatric patients and can affect any portion of the gastrointestinal tract (GIT) from the mouth to the anus. Alimentary tract duplications typically share a common wall and vascular supply and are either cystic or tubular structures (1, 2). Duplications most commonly affect the esophagus and ileum (the most common location) with only 4% to 18% affecting the colon (2). Their etiology is of embryonic origin and several theories have been proposed. However, none of these theories alone is able to explain the full diversity of these lesions. Although they can present at any age, more than 80% of the cases present before the age of 2 as an acute abdomen or bowel obstruction (3, 4).

GIT duplications are most single, tubular, or cystic and most often located on the mesenteric side of the native alimentary tract structure. The types of duplication cysts; 1-cystic (80%) spherical in shape and no communication with the bowel lumen. 2-tubular duplications (20%) may communicate with the normal intestine at one or several points along the common wall and usually extend above and below the diaphragm (5). The symptoms are often related to the location of the duplication: oral and esophageal lesions can create respiratory difficulties, whereas, lower GI lesions may cause nausea, vomiting, bleeding, perforation, or obstruction (6, 7).

Case Presentation

Case 1: A 2 year old boy was presented to clinic with abdominal protrusion, difficulty to defecate, chronic constipation and prolapse of mucosa covered bulging (rectocele) since he was 6 months old. In the examination, the abdomen was soft with palpable pelvic mass with doughy consistency. The rectal exam confirmed perirectal mass with soft consistency.

The plain abdominal x-ray showed dilated bowel loops and soft tissue pelvic mass that pushed the rectum anteriorly and the cervical hemi-vertebra.

Barium enema and abdominal and pelvic CT scan confirmed the same finding. Ultrasonography revealed the absence of right kidney that was confirmed by DMSA scan, VCUG was normal.

Laparotomy was performed, there was a complete colorectal duplication with one blinded end and accumulation of huge amount of stool that protruded as mucosa covered bulging during defecation (rectocele). The duplicated colon had common mesentery and resection of one part was impossible (figure 1).



Figure 1. Total colo-rectal duplication of colon in case 1.

The mucosal web was resected and the two ends of duplicated colon were fenestrated to each other and opened to common anal canal (figure 2), all fecal impaction was evacuated (figure 3), the patient had a very good post operation recovery and was discharged from the hospital in a week time.

In a two-year follow up, he had normal defecation and good weight gain without any problem with duplicated colon.



Figure 2. Fenestration of two end of duplicated colon.



Figure 3. Post evacuation of duplicated colon in our case 1 patient.

Case 2: A 2- day old male newborn was referred to us because of distended abdomen, abnormal genitalia and imperforate anus. He was the first child of a family of 10 years marriage. The birth history was normal. The parents were relatives. Examination of genitalia revealed a well formed double penis with normally located and functioning urethra, left penis had proximal hypospadias. There were normal penile shaft and bifid scrotum with each compartment containing a testicle. Perineal examination showed imperforate anus. The values of blood analysis were within normal ranges. Abdominal sonography showed distended bowel loops and bilateral normal kidneys. The day after admission, he underwent laparotomy and colostomy, the colon was distended and duplicated at the level of the recto- sigmoid colon (figure 4).

Two months later, we performed an intravenous pyelography which showed normal kidneys and ureters. Voiding-cystourethrography revealed double bladder and urethra, but no vesicourethralreflux. At 4 months of age, the

surgical exploration of genitourinary tract showed abnormal position of bladder and urethra .We performed cystoplasty and reimplantation of left ureter in a single bladder, left side urethra with hypospadias was resected and bifid scrotum repaired. The post- operative days passed without complications, so the patient was discharged 2 weeks later. At one year old after colostogram (figure 5), abdominal CTS (figure 6) and total colon evaluation, laparotomy, resection of duplicated recto-sigmoid colon and pull-through was performed, three months later colostomy closure was carried out. He was under observation until the age of 4 when his parents decided to leave Iran due to family problems.



Figure 4. Distended and duplicated colon.



Figure 5. Colostogram in case number two.

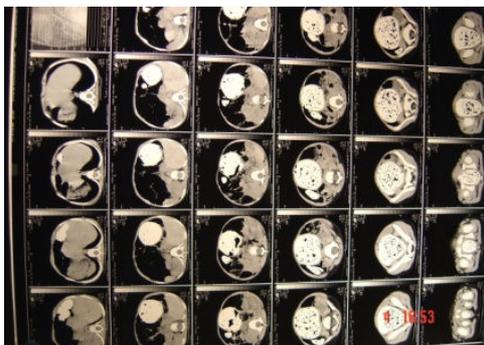


Figure 6. Abdominal CTS in case 2.

Discussion

Gastrointestinal (GI) duplications are congenital anomalies of partial or complete parallel growth of a twin segment of intestine that may be cystic or tubular in form. One of the most widely accepted theories is the abnormality of the embryonic gut that results in the formation of a diverticulum, a cyst, or twinning of a bowel segment (8). GI duplication is seen in approximately 1: 4500 autopsies. The distal esophagus and distal ileum are the most frequent sites involved; however, GI duplications can occur anywhere in the GI tract (9). Synchronous gastrointestinal duplications occur in as many as 15% of patients.

The presence of heterotopic mucosa (eg.gastric mucosa) in duplication can lead to peptic ulcerations, bleeding, and perforation with peritonitis. Neoplastic changes have been reported in gastrointestinal duplications. Colonic duplications represent 15% of duplications and may be seen in either cystic or tubular forms. Cystic colonic form has variable presentations (asymptomatic, volvulus, intestinal obstruction), it may be isolated or present with a fistula to the skin, urinary tract, or normal colon (7). Tubular colonic duplication is usually asymptomatic (with the exception of the cosmetic problem of duplicated genitalia), this type is often associated with duplication of the anus, vagina, and penis as we presented our case number two (9, 10). Patients presenting with complex tubular colonic duplications may not require a surgical approach if internal communication of the duplication is adequate and the colon is normal (11).

Symptoms are determined by the location of the cyst. In the colon, symptoms include constipation, hematochezia, rectal prolapsed, fistula, hemorrhoids and abscess. We had some of these symptoms in our case one (5). Preoperative diagnosis of alimentary tract duplications is often difficult. Ultrasonography is also helpful to establish a preoperative diagnosis and may similarly be used as a screening tool to address the 10-20% of multiple lesions (6). CT scanning of the chest or abdomen is helpful in establishing a diagnosis. Although preoperative diagnosis has been made with the aid of radiological studies such as a barium enema; the majority of cases have been diagnosed at the surgery or upon pathological examination. Barium enema studies are considered essential for the diagnosis of tubular colonic duplication, with opacification of two colons being the diagnostic sign (8, 12).

Barium enema and abdominal and pelvic CT confirmed the diagnosis in our case one. Most authors recommend that

once the diagnosis is made, an elective surgical procedure should be performed to avoid complications and to perform the procedure in an optimal state of the patient, but few think that only symptomatic duplications should be surgically treated (3). The recommended surgical procedure is excision of the duplication (3). Colorectal duplications are benign lesions. For this reason, surgical excision should not be radical but complete resection of the duplication along with the part of colon involved should be made (13). Treatment, usually in the form of surgical resection, is reserved for symptomatic cases (14). Indications for surgical intervention depend on acute setting, symptoms, types of duplications and associated anomalies.

In conclusion, the patients with complete duplication have to be examined carefully because of the high incidence of other systemic anomalies. Treatment includes simple resection of distal common wall, fenestration, and repair of the other associated anomalies.

Acknowledgments

The authors would like to thank the Pediatric Surgery Research Center of Shahid Beheshti University of Medical Sciences for their help in preparing the data.

Conflict of interest: There was no conflict of interest.

References

1. Limas C, Soutanidis C, Kirmanidis MA, Tsigalou C, Tsirogianni O. Abscess formation of spherical-shape duplication in the splenic flexure of the colon: case report and review of the literature. *Cases J* 2009; 2: 158.
2. Bissler JJ, Klein RL. Alimentary tract duplications in children: case and literature review. *Clin Pediatr* 1988; 27: 152-7.
3. Kekez T, Augustin G, Hrstic I, et al. Colonic duplication in an adult who presented with chronic constipation attributed to hypothyroidism. *World J Gastroenterol* 2008; 14: 644-6.
4. Merrot T, Anastasescu R, Pankevych T, et al. Duodenal duplications: clinical characteristics, embryological hypothesis, histological findings, treatment. *Eur J Pediatric Surg* 2006; 16: 18-23.
5. Ha J, Uy K, Gerlach K, et al. 4 year old with hematochezia. VCU Health System, Virginia Commonwealth University, Pediatric Radiology, Case of the Week. Answer to case of the week: Oct 14-Oct 21 2010. Available at: URL: <http://www.pedsradiology.com/Historyanswer.aspx?xid=413&fid=1>. Accessed Dec 19, 2011.
6. Sexena AK. Gastrointestinal duplications. 2010; Available at: www.emedicine.medscape.com/article/936799-overview. Accessed Feb 22, 2011.
7. Aggarwal H, Singh S, Patidar N. Multiple intraabdominal cysts could be a duplication of intestine: a cases report and treatment protocol. *Internet J Surg* 2008; 15. Available at: URL: <http://www.ispub.com/journal/the-internet-journal-of-surgery/volume-15-number-2/multiple-intraabdominal-cysts-could-be-a-duplication-of-intestine-a-case-report-and-treatment-protocol.html>. Accessed Nov 17, 2011.
8. Kabay S, Yucel M, Yaylak F, et al. Combined duplication of the colon and vermiform appendix in an adult patient. *World J Gastroenterol* 2008; 14: 641-3.
9. Mirshemirani A, Roshanzamir F, Shayeghi SH, Mohajezadeh L, Hasas-yeganeh SH. Diphallus with imperforate anus and complete duplication of recto-sigmoid colon and lower urinary tract. *Iran J Pediatr* 2010; 20: 229-32.
10. Miki T, Seiich H, Motofumi T, Ikkei T. An experience of tubular colonic duplication with imperforate anus. *J Japanese Society Pediatr Surg* 2005; 41: 212-5.
11. Clonic duplication. 2009; Available at: URL: <http://rad.usuhs.edu/medpix/new-topic.Html?Mode=single&recnum=9123&quiz=#pic>. Accessed Dec 26, 2011.
12. Singhal A, Gothi R, Singhal S. Radiological quiz- Abdomen. *Indian J Radiol Imag* 2006; 16: 965-7.
13. Holcomb GW 3rd, Gheissari A, O'Neill JA Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. *Ann Surg* 1989; 209: 167-74.
14. Abeygunasekera AM, Fathiehu F, Duminda MT, Jayasinghe R. A case of bladder and colonic duplication. *Saudi J Kidney Dis Transpl* 2008; 19: 87-9.