CASE REPORT

A case of sigmoid colon duplication in an adult woman

Abdulla Hassan Al-Jarroof,¹ Faisal Al-Zayer,² Abdul-Wahed Nasir Meshikhes³¹Surgery Department, Qatif Central Hospital, Qatif, Eastern Province, Saudi Arabia²Radiology Department, Qatif Central Hospital, Qatif, Eastern Province, Saudi Arabia³Department of Surgery, King Fahad Specialist Hospital, Dammam, Eastern Province, Saudi Arabia**Correspondence to**

Dr Abdul-Wahed Nasir Meshikhes, meshikhes@gmail.com

Accepted 21 July 2014

SUMMARY

Colonic duplication is a rare congenital anomaly that is often diagnosed in childhood, but may go unrecognised until adulthood. It often presents with chronic abdominal pain and constipation, and the preoperative diagnosis may be difficult. We present a case of sigmoid duplication in a 33-year-old Indonesian woman who presented with right-sided colicky abdominal pain and vomiting. Clinical examination was unremarkable and radiological investigations raised the possibility of a giant colon diverticulum. The patient underwent exploratory laparotomy that revealed a tubular sigmoid duplication. A sigmoid colectomy with end-to-end anastomosis was performed. She was discharged a week later and remained well at 1 year follow-up. Colon duplications rarely present in adult life and the accurate diagnosis is often made at laparotomy.

BACKGROUND

Colonic duplication is a rare congenital anomaly. It can either be spherical or tubular and may or may not communicate with the bowel.¹ The condition is often diagnosed in childhood, but may go unrecognised until adulthood when it presents with chronic abdominal pain and constipation. It rarely presents acutely due to obstruction, volvulus, perforation or bleeding.² The diagnosis is usually not entertained preoperatively and only made at exploratory laparotomy.¹ The majority of reported cases are in children and, hence, the number of reported cases in the non-paediatric population is limited. We report a case of sigmoid colon duplication in an adult woman. The preoperative diagnosis was mistaken for giant colonic diverticulum and the definitive diagnosis was only made at laparotomy.

CASE PRESENTATION

A 33-year-old Indonesian woman presented as an emergency with a 1-day history of right-sided colicky abdominal pain that increased in severity for 2 days prior to the presentation and was not associated with nausea, vomiting or aggravating/relieving factors. She denied any history of chronic constipation, except for during the past 4 days. Her surgical history was unremarkable except for an appendectomy 4 years earlier.

Clinical examination revealed normal vital signs (temperature 37.0°C, pulse 80 bpm and a blood pressure of 124/84 mm Hg). The abdomen was soft, lax and a fusiform mass was felt occupying the right side of the abdomen, extending from the right hypochondrium to the left pelvis. The upper part of the mass was slightly tender on deep

palpation and was firm in consistency and non-indentable. Bowel sounds were slightly increased but non-obstructive and rectal examination was normal with an empty rectum.

INVESTIGATIONS

Routine blood tests showed anaemia (haemoglobin 10.5 g%) and leucocytosis ($15.2 \times 10^3 \mu\text{L}$).

Plain abdomen X-ray was inconclusive, but demonstrated a large air-filled structure in the right upper quadrant. CT of the abdomen revealed a 9×8 cm structure arising from the sigmoid colon and crossing to the right side and extending up adjacent to the hepatic flexure and the edge of the liver, with an air-fluid level (figure 1, left panel). MRI revealed a huge extrabowel loop arising from the sigmoid colon and extending to the right side and running up lateral to the ascending colon to the liver (figure 1, left panel).

It was filled with gas and faecal matter. No colonoscopy was performed, but barium enema showed an extrabowel loop arising through a short narrowed neck from the sigmoid colon (figure 2).

DIFFERENTIAL DIAGNOSIS

- Giant colonic diverticulum
- Colonic duplication
- Colonic volvulus
- Duplication cyst

TREATMENT

The patient underwent exploratory laparotomy, which revealed a tubular sigmoid duplication that was adherent to the sigmoid colon and extending to the right side of the abdomen (figure 3). This duplication and the involved sigmoid colon were excised surgically by sigmoid colectomy and stapled primary end-to-end colocolic anastomosis.

The histopathology revealed a segment of sigmoid colon with duplication. The blind segment was covered by serosa with prominent blood vessels and measured 31 cm in length and 5–9 cm in diameter. It had a thick wall and ran parallel to the resected segment. Part of the duplication was adherent to the original sigmoid colon without communicating with it except through a single ostium (figure 3). The duplicated segment ended blindly and was filled with faecal matter without evidence of mucosal dysplasia or malignancy.

OUTCOME AND FOLLOW-UP

The patient's postoperative recovery was uneventful and she was discharged home 7 days later in good

CrossMark

To cite: Al-Jarroof AH, Al-Zayer F, Meshikhes A-WN. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-203874

Figure 1 CT of the abdomen (transverse view) showing the air-filled cavity in the right upper quadrant, which was mistaken for giant colon diverticulum (left panel). A single coronal view of MRI film showing the duplicated colon (right panel).

general condition. She remained well and symptom free at 1 year follow-up.

DISCUSSION

Alimentary tract duplication is a very rare congenital malformation that occurs most commonly in the small bowel of the gastrointestinal tract. It has a smooth muscle coat and an epithelial mucosal lining.³ It is cystic in more than 80% of cases and tubular in the rest.⁴ Colorectal duplication is the least common, representing only 13% of all gastrointestinal duplications.⁵ Mourra *et al*⁶ reported a series of seven cases of colorectal duplications that presented with abdominal pain (n=4) and obstruction (n=3); only one duplication was located in the sigmoid colon and another one in the transverse colon while the remaining five were arising from the caecum.

Tubular colon duplications are often associated with abnormalities of the anus, vagina or penis.^{7 8} Also, there may be an

association between colon duplication and colovesical fistula.^{9 10} In this case, although the duplication was tubular, it was not associated with any other congenital anomaly.

Colonic duplication presents with a wide spectrum of clinical and radiological features, creating diagnostic difficulties for the clinician and radiologist.¹¹ However, it presents commonly with chronic colicky abdominal pain and long-standing constipation with or without abdominal mass or distension. In recent years, the liberal use of colonoscopy and abdominal CT scans may have helped to establish the diagnosis preoperatively with greater confidence.¹¹ Colonoscopy is not considered helpful except in cases with a large ostium.¹²

Duplication may clinically present with volvulus, perforation or, most commonly, intestinal obstruction due to compression of the normal adjacent bowel by the expanding blind end of the duplication. It may also present with signs and symptoms of diverticulitis.¹³ However, it rarely presents with peritonitis due

Figure 2 Barium enema showing the duplicated colon filled with air and faeces.

Figure 3 Operative view of the sigmoid colon duplication (upper panel). Note its close attachment to the sigmoid colon, but without communication except through a single ostium. The lower panel shows the resected specimen of the duplicated colon and the involved sigmoid colon.

to perforation or bleeding,² unless the mucosa has an ectopic gastric mucosa that ulcerates, with subsequent bleeding or perforation.^{13 14}

Duplication is often diagnosed in the first 2 years of childhood, but in some cases it may escape detection due to the vague and mild nature of the symptoms. However, some cases of duplication in adults are found either incidentally or—as in this case—at workup for some vague and non-specific abdominal symptoms. Generally speaking, if there are no other associated malformations, colon duplications may go undiagnosed until complications develop in later adult life.

In this case, the expanded air-filled blind end of the duplication led to the common mistaken preoperative diagnosis of giant colon diverticulum. This is a rare clinical entity which is defined as a diverticulum equal to or larger than 4 cm in size and most commonly arises from the sigmoid colon and in the majority of cases is associated with diverticular disease.¹⁵

Based on their aetiological origin, McNutt *et al*¹⁶ divided giant colon diverticula into three types. Type I: pseudodiverticulum with a wall consisting mainly of granulation tissue.¹⁷ Type II: inflammatory diverticulum, which is secondary to a perforation of the mucosa and submucosa with a walled-off abscess cavity that communicates with the lumen. It is usually lined by fibrous tissue without any intestinal layer, making its differentiation from the duplication easy. Type III: true diverticulum, which contains

all bowel layers and communicates with the main lumen.¹⁸ Type III is most likely to represent a communicating duplication cyst and if it is large and elongated it is indistinguishable from the duplication. Choong and Frizelle¹⁹ divided giant diverticula into 2 types depending on the disease histology. Type 1 is a pseudodiverticulum with a fibrous wall with no muscular layer and it is acquired as it is associated with diverticular disease. While, type 2 is a true (congenital) diverticulum with a muscular wall, and is equivalent to McNutt's type III. Regardless of which classification is applied, colonic duplication may be indistinguishable from either McNutt's type III or Choong and Frizelle's type II giant colonic diverticulum, especially if it becomes elongated. This case was considered a duplication since there was no associated diverticular disease and it was very elongated (figure 3) to a degree that a diverticulum cannot reach; the latter most often tends to achieve a spherical shape. Moreover, in this case the duplication was lined by a mucosal layer which proved to be histologically free of dysplasia or malignancy.

In adults, colon duplication may also be misdiagnosed as carcinoma; Roberts *et al*²⁰ reported two cases of sigmoid colon tubular duplication in two elderly men in their 60s.

The diagnosis was mistaken for cancer preoperatively; even during surgery the differentiation between benign and malignant conditions was difficult due to extensive inflammatory reaction. The diagnosis was performed with confidence by histopathology.

Both were treated by resection and primary anastomosis.²⁰ Other preoperative differential diagnoses of colonic duplication include volvulus, intussusception and duplication cyst.²¹

Once the diagnosis is suspected, the recommended treatment is surgical resection of the duplication together with the attached normal colon segment. This eliminates the small but potential risk of future cancer development in the duplicated colon.²² Currently, the widespread use of laparoscopic colorectal surgery for benign and malignant conditions encouraged surgeons to apply this modality successfully in the surgical resection of colonic duplication.^{23–26}

We report this case of sigmoid duplication to highlight its vague presentation and elusive diagnosis in adult life, having been unrecognised or missed in childhood. Suspicion of the diagnosis should be borne in mind when treating an adult patient with chronic colicky abdominal pain and constipation. The appropriate treatment is either open or laparoscopic resection of the duplication together with the involved colon and primary anastomosis.

Learning points

- Colonic duplication is a rare congenital anomaly which is often diagnosed in childhood, but may go unrecognised until adulthood, especially if it is not associated with other congenital anomalies.
- It presents commonly with chronic colicky abdominal pain and long-standing constipation with or without abdominal mass or distension. The acute presentation is rare.
- The wide spectra of clinical and radiological presentations make preoperative diagnosis of colon duplication difficult.
- Its differentiation from McNutt's type III or Choong and Frizelle's type II giant colon diverticulum is difficult especially if the duplication is elongated.
- The recommended treatment is either open or laparoscopic surgical resection of the duplication together with the attached normal colon segment.

Contributors AHAI-J managed the case and contributed to the final draft. FA-Z did the radiological investigations and interpretations. He also contributed to the final draft. A-WNM searched the literature and wrote the manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Gross RE, Holcomb GW Jr, Farber S. Duplications of the alimentary tract. *Pediatrics* 1952;9:448–68.
- 2 Fotiadis C, Genetzakis M, Papandreou I, *et al.* Colonic duplication in adults: report of two cases presenting with rectal bleeding. *World J Gastroenterol* 2005;11:5072–4.
- 3 Shin KS, Lee NH, Kim SY. An unusual case of colonic duplication causing chronic constipation in a child. *J Pediatr Surg* 1999;34:1410–12.
- 4 Stern LE, Warner BW. Gastrointestinal duplications. *Semin Pediatr Surg* 2000;9:135–40.
- 5 Puligandla PS, Nguyen LT, St-Vil D, *et al.* Gastrointestinal duplications. *J Pediatr Surg* 2003;38:740–4.
- 6 Mourra N, Chafai N, Bessoud B, *et al.* Colorectal duplication in adults: report of seven cases and review of the literature. *J Clin Pathol* 2010;63:1080–3.
- 7 Banu T, Chowdhury TK, Hoque M, *et al.* Congenital double anus with total colon duplication: a case report. *J Pediatr Surg* 2007;42:E1–2.
- 8 Rodesch G, Dargent JL, Haller A, *et al.* An unusual presentation of a cystic duplication of the sigmoid colon entirely lined with squamous epithelium. *J Pediatr Surg* 2009;44:1831–4.
- 9 Prasil P, Nguyen LT, Laberge JM. Delayed presentation of a congenital recto-vaginal fistula associated with a recto-sigmoid tubular duplication and spinal cord and vertebral anomalies. *J Pediatr Surg* 2000;35:733–5.
- 10 Payne CE, Deshon GE Jr, Kroll JD, *et al.* Colonic duplication: an unusual cause of enterovesical fistula. *Urology* 1995;46:726–8.
- 11 Carr SL, Shaffer HA Jr, de Lange EE. Duplication of the colon: varied presentations of a rare congenital anomaly. *Can Assoc Radiol J* 1988;39:29–32.
- 12 Steenvoorde P, Vogelaar FJ, Oskam J, *et al.* Giant colonic diverticula. Review of diagnostic and therapeutic options. *Dig Surg* 2004;21:1–6.
- 13 Paulson EC, Mahmoud NN. Sigmoid colon duplication cysts. *Am Surg* 2008;74:250–2.
- 14 Correia-Pinto J, Romero R, Carvalho JL, *et al.* Neonatal perforation of a Y-shaped sigmoid duplication. *J Pediatr Surg* 2001;36:1422–4.
- 15 Zeina AR, Nachtiga A, Matter I, *et al.* Giant colon diverticulum: clinical and imaging findings in 17 patients with emphasis on CT criteria. *Clin Imaging* 2013;37:704–10.
- 16 McNutt R, Schmitt D, Schulte W. Giant colonic diverticula—three distinct entities. Report of a case. *Dis Colon Rectum* 1988;31:624–8.
- 17 Gallagher JJ, Welch JP. Giant diverticular of the sigmoid colon: a review of differential diagnosis and operative management. *Arch Surg* 1979;114:1079–83.
- 18 Saha SP, Roesch CB. A giant sigmoid diverticulum: report of a case. *Dis Colon Rectum* 1972;15:63–5.
- 19 Choong CK, Frizelle FA. Giant colonic diverticulum: report of four cases and review of the literature. *Dis Colon Rectum* 1998;41:1178–86.
- 20 Roberts M, Rabinovitch J, Felton CM, *et al.* Duplication of the sigmoid colon. *Ann Surg* 1959;150:904–8.
- 21 Domajnko B, Salloum RM. Duplication cyst of the sigmoid colon. *Gastroenterol Res Pract* 2009;2009:918401.
- 22 Heiberg ML, Marshall KG, Himel HS. Carcinoma arising in a duplicated colon. Case report and review of literature. *Br J Surg* 1973;60:981–2.
- 23 Park YA, Jung EJ, Han SJ. Laparoscopic resection of duplicated sigmoid colon under the guidance of intraoperative colonoscopy. *Surg Laparosc Endosc Percutan Tech* 2005;15:299–301.
- 24 Chang YT, Lee JY, Liao YM, *et al.* Laparoscopic resection of a giant retroperitoneal T-shaped duplication of descending colon. *J Pediatr Surg* 2008;43:401–4.
- 25 Kiu V, Liang JT. Laparoscopic resection of Y-shaped tubular duplication of the sigmoid colon: report of a case. *Dis Colon Rectum* 2010;53:949–52.
- 26 Yong YG, Jung KU, Cho YB, *et al.* Large tubular colonic duplication in an adult treated with a small midline incision. *J Korean Surg Soc* 2012;82:190–4.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow