

Antoine KACHI<sup>1</sup>, Fadi HADDAD<sup>2</sup>, Antoine GEAGEA<sup>3</sup>, Maroon J. TOHMEH<sup>4</sup>

Kachi A, Haddad F, Geagea A, Tohmeh MJ. Unusual case of jejunal duplication in a 64-year-old female. *J Med Liban* 2016; 64 (1): 50-53.

**ABSTRACT** • Gastrointestinal duplications are rare congenital malformations, which may occur anywhere in the gastrointestinal tract, more commonly in the ileum. Most cases appear during the first two years of life. Usually they are located at the paramesenteric side of the bowel and they share with the latter the same blood supply. Vague abdominal pain and distention are common presentations of duplications in adults, although many of them remain asymptomatic. We report a rare case of a jejunal duplication cyst in an elderly female patient, with atypical anatomic features.

Keywords: gastrointestinal duplications, paramesenteric side

#### INTRODUCTION

Gastrointestinal duplications are rare congenital malformations that may be cystic, tubular, or mixed [1].

The ileum is most commonly involved, however duplications may occur anywhere in the gastrointestinal tract, from the mouth to the anus [2]. Duplication cysts are composed of all the bowel wall layers (submucosa and muscularis propria) [3].

They tend to be at the paramesenteric side [4], sharing most of the time the same blood supply with the associated native bowel [5], and they rarely communicate with it [6].

Most cases appear in the first two years of life, but some of them may appear at an older age [7,8]. They are mostly encountered during laparotomy and misdiagnosed as intussusception, appendicitis, or gastrointestinal bleeding [5].

Clinical presentation depends on the site and size of the duplication [9]. Vague abdominal pain and distention are common presentations of duplications in adults, although many of them remain asymptomatic [10].

Few cases of jejunal duplications in adults were published. Clinical presentation, diagnostic modality, man-

Kachi A, Haddad F, Geagea A, Tohmeh MJ. Un cas inhabituel de duplication jéjunale chez une femme âgée de 64 ans. *J Med Liban* 2016; 64 (1): 50-53.

**RÉSUMÉ** • Les duplications gastro-intestinales sont des malformations congénitales rares, qui peuvent apparaître n'importe où dans le tractus gastro-intestinal, le plus souvent dans l'iléon. La plupart des cas apparaissent au cours des deux premières années de vie. Habituellement, les duplications sont situées du côté paramésentérique de l'intestin, et partagent avec ce dernier le même apport de sang. La douleur abdominale vague et la distension sont des présentations courantes de duplications chez les adultes, bien que beaucoup d'entre elles restent asymptomatiques. Nous rapportons un cas rare de duplication kystique du jéjunum chez une patiente âgée se présentant avec des caractéristiques anatomiques atypiques.

agement, and outcomes of each of the published cases are resumed (See Annex).

We report an exceptionally rare case of a jejunal duplication cyst in a 64-year-old female patient.

#### CASE REPORT

##### History of present illness

A 64-year-old female Lebanese patient presented to her gastroenterologist with a one-day history of abdominal pain, located at the right upper quadrant, crampy in nature, associated with postprandial nausea. Based on the physical exam with a positive Murphy sign, the physician ordered an abdominal ultrasound to rule out cholecystitis.

The patient returned to her doctor to show him the result of the ultrasound, which was a distended gallbladder with multiple small calculi, the largest one of 4 mm in size, but she was complaining of obstipation for the last two days. A contrast enhanced (IV and PO) CT scan for the abdomen and pelvis was ordered by the gastroenterologist, and showed dilated subumbilical small intestines of 5.5 cm caliber with a "feces sign" at this level, collapsed small bowel loops distal to the latter were also seen (Figure 1).

The patient was referred to our surgery unit after this presentation. A detailed encounter was done with her, where she reported a 15-year history of on-and-off obstipations that lasted each time for two to three days with recuperation of normal bowel movements after that.

*Past medical history:* hypertension and diabetes mellitus.

*Past surgical history:* no surgeries done before.

<sup>1</sup>Faculty of Medical Sciences, Lebanese University (FMS/LU), Laparoscopic Surgery, Beirut, Lebanon.

<sup>2</sup>Faculty of Medicine/Internal Medicine, St Joseph University, Beirut, Lebanon.

<sup>3</sup>Gastroenterology Department, Clinique du Levant, Beirut, Lebanon.

<sup>4</sup>General Surgery Resident, FMS/LU, Beirut, Lebanon.

Correspondance: *Antoine Kachi, MD.*  
e-mail: tonikachi@hotmail.com



**FIGURE 1**

CT scan of the abdomen showing the "feces sign".

*Physical exam:* distended abdomen, tympanic on percussion, and diffusely tender on palpation.

*Blood work* was done: CBCD, LFTs, amylase, lipase, LDH, CRP, urinalysis were all within normal limits.

### Management

Based on a diagnosis suspicious for small bowel obstruction, laparoscopy was scheduled urgently for cholecystectomy and exploration of the abdomen. The patient passed flatus few minutes before the operation.

Upon laparoscopy, cholecystectomy was done first, and a thorough inspection of the intestines starting from the ileo-cecal valve to the angle of Treitz showed a mid jejunal cystic mass of 3 to 4 cm diameter that was soft, located at the antimesenteric side (Figure 2). Dilated bowels proximal to it and collapsed intestines distal to the cystic malformation were also seen.

A 5 cm midline incision was performed superior to the umbilicus for resection of the cystic malformation with the adjacent intestine, followed by a latero-lateral anastomosis.

We considered the resected intestine as a jejunal diverticulum, and the piece was sent to the pathologist for further examination and diagnosis.

The patient was discharged four days after an uneventful postoperative course and recuperation of normal bowel movements.

### Histology

Section showed that the grossly described intraparietal tubular formation of the jejunum was represented by a duplicated segment, totally independent from the normal intestine, but sharing with it a common muscular layer. The lumen was covered by a normal mucosa and muscularis mucosa. No inflammatory changes. The rest of the

jejunum, including both surgical margins, was unremarkable. The intestinal duplication was of 3 cm in size.

Cholecystitis with cholesterosis were shown on gallbladder histology.

### DISCUSSION

Alimentary tract duplications are rare congenital anomalies that arise at about 8-9 weeks of gestation [3]. They occur in one of every 4000-5000 live births [6].

There are several theories explaining the duplication process, and these including the aberrant luminal recanalization theory, the diverticular theory, and the notochordal split theory which consists on neurenteric duplications associated with vertebral anomalies [11]. Duplications may be cystic, tubular, or mixed and can occur in any part of the gastrointestinal tract, but the terminal ileum is more commonly affected. They are adherent to the normal bowel, sharing the same blood supply usually, and rarely communicating with it [12]. They tend to occur more at the paramesenteric side [4].

Duplications usually appear clinically within the first two years of life when infants present with abdominal pain, occlusion of the intestines, or a palpable mass. However, they may be asymptomatic in some adults and remain undiagnosed for years because of their variable clinical presentation. Symptomatic adult patients present most commonly with a palpable mass, vague abdominal pain, distention, intestinal obstruction, and bleeding, but most of the patients remain asymptomatic [6,10,13]. They may be found incidentally during surgery [11].

In our case, the patient reported a 15-year history of on-and-off obstipations, and few days duration of right upper quadrant pain, that was related after investigations to cholecystitis.

However, the symptoms are related strictly to the site, type, shape, and size of the duplication [11-13]. In the jejunum, tubular duplication with a common lumen is more frequently seen, in contrast to the ileum where it looks like a diverticulum.

Distal ileal duplication should be differentiated from Meckel's diverticulum, even though the latter is usually



**FIGURE 2**

Jejunal duplication at the antimesenteric side.

present on the antimesenteric side of the intestine [13].

It is not easy to diagnose an enteric duplication cyst, because of the wide differential diagnosis that includes mesenteric and omental cysts, ovarian cysts, pancreatic cysts, and all intra-abdominal cystic masses [6].

The main preoperative diagnostic modalities of duplication consist on ultrasonography, barium radiography, CT scan, and MRI. At acute presentation the laparotomy/laparoscopy is the mainstay of diagnosis [3].

However, ultrasonography is important in differentiating the cystic consistency of duplications from solid tumors. It shows an inner echogenic rim of intestinal mucosa surrounded by hypo-echogenic rim of muscular wall. CT scan can define their anatomic location, and MRI can demonstrate their cystic nature [12].

The treatment of choice of small enteric duplications is surgical excision and small bowel resection with primary re-anastomosis [3]. Partial excision and stripping of the mucosa within the duplication is an option in longer tubular duplications, and that's to avoid short bowel syndrome. However, there is no consensus yet for the treatment of asymptomatic duplication cysts. But to avoid complications, including malignancy, surgical management is recommended for asymptomatic adults. Simple ligation and pedicle division without bowel resection is done for completely isolated duplication cysts [6].

In the present case, the patient was an elderly woman in contrast with the typical young age of presentation, with a history of obstipations for the last 15 years, presented for acute onset symptoms of right upper quadrant pain and nausea of one day duration related to cholecystitis diagnosed by ultrasound. We had "feces sign" on contrast enhanced CT scan that was done after the patient reported two days history of obstipation, and found to have a distended and diffusely tender abdomen on physical exam. What does "feces sign" mean?

It is defined as the presence of feculent material mingled with gas bubbles in the small bowel, due to delayed intestinal transit caused by undigested food, bacterial overgrowth, or increase in water absorption due to obstruction of the distal small bowel. This sign is the result of small bowel obstruction or severe small bowel metabolic or infectious diseases [14].

Surgery was planned regarding the small bowel obstruction picture, clinically and radiologically.

Laparoscopy showed us the atypical location of the duplication, which we considered as a diverticulum before the histological diagnosis was done. The cystic mass was located in the mid jejunum, on the antimesenteric side, in contrast with the literature where the duplication's typical location is on the paramesenteric side of the distal ileum. Complete resection of the cyst and the adjacent intestine was done with a primary side to side anastomosis, and the patient was discharged four days after a smooth postoperative course.

The on-and-off obstipations for the last 15 years were due to the food stagnation in the duplicated jejunal segment, and that was proven true after the total disappear-

ance of obstipations postoperatively and the resorption of distention thereafter.

## CONCLUSION

Although intestinal duplications are rare and occur more commonly in children, they should be considered in our differential diagnosis of small bowel obstruction with a radiologic "feces sign" in adults. Diagnosis done mainly by ultrasonography and CT scan. Surgical management by excision of the duplication or stripping of its mucosa is the mainstay of treatment. In our case, surgery was both curative and diagnostic.

## REFERENCES

1. Rasool N, Safdar CA, Ahmad A, Kanwal S. Enteric duplication in children: clinical presentation and outcome. *Singapore Med J* 2013; 54 (6): 343-6.
2. Simsek A, Zeybek N, Yagci G et al. Enteric and rectal duplications and duplication cysts in the adult. *ANZ J Surg* 2005 Mar; 75 (3): 174-6.
3. Tamvakopoulos GS, Sams V, Preston P, Stebbings WS. Iron-deficiency anaemia caused by an enterolith-filled jejunal duplication cyst. *Ann R Coll Surg Engl* 2004 Nov; 86 (6): W49-51.
4. Holcomb GW 3rd, Gheissari A, O'Neill JA Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. *Ann Surg* Feb 1989; 209 (2): 167-74.
5. Ildstad ST, Tollerud DJ, Weiss RG, Ryan DP, McGowan MA, Martin LW. Duplications of the alimentary tract. Clinical characteristics, preferred treatment, and associated malformations. *Ann Surg* 1988 Aug; 208 (2): 184-9.
6. Park JY, Her KH, Kim BS, Maeng YH. A completely isolated intestinal duplication cyst mimicking ovarian cyst torsion in an adult. *World J Gastroenterol* 2014 Jan 14; 20 (2): 603-6.
7. Ekbote G, Pokharkar AB, Moon P. A rare case of perforated tubular ileal duplication in 72-year-old male. *Indian J Surg* 2013 Jun; 75 (Suppl 1): 418-20.
8. Lima M, Molinaro F, Ruggeri G, Gargano T, Randi B. Role of mini-invasive surgery in the treatment of enteric duplications in paediatric age: a survey of 15 years. *Pediatr Med Chir* 2012 Sep-Oct; 34 (5): 217-22.
9. Peiper M, Zornig C. [Jejunal small intestine duplication]. [Article in German]. *Langenbecks Arch Chir* 1994; 379 (6): 358-60.
10. Goyal M, Saucedo-Crespo HI, Welsh FJ. Congenital intestinal duplication in an adult. *J Gastrointest Surg* 2014 Oct; 18 (10): 1876-7.
11. Gebesce A, Korkmaz M, Keles E, Korkmaz F, Mahmutyazicioglu K, Yazgan H. Importance of the ultrasonography in diagnosis of ileal duplication cyst. *Gastroenterol Res Pract* 2013; doi:10.1155/2013/248625. Epub 2013 Nov 4.
12. Atalar MH, Cankorkmaz L, Ozer H, Koyluoglu G. A huge duplication cyst of the ileum. *Pol J Radiol* 2013 Jul; 78 (3): 70-3.
13. Fiorani C, Scaramuzza R, Lazzaro A et al. Intestinal duplication in adulthood: A rare entity, difficult to diagnose. *World J Gastrointest Surg* 2011 Aug 27; 3 (8): 128-30.
14. Fuchsjäger MH. The small-bowel feces sign. *Radiology*

- 2002 Nov; 225 (2): 378-9.
15. Johnstone MJ, Clegg JF. Gastrointestinal haemorrhage from small bowel duplication. *Postgrad Med J* 1977 Nov; 53 (625): 700-2.
  16. Kusunoki N, Shimada Y, Fukumoto S et al. Adenocarcinoma arising in a tubular duplication of the jejunum. *J Gastroenterol* 2003; 38 (8): 781-5.
  17. Kim TH, Kim JK, Jang EH, Lee JH, Kim YB. Papillary adenocarcinoma arising in a tubular duplication of the jejunum. *Br J Radiol* 2010 Mar; 83 (987): e61-4.
  18. Wan XY, Deng T, Luo HS.. Partial intestinal obstruction secondary to multiple lipomas within jejunal duplication cyst: A case report. *World J Gastroenterol* 2010 May 7; 16 (17): 2190-2.

#### ANNEX

##### PUBLISHED CASE PRESENTATIONS OF JEJUNAL DUPLICATIONS IN ADULTS.

| Author/Year   | Clinical presentation   | Diagnostic modality  | Management | Outcome  |
|---|---|--|------------|--|
| <b>Gastrointestinal haemorrhage from small bowel duplication</b>  |   |  |            |  |
| M.J. Johnstone, J.F. Clegg<br>1977<br>[15]  | 8-day history of passing melena stools and feeling faint.   | <i>Gastroduodenoscopy, barium meal and enema</i>   | Laparotomy | <i>Duplication of 11.5 cm of the proximal jejunum with ectopic pancreatic tissue and small ulcer</i>   |
| <b>Adenocarcinoma arising in a tubular duplication of the jejunum</b>   |   |  |            |  |
| N. Kusunoki <i>et al.</i><br>2003<br>[16]   | Abdominal distention and intermittent pain  | <i>Upper GI series, barium enema, gastroscopy &amp; colonoscopy, CT scan.</i>            | Laparotomy | <i>Histology: adenocarcinoma arising in a duplication of the jejunum</i>   |
| <b>Iron-deficiency anaemia caused by an enterolith-filled jejunal duplication</b>   |   |  |            |  |
| G.S. Tamvakopoulos <i>et al.</i><br>2004<br>[3]   | 3-month history of loin pain, anemia with low MCV.  | <i>Barium swallow (Upper GI series)</i>  | Laparotomy | <i>10-cm diameter cystic mass containing enteroliths on the mesenteric border at the level of mid-jejunum</i>  |
| <b>Papillary adenocarcinoma arising in a tubular duplication of the jejunum</b>   |   |  |            |  |
| T.H. Kim <i>et al.</i><br>2010<br>[17]  | Lower abdominal pain and vomiting for 3 days  | <i>Small bowel series, contrast-enhanced CT, small bowel enteroscopy, CEA and CA19-9</i> | Laparotomy | <i>Histology: tubular duplication arising in the jejunum. A poorly differentiated papillary adenocarcinoma was found within the tubular duplication invading subserosa, metastases to regional lymph nodes &amp; mesenteric lymph nodes of SMA</i> |
| <b>Partial intestinal obstruction secondary to multiple lipomas within jejunal duplication cyst: A case report</b>                          |   |  |            |  |
| X.Y. Wan <i>et al.</i><br>2010<br>[18]  | Abdominal bloating after meals for the last 10 days, history of chronic constipation for 6 years. | <i>Abdominal X ray, double-balloon endoscopy</i>   | Laparotomy | <i>Jejunal duplication having seven cysts ranging from 5.5 cm x 6 cm to 8.5 cm x 7.5 cm, with multiple submucosal lipomas lining on its walls</i>  |
| <b>GI:</b> gastrointestinal <b>MCV:</b> mean corpuscular volume <b>CEA:</b> carcinoembryonic antigen <b>SMA:</b> superior mesenteric artery |   |  |            |  |