

A RARE PRESENTATION OF COLONIC DUPLICATION CYST: REPORT OF A CASE AND REVIEW OF LITERATURE

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Duplication cyst is an uncommon congenital abnormality of the alimentary tract. It can occur anywhere in the alimentary tract with the ileum and the jejunum representing the most common sites of duplication. Most often the patients are asymptomatic and colonic duplication cysts remain undiagnosed for years. In this case report we present an unusual case of colonic duplication cyst with a transverse colon location. We present the radiological findings of this rare congenital malformation in order to be included in the differential diagnosis of cystic masses of the gastrointestinal tract.

Key-word: Colon, abnormalities.

Duplication cyst is an extremely rare congenital malformation of the alimentary tract. It occurs most often in the ileum, accounting for over 60% of cases followed by the jejunum and the duodenum (1). The colon is the least common site of enteric duplication. In fact, in a review of 495 alimentary tract duplications only 7% of the duplications involved the colon (2). To our knowledge less than 100 cases have been described in the published literature (3).

Case report

We present an unusual case of colonic duplication cyst in a 45-year-old Caucasian man who presented to our hospital with lumbar pain. The patient had a medical history of constipation. On admission, the patient was afebrile. Physical examination revealed a large mass in the left upper quadrant with mild diffuse tenderness and no peritoneal signs. Blood laboratory tests were within normal limits. An initial plain X-ray was performed with no abnormal findings. Ultrasonography was insignificant due to gas filled bowels. After the first inconclusive radiological exams, a contrast Barium Enema study was decided to be performed. Barium Enema showed a large air-filled tubular structure in the left upper quadrant containing an air-fluid level (Fig. 1). The same cystic structure was partially filled with contrast due to communication with the transverse colon at the splenic flexure. The shape of the cystic structure changed with peristalsis and the position of the patient

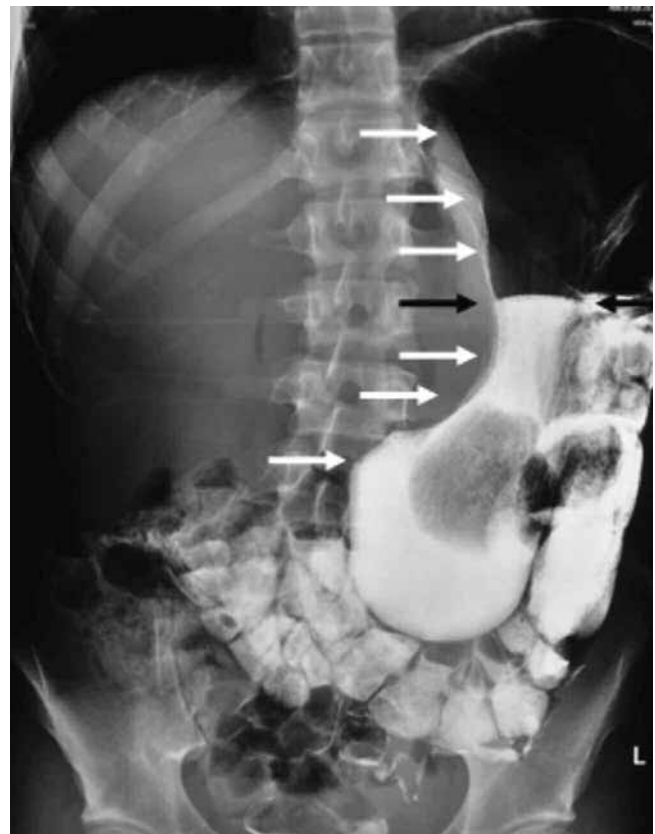


Fig. 1. — Abdominal X-ray following the contrast barium enema study in upright position. It shows a large structure (white arrows) with contrast due to communication with the transverse colon with air-fluid level (black arrows).

(Fig. 2). At the same examination the gastric antrum was recognized as a smaller air-filled structure adjacent to the same tubular structure. Furthermore, Computed Tomography (CT) of the abdomen with

oral contrast material was performed. It demonstrated a large air-filled 14 × 7 cm structure containing oral contrast in the left upper quadrant (Fig. 3). CT confirmed the findings of contrast Barium Enema study and a possible diagnosis of colonic duplication of the transverse common was concluded. A surgical intervention was decided in order to prevent complications of the colonic duplication cyst. At operation, the cyst was excised with a segment of

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Fig. 2. — Abdominal X-ray following the contrast Barium Enema study in supine position demonstrates the change of the shape of the cystic air-filled structure (arrowheads).

transverse colon and a colocolostomy was performed (Fig. 4). The duplication cyst was attached to the transverse colon with the presence of a small communication with the colonic lumen. Post-operative recovery was uneventful.

Discussion

Enteric duplications cysts are uncommon congenital malformations (4). They are usually discovered in infancy and childhood, but they may be discovered at any period of life. They occur anywhere along the length of the alimentary tract on the mesenteric side. The location at the transverse colon is extremely rare (2). Their walls may contain all of the normal bowel layers, including the mucosa, submucosa and muscularis. They may appear as cystic or tubular malformations. While duplication cysts typically do not communicate with the adjacent bowel lumen, tubular lesions, which usually arise near the colon, may communicate.

Most colonic duplication cysts are asymptomatic and remain undiagnosed for years (5). If symptomatic, they manifest obstruction, bleeding,

infection or constipation (3, 6-9). In one rare case of combined duplication of the colon and vermiform appendix, it was presented with hydronephrotic atrophy of the kidney (10). Also patients can present a variety of non-specific signs and symptoms like abdominal pain. Resection of the duplication cyst and the adjacent bowel is recommended because of the possibility of malignant changes and the risk of gastrointestinal ulceration and haemorrhage due to ectopic gastric mucosa (11). To avoid any future complications, cyst resection is indicated even in the asymptomatic patient (12-14).

The diagnosis of a duplication cyst is difficult to be made clinically. Radiological studies play an important role in the detection and diagnosis of the duplication cysts. Plain film



Fig. 3. — Computed Tomography of the abdomen demonstrating the cystic structure (arrows) containing an air-fluid level.

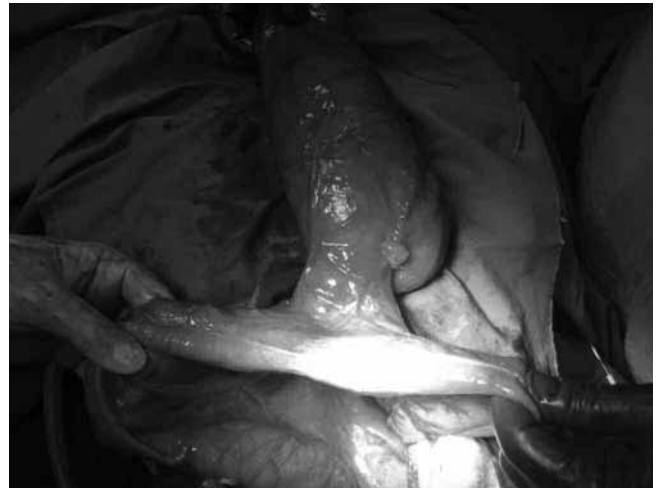


Fig. 4. — Surgical specimen including part of the transverse colon with the duplication.

radiography may be normal or may show a soft tissue mass with/without displacement of adjacent bowel or evidence of intestinal obstruction. Ultrasound is particularly well suited for the identification and characterization of duplication cysts, although in our case it was inconclusive (15-16). Contrast examinations of the gastrointestinal tract can be useful in order to demonstrate displaced loops of bowel surrounding the presumed cyst and depict the communication with the gastrointestinal tract (17). In our case the use of contrast in the Barium Enema study revealed the origin of the cystic structure and depicted the communication with the transverse colon. In difficult cases which require a multi-planar approach to delineate the relationship between the cystic and peripheral structures, CT and MRI

may be helpful. However, the contribution of sonography, CT and MRI has to be developed, in order to help radiologists to suggest such a disorder in their daily practise.

In summary, colonic duplication share an extremely variable clinical presentation. The differential diagnosis of a cystic abdominal mass should always include enteric duplication, especially when associated with intestinal symptoms.

References

1. Pulingandla P.S., Nguyen L.T., St-Vil D., et al.: Gastrointestinal duplications. *J Pediatr Surg*, 2003, 38: 740-744.
2. Stringer M.D., Spitz L., Abel R., et al.: Management of alimentary tract duplication in children. *Br J Surg*, 1995, 82: 74-78.
3. Fotiadis C., Genetzakis M., Papandreou I., Misiakos M.P., Agapitos E., Zografos G.C.: Colonic duplication in adults: report of two cases presenting with rectal bleeding. *World J Gastroenterol*, 2005, 11: 5072-5074.
4. Macpherson R.I.: Gastrointestinal duplications: clinical, pathologic, etiologic, and radiologic considerations. *Radiographics*, 1993, 13: 1063-1080.
5. Favara B.E., Franciosi R.A., Akers D.R.: Enteric duplications: thirty-seven cases – a vascular theory of pathogenesis. *Am J Dis Child*, 1971, 122: 501-506.
6. 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: 644-646.
7. Otter M.I., Marks C.G., Cook M.G.: An unusual presentation of intestinal duplication with a literature review. *Dig Dis Sci*, 1996, 41: 627-629.
8. Van Elst F., Hubens A.: Duplication of the colon in the adult. *Acta Chir Belg*, 1978, 77: 335-342.
9. Bremer J.L.: Diverticula and duplications of the intestinal tract. *Arch Pathol*, 1944, 38: 132-140.
10. 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: 641-643.
11. Horie H., Iwasaki I., Takahashi H.: Carcinoid in a gastrointestinal duplication. *J Pediatr Surg*, 1986, 21: 902-904.
12. Kim Y.W., Kim J., Lee K.Y., Kim N.K., Cho C.H.: Asymptomatic tubular duplication of the transverse colon in an adult. *Yonsei Med J*, 2005, 46: 189-191.
13. Holcomb G.W., Gheissari A., O'Neill J.A. Jr.: Surgical management of alimentary tract duplications. *Ann Surg*, 1989, 209: 167-174.
14. Cavar S., Bogovic M., Luetic T., Antabak A., Batinica S.: Intestinal duplications – experience in 6 cases. *Eur Surg Res*, 2006, 38: 329-332.
15. Kangarloo H., Sample W.F., Hasen G., Robinson J.S., Sarti D.: Ultrasonic evaluation of abdominal gastrointestinal tract duplication in children. *Radiology*, 1979, 131: 191-194.
16. Hayden C.K. Jr. Ultrasonography of the gastrointestinal tract in infants and children. *Abdom Imag*, 1996, 21: 9-20.
17. Rathi V., Singh S., Bhargava S.K., Kaur N., Seth N.: Diagnosis of tubular colonic duplication by barium follow-through study. *Australas Radiol*, 2005, 49: 157-159.