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Rectal duplication, rare cause of constipation – case report

Case report

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Abstract: Digestive tract duplications are uncommon congenital anomalies, encountered mostly in the first 3 months of life. Overall the rectum is the least common site of alimentary duplications. We represent a case of large cystic non-communicating duplication that manifested with constipation and profuse rectal bleeding. When diagnosis was established, surgery was planned and the cyst was enucleated completely. Histopathology examination confirmed the diagnosis.

Keywords: *Digestive • Tract • Duplication • Children • Treatment*

1. Introduction

Digestive tract duplications are the uncommon congenital anomalies, mostly encountered in the first 3 months of life. Fitz in 1884 [1], and Ladd [2] in 1937, first described duplications and their clinical symptoms. In 1961, Potter reported 2 cases in more than 9,000 fetal and neonatal autopsies [3]. Duplications can occur along the entire digestive tract, but mostly in the ileum [4]. Overall, the rectum is the least common site of alimentary tract duplications [5]. Duplications have disposition to be located mesenterially, firmly attached to an adjacent segment of normal intestine (sharing a common wall and mesenteric blood supply).

2. Case report

An 11-month-old male patient was admitted to the Pediatric Surgery Department due to signs of acute abdomen and rectal bleeding. The patient also had a medical history of a five-month chronic constipation. The hematological and biochemical profile was within normal range. Significant anemia (Er=2,62, Hgb=8,2g/dl) was found. Rectal examination revealed a cystic mass in the rectal lumen causing obstruction. Plain abdominal films detected a dilated intestinal loop, presumably colon with fecal masses and a few air fluid levels. CT examination revealed a well formed, unilocular cystic formation 33x33 mm in size, located immediately in front of the urinary bladder and vesiculae seminales, extending along retro-rectal space. (Figure 1).

PSARP (posterior sagital anorectoplasty) has been used to expose the cyst (Figure 2); complete cystic enucleation followed. The parasagital muscle complex was anatomically reconstructed in order to provide satisfactory sphincter function and continence. Functionality of the rectum was preserved and the rectal wall remained intact. The final cosmetic result was excellent. Postoperative recovery was uneventful. Histopathology examination revealed the rectal mucosa in the cystic wall, with sub-mucous glands and muscle coat, as confirmation of rectal duplication (Figure 3). To date, three years after the procedure, the patient has no defection difficulties, sphincter function is preserved and he is continent.

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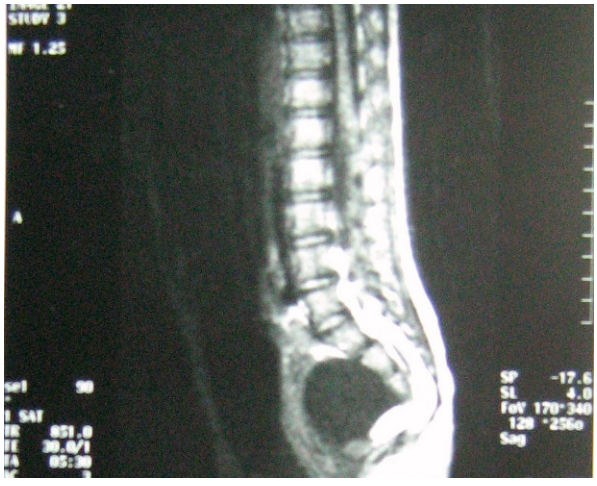

Figure 1. NMR revailed well formed cystic formation 33x33 mm in size in front of urinary bladder extending to retrorectal space

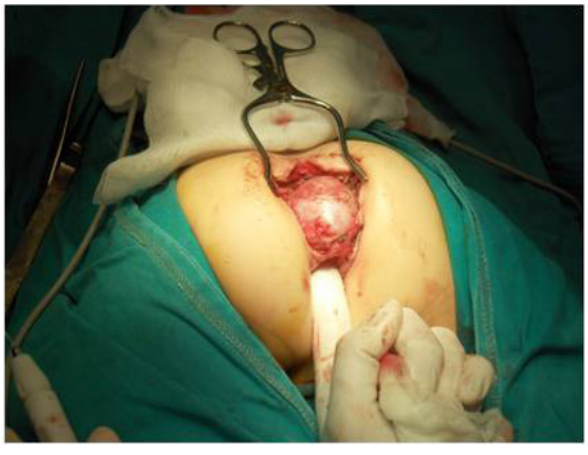

Figure 2. Cystic formation located in the rectorectal space

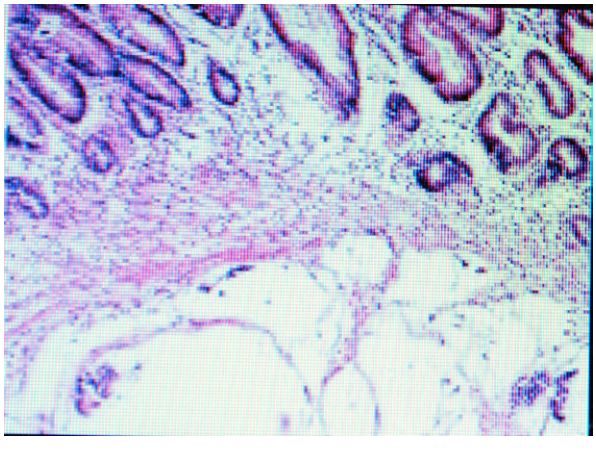

Figure 3. Pathohystology examination revealed rectal mucosa in cystic wall with smooth muscle layers.

3. Discussion

Congenital alimentary tract duplications are considered to be benign lesions, which may result in significant morbidity and mortality if left untreated. With 2-4% of all GI duplications, rectal duplications are extremely rare [6,7] and they are located mostly in retrorectal space [8]. It is generally assumed that duplications are the result of an error in notochord development (split notochord theory), with coexisting vertebral and genitourinary malformations or intestinal atresias. Other theories explain duplications as a defect named “aberrant luminal intestinal recanalization” in the early, gastrulation stage of development [9].

The clinical presentation depends on size, localization, extend of the lesion, mucosal lining and luminal communication if it is present. Symptoms include abdominal distension and chronic constipation, rectal bleeding, intussusceptions, vomiting, palpable mass, respiratory distress or bladder outlet obstruction.

In all children profuse rectal bleeding associated with chronic constipation disorders, should be suspected for rectal duplication. Digital rectal examination can reveal prominence in the rectal lumen. Careful rectal examination should be the first diagnostic step. Rectal biopsy is not recommended, because M. Hirschsprung is extremely rare associated with rectal duplication.

Without the pathognomonic signs, radiologic diagnosis is unlikely. Irigography is demonstrating the filling defect. CT scan and MRI are the most precise, pointing to the well-formed cystic formation firmly attached to the part of intestine, and delineating surrounding structures.

Surgical treatment is largely dictated by the specific anatomic location of duplication, its relation to normal anatomic structures, and mucosal lining. In general, surgical excision is the preferred treatment as it has good curative features. Small cysts can be completely excised by enucleation, without compromising the adjacent bowel vascularisation. This approach is not always feasible, especially in cases of duplications that are very large and firmly attached to the rectum. Partial resection, marsupialization, or multiple repeated mucosal stripping are treatments of choice [10].

Although these lesions are usually benign, surgical resection is the most feasible method of treatment in order to prevent future complications: massive hemorrhage, cystic infection, short bowel Sy, mass effect obstruction and rarely cancerous degeneration [11]. Malignant degeneration (adenocarcinomas) has been reported in patients with untreated rectal duplications, by Ballantyne in 1932 [12].

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