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ENTERIC DUPLICATION CYST IN CHILD: A CASE REPORT

CISTO DE DUPLICAÇÃO INTESTINAL NA CRIANÇA: RELATO DE CASO

QUISTE DE DUPLICACIÓN INTESTINAL EN UN NIÑO: REPORTE DE UN CASO

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ABSTRACT:

Enteric duplication cyst is a rare pathology, it affects the whole gastrointestinal tract, although it has a greater incidence in the small intestine. It is distributed equally in both sexes. It has variable clinical presentations, being asymptomatic or symptomatic, sometimes leading to serious complications. The enteric duplication cyst is a complex diagnosis pathology, and the surgical treatment has a diagnostic and therapeutic role, with an excellent prognosis. This study consists of a case report about a four-year-old patient, with enteric duplication cyst in the small intestine, treated at a reference Pediatric Hospital in Santa Catarina State, Brazil. Data were collected for medical records and interviews with relatives. The decision for the surgical treatment provided a referred symptoms improvement, as well as prevented posterior severe complications.

KEYWORDS: Gastrointestinal Tract; Intestinal Duplication Cyst; Intestinal Cyst; Congenital Bowel Anomalies, Pediatrics

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RESUMEN: Los quistes intestinales por duplicación son una patología rara que afecta a todo el tracto gastrointestinal, aunque tienen una mayor incidencia en el intestino delgado. Se distribuyen uniformemente entre ambos sexos. Tiene una clínica variable, que puede ser asintomática o sintomática, dando lugar en ocasiones a complicaciones graves. Patología de difícil diagnóstico, con tratamiento quirúrgico que tiene un papel diagnóstico y terapéutico, con excelente pronóstico. Este estudio consiste en un reporte de caso de un paciente de cuatro años con quiste de duplicación intestinal en el intestino delgado, atendido en un Hospital de Niños de referencia en el estado de Santa Catarina/Brasil. La información se recopiló de historias clínicas y entrevistas con familiares. La decisión de tratamiento quirúrgico proporcionó una mejoría de los síntomas referidos, así como la prevención de complicaciones posteriores de mayor gravedad.

PALABRAS CLAVE: Tracto Gastrointestinal; Quiste de duplicación intestinal; quiste intestinal; Anomalías Intestinales Congénitas, Pediatría

RESUMO

Os cistos de duplicação intestinal são uma patologia rara, que acometem todo o trato gastrointestinal, embora tenham maior incidência no intestino delgado. Estão distribuídos igualmente entre ambos os sexos. Possui uma clínica variável, que pode ser assintomática ou sintomática, por vezes levando a complicações graves. Patologia de difícil diagnóstico, tendo o tratamento cirúrgico um papel diagnóstico e terapêutico, com excelente prognóstico. Esse estudo consiste em um relato de caso de paciente com quatro anos de idade, com cisto de duplicação intestinal em delgado, atendido em um Hospital Infantil de referência no estado de Santa Catarina/ Brasil. Foram realizadas coletas das informações em prontuário e entrevista com familiares. A decisão pelo tratamento cirúrgico proporcionou uma melhora dos sintomas referidos, como também, preveniu complicações posteriores de maior gravidade.

Palavras-chave: Trato Gastrointestinal. Cisto de Duplicação Intestinal. Cisto intestinal. Anomalias Intestinais Congênitas. Pediatria.

INTRODUCTION

Enteric duplication cysts encompass a group of congenital malformations composed of a smooth muscular layer, which can be tubular, spherical and of variable size. The embryological origin for these duplications' emergence remains controversial, there are many theories, and among them, the most accepted is the "intrauterine vascular accident" [1,2].

This pathology is considered rare, occurring circa 1 in every 4500 births, "with a slight predominance of the male sex" and greater occurrence in the small intestine [3]. The clinical presentation is variable, being asymptomatic or symptomatic. Image exams can detect the disease, but the surgical approach also has a diagnostical and therapeutical role, presenting excellent results [1].

With this perspective, this study aims to report a case of enteric duplication cyst assisted by the Pediatric Surgery Service at a Reference Hospital in Pediatrics in Santa Catarina.

METHODOLOGY

This piece analyses a patient's case of enteric duplication cyst assisted by the Pediatric Surgery Service at a Reference Hospital in Pediatrics in Santa Catarina. The case happened in 2019, the medical data was collected as well as an interview with the minor's family.

The references used in this study were searched using the following words: enteric duplication cyst, enteric cyst, and congenital enteric anomalies.

The study followed the requirements related to the ethical aspects of Resolution n. 466/2012 by the National Health Council, guaranteeing the right to secrecy, anonymity, and impersonality about the information. The Free and Informed Consent and Assent Terms were applied. The study was carried out in a teaching activity within the scope of Medical Residency.

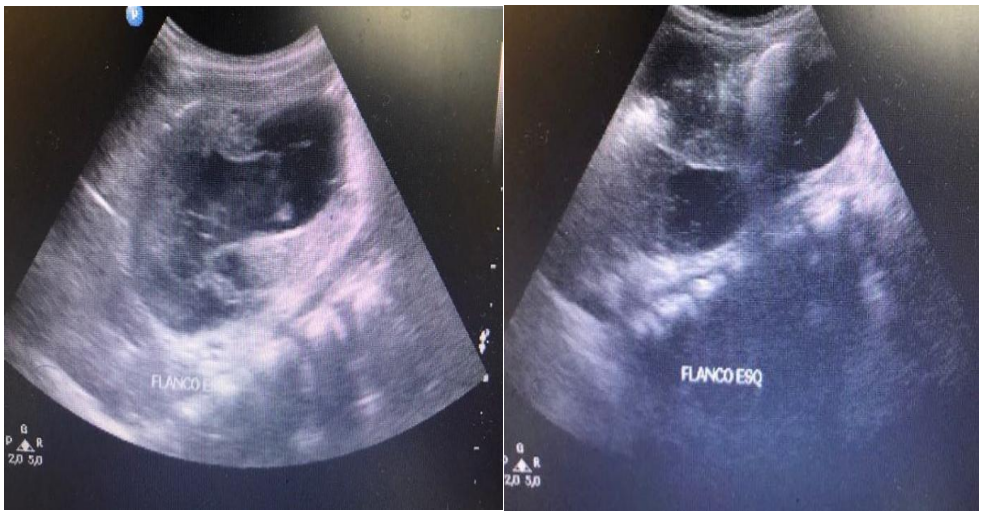
CASE REPORT

A four-year-old male patient sought hospital care due to vomiting for four days associated with distention and diffuse abdominal pain, with worsening when walking, constipation, drowsiness, and prostration; maintained food acceptance. The mother related recent intestinal alterations; diarrhea and constipation periods were interpolated. He was previously healthy, without complications during pregnancy and birth: birth weight 3.470 kg and Apgar 9/10. The physical examination showed good condition in general, blushed, anicteric, and afebrile. The cardiorespiratory exam did not show alterations. In the abdominal exam, an important abdominal distention was identified, hypertympanism on percussion and pain on superficial and deep palpation, small palpable mass in the mesogastrium.

During the patient's admission, laboratory exams (hidden blood and faecal leucocytes) were required, which were normal. Given the patient's clinical condition, hospitalization was established for diagnostic clarification. The total abdomen ultrasound (Figure 1) required during the hospitalization showed an anechoic image of thickened walls, multiple septations, evolving left flank, anterior abdomen, and pelvis, which may correspond to an expansive

process at retroperitoneum. For diagnostic additions, it was requested a total abdomen CT scan (Figure 2) which revealed a voluminous cystic formation adjacent to the small intestine, accompanying the fourth portion of the duodenum and proximal loops of the jejunum, thin-walled and homogeneous content with thick fluid density, without signs of contrast enhancement, causing extrinsic compression of the intestinal loops described above, measuring about 18.5 cm in length x 4.1 cm in diameter, which may correspond to intestinal duplication cyst. In view of the above, surgical treatment was chosen.

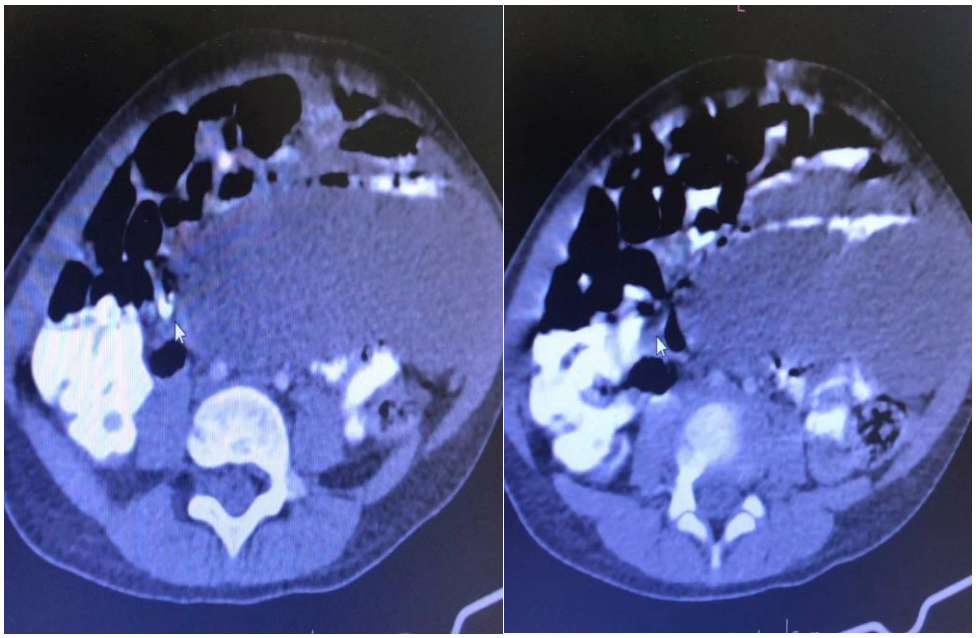
Figure 1: Total abdomen USG showing anechoic and septate image on the left flank



Source: Primary data (2019).

The patient underwent a right supraumbilical transverse laparotomy with exposure of the intestinal loops, which identified an intestinal duplication that started in the fixed loop of the mesentery, in the transition from the end of the duodenum to the beginning of the jejunum. The duplicated part was significantly dilated due to a moderate to large hematoma, caused by old bleeding, which compressed the intestinal passage, causing intestinal occlusion.

Figure 2: Total abdominal tomography demonstrating intestinal cyst causing extrinsic compression of intestinal loops



Source: Primary data (2019).

In the intraoperative period, we opted for enterectomy of the duplicated duodenal portion, due to the impossibility of excision of the lesion, followed by manual end-to-end enteroanastomosis in two planes. No other intra-abdominal malformations or anatomical changes were seen. Then, the abdominal cavity was thoroughly washed with saline solution of 0.9%, as well as the review of hemostasis and placement of a Penrose drain close to the intestinal anastomosis. The closure occurred in layers and the resected material was sent for anatomopathological study. There were no intraoperative complications.

In the postoperative period, the patient remained without an oral diet for five days and with an open nasogastric tube in a bottle for two days. Oral diet was reintroduced on the fifth postoperative day, starting with a liquid diet and progressively escalating to free diet. He was discharged on the eighth postoperative day in good general condition, without vomiting or complications and accepting the proposed diet. Patient discharged from pediatric surgery in return visit, with acceptance of the proposed diet and preserved intestinal transit.

DISCUSSION

Enteric duplication cysts encompass a group of congenital malformations composed of a smooth muscular layer, which can be tubular, spherical and of variable size [(2)]. The embryological origin for these duplications' emergence remains controversial [(4)].

Anomalous cysts in the gastrointestinal tract are placed mainly in the small intestine (47%), colon (20%), esophagus (17%), stomach (8%) and duodenum (2-12%) [5, 6, 4]. The cyst's muscular wall, in most cases, is the same as the intestine, and might or might not communicate with its lumen, which usually does not occur. The anatomical positioning of this duplication in this patient matches the most reported cases. Regarding the Histology, in up to 25 to 30% of the cases, the cyst can be overlayed by ectopic tissue, more frequently gastric tissue, predisposing ulcer formation, bleedings and perforations [7,2]. Besides that, the cyst can also be revested by exocrine and endocrine pancreatic mucosa tissue [6]. In view of the above, three anatomical criteria are used to define intestinal duplications, namely: intimate connection to the gastrointestinal tract, mucosal lining similar to that of the gastrointestinal tract, and the presence of a smooth muscle wall [8].

The clinical presentation is variable, and it may present with non-specific signs and symptoms, such as recurrent abdominal pain, nausea, vomiting or abdominal mass, which is a frequent finding on physical examination [8,9]. Sometimes, it manifests itself through its clinical complications, among them gastrointestinal hemorrhage, intussusception, perforation, obstruction, jaundice, pancreatitis and infection [9,2]. Complications of malignancy include carcinoid tumors, squamous cell carcinomas, and common adenocarcinomas [5,4]. In up to 80% of the cases, the cysts present before the age of two, associated with other abdominal malformations [6].

Gastrointestinal hemorrhage is a frequent complication that results from erosion of the mucosa of the duplication and/or adjacent intestine, secondary to the acid produced by the ectopic gastric mucosa [8,9]. Intestinal obstruction, although uncommon in these cases, was present in this patient's clinic, because of extrinsic compression of the cyst. However, intestinal obstruction can also result from intestinal invagination or volvulus. Intestinal perforation is rare and may be secondary to gangrene of a secretory and non-communicating duplication or secondary to peptic ulceration [9].

The imaging methods most used for the diagnosis of intestinal duplication are abdominal ultrasonography and computed tomography of the abdomen, which may demonstrate a cystic or elongated structure with a thick wall [8,9]. Despite this, the preoperative diagnosis is difficult, and they are often diagnosed intraoperatively from a suspected diagnosis or even as an accidental finding [7].

The surgical procedure has a diagnostic and therapeutic role, with an excellent prognosis [7,8], always focusing on patient safety [10]. However, one should opt for the most conservative treatment possible, that is, with lesional excision, when possible, or segmental enterectomy in cases where excision is not feasible. Wide resections should be avoided in order to avoid short bowel syndrome. Enterectomy can be avoided, in selected cases, by excising most of the circumference of the duplication, leaving only the common wall with the adjacent normal bowel. However, this is feasible as long as it is associated with the destruction, by mechanical or chemical means, of the remaining mucosa. Mucosectomy is another therapeutic possibility, being performed through one or multiple transverse incisions along the duplication. In cases of duplication resection, careful dissection of the vessels should be performed to ensure vascularization of the adjacent gastrointestinal tract [8].

CONCLUSIONS

Intestinal duplication cysts are rare and often difficult to diagnose clinically and complementary. However, they should be explored if clinically suspected, with surgery having a diagnostic and therapeutic role. In the hands of experienced surgeons and early diagnosis, they have an excellent clinical prognosis.

In relation to the case reported, we can see compliance with the general picture of the evolution of the disease described in the literature. The decision for surgical treatment provided an improvement in the referred symptoms, as well as prevented later complications of greater severity and morbidity and mortality.

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