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Late-Onset Ileal Duplication Complicated by Occlusive Syndrome

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Abstract **Case Report**

Gastrointestinal duplications are rare congenital malformations that can involve all digestive segments, from the mouth to the anus. These duplications can be cystic or tubular, communicating or not with the adjacent digestive tract segment. The diagnosis is increasingly suspected antenatally. In other cases, the clinical symptoms are often aspecific, depending on the anatomical region concerned and the possible occurrence of revealing complications. Imaging, and in particular ultrasound, allows the diagnosis to be evoked and to look for associated anomalies, which condition the prognosis and management. The diagnosis of certainty is most often confirmed only after excision and anatomical anatomopathological examination. Surgery is required and must be complete at best, to avoid any risk of complication or risk of complication or malignant degeneration. We report the observation of an infant who presented with a late-onset cystic ileal duplication complicated by an occlusive syndrome.

Key words: Cystic lesion, Digestive duplication, CT.

INTRODUCTION

Gastrointestinal duplications are a group of various congenital malformations of the digestive tract, defined as tubular or cystic malformations, located on a segment of the digestive tract, from the oral cavity to the anus. They represent a rare entity. Antenatal diagnosis is possible.

Symptoms appear most often in the newborn (25% in the first months of life) or in the first years of life (70% in the first 2 years).

Imaging is a key step in the diagnosis, especially for the search of arguments of positive or differential diagnosis, but also for the search of associated anomalies.

We report the observation of an infant who presented with a late-onset cystic ileal duplication complicated by an occlusive syndrome.

CASE REPORT

We examined an 18-month-old infant, male, with no particular pathological history, who presented with progressive abdominal distension associated with diffuse abdominal pain and food vomiting that was relieved by symptomatic treatment. The evolution was marked by the installation of an occlusive syndrome made of a stop of matter and gas and bilious vomiting what motivated the family to consult the pediatric emergency room for adequate management. On clinical examination, he was apyretic with abdominal defense under the umbilical.

The initial radiological workup, which consisted of an abdominal ultrasound scan, revealed significant distension of the digestive tract, measured at 6 cm in diameter, with fluid and aeriform contents of a grelicular appearance (Figure 1).

The workup was completed by an abdominal and pelvic CT scan without and with injection of PDC showing a voluminous intraperitoneal abdominal and pelvic formation of tubular liquid density, serpiginous, having the same shape as a dilated digestive loop, with a non-enhanced wall after injection of PDC and whose two legs are located in the intra-pelvic area (figure 2).

The diagnosis of tubular digestive duplication was evoked in the first place. Cystic lymphangioma was a diagnosis not to be eliminated in front of this scannographic aspect.

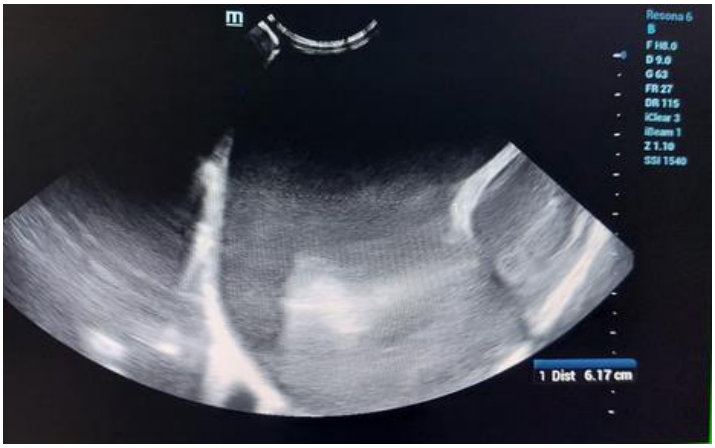

Fig-1: Ultrasound image showing significant distension of the gall coves measured at 6cm

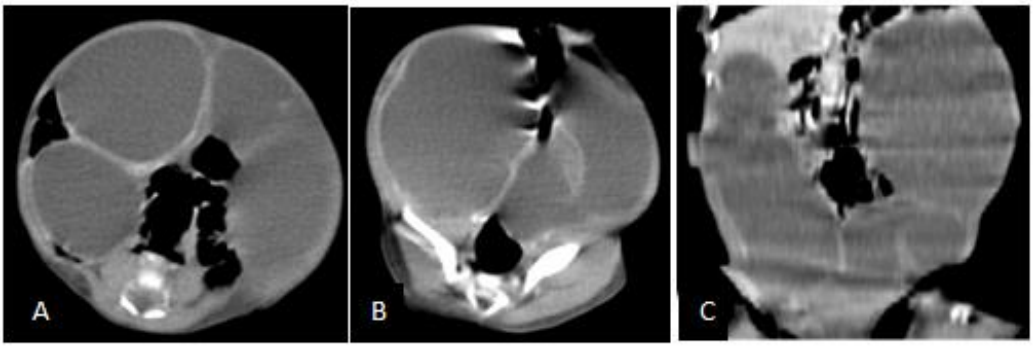

Fig-2: Abdominal-pelvic CT in axial (A and B) and coronal (C) sections with PDC injection

Voluminous intra peritoneal abdomino-pelvic formation of tubular, serpiginous fluid density, having the same shape as a dilated digestive loop, with non-enhanced wall after PDC injection exerting a mass effect on the ascending and descending colon.

The infant was operated on the same day (figure 3) by a transverse incision under the umbilicus at the opening of the peritoneum, showing an ileal duplication of the bauhin valve.

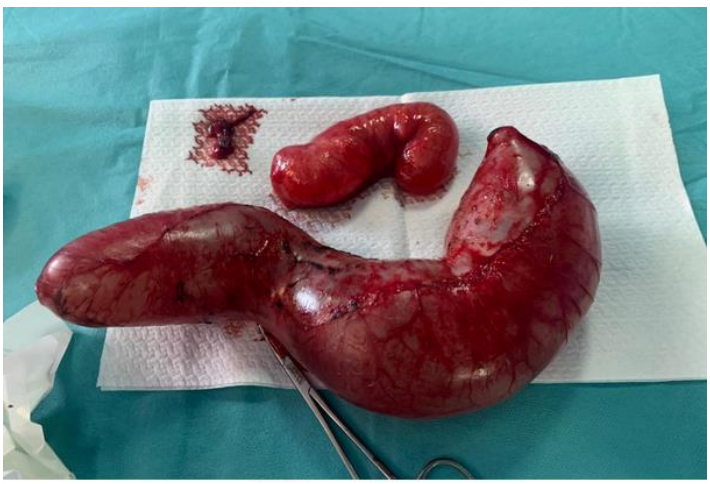

Fig-3: The ileo-caecal resection specimen with the cystic duplication in section

DISCUSSION

Gastrointestinal duplications are rare malformations with an incidence of 1 per 4500 births [2]. They can be located anywhere from the esophagus to the rectum. The most frequent location is the terminal ileum as in the presented case. The other classic locations are, in decreasing order of frequency, the esophagus, the colon, the jejunum, the stomach and the duodenum [1].

The cystic form (found in our patient), represents about 82% of the forms, while the tubular form is present in the remaining 18% of cases [5].

The diagnostic difficulty varies according to the type of duplication, cystic or tubular, its location and the existence of a communication with the digestive lumen

Duplications of the gastro intestinal tract are characterised by the following histological features two muscular layers with myenteric cells inside. Cystic or tubular, they are always situated on the mesenteric border of the gastro intestinal tract with a common blood supply. The pathogenesis of duplication affects early fetal development via various mechanisms [3].

It is a congenital pathology that is often diagnosed in the neonatal period during the first ultrasound at 12 weeks of amenorrhea in the form of acystic image of variable topography or indirect signs such as hydramnios, ascites or pleural effusion [4, 7].

In our patient, the diagnosis was late because the pregnancy was poorly monitored. The condition may be revealed by vomiting (24%), abdominal pain (34%), and a palpable mass (10.5%) [4], sometimes by a revealing complication: occlusive syndrome, hemorrhage or perforation [7].

Abdominal ultrasonography and CT scan may suggest the diagnosis in the presence of an intraperitoneal cystic image in children. However, in the majority of reported series, the diagnosis is made intraoperatively [6]. In our case, the abdominal CT scan allowed the objectification of the cystic lesion without being able to affirm its origin.

The differential diagnosis, usually easy, is made with cystic lymphangioma, ovarian cyst, and choledochal cyst which is more posterior and has a thinner wall [4].

The treatment of these conditions consists of ileal resection removing the malformation [8]. Digestive anastomosis is performed at the same time if there are no contraindications such as the presence of peritonitis [5].

CONCLUSION

Gastrointestinal duplications are most often diagnosed in infants. They have a clinical polymorphism which makes their diagnosis difficult. Imaging is a key step in the diagnosis, especially for the search of arguments of positive or differential diagnosis, but also for the search of associated anomalies. Antenatal diagnosis thus helps to prepare the family and the medical teams for rapid and targeted treatment. This pathology is rare, has a good prognosis, and its treatment is always surgical by intestinal resection removing the lesion.

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