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Duplication of the small intestine manifested by obstruction of the gastrointestinal tract – case report

Zdwojenie jelita cienkiego objawiające się niedrożnością przewodu pokarmowego – opis przypadku

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Abstract We present the case report of a 24-day-old infant admitted to the Department of Paediatric Surgery in order to treat suspected obstruction of the gastrointestinal tract. For two days prior, the patient had experienced gas and stool retention. On the day of admission, vomiting occurred with gastric contents, later followed by faecal. Ultrasound and scout film of the abdomen were performed, showing radiological features of obstruction of the gastrointestinal tract. The newborn was urgently referred for surgery. The procedure was performed under general anaesthesia. Intraoperatively, at the level of the ileum, 5 cm before the cecum, obstruction of the gastrointestinal tract was found in the form of a lesion resembling Meckel's diverticulum. Resection of the lesion was performed and end-to-end ileoileal anastomosis was formed. The resected lesion was sent for histopathological examination, which revealed a cystic duplication of the gastrointestinal tract.

Keywords: obstruction of the gastrointestinal tract, duplication of the gastrointestinal tract, prenatal diagnosis

Streszczenie

Przedstawiono przypadek 24-dniowego noworodka przyjętego na Oddział Chirurgii Dziecięcej celem leczenia podejrzenia niedrożności przewodu pokarmowego. U pacjentki obserwowano zatrzymanie gazów i stolca od dwóch dni, a w dniu przyjęcia wymioty treścią pokarmową, a następnie kałową. Wykonano badanie ultrasonograficzne oraz radiologiczne przeglądowe jamy brzusznej, stwierdzając radiologiczne cechy wysokiej niedrożności przewodu pokarmowego. Zakwalifikowano noworodka w trybie pilnym do operacji. Zabieg wykonano w znieczuleniu ogólnym. Śródoperacyjnie stwierdzono niedrożność przewodu pokarmowego na wysokości jelita krętego 5 cm przed kątnicą, w postaci zmiany przypominającej uchyłek Meckela. Wykonano resekcję zmiany z zespoleniem jelita krętego koniec do końca. Resekowaną zmianę wysłano do badania histopatologicznego, w którym stwierdzono torbielowate zdwojenie przewodu pokarmowego.

Słowa kluczowe: niedrożność przewodu pokarmowego, zdwojenie jelita cienkiego, diagnostyka prenatalna

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INTRODUCTION

astrointestinal duplication is a foetal anomaly in which three characteristic features are distinguished. First and the most frequent one: it can be linked to any part of the gastrointestinal tract, second: it has an epithelial lining similar to the mucous membrane which can be found in the gastrointestinal tract, and lastly it has a smooth muscle layer. The cysts can be of different sizes, containing the above-mentioned elements, and they can be located in any part of the digestive tract, from the oral cavity to the anus⁽¹⁻⁷⁾. Nowadays, most of the cysts are diagnosed at the stage of prenatal diagnosis during foetal ultrasound⁽³⁾. The most common symptoms of gastrointestinal duplications include abdominal pain and vomiting. Often, distension of the duplicated part occurs due to secretion to its lumen, potentially causing compression of adjacent structures, which can lead to bowel obstruction^(1,2,4,5). The most common location of duplication is the small intestine, which accounts for as many as 45% of all described cases. Surgery is the treatment of choice, consisting in segmental resection along with adjacent lesion.

CASE REPORT

A 24-day-old newborn was admitted to the Department of Paediatric Surgery in a good general condition with suspected obstruction of the gastrointestinal tract. Stool and gas retention had been observed for two days. Moreover, vomiting with food content and then faecal contents appeared on the day of admission. After the birth of the baby, low weight gain and frequent regurgitation after feeding were observed. On physical examination, the abdomen arched above the chest level was found tense, tender on palpation, with a pain reaction without palpable pathological resistance. Intestinal peristalsis was asymmetrical, obstructive, audible only in the right mid-abdomen, and retention in the gastric probe was light brown. In laboratory tests, only slightly elevated markers of inflammation were found; the remaining parameters were without deviations from the norm. A hanging X-ray of the abdominal cavity was taken, revealing dilated loops of the small intestine with fluid levels in the left epigastric region. Abdominal ultrasound found dilated loops of the small intestine, filled with liquid digestive contents, while the colon loops were visible only segmentally, with a collapsed sigmoid colon. In the right upper abdomen, a limited cystic fluid structure measuring 21×12 mm was visible (Fig. 1). Radiological diagnostics showed the features of high obstruction of the gastrointestinal tract. Due to the abovementioned symptoms, the child was urgently referred for surgery. Intraoperatively, obstruction of the gastrointestinal tract was found at the level of the ileum, 5 cm before the caecum, in the form of a lesion resembling Meckel's diverticulum. The change presented as a hard tumour, partially closing the lumen of the small intestine. Resection of the lesion with end-to-end ileal anastomosis was performed.



Fig. 1. Abdominal X-ray of the hanging-dilated loops of the small intestine with fluid levels in the left epigastrium

Inspection of the abdominal cavity did not reveal other abnormalities; the appendix was not removed (Figs. 2, 3). Intraoperatively collected material was submitted for histopathological examination. Macroscopically, in the intestinal wall, at a length of 2 cm, a cystic structure was found with a wall thickness of up to 0.2 cm, filled with watery fluid. The cyst significantly narrowed the lumen of the small intestine and modelled the serum. In contrast, microscopically, the duplication of the small intestine was seen, partly by the intestinal and gastric type epithelium, and the muscle membrane wall had nerve ganglia. No cancerous tissue was found in the tested material. Immediately after the surgery, the newborn was treated in the Children's Intensive Care Unit, and then in the Department of Paediatric Surgery. Treatment included broad-spectrum antibiotic therapy and parenteral nutrition up to and including five days. On the third day after the operation, the child passed stool. The course of treatment was uncomplicated. On the 9th day of hospitalisation, the wound was healed, and the continuous suture was removed. The girl was discharged home in a good general condition.

DISCUSSION

Gastrointestinal doubling is a rare developmental disorder with a frequency estimated to be $1:4,500^{(1,2)}$. It can occur in any part of the digestive tract, but most commonly it involves the small intestine^(1,2). Small intestine doubling accounts for as much as 45% of all reported cases of doubling⁽¹⁾.



Fig. 2. Intraoperatively visible change resembling Meckel's diverticulum



Fig. 3. In the removed fragment of the small intestine, it can be seen that the lesion only partially closed its lumen for the surgical instrument

It arises as a result of unsuccessful recanalisation of the digestive tube^(1,3). The doubling usually takes aspherical or tubular form, and often communicates with the digestive tract. It is located along the mesentery of the intestine and usually manifests itself as a thick-walled cystic structure, often palpable as a pathological resistance examined by the integuments^(2,3,5,7). Most typically, the diagnosis is established based on prenatal diagnosis of the foetus, while undiagnosed doubling manifests itself within the first two years of the child's life^(1,3). Mostly, these are isolated defects, but there are known cases of coexistence of other malformations such as congenital malformations of the anus, rectum, bile duct cysts, and liver cysts including intrauterine death of the foetus⁽³⁾. The most common symptoms of doubling include signs of obstruction of the gastrointestinal tract in the form of abdominal pain and vomiting with gas and stool retention. There may also be bleeding into the gastrointestinal tract and, less often, features of acute abdomen caused by intestinal torsion, intussusception or developing inflammation^(1,2,4,5,6). This doubling often widens significantly, resulting from the secretion of the ectopic mucous membrane of the stomach or intestines. If gastric mucosa is present, bleeding or perforation of the doubled fragment often occurs⁽¹⁾.

In the diagnostic process, an ultrasound examination of the abdominal cavity is performed, with a typical picture in children including the thickening of the cyst wall to 2-3 mm, which is associated with the presence of a smooth muscle layer and mucous membrane. This allows differentiation of the doubling from, for example, ovarian or pancreatic cysts, which usually consist of only one layer⁽¹⁻⁵⁾. The examination of choice is ultrasonography due to reduced exposure to X-rays. However, if ultrasound does not allow to establish the diagnosis, computed tomography or magnetic resonance imaging should be performed⁽⁵⁾. Diagnosis is challenging, and often causes difficulties even for experienced clinicians. Imaging studies can only establish a diagnosis in 25% of cases^(2,8). Due to the possible complications associated with this defect, prenatal diagnosis is extremely important. It allows for careful planning of surgical treatment, aimed at preventing complications later in the child's life, but a certain diagnosis can be obtained based on histopathological examination of a fragment taken surgically due to the possibility of locating the gastric mucosa in the doubling. In such cases, it should be differentiated with Meckel diverticulum, which can also be lined with it⁽¹⁾.

The treatment of choice is surgical resection of doubling^(1,2,3,5). Patients with asymptomatic doubling should also be referred for surgical treatment due to possible complications manifesting later in life. The scope of the operation depends on the size of the lesion and the topography of its location in relation to other important anatomical structures. Treatment outcomes are good, and mortality does not exceed 20%⁽²⁾.

CONCLUSIONS

Due to the clinical picture of gastrointestinal doubling and diagnostic difficulties, prenatal diagnosis is extremely important. Making a diagnosis already at the stage of foetal life provides sufficient time to properly plan treatment and avoid life-threatening complications. Early surgical intervention significantly reduces perioperative risk and thus accelerates the patient's recovery.

Conflict of interest

The authors do not report any financial or personal affiliation with other persons or organisations that could adversely affect the content of the publication and claim the right to this publication.

Author contributions

Original concept of study: MPK. Collection, recording and/or compilation of data: AK, MPK. Analysis and interpretation of data: AK. Writing of manuscript: AK, MPK, IM, MN, BP. Critical review of manuscript: MPK. Final approval of manuscript: AK.

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