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Rectal gastric heteroptopia in a child. Case-report of casuistic pathology

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ABSTRACT *Heterotopy of gastric mucosa in the rectum in children is a rare malformation to keep in mind when examining a child with a rectal bleeding. About 5 such clinical cases in children were described in the literature over the past 10 years. This condition is congenital, due to impaired tissue differentiation during embryogenesis. This case-report demonstrates the diagnostics and treatment of a child with rectal gastric heteroptopia.*

KEYWORDS: *heterotopic gastric mucosa, rectum, children*

CONFLICT OF INTEREST: *The authors declare no conflict of interest*

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In the practice of pediatric surgeons, heterotopia is quite common. Diseases such as Meckel's diverticulum, duplication of the gastrointestinal tract (GIT), as a rule, are accompanied by the presence of a focus of heterotopia of cells of the gastric mucosa, less often of the pancreas or epithelium of the respiratory tract [1,2]. However, isolated gastric heterotopia into the rectum, both in the pediatric population and in adult patients, is an extremely rare abnormality. It is known that heterotopia is a congenital pathology, in contrast to metaplasia — the transformation of the epithelium in the process of vital activity. The term 'heterotopia' is the presence of a morphologically normal type of tissue in a non-physiological area for it. Heterotopia is more common in the anterior intestine, which is explained by a violation of the migration of endoderm cells developing from a common germ during embryogenesis [3]. However, variants of heterotopia site outside the gastrointestinal tract are described: in the mediastinum, scrotum or biliary tract [3,4]. Such a diverse localization is explained by the fact that the pluripotent stem cells of the endoderm are able to differentiate into all tissues of the gastrointestinal tract. The error of this process leads to the fact that the gastric mucosa has the most diverse localization [3].

Since the first publication of this pathology by Ewell G.H., Jackson R.H. in 1939, about 30 cases of isolated heterotopy into the rectum have been described in the literature [5]. Over the past decade, only 5 similar clinical cases in children have been found in the available literature [5–9]. Domestic papers describing isolated heterotopia in the rectum in children could not be found.

Due to the lack of observational studies on this topic and the presence of only a small number of published clinical cases, indicators such as morbidity and prevalence of pathology are currently unknown. In the literature review by Iacopini, F. et al., which included 72 cases of various heterotopias in adults and children, this pathology was more common in men — 63% than in women — 37%.

The authors also traced the age at the time of pathology detection, the median of which was 22 years (1 day — 69 years) [10].

The clinical picture of gastric epithelium heterotopia in the rectum includes symptoms such

as rectal bleeding, often in combination with diarrhea (22%), tenesmus (22%), and abdominal pain (55%). In 20% of cases, an asymptomatic course of the disease is described [3]. In addition, the production of hydrochloric acid by displaced stomach cells leads to damage to adjacent tissues and the occurrence of complications such as bleeding, the occurrence of fistulas (recto-vesical, -vaginal, -perineal), perforation of the rectum into the free abdominal cavity or the pelvic cavity [10]. In addition, to date, there have been isolated publications describing the appearance of malignant formations against the background of existing heterotopia, including those with an asymptomatic course [11]. Thus, the issue of malignancy at the moment remains open for research and discussion.

Rectal bleeding is the most common symptom of gastric heterotopia in the rectum [10]. However, it is important to remember about a wide range of diseases accompanied by the clinic of gastrointestinal bleeding, such as Meckel's diverticulum, gastrointestinal duplication of various localization, juvenile polyps and other less common polypous syndromes, anal fissures, inflammatory bowel diseases (ulcerative colitis, Crohn's disease), as well as various variants of vascular malformations. Such a variety of diseases dictates the need for a thorough clinical and laboratory check-up of a patient with the onset of gastrointestinal bleeding at any age using instrumental research methods (esophagoduodenoscopy, ileocolonoscopy, videocapsular endoscopy, radioisotope examination).

It should be noted the need for performing a biopsy of the formation and mucous membrane of the intestinal tract for histological verification, which plays a crucial role in making a final diagnosis.

It is important that some imaging methods such as CT and MRI do not have sufficient sensitivity and specificity in case of suspected gastrointestinal heterotopia.

The described methods of conservative therapy of the disease — proton pump blockers, antibacterial therapy for the purpose of eradication of Helicobacterpylori (if the latter is detected in a biopsy), may for some time lead to the relief of bleeding and the inflammatory process, and even to the healing of ulcers. However, in 62% of cases, within 3 months after discontinuation of treatment, there was a resumption of symptoms, which required surgery. Only radical removal of the heterotopy focus led to a complete recovery [10].

There have been significant changes in surgery in recent years: from resection of the affected part of the intestine to precision removal of the lesion under endoscopic control. To date, in the practice of colorectal surgeons, preference is given to endoscopic methods of treatment — loop resection, including polypectomy, endoscopic mucosal resection (EMR), argonoplasmic ablation of the residual zone, endoscopic submucosal dissection (ESD). Despite the fact that in 17% of cases after endoscopic resection of the mucous membrane, residual zones of heterotopia were revealed, this method in the hands of experienced specialists can certainly be called the gold standard of treatment [8,10].

It is important to note that most pediatric surgeons have no experience in performing such endoscopic procedures, given the rarity of pathology. Fibrosis of the submucosa, which is naturally observed with this anomaly, as a result of a chronic inflammatory process, can also cause significant difficulties when performing surgery.

Our clinical case demonstrates a rare congenital pathology and the choice of treatment method in a patient with isolated gastric heterotopia.

The boy V., aged 10 years old, was admitted to pediatric coloproctology unit with complaints of an admixture of blood and mucus in the stool. When collecting history of the disease, it was noted that the above complaints periodically disturbed the child from the age of 2 years old. During the initial check-up at the place of residence, an anal fissure was revealed, which explained the hematocheesia clinic.

Conservative topical therapy (anti-inflammatory suppositories) was ineffective, relapses of rectal bleeding persisted. No additional check-up was carried out. For the first time at the age of 9 years old, after an episode of massive rectal bleeding, the child underwent a proctoscopy: on the back wall of the rectum 10 cm from the anal verge, a rounded, diverticular formation

with a diameter of about 2 cm with pronounced mucosal hyperemia along the circumference and contact bleeding was visualized. For further examination and determination of treatment approach, the child was transferred to a federal institution.

Upon admission to the clinic, the general condition of the patient was of moderate severity. When clarifying the clinical and anamnestic data, it was noted that there were no other symptoms, the hereditary anamnesis was not burdened. His physical development corresponded to his age. Ileocolonoscopy was performed, according to which the mucous membrane of the bowel was pink, smooth throughout; the vascular pattern was clear and uniform. Along the posterior wall of the rectum at a distance of 10 cm from the anal verge, a crater-like depression up to 2 cm in diameter with pronounced contact bleeding was visualized (Fig.1). With a digital rectal examination, it was possible to palpate only the lower pole of this lesion.

To clarify the anatomical features, an MRI of the pelvis was performed, the results of which described the presence of an uneven thickening of the mucosa up to 35–40 mm in length up to 80–90 mm along the posterior wall of the rectum, above the level of the anal margin by 9 cm. It was difficult to assess the depth and extent of the lesion (Fig.2).

Considering the extreme rarity of such neoplasms in surgical practice, the contradictory data of the check-up on the depth and prevalence of heterotopia in the rectal wall, as well as the lack of experience in endoscopic dissection in children, it was decided to refrain from using this technique.

It was decided to perform a resection of the rectum.

The child underwent a low anterior rectal resection of the rectum with the stapler anastomosis at a height of 3 cm from the dentate line with intraoperative proctoscopy to determine the distal resection margin, without the preventive ileostomy.

According to the histology of the removed specimen, fragments of the gastric mucosa of the fundal type were detected.

The lamina propria of the mucosa was edematous, with weakly expressed lymphoplasmocytic infiltration with an admixture of eosinophils (Fig. 3).

No complications occurred after the surgery. First defecation was on the 4th day. On the 10th postoperative day, the child was discharged home in a stable condition. With catamnestic observation after 1 month, the child's condition

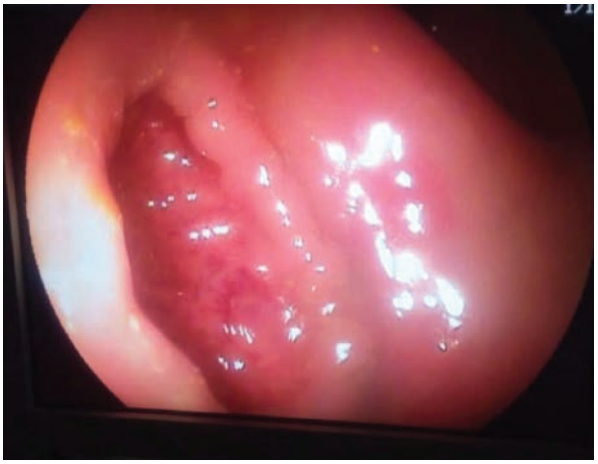

Figure 1. *Endoscopy image of gastric heterotopy of the rectum — lesion with a funnel-shaped depression in the center and with margins slightly raised*

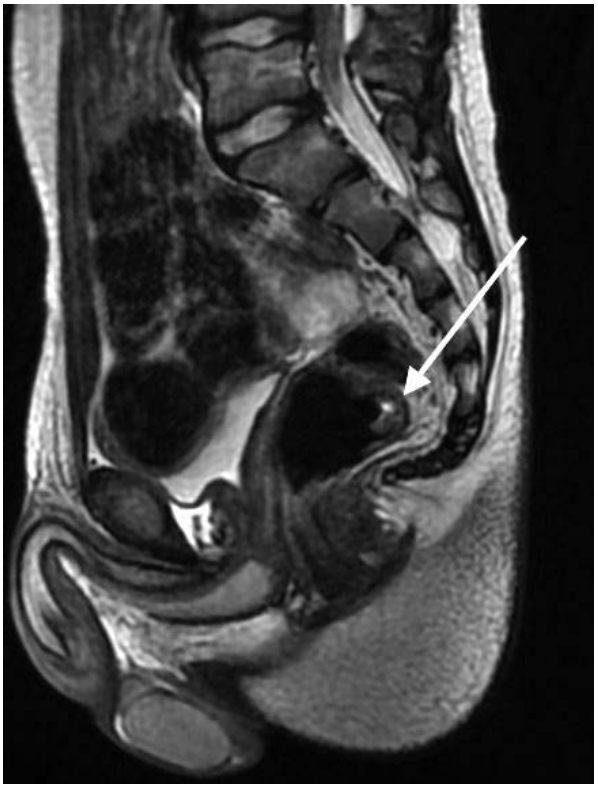

Figure 2. *Pelvic MRI of a patient with gastric heterotopy of the rectum. Local thickening of the rectal mucosa (arrow)*

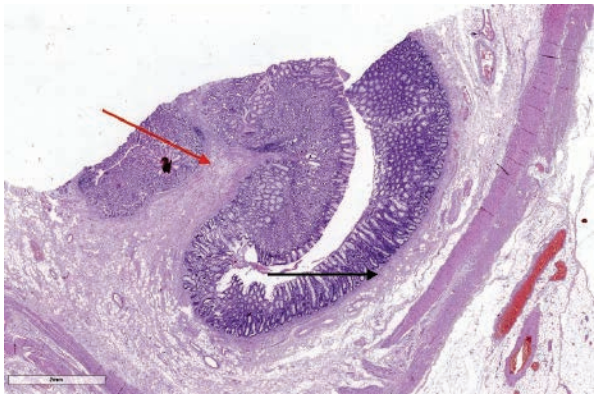

Figure 3. *Histology of gastric heterotopy. The site of heterotopy, represented by the cells of the fundus of the stomach (red arrow). Normal rectal mucosa (black arrow). Hematoxylin and eosin staining, ×11 increase*

was satisfactory. For 1.5 months after surgery, there was a syndrome of low anterior resection, with defecations 4–5 times a day.

During the control proctoscopy after 2 months, the rectal mucosa and the zone of anal sphincter did not change.

Despite the rarity of this disease, heterotopy should be included in the differential diagnostics when examining a child with a gastrointestinal bleeding clinic, of course, after excluding other more common diseases in childhood. This clinical case demonstrates the difficult path of finding a definitive diagnosis and choosing treatment approach for a child with an extremely rare congenital abnormality.

Nevertheless, such rather rare and casuistic clinical cases dictate the need to introduce new operational skills into the practice of pediatric surgeons to improve the quality of medical care for children.

AUTHORS CONTRIBUTION

Concept and design of the study: *Olga V. Shcherbakova, Linara R. Khabibullina*
Collection and processing of the material: *LinaraxR. Khabibullina*
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