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Case Report

A Rare Form of Rectal Atresia Associated with Rectovestibular Fistula

Evgeniy A Okulov[1], Aleksey A Gusev[1,2]*, Aleksey V Dotsenko[1], Elena Yu Dyakonova[1,2] and Sergey P Yatsyk[1]

[1]*National Medical Research Center for Children's Health, Russia*

[2]*RUDN University (Peoples' Friendship University of Russia), Moscow, Russia*

***Corresponding Author:** Aleksey A Gusev, National Medical Research Center for Children's Health, Moscow, Russia.

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Abstract

A clinical case of a girl with a rare form of rectal atresia in combination with a rectovestibular fistula, radically operated on at the age of 7 months is presented.

The elimination of the vestibular fistula was performed with the imposition of an "end-to-end" anastomosis of the atrezed parts of the rectal ampulla by posterior sagittal access.

In the postoperative period, the child had an independent regular satisfactory defecation with a moderate tendency to chronic constipation once every two days, stool retention up to 2-3 days was periodically noted.

The article presents the results of surgical treatment and the protocol of the operation itself in a rare clinical case.

Keywords: Rectovestibular Fistula; Rectal Atresia; Child; Anorectal Malformations

Introduction

In the practice of a pediatric surgeon, malformations of the anorectal region include a wide range of pathologies from ectopia of the anus to combined anomalies of the rectum and genitourinary tract. Many forms require emergency diagnosis and surgical treatment in order to avoid severe complications and death of the child. On average, the disease occurs in 1:3500 - 5000 newborns, more often in boys (up to 55-70%).

Anorectal anomalies in newborns are extremely diverse, so there are many classifications of these defects. Of the "usual" defects belonging to the main clinical group (according to the Conciliatory classification of anorectal defects Krikkenbek, adopted in the Russian Federation (Germany, 2005).

Of practical importance is the allocation of two types of anorectal atresia:

- Atresia of the anus and rectum without fistulas.
- Atresia of the anus and rectum with fistulas.

Anomalies of the second group, in turn, can be divided into:

- Anomalies with external fistulas (perineal fistulas; in girls - vestibular);
- Anomalies with internal fistulas (all fistulas with the urinary system, in girls - with the genitals).

Annually, up to 30 primary patients with anorectal malformations undergo an average of 30 primary patients with anorectal malformations in the surgery department of the National Medical Research Center for Healthcare of the Ministry of Health of the Russian Federation.

Of the "routine" defects belonging to the main clinical group (according to the Conciliatory Krickenbeck classification of

anorectal defects adopted in the Russian Federation (Germany, 2005), patients with rare anomalies occasionally appear in the department.

A clinical case describes rectal atresia with rectovestibular fistula.

Clinical Case

Girl M. was born from the first pregnancy, which proceeded against the background of anemia, edema and spontaneous urgent delivery. The malformation was diagnosed immediately after birth. When examined in the maternity hospital, the anus is located in the usual place, the catheter passes to a depth of 4 cm, then an obstruction is felt. On the eve of the vagina - a fistula through which meconium abundantly departs. The fistula does not communicate with the anus. A double-barreled colostomy was placed on the first day of life.

On examination in the department at the age of 7 months locally: the intergluteal fold is underlined. Large and small labia are well developed. The external opening of the urethra and anus in the proper place. When bougienage of the anus, Gegar's bougie No.12 freely passes up to an obstacle 4 cm from the anoderm (Figure 1, 2).

Figure 1: local status of the patient (photo) - explanations in the text.

Figure 2: local status of the patient (photo) - a plastic catheter was inserted into the vestibular fistula.

Examination carried out

Ultrasound of the kidneys and bladder revealed signs of transverse iliac dystopia of the right kidney.

Distal colostomy - through the excretory section of the colostomy, 50 ml of a water-soluble contrast agent was introduced, loops of the excretory section of the large intestine (sigmoid and rectum) were made. The ampulla of the rectum is dilated. The distance from the radiograph to the rectum is 2 cm. The communication of the rectum with the vagina is determined (Figure 3, 4).

Figure 3: Colostography in direct projection (X-ray) - radi-opaque mark in the anus.

Figure 4: Colostomy in the lateral projection (X-ray) - radiopaque mark in the anus.

MRI of the small pelvis - on the obtained images, hypoplasia of the typically located left kidney is determined - its vertical dimensions are 30 mm, transverse 18x13 mm, with the presence of cystic inclusions in its parenchyma. The differentiation of the kidney is practically not determined. The right kidney is dystopic in the lumbar region, located prevertebral at the level of L2-S1. The kidney is irregular in shape, enlarged, deformed, the differentiation of the layers of the kidney is not disturbed (Figure 5).

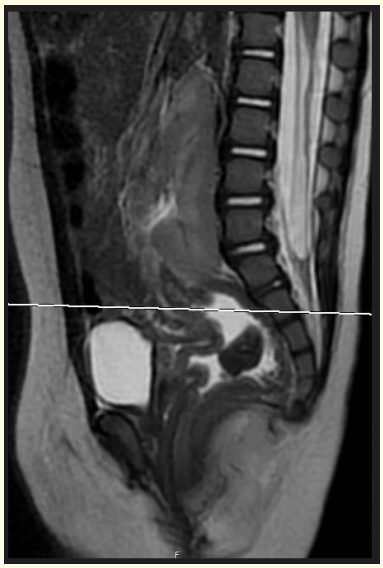

Figure 5: Magnetic resonance imaging - explanations in the text.

Computed tomography of the pelvic organs with vascular contrasting - the contrast agent was injected through the drainage tubes (into the cavity of the bladder, into the outlet end of the colostomy, into the ampoule of the rectum and the lumen of the fistulous tract) and intravenously. After the introduction of a contrast agent, the fistulous tract is contrasted for up to 36 mm to the level of S4-S5 vertebrae, connecting the lumen of the distal part of the blindly closed intestine and the area of the vestibule of the vagina, up to 2 mm in diameter. Presacral at the level of the S5 vertebra, a blindly closed ampulla of the rectum is determined (3 mm below the mouth of the fistulous tract). The left kidney is deformed, hypoplastic, vertical size up to 27 mm. The accumulation of contrast agent is reduced (Figure 6).

Figure 6: Computed tomography - side view (explanations in the text).

On the basis of examinations, the diagnosis was made: rectal atresia with rectovestibular fistula, megarectum, carriage of a double-barreled colostomy. Hypoplasia of the coccyx. Syringomyelic cyst of the conus of the spinal cord. Lumbar dystopia of the hypertrophied right kidney, hypoplasia of the left kidney. MRI data for the presence of additional formations of the small pelvis and presacral region were not obtained.

On March 22, 2019, the vestibular fistula was surgically eliminated with the anastomosis of the atrezed parts of the rectal ampulla end-to-end from the posterior sagittal approach.

After anesthesia, the bladder was drained with a Ch 8 Foley catheter and the existing rectovestibular fistula was drained with a Ch 6 silicone catheter.

Next, the child is placed in the position of a folding knife. An incision was made along the intergluteal fold. Basically, a blindly ending section of the rectum and a part of the rectum with a rectovestibular fistula within the Denon-Villiers capsule were isolated in a sharp way. The seams between them are very tight. It was also possible to visualize and mobilize the anterior wall of the blind zone without damaging the vagina. A fistula leading to the vestibule of the vagina was revealed. The latter was extirpated, leaving a passage to the navicular fossa up to 1.5 cm long due to the risk of microcirculation disturbance in the caecum of the rectum. The latter has a well-defined muscular wall, is recognized as viable and functionally fit. The proximal and distal parts of the rectum were resected sparingly and end-to-end anastomosis was performed with a single-row continuous 4/0 Vicryl suture. Then levatoroplasty was performed. The anus with a well-formed dentate line, due to the absence of muscles of the sphincter ring along the anterior semicircle, was mobilized to a depth of 1.5 cm, after which an anterior sphincteroplasty was performed, the anus was displaced posteriorly by 1 cm. figure 7, 8).

Figure 7: The stage of the operation, during which the distal and proximal parts of the rectum are isolated, with a rectovestibular fistula.

Figure 8: Stage of the operation - performed single-row anastomosis, rear view.

Results

The postoperative period proceeded smoothly. The colostomy was closed 3 months after radical surgery on 06/25/2019.

In the interhospital period, the child had an independent regular satisfactory defecation with a moderate tendency to chronic constipation once every two days, stool retention up to 2-3 days was periodically noted. At the same time, cleansing enemas were performed with a minimum volume, against which a satisfactory emptying was noted. With stool retention up to 2 days, the child had calomasia. The urge to defecate is determined (Figure 9,10).

Figure 9: Appearance of the perineum after surgery (Photo).

Figure 10: X-ray contrast examination of the colon (X-ray).

Conclusion

Thus, this clinical example emphasizes the diversity of anorectal malformations, once again proves the need for a comprehensive preoperative examination, as well as the need for long-term rehabilitation of children with anorectal malformations in the postoperative period [1-7].

Conflicts of Interest

We have no conflicts of interest and nothing to disclose.

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