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Rectourethral Fistula in Children with Anorectal Malformation

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Abstract

The article presents the clinical manifestation and data of special research methods for rectourethral fistula of 52 boys with ARM observed in the clinic in 2009-2019. Rectourethral communications can be congenital or acquired (iatrogenic origin); their identification is difficult. In some patients, the fistula remains undetermined before - and during the operation. The high frequency of concomitant anomalies requires a comprehensive examination of patients to reveal associated anomalies.

Keywords: *ARM, rectourethral fistula, boys, diagnosis.*

Relevance

Anatomical and topographic features of the pelvic organs and differences in the genital organs in boys and girls cause a different frequency of anorectal malformations(ARM). Rectourethral fistulas (RUF) in rectal atresia observed in boys are localized in the prostatic and bulbar parts of the urethra. Rectourethral and rectoperineal fistulas are rarely found with a normally formed anus. (fig. 1). According to various authors, the frequency of RUF among ARM in boys is 14,5% - 25,9% [2,3,7,10]. This variability can be explained by the fact that fistulas of this localization in some cases stay at the stage of diagnosis and during the initial operation. Mostly, they are considered to be no fistula forms of ARM. In some patients, fistula may form due to injury of the urethra during corrective surgery [8,9]. Thus, rectourethral fistulas can be congenital or acquired - of iatrogenic origin. In practical terms, it is important not only to ascertain the presence of a fistula, to determine its origin, but also to clarify the nature of concomitant anomalies, assess the condition of the distal colon and anatomical structures that provide anorectal retention, this is important when choosing therapy [1,4,5,6,13,14].

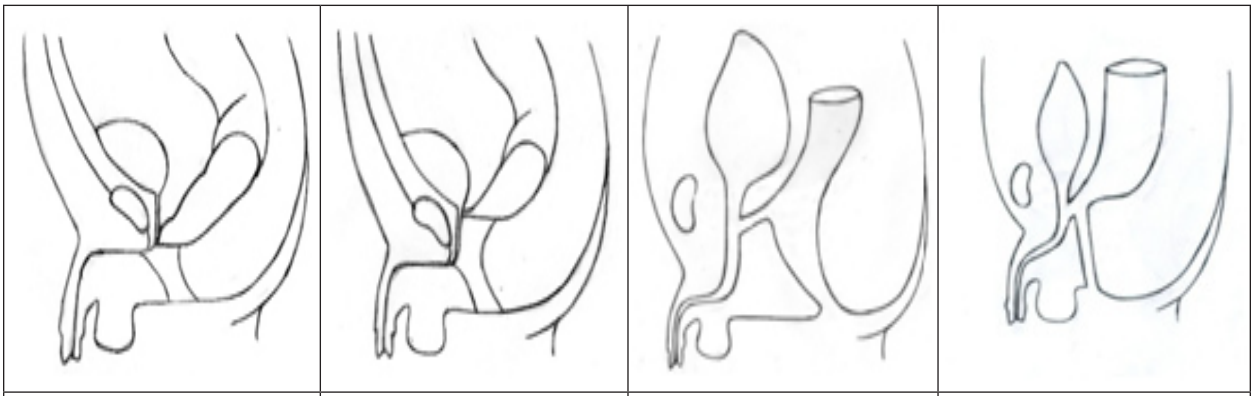

Fig1. *Types of rectourethral fistula: a) rectobulbar; b) rectoprostatic c) H-form of rectourethral fistula; d) rectourethral and perineal fistula. Scheme of drawings from the manual Holschneider M.A., Hutson M.J., 2006.*

The purpose of the work is to determine the rate, clinical and anatomical features, diagnosis of rectourethral fistulas in boys using the material of the clinic.

Material and Methods

In the clinics bases of the Department of Hospital Pediatric Surgery of TashPMI in 2009 - 2019, we were treated among 495 patients aged 1 day to 15 years with ARM. In 246 (49,7%) boys, various forms of anomalies were established. Patients have been distributed in accordance with the International classification adopted in Krikenbek [10]. The following forms of ARM were diagnosed: anorectal agenesis without fistula in 80 (32,5%) patients, perineal fistula in 65 (26,4%), anorectal agenesis with rectourethral fistula (29 rectoprostatic fistula and 22 rectobulbar fistula) - 51 (20,7%), rectourethral fistula with a normally formed anus "H-form" - in 1 (0,4%), rectovesical fistula - in 9 (3,7%), anal stenosis - in 14 (5,7%), covered anus in 15 (6,1%), ectopic anus in 5 (2,1%), Pouch colon - 6 (2,4%) children.

To clarify the anatomical form of the ARM and reveal concomitant defects of other organs and systems, a comprehensive examination has been performed, the state of the perineum was assessed, the anatomy and topography of the external genital organs were determined; ultrasound, radiological, CT and MRI studies have been done. The results of clinical and instrumental studies have been proved byoperatios data.

Result and Discussion

When examining newborns with no anus, an examination was performed to assess the general condition and identify associated abnormalities. Endoscopy of the stomach was performed to exclude atresia of the esophagus, to determine the amount and character of the gastric contents, to detect intestinal obstruction. Taking into account the rarity of the without fistulous forms of rectal agenesis in the ARM, we carefully examined the children to identify the "invisible", difficult to diagnose pathological anastomoses of the rectum with adjacent organs, including the urethra.

In newborns with rectal atresia, there is no anal opening, where a small depression or hyperpigmented cushion is seen. The middle suture of the perineum at this point is discontinued. Irritation of this area with a sharp object or electrical stimulator causes anal reflex. This indirectly indicates the safety of the anatomical structures that provide the functions of the obstructive mechanism of the rectum. The results of the anal reflex were different: from barely noticeable to normal. This is because of the severity of the anal reflex depending not only on the condition of the obturator apparatus of the rectum, but also on the somatic background caused by various attendant diseases, the degree of maturity of the newborn. In doubtful cases or with a negative reaction of the anal reflex during the observation, some of the children in subsequent years show positive dynamics. It is very difficult clinically to evaluate the persistent functional impairment caused by the underdevelopment of the muscular complex and abnormalities in the development of the sacrum and coccix, to establish the height of the atresized end of the rectum relatively to the normal location, therefore special examination methods are needed.

Pathognomonic manifestations of atresia of the rectum with a fistula with the genitourinary system are meconuria and pneumouria, detected within a few hours to 2 days after the birth of the baby. With an urethral fistula during urination, the urine is initially cloudy, then it becomes clearer; periodically, regardless of the act of urination, gases and feces are excreted. With rectovesical fistula, intestinal contents are constantly mixed with cloudy urine, throughout urination. The absence of these signs does not exclude the fistulous form of atresia since they depend on the degree of pneumatization of the intestine and the filling of the blind sac of the terminal section of the intestinal tube, which is necessary to increase the pressure in it to a threshold level. The anatomical and morphological features (diameter, extent, direction) of the fistulous course are also important. Narrow fistulas can be blocked by viscous meconium or mucus. These changes may be due to impaired passability of the overlying sections of the intestinal tube (atresia of the esophagus, duodenum), birth injury, and prematurity. Under these conditions, signs of complete or partial obstruction of the fistula are more pronounced with a narrow course. In the course of active observation, measures are needed aimed at restoring the water-electrolyte balance, correction of hemodynamic disturbances, contributing to an increase in intestinal contents, liquefaction of meconium, movement in the intestine and increase in pressure in its final section. While diagnosing, catheterization, washing of the

bladder, repeated microscopic analyzes of urine to identify pathological impurities are helpfull.

With atresia of the without fistulous forms, in order to determine the level of the blind segment of the rectum in newborns, an invertogram is performed in the lying position with a stomach raised on the roller and bent hips at 45 degrees for 16-20 hours after birth. As our studies show, an echoscopic scan of the perineum performed by 24 patients is informative simple diagnostic method which, along with determining the height of atresia in some cases, allows you to identify the fistulous course.

Analysis of clinical material shows that rectourethral fistula is mainly observed with rectal atresia (51 out of 52). H-forms of messages with the formed anus are an extremely rare type of anomaly (1 – 0,4%), which corresponds to published data. In inadequate diagnostics and imperfect operational tactics, a rectourethral fistula is often mistaken for a without fistulous form. Of the 52 patients with rectourethral fistulas, the correct preoperative diagnosis was made during the neonatal period in 4 (7,7%) patients who underwent early perinealproctoplasty with elimination of the fistula, and 15 (28,8%) after perineal proctoplasty. In 26 (78,8%) of 33 patients who underwent sigmoid placement, the pathology was regarded as a non-fistulous form. The presence of rectourethral connections was established in the clinic during a comprehensive examination. In some of these children, the risk of iatrogenic damage to the urethra during an initial operation by an insufficiently qualified surgeon is not excluded. The performed of a divided preventive sigmastoma or perineal access surgery without a thorough revision during the neonatal period intricates the identification of rectourethral connections without special research methods such as ascending urethrocystography and distal colostography under increased pressure with a water-soluble contrast agent.

Examination of children in the presence of H-forms of fistulas includes the study of the anatomical and functional features of the anorectal zone and localization of the fistula. The simplest and most informative diagnostic technique is the examination of the rectum using a mirror with the introduction of methylene blue through the external opening of the urethra. It is also necessary to conduct urethrocystoscopy, urethrocystography, antegrade and retrograde irrigography with a water-soluble contrast medium and excretory urography. Valuable information for diagnosis can be obtained by catheterization of the fistulous course when performing urethrocystoscopy under medicated potentiation. A comprehensive examination allows not only to identify the fistula, but also to clarify its anatomical version, localization; propose of changes in the urethra, distal intestines; determine the nature of the associated pathology of the urinary tract. The data of a comprehensive study aimed at identifying concomitant anomalies and assessing the anatomical and functional state of the anorectal zone allow us to determine the tactics and method of treatment.

As a rule, H-forms of rectourethral fistulas of congenital and iatrogenic origin or with relapse due to underdevelopment of the stump of a crossed fistula (6-11,5%) after primary operations are detected in children in infancy or later. Patients have non-physiological options for the discharge of urine or intestinal contents (through the urethra and anus) of varying intensity, depending on the onset of the fistula in an obliquely descending direction from the urethra or rectum. With fistulas starting from the urethra, when urinating, urine is excreted from the urethra and through the anus 5 (9,6%). Over time, the amount of urine released naturally decreases. Apparently, this is due to the gradual stenosis of the distal urethra in relation to the localization of the fistula. In 6 (11,5%) children with a fistula of oblique direction from the rectum, the intestinal contents of the gas from the urethra have been noted. In 10 (19,2%) cases, when urinating, urine is evenly excreted from the urethra and rectum. Occasionally, feces and gases escaped through the external opening of the urethra. This picture is due to the extended connection of the rectum with the urethra damaged during proctoplasty. In some children, after palliative intervention (sigmastoma application), the ingestion of intestinal contents into the urinary tract is excluded or sharply reduced, which makes it difficult to identify the fistula. In some patients, after proctoplasty and / or stoma of the colon, complications such as fecal incontinence of various intensities, cicatricial deformities of the anus, or their combinations are developed. Under these conditions, it is difficult to identify the nature of the fistula, although changes that are uncharacteristic for congenital and acquired rectourethralconnections can be distinguished. Congenital fistulas, as a rule,

open in the prostatic and bulbar parts of the urethra, have a more or less pronounced transitional channel. Fistulas of iatrogenic origin are often localized in the membranous part with soldering to the wall of the intestine and urethra, which is deformed and stenosed at the site of the anastomosis. With rectourethral fistulas, an ascending urinary tract infection often develops. Infection occurs the easier, the larger the fistulous opening and the stool in the rectum lingers longer due to cicatricial narrowing of the anus. Leakage of urine through the rectum causes the child additional suffering, supports maceration of the perineal skin and itching in this area. Complications after perineal operations in some patients are manifested by violations in the form of insufficiency of the anus with fecal incontinence or chronic constipation of varying intensity. With decompensated colostasis, the distal colon gradually expands, megacolon syndrome occurs. The indicated condition, which occurs with asthenisation, anemia, and hypotrophy, is more pronounced when anorectal anomaly is combined with lengthening of various parts of the large intestine and a megarectosigmoid, established by contrast irrigography.

A comprehensive examination of 38 (73,1%) patients revealed 65 association malformations. In 15 (39,5%) patients, association anomalies were isolated. Associated anomalies of two or more systems - multiple malformations - were found in 23 (60,5%) children. Along with visible anomalies during a comprehensive examination, malformations of other organs and systems were established. In their structure, spinal pathology predominated significantly - in 25 (65,8%); abnormalities of the urinary systems - 23 (60,5%), the gastrointestinal tract - in 8 (21,0%), the cardiovascular system - in 4 (10,5%). The number of anomalies prevailed in patients with rectoprostatic fistula (26-68,4%) compared with patients with rectobulbar fistula (12-31,6%), which coincides with the data of individual authors and is reflected in the results of treatment [4,5,11,12]. Our data on the fact that the clinical examination and examination methods for newborns with ARM in most cases allow us to establish the type of anomaly and decide on the tactical approach correlate with the literary ones. The exception is rectourethral fistula, which is difficult not only at the stage of diagnosis but also during surgery.

Conclusions

Rectourethral fistula in the structure of the ARM in boys compose 20,7%, mainly observed with atresia of the rectum and is localized in the prostatic and bulbar parts of the urethra.

The H-form of a rectourethral fistula with a formed anus is rare. Such anastomoses detected after procto plasty or the application of a preventive stoma are not diagnosed as iatrogenic injuries during surgical correction.

Diagnosis of rectourethral fistula is complicated. To prove the presence or absence of such connections, the identification of concomitant pathology requires special diagnostic methods.

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