X-ray analysis of published studies of rectal atresia. Review.

Abstract

It is known that patients with rectal atresia (RA) or stenosis (RS) have a normal anal canal. The narrowing or complete atresia is located a few centimeters proximal to the dentate line. The ARM-Net Consortium authors analyzed 36 (1.3%) of 2619 patients with anorectal malformations: RA (n = 18) or RS (n = 18). They emphasize that due to the low level of evidence, it is impossible to judge the benefits of a particular treatment method. The purpose of this study is to review the diagnostic methods used to evaluate RA/RS. A total of 18 radiological studies of RA/RS were identified, but 4 were excluded because the diagnosis was not consistent with the clinical and/or radiological findings. We used a previously developed x-ray method, as well as studies on the anatomy and physiology of the anorectal zone. In 8 cases, radiological data indicated the presence of a septum not in the rectum, but in the anal canal approximately 1 cm from the anus. In 6 cases, on radiographs, the distal contour of the intestine was located at the level of the pubococcygeus line, as is normal. Consequently, the obstruction was in the anal canal, but since conditions were not created to open the upper part of the anal canal, it was impossible to judge the level of obstruction. Conclusion. The study showed that RA/RS is not located in the rectum, but in the anal canal approximately 1 cm from the anus and is represented by the septum. This makes it possible to excise the septum through anal access without damaging the anal canal. Methods of diagnosis and treatment are described. It has been hypothesized that RA/RS occurs due to disruption of the fusion of the endodermal and ectodermal primordia of the anal canal.

Keywords: anorectal malformations; anorectal physiology; rectal atresia; rectal stenosis; x-ray study; anorectal embryology.

Patients with rectal atresia (RA) and stenosis (RS) have a normal anal canal. Stenosis or complete atresia is located several centimeters proximal to the dentate line [1]. The authors of the ARM-Net Consortium identified and analyzed 36 (1.3%) of 2619 patients with ARM: with RA (n = 18) and RS (n = 18) [2]. They emphasize that the Sharma and Gupta classification is not yet generally accepted, and it had not been applied to most patients. Low level of evidence does not allow for the assessment of different types of RA/RS and the advantage of any of the operations. In the article by the authors of the Consortium there is no analysis of methods for diagnosing RA/RS.

The purpose of this study is to review the diagnostic methods used to evaluate PA/PC.

Material and methods. We searched for articles published in PubMed describing and illustrating the diagnosis of PA/PC. A total of 18 such works were discovered. However, 4 of them were excluded from the analysis since the clinical data did not correspond to the diagnostic data.

Methods. X-ray analysis is based on the normal anatomy and physiology of the anorectum [5]. To obtain true dimensions, we use either contrast markers of known size or previously calculated height of L-1 in children of different ages **(Table 1)** [3,4].

Table 1. Height L-1 depending on age.

Age (years)	1	2	3	4	5	6	7	8	9	10	11	12	13	14-15
Height L-1(cm)	1.3	1.4	1.4	1.5	1.5	1.6	1.7	1.8	1.8	1.8	1.9	2.0	2.1	2.2

The starting point can be the length of the anal canal from the pubococcygeal line to the marker near the anal dimple. Since the standards for the anal canal length in children of different ages are known (**Table 2**) [3,5], all other parameters can be calculated on the radiograph.

Table 2. The true rectal width and anal canal length depending on age.

Age	n	Rectal width (cm)	n	Anal canal length (cm)
5 days-11 months	12	1,3-3.0 (2.24±0.09)	7	1.7-2.5 (2.21±0.15)
1-3 years	9	3.0-3.7 (3.21±0.11)	7	2.3-2.8 (2.55±0.10)
4 – 7 years	9	3.0-3.9 (3.43±0.14)	8	2.3-3.6 (3.11±0.10)
8 – 10 years	9	3.2-4.1(3.70±0.06)	8	2.6-3.7 (3.07±0.11)
11 – 15 years	19	3.6-4.6 (39.5±0.07)	18	3.1-3.9 (3.43±0.10)

In full-term newborns, the length of the anal canal is 1.7 cm, and in premature ones 1.5 cm [3].

Results. In 8 patients the atresia was in the anal canal and in these cases, it was possible to measure the approximate length of the anal canal above and below the level of obstruction. This was shown best in the article by Stenström et al [6]. **(Figure 1**).

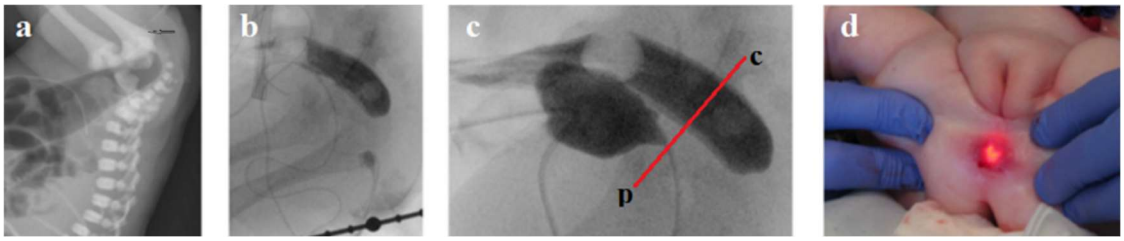

Figure 1. (a-b). «The distance between the endings was approximated to 2 cm» [6]. The black arrow (a) shows the end of the rectal probe. (c). During a combination of colostogram and contrast in the urinary bladder, there was an opening of the upper part of the anal canal distal to the pubococcygeal line drawn by me. (d). "The endoscope was pressed against the rectal atresia, and with the help of external pressure, the endoscope could be seen 1 cm up in the anal canal" [6].

This is one of the best studies. Its positive aspects: (1). The authors took lateral radiographs, which can be used to determine the condition of the rectum and anal canal. (2). The rectum was washed repeatedly so that thick stool did not interfere with the definition of the contour of the upper segment. (3) Colostogram (c) was made with high rectal pressure, which caused the anal canal to open above the septa. This study would have been more informative if the authors had used a contrast mark near the anus or coated the perineum between the buttocks with barium paste. On radiographs (a,b), the intestine, as normal, is located above the pubococcygeal line. The large gap between it and the end of the anal catheter is explained by the fact that the upper part of the anal canal, as normal, is in a closed state. The authors state that "The 2 cm thick wall between the endings of the rectum was divided with a diathermy between two stay sutures after which the coloscope came through the anus." First, the authors refer to the anal canal as the rectum. Secondly, it is known that at the age of 4 months the length of the anal canal is approximately 2 cm. Therefore, the septum cannot be that thick, i.e., occupy the entire anal canal. Moreover, about 1 cm of the anal canal, according to the authors, was distal to the septum, and at least 1 cm below the pubococcygeal line. From which it follows that the partition was very thin since the true length of the anal canal is about 2 cm. Probably, in the closed anal canal the surface is much smaller than in the open state and the excess of the mucous membrane created the appearance of great thickness. The described operation opens wide possibilities for saving the anal canal, which significantly distinguishes it from pull-through operations.

Atresia of the anal canal in the form of a septum was found in 8 cases [6, 7,8,9,10,11,12,13]. The approximate dimensions of the upper and lower parts of the anal canal, i.e., above and below the septum, are given in **Table 3.**

Table 3. Approximate dimensions of the anal canal above and below the anal septum.

Reference (#)	6	7	8	9	10	11	12	13
Upper AC	1.0	0.8	1	0.8	0.3	0.8	1.1	0.7
Lower AC	0.7	0.9	3	0.8	1.4	0.9	1.2	1

In 2 of 8 cases [8, 12] had a hole in the anal membrane, consistent with the diagnosis of rectal stenosis (RS).

All these observations are characterized by the presence of a septum below the pubococcygeal line, where the length of the anal canal above and below the septum could be measured. However, the numbers in Table 3 are not accurate because the measuring instruments were not accurate (fingers, the projection magnification on radiographs was not considered). In addition, only frontal radiographs were more often taken, на которых невозможно точно определять размеры прямой кишки и анального канала. The use of solid rectal catheters or Hegar dilators cause elevation of the pelvic floor and septum, that results in a decrease in the length of the upper part of the anal canal (**Figure 2**).

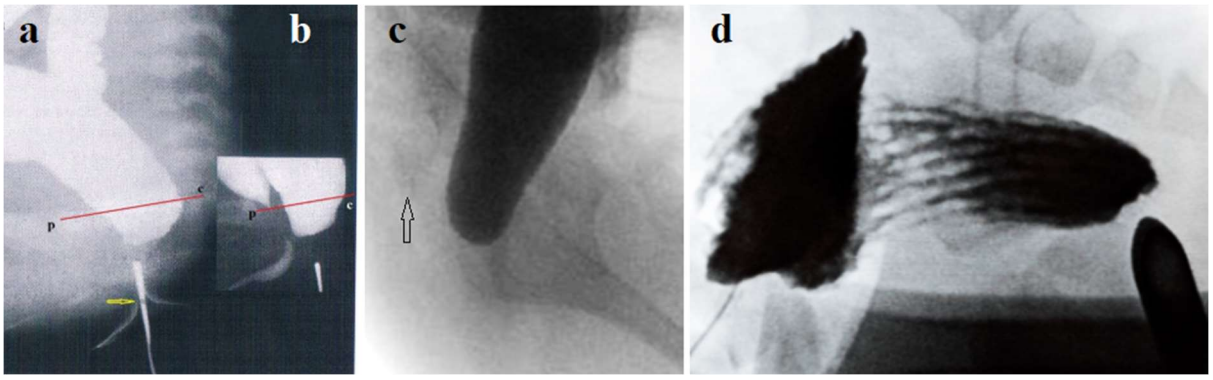

Figure 2.

(a-b). From the article by Kobayashi et al [7], where it is said that in a 3-day-old male “a firm 8-Fr catheter could be introduced only 2 cm into the anal canal from the anal verge” [7]. Since in newborns the true length of the anal canal between the pubococcygeal **(p-c)** line and the anus is 1.7 cm, the true length between the anus (yellow arrow) and the septum is 0.7 cm. The difference in measurement is due, firstly, to the lack of marking of the anus, and -secondly,

pushing a firm 8-Fr catheter causes the pelvic floor to rise, which increases the perception of the length of the anal canal below the septum. The diastasis between the upper and lower segments of the anal canal depends on the degree of opening of the upper part of the anal canal.

(c-d). Colostogram from Kurashima et al [11] shows (c) that the distal contour of the rectum is just below the last coccygeal vertebra (arrow). (d). During the insertion of the Hegar dilator, the rectum moved upward.

In 6 cases, the rectum was located at the level of the p-c -line, as is normal [13,14,15,16,17,18]. This means that atresia can only be located caudal to the rectum, i.e., in the anal canal. However, no evidence of either the presence of atresia itself or the location of the septum in the anal canal was found in the drawings in these articles (**Figure 3).** When analyzing 14 observations, not a single case was identified where intestinal obstruction could be localized in the rectum. We found only thin-walled partitions. Other models described by Sharma and Gupta [19] were not found. In 3 (21%) of 14 studies, anal canal membrane was combined with other anorectal malformations [7,9,18].

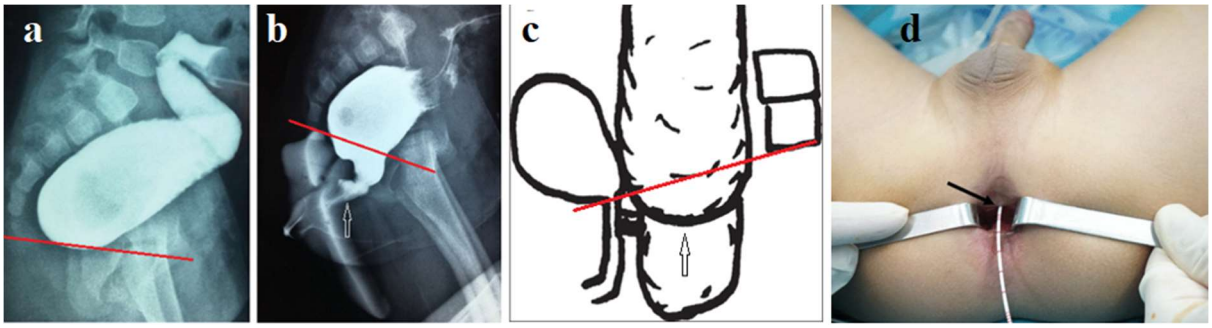

Figure 3. (a). On colostogram [14] the wide rectum is located at the level of the p-c line. A megarectum had already arisen by the time of the surgery due to poor cleansing of the rectum. Low rectal pressure does not lead to the opening of the anal canal, so it is impossible to judge the presence and level of the diaphragm in the anal canal. **(b).** Contrast study after transanal recto-anal anastomosis [14] showing megarectum and serious damage to the puborectalis muscle with descending perineum syndrome, which inevitably leads to chronic constipation and fecal incontinence. **(c).** From the article by Thakur et al "Schematic diagram of rectal diaphragm in imperforate anus with delineating fistula and only two sacral pieces" [18]. **(d).** From the article Huang et al the rectourethral fistula (arrow) was located 1 cm above the anus [20].

All of the above observations indicate the presence of a thin septum in the anal canal and its accessibility for excision from the anal access.

Discussion

About the scientific level of articles

In contrast to the ARM-Net Consortium, which assessed radiology reports for the presence of additional brain, cardiac, renal, vertebral, skeletal and spinal cord anomalies [2], we performed a radiographic analysis of anorectal studies, comparing them with the standards described in our articles [3,4,5] and with clinical data. Our analysis confirms the Consortium's conclusion that the majority of studies are of low quality. In addition to typical cases of RA, observations have been published, the publication of which leads to unfounded theoretical conclusions. These cases need to be examined to protect the important patterns specific to ARM. The article by Sharma et al presents a case of rectal atresia with left rectolabial fistula [21]. Invertogram along with feeding tube in situ "suggested membranous rectal atresia" to the authors. «The membrane was perforated blindly by using Hegar's dilator following which meconium was coming from the fistulous opening» (**Figure 4).**

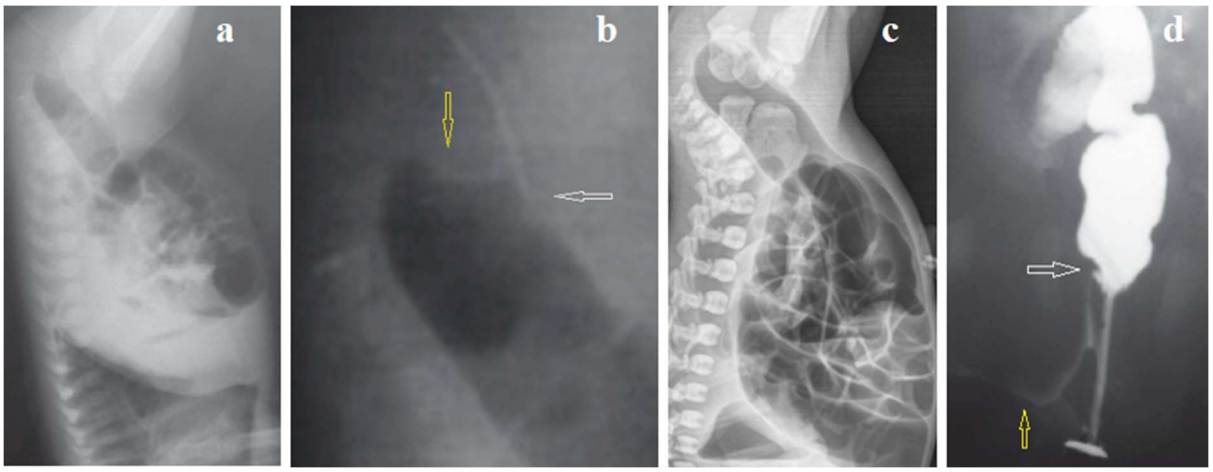

Figure 4. (a). Invertogram by Sharma et al [21]. **(b).** Enlarged and edited image of the rectum. The end of the feeding tube is above the distal contour (white arrow). There is concavity of the anterior wall of the proximal segment (yellow arrow), formed by external pressure. **(c).** This is not typical for normal invertogram images (see Figure 1) [6]. **(d).** The fistulogram shows the connection between the fistula channel and the anal canal, as well as with the rectum, where there is a depression and a break in the contour (white arrow). The length of the contrast channel, between the rectum and the transverse segment, which usually serves as a contrast marker for marking the anus, is 2 times the width of the rectum. The transverse marker, which

would normally be near the anus, in this image protrudes above the fistula in the labial (yellow arrow). This means that the marker is away from the anus.

1). The explanation for this case contradicts the known pattern of congenital ARM. Ectopia of the anus always occurs strictly in the central sagittal plane. Urethral fistulas always penetrate the urethra. Long subcutaneous fistulas in boys sometimes reach the root of the scrotum, but do not move to the sides. 2). There are no signs of rectal atresia either in the description or on the radiographs. 3). The fistulogram clearly shows two acquired fistulas. 4). Deformation of the rectal contour is caused by a hematoma due to trauma. 5). In all age groups, the length of the anal canal is approximately equal to the width of the rectum (**see Table 2**). The increase in the length of the anal canal by 2 times compared to the norm (the width of the rectum), considering all the previous discrepancies, can be explained by the fact that it was drawn. X-ray analysis and comparison with scant clinical data indicate acquired fistulas. This article should not be considered for scientific research as it misrepresents the scientific understanding of APM.

Articles by Lane et al [17] and Hamrick et al [22] state that rectal atresia is also associated with the presence of a presacral mass and sacral abnormalities. In none of the 14 analyzed cases was the combination of RA/RS with a presacral mass described. Below is an observation that may clarify this issue (**Figure 5**).

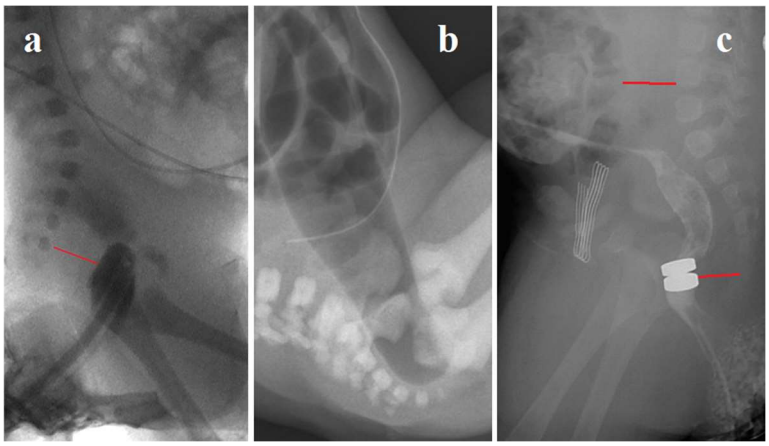

Figure 5. The authors of the article assessed the results of the examination as follows: clinical examination of a **1680 g** male revealed a normal appearing anus but there was failure to pass meconium. On **digital rectal examination** there was a blind ending rectum 2 cm above the dentate line. A plain abdominal X-ray revealed dilated loops of bowel, and a contrast enema demonstrated rectal atresia **(a)** [23]. However, the description **(a)** does not correspond to reality. Firstly, the sharp expansion of the retro-rectal space is noteworthy. Secondly, there is no

dilation of the intestinal loops. Third, there is no dilation of the rectum over the supposed site of obstruction, as can be seen in the example **(b)** from the article by Stenström et al [6]. In Figure **(c)** can see the forward displacement of the magnets caused by the formation located behind the rectum. At the same time, in the presence of a colostomy, intestinal aeration is not much different from the first radiograph **(a).**

Analysis of this case indicates the absence of rectal atresia. The disruption of passage was caused by compression of the rectum by the pre-sacral mass. However, the idea of using magnamosis to treat rectal atresia is quite reasonable.

About the anatomy and physiology of RA/RS. Errors in diagnosis and choice of treatment methods are primarily due to a lack of knowledge about the anatomy and physiology of the anorectal zone. Our analysis suggests that (1) the area of obstruction is not in the rectum, but in the anal canal; (2) presumably it is 1 cm from the anus; (3) all data indicate the presence of a membrane; (4) RS is a perforated membrane; (5) the high percentage (21%) of the combination of RS with other ARMs suggests their common origin; (6) the pre-sacral mass disrupts rectal patency, mimicking RA/RS. From the above it follows that for the name to correspond to the anatomical essence of the defect, it should be called “anal canal atresia”.

Diagnosis of RA/RS. Most pediatric surgeons have performed an invertogram, which is based on the false assumption that gas in the intestine moves upward. Firstly, any bolus moves through the intestines only because of peristalsis. Secondly, at low ARM [24], as well as at RA/RS, the anal canal at rest is in a closed state, as well as normally [3]. It opens only when rectal pressure rises to a certain level. Then the rectum, adapting to the new volume, relaxes, which leads to a decrease in rectal pressure. This causes the anal canal to contract, which pushes gas back into the rectum. Therefore, to accurately record the opening of the anal canal, fluoroscopic observation is necessary.

Method of X-ray examination in newborns. If the catheter or thermometer is delayed not far from the anus, it is advisable to lubricate the perineum near the anus with a paste containing a contrast agent. To accurately determine the location of the septum, a thick tube is inserted into the anal canal until it stops and fixed it. The radiograph is taken during fluoroscopy in the lateral position of the newborn, when compression of the abdomen between the palms of the doctor causes the opening of the anal canal **(Figure 6)** [24].

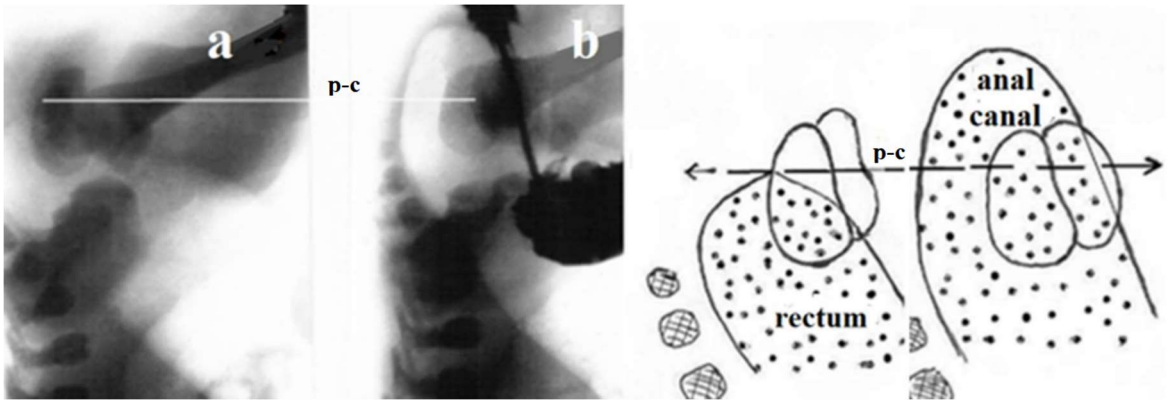

Figure 6. X-ray examination of a newborn with ARM without a visible fistula. **(a).** At rest, the gas was in the rectum at the level of the pubococcygeal line (p-c). (b). The anal canal opened, and gas approached the skin of the perineum when the rectal pressure increased to a certain level.

Surgery. Once the diagnosis is made, a sigmoid divided colostomy is performed. The final correction should be postponed for several months. During this period, regular cleansing of the distal intestine is necessary to prevent the accumulation of large volumes of feces and the development of a megarectum. Since the septum is in the anal canal at about 1 cm from the anal verge, it is accessible for excision through the anal approach. Insertion of a Hegar dilator or endoscope into the distal bowel, as shown by Stenström et al [6], allows the septum to be brought closer to the anus and facilitates its excision. There is no point in suturing the mucous membrane, especially since this can contribute to the development of stenosis. We recommend inserting a tracheostomy tube with a diameter of 1.0-1.3 cm into the rectum. Its balloon, inflated in the rectum, will hold the tube in place for a week. Emptying the rectum occurs through the airway tube. After 7-10 days, the diastasis between the ends of the mucosa will disappear because of the reparative process. Complete preservation of all structures of the anal canal will ensure its normal functioning.

The constant magnetic technique (magnamosis) is theoretically justified [25]. All pull-through operations, especially from the posterior sagittal approach, destroy the anal canal to varying degrees and inevitably lead to constipation and fecal incontinence.

The RA/RS hypothesis of embryonic development.

With the exception of the true cloaca, which is formed as a result of the absence of division of the cloaca into the urogenital sinus and anorectum, all other ARM develop in a later period as a result of disruption of the formation of the ectodermal part of the anal canal, as evidenced

by the absence of the anus. “Normally, in the post cloacal period, the endodermal internal anal sphincter migrates in the craniocaudal direction to meet with the ectodermal portion” [26]. It reaches the subcutaneous tissue above the anal dimple, but without meeting the ectodermal rudiment on its way, it forms fistulous connections (anal stenosis) or, moving forward and upward, forms an ectopic anus in the form of a narrow fibrous ring (ectopia on the perineum, vestibule, vagina, urethra).

As shown by x-ray analysis of 14 published studies, the septum causing intestinal obstruction is located below the pubococcygeal line and is always approximately 1 cm from the anal fossa. The high frequency (21%) of other ARMs suggests a common cause. We hypothesize that the septum arises because of disruption of the connection between the endo- and ectodermal primordia of the anal canal. This is evidenced by the work of Kromer and Korzeniowska-Kromer [27].

Sharma and Gupta [19] describe an assumption, i.e., a hypothesis that is mistakenly called a theory. Meanwhile, theories are considered ideas that do not have contradictions. If at least one reliable fact contradicts the hypothesis, it should be rejected as incorrect. The hypothesis of the origin of RA/RS by Sharma and Gupta is not correct due to the following contradictions:

1). The septum is not at the level of the pubococcygeal line, but approximately in the middle of the anal canal.

2) The rectum of the fetus and newborn has no bends, as Sharma and Gupta write. The bends occur during ontogenesis. Therefore, valves are not relevant to the development of RA/RS.

3). The RA/RS classification proposed by Sharma and Gupta was based on an analysis of published radiographs. (A). The authors did not consider that in most cases the upper part of the anal canal was in a closed state. (B). They included 3 cases in the analysis that were misdiagnosed as RA/RS. (C). The diagrams drawn from radiographs do not correspond to the internal lumen of the anal canal. As a result of these errors, a classification arose, which in the 14 observations described was not confirmed by the authors of the articles, except for 3 cases where a membrane was described. Our study convincingly proved that all patients had a diaphragm. Thus, neither the hypothesis nor the classification of these authors can be used in scientific research. This classification, which assumes a long segment lesion, leads pediatric surgeons to conclude that it is necessary to resect a portion of the bowel using a posterior sagittal approach or endorectal traction, which damages the anal canal and leads to chronic

constipation and fecal incontinence. Unfortunately, case reports with the use of these operations describe only constipation. But long-term results have not been described and objective assessments of anorectal function after these operations have not been used.

Conclusion The study showed that PA/PC is not located in the rectum, but in the anal canal approximately 1 cm from the anus and is represented by the septum. This makes it possible to excise the septum through the anal approach without damaging the anal canal. Methods of diagnosis and treatment are described. A hypothesis has been proposed that PA/PC arises because of disruption of the fusion of the endodermal and ectodermal primordia of the anal canal.

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