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To The Pathological Anatomy of Atresia and Stenosis of the Rectum. Review

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ABSTRACT

Introduction: The traditional concept of rectal atresia and rectal stenosis (RA/RS) assumes that the obstruction is in the rectum proximal to the dentate line, with preservation of a normal anal canal. Objective. To study the anatomy and physiology of RA/RS.

Material and Methods: Twenty-eight open-access articles describing RA/RS that included radiographic data were found in PubMed. Radiographs were evaluated using the radiometric method described in previous articles. The diagnosis of RA/RS was confirmed in 16 of 28 cases. In nine cases, RA/RS was excluded, including five patients with retrorectal space-occupying lesion, where the obstruction was caused by rectal torsion, and in seven cases, bowel obstruction was of a different nature or was absent altogether.

Results: In all 16 of 28 cases with RA/RS, the obstruction was localized not in the rectum, but in the anal canal. In eight of these, the membrane was located in the middle of the anal canal, likely at the level of the dentate line. In six cases, the exact location of the membrane in the anal canal could not be determined. In 2 cases of late-diagnosed RS, it was demonstrated that with a relatively large membrane opening, patients can suffer from organic constipation for many years but be mistakenly treated as suffering from functional constipation. Diagnostic methods are proposed to enable a timely and accurate diagnosis. Treatment methods preserving the anal canal are described to prevent the development of chronic constipation and fecal incontinence.

Conclusion: The proposed hypothesis, which challenges traditional classification, has direct surgical implications. If the obstruction is in the anal canal, transanal membrane excision may preserve the anorectal structures and avoid the functional consequences associated with more invasive procedures. Although the conclusions are limited by a retrospective analysis and a literature-based approach, according to mathematical analysis, the consistency of the results justifies a revision of the current paradigm and supports further prospective studies.

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INTRODUCTION

Rectal atresia/stenosis (RA/RS) is a rare disorder. Hamrick et al. [1] believe that these patients are born with a normal anal canal but have a stricture or complete atresia located a few centimeters proximal to the dentate line. The authors used a posterior sagittal approach with a recto-rectal anastomosis. It is clear from the description that they mistakenly referred to the mucocutaneous junction of the anus as the dentate line. All patients over 3 years of age demonstrated the ability to voluntarily empty their bowels [1]. This is the common term for a successful outcome after posterior sagittal anorectoplasty (PSARP), which results in varying combinations of fecal incontinence and chronic constipation. Because the posterior sagittal approach destroys the anal canal.

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 [2]. Saenz et al., [3] on behalf of the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC), believe that "Congenital rectal stenosis can be defined as a well-developed, normally positioned anus within an intact sphincter complex but with a pathological narrowing located proximally to the dentate line" [3]. There is no evidence for this definition in the article by these authors, nor in the references they cite. Moreover, congenital rectal stenosis has no clear localization, since two-thirds of the anal canal are located above the dentate line, and the true length of the anal canal in newborns is approximately 1.7 cm [4,5]. If RA/RS is located up to 1 cm above the dentate line, then it is in the anal canal, and if several centimeters, then in the rectum. In none of the articles devoted to RA/RS did I find studies that precisely defined the pathological anatomy of this type of ARMs. All authors cite articles that made assertions without evidence.

The purpose of this article is to determine the pathological anatomy of RA/RS.

MATERIALS AND METHODS

Twenty-eight open-access articles describing RA/RS that included radiographic data were found in PubMed. This paper presents a mathematical analysis of 16 cases in which the clinical and radiographic findings were consistent with RA/RS. In nine patients, the diagnosis of RA/RS was excluded, including five patients with retrorectal space-occupying lesion, where the obstruction was caused by rectal torsion. In seven cases, bowel obstruction was due to various diseases or was absent.

Method of X-ray Analysis of Radiographs

To obtain true dimensions, we use either a contrast marker of known size located near the anal dimple or previously calculated height of L-1 in children of different ages (Table 1) [5,6]. In the absence of these markers, the true length of the anal canal was used to calculate the true dimensions.

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 anus. The standards for the anal canal length in children of different ages were calculated earlier (Table 2) [5,6]. Megacolon is diagnosed when the width of the rectum exceeds the maximum normal values.

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 [6].

The true length of the anal canal, like any dimension on a radiograph, can be calculated using the formula: s = S x m/M, where: s - the true length of the anal canal (cm), S- its length measured on the radiograph, m - the true height of L-1 from table 1, M - the height of L-1 measured on the radiograph.

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. [7]. **(Figure 1).** In a newborn girl, "a digital rectal examination showed a blind ending of the anal canal 2 cm above the dentate line".

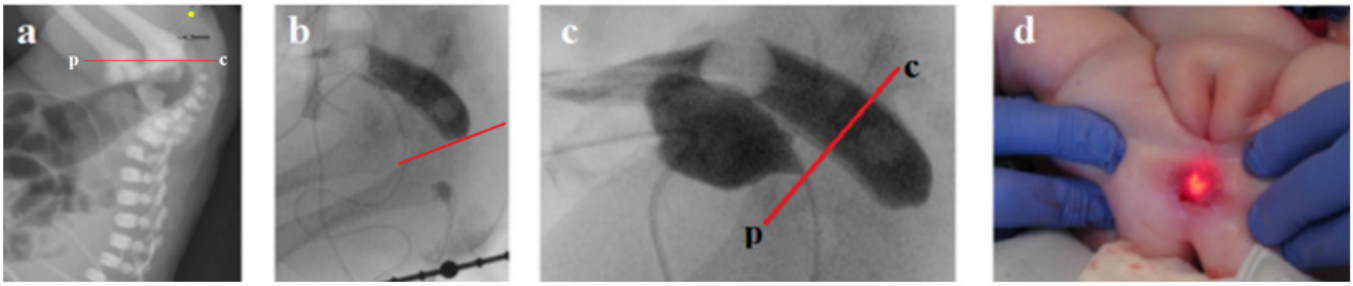


Figure 1. A digital rectal examination

(a) The authors wrote that the distance from the black arrow, which shows the end of the rectal probe, and the gas in the intestine is 2 cm. **(b)** The authors believe that "a distance of 2 cm between the rectal endings". **(c)** During a combination of the high pressure colostogram with contrast in the urinary systems, the upper part of the anal canal opens distally 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" [7].

An analysis of this study suggests that the authors' measurement instrument was not the most accurate. They determined through digital examination that the blind end was 2 cm above the dentate line. Since the anal canal length in newborns is approximately 1.7 cm, with the dentate line located approximately 0.7 cm from the anal verge, the blind end should be located 2.7 cm from the anal verge, i.e., in the rectum. However, on radiographs (a, b), the visible intestine is located at the level of the pubococcygeal line (red). The large gap between it and the end of the anal catheter (arrow) is explained by the fact that the upper part of the anal canal is in a contracted state. The yellow circle indicates the approximate location of the anus. Colostogram was made with high rectal pressure, which caused the anal canal to open above the septum. Light from the endoscope inserted through the colostomy was seen 1 cm up in the anal canal" [7]. Since the length of the anal canal in a 4-month-old child is approximately 2 cm, then with a wide opening of the upper and lower parts of the anal canal, the narrow septum was located somewhere in the middle of the anal canal, i.e., about 1 cm from the anal verge. This calculation is approximate, since the authors did not use a marker near the anus and did not use other instruments to measure distances. Radiometric analysis of the study indicates that the obstruction (atresia) was not in the rectum, but approximately in the middle of the anal canal at 0.7 cm in the neonatal period and about 1 cm at the age of 4 months. The presence of a membrane in the anal canal was proven during surgery. A video endoscope through the sigmoidostomy down to the blind end of the rectum and telescoped through the distance of 2 cm and then almost

protruded through the anus. 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.

Atresia of the anal canal in the form of a septum was found in 8 cases [7-14]. 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 (#)	7	8	9	10	11	12	13	14 (1st)
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

AC – Anal Canal

In 2 of 8 cases [10,14] 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 were not considered). In addition, only frontal radiographs were more often taken, which make it impossible to accurately determine the size of the rectum and anal canal. 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 [8,12].

In 6 cases, the rectum was located at the level of the p-c -line, as is normal [2,14-19]. 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. When analyzing 14 observations, not a single case was identified where intestinal obstruction could be localized in the rectum. We found only thin-walled membranes. The thickness of the membrane depends on the width of the anal canal. With an enlarged anal canal and a stretched membrane, as in Figure 1, it looks thin. When the anal canal is closed, as seen in the description of Chowdhury et al. [9], it appears thick. Other models described by Sharma and Gupta [20] were not found. In 3 (21%) of 14 studies, anal canal membrane was combined with other anorectal malformations [8,10,19].

Late Diagnosis of Rectal (Anal) Stenosis

A 78-year-old woman with a history of severe constipation dating back to early school age was examined. All her life she was helped only by cleansing enemas and digital removal of feces. Defection was always painless. With age, problems with defecation increased so much that the patient began to have suicidal thoughts. Upon examination, a large belly is noticeable. A digital rectal examination performed because of prolonged defecation retention revealed a narrow, hard, thin-walled ring in the anal canal a few centimeters from the anus. The hole in the ring made it possible to insert an index finger about 2 cm wide into the rectum. A large, very dense fecal stone was felt above the membrane, which, during manual examination, was broken into fragments and removed. In our case, the perineum in front and behind the anus was smeared with barium paste to determine the location of the anus as the point of intersection of the enema tip with the contrast line on the perineum. 300 ml of barium suspension was injected into the rectum (Figure 2a). After removing the enema tip, a No. 20 Foley catheter was inserted into the rectum without any effort. The balloon was inflating to a diameter of ≈ 2.5 cm and pulled into the anal canal as far as it would go. The balloon stopped over the constriction at 1.9 cm from the anus (Figure 2b).

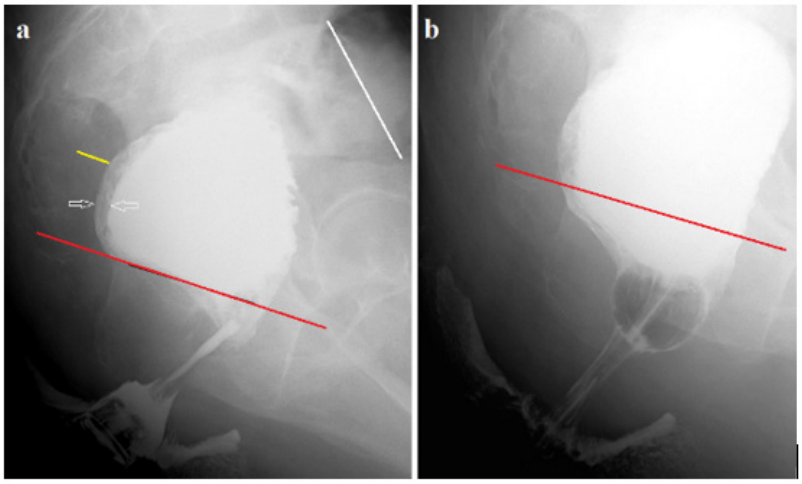


Figure 2. The study was carried out after colon cleansing.

(a) A lateral radiograph of the anorectum was taken after administration of 300 ml of barium. It is known that the true length of the anal canal between the anus and the pubococcygeal line (red) in adults is ≈ 4 cm. The rectum is only partially filled, as the retrorectal space (yellow line) is expanded and barium is in the folds of the mucosa between the white arrows. The presence of barium below the p-c line indicates insufficiency of the puborectalis muscle (descending perineum syndrome). The sigmoid colon (white line) is significantly dilated to 4 cm. **(b)** The Foley catheter balloon inflated in the rectum to a diameter of ≈ 2.5 cm, became lodged in the anal canal above the stenosis, 1.9 cm from the anus.

The delay in diagnosis of anal stenosis in both this case and the case described by Chowdhury et al. [9] is due to the relatively wide opening in the membrane. The walls of the opening are rigid and do not change with age. In childhood, the opening's width can allow for normal bowel movements. However, with age, the width of the rectum increases, and feces form within it in proportion to its width, inevitably leading to severe constipation.

DISCUSSION

Based on a mathematical analysis, the cause of the pathology was found in the anal canal in each of the 15 cases classified as atresia or stenosis according to the Krickenbeck classification. In 7 of the 14 newborns, the site of atresia/stenosis was more precisely localized. It was located approximately midway between the anal verge and the pubococcygeal line, corresponding to the location of the dentate line. Six of these infants had atresia, and two had stenosis. In six cases, convincing evidence was obtained that the obstruction was located below the pubococcygeal line, i.e., in the anal canal. However, a precise localization was not possible. In two patients with late diagnosis of stenosis, the perforated membrane was in the projection of the dentate line. The late diagnosis was because the opening in the membrane was relatively wide and did not cause clinical obstruction in childhood.

About the Anatomy and Embryology of the so-called RA/RS

Although in 6 patients, due to poor radiographic examination, it was not possible to accurately localize the obstruction in the anal canal, it seems very likely that they had the same location as 8 patients, in whom the location of the obstruction was near the dentate line area, presumably 1 cm from the anus. All data indicate the presence of a membrane, which in всех 4 cases had a central opening with rigid walls (stenosis). The location of the membrane corresponds to the junction of the endodermal and ectodermal primordia of the IAS and coincides with the location of the anal membrane in the embryonic period. According to Nobles (1984), normally the anal membrane ruptures during embryos with a length of 13.5-135 cm [21]. I propose the hypothesis that the absence of the anal membrane rupture during the embryonal period leads to anal canal atresia, and partial membrane resorption to the anal canal stenosis. 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" or "anal canal stenosis (ACA/ACS). This is of great practical importance, as the presence of a membrane in the rectum requires complex surgical interventions that damage the anal canal to varying degrees (PSARP, anoplastica), whereas the anal membrane can be removed through an anal approach without damaging

the anal canal, as shown in two articles [7,9].

In the present study, the number of atresia was almost 4 times higher than stenosis. There is reason to believe that a significant number of patients with congenital anal stenosis, in whom the orifice in the membrane is wide enough, can be treated under the mask of functional constipation (FC). For example, Clayden and Lawson, during anal dilatation, which was performed under general anesthesia by inserting up to 4 fingers of the operator's hand, in 4 of 79 patients with FC they were unable to insert more than 2 fingers due to the presence of a rigid ring that was 1 cm from the anal verge [22]. Duhamel, with histological examination of the IAS found in some patient fibrous changes in the anal canal, which were like congenital anal stenosis [23]. It follows from this that when examining patients with megacolon, one must consider the possibility of relatively wide anal stenoses, which cannot be determined by bougienage. Thus, anal distension, which is effective in treating FC, may also be useful for diagnosing secondary megacolon because of congenital stenosis.

To Diagnose and Treat

Congenital ACA/ACS, it's important to understand the function of the normal anal canal, which is present in these patients [24]. With low rectal pressure, it remains permanently contracted. For 24-34 hours of life, it does not allow gas or contrast to pass through and therefore is not visible on radiographs. It can be measured as the distance between the rectum and a marker near the anal dimple (Figure 3a). To visualize the anal canal, higher rectal pressure must be created. When this pressure reaches the threshold for the defecation reflex, the levator plates will open it up to the width of the rectum, allowing gas to enter the anal canal. To achieve the threshold pressure for defecation, the examination should be performed no earlier than 30 hours after birth, allowing swallowed air and meconium to fill the rectum. During fluoroscopy, the abdomen should be compressed between the doctor's palms in a lateral position. Relaxation of the anal canal is manifested by the gas approaches the anus (Figure 3b). This can only be done with atresia, because with stenosis, gas quickly escapes and it is impossible to create a threshold pressure for stable opening of the anal canal.

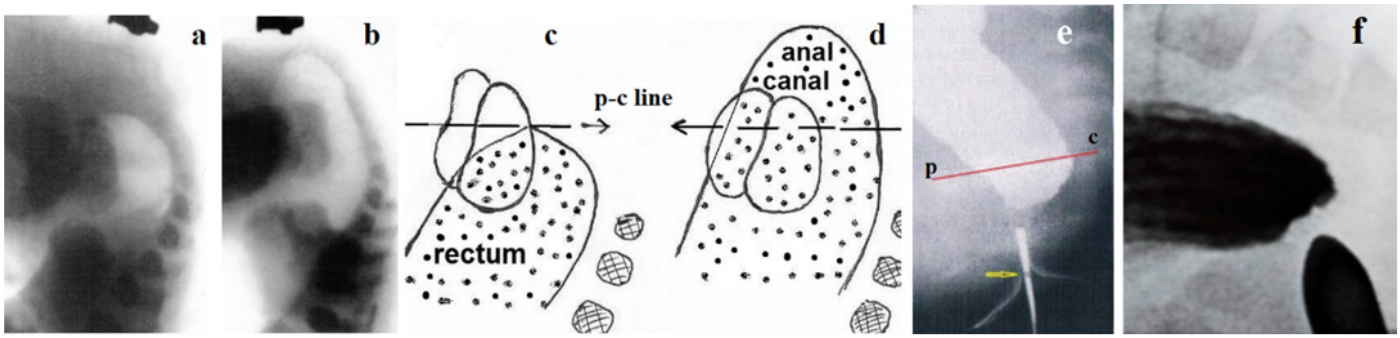


Figure 3. X-ray examination of a newborn with ARM without a visible fistula and its diagram.

(a) Thirty hours after birth, without abdominal compression. A metal marker is glued to the anal dimple. Gas is in the rectum, since the contracted anal canal does not allow gas to pass through. **(b)** During abdominal compression, when the pressure in the rectum increased to the threshold pressure for defecation, the anal canal opened and the gas approached the marker on the perineum**. (c-d)** Diagrams for figures (a) and (b). p-c line - pubococcygeal line. **(e)** From Kobayashi et al. [8]. The membrane is visible as a gap between the catheter inserted through the anus and the contrast medium injected under pressure from the colostomy. **(f)** From Kurashima et al. [12]. The membrane is shown as a gap without contrast medium between the bougie inserted through the anus and the contrast medium injected under pressure from the colostomy.

The opening of the anal canal above the membrane due to the increase in pressure in the rectum makes it possible to diagnose atresia of the anal canal in newborns and children with a colostomy. This method is also applicable for transanal excision of the membrane, which allows preserving all elements of the anal canal.

CONCLUSION

The radiometric analysis suggests that, in the examined cases, which were rectal atresia and stenosis, the obstruction is localized within the anal canal, most commonly at or near the dentate line, and is morphologically compatible with a membranous structure. This interpretation challenges the traditional classification and has direct surgical implications. If the obstruction is indeed confined to the anal canal, transanal membrane excision may allow preservation of native anorectal structures and avoid the functional consequences associated with more invasive procedures. While the conclusions are limited by the retrospective and literature-based nature of the analysis, the consistency of findings warrants reconsideration of the current paradigm and supports further prospective investigation.

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