8-2024

Can Anorectal Stenosis be Managed With Dilations Alone? A PCPLC Review.

Zoe M. Saenz

Kelly Austin

Jeffrey R. Avansino

Andrea Badillo

Casey M. Calkins

See next page for additional authors

Recommended Citation

Saenz ZM, Austin K, Avansino JR, et al. Can Anorectal Stenosis be Managed With Dilations Alone? A PCPLC Review. J Pediatr Surg. 2024;59(8):1652-1656. doi:10.1016/j.jpedsurg.2024.04.007

Journal of Pediatric Surgery 59 (2024) 1652–1656

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.sciencedirect.com/journal/journal-of-pediatric-surgery

Can Anorectal Stenosis be Managed With Dilations Alone? A PCPLC Review

Zoe M. Saenz [a, b], Kelly Austin [c], Jeffrey R. Avansino [d], Andrea Badillo [e], Casey M. Calkins [f], Megan M. Durham [g], Megan K. Fuller [h], Ankur Rana [i], Ron W. Reeder [j], Rebecca M. Rentea [k], Michael D. Rollins [l], K Elizabeth Speck [m], Richard J. Wood [n], Jamie C. Harris [o], Jamie Anderson [a, b], Maheen Hassan [a, b], Payam Saadai [a, b, *], on behalf of the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC)

[a] Department of Surgery, UC Davis Children's Hospital, University of California Davis, Sacramento, CA, USA
[b] Department of Surgery, Shriners Hospital for Children-Northern California, Sacramento, CA, USA
[c] Department of Surgery, UPMC Children's Hospital of Pittsburgh, University of Pittsburgh, Pittsburgh, PA, USA
[d] Department of Surgery, Seattle Children's Hospital, University of Washington, Seattle, WA, USA
[e] Division of Colorectal and Pelvic Reconstruction, Children's National Hospital, George Washington University, Washington, DC, USA
[f] Department of Surgery, Children's Wisconsin, Medical College of Wisconsin, Milwaukee, WI, USA
[g] Department of Surgery, Emory + Childrens Pediatric Institute Atlanta, GA, USA
[h] Department of Surgery, Boys Town National Research Hospital, University of Nebraska Medical Center, Boys Town, NE, USA
[i] Department of Surgery and Perioperative Care, Dell Children's Medical Center, University of Texas Austin, Austin, TX, USA
[j] Department of Pediatrics, University of Utah, Salt Lake City, UT, USA
[k] Department of Surgery, Children's Mercy Hospital, University of Missouri-Kansas City, Kansas City, MO, USA
[l] Department of Surgery, Primary Children's Hospital, University of Utah, Salt Lake City, UT, USA
[m] Division of Pediatric Surgery, C.S. Mott Children's Hospital, University of Michigan, Ann Arbor, MI, USA
[n] Department of Surgery, Nationwide Children's Hospital, The Ohio State University, Columbus, OH, USA
[o] Department of Surgery, Phoenix Children's Hospital, University of Arizona, Phoenix, AZ, USA

ARTICLE INFO

Article history:
Received 12 April 2024
Accepted 15 April 2024

Keywords:
Congenital anal stenosis
Congenital rectal stenosis
Dilations
Surgery
Anorectal malformation
Presacral mass
Currarino

ABSTRACT

Purpose: Congenital anorectal stenosis is managed by dilations or operative repair. Recent studies now propose use of dilations as the primary treatment modality to potentially defer or eliminate the need for surgical repair. We aim to characterize the management and outcomes of these patients via a multi-institutional review using the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC) registry.
Methods: A retrospective database review was performed using the PCPLC registry. The patients were evaluated for demographics, co-morbidities, diagnostic work-up, surgical intervention, current bowel management, and complications.
Results: 64 patients with anal or rectal stenosis were identified (57 anal, 7 rectal) from a total of 14 hospital centers. 59.6% (anal) and 42.9% (rectal) were male. The median age was 3.2 (anal) and 1.9 years (rectal). 11 patients with anal stenosis also had Currarino Syndrome with 10 of the 11 patients diagnosed with a presacral mass compared to only one rectal stenosis with Currarino Syndrome and a presacral mass. 13 patients (22.8%, anal) and one (14.3%, rectal) underwent surgical correction. Nine patients (8 anal, 1 rectal) underwent PSARP. Other procedures performed were cutback anoplasty and anterior anorectoplasty. The median age at repair was 8.4 months (anal) and 10 days old (rectal). One patient had a wound complication in the anal stenosis group. Bowel management at last visit showed little differences between groups or treatment approach.
Conclusion: The PCPLC registry demonstrated that these patients can often be managed successfully with dilations alone. PSARP is the most common surgical repair chosen for those who undergo surgical repair.
Level of Evidence: III.

Abbreviations: PCPLC, Pediatric Colorectal and Pelvic Learning Consortium; ARM, anorectal malformation; VACTERAL, (Vertebral, Anal, Cardiac, Tracheo-Esophageal, Renal and Limb); DCC, Data Coordinating Center; MRI, magnetic resonance imaging; PSARP, posterior sagittal anorectoplasty.

* Corresponding author. Department of Surgery, Shriners Children's Northern California, 2425 Stockton Boulevard, Sacramento, CA 95817, USA.
E-mail address: psaadai@ucdavis.edu (P. Saadai).

https://doi.org/10.1016/j.jpedsurg.2024.04.007

1. Introduction

Anorectal malformations (ARM) are a rare congenital condition occurring in 1 in 5000 births with a slight male to female predominance [1]. Anorectal stenosis comprises only 1% of anorectal malformations. According to the Krickenbeck classification of anorectal malformations, congenital anal stenosis is a major clinical group, and rectal stenosis is categorized as a rare variant. Congenital anal stenosis is defined as an anus that lies within an intact sphincter muscle complex but is pathologically narrow. The narrowing of the anal canal is usually located at the dentate line and patients often present with a skin-lined, 'funnel anus' and is frequently associated with a sacral anomaly [2,3]. Congenital rectal stenosis can be similarly defined as a well-developed, normally positioned anus within an intact sphincter complex but with a pathological narrowing located proximal to the dentate line [4].

While one third of anorectal stenosis are isolated malformations, the remainder present with associated congenital anomalies. These anomalies affect a wide variety of systems including the cardiovascular, gastrointestinal, and urogenital systems [5]. VACTERL (Vertebral, Anorectal, Cardiac, Tracheo-Esophageal, Renal and Limb) associations are readily identified in many patients with a confirmed ARM. Another important association with anorectal stenosis is Currarino syndrome. Currarino syndrome is characterized by a triad of sacral agenesis, a presacral mass, and anorectal malformation. Interestingly, all three features occur in only 20% of cases [6]. Given that the presacral mass may include neoplastic or malignant components, all patients with anorectal stenosis require adequate presacral imaging to evaluate for the presence of a mass [4,7–9]. The etiology and contributing epigenetic factors, genetics, and pathogenesis of anorectal stenosis continue to be investigated.

Given the rarity of the disease, there is limited literature published on the treatment of congenital anorectal stenoses. Traditionally, definitive surgical repair has been the gold standard with dilations used as adjuncts. Recent studies have now proposed the use of dilation as the primary treatment modality to potentially defer or eliminate the need for surgical repair [4]. In some cases, dilations might be able to treat the stenosis alone, thus making a definitive surgical repair unnecessary. While definitive surgical repair remains are indicated in cases where the stenosis is refractory to dilation, there is little long-term and/or multi-institutional data describing the incidence of dilation-refractory anomalies or comparing long-term outcomes with surgery versus serial dilation.

We performed a multi-institutional retrospective review of patients with anorectal stenosis to characterize management and describe outcomes using the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC) registry.

2. Methods

The PCPLC is a national registry of pediatric colorectal patients in the United States. We queried all cases of anal and rectal stenosis in the PCPLC registry from 14 participating children's hospitals between November 22, 2016 to July 10, 2023. The institutional review boards at each participating site approved participation in the PCPLC prior to submission of data. PCPLC member sites entered de-identified patient data into a centralized database using a secure online data capture system. The Data Coordinating Center (DCC) monitors completeness and validity of the data in real time [10,11]. Patients with a diagnosis of anorectal malformation (ARM) without a specific diagnosis of either anal or rectal stenosis were excluded.

Variables collected included demographics, age at most recent visit, associated diagnoses, diagnostic work-up, surgical intervention, complications after surgery, and bowel management regimen at the most recent visit. The following complications within 30 days of surgical intervention were queried: dehiscence of the perineum, acute renal failure, anastomotic leak, infection, rectal prolapse, and stenosis. Bowel management methods queried included enemas, laxatives, and fiber. When multiple methods of bowel management were used, the most invasive was the method reported. Descriptive statistical analyses were performed for all variables collected.

3. Results

Sixty-four patients with a diagnosis of congenital anal or rectal stenosis were identified of which 57 had anal stenosis and 7 had rectal stenosis. Over half (n = 34, 59.6%) were male in the anal stenosis group and 42.9% (n = 3) were male in the rectal stenosis group. The median age in years at the most recent follow up was 3.2 years (anal stenosis) and 1.9 years (rectal stenosis).

3.1. Currarino Syndrome and other associated anomalies

Eleven (19.3%) patients with anal stenosis were diagnosed with Currarino Syndrome. Investigation of surgical management for correction of either anal or rectal stenosis within this cohort were then analyzed. Though presacral mass resection was not queried in our study, it was assumed that Currarino patients diagnosed with a presacral mass underwent appropriate resection and management. Of these eleven patients, there was a slight predominance that underwent surgical repair for their stenosis (23.1%) compared to dilations alone (18.2%). Ten patients with anal stenosis were identified as having a presacral mass (17.5%). Patients with a presacral mass also demonstrated a continued trend of undergoing surgical correction of their anorectal malformation (30.8%) versus dilations alone (13.6%). Seven anal stenosis patients were confirmed to have sacral dysplasia (12.3%) with 23.1% of those patients managed with surgery. In the rectal stenosis group, only one patient (14.3%) was diagnosed with Currarino Syndrome. Of other associated anomalies, the most common were renal, cardiovascular, neurologic, head/neck, and gastrointestinal abnormalities (Table 1).

3.2. Spinal imaging

Imaging of the spine and spinal cord was variable (Table 2). Of the 57 patients with anal stenosis, 34 patients (59.6%) underwent spinal MRI and 17 patients (29.8%) underwent spinal ultrasound. Nine patients (15.8%) had both spinal US and MRI. Eight (14.0%) patients received a spinal ultrasound only with one patient found to have an anomaly identified in that group. A larger percentage of patients (n = 25, 43.9%) had a spinal MRI only and 11 of those patients were found to have a spinal anomaly. Of the subset of 9 patients who had both spinal US and MRI, one had an anomaly identified on ultrasound only, two patients had anomalies on MRI only, four patients had anomalies seen on both ultrasound and MRI, and two patients had no anomalies identified. In the rectal stenosis group, six of the seven patients (85.7%) underwent a spinal MRI, and four of the six MRIs identified anomalies. Two of the seven patients (28.6%) had a spinal ultrasound performed.

Overall, for the total cohort of both anal and rectal stenosis, nearly all patients who underwent surgery had a spinal MRI performed (84.6%, anal stenosis; 100%, rectal stenosis). Notably, patients who were managed with dilations alone had a lower rate of spinal MRIs: (52.3%, anal stenosis; 83.3%, rectal stenosis). However,

Table 1
Demographics and associated diagnoses for patients with anal and rectal stenosis.

Anal Stenosis	Management Approach		Overall (N = 57)
	Dilations Alone (N = 44)	Surgical (N = 13)	
Age at the most recent visit (years)	2.5 [0.9, 6.2]	4.6 [3.2, 11.2]	3.2 [1.2, 6.8]
Sex			
Male	27 (61.4%)	7 (53.8%)	34 (59.6%)
Female	17 (38.6%)	6 (46.2%)	23 (40.4%)
Race			
White	35 (79.5%)	7 (53.8%)	42 (73.7%)
Unknown	4 (9.1%)	3 (23.1%)	7 (12.3%)
Black or African American	3 (6.8%)	2 (15.4%)	5 (8.8%)
Other	2 (4.5%)	1 (7.7%)	3 (5.3%)
Ethnicity			
Not Hispanic or Latino	35 (79.5%)	9 (69.2%)	44 (77.2%)
Hispanic or Latino	7 (15.9%)	3 (23.1%)	10 (17.5%)
Unknown	2 (4.5%)	1 (7.7%)	3 (5.3%)
Associated diagnoses			
Presacral mass/tumor	6 (13.6%)	4 (30.8%)	10 (17.5%)
Sacral dysplasia	4 (9.1%)	3 (23.1%)	7 (12.3%)
Chromosomal	11 (25.0%)	5 (38.5%)	16 (28.1%)
Currarino syndrome	8 (18.2%)	3 (23.1%)	11 (19.3%)
Renal	9 (20.5%)	5 (38.5%)	14 (24.6%)
Cardiovascular	5 (11.4%)	6 (46.2%)	11 (19.3%)
Neurologic	3 (6.8%)	2 (15.4%)	5 (8.8%)
Head/neck	2 (4.5%)	3 (23.1%)	5 (8.8%)
Gastrointestinal	3 (6.8%)	0 (0.0%)	3 (5.3%)
Rectal Stenosis	Dilations Alone (N = 6)	Surgical (N = 1)	Overall (N = 7)
Age at the most recent visit (years)	5.1 [0.9, 14.8]	0.0 [0.0, 0.0]	1.9 [0.6, 14.8]
Sex			
Male	3 (50.0%)	0 (0.0%)	3 (42.9%)
Female	3 (50.0%)	1 (100.0%)	4 (57.1%)
Race			
White	5 (83.3%)	0 (0.0%)	5 (71.4%)
Black or African American	1 (16.7%)	1 (100.0%)	2 (28.6%)
Ethnicity			
Not Hispanic or Latino	6 (100.0%)	1 (100.0%)	7 (100.0%)
Associated diagnoses			
Presacral mass/tumor	1 (16.7%)	0 (0.0%)	1 (14.3%)
Sacral dysplasia	1 (16.7%)	0 (0.0%)	1 (14.3%)
Chromosomal	1 (16.7%)	1 (100.0%)	2 (28.6%)
Currarino syndrome	1 (16.7%)	0 (0.0%)	1 (14.3%)
Renal	1 (16.7%)	0 (0.0%)	1 (14.3%)
Cardiovascular	1 (16.7%)	1 (100.0%)	2 (28.6%)
Neurologic	2 (33.3%)	0 (0.0%)	2 (28.6%)
Head/neck	1 (16.7%)	1 (100.0%)	2 (28.6%)
Gastrointestinal	1 (16.7%)	1 (100.0%)	2 (28.6%)

there appeared to be no difference in the incidence of abnormal findings on spinal MRI in the dilation cohort versus the surgery cohort, although no formal statistics were performed due to inadequate power (Table 2).

3.3. Surgery

Surgical intervention for correction of either anal or rectal stenosis was performed in 13 (22.8%) patients with anal stenosis and one (14.3%) patient with rectal stenosis. The three procedures performed in the cohort included the anterior anorectoplasty, cutback anoplasty, and the posterior sagittal anorectoplasty (PSARP). PSARP was the most common surgery, performed in 8 (61.5%) patients with anal stenosis and one with rectal stenosis patient (Table 3). The median age of repair was 8.4 months in the anal stenosis group, whereas the rectal stenosis patient was repaired much earlier at only 10 days old. One patient (7.7%) in the anal stenosis group was noted to have dehiscence of the perineum. The other 30-day complications queried included acute renal failure, anastomotic leak, infection, rectal prolapse, and stenosis. None of these complications were seen in either the anal or rectal stenosis group.

3.4. Bowel management

Overall, bowel management requirements were similar in the anal and rectal stenosis cohorts when comparing those who received dilations alone or surgical intervention. Bowel management regimens were divided into laxatives or enemas. Laxatives were the most utilized for management in both the anal stenosis and rectal stenosis group at 45.6% and 42.9%, respectively, regardless of if these patients had surgery or dilations alone (Table 4).

4. Discussion

In this multi-institutional retrospective review, most patients with anal stenosis (77.2%) and rectal stenosis (85.7%) were successfully treated with dilations alone without requiring surgical intervention during the follow up period. This finding is consistent with the evolution in the management of ARMs, whereby not all patients require more invasive interventions such as colostomies or extensive surgical repair [10].

There is wide variability of anal and rectal stenosis phenotypes, and these may respond differently to dilations. In our experience, some stenoses may be more narrow or fibrotic than others. For example, those with more severe stenosis, more than 50% narrower than would be expected for patient's age when sized with standard dilators, may be less likely to respond to dilations than those that are only slightly smaller than normal. In addition, very fibrotic stenoses that are not pliable are unlikely to respond to dilations, and continuing dilations despite lack of improvement in anorectal canal size can not only delay definitive care but also lead to significant pain to the patient and be challenging for the caregiver.

Table 2
Diagnostic workup of patients with anal and rectal stenosis.

Anal Stenosis	Management Approach		Overall (N = 57)
	Dilations Alone (N = 44)	Surgical (N = 13)	
Renal ultrasound	25 (56.8%)	8 (61.5%)	33 (57.9%)
Sacral x-ray	13 (29.5%)	1 (7.7%)	14 (24.6%)
Spinal ultrasound	14 (31.8%)	3 (23.1%)	17 (29.8%)
Spinal MRI	23 (52.3%)	11 (84.6%)	34 (59.6%)
Abnormal findings on spinal MRI	13 (29.5%)	4 (30.8%)	17 (29.8%)
Rectal Stenosis	Dilations Alone (N = 6)	Surgical (N = 1)	Overall (N = 7)
Renal ultrasound	3 (50.0%)	1 (100.0%)	4 (57.1%)
Spinal ultrasound	1 (16.7%)	1 (100.0%)	2 (28.6%)
Spinal MRI	5 (83.3%)	1 (100.0%)	6 (85.7%)
Abnormal findings on spinal MRI	3 (50.0%)	1 (100.0%)	4 (57.1%)

Table 3
Surgical Intervention of patients with anal and rectal stenosis.

	Anal Stenosis (N = 13)	Rectal Stenosis (N = 1)
Anterior anorectoplasty	1 (7.7%)	0 (0.0%)
Cutback anorectoplasty	4 (30.8%)	0 (0.0%)
Posterior sagittal anorectoplasty (PSARP)	8 (61.5%)	1 (100.0%)

Table 4
Bowel management at most recent follow up for patients with anal and rectal stenosis.

Anal Stenosis	Management Approach		Overall (N = 57)
	Dilations Alone (N = 44)	Surgical (N = 13)	
Enemas	5 (11.4%)	5 (38.5%)	10 (17.5%)
Laxatives	21 (47.7%)	5 (38.5%)	26 (45.6%)
None of the above	16 (36.4%)	2 (15.4%)	18 (31.6%)
Rectal Stenosis	Dilations Alone (N = 6)	Surgical (N = 1)	Overall (N = 7)
Enemas	1 (16.7%)	0 (0.0%)	1 (14.3%)
Laxatives	3 (50.0%)	0 (0.0%)	3 (42.9%)
None of the above	2 (33.3%)	1 (100.0%)	3 (42.9%)

Patients who are likely to respond to dilations tend to have pliable tissue that can be dilated weekly or every other week without significant or disproportionate discomfort to the patient. Once a desired size has been reached, dilations can be slowly weaned as per traditional ARM protocols while monitoring for refractory stenosis. It is worth noting that while there are widely referenced norms for anal canal size based on a child's age, with a newborn full-term infant being able to accommodate a 12 mm dilator for example, these norms have not been validated. As with all ARM patients, those who do respond to dilations must continue to receive long-term follow-up for their ARM and appropriate screening for associated urological, gynecological, and neurosurgical anomalies, amongst others. Monitoring for re-stenosis and follow up for long-term fecal continence is also recommended given the rarity of these disorders and the fact that long-term outcomes are unknown.

If surgical intervention is required, a modified PSARP has been described as one approach for anal or rectal stenosis [9]. For anal stenosis, the patient is placed in a prone position, and a posterior sagittal incision is made from the level of the coccyx down to the level of the anus. The intended goal is to split the anal canal in the posterior midline. Dissection is taken down to the level of the presacral fascia where the rectum is then mobilized on the posterior 180° only. It is not necessary to dissect the anterior rectal wall as this dissection allows for posterior advancement of the rectum. Next, the anal canal is opened in the posterior midline, specifically through the stenotic segment into the normal caliber lumen. This should allow for the passage of a size 15 Hegar dilator. The “open book” dentate line is now ready for anastomosis, which is where this modified approach differs from the standard PSARP. The anterior 180° is the original anal canal and the posterior aspect of rectal mucosa is sutured directly to skin, which effectively doubles the size of the lumen.

The operative approach for rectal stenosis is similar in the prone position, posterior sagittal incision, dissection to the white fascia of the posterior rectum. Before further mobilizing the posterior wall of the rectum, care should be taken to place silk sutures on the posterior aspect of the distal anal canal and ensure preservation of the dentate line. Again, as in the approach for anal stenosis there should be no dissection of the anterior wall. The posterior distal anal canal is opened through the level of the stenosis. The posterior rectal wall after appropriate mobilization is then brought down and anastomosed to the anoderm along the posterior 180°. The posterior rectal wall is sutured to the muscle complex and posterior sagittal incision closed in the standard fashion [12].

Patients with anorectal stenosis and a presacral mass are worth special consideration. It is crucial to emphasize that all patients with anal or rectal stenosis must obtain spinal imaging to evaluate for the presence of a presacral mass, even if that patient's stenosis is responsive to dilations alone. If a patient does have a presacral mass and their stenosis is not responsive to dilations alone, excision of the mass can be timed with surgical repair of the anorectal stenosis in most patients [9]. The type of spinal imaging varied within the groups. We recommend that all anorectal stenosis patients undergo standard screening for all anomalies associated with ARM's per ARM protocols. Although there is no consensus on what type of spinal imaging should be performed, a presacral mass must be definitively ruled out, and an MRI spine and/or pelvis is highly recommended when possible. The sensitivity of newborn spinal ultrasound for presacral mass screening is unknown and may be adequate screening to rule out a mass, but there is little existing literature that directly compares the two imaging modalities.

Evaluating for other comorbidities is also critical. As with all ARM patients, appropriate screening for associated malformation including VACTERL (Vertebral, Anorectal, Cardiac, Tracheo-Esophageal, Renal and Limb) anomalies amongst others should be assessed using standard ARM protocols. Our findings in both anal and rectal stenosis groups regarding renal imaging via renal ultrasound seen in Table 2 was 57.9% and 57.1% respectively. These numbers were surprisingly low, and it is unclear whether this was due to limitations of the database versus a “true” incomplete work-up. Although cardiac echocardiography is often performed in the newborn period to assess for cardiac anomalies, this was not investigated in our study to compare if there were similar appearing low incidence of work-up or diagnosis. Therefore, we felt it of great important to highlight that proper screening, especially for anal stenosis patients, that these VACTERL associations are not uncommon and should not be missed.

Our study had several limitations, due both to its retrospective nature and the limits of the using a large database. The registry is unable to select for more granular data and details regarding patient management. For example, the database was able to identify the presence or absence of abnormality on spinal MRI, but not the specific diagnosis. Specific details such as initial and final dilation size and duration of intervention before considered a failure of management were not available. In addition, due to the relatively low number of patients with these disorders, especially patients diagnosed with rectal stenosis, we did not have the statistical power to compare the two groups of patients and were only able to apply descriptive statistics.

In summary, our study of anorectal stenoses using the multi-institutional PCPLC registry demonstrates that patients with anal and rectal stenosis can often be managed successfully with dilations alone. When surgery is required, PSARP remains the standard and most commonly performed repair chosen for these patients. Providers can consider dilations as a potential primary intervention rather than an adjunct in patients with anorectal stenosis: surgical intervention can be pursued in those who are refractory to dilation. Further investigation is warranted to assess factors that lead to successful clinical outcomes in patients managed with dilations alone and risk factors leading to surgery. Given the rarity of anorectal stenoses, multi-center collaboration is needed to examine patient level factors and guide future management.

Funding

This study was approved by the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC) for use of the national PCPLC database for data acquisition. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

Many authors are members of this consortium but have no financial or further competing interests in the outcomes of the study. All other authors have nothing to declare.

References

[1] Wood RJ, Levitt MA. Anorectal malformations. Clin Colon Rectal Surg 2018;31:61–70. https://doi.org/10.1055/s-0037-1609020.

[2] Amerstorfer EE, Schmiedeke E, Samuk I, Sloots CEJ, van Rooij IALM, Jenetzky E, et al. Clinical differentiation between a normal anus, anterior anus, congenital anal stenosis, and perineal fistula: definitions and consequences—the ARM-net consortium consensus. Children 2022;9. https://doi.org/10.3390/children9060831.

[3] King SK, Krois W, Lacher M, Saadai P, Armon Y, Midrio P. Optimal management of the newborn with an anorectal malformation and evaluation of their continence potential. Semin Pediatr Surg 2020;29. https://doi.org/10.1016/j.sempedsurg.2020.150996.

[4] Gangopadhyay AN, Sinha CK, Sahoo SP. Combined rectal atresia and rectal stenosis. Pediatr Surg Int 1997;12(8):605–6. https://doi.org/10.1007/BF0131910. PMID: 9354736.

[5] Wang C, Li L, Cheng W. Anorectal malformation: the etiological factors. Pediatr Surg Int 2015;31:795–804. https://doi.org/10.1007/s00383-015-3685-0.

[6] Coleman John. Currarino syndrome. Orphanet encyclopedia. April 2023. http://www.orpha.net/consor/cgi-bin/OC_Exp.php?lng=en&Expert=1552.

[7] Koskay G, Opperman P, Mezzacappa FM, Menousek J, Fuller MK, Fornoff L, et al. Presacral mature cystic teratoma associated with Currarino syndrome in an adolescent with androgen insensitivity: illustrative case. J Neurosurg: Case Lessons 2022;4. https://doi.org/10.3171/CASE22351.

[8] Dworschak GC, Reutter HM, Ludwig M. Currarino syndrome: a comprehensive genetic review of a rare congenital disorder. Orphanet J Rare Dis 2021;16. https://doi.org/10.1186/s13023-021-01799-0.

[9] Lane VA, Wood RJ, Reck C, Skerritt C, Levitt MA. Rectal atresia and anal stenosis: the difference in the operative technique for these two distinct congenital anorectal malformations. Tech Coloproctol 2016;20:249–54. https://doi.org/10.1007/s10151-016-1435-5.

[10] Short SS, Reeder RW, Lewis KE, Dickie B, Grabowski J, Sepuha T, et al. The presence of a neurodiverse disorder is associated with increased use of antegrade enema therapy in children with severe constipation: a study from the Pediatric Colorectal and Pelvic Learning Consortium (PCPLC). J Pediatr Surg 2022;57:1676–80. https://doi.org/10.1016/j.jpedsurg.2022.04.008.

[11] Reeder RW, Wood RJ, Avansino JR, Levitt MA, Durham MM, Sutcliffe J, et al. The pediatric colorectal and pelvic learning consortium (PCPLC): rationale, infrastructure, and initial steps. Tech Coloproctol 2018 May;22(5):395–9. https://doi.org/10.1007/s10151-018-1782-5. Epub 2018 May 4. PMID: 29725784.

[12] Badillo A, Tiusaba L, Jacobs SE, Al-Shamaileh T, Feng C, Russell TL, Bokova E, Sandler A, Levitt MA. Sparing the perineal body: a modification of the posterior sagittal anorectoplasty for anorectal malformations with rectovestibular fistulae. Eur J Pediatr Surg 2023 Dec;33(6):463–8. https://doi.org/10.1055/a-1976-3611. Epub 2022 Nov 10. PMID: 36356590.