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Pediatric Colorectal and Pelvic Learning Consortium

THE PEDIATRIC COLORECTAL AND PELVIC RECONSTRUCTION CONFERENCE

Surgical, Clinical, and Scientific Programs

November 13-15, 2024
Emory Conference Center Hotel
Atlanta, Georgia

ABSTRACT BOOK

PHENOTYPIC VARIATION OF HIRSCHSPRUNG DISEASE BY GENDER WITHIN A FAMILY WITH A PATERNAL RET MUTATION

Papillon SC, Gilliam E, Maselli KM, Speck KE

Hirschsprung disease (HD) is a common congenital cause of intestinal obstruction in the neonatal period. While the majority of cases are sporadic affecting the rectosigmoid colon with a male predominance, approximately 5-20% are familial with a higher incidence of long-segment or total-colonic HD and lower male to female ratio. Familial HD includes a diverse number of mutations with variable penetrance with a reported rate of familial occurrence in up to 8% of siblings.

We report a case of the first female born to a family with a paternally inherited RET proto-oncogene mutation affecting the majority of males in multiple generations, including her older brother (Figure). Her father has long-segment HD and her brother has near total-colonic HD. Additionally, one paternal uncle and two of his three sons have total-colonic HD while the other paternal uncle was unaffected. Prenatal genetic testing via amniocentesis identified the same RET mutation involving a 12kb microdeletion of chromosome 10q11.21 (exon 1) noted in her brother. She was born at term via C-section and passed meconium within the first hour of life and had multiple subsequent stools. After a decrease in spontaneous stooling over the weekend, a planned suction rectal biopsy was performed on day of life three. HD was confirmed on histopathology and no transition zone was apparent on contrast enema. Rectal irrigations were initiated and continued every 6 hours until surgery. At 2 months, she underwent laparoscopic leveling biopsies with an endorectal pull-through. Pathology confirmed short-segment disease, with a short transition zone and normal biopsy in the mid rectum. This interesting case underscores the complex disease heritability and phenotypic variation in the setting of an identical inherited mutation.

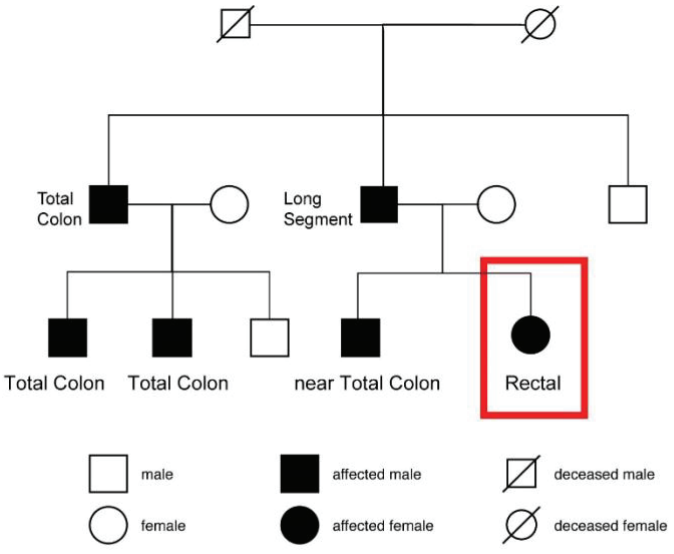


Figure. Familial pedigree of the patient (red rectangle) with annotations regarding disease severity for the affected individuals.

MULTIDISCIPLINARY MANAGEMENT OF A SHORT UROGENITAL SINUS AND PERINEAL FISTULA

Kate McNevin, MD, Angela Hernandez, MD, Jen Ahn, MD, MS, Caitlin Smith, MD

Case:
The patient is an 8-month-old girl who was born at 35 5/7 gestational age with prenatally diagnosed Tetralogy of Fallot. She underwent repair of her congenital cardiac diagnoses with transannular repair of TOF with VSD closure, PDA ligation, central PA augmentation, and PFO left open. It was noted that she had ambiguous genitalia with clitoromegaly and two perineal openings. No pathologic variants on her disorders of sex development panel were identified and she had a normal endocrine evaluation. She was urinating and stooling appropriately and ultimately presented to Reconstructive Pelvic Medicine Clinic at 7 months of age. To clarify her anatomy, she was taken to the operating room with general surgery, urology, and gynecology. Intraoperatively, she was noted to have two perineal openings, separated by 3mm. The posterior opening was sized to a 12 Hegar dilator. The muscle complex contracted mostly posterior to the anal opening. The anterior opening had a common wall with the posterior opening and was found to be a urogenital sinus with a very small opening to the bladder. The common channel measures 1.3cm.

Summary:
This 8-month-old girl has a UG sinus with a perineal fistula but with very little space on the perineum for separation due to location of the pubic symphysis. Additionally, she has a very small (3 Fr) urethra from the UG sinus to the bladder. Currently she has no hydronephrosis. The discussion questions relate to timing of surgical intervention, type of surgical intervention with no space on the perineum for separation as well as longer term management of the urinary system.

TRANS-ANAL VACUUM-ASSISTED THERAPY FOR MANAGEMENT OF LEAK AFTER ILEAL POUCH ANAL ANASTOMOSIS

Megan Read, MD, Steven Scoville, MD, PhD, Benedict Nwomeh, MD, Ihab Halaweish, MD

Case: An 18-year-old female with a history of medically refractory ulcerative colitis presented for laparoscopic completion proctectomy, Ileal J-Pouch Anal Anastomosis (IPAA), and diverting loop ileostomy. She had previously undergone laparoscopic colectomy 6 months prior with an uneventful postoperative course. The patient had a BMI of 28 kg/m^2 (91% percentile). During the procedure, the J pouch was created and confirmed to reach the pelvis followed by completion proctectomy using an eversion technique and a linear stapler for transection of the rectum. The IPAA was completed using a circular stapling device with minimal tension. Due to the patient's body habitus, it was not feasible to obtain adequate reach of the distal ileum for a diverting loop ileostomy. Due to persistent tachycardia on postoperative day 5, a computed tomography scan was obtained which showed extensive abdominal fluid collections. An exploratory laparotomy revealed gross abdominal contamination. Flexible endoscopy showed dehiscence of the posterior half of the IPAA (Fig 1). A diverting loop ileostomy was created with difficulty and sutured flush to the skin, and trans-abdominal drain was placed into the pelvis. A trans-anal vacuum dressing was fashioned (Fig 2) and placed through the anastomosis and into the presacral space. Over the subsequent 22 days, the patient underwent 6 wound vac changes with eventual complete closure of the presacral space and healing of the anastomosis (Fig 3). She underwent successful ileostomy closure 6 weeks later with an uneventful course.

Summary: Anastomotic IPAA leak is associated with significant morbidity including pelvic sepsis, chronic presacral sinus or pouch fistula, pouch failure, and prolonged need for ileostomy. Maximal efforts should be used to salvage IPAA leak and prevent potential complications. The use of trans-anal vacuum assisted therapy presents a useful and safe tool in this setting.

Figure 1: Dehiscence of the posterior half of the IPAA on flexible endoscopy.

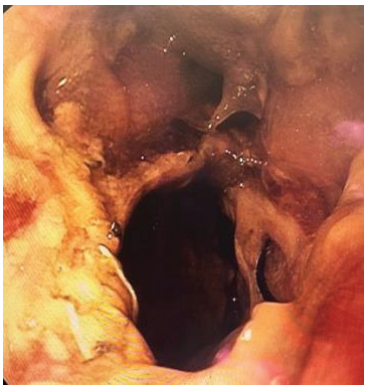


Figure 2: Trans-anal vacuum dressing

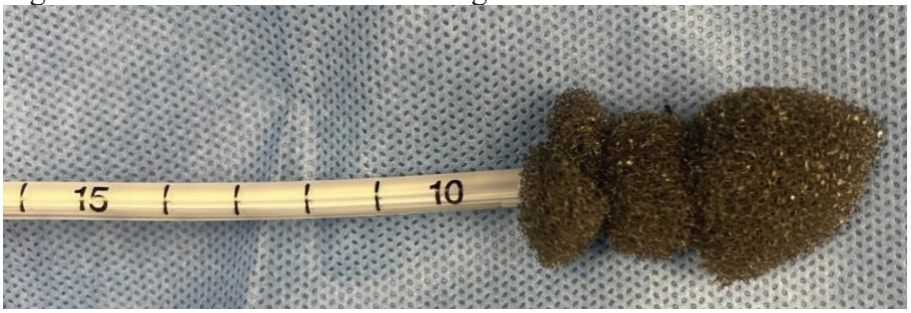
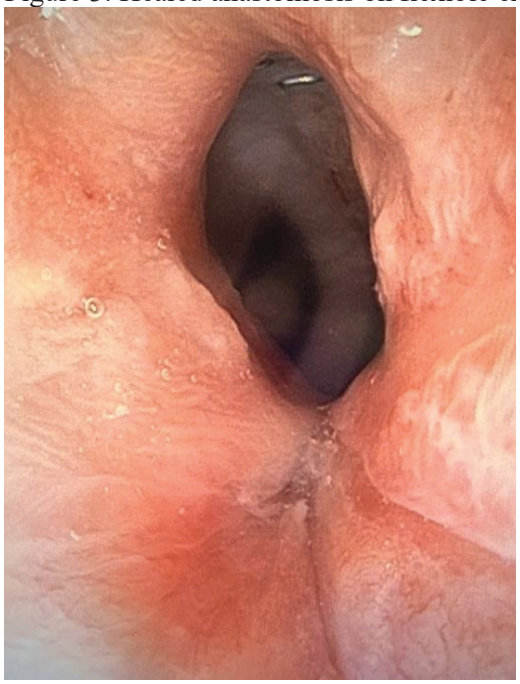


Figure 3: Healed anastomosis on flexible en



COLLIS-TYPE CECOPLASTY FOR GAINING LENGTH FOR MALONE APPENDICOSTOMY

Kristine L. Griffin, MD, Richard J. Wood, MD

Case: Splitting the appendix is an ideal technique with low morbidity for simultaneous Malone appendicostomy and Mitrofanoff appendicovesicostomy procedures. Generally, this can be achieved in most patients if the appendix is at least 7cm in length. We propose a new technique, similar to a Collis gastroplasty, used in 2 patients when the appendiceal length was inadequate to share for bowel and bladder management.
Technique: The distal appendix is harvested for Mitrofanoff. A 10Fr council tip foley catheter is inserted into the remaining appendix. An EndoGIA stapler is fired at the base of the appendix, parallel to the foley catheter, to gain length for the appendix. The resulting staple line is wrapped around the new appendiceal base and secured with silk sutures, creating a valve to prevent stool leakage. The Malone is then matured at the chosen site on the abdominal wall.
Patient 1 is a 5-year-old female with a history of anorectal malformation. She was scheduled for laparoscopic Malone appendicostomy and Mitrofanoff or Monti by urology. Intraoperatively, the appendix was found to be only 2.5 cm in length. The patient was incidentally noted to have a Meckel's diverticulum. A bowel resection of the Meckel's was performed and an adjacent bowel harvest for Monti creation. An EndoGIA stapler was fired parallel to the appendix to gain an additional 3cm of length for the Malone. The cecal wrap was made and the Malone was matured in the right lower quadrant abdominal wall.
Patient 2 is an 18-year-old male with a history of neurogenic bowel and bladder from a spinal cord injury. He underwent robotic-assisted Malone and Mitrofanoff. Intraoperatively, the appendix was found to be 13 cm long. The patient's abdominal wall was thick, so the distal 10cm was used for the Mitrofanoff. A Collis cecoplasty was performed to gain an additional 4.5cm for the Malone to reach the abdominal wall, where it was matured in the right lower quadrant.
Both patients were started on full-volume Malone flushes once tolerating a diet postoperatively.
Summary: The Collis-type cecoplasty is a feasible technique for gaining appendiceal length, especially when sharing a split appendix. It should be considered as an alternative to the Neomalone appendicostomy.

HIGH-CONFLUENCE CLOACA WITH ATRETIC COMMON CHANNEL PRESENTING WITH LOWER URINARY TRACT OBSTRUCTION.

Raymond Yong, MD, Ana Cisneros-Camacho, MD, Olivia Winfrey, MD, Kristy Rialon, MD, Andrea Balthazar, MD, Niccolo Passoni, MD

Case report

The patient is a girl born at 29 weeks. Prenatal MRI was consistent with cloacal malformation with phallic variant (megalourethra), duplicated hemivaginas (hydrometrocolpos), no hydroureteronephrosis, oligohydramnios and pulmonary hypoplasia. Patient required two peritoneal-amniotic shunts. At birth, she was intubated, and underwent exploratory laparoscopy with mucous fistula creation and placement of vaginostomy drain in right hemivagina. Eventually required ultrasound placement of drain in left hemivagina.

Had complicated NICU stay including prolonged intubation, pneumothorax, acute hypoxic respiratory decompensation requiring ECMO, left ischemic cerebral infarct, C-diff infection, and two pseudomonas urosepsis. Has normal spinal and renal anatomy.

Prior to discharge home, we attempted vaginostomy creation, to remove vaginostomy tube and reduce risk of infection. However, due to chronic decompression of the vagina, we were not able to perform vaginostomy and just performed a vesicostomy. All vaginal tubes were removed. The pseudophallus was also incised ventrally, exposing a very tight common channel that would not accommodate a 5 Fr catheter.

In the following months, despite having drainage of urine from the vesicostomy, her abdomen became distended, causing prolapse of vesicostomy and feeding issues. Repeat imaging showed re-accumulation of severe hydrometrocolpos requiring placement of vaginostomy tube.

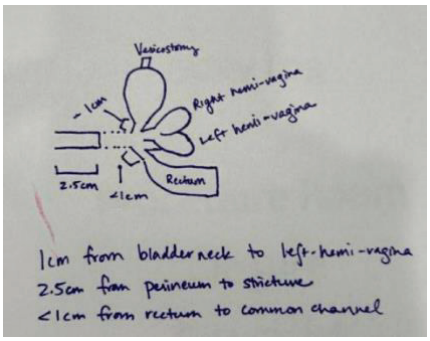
At 9 mo (6 mo corrected) she underwent exam under anesthesia with endoscopy and 3D-cloacogram (Figure). This showed a 5 cm common channel, with a 2.5 cm atretic segment with barely no passage of contrast and a 1 cm urethra. Reconstruction is planned around 1 year of age.

Summary

Girl with complex cloacal anomaly, 5 cm common channel with intrinsic atresia causing lower urinary tract obstruction. Normal spine anatomy but suffered a left ischemic cerebral stroke.

- Given her complex anatomy, how do the experts recommend approaching reconstruction of her genitourinary tract?
- What is the best management of hydrometrocolpos in complex cloacas?

Figure



CLOACAL MALFORMATION AND MULLERIAN AGENESIS: MANAGEMENT OF THE GYNECOLOGIC RECONSTRUCTION FOR THIS RARE SITUATION

Eugenie Lehembre-Shiah, Kirsten Das, Thomas Xu, Inbal Samuk, Christina Ho, Andrea T. Badillo, Marc A. Levitt, Allison C. Mayhew

Case

A 3 year old female with VACTE-GRLS (vertebral, anal, cardiac, tracheoesophageal, gynecologic, renal, limb, spinal) association including cloacal malformation, solitary right kidney, and sacral agenesis underwent a colostomy at birth and then presented to us for cloacal reconstruction. There was no hydrocolpos or hydronephrosis noted in the newborn period. The baby spontaneously voided and had no UTIs. We performed an examination under anesthesia, cystoscopy, and cloacagram which demonstrated a single perineal orifice with a 2.5 cm common channel and a 1.3 cm urethra. There was a rectal fistula to the common channel and no vaginal opening seen on endoscopy or the contrast study. Diagnostic laparoscopy demonstrated no midline Mullerian structures, absent right Mullerian structures, no right ovary, left rudimentary Mullerian structures, and a left ovary at the left inguinal canal without dissensus to the midline. Given the lack of connection of Mullerian structures to the common channel and urologic system with no hydronephrosis or UTIs, the decision was made to proceed with posterior sagittal anorectoplasty only, and to delay surgical reconstruction of the Mullerian system until after menarche.

Summary

In patients born with cloacal malformations, reconstructive surgery has historically included urethral, vaginal, and rectal reconstruction to restore typical anatomy and preserve urinary, gynecologic, and fecal function. However, in rare cases of cloaca and Mullerian agenesis where a connection between the gynecologic tract and the urinary/fecal tract is not demonstrated, the key question is: can gynecologic reconstruction can be delayed? Such a delay allows for assessment of Mullerian structures after pubertal growth, prioritizes pubertal development in the reconstruction, and allows for patient participation and autonomy in reconstructive decision-making. If delay in gynecologic reconstruction is considered, which patients qualify? Are there long-term risks to a delay in gynecologic reconstruction? What is the appropriate timing then for management of the Mullerian system?

ANTERIOR CLOACA WITH BLIND ENDING ANUS, INCOMPETENT BLADDER NECK AND ECTOPIC URETERS.

Raymond Yong, MD, Ana Cisneros-Camacho, MD, Olivia Winfrey, MD, Kristy Rialon, MD, Andrea Balthazar, MD, Niccolo Passoni, MD

Case report

Patient is a baby girl born at 33 weeks of gestation with VACTERL and prenatal diagnosis of clocal malformation. Her gestation was complicated by oligohydramnios and pulmonary hypoplasia, requiring intubation after delivery. On day of life 2 she underwent diverting colostomy as well as vesicostomy creation due to inability to catheterize common channel.

Other congenital malformations include double inlet single left ventricle, pulmonary stenosis, horseshoe kidney with cystic dysplasia of her left moiety secondary to reflux nephropathy and vertebral fusion anomaly without specific abnormality of her spinal canal.

Her exam under anesthesia (Figure) at 15 mo showed two perineal openings: a posterior blind ending orifice (3 cm deep), located in the anal sphincteric complex, and an anterior opening just below the clitoris that led to a 3 cm common channel. She had vaginal duplication and a very short urethra, less than 1 cm in length, with a gaping bladder neck. Her ureteral orifices were not visualized in the bladder and likely ectopic.

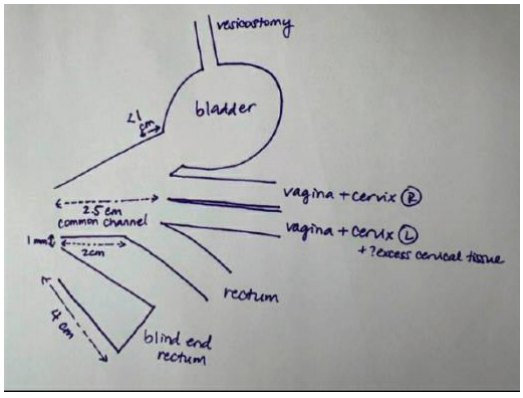
After discussion with family, she was taken to the OR for reconstruction at 19mo. Pediatric surgery performed a PSARP. Her blind ending rectal canal was too dysplastic to be used for reconstruction. Urology performed bladder neck closure with bilateral ureteral reimplant of ectopic ureters (left opened at the bladder neck and right orifice was never identified). Her vesicostomy was maintained. Her vaginal defect (after bladder neck closure) was repaired, and her common channel was mobilized (TUMS) and moved more inferiorly in the perineum.

Summary

Girl with complex cloacal anomaly, anteriorly displaced common channel, blind ending anal pouch and incompetent bladder neck:

- How would you manage her urinary tract at birth and at reconstruction?
- Is the common channel a valid temporary vaginal canal that can be later dilated or augmented?

Figure



CHALLENGES OF A RECTOVESTIBULAR FISTULA WITH BLADDER AND VAGINAL AGENESIS

Emmanuel L. Abebrese, MD, MS[1], Casey M. Calkins, MD[1] John G. Schneider, MD[1]

[1] Department of Surgery, Medical College of Wisconsin, Milwaukee, WI.

Case Background

AM is a former 35 week premature infant girl with a prenatal diagnosis of bladder exstrophy. Pediatric urology was consulted immediately after birth. Initial examination was concerning for a cloacal malformation. Yet, she began passing meconium after ~ 8 hours of life. Pediatric surgery exam demonstrated a rectovestibular fistula. Relevant screening studies demonstrated thoracic vertebral and sacral anomalies, large ASD with aberrant right subclavian artery and bilateral SVC; bladder non-visualization, small bilateral kidneys with left side in pelvis; and absent septum pellucidum. Further evaluation with brain and spinal MRI demonstrated thin bilateral optic nerves suggestive of septo-optic dysplasia. Fistula dilations were initiated and serially increased up to # 8 Hegar. Respiratory support was gradually weaned. Enteral feeds were initiated and steadily advanced. Discharged home on DOL 20.

Outpatient Follow-up

Urology appreciated continuous urine leakage and could not visualize a vaginal opening, and suspected ectopic ureters draining into micro bladder. She continued to do well - defecating via vestibular fistula both spontaneously and with dilations – when seen in surgery clinic. Multidisciplinary cystovaginoscopy and anorectal EUA were recommended and arranged to define anatomy and plan further interventions.

Results

Normal, symmetric labia majora and minora, and clitoris. Rectovestibular fistula – accepts up to #10 Hegar. Symmetric sphincter complex with center 1.2 cm posterior to labial commissure. Urogenital vestibule contains multiple small pits and midline opening with distal membrane with two proximal orifices. Urogenital common channel ~ 4 cm in length. No vaginal opening nor cervix identified.

Summary

This infant girl has a unique rectovestibular fistula associated with bladder and vaginal agenesis. She is growing and defecating well thus far as treated with fistula dilations. Uncertainties in her care include need for fecal diversion, colorectal reconstruction challenges, as well as urologic and gynecologic reconstruction timing.

DISTAL URETHRAL STENOSIS WITH ACQUIRED PROXIMAL RECTOURETHRAL FISTULA AFTER PRIMARY PSARP FOR PERINEAL FISTULA: COLLABORATIVE APPROACH FOR REPAIR

Authors: Briony K. Varda, Inbal Samuk, Andrea T. Badillo, Marc A. Levitt

Case Description

A 4-month-old male was referred after a perineal fistula repair that was complicated by urethral injury. He had been appropriately diverted with vesicostomy and end colostomy with Hartman's pouch. He drains urine via his anus with valsalva. Cystoscopy demonstrated an obliterated distal urethral limb at the bulbar urethra. Up-and-down study showed a gap between the distal bulbar urethra and the upper prostatic urethra, as well as an acquired fistula to the distal rectum (Figure 1). Exam showed funneling around the anus and stricture on digital exam.

Challenges

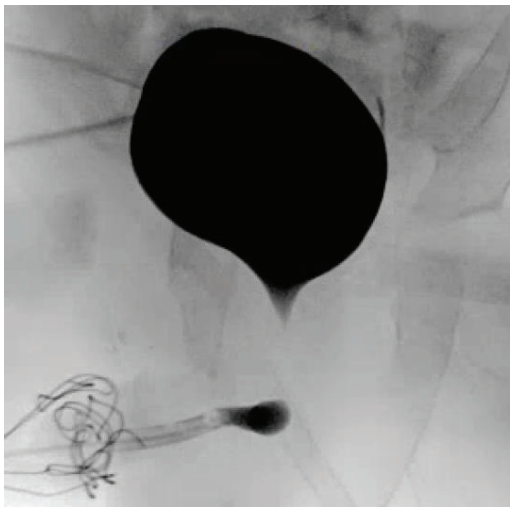
A redo PSARP was performed. The stenosed anus was released circumferentially, the fistula divided, and the distal rectum flipped cephalad exposing the posterior urethra. In so doing, both vas deferens were found dismembered from the urethra, densely embedded in scar tissue on the mobilized rectum. Mobilization of the distal urethra to the corporal crus provided mobility. Cystoscopy of the proximal side showed coaptation of the bladder neck just above the tissue edges. End primary anastomosis was completed in two layers. The repair was covered with an ischiorectal adipose flap for interposition. Redo pull-through of the rectum and anoplasty were completed. The vesicostomy and colostomy retained.

Summary

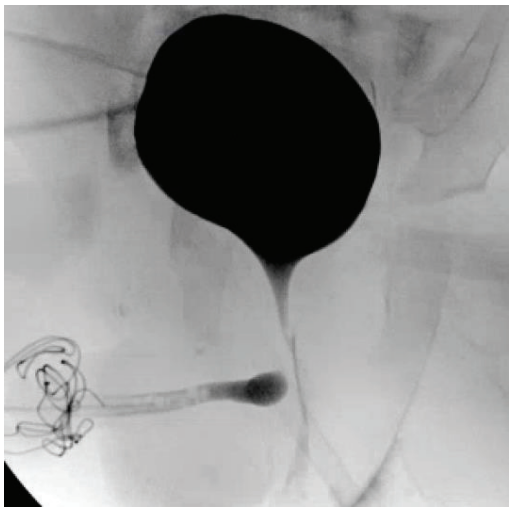
Perineal fistula is considered a 'simple' anorectal malformation, however complications can occur. The value of a collaborative surgical approach between a urologist and colorectal surgeon is underscored by this case. There were multiple urologic considerations: What is the best repair for this urethral injury and potential risks? What are your options if end-primary-anastomosis is not possible? After repair, what is your drainage plan? How do manage the vas deferentia?

Figure 1. Up-and-down study showing obliterated distal urethra and acquired fistula from prostatic urethra to Hartman's pouch after complication during PSARP for perineal fistula.

a) Gap



b) Fistula



ANNUAL RENAL SURVEILLANCE WITH ULTRASOUND IN MILD ANORECTAL MALFORMATIONS MAY NOT BE NECESSARY

Shruthi Srinivas, MD, MPH, Kristina Booth, FNP, Susan Kasubick-Tillman, PA, Molly Huskisson, CPNP-AC, Kristine L Griffin, MD, Elizabeth Thomas, MS, Madeline Su, BS, Richard J Wood, MD, Daniel G DaJusta, MD, Molly E Fuchs, MD

Purpose

Patients with anorectal malformations (ARMs) are at risk of renal anomalies. As a result, we obtain annual renal ultrasounds (RUS) regardless of symptoms or complexity. It is unknown if such aggressive screening is necessary for patients with mild ARM. Thus, we aimed to determine outcomes in patients with mild ARM on this strict protocol.

Methods

We performed a single-institution review of patients managed at a large pediatric colorectal center from 2008 – 2023 with mild ARM (rectoperineal fistula, rectovaginal fistula, rectovestibular fistula, rectal/anal stenosis, and rectal atresia). We evaluated all RUS before and after surgical repair. Only patients with a RUS performed within 2 years of their primary surgical repair were included. Demographics and clinical variables were collected, and descriptive statistics were computed.

Results

There were 333 patients with mild ARMs, of whom 197 had a postoperative RUS within the first two years, obtained at a median of 3.9 months after anoplasty [IQR: 2.4 – 11.0] (**Figure 1**). Most patients (70.1%) had a normal first postoperative RUS, and only four (2.0%) became abnormal. Only one patient (0.5%) was asymptomatic at the time of RUS and had a finding necessitating further intervention. Patients with an initially normal RUS underwent a median of 3 further RUS [IQR: 2-4], with 33 patients (24.6%) undergoing five or more normal RUS. Overall, 197 patients with a normal postoperative RUS needed to be screened yearly to find one asymptomatic renal anomaly.

Conclusion

In a large cohort of patients with mild ARM, only one patient (0.5%) benefited from screening RUS with a renal anomaly requiring intervention, suggesting that if a RUS after the primary repair of a mild ARM is normal, additional screening of the asymptomatic patient is not necessary.

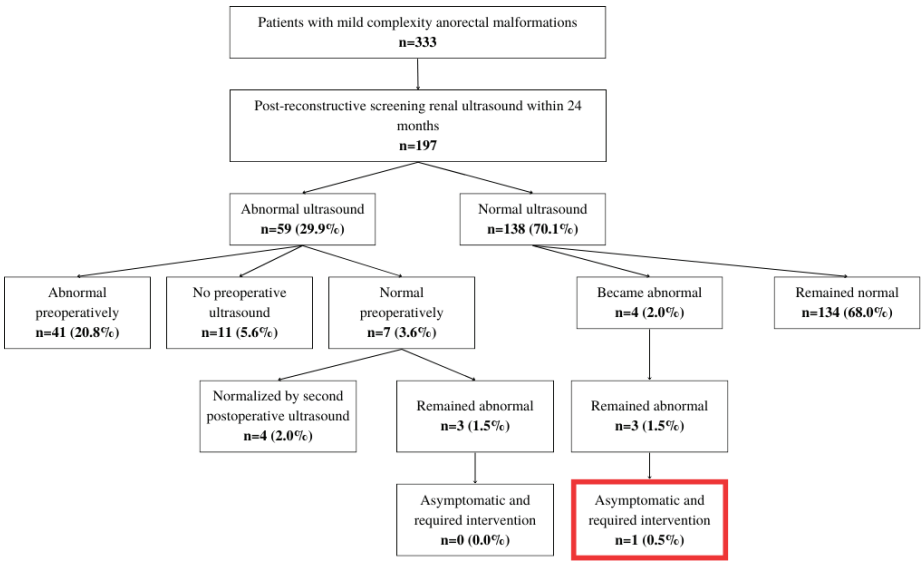


Figure 1. Flow chart describing the utility of postoperative ultrasounds in patients with mild anorectal malformations. Only one patient had a renal anomaly that was asymptomatic and detected on screening ultrasound that required intervention, representing a detection rate of 0.5%.

MORBIDITY OF RECTAL PROLAPSE REPAIR AFTER SURGERY FOR ANORECTAL MALFORMATION

Megan A Read, MD, Kristine L Griffin, MD, Brenna Rachwal, Richard J Wood, MD, Alessandra Gasior, DO

Purpose: Rectal prolapse is a known complication of surgery for anorectal malformations (ARM), however morbidity of prolapse repair and long-term outcomes are not well-described.

Methods: We performed a single-institution retrospective review of patients who underwent surgery for ARM then were treated for rectal prolapse, defined as at least partial thickness mucosal prolapse, at our institution from 2014-2024. Demographics, clinical characteristics, and surgical outcomes were assessed, and compared using Fisher's exact testing.

Results: Of the 1,275 patients with ARM treated at our institution during this period, 99 patients with previously repaired ARM underwent rectal prolapse repair (66 males, 66.7%). Median age at initial surgery for rectal prolapse was 3 years (IQR 1.5 – 7.0). Median follow-up time was 3 years (IQR 1.0 – 5.75). The most common ARM subtypes were cloaca (25.2%) and rectoprostatic fistula (17.2%). Most patients (66.7%) were asymptomatic, with prolapse noted either by parents or on routine post-operative exam.

Clinically significant post-operative stricture was identified in 29 patients, of which 25 required Heineke-Mikulicz stricturoplasty (25.25%). There were no statistically significant differences in stricture formation based on any of the assessed clinical characteristics, including type of ARM, number of prior revisions, indication for repair, or the circumference of prolapse and repair. Notably, 23 patients who developed stricture were asymptomatic prior to surgery (79.3%), which reflects a 34.8% rate of stricture formation for asymptomatic patients, as compared to 18.2% for symptomatic patients. Although not statistically significant ($p = 0.133$), this difference has been practice-changing for our institution, leading to fewer asymptomatic repairs performed and lower burden of stricture requiring additional surgery in asymptomatic patients.

Conclusion: Morbidity from post-operative stricture is not an insignificant concern after prolapse repair, and merits close monitoring and follow-up. Judicious patient selection is critical to minimizing morbidity – we propose that asymptomatic patients should be treated expectantly.

HOSPITAL VOLUME AND REGION ASSOCIATED WITH VARIATION IN VACTERL SCREENING AMONG NEWBORNS WITH ANORECTAL MALFORMATIONS – AN OPPORTUNITY TO OPTIMIZE SCREENING PRACTICES

Thomas O. Xu, MD, Rachel E. Hanke, MD, Butool Hisam, MBBS, Melanie Bowser, BS Inbal Samuk, MD, Erin Teeple, MD, John S. Myseros, MD, Andrea Badillo, MD, Briony K. Varda, MD, MPH, Marc A. Levitt, MD, Christina Feng, MD

Purpose:
VACTERL screening protocols are not standardized, therefore we aimed to evaluate the rate of VACTERL screening among newborns born with anorectal malformations (ARM) across neonatal intensive care units (NICU) in the United States.

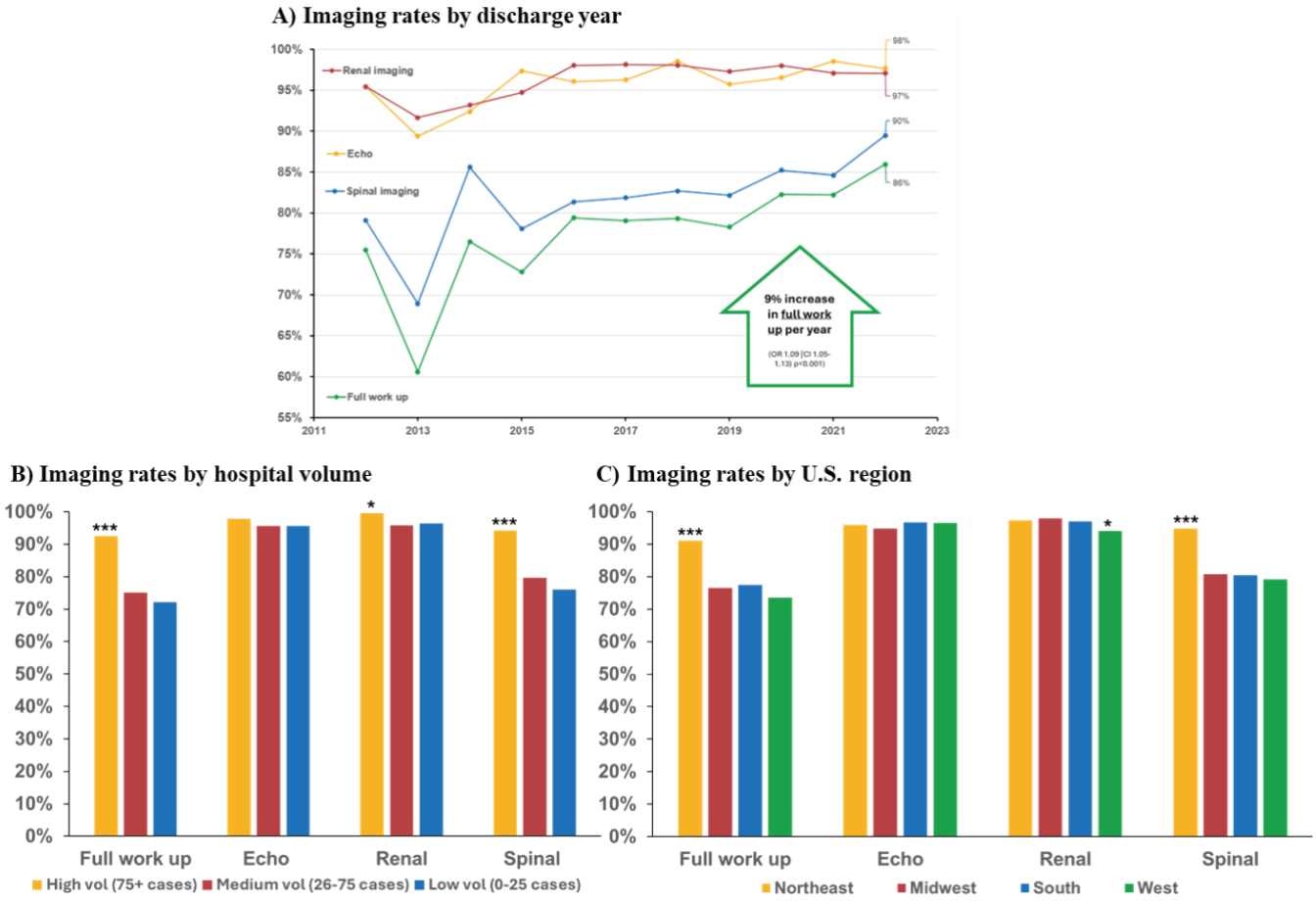
Methods:
A retrospective, multi-institutional cohort study between 1/2012-12/2022 was performed using the Pediatric Health Information Systems (PHIS) database. Relevant ICD9/10 diagnostic and procedure codes were used to identify ARM patients. The primary outcome was completion of a VACTERL workup (echocardiogram + renal imaging + spinal cord imaging). Hospital volume was stratified by the number of total ARM admissions during the study period (low ≤25, medium 26-75, high >75). Descriptive statistics and multivariable regression analysis were performed.

Results:
Between 1/2012-12/2022, 1,955 newborns (35% female) with ARM were admitted across 44 PHIS hospitals. Overall, the full VACTERL screening rate was 78% (96% echo, 97% renal imaging, 82% spinal imaging) however screening rates increased over time with full screening rates approaching 86% by 2022. (Fig 1a). Hospitals that were high volume or located in the Northeast region had higher rates of VACTERL screening (Fig 1b, c). Factors independently associated with completion of screening were high hospital [aOR 4.1, 95% CI 2.6-3.6, p <0.001] and if length of stay was greater than week [aOR 1.6, 95% CI 1.2-2.2, p <0.001]. Odds of full screening were 50% lower in hospitals in the Midwest, South, and West, compared to the Northeast [all p<0.001]. Patients who required mechanical ventilation also had lower odds of full screening [aOR 0.72, CI 0.55-0.93, p=0.01].

Conclusion:
There is still variation in the rate of which ARM patients receive a full VACTERL work up across time, region, and hospital volume, especially with respect to spinal cord screening. Implementation of standardized guidelines or provider education may reduce this variation and improve care.

Disclosure: This abstract will be presented as a poster resentation at AAP Section on Surgery, September 2024.

Figure 1: A) VACTERL screening rates across PHIS hospitals by year, B) VACTERL screening rates by hospital volume C) VACTERL screening rates by Hospital U.S Region *Transition from ICD9 to ICD10 occurred in 2016, OR = odds ratio, CI = 95% confidence interval * = p<0.05, *** = p <0.001



LOWER CHILDHOOD OPPORTUNITY INDEX IS ASSOCIATED WITH DELAYED DIAGNOSIS OF ANORECTAL MALFORMATIONS

Kristine L. Griffin, MD, Shruthi Srinivas, MD, MPH, Megan Read, MD, Jeremy Lun, Alessandra C. Gasior, DO, Richard J. Wood, MD, Ihab Halaweish, MD

Purpose:
Delay in diagnosis of anorectal malformations (ARM) can potentially lead to adverse events. Perineal and rectovestibular fistulas are more likely to be missed on exam compared to other ARM subtypes. We aimed to identify which clinical factors and social determinants of health (SDH) may be associated with missed diagnosis of these ARMs.

Methods:
A retrospective review was performed of all infants with perineal or rectovestibular fistulas seen at a single center from 2014 to 2023. Delayed diagnosis was defined as those made beyond the second day of life. Data were collected regarding birth hospital NICU acuity, location of diagnosis, clinical factors, adverse sequelae, and SDH, including Child Opportunity Index (COI). Categorical variables were analyzed via Fisher's Exact Test. Ordinal variables were analyzed using Cochran-Armitage Test for Trend.

Results:
A total of 171 patients were diagnosed with perineal or rectovestibular fistula during the time period, of which 31.6% had delay in diagnosis (Table 1). There was no significant difference in acuity level of the birth hospital, race, ethnicity, insurance type, need for interpreter, or rural vs. urban setting between delayed and timely diagnosis. As COI increased in the cohort, there was a significant trend toward lower association with delayed diagnosis (Table 1). Those with delayed diagnosis were more likely to present with fecal impaction or have an unplanned pre-operative admission ($p<0.001$). There was no increased risk of colostomy pre-operatively or post-operative complications in patients with delayed diagnosis of ARM.

Conclusion:
In this study, patients with lower COI experienced higher likelihood of delayed diagnosis of ARM. Efforts should be made to improve prompt diagnosis in this at-risk population.

Table 1. Social Determinants of Health in Timing of ARM Diagnosis

		All n = 171	**Timely Diagnosis** n = 117 (68.4)	**Delayed Diagnosis** n = 54 (31.6)	***p*-value**
Demographics	Age at diagnosis, days	0 [0 – 4.1]	0 [0 – 0]	31.9 [5.1 – 182.5]	**<0.001**
	White race	126 (73.7)	87 (74.4)	39 (72.2)	0.51
	Non-white race	45 (26.3)	30 (25.6)	15 (27.8)	
Insurance Type	Public Insurance	85 (49.7)	57 (48.7)	28 (51.9)	0.60
Child Opportunity Index	Very Low	21 (13.0)	11 (9.9)	10 (19.6)	**0.03**
	Low	35 (21.6)	22 (19.8)	13 (25.5)	
	Moderate	24 (14.8)	17 (15.3)	7 (13.7)	
	High	35 (21.6)	24 (21.6)	11 (21.6)	
	Very High	47 (29.0)	37 (33.3)	10 (19.6)	

GYNECOLOGIC ANOMALIES IN PATIENTS WITH ANORECTAL MALFORMATIONS AND ASSOCIATED ANOMALIES: A NATIONAL RETROSPECTIVE COHORT FROM 2016-2023

Kirsten J.H. Das, Thomas O. Xu, Butool Hisam, Melanie Bowser, Inbal Samuk, Erin Teeple, John S. Myseros, Briony K. Varda, Christina Ho, Christina Fang, Andrea T. Badillo, Marc A. Levitt, Allison C. Mayhew

Purpose: Gynecologic anomalies are present at higher rates in patients with anorectal malformations (ARM). However, little is known about specific types of gynecologic anomalies and other co-occurring ARM anomalies. We aimed to assess the co-occurring anomalies present in patients with anorectal malformations and specific gynecologic anomalies.

Methods: A retrospective cohort was analyzed through the Pediatric Health Information Systems (PHIS) database of female patients with an ARM diagnosis. Inclusion criteria included patients who are female sex, admitted within day of life 1 from January 2016 to December 2023 and had either an ICD-9 or ICD-10 diagnosis ARM code. Utilizing ICD-9 and ICD-10 diagnosis codes, we then evaluated gynecologic anomalies and compared rates of other known co-occurring anomalies (e.g., vertebral, cardiac, tracheoesophageal fistulas, renal, and limb anomalies).

Results: Among the 3,078 female patients with anorectal malformation in this cohort, 1,064 (34.6%) had a gynecologic anomaly diagnosis. 378 (35.2%) had a uterine anomaly, 337 (31.7%) had Mullerian agenesis (uterine, cervical, or vaginal agenesis), 136 (12.8%) had a vaginal anomaly (e.g., longitudinal septum), 23 (2.1%) had an ovarian anomaly (e.g., streak ovary or absence of one or both ovaries), and 559 (52.5%) had another gynecologic or unspecified anomaly (e.g., unspecified congenital anomaly of cervix, vagina or malformation of vulva, malformation of clitoris). Rates of other co-occurring anomalies that are associated with ARM (e.g., VACTERL) were present in similar rates across the different gynecologic anomalies. For example, vertebral anomalies were present in 23-27% of the various gynecologic anomalies and tracheoesophageal fistulas (TEF) were present in 3-4% (Table 1).

Conclusions: More than one third of patients with anorectal malformations had an associated gynecologic anomaly. There were no differences in co-occurring anomalies among the different types of gynecologic anomalies. These data helps to emphasize the importance of gynecologic screening in anorectal malformations.

Table 1: Rates of Co-Occurring Anomalies in Patients with Gynecologic Anomalies & Anorectal Malformations

	Vertebral	Renal	TEF	Cardiac	Limb	Total
Uterine Anomaly	258 (25%)	310 (30%)	35 (3%)	242 (24%)	175 (17%)	1020
Mullerian Agenesis	240 (25%)	275 (29%)	42 (4%)	233 (24%)	164 (17%)	954
Vaginal Anomaly	91 (27%)	105 (31%)	11 (3%)	76 (22%)	56 (17%)	339
Ovarian Anomaly	17 (23%)	20 (31%)	2 (3%)	12 (19%)	13 (20%)	64
Other Genital/Unspecified Anomaly	332 (23%)	385 (31%)	385 (3%)	381 (19%)	219 (20%)	1702

ROUTINE POST-OPERATIVE ANAL DILATATIONS FOR THE PREVENTION OF ANAL STRICTURES IN CHILDREN WITH ANORECTAL MALFORMATIONS: A SYSTEMATIC REVIEW

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Abstract

Background:

Despite surgical advances, children with an anorectal malformation typically follow an anal dilatation practice that has changed little since first described by Pena and de Vries in 1982. The role of anal dilatations has been questioned due to variable effectiveness and parental burden; however, the current knowledge has not been consolidated. We aimed to synthesize the available literature on the effectiveness of routine post-surgical dilatations.

Method:

This systematic review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) protocol and prospectively registered (PROSPERO: CRD42022345670). English articles (01/1990 – 06/2023) in PubMed, Medline and EMBASE databases were reviewed. Included studies reported on children with an anorectal malformation (0-18 years) who had reconstructive surgery, followed by routine anal dilatations and an assessment of outcome. Studies that did not describe the dilatation regimen, nor assessed for stricture, were excluded.

Results:

Seven studies (published 2012-2023) met the inclusion criteria, with a combined cohort of 400 patients. The primary aim for most studies was to evaluate if routine, post-operative dilatations reduced anal stricture incidence. 311 (78%) patients received routine post-operative anal dilatations. Dilatation regimens typically followed the Pena protocol, commencing two weeks post-operatively until adequate anal caliber relative to the child's age was reached. Duration of dilatations differed between each study, while dilatation effectiveness, if described, was assessed by clinician experience and anal caliber. A total of 45 of 311 patients (0-39%) developed an anal stricture. Post-surgical stricture rates were similar between those prescribed the Pena protocol and those that received weekly dilatations by a clinician.

Conclusion:

The current understanding of the effectiveness of routine post-operative anal dilatations in preventing anal strictures following surgery for anorectal malformations is limited. Parental burden, age of the child, and psychosocial implications all need to be considered in this patient cohort.

PTSD AMONG PATIENTS WITH ANORECTAL MALFORMATION AND HIRSCHSPRUNG DISEASE

Andreina Giron, MD, Zoe Flyer, DO, John Schomberg, PhD, MPH, Ashley Bone, MSN, CPNP, Andreina Urrutia Gonzales, BS, Elicia Wartman, PhD, Ashish Chogle, MD, MPH, Peter Yu, MD, MPH, Donald Shaul, MD, Hira Ahmad, MD

Purpose: Management of patients with anorectal malformations (ARM) and Hirschsprung Disease (HD) requires multiple interventions and lifelong follow up. We aimed to determine the prevalence of Post-Traumatic Stress Disorder (PTSD) among patients with ARM and HD.

Methods: After IRB approval, a validated self-rating *DSM-IV* PTSD (PCL-5) checklist was administered to 30 patients with ARM and HD to assess the presence and severity of PTSD symptoms. The PCL-5 is a 20-item questionnaire, corresponding to the DSM-5 symptom criteria for PTSD. The survey (likert scale) is a provisional diagnostic tool to connect patients to appropriate mental health care resources. PCL-5 cutoff score between 31-33 is indicative of probable PTSD.

Results: All 30 patients completed the survey; the participants had even gender distribution, a median age of 12-64 years and majority were White (76.6%). Education levels varied, with 30% holding a Bachelor's degree and 20% having less than a high school education; employment status varied across as well, with only 3 (10%) reported being unable to work. 27 patients had a diagnosis of ARM (90%). Among the 30 respondents, 12 (44.4%) scored >31 on the PCL-5 score; 17 (62.9%) of patients reported "moderate" or greater in all PCL-5 domains. When asked about highest stressors, 13 (43.2%) ranked anal dilation/enema treatment as their highest stressor, while 12 (39.9%) reported effect of diagnosis on body image and social life as the highest stressor.

Conclusions: The high prevalence of PTSD among patients with ARM and HD underscores the need for mental health support. About half of the patients met the provisional PTSD diagnosis based on PCL-5, with a significant proportion in the moderate to severe category. These findings highlight the critical role of early psychological involvement and availability of mental health care resources for these patients.

A QUALITATIVE ASSESSMENT OF TRANSITION OF CARE FOR THE ANORECTAL MALFORMATION PATIENT

Authors: Yvanna Bidart MD, Nutan B. Hebballi BDS, MPH, PhD, Preethi Kannan BS, Maryam Broussard MHA, CCRP, Maximilian Selbst BS, Danielle M. Hsu MD, KuoJen Tsao MD, Akemi L. Kawaguchi MD, MS

Introduction: Given the complexity of care and the long-term health complications for patients with anorectal malformations (ARM), the need for comprehensive transitional services is a priority. We aim to identify the challenges faced by patients with ARM during the transition of care in order to develop a program to improve their quality of care and decrease long-term health complications.

Methods: A qualitative study was conducted between June 2023 and April 2024 with ARM patients (≥ 18 years), who have already transitioned or are actively transitioning to adult providers. Participants were recruited through the Comprehensive Congenital Colorectal Program and via social media. Semi-structured interviews were conducted by a pediatric surgeon using an online platform (WebEx). Interviews were recorded and transcribed, then coded by a 4-member team. Thematic analysis was performed to identify themes across patient experiences.

Results: Of the fourteen participants included, the median age was 48 years (range: 18 – 70) with twelve female and two male patients. Nine patients had imperforate anus and five patients had cloaca. Patient experiences were centered around six themes: gaps in health care (difficulty finding adult providers, lack of continuity of care), gaps in knowledge (from the patient, parents, and providers), medical-related trauma (physical, emotional), mental health (anxiety, depression, therapy/counseling/support), social maltreatment (by peers, parents, spouse/partners), and quality of life (health-related, psychosocial) (Figure).

Conclusion: Patients with ARM face emotional, social, and practical challenges that impact their lives into adulthood. Educating patients, parents, and adult providers about the complexities of these conditions; identifying qualified adult providers; and extending psychosocial support to patients and their families should be prioritized during the transition of care.

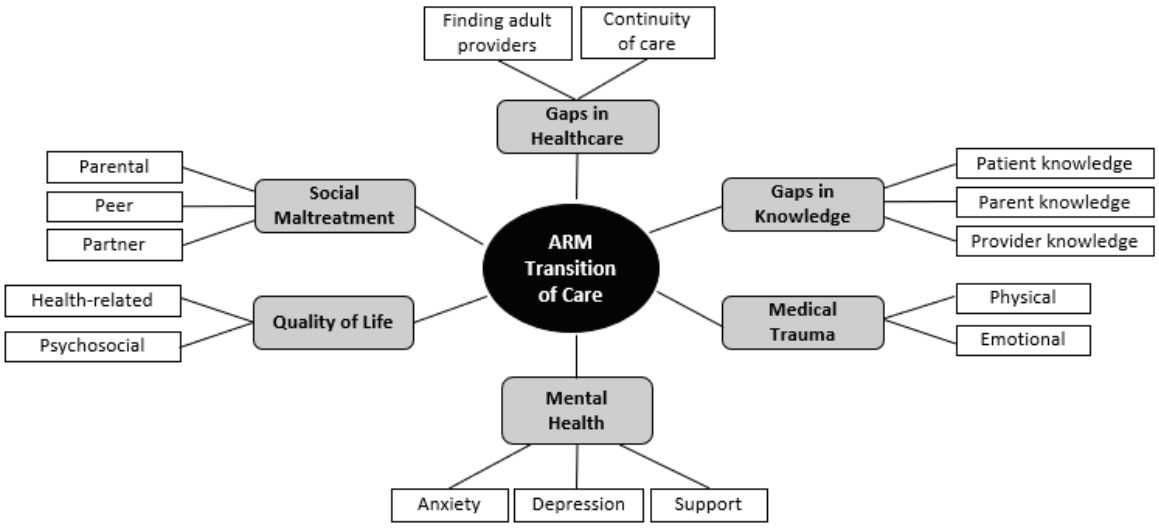

Figure. Thematic map of transition from pediatric to adult care in ARM patients

EVALUATING THE IMPACT OF THE PCPLC LEARNING COURSE ON PEDIATRIC SURGERY FELLOW'S CLINICAL PRACTICE

Authors; Rebecca M. Rentea, Liza Bokova, Casey Calkins, Belinda Dickie, Chris Gayer, Richard Wood, Elizabeth Speck, Marc A. Levitt, Payam Saadai

Members of the PCPLC consortium

Purpose:

Pediatric surgery fellows often receive limited and inconsistent exposure to pediatric colorectal cases, with variations depending on departmental practices and center volume. To address this educational gap, the Pediatric Colorectal and Pelvic Learning Collaborative (PCPLC) was established to enhance education in pediatric colorectal surgery through specialized courses. This study aims to assess the impact of a dedicated PCPLC course on the clinical practices of pediatric surgery fellows.

Methods: A survey was distributed to participants registered as fellows in the PCPLC course, which was conducted from November 2023 to July 2024. The course included didactic and scenario-based components, video materials, and clinical case presentations with interactive questions. Surveys were administered to pediatric surgery fellows both before and after the course. The questionnaires were divided into four sections: demographics, management of anorectal malformations (ARMs), approach to Hirschsprung disease (HD) and functional constipation, and overall course impressions. Quantitative statistical analysis and qualitative thematic analysis were performed to assess changes in clinical practices following the course.

Results: The response rate was pre 50% (n=21) and post 24% (n=10). Participants had a median of 1.5 years (IQR 1-2) of pediatric surgery training following 6 years (IQR 5-7) in general surgery, predominantly in the US (n=17, 85%) and Canada (n=3, 15%). Most had dedicated colorectal centers (n=16, 76%) and specialized staff (n=17, 85%) at their institutions. Post-course, routine post-posterior sagittal anorectoplasty (PSARP) dilations decreased from 61% (n=11) to 30% (n=3), p=0.06. Preference for perineal body sparing PSARP increased from 26% (n=5) to 80% (n=8), p<0.01. Routine use of MiraLAX for ARMs decreased (29% vs. 10%, p=0.1). HAEC management shifted from outpatient initial treatment to preferring immediate admission (48% vs. 70%, p=0.13). Use of a checklist for HAEC diagnosis increased from 24% (n=5) to 100% (n=10), p<0.01. Colonic mapping for HD cases with proximal transition zones increased from 67% to 100% (n=14 to 10), p<0.01. The practice of performing an antegrade continence enema (ACE) before managing severe constipation after excluding HD rose from 10% (n=2) to 60% (n=6), p<0.01.

Conclusion: The PCPLC course significantly influenced pediatric surgery fellows' clinical practices, showing notable shifts in management approaches, including reduced routine post-PSARP dilations, increased preference for perineal body sparing PSARP, greater use of diagnostic checklists for HAEC, initial colonic mapping and ostomy creation for long-segment HD, and increased use of ACE in managing severe constipation.

EVALUATING THE IMPACT OF THE PCPLC LEARNING COURSE ON PEDIATRIC SURGERY FELLOW'S CLINICAL PRACTICE

Authors; Rebecca M. Rentea, Liza Bokova, Casey Calkins, Belinda Dickie, Chris Gayer, Richard Wood, Elizabeth Speck, Marc A. Levitt, Payam Saadai

Table 1. Clinical Practices Before and After the Course Completion.

Variables	**Pre-course** n=21	**Post-course** n=10	**p-value**
Anorectal Malformations			
Routine post-PSARP dilations	11/18 (61%)	3 (30%)	0.06
Preferring perineal body sparing PSARP	5/19 (26%)	8 (80%)	**<0.01**
Routine use of MiraLAX in ARM patients	6 (29%)	1 (10%)	0.1
Hirschsprung Disease and Functional Constipation			
HAEC managed outpatient first and admitted afterward if the symptoms worsen	10 (48%)	7 (70%)	0.13
Using a checklist for HAEC diagnosis	5 (24%)	10 (100%)	**<0.01**
Performing AMAN prior to management of severe constipation after HD ruled out	2 (10%)	6 (60%)	**<0.01**

FREQUENCY OF VACTE(G)RLS ASSOCIATED ANOMALIES IN NEWBORNS WITH ANORECTAL MALFORMATIONS ACROSS HOSPITALS WITHIN THE PEDIATRIC HEALTH INFORMATION SYSTEMS DATABASE: NEONATAL DIAGNOSES AND BEYOND

Thomas O. Xu, MD, Rachel E. Hanke, MD, Kirsten Das, MD, Butool Hisam, MBBS, Melanie Bowser, BS, Inbal Samuk, MD, Erin Teeple, MD, Allison Mayhew, MD, John S. Myseros, MD, Andrea Badillo, MD, Briony K. Varda, MD, MPH, Marc A. Levitt, MD, Christina Feng, MD

Purpose:
The purpose of this study was to report rates of associated congenital diagnoses found in patients with ARM in the United States using the Pediatric Health Information Systems (PHIS) database.

Methods:
A retrospective multi-institutional cohort study was performed using the PHIS database for patients born with ARM admitted to a neonatal intensive care unit (NICU) between 1/2016-12/2022 across 44 US children's hospitals. Relevant ICD10 diagnostic and procedure codes were used to identify ARM patients. ICD10 codes were then used to identify VACTERL anomalies associated with ARM, including spinal cord and gynecologic malformations (Table 1). We also assessed the rate of diagnosis at a patient's initial NICU admission as well as the rate of diagnosis across all subsequent hospital encounters to identify anomalies that may be diagnosed beyond the neonatal period.

Results:
1,467 patients met inclusion criteria, 496 (34%) were female. The rates of an associated VACTERL diagnoses while admitted to the NICU were 35% vertebral/spinal, 73% cardiac, 10% TEF, 35% renal, and 13% limb. The rates of diagnoses across all hospital encounters for each patient were all higher with 45% vertebral/spinal, 77% cardiac, 10% TEF, 40% renal, and 16% limb which may indicated conditions with delayed presentation or diagnosis. Notably, spinal cord malformations had a discrepancy of +14% rate of prevalence between index NICU admission and subsequent hospital encounters. Among female patients, 16% had an associated congenital gynecologic malformation diagnosis at birth but 26% had a gynecologic malformation diagnosis across all hospital encounters.

Conclusion:
The most commonly associated anomalies with ARM are cardiac, vertebral/spinal, renal, and gynecologic (in females). Discrepancies in diagnosis rates at birth compared to all encounters suggests potential for improvement in screening for certain diagnoses. We highlight spinal cord and gynecologic malformations as additional "VACTERL" anomalies and suggest changing the acronym to VACTE(G)RLS

Disclosure: This abstract will be presented as an **oral** presentation at AAP Section on Surgery, September 2024.

Category (N=1467)	NICU	All Encounters	Difference
Vertebral/Spinal malformations[1]	514 (35.0%)	666 (45.4%)	152 (10.4%)
Cardiac malformations[2]	1070 (72.9%)	1136 (77.4%)	66 (4.5%)
Tracheoesophageal fistula	140 (9.5%)	150 (10.2%)	10 (0.7%)
Gynecologic malformations[3] (*N=496)	80 (16.1%)	128 (25.8%)	48 (9.7%)
Renal/urinary system malformations[3]	509 (34.7%)	586 (39.9%)	77 (5.2%)
Limb malformations[4]	195 (13.3%)	230 (15.7%)	35 (2.4%)
Spinal cord malformations	246 (16.8%)	418 (28.5%)	172 (11.7%)

Table 1: Rates of VACTE(G)RLS diagnoses for patients born with anorectal malformation at time of index neonatal intensive care unit admission and across all subsequent hospital encounters between 1/2016-12/2022. 1. Inclusive of bony vertebral malformation, spinal cord malformations, spina bifida, rib malformation, Arnold-Chiari syndrome. 2. Inclusive of atrial/septal defect, malformation of great arteries, congenital valve malformations, Tetralogy of Fallot, dextrocardia, levocardia, cor triatriatrum, pulmonary infundibular stenosis, congenital subaortic stenosis, malformation of coronary vessels, congenital heart block, other specified congenital malformations of the heart, common atrial trunk, discordant ventriculoarterial connection, double inlet ventricle, isomerism of atrial appendages, and septal malformations. 3. Inclusive of upper and lower Mullerian tract anomalies, ovarian/fallopian tube anomalies. *out of female patients. 4. Inclusive of foot, hand, upper, lower, and other non-specified limb anomalies.

MALONE ANTEGRADE CONTINENCE ENEMA (MACE) - REASSESSMENT OF COMPLICATIONS WITH ADVANCEMENTS IN TECHNIQUE

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Purpose:
Complications following the Malone antegrade continence enema (MACE) procedure have been described to be as high as 30%. We aimed to reassess the incidence of complications for the MACE procedure as experience has been gained and the technique has undergone modifications.

Methods:
We performed a single institution, retrospective review (from 1/2010 to 1/2023) of all patients who underwent MACE for treatment refractory constipation and/or soiling. Our primary outcome was any complication that involved either a procedural intervention (operative and/or interventional radiology (IR)) or channel loss. Patient and clinical covariables were compared between patients with and without complications using a univariate analysis. Advancements have included changes to the surgical technique, post-operative management protocols, and routine collaboration with IR.

Results:
188 patients underwent a MACE (176 appendicostomy, 12 neo-appendicostomy; 25 done as a split appendix with a Mitrofanoff) with a median follow up of 17 months (range: 11–26 months). 35 (19%) patients had a major complication including stenosis (8%), device loss (3%), prolapse (3%), leakage (2%), and other (3%). 20 (10%) patients underwent an IR intervention and 17 (9%) required operative intervention (Figure 1). Of those with complications, 22 (63%) patients required only an IR intervention or a minor skin-level revision. Six (3%) patients ultimately experienced channel loss. On univariate analysis, there was no difference in the complication rate when the underlying diagnosis, type of MACE, or use of laparoscopy was considered (all $p>0.05$).

Conclusion:
In this single institution retrospective study, one in five patients undergoing MACE may experience a major complication. Fortunately, most were amendable to skin level repairs or IR salvage procedures. This apparent improvement in complication rates compared to historical rates may be due to refinements of surgical technique, changes to post-operative management, and collaboration with IR.

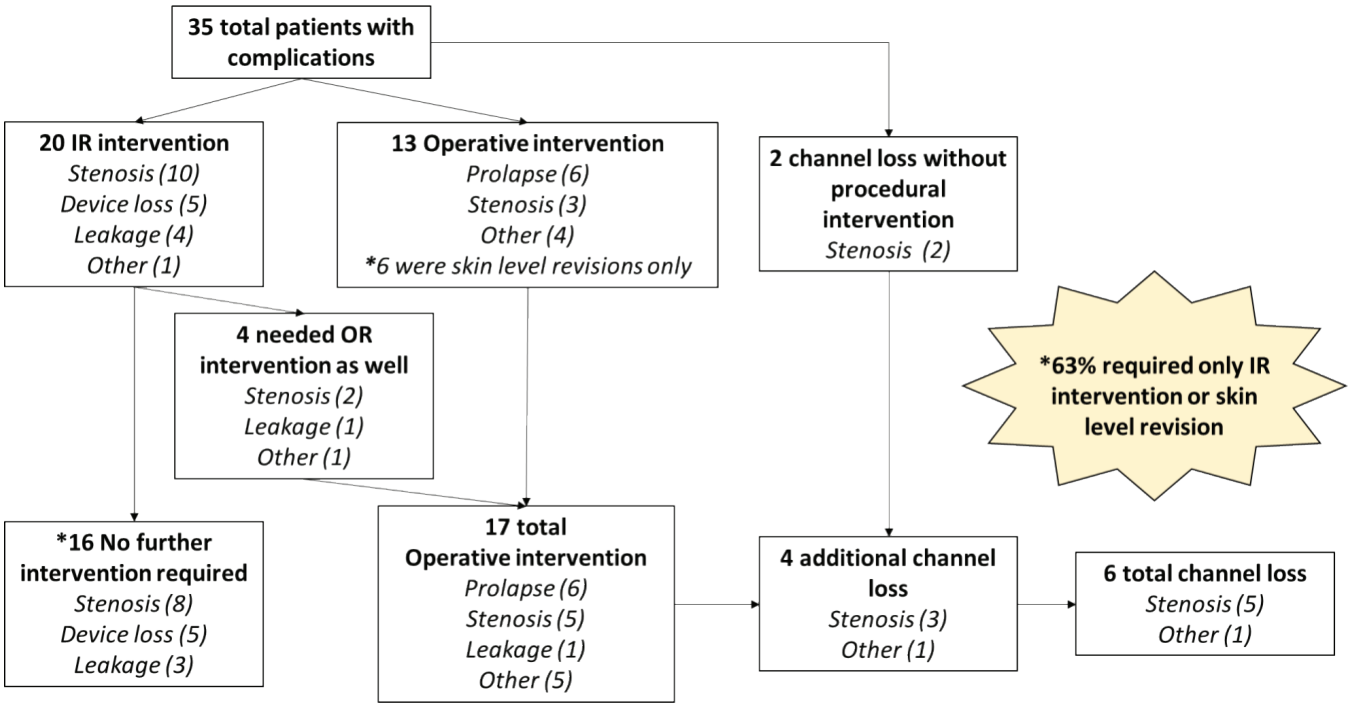
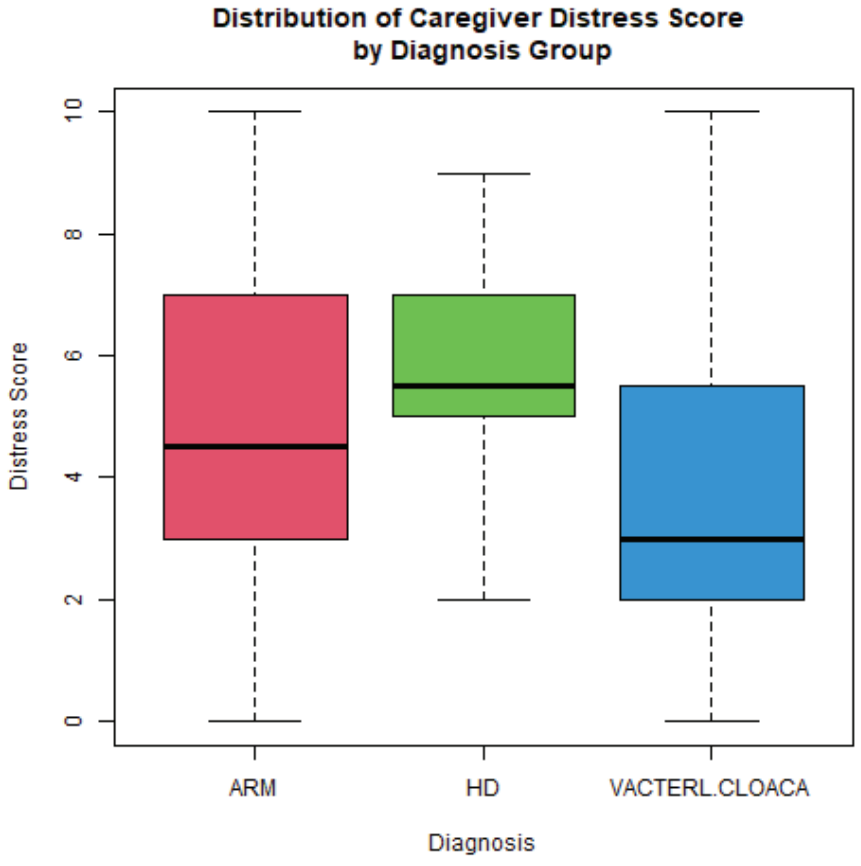


Figure 1. Complications of patients who underwent Malone antegrade continence enema (MACE) procedure. IR = Interventional radiology.

DISTRESS AMONG PARENTS AND CAREGIVERS OF PATIENTS WITH ANORECTAL MALFORMATION AND HIRSCHSPRUNG DISEASE

Andreina Giron, MD, Zoe Flyer, DO, John Schomberg, PhD, MPH, Ahsley Bone, MSN, CPNP, Andreina Urrutia Gonzalez, BS, Ashish Chogle, MD, MPH, Donald Shaul, MD, Hira Ahmad, MD

Purpose: Hospital anxiety and depression occurs commonly among family members who care for patients with congenital anorectal disorders. Better understanding of the prevalence is needed.
Methods: After IRB approval, a validated DT-P (Distress thermometer for parents) was given to 98 caregivers of the above patients at the Pull-through Network (PTN) conference, asking them to rank their degree of distress from 0 (no distress) to 10 (extreme distress) within the past week in a variety of areas including practical, emotional, family/social, physical, support, and communication categories. Scores greater than 4 strongly correlate to hospital anxiety, depression and parenting stress index. Diagnosis and demographic data were also collected. Participants were also asked to rank their most significant stressors with 1 being the most significant stressor and 5 was least significant.
Results: Among 98 caregivers that completed the survey, 90% of respondents were parents, 68% were female, and 61% were 35-54 years old. The diagnosis groups included isolated ARM (n=61), Cloaca (n=11), HD (n=9), and VACTERL (n=17). The DT-P scores over 4 occurred in 50% of ARM/cloaca, 35% of VACTERL and 77% of HD caregivers, p=0.03, among those completing the scoring. High proportions of responders reported high stress (score of 1-2): anal invasive treatment such as dilations, enemas, irrigations (36%) and unexpected diagnosis (46.9%).
Conclusions: Anxiety and/or depression was present in over half of caregivers of congenital anorectal disorders even though respondents were asked to simply quantify their stress over the past week. Unexpected ARM or HD diagnosis in the newborn period was a source of a significant stressor in almost half of the caregivers. These findings suggest that mental health interventions should be utilized early, perhaps as soon as the diagnosis of an ARM or HD is made.



USE OF COLONIC MANOMETRY IN PATIENTS WITH HIRSCHSPRUNG DISEASE FOLLOWING PULL-THROUGH

Kristine L. Griffin, MD, Shruthi Srinivas, MD, MPH, Raul E. Sanchez, MD, Peter L. Lu, MD, MS, Richard. J. Wood, MD

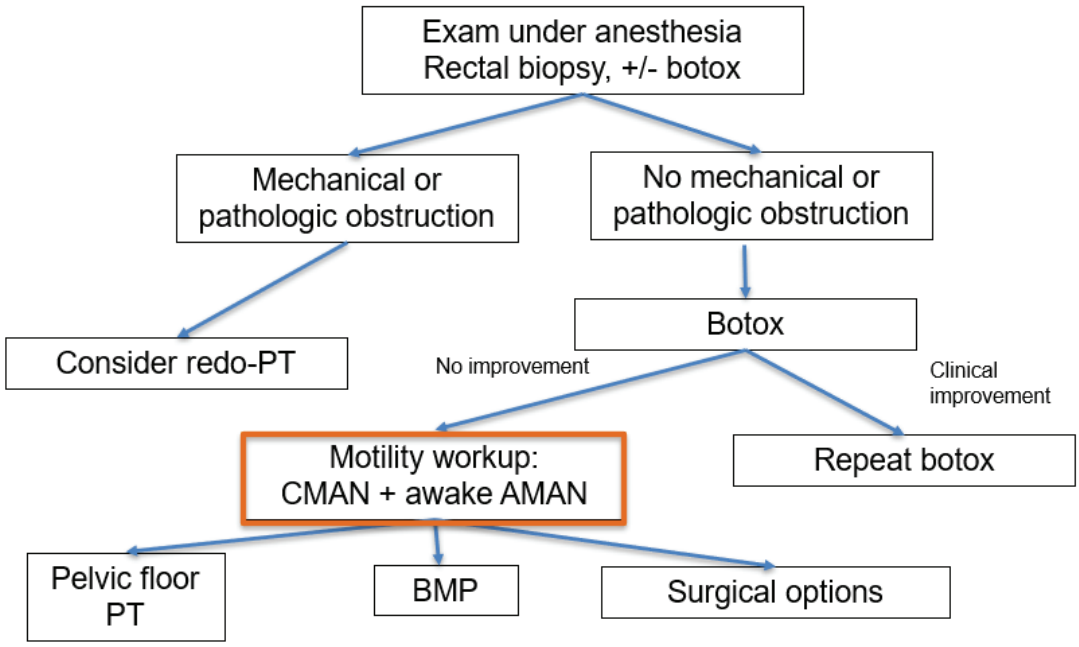
Purpose: We identified patients with Hirschsprung disease (HD) who presented with obstructive symptoms concerning for dysmotility following either primary or redo pull-through (PT). Of the patients who underwent colonic manometry (CMAN), some demonstrated significant colonic dysmotility. We sought to describe these patients and determine where CMAN should fit into our workup algorithm for patients prior to undergoing surgical intervention (**Figure 1**).

Methods: A query was performed using our CMAN database to identify all patients with a history of HD who underwent CMAN at our center from 2015 to 2023. Patients were analyzed for surgical and medical history, CMAN results, and subsequent management.

Results: Forty-one patients with obstructive symptoms underwent CMAN during the study period. The majority of patients (70.7%) originally had rectosigmoid transition zone. There were 21 patients (51.2%) who had normal motility and 20 (48.8%) who had abnormal motility. Among those with normal motility, 18 (85.7%) were managed with nonoperative measures such as pelvic floor physical therapy (PFPT), anal sphincter botox, and bowel management. The 3 patients who underwent surgery had redo PT for retained transition zone. Of the 20 patients with abnormal motility, 8 (40%) were managed operatively with either redo PT or temporary ileostomy with plans to repeat CMAN in 6-12 months. There was no significant difference in motility status between those managed operatively versus nonoperatively (p=0.085). Nine patients underwent redo PT following CMAN testing. Most (66.7%) of these patients had retained transition zone, however 3 of the patients with distal colonic dysmotility had normal pathology and redo was done to remove the nonfunctioning colon.

Conclusion: CMAN testing can guide treatment in HD patients with obstructive symptoms post-PT. Most patients with normal motility can be managed nonoperatively. CMAN can help guide care in patients in whom mechanical or pathological obstructions have been ruled out.

Figure 1. Workup algorithm for patients with obstructive symptoms following pull-through



FREQUENCY AND SEVERITY OF DIAPER DERMATITIS IN CHILDREN FOLLOWING STOMA CLOSURE

Corresponding Author: Jessica McAuliffe FNP-BC Co-Authors: Lindsay Pesacreta, FNP.; Lindsay Clarke, PA.; Krystal Artis, PA.; Julie Choueiki; Marc Levitt, MD; Emma Varner, DNP; Katherine Worst, PNP; Justine Gagnon RN

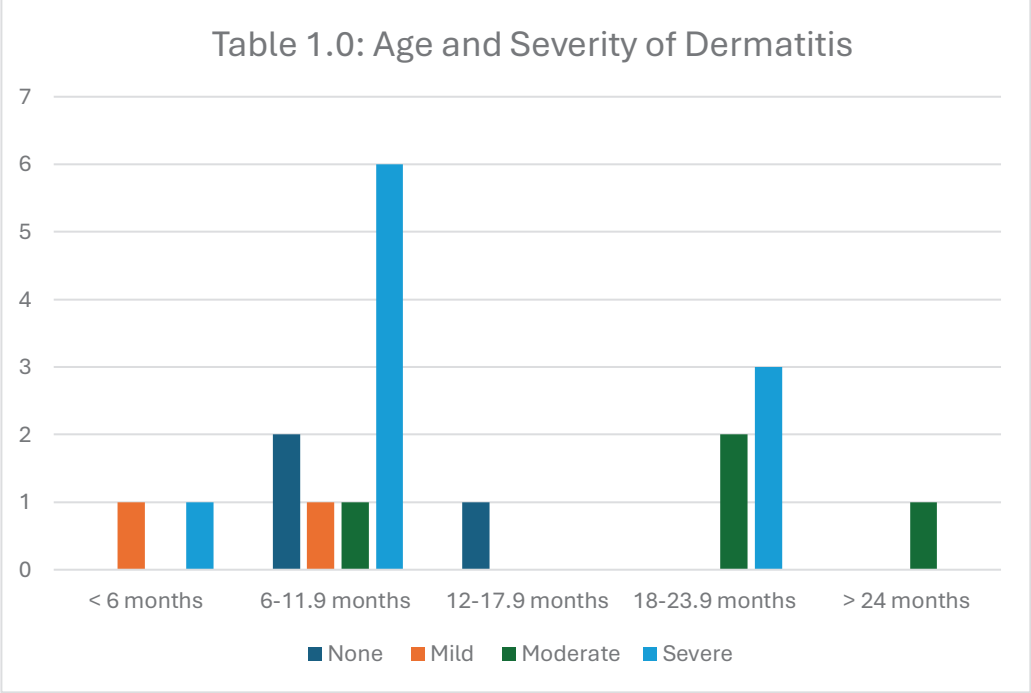
Purpose: Development of a randomized controlled trial (RCT) to determine if applying stool to the perianal area prior to ostomy closure decreases the incidence of diaper dermatitis.

Method: Prior to an IRB application for an RCT, we performed a review of our patients (IRB Pro00016402: Colorectal Patient Registry) to assess the frequency and severity (graded as: none, mild, moderate, or server) of diaper dermatitis after ostomy closure. Our routine treatment included zinc oxide for mild dermatitis, triple paste (zinc oxide, cholestyramine, and nystatin) for moderate, and Marathon or Cavilon for severe. No patient received the proposed intervention that will be the protocol for the RCT.

Results: 28 children (age 5 months to 8 years, median age of 11 months) had their ostomy closed between July 2023 and December 2023 were reviewed. The original surgery included a primary PSARP in 19, a pull-through in 7 (6 for HD, 1 with colonic atresia), and a cloacal repair in 2. 23 had a colostomy closure and 5 an ileostomy closure. To prevent constipation, 17 (61%) were discharged after their stoma closure on laxatives and 3 were treated for hypermotility. 2 patients were discharged on antegrade flushes.

Of 19 with a documented diaper rash plan, dermatitis was severe in 10 (35%) moderate in 4 (14%) mild in 2 (7%), and 3 (10%) had no dermatitis. All cases of severe dermatitis occurred in children <= 24 months. (table 1.0).

Conclusion: Diaper dermatitis affected more than half of patients after a stoma closure. Based on the severity of this problem, we plan to develop a RCT to study the impact of applying stool to the perianal area in advance of ostomy closure to see its effect on post stoma closure dermatitis. We aim to compare rates of this treatment to our current protocol.



COMPLICATIONS AND LONG-TERM OUTCOMES OF PATIENTS WITH CLOACAL MALFORMATION UNDERGOING NEOVAGINA CREATION

Wendy Jo Svetanoff, MD MPH, Kristine L Griffin, MD, Shruthi Srinivas, MD MPH, Frances Fei, MD, Geri Hewitt, MD, Richard J Wood, MD, Chelsea Kebodeaux, MD

INTRODUCTION: Up to 60% of patients with a cloacal malformation have a Mullerian anomaly. In cases where the vagina is absent or cannot reach the perineum, a neovagina is often created. Long-term complications and outcomes of this are not well described. We aimed to describe the gynecologic outcomes of patients who underwent neovagina creation with follow-up at our institution.

METHODS: After IRB approval, a single institution retrospective review of patients ≥ 8 years old with cloacal malformation who underwent intestinal neovagina creation. Demographics, surgical intervention, complications, and long-term gynecologic function were assessed.

RESULTS: Forty patients were included. The median age at neovagina creation was 3.95 years (IQR 1.40, 7.61). Seventeen (43%) patients had an absent vagina, and 12 (30%) had an absent uterus. 65% had a colonic neovagina creation, while 28% received a small bowel neovagina. Median age at most recent follow up was 9.7 years (IQR 7.6, 14.1).

There was no difference in 30-day complications, incidence of vaginal prolapse, introital stenosis, or graft stenosis between those undergoing small bowel vs colonic neovagina (Table 1). More colonic neovagina patients required neovagina revision (14.8% vs 8.3%, p=0.58) or removal (7.4% vs 0%, p=.34), but this was not statistically significant. Of those with neovagina prolapse, 1 small bowel (8.3%) and 2 colonic grafts (7.4%) required prolapse repair. One small bowel (8.3%) and 7 colonic (26%) patients underwent introitoplasty (Table 1). Eight patients (20%) required vaginal dilations during follow-up and 10 patients reported bothersome vaginal discharge. Of the 4 patients who disclosed having penetrative sex (3-colon, 1-unknown), 3 reported dyspareunia (2-colon, 1-unknown). No patients have become pregnant.

CONCLUSION: Creation of a neovagina requires long-term follow-up and multi-disciplinary collaboration with gynecology to address gynecologic concerns as the patient matures and to educate patients on potential options available to improve their quality of life.

Table 1: 30-Day Complications and Long-Term Vaginal Complications after Neovagina Creation. The incidence of 30-day and long-term complications was compared between patients who underwent small bowel vs colonic neovagina creation.

Complications	Small Bowel Neovagina (n=12)	Colon Neovagina (n=27)	p-value
Dehiscence	0 (0%)	3 (11.1%)	0.24
Wound Infection	0 (0%)	2 (7.4%)	0.34
Readmission in 30 Days	1 (8.3%)	2 (7.4%)	0.92
Return to OR in 30 Days	1 (8.3%)	1 (3.7%)	0.55
Long-term Complications			
Vaginal Prolapse	2 (16.7%)	4 (14.8%)	0.88
Introital Stenosis	3 (25%)	7 (26%)	0.95
Vaginal Dilations Needed*	2 (16.7%)	5 (18.5%)	0.89
Known Menstrual Obstruction	0 (0%)	3 (11.1%)	0.24

*One patient (aged 46yo) had her reconstruction done elsewhere and operative records were not available to determine type of neovagina creation.

DESCRIPTIVE ANALYSIS OF PATIENTS WITH DELAYED DIAGNOSIS OF ANORECTAL MALFORMATION IN A MULTICENTER NATIONAL DATABASE

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Purpose: The aim of this study was to describe the population of patients in the United States diagnosed with anorectal malformations (ARMs) in a delayed fashion, their management, the effect on surgical outcomes and complications, and long-term bowel function.

Methods: A retrospective cohort study was conducted using the PCPLC data registry from January 2017-July 2024. Patients were included in the study if they were diagnosed with ARM after 180 days of life and had appropriate enrollment forms complete.

Results: A total of 87 patients met inclusion criteria. The median age at diagnosis was 13 months. Fifty two percent of patients were female and 71.3% of patients were white. The most common ARM type was perineal fistula (62.1%). Forty percent of patients underwent a full VACTERL work up. Overall, associated diagnoses were rare. Fifty two of the 87 patients underwent primary ARM repair at a PCPLC center. Most patients with underwent a posterior sagittal anorectoplasty (PSARP) (73.1%). Of those with documented surgical repair, 75% did not require diverting ostomy before or after their procedure. Complication rates were low but included dehiscence of anoplasty (1.9%), dehiscence of perineum (7.7%), and infection of deep surgical site (1.9%). At the one-year post-operative visit, 37.9% of patients had no specific bowel management plan while another 39.1% were on laxatives. Additionally, 44.8% of patients were not toilet trained for stool, though 74% of these patients were less than 2 years of age.

Conclusion: The subset of patients who are born with ARMs but diagnosed more than 180 days after birth are less likely to receive a full VACTERL screening. Of those who do undergo screening, only a small percentage have associated diagnoses. Three quarters of patients did not require diverting ostomy at the time of repair and post operative complication rates were low.

PREDICTING THE NEED FOR VAGINAL AUGMENTATION IN PATIENTS UNDERGOING CLOACAL RECONSTRUCTION

Mariana Moncada-Madrazo, MD, MEd., Kirsten Jay Hartwick Das, MD., Marc A. Levitt, MD., Briony Varda, MD., Christina Ho, MD., Christina Feng, MD., Andrea Badillo, MD., Veronica Gomez-Lobo, MD., Allison C. Mayhew, MD.

Purpose To predict the need for vaginal augmentation during cloacal reconstruction when a tension-free closure cannot be achieved and to assess for post-operative complications such as vaginal stenosis.

Methods A retrospective cohort study was conducted of all patients who underwent primary cloaca repair at a colorectal center (2020 to 2023). Data was analyzed utilizing descriptive statistics, Mann-Whitney U, and Chi Square test. This study was classified as IRB exempt.

Results 30 patients with a mean age at surgery of 14 months (range 4-77 months) formed our cohort. Before reconstructive surgery, all patients underwent an exam under anesthesia, with cysto-vaginoscopy and cloacagram (Table 1). For reconstruction, 15 underwent total urogenital mobilization (TUM), 13 required urogenital separation (UGS), and two patients with Mullerian agenesis underwent reconstructive surgery for the rectum only without vaginal reconstruction. For vaginal reconstruction, 24/28 (86%) had a native vagina pull-through and 4/28 (14%) required a transposition graft (2 with colon, 1 with rectum, and 1 with small bowel). 8 (50%) who underwent native pull through and 2 (66%) who had a transposition graft had a post operative exam which showed some degree of stenosis. There was a statistically significant difference in common channel length on cloacagram in patients without stenosis 2.35cm (1.5-4.4cm) compared with those with stenosis in the post operative period 4.10cm (1.61-5.2cm, p=0.026). No difference was observed in the preoperative radiologic vaginal length in the same study 4.45cm (3.4-6.5cm) vs. 5.19cm (2.3-9.2cm, p=0.67). Patients who required UGS had a greater incidence of postoperative stenosis (88%) vs TUM (40%) p=0.03.

Conclusions A tension-free vaginoplasty was able to be performed in both short and long common channel cloacas. Patients with stenosis had a longer common channel. For longer common channels an interposition graph should be considered to avoid postoperative stenosis.

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Table 1
Preoperative measurements in cm

			Native Pull-through (N=24)	Transposition Graft (N=3)	Complete vaginal replacement (N=1)	No reconstruction (N=2)
	Cysto-vaginoscopy					
		Length of common channel	2.16 (0.8 – 5)	4.4 (4 – 4.8)	1	2
		Length of urethra	1.89 (0.5 – 3)	1.2 (0 – 1.2)	2.3	1.5
		Length of vagina	4.11 (2 – 6.3)	3.9 (1.6 –6.2)	Not assessed/vaginal opening not present	0
	Cloacagram					
		Length of common channel	2.58 (0.8 – 5.2)	4.55 (0 – 5.1)	1	4.45 (1.9 – 7)
		Length of urethra	2.0 (0.5 – 3.2)	1.75 (0 – 1.75)	2.5	1 (0.5 – 1.5)
		Length of vagina	4.77 (2.3 – 4.12)	3.9 (1.6 – 6.2)	High vagina above bladder neck	0

ASSOCIATION OF SOCIAL DETERMINANTS OF HEALTH AND HIRSCHSPRUNG-ASSOCIATED ENTEROCOLITIS IN CHILDREN

Swathi Raikot, MBBS, Goeto Dantes, MD, Afrin Jahan, Nikolay Barykov, Jonathan Beus, MD, MS, Allison F. Linden, Hanna Alemayehu, MD

Purpose:
Hirschsprung-associated enterocolitis (HAEC) is the leading cause of morbidity and mortality in children with Hirschsprung disease (HD). Social determinants of health (SDOH) play an important role in healthcare outcomes. The goal of this study was to determine if SDOH are associated with HD-related outcomes including recurrent HAEC.

Methods:
An IRB-approved, single institutional, retrospective study was performed of children with HD diagnosed between 2015 - 2022 who underwent pull-through surgery within the first year of life. Patients were identified using ICD10 code Q43.1 matched with pull-through procedure names. Demographics, timing of diagnosis to surgery, and HAEC admissions within 1year of pull-through were collected. Childhood opportunity index (COI), area deprivation index (ADI), and social vulnerability index (SVI), composite measures of SDOH, were calculated for each patient. Outcomes were compared by SDOH using Chi-square tests. P-values<0.05 were considered statistically significant.

Results:
A total of 158 patients with HD underwent pull-through surgery within 1year of age. Seventeen (11%) had ≥2 HAEC readmissions and 141 (89%) had ≤1. There was no significant difference in race, ethnicity, language, insurance type, COI, SVI, and ADI levels between those with ≥2 vs. ≤ 1 HAEC readmissions (Table 1). COI, ADI, and SVI levels were also not significantly associated with any HAEC admission within 30 days, 90 days, and 1year of pull-through. There was no significant difference in COI or SVI levels when comparing those admitted to the general floor vs. ICU (p=0.56 and 0.69, respectively) and those with short vs. long (≥5days) LOS during HAEC admission (p=0.37and 0.13, respectively).

Conclusion:
Social determinants of health, as measured by traditional demographics and neighborhood-based composite indices, may not be associated with HAEC, recurrent HAEC, or HAEC severity in children. Use of a more patient-centric evaluation of social determinants may elucidate different outcomes.

ASSOCIATION OF SOCIAL DETERMINANTS OF HEALTH AND HIRSCHSPRUNG-ASSOCIATED ENTEROCOLITIS IN CHILDREN

Swathi Raikot, MBBS, Goeto Dantes, MD, Afrin Jahan, Nikolay Barykov, Jonathan Beus, MD, MS, Allison F. Linden, Hanna Alemayehu, MD

Table 1: Comparison of Social Determinants of Health and Hirschsprung-associated Enterocolitis (HAEC) Admissions.

Characteristic	Total, n=158	≥2 HAEC admissions, n=17 (11%)	0-1 HAEC admissions, n=141 (89%)	p-value
Race:				
Black	71 (45%)	5 (29%)	66 (47%)	0.20
White	69 (44%)	11 (65%)	58 (41%)	
Others	18 (11%)	1 (6%)	17 (12%)	
Ethnicity:				
Hispanic or Latino	14 (9%)	2 (12%)	12 (9%)	0.65
Non-Hispanic or Non-Latino	140 (91%)	15 (88%)	125 (91%)	
Insurance type:				
Medicaid	95 (60%)	14 (82%)	81 (57%)	0.15
Managed Care	57 (36%)	3 (18%)	54 (38%)	
Others	6 (4%)	0 (0%)	6 (4%)	
COI:**				
Very high	26 (17%)	2 (12%)	24 (18%)	0.73
High	28 (18%)	4 (24%)	24 (18%)	
Moderate	32 (21%)	4 (24%)	28 (20%)	
Low	34 (22%)	2 (12%)	32 (23%)	
Very low	34 (22%)	5 (29%)	29 (21%)	
SVI:**				
High	60 (39%)	7 (41%)	53 (39%)	0.47
Medium	43 (28%)	7 (41%)	36 (26%)	
Low	38 (25%)	2 (12%)	36 (26%)	
Unknown*	13 (8%)	1 (6%)	12 (9%)	
ADI:**				
High	49 (32%)	5 (29%)	44 (32%)	0.78
Medium	72 (47%)	10 (59%)	62 (45%)	
Low	20 (13%)	1 (6%)	19 (14%)	
Unknown*	13 (8%)	1 (6%)	12 (9%)	
Biopsy within 14 days prior to surgery (Yes)	100 (63%)	11 (65%)	89 (63%)	1.00
Ostomy creation (Yes)	28 (18%)	3 (18%)	25 (18%)	1.00

*Unknown category used when patient address could not be geocoded to extract a Census Tract or Block Group; For COI, ZIP-code was used to fill in unknown addresses. **Lower COI, higher ADI, and higher SVI levels represent higher greater social disadvantage.

MRI-GUIDED LAPAROSCOPIC ANORECTAL MALFORMATION REPAIR IS TRANSFERABLE TO A COMMUNITY-BASED HOSPITAL SETTING

Maria Kenner MD, Matthew Ralls MD, Justin Johnson, Chad Thebeau, Kelsey Powell, Micheal Walsh, Marcus Jarboe, MD

Despite meticulous surgical technique, the majority of patients who undergo anorectal malformation (ARM) repair face functional bowel issues. The introduction of laparoscopic assisted anoplasty has failed to improve outcomes. Regardless of technique, over 70% of school aged children experience constipation and/or fecal incontinence refractory to oral medication. These children require enema therapy to maintain social continence. Reports of MRI-guided needle placement with laparoscopic ARM repair have been recently published that suggest improved outcomes over traditional techniques. However, this study was a single center study performed at tertiary children's hospital with MRI equipped operating room. Further study is needed to confirm the preliminary findings of improved outcomes. More importantly, it is unclear whether this technique is transferable to other medical center environments to allow multicenter study. This study reports successful performance of the MRI-guided laparoscopic ARM repair in a community pediatric hospital environment.

METHODS

Several steps were necessary to transfer the MR guided technique. First, MR sequences had to be developed since a different MRI in a different facility was used. Second, different bore size and MRI flex coils necessitated a custom MRI compatible patient positioning device. Logistics of using a diagnostic MRI spatially separated from the operating room had to be solved and planned before the operation.

RESULTS

The process of patient transfer from anesthesia induction room to MR to OR were all successful. Sequence modification of 3D T2 sequence and T1 based real-time sequence produced good image quality at 3 frames per second. The modified patient positioning device successfully held the unique coils for image optimization and allowed safe lithotomy positioning within the bore. The single procedure was completed in a total of 321 minutes compared to 240 minutes of the previous experience.

CONCLUSIONS

MRI guided laparoscopic ARM can successfully be performed in a non-tertiary pediatric hospital environment.

ANTEGRADE CONTINENCE ENEMAS FOR PATIENTS WITH SPINA BIFIDA TO TREAT FECAL INCONTINENCE, A COMPARISON WITH PATIENTS WITH NON-NEUROGENIC CAUSES

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Purpose:
We sought to compare the rate and timing of becoming clean of stool with antegrade continence enemas (ACE) in patients with spina bifida (SB) vs patients with other causes of fecal incontinence as there is variability in offering ACE to SB patients due to perceived ineffectiveness.

Methods:
We performed a single institution retrospective study (1/2010-1/2023) of patients who underwent ACE surgery between age 3-31. SB patients were compared to those with non-neurogenic diagnoses including anorectal malformation (ARM), functional constipation (FC), and Hirschsprung disease (HD). The primary outcome was time to become "clean of stool" – defined as having ≥1 bowel movement/day, ≤1 stool accident/week, and no significant radiographic stool burden.

Results:
Of 175 total patients, 44 (24.0%) had SB, 42 (35.5%) had ARM, 62 (25.1%) had FC, and 27 (15.4%) had HD. SB patients were more likely to be female (54 v 33%, p = 0.01) and less likely to have any behavioral/psychiatric condition (16 vs 39%, p=0.005). Overall, 148 (85%) became clean of stool. SB patients became clean at the same rate as those with other diagnosis (88 vs 83%, p=0.5) and there was no difference in median time to cleanliness (2.2 vs 1.9 months, p=0.7). When comparing SB patients to individual diagnoses, there was still no difference in time to cleanliness [2.2 months (SB) vs 1.5 months (FC), 4.3 months (HD), 4.6 months (ARM), all p>0.05]. On Cox regression analysis, there were no demographic or clinical factors that were associated with the timing of becoming clean of stool.

Conclusion:
ACEs are similarly effective at providing patients with spina bifida the ability to achieve fecal continence compared to those with non-neurogenic causes of constipation and should be offered as a bowel management option for these patients through greater collaboration with spina bifida providers

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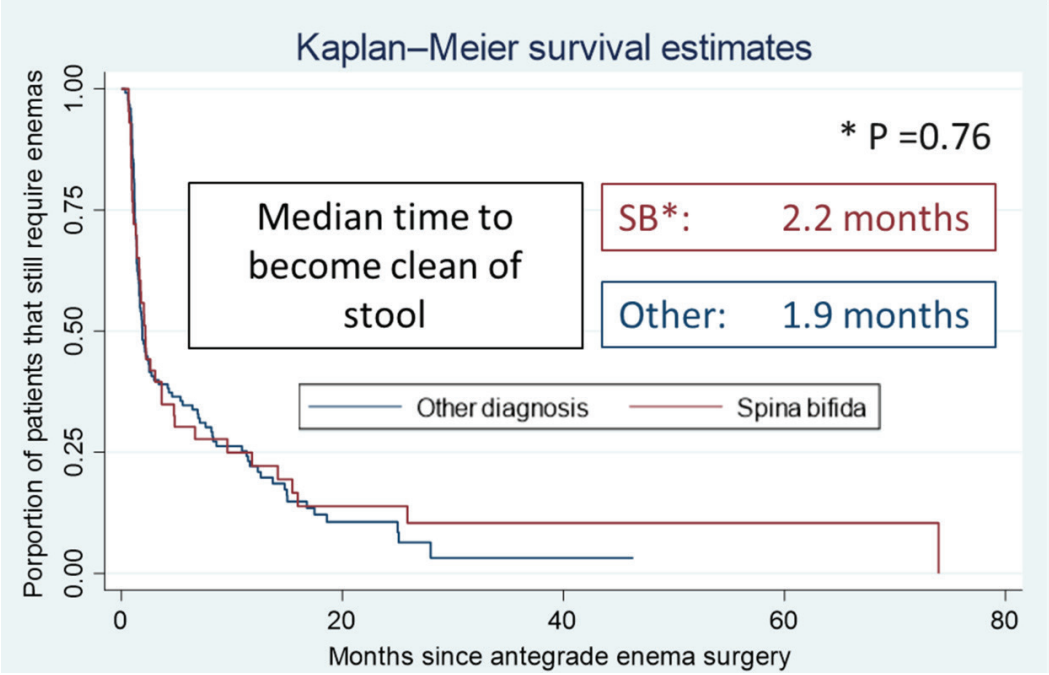
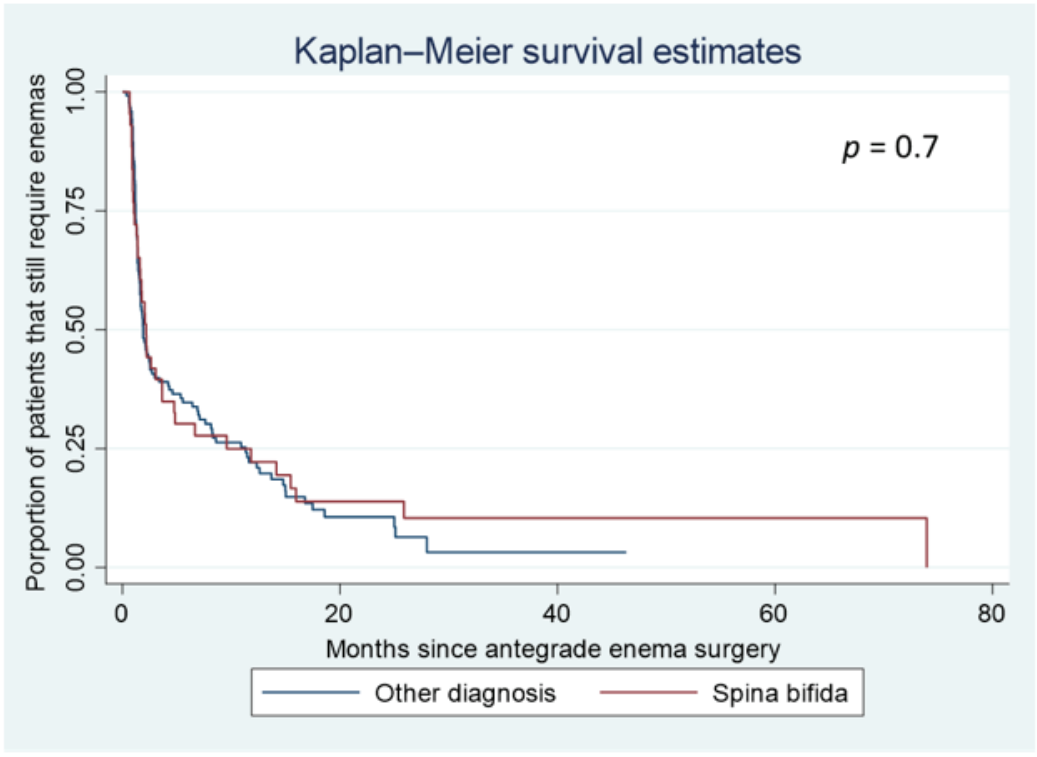


Figure 1: Kaplan-Meier curve for time to becoming clean of stool using antegrade continence enemas for patients with spina bifida (SB) vs other, non-neurogenic causes of constipation. Median time to achieve cleanliness for SB vs other diagnosis was 2.2 vs 1.9 months (p=0.7)



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DIFFERENCES IN CHILD OPPORTUNITY INDEX FOR PEDIATRIC PATIENTS WHO TRAVEL FOR SUBSPECIALTY COLORECTAL CARE

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Differences in Child Opportunity Index for pediatric patients who travel for subspecialty colorectal care

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Purpose: Social determinants of health are widely recognized as fundamental drivers of healthcare inequities. The childhood opportunity index (COI) measures the quality of resources available to a child based on the neighborhood in which they live and can be used to help understand community level impact on surgical outcomes. We aim to compare the COI of patients who traveled for treatment to those who did not travel for treatment.

Methods: This was a retrospective cohort study evaluating patients who were enrolled in the PCPLC data registry with a valid zip code. The COI was determined using zip code. Travel was defined as living at least 50 miles (as the crow flies) from the PCPLC center in which one received care. Demographic data between the group who traveled for treatment and those who did not travel was evaluated using chi square. The COI distribution between the groups was evaluated using the Cochran-armitage trend test.

Results: Of the 2633 patients who met inclusion criteria, 1535 patients did not require travel, and 1098 patients did require travel for care. When the overall COI was compared between groups, those who required travel were significantly more likely to be from low or moderate COI neighborhoods (20.5% and 26.4% respectively). Those who did not require travel were significantly more likely to be from very high COI communities (29.6%).

Conclusion: Differences were seen in distribution of COI between patients who traveled more than 50 miles compared to those patients who lived closer to the center in which they received specialty colorectal care. More work is needed to further clarify if those who travel are from urban or rural environments and if there are differences in COI between the two. Screening for resource support is essential to avoid healthcare disparities in these communities.