



# Assessing the benefit of reoperations in patients who suffer from fecal incontinence after repair of their anorectal malformation

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## ABSTRACT

**Background and Aim:** Patients with a previously repaired anorectal malformation (ARM) can suffer from complications which lead to incontinence. Reoperation can improve the anatomic result, but its impact on functional outcomes is unclear.

**Methods:** We performed a retrospective cohort study of patients with a previously repaired ARM who underwent redo PSARP at our Center and compared results at initial assessment and 12 months after redo.

**Results:** One hundred fifty-three patients underwent a redo PSARP for anoplasty mislocation ( $n=93$ , 61%), stricture ( $n=55$ , 36%), remnant of the original fistula ( $n=28$ , 18%), or rectal prolapse ( $n=11$ , 7%). Post-redo complications included stricture ( $n=33$ , 22%) and dehiscence ( $n=5$ , 3%). At 1-year post-redo, 75/153 (49%) are on laxatives only, of whom 57 (76%) are continent of stool. Of the remaining 78 (51%) patients, 61 (78%) are clean ( $\leq 1$  accident per week) on enemas. Interestingly, 16/79 (20%) of patients with expected poor continence potential were continent of stool on laxatives. Overall, 118/153 (77%) are clean after their redo. Quality of life (76.7 vs. 83.8,  $p=0.05$ ) and Baylor continence (29.2 vs. 17.7,  $p<0.0001$ ) scores improved.

**Conclusion:** Patients with fecal incontinence after an ARM repair can, with a reoperation, have their anatomy corrected which can restore continence for many, and improve their quality of life.

**Level of Evidence:** IV.

**Type of study:** Retrospective cohort study.

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Anorectal malformations (ARMs) are complex congenital malformations that result from the abnormal formation of the developing hindgut [1,2]. The care of this condition was revolutionized in 1982 with the description of the posterior sagittal anorectoplasty (PSARP) which for the first time afforded an approach for a precise anatomic reconstruction as well as stratification of the spectrum of anomalies [3]. The ultimate goals of the surgical repair are to disconnect the rectum from the urinary or genital tract if a fistula is present and create an adequately sized anal opening centered within the sphincter complex. These surgical principles maximize the chance the child will be continent of stool later in life [3,4]. Although the functional outcomes in ARM are generally good, particularly if the sacrum and spine are normal, a proportion of patients remain fecally incontinent, and their optimal management provides a challenge to the surgeon [5–9].

Fecal incontinence after PSARP is usually due to poor pelvic muscular and sensory development which can impair the continence mechanisms [1,10,11]. For a child with good continence potential, incontinence can

result from a technical complication related to their original reconstruction. Such complications include mislocation of the anoplasty, stricture, remnant of the original fistula (ROOF), or rectal prolapse, which might not be discovered until years later when the child attempts to potty train [12,13]. The clinician is then tasked with the decision of whether or not to offer the child a redo procedure to correct the anatomy. Traditionally, a redo operation was offered only to patients with good potential for bowel control [12], but that assessment is subjective and potentially limiting of candidates who might be able to translate improved anatomy into voluntary bowel control. Reoperation may indeed improve the anatomic result, but the ultimate impact of this intervention on continence is unclear. The aim of our study therefore was to assess the benefit of re-operative PSARP in patients suffering from fecal incontinence after previous ARM repair.

## 1. Methods

### 1.1. Cohort identification

We retrospectively identified all patients with a previously repaired ARM who were referred to us for evaluation of fecal incontinence between 2014 and 2018. Those who underwent redo a PSARP at our center

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were considered eligible for analysis. Patients with cloacal malformations were excluded from this study and will be considered in a separate report.

### 1.2. Intake process

All patients referred to our center undergo a formal intake process. The full intake has been detailed previously and is aimed to obtain data concerning the patient's medical and surgical history, fecal and urinary symptoms, and attempted interventions prior to the child's first clinic appointment [14,15]. Additionally, families (or the patient if >18 years of age) complete validated surveys which include the 23 item Baylor Continence Scale (Baylor) (if >4 years) [16], Cleveland Constipation Scoring System (Cleveland) (if >3 years) [17], Vancouver Symptoms Score (Vancouver) (if >3 years) [18], and the age-appropriate PedsQL Measurement Model (PedsQL) [19]. The intake process is performed using REDCap software [20].

### 1.3. Preoperative assessment

The preoperative assessment of both males and females who previously underwent ARM repair were conducted in a uniform manner and our algorithm can be seen in Figs. 1 (males) and 2 (females). Patients determined to have a normal spine and sacrum (sacral ratio  $\geq 0.70$ ) were considered to have "good" continence potential after reoperation while those with a spinal abnormality or underdeveloped sacrum (sacral ratio  $< 0.70$ ) were considered to have "poor" continence potential after reoperation.

### 1.4. Patient selection

If complications from the preceding PSARP were identified and determined to be contributing to the patient's incontinence, then a redo PSARP was offered. For patients with a mislocated anoplasty, reoperation was

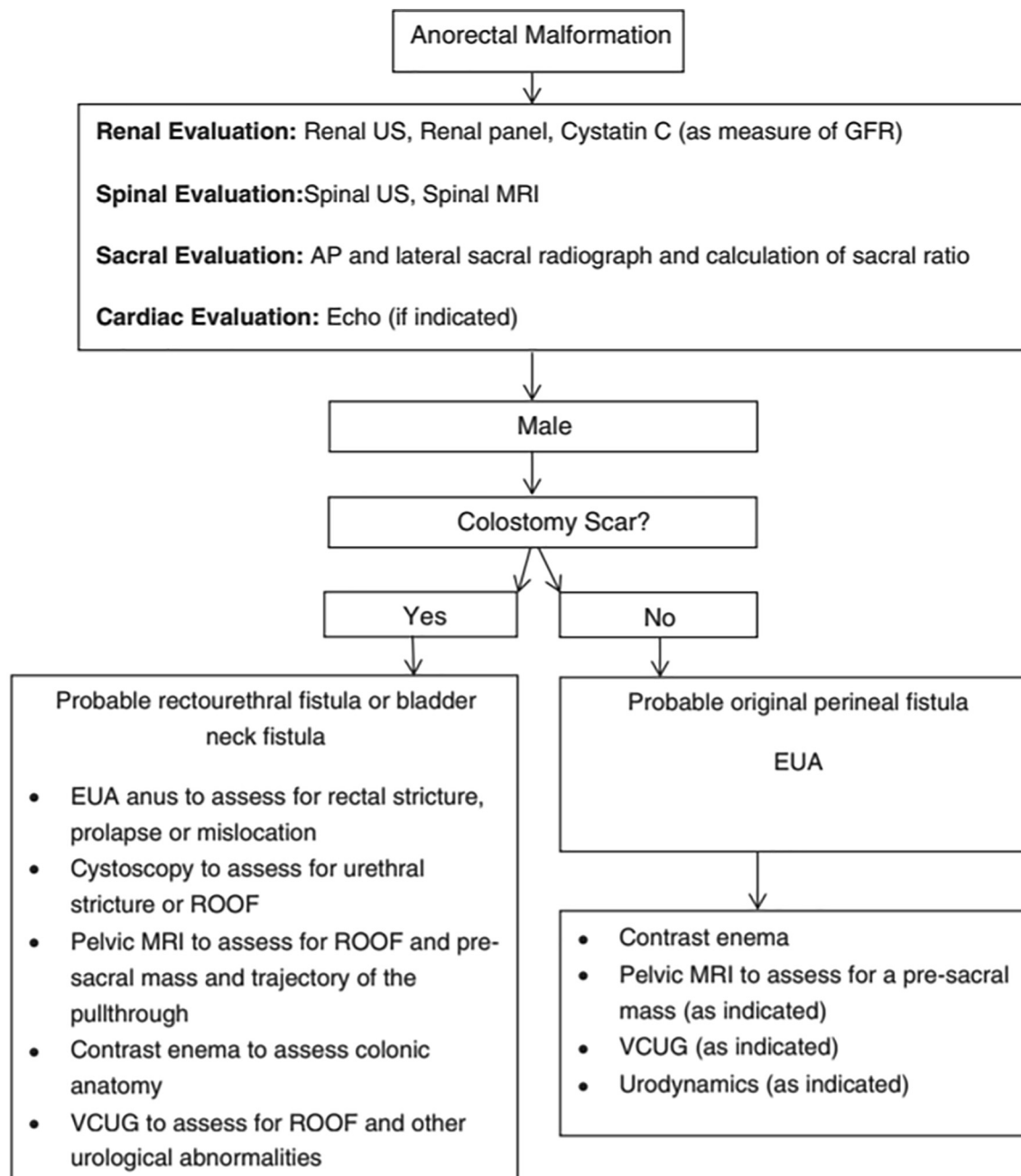


Fig. 1. Preoperative assessment of the male patient with a previously repaired ARM and incontinence [14].

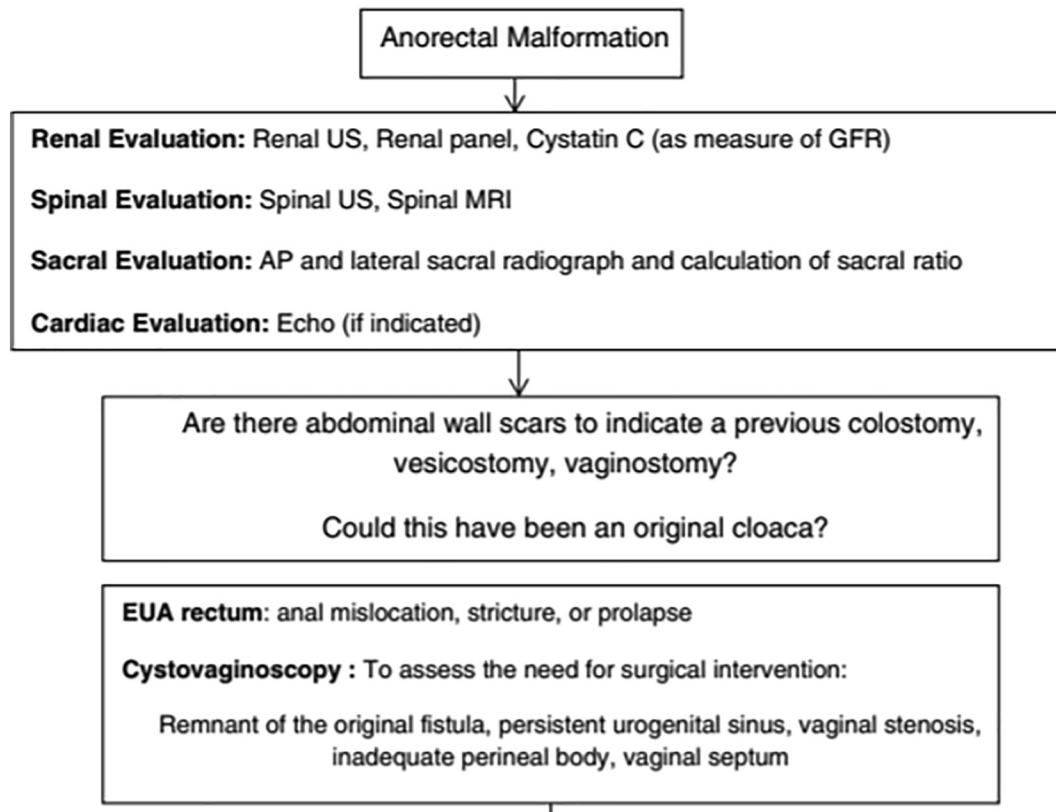


Fig. 2. Preoperative assessment of the female patient with a previously repaired ARM and incontinence [14].

offered if >50% of the anoplasty was outside of the sphincter complex. The offer for reoperation was made independent of spine or sacral status or the patient's developmental status. Reoperations were scheduled on an elective basis. If the patient was deemed to have an anatomically sound repair, the patient was started on either a laxative or enema regimen as part of our bowel management program, the choice of which is based on their underlying potential for bowel control. Those with a normal sacrum and spine were started on a laxative-based regimen while those with spine or sacral anomalies were started on an enema-based regimen.

Reoperations were done without a covering colostomy with the exception of 11 patients who came to us with a stoma. Patients were admitted one day before surgery and given GoLYTELY until stools are clear. Three doses of preoperative antibiotics are given. Clear liquids only were given postoperatively for five to seven days to avoid solid stool, and diet was advanced once perineal healing was confirmed [21]. No routine postoperative dilations were performed.

### 1.5. Postoperative assessment

After reoperation, patients are evaluated for follow-up at one, three, six, and twelve months. Patients are administered the Baylor, Cleveland, Vancouver, and PedsQL surveys at each time point, and a determination of the patient's bowel regimen and continence or cleanliness of stool is made. All postoperative assessments are performed using surveys administered using REDCap software either through a link e-mailed to the caregiver or in person using an iPad in the clinic, depending on caregiver preference [20]. To ensure validity, surveys filled out in the clinic were done privately without a member of the care team present.

### 1.6. Statistical analysis

Categorical variables were reported using frequencies and percentages, and continuous variables were assessed using medians and interquartile ranges (IQR). A paired t-test was performed to analyze changes

between the intake and 12 months postoperative Baylor, Cleveland, Vancouver, and PedsQL scores for patients with available data at both time points. The bowel regimen (laxatives or enemas) and the frequency of accidents at 12 months postoperatively were compared to the regimen and continence status at intake. A p-value of <0.05 was considered statistically significant. All statistical analyses for this study were performed using Stata 14.0 (StataCorp LP, College Station, TX, USA).

## 2. Results

Of 682 patients referred with fecal incontinence, 153 (22%) underwent a redo PSARP to correct their anatomy in an attempt to improve their continence. 511 (75%) had an anatomically correct repair and underwent a bowel management program which will be the subject of a future report, while 18 (3%) had a superficial stenosis that was amenable to a skin-level procedure (Heineke Mikulicz type anoplasty). Patients underwent redo PSARP at a median of 3.7 years of age (IQR 2.5, 5.9). The most common ARM subtypes of patients undergoing reoperation included vestibular fistula ( $n=20$ , 13%) and perineal fistula ( $n=17$ , 11%). Patients whose precise original anomaly could not be determined accounted for 50% ( $n=77$ ) of patients. The majority of patients had a normal spine ( $n=93$ , 61%), while 54 (35%) had a tethered cord or fatty filum and 6 had myelomeningocele. Eighty-nine (58%) patients had normal sacral development (lateral sacral ratio  $\geq 0.70$ ), 47 (31%) had a moderately developed sacrum (0.41–0.69), and the remaining 17 patients (11%) had a poorly developed sacrum ( $\leq 0.40$ ). 74 patients (48%) had both normal spinal and sacral development. Patient characteristics at the time of redo PSARP are shown in Table 1.

The most common indications for reoperation included anoplasty mislocation ( $n=93$ , 61%) [Fig. 3], stricture ( $n=55$ , 36%) [Fig. 4], ROOF ( $n=17$ , 11%), and rectal prolapse ( $n=11$ , 7%) [Fig. 5]. The surgical approach included a posterior sagittal incision only in 138 (90%) cases, with the remaining patients requiring the addition of laparotomy

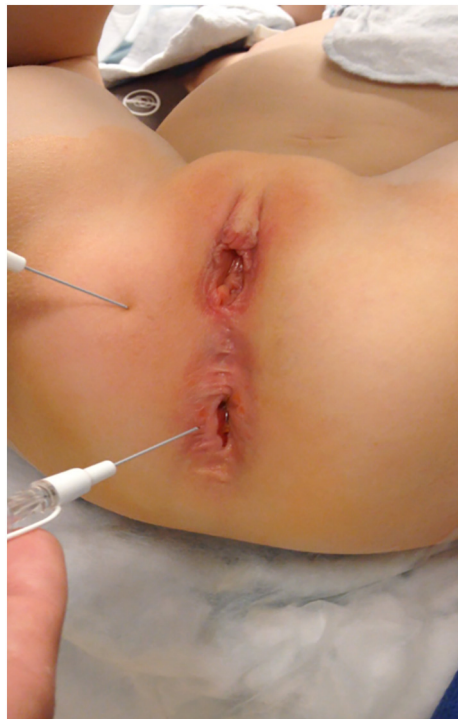
**Table 1**

Patient characteristics at the time of redo PSARP (N = 153).

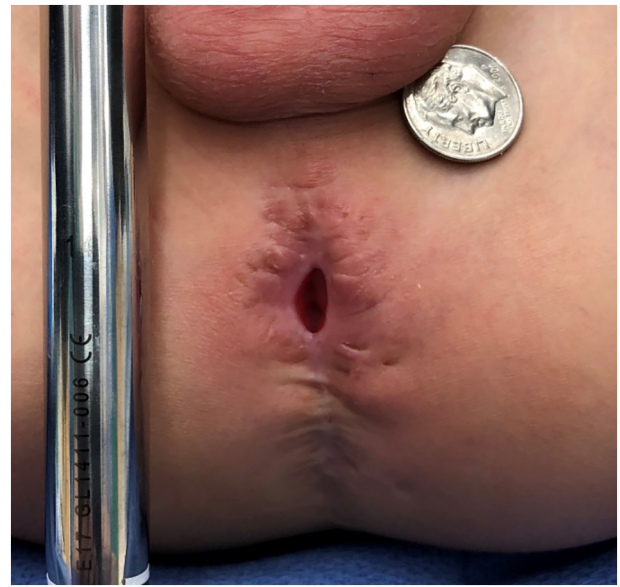
Male, n (%)	109 (71%)
Age at redo, years (median, IQR)	3.7 (2.5, 5.9)
ARM subtype	N (%)
Perineal fistula	17 (11%)
Vestibular fistula	20 (13%)
Vaginal fistula	2 (1%)
Bulbar fistula	6 (4%)
Prostatic fistula	9 (6%)
Bladder neck fistula	8 (5%)
Urethral fistula unknown level	40 (26%)
No fistula	10 (7%)
H-type fistula	1 (1%)
Rectal atresia	3 (2%)
Unknown	37 (24%)
Spinal status	
Normal/low conus	93 (61%)
Tethered cord/fatty filum	54 (35%)
Myelomeningocele	6 (4%)
Sacral ratio	
≥0.70	89 (58%)
0.41–0.69	47 (31%)
≤0.40	17 (11%)
Preoperative bowel regimen	
Enemas	77 (50%)
Retrograde	68 (88%)
Antegrade	9 (12%)
Laxatives	64 (42%)
Diverted	11 (7%)
Duration of follow up, years (median, IQR)	1.9 (0.9, 3.3)

(n = 11, 7%) or laparoscopy (n = 3, 2%). 83 patients (54%) underwent a combined procedure in addition to redo PSARP, such as creation of a Malone appendicostomy (n = 52, 34%) or colostomy revision (n = 6, 4%). The median duration of follow up was 1.9 years. These data are summarized in Table 2.

Table 3 details the surgical and functional outcomes of the patients. Postoperative complications were seen in 54 (35%) patients and included anal stricture (n = 37, 24%), rectal prolapse (n = 6, 4%), urinary



**Fig. 3.** Posterior mislocation of an anoplasty. (Arrow demonstrates the center of the sphincteric contraction.)



**Fig. 4.** Stricture of an anoplasty (Hegar dilator: size 14).

tract infection (n = 5, 3%), wound dehiscence (n = 5, 3%), and intraabdominal abscess (n = 1, 1%). Most anal strictures were managed with an out-patient skin-level procedure (n = 29, 78%) or dilation under anesthesia (n = 5, 14%). Three patients (5%) with deeper strictures required a subsequent redo PSARP. No patients required creation of a post-redo colostomy.

At 12 months after the redo procedure, 75 (49%) patients were on a laxative-based bowel regimen while the remaining 78 (51%) were managed with enemas. Three-quarters of patients on laxatives (n = 57, 76%) were continent of stool with regular voluntary bowel movements and one or fewer accidents per week. Of patients on enemas, 68 (87%) were on antegrade and 10 (13%) were on retrograde flushes. The majority of patients on enemas (n = 61, 67%) were clean on their flush regimen with less than one accident per week. Overall, 118 (77%) were completely clean of stool.

Of the 74 patients who were predicted to have excellent continence potential based on the presence of a normal sacrum and spine, 51 (69%)



**Fig. 5.** Circumferential rectal prolapse following PSARP.



**Table 2**

Surgical details of patients undergoing redo PSARP.

Indication for redo	
Mislocation	93 (61%)
Anterior	31 (33%)
Posterior	56 (60%)
Lateral	6 (6%)
Stricture	55 (36%)
Remnant of original fistula or acquired fistula	28 (18%)
Rectal prolapse	11 (7%)
Inadequate perineal body	6 (4%)
Dehiscence of anoplasty	5 (3%)
Surgical approach	
Posterior sagittal only	138 (90%)
Posterior sagittal plus laparotomy	11 (7%)
Posterior sagittal plus laparoscopy	3 (2%)
Combined procedure	83 (54%)
Antegrade enema access	52 (34%)
Excision of ROOF (with another redo indication)	17 (11%)
Colostomy revision	6 (4%)
Excision of presacral mass	1 (1%)
Additional *urologic or †gynecologic procedure	20 (13%)
#Other	5 (3%)

\* Hypospadias repair (3), circumcision (7), penoscrotal transposition (2), orchiopexy (1), urethroplasty (1), buried penis repair (1), chordae repair (1), bladder neck reconstruction (1), bladder neck closure (1), Mitrofanoff revision (1), ureteral reimplantation (1), suprapubic tube placement.

† Removal of vaginal septum (5), vaginal replacement with colon (1).

# Small bowel fistula repair (2), umbilical hernia repair (1), inguinal hernia repair (1), gastrocutaneous fistula takedown (1).

were on a laxative regimen 12 months after their reoperation. Of those, 41 (80%) were continent of stool and had normal, voluntary bowel movements. 23 patients were on an enema regimen, of whom 19 (83%) were clean of stool. 79 patients were predicted to have poor continence potential as a result of an underdeveloped sacrum or spinal anomaly. Of the 24 (30%) patients with poor continence potential on a laxative regimen, 16 (67%) were continent on a laxative regimen. Of the 55 patients with poor continence potential on an enema regimen, 42 (76%) were clean on an enema regimen. Fig. 6 details the bowel regimen and outcomes of these patients by their sacral and spinal status.

Table 4 details the change in validated scores from intake to 12 months postoperatively. Full survey data was completed in 137/153 (90%) patients for Peds QL, 123/153 (80%) patients for Vancouver, 92/153 (60%) patients for Cleveland, and 76/153 (48%) patients for Baylor. Statistically significant improvements were seen in the Baylor continence (29.2 to 17.7,  $p < 0.0001$ ) and PedsQL (76.7 vs. 83.8,  $p = 0.05$ ) scores. No statistical change was seen in the Cleveland (8.5 vs. 8.4,  $p = 0.94$ ) or Vancouver (12.1 vs. 10.7,  $p = 0.14$ ) scores.

**Table 3**

Functional outcomes of patients undergoing redo posterior sagittal anorectoplasty (N = 153).

Postoperative bowel regimen	
Laxatives	75 (49%)
Continent	57 (76%)
Not continent	18 (24%)
Enemas*	78 (51%)
Clean	61 (78%)
Not clean	17 (22%)
Complications	
Stricture	37 (24%)
Dilation under anesthesia	5 (14%)
Heineke-Mikulicz anoplasty	29 (78%)
Redo PSARP	3 (8%)
Prolapse	6 (4%)
Urinary tract infection	5 (3%)
Wound dehiscence	5 (3%)
Intraabdominal abscess	1 (1%)

\* Antegrade enemas (n = 68, 87%), retrograde enemas (n = 10, 13%).

### 3. Discussion

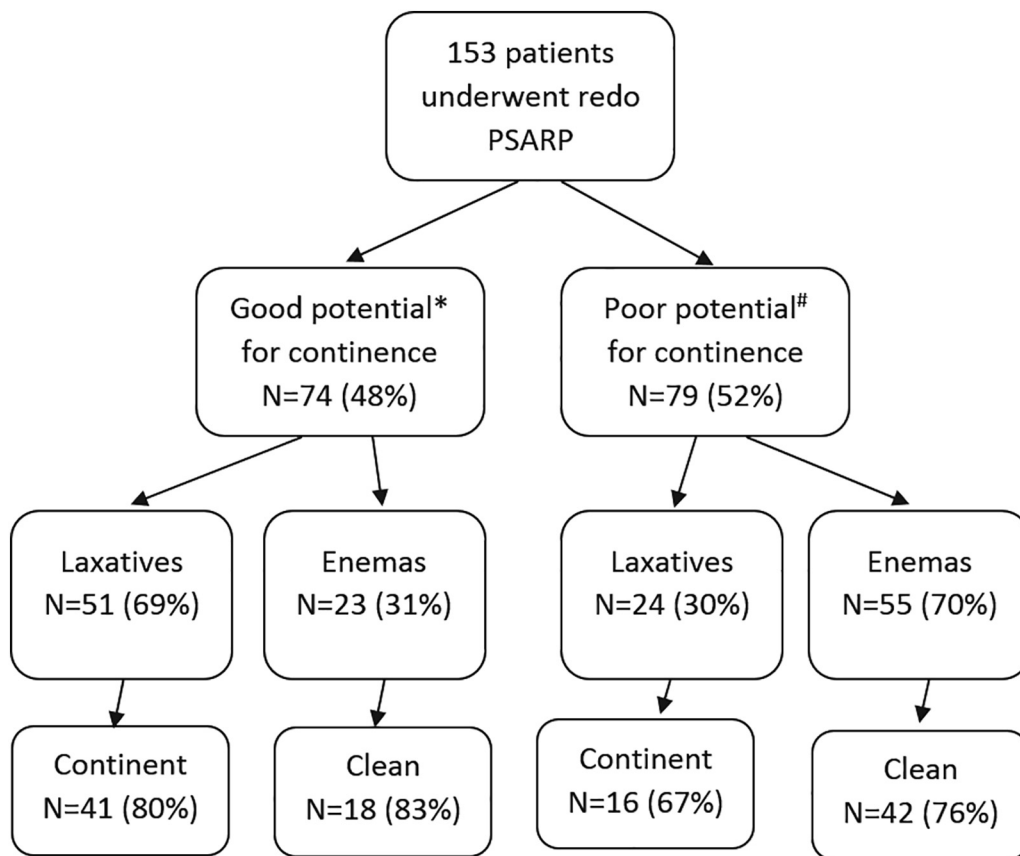
Soiling after ARM repair is a source of major morbidity in children born with an ARM [22–25]. These symptoms often do not present until years after the initial PSARP when it is discovered that the child is unable to successfully potty train. The clinician is then tasked to identify the cause of the patient's incontinence and determine whether the cause is ideally addressed with a medical (e.g. bowel management) or surgical solution. This is best ascertained with a detailed anatomic assessment such as the one outlined in Figs. 1, 2. The goal of the assessment is to identify whether an anatomic source for soiling is present, as a medical solution is less likely to be successful, for example in a patient with a strictured or mislocated anoplasty. Furthermore, attempts to treat a child with laxatives or enemas without a detailed anorectal examination may delay diagnosis of an anatomic cause of the incontinence. Thus any provider caring for such patients must first make an anatomic assessment of the surgical repair before treating the functional problem of soiling [14].

Traditionally, the decision to offer a reoperation was heavily influenced by the patient's perceived continence potential [12]. This dogma is rooted in the belief that patients with a poorly developed sacrum or an associated spinal anomaly are less likely to achieve future bowel control and thus an adequately-sized perineal opening to empty the colon once daily with an enema flush is sufficient to manage their stooling. Our data suggests that reoperations can be beneficial not only to patients with good predicted continence potential, but also for patients who might have previously been considered poor candidates for reoperation. In our study, we were surprised to find that 20% of such patients are fully continent on a laxative-based regimen. This suggests that the earlier dogma stating that reoperations should be offered only to patients with good potential for bowel control left many subjected to retrograde or antegrade enema flushes, never able to discover their continence potential. Study of the long-term outcomes of patients clean but still on enemas (76%) after reoperation is ongoing, and it is reasonable to expect that with improved anatomy many more will successfully transition to a laxative-based regimen as they progress through childhood and into early adulthood.

Despite an anatomically sound repair however, approximately one quarter of patients continue to have accidents at 12 months after their reoperation. These represent a significant bowel management challenge and remain the subject of our ongoing longitudinal study of our bowel management program.

Anoplasty mislocation was the most common indication for reoperation in our study and causes fecal incontinence by preventing the use of the external anal sphincter to close the anus. Proper placement of the anus within the sphincter complex therefore is critical in this patient population, as patients with an ARM are born without an anal canal and dentate line which is an important component in the continence mechanism of a normal child [26]. This is a potentially preventable complication that is avoided by clearly identifying and marking the location of the sphincters at the start of the operation, prior to starting the incision, and reconstructing the sphincter complex accurately around the pulled through rectum. Although in our study we noted the direction of the mislocation (e.g. anterior, posterior, or lateral), the degree of mislocation was not recorded, and it could be hypothesized that those with a greater degree of mislocation would derive greater functional benefit from a relocation of the anoplasty to its appropriate location within the sphincters. This represents an important area of future study.

Stricture of the anoplasty was another common indication for reoperation in our cohort. A thorough examination is needed in these cases because the depth of stricture will determine the appropriate repair. We previously reported our experience with a Heineke-Mikulicz-like anoplasty for superficial, skin-level strictures which can be performed safely and with durable results [27]. Deeper strictures are likely due to inadequate perfusion to the distal bowel or excessive anastomotic



**Fig. 6.** Twelve-month functional outcomes of patients after redo PSARP. \*Patients with good potential for continence include patients with a normally developed sacrum ( $SR \geq 0.70$ ) and no spinal anomaly. Patients considered as having poor potential for continence include those with sacral hypo-development ( $SR < 0.70$ ), an associated spinal anomaly (e.g. tethered cord, fatty filum, or myelomeningocele), or both.

tension require a formal reoperation in order to reach healthy bowel which can then be brought down to the skin in a tension-free manner.

Remnant of the original fistula (ROOF) is another common indication for reoperation. We recently described our experience with ROOFs in male patients with rectourethral fistula [28]. In addition to a ROOF, persistent or recurrent fistulae can occur and these result from failure to identify or sufficiently manage the rectal fistula at the primary operation. Acquired fistulas can be seen in female patients with rectovaginal fistula after repair of a recto-vestibular fistula, or male patients with a perineal fistula who suffer a urethral injury at the time of primary repair. In these cases, an understanding of the type and cause of the fistula is needed to ensure that the error that led to the persistent, recurrent, or acquired fistula is not repeated, including 1) proper distal colostogram to identify the anatomy of the rectourethral fistula in males and 2) surgical planning (e.g. PSARP vs. laparoscopy) [29]. Irrespective of the cause, ROOFs and fistulae can be effectively approached with a posterior sagittal incision and visualized by opening the posterior rectal wall to adequately visualize and ligate the fistula.

Rectal prolapse is seen following PSARPs, and is more common in more complex malformations [30]. Poor muscle tone and constipation

are believed to be factors that predispose the patient to this complication. The optimal management is dictated by the degree of prolapse. If relatively minor ( $< 5$  mm), a perineal-only resection of the prolapsing mucosa is adequate. However, in cases of more severe prolapse, a formal reoperation is needed to adequately secure the rectum to the posterior limit of the muscle complex. This was the indication for redo PSARP in 11 (7%) patients, none of whom later went on to develop recurrent prolapse. Even minor prolapses may impact bowel control and their repair should be considered.

Our results demonstrate that reoperations can be performed with acceptable surgical morbidity. Stricture was the most common complication seen after our reoperations and occurred in nearly one in four patients. This is higher than the up to 20% stricture rate reported in the literature for primary repair, although it should be noted that our cohort of patients represents a very different patient population [31]. Additionally, no routine dilations were performed in this patient cohort. The likely cause for the higher rate of stricture is damage to the intramural blood supply resulting from a previous rectal dissection. All patients in our cohort had undergone at least one previous attempt at repair. In many cases, the precise surgical history could not be determined. The most important determination when faced with a patient with an anal stricture is to assess the depth, which determines the appropriate management. Superficial, skin-level strictures can be managed with a skin-level procedure and were successful in doing this in over three-quarters of post-operative strictures in our cohort, with a minority successfully managed with an anal dilation under anesthesia [27]. Deeper strictures however require a formal redo procedure to reach healthy bowel, which was needed in three cases.

Wound dehiscence was also rare complication in this cohort at 3%. It should be noted that most patients underwent reoperation without a

**Table 4**  
Quality of life scores in patients at intake and 12 months after redo PSARP.

	Intake	12 Months	95% CI	p
	Mean	Mean		
Peds QL (n = 137)	76.7	83.8	0.4, 13.4	<b>0.048</b>
Baylor (n = 76)	29.2	17.7	-15.0, -7.9	<b>&lt;0.0001</b>
Cleveland (n = 92)	8.5	8.4	-1.4, 1.3	0.94
Vancouver (n = 123)	12.1	10.7	-4.2, 1.3	0.14

diverting ostomy with the exception of 11 patients who arrived with a stoma in place. None of the cases of wound dehiscence required subsequent diversion of the fecal stream.

The decision to perform a simultaneous Malone appendicostomy at the time of redo PSARP should be individualized for each patient. This was performed in nearly one third of patients in our series, and should be considered in patients with poor anatomic predictors of continence, those who are likely to require long-term enema treatment, or who may require a short duration of flushes before transitioning to laxatives but do not tolerate retrograde enemas. In general, our threshold to perform a Malone is lower if the patient requires an abdominal component of their operation. Prior to utilizing the appendix for an appendicostomy however, the patient's future urologic needs should be considered, as a combined approach with colleagues in urology permits sharing of the appendix for bladder reconstruction if needed [32]. Also helpful is to start the patient after the redo on antegrade flushes. This idea affords them the opportunity to practice holding in the liquid and releasing it on command. If they can improve their sphincter control this way, it is a good sign for success in fully transitioning them off of flushes to their own voluntary bowel control.

Invariably there will be patients in whom a reoperation might not offer significant improvement in continence potential or quality of life. An example of this would be patients with severe cognitive or developmental delay. In this complex group of patients, a decision might be made to not perform a redo operation if a minor anatomic complication is identified (e.g. mislocation just outside the sphincter complex or a mild stricture). However, at present there are no clear guidelines on which patients with developmental or cognitive delays would most benefit from a reoperation for an anatomic complication, although this is an important area of future research. An honest conversation with caregivers about their child's ability to be continent of stool and the role of surgery in achieving that goal is vital. These patients might instead derive more benefit from a bowel management program to empty the colon on a predictable schedule without the surgical morbidity of a reoperation. For such patients, only an anal opening through which to flush the colon is needed.

The main limitation of our study is that many of the children in our cohort were not of potty training age at the time of their repair, and thus it is unknown whether some of these patients with relatively minor complications would have been able to overcome their imperfect anatomy and achieve continence of stool without the need for a redo procedure. We chose to do redos in such patients in order to give them the best potential to achieve success during the potty training process. Furthermore, all patients were placed on an aggressive bowel management program with either a laxative or enema-based regimen aimed to optimize the patient's functional outcome. The degree to which bowel management contributed to the functional improvements seen cannot be known, and whether or not children with more minor complications (e.g. a slight mislocation) might achieve an adequate functional status is also not known. Without a pure control group (e.g. a cohort of patients with known complication after PSARP who do not undergo corrective surgery), the true benefit of reoperations in these patients cannot be known for certain. Importantly, all patients evaluated, were operated on at a high volume referral center with experience in redo ARM surgery, thus potentially limiting generalizability of these results. Despite these limitations, we are encouraged by the improvement in continence, cleanliness, and quality of life that followed from this intervention.

#### 4. Conclusion

The results of our study demonstrate that a redo procedure in patients with a previously repaired ARM along with intensive bowel management can significantly improve both fecal continence and cleanliness for stool

in a majority of patients. Furthermore, we show that a significant improvement can be seen in validated fecal incontinence and quality of life scores as a result of these interventions.

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