

Reoperative Surgery for Anorectal Anomalies

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Complications occur during the repair of anorectal malformations relatively frequently. Unfortunately, these complications are often preventable. Furthermore, the consequences of these complications are significant. Not only do patients experience unnecessary pain and suffering, but a secondary operation always renders less optimal functional results. A 20-year experience in the care of children with anorectal malformations was retrospectively analyzed. Patients who previously underwent surgical repair at other institutions, and subsequently required secondary surgery by the primary author were evaluated; 334 patients were identified. Reasons for reoperation included fecal incontinence in 77 patients; dehiscence and retraction in 96; recto-genito-urinary fistulae in 55; persistent urogenital sinus in 31 cloaca patients; acquired vaginal atresia in 21; acquired urethral atresia in 9; posterior urethral diverticulum in 20; and overflow pseudo incontinence in 25 patients. Except for fecal incontinence, all other complications are considered preventable. The source of the complications in almost all other settings are technical errors at the time of the primary repair. Recommendations are presented to help prevent these complications, and suggestions are made on how to treat them when they occur.

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IN SPITE of the technical advances in the surgical repair of anorectal malformations that have occurred over the last 20 years, operations to revise, redo, or otherwise repair the results of initial procedures still are not that uncommon. Surprisingly, this phenomenon is not isolated to the group of patients with very complex malformations. Instead, it spans the entire spectrum of malformations seen.

Reoperative surgery may be considered for several reasons. Suboptimal results as far as bowel and urinary control may have been achieved after the first operation, and a surgeon may wish to attempt to improve on the results. Other patients may have suffered significant, sometimes catastrophic, complications because of technical misadventures and require revisional surgery to alleviate pain, discomfort, and other sequelae. However, it is clear that a patient's best chance for a good functional result is when the proper operation is performed during the first definitive procedure, and complications are avoided.¹ This is especially true in those patients born with a good-prognosis defect. It is unfortunate when these patients end up with fecal or urinary incontinence resulting from avoidable complications of the surgical repair. We do recognize, however, that complications will occur, and patients will continue to require revisional surgery. The purpose of this retrospective review is not only to detail the best way to approach these complications, but to also review some of the reasons they occur and how they can be prevented.

MATERIALS AND METHODS

A retrospective review of our experience with patients with anorectal malformations who underwent primary repair at other Institutions and then required a secondary procedure at our institution was performed. Personal interviews as well as telephone conferences allowed us to determine the current condition of each patient. The operative reports from the primary operations were reviewed in an effort to determine the specific type of malformation with which the patient was originally born. A correlation between the description of the original operation and the anatomic findings during the secondary procedure was sought to try to elucidate the cause of the complication. The final functional results in all patients were evaluated with emphasis on the presence or absence of voluntary bowel movements, soiling of the underwear, constipation, and urinary control.

From 1980 through April 2002, we performed surgery on 1,450 patients who had anorectal malformations. Of these, 334 were secondary procedures (Table 1). These 334 patients were divided into 3 main groups.

Group A includes 77 patients with fecal incontinence who underwent posterior sagittal anorectoplasty (PSARP), in an attempt to improve their bowel control. Group B includes 232 patients who sustained some sort of a catastrophic complication during or shortly after the first operation and required a complete reoperation. This group of patients was subdivided according to the specific type of complication that occurred. Group B1 includes 96 patients with dehiscence, retraction, infection, or acquired atresia of the rectum. Group B2 includes 55 patients with recto-genitourinary fistula complications. These complications include persistent fistulae (22 patients), in which the original rectourethral fistula remained untouched, even when the rectum was repaired; recurrent fistulae (10 patients), in which the surgeon repaired the fistula but it reopened; acquired rectourethral fistulae (5 patients), in which the fistula was created during the repair of a benign malformation; acquired rectovaginal fistula (18 patients), created during an attempted failed repair of a rectovestibular fistula.

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Table 1. Reoperative Surgery for Anorectal Malformations

(Total Series, 1,450 Patients)	
Primary, 1,116	
Secondary, 334	
Group A, 77 patients, fecally incontinent	
Group B, 232 patients, catastrophic complications	
B1, 96 patients, dehiscence, retraction, infection, acquired atresia	
B2, 55 patients, rectogenito urinary fistula	
B3, 31 patients, persistent urogenital sinus (cloaca patients)	
B4, 21 patients, acquired vaginal atresia (cloaca patients)	
B5, 9 patients, acquired urethral atresia	
B6, 20 patients, posterior urethral diverticulum	
Group C, 25 patients, pseudoincontinent patients	
12 sigmoid resection	
13 medical management	

Group B3 includes 31 patients with persistent urogenital sinus. These patients were all born with a cloaca and underwent an operation in which the rectal component of the malformation was repaired, but the urogenital sinus was ignored. Group B4 includes 21 patients born with a cloaca, who underwent an attempted repair, and suffered from an acquired vaginal atresia. Group B5 includes 9 patients with acquired urethral atresia. Group B6 includes 20 male patients with a posterior urethral diverticulum. These complications occur when a retained portion of the rectum is left attached to the posterior urethra.

Group C includes 25 patients referred to our Institution because of "fecal incontinence." Further evaluation proved that they were actually severely constipated, chronically impacted, and suffered from pseudoincontinence. All these patients had several factors in common. All were born with a malformation with good functional prognosis, and all underwent a technically correct, successful operation. Postoperatively, they all had severe constipation, and had megasigmoid and chronic fecal impaction. Because they never received appropriate treatment for their constipation, pseudoincontinence in all of them developed. Adequate treatment of their constipation, with or without a sigmoid resection, rendered them fecally continent.

Indications for Surgery

Group A. During the first 5 years of our experience, reoperative surgery was performed on every patient we evaluated who underwent a repair at another institution and subsequently had fecal incontinence. During those years, we hoped that the "new posterior sagittal approach" would give these patients an opportunity to recover fecal control. When the results were evaluated^{2,3} only 30% of those patients experienced a significant improvement. Therefore, the indications for surgery were modified. Currently, reoperation for fecal incontinence is recommended only for patients with very

special criteria. They should have been born with a malformation associated with a good prognosis, the rectosigmoid should be intact, and the sacrum normal and the sphincter mechanism should be intact. The rectal location can be evaluated with magnetic resonance imaging (MRI), and if it is significantly mislocated, a secondary operation can be offered.

Group B. All patients that suffered catastrophic complications were accepted for surgery for obvious reasons.

Group C. All patients in group C underwent a "laxative test" to determine if they were fecally continent. Large volume enemas were administered until the patient's colon was clean. Daily laxatives then were administered, increasing the amount each day until the amount necessary to produce colonic evacuation was determined. A plain abdominal x-ray was obtained every day to assess the colonic emptying. If the patient showed the capacity to feel the stool in the rectum, reach the bathroom, have voluntary bowel movements, and remain clean every day, the patient was considered continent. The patient then was offered the option of continuing the treatment with large quantities of laxative for an indefinite period or a sigmoid resection to make the constipation more manageable and thereby decreasing the laxative requirement. Twelve patients elected to have a sigmoid resection, and the remaining 13 decided to continue on laxatives.

Operative Approach

All patients except for Group C were approached posterior sagittally.

Group A. The first 62 patients were operated on in 3 stages including a colostomy, posterior sagittal reoperation, and colostomy closure. In the last 15 cases, all were operated on in a single stage without a protective colostomy. All were admitted to the hospital the day before surgery and underwent a mechanical bowel preparation utilizing a solution of ethylene glycol and balanced electrolytes (Golytely [Golytely-PEG-3350 & Electrolytes for oral solution. Braintree Laboratories Inc, Braintree, MA]) administered via a nasogastric tube at a rate of 25 mL/kg/h until the bowel was clean. In the operating room, a central venous line was inserted, the patients received parenteral nutrition, and were kept fasting for 7 to 10 days.

The rectum was approached posteriorly.²⁻⁴ Multiple silk stitches were placed at the mucocutaneous margin to apply uniform traction to facilitate the dissection and mobilization of the rectum. A full rectal dissection and mobilization was performed, staying as close as possible to the bowel wall but avoiding injury.

The limits of the sphincters, including the parasagittal fibers, muscle complex, and levator muscle were deter-

Table 2. Results

Complication and Surgical Indication	No.	Voluntary Bowel Movement (%)	Soiling (%)	Totally Continent (%)	Urinary Continent (%)
A, Incontinent	77	52	85	15	91
B1, Dehiscence, retraction	96	67	62	19	92
B2, Fistulae complications	55	71	63	32	78
Rectourinary Fistulae					
Persistent	22	64	43	43	62
Recurrent	10	83	86	13	100
Acquired	5	67	67	50	100
Rectovaginal acquired	18	75	71	31	79
B3, Persistent U/G sinus	31	58	77	25	76
B4, Acquired vaginal atresia	21	67	40	50	83
B5, Acquired urethral atresia	9	67	75	33	None
B6, Posterior urethral diverticula	20	33	100	None	77

mined by electrical stimulation. The rectum was repositioned within the limits of the sphincters. In many cases, the patient was found to have had colon, and not rectum pulled down. The presence of a mesentery attached to the bowel confirmed this. In those cases, the mesenteric fat was trimmed from the last few centimeters of the rectum to achieve direct contact between the sphincter mechanism and the colonic wall. An anoplasty then was performed within the limits of the sphincter mechanism.

Group B. These patients were also approached posterior sagittally. In cases of retraction, dehiscence, and acquired atresia, the rectum was usually located somewhere high in the pelvis surrounded by a significant amount of fibrous tissue. Multiple 6-0 silk sutures were placed in the rectal wall to exert uniform traction and facilitate a circumferential dissection of the rectum, again trying to stay as close as possible to the rectal wall without injuring it. Bands and extrinsic vessels surrounding the rectum were divided and cauterized circumferentially until enough rectal length was gained as to place the rectum within the limits of the sphincter mechanism.

Short ringlike rectal strictures were treated with a Heineke-Mikulicz type of plasty; strictures longer than 1 cm were resected. The rectum was mobilized as previously described, until the fibrotic portion could be removed and a fresh nonscarred portion of the rectum could be pulled down, creating a new anus.

Fistulous complications were also approached posterior sagittally. The posterior rectal wall was opened, and the fistulae was identified and closed. The rectum then was separated from the urinary tract or the vagina and was mobilized as previously described so as to be sure that a completely normal anterior rectal wall was left in front of the urethral or vaginal suture line.

Those patients with persistent urogenital sinus also were approached posterior sagittally. The rectum was dissected completely and reflected out of the way. This allowed exposure of the urogenital sinus, which was repaired using the same technique that is used during the

treatment of a cloaca.^{5,6} In the first 23 cases, a separation of the vagina from the urinary tract was performed. The urethra was reconstructed, and the vagina was then mobilized, reconstructed, and placed behind the urethra. In the last 8 patients, we utilized the total urogenital mobilization technique to repair the urogenital sinus.⁷ This technique significantly shortens the operative time and results in a much better anatomic reconstruction. Those patients with acquired vaginal atresia were treated using the same surgical approach as for persistent urogenital sinus.

For those patients with acquired urethral atresia, the rectum was mobilized to expose the urethral area. Both urethral ends were identified, dissected, and mobilized enough to perform a tension-free end-to-end anastomosis.

The same approach was utilized to treat those patients with posterior urethral diverticulae. After a posterior sagittal incision, the rectum was mobilized to expose the posterior aspect of the urinary tract. The diverticulum was identified and dissected all the way down to the urethral site. The diverticulum was separated from the urethra in the same manner as in primary repair of anorectal anomalies.⁸ The urethra was closed, and the diverticulum was resected.

Group C. Those patients with megasigmoid and pseudo incontinence who elected to undergo a sigmoid resection were treated with a standard colon resection. The operation involved resecting the most dilated portion of the sigmoid.⁹ An anastomosis was created between the descending, normal caliber descending colon and the rectum.

RESULTS

The detailed results are presented in Table 2. In group A (reoperations for fecal incontinence), 52% of all patients had voluntary bowel movements. However, 85% of them soiled their underwear occasionally; therefore, only 15% of them were totally continent (had voluntary

bowel movements and never soiled). In 91% of all these patients urinary control developed.

In group B (reoperations for patients who suffered catastrophic complications), between 64% and 83% (depending on the specific group of these patients) had voluntary bowel movements. Between 43% and 86% of the patients soiled; therefore, the totally continent fraction was only between 13% and 43% depending on the specific group. Most patients had good urinary control except for the group that had a persistent rectourinary fistula; only 62% of them achieved urinary continence.

In group B3, 58% of cloaca patients with a persistent urogenital sinus had voluntary bowel movements, 77% of them soiled, and, therefore, only 25% of them were totally continent; 76% of them had urinary control.

In group B4, 67% of patients with an acquired vaginal atresia had voluntary bowel movements, 40% of them soiled, 50% of them were totally continent, and 83% of them had urinary control.

In group B5, 67% of patients with acquired urethral atresia had voluntary bowel movements, 75% of them soiled, 33% of them were totally continent, and none of them had urinary control.

In group B6, 33% of patients with posterior urethral diverticulum had voluntary bowel movements and 100% of them soiled; therefore, there were no totally continent patients. Seventy-seven percent of them had urinary control.

In Group C, all 25 patients had voluntary bowel movements. They occasionally suffer soiling when they are not compliant with their laxative regimen. The 12 operated patients significantly reduced the amount of laxative necessary for them to have bowel control. Thirteen non-operated patients still receive a large amount of laxatives to avoid impaction and remain continent.

DISCUSSION

The results shown in Table 2 in terms of voluntary bowel movements and totally continent patients, as well as urinary control, seem to be notoriously worse than those obtained in primary cases.¹ To have a more objective view of this it would be necessary to know the specific anatomic diagnosis of each one of these patients to compare with a similar group operated on primarily. We were unable to do this because many times it was impossible to find out the type of specific birth defect of the patient.

The number of patients that require a reoperation for fecal incontinence has decreased significantly over the years. We suspect that this is because of the increased use of the posterior sagittal approach, which provides superior exposure and prevents the complete mislocation of the rectum that was seen when the procedure was done blindly.

Years ago, many patients also underwent abdominal perineal pull-throughs with endorectal dissections of the rectosigmoid.¹⁰ This procedure essentially resulted in loss of the rectosigmoid. These patients do not suffer from constipation. Instead, they suffer from increased colonic motility and a tendency to diarrhea. It took us several years to recognize this specific group of patients, and, today, revisional surgery is not offered to them, because it is clear that they never regain bowel control. Fortunately, endorectal pull-throughs for anorectal malformations are no longer done, and it is rather unusual to see these patients.

In addition to the above group, those patients born with poor-prognosis defects and fecal incontinence are also considered inappropriate candidates for reoperation. These patients typically have an abnormal sacrum, flat perineum, and poor sphincters. There usually is evidence that they were born with a recto prostatic, a recto bladder neck fistula, or a cloaca with a common channel longer than 3 cm. Their sacral ratio is almost always less than 0.4. We do not reoperate on these patients, even if they have a completely mislocated rectum, because they do not improve after reoperation. Instead, they are offered a bowel management program¹¹ to prevent soiling and to keep them completely clean. The parents are taught to clean the colon of the patient every day, usually with an enema or colonic irrigation. It takes one week to implement the bowel management program. The patients come to the hospital every day, and a plain abdominal radiograph is taken to evaluate the amount of stool left in the colon after the enema. The volume and concentration of the enema is adjusted by trial and error, until the enema that is capable of cleaning the colon is found. The technique of administering the enema is demonstrated by an expert nurse. This program is successful in 95% of the patients.¹¹

When revisional surgery for fecal incontinence is offered, the likelihood of the patient regaining bowel control is reviewed with the family. Even with those patients who are expected to improve, the bowel management program is implemented before surgery. If it turns out that the patient does not improve enough to avoid enemas, the original bowel management is reinstituted.

When the patient complains about the administration of an enema and the intrusion on his privacy from the parents giving enemas, and if the patient is old enough to become independent, the Malone procedure (continent appendicostomy or continent neo-appendicostomy, when they do not have an appendix) is offered.^{12,13} We *only* offer that operation when we have shown that the bowel management works, because these operations only represent a different route of administration of an enema. Most likely, if enemas do not work when given through

the rectum, they will not work when given from above in an antegrade fashion.

The most rewarding group of patients to treat are those who had suffered previous catastrophic complications, because they enjoyed the greatest benefit from reoperation. The closure of fistulas, removal of diverticulums, and the opening of an acquired atretic urethra, rectum, or vagina resulted in dramatic improvements in the patients quality of life. The functional results were also better because we usually were dealing with patients who had better potential for bowel control.

The existence of this group of patients with catastrophic complications highlights the need for pediatric surgeons, as a group, to improve the surgical technique and approach used to repair anorectal anomalies. It is our contention that these complications can be avoided by a meticulous and educated operative technique.

Based on our anatomic findings during these reoperations, we speculate that retraction, dehiscence, and acquired rectal atresia, were most likely caused by a poor technique used to mobilize the rectum. The rectum, when seen posterior sagittally, is covered by a very characteristic white fascia that provides support as well as contains vessels to the rectum. This fascia must be divided, and the surgeon must dissect the rectum remaining as close as possible to the rectal wall. Uniform traction provided by multiple silk sutures is imperative to facilitate the dissection. Bands and extrinsic rectal blood supply must be divided to gain rectal length. We have learned that the intramural blood supply of the rectum is excellent; we can dissect and gain significant length of rectum provided the rectal wall is not injured. We believe that the most likely cause for difficulty in dissection of the rectum is working outside the fascia. Alternatively, dissection too close to the rectum injures the rectal wall, interferes with the intramural blood supply, and provokes ischemia. The result of all this is an incomplete mobilization, rectal ischemia, and a rectal-skin anastomosis under tension, which may explain most of these complications.

We speculate that rectal strictures are also most likely caused by ischemia of the distal part of the rectum. When the rectum is correctly mobilized, and the blood supply kept intact, it is extremely unlikely to see anal stenosis. A few patients of ours who were operated on primarily and failed to follow our protocol of dilatations, returned to our clinic months after their operation with strictures. These patients had a thin fibrotic ring in the area of the anoplasty, which was easy to treat either with an anoplasty or dilatations. When one finds a rather long narrow stricture, most likely it is caused by rectal ischemia.

Some surgeons do not like to follow a protocol of anal dilatations. To avoid painful maneuvers to the patient, they follow a specific plan consisting of taking the patient to the operating room every week and, under anes-

thesia, performing forceful dilatations. We think that those dilatations actually provoke lacerations in the anal verge, which then heal with scarring, only to be reopened during the next forceful dilatation. This creates an intractable ring of fibrosis.

Persistent rectourethral fistulae occurred in patients who were born with a rectourethral bulbar fistula and underwent a repair that did not address the fistula. We speculate that surgeons following the old diagnostic approach performed an invertogram and found the bubble of rectal air close to the skin. This may have led to an approach through the perineum, with identification of the rectum and subsequent pull-through and anoplasty. Because the surgeon was completely unaware of the low rectourethral bulbar fistula, it was not repaired.

Recurrent recto-urethral fistulae may result if the fistula is closed, but the rectum is not mobilized adequately, leaving the anterior wall under tension. A dehiscence of the anterior rectal wall may explain the recurrence of the fistula. Also, leaving sutures in the rectum adjacent to the sutures in the urethra may lead to formation of a recurrent fistula. Consequently, an injured rectum that requires a repair to the anterior wall may create a situation for the development of a recurrent fistula. The same explanation may apply to rectovaginal fistulae.

Acquired rectourethral fistulae occurred in boys that were born with rectoperineal fistulae. These patients underwent their first operation without the benefit of a Foley catheter in the urethra. During the mobilization of the anterior wall of the rectum, an unrecognized urethral injury occurs, and, if not mobilized to leave normal rectal wall in front of the urethral injury, an acquired rectourethral fistula will form.

More disturbing is the group of cloaca patients with persistent urogenital sinus. All these patients came to us with an original diagnosis of "rectovaginal fistula." The word *cloaca* was never mentioned in the operative report. Only the rectal component of the malformation was repaired and the urogenital sinus was left unattended. In our experience, a true congenital rectovaginal fistula is an extremely unusual defect.¹ Yet, in the literature from previous years, we have found frequent mention of this malformation and very little reference to cloacas.¹⁴⁻²⁰ In the recent literature, there is an increase in the diagnosis of cloacas and less mention of rectovaginal fistulae, which, we think, reflects the improved understanding of the true anatomy of these lesions.²¹⁻²⁴

Acquired vaginal atresia is a problem that may occur when the vagina is separated from the urethra during the repair of cloacas. The separation of the vagina from the urinary tract is not an easy maneuver. The vagina becomes devascularized very easily and, as a consequence, patients get ischemic vaginal atresia. This happened in 3 of our own cases before the use of total urogenital mobilization.

The group of patients with posterior urethral diverticulum is a very interesting one. All these patients were born with a rectourethral bulbar fistula. All of them were operated on transabdominally. It is easy to understand that the surgeon was unable to reach the fistula site through the abdomen. Consequently, the rectum was amputated, leaving a piece of rectum attached to the urethra. The patients, although initially asymptomatic, after years had symptoms such as passing mucous through the urethra, orchioepididimitis, urinary tract infection, and urinary pseudoincontinence. In addition, we saw one of these patients who, after 30 years, had adenocarcinoma in the piece of rectum left attached to the urethra.

Finally, the group of patients suffering from pseudo incontinence is extremely important. All of the pediatric surgical community must be aware of this problem. Some of these patients may be diagnosed wrongly as suffering from real fecal incontinence and even undergo reoperations such as gracilis muscle or artificial sphincters that actually can make the patient worse. This problem should be suspected when one sees a patient that was born with a benign malformation, who underwent a technically correct operation, but was not treated correctly for constipation.

Despite significant technical improvements in the treatment of anorectal malformations, there still is a

group of patients that are born with poor anatomy and will suffer from fecal and sometimes urinary incontinence. At least 25% of all our patients treated posterior sagittally still suffer from fecal incontinence.¹ We can predict early on which patients will belong to that group, and we are able to offer them a bowel management program starting from the age of 3 years and keep them completely clean. Our goal in the management of anorectal malformations is to have all patients clean and dry either because they are fecally and urinary continent or because we keep them artificially clean and dry with an efficient bowel management and urinary management program.

Unfortunately, despite great advances in pediatric surgical care, there remains a significant number of patients who undergo attempted anorectal repairs with catastrophic complications. We believe that the majority of these complications are preventable. One must have a thorough understanding of these malformations, and utilize meticulous technique, delicate dissection, and preservation of the blood supply, if repair is to be attempted. These basic fundamentals need to be emphasized in the training of young pediatric surgeons, so as to improve the outlook for children born with anorectal malformations.

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