



Review articles

Laparoscopy and its use in the repair of anorectal malformations

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Received 4 October 2010; revised 25 March 2011; accepted 25 March 2011

Key words:

Laparoscopy;
Anorectal malformation;
Imperforate anus

Abstract

Introduction: Laparoscopy has been used for the treatment of anorectal malformations (ARMs) in an attempt to be less invasive and with the hope that it would result in a better functional outcome. There remains a significant debate about whether these expectations have been fulfilled.

Methods: Seventeen patients with ARM for whom laparoscopy was used were retrospectively reviewed. Six were operated on primarily by the authors, and 11 cases were referred after a laparoscopic repair performed elsewhere. In addition, a literature review was performed looking for evidence of less invasiveness and improved functional results in patients operated on laparoscopically.

Results: The diagnosis was imperforate anus with a rectobladder neck fistula in our 6 cases with the fistula ligated laparoscopically in each case. In 1 patient, the malformation was repaired entirely using laparoscopic technique. The other 5 patients had a laparoscopically assisted repair because we had to open the abdomen to taper a dilated rectum in 2, mobilize a very high rectum in 2, and take down a distal colostomy stoma in 1. Eleven patients were referred with a variety of problems after a laparoscopic repair done elsewhere for rectal stricture (5), rectal prolapse (4), recurrent rectourethral fistula (3), rectal mislocation (3), failed attempted repair leading to fecal incontinence (1), and a posterior urethral diverticulum (1). Our literature review included 47 references (involving 323 patients) published between 1998 and 2010. All studies showed that laparoscopic repair of ARMs is feasible. The review, however, did not provide evidence of less invasiveness or improved functional results.

Conclusions: Laparoscopy for ARM is a less invasive procedure when compared with those operations that would have previously required a laparotomy (rectobladder neck fistula). In cases of rectoprostatic fistulae, the laparoscopic approach is feasible and avoids a lengthy posterior sagittal incision. There is no evidence that the laparoscopic approach is a less invasive procedure for other types of ARMs. In cases of rectobulbar fistula, congenital anal stenosis, perineal fistula, ARM without fistula, the evidence suggests that it may lead to more complications. There is no evidence in the literature demonstrating better functional results in cases of ARM operated on laparoscopically.

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Laparoscopy was originally introduced for the treatment of anorectal malformations [1] in an attempt to be less invasive and with the hope that it would result in a better functional prognosis. In the 12 years since the introduction of

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the technique [1] and its popularization [2], there remains a significant debate about whether those expectations have been realized.

Our aim in this review was to determine if there is evidence in the available literature to affirm that laparoscopy is less invasive and achieves better functional results as compared with other approaches. In addition, based on the evidence found, we tried to determine the precise indications for laparoscopy to treat the different defects in the spectrum of anorectal malformations and to assess the risk and complications associated with the laparoscopic approach. The clinical experience of the authors with this therapeutic modality is limited and is presented as a reference to convey some technical points we have learned.

1. Methods

The medical records of 17 patients were retrospectively reviewed. Six with rectobladder neck fistula were operated on by us laparoscopically, and 11 cases were referred to us after a laparoscopic repair of an anorectal malformation was performed elsewhere.

Our emphasis was on the original anatomical diagnosis, technical challenges in the cases operated on by us, length of hospital stay, and intraoperative or postoperative complications that occurred related to the laparoscopic repair.

In addition, a literature review was performed looking for the reported number of patients with anorectal malformation operated on laparoscopically, the type of malformation, indications for the procedure, length of hospital stay, short-term and long-term follow-up, complications, evidence of less invasiveness, and/or improved functional results in patients operated on laparoscopically.

2. Results

Our 6 cases included only patients with rectobladder neck fistulas. In all 6 cases, the fistula was ligated laparoscopically, but only in 1 patient was the entire malformation repaired laparoscopically. The other 5 cases were considered laparoscopically assisted repairs because we felt obligated to open the abdomen to taper a dilated rectum (2 cases), to mobilize a very high rectum (2 cases), and to take down the distal stoma because of a very short available distal colon (1 case).

Eleven patients were referred to us with problems after a laparoscopic repair or attempted repair done elsewhere. Their original diagnoses were rectoprostatic fistula (5), rectobulbar fistula (2), unknown (2), anorectal malformation without fistula (1), and anal stenosis (1). The complications that led to referral to our center included rectal stricture (5), rectal prolapse (4), recurrent rectourethral fistula (3), rectal mislocation (3), fecal incontinence (1) (after an aborted

laparoscopic attempt to repair a rectourethral bulbar fistula), and a posterior urethral diverticulum (1).

The literature review included 47 references [1–47] related to the laparoscopic repair in 323 patients with anorectal malformation, published between 1998 and 2010. The diagnoses were rectoprostatic fistula (98), no diagnosis noted (75), rectobulbar fistula (36), rectovaginal fistula (26), “rectourethral” fistula (20), rectobladder neck fistula (20), cloaca (19), imperforate anus with no fistula (11), rectoves-tibular fistula (8), rectal atresia (5), rectoperineal fistula (4), and congenital rectal stenosis (1) (Table 1).

Seventy-four (22.9%) complications were reported in the literature after a laparoscopic repair (Table 2), and they included rectal prolapse (21) anal stenosis (11), posterior urethral diverticulum that required reoperation (8), other diverticula that were detected by magnetic resonance imaging and are still under observation (8), peritoneal fecal contaminations (6), perineal reoperations (4), urethral injury (2), conversion to laparotomy (2), temporary neurogenic bladder (2), peritonitis (2), postoperative dysuria (2), colitis (1), bowel perforation (1), perineal infection (1), evisceration at the trocar site (1), partial dehiscence of the anoplasty (1), and intestinal obstruction owing to adhesions (1).

The length of stay was mentioned in only 4 articles, and it was reported as 48 hours [5], varied from 3 to 12 days [22], an average of 6 days [37], and a mean of 14.3 days [41].

Of the 323 patients operated on laparoscopically, clinical results were only mentioned for 107 patients (33%) (Table 3). In 85 cases, the authors used a subjective evaluation, with terminology such as “satisfactory bowel activity” (7 cases) [19], “acceptable defecation status” (10 cases) [15], “acceptable frequency (1 to 4) of bowel openings per day” (7 cases) [12], “less than 1 bowel opening per day” (1 case), and “more than 5 bowel openings per day” (1 case) [12]. Of these 85 patients, 35 were evaluated using a subjective score that included parameters such as frequency of defecation, staining, perianal erosion, anal shape, and medication used [28,47]. Twenty-four patients were evaluated by another subjective method (Kelly score), with the authors noting that their patients had a score of 3.8 [16,41]. In 22 cases, the authors used what we considered a clear description of the clinical results: 15 patients had voluntary bowel movements, with 2 of them having minimal, occasional soiling [25,33,46]. Included in these 15 cases are 9 patients with bladder neck fistulas [33], 4 reoperations [25], and 2 rectovaginal fistulas [46]. Finally, included in these 22 cases are 7 cases reported as being fecally incontinent [2,13,46].

Five references [12,15,16,28,41] are frequently quoted, suggesting that laparoscopy produces better functional results. Two of those reports contain manometric studies comparing patients operated on laparoscopically with those who underwent a posterior sagittal anorectoplasty (PSARP). In the first article [12], there were 9 patients in the laparoscopic group and 13 in the PSARP group. There was no statistical difference between the 2 groups in frequency of bowel movements and resting sphincteric pressure. Presence

Table 1 Summary of patients with anorectal malformation found in the literature who were operated on laparoscopically, their diagnosis, and analysis of the indication for the approach

323 patients with anorectal malformations operated laparoscopically	{	Bladderneck 20 - indicated	}	84 Non indicated
		Prostatic 98 - controversial		
		Bulbar 36		
		Cloaca 19		
		No fistula 11		
		Vestibular 8		
		Perineal 4		
		Rectal Atresia 5		
		Rectal Stenosis 1		
		Unknown 75		121 ?
		Vaginal 26		
		"Recto-urethral" 20		

of rectoanal relaxation reflex was higher and occurred earlier in the laparoscopic group, but the clinical relevance of this fact is undetermined. All patients were still too young to be evaluated in terms of bowel control (average age in the laparoscopic group was 16.2 months, and in the PSARP group, 17.1), and the only clinical parameter analyzed was "frequency of bowel movements," which did not show statistical difference between the 2 groups.

In the second article [42], there were 11 patients in the laparoscopic group and 12 patients in the PSARP group. There were no differences in the Kelly score. The length of stay was shorter in the laparoscopic group (10 vs 14 days). There were no differences in the "rectal anal inhibitory reflex" and in the "high pressure zone length." The only

statistical difference was in the anal canal resting pressure (29.4 mm Hg laparoscopic repair [LAR] vs 23.4 mm Hg), with no clinical implications noted. All patients were younger than 3 years at the time of evaluation.

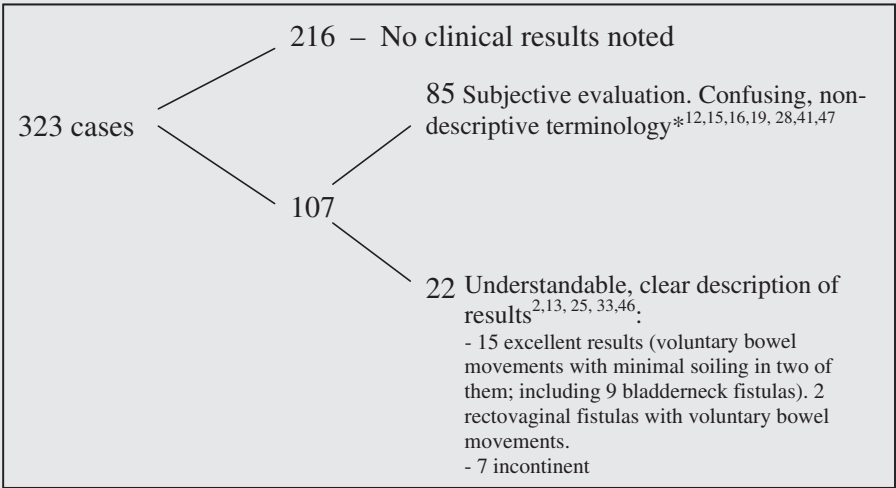
The third article [16] also used the Kelly score and rectal manometry to evaluate 13 patients operated on laparoscopically and 7 patients repaired by PSARP. The difference between the 2 groups did not reach statistical significance.

The fourth reference [15] relates to a magnetic resonance evaluation of 10 patients operated on laparoscopically and 8 patients repaired posterior sagittally. There was "less sphincter asymmetry" and "less peri-rectal fibrosis" in the laparoscopic group. The position of the rectum was central in both groups. No statistical correlation between "defecation

Table 2 Complications reported in the literature after the laparoscopic repair for anorectal malformations

74 complications	21 rectal prolapse
	16 posterior diverticulum
	11 anal stenosis
	6 peritoneal contamination with fecal material
	4 perineal reoperations
	2 urethral leak
	2 conversion to laparotomy
	2 temporary neurogenic bladder
	2 peritonitis
	2 post-operative dysuria
	1 of each: colitis, bowel perforation, perineal infection,
	evisceration at the trocar site, partial dehiscence,
	intestinal obstruction due to adhesions.

Table 3 Clinical results in patients with anorectal malformation repaired laparoscopically



* “satisfactory bowel activity” (7 cases) [19], “acceptable defecation status” (10 cases) [15], “acceptable frequency 1–4 bowel openings per day” (7 cases) [12], “less than one bowel opening per day (1 case), and more than 5 bowel openings per day (1 case)” [12], evaluation by an arbitrary score (35 cases) [28,47], Kelly score of 3.8 (13 cases) [16], and Kelly score of 3.8 and anorectal manometry in patients younger than 3 years [41].

status,” “degree of sphincter asymmetry,” or “peri-rectal fibrosis” was found.

The fifth article [28] refers to an endosonographic and magnetic resonance imaging evaluation, as well as continence evaluation of 15 patients operated on laparoscopically and 9 patients repaired posterior sagittally. Three patients in the posterior sagittal group were previously operated on laparoscopically and had been left with a posterior urethral diverticulum that was repaired posterior sagittally. There was no statistical difference when “surgical stress” and “muscle thickness” were compared between groups. Analyzing the continence questionnaire, there was no statistical difference when comparing groups at ages 1, 2, and 5 years. There was a statistical difference at ages 3 and 4 years.

A recent article [33] worth mentioning because it is the only one with long-term results reported 9 patients operated on laparoscopically with rectobladder neck fistulas older than 3 years. According to the authors, 6 patients have voluntary bowel movements, are clean, and occasionally need a rectal suppository for evacuation. The remaining 3 are managed with diet and are clean with minimal soiling.

The literature review demonstrated that the laparoscopic repair of anorectal malformation is feasible. It did not show evidence of improved functional results.

3. Discussion

A universally accepted indication for laparoscopy is to replace a laparotomy. A transabdominal approach to reach the rectum in male patients with an anorectal malformation is required only in patients with rectobladder neck fistula (the

rectum is located at or above the peritoneal reflection), which represent 10% of all male cases. Rectums in lower positions are reachable through a perineal approach. In cases of rectobladder neck fistula, the fistula ligation can be effectively done laparoscopically because the rectum and the bladder do not share a common wall, and the rectum joins the bladder neck in a T fashion, which makes the laparoscopic dissection straight forward. In all other malformations of male patients, the common wall between the rectum and urinary tract is longer when the malformation is lower. Trying to make 2 walls, out of a thin common wall down in the pelvis, below the peritoneal reflection, risks damage to the urinary tract.

In our limited experience with bladder neck fistulas, we converted to a laparotomy in 5 of our 6 cases. One of the reasons for conversion to laparotomy was the need to taper a very dilated rectum (Fig. 1) before the pull-through. To be able to place the rectum within the limits of the sphincter, we felt that tapering was required, and we did not feel comfortable attempting the tapering laparoscopically. We recognize that this may be only a reflection of our lack of experience and others may taper the rectum laparoscopically, although we were not able to find a mention of that maneuver in the literature. In 2 patients, the rectal mobilization required a very accurate and selective ligation of the mesenteric vessels, and we did not feel that it was safe to attempt such maneuver using the cautery laparoscopically for fear of devascularizing the distal segment. This anatomical challenge of mobilizing a very high rectum is particularly relevant for bladder neck fistulas as compared with prostatic fistulas. Fig. 2 shows the distal end of the bowel already separated from the bladder neck and the entire delicate blood supply of the rectosigmoid. The goal in this situation is to

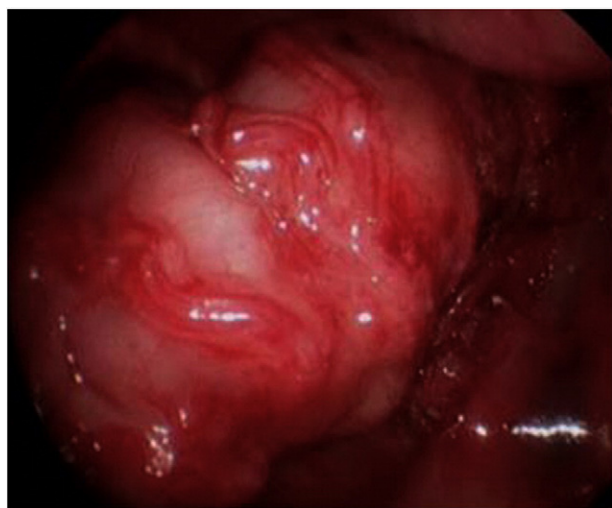


Fig. 1 Laparoscopic view of a dilated rectum that needed to be tapered.

ligate only those vessels that interfere with the pull-through, preserving those that are needed for perfusion of the bowel (Fig. 2) (preservation of the rectum is very important to the child for future continence or success with a bowel management program). We felt that an open approach was safer for this maneuver than cautery coagulation done laparoscopically. We recognize that this maneuver may be done effectively laparoscopically by a more experienced operator. For a prostatic fistula, once the rectum is freed, it

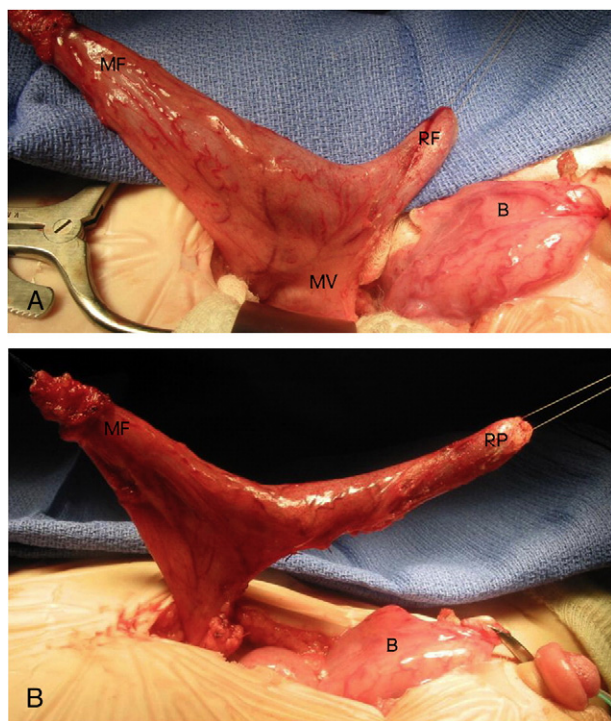


Fig. 2 A and B, Gaining length via laparotomy on a very high rectum to accomplish the pull-through. MF indicates mucous fistula; RF, rectal fistula; MV, mesenteric vessels; B, bladder; RP, rectum ready for the pull-through.

reaches the perineum with less manipulation of the blood supply. The last reason for conversion in our series was a colostomy placed too distal in the rectosigmoid (Fig. 3), interfering with the pull-through. This patient required a laparotomy to mobilize the distal stoma to a more central and lower position, allowing the pull-through to be completed: a problem that we have previously presented [39]. This problem is not attributable to laparoscopy, but it represents rather a general error when making a newborn colostomy in anorectal malformation. Proper placement of the newborn colostomy in the proximal sigmoid/descending colon would have avoided this conversion to an open approach.

We acknowledge that the complications observed in patients who were referred to us after a laparoscopic repair done elsewhere may not be representative of all patients operated on laparoscopically. In addition, they are not necessarily a consequence of the laparoscopy because they may occur with other approaches (our redo reference), and instead, they rather reflect a technical problem.

We were very impressed by the fact that only 20 patients (6%) of 323 had a rectobladder neck fistula, which we believe (with these described limitations) represents a clear indication for a laparoscopy because these all would have required a laparotomy to find and dissect the distal rectum.

Ninety-eight patients (30%) in the reported literature had a prostatic fistula. Even when we repair prostatic fistulas without the need of a laparotomy or laparoscopy, we recognize that prostatic fistulae sometimes require a technically demanding posterior sagittal operation. Therefore, it is conceivable that a well-trained laparoscopist would

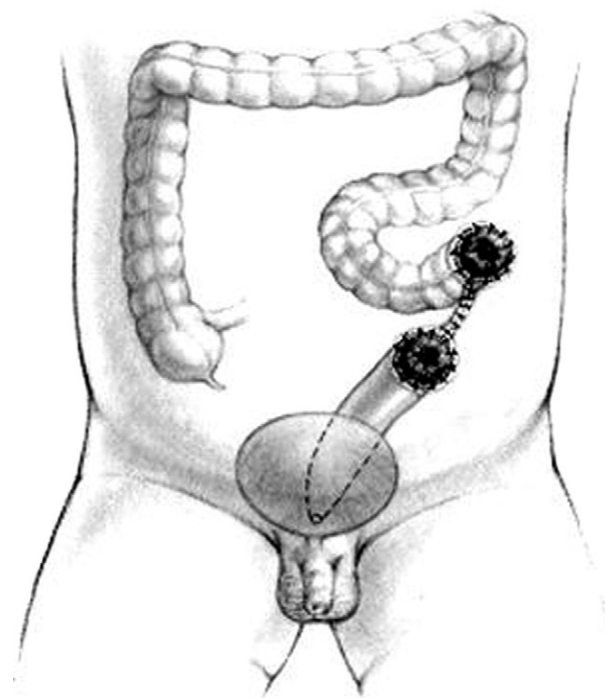


Fig. 3 Colostomy placed too distal in the rectosigmoid, interfering with the pull-through.

find it easier to repair such malformation laparoscopically. We therefore acknowledge that the laparoscopic approach to repair rectoprostatic fistulae is reasonable.

In the reviewed articles, 36 patients with bulbar fistula were operated on laparoscopically. We consider laparoscopy unnecessary in the repair of this specific variant of anorectal malformation. The rectum in a bulbar fistula and also in a defect without fistula is located well below the peritoneal reflection. Therefore, a transabdominal (laparoscopic) approach requires an extensive unnecessary dissection of the rectum and involves a long common wall that exists between rectum and urethra.

Urethral complications associated with a laparoscopic approach, including recurrent fistula and posterior urethral diverticulum (Fig. 4), occurred in cases of rectourethral bulbar fistulae. Conceivably, the surgeons had difficulty separating the rectum from the urethra deep in the pelvis. The common wall between urethra and rectum is longer when the malformation is lower. Fistulae likely occurred because of damage to the posterior urethra and anterior rectal wall.

Posterior urethral diverticulum is a complication that we have seen in the past [51] and now may be increasing in number because of the use of laparoscopy in cases with rectourethral bulbar fistula. Trying to dissect the rectum laparoscopically in a patient with a rectourethral bulbar fistula may be an extremely difficult task, which perhaps induced surgeons to stop the dissection without having reached the fistula site, resulting in leaving behind the distal most rectum, leading to a posterior urethral diverticulum [51].

Nineteen patients with a cloaca were reported in 9 articles to have a laparoscopic repair [4,5,8,11,13,16,28,35,47]. We were interested in learning about the technical details of those operations, but we found that 5 reports did not describe the technique used [5,16,28,35,47], and in the remaining reports, the patients only underwent the laparoscopic repair of the rectal component of their defect and underwent another procedure (posterior skin vaginoplasty [8,11], total urogenital mobilization [4], or posterior sagittal anorecto-vaginal-urethral plasty [13]) to repair the urogenital component of the malformation. In cloacas with a low urogenital confluence and a very high rectum, which is a rare circumstance, laparoscopy may be useful to mobilize the rectum and avoid a laparotomy. However, these authors cannot really claim that they have repaired a cloaca laparoscopically.

We could not understand why traditionally benign malformations (imperforate anus without fistula, 11 cases; rectovestibular fistula, 6 cases; rectal atresia, 5 cases; and rectoperineal fistula, 4 cases) were repaired laparoscopically. These are defects that can be repaired with a perineal approach in operations that take 1 to 2 hours; patients have an excellent functional prognosis and minimal postoperative pain. Two patients with rectovestibular fistulas and an associated absent vagina underwent a laparoscopic repair [31,47].

The literature review showed that 10 authors [1,13,15,17-19,21,25,30,32] did not specify the type of malformation that they operated on. Seventy-five patients were operated on laparoscopically for an “unknown” type of defect, which makes the results impossible to analyze.

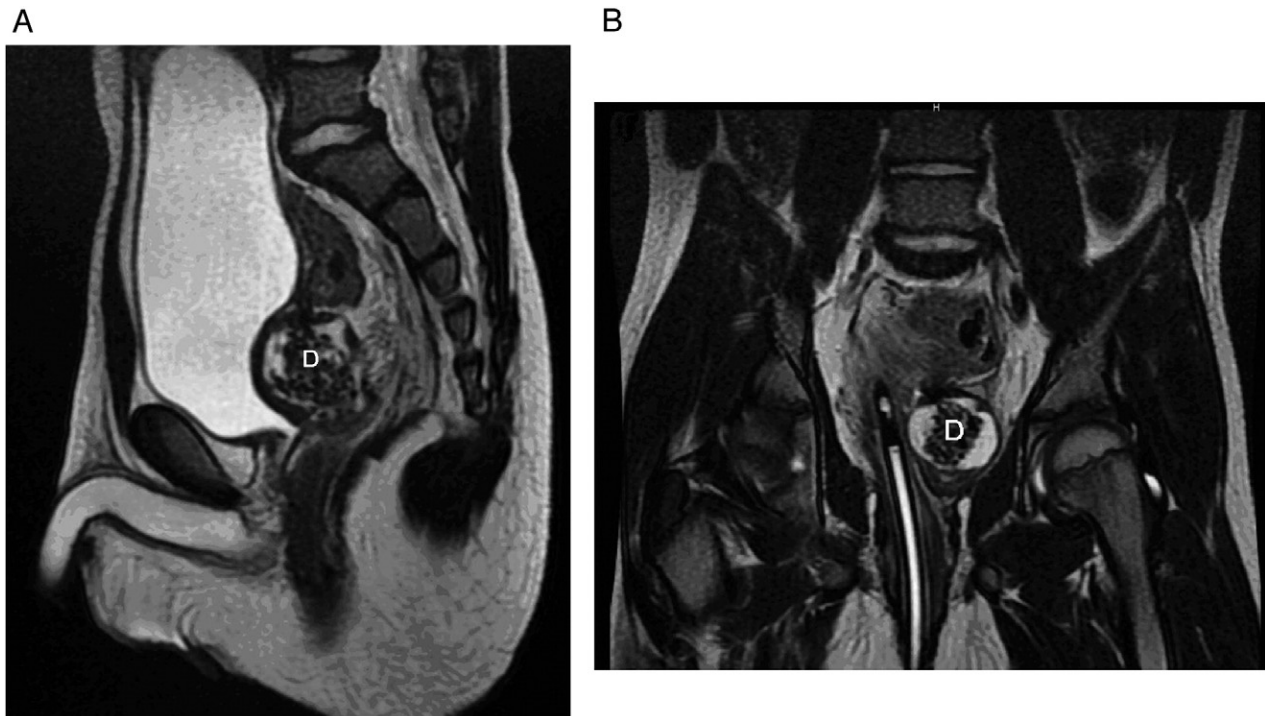


Fig. 4 A and B, Posterior urethral diverticulum (D) after laparoscopic approach. Foley catheter is used to delineate the rectal lumen.

We urge future authors to be specific about the anatomical diagnosis to ensure valid comparisons, results, and conclusions. When authors mention “high anorectal malformation,” some are referring to bladder neck fistula only, others include prostatic fistulas in this group, and others also include bulbar fistula and patients without fistula. The same problem was found in the female group with some authors considering all cloacas “high” without noting the length of the common channel. In addition, the status of the sacrum and spine, vital aspects to predict functional results, should be noted on each patient. This lack of agreement and consistency in reporting results makes it impossible to analyze and compare series.

Because we are trying to compare results between different techniques used to repair different defects that form a wide spectrum of malformations with different prognosis for bowel control, it is essential to describe the exact type of malformation.

We were impressed by the extremely high number of “vaginal fistulas” operated on laparoscopically (26 cases). From our experience, this is an extremely rare malformation [48]. Most of the cases referred to us with the diagnosis of “vaginal fistula” were actually cloacas or redo vestibular fistulas [50]. In addition, in our 30 years of experience (1100 female cases), we have only seen 8 documented cases of true rectovaginal fistulae. This observation, of course, increased our skepticism about the accuracy of the diagnoses reported in the literature.

Advantages of laparoscopy that are commonly noted are less invasiveness, which translates into less postoperative pain; earlier discharge; and earlier return to activities. Interestingly, only 4 articles mentioned the length of stay [5,22,37,41], which is one of the accepted criteria to assess invasiveness, and actually, the length of stay in those series was much longer than expected for a minimally invasive procedure.

This seems to be true in those patients in whom the laparoscopic repair replaced a laparotomy. In all other male cases, a laparoscopic repair actually means replacing an extraperitoneal limited rectal dissection by a transperitoneal extended rectal dissection. We wonder in this context if laparoscopy should still be considered minimally invasive.

Several authors [2,12-14,20,23,47] referred to the potential damage or disadvantages that the posterior sagittal incision may cause to the sphincters to justify the use of the laparoscopic approach. So far, none of the existing publications presented evidence of such damage. We specifically recommend the reading of 2 articles [49,52]. One of them [49] is an experimental study that demonstrated that the posterior sagittal incision alone did not affect bowel control. The other [52] presents a large clinical experience with the use of a posterior sagittal incision, dividing the entire sphincter mechanism, for the treatment of tumors, sequela posttrauma, urethral problems, and patients with urogenital sinus. All of these patients had a normal anorectum, and the approach of incising and then recon-

structing the sphincter did not affect their bowel control. Therefore, we believe that the criticism that the posterior sagittal approach “cuts the sphincter” and thereby damages it is not valid.

Laparoscopy has been used as a primary procedure during the newborn period, avoiding a colostomy. That has been considered as evidence of less invasiveness. Actually, the primary repair has been previously reported without the need of laparoscopy [53-56]. The potential advantages of a primary repair in the newborn are not unique to the laparoscopic approach. The clinician must keep in mind that the primary repair has very specific indications and contraindications. Ignoring those contraindications has led to catastrophic, otherwise preventable complications. In addition, bowel distension, as seen in newborns with anorectal malformations, may be a serious disadvantage for a laparoscopic approach [10].

The article that suggested that the laparoscopic repair could render better functional results than the traditional PSARP [12,15,16,28,41] failed to support that premise. The numbers of reported cases were very small, and the results were either not statistically significant or had no clinical implications. We question, for example, how better manometric values without a clinical correlation impact the patient’s clinical result. In addition, the authors use subjective and unreliable parameters such as “frequency of bowel movements” to evaluate the functional results.

We are skeptical about the study with bladder neck fistulas that showed extraordinarily good results [33]. This is the malformation with the worse prognosis [48], and the authors reported 100% bowel control.

4. Conclusions

Laparoscopy is a less invasive procedure when compared with those operations that require a laparotomy. There is no evidence in the literature indicating that it is a less invasive procedure in other types of malformations, and in some (rectums located below the peritoneal reflection), it may be more invasive and lead to complications. In rectoprostatic fistulas, its use seems reasonable because the fistula can be reached easily by an experienced laparoscopist and will avoid a large posterior incision. Laparoscopic cases we reviewed resulted in a significant number of complications, but this was not necessarily related to the approach. There is no evidence of better functional results in cases operated on laparoscopically.

In females, the laparoscopic approach may be useful to visualize the internal gynecologic anatomy for diagnostic purposes and to mobilize the rectum in cases of high implantation of the rectum in the vagina in patients with or without a cloaca.

In instances of rectobulbar urethral fistula, vestibular fistula, and anorectal malformation without a fistula, we believe that laparoscopy is unnecessary. We recommend that authors publishing material comparing results of new

surgical approaches for anorectal malformations that report clinical outcomes be specific in terms of diagnosis (avoiding old, misleading terminology such as *high*, *intermediate*, and *low*) and include data on the status of the patients' sacrum and spine.

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