

Laparoscopic approach in the management of anorectal malformations

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Abstract Seventeen years have passed since the first description of the laparoscopic approach for anorectal malformation and approximately 68 articles have been published on the subject. In this review article, we aim to describe the advantages as well as the indications and contraindications of this approach when dealing with each specific type of anorectal malformation, according to what has been described in the literature and to our own experience. The ideal and undisputable indication for laparoscopy remains for cases in which the abdomen needs to be entered to repair the malformation. Only 10 % of male patients with anorectal malformation are born with a recto-bladder neck fistula that requires an abdominal approach, this represents an ideal indication for laparoscopy. In females, only the complex cloacae with a common channel length greater than 3 cm are the ones that require a laparotomy; they represent about 30 % of the cloacae. However, the repair of this type of cloacae also requires sophisticated and technically demanding maneuvers that have never been done laparoscopically. In cases of recto-urethral prostatic fistulas the malformation can be repaired either way: laparoscopically or posterior sagittally. In all other malformations: recto-perineal fistula, recto-urethral bulbar fistula, anorectal malformation without fistula, rectal atresia, recto-vestibular fistula; no justification for laparoscopy could be found; and in some cases, laparoscopy is contraindicated. In the published reports, there is no evidence supporting the idea that laparoscopic repair results in better functional results when compared with non-

laparoscopic operation; there is a tendency to omit information relevant to bowel control such as the characteristics of the sacrum and the presence or absence of tethered cord; and most authors do not compare results between comparable malformations.

Keywords Anorectal malformation · Imperforate anus · Laparoscopic repair

Introduction

Laparoscopy to repair anorectal malformations has been widely used by pediatric surgeons, in an attempt to be less invasive, to offer better cosmetic results, to have a faster return in bowel function, to decrease hospital length and pain and lately, to improve functional results.

After 17 years since the first description of the laparoscopic approach for anorectal malformation [1], approximately 68 articles [1–68] have been published on the subject, 6 of them being review articles [36, 44, 59, 61, 62, 68], and 10 of them attempting to compare laparoscopic versus posterior sagittal approach [9, 14, 16, 35, 40, 43, 45, 49, 64, 66].

This review represents an attempt to evaluate what are the advantages, indications, and contraindications for laparoscopy in the management of anorectal malformations.

Methods

A literature review was performed looking for articles related with the laparoscopic repair of anorectal malformations, including comparative studies between PSARP (posterior sagittal anorectoplasty) and laparoscopy,

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laparoscopic case reports, and review articles. Emphasis was placed on functional results, length of stay, and complications. Articles that were unrelated to the main repair of the malformation as well as abstract publications were excluded. The study period ranged from 1998 until 2015.

Results

622 patients with anorectal malformation have been reportedly operated on, with the use of laparoscopy since 1998. All types of anorectal malformation have been repaired partially or totally by laparoscopy.

The specific types of anorectal malformations were: recto-urethral prostatic fistula (235 cases), recto-bladder neck fistula (112), recto-urethral bulbar fistula (107), not specified or the authors used the old terminology of “high, intermediate, and low” (45), described as “recto-vaginal fistula” (31), described only as “recto-urethral fistula” without specification of the level (30), cloaca (23), imperforate anus without fistula (22), recto-vestibular fistula (13), rectal atresia (2), and recto-perineal fistula (2).

Ten articles [8, 10, 12, 14, 25, 28, 32, 38, 40, 54] reported 23 cloaca cases in whom laparoscopy was used. In ten cases, only the rectal component of the malformation was repaired and the urogenital sinus was left intact for future repair [54]. In ten cases [14, 25, 28, 32, 38, 40], the surgical technique was simply not described. In two cases, besides the laparoscopic rectal repair, the surgeons performed a “posterior flap vaginoplasty” [8, 10]. In the remaining case, the authors performed a posterior sagittal anorectovaginourethroplasty [12], in addition to the laparoscopy. Interestingly, the length of the common channel was only mentioned in 7 cases [8, 10, 12, 54] and it was shorter than 3 cm in all of them. The pre-operative location of the rectum in these cloaca patients was described as: “above pubo-coccygeal line” in 7 cases, “below pubo-coccygeal line” in 3 cases [54], “high” in 2 cases [8, 12]. This information was not provided in the remaining 11 cases [10, 14, 25, 28, 32, 38, 40].

Thirteen articles mentioned the sacrum [5, 17, 19, 37, 43, 46, 55, 58, 60, 64–67] but only in 5 of them [43, 55, 64–66] the sacral ratio was calculated. Only 6 articles mentioned the status of the cord [2, 5, 37, 41, 55, 60].

Complications were reported in 34 articles [7, 10–12, 14, 15, 18, 21, 25, 29–33, 35, 37, 38, 40, 41, 43–48, 50, 52, 55, 56, 60, 63, 64, 66, 67] and they included: rectal prolapse (58 cases), anal stenosis (25), posterior urethral diverticulum (13), bowel obstruction (4), leakage of stool into the abdominal cavity (5), wound infection or dehiscence (5), urinary retention (4), recurrent fistula (4), hernia at the port site (3), rectal mislocation (3), extravasation of urine into the abdominal cavity (2), pelvic sepsis (2),

urethral injury (2), retraction (1), urethral mucocele (1), transected ureter (1), and vas deferens injury (1).

Ten papers attempted to compare results between laparoscopy and posterior sagittal approach [9, 14, 16, 35, 40, 43, 45, 49, 64, 66]. Together those papers represent 161 patients operated by laparoscopy (LAARP) and 130 repaired with a posterior sagittal approach (PSARP). The parameters used by the authors to compare both techniques were: clinical results, rectal manometry, MRI, defecogram, length of stay, and complications.

- Clinical results: 7 papers [14, 35, 40, 45, 49, 64, 66] compared the clinical results using scores that included parameters such as “frequency of bowel movements”, “constipation”, and “perception of the anal tone”. 3 papers [14, 35, 64] reported better results with LAARP, other 4 [40, 45, 49, 66] found no differences.
- Rectal manometry: 3 papers [9, 14, 45] compared the manometric results between both techniques. The results were contradictory and, therefore, no valid conclusions could be reached.
- MRI: 4 papers [16, 25, 29, 64] compared the images obtained with MRI studies. One of them [16] reported “better symmetry” and “less fibrosis” in the location of the rectum. The other three [25, 29, 64] did not report significant differences.
- Defecogram: 2 papers [40, 64] compared the anorectal angle measured on the images of defecograms and found no difference between both techniques.
- Length of stay: 1 paper [66] reported a shorter length of stay in LAARP cases as compared to PSARP (5.8 vs. 8.4 days). However, sixteen papers [19, 21, 22, 31, 35, 41, 46–48, 53, 54, 56, 57, 60, 66, 67] reported an average length of stay in laparoscopic cases of 7.6 days with a range of 4–21 days.

Five review articles addressed the laparoscopic repair of anorectal malformation [36, 44, 61, 62, 68]. The consensus on these papers is the need for standardization and improvement in the quality of reporting; and that there is currently no evidence of better functional results in cases of anorectal malformation operated on laparoscopically.

The theoretical potential “detrimental effect of the posterior sagittal anorectoplasty on the sphincter mechanism” or the “advantage of not having to divide the muscle complex” was mentioned but not demonstrated in 30 articles [2, 3, 6, 7, 9–11, 13, 16–18, 20, 22, 24, 34, 35, 40, 46, 48–50, 52, 56, 58, 61–66].

Discussion

It is important to emphasize that, when reviewing published articles, it is not possible to determine if multiple

publications by the same authors are mentioning different patients or if the same patient is presented more than once; therefore, all numbers mentioned in this review should be analyzed with this potential bias in mind, this represents a limitation of this study. Specifically, when we analyzed the numbers of cases reported to be operated on laparoscopically, as well as the type of malformation and the complications, we did not include the patients reported on the multicenter retrospective study [58] in an attempt to avoid this bias.

Laparoscopy was mainly used in recto-bladder neck fistula and recto-prostatic fistula (347 cases). We consider that in recto-bladder neck fistulas, laparoscopy is indicated. In recto-prostatic fistula, we believe that the malformation can be repaired both ways (laparoscopically or posterior sagittally). The decision must be taken based on the experience of the surgeon.

In recto-urethral bulbar fistula, the rectum can be easily found through a posterior sagittal approach as it is located within a short distance from the perineum, but the most important anatomic fact is that there is a long common wall between the rectum and the urethra making its separation very challenging during an abdominal approach with laparoscopy or laparotomy. When the patient is operated posterior sagittally the operation takes approximately 2 h, pain is minimal, the patient can be discharged 48 h after the procedure, and 85 % of the patients have bowel control [69]. In contrast, this is the group of cases in whom a posterior urethral diverticulum occurred when the repair was attempted laparoscopically [15, 32, 44, 70]. In addition, the laparoscopic repair of a recto-urethral bulbar fistula requires the dissection of the entire rectum; a maneuver that is not indicated nor required.

The old terminology of “high, intermediate, and low” malformation, described in 45 cases is inadequate and misleading. Within the so-called “high” category, different authors include different types of defects with different functional prognosis and treatment modalities. The same problem happens when authors reported “recto-urethral fistula” in 30 cases without further definition of the type of defect. The differentiation between a recto-urethral bulbar fistula and a recto-urethral prostatic fistula has important therapeutic and prognostic implications, regardless of the type of approach used to repair the malformation [69].

We were surprised to find that 31 cases were described as having a recto-vaginal fistula. The majority of the authors did not present a graphic evidence of the location of the fistula (photograph or distal colostogram). In our own database of 1100 cases of female anorectal malformations, we only found 7 documented cases of recto-vaginal fistula [71]. In a series of 65 female patients operated on at another institution due to a “recto-vaginal” fistula that required a reoperation for different reasons, we found that

none of those actually had a recto-vaginal fistula; instead, they were born with a cloaca (they still had a urogenital sinus) or a vestibular fistula (they still had a small pouch in the vestibule). Therefore, we have a reason to suspect that some or most of those cases labeled as “recto-vaginal fistula” were misdiagnosis. When the diagnosis is correctly made, in the majority of the cases, the rectum opens very low in the vagina and shares a long common wall like in cases of recto-vestibular fistulas.

Anorectal malformations without fistula can happen in males and females. In male patients, the rectum is located at the level of the bulbar urethra and in females, approximately 2 cm from the perineum [72]. The laparoscopic repair of this malformation includes an otherwise unnecessary peritoneal invasion and a full dissection of a rectum that can be repaired with minimal dissection. There is a very rare group of patients that we call complex malformations (since they cannot be compared with patients with classic anorectal malformation without fistula) that are born without fistula and the rectum can be found floating in the abdominal cavity that would represent a good indication for laparoscopy. In our database that has over 2300 cases, we could only find 5 of those patients emphasizing how rare this anomaly is.

Recto-perineal fistulas can also happen in males and females. The mobilization required to place the rectum within the limits of the sphincter is minimal. The post-operative pain is also minimal and 100 % of the patients have bowel control [69]. For the same reason described above, in cases of anorectal malformation without fistula, this defect should not be repaired laparoscopically.

Recto-vestibular fistula is the most common type of anorectal malformation seen in females. The most important anatomic characteristic is that the vagina and the rectum share a long common wall that can be carefully and meticulously separated through a posterior sagittal approach. Operative time is 2 h, there is minimal pain, if the patient has a colostomy she stays in the hospital 48 h, 95 % of the patients have bowel control [69]. It is difficult to find a justification for a laparoscopic approach in this malformation.

There is a rare group of patients with vestibular fistula and absent vagina [73] who could benefit from a laparoscopic approach to evaluate the presence of Mullerian structures. In the event that no Mullerian structures are found and when simultaneously the patient presents with poor prognosis for bowel control (sacral ratio <0.4 and presence of tethered cord), laparoscopically, the rectum could be amputated and left as neo-vagina and more proximal bowel could be mobilized and pulled through as a neo-anus. Tei [12] has reported this technique but failed to mention the characteristics of the sacrum and the status of the cord in his patients, which is essential before using this

technique. When the patient has a good potential for bowel control, the rectum should be left intact as rectum and a more proximal portion of the colon should be used as neo-vagina, since patients with anorectal malformation are very sensitive to diarrhea which can occur after removal of the recto-sigmoid (used as neo-vagina).

Rectal atresia only happens in 1 % of the cases [69]. When this defect is repaired via a posterior sagittal approach, the mobilization required is minimal as the proximal and distal bowels are almost touching each other, very close to the pectinate line level. The posterior sagittal approach allows an easy and quick repair with excellent functional results. Laparoscopy is not justified.

Cloacae with a common channel length shorter than 3 cm can be totally repaired posterior sagittally [74] with an operation that takes approximately 3 h. Patients stay in the hospital for 48 h and the functional prognosis is good. The only reason to approach the abdomen in a case of a cloaca with a common channel shorter than 3 cm would be the extremely unusual combination of a short common channel and a very high rectum, not reachable from below. That association has been seen by us in 4 % of our cloacae; therefore, we suspect that some authors entered through the abdomen in a number of cases that could be easily repaired posterior sagittally.

In cloacae with a common channel length of more than 3 cm, frequently an abdominal approach is required for the separation of the vagina(s) from the urinary tract, a maneuver that is technically demanding, since there is no plane of separation and, in addition, the ureters run through that common wall. To achieve the separation of the vaginas from the urinary tract, the bladder must be opened, the ureters must be catheterized and a meticulous, tedious dissection is performed trying to make two walls out of one. The vagina(s) and the urinary tract share a wide common wall, heavily vascularized. This maneuver has never been done laparoscopically.

The presence or absence of the sacrum, as well as its characteristics, is the most important factor which determines the functional prognosis for bowel control. It is not conceivable to discuss bowel control without taking into consideration the sacrum. Terms such as “hypoplasia” or “deformity” must be avoided since they do not allow to know the magnitude of the sacral deficiency. That was the motivation to create the sacral ratio [75], which is an attempt to have an objective, numerical description of the sacrum. This literature review showed that only 13 articles mentioned the sacrum, but the sacral ratio was disclosed in 5 publications [43, 55, 64–66].

The presence or absence of tethered cord is widely recognized as a prognostic indicator mainly for urinary function [76]. Only six papers mentioned the status of the cord [2, 5, 37, 41, 55, 60].

The complications mentioned in 34 articles [7, 10–12, 14, 15, 18, 21, 25, 29–33, 35, 37, 38, 40, 41, 43–48, 50, 52, 55, 56, 60, 63, 64, 66, 67] were not impressive, since the authors described complications that occurred frequently with techniques other than laparoscopy. Two complications are worth discussing; one is the rectal prolapse, which has been frequently described as severe, following a laparoscopic repair of anorectal malformations. We speculate that the full dissection of the rectum and the lack of anchoring stitches may contribute to explain the complication. Such complication is frequently seen in cases of recto-bladder neck fistula and in cases of poor sphincters. In other malformations, the posterior sagittal approach, allows to anchor the rectum to the neighbor structures; in addition, the rectum is still fixed to the pelvic diaphragm because only the lower part is dissected. The other worth discussing complication is the posterior urethral diverticulum, since it is totally preventable when the malformation is repaired posterior sagittally. This complication occurs when the surgeons tried to repair a recto-urethral bulbar fistula laparoscopically. As they progress in the dissection of the distal rectum, it becomes more difficult to separate the anterior rectal wall from the posterior urethral wall. In addition, it became increasingly difficult to dissect the lowest part of the rectum; consequently, the surgeon is obligated to amputate the rectum, leaving a distal portion attached to the bulbar urethra. This has important serious, negative implications for the patient [44, 70].

It seems to us that the 10 papers that compared LAARP to PSARP could not substantiate an advantage of one technique over the other. When they compare clinical results, they used old scores that do not reflex the degree of bowel control of the patient. The correlation between rectal manometry and bowel control has never been objectively demonstrated. In addition, the three papers that compared manometric results reported very controversial results. The observations made by those who performed MRI studies describing “better symmetry” and “less fibrosis” were not clinically correlated. The “anorectal angle”, showed no difference between both techniques, furthermore such angle has not been correlated with the degree of bowel control.

Decreasing the length of stay is one of the reported advantages of all laparoscopic operations. In our review, we found one paper reporting a shorter length of stay for laparoscopy as compared with PSARP. Yet, we were surprised that the average length of stay in LAARP cases, as reported by 16 other papers was 7.6 days. Patients with a colostomy subjected to a PSARP usually stay in the hospital for 48 h [69]. Patients without a colostomy usually remain in the hospital for 7–10 days because they remain fasting, receiving parenteral nutrition. The authors do not

explain the reason why patients operated laparoscopically remain in the hospital for such a long time.

About the theoretical concern of dividing the muscle complex during a posterior sagittal approach, the authors failed to mention an experimental study which demonstrated that the posterior sagittal division does not interfere with bowel control [77]. In addition, none of the authors mentioned another publication [78] reporting the clinical experience with the posterior sagittal division of the sphincter mechanism in patients with normal rectum with post-operative preservation of bowel control.

Conclusion

In the treatment of anorectal malformations there is evidence of benefit when laparoscopy is used to replace a laparotomy (less surgical trauma and better cosmetic results) in cases of recto-bladder neck fistula.

Laparoscopy can be justified in some recto-prostatic fistulas, rare cloaca cases in which there is a short common channel between the urethra and the vagina and a very high rectum, rare cases of recto-vestibular fistulas with absent vagina with poor prognosis for bowel control, and rare cases of complex anomaly when the rectum can be found floating in the peritoneal cavity.

Laparoscopy is not indicated as it is not less invasive, in cases of recto-perineal fistula, recto-urethral bulbar fistula, recto-vestibular fistula, rectal atresia, most cloacae, and in anorectal malformation without fistula.

There is no evidence of a functional superiority of laparoscopy when compared to posterior sagittal approach.

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