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REVIEW ARTICLE

Laparoscopy-assisted surgery for male imperforate anus with rectourethral fistula

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Abstract Laparoscopically assisted anorectal pull-through (LAARP), first described by Georgeson, is now considered to be the radical surgical treatment of choice for rectourethral fistula (RUF) in boys with high/intermediate-type imperforate anus. Accurate positioning of the pull-through canal, with pelvic floor muscles surrounding it symmetrically, is well recognized as the most important prognostic factor irrespective of the procedure performed. Surgical intervention should be LAARP with intraoperative measurement of the RUF, with follow-up focused on bowel habit. Complications such as diverticulum formation, have been reported with increasing frequency after LAARP and are most likely related to incomplete excision of the RUF, especially in bulbar cases. Thus, complete excision, while technically challenging, is crucial. Based on the results of a multicenter study comparing LAARP with other surgery, the most reliable investigation for detecting the presence of a diverticulum is MRI. At Juntendo University Hospital in Tokyo, Japan, blunt dissection with mosquito forceps to identify the potential pull-through canal, measuring the length of the RUF directly, and closer placement of trocars (in bulbar fistula cases) are homegrown refinements that we feel improve outcome and we present a review of our approach to the surgical management of ARM.

Keywords Imperforate anus · Rectourethral fistula · Anorectal malformations · Laparoscopy-assisted anorectal pull-through · Diverticulum

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Introduction

Male imperforate anus (MIA) with rectourethral fistula (RUF) still presents surgical challenges even with the most up to date techniques and procedures. There are certain outstanding issues involving classification of anorectal malformations (high, intermediate and low) and most cases of MIA are regarded to be high/intermediate malformations. In this review, we discuss surgical procedures, outcome, and complications with respect to the type of RUF present.

Historically, the Pena procedure pioneered an era of greatly improved surgical intervention for RUF excision in 1982 involving division of pelvic floor muscles and sphincter muscles exactly in the midline through a posterior sagittal approach, thus allowing the junction between the urethra and the RUF to be observed under direct vision [1].

The next revolution was instigated by Georgeson et al. [2] in 2000 who combined careful application of minimally invasive surgery for dissection/division of the RUF, followed by colon pull-through without dividing the pelvic floor muscles and sphincter muscles. This procedure, called laparoscopy-assisted anorectal pull-through (LAARP) has allowed certain centers to achieve encouraging results, although there are also increasing numbers of reports of postoperative complications such as diverticulum formation due to residual fistula appearing in the literature [3, 4].

The aim of this review paper is to introduce current treatment techniques including technical refinements, introduce an effective system for assessing outcome based on postoperative continence, discuss the relevance of postoperative radiologic findings, and describe postoperative complications of LAARP likely to arise when MIA with RUF is treated surgically. We would also like to introduce some novel procedures we developed at Juntendo University Hospital in Tokyo, Japan, which we feel would

improve outcome and present an overview of our protocol for the surgical management of MIA with RUF.

LAARP

Laparoscopically assisted anorectal pull-through (LAARP) is now considered to be the radical surgical treatment of choice for MIA with RUF in many centers, predominantly indicated for patients with "high/intermediate" ARM [5]. Even Pena's group recently acknowledged that they would consider using a laparoscopic approach to repair prostatic RUF [6]. When Pena's group surgeons repair prostatic RUF without the need for a laparotomy or laparoscopy, the posterior sagittal procedure is technically demanding, therefore, it is conceivable that a well-trained laparoscopist would find it easier to repair such a malformation laparoscopically.

In contrast to prostatic RUF, Pena's group mentioned that the rectum in a case with bulbar RUF is located well below the peritoneal reflection, therefore, a laparoscopic approach requires extensive unnecessary dissection of the rectum and involves a long common wall that exists between rectum and urethra [6]. Thus, they consider laparoscopy unnecessary for the repair of bulbar RUF but would use it for repairing prostatic RUF, a major change of heart that ensures that LAARP is the procedure of choice for treating MIA with RUF.

Creation of the pull-through canal

Accurate positioning of the pull-through canal, with pelvic floor muscles surrounding it symmetrically, is well recognized as the most important prognostic factor irrespective of the procedure performed. We believe that a potential rectal canal already exists when the anorectum develops. However, in patients with imperforate anus, the rectum does not develop normally within the pelvic floor muscles, and the potential rectal canal becomes filled with loose connective tissues and is obscured [7]. In other words, the potential rectal canal that develops in sync with the rectum, as well as the pelvic floor muscles in normal babies, is also present embryologically, even in cases of imperforate anus. According to the literature, the majority of surgeons insert a needle into the center of the external sphincter as a guide when creating a pull-through canal [8]. However, this "needle technique" may have a risk for incidental complications, such as injury to the urethra, prostate, or bladder, as well as risk for the needle to go outside the potential rectal canal, and furthermore, the pull-through route may be unnaturally straight in contrast to a natural anorectum that has a certain degree of curvature. Interestingly, Raschbaum et al. [9] recently reported magnetic resonance imaging-guided laparoscopic-assisted anorectoplasty for MIA with RUF, concluding that their technique results in anatomically correct placement of the rectum within the vertical muscle complex and noted that straight needle advancement techniques in LAARP could result in a deviation of the pulled-through rectum from the central muscle path. According to a recent systematic review of 17 reports in the English literature there was some confusion about how to assess outcome [5]. We too were initially very focused on ensuring that the pull-through canal was as central as possible and published some reports about intraoperative ultrasonography [10], and intraoperative muscle stimulation [11, 12], but in the end, we found using diagnostic imaging for confirming the position of the pull-through canal was unnecessarily tedious, given the fact that we came to realize that there is a residual plane in MIA patients that can be easily followed to position the pull-through canal [7]. Thus, by simply dissecting the pelvic floor bluntly following this loose connective tissue plane with a pair of mosquito forceps, a pull-through canal can be created with adequate anorectal angle that is compatible with the anorectal angle produced after a Pena procedure [13]. Our "mosquito technique" [7], which is essentially blind, will create a pull-through canal that is accurately placed if performed gently and can be confirmed at any time using intraoperative endosonography for the less experienced [10].

Several authors, including us, believe that the posterior sagittal incision is detrimental enough and has the potential to disrupt sphincter function to such an extent that a laparoscopic approach would seem less invasive, although to date, there have been no reports presenting clear evidence of such damage [6].

Postoperative radiologic findings

After LAARP with creation of the pull-though canal using the "mosquito technique", magnetic resonance image (MRI) and proctoscopic ultrasonography probe can be used to show that the anorectum is located in the center of the pelvic floor muscles and sphincter muscles, although they are quite thin [10, 14].

Wong et al. [15] reported post-operative MRI evaluation of children with imperforate anus after LAARP and demonstrated that LAARP resulted in less fibrosis and more sphincter symmetry than Pena procedure.

Assessing outcome

A recent systematic review of 17 studies in the English literature conducted by Al-Hozaim et al. [5] found there to be no standardized approach to assessing outcome. Fecal

continence, presence of rectal prolapse, position of the rectum, manometry, contrast enema, endosonography, and magnetic imaging have all been reported. One must be constantly aware when assessing postoperative outcome that the presence of associated anomalies such as deformities in the gastrointestinal tract and, more importantly, deformities of the sacrum can affect postoperative prognosis. Thus, in patients with cord tethering, sacral anomalies, mental retardation as associated etiologic factors, outcome is worse, and any accurate assessment of surgical intervention should separate such patients to generate a truer picture of the benefit of the surgical procedure performed.

Reports concluding that LAARP could either render better functional results than the Pena procedure or be similar to the Pena procedure [16–18] tend to involve only small numbers of cases, and the results are often not statistically significant. However, the most obvious difference between LAARP and the Pena procedure is that the pelvic floor muscles and sphincter muscles do not have to be divided, thus there should be less local trauma and better continence.

Recently, Yang et al. [19] used anorectal manometry to show that LAARP can significantly improve anal canal resting pressure although there were no significant differences to be noted on clinical scoring of the Pena and LAARP procedures. This was postulated to be related to there being less damage to pelvic floor muscles and sphincters associated with LAARP, in other words, a reflection of structural damage caused by the posterior sagittal incision. Tong et al. [18] also reported using anorectal vector volume manometry that compared with Pena procedure, lower asymmetric index, larger vector volume, and higher anal canal pressure at rest and during voluntary squeeze were observed in LAARP, which were statistically significant in all. Lin et al. [20] reported earlier appearance and higher incidence of the rectoanal relaxation reflex in 9 patients with high/intermediate imperforate anus repaired with LAARP, compared with 13 aged-matched patients repaired with the Pena procedure, concluding that in the early postoperative stage, patients repaired with LAARP had more favorable findings in anorectal manometry than patients repaired with Pena procedure.

We assessed parameters of surgical stress such as mean febrile period, duration of raised white blood cell count, and peak C-reactive protein level as indicators of outcome but found no significant differences between LAARP and Pena patients [21].

Our assessment of outcome is focused almost exclusively on fecal continence as a physiological marker of successful surgical intervention [21]. Our simplest technique is a continence evaluation questionnaire (CEQ) comprising five parameters each scored 0–2 to give a maximum score of 10. The parameters are: frequency of motions, severity of staining/soiling, severity of perianal erosions, anal shape, and requirement for medications. We compared LAARP cases with Pena cases and found that LAARP had higher scores consistently throughout the study but which were only statistically significant at 3 and 4 years after surgery.

Postoperative complications

Complications such as diverticulum formation, overcome by the Pena procedure, have been reported with increasing frequency after LAARP [3, 4, 22, 23] and are most likely related to incomplete excision of the RUF and, as a result, we must emphasize that complete excision, while technically challenging, is crucial, especially for bulbar RUF. However, most diverticulae are asymptomatic and the indication for resection is unknown, although risk for malignant transformation would suggest excision is prudent [23]. But there is no established convention at present.

During dissection of the fistula in boys with both prostatic and bulbar RUF, there is potential risk for posterior urethral diverticulum to develop because complete excision of any RUF is not easy, even if it is prostatic; if it is bulbar, it is even more difficult, as previously described [3], because the rectum and the prostate/urethra at the fistula share a common wall. Most pediatric surgeons tend not to dissect the distal part of an RUF aggressively, since they fear urethral injury/stenosis on dividing the fistula too close to the urethra, with a potential risk of a small diverticulum developing because of incomplete excision. To dissect the RUF more accurately, we previously reported the use of intraoperative colonoscopy during LAARP to observe the size of the RUF orifice and monitor progress of dissection distally [24]. Using a fine flexible endoscope introduced through an opening made in the anterior rectal wall, laparoscopic dissection could also be monitored intraluminally, assisting the laparoscopic surgeon to dissect the RUF closer toward the distal end. Another reported procedure involves subseromuscular dissection of the rectum up to where the fistula joins with the urinary tract during LAARP [25], however, based on our experiences, submuscular dissection often cause mucosal perforation of the RUF. And both these techniques could still cause injury to the urethra or still may leave excessive length of fistula behind because dissection progresses essentially by trial and error as the surgeon has no exact indication of how far to dissect between the rectal and urethral orifices, in other words, no landmark to use to judge how far to go distally. With this in mind, we thought of measuring the length of the RUF using a calibrated catheter [26]. Hardly a complicated concept, but the exact length of the RUF can be obtained under

direct vision, enabling the RUF to be dissected distally with confidence and without any risk for injury to genitourinary structures. Briefly, a fine flexible colonoscope is inserted into the anterior rectal wall to allow both the fistula orifice and the level of laparoscopic dissection to be observed intraluminally. Next, a fine calibrated catheter is inserted through the opening by the laparoscopic surgeon while another surgeon performing cystoscopy observes how far it emerges at or near the verumontanum. By doing so, the laparoscopic surgeon can measure the inside length of the fistula between the fistula opening and the urethral site orifice. If the length of the residual fistula is longer than 5 mm, the rectal end is dissected further toward the urethra using mucosectomy. This procedure is repeated until the length of the residual fistula is shorter than 5 mm. The fistula is then tied and excised. No matter how skilled, a surgeon will perform better with accurate landmarks; and with experience, a surgeon can develop a feel for how far to dissect. In all cases, we measured the length of the residual fistula and the length was longer than expected, ranging from 5 to 21 mm [26]. Interestingly, Rollins et al. [27] have reported that simple division of the RUF is safe and successful by dividing the RUF flush with the urethra without using ties; but extended urethral catheterization was required (6–40 days) compared with our mean of 4.8 [3–7] days, and cystourethrography was needed for removal of the urinary catheter and was repeated in some cases to confirm that there was no leakage of urine into the pelvic cavity after LAARP. We consider application of the Rollins approach to be risky on its own, but regard a combination of measurement and Rollins to be reasonable.

From experience, cases of bulbar RUF are more prone to diverticulum formation [3], however, we feel that bulbar RUF can be treated safely by LAARP if measurement of the fistula is performed intraoperatively as described above, although many centers do not consider bulbar RUF as an indication for LAARP. Prostatic RUF, however, is generally shorter and shallower than bulbar RUF and does not require intraoperative measurement of the fistula in the hands of experienced surgeons.

Homegrown refinements for treating bulbar RUF

Dissecting the rectum laparoscopically in MIA cases with bulbar RUF can be so difficult that surgeons are tempted to abandon dissection early, leaving the most distal part of the bulbar RUF behind with great likelihood of it becoming a posterior urethral diverticulum. To overcome such frustration with dissection, surgeons would benefit from: (1) refined trocar placement in bulbar RUF patients. The right and left trocars are placed much closer to the laparoscope so that their ends can reach the distal end of the bulbar RUF. (2) Using an EndoCAMeleon® (STORZ) device. This device allows the laparoscope to be adjusted from 0° to 120° intraoperatively, eliminating the need to choose a type of laparoscope in advance or be limited to a fixed view. (3) Insertion of a suprapubic catheter into the bladder with continuous suction of urine to decompress the bladder, improving exposure of the distal part of the bulbar RUF located deep in the pelvis. (4) Direct RUF measurement [24]. This has already been described in the previous section. Briefly, the RUF is dissected close to the urethra, opened, and a fine catheter with 10-mm calibrations is inserted until it emerges distal to the verumontanum under cystoscopic control. The distance from the point where dissection was ceased at the rectal end to the urethral orifice is measured. The RUF is further dissected free for exactly this length, tied and excised.

Primary laparoscopic repair of prostatic RUF

Vick et al. [28] reported in 2007 that three MIA with bladder neck fistulas and three MIA with prostatic RUF had primary laparoscopic repair on days 1–2 of life, and that all patients passed stools within the first 72 h of surgery. Although long-term follow-up will be needed to assess outcomes and continence rates, the avoidance of a colostomy is a major advantage of this technique. With this method, there are virtually no abdominal scars because it is a primary laparoscopic procedure and there is no risk for adhesions secondary to the colostomy.

Recommendations

Surgical intervention should be LAARP with intraoperative measurement of the RUF, with follow-up focused on bowel habit. Based on the results of a multicenter study comparing LAARP with other surgery [23], the most reliable investigation for detecting the presence of a diverticulum is MRI.

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