

The midterm outcomes of 1-stage versus 3-stage laparoscopic-assisted anorectoplasty in anorectal malformations with rectoprostatic fistula and rectobulbar fistula

A retrospective cohort study

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Abstract

The aim of this study was to compare the midterm outcomes of 1-stage and 3-stage surgical procedures to treat anorectal malformations (ARMs) with rectoprostatic and rectobulbar fistula using laparoscopic-assisted anorectoplasty (LAARP).

A total of 56 patients with ARMs and rectoprostatic and rectobulbar fistula who underwent LAARP from January 2011 to May 2014 in our institution were included in the study. They were divided into 2 groups according to the stage of procedure. The patients' data and postoperative complications were compared between the 2 groups. The Krickenbeck classification was used for assessing the bowel functions.

About 20 ARM newborns (rectoprostatic fistula [12], rectobulbar fistula [8]) successfully underwent a 1-stage LAARP, and about 36 ARM children (rectoprostatic fistula [20], rectobulbar fistula [16]) underwent a 3-stage LAARP (colostomy, LAARP, and closure of colostomy). The average age at the LAARP procedure in 1-stage group was significantly lower than that in 3-stage group (39.8 ± 8.1 hours vs 4.9 ± 1.2 months; $P = .00$). The average operative time during the definitive procedure was 132.2 ± 15.9 minutes in the 1-stage group and 120.5 ± 12.7 minutes in the 3-stage group ($P = .13$). There was only 5 to 10 mL of blood loss during the LAARP procedure both the groups ($P = .75$). There were no significant differences between the 2 groups in postoperative hospital stay during the definitive procedure (10.2 ± 2.3 days vs 8.5 ± 2.2 days; $P = .22$). The rate of surgical site infection and dehiscence was 5% (1/20) in the 1-stage group and 5.6% (2/36) in 3-stage group ($P = 1.00$). During the period of follow-up, the rate of voluntary bowel movement was 90% (18/20) in 1-stage group and 94.4% (34/36) in 3-stage group ($P = .94$). Free from soiling or grade I soiling was 80% (16/20) in 1-stage group and 83.3% (30/36) in 3-stage group ($P = 1.00$); grade II soiling was found in 3 (10%) patients in 1-stage group and 85.7% in 3-stage group ($P = .75$); grade III soiling was found in 3 (10%) patients in 1-stage group and 85.7% in 3-stage group ($P = 1.00$). Three patients (15%) in 1-stage group and 5 patients (13.9%) in 3-stage group suffered from grade I constipation ($P = 1.00$); while 3 (15%) patients in 1-stage group and 4 patients (11.1%) in 3-stage group had grade II constipation ($P = 1.00$); no patients in the 2 groups suffered from grade III constipation.

The 1-stage LAARP procedure for neonate with rectoprostatic and rectobulbar fistula can achieve comparable midterm outcomes as the conventional 3-stage LAARP procedure. It provides an alternative method to rectify the ARMs with rectoprostatic fistula and rectobulbar fistula without colostomy.

Abbreviations: ARMs = anorectal malformations, LAARP = laparoscopic-assisted anorectoplasty, SR = sacral ratio, SSI = surgical-site infection.

Keywords: anorectal malformations, 1-stage, single-incision laparoscopic anorectoplasty, 3-stage

1. Introduction

The conventional 3-stage anorectoplasty for the treatment of anorectal malformations (ARMs) that contain a divided colostomy, definitive operation, and colostomy closure has been widely

accepted.^[1,2] Because the existence of a colostomy stoma is beneficial to protect the definitive repair in the staged procedure. Meanwhile, the colostograms can be easily done through colostomy to recognize the location of fistula precisely, and it is

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also convenient to decompress the meconium through the distal colostomy. But with the advancement in imaging, scopes, and surgical technique, the procedure of 1-stage posterior sagittal anorectoplasty for the treatment of neonates with ARMs has been reported in many centers and achieved good clinical outcomes.^[3–5] The 1-stage laparoscopic-assisted anorectoplasty (LAARP) for neonates with rectoprostatic and rectobulbar fistula was utilized since 2013 in our institution.^[6] There is a paucity of literature on the application of 1-stage LAARP in other institutions for neonates with ARMs and the functional benefit of 1-stage LAARP remains unclear when in contrast to the procedure of 3-stage LAARP. Hence the purpose of this study is to compare the midterm outcomes of 1-stage and 3-stage LAARP procedures for the ARMs with rectoprostatic and rectobulbar fistula.

2. Materials and methods

2.1. Patients demographics

This was a single-centered retrospective study of 56 consecutive patients who had ARMs with a rectoprostatic fistula or a rectobulbar fistula in Capital Institute of Pediatric between January 2011 and May 2014. Twenty neonatal patients with ARM successfully underwent the procedure of 1-stage LAARP, and the remaining 36 patients underwent the procedure of 3-stage LAARP. There were no conversions in the both groups. The types of malformations were categorized according to the Krickbeck classification.^[7] The sacral ratio, age at operation, operative time, postoperative hospital stay, and complications were compared between the 2 groups. We have to claim that it is not entirely depends on us to perform 1-stage laparoscopic repair, as most of ARM newborns in rural areas undergo a colostomy before being transferred to our center. Hence only for those newborns transferred to us directly after birth, we can perform 1-stage laparoscopic corrections without colostomy. In addition, we think it is not suitable and safe to perform 1-stage LAARP for rectobladder neck fistula for the location of fistula is too high. In the 1-stage group, the preoperative invertograms, voiding cystourethrogram, and pelvic magnetic resonance imaging (MRI) were used as routine methods to identify the location of fistula, while the colostograms was the main method to distinguish the types of malformations in the 3-stage group. Ethical approval from the Ethics Committee of the Capital Institute of Pediatrics was obtained. Written informed consents were obtained from the parents prior to the surgery.

2.2. Bowel function evaluation

Functional results of patients who were older than 2 years in the 2 groups were analyzed. Bowel functions including presence of voluntary bowel movements, soiling and constipation, were evaluated according to the Krickbeck classification.^[7]

2.3. Follow-up

The patients were followed up in our clinic 1, 3, 6, and 12 months postoperatively and 6 months thereafter. Physical examination, pelvic MRI, and a voiding cystourethrogram were carried out.

2.4. Statistical analysis

The data were analyzed using SPSS 21.0 package. Student *t* test was used to compare the age at operation, operative blood loss, operative time, and postoperative hospital stay between the 2 groups. Chi-squared tests were applied to compare the postoperative complications and postoperative bowel functions

between the 2 groups. $P < .05$ was considered to be statistically significant.

3. Results

3.1. Surgical characteristics of the 2 groups

The age when the LAARP procedure was performed for the 1-stage group was 39.8 ± 8.1 hours (30–52 hours) and the age for the 3-stage group at time of LAARP procedure was 4.9 ± 1.2 months (3–7 months). There was statistically significant difference in regard to the average ages during the LAARP procedure between the 2 groups ($P = .00$). The 1-stage group took 132.2 ± 10.2 minutes to complete and the 3-stage group took 113.5 ± 12.7 minutes to complete the definitive surgery ($P = .13$). There was only 5 to 10 mL of blood loss during the LAARP procedure both the groups ($P > .05$). The hospital stay for the 1-stage group was 10.2 ± 2.3 days and the hospital stay for the definitive operation in the 3-stage group was 8.5 ± 2.2 days ($P = .22$). The detailed items are summarized in Table 1. The associated anomalies between the 2 groups are listed in Table 2. The spinal anomalies (includes partial sacral agenesis, tethered cord, and spinal bifida occulta), cardiothoracic anomalies, and genitourinary anomalies (including vesicoureteral reflux, hydronephrosis, hypospadias, and undescended testis) of the 2 groups were similar ($P > .05$).

3.2. Complications between the 2 groups

The rate of surgical site infection (SSI) and dehiscence was 5% (1/20) in 1-stage group and 5.6% (2/36) in 3-stage group ($P = 1.00$), and the SSI involves only skin and subcutaneous tissue of incision in the 2 groups, it can be treated conservatively without reoperation. Rectal prolapse was found in 10% (2/20) of patients in 1-stage group, which is similar to that 8.3% (3/36) in 3-stage group ($P = 1.00$). The complications associated with colostomy and colostomy closure were (16.7%, 6/36) in the 3-stage group. The detailed information are listed in Table 3. No patients experienced recurrent fistula and urethral diverticulum in either group according to postoperative voiding cystourethrogram and MRI. Postoperative MRI verified the centrally placed rectum within the pelvic muscle complex, no asymmetrical muscle complex seen in MRI (Fig. 1).

3.3. Functional results between the 2 groups

The functional results of the 2 groups are summarized in Table 4. All the patients were followed up for more than 2 years, because only for the patients that older than 2 years can we make an

Table 1
Numbers of patients that associated anomalies in the 2 groups.

Associated anomalies	1-stage (20)	3-stage (36)
Vesicoureteral reflux	2	5
Undescended testis	1	5
Hypospadias	1	2
Spinal bifida occulta	2	4
Hydronephrosis	2	6
Cardiac anomalies	10	15
Partial sacral agenesis with four remaining sacral vertebrae	4	5
Tethered cord	0	1

Table 2

Patients and surgical characteristics of the 2 groups.

	1-stage group (20), mean ± SD	3-stage group (36), mean ± SD	P
Sacral ratio	0.53 ± 0.19	0.61 ± 0.12	.54
Rectoprostatic fistula	12	20	.75
Rectobulbar fistula	8	16	.75
Age at definitive operation, mo	39.8 ± 8.1 (30–52 h)	4.9 ± 1.2 (3–7 mo)	<.001
Minutes for definitive operation, min	132.2 ± 15.9	120.5 ± 12.7	.13
Blood loss during definitive operation, mL	9.3 ± 1.7	8.5 ± 1.8	.75
Postoperative hospital stay during definitive surgery, d	10.2 ± 2.3 (7–13 d)	8.5 ± 2.2 (5–12)	.22

relatively accurate assessment about the bowel function. The median follow-up period is 39 months (range 24–50 months) in the 1-stage group and the median follow-up period is 45 months (range 30–70 months) in the 3-stage group. The rate of voluntary bowel movement was 90% (18/20) in the 1-stage group, while it was 94.4% (34/36) in the 3-stage group (*P* = .94). Free from soiling or grade I soiling was 80% (16/20) in the 1-stage group and 83.3% (30/36) in the 3-stage group (*P* = 1.00); grade II soiling was 15% (3/20) in the 1-stage group and 8.3% (3/36) in the 3-stage group (*P* = .75); grade III soiling was 5% (1/20) in the 1-stage group and 8.3% (3/36) in the 3-stage group (*P* = 1.00). Three patients (15%) in the 1-stage group and 5 patients (13.9%) in the 3-stage group suffered from grade I constipation (*P* = 1.00), while 3 (15%) patients in the 1-stage group and 4 patients (11.1%) in the 3-stage group had grade II constipation (*P* = 1.00); no patients in the 2 groups suffered from grade III constipation. Throughout the follow-up period, the functional results improve with time, both the 2 groups appear to deliver better results.

4. Discussion

The LAARP has been increasingly adopted in the management of patients with ARMs, and the conventional LAARP procedure is divided into 3 stages of colostomy, LAARP, and followed colostomy closure. The conventional laparoscopic correction of ARM with rectoprostatic fistula and rectobulbar fistula has been utilized in many centers and confirmed to be an effective and safe method during the past few years.^[8,9] However, it still remains controversy whether the 1-stage or the 3-stage LAARP should be adopted to treat this anomaly. A host of pediatric surgeons harbor the idea that the formation of initial colostomy for the ARM children, because it can not only solve the problem of decompression in the neonatal period, but also protect the subsequent definitive operation.^[10,11] Meanwhile, the types of malformations and the precise location of fistula can be easily

recognized through a good distal colostogram. On the contrary, there are several reasons behind the preference of 1-stage LAARP procedure, for it can prevent repeated anesthesia and surgeries and the high complications associated with colostomy and colostomy closure. But apart from that we maybe more concerned with the bowel function after definitive procedure. Hence, the purpose of this study was to compare the midterm outcomes of 1-stage and 3-stage LAARP to treat ARMs with rectoprostatic and rectobulbar fistula.

The meconium cannot defecate through the fistula because the type of malformations was either rectoprostatic or rectobulbar fistula, but no perforations occurred in the 1-stage group in our study. Our experience suggest that the definitive surgery should be performed as soon as possible, usually within 48 hours after birth, because the obstructive symptoms will be aggravated in neonates with rectoprostatic fistula or rectobulbar fistula if not be treated promptly. In our study, the age at operation in the 1-stage group was 39.8 ± 8.1 (30–52 hours).

The SSI is a common postoperative complication. SSI is often defined as superficial or deep. The superficial incisional SSI involves only skin and subcutaneous tissue of incision. The deep incisional SSI involves deep tissues, such as facial and muscle layer. It has been widely accepted that the 3-stage repair procedure is associated with lesser risk of SSI because of fecal diversion by the colostomy. In our study, the rate of SSI and

Table 3

The postoperative outcomes and complications between the 2 groups.

	1-stage group (20)	3-stage group (36)	P
SSI/dehiscence, n (%)	5% (1/20)	5.6% (2/36)	1.00
Rectal retraction, n (%)	0	0	—
Recurrent fistula, n (%)	0	0	—
Urethral diverticulum, n (%)	0	0	—
Anal stenosis, n (%)	0	0	—
Rectal prolapse, n (%)	10% (2/20)	8.3% (3/36)	1.00
Colostomy complications	0	6/36 (16.7%)	.08

SSI = surgical site infection.

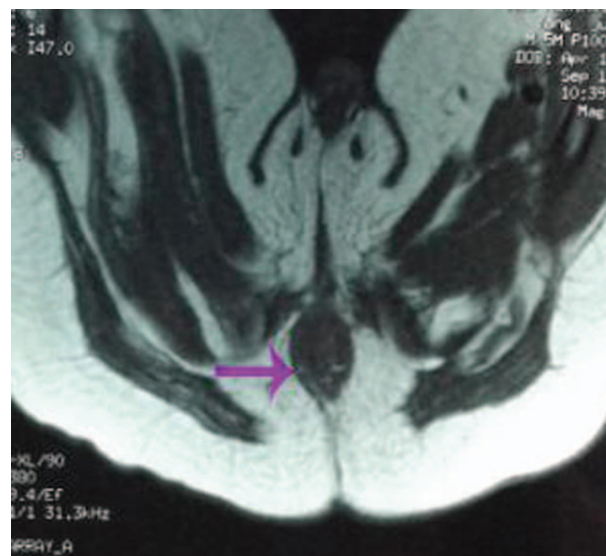


Figure 1. Postoperative magnetic resonance imaging (MRI) verified the centrally placed rectum within the pelvic muscle complex, no asymmetrical muscle complex seen in MRI.

Table 4
Functional results according to Krickenbeck score system.

	1-stage group (20)	3-stage group (36)	P
Follow-up period (median, range)	39 months (24–50 months)	45 months (30–70 months)	.71
Voluntary bowel movements	90% (18/20)	94.4% (34/36)	.94
Soiling			
Grade I (occasionally)	80% (16/20)	83.3% (30/36)	1.00
Grade II (every day)	15% (3/20)	8.3% (3/36)	.75
Grade III (constant)	5% (1/20)	8.3% (3/36)	1.00
Constipation			
Grade I (manageable by changes in diet)	15% (3/20)	13.9% (5/36)	1.00
Grade II (required laxatives)	15% (3/20)	11.1% (4/36)	1.00
Grade III (resistant to laxatives and diet)	0	0	–

dehiscence was 5% (1/20) in the 1-stage group and 5.6% (2/36) in the 3-stage group ($P = 1.00$). But the SSI involves only skin and subcutaneous tissue of incision in the 2 groups, it can be cured conservatively without reoperation.

In our study, rectal prolapse was found in 10% (2/20) of patients in the 1-stage group, which is similar to that 8.3% (3/36) in the 3-stage group ($P = 1.00$). Rectal prolapse has been reported to be 8.8% to 46% following LAARP.^[12,13] Previous studies of ARMs suggest that the patients with higher malformations with sacral agenesis and pelvic musculature were more likely to suffer from rectal prolapse.^[14] Among the 5 patients who underwent rectal prolapse in our study, 3 patients had partial sacral agenesis. To prevent the occurrence of rectal prolapse, the distal rectum should not be dissected excessively. In addition, it is overwhelmingly important to pull the rectum through the center of the striated muscle complex without any resistance, thus it can effectively reduce the incidence of anorectal retraction or stricture. The tunnel of sphincter muscle complex should better be moderate enough to allow the rectum to be pulled through without any resistance; otherwise, it can easily result in narrowing not only at the anocutaneous junction but also at the deeper level.

No patients suffered from injury of urethra during operations in the 2 groups. No patient experienced recurrent fistula and urethral diverticulum according to voiding cystourethrogram and MRI in the 2 groups. MRI showed a centrally placed rectum within the muscle complex in all cases. However, there exists a major problem that the complications associated with colostomy and colostomy closure were very high in the 3-stage group (16.7%, 6/36).

And above all, bowel functional results were the most important for patients with ARMs after anorectoplasty. Our results showed that the rate of voluntary bowel movement was 90% (18/20) in the 1-stage group and 94.4% (34/36) in the 3-stage group ($P = .94$). Grade III soiling was 5% (1/20) in the 1-stage group, while it was slightly lower than that in the 3-stage group (8.3%, 3/36; $P = 1.00$). No patients in the 2 groups suffered from grade III constipation. After the treatment such as counseling, toilet training, and dietary modification, most patients especially in the 1-stage group have experienced striking improvement of soiling. There was no significant difference between the 2 groups in regard to the bowel function ($P > .05$). In addition, as functional results improve with time, both groups appear to deliver better outcomes. Hence, it is indispensable that the long-term follow-up study should be conducted to observe the development of functional results.

It is well known that the level of the anomaly impacts on the continence prognosis. Although different repair procedure for

ARMs may lead to differences in functional outcomes, the primary differences in outcome are often attributed to the type of malformation, location of fistula, and quality of the sacrum and spine. As there were seldom patients with rectoprostatic and rectobulbar fistula accompanied with severe sacral abnormalities in the 2 groups, and the type of malformations and laparoscopic procedure between the 2 groups were similar. Hence, the functional outcomes in the 2 groups mainly be attributed to the timing of intervention in the management of ARMs.

Several drawbacks also existed in our study. First, the results may have confounding bias for its retrospective nature. Second, the Krickenbeck classification for postoperative results were notoriously inaccurate because it mainly obtained from patients' parents and is inevitably subjective. The absence of standardized recommendations for follow-up study is the greatest limiting factor in definitively comparing 1-stage and 3-stage LAARP. So the Krickenbeck score combined with clinical, manometric, and radiologic studies should be conducted to evaluate the bowel function comprehensively.

5. Conclusion

The 1-stage LAARP procedure for neonate with rectoprostatic and rectobulbar fistula can achieve comparable midterm outcomes as the conventional 3-stage LAARP procedure. It provides an alternative method to rectify the ARMs with rectoprostatic fistula and rectobulbar fistula without colostomy.

Author contributions

Authors' contributions: Hui Xiao carried out the entire procedure including the literature search, data extraction, performed the statistical analysis, drafted the manuscript, revised submitted the manuscript. Long Li, Mei Diao, and Wei Cheng conceived of the study, coordinated and participated in the entire process of drafting and revised the manuscript. Rui Huang, and Long Chen contributed to statistical analysis and revision the manuscript. Xiao-Dai Cui, Wei Cheng and Long Li contributed to the revisions of the manuscript. All authors have contributed significantly. All authors read and approved the final manuscript.

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References

- [1] Peña A, De Vries P. Posterior sagittal anorectoplasty: important technical considerations and new applications. *J Pediatr Surg* 1982;17:796–811.
- [2] Peña A, Hong A. Advance in the management of anorectal malformations. *Am J Surg* 2000;180:370–6.
- [3] Liu G, Yuan J, Geng J, et al. The treatment of high and intermediate anorectal malformations: one stage or three procedures? *J Pediatr Surg* 2004;39:1466–71.
- [4] Adeniran JO. One-stage correction of imperforate anus and rectovestibular fistula in girls: preliminary results. *J Pediatr Surg* 2002;37:E16.
- [5] Upadhyaya VD, Gopal SC, Gupta DK, et al. Single stage repair of anovestibular fistula in neonate. *Pediatr Surg Int* 2007;23:737–40.
- [6] Diao M, Li L, Ye M, et al. Congenital anomaly rectified at birth: one-stage single-incision laparoscopic-assisted anorectoplasty for newborns with anorectal malformations and recto-urethral fistula. *Surg Endosc* 2016;30:5156–64.
- [7] Holschneider A, Hutson J, Peña A, et al. Preliminary report on the International Conference for the Development of Standards for the Treatment of Anorectal Malformations. *J Pediatr Surg* 2005;40:1521–6.
- [8] Sydorak RM, Albanese CT. Laparoscopic repair of high imperforate anus. *Semin Pediatr Surg* 2002;11:217–25.
- [9] Georgeson KE, Inge TH, Albanese CT. Laparoscopically assisted anorectal pull through for high imperforate anus—a new technique. *J Pediatr Surg* 2000;35:927–30.
- [10] Georgeson K. Laparoscopic-assisted anorectal pull-through. *Semin Pediatr Surg* 2007;16:266–9.
- [11] Heinen F. The surgical treatment of low anal defects and vestibular fistulas. *Semin Pediatr Surg* 1997;6:204–16.
- [12] Podevin G, Petit T, Mure PY, et al. Minimally invasive surgery for anorectal malformation in boys: a multicenter study. *J Laparoendosc Adv Surg Tech A* 2009;19(Suppl 1):S233–5.
- [13] Kudou S, Iwanaka T, Kawashima H, et al. Midterm follow-up study of high-type imperforate anus after laparoscopically assisted anorectoplasty. *J Pediatr Surg* 2005;40:1923–6.
- [14] Belizon A, Levitt M, Shoshany G, et al. Rectal prolapse following posterior sagittal anorectoplasty for anorectal malformations. *J Pediatr Surg* 2005;40:192–6.