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ORIGINAL ARTICLE

Single-incision laparoscopic-assisted anorectoplasty for treating children with intermediate-type anorectal malformations and rectobulbar fistula: a comparative study

Xianghai Ren[1,2] · Hang Xu[2] · Qi Jiang[3] · Mei Diao[2] · Xu Li[2] · Long Li[1,2]

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Abstract

Purpose Single-incision laparoscopic-assisted anorectoplasty (SILAARP) was compared to posterior sagittal anorectoplasty (PSARP) for treating intermediate-type anorectal malformations (ARMs) with rectobulbar fistula.

Methods Between December 2011 and January 2016, 48 children with intermediate-type ARMS were treated with SILAARP ($n=34$) or PSARP ($n=14$) in our centre. Data including demographics, complications, and long-term outcomes were retrospectively compared.

Results No significant difference was observed between both groups in terms of median operative time and complications. The length of postoperative hospital stay was shorter in the SILAARP group than in the PSARP group (6.15 ± 1.10 vs 9.64 ± 4.13 days; $p=0.008$). After a mean follow-up of 59.38 ± 13.68 months, the rates of voluntary bowel movements, soiling, and constipation were similar in both groups. Anorectal manometry was performed in 15 and 7 children from the SILAARP and PSARP groups, respectively. Although there were no significant differences in the presence of rectoanal relaxation reflex and high-pressure-zone length, anal canal resting pressure was higher in the SILAARP group than in the PSARP group (33.35 ± 12.95 vs 23.06 ± 8.40 mmHg; $p=0.039$).

Conclusions Both SILAARP and PSARP seemed feasible and effective for treating intermediate-type ARMs with rectobulbar fistula in children. However, SILAARP significantly reduced the length of postoperative hospital stay and improved anal canal resting pressure.

Keywords Laparoscopy-assisted anorectoplasty · Posterior sagittal anorectoplasty · Anorectal malformations · Levator ani · Terminal rectum

Introduction

Anorectal malformations (ARMs) are one of the most common congenital gastrointestinal anomalies [1]. A recent study showed that the incidence of ARMs from 1973 to 2014 was 1 in 3000 live births [2]. ARMs are divided into high, intermediate, and low types based on the positional relationship between levator ani and the terminal rectum. This classification can effectively guide the choice of surgical approaches and remains widely accepted by paediatric surgeons.

For the treatment of high/intermediate ARMS, posterior sagittal anorectoplasty (PSARP), introduced by Peña and deVries, is regarded as a standard operation [3]. Laparoscopic-assisted anorectoplasty (LAARP) is another commonly used procedure for ARMs since it was first reported by Georgeson et al. in 2000 for treating high-type ARMs

Xianghai Ren and Hang Xu contributed equally to this work.

✉ Long Li
lilong3@hotmail.com

1 Graduate School of Peking Union Medical College, Chinese Academy of Medical Sciences, Beijing 100730, People's Republic of China

2 Department of Pediatric Surgery, Capital Institute of Pediatrics, No. 2 Yabao Road, Chaoyang District, Beijing 100020, People's Republic of China

3 Department of Pathology and Pathophysiology, Hubei Provincial Key Laboratory of Developmentally Originated Disease, School of Basic Medical Sciences, Wuhan University, Wuhan 430071, Hubei, People's Republic of China

[4]. It is becoming increasingly adopted for selected patients with ARM. For some high-type ARMs, it might be difficult to use PSARP alone for definitive malformation repair without entering the abdomen. Therefore, it is generally accepted that LAARP is an effective and safe option for children with high-type anomalies [5–7]. Nevertheless, studies have shown that LAARP appears to be associated with the formation of a urethral diverticulum when performed for patients with ARMs whose rectum is at the bulbar level [8, 9]. As a minimally invasive type of LAARP, single-incision laparoscopic-assisted anorectoplasty (SILAARP) has been used to treat ARMs [10, 11]. However, it is unclear whether SILAARP is suitable for intermediate-type ARMs with rectobulbar fistula. This study aimed to compare long-term outcomes between SILAARP and PSARP for children with intermediate ARMs and rectobulbar fistula.

Materials and methods

Patient demographics

The records of all patients with ARMs treated from December 2011 and January 2016 in our hospital were reviewed. The pubococcygeal line (PCL) and the parallel line from the lowest point of the ischial tuberositym (I-line) were used for classification [12, 13]. In intermediate-type ARM, the patient's rectal pouch lies between PCL and I-line (Fig. 1). The inclusion criteria for this study were as follows: patients with ARM who (1) underwent SILAARP or PSARP in our centre; (2) had intermediate-type ARMs with rectobulbar fistula; and (3) had colostomy was performed before anorectoplasty, and those (4) whose parents agreed to their completing this study. The exclusion criteria were as follows: patients with (1) any contraindication for SILAARP or PSARP and (2) other surgical procedures performed on the anus or rectal before anorectoplasty. Ultimately, 48 patients were available for analysis, of which 34 patients were allocated to the SILAARP group and 14 patients were assigned to the PSARP group. Radiography of the sacrococcygeal region, micturating cystography, pelvic magnetic resonance imaging (MRI) and cardiovascular ultrasound was performed to determine the type of ARMs and associated anomalies.

In the SILAARP group, the mean age of patients at the time of anorectoplasty was 5.01 ± 1.57 months. Associated anomalies were present in 16 patients (47.1%), including spinal bifida occulta (6 patients, 17.6%), vesicoureteral reflux (4 patients, 11.8%), patent foramen ovale (4 patients, 11.8%), patent ductus arteriosus (3 patients, 8.8%), partial sacral agenesis with four remaining sacral vertebrae (3 patients, 8.8%), mild scoliosis (3 patients, 8.8%), hydronephrosis (2 patients, 5.9%), unilateral kidney (2 patients, 5.9%), renal malrotation (2 patients, 5.9%), megaureter (2 patients, 5.9%), and renal duplication (1 patient, 2.9%). Multiple anomalies were detected in 7 patients.

Fig. 1 Radiological image of a child with intermediate-type anorectal malformations and rectobulbar fistula

In the PSARP group, the mean age of patients at the time of anorectoplasty was 5.21 ± 1.74 months. Associated anomalies were present in 8 patients (57.1%), including spatial sacral agenesis with four remaining sacral vertebrae (4 patients, 28.6%), spina bifida occulta (2 patients, 14.3%), vesicoureteral reflux (2 patients, 14.3%), patent foramen ovale (2 patients, 14.3%), patent ductus arteriosus (2 patients, 14.3%), and solitary kidney (1 patient, 7.1%). Multiple anomalies were detected in 4 patients (28.6%).

Ethical approval was obtained from the Ethics Committee of the Capital Institute of Pediatrics. Written informed consents were obtained from the parents before anorectoplasty.

Surgical technique

The procedure of SILAARP has already been described [14, 15]. Briefly, an umbilical incision was performed and stretched horizontally for the insertion of a 5-mm 30° laparoscope and two 3-mm lateral ports. A transabdominal retraction suture was placed through the posterior bladder or uterine wall to expand the operating field. Laparoscopic circumferential rectal dissection was performed from the peritoneal reflection to the fistula. The terminal rectal pouch was dissected along the submucosal layer using a 3-mm cautery hook, 0.5 cm proximal to the posterior urethra wall. The mucosa of the fistula was completely removed. Thereafter, the fistula was ligated with 5-0 PDS horizontal running

suture. A 1-cm midline skin incision was made at the site of the future anal orifice and the centre of the external sphincter was mapped with a transcutaneous electro-stimulator under laparoscopic guidance. Next, a ' 'potential tunnel'' penetrating the centre of the sphincter complex to the pelvis was created and dilated up to about 12 mm. The separated rectal pouch was pulled down through the muscle complex to the anal site. The megarectosigmoid with weak peristaltic function was resected. Anastomosis was performed without tension using 5-0 PDS sutures. The PSARP procedure was performed as previously described [16]; the centre of the external sphincter and the appropriate anal site were determined using a transcutaneous electro-stimulator. Surgeries in both groups were performed by a fixed surgical team.

Anorectal dilation was started at 2 weeks after surgery and lasted for at least 3 months. The size of the dilator was gradually increased until it reached a suitable size. The colostomy was closed after the anus had reached the desired size.

Follow-up

All children were followed up in our clinic at 1, 3, 6, and 12 months postoperatively and every 6 months, thereafter. Before colostomy closure, physical examination, distal colostography, and micturating cystography were routinely performed. The pelvic magnetic resonance imaging was performed in all patients to assess the position of the rectum within the muscle complex. Perioperative data, including age at surgery, operative time, time to resumption of diet, length of hospital stay, and intraoperative complications were evaluated. Children's postoperative complications and postoperative bowel functions (Krickenbeck classification) were retrospectively reviewed to evaluate the long-term outcome of each procedure [13]. As a non-mandatory examination, anorectal manometry with an anorectal pressure monitoring system (Solar GI; Medical Measurement Systems, Dover, NH) was also performed to evaluate postoperative anorectal function. After an oral dose of 6% chloral hydrate (1 ml/kg), patients were sedated and the rectal anal inhibitory reflex (RAIR), anal canal resting pressure (ACRP), and high-pressure-zone length (HPZL) were evaluated (Fig. 2).

Statistical analyses

All statistical analyses were performed using SPSS version 17.0 software (SPSS, Chicago, IL). Continuous variables are reported as mean ± SD, while prevalence is presented as percentages. The Student's *t* test, Chi-squared test, Mann–Whitney *U* test, or Fisher's exact test was used when appropriate to compare characteristics between the groups. All *p* values were derived from two-tailed analyses, with significance accepted at $p < 0.05$.

Results

The demographic and clinical characteristics of the two groups were comparable with respect to age, sacral ratio (SR), and associated anomalies (Table 1).

Surgeries were successful in all cases and intraoperative blood loss was minimal. No patient in the SILAARP group required an additional extra-incisional trocar. MRI showed a centrally placed rectum within the muscle complex in all patients of the two groups.

As shown in Table 1, there was no statistically significant differences between the SILAARP and PSARP groups in terms of median operating time (96.79 ± 12.23 vs 104.71 ± 18.78 min; $p = 0.163$), transfusion rate (0% for both; $p > 0.99$), intraoperative complications (0% for both; $p > 0.99$), or time to resumption of diet (10.35 ± 2.24 vs 9.36 ± 2.50 h; $p = 0.210$). However, the postoperative hospital stay in the LAARP group was significantly less than that in the PSARP group (6.15 ± 1.10 vs 9.64 ± 4.13 days; $p = 0.008$).

Postoperative complications did not differ significantly between both groups (Table 1). Two children (14.3%) in the PSARP group developed wound infection on the fifth and seventh days after surgery and successfully recovered with conservative therapy (antibiotics and dressing). One child (7.1%) in the PSARP group developed mild mucosal prolapse, whereas, in the SILAARP group, mucosal prolapse was noted in six children (17.6%), but none required surgical intervention. Among these, children with sacral and/or spinal malformations showed a higher incidence of mucosal prolapse compared with those without sacral and/or spinal malformations. However, the difference was not statistically significant (27.8% vs 6.7%; $p = 0.113$). There was no rectal retraction, port-site hernia, recurrent fistula, urethral diverticulum, acquired anorectal atresia, or urethral injury in either group.

The postoperative bowel function was assessed with the Krickenbeck classification. The mean follow-up time for all patients was 59.38 ± 13.68 months; 59.65 ± 12.62 months in the SILAARP group and 58.71 ± 16.50 months in the PSARP group ($p = 0.851$). The mean age at the time of evaluation for patients in the SILAARP and PSARP groups was 64.69 ± 13.03 and 63.93 ± 16.72 months ($p = 0.881$), respectively. At this time, all children were beyond 3 years old and had finished potty training.

There were no significant differences with respect to voluntary bowel movement, soiling, and constipation between the two groups (Table 2). The rate of voluntary bowel movement in SILAARP group was 94.1%, while in the PSARP group, it was 85.7%. Patients presenting with Grade 3 soiling were considered to have faecal incontinence; however, no patient in either group had this grade

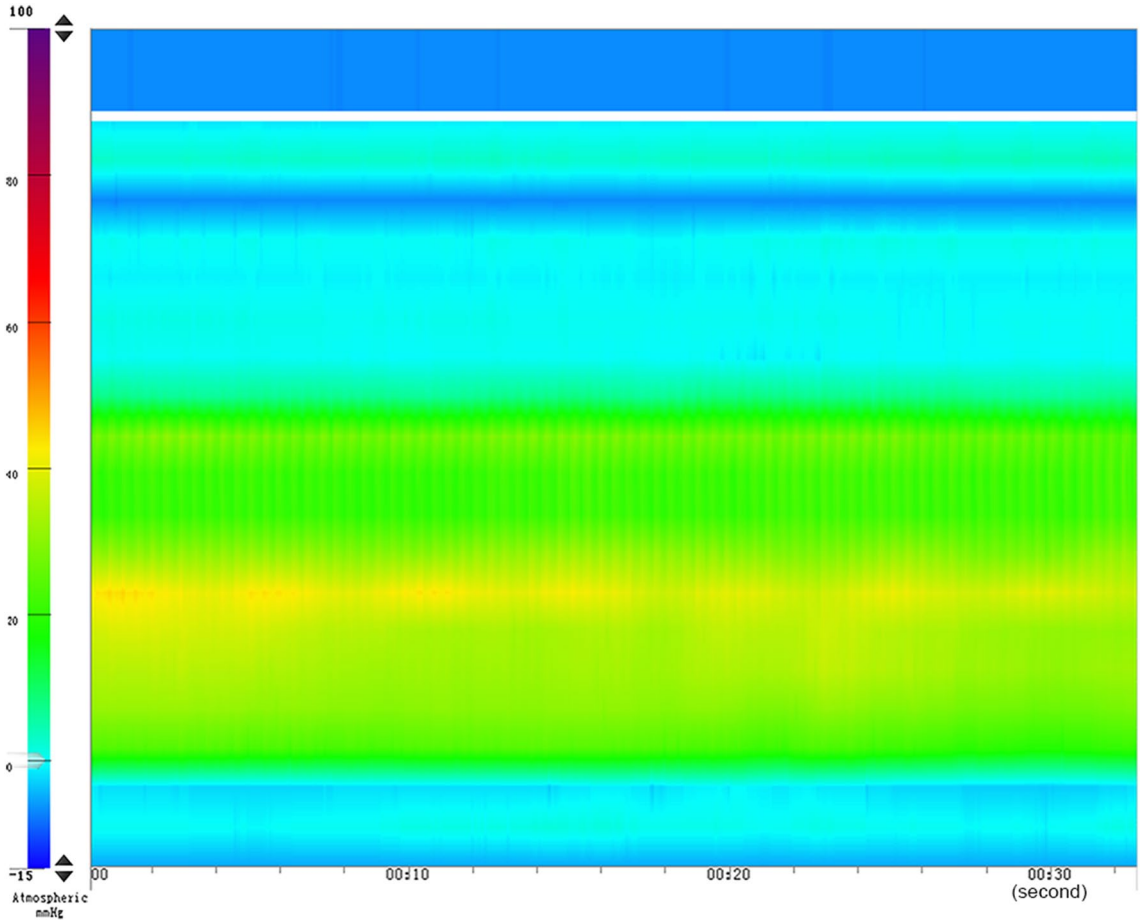

Fig. 2 The high resolution manometry image (anal canal resting pressure)

Table 1 Patient characteristics, perioperative variables and complications between the two groups

Parameter	SILAARP ($n=34$)	PSARP ($n=14$)	p
Age (months)	5.01 ± 1.57	5.21 ± 1.74	0.710
Sacral ratios	0.59 ± 0.13	0.61 ± 0.13	0.684
Associated anomalies			
Lumbosacral anomalies	11 (53.8)	6 (42.9)	0.719
Urologic abnormalities	4 (11.8)	3 (21.4)	0.680
Cardiovascular anomalies	4 (11.8)	2 (14.3)	1.000
Operating time (min)	96.79 ± 12.23	104.71 ± 18.78	0.163
Transfusion	0 (0)	0 (0)	1.000
Intraoperative complication	0 (0)	0 (0)	1.000
Time to resume diet (h)	10.35 ± 2.24	9.36 ± 2.50	0.210
Postoperative hospital stay (days)	6.15 ± 1.10	9.64 ± 4.13	0.008
Complications			
Wound infection	0 (0)	2 (14.3)	0.081
Mucosal prolapse	6 (17.6)	1 (7.1)	0.626
Urethral diverticulum	0 (0)	0 (0)	1.000
Recurrent fistula	0 (0)	0 (0)	1.000

Data are presented as the mean ± SD or number (percentage)

PSARP posterior sagittal anorectoplasty, *SILAARP* single-incision laparoscopic-assisted anorectoplasty

Table 2 Functional results (Krickenbeck classification) of patients between the two groups

Parameter	SILAARP ($n=34$)	PSARP ($n=14$)	p
Voluntary bowel movements	32 (94.1)	12 (85.7)	0.702
Soiling			0.370
No	28 (82.4)	10 (71.4)	
Grade 1	4 (11.8)	2 (14.3)	
Grade 2	2 (5.9)	2 (14.3)	
Grade 3	0 (0)	0 (0)	
Constipation			0.347
No	24 (70.6)	8 (57.1)	
Grade 1	7 (20.6)	4 (28.6)	
Grade 2	3 (8.8)	1 (7.1)	
Grade 3	0 (0)	1 (7.1)	

Data are presented as number (percentage)

PSARP posterior sagittal anorectoplasty, *SILAARP* single-incision laparoscopic-assisted anorectoplasty

of soiling. Although obvious incontinence was not found in these two groups, daily or occasionally soiling was detected in 6 patients (17.6%) in the SILAARP group and in 4 patients (28.6%) in the PSARP group. A total of 16 patients (10 patients [29.4%] in the SILAARP group and 6 patients [42.9%] in the PSARP group) developed varying degrees of constipation. In all patients in the SILAARP group and 5 patients in the PSARP group, symptoms could be alleviated through laxative and diet. The remaining one patient (SR = 0.38) with rectobulbar fistula and spina bifida occulta in the PSARP group was nearly four years old and required enema therapy to assist defaecation.

Anorectal manometry was performed in 15 patients from the SILAARP group and 7 patients from the PSARP group (Table 3). There were no significant differences regarding age at examination, SR, and associated anomalies between the two groups. ACRP was significantly higher in the SILAARP group than in the PSARP subgroup (33.35 ± 12.95 vs 23.06 ± 8.40 mmHg; $p=0.039$). However, there was no significant difference between the two subgroups with respect to the rate of positive RAIR (86.7% vs 100%; $p>0.99$) and HPZL results (17.60 ± 3.64 vs16.79 ± 4.36 mm; $p=0.677$).

Discussion

From the simple incision of a thin anal membrane in the second century AD to the first proctoplasty performed by suturing the opened rectal atresia to the perineum in 1835, paediatricians have attempted to treat children with ARMs for centuries. In the modern era, PSARP was considered as a new landmark procedure and gained wide acceptance [3, 16]. As an excellent alternative to PSARP, LAARP has been adopted in many paediatric centres treating high and intermediate-type ARMs [4–7]. Three control studies in early 2010 showed that both LAARP and PSARP could successfully treat high and intermediate-type ARMs with, at least, similar outcomes [17–19]. However, these observations failed to focus on comparisons between groups with intermediate ARMs. A more recent study from Tainaka et al. compared 2 patients with intermediate-type ARMs and rectobulbar fistula that underwent LAARP to 6 patients that underwent PSARP. They concluded that the faecal continence after LAARP was comparable to that after PSARP [20]. Because of the low number of patients in the LAARP group, this report seemed to have insufficient power to allow firm conclusions. Hence, we conducted a retrospective

Table 3 Results of anorectal manometry between the two groups

Parameter	SILAARP ($n=15$)	PSARP ($n=7$)	p
Age at anorectoplasty (months)	5.13 ± 1.71	5.50 ± 1.78	0.651
Age at examination (months)	29.20 ± 10.21	32.07 ± 10.54	0.559
Sacral ratios	0.59 ± 0.14	0.61 ± 0.12	0.682
Associated anomalies			
Lumbosacral anomalies	5 (33.3)	3 (42.9)	1.000
Urologic abnormalities	1 (6.7)	1 (14.3)	1.000
Cardiovascular anomalies	2 (13.3)	0 (0)	1.000
RAIR			1.000
+	13 (86.7)	7 (100)	
−	2 (13.3)	0 (0)	
ACRP (mmHg)	33.35 ± 12.95	23.06 ± 8.40	0.039
HPZL (mm)	17.60 ± 3.64	16.79 ± 4.36	0.677

Data are presented as the mean ± SD or number (percentage)

ACRP anal canal resting pressure, *HPZL* high-pressure zone length, *PSARP* posterior sagittal anorectoplasty, *RAIR* rectal anal inhibitory reflex, *SILAARP* single-incision laparoscopic-assisted anorectoplasty

study to compare the long-term outcomes of SILAARP and PSARP in patients with intermediate-type ARMs and rectobulbar fistula.

Although a new international classification simplifies the process of differentiating the type of ARMs, the Wingspread classification is still useful for guiding clinical work [13]. The latter distinguishes different ARM types according to PCL and the lowest point of the ischial tuberosity, and according to the levator ani muscles roughly located in the position between PCL and I-line [12, 13]. Traditionally, repairing intermediate-type malformations seems more suitable with PSARP, mainly because a laparoscopic procedure appears to be associated with the risk of inadequate exposure of the deep pelvic cavity and inconvenient dissection of the rectal pouch at the level of levator ani. In our study, however, SILAARP was successfully preformed for intermediate-type ARMs without conversions and with a similar operative time compared to PSARP. These evidences seem to demonstrate that the SILAARP is also a feasible technique for treating children with intermediate-type ARMs and rectobulbar fistula. The SILAARP procedure may have disadvantages, such as a challenging work angle, the loss of triangulation with instruments, and the relatively small working space; however, these disadvantages do not affect the SILAARP procedure because the procedure is performed by experienced paediatricians. In our technique, the transabdominal suture retraction from the posterior bladder wall to the abdominal wall allowed a better laparoscopic view of the pelvic cavity and provided the appropriate tension for dissection along the plane between the posterior urethra wall and terminal rectum. Turning the 30°-angled laparoscopic camera upward and using the magnified view were also critical for visualizing the bulbar fistula in detail, aiding the identification of the pelvic portion of the longitudinal muscle tube and the complete excision of the rectal mucosa (Fig. 3).

The incidence of postoperative complications was not significantly different between both groups. However, rectal prolapse was more prevalent in SILAARP, while wound infection was more common in PSARP. The increased rate of rectal prolapse in the SILAARP group might be explained by the fact that the rectum is anatomized and dissected widely from the peritoneal reflection to the fistula to gain adequate pulling bowel length, whereas, the rectum is dissected just enough to reach the anus in PSARP. Moreover, the rate of rectal prolapse might be higher in patients with associated lumbosacral anomaly and poor pelvic musculature [21]. In the current study, patients with sacral and/or spinal malformations showed a high incidence of rectal prolapse compared to those without such malformations, however, the difference was not statistically significant, possibly due to the small sample size. Therefore, when performing laparoscopic surgery on patients, especially those with lumbosacral anomaly, we recommend the careful suturing of the seromuscular layer of the rectum to the presacral fascia and the avoidance of over-extensive dissection of the mesorectum and pelvic floor muscles.

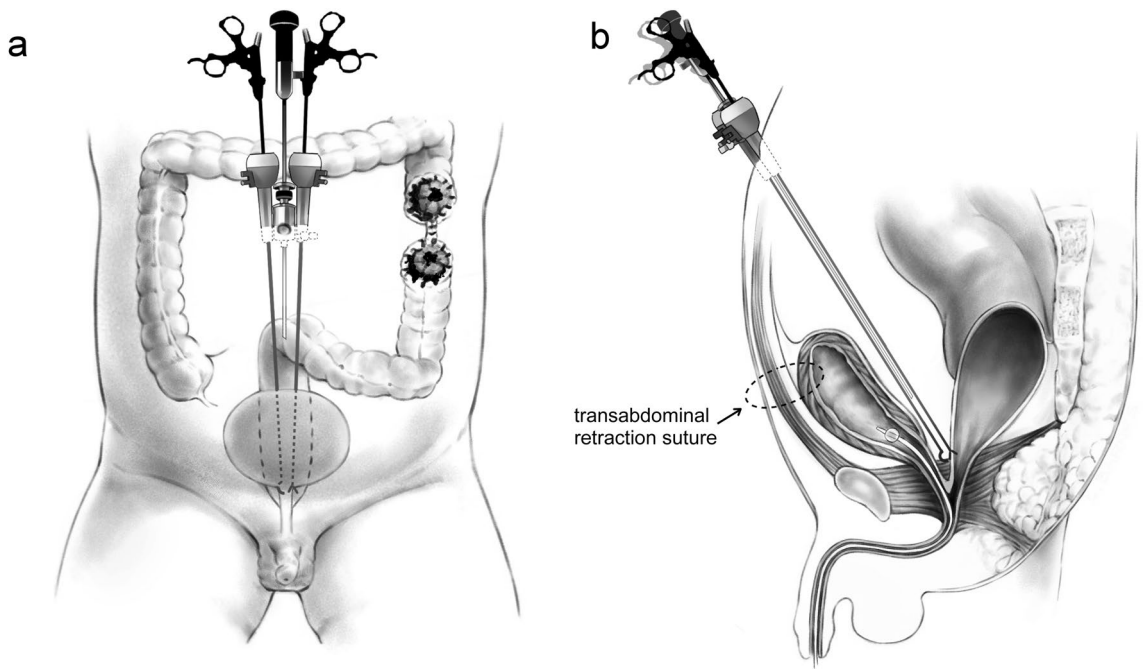

Fig.3 **a** Instrument arrangements of single-incision laparoscopicassisted anorectoplasty. **b** Illustration of single-incision laparoscopic-assisted anorectoplasty for children with intermediate-type anorectal

The incidence of urethral diverticulum after laparoscopy is also a concern for paediatricians. Previous studies indicate that posterior urethral diverticula might be associated with the use of laparoscopy [8, 9]. Dissecting of the deep fistula is a challenging part of laparoscopy. A histopathologic study of resected urethral diverticula showed colon-type mucosa [22]. When excess residual stump of the rectourethral fistula is unresected during the laparoscopic procedure, it may lead to urethral diverticulum. Alam et al. reviewed 29 patients with posterior urethral diverticulum after anorectoplasty, and two of these underwent LAARP [9]. In a study by Tainaka et al., 45 patients with high and intermediate-type ARMs underwent LAARP or PSARP; nine of 29 patients developed urethral diverticulum postoperatively after LAARP compared to 1 of 16 patients after PSARP [20]. Notably, in our series, there was no urethral diverticulum or urethral injury in either group. This might be attributed to the "internal closure" technique used in our centre. Because the urethra and distal rectum might share a common muscular wall, a dissection was made along the submucosa layer of the rectal pouch and the anterior mucosal layer of the terminal rectum was opened to identify the orifice of fistula from the inside. The rectal mucosa usually becomes columnar and radial at the point of the fistula, which is defined as the junction between the urethral squamous mucosa and rectal columnar mucosa. The mucosal layer of the rectum was transected at the transitional epithelium between the rectal mucosa and fistula mucosa. After the rectal mucosa was removed from the hypertrophic muscular cuff via a 3-mm hook cautery, the fistula mucosa retracted back into the fistula stump. The fistula was closed with 5-0 PDS horizontal running suture across the muscular cuff [14]. Hence, the colon-type mucosa of the fistula, which may be responsible for urethral diverticulum, was completely removed.

To achieve adequate anorectal function is crucial for surgical reconstruction of ARMs. For the treatment of high ARMs, it is generally agreed that laparoscopic surgery provides better functional performance than PSARP [5, 23, 24]. Similarly, for intermediate-type ARMs, our results showed that the rate of voluntary bowel movement was higher, while the rate of patients with some degree of soiling and constipation was lower in the SILAARP group, although these differences were not statistically significant, possibly because of the small sample size. Previous studies also showed that the results of anorectal manometry in high-type ARMs treated with LAARP were similar or better than those of patients treated with PSARP [25–27]. Not surprisingly, in the treatment of intermediate-type malformations, our results also showed that patients in the SILAARP group had higher ACRP than patients in the PSARP group, although the HPZL and RAIR findings were comparable between both groups. This may be due to the fact that apart from the traditional minimally invasive advantages of laparoscopy, SILAARP has an advantage over PSARP as the former procedure splits the anal sphincter musculature rather than incise it [11]. Although the rectal pouch is located at the level of levator ani in intermediate-type ARMs, single-port laparoscopy provides surgeons an excellent magnified view of the anatomical structures of the pelvis which helps them dissect the rectal fistula more meticulously. Therefore, SILAARP (circumferential dissection towards the fistula was made as close as possible to the longitudinal fibre of the rectal wall) minimized both scarring of the anal sphincter muscle and damage to the nerve fibres, which might result in maximal preservation of anal sphincter and further acquire better anorectal function [28]. Kudou et al. suggested that the anorectal function of patients who undergo laparoscopy may acquire degrees of improvement with age [27]. Additionally, the advantages of laparoscopy might be underestimated since it is commonly used to treat higher-type ARMs which are often associated with relatively severe pelvic floor dysplasia. When compared with higher-type ARMs, the degrees of dysplasia might be relatively mild in intermediate-type malformations, which in turn could result in better anal functions [21]. Hence, studies with longer follow-up are needed to further validate the benefits of SILAARP technique for the treatment of intermediate-type malformations with rectobulbar fistula.

The present study has several limitations. First, it was a single-centre retrospective study, with possible selection biases. Second, the relatively small number of patients who underwent PSARP may have prevented the identification of some differences between the groups. Despite these limitations, this study compared patients who underwent SILAARP and PSARP for intermediate-type ARMs with rectobulbar fistula in a way that provides evidence for the feasibility and safety of single-port laparoscopy for treating these selected patients.

Conclusion

Both SILAARP and PSARP seemed safe, feasible, and effective for children with intermediate-type ARMs and rectobulbar fistula. SILAARP appears to be associated with shorter postoperative hospital stays and higher ACRP when compared with PSARP. However, more high-quality, multicentre, large-sample randomized controlled trials are needed to compare SILAARP and PSARP for children with intermediate-type ARMs and rectobulbar fistula. Moreover, a longer follow-up time is required to confirm the true benefits of single-port laparoscopy.

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Compliance with ethical standards

Conflict of interest The authors have no conflict of interest to disclose.

Research involving human participants Ethical approval was obtained from the Ethics Committee of the Capital Institute of Pediatrics.

Informed consent Written informed consents were obtained from the parents before anorectoplasty.

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