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Retrospective comparative study

Lessons learned from lower urinary tract complications of anorectoplasty for imperforate anus with rectourethral/rectovesical fistula: Laparoscopy-assisted versus posterior sagittal approaches

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ABSTRACT

Purpose: To report the sequelae of and preventive strategies for selected lower urinary tract (LUT) complications, i.e., posterior urethral diverticulum (PUD), intraoperative LUT injuries, postoperative dysuria, and fistula recurrence in male imperforate anus (IA) with rectourethral/rectovesical (RU/RV) fistula after laparoscopy-assisted anorectoplasty (LAARP) or posterior sagittal anorectoplasty (PSARP).
Methods: 153 boys with IA and RU/RV fistula treated 1986–2019 by LAARP ($n = 56$) or PSARP ($n = 97$) at two unrelated institutes were studied retrospectively.
Results: After mean follow-up of 17.0 years (range: 36.5 days-32.0 years), the overall incidences of LUT complications were: LAARP (6/56; 10.7%); PSARP (7/97; 7.2%); $p = 0.55$, comprising PUD: LAARP ($n = 5$), PSARP ($n = 0$); $p = 0.006$; injuries: LAARP ($n = 0$), PSARP ($n = 5$); $p = 0.16$; dysuria: LAARP ($n = 1$), PSARP ($n = 1$); $p > 0.999$; and recurrence: LAARP ($n = 0$), PSARP ($n = 1$); $p > 0.999$. Mean onset of PUD was 5.1 years (range: 1.0–15.1 years). Treatment: PUD: surgery ($n = 2/5$), conservative ($n = 3/5$); injuries: intraoperative repair ($n = 5/5$); dysuria: conservative ($n = 2/2$), and recurrence: redo PSARP ($n = 1/1$).
Conclusions: Strategies devised to improve dissection accuracy resolved the specific technical issues causing LUT complications (remnant RU fistula dissection in LAARP and blind posterior access in PSARP). Currently, the incidence of new cases of PUD and LUT injuries is zero.
Level of Evidence: Level III

1. Introduction

Laparoscopy-assisted anorectoplasty (LAARP) [1] during which the levator ani muscle is preserved in order to achieve better postoperative bowel function, has been our procedure of choice for treating male imperforate anus (IA) with rectourethral/rectovesical (RU/RV) fistula since 2000. Whether LAARP or posterior sagittal anorectoplasty (PSARP) is chosen, RU fistula repair is extremely challenging and from experience, the types of postoperative lower urinary tract (LUT) complications that occur would appear to differ with respect to technique. For example, posterior urethral diverticulum (PUD) which is considered to be one of the serious LUT complications associated with anorectoplasty and RU fistula (especially bulbar) repair seems to be associated exclusively with LAARP [2], while other LUT complications, notably, LUT injuries, seem to occur more often during PSARP [3,4].

Because of these observations, we conducted a study on the incidence, outcome, and sequelae of LUT complications arising during/after anorectoplasty and RU/RV fistula repair, comparing LAARP with PSARP, focusing specifically on the incidence of typical LUT complications such as PUD, intraoperative LUT injuries (prostate, seminal vesicles, prostatic utricle, urethra), postoperative dysuria, and fistula recurrence and present technical strategies devised for their prevention.

2. Methods

The present study was an observational retrospective review of the case notes and medical records of 153 boys with IA and RU/RV fistula treated at Juntendo University Hospital in Japan ($n = 54$) and Children's Hospital, Helsinki University Hospital in Finland ($n = 99$), between 1986 and 2019. Two cases with primary neurogenic bladder due to myelomeningocele ($n = 1$) and diastematomyelia ($n = 1$) at Juntendo and 4 cases with primary

Abbreviations: PUD, Posterior urethral diverticulum; LUT, Lower urinary tract; RU, Rectourethral; RV, Rectovesical; IA, Imperforate anus; LAARP, Laparoscopy assisted anorectoplasty; PSARP, Posterior sagittal anorectoplasty.

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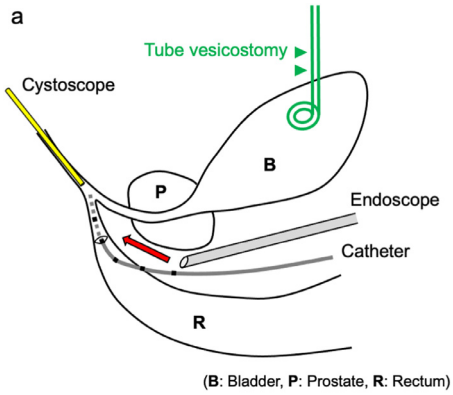

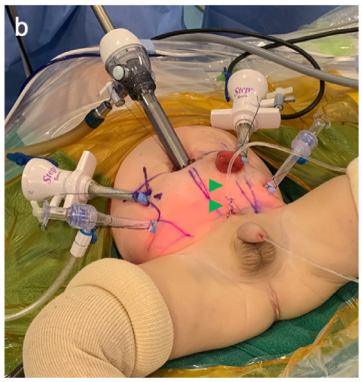

Fig. 1. a: Good visualization of the anterior wall of a fistula and surrounding structures during LAARP. A 30° or 45° endoscope is recommended. After opening the anterior wall of the rectum, a fine catheter is inserted into the fistula by the laparoscopic surgeon until it emerges in the urethra, and can be observed with a cystoscope. The exact length of the remaining fistula is measured and dissected further, remeasured, redissected until it is less than 5 mm long, and ligated. b: A patient prepared for LAARP with a tube vesicostomy. A suprapubic bladder catheter (arrowheads) allows continuous bladder decompression during cystoscopic examination to enable direct vision of the fistula during LAARP especially in patients with bulbar RU fistula.

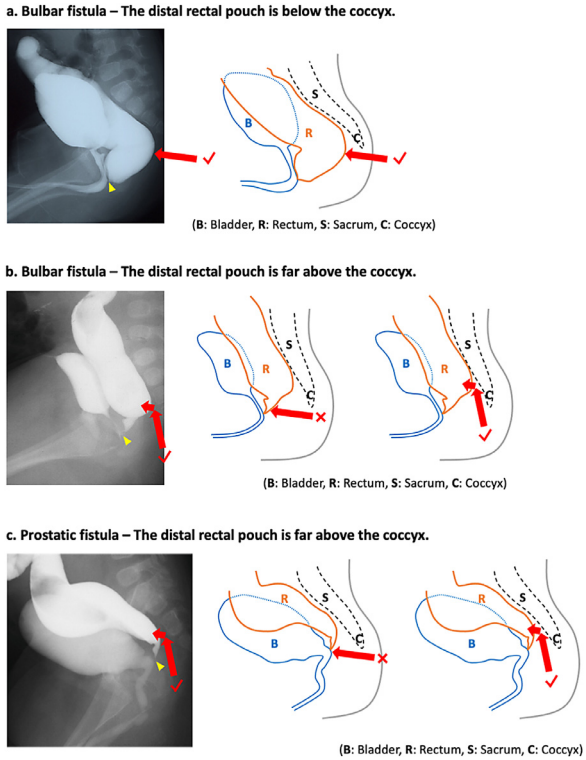

Fig. 2. a: Bulbar RU fistula. The distal rectal pouch is below the coccyx. Surgeons can identify the distal rectal pouch easily by starting to dissect directly downward. b: Bulbar RU fistula. The distal rectal pouch is far above the coccyx. c: Prostatic RU fistula. The distal rectal pouch is far above the coccyx. Surgeons should start dissecting diagonally upwards, aiming for the coccyx and following it inferiorly. The distal rectal pouch will be found lying almost parallel to the sacrum. If any other approach to dissection is used, surgeons are likely to get disorientated and injure the urethra or fistula before finding the distal rectal pouch. Arrowheads indicate a fistula in a distal colostogram. The big red arrows indicate the right and wrong directions for dissection.

neurogenic bladder due to severe sacral agenesis ($n = 3$) and presacral lipomeningocele ($n = 1$) at Helsinki University Hospital were excluded. One case with asymptomatic tethered cord syndrome at Juntendo was included in this study. The incidence, outcome, and sequelae of selected LUT complications (PUD, intraoperative LUT injuries, postoperative dysuria, and fistula recurrence) were compared between LAARP and PSARP.

LAARP has been indicated for RV and prostatic RU fistula patients at Helsinki University Hospital since 2005 and for all types of RU/RV fistula (including bulbar), at Juntendo since 2000. PSARP is indicated for technical reasons or because of the operating surgeon's preference. At Helsinki University Hospital, all bulbar RU fistula patients were treated by PSARP.

During the study period, technical strategies were devised to help prevent LUT complications. Incomplete excision of an RU fistula during LAARP was considered to be a serious problem so a novel technique for the intraoperative measurement of the remaining length of an RU fistula was devised at Juntendo [5,6,7]. Briefly, the fistula is dissected close to the urethra, opened, and a fine catheter with 10 mm calibrations is inserted until it is seen emerging under cystoscopic control. The distance from the point where dissection was ceased at the rectal end to the urethral orifice is measured (Fig. 1a). The fistula is further dissected free for exactly this length, remeasured, and if satisfactory, is tied and excised [6]. In addition, to improve visualization in bulbar RU fistula patients, continuous decompression of the bladder with a tube vesicostomy is mandatory to enable the distal end of the bulbar fistula to be viewed directly and facilitate measuring its length during cystoscopic examination, because there is no urethral catheter during cystoscopy (Fig. 1b). At Helsinki University Hospital, cystoscopy is not used for the identification of a fistula, and the rectovesical or rectoprostatic communication is transected flush to the posterior prostatic capsule or bladder neck without ligating the fistula.

For PSARP, a distal colostogram is mandatory for definitive preoperative surgical planning. The technical strategy devised to help prevent LUT complications during PSARP involves improving the accuracy of dissection. To dissect safely, surgeons must be familiar with the level of the distal rectal pouch in order to plan the best angle of approach for dissection. In patients with a bulbar RU fistula and a distal pouch below the coccyx (Fig. 2a), surgeons will be able to identify the distal pouch by dissecting directly downward and for patients with a bulbar RU fistula and a distal pouch at the level or above the coccyx (Fig. 2b), or a prostatic RU fistula (Fig. 2c), surgeons should dissect diagonally upwards to the coccyx, then follow the coccyx inferiorly to find the distal pouch lying almost parallel to the sacrum. If the approach to dissection is directly downwards when the distal rectal pouch is above the coccyx, the surgeon may become disorientated and injure the urethra, prostate, seminal vesicles, or fistula before identifying the distal pouch.

Statistical analyses were performed using GraphPad Prism software version 8.0 (GraphPad Software). The Student *t*-test or Fisher's exact test were used to assess significance between two

Table 1
Types of imperforate anus.

IA with RU/RV fistula	LAARP ($n = 56$)	PSARP ($n = 97$)
RV fistula ($n = 12$)	$n = 6$	$n = 6$
Prostatic RU fistula ($n = 77$)	$n = 26$	$n = 51$
Bulbar RU fistula ($n = 64$)	$n = 24$	$n = 40$

IA: Imperforate anus.
RU: Rectourethral.
RV: Rectovesical.
LAARP: Laparoscopy assisted anorectoplasty.
PSARP: Posterior sagittal anorectoplasty.

Table 2
Change in the incidence of LUT complications.

	LAARP ($n = 56$)	PSARP ($n = 97$)
Pre*	PUD ($n = 5$) Dysuria ($n = 1$)	LUT injuries ($n = 5$) Dysuria ($n = 1$) Fistula recurrence ($n = 1$)
Post*	0	0

LUT: Lower urinary tract.
LAARP: Laparoscopy assisted anorectoplasty.
PSARP: Posterior sagittal anorectoplasty.
PUD: Posterior urethral diverticulum.
*: Adoption of preventive technical strategies.

groups. Data were presented as mean±SD and the standard of statistical significance was defined as $p = 0.05$.

This study was approved by the Ethics Committee of Juntendo University School of Medicine and the University of Helsinki, and complies with the Helsinki Declaration of 1975 (revised 1983).

3. Results

Types of RU/RV fistula were: prostatic RU ($n = 77$), bulbar RU ($n = 64$), and RV ($n = 12$). Surgical repairs were either LAARP ($n = 56$) or PSARP ($n = 97$). Surgical repair performed with respect to type of fistula were: prostatic RU: (LAARP: 26/77, PSARP: 51/77), bulbar RU: (LAARP: 24/64, PSARP: 40/64), and RV: (LAARP: 6/12, PSARP: 6/12); (Table 1).

After mean follow-up of 17.0 years (range: 36.5 days-32.0 years), the overall incidence of LUT complications was 6/56 (10.7%) in LAARP cases, and 7/97 (7.2%) in PSARP cases ($p = 0.55$) (Table 2). Specifically, PUD: LAARP ($n = 5$) versus PSARP ($n = 0$); ($p = 0.006$); intraoperative LUT injuries: LAARP ($n = 0$) versus PSARP ($n = 5$; prostate=2, seminal vesicles=2, and prostatic utricle=1); ($p = 0.16$); postoperative dysuria: LAARP ($n = 1$) versus PSARP ($n = 1$); ($p>0.999$), and fistula recurrence: LAARP ($n = 0$) versus PSARP ($n = 1$); ($p>0.999$). At Juntendo, no subject developed neurogenic bladder secondary to surgery, however, at Helsinki University Hospital, 3 cases developed chronic postoperative neurovesical dysfunction. There were no perioperative urinary tract infections secondary to vesicoureteral reflux recorded at both centers because of prophylactic antibiotic administration.

The incidence of PUD after LAARP was (5/56; 8.9%) and the incidence of LUT injuries in PSARP was (5/97; 5.2%). However, the incidence of PUD after LAARP and LUT injuries during PSARP has been zero, respectively, since adopting the preventive technical strategies mentioned earlier (Table 2).

All PUD ($n = 5$) presented with difficult urination, sensation of incomplete voiding, urinary incontinence, or dribbling of urine retained in the PUD, after a mean follow-up of 5.1 years (range: 1.0–15.1 years). Treatment was surgical repair ($n = 2$) or observation in PUD patients who became asymptomatic ($n = 3$). All intraoperative LUT injuries ($n = 5$) were repaired immediately, intraoperatively. Fistula recurrence ($n = 1$) was repaired by redo PSARP. Details of LUT complications and their sequelae are summarized in Table 3a for LAARP and Table 3b for PSARP.

Brief descriptions of representative cases of LUT complications are presented. A case of PUD (Case 3 in Table 3a) after LAARP for IA with bulbar RU fistula, reported previously elsewhere [2], an early case in our series, was treated before we started measuring the residual fistula with a catheter under cystoscopic control. PUD was caused by incomplete fistula excision and diagnosed when 1.9 years old. Excision of PUD was performed and the patient is now 19 years old with no urinary symptoms, although he had stress incontinence until he was 15 years old that resolved spontaneously. A case of prostate injury (Case 1 in Table 3b) after PSARP for IA with prostatic RU fistula was most likely caused by the operating surgeon getting disorientated (Fig. 2c), leading to injury of the prostate during dissection of the distal end of the rectal pouch.

4. Discussion

Male IA with RU/RV fistula still presents a surgical challenge even with most up to date equipment and surgical experience. In fact, the sequelae of anorectoplasty related LUT complications can totally erode the quality of life a child may achieve following appropriate anorectal reconstruction.

Various LUT complications have been reported after LAARP as well as PSARP [3,4,8,9,10]. Hong et al. [3] reported about urologic injuries associated with PSARP in a large series. At their institute, they experienced 19/572 (3.3%) urologic injuries including 7 urethral injuries during PSARP [3], a figure that is not as high as reported by other centers [8,11,12]. Hong et al. [3] also recommend performing a distal colostogram preoperatively to show the level of a fistula to help prevent urologic injuries. A Japanese multicenter study group on male IA with prostatic RU fistula where 81 patients were treated by PSARP ($n = 21$), abdominoperineal pull-through ($n = 15$), or LAARP ($n = 45$), reported no intraoperative LUT injuries [13]. In this series, PUD was detected later by cystourethrography and/or magnetic resonance imaging (MRI) in 15 LAARP patients and 2 PSARP or abdominoperineal pull-through patients, but only one of these 17 patients was actually symptomatic [13]. Uchida et al. [10] also investigated the complications of LAARP for IA with prostatic RU fistula ($n = 15$) and RV fistula ($n = 2$) and reported no urethral injuries but 9 patients had PUD on routine follow-up MRI. In our series, all patients had MRI performed after anorectoplasty but all our PUD patients were symptomatic. Of the 5, 2 had surgical excision and 3 are under observation after becoming asymptomatic.

This series identified that certain LUT complications are procedure-related; in other words, there is a risk for PUD if a fistula is inadequately excised during LAARP and a risk for LUT injuries during PSARP. With LAARP, visualization of an RU fistula, surrounding structures, and the posterior wall of the urethra is excellent and probably the most obvious feature that prevents injury to the urethra (Fig. 1a). Our 5 PUD cases comprised 4 in 36 LAARP cases at Juntendo, and 1 in 20 LAARP cases at Helsinki University Hospital. All 4 cases at Juntendo were early cases, treated before adopting the preventive technical strategy of measuring the length of the remaining fistula intraoperatively. Since then, the incidence of PUD has been zero, as mentioned earlier (Table 2). Thus, we strongly recommend the introduction of an intraoperative measuring technique to ensure complete excision of a bulbar RU fistula, in particular.

In contrast, all 5 LUT injuries experienced in the 97 PSARP cases in this series (3/18 cases at Juntendo and 2/79 cases at Helsinki University Hospital) were early cases treated before the preventive strategy mentioned earlier was adopted. Case 4 in Table 3b, a prostatic utricle injury during PSARP, is a good example of a lesson learnt from this study. Despite exhaustive preoperative investiga-

Table 3a
LAARP LUT complications.

Case	Fistula	Diagnosis	Age at onset	Current age	Symptoms	Treatment→Outcome	Remarks
1	Bulbar	PUD (Size: 37 × 35 × 32 mm)	15.2 years	18.7 years	Sensation of incomplete voiding	Conservative management →Spontaneous resolution	Nil
2	Prostatic	PUD (Size: 40 mm)	3.8 years	19.0 years	Sensation of incomplete voiding	Conservative management →Spontaneous resolution	Nil
3	Bulbar	PUD (Size: 46 mm)	1.9 years	19.0 years	Difficult urination, dysuria	Excision of PUD →Stress incontinence until 15 years old that resolved spontaneously	Trisomy 21
4	Bulbar	PUD (Size: 30 × 50 × 40 mm)	2.7 years	17.8 years	Sensation of incomplete voiding	Conservative management →Spontaneous resolution	Nil
5	Vesical	Dysuria (transient)	10 months (immediately postoperative)	16.6 years	Dysuria	Conservative management →Spontaneous resolution	Nil
6	Prostatic	PUD (Size: Not available)	5.0 years	8.4 years	Urinary incontinence	Excision of PUD →Mild dribbling of urine	Nil

LAARP: Laparoscopy assisted anorectoplasty, LUT: Lower urinary tract, PUD: Posterior urethral diverticulum.

Table 3b
PSARP LUT complications.

Case	Fistula	Diagnosis	Age at onset	Current age	Symptoms	Treatment→Outcome	Remarks
1	Prostatic	Injury (prostate)	2 months (Intraoperative)	31.1 years	Nil	Immediate intraoperative repair →Epididymitis	Chronic epididymitis
2	Prostatic	Injury (seminal vesicle)	2 months Intraoperative)	28.5 years	Nil	Immediate intraoperative repair →Unremarkable recovery	Nil
3	Bulbar	Dysuria (transient)	4 months (Immediately Postoperative)	27.1 years	Dysuria	Conservative management →Spontaneous voiding	CIC until 8 years old. Must sit to void.
4	Bulbar	Injury (prostatic utricle)	9 months (Intraoperative)	23.6 years	Nil	Immediate intraoperative repair →Unremarkable recovery	Intellectual disability
5	Prostatic	Injury (prostate)	3 months (Intraoperative)	14.0 years	Nil	Immediate intraoperative repair →Unremarkable recovery	Nil
6	Prostatic	Injury (seminal vesicle)	4 months (Intraoperative)	13.9 years	Nil	Immediate intraoperative repair →Unremarkable recovery	Nil
7	Bulbar	Recurrence (fistula)	6 months (2 months postoperatively)	10.6 years	Dribbling of urine from the anus	Redo PSARP →Unremarkable recovery	Nil

PSARP: Posterior sagittal anorectoplasty, LUT: Lower urinary tract, CIC: Clean intermittent catheterization.

tions such as colostogram, voiding cystourethrography, ultrasonography, and cystoscopy, the operating surgeon had no inkling of the presence of a prostatic utricle. Surgeons must be constantly on the alert for the unexpected and be wary of anomalies of the posterior urethra and vas deferens as reported by Mickelson et al. [11] to prevent inadvertent injuries.

This study has some limitations because of its design. Being retrospective is one limitation as well as it being a joint study between two institutes with different routines in terms of surgical technique and postoperative follow-up. Furthermore, there might be potential bias in reporting postoperative LUT symptoms in medical records, especially with respect to subjective symptoms such as dysuria, urinary incontinence and problems in bladder emptying which appeared to be rare in this study but in reality are quite frequent, although temporary in most cases.

5. Conclusions

This study reported that LUT complications during/after both LAARP and PSARP are technique related, and presented novel technical strategies to prevent them. If appropriate surgical techniques are used, LUT complications are uncommon, readily treatable, and unlikely to be associated with long-term morbidity.

Disclosures

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Declaration of Competing Interest

The authors have no financial conflicts of interest.

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