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ORIGINAL ARTICLE

Fecal and urinary continence after scope-assisted anorectovaginoplasty for female anorectal malformation

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Abstract
Aim We assessed continence after scope-assisted anorectovaginoplasty (SARVP) for female anorectal malformation (FARM).
Methods Five FARM cases were assessed; cases 1 and 2: cloacal malformation; case 3: urogenital sinus, and rectovestibular fistula (RF); case 4: RF, absent vagina, and sacral anomaly; case 5: covered cloacal exstrophy. Treatment was SARVP in all cases, with perineal vaginoplasty (case 1), vagina pull-through (PT) similar to Georgeson's colon PT (case 2), and the use of the native RF/cloaca channel as a vagina (cases 3–5). Continence was assessed pre and postoperatively.
Results SARVP was performed in the lithotomy position without repositioning. Mean age at surgery was 3.2 (1.7–5.5) years. Current mean age: 8.8 years (range 7.5–12.2). Mean follow-up: 5.7 years. Preoperative continence: fecal: all had stomas; urinary: cases 1 and 2: continent; cases 3–5: incontinent. Postoperative continence: fecal: cases 1–3: continent; case 4: incontinent; case 5: awaiting stoma closure; urinary: cases 1 and 2: continent; cases 3 and 4: incontinent; case 5: continent (intermittent catheterization). Fetal continence evaluation questionnaire (CEQ) scores for cases 1–4 were 7.5, 9, 10, and 2 (maximum score 10, mean 7.1).
Conclusion Scope assistance improves visualization, thus pelvic sphincter dissection/division is minimized with less detrimental impact on postoperative continence.

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Keywords Anorectal malformation · Cloaca · Imperforate anus · Laparoscopically assisted anorectovaginoplasty · Laparoscopy

Introduction

Georgeson et al. [1] reported a new technique for repairing anorectal malformation (ARM) using laparoscopy in 2000. This procedure provides excellent visualization of the pelvic floor sphincter muscles, and allows accurate placement of pulled-through intestine in the center of the levator sling without dividing the sphincter muscles. While there are more reports of laparoscopy-assisted Georgeson's technique being performed under pneumoperitoneum in female ARM (FARM) [1–8] in the literature, to the best of our knowledge, there are no reports of anorectovaginoplasty (ARVP) using Georgeson's pull-through (PT) technique being performed through a Pfannenstiel incision with scope assistance without insufflation, i.e., a laparoscope is inserted directly into the pelvic cavity.

Here we report a preliminary evaluation of scope-assisted anorectovaginoplasty (SARVP) using Georgeson's technique for the repair of FARM including cloacal malformation.

Materials and methods

Five cases of FARM, who underwent SARVP between June 2005 and April 2007 were reviewed. Follow-up data were obtained by reviewing the outpatient medical records. Cases 1 and 2 had cloacal malformation, case 3 had urogenital sinus and rectovestibular fistula (RF), case 4 had RF

with absent vagina and sacral anomaly, and case 5 had covered cloacal exstrophy. Anorectoplasty was performed using Georgeson's technique through a Pfannenstiel incision with scope assistance, i.e., insertion of a laparoscope into the pelvic cavity without insufflation (Fig. 1, left). When we perform Georgeson's procedure, we use a pair of mosquito forceps and blunt dissection to create the PT route from the perineum to the pelvic floor [2–5]. The mosquito forceps inserted into the perineum can be observed emerging in the center of the pelvic floor muscles through the scope (Fig. 1, right). Vaginoplasty in case 1 was perineal, case 2 had vaginal PT with scope assistance performed in a similar way to Georgeson's colon PT, and cases 3–5 had a vagina created from the native RF/cloacal channel (Fig. 2). In case 2, the PT route for the vagina was created from the perineum in the same as Georgeson's colonic PT for anorectoplasty.

Postoperative fecal continence was assessed over time using a continence evaluation questionnaire (CEQ). Five parameters were scored on a scale of 0–2, giving a maximum score of 10 (Table 1). In this study, we used "staining" to refer to fecal markings on underclothing, and "soiling" to mean actual feces on underclothing. CEQ scores were determined, and urinary continence were also compared pre and postoperatively.

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

Results

SARVP was performed entirely in the lithotomy position without repositioning. Mean age at SARVP was 3.2 years (range 1.7–5.5). Mean operative weight at SARVP was 12.4 kg (range 9.7–16.3). Mean current age: 8.8 years (range 7.5–12.2); mean follow-up: 5.7 years (range 4.9–6.1). Postoperative fecal and urinary continence are summarized in Table 2. At last follow-up, post-SARVP, cases 1–3 were continent of feces; case 4 was incontinent with spina bifida, and case 5 was awaiting stoma closure. CEQ scores for cases 1–4 were 7.5, 9, 10, and 2, respectively (mean score 7.1). Cases 1 and 2 were continent of urine preoperatively and post-SARVP. Cases 3 and 4 had persistence of preoperative urinary incontinence postoperatively due to the absence of urethral sphincters. Case 5 was continent of urine with intermittent catheterization.

Case 1

Cloaca was diagnosed at birth, and sigmoid colostomy was created the next day elsewhere. Six days later, she was referred to our hospital for surgical treatment of cloaca. At the age of 20 months, she underwent SARVP and closure of sigmoid colostomy. After induction of general anesthesia, a 5 mm trocar was inserted through the superior margin of the umbilicus, and carbon dioxide was insufflated to 8 mmHg. However, laparoscopy was complicated by small intestine containing swallowed air, requiring the conversion to open surgery through a Pfannenstiel incision. The rectum was dissected from the vagina with a bicornuate uterus. Cystoscopy showed that the vagina was divided by a septum with a rectovaginal fistula opening on the septum. The distal stump was ligated using endoloops just at the common wall of the rectum and vagina. The sigmoid colostomy was then taken down, and colocolostomy was created. The pelvic floor was dissected until the pelvic floor muscles were identified. Colon was pulled-through using Georgeson's technique with scope assistance. The common channel was 1.5 cm and perineal Y-V plasty was performed for vaginoplasty. Eleven months after SARVP, she underwent a Ganz-Miwa operation for anal mucosal prolapse. She is currently 7.6 years old, and has fecal and urinary continence without glycerin enemas.

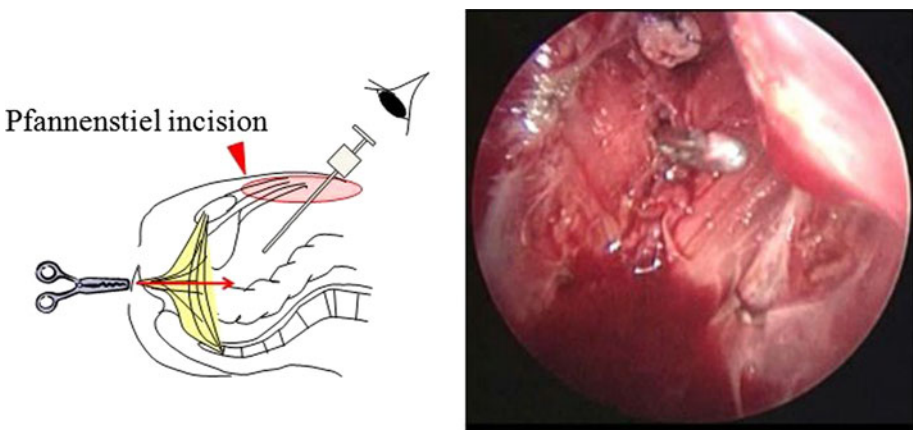

Fig. 1 Scope-assisted anorectovaginoplasty. *Left* A scope has been inserted through the Pfannenstiel incision. *Right* An intraoperative photo showing a pair of mosquito forces emerging in the center of the pelvic floor muscle

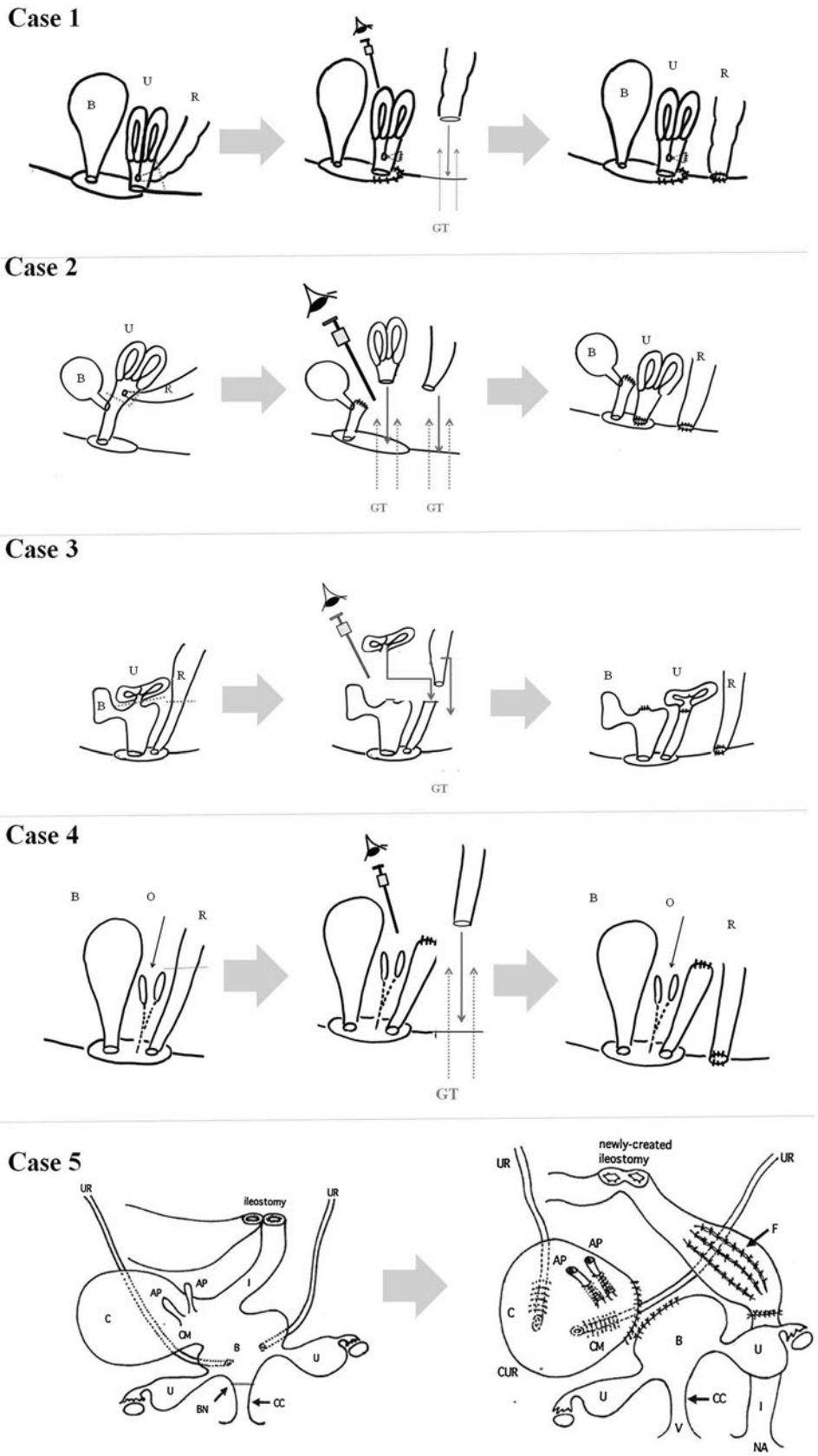

Fig. 2 Diagrams of cases 1–5. *B* bladder, *U* uterus, *R* rectum, *GT* Georgeson's technique, *CC* cloacal channel, *BN* bladder neck, *AP* appendix, *C* colon, *V* vagina, *CM* cecum, *I* ileum, *UR* ureter, *CUR* continent urinary reservoir, *NA* neoanus, *F* folded

Table 1 Continence evaluation questionnaire

	Score
Frequency of defecation/urination	
1–2/day	2
3–5/day	1
≥6/day	0
Staining/soiling	
None	2
Staining occasionally	1.5
Staining often	1
Staining every motion	0.5
Soiling	0
Perianal erosion	
Nil	2
Occasionally	1
Often	0
Anal shape	
Normal appearance	2
Scar or skin-tag visible	1.5
Mucosa visible	1
Prolapsed requiring intervention	0
Medication	
Nil	2
Laxative/enema/suppository	1
Antidiarrheals needed	0
Maximum score = 10	

Case 2

Prenatal diagnosis of hydrocolpos and bilateral hydronephrosis was made, and cloaca was diagnosed at birth. Left transverse colostomy and cystostomy were created soon after birth. When 16 months old, she had radical surgery for a ventroseptal defect performed by cardiothoracic surgeons, then antireflux surgery (Cohen procedure) for bilateral vesicoureteral reflux when she was 32 months old. At the age of 42 months, SARVP was performed. Cystoscopy showed that the common channel was 3.5 cm in length, and a fistula between the rectum and vagina opened 4.7 cm from the cloacal orifice. A Pfannenstiel incision was made, and the rectum was dissected from a dilated vagina with a bicornuate uterus. The rectum was dissected as it tapered distally, and the fistula ligated. The vagina was then dissected from the bladder, and the distal stump ligated. The cloacal channel was used as a neourethra. The PT route for the anorectum was created using scope-assisted Georgeson's technique, and the PT route for the vagina was created from the perineum using scope assistance in a similar way to Georgeson's technique, which we report here as a new technique. Thus, both colon and vagina PT were performed without cutting the pelvic floor muscles, in contrast to posterior sagittal anorectovaginoplasty. Anoplasty and vaginoplasty were performed in appropriate positions in the perineum. Some 12 months after SARVP, redo anoplasty and vaginoplasty were required from a perineal approach because of stenosed openings. Colostomy closure was performed 32 months after SARVP. She is currently 8.5 years old, and is continent of feces and urine.

Case 3

This case was diagnosed prenatally with duodenal atresia, and a diamond-shaped anastomosis was performed on day 7 of life. After birth, urogenital sinus with RF and bilateral vesicoureteral reflux with a fused kidney were also

Table 2 Continence status pre/post scope-assisted anorectovaginoplasty

Patient no.	FARM type	Vaginoplasty	Fecal continence		Urinary continence
			Pre/post	CEQ score	Pre/post
1	Cloaca (1.5 cm)[a]	Perineal Y-V plasty	Stoma/continent	7.5	Continent/continent
2	Cloaca (3.5 cm)[a]	Vaginal pull-through	Stoma/continent	9	Continent/continent
3	Urogenital sinus with rectovestibular fistula	Distal colon used	Stoma/continent	10	Incontinent[c]/ incontinent[c]
4	Rectovestibular fistula with absent vagina	Distal colon used	Stoma/ incontinent[b]	2	Incontinent[c]/ incontinent[c]
5	Covered cloacal exstrophy	Cloacal channel used	Stoma/continent	–[b]	Incontinent/continent[d]

All cases: Pfannenstiel incision with scope-assisted Georgeson's technique

[a] Length of common channel

[b] Sacral anomaly

[c] Absence of sphincter muscles

[d] Colon reservoir with appendicostomy

diagnosed. An ileostomy was created in the right lower quadrant when 17 months old, and SARVP was performed when 33 months old. The pelvis was examined through a Pfannenstiel incision, revealing a very small bladder and a bicornuate uterus fused to a dilated urogenital sinus. This dilated urogenital sinus was used as a neobladder. After both ureters were divided from the bladder, the anterior wall of the urogenital sinus was opened. Both ureters were reimplanted through 20 mm of subepithelial tunnels using Cohen's technique. The distal rectum was divided, and used to create a neovagina. The uterus was anastomosed to the distal rectum. The proximal rectum was then mobilized and brought through the pelvic floor sphincter muscles using Georgeson's technique with scope assistance. Some 10 months after SARVP, bladder augmentation of the neobladder was performed using a segment of ileocecum with the ileocecal valve to prevent reflux. Both ureters were reimplanted in the ileum, and appendicostomy was created for intermittent catheterization. Closure of ileostomy was performed 25 months after SARVP. She is currently 8.0 years old, and continent of feces with a regular bowel habit. However, she has persistence of preoperative urinary incontinence postoperatively due to the absence of urethral sphincters.

Case 4

Rectovestibular fistula was suspected in a 1-day-old child who was transferred to our hospital for assessment. On day 4 of life, a right transverse colostomy was created and at surgery it was noted she had bilateral ovaries, but no uterus. Further investigations proved that she had uterovaginal agenesis (Mayer-Rokitansky-Kuster-Hauser syndrome [4]) associated with RF. SARVP was performed through a Pfannenstiel incision when she was 20 months old. Because both ureters opened at the bladder neck, there was bilateral vesicoureteral reflux, and the ureters were divided and reimplanted in the bladder. The distal rectum was divided and closed at its stump, and used to create a neovagina. The proximal rectum was then mobilized and brought through the pelvic floor sphincter muscles using Georgeson's technique with scope assistance. She is currently 7.5 years old and incontinent of feces and urine due to the absence of urethral sphincters and sacral anomaly.

Case 5

This case has been reported previously [9]. Imperforate anus with cloacal fistula was diagnosed, and an ileostomy was performed elsewhere. She was referred to us for further management of covered cloacal exstrophy when she was 5 years old. Cystoscopy showed a long, wide cloacal channel connected to an extremely broad bladder neck with no urinary sphincter, rendering her totally incontinent. The cecum, 2 uteri, and 2 ureters all opened into the bladder. The cecum had 2 appendices, and the colon was short, blind-ended and hugely dilated. Function of both kidneys was normal, and both ovaries were detected on magnetic resonance imaging. SARVP was performed when she was 66 months old. The cloacal channel and bladder were used as a vagina, and the size of the orifice of the cloacal channel appeared to be adequate enough for future sexual intercourse. Both ureters were ligated and divided at their distal ends, and the cecum was excised from the bladder to create a continent urinary reservoir (CUR) with the blind-ended colon. The left ureter was reimplanted into the CUR. The right ureter was exteriorized as an ureterostomy in the first instance to minimize any risk for decrease in renal function due to obstruction that might occur if both ureters were reimplanted into the CUR at the same time. The proximal one-third of each appendix was rotated and embedded in the seromuscular layer of the cecum to create an antireflux mechanism to prevent leakage of urine through the appendix. The distal end of each appendix was then exteriorized to the abdominal wall for catheterization to control urine excretion. The ileum transected from the cecum was pulled-through using SARVP to create a neoanus. The postoperative course was uneventful and both appendices were used successfully for intermittent catheterization to control urine excretion. Eleven months later, the right ureter was reimplanted into the CUR. She is currently 12.2 years old and is continent of urine by catheterizing the appendices.

Discussion

Posterior sagittal anorectoplasty (PSARP) is the standard procedure for surgical repair of ARM, affording excellent exposure and allowing exact placement of the rectum within the pelvic muscle complex in the majority of centers [10, 11]. Pena et al. [12] reviewed 339 patients with cloacal malformations treated by PSARP, and reported that 94 of 156 patients (60 %) had voluntary bowel movements, but that the remaining 62 patients (40 %) had fecal incontinence, while 104 of 193 patients (54 %) had urinary continence. In 19 patients, with a short common channel (1 cm), 77 % of patients had voluntary bowel movements, and all had urinary continence. However, PSARP requires extended dissection of the perineal region, including division of the levator ani muscle sling. We believe that if the pelvic floor muscles and anal sphincter muscles are cut, the injury will compromise urethral and anal sphincter control (urethra and anal and should be avoided at any cost). Thus, we recommend Georgeson's technique to prevent injury to the nerves and muscles of the anal sphincter, to preserve sphincter control.

Georgeson's technique involves minimal perineal dissection without division of pelvic muscles with accurate placement of the rectum within the levator ani and pelvic muscle complex [1], and according to our experience, simple blunt dissection of the pelvic floor is more than sufficient to create a pull-through canal without requiring anal endosonography for adequate positioning, because there appears to be a potential canal present naturally that is filled with loose connective tissue [5].

In our series, cases 3–5 had concurrent anomalies such as absence of sphincter muscles, and sacral anomaly that we knew would complicate postoperative fecal and urinary continence, however, we found SARVP was very useful because there was no need to reposition the patient during surgery; thus reducing the risk for problems with intubation, shortening operating time, minimizing the potential for wound infections, as well as eliminating the need to approximate pelvic floor/sphincter muscles divided by posterior sagittal incision, which is necessary with PSARP.

Despite common channel lengths of 1.5 and 3.5 cm in cases 1 and 2, respectively, both have voluntary bowel movements with high CEQ scores, and are continent of urine without intermittent catheterization. We attribute this to scope assistance enhancing the view of the pelvic floor, allowing blunt dissection of the pelvic floor with a pair of mosquito forceps to be observed, thus facilitating accurate placement of the anorectal/vaginal PT without dividing the pelvic floor sphincter muscles. Scope backlighting was also useful as a guide to indicate the direction in which to progress with blunt dissection in all cases.

This is the first report of our new procedure, scope-assisted anorectovaginoplasty (SARVP) using Georgeson's technique being performed for the repair of FARM including cloacal malformation. It has the distinguishing features of not incising the pelvic floor muscles for creating the PT route for the vagina according to Georgeson's technique, with access to the abdomen through a Pfannenstiel incision. With PSARP, the pelvic floor muscles need to be incised, and with laparoscopy assisted Georgeson's PT, the whole procedure is performed laparoscopically.

Although patient numbers in this study are very small to generalize, and the types of anorectal malformation treated are different, it would appear that our approach of using a laparoscope inserted through a Pfannenstiel incision allowing us to perform colon PT using Georgeson's technique may appear to be associated with less postoperative incontinence in certain subtypes of FARM, since the pelvic floor muscles are not incised. Further investigation is necessary to compare the outcome of PSARP and SARVP using Georgeson's technique.

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