

## Original Article

# A follow-up study on postoperative function after a modified semi-posterior sagittal one-stage anorectoplasty for female imperforate anus with a rectovesibula fistula

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Received September 4, 2016; Accepted November 2, 2016; Epub January 15, 2017; Published January 30, 2017

**Abstract:** Objective: We evaluated the clinical efficacy of a one-stage modified semi-posterior sagittal anorectoplasty (modified Peña procedure) for radical treatment of female imperforate anus with a rectovesibula fistula. Methods: A total of 89 female neonates who had imperforate anus with a rectovesibula fistula and underwent treatment in our hospital between January 2003 and December 2013 were enrolled in this study, including 39 who underwent the modified Peña procedure (modified Peña procedure group), namely keeping the levator ani muscle and ischiorectal muscle complex intact, and 50 who underwent transperineal anal transposition (transperineal anal transposition group). Postoperative bowel movements were assessed and compared using the Krickbeck classification and anorectal manometry at follow-up. Results: A significantly lower age at operation and incidence of preoperative rectal dilation was observed in the modified Peña procedure group than in the transperineal anal transposition group ( $P < 0.001$ ). A higher incidence of postoperative soiling was observed in the transperineal anal transposition group compared with the modified Peña procedure group ( $P = 0.049$ ). There was no significant difference in the incidence of grade I postoperative constipation between groups ( $P > 0.05$ ), while a significantly higher incidence of grade II and III postoperative constipation was found in the transperineal anal transposition group ( $P = 0.049$  and  $0.026$ , respectively). Anorectal manometry at the 12 month postoperative follow up visit revealed no induction of the rectoanal inhibitory reflex in either group; there were also no significant differences in the resting rectal pressure or active systolic blood pressure between groups. A lower rectal compliance was observed in the transperineal anal transposition group compared with the modified Peña procedure group, although this finding was not statistically significant ( $P > 0.05$ ). Conclusions: The one-stage modified Peña procedure for congenital imperforate anus with a rectovesibula fistula may be associated with a good long-term ability to control postoperative bowel movements.

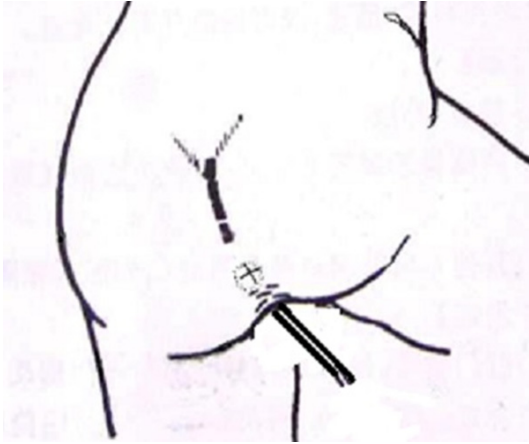
**Keywords:** Imperforate anus, rectovestibular fistula, posterior sagittal anorectoplasty, surgery, follow-up study

## Introduction

As the most common type of congenital anorectal malformation (ARM) seen in imperforate anus among female infants, rectovestibular fistula (RVF) is present in 10% to 30% of all forms of ARM [1]. Until now, there has been no consensus on the approach of anorectoplasty for RVF [2-4]. Transperineal anal transposition is a widely used procedure for the patients aged 6 to 8 months in most of the hospitals. However, this surgery results in a high recurrence of postoperative rectovaginal fistula and unsatisfactory postoperative bowel movements [4]. Since posterior sagittal anorectoplasty (PSARP) was introduced in 1982 [5], the precise anatomy

and reconstruction of the anorectum and sphincter greatly improves postoperative quality of life in neonates with congenital ARM. Since the efficacy of one-stage anorectoplasty without colostomy for treatment of neonatal RVF has been widely recognized worldwide [6-11], so modified PSARP [6], a vertical incision was made to explore the skin, sacrorectal muscle, and levator ani muscle, with preservation of the intact levator ani muscle and ischiorectal muscle complex, unlike complete incision of the levator ani muscle and ischiorectal muscle complex of PSARP, was proposed for treatment of RVF. We aimed to assess the clinical efficacy of one-stage modified semi (namely keeping the levator ani muscle and ischiorectal muscle

## One-stage postoperative function of RVF



**Figure 1.** A sagittal incision from 1 cm superior to the tip of the coccyx to 1 cm anterior to the anal crypts at the middle sacral region.



**Figure 2.** Intraoperative vertical incision of skin, coccygeal rectus muscle, and levator ani muscle, and preservation of the intact levator ani muscle and ischioanal muscle complex.

complex intact)-PSARP (the modified Peña procedure) for radical treatment (namely without colostomy) of neonatal imperforate anus with RVF in our hospital, between January 2007 and December 2013, and compared the effectiveness of this surgery with one-stage transperineal anal transposition performed in our hospital between January 2001 and December 2006, so as to summarize the experiences of the modified Peña procedure for radical therapy of congenital RVF.

### Mertirals and methods

#### Subjects

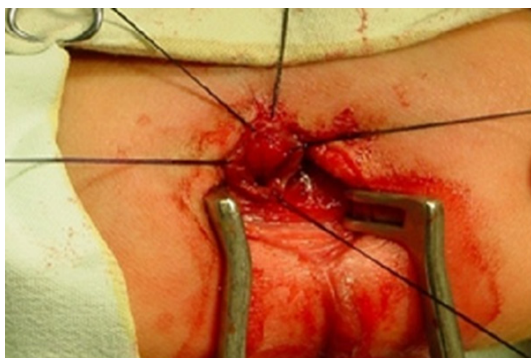
A total of 91 neonates who had congenital imperforate anus with a RVF underwent surgi-

cal treatment in our hospital between January 2001 and December 2013. All diagnoses were made by clinical examination in the neonatal period. Preoperative abdominal B-mode ultrasonography was performed to detect renal malformations and presacral mass, cardiac ultrasound to detect cardiac malformations, MRI of the lumbosacral vertebrae to detect spinal deformity and tethered spinal cord, and trans-fistular radiography to determine the degree of the rectal dilation. All patients were full-term infants. One patient had bilateral renal duplication, hemivertebra deformity of the sacrococcygeal vertebrae, and vaginal septum deformity; one had vaginal atresia and double uterus malformation. These two cases were not included in this study. None of the other cases had severe cardiac/renal malformations, presacral mass, apparent sacral dysplasia, or tethered spinal cord.

Prior to 2007, early-stage dilation of fistula followed by transperineal anal transposition 6 to 8 months after birth was employed for treatment of RVF in our hospital; after 2007, the one-stage modified Peña procedure was used for radical therapy of female imperforate anus with RVF. The 89 neonates with RVF were classified into two groups according to surgical approach. Patients in the modified Peña procedure group (n=39) underwent the modified Peña procedure; neonates in the transperineal anal transposition group (n=50) underwent transperineal anal transposition. All cases had total correction without colostomy.

#### Surgical procedures

The modified Peña procedure was performed as follows. Preoperative enema was done with warm physiologic saline via the fistula until the colon was clean. Broad-spectrum antibiotics (ceftriaxone and metronidazole) were administered for preoperative preparation. General endotracheal anesthesia was induced and gastric tubes and urinary catheters were implanted. Neonates were positioned prone with buttocks elevated. The perineal region was stimulated using a muscular stimulator to identify the position of anus. A sagittal incision was made 1 cm superior to the tip of the coccyx, to 1 cm anterior to the anal crypts at the middle sacral region (**Figure 1**). Intraoperatively, the position of muscular groups was identified using muscle stimulators and a vertical incision



**Figure 3.** Anatomic separation of the blind end of the rectum and preservation of the intact levator ani muscle and ischiorectal muscle complex.

was made to explore the skin, coccygeal rectus muscle, and levator ani muscle, preserving the intact levator ani muscle and ischiorectal muscle complex (**Figures 2 and 3**). A precise anatomic dissection of the blind end of the rectum and management of the fistula was performed under the microscope. Then, the blind end of the rectum was fully dissociated, and the rectum was pulled through the middle of the ischiorectal muscle complex and external anal sphincter. If the rectum was dilated, the dilated intestinal tube was removed and the terminus of the intestinal tube was sutured with the anal skin.

Preoperative preparation and induction of anesthesia of the transperineal anal transposition was the same with the modified Peña procedure. Neonates were placed in the supine position. Then, the deep tissues were dissociated along the fistula to separate the rectum and posterior vaginal wall. The integrity of the posterior commissure of the labia was preserved during the surgery and the levator ani muscle and ischiorectal muscle complex were not incised, the same as in the modified Peña procedure.

The anus was dilated for 3 to 6 months in all neonates 2 weeks post-surgery and postoperative follow-up was performed for 1 to 10 years.

### Observations

The following parameters were observed and compared between the two groups. (1) Incidence of complicating malformations and age at operation; (2) Therapeutic efficacy in the perioperative period, including duration of preop-

erative preparation, duration of operation, intraoperative blood transfusion volume, duration of postoperative fasting, duration of antibiotic use, length of hospital stay, cost, incidence of short- and long-term postoperative complications, and reoperation rate; (3) Assessment of bowel movements 36 months postoperatively according to the Krickenbeck classification [12]; and (4) Anorectal manometry 12 months postoperatively. Postoperative bowel movements were evaluated with a high-resolution, multi-channel manometer for detecting gastrointestinal function (Polygram HR, Denmark) and detection parameters included rectoanal inhibitory reflex (RAIR), resting rectal pressure, active systolic blood pressure, and rectal compliance.

### Statistical analysis

Comparisons were performed with the student's t-test, while differences of proportions were tested for statistical significance with the chi-squared test. A *p*-value of less than 0.05 was considered statistically significant.

### Results

Screening for the VACTERL syndrome (vertebra/anus/cardiac/trachea/esophagus/radius/renal/limb anomalies) demonstrated that, of the 39 neonates with RVF undergoing the modified Peña procedure, 11 patients had mild cardiac malformations (9 with patent ductus arteriosus, one with an interventricular septal defect, and one with an atrial septal defect), 3 patients with urinary system malformation (2 mild uronephrosis and one solitary kidney), 2 patients with esophageal atresia, and one patient with polydactylism. Among the 50 neonates with RVF undergoing transperineal anal transposition, 16 had mild cardiac malformations (12 patent ductus arteriosus, 3 interventricular septal defects, and one atrial septal defect), 7 urinary system malformations (4 mild uronephrosis, one solitary kidney, one renal dysplasia, and one renal duplication), one patient with absence of the coccyx, and one case with sacrococcygeal pilonidal sinus. There was no significant difference in the incidence of complicating malformations between groups.

**Table 1** describes the clinical characteristics of the study subjects. The age of the neonates undergoing the modified Peña procedure ra-

## One-stage postoperative function of RVF

**Table 1.** Comparison of the therapeutic efficacy between the modified Peña procedure and the transperineal anal transposition groups during the perioperative period

Group	Age at surgery (month)	Proctectasia (count)	Preoperative preparation (day)	Duration of operation (minute)	Intraoperative blood transfusion (count)	Postoperative fasting (day)	Antibiotic usage (day)	Total length of hospitalization (day)	Cost of hospitalization (Yuan)
Modified Peña procedure (n=39)	1.6±0.6***	0***	2	76±15	0	4	4±1	11.0±2.6	14213.24±2548.78
Transperineal anal transposition (n=50)	4.1±1.0	31	2	82±20	0	4	4±1	12.0±3.0	13509.37±2371.89

Note: \*\*\*P<0.001.

## One-stage postoperative function of RVF

**Table 2.** Comparison of bowel movements between the modified Peña procedure and the transperineal anal transposition groups 36 months postoperatively

Group/Number	Voluntary defecation	Soiling			Constipation		
		Grade I	Grade II	Grade III	Grade I	Grade II	Grade III
Modified Peña procedure (n=39)	24	4*	0	0	6	4*	1*
Transperineal anal transposition (n=50)	26	3	2	0	5	11	3

Note: \*P<0.05.

**Table 3.** Comparison of anorectal manometric results between the modified Peña procedure and the transperineal anal transposition groups 12 months postoperatively

Group	Rectoanal inhibitory reflex (count)	Rectal resting pressure (mmHg)	Active systolic blood pressure (mmHg)	Rectal compliance (mL/mmHg)
Modified Peña procedure (n=17)	0	50.8±1.1	52.5±1.4	5.5±1.2
Transperineal anal transposition (n=26)	0	52.2±1.3	52.4±3.0	3.5±1.1

nged from 9 days to 2.5 months, with a mean age of 1.6 months. The age of the neonates undergoing transperineal anal transposition ranged from 5 to 8 months, with a mean age of 6.4 months. The mean age at surgery was significantly lower in the modified Peña procedure group compared with the transperineal anal transposition group (P<0.001). Preoperative transfistular radiography and intraoperative examinations showed remarkably dilated rectum in 31 cases undergoing transperineal anal transposition (62.0%, 31/50), and no cases with proctectasia in the modified Peña procedure group; there was a significant difference in the incidence of rectal dilation between groups (P<0.001). However, there were no significant differences found in the duration of preoperative preparation, duration of operation, intraoperative blood transfusion volume, duration of postoperative fasting, duration of antibiotic use, length of hospital stay, or cost between groups (P>0.05).

Among the 39 neonates undergoing the modified Peña procedure, there were 4 cases of postoperative superficial sacral wound infection or dehiscence (10.3%, 4/39), which resolved with conservative treatment. One patient developed an anal stricture (2.6%, 1/39) which required a second surgery after 3 months, for a 2.6% (1/39) reoperation rate. Of the 50 neonates undergoing transperineal anal transposition, three developed postoperative rectal retraction (6.0%, 3/50), one fistula recurrence (2.0%, 1/50), and one anal stricture (2.0%, 1/50). All of these 5 patients underwent a second surgery after 3 to 5 months, for a 10.0% (5/50) reoperation rate. There was no signifi-

cant difference in the incidence of short-term postoperative complications between groups (P>0.05); however, the reoperation rate was significantly higher in the transperineal anal transposition group (P=0.0073). All neonates underwent postoperative follow-up for 1 to 10 years. Assessment of bowel movements 36 months after surgery showed a significantly lower incidence of postoperative soiling in the modified Peña procedure group (P=0.049) and no significant difference in the incidence of grade I postoperative constipation between groups (P>0.05); however, a significantly higher incidence of grade II and III postoperative constipation was found in the transperineal anal transposition group compared with the modified Peña procedure group (P=0.049 and 0.026, respectively; **Table 2**).

Seventeen of 39 neonates in the modified Peña procedure group and 26 of 50 in the transperineal anal transposition group received anorectal manometric assessment 12 months postoperatively; anorectal manometry revealed no induction of rectoanal inhibitory reflex in either group. There were no significant differences in resting rectal pressure and active systolic blood pressure between groups (P>0.05); a significantly lower rectal compliance was observed in the transperineal anal transposition group compared with the modified Peña procedure group. However, this difference was not found to be significant (**Table 3**).

### Discussion

Currently, there are multiple surgical options used to treat congenital imperforate anus with

## One-stage postoperative function of RVF

RVF, including early-stage cutback surgery, anal transposition, Y-V plasty, X-Z plasty, and the recently developed PSARP and anterior sagittal anorectoplasty (ASARP) [2-4]. Transperineal anal transposition, which is also termed neutral sagittal anorectoplasty [13], was employed for treatment of RVF in our hospital before 2007. Such a surgery preserves the integrity of the posterior commissure of the labia between the vagina and anus. In the current study, three patients with postoperative rectal retraction were observed following transperineal anal transposition. It has been shown that the transperineal approach is difficult to perform for surgical therapy of RVF (notably, for RVF complicated by proctectasia) and that inadequate dissociation of the rectum results in a high incidence of postoperative complications (10.0%) and reoperation (10.0%). Therefore, the modified semi-PSARP (modified Peña procedure) was first used for treatment of neonatal RVF in our hospital. Unlike the classical PSARP, this surgery preserves the levator ani muscle and ischiorectal muscle complex while the PSARP completely incises the levator ani muscle and ischiorectal muscle complex. We used the one-stage modified Peña procedure for radical treatment of RVF in neonates, and achieved a high therapeutic efficacy [6]. In the current study, we aimed to evaluate the clinical efficacy of the one-stage modified Peña procedure for therapy of RVF in 39 neonates, so as to summarize the clinical experiences of the one-stage modified Peña procedure for the treatment of RVF.

Generally, neonates with RVF have a well developed sacrum and anal sphincter. Among the 91 neonates with RVF admitted to our hospital during the 13-year period between 2001 and 2013, only 1 patient (1.1%, 1/91) had a severe hemivertebral deformity of the sacrococcygeal vertebrae (this patient was not enrolled in our study). Therefore, except for a small minority of neonates with RVF complicated by poor development of the perineal region and a sacral anomaly who have no ability to control bowel movements, the majority of the neonates with RVF have a potential ability to control bowel movements at birth (93%) and may have voluntary bowel movements beginning at age 3 years [14], indicating that neonates with RVF have a good postoperative prognosis with regards to bowel movements and social com-

munication. This finding suggests that any complication (such as wound infection or dehiscence) postoperatively is not acceptable, since these complications may alter the final functional prognosis. In our cohort, four of 39 neonates undergoing the modified Peña procedure for treatment of RVF had postoperative superficial sacral wound infection or dehiscence which did not involve the deep muscle tissues, were cured following conservative therapy. Wound infection or dehiscence may be associated with failure to place the neonate in a warmer postoperatively, or inadequate care of the unexposed sacral incision. In addition, one of the 39 neonates who underwent the modified Peña procedure and one of the 50 patients who underwent transperineal anal transposition developed a postoperative anal stricture; in these cases, the parents did not persist with anal dilation because their children cried during anal dilation. Therefore, one-stage surgery is currently recommended for the treatment of all forms of ARM in neonates [15]. The superiority of this surgical option is the neonates' small age and absence of memory which is easier for anal dilation and early-stage formation of new synapses surrounding the rectum, thereby facilitating control of bowel movements. Compared with the modified Peña group, higher reoperation rate in the transperineal anal transposition group was closely related to older ages and the degree of the rectal dilation. We found that the mean age of the neonates at surgery was significantly lower in the modified Peña procedure group compared with the transperineal anal transposition group; none of the neonates in the modified Peña procedure group developed proctectasia due to small age. Furthermore, one of the 50 neonates with RVF who underwent transperineal anal transposition developed fistula recurrence from an unknown cause. Review of the neonates' medical records revealed successful surgical procedures and postoperative recovery and that anal dilation was initiated 2 weeks post-surgery, with fistula recurrence seen 3 weeks post-surgery. Fistula recurrence may be associated with ischemia and injury of the anterior rectal wall due to excessive electrocautery. Wakhlu et al. [16] also reported a similar iatrogenic injury. Thus, surgeons must not only be familiar with the anatomy of the tissues and have abundant surgical experience, but also fully consider every

perioperative pitfall so as to avoid compromising the final functional prognosis.

Since PSARP was introduced in 1982, the major challenge for postoperative management for ARM is not fecal incontinence, but chronic constipation caused by bowel dysfunction, which notably affects neonates with RVF [5]. We found a 28.2% (11/39) incidence of postoperative constipation in neonates with RVF undergoing the modified Peña procedure, which was significantly lower than those with transperineal anal transposition (38.0%, 19/50) and classical PSARP (40%) [17]. We also found a significantly higher incidence of grade II and III postoperative constipation in the transperineal anal transposition group, which may be associated with the development of megarectum and incomplete intraoperative incision of the dilated intestinal tube. Peña [5] reported that excessive preservation of the blind end of the rectum may increase the incidence of postoperative constipation in neonates with a moderate- or low-type ARM, which was directly related to the degree of preoperative rectosigmoid dilation [18]. In addition, a lower incidence of postoperative soiling in the modified Peña group may be associated with normal sacrum and spine of the patients.

In conclusion, the one-stage modified Peña procedure results in better long-term control of postoperative bowel movements compared with transperineal anal transposition for congenital neonatal imperforate anus with RVF, which may be not only associated with the technique, but also the younger patients in the former group; however, further studies with a larger sample size and long-term follow-up studies are required to guide recommendations for clinical application.

### Acknowledgements

We acknowledge the work of all the surgeons involved in the study in our hospital. Supported by National Natural Science Foundation of China (81401243), Shanghai Health Bureau "Scientific research Foundation" (2014-2016), Shanghai Fudan University "Excellent Study Plan" (2015-2018).

### Disclosure of conflict of interest

None.

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