Long-Term Outcome of the Pippi Salle Procedure for Intractable Urinary Incontinence in Patients with Severe Intrinsic Urethral Sphincter Deficiency

Shigeru Nakamura,* Taiju Hyuga, Shina Kawai and Hideo Nakai

Department of Pediatric Urology, Jichi Medical University, Children's Medical Center Tochigi, Tochigi, Japan

Abbreviations and Acronyms

A-ISD = anatomical intrinsic sphincter deficiency
AC = augmentation cystoplasty
BNR = bladder neck reconstruction
CIC = clean intermittent catheterization
Dx/HA = dextranomer-hyaluronic acid copolymer
FS = fascial sling
IBA = injection of bulking agent
ISD = intrinsic sphincter deficiency
N-ISD = neurogenic intrinsic sphincter deficiency
PSP = Pippi Salle procedure
TUI = transurethral incision of urethra
UDS = urodynamic studies
UTI = urinary tract infection
VUR = vesicoureteral reflux

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* Correspondence: Department of Pediatric Urology, Jichi Medical University, Children's Medical Center Tochigi, 3311-1, Yakushiji, Shimotuke, Tochigi 329-0498, Japan (telephone: 81-285-44-2111; FAX: 81-285-44-8452; e-mail: naka251148@gmail.com).

Purpose: We evaluated the long-term outcome of the Pippi Salle procedure in patients with severe intrinsic urethral sphincter deficiency.

Materials and Methods: We performed the Pippi Salle procedure in 6 males and 6 females with severe intrinsic sphincter deficiency between March 2003 and August 2013. Median patient age was 15 years (range 6 to 45). Mean followup was 75 months (range 17 to 142). Six males and 3 females had neurogenic intrinsic sphincter deficiency (spina bifida in 8 and spinal cord injury in 1). Three females had anatomical intrinsic sphincter deficiency (idiopathic bladder hypoplasia in 2 and pseudo-ureterocele in 1). Four patients had previously undergone bladder neck surgery, 3 had been treated with endoscopic injection of collagen, 2 had undergone fascial sling and 1 had been treated with tension-free vaginal tape surgery. The Pippi Salle procedure was performed alone (2 patients), or in combination with bladder augmentation (4) or catheterizable abdominal stoma (1), or both (5).

Results: Complete dryness was achieved in 7 patients (58%). Of 9 patients with neurogenic intrinsic sphincter deficiency 7 (78%) achieved complete dryness. Eight patients experienced complications, including continued urinary incontinence (5), difficulty catheterizing per urethra (3) and urinary calculi (1). These 8 patients were successfully treated with additional endoscopic interventions, including injection of collagen in 4, injection of dextranomer-hyaluronic acid in 1, transurethral incision of urethral kink in 3 and vesicolithotripsy in 1. After these simple interventions complete dryness was achieved in all 12 patients.

Conclusions: Although we experienced some minor complications in the short term, most patients were simply and successfully treated with endoscopic surgery. The long-term results of the Pippi Salle procedure are promising.

Key Words: urethra, urinary bladder, urinary incontinence, urologic surgical procedures

Intrinsic urethral sphincter deficiency associated with refractory urinary incontinence includes neurogenic and anatomical varieties. Neurogenic disease is associated with spina bifida, while anatomical disease is associated with bladder exstrophy, epispadias, bilateral ectopic ureters, ureterocele, pseudo-ureterocele and other conditions. The treatment for intrinsic sphincter deficiency is extremely challenging for urologists and involves methods to improve bladder outlet resistance, such as injection of a bulking agent, fascial sling, bladder neck reconstruction,

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artificial urinary sphincter and bladder neck closure. Because each of these methods has its own merits and drawbacks, no single method is effective for all patients with severe intrinsic sphincter deficiency.

We perform fascial sling as the first choice in females with neurogenic ISD using CIC to evacuate urine, because this procedure has frequently been reported as effective in these patients.[1–4] Since 2003, we have performed the Pippi Salle procedure in males with neurogenic ISD, patients with anatomical ISD and patients undergoing bladder neck reoperation.

There are few previous studies of PSP, and only short-term treatment outcomes (average up to 2 years) have been reported.[5–9] To our knowledge, no study of long-term outcomes exists. We examined the long-term treatment outcomes of PSP, including postoperative complications and outcomes of additional treatment for complications.

MATERIALS AND METHODS

Patients

A total of 18 patients underwent surgery for ISD at our institution between March 2003 and August 2013. Patients with N-ISD underwent FS (4 females) or PSP (6 males and 3 females). The Leadbetter procedure (trigonal tubularization) was performed in 1 male and 1 female with A-ISD (covered cloacal exstrophy and giant pseudo-ureterocele, respectively), who were deemed able to void spontaneously. Three patients with A-ISD requiring CIC for urine evacuation underwent PSP.

We retrospectively evaluated the 12 patients who underwent PSP during the study period. Informed consent was obtained from all patients and their parents before beginning the study, which was approved by the ethics committee of our institution.

Table 1 shows the characteristics of the 12 patients by date of surgery (2003 to 2008 in patients 1 to 5 and 2009 to 2013 in patients 6 to 12). There were 6 males and 6 females, and median age at surgery was 15 years (range 6 to 45). Six males and 3 females had N-ISD, with primary disease consisting of spina bifida (8 patients) and spinal cord injury resulting from traffic injury (1). Three females had A-ISD, with primary disease consisting of bladder hypoplasia of unknown cause (2 patients) and pseudo-ureterocele (1). Four females had undergone bladder neck surgery, while 3 patients had undergone IBA (collagen), 2 had undergone FS and 1 had undergone tension-free vaginal tape surgery. Two patients (Nos. 3 and 6) had a history of UTI preoperatively, both of whom presented with right VUR and ipsilateral renal scarring.

All patients underwent preoperative examination, which included urinalysis, urine culture, ultrasound, cystography, dimercaptosuccinic acid renal imaging and UDS. Patients underwent followup consisting of urinalysis every 1 to 3 months, ultrasound and abdominal radiography every 6 months, and cystography and UDS at 3 to 4 months, 1 year, 3 years and 5 years postoperatively. Indications for PSP were 1) open bladder neck on cystography during low pressure urine storage and 2) abdominal leak point pressure less than 40 cm H_2O based on UDS.

Surgery

Urethral lengthening was performed according to the modified PSP.[8] For patients 1 to 5 (first half of study) the size of the anterior bladder wall flap and the urethral width at the trigone and posterior bladder wall were set according to the original procedure. Thus, the width of the base and tip of the anterior bladder wall flap were 25 and 15 mm, respectively, the length of the flap was 50 mm and the urethral width at the posterior bladder wall was 10 mm. As a result, urinary continence status was insufficient. Thus, for patients 6 to 12 (second half of study) the width of the base and tip of the anterior bladder wall flap were changed to 25 and 10 mm, respectively, the length of the flap was changed to 60 to 80 mm and the urethral width of the posterior bladder wall was changed to 7 to 8 mm. A neourethra was reconstructed to be smaller in diameter and longer compared to the original procedure.

Patients who underwent lower urinary tract reconstruction simultaneously with PSP are indicated in table 1. Nine patients with poor vesical compliance even after administration of anticholinergic drugs underwent augmentation cystoplasty using the intestine (small intestine in 4 and sigmoid colon in 5). Six patients who had urethral pain during catheterization or difficulty performing CIC through the urethra due to wheelchair confinement underwent creation of a continent catheterizable abdominal stoma (Mitrofanoff in 4 and Yang-Monti in 2). Seven patients underwent ureteral reimplantation. Of these patients 2 (Nos. 3 and 6) presented with VUR, history of UTI and renal scarring. For the remaining 5 patients the ureteral orifice had to be relocated during intraoperative urethral lengthening.

Study Outcomes

The short-term (less than 6 months postoperatively) and long-term outcomes of PSP were evaluated in terms of urinary continence status and postoperative complications. "Dry" status was defined as being completely free from urinary incontinence during the day and night, or with a small amount of urine leakage not requiring use of a pad with CIC performed every 4 hours during the day. "Wet" status was defined as all other conditions. No control group was applicable, nor was statistical analysis.

RESULTS

Short-Term Outcomes

Among patients treated in the first half of the study (Nos. 1 to 5) 4 were "wet" (tables 1 and 2). Of these patients 3 had A-ISD. All 7 patients treated in the second half of the study (Nos. 6 to 12) had N-ISD. The urethra was reconstructed to be smaller in diameter and longer than in the original procedure. As a result, 6 of the 7 patients achieved "dry" status. Overall 7 patients (58%) achieved "dry" status in the