



Case report

Single-stage repair for female epispadias with urinary incontinence: A case report and literature review

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ABSTRACT

Background: Epispadias is a rare condition. Epispadias in females is two times less common than in males. Female epispadias range from 1 in 160,000 to 480,000 live births. Epispadias can be diagnosed through careful physical examination of the genital. Surgery is the management of epispadias. Surgical management of epispadias is quite tricky and requires expertise. The literature that discusses female epispadias is challenging to be found. In this paper, we would like to report surgical management of isolated female epispadias in Cipto Mangunkusumo Hospital, Jakarta.

Case presentation: A 7-year-old girl was admitted with a chief complaint of urinary incontinence since birth, during daytime and nighttime. Urinary incontinence was not induced by activities nor worsened by eating/drinking. Physical examination showed that the patient's external genitalia has underdeveloped labia minora, patulous urethra, bifid clitoris, and multiple hypopigmentation lesions. Laboratory results were in the normal range. The voiding cystourethrography (VCU) result revealed urine leaks during the filling phase. The bladder wall was normal, and no vesicoureteral reflux (VUR) appeared. The urethrocystoscopy shows a more vertical OUE, a wide-open bladder neck, and a urethral length of 1.5 cm. The patient underwent single-stage surgical procedures that consist of epispadias repair and bladder neck reconstruction through a subsymphyseal perineal approach. No complications occurred intra-operative and post-operative. At the 1-week and 6-months follow-up, the patient achieved urinary continence, and the surgical wound healed normally.

Discussion: Epispadias is a rare condition that could occur in various degrees, from mild to severe degrees. To a severe degree, there is a split at the entire urethral and involves the bladder neck causing constant incontinence for the patient. Epispadias cases are quite challenging to diagnose. The physician needs to separate the labia majora and perform the physical examination carefully. The objective goals of surgical management of epispadias are to achieve urinary continence, restoration of anatomy, function, and cosmetic appearance of urethra and genitalia. Single-stage reconstruction is the current surgical method used to repair isolated female epispadias.

Conclusion: Females epispadias is a rare congenital anomaly that is quite often to miss diagnosed. The single-stage technique is the recommended surgical technique for isolated female epispadias. This patient has achieved urinary continence, and no complications occurred during and post-surgery.

1. Background

Epispadias is a condition when the urethral plate is wide-open, and the opening of the urethral is located in an abnormal location [1,2]. In females, the opening is usually located between the clitoris and labia or located in the belly area [2]. Epispadias condition caused by the urethral tube failing to tubularize on the dorsal aspect. There are varying degrees of epispadias abnormality. Epispadias can be found isolated or with bladder exstrophy. In females with epispadias, a bifid clitoris, vaginal

opening displaced in the anterior part, patulous urethral opening, an absent or abnormal form of mons, with varying degrees of urine incontinence can be found [2].

Epispadias is a rare condition. The prevalence of epispadias is estimated at around 1 in 10,000 to 50,000 live births [1]. Epispadias in females is two times less common than in males. Female epispadias range from 1 in 160,000 to 480,000 live births [3]. More than 60% of cases of epispadias is an accounted for by isolated epispadias [4]. Epispadias can be diagnosed with the clinical investigation, but

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Fig. 1. External genitalia examination showed under-developed labia minora, patulous urethra, no clitoris, multiple pustules around the paravaginal area.

additional investigations such as USG genitourinary system and cystourethrography are needed to exclude anomalies in other organs [1]. The management of epispadias is through surgery. Surgical management of epispadias is quite tricky and requires expertise. The goals of the surgery are to reconstruct and provide optimal function and cosmetics for the genital and urethra [1].

The literature that discusses female epispadias was difficult to be found. Only a few kinds of literature discuss female epispadias and mostly are case report articles. Several case reports recommended the best approach and management of isolated female epispadias [5–9]. Clinical approach and evaluation of epispadias are through careful physical examination of genital, radiology evaluation (ultrasound and intravenous urography to evaluate other anomalies in genitourinary system), voiding cystourethrogram (VCUG) to evaluate bladder capacity, vesicoureteral reflux, and urethra, lab and urine analysis (to evaluate biochemical parameters and urinary tract infection or kidney disease), and urethro-cystoscopy to evaluate the urethra length, roof, and bladder neck [5,6]. The recommended surgery method is a single step of urethroplasty, bladder neck reconstruction, and perineoplasty surgery [5,9]. In this paper, we would like to report on surgical

management of isolated female epispadias and a literature review of the development of surgical techniques. We hope that with this case report, researchers could extend their knowledge about treatment and management of female epispadias.

2. Case presentation

A 7-year-old girl was admitted with a chief complaint of urinary incontinence since birth, during daytime and nighttime. Urinary incontinence did not induce by activities nor worsened by eating/drinking. There was no history of fever, cloudy urine, hematuria, passing stone, dysuria, and flank pain. The patient drank around 1000 mL of bottled mineral water per day and changed diapers 4 times/day. Urine color was clear yellow, urine volume estimated around 600–700 mL/day, and have normal defecation. The patient has a history of a skin rash around the paravaginal area and has been treated with cauterization by a dermato-venereologist in April 2020. There is no history of spinal trauma. The patient was birth through vaginal delivery, a term birth, birth weight of 2800 g. The patient also has normal developmental milestones. The patient is the second child in the family, and her sister has no similar complaint and no familial history related to any congenital anomalies. The patient was first diagnosed with epispadias and received bladder training from a urologist at the district hospital for three months, without any improvement being noted. Then, she was referred to Cipto Mangunkusumo General Hospital for repair surgery.

Physical examination showed underdeveloped labia minora, patulous urethra, bifid clitoris, and multiple hypopigmentation lesions (Fig. 1). The patient has a normal position of the anus, and there is no mass or bulging in the flank area, no anal dimple, or other abnormalities as a sign of spina bifida. The vital sign was in the normal range. The patient has a bodyweight of 29 kg, height of 126 cm, and BMI of 18.3 kg/m².

Laboratory results were in a normal range. However, at urinalysis tests were found cloudy yellow color urine, leucocyte 6–8/HPF, erythrocyte 0–1/HPF, bacteria (+), pH 8.5, SG \leq 1.005, nitrite (+), LE (–). Although the bacteria and nitrite were positive, this could not be diagnosed as a UTI because the leucocyte esterase was negative and the patient did not have clinical symptoms of UTI [10].

At the prior hospital, a CT Whole Abdomen contrast was done and showed normal shape and size of the right and left kidney, and there was neither hydronephrosis, mass, stone, nor cyst. The cystoscopy was also done, showing that the external orifice urethra was located more vertically, and the bladder neck was widely open.

At Cipto Mangunkusumo Hospital, voiding cystourethrography (VCUG) was done to evaluate the bladder capacity, vesicoureteral reflux, and urethra (Fig. 2). During VCUG examination, the patient had perceived the desire to void. A continuous urine leak was observed during the filling phase after injection of 50 mL of contrast agent. The bladder wall was regular, without the appearance of filling defect, Christmas tree appearance, and vesicoureteral reflux (VUR). The patient was able to empty her bladder completely.



Fig. 2. Voiding cystourethrography (VCU) result shows incontinence during filling. Normal bladder wall and no vesicoureteral reflux (VUR).



Fig. 3. The urethroscopy result showed a urethral length of 1.5 cm, the bladder neck was widely open (patulous urethra) causing urinary incontinence.

The surgical procedures for this patient were epispadias repair and infrasympysis bladder neck reconstruction. The operation was performed by Dr. Irfan Wahyudi, M.D. The patient was in the lithotomy position. Initially, before the surgical procedure, the urethroscopy was done to evaluate the urethra and bladder neck. The urethroscopy was done with a sheath 9 Fr and 0° lenses inserted through the orificium urethra externum (OUE) to the bladder. Urethroscopy showed the urethral length (bladder neck to OUE) was 1.5 cm (Fig. 3). The bladder could not expand maximally due to a wide-open bladder neck.

The surgery procedure began with incisions at OUE and mons pubic in the area that has been marked before (Fig. 4). The urethral incision at 12 o'clock direction and an intersymphyseal band were incised to expose the bladder structure. The incision continued to the mucosa layer and seromuscular layer of the bladder. Then, the open cystostomy was set up with a Foley catheter 12 Fr, and the balloon was inflated with 10 mL of water. Urethral lengthening and bladder neck reconstruction was done simultaneously. Urethral lengthening was done with tubularization technique over an 8 Fr Silastic stent. The use of a Silastic stent 8 Fr was for urethral splinting. The anterior bladder wall and the bladder neck were sutured to achieve a funneling configuration. The step was done by PDS 5.0 interrupted suture. Then the bladder was closed with a 2-layer of the interrupted suture. After the bleeding control was done, there is no active bleeding that comes out from the wound, then the operation was finished. There is no reconstruction of the labia minora procedure in this patient. During the surgery, the bleeding was minimal, and there are no intraoperative complications. The follow-up was done at 1-week and 6-months after the surgical procedure. At 1-week post-surgery follow-up, the physical examination shows there is no bleeding or infection at the surgical wound suture. The patient also achieves normal urination and urinary continence, and the vital signs are in the normal range. At 6-months post-surgery, follow-up also shows no complication, and the wound normally heals to scar (Fig. 5).

3. Discussion

Epispadias in females is two times rarer than in males and 1,4 times rarer for isolated epispadias cases [4]. Epispadias in females occur one around 484,000 live births [11], but Allen et al.'s study estimated that because of the missed diagnosis, the epispadias in females is three times more common than that has been estimated, which is one in 160,000 female live birth [3]. Isolated epispadias account for more than 60% of epispadias cases [12]. The embryogenesis and organogenesis of the female genitourinary system are complex. The problem that happens during the embryogenesis process may cause abnormal development in urinary and genital tracts [5]. Epispadias could occur because of genital tubercle malposition, which made tubularization of the urethral incomplete [13].

Epispadias could occur in various degrees. Based on Davis's classification, epispadias can be classified into mild to severe degrees. In the mild degree, the urethral orifice is patulous. In a moderate degree, there is a dorsal split at the urethral, and in severe degrees, the split is at the entire urethral and involves the bladder neck causing constant incontinence for the patient [7,14]. But another study also states that there is no correlation between anatomical abnormality and incontinence degree because the correlation between abnormal sphincter and urethral abnormality is not absolute [3]. Bladder neck abnormality, low bladder capacity, or low urethral pressure are the frequent causes of incontinence in isolated female epispadias [15]. Epispadias patients with urinary incontinence may have skin eruption around the perineum because of the continued use of the diaper [9].

Epispadias cases are quite challenging to diagnose when there are no pelvic or spinal abnormalities. The physician needs to separate the labia majora and perform the physical examination carefully so the isolated female epispadias would not be missed [9]. In isolated female epispadias, the vagina, and internal genitals are usually normal. Radiological evaluation (voiding cystourethrogram) is needed to evaluate bladder reflux and exclude other causes of incontinence. Cystourethroscopy or cystoscopy is also needed to evaluate the bladder capacity and

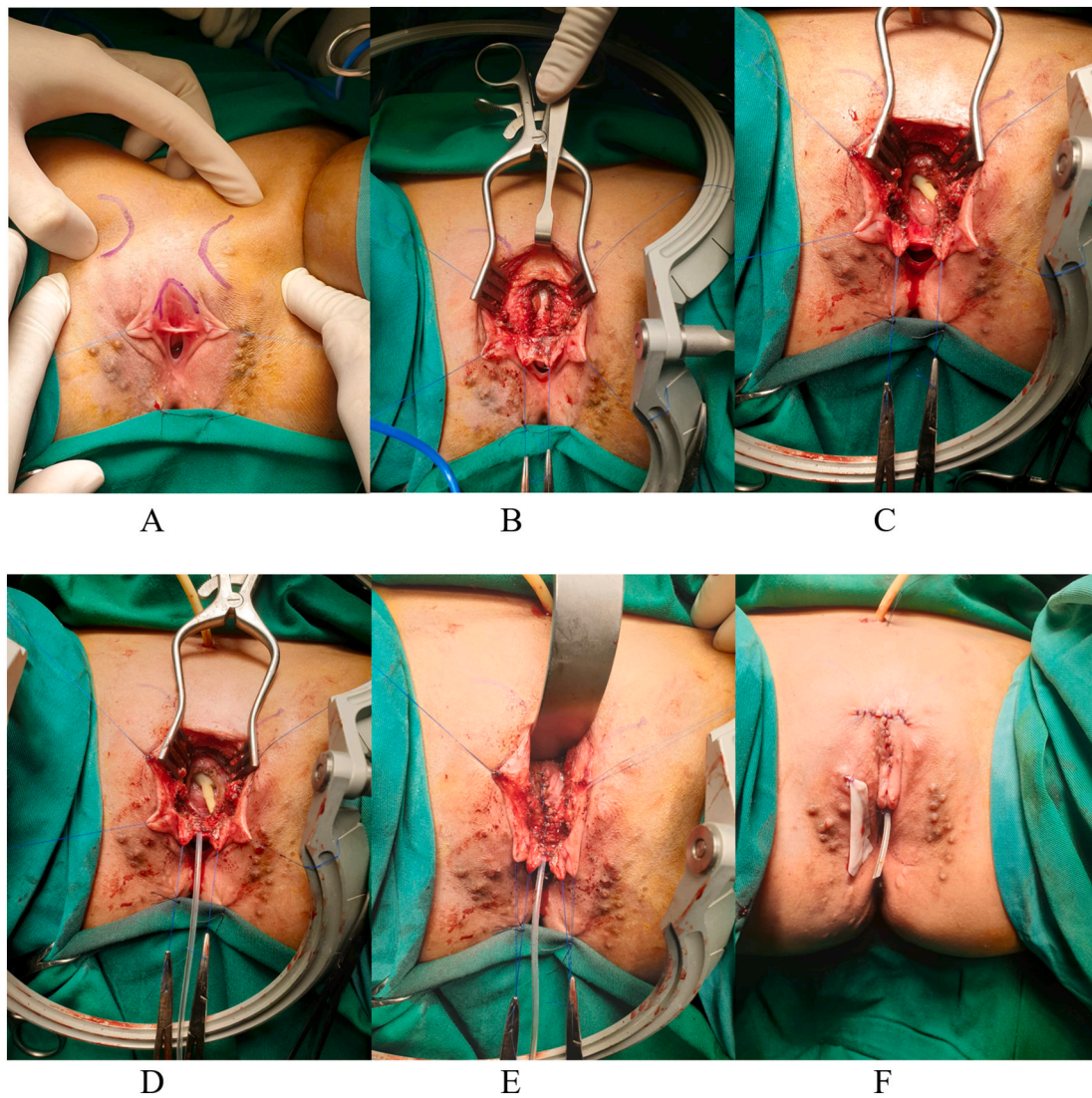


Fig. 4. The surgical procedure of epispadias repair and bladder reconstruction.

Pre-operative condition showed the incision mark and sutures traction at 9 and 3 o'clock (A). The incision of mucosa and the seromuscular layer of the bladder (B). The setting up of open cystostomy (C). Urethral lengthening with Silastic stent guiding and bladder neck reconstruction (D). Complete bladder closure (E). Complete mons closure (F).



Fig. 5. External genitalia condition 1-week post-surgical procedure (left) and 6-months post-surgical procedure (right).

urethral orifices [7].

Based on Davis classification, this patient can be classified into severe cases because there was bladder neck involvement, and the urinary incontinence occurred since birth. The Radiological evaluation (voiding cystourethrogram) and cystourethroscopy were done on this patient to evaluate the vesicoureteral reflux, bladder capacity, urethral length, and urethral orifice position. This patient has the desire to void, able to empty the bladder, has no vesicoureteral reflux, widely open bladder neck, and short urethral length. Single-stage surgery was done to repair the epispadias and bladder reconstruction.

The objective goals of surgical management of epispadias are to achieve urinary continence, restoration of anatomy, function, and cosmetic appearance of urethra and genitalia. There are two kinds of surgical methods of epispadias two stages and single-stage reconstruction. The oldest method was the two-stage reconstruction that was developed by Young in 1908 [9]. The technique has been modified by Dees and Leadbetter [30]. The Young-Dees-Leadbetter technique is the technique commonly used and the gold standard technique for bladder neck reconstruction. The operation technique has continence rates around 60% to 85.5% [15]. However, the technique reduces the bladder capacity, and it is difficult to achieve continence in small bladder

Table 1
List of techniques for epispadias reconstructive procedure.

Authors	Kind of technique	Technique	Outcome
Stages Young, 1922 [16]	Two-stages reconstruction	Radical urethral and genital reconstruction delayed bladder neck reconstruction at the age of social continence. Double sphincter technique (excision of wedge tissue of anterior bladder neck and removal of the wedge tissue at proximal of the external sphincter).	
Young-Dees, 1949 [17]	Two-stages reconstruction	Modified Young technique, Dees added urethra lengthening to narrowing and revise proximal tube	
Young-Dees-Leadbetter, 1964 [17]	Two-stages reconstruction	Modified Young-Dees technique Leadbetter added muscular flaps at the proximal tube and placed ureters higher in the bladder (ureters reimplantation) to allow further trigonal tube elongation.	
Marshall-Marchetti, 1949 [18]	Bladder Neck Reconstruction (BNR): Vesicourethral suspension	Reconstruction of the urethra and bladder neck by simple elevation and fixation of the bladder neck and urethra to the pubis and rectus muscles.	89% of 44 patient has a significant improvement in urinary control 11% of the patient has no improvement in urinary control
King and Wendel, 1970 [19]	BNR: Transvaginal plication	Reconstruction of the urethra and bladder neck with layers of plicating sutures with urethroscopy assistance until a view of bladder neck sphincter closing can be seen.	2 of 6 patients achieve continence. 2 of 6 patients achieve continence after the subsequent Leadbetter procedure. The rest patient achieves continent for a short time and continent in some urgency.
Tanagho, 1981 [20]	BNR: Bladder flap procedures	Anterior tube reconstruction, separation of bladder and urethra, developed 1-in. long and wide of anterior bladder flap.	70% success rate of urinary continence
Gearhart, 1993 [21]	Two-stages reconstruction	Primary closure of epispadias to increase bladder capacity. Urethral and genital reconstruction at 18–24 months, Young-Dees-Leadbetter bladder neck reconstruction at 4–5 years old.	Bladder capacity >80 cc, continence rate 85%
Single Hendren, 1981 [22]	Single-stage perineal and	Consists of excision of the abnormal wide roof of the distal urethra,	Complete repair of the abnormal wide urethra

Table 1 (continued)

Authors	Kind of technique	Technique	Outcome
	transvesical approach	narrowing urethral caliber from below, narrowing the upper urethra and bladder neck from above, and ureteric reimplantation.	which is better than other alternatives procedures
Kramer and Kelalis, 1982 [23]	One-stage operation	Single stages procedure of Young-Dees-Leadbetter and Transvaginal plication technique. The minimum age of surgery is 3 years old.	83% of patients achieve urinary continence
Kelly, 1995 [24]	Radical soft-tissue mobilization	Pelvic and perineal approaches, consist of excision of external full-thickness triangles to perform bladder neck funneling, wrapping the bladder neck and upper urethral with muscular fibers, urethral tubularization reconstruction, and corporoplasty. Ureteral submucosal cranial reimplantation was performed if necessary.	100% diurnal continence, 37% full continence at 5 years old
De Jong, 2000 [25]	Single-stage urethral reconstruction with the transperineal approach	Combined urethroplasty with percutaneous bladder neck suspension that moved the bladder neck and proximal urethra into intra-abdominal positions and reconstruction of pelvic floor	3 of 4 patients achieve continence 1 patient had a recurrent urinary tract infection No major bladder neck surgery or ureteral reimplantation is needed in the absence of vesicoureteral reflux.
Manzoni and Ransley, 2007 [26]	Single-stage perineal approach	The perineal approach of urethral reduction and angulation, urogenital diaphragm repair, and external cosmetic correction	Increase outflow resistance and bladder capacity. Initial procedure if continence surgery may be needed later. 60–85% diurnal continence <50% night-time continence Expected ladder capacity 55% and 58%
Bhat, 2008 [15]	Single-stage perineal approach	Perineal urethroplasty, double breasting of the urethra and bladder neck, and sphincteroplasty.	Increase urethral and bladder neck resistance and bladder capacity. 3 of 4 patients achieve continence with dry intervals of 4–5 h
Kajbafzaden, 2011 [27]	Single-stage perineal approach	Subsymphyseal cystoscopic-guided of bladder neck plication and urethrogenitoplasty	10/10 patients achieve dry periods >3 h of diurnal time 6/10 patients achieve full continence

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Table 1 (continued)

Authors	Kind of technique	Technique	Outcome
Macedo, 2015 [28]	Single-stage perineal approach	Perineal infra pubic approach, bladder neck, and proximal urethra tightening, urethral tubularization from the mucosal flap, and 6F catheter guided. Labia minora plasty, mentoplasty, and vulvoplasty.	Has the possibility in restoring cosmetics and providing resistance, preventing the need for abdominal bladder neck surgery
Yadav, 2017 [29]	Single-stage perineal approach	Infrasymphyseal bladder neck plication over a catheterised urethra to outward traction during bladder neck plication and suspension of bladder neck bilaterally from the pubic bone to maintain anatomical urethrovascular angulation and urethrogenitoplasty. Epispadias with associated VUR cannot be corrected	6/6 patient achieved continence after 4 months follow-up 6 patient grade 3
Alyami, 2017 [14]	Single-stage perineal approach	Perform urethral tubularization and lengthening, and the excess lateral urethral plate, wrapped over the neourethra. Without ureter reimplantation. Anterior bladder wall and the bladder neck were tailored and sutured to achieve a funneling configuration.	4/7 patient achieved diurnal continence and no postoperative complication occurred.

capacity, so the BNR surgery was delayed at the age that the kids can accept the essential instruction of toilet training and the bladder has enough capacity for surgery [15,30]. Later, Gearhart proposed that the urethral and genital reconstruction be done at age 1.5–2 years old and bladder neck reconstruction at 4 to 5 years old [15]. Although this technique achieves good continence in 87.5%, this technique requires multiple sessions of surgery and is associated with higher morbidity [5]. Other BNR techniques such as vesicourethral suspension, transvaginal plication, and bladder flap procedures also developed for epispadias repair can be seen in Table 1.

The current use surgical method for isolated female epispadias is the single-stage reconstruction [5]. The urethral repair and bladder reconstruction were done in one operation [9]. The first single-stage reconstruction was suggested by Hendren. It combined the perineal and transvesical approach with ureteric reimplantation [29]. The technique requires extensive tissue dissection and has a long duration of surgery and has satisfactory results. Since then, there are multiple modifications have been developed in single-stage reconstruction, which can be seen in Table 1 [29]. The literature that did the comparison of some techniques can be seen in Table 2. The selection of a single-staged reconstruction method is based on surgeon preference and patient condition. The perineal approach procedure is not suggested to be performed on patient associated with VUR who need ureters reimplantation. The more severe cases need a more radical technique to achieve a better continence outcome. For this patient, we performed a single-stage reconstruction of epispadias repair and bladder neck reconstruction through a

Table 2

Comparison of technique for epispadias reconstructive procedure.

Authors	Technique	Sample	Outcome
Kramer and Kelalis, 1982 [23]	Single stages procedure of Young-Dees, Leadbetter and Transvaginal plication technique. The minimum age of surgery is 3 years old.	12 patient	5/6 patients achieved complete continence in the Leadbetter group 2/2 patients achieved complete continence in Young-Dees 3/4 patients achieved complete continence in the Transvaginal plication group
Cheikhelard, 2009 [31]	Comparison of Young-Dees technique and Manzoni Ransley technique	14 patients, Younger patients for Manzoni Ransley technique	Daytime continence rate similar in groups 1 and 2 Yong dees group: 87,5% 5/7 full continence, 1/7 day time continence, bladder capacity 175 mL, the complication of upper tract dilatation, ureterovesical reimplantation stenosis Manzoni Ransley group: 85%, full continence 3/7 patient, daytime continence 3/7 patient, bladder capacity 150 mL, no complication
Leclair, 2017 [24]	Comparison of perineal urethrocervicoplasty (PUCP) and Kelly repair. Patients with normal bladder capacity were treated with PUCP and patients with a small bladder and/or VUR were treated with Kelly repair.	16 patient	In the PUCP group: 4/7 patients achieved full continence, 1/7 patients achieved diurnal continence, and 3/5 continence patients need bladder-neck injection due to stress incontinence In the Kelly repair group: 3/11 patients achieved full continence, 5/11 patients achieved diurnal continence, 1/8 of continence patients had obstructive micturition
Alyami, 2017 [14]	Comparison of Young-Dees-Leadbetter (YDL) and Alyami's PUCP procedure	3 patients of YDL procedure and 7 patients of PUCP procedure	YDL group: all patient incontinent, PUCP group: 4/7 full continence, 2/7 patient achieved continence with bulking agent injection and there was no postoperative complication

perineal approach following the technique mentioned by Alyami F et al. The surgery is successful because the patient has achieved urinary continence, and there is no complications happen during and post-surgery.

4. Conclusion

Females epispadias is a rare congenital anomaly that is quite often to miss diagnosed. To diagnose the epispadias need a careful physical examination. Surgical management is the option for isolated female epispadias. The single-stage technique is the recommended surgical technique for isolated female epispadias. The goals of the surgery are to achieve urinary continence, urethra and genitalia anatomy, function, and cosmetic restoration. This patient has achieved urinary continence, and there is no complication that occur during and post-surgery.

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Ethical approval

Ethical Approval granted from Ethical Committee of Faculty of Medicine, Universitas Indonesia, protocol number 22-01-0118.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ervandy Rangganata: data collection, writing the paper.
Irfan Wahyudi: study concept, data interpretation.

Registration of research studies

Not applicable.

Guarantor

Irfan Wahyudi as the guarantor of this work of case report.

Declaration of competing interest

None.

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