Isolated Penopubic Continent Epispadias in Children. A Case Series.

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Abstract

Isolated male epispadias (IME) is a rare entity with incidence of approximately 1 in 120,000 live births. Continent epispadias is an extremely rare entity with an incidence of less than 10percent of all cases. It is characterized by failure of the urethral plate to tabularize on the dorsum of the penis with defect ranging from glandular to penopubic region.The greatest challenge in treating epispadias is creating adequate penile length, straightening of penis and a penopubic angle that would allow comfortable penile penetration. A total of three male patient aged 6, 8 and 9 years with continent isolated penopubic epispadias underwent surgical repair with Modified Cantwell-Ransley reconstruction procedure. Postoperatively, there was no fistula and urine stream through neomeatus was adequate and the shape of the penis was cosmetically acceptable.

Keywords

- Isolated male epispadias (ime)
- Modified cantwell-ransley repair
- Bladder exostrophy epispadias complex (beec)

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Introduction

Isolated male epispadias (IME) is a rare entity with incidence of approximately 1 in 120,000 live births.[1] Continent isolated epispadias is an extremely rare entity with an incidence of less than 10percent of all cases. It is characterized by failure of the urethral plate to tubularize on the dorsum of the penis with defect ranging from glandular to penopubic region. Epispadias usually presents with phimotic prepurtial orifice where glans is not visible and is known as concealed epispadias.[2] Like hypospadias, it covers wide spectrum in which the meatal orifice can be located anywhere from distal penile shaft to the pubic area. Unlike in hypospadias, the severity of the condition is related not only to the meatal position but also to the degree of incontinence associated with the meatal position, as the bladder neck might be involved in more proximal variants of isolated male epispadias (IME).[3-4] The greatest challenge in treating epispadias is creating adequate penile length, straightening of penis and a penopubic angle that would allow comfortable penile erection/ penetration. Another challenge is to provide adequate skin when straightening and lengthening the penis is done.[5] Many surgical techniques have been described for the repair of male epispadias, of which Modified Cantwell-Ransley repair has become the procedure of choice.[6]

Case presentation

A total of three male patient aged 6, 8 and 9 years presented with complaints of urinary meatal opening on the dorsal aspect at the base of the penis. It was present since birth with passage of clear urine, urinating forward with history of straining. 2 patients had mild stress urinary dribbling and spraying of the urine stream. There was no history of any discharge or any apparent bending (dorsal chordee) of the penis. The antenatal and birth history was also normal. General and systemic examination did not reveal any abnormality. Urological examination showed no mass in the flank, empty bladder, male external genitalia normal and the only abnormality seen was external urethral meatus found on the dorsum of the penis in the penopubic region without any dorsal chordee in any patient. There was no abnormal widening of the pubic symphisis in any of the patient. Both testicles were palpable with normal shape and size. Penile length was assessed preoperatively and 3 months after surgery by using Wilcoxon signed rank test. All

baseline blood investigations were found to be normal. Abdominal ultrasound (USG KUB) was normal. The patient was diagnosed with continent penopubic epispadias and planned for urethroscystoscopy. Urethrocystosscopy revealed normal sphincter, bladder neck and bladder. Surgical repair was done with Modified Cantwell-Ransley reconstruction procedure. All cases underwent repair under general anaesthesia by modified Cantwell-Ransley technique. The surgical procedure involves placing stay sutures separately on the two hemiglans 9Fr feeding tube was inserted as shown in **(Figure 1)**. Incision made around lateral edges of urethral plate and around epispadias meatus and mobilization done. Skin dissected and deglovingdone and glans wings created.**(Figure 2)** The urethral tubularization done by continuous suture over the 9Fr feeding tube using 5-0 round body vicryl suture.**(Figure 3)** Both the corpora were separated, mobilised and incisions were made dorsally on the corporal bodies at a point of maximum curvature that provide diamond shaped extensions of the incisions at the time of closure. Both corpora were closed with each other over the neo-urethra with continuous sutures and the diamond shaped extension of the incisions on the adjacent corpora is sutured to each other. **(Figure 3 and 4)** The glans wings were held together with interrupted suture, granuloplasty was done and urethral meatal opening made at the tip of the penis. **(Figure 5)** Skin closure with Z-plasty done. **(Figure 6)** Feeding tube was removed on day 12 postoperatively in all patients. Uroflowmetry was done 4 weeks after catheter removal. There was no fistula and urinary stream through neomeatus was adequate, thick and good and the shape of the penis was cosmetically acceptable. The patients were followed at 3rd and 6th months and 1 year for any thinning of the stream and need for meatal calibration and urethral dilatation. Urine output through the external meatus was adequate with thick and good stream and only one patient needed multiple sittings of meatal calibration up to 6 months postoperatively. Penile shape was cosmetically acceptable. The postoperative outcome is shown in **(Table 1)**.

Figure 1: Placing stay sutures and feeding tube

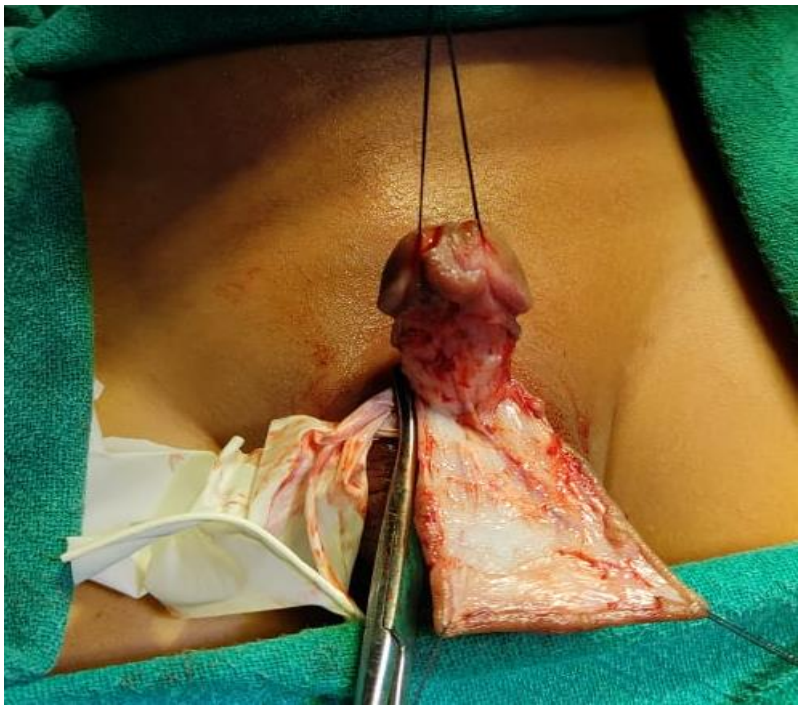

Figure 2: Degloving

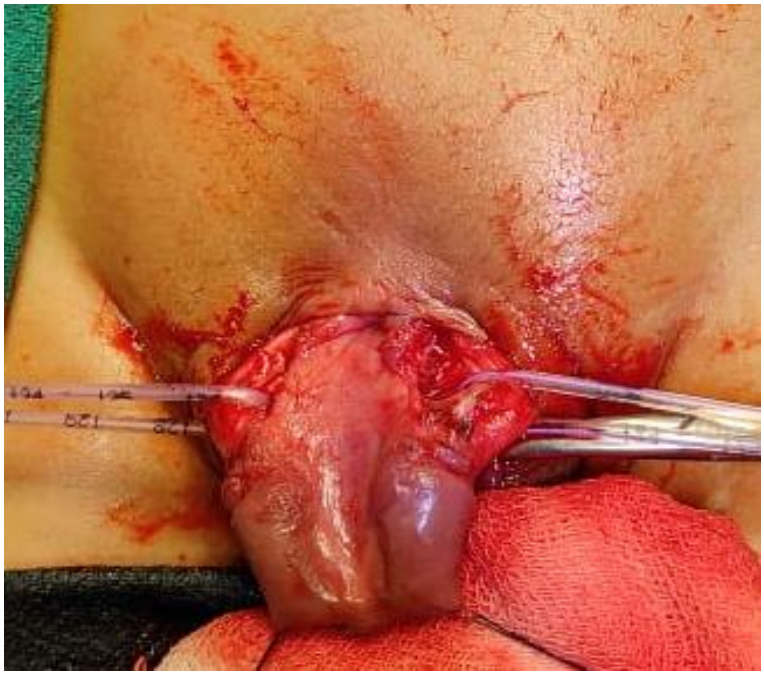

Figure 3: mobilization of urethral plate

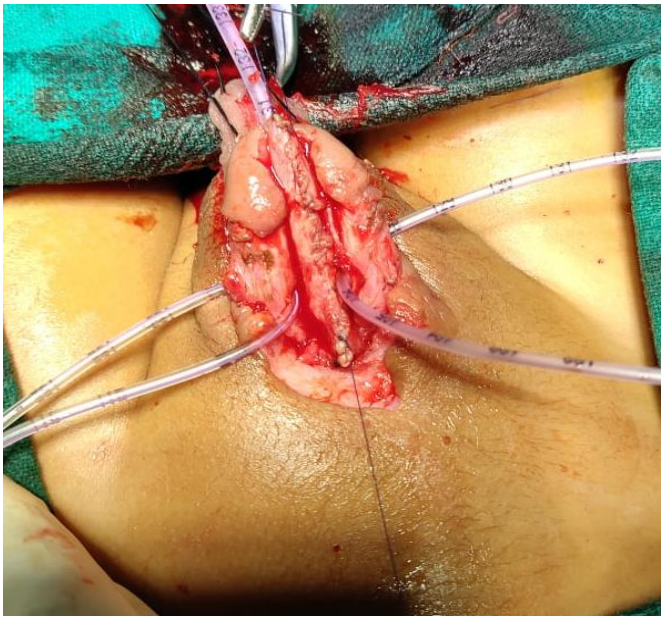

Figure 4: Tubularisation of urethra over feeding tube.

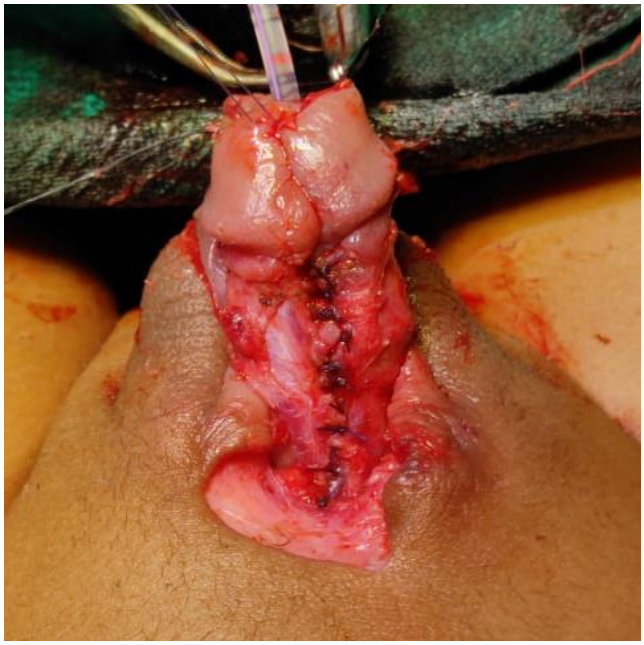

Figure 5: Corporisation over urethra

Figure 6: Skin coverage with Z plasty.

Table-1: Postoperative subjective and objective outcome.

Outcome Parameters	Case -1	Case-2	Case -3
Patient / Parent Satisfaction	Excellent	Excellent	Good
Penile Size Preoperative Postoperative	2.2cm 4.3cm	1.9cm 3.6cm	2.3cm 3.5cm
Glans Shape	Conical	Conical	Conical
Urethrocutaneous fistula (UCF)	None	None	None
Urinary Continence	Continent	Continent	Continent
Urinary stream	Thick and good	Thick and good	Satisfactory
Appearance (cosmesis)	Excellent	Excellent	Excellent

Discussion

Epispadias represents the one end of spectrum of Bladder extrophy-epispadias complex (BEEC). The combined incidence of BEEC spectrum is 1 in 10,000 births approximately while the isolated male epispadias is present in 10 percent cases of BEEC. Most male epispadias (about 70%) have complete epispadias with urinary incontinence and only 30% are continent.[7] In our series all patients were continent. Several surgical procedures have been described for the repair of either complete epispadias or bladder extrophy. The Cantwell-Ransley technique is the most popular and widely used procedure for epispadias repair.[8] Urethrocutaneuos fistulas and urethral strictures are the most common complications following modified Cantwell-Ransley procedure. Most patients with penopubic epispadias (upto 70%), the bones of the pelvis are widely separated which affects the bladder neck and external sphincter leading to sphincter incompetence and constant or stress urinary dribbling. There was no pubic

diastasis in the present series and none of the patient had urinary incontinence.

There are several procedures for epispadias repair but mainly two procedures are practised, the Modified Cantwell Ransley and Mitchell Bagali repair. Modified Cantwell involves partial disassembly of penis, while as Mitchell technique involves complete penile disassembly.[9] The choice of the procedure depends upon the surgeon's choice and expertise. Mitchell repair is preferred in bladder exostrophy epispadias complex (BEEC) and incontinent epispadias.[10] The most common complication following epispadias repair is urethrocutaneous fistula, persistence of chordee, difficult urethral catheterization and penile dysfunction. In our series all patients had mild or no chordee, and there was no urethrocutaneous fistula formation in any patient and no other complication. The main objective of the repair is functional and cosmetic appearance of the penis. As far as the functionality is concerned, the urinary stream should be good, thick and single. The cosmetic appearance involves a good penile length, conical glans and urinary meatus at tip. The cosmetic appearance however is subjective and may differ between surgeon and parents/patient. In our series the functionality and cosmesis was good and satisfactory.

Conclusion

Isolated continent penopubic epispadias although rare but is not uncommon. Modified Cantwell-Ransley technique is best suited for such patients and found to have satisfactory surgical and cosmetic outcome.

Ethical Consideration

Informed consent was obtained from all individual participants included in the study.

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Conflict of interests

There is no conflict of interest

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