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Isolated female epispadias: A rare case report and surgical outcome

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ABSTRACT

Isolated female epispadias without bladder exstrophy is a rare anomaly. We report a six-year-old girl who presented with urinary incontinence since birth and recurrent urinary tract infections. Examination revealed a bifid clitoris and a wide urethral opening. She underwent single-stage surgical repair via a perineal approach. Postoperatively, she achieved urinary continence with a good stream and could retain urine for 90–120 minutes. Female epispadias is often misdiagnosed as refractory enuresis or recurrent urinary tract infections. Early recognition and timely surgical correction significantly improve continence and quality of life.

1. Introduction

Isolated female epispadias without bladder exstrophy is a rare anomaly with an incidence of approximately 1 in 4,80,000 live births.[1] It usually presents with incomplete bladder incontinence and small-for-age bladder capacity. Due to the straight course of the ureters opening laterally, vesicoureteral reflux is seen in 30-75% of patients. Achieving continence with functionally and cosmetically acceptable genitalia is the main goal of management in these patients.[2,3]

This case report was prepared following the CARE guidelines [https://www.care-statement.org].

2. Case presentation

A six-year-old female child presented with urinary incontinence since birth associated with recurrent urinary tract infections. On examination, the child had a bifid clitoris with a wide-mouthed urethral opening [Fig. 1]. Vagina and anal opening were normal. Ultrasonography showed mild pelvicalyceal fullness. On a micturating cystourethrogram, grade II vesicoureteral reflux was seen on the left side with a bladder capacity of 20 cc. Magnetic resonance imaging of the pelvis was performed to delineate the anatomy [Fig. 2]. Renal scintigraphy showed normal bilateral renal function.

2.1. Operative technique and outcome

An inverted Y-shaped incision was made starting from the pubis and incorporating the urethral mucosa between the limbs of the Y [Fig. 3]. The incision was deepened to reach the pubic bone. The urethral mucosa, along with the spongiosum, was freed from surrounding tissue. Dissection was continued in the retropubic area to reach the bladder. The inter-symphyseal band of the pubis was cut in the midline to expose the anteroinferior aspect of the bladder and bladder neck. Stay sutures were placed at the apex of the planned cranial end of the bladder neck repair. The urethra was cut ventrally in the midline up to the bladder neck. Excess urethral mucosa was removed, and urethroplasty was performed. The inferior aspect of the bladder was excised to create a funnel-shaped bladder neck. The mucosa and bladder muscles were approximated in layers. The neo-urethra was cannulated with an infant feeding tube number 8, and the labia minora were reconstructed. The pubic incision was closed in layers with a supra-pubic cystostomy catheter in situ [Fig. 4].

2.2. Postoperative course

After intermittent clamping, the catheters were removed on postoperative day 18. At discharge, the patient was passing urine in a stream, was continent, and could hold urine for 90–120 minutes. At six months follow-up, the patient had stress incontinence which was managed

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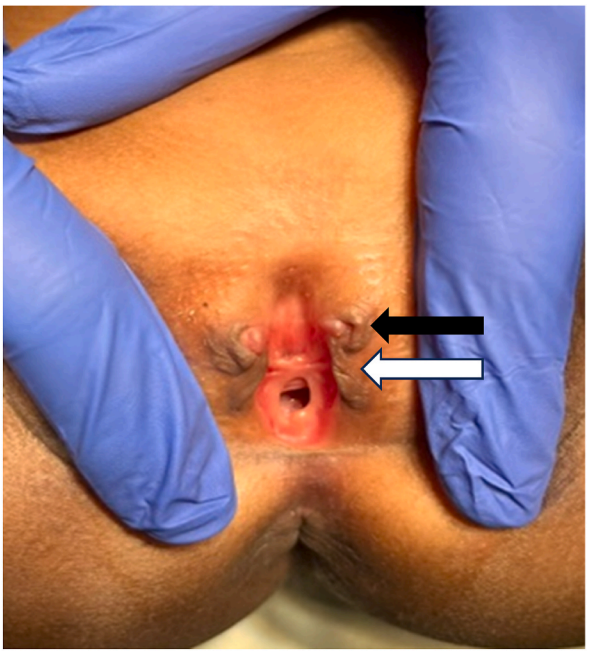

Fig. 1. Clinical image showing a bifid clitoris(black arrow), ill-developed labia majora and minora (white arrow) with a patulous urethral opening.

conservatively with physiotherapy, Kegel's exercises and faradic stimulation.

3. Discussion

Urinary anomalies are the third most common congenital anomalies.[4] Female epispadias patients present with continuous dribbling of urine. They are usually unrecognised as surgical anomalies and are mis-treated for chronic bedwetting and urinary tract infections. A history of incontinence that is resistant to medical management is present. General physical examination provides characteristic findings like a bifid clitoris, depressed mons pubis covered by thin hairless skin, and anterior separation of labia majora and minora. Labia minora are ill-defined. Diastasis of the pubic symphysis and an abnormal oval-shaped external urethral meatus are present. They also have low bladder leak pressure on cystourethrography.[5]

The aims of management include achieving continence, preservation of the upper tract and reconstruction of functional and cosmetically acceptable genitalia· Young first described the need for radical revision of the urethra and bladder neck to achieve continence. For the widened bladder neck, Dees proposed the cranial extension of the urethra and narrowing the bladder neck.[6] Leadbetter emphasised the importance of ureteral re-implantation to attain space to extend the posterior urethra cephalad.[7] Other proposed procedures are transvaginal urethral and bladder neck plication, urethral reefing, muscle transplantations, uterus interposition, and Marshall-Marchetti vesicourethral suspension. All these methods partially correct incontinence, but not the anatomy of the external genitalia and the lower urinary tract.

Staged procedures for epispadias consist of urethral and vulva reconstruction at stage 1 at 1-1.5 years, and then followed by bladder neck reconstruction at 4-5 years. This delay of reconstruction was proposed to allow the bladder to adapt and increase in size post-urethral and genital reconstruction, and wait for the child to attain the age of toilet training. These procedures usually have suboptimal results pertaining to urinary continence and are associated with higher morbidity. Hendren performed single stage repair combining abdominal-perineal reconstruction. This is the most commonly performed procedure and is also associated with good continence results.[6]

A small bladder capacity need not necessarily mean a lesser chance of achieving continence without augmentation. A better quality of life can be provided with timely, precise surgical correction and with lesser morbidity in a single-staged repair. We achieved similar results in our patient with the single-stage repair of epispadias and a functional and cosmetically acceptable outcome.

4. Conclusion

Female epispadias is a rare entity. Single-stage/staged surgical repairs provide functional and cosmetically accepted outcomes, improving the quality of life. Recognition of the milder clinical forms is essential to avoid mis-treatment as medical treatment-resistant enuresis and urinary tract infections.

CRediT authorship contribution statement

Sushma Nakka: Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Formal analysis, Data curation. **Aditi Dalvi:** Writing – review & editing, Visualization, Supervision. **Maitreyee Save:** Writing – review & editing, Visualization. **Dravina Shetty:** Writing – review & editing, Validation. **Shahaji Deshmukh:** Writing – review & editing, Validation, Supervision. **Abhaya Gupta:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Project administration, Conceptualization. **Paras Kothari:** Validation, Supervision.

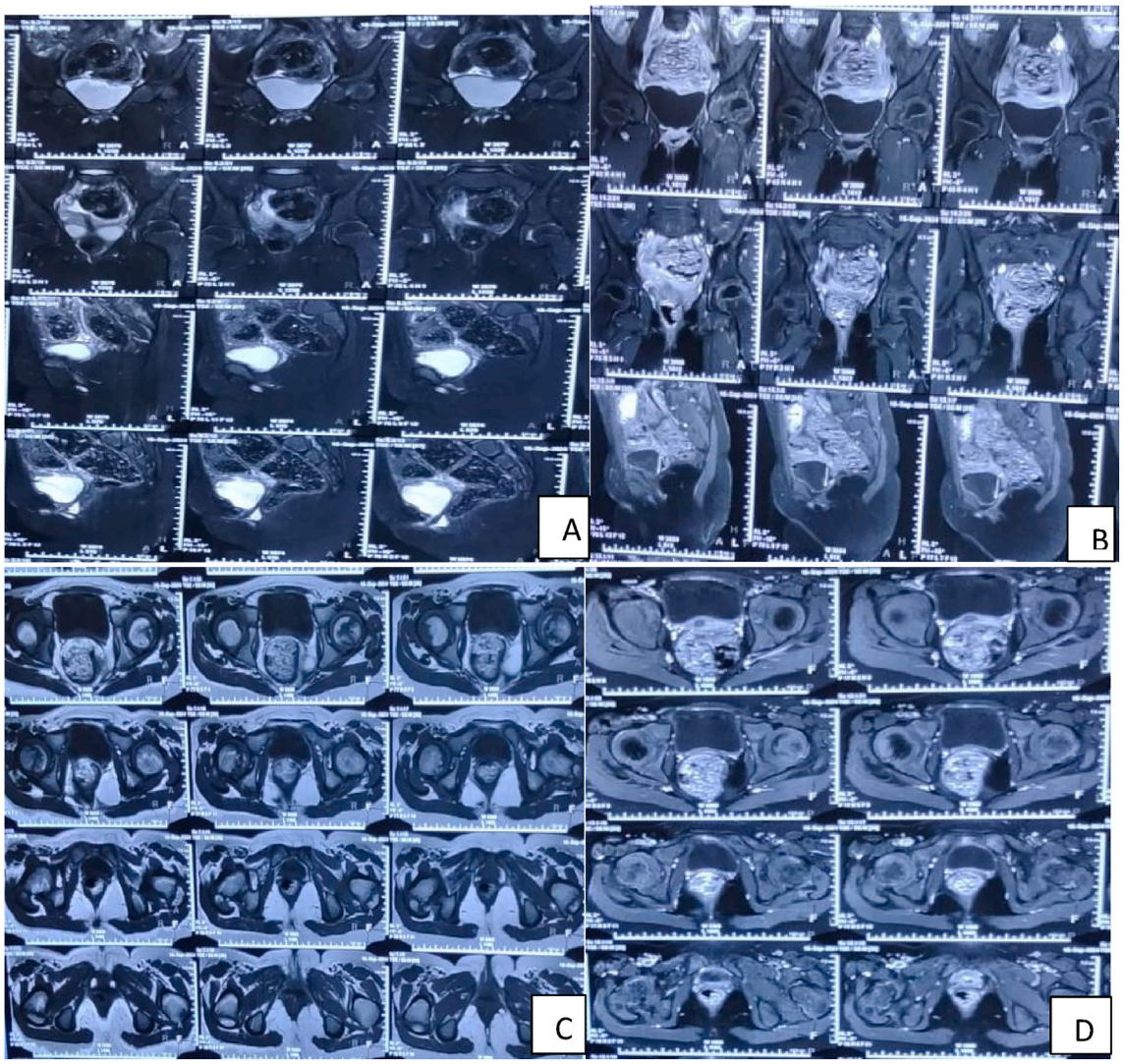

Fig. 2. Multi-planar MRI of the pelvis. (A, B) Coronal and Sagittal sections demonstrate the anatomical relationship between the low-lying bladder and the symphysis pubis, highlighting a short, patulous urethra and a wide bladder neck. (C, D) Axial sections clearly delineate the characteristic diastasis of the pubic symphysis and the lateral divergence of the pubic bones. No associated Mullerian duct anomalies or significant upper urinary tract abnormalities are visualised, confirming the diagnosis of isolated female epispadias.

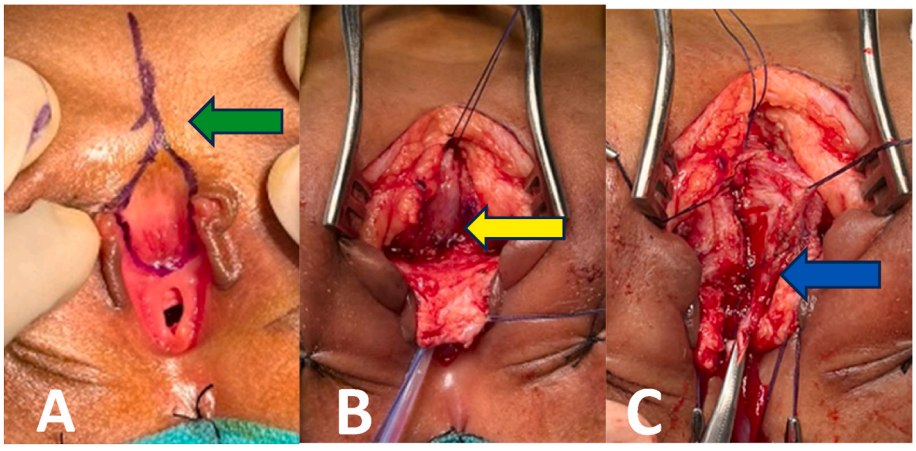

Fig. 3. Intra-operative pictures (A) showing the marked incision (green arrow), (B) showing the exposed bladder neck (yellow arrow), and (C) showing the cut-opened urethra (blue arrow).

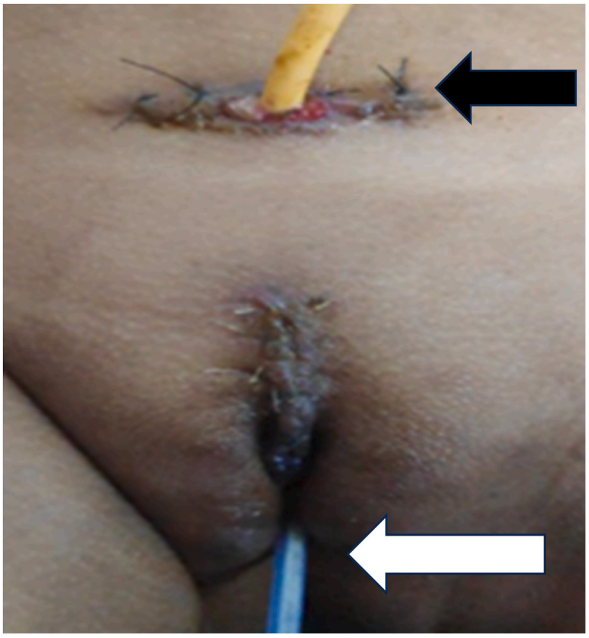

Fig. 4. Post-operative picture taken on postoperative day 12 with an indwelling supra-pubic Foley catheter (black arrow) and per urethral infant feeding tube (white arrow).

Informed consent

Informed consent for publication of this case report and accompanying images was obtained from the patient's parents.

Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

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