Case Report

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Experience with isolated male epispadias repair: case report from Northern Tanzania

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Introduction: Isolated male epispadias is a rare congenital condition where the urethral opening is abnormally on the upper penis side. Its severity varies, affecting continence and often requiring surgery. Outcomes depend on severity, voiding problems, and the surgeon's expertise.

Case presentation: Two boys, aged 7 and 9, each presented with abnormal urethral opening on the dorsum of the penis, with abnormal urinary stream (bent upwards), without a history of prior surgeries, which was consistent with isolated epispadias. Clinical evaluations revealed a subcoronal meatus on the dorsum of the penis for the first case, and the coronal meatus for the second. The preoperative workups were normal, and there was no pubic symphysis diastasis for either case, and both were continent. The surgical repair involved urethral plate tubulization, neourethra ventralization, glanuloplasty, and circumcision using the modified Cantwell–Ransley technique. Postoperative outcome was successful, with good functional and cosmetic results.

Clinical discussion: About 90% of congenital anomalies occur in low- and middle-income countries, including Tanzania, where urethral anomalies affect 24% of children and 0.5% present with epispadias. Diagnosis relies on clinical evaluation, while management involves surgical correction, most effectively using the modified Cantwell–Ranseley technique, which preserves urethral blood supply and achieves excellent functional outcomes.

Conclusion: Early diagnosis and surgical repair of isolated male epispadias, even in resource-limited settings, can result in excellent functional and cosmetic outcomes using standard reconstructive techniques.

Keywords: Cantwell–Ransley technique, case report, congenital anomaly, epispadias, urethroplasty

Introduction

Isolated male epispadias is a rare congenital anomaly, characterized by the urethral opening located on the upper aspect of the penis. This condition occurs when the urethra fails to close properly, resulting in a flat and exposed inner lining[1]. This spectrum of disease varies from the most severe, bladder exstrophy epispadias complex, to simpler isolated glanular epispadias. The abdominal wall is entirely normal, and the symphysis is closed or slightly separated in isolated epispadias[1].

Isolated epispadias is more common in males than in females, with an incidence of 1 in 117 000 in men[1]. It is divided into three types based on the position of the urethral meatus: penopubic, penile, and glanular, with the first being the most prevalent. Most of these cases tend to be overlooked at birth due to their minimal abnormalities[2], together with a weak neonatal screening strategy, as in many developing countries, which contributes to their late presentations[3].

The primary aims of treatment are restoring urinary control and creating a straight, cosmetically acceptable, functional penis with sufficient length[1,2,4]. Key factors influencing postoperative continence and appearance include initial anatomical abnormalities, the extent of voiding dysfunction, the surgeon's experience and skill, and the need for reoperation[5-7].

Isolated epispadias is a rare condition affecting both sexes and often needs timely intervention. We present two cases of isolated male epispadias, detailing their presentation, investigations, and treatments, while illustrating the modified Cantwell–Ransley technique, which is commonly used for managing these cases, in accordance with the SCARE guidelines for case reporting[5].

HIGHLIGHTS

- A case report of isolated male epispadias.
- Examination revealed subcoronal epispadias in the first case and glanular epispadias in the second.
- In both cases, there was no significant chordee; however, the dorsal skin was deficient in the first case.
- Defects were corrected using a modified Cantwell–Ransley technique.
- Upon follow-up, they were able to void with a forward stream and exhibited an acceptable cosmetic appearance.

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Case presentation

First case

A 9-year-old boy presented with a congenital abnormality of the urethral opening, located on the upper part of the penis, which caused a spraying of urine and wetting himself during micturition. He was born via spontaneous vaginal delivery at full term, without any complications.

According to his mother, she attended all antenatal visits and took the recommended supplements. The boy received all vaccinations as scheduled and has no previous history of surgery or chronic illnesses. He is the firstborn in a family of four, with no history of a similar condition.

He was fully conscious with normal vital signs. His abdomen was normal, and there were no anterior wall defects. He had an uncircumcised penis with ventral redundant prepuce (Fig. 1A) and a urethral defect on the dorsum of the glans penis. The meatus was located below the corona, with a broad, healthy-quality urethral plate (Fig. 1B). Both testes were palpable, in their normal position, with healthy skin.

The baseline test showed a hemoglobin level of 11.9 g/dL, a platelet count of 273 × 10^9/L, a serum creatinine of 34 µmol/L, blood group A Rh positive, no abnormalities on urinalysis, and pelvic radiography was normal (Fig. 3A). The patient was scheduled and prepared for epispadias repair.

Intraoperatively, a "U" incision was made around the urethral plate toward the glans (Fig. 1C). The penis was degloved, and tubulization of the urethral plate was performed over a 12-Fr Foley catheter to create the neourethra (Fig. 1D). The corpora cavernosa separated from the midline, not completely, and then approximated at the midline at the glans using Monocryl 6/0, while the neourethra was ventralized.

Glanuloplasty and meatoplasty were performed to centralize the neourethra and relocate the meatus to the tip of the penis in a conical glans. The penile skin was then reconstructed, and circumcision was carried out using Monocryl 6/0 (Fig. 1E). A non-adhesive Vaseline gauze was placed around the wound for compression during dressing.

Postoperatively, the wound was examined after five days; it was clean and dry, with intact sutures and minimal tenderness (Fig. 1F). The urethral catheter was removed on the seventh day, and the boy was able to void with a strong, forward stream. At follow-up, the patient was doing well, remained continent, and had a satisfactory result.

Second case

A 7-year-old boy presented with a congenital abnormal opening on the dorsum of the glans penis. This was observed during the physical examination before his routine circumcision. It is associated with an upwardly deflected and spraying urine stream; however, there was no urine leakage.

The prenatal period was uneventful, and he was born at full term via spontaneous vaginal delivery. He was vaccinated according to the required schedule and has had an unremarkable postnatal and early childhood history. There was no history of previous surgery.

Upon examination, he was fully conscious with normal vital signs. The abdomen was of normal contour, and there were no defects on the anterior wall. He had an uncircumcised penis with a retractable prepuce, a urethral defect on the dorsum of the glans penis, and the meatus at the level of the corona (Fig. 2A). Both testes were palpable in their respective hemiscrotum.

Baseline tests revealed a haemoglobin level of 13.0 g/dL, a platelet count of 252 x 10^9/L, serum creatinine of 41 µmol/L, blood group A, Rh positive, with no bacteria isolated from the urine culture. Routine pelvic radiography was normal (Fig. 3A), and an echocardiogram showed an ejection fraction

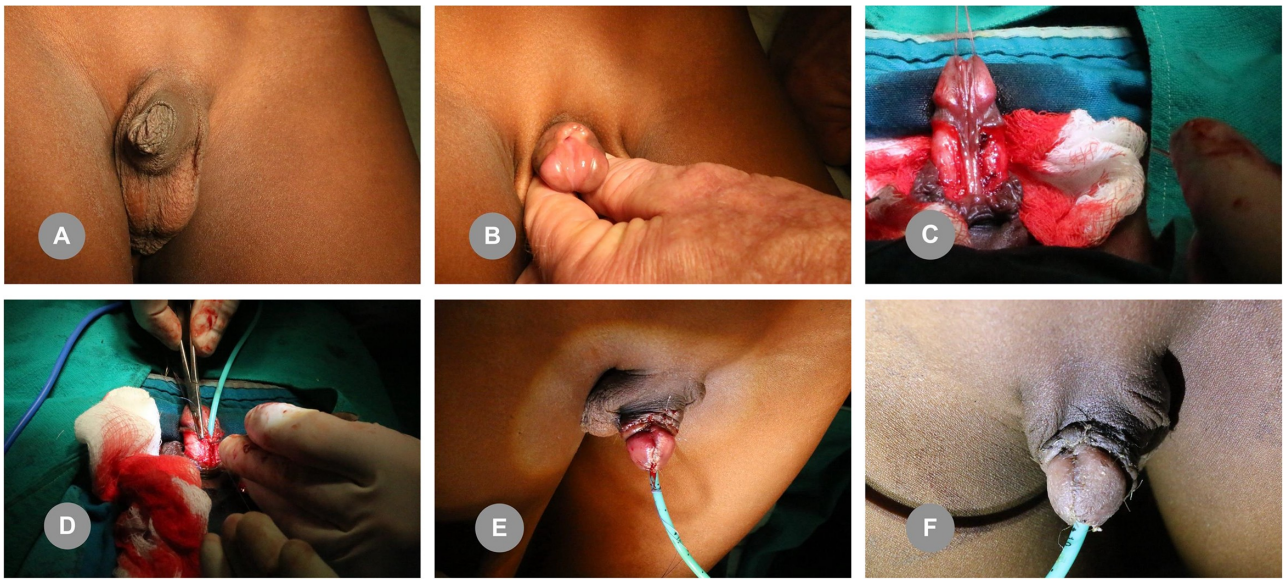


Figure 1. Depicts the peripubertal external genitalia of a boy with epispadias, featuring an uncircumcised penis with redundant prepuce, particularly ventrally (A), a retracted prepuce with a defect on the dorsum of the penis, where the urethral meatus is located at a subcoronal level, comprising a wide and fairly well-formed urethral plate (B). This is followed by a "U" incision to trim the urethral plate and degloving of the penile skin (C), then tubularization of the plate over a 12-Fr Foley catheter (D), completion of circumcision (E), and a clean, dry wound after 5 days (F).

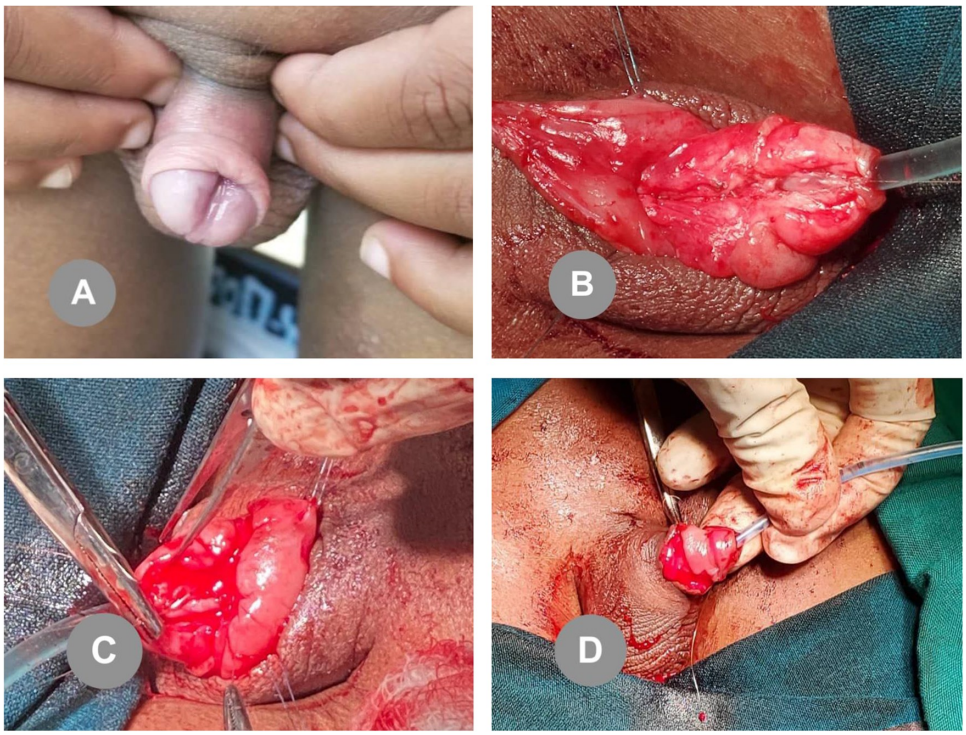


Figure 2. Illustrates the external genitalia and operative reconstruction. The uncircumcised penis shows a visible defect on the dorsum of the glans and the meatus at the corona, noted following preputial reduction (A). The ventralized neourethra is created by approximating the corpora cavernosa at the midline, accompanied by a conical reconstructed glans (B). Circumcision is performed to restore penile coverage (C), and a 10Fr feeding tube is inserted transurethrally for stenting (D).

of 62%. The patient was scheduled and prepared for epispadias repair.

Intraoperatively, a "U" incision was made around the urethral plate, deepening toward the glans and creating granular wings on both sides. The penis was degloved, and tubulization of the urethral plate was performed over a 12-Fr feeding tube to create the neourethra.

The corpora cavernosa were separated from the midline, with the neurovascular bundle preserved, and then brought together toward the midline at the glans using Monocryl 6/0. Simultaneously, the neourethra was ventralized (Fig. 2B). Glanuloplasty and meatoplasty were completed, after which the penile skin was restored.

Circumcision was carried out using Monocryl 6/0 (Fig. 2C). A 10-Fr feeding tube was inserted transurethrally to replace the previous 12-Fr feeding tube, thus stenting the repair (Fig. 2D). Vaseline gauze was wrapped around the wound, and a compressive dressing was applied.

Postoperatively, reconstruction was complicated by penile edema, which gradually resolved. By day 5, the wound was clean and dry, and the urethral stent was removed on day 7. The boy voided well with a strong stream and no leakage. At 6 months, he

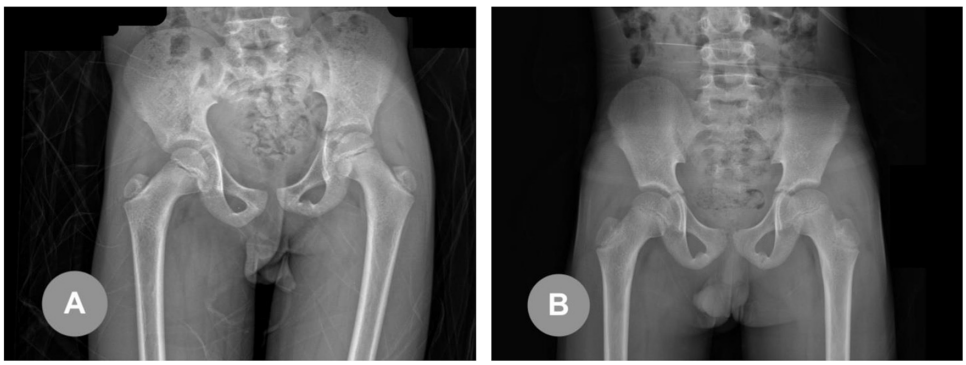


Figure 3. Illustrates the plain pelvic radiography of a 7-year-old boy (case 1, A) and a 9-year-old boy (case 2, B), both exhibiting normal pelvic bones.

continued to void well, remained continent, and had a satisfactory cosmetic outcome.

Discussion

Approximately 90% of congenital abnormalities occur in low- and middle-income countries, likely due to high fertility, low pregnancy termination, nutritional deficiencies, infections, and teratogens[6,8]. In Tanzania, efforts to improve early neonatal screening continue, focusing on detecting urological anomalies and other conditions[3]. Kahuruta *et al* reported a 24% prevalence of urethral anomalies among Tanzanian children, including 0.5% with epispadias[9].

Diagnosis of isolated epispadias in boys mainly depends on history and physical examination; imaging, cystoscopy, and urodynamics are used when associated abnormalities are present[4,7,10]. Pelvic radiography is performed to assess for pubic symphysis diastasis, which is more common in the bladder–exstrophy–epispadias complex than in isolated epispadias.

The boys with isolated epispadias have a short, pendulous penis, along with a variable degree of dorsa chordee and extragenital abnormalities, such as vesicoureteric reflux and impaired continence mechanisms seen in some cases[1,11]. The urinary incontinence is due to the sphincter involvement as seen in complete epispadias, whereas distal epispadias are typically continent, as the sphincter mechanisms remain intact[1,12].

The defect requires surgical correction. Numerous techniques have been described in the existing literature, but the Mitchell-Bagli (1996) and modified Cantwell–Ransley repair (1986) methods are widely employed for repairing these defects[1,13–16]. The main distinction between these techniques is the complete disassembly of the penis used in the former method[16,17].

According to Bhat *et al*, there is preservation of dual blood supply to the urethra and hence reduced ischemia with the partial penile disassembly during the modified Cantwell–Ransley technique, and this is the main advantage over other techniques[18]. Moreover, Nerli *et al*, in their follow-up study, reported an acceptable anatomical and functional outcome with this

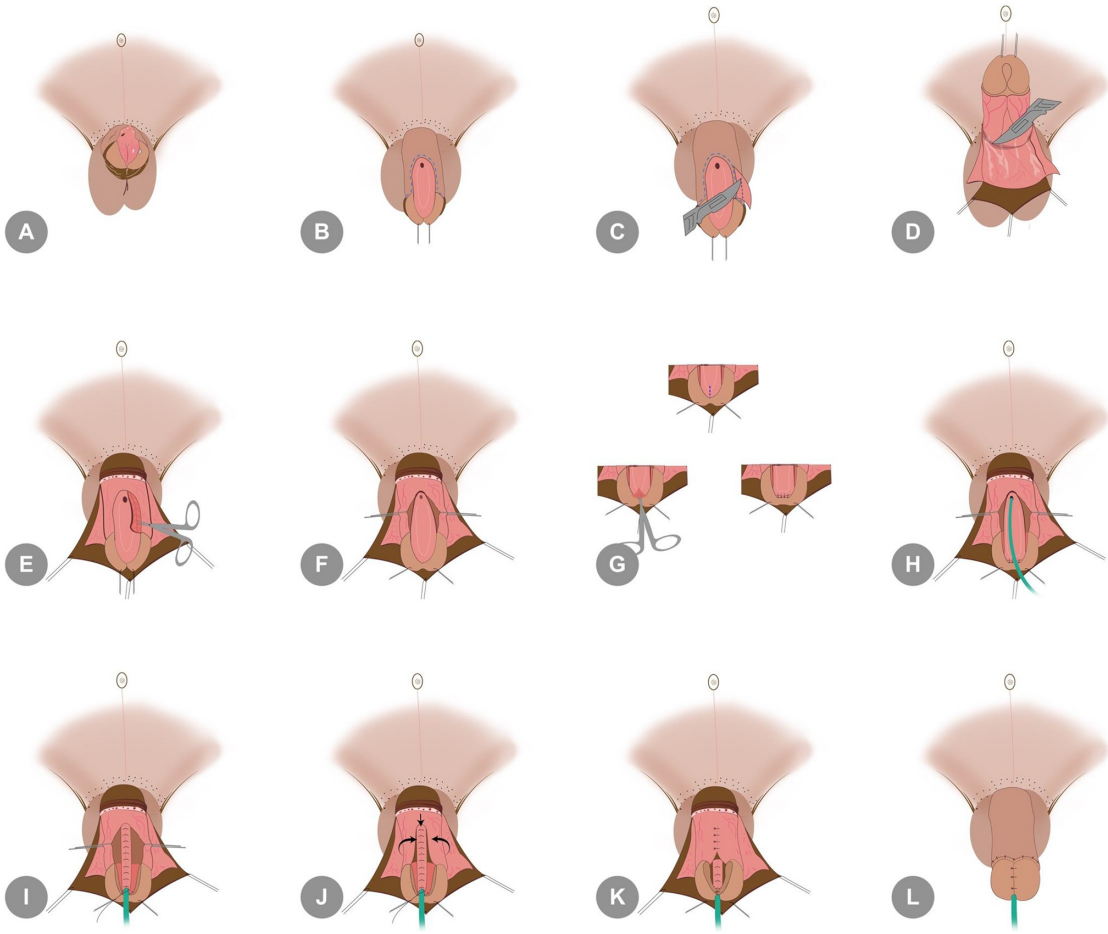


Figure 4. Demonstrates the surgical steps as performed in the present cases, similar to a modified Cantwell–Ransley repair. The meatal opening is located dorsally on the penile shaft (A), indicating where to make the "U" incision to mobilize the urethra (B). The U incision is made (C), followed by a circumferential subcoronal incision to deglove the penis (D). The urethral plate is dissected away from the corpora (E). The mesentery of the urethral plate is released to free the plate at the midpoint of the penile shaft (F). A deep vertical incision is made in the distal urethral plate, then closed transversely (G). A 12-Fr Foley catheter is inserted (H). Tubularization is performed using a running subcuticular suture (I). The neourethra is mobilized ventrally, and the corpora cavernosa are rotated internally (J). The corpora cavernosa are approximated using absorbable suture (K). Skin closure is completed with a circumcision (L).

technique, at a success rate of 90% and minimal complications at least with experienced surgeons on long-term follow-up[19]. Our cases were limited by a short-term period of follow-up.

This report involves two boys with isolated epispadias, conditions that are often diagnosed late in developing countries, typically during circumcision, with some patients living with the condition for a long period[20,21]. In ideal circumstances, repairs can be carried out a few weeks after birth. Bar-Yosef *et al* observed that these cases are operable after 6 months with positive outcomes[15,22].

The treatment involves corrective surgery using the modified Cantwell–Ransley repair, as described by Bar-Yosef *et al*[15]. John Phillip Gearhart's modification detached the urethral plate from the corpora except the distal part[23], to ensure a deeper urethra beneath the corpora at the glans level, enhancing both cosmetic and functional outcomes[15,16,24,25]. The surgical steps are as follows:

Dissection of the urethral plate

A "U" incision is made at the urethral plate (Fig. 4B,C), followed by a circumferential incision beneath the corona and complete degloving of the penile skin (Fig. 4D). The urethral plate is carefully dissected away from the corpora, leaving a small distal section intact (Fig. 4E). This dissection helps in mobilizing the urethral plate.

Mobilization of the dorsal urethral plate

The mesentery of the urethral plate is accessed from a ventral approach at the midpoint of the shaft (Fig. 4F) and separated from the corporal bodies. The dissection of the urethral plate continued up to the level of the epispadic meatus and distally to the tip of the glans penis, where the glanular wings are dissected (Fig. 4H).

Tubularization of the urethral plate

The glanular urethra and meatus are formed by reversing the meatal advancement and glanuloplasty maneuver (Fig. 4G), involving a deep vertical incision in the distal urethral plate and transverse closure with absorbable sutures. The urethra is tubularized over a 12-Fr stent with a running absorbable suture (Fig. 4H,I). Redundant glans skin is excised from the glanular wings, which are closed with interrupted absorbable sutures.

Repositioning of the neourethra and endorotation of the corpora cavernosa

The reconstructed urethra is then mobilized ventrally beneath the internally rotated corpora cavernosa (Fig. 4J). The corpora are approximated with absorbable sutures, without cavernocavernosal anastomosis (Fig. 4K).

Skin Closure and Circumcision

Skin coverage is secured with interrupted absorbable sutures, excess skin trimmed, and circumcision completed (Fig. 4L). The urethral stent is left for 10 days with prophylactic antibiotics during this period[26]. Follow-up is set at 2 weeks post-stent removal, then three months and annually[27,28].

Conclusion

Isolated male epispadias, although rare and affecting appearance, can be effectively treated with prompt surgery. The two cases demonstrated successful outcomes using the modified Cantwell–Ransley repair, emphasizing early diagnosis and intervention. Positive results are possible even in resource-limited settings, underscoring the importance of awareness and proper management to enhance quality of life.

Ethical approval

The hospital's ethics review panel determined that this case report did not require ethical approval, as it reports cases that occurred during routine surgical practice.

Consent

Written informed consent was obtained from the patients' parents for publication and any accompanying images. A copy of the consent is available for the Editor-in-Chief of this journal to review upon request.

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Author contributions

D.D.L.: Conceptualization, Writing – original draft, Writing – review & editing, Visualization, Resources, Software. B.N.N.: Conceptualization, Resources, Project administration. J.B, D. M., E.J.A.: Conceptualization, Writing – review & editing, Resources. O.J.M.: Writing – review & editing, Supervision, Resources, Conceptualization.

Conflicts of interest disclosure

The authors disclose no relevant financial or nonfinancial interests.

Guarantor

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Registration unique identifying number (UIN)

Name of the registry: Unique identifying number or registration ID: Hyperlink to your specific registration (must be publicly accessible and will be checked).

Data availability statement

No data were produced from this study.

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