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The so-called Y-type urethral duplication: anatomical insights through controversial terminology

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Abstract

Background Y-type urethral duplication is a term frequently used to describe a rare condition in the male associated with double urinary stream: a weak interrupted urinary stream through a hypoplastic penile urethra, while the main urine flow is through the anus/perineum via accessory posterior channel. Fortunately, the advent of MRI has provided a powerful tool to study these anomalies in depth and on multiple planes.

The study included boys presenting with abnormal micturition through the anus beside weak interrupted urine stream through the penis. Investigations included pelviabdominal ultrasound to screen for possible associated upper urinary tract anomalies, conventional contrast X-ray studies, and pelvic MRI.

Results During the period between 2016 through 2021, the study included two newly diagnosed cases with abnormal communication between the lower urinary tract and the anus (the so-called Y-type urethral duplication), in addition to one previously reported case whose preoperative imaging studies (including pelvic MRI) were available for analysis. Different surgical solutions have been applied for each case. Hypoplasia was not only restricted to the penile urethra but also affected the corporeal bodies of the penis with variable degrees. In two cases, only a single corporeal body could be identified. In another case, the three corporeal bodies were present but appeared distorted (irregularities and interruption of corporeal bodies).

Conclusion In our present study, pelvic MRI examination clearly unveiled significant degrees of corporeal dysgenesis among cases diagnosed as Y-type urethral duplication that would strongly suggest these cases to belong to the same disease spectrum of penile dysgenesis/agenesis.

Keywords Penile dysgenesis, Anorectal malformations, Aphallia, Ambiguous genitalia

Background

Y-type (or ℶ-type) urethral duplication is a term frequently used to describe a rare condition in the male associated with double urinary stream [1–3]: a weak interrupted urinary stream through a hypoplastic penile urethra, while the main urine flow is through the anus/perineum via accessory posterior channel. Occasionally, some cases may present with passage of urine through the anus during otherwise normal micturition through functioning penile urethra [2, 4]. Other reports described this condition as congenital "H-type" urethro-anal fistula analogous to the H-type trachea-esophageal fistula at the other (proximal) end of the gastrointestinal tract [5, 6]. In a previous report, we have brought several observations that support the diagnosis of this abnormal uro-anal communication as a "fistula" rather than a "true" urethral duplication [7].

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Apart from controversial terminology, this condition represents one of the most challenging reconstructive surgeries of the lower urinary tract [2, 3, 8, 9]. Several reports have highlighted the need for multiple procedures, high risk of stenosis, and failure even with 2-stage strategy [10, 11]. Many patients would end with supra-vesical channel (Mitrofanoff catheterizable port) to help to evacuate the bladder and protect the upper urinary tract [2, 3, 10]. The reported less satisfactory surgical outcomes would call for better understanding of the anomaly and associated deviation from the norm, which can be difficult to perceive with surgical practice alone. The advent of cross-sectional imaging (MRI) has provided a powerful tool to study these anomalies in depth and on multiple planes.

Based on previous experience gained from studying pelvic MRI anatomy of other congenital disorders [12], we present this study aiming to uncover some characteristic features of this rare anomaly. This may help surgeons make correct planning before embarking on complex reconstructive procedures [11].

Methods

The study included boys presenting with abnormal micturition through the anus beside weak interrupted urine stream through the penis. These boys were diagnosed as having a congenital abnormal communication between the lower urinary tract and the anus, the so-called Y-type urethral duplication [1–3]. Investigations included pelviabdominal ultrasound to screen for possible associated upper urinary tract anomalies, conventional contrast X-ray studies (micturating cystourethrogram), and pelvic MRI. For cases presenting with signs of obstruction (distended bladder, bilateral hydroureteronephrosis), early supra-vesical diversion via a temporary suprapubic cystostomy was established [3]. Later, conversion to permanent Mitrofanoff self-catheterizable port was considered a useful adjunctive procedure to ensure adequate bladder drainage and protect the upper tract during attempts of reconstruction of the lower urinary tract, as the latter is liable for strictures and reoperations [2, 3, 10, 11].

Micturating cystourethrogram (Fig. 1)

This is preferably performed through a suprapubic access [3]. The penile urethra is often too narrow/irregular for performing the study, while the accessory posterior uro-anal channel may be difficult to cannulize except under anesthesia. Moreover, a catheter introduced through the accessory posterior channel usually fails to enter the urinary bladder.

MRI technique

Non-contrast pelvic MRI examination was performed using 1.5 T magnet (Achieva, Philips medical system; The Netherlands). T2- and T1-weighted images were obtained in multiple planes (axial, coronal, and sagittal). The workstation was a personal computer; the authors reviewed the images together by consensus using the DICOM viewer (K-PACS, software version V 1.6.0). A special attention was given to study the degree of penile development in these cases with hypoplastic penile urethra. The penile corporeal tissue is well demonstrated in MRI by its hyper intense signal in T2WI [12]. For clarification, the study group was compared to another control group of boys who underwent pelvic MRI for other indications (e.g., perianal fistula). Normally, the penis is formed of three corporeal bodies: two dorsal corpora cavernosa and one ventral corpus spongiosum surrounding the penile urethra (Fig. 2).

Results

During the period between 2016 and 2021, the study included two newly diagnosed cases with abnormal communication between the lower urinary tract and the anus (the so-called Y-type urethral duplication), in addition to one previously reported case [7] whose preoperative imaging studies (including pelvic MRI) were available for analysis. Different surgical solutions have been applied for each case. Dilatation of the anterior penile urethra was tried in the first case, followed by mobilization and transfer of the ectopic anal urinary meatus to the perineum. In the second case, reconstruction of the urethra was performed on three stages (Fig. 3): (1) ligation/excision of accessory urethro-anal channel + perineal urethrotomy (orthotopic posterior urethra); (2) tubularized inner preputial island flap to replace hypoplastic penile urethra; (3) anastomosis between perineal urethrotomy and anterior neo-urethra. After counseling with parents, the third case was managed conservatively. Table 1 summarizes the clinical presentation, investigations, management, and follow-up of the three cases included in the study.

We report the following observations from studying the following three cases.

The penile urinary meatus (Fig. 1)

This was a narrow "hypoplastic" meatus in the three cases, consistent with the associated hypoplastic penile urethra. Although located in the glans, yet the meatus appeared slightly dorsally displaced (epispadiac).

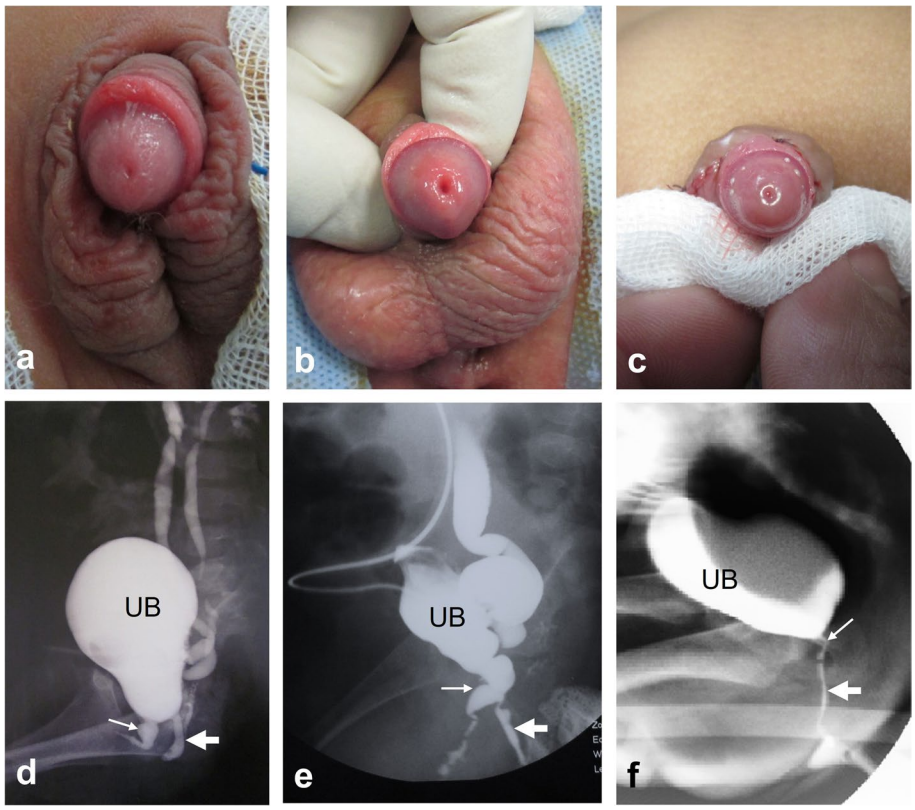

Fig. 1 Three cases with abnormal communication between the lower urinary tract and the anus (the so-called Y-type urethral duplication). **a**–**c** The hypoplastic penile urinary meatus in the three cases and their corresponding cystograms (**d**, **e**, and **f**, respectively). UB, urinary bladder; thin white arrow: patent orthotopic posterior urethra; thick white arrow: accessory uro-anal tract. Note that the cystogram was performed via suprapubic route in d and e, while the study was performed via accessory anal tract in f. Also, note that the accessory anal tract commonly originates from orthotopic posterior urethra (**e** and **f**), while it originated directly from the urinary bladder in the first case (**d**)

The ectopic (anal) urinary meatus (Fig. 4)

In two cases, this was located inside the anal canal on its anterior wall below the dentate line, while in the 3rd case, it was located just outside the anal canal on its anterior margin.

Urethral hypoplasia (Fig. 5a)

Hypoplasia was affecting the anterior (penile) urethra in the three cases, while the posterior urethra was always spared.

Associated penile dysgenesis

Hypoplasia was not only restricted to the penile urethra but also affected the corporeal bodies of the penis with variable degrees. In two cases, only a single corporeal body could be identified (Fig. 2). The latter was in the position and most probably replacing the two corpora cavernosa; neither the bulb nor the corpus spongiosum could be identified in these two cases. In another case (Fig. 5), the three corporeal bodies were present but appeared distorted (irregularities and interruption of corporeal bodies).

Associated urinary tract anomalies

The urinary bladder appeared not uniform with thickened wall reflecting some degree of impaired drainage (Fig. 1). Commonly, it was associated with vesico-ureteric reflux. Para-ureteric bladder diverticula (hutch diverticula) were noticed in one case (Fig. 6), which was also associated with small dysplastic left kidney.

Associated anorectal anomalies

Two cases had mild anterior anal misplacement. One of them suffered constipation with the start of weaning at the age of 6 months, for which he underwent simple "cut-back" anoplasty (Fig. 7).

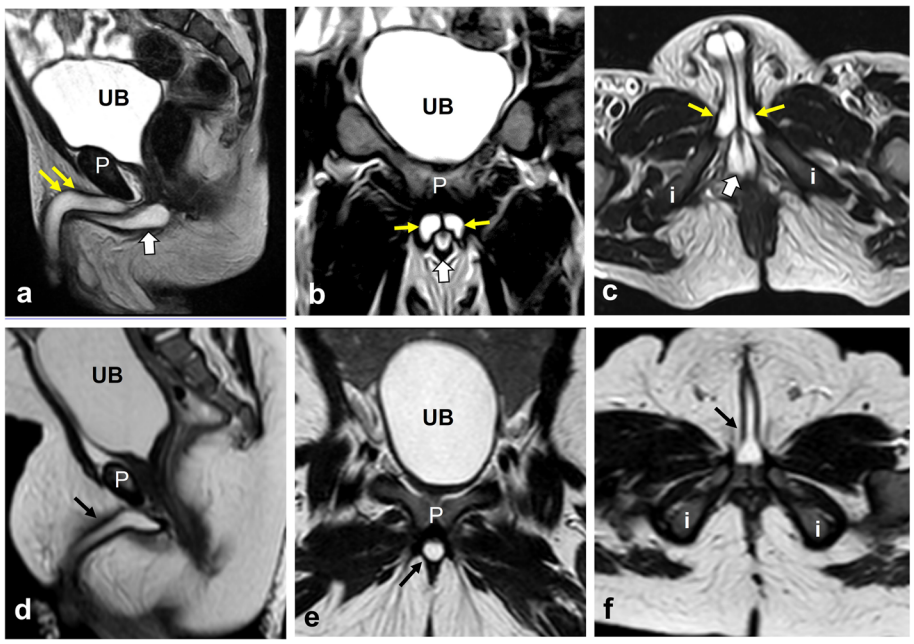

Fig. 2 Penile corporeal dysgenesis in the so-called Y-type urethral duplication compared to normal control. Upper row (**a**, **b**, and **c**): Pelvic MRI (sagittal, coronal, and axial, respectively) of a 24-month-old boy (control) demonstrating normal anatomy: two dorsal corpora cavernosa (yellow arrows) and ventral corpus spongiosum (white arrow). Lower row (**d**, **e**, and **f**): Pelvic MRI (sagittal, coronal, and axial, respectively) of a 5-month-old boy with accessory urethro-anal tract. Note that the three corporeal bodies in the control (upper row) are replaced by a single corporeal body (single black arrow) in the case with accessory urethro-anal tract (lower row). UB, urinary bladder; P, pubic symphysis; i, ischial tuberosity

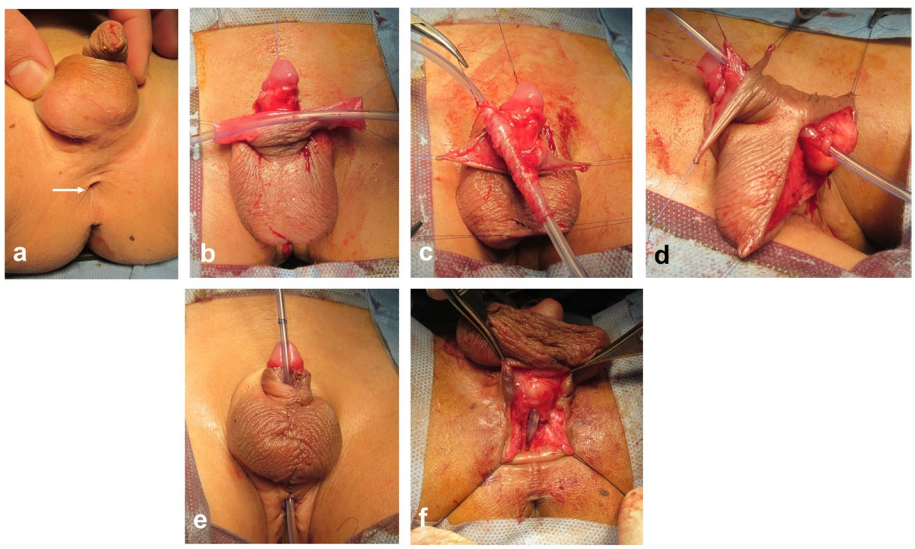

Fig. 3 Demonstration of the 3-stage reconstruction of the urethra in second case. **a** At first, a perineal urethrotomy was performed (white arrow) using the orthotopic patent posterior urethra. **b**–**e** 2nd stage: Tubularized inner preputial island flap to replace hypoplastic penile urethra. **f** 3rd stage: anastomosis between perineal urethrotomy (orthotopic posterior urethra) and anterior neo-urethra

Discussion

Congenital anomalies usually present as a spectrum with variable expression and different degrees of severity; these are best understood through studying embryology. This report discusses one of the rare congenital anomalies where there is abnormal communication between the lower urinary tract and the anus. This condition may be inadvertently classified as a type of urethral duplication

Table 1 Summary of the clinical presentation, investigations, management, and follow-up of the three cases with abnormal communication between the lower urinary tract and the anus (the so-called Y-type urethral duplication)

	Clinical presentation	Renal ultrasound/renal isotopic scan	Micturating cystourethrogram	MRI: developmental status of the penis	Management	Follow-up
Case 1	- Urine flow through the anus; few drops through penile urethra (Fig. 1a) - Recurrent epididymo-orchitis - Slight anterior anal misplacement - Congenital deafness	- Bilateral mild hydroureteronephrosis (more on left) - Increased wall thickness of urinary bladder - Bilateral cortical scaring in DMSA renal isotopic scan	- Non uniform bladder contour (pear-shaped) - Abnormal accessory channel communicating between urinary bladder and anus (Fig. 1d) - Hypoplastic anterior urethra; patent posterior urethra - Bilateral vesico-ureteric reflux (grade III)	Penile corporeal dysgenesis: penis is formed of a single corporeal body	- Trial of dilatation of hypoplastic penile urethra (PADUA)→failed - Permanent supra-vesical port for CIC (Mitrofanoff) - Separation and transfer of distal end of posterior accessory channel from the anus to the perineum anteriorly - Recurrent stricture of perineal meatus that required reoperation twice	- Follow-up duration: 10 years - The patient passes urine through perineal meatus, yet we opt for CIC (supra-vesical port) to ensure adequate bladder drainage - The patient is kept on prophylactic antibiotic and anticholinergic drugs to control recurrent UTIs
Case 2	- Urine flow through the anus; few drops through penile urethra (Fig. 1b) - Marked bilateral hydronephrosis	- Left kidney is small in size with poor corticomedullary differentiation and thinned parenchyma - Increased wall thickness of urinary bladder - Bilateral cortical scaring in DMSA renal isotopic scan	- Non uniform bladder contour with posterior paraureteric diverticulae (hutch diverticulae) - Abnormal posterior channel communicating between posterior urethra and anus (Fig. 1e) - Hypoplastic irregular anterior urethra; patent posterior urethra - Bilateral vesico-ureteric reflux (grade VI)	Although the penis was formed of three corporeal bodies, yet it showed some degree of disorganization (irregularities and interruption of corporeal bodies)	- Initially, suprapubic cystostomy at presentation - Ligation and excision of accessory posterior channel + perineal urethrotomy using the distal patent end of orthotopic posterior urethra (1st stage) - Ureteric reimplantation + Permanent supra-vesical port for CIC - Reconstruction of anterior urethra using inner preputial island tube (2nd stage) - Anastomosing the reconstructed anterior urethra to the perineal urethrotomy (3rd stage)	- Follow-up duration: 6 years - Weak urinary stream through reconstructed neo-urethra (meatus is distal penile) - Patient is kept on regular CIC (supra-vesical port) to ensure adequate bladder drainage and protect upper tract

Table 1 (continued)

	Clinical presentation	Renal ultrasound/renal isotopic scan	Micturating cystourethrogram	MRI: developmental status of the penis	Management	Follow-up
Case 3	- Urine flow through abnormal orifice on the anterior margin of the anus; few drops through penile urethra (Fig. 1c) - Slight anterior anal misplacement + constipation	- Normal renal ultrasound - Normal renal scan	- Abnormal posterior channel communicating between posterior urethra and anus - Hypoplastic penile urethra; patent posterior urethra	Penile corporeal dysgenesis: penis is formed of a single corporeal body (Fig. 2)	- After counseling with parents, we decided not to perform reconstruction of the lower urinary tract as far as the urinary system is in equilibrium - Instead of circumcision, a dorsal vertical split of the prepuce (preputeoplasty) was performed to spare preputial tissues for possible reconstruction of anterior urethra (if the patient or parents would change their mind) - Cut-back anoplasty for constipation	- Follow-up duration: 12 months - He is urinating through ectopic perineal orifice on anterior anal margin - Close follow-up to ensure adequate bladder evacuation - The option of shifting to a permanent supra-vesical port for CIC is explained to parents if turns to be necessary

PADUA Progressive augmentation by dilating urethra anterior, *CIC* Clean intermittent catheterization, *UTIs* Urinary tract infections

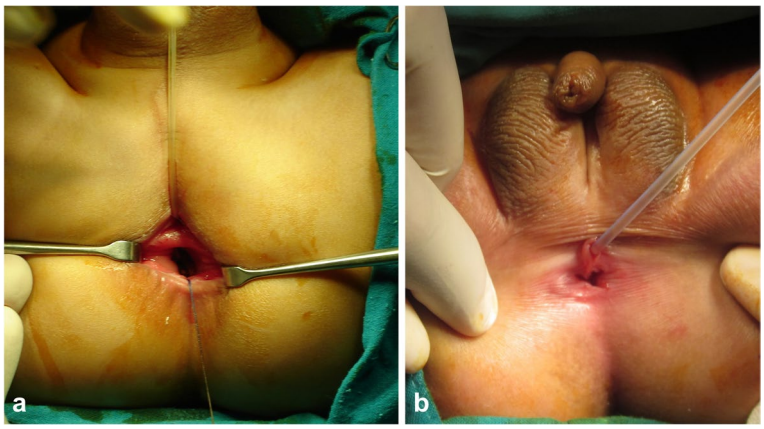

Fig. 4 The ectopic (anal) urinary meatus was located inside the anal canal on its anterior wall below the dentate line (a); while in the 3rd case (b), it was located just outside the anal canal on its anterior margin

[7]. We believe that proper identification of the true nature of the disease can help to improve and standardize surgical management.

During the early embryonic period (4–6 weeks after fertilization), the developing uro-rectal septum starts dividing the embryonic "cloaca" into anterior urogenital and posterior anorectal components [5, 6]. The urogenital component will give rise to the urinary bladder and the posterior urethra in the male. The process of complete separation of the anorectum from the lower urinary tract may become interrupted by the presence of obstruction (atresia) either at the anal side or the urethra; this would result in persistence of abnormal communication between both tracts (persistent cloacal duct of Reichel) [5, 6, 13]. In the literature, it is well documented among boys with imperforate anus that the rectum usually remains communicating via a fistula with the urinary tract (recto-bulbar/prostatic/bladder neck) [7]. On the other hand, urethral atresia is a much less common disorder; the condition is usually lethal causing fetal death due to severe renal dysplasia and lung hypoplasia unless urine can find its way out (escape) through a patent urachus or persistent communication with the anus [14]. A famous example for the latter condition is aphallia (boys presenting by absent penis and urination through the anus) [10].

In a previous report [10], Macedo et al. referred to the similarity between cases of aphallia (penile agenesis) and the so-called Y-type urethral duplication when they used the same surgical strategy for reconstruction of the lower urinary tract in both groups. In our present study, pelvic MRI examination clearly unveiled significant degrees of corporeal dysgenesis among cases diagnosed as Y-type urethral duplication that would strongly suggest these cases to be part of the same disease spectrum "penile dysgenesis/agenesis".

Based on embryological explanations and current MRI findings, we may suggest penile dysgenesis to be the primary pathology in cases described as Y-type urethral duplication. The anterior urethra lies within the penis and therefore will be affected by its defective development (penile dysgenesis). This may reach up to complete absence in cases of aphallia (penile agenesis) at the extreme end of spectrum. On the other hand, the posterior urethra was always spared due to different embryologic origin. Significant hypoplasia/atresia of the anterior urethra will redirect the main urinary stream backwards via persistent abnormal communication between the posterior urethra and the anus (persistent cloacal duct of Reichel) [5]. Through studying an autopsy specimen [6], Stephens and Donnellan showed this urethro-anal tract to be devoid of muscle coat; moreover, it showed severe shrinkage during specimen preparation. This may explain the reported high incidence of strictures and failure when using the accessory uro-anal tract in reconstruction of the urethra and would indicate for its true fistulous nature [7]. Sinha et al. reported using a strip of rectal wall in continuity with the posterior ectopic urinary meatus adding extra length to the uro-anal tract during its mobilization to the anterior perineum [15]. Others recommended discarding the accessory uro-anal tract, and instead, to incorporate the orthotopic patent posterior urethra in the repair [6].

The hypoplastic penile urethra is usually reconstructed/replaced by applying the principles of hypospadias surgery: preputial island flaps or grafts (buccal mucosa). Trials of progressive dilatation of the hypoplastic penile urethra (PADUA) [16] are usually prone to failure [3, 9]. This may be related to the dysgenetic nature of penile urethra in these cases [1, 9]. Moreover, the penile urethra may not be perfectly orthotopic within

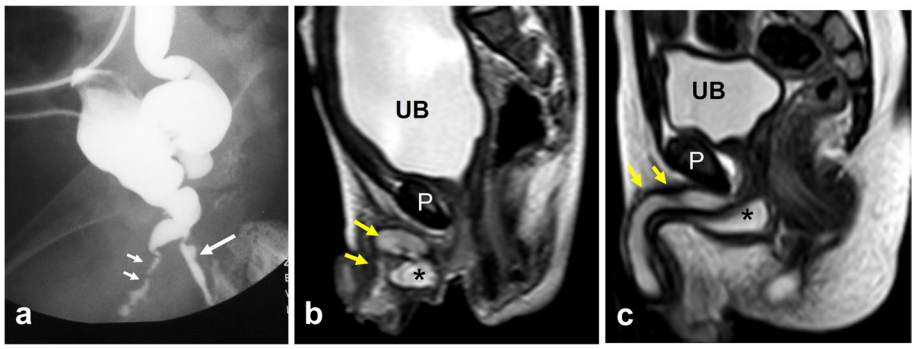

Fig. 5 **a**, **b** Second case with abnormal accessory urethro-anal tract: **a** micturating cystogram demonstrating hypoplasia of penile urethra (short white arrows) + accessory tract between posterior urethra and anal canal (long white arrow); **b** pelvic MRI (mid sagittal T2WI) of the same case demonstrating associated corporeal dysgenesis. **c** Control group for comparison: pelvic MRI (mid sagittal T2WI) of a 33-month-old boy demonstrating normal penile corporeal anatomy. UB, urinary bladder; P, pubic symphysis; asterisk: corpus spongiosum; double yellow arrows: corpus cavernosum

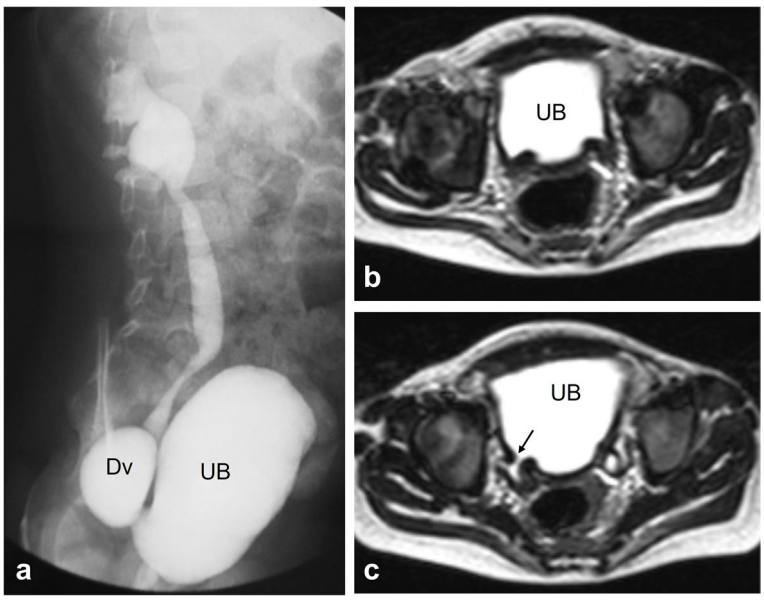

Fig. 6 Second case with accessory communication between the lower urinary tract and the anus. **a** Cystogram demonstrating vesico-ureteric reflux and posterior bladder diverticulum (hutch diverticulum). **b**, **c** Axial pelvic MRI (T2WI) demonstrating the opening of diverticulum (arrow). UB, urinary bladder; Dv, bladder diverticulum

the corpus spongiosum as expected but rather running an ectopic course on the dorsum of the penis [3]. On cystoscopy, Macedo et al. reported ectopic implantation of the hypoplastic urethra into the posterior urethra [8]. The hypoplasia together with the dorsal ectopic course may indicate this penile urethra to be rather an accessory urethra; the latter might have been formed after occurrence of a major vascular accident interrupting the development of corpus spongiosum and the orthotopic penile urethra inside [13]. A similar condition has been described in the female (posterior cloaca), where the urogenital sinus is directed backwards to open through the anus; an accessory urethra running through the tip of the clitoris has similarly been described [17].

The lack of consensus on the best surgical treatment, besides the overall less satisfactory results in the literature, may explain the different treatment options applied for each case in this study. Macedo argued the risk/benefit of complex procedures for complete reconstruction of the urethra up to the tip of penis [8, 11]. Instead, he suggested a permanent perineal urethrotomy for voiding and a functional penis for sexual intercourse [11]. Similarly,

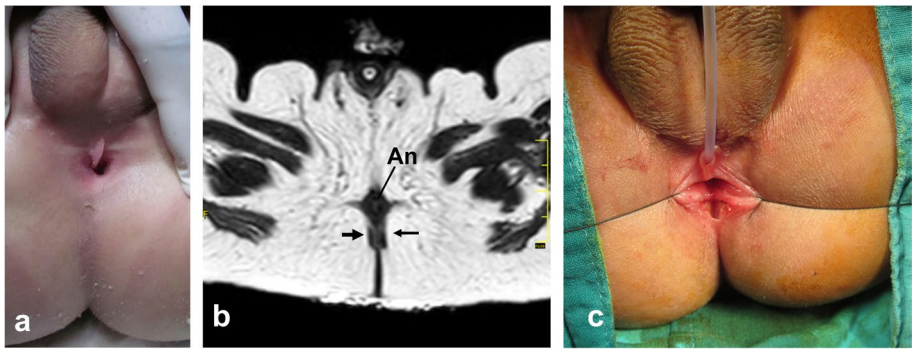

Fig. 7 A case with abnormal communication between the lower urinary tract and the anus (3rd case). **a** Associated mild anterior anal misplacement. **b** Confirmation of anterior anal misplacement in MRI. An: anal canal, black arrows point to normal anal position. **c** Simple posterior anoplasty (cut-back) to manage constipation in this case

Lorenz and colleagues raised a question whether we really need to reconstruct the lower urinary tract in these patients [3]. When adequate bladder drainage is questionable, a permanent supra-vesical channel for clean intermittent catheterization may be a better alternative for these patients and their families; this may reduce number of surgical interventions, shorten hospital stay, and facilitate follow-up [3]. Anyway, the available surgical options and limitations should be clearly and objectively discussed with parents [3, 11]. Whatever was the extent and type of surgical reconstruction, it is of utmost importance to protect the vulnerable upper urinary tract in these patients by keeping them on close long-term follow-up [3]. This can help to avoid terrible situations seen in these cases ending by renal failure [2, 11].

Although this study included only three cases, yet associated penile corporeal dysgenesis was quite evident and well demonstrated in MRI. Further studies on more cases and including other disease variants with functioning penile urethra can help to complete the picture and reach a consensus on the true nature of the disease whether a 'fistula' or a 'duplication' of the urethra [18]. We believe that proper understanding of the pathology is important to make correct surgical planning and improve outcomes.

Conclusion

In our present study, pelvic MRI examination clearly unveiled significant degrees of corporeal dysgenesis among cases diagnosed as Y-type urethral duplication that would strongly suggest these cases to belong to the same disease spectrum of penile dysgenesis/agenesis.

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None.

Authors' contributions
Data acquisition, analysis, and interpretation were performed by both authors. AAA made the drafting of the manuscript. Both authors have read and approved the final manuscript.

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None.

Availability of data and materials
The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
A written parental consent was taken before operation in all cases. Owing to the retrospective nature of the study, an IRB number was not required, and the study was approved through expedited review by the scientific/ethical committee of the Pediatric Surgery Department (Faculty of Medicine; Ain-Shams University).

Consent for publication
Patient identity did not appear in any part of the manuscript; therefore, consent for publication was not required.

Competing interests
The authors read and approved the final manuscript.

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