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ABOUT COVER
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RESPONSIBLE EDITORS FOR THIS ISSUE
Production Editor: Ying-Yi Yuan; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lei Wang.
Bilateral ureteral reimplantation in a patient with an intraperitoneal ectopic bipenis: A case report

Ya-Tao Jia, Bao-Lei Shi, Jie Zhang, Ying-Yi Li, Jiang Zhu

BACKGROUND
Diphallia is a highly uncommon congenital urogenital abnormality and a few connected reports have been published. However, no case of intraabdominal heterotopic diphallus has been documented to date. In the present study, we present a rare case of intraperitoneal ectopic bipenis.

CASE SUMMARY
A 49-year-old man was hospitalized with the chief complaint of hydronephrosis of both kidneys, which was discovered three days earlier through regular physical examination performed using urological ultrasound without significant lumbar or abdominal pain or bladder irritation. Physical examination showed normal external penile development, bilateral testes located on the left side of the scrotum, and a fused epididymis. Urological plain and enhanced computed tomography suggested bilateral hydronephrosis, bilateral ureters opened to the left side of the bladder wall; an intrapelvic soft tissue shadow on the left side of the bladder was considered a germline malformation called bipenis (hidden penis in the abdominal cavity). Based on the urological plain and enhanced computed tomography results, a 49-year-old man was diagnosed with bipenis (one hidden in the abdominal cavity). Ectopic penile compression produced bilateral ureteral dilatation and hydronephrosis. The ectopic penis was amputated and partially removed during surgery, and bilateral ureteral reimplantation was successfully performed. At a 2-mo follow-up, the patient was very satisfied with the operation, there was no significant hydronephrosis in both kidneys, and urination and erectile function were normal.

CONCLUSION
To our knowledge, this is the first report of diphallia with an intraperitoneal ectopic penis. Computed tomography or magnetic resonance imaging can be used to assess the associated internal anomalies before surgery. Postoperative pathological findings are the gold standard for the diagnosis.
INTRODUCTION
Diphallia (penile duplication) is an extremely rare congenital urogenital anomaly, with an incidence of approximately one in 5-6 million births\[1,2\]. Since Wecker reported the first case in 1609, reports on more than 100 cases have been published\[3\]. Most of the available reports are about penile duplication in the perineum, which is visible to the naked eye. This is the first report of a patient with intraperitoneal ectopic bipenis to our knowledge, mainly based on the literature review on PubMed database.

CASE PRESENTATION

Chief complaints
A man in his 40s was admitted to the hospital with the chief complaint of hydronephrosis in both kidneys for 3 d.

History of present illness
Without significant lumbar or abdominal pain or bladder irritation.

History of past illness
He had previously undergone ureteral stent placement on the right side due to bilateral hydronephrosis in January 2020; however, ureteroscopic placement on the left side failed. Left ureterotomy and ureteral stent implantation were performed accordingly. The bilateral ureteral stents were removed 3 mo postoperatively. Other past history were all negative.

Personal and family history
The patient had no relevant personal and family history.

Physical examination
Physical examination showed normal external penile development, bilateral testes located on the left side of the scrotum, and a fused epididymis. The left-sided scrotal swelling that transilluminated upon application of light suggested a hydrocele.

Laboratory examinations
Biochemical blood tests revealed no apparent abnormalities.

Imaging examinations
A previous ureteroscopy for bilateral hydronephrosis revealed bilateral ureteral malformation, derangement, and displacement of the seminal vesicle glands to the left side. Urological plain and enhanced computed tomography suggested bilateral hydronephrosis, ureteral dilatation, and ureteral confluence on the left side of the bladder, a malformation opening on the left side of the bladder wall (Figure 1), and an intrapelvic soft tissue shadow on the left side of the bladder, was considered a germline malformation, namely a bipenis (hidden penis in the abdominal cavity) (Figure 2).
Figure 1 Urological plain and enhanced computed tomography coronal images. A: A columnar soft tissue-like density shadow of around 8.0 cm × 2.0 cm in length on the left side of the pelvis during urological computed tomography (CT) (orange arrow); B: Enhanced CT showing mild homogeneous enhancement of the mass (orange arrow).

Figure 2 Urological scan and enhanced computed tomography sagittal images. A: Left ureteral orifice (orange arrow); B: Ectopic opening of the right ureter opens on the left side of the bladder wall (orange arrow); soft tissue shadow on the left side of the bladder is identified as intra-abdominal ectopic penis (blue arrow).

FINAL DIAGNOSIS
Postoperative pathology suggested that the urethra and urethral and penile corpus cavernosum tissues were visible in the specimen sent for examination, which was consistent with an ectopic penile morphology (Figure 3).

TREATMENT
Intraoperative exploration around the bladder revealed 8.0 cm × 2.0 cm columnar solid tissue outside the left posterior wall of the bladder, with tough texture, reaching down to the anterior rectal wall and extending upward to the pelvic wall (Supplementary Figure 1). No seminal vesicle was seen, and the lumen of the left ureter was dilated, with a diameter of about 1.5 cm, passing behind the columnar tissue. The right ureter was carried out in the cysto-rectal space, with a lumen diameter of approximately 1.0 cm; the pelvic columnar neoplasm was considered a repeat ectopic penis. Due to serious adhesions, partial excision of the pelvic mass was performed, with a length of approximately 2.0 cm excised from the middle section, the smooth muscle tissue visible inside, and a tubular channel visible in the middle. The 7-0 silk sutures were wrapped and sutured at the stump, whereas the right and left ureters were loosened. Next, the ureters were cut short at the lowest point and reimplanted on the left and right anterior walls of the bladder, respectively, while bilateral ureteral stents were left in place.
OUTCOME AND FOLLOW-UP

The patient was discharged with no postoperative events. Follow-up was performed for 2 mo, with no reported adverse events. The patient was advised to visit regularly, especially within 1-2 years after surgery.

DISCUSSION

Penile duplication is an extremely rare congenital anomaly that varies in presentation and location from case to case[1,4]. Existing reports in the literature refer mainly to external penile duplication, whereas intra-abdominal ectopic penis has not been reported to date.

The etiology of diphallia remains unclear; however, there are many possible embryological explanations. Duplication of the penis is caused by a lack of fusion of the paired mesodermal anlagen of the genital tubercle by the 15th week of gestation[5]. Many genetic alterations have been implicated in the development of supernumerary penises, some of which include genes that encode androgen receptors and are linked to the development of male external genitalia[3,4]. Diphallia is not considered familial or hereditary[6].

There are many clinical classifications for these conditions, and some authors[2] classify them as follows: (1) True diphallia, (A) complete or (B) partial; (2) Bifid phallus: (A) partial (a) bifid glans (b) bifid shaft, or (B) complete. In 2017, Jesus et al[7] proposed a new simplified classification based on the clinical and surgical implications of each type: (1) True penile duplication (each duplicate penis has two corpora and one spongiosum); (2) Hemiphalluses (each penis has corpora and a hemiglans); (3) Pseudoduplication (normal penis with accessory penis-like tissue); and (4) Partial duplication (duplication involving only the distal penis). Our case had true penile duplication. True duplication is associated with other congenital defects[4,8,9]. Our case also involved anomalies, such as bilateral ureteral opening malformations, bilateral testicular ectopia, and bilateral epididymal fusion.

In general, ultrasonography helps to confirm the diagnosis by detecting the presence or absence of the corpus cavernosum or spongiosum and its number. It can also detect other associated abnormalities. However, this case was unique because the ectopic penis was located in the abdominal cavity. Furthermore, inexperienced sonographers may not always be able to identify this malformation. Magnetic resonance imaging (MRI) is a valuable method to accurately diagnose diphallia and associated malformations because T2-W images have the appropriate contrast resolution[1,10,11]. One limitation of the study is that no further MRI was performed in this case to confirm the diagnosis.

Our case was first identified by radiologists, and the possibility of an intra-abdominal ectopic penis was considered based on plain and enhanced computed tomography of the urinary tract, which was dependent upon the radiologists’ experience in reading the films and provided important information for the physicians.

In general, the associated malformations should be treated first[12]. In the case of actual penile duplication, partial duplication, or pseudo-duplication, most surgeons choose to resect the hypoplastic duplicate penis, glans, or accessory penile-like tissues to maintain the main urethra[13]. In this case, the ectopic penis was located in the abdominal cavity and crossed the left ureter anteriorly, which caused bilateral ureteral dilatation and hydronephrosis. Bilateral ureteral reimplantation is required to further protect renal function, and dissociation of the ectopic penis is also compulsory.

As the patient was a middle-aged male, he had no reproductive needs for the time being, but the status of the patient's sexual activity still requires further attention after surgery.
CONCLUSION

To date, intra-abdominal ectopic bipenis has not been reported in humans. Penile duplication has a unique presentation in each patient, in which the position of the penis can be either ectopic or orthotopic. Depending on the corporal development and anatomy of the urethra, excision or reconstruction of the duplicate penis is required.

FOOTNOTES

Author contributions: Li YY and Zhu J designed the research study; Jia YT and Zhju J performed the research; Jia YT, Shi BL, and Zhang J Searched relevant literature and review; Jia YT and Zhu J sort out the information and wrote the manuscript; all authors have read and approve the final manuscript.

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Conflict-of-interest statement: All authors declare that they have no potential conflicts of interest.

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