Case Report

Ureteric bud remnant with renal agenesis

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Abbreviation & Acronym
RET = rearranged during transfection

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Introduction: Ureteric bud remnant with renal agenesis is a rare disorder when urogenic system develops. Because of no obvious symptoms, it is usually explored incidentally.

Case presentation: A 41-year-old male presented with intermittent discharge of turbid fluid from his scrotum. A pinhole was noted in his left scrotum, and an infectious sinus or fistula was impressed. After serial studies, computed tomography revealed agenesis of his left kidney and a cystic lesion over his left scrotum. He underwent resection of the infectious sinus. Near the tail of the sinus, a connection was found to a channel-like structure. Contrast medium was injected which showed a blind end of this channel-like structure. The tube was ligated, and the cut end was sent for surgical pathology, which confirmed a left ureteric bud remnant.

Conclusion: Complete imaging studies make this diagnosis clearly.

Key words: renal agenesis, ureteric bud remnant.

Keynote message
Ureteric bud remnant with renal agenesis is rare. Full imaging examination should be done if it is suspected.

Case presentation
A 41-year-old man presented to our outpatient department with intermittent discharge of a turbid fluid (<1 mL) from his scrotum. On physical examination, a pinhole was noted over his left scrotum with squeezing pain and discharge. Infectious sinusitis or a fistula was impressed initially. An anal fistula was ruled out by digital rectal examination, and a urethra-cutaneous fistula was also excluded by cystourethroscopy. However, the cystourethroscopy examination revealed that there was no left ureteral orifice.

Computed tomography revealed agenesis of his left kidney (Fig. 1a) and a cystic lesion over his left scrotum (Fig. 1b). Because of a recurrent local heat sensation, turbid discharge, tenderness, and infection, he underwent resection of the infectious sinus (Fig. 2a). During surgery, the wound was deepened along with the sinus to the retropubic bone. Near the tail of the sinus, a connection was noted to a channel-like structure. For further investigation, patient was placed at supine position, and mobile C-arm X-ray system was setting for image study. Contrast medium was injected and revealed a channel-like structure about 15 cm in length with a blind end in an upward direction (Fig. 2b). The setting of mobile C-arm X-ray system is conversely, so the picture showed mirror image. The direction of this tubule was compatible with the development of the left ureter. The tube was ligated and the cut end was sent for surgical pathology. The specimen is confirmed as ureter histologically (Fig. 2c).

Discussion
Ureteric bud remnant with renal agenesis is a rare congenital disease that has only been reported in a few case reports.1 It is usually discovered incidentally,2 and computed tomography and ultrasonography have been reported to be of limited use in its detection.3 Although magnetic resonance imaging has been reported to be more advantageous than computed
tomography or ultrasonography, it is not routinely used to identify this disease. Our young male patient suffered from scrotal discharge, which is not usually associated with renal agenesis. A previous study reported that the incidence of ipsilateral renal agenesis or dysplasia was only 0.00464% when incidentally noted in ultrasonography and thought to be a pelvic cyst.4

The pathophysiology of this rare case is unknown. The ureter bud remnant may process initially when fetal development. However, it stopped due to unknown reason. Some metanephric mesenchyme development disorder may cause this situation. The different interactions between the epithelial ureteric bud and the metanephric mesenchyme cause congenital abnormalities, such as Zinner’s syndrome, which shows renal agenesis with ipsilateral seminal vesicle cyst, and ejaculatory duct obstruction. In our case, we only see left renal agenesis without ipsilateral seminal vesicle cyst (Fig. 1c).

In gene level, some proto-oncogenes have also been reported. A ureteric bud remnant with renal agenesis is formed due to a mutation of the proto-oncogene. RET is a receptor tyrosine kinase which can induce cell growth and differentiation. A previous animal study reported abnormal kidney and ureter development in around 90% of RET−/− mice, with a ureteric bud remnant with renal agenesis in about 2% of the RET−/− mice.5

There are some limitations in this case report. The diagnosis of ureteric bud remnant with renal agenesis is not suspected initially, so complete imaging study is not doing well. The definite mechanism of this patient’s disease is not sure. It may be due to some mistakes occurred during congenital renal development. The reason of proto-oncogene cannot be confirmed either.

In such rare cases with a diagnosis of infectious sinusitis, complete imaging studies such as computed tomography, ultrasonography, or magnetic resonance imaging should be done preoperatively. Differential diagnoses may include renal agenesis or dysplasia, ectopic ureter, infectious sinusitis, urethral fistula, or anal fistula. Complete resection of the sinus should be considered to confirm the definite diagnosis.

Conflict of interest
The authors declare no conflict of interest.

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