Urethral calculi with a urethral fistula: a case report and review of the literature

Mingqiang Zeng1,2, Fanchang Zeng2,3, Zhao Wang2, Ruizhi Xue2, Liang Huang2, Xuyu Xiang2, Zhi Chen2 and Zhengyan Tang2*

Abstract

Background: To explore and summarize the reasons why urethral calculi cause a urethral fistula.

Case presentation: We retrospectively studied 1 patient in Xiangya hospital and all relevant literature published in English between 1989 and 2015. The patients (including those reported in the literature) were characterized by age, origin, location of calculus, size of calculus, fistulous track, and etiological factors. Most of urethral calculi associated with a urethral fistula were native generated. Urethral calculi can be formed in various locations of the urethra, and the size of the calculus ranged from small (multiple) calculi to giant stones. The fistula external orifice located at the root of the penis was relatively common, and there were various etiological factors, such as urethral strictures, urethral trauma induced by long-term catheterization, lumbar fractures, and congenital anomaly factors. They were managed by the excision of the fistulous tract, retrieval of the urethral stones, and/or debridement and pus drainage operations.

Conclusion: Some elements, such as trauma, recurrent urinary tract infections, abscess formation induced by long-term catheterization, and urethral calculus, may be the risk factors for a urethral fistula.

Keywords: Urethral calculus, Urethral fistula, Urinary tract infections, Suprapubic catheter
was found, with the complete obliteration in the anterior urethra. The patient was administered intravenous fluids and antibiotics in the perioperative period. After recovery, he returned to the hospital monthly to replace the SPC. The patient’s condition was followed for 6 months, during which time the SPC blockage, severe urinary tract infections (UTIs), urinary calculi, and renal damage did not recur.

**Literature review**

We searched PubMed (http://www.ncbi.nlm.nih.gov/pubmed) for articles published in English between 1989 and December 2015 with the terms “urethral calculus”, “urethral steinstrasse”, “fistula”, “urethral fistula”, “urethrocutaneous fistula”, or “recto-urethral fistula”. There were 5 reports included in our study (Table 1). In these studies, all the urethral calculi were native generated (they were formed de novo in the urethra) and formed in different locations of the urethra (in the proximal penile, bulbar, and posterior urethra regions), the size of the calculi ranged from small (the size of a grain of sand) to massive (stones ~6 × 5 cm), and it was common to find the fistula external orifice at the root of the penis. There were also various etiological factors, such as the urethral stricture, urethral trauma induced by long-term catheterization, lumbar fractures, and congenital anomaly factors. Some cases exhibited anatomic abnormalities, such as having a stone in the large diverticulum. Initially, these cases were managed with suprapubic cystostomy and intravenous antibiotics. Later, the patients underwent external urethrotomy and stone retrieval.

**Discussion**

Urethral calculi accounted for only 1–2% of the urinary tract stones. Urethral calculi that cause urethral fistula are extremely rare [1], but they still cause serious
discomfort in many patients with the disease. In order to obtain a deep understanding of the disease, we analyzed the origin of the urethral calculi and summarized the causes of the urethral calculi that result in a urethral fistula.

The origin and management of the urethral calculi
Urethral calculi are either formed in the native urethra or migrate from the upper urinary tract [7]. Primary native calculi (those formed de novo in the urethra) are usually small and come in multiples, and secondary migratory calculi (those formed in the upper urinary tract with secondary downward descent) are usually large [8]. We found that the size of the primary native urethral calculi ranged from 2 to 3 mm to 6 cm, larger calculi usually occur in the posterior urethra and vesico-prostatic urethra, and a small calculus is commonly found in the anterior urethra. Primary urethral stones are generally composed of magnesium ammonium phosphate (struvite) [8]. This finding is consistent with our investigation of the stone components. The stones are formed in the urethra either behind some stricture or within a poorly drained communicating cavity, with an obstruction, stagnation, infection, and/or inflammation acting as the predisposing factor [9]. The above predisposing factors exactly explain the formation of various urethral stones in the studies. Secondary or migratory stones are usually composed of calcium oxalate or citrate [3], and they are very common. Migratory stones are most often encountered in association with urethral stricture disease or other forms of urethral obstruction [10].

The main symptoms are acute urinary retention, frequency, a burning sensation in the urethra during urination, a burning sensation in the perineum and/or rectum, or a stinging in the anus. Other less common symptoms included haematuria, dribbling or incontinence, interruption of the urinary stream, and a history of having passed a stone. Management of urethral calculi varied according to the site, size, and associated urethral disease. Retrograde manipulation into the urinary bladder followed by litholapaxy or lithotripsy is a suitable procedure for small urethral calculi. Anterior urethral calculi can be removed with surface anesthesia, endoscopic removal, or ventral meatotomy [11].

Etiology and treatment of the urethral fistula
Urethral fistulas may be acquired or congenital. Acquired urethral fistulas may be neoplastic, traumatic, or caused by a urethral foreign body or infection [12–15]. Urethral fistulas have been found after a straddle injury [16] and blunt penile trauma [17]. They can also occur as a complication of penile surgery. Urethral fistulas have been described after circumcision [18] and operations for priapism [19]. In this study, we found that the majority of the urethral fistulas were found secondary to urethral calculi complicated by trauma and infection. They may have occurred as a result of a penile-scrotum abscess. The patients with primary urethral stones were usually asymptomatic or had chronic voiding problems. An important cause of a urethral fistula may be related to delayed treatment. A congenital urethral fistula represents either an embryonic urethral blowout behind a distal congenital obstruction or segmental embryonic arrest so that the mesoderm fails to encircle the developing groove at the site of the fistula [12]. Congenital urethral fistulas may be associated with anorectal atretic

| No. | Case load | Age   | Ethnicity | Native/migratorya | Location of calculus | Size of calculus (cm) | Fistulous track | Etiological factors |
|-----|-----------|-------|-----------|-------------------|----------------------|----------------------|------------------|--------------------|
| 1   | 1         | 38-year-old Indian Native | Penobulbar junction | 6 × 5 | Near the penoscrotal junction | Stricture of penile urethra distalb |
| 2   | 1         | 41-year-old Osmanli Native | At the prostatic urethra | 5.9 × 3.2 × 2.8 | Below the radix of penis | Urethral trauma as a result of long-standing urethral catheter drainage |
| 3   | 1         | 30-year-old English Native | Recto-urethral fistula | 5 × 3.5 | The membranous urethra | Born with abnormalities |
| 4   | 1         | 40-year-old Indian Native | In proximal penile, bulbar, and posterior urethra | Multiple calculi | At the root of the penis | Urethral stricture |
| 5   | 1         | 37-year-old Japanese Native | Prostatic urethral | 4.2 × 3.4 × 2.1 | Urethrocutaneous fistula in the perineal region | A lumbar vertebral fracture |

a Native: those formed de novo in the urethra. Migratory: those formed in the bladder or kidney with secondary descent
b Existed anatomic abnormality, the stone in large diverticulum
malformations, which is consistent with the No. 3 case in Table 1. The fistula of the membranous urethra extended into an abscess cavity which contained the calculus and communicated with the rectum. In the most recent studies, all patients underwent the excision of the fistulous tract and retrieval of the urethral stones [2–6], while only some of the patients with a penile-scrotum abscess underwent a debridement and pus drainage operation. It was necessary to administer antibiotics to the patients during the perioperative period.

**Urethral steinstrasse causing urethrocutaneous fistula with a long-term suprapubic catheter**

The SPC is a popular method for managing long-term bladder drainage in voiding dysfunction [20]. Although a SPC has a high success rate, there can be several complications from its use, including UTIs, bladder stones, upper tract calculi, renal scarring, vesicoureteral reflux, urethral incontinence, and even bladder cancer [21, 22]. Urethral steinstrasse was likely caused by repeated UTIs with a long-term SPC. These stones formed in the bladder with a secondary migration to the urethra that was complicated with trauma and infection, which ultimately resulted in the urethral fistula. There were 2 concurrent fistulas in the patient involved in this study, the penile fistula caused by the calculus injuries and the scrotal fistula caused by a serious fascia space infection.

**Conclusions**

This study identified some predisposing factors of urethral calculi with a urethral fistula. Patients suffering from this problem can be treated with the excision of the fistulous tract, retrieval of the urethral stones, and/or a debridement and pus drainage operation. The complications seriously affect the quality of life and even cause more serious consequences, so clinicians and patients should be aware of them.

**Abbreviations**

SPC: suprapubic catheter; CT: computed tomography; UTIs: urinary tract infections.

**Authors’ contributions**

ZT participated in the design of this study, RX performed the statistical analysis. LH carried out the study, together with FZ, XX, ZC, and collected important background information. ZW drafted the manuscript. MZ conceived of this study, and participated in the design and helped to draft the manuscript. All authors read and approved the final manuscript.

**Author details**

1. Department of Urology, Hunan Provincial People’s Hospital, The First Affiliated Hospital of Hunan Normal University, Changsha, China. 2. Department of Urology, Xiangya Hospital, Central South University, 87 Xiangya Road, Changsha 410008, Hunan, China. 3. Department of Urology, Hainan General Hospital, Haikou, China.

**Acknowledgements**

The authors thank the patient for allowing us to publish this case report.

**Competing interests**

The authors declare that they have no competing interests.

**Availability of data and materials**

All relevant data are within the paper.

**Consent for publication**

Written informed consent was obtained from the wife of the patient for publication of this case report and any accompanying images.

**Ethics approval and consent to participate**

This study was approved by the Medical Ethics Committee of the Xiangya Hospital (Approval Number: 201512532). Written informed consent was obtained from the patient.

**Funding**

This project was supported by the National Natural Science Foundation of China (NSFC) (Grant Number 81570627) and the Key Project of Science and Technology of Hunan Province (2016C2044). The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

**Publisher’s Note**

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

**Received:** 5 April 2016   **Accepted:** 31 August 2017

**Published online:** 06 September 2017

**References**

1. Koga S, Arakaki Y, Matsuoka M, Ohyama C. Urethral calculi. Br J Urol. 1990;65(3):288–9.
2. Prakash J, Sharma P, Sankhwar S, Goel A. Large anterior urethral calculus presented as scrotal mass with urethrocutaneous fistula. BMJ Case Rep. 2013. doi:10.1136/bcr-2013-200805.
3. Kaplan M, Atakan IH, Kaya E, Aktoz T, Inci O. Giant prostatic urethral calculus and urethrocutaneous fistula. Int J Urol. 2006;13(5):643–4.
4. Rowe PH, Taylor PR, Mason RC, Joyce MR, McColl I. Calculus in a congenital recto-urethral fistula: a late presentation. J R Soc Med. 1988;81:422–3.
5. Kumar S, Sharma S, Ganeshamoni R, Singh SK. Urethral steinstrasse with urethrocutaneous fistula. Urology. 2012;79(2):e1–2.
6. Toyoshima Y, Hosokawa Y, Hashimura M, Takada S, Hayashi Y, Fujimoto K, et al. A case of prostatic urethral calculus associated with perineal abscess and urethrocutaneous fistula. Hinyokika Ky o. 2012;58(1):35–8.
7. Shanmugam TV, Dhanapal V, Rajaraman T, Chandrasekar CP, Balashanmugam KP. Giant urethral calculus. Hosp Med. 2000;61(8):582.
8. Usta MF, Baykara M, Erdogru T, Koksal IT. Idiopathic prostatic giant calculus in a young male patient. Int Urol Nephrol. 2005;37:295–7.
9. Lowes OS, Kirw JJ. Clinical urology, vol. 1. 3rd ed. Baltimore: Williams and Wilkins Company; 1956.
10. McCall RW, Bann MP. Lower urinary tract calculus and alcalifications. In: Pollak HM, editor. Clinical urography. Saunders: USA; 1992. p. 1904–15.
11. Wollin TA, Singal RK, Whelan T, Dicecco R, Razvi HA, Denstedt JD. Percutaneous suprapubic cystolithotripsy for treatment of large bladder calculi. J Endourol. 1999;13(10):739–44.
12. Horton CE, Devine CJ, Graham JK. Fistulas of the penile urethra. Plas Reconstr Surg. 1980;66(3):407–18.
13. Nakaawawa S, Uemura M, Miyagawa Y, Tsujimoto A, Nonomura N. Urethral fistula and scrotal abscess associated with colovesical fistula due to the sigmoid colon cancer. Hinyokika Ky o. 2015;61(9):375–8.
14. Ramirez Martin D, Aragón Chamizo J, Jara Rascón J, Ogaya Piniés G, Piñero J, Herranz Amo F, et al. Recto-urethral fistulasecondary to prostate cancer. Arch Esp Urol. 2014;67(1):92–103.
15. Singh O, Gupta SS. Urethral foreign body causing urethral fistula. J Urol. 2012;9(1):430–2.
16. Ochsner MG, Joshi PN. Urethro-cavernous fistula. J Urol. 1982;127(6):1190.
17. Palaniswamy R, Rao MS, Bapna BC, Chary KS. Urethro-cavernous fistula from blunt penile trauma. J Trauma. 1981;21(3):242–3.
18. Ceylan K, Burhan K, Yilmaz Y, Can S, Küş A, Mustafa G. Severe complications of circumcision: an analysis of 48 cases. J Pediatr Urol. 2007;3(1):32–5.
19. Zheng DC, Yao HJ, Zhang K, Xu MX, Chen Q, Chen YB, et al. Unsatisfactory outcomes of prolonged ischemic priapism without early surgical shunts: our clinical experience and a review of the literature. Asian J Androl. 2013;15(1):75–8.
20. Jamison J, Maguire S, McCann J. Catheter policies for management of long term voiding problems in adults with neurogenic bladder disorders. Cochrane Database Syst Rev. 2013;11:CD004375. doi:10.1002/14651858.CD004375.pub4.
21. Sheriff MK, Foley S, McFarlane J, Nauth-Misir R, Crapps M, Shah PJ. Long-term suprapubic catheterisation: clinical outcome and satisfaction survey. Spinal Cord. 1998;36(3):171–6.
22. Sugimura T, Arnold E, English S, Moore J. Chronic suprapubic catheterization in the management of patients with spinal cord injuries: analysis of upper and lower urinary tract complications. BJU Int. 2008;101(11):1396–400.