Case Report **Open Access**

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Urethral stone of unexpected size: case report and short literature review

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Abstract: Urolithiasis is a well-known disease. Stones can form in all parts of the urinary tract. Diagnosis and treatment usually do not create clinical problems. However from time to time in daily practice doctors are confronted with casuistic cases. Here, we report a 54-year-old man with a large stone localized to the urethra. Because of the size of the stone, confirmed on radiography, and the presence of a cutaneous fistula, open surgery was performed. Due to urethral stricture, the patient required a second stage of reconstructive surgery. This case emphasizes that unusual presentation of urolithiasis may occur, and different surgical approaches as well as other surgical manipulations in such cases must be considered.

Keywords: calculi, diverticulum, urethra, urolithiasis

1 Introduction

Lower urinary tract calculi are observed in the bladder, prostate, and urethra. The urethra is an infrequent location, accounting for no more than 0.3% of urinary calculi disease cases [1]. Approximately 88% of urethral stones are localized to the posterior urethra [2]. The frequency of urethral stones varies with geographical location: in Western countries this disease is diagnosed only occasionally, whereas it is endemic in the Middle East and Asia [1]. In developing countries, urethral calculi typically consist of struvite and uric acid, whereas in industrialized societies calcium oxalate, and cystine are dominant [2]. Two peaks of incidence have been reported for urethral calculi: the first occurs in early childhood, but recently the number of pediatric urolithiasis cases has decreased [1-3]; the second peak is in approximately the fourth decade of life [1]. The infrequency of urethral calculi in the second and third decades of life is potentially caused by higher urinary peak flow rates in this age range [4]. The flow rate for this group reaches 35 ml/s and decreases to 15 ml/s in the fourth decade of life. The relatively longer urethral length in males at those ages is the reason that urethral stones are more commonly reported in that age group as compared with younger males [5]. Stones in the urethra are usually small in size; however, very large stones are occasionally diagnosed that present a challenge for the treatment process.

2 Case report

A 54-year-old Caucasian man presented at the Emergency Department of our institution with symptoms of suprapubic pain and leakage of urine from the cutaneous fistula. His medical history stated that as a 7-year-old boy he had an accident involving construction equipment and underwent surgery for numerous fractures of the pelvis, pelvic exenteration, and rupture of the bladder. Several years after the urinary tract reconstruction, the patient developed a bladder stone that was removed by open surgery. Following that operation, the patient experienced urinary retention, and a Foley catheter was inserted. Subsequently, because of urethral stricture, the patient was treated at a tertiary referral center for urological surgery, where urethral reconstruction was performed.

Since the last operation, the patient had noticed spontaneous expulsion of a few stones. On admission, the patient's temperature was normal. The abdomen was soft and non-tender. Goldflam's sign was negative bilaterally. Palpation of the perineum failed to reveal any pathology. Examination of the scrotum and perineum revealed a fistula with urine discharge, and a large, solid mass was noticeable under the skin of the perineum. The mass was localized to the base of the scrotum in the anterior part of

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the perineum. The patient stated that the mass had existed for 15 years. The prostate was not identified on digital rectal examination. Ultrasonography indicated that the kidneys were of the correct shape, size, and localization. Urinalysis showed 10 white blood cells per high-powered field. X-ray visualization of the pelvis was ordered because of suspicion of a stone in the urethra. Radiologic evaluation confirmed the diagnosis (Figure 1), and the patient was admitted to the urology department and offered surgical treatment.

During the exploratory procedure, a primary incision was made near the fistula and above the palpable mass on the peritoneum. After the urethra with diverticulum was exposed and external incision of the urethra was performed, one large stone and several smaller stones were removed (Figure 2). The largest stone measured 75 mm high and 65 mm wide (Figure 3). Subsequently, a new fistula between the urethra and skin was created in place of the previous fistula. The diverticulum was left open because of the local inflammatory process and lack of possibility of catheterization resulting from stricture of the urethra distal to the diverticulum. Repair of the urethra was planned for a second stage.

After six months, when the wound-healing process was complete, the patient was readmitted for reconstruction of the urethra. Before this operation, a retrograde urethrogram was ordered; it confirmed a critical stricture of the urethra distal to the diverticulum (Figure 4). In the first step of reconstruction, the stenotic part of the urethra, measuring 15 mm, was resected, and then end-to-end anastomosis of the urethra was performed. During subsequent urethroscopy, a giant diverticulum in the bulbous urethra was found. The volume of the diverticulum was

Figure 1: X-ray examination of the pelvis showing a calcareous giant shadow in the view of the urethra suggestive of a stone.

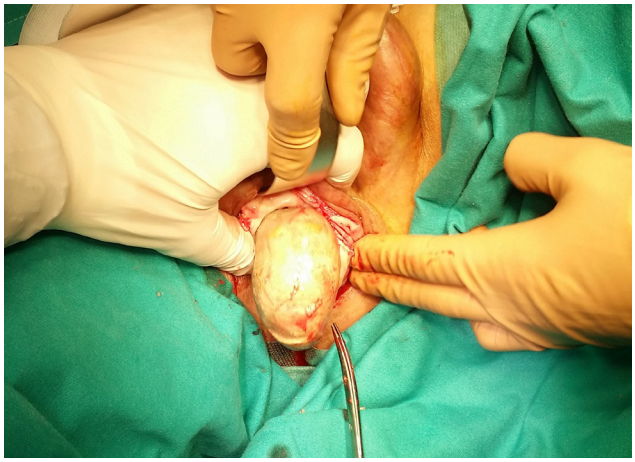

Figure 2: Final stage of the operation.

Figure 3: The calculi removed from the urethra.

Figure 4: Retrograde urethrogram showing critical stricture of the urethra distally from the diverticulum, with fistula to the skin of the perineum.

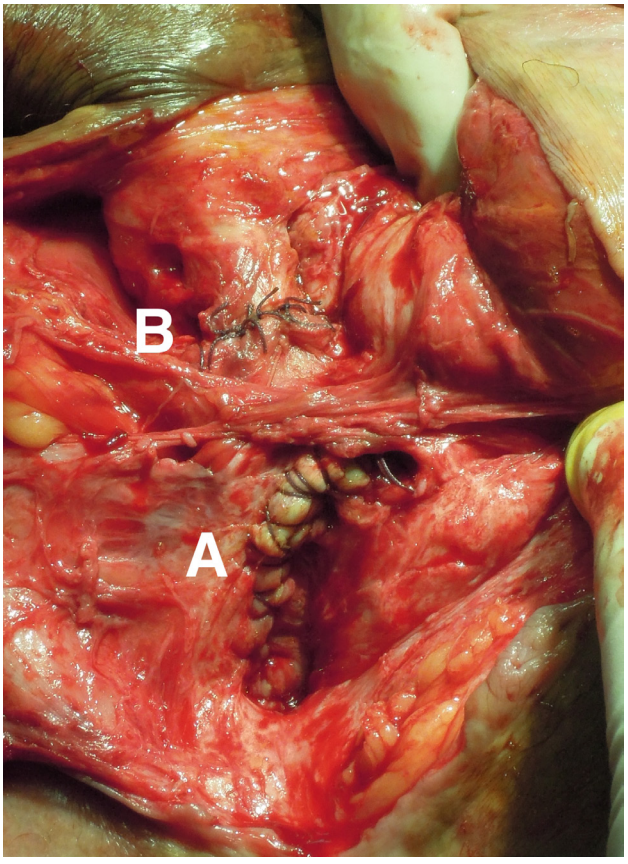

Figure 5: A: Sutured urethra after excision of the diverticulum; B: End-to-end anastomosis after excision stricture of the urethra distal to the diverticulum..

approximately 70 ml, and it communicated with the skin of the perineum. Finally the diverticulum was dissected, much reduced, and completely closed (Figure 5). Continuity of the urethra was restored. A sterile 20-French Foley catheter was inserted.

Ethical approval: The research related to human use has been complied with all the relevant national regulations, institutional policies and in accordance the tenets of the Helsinki Declaration, and has been approved by the authors' institutional review board or equivalent committee.

Informed consent: Informed consent has been obtained from all individuals included in this study.

3 Discussion

Large urethral stones are rarely diagnosed. In the literature, a limited number of cases have been described, which typically led to clinical dilemmas. In most of those situations, typical minimally invasive endoscopic treatment, such as forceps or basket extraction or endoscopic push-back with lithotripsy, is not possible. In general, open surgery is recommended, but some exceptions have been described. For instance, Demir et al. successfully treated a patient with a stone in the prostatic urethra measuring 65x70x60 mm by endoscopic procedures such as laser and pneumatic lithotripter [5]. In our case, the stone was localized to the anterior urethra, and therefore a straightforward open approach was performed. Additional a recurrent, critical stricture of urethra required two-stage treatment with open urethroplasty. Thereby, complete urinary tract reconstruction was obtained. A similar technique was used by Lubana et al., who described a case of a large urethral stone removed by urethrotomy [6]. Prabhuswamy et al. reported a dumbbell-shaped vesico-prostatic urethral stone measuring 102x35x45 mm that was removed by an open transvesical approach [7].

Another problem in stone removal is large stone occurrence in the diverticulum, where open surgery with incision of the diverticulum is usually required. However, an exception for this situation has been also reported: Susco et al. described a female patient with large stone localized to a urethral diverticulum treated successfully by litholapaxy [8]. The shorter length of the female urethra affords easier access to stones in women.

Large urethral stones are not observed only in adult patients. In a very unusual case reported by Rivilla et al., a 6-year-old girl presented with a urethral stone 58´25´21 mm in size. In this immature patient, a suprapubic approach with a small bladder incision was chosen [9]. In our case, reconstructive treatment of the urethral stricture was required. This case emphasized that a rare pathology, such as a stone localized to the urethra, can coexist with other pathologies of the urinary tract. This possibility should be taken into account in the early phase of diagnosis and further in the treatment planning process.

4 Conclusion

Large urethral stones are atypical situations that frequently coexist with other pathology of the urinary tract. The choice of a surgical technique is not always obvious. In daily practice, every case should be discussed, and the most appropriate treatment performed.

Conflict of Interest: The authors declare that they have no conflict of interests.

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