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ORIGINAL PAPER

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Outcomes of urethral calculi patients in an endemic region and an undiagnosed primary fossa navicularis calculus

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Abstract Urethral calculus is a rare form of urolithiasis with an incidence lower than 0.3%. We determined the outcomes of 15 patients with urethral stone, of which 8 were pediatric, including an undiagnosed primary fossa navicularis calculus. Fifteen consecutive male patients, of whom eight were children, with urethral calculi were assessed between 2000 and 2005 with a mean of 19 months' follow-up. All stones were fusiform in shape and solitary. Acute urinary retention, interrupted or weak stream, pain (penile, urethral, perineal) and gross hematuria were the main presenting symptoms in 7 (46.7%), 4 (26.7%), 3 (20%) and 1 (6.6%) patient, respectively. Six of them had accompanying urethral pathologies such as stenosis (primary or with hypospadias) and diverticulum. Two patients were associated with upper urinary tract calculi but none of them secondary to bladder calculi. A 50-year-old patient with a primary urethral stone disease had urethral meatal stenosis accompanied by lifelong lower urinary tract symptoms. Unlike the past reports, urethral stones secondary to bladder calculi were decreasing, especially in the pediatric population. However, the pediatric patients in their first decade are still under risk secondary to the upper urinary tract calculi or the primary ones.

Keywords Urethral stones · Primary · Outcome · Treatment

Introduction

Urethral calculus is a rare urological phenomenon in the Western world, especially in North America; however, the Middle East and Asia have relatively high incidences [1–3]. Turkey, especially its southeast, is also regarded as an endemic region. Urethral calculi are estimated to represent 0.3% of all urinary stone diseases [3]. Owing to the high incidence of bladder calculi, in the literature urethral calculi have been generally reported from developing countries especially among childhood populations; however, the incidence of pediatric urolithiasis is now thought to be decreasing [3]. In a recent, large prospective urethral calculi series of Kamal et al. [4], pediatric urethral calculi were only 2% and upper urinary stone disease now became the main source, other than bladder calculi, depending on the compositions of the stones. In developing countries, while the stone composition of most urethral stones was reported as struvite and uric acid, calcium oxalate and cystine were the most common results of stone analyses in the developed communities [1]. In this study we discussed the outcomes of 15 patients with urethral stone, of which 8 were pediatric—including a 50-year-old patient with an undiagnosed primary fossa navicularis (FN) calculus—who were admitted to our institute over a period of 5 years.

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Patients and methods

Between February 2000 and February 2005, patients admitted to the Urology and Pediatric Surgery departments of our hospital were assessed in a retrospective study. Their family details and full medical history were evaluated. A urogenital physical examination, including palpation of the penis and perineum, and digital rectal examination were performed. All patients were evaluated with X-rays of the abdomen and pelvis. Urethroscopy was used to confirm the diagnosis of all patients except for the four in whom the stones were located in the FN. The number, size, shape and location of associated urinary tract calculi were reported. The presence of associated urinary tract calculi and congenital anomalies were evaluated. The size of these calculi was measured on plain X-rays and directly after the extraction of the stones in some. From noninvasive to invasive, milking, manual or endoscopic forceps extractions, endoscopic push-back, electrohydraulic lithotripsy

(EHL), and open surgery (meatotomy, cystolitotomy) were the selected approaches. Milking was done by the application of 2% lidocain jelly preparations. Pushed-back and additional procedures such as EHL and cystholitotomy were reserved for the posterior urethral calculi. Stone analyses were collected in 12 patients. Periprocedure urinary microbiologic examinations were done for all patients and posttreatment control was carried out for three.

Results

Among 15 male patients (mean age 26.2 ± SD 24, 76, range 4–68), 8 were children between the age of 4 and 10. The mean follow-up time was 19 ± SD 19, 13 (range 1–60) months. Acute urinary retention, interrupted or weak stream, pain (penile, urethral, perineal) and gross hematuria were the main presenting symptoms in 7 (46.7%), 4 (26.7%), 3 (20%) and 1 (6.6%) patient, respectively. Seven patients who suffered from acute urinary retention were admitted to the emergency department and four (57.1%) of them had posterior urethral calculi. Any patient who reported previous stone disease and family history was negative for calculi. All 9 anterior urethral (penile + FN) stones were palpable. The location of the stone was represented as being posterior, penile urethra and FN for 6 (40%), 5 (33.3%) and 4 (26.7%) patients, respectively. According to the location (posterior, penile urethra and FN) of the calculi, the mean (range) width of the stones was 8 (5–10), 8 (4–10) and 6 (5–8) mm, respectively. All stones were fusiform in shape and solitary. As for the accompanying urethral pathologies, primary stricture of urethral meatus, urethral meatus stenosis due to the coronal hypospadias, urethral diverticulum and benign prostate hyperplasia (BPH) were present in 2, 2, 1 and 1 patient, respectively. Two of the patients were supposed to be associated with primary urethral stone disease, accompanied with mild urethral meatal stenosis and a long period of lower urinary tract symptoms (LUTS), and urethral diverticulum. Two patients were associated with upper urinary tract calculi. One patient was diagnosed with three urologic diseases combined—bladder tumor, BPH and posterior urethral calculi—while his gross hematuria was being evaluated. All the patients were diagnosed using uroradiography alone. Urethroscopy had been used for confirmation for all patients except for the four patients in whom the stones were located in the FN.

Among the six posterior urethral calculi, four stones were relieved by endoscopic push-back + EHL (66%), including the one patient who underwent TUR of tumor during the same session and two patients who underwent the endoscopic push-back + open cystolithotomy procedure due to failed EHL (33%).

Only one patient with a midpenile urethra stone, among the nine anterior urethral [penile (5) + FN (4)] stones, was successfully managed by milking + forceps extractions. Endoscopic procedures such as in situ lithotripsy with EHL and basket were the selected stone extraction ways for penile stones in 2 and 1 patient, respectively. Open stone extraction and tubularized incised plate urethroplasty (TIPU) were performed during the same session for the one remaining penile stones group with subcoronal hypospadias in an elective condition whose symptom was penile pain.

All four patients with stones in FN underwent meatotomy and one had coronal hypospadias (classified under the FN group); however, the repair of the hypospadias was left to be done at another session because he was suffering from acute retention of urine and urethral edema was thought to be possible.

The component of the stones in all our patients (12) who underwent the stone analyses was reported as calcium oxalate. No stone component data were available for three patients. Urinary tract infection with *Escherichia coli* more than 10^5 cfu occurred in three patients

Table 1 The summary of the results of the urethral stones

No.	Age	Width (mm)	Location	Symptom	Accompanying pathology	Treatment
1	4	5	Fossa navicularis	Pain		Meatotomy
2	5	5	Fossa navicularis	Acute retention	Coronal hypospadias	Meatotomy
3	5	10	Penile urethra	Pain	Subcoronal hypospadias	Urethraplasty
4	5	10	Penile urethra	Acute retention		EHL
5	5	5	Posterior urethra	Pain		PB + EHL
6	5	6	Penile urethra	Weak stream	Renal calculi	Milking
7	7	10	Posterior urethra	Acute retention		PB + cystolithotomy
8	10	5	Posterior urethra	Acute retention		PB + EHL
9	34	10	Posterior urethra	Acute retention		PB + EHL
10	35	10	Penile urethra	Weak stream		Basket
11	35	6	Fossa navicularis	Weak stream	Meatal stenosis	Meatotomy
12	49	4	Penile urethra	Weak stream	Diverticulum	EHL
13	50	8	Fossa navicularis	Acute retention	Meatal stenosis	Meatotomy
14	66	9	Posterior urethra	Acute retention	Renal calculi	PB + cystolithotomy
15	68	10	Posterior urethra	Hematuria	Bladder tumor + BPH	PB + EHL

Patients were arranged in an order with increasing age
PB push-back, *EHL* electrohydrolic lithotripsy

and after a 5-day cephalosporin/flouroqinolone treatment, all three patients were relieved by posttreatment microbiologic control. Results are summarized in Table 1.

Discussion

Owing to anatomic reasons, parallel with the previous studies, no female patient with urethral calculi was reported in the present study [1, 4]. In the early studies, pediatric urethral stones secondary to bladder stones occupied a high incidence among urethral calculi in endemic regions. The reason for this is that most bladder stones were diagnosed among children of developing countries and the nutritional habits of those regions were mostly dominant with cereal foods [1]. However, a recent, large retrospective study has confirmed the reduction of the bladder stones that were the main source of the urethral stones and hoped that bladder and urethral stones may be totally eradicated in the near future [3]. Furthermore, in their large prospective urethral stone study with 51 patients, Kamal et al. [4] reported only one child of 6 years of age (2%). Unfortunately, unlike these studies, we still had quite a number of children (53.3%) in their first decade, with urethral stones but, interestingly, not secondary to bladder calculi. We believe that nutritional habit is not the only explanation of this high pediatric rate because dietary habits are also changing from cereal foods to protein-enriched ones in our territory owing to the industrialized type of life and economic development. On the other hand, urolithiasis was supposed to be a disease with polygenic defect with partial penetrance [1]. However, due to the lack of family history in any of our patients, we concluded that the genetic theory also could not explain the high pediatric rate in our urethral stone study. We had no patient between 10 and 34; thus, we believed that people in their second and third decades had a low risk of urethral stones. Kamal et al. [4] reported the rates as being 0 and 18% in these decades, respectively. Depending on the knowledge that the maximum flow rate decreases from 35 to 15–20 ml/s in men through 14–50 years, the power of the urinary stream that can propel the stone may be responsible for these low rates at this period [5].

Every stone in this study was radio-opaque and X-ray was adequate to confirm the diagnosis. Despite the knowledge of early studies that the uroradiographic diagnosis could be possible in only 40% of cases, the more recent series confirmed our results with a rate of 98% [4]. Ultrasonography as a noninvasive screening technique may be useful, especially for the non-opaque posterior urethral calculi [6]. The dominance of radio-opaque stones in the recent literature was also further evidence that today's urethral stones originate from the upper urinary tract unlike their similar forms in the past that were mostly composed of struvite or uric acid secondary to the bladder stones. While two of our patients had associated with lower pole renal calculi; any patient had bladder calculi. In addition, the shape of all the stones was fusiform and solitary. Nevertheless these were all evidence that they were the primary or associated with the upper urinary tract disease other than secondary to the bladder calculi. Similarly, Kamal et al. [4] evaluated the stone compositions of their patients, and concluded that most of the urethral stones should come from the upper urinary tract due to the fact that the struvite and uric acid stones were reported to originate mostly from the bladder, whereas calcium oxalate and cystine stones originate mostly from kidneys. All our 12 patients who had the stone analyses had calcium oxalate stones.

We strongly believed that one of our patients (50 years old, no.13, Table 1) was associated with a primary FN calculus (calcium oxalate) that was skipped off in medical controls because of the following reasons: due to his lifelong LUTS, no evidence of urinary calculus disease, including no report of colic episode in his medical history and the shape of the calculi that appeared just as the original shape of the FN (Fig. 1). All of these were strong pieces of evidence of its diagnosis as primary. Nevertheless, a small calculus in the early childhood with no clinical symptoms that descended from upper urinary tract (antegrade) or a small foreign body (retrograde) that then grew in the shape of FN should be taken into account in the stone occurrence hypothesis in this case. In his previous hospital admissions, we believed that the diagnosis had always failed due to the improper physical examinations and plain X-ray that was just ended caudally at the symphysis pubis level. Long duration of the LUTS gave strong evidence that the beginning of the event was in his childhood. Then, after many years, LUTS ended with acute urinary retention by the growing of the stone as in the shape of the FN. Thus primary urethral stones should be considered as a possible etiology of LUTS even in lifelong

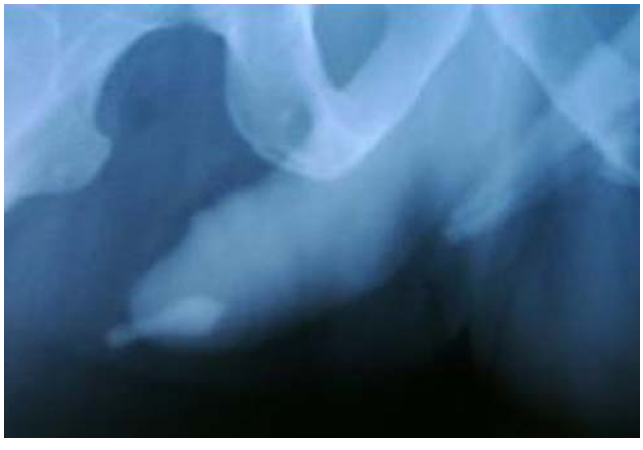

Fig. 1 X-ray appearance of an undiagnosed primary fossa navicularis calculus as in the original shape of the fossa navicularis in a 50-year-old patient who was suffering from lifelong lower urinary tract symptoms (LUTS)

ones. Mild meatal stenosis was the possible reason for the stone in this patient. Urethral stricture and the diverticulum were the main reasons for the primary cases and two female giant primary urethral calculi cases in the diverticulum were also reported [7, 8]. In the literature there were some other extremely rare causes of primary urethral stones such as Munchausen syndrome and safety pin as a foreign body [9, 10]. Unlike these extreme cases that were reported, except for the mild meatal stenosis, there was no associated pathology in our primary FN calculus and it was unique among the literature due to its occult clinical nature and the shape that just fits FN.

With regard to the anatomic location, such as the posterior, penile urethra, and FN, the rate of the stones was equally distributed (27–40%). Bedii [11] supported our results in his study with 60 patients. However, posterior urethra was the commonest location in the other large series with about 50 subjects [4, 12]. Nevertheless, we believed that the urethral stones can be entrapped anywhere in the male urethra, but the ones located in the posterior caused acute urinary retention more frequently (57%).

Treatment of the urethral stones mostly depends on the anatomic location and the width of the stone and on the associated anatomical pathology of the urethra. Milking, forceps extractions (manual or endoscopic) or basket, endoscopic push-back, EHL and surgery (internal urethrotomy, meatotomy, percutaneous and open cystolitotomy) are the treatment options. Meatotomy, EHL and push-back + (EHL) seem to be the golden standard for the FN, penile urethra and posterior stones, respectively. Laser should also be taken into account as being a safe procedure during lithotripsy for the urethral stones [13]. Push-back + EHL success was 66% in our patients with posterior urethra calculi and this is relatively lower than the rate of 86% reported in the study of Kamal et al. [4]. We used cystolithotomy for the extraction of the failed ones; however, a percutaneous approach as a noninvasive procedure would surely be advisable. Milking with lidocain jelly as a conservative procedure was not so successful for our FN and penile urethral stones, except in one case (6.7%). While Sherif [14] reported a success rate of 78% for the same procedure, Kamal et al. [4] consented with our results with a rate of 13%. An open surgical procedure to extract the urethral calculi was used for only one elective patient of 5 years of age with subcoronal hypospadias and no fistula was observed in 34 months' follow-up. Thus, we believed that the stone extraction and hypospadias repair can be performed at the same session especially for patients who have no acute urinary retention.

Summing up, from past to present, depending on our results and the recent literature, the incidence of urethral stones secondary to bladder calculi is decreasing. Upper urinary tract calculi and the primary ones now became the main reason for the urethral stones. On the contrary, unlike in the recent literature, we found out that pediatric patients especially the ones in their first decade are still under the risk for urethral stones and that calcium oxalate was the main stone component. Owing to the development of the condition of nutrition, although the bladder stones and urethral stones related with bladder calculi in pediatric population are supposed to be eradicated in endemic regions in the developing countries in the near future by some authors, we think that urethral stones secondary to upper urinary system and the primary ones still maintain their importance in the pediatric urological practice.

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