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A Rare Case Report of Infertility Due to a Giant Hair-bearing Urethral Stone in a Urethral Diverticulum

Üretral Divertikülde Saçlı Dev Üretral Taşa Bağlı Nadir Bir İnfertilite Olgu Sunumu

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Abstract

Urethral stones are rare, accounting for less than 1% of all urinary system stones. These stones may present with obstructive symptoms or remain asymptomatic. Hair-bearing urethral diverticula, which can form after surgical interventions such as hypospadias repair, are an uncommon cause of urethral stone formation. However, urethral stones leading to infertility are extremely rare.

A 38-year-old male patient presented to the urology outpatient clinic with complaints of infertility. Physical examination revealed a palpable mass in the penoscrotal region. Further evaluations and imaging identified this mass as a urethral stone within a hair-bearing urethral diverticulum. The patient's history revealed a childhood hypospadias repair. Open surgery was performed for stone removal and diverticulectomy. Semen analysis at the six-month postoperative follow-up showed an improvement in semen volume from 1 ml preoperatively to 2.5 ml, reaching normal levels. Additionally, nine months after the procedure, the patient's spouse was confirmed to be pregnant. Long-term follow-up revealed no postoperative complications.

This case highlights the importance of considering urethral pathologies in infertile patients with a history of urethral surgery. Such conditions can be effectively treated with open surgery, potentially restoring fertility.

Keywords: urethral stone, urethral diverticulum, hairy urethra, case report

Özet

Üretra taşları, üriner sistem taşlarının %1'inden azını oluşturan nadir taşlardır. Bu taşlar obstrüktif semptomlarla ortaya çıkabileceği gibi asemptomatik de kalabilir. Hipospadias onarımı gibi cerrahi müdahaleler sonrasında oluşan saçlı üretra divertikülleri üretra taşı oluşumuna neden olabilecek nadir sebeplerdendir. Üretra taşlarının infertiliteye neden olması ise oldukça nadirdir.

İnfertilite şikayeti ile üroloji polikliniğine başvuran 38 yaşında erkek hastanın muayenesinde penoskrotal bölgede ele gelen oluşum tespit edildi. İleri değerlendirmeler ve görüntülemerle bu oluşumun üretra divertikülü içindeki saçlı üretra taşından kaynaklandığı tespit edildi. Hastanın hikayesinden çocukluk döneminde hipospadias onarımı operasyonu geçirdiği anlaşıldı. Taşın çıkarılması ve divertikülün eksizyonu için açık cerrahi yapıldı. Operasyon öncesi 1 ml olan semen hacminin, operasyon sonrası altıncı ay takibinde yapılan semen analizinde normale döndüğü (2,5 ml) görüldü. Operasyondan dokuz ay sonra ise hastanın eşinin gebe kaldığı öğrenildi. Uzun dönem takipte hastada herhangi bir komplikasyon görülmedi.

Bu olgu, üretral cerrahi öyküsü olan infertil hastalarda üretral patolojilerin dikkate alınmasının önemini vurgulamaktadır. Bu durum açık cerrahi ile etkili bir şekilde tedavi edebilir ve potansiyel olarak fertiliteyi geri kazandırabilir.

Anahtar kelimeler: üretra taşı, üretra divertikülü, saçlı üretra, olgu sunumu

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Introduction

Urethra is a rare location for urinary system stones, accounting for less than 1% of all cases [1]. The majority of urethral stones are found in the posterior urethra [2]. These stones can be asymptomatic, but they may also present with obstructive symptoms, recurrent urinary tract infections, and even acute renal failure [3, 4]. One of the rare causes of urethral stones is urethral diverticula, which can occur as long-term complications after hypospadias repair [4]. While hypospadias surgery is the most common cause of acquired diverticulum in children, it is not the leading cause in adults [5]. Diverticula that develop following hypospadias repair, especially when flaps from the scrotum or penile skin are used, may contain hair-bearing urethra. The stones found in these hair-bearing urethral diverticula can grow to very large sizes and may be asymptomatic. In the existing literature, there have been no reports of urethral stones causing infertility. Additionally, only one documented case of infertility caused by a urethral diverticulum has been reported, and that involved a congenital diverticulum. Our case, involving a secondary diverticulum with a giant urethral stone leading to infertility, presents a unique scenario that has not been previously described. To our knowledge, this is the first case report of urethral stone in a urethral diverticulum causing infertility. We present the following case in accordance with the CARE reporting checklist.

Case

A 38-year-old male patient presented to the clinic with complaints of infertility. He reported that despite engaging in unprotected intercourse for two years, he had not been able to have a child. The patient's medical history revealed that he had undergone right orchiopexy and hypospadias repair surgery at the age of five. Physical examination revealed a subglandular urethral meatus and a significant reduction in the volume of the right testis. Additionally, a palpable formation of approximately 4 cm was detected in the midline at the junction of the scrotum and the penile shaft. Upon further questioning, the patient indicated that this mass had been present for years, but he had never sought medical attention for it. Aside from post-micturition dribbling, the patient reported no other lower urinary tract symptoms. Semen analysis revealed low sperm volume (1 ml). Ultrasound targeting the midline formation revealed a calcified lesion approximately 3 cm in diameter, located extratesticularly near the base of the penis, casting a posterior shadow. Magnetic resonance imaging at the level of the penile root showed a well-defined lesion measuring 30x36 mm, hypointense on T1 and T2-weighted images, non-enhancing on post-contrast sequences, and hypointense on fat-suppressed sequences. Retrograde urethrography revealed a well-defined opacity of approximately 4 cm at the level of the penile root, with contrast material surrounding the opacity **(Figure 1a).** Urethroscopy confirmed the presence of a hairy urethral diverticulum with a stone inside **(Figure 1b).** However, no urethral stricture was observed distally to the diverticulum. It was understood that the patient had undergone hypospadias repair at the age of five using an onlay flap from scrotal skin. Open surgery was planned to remove the stone and repair the diverticulum. The operation began with a vertical incision at the penoscrotal junction. The diverticulum was accessed, and the stone, covered with hair, was removed through a vertical incision **(Figure 1c).** The excess tissue in the diverticulum was excised, and the urethral lumen was restored to its normal calibration **(Figure 1d).** The remaining hair follicles in the urethral tissue were ablated using a holmium laser. The urethra was closed in a single layer over a 20 fr urethral catheter using 4-0 Vicryl sutures **(Figure 1e).** The urethral catheter was removed on the

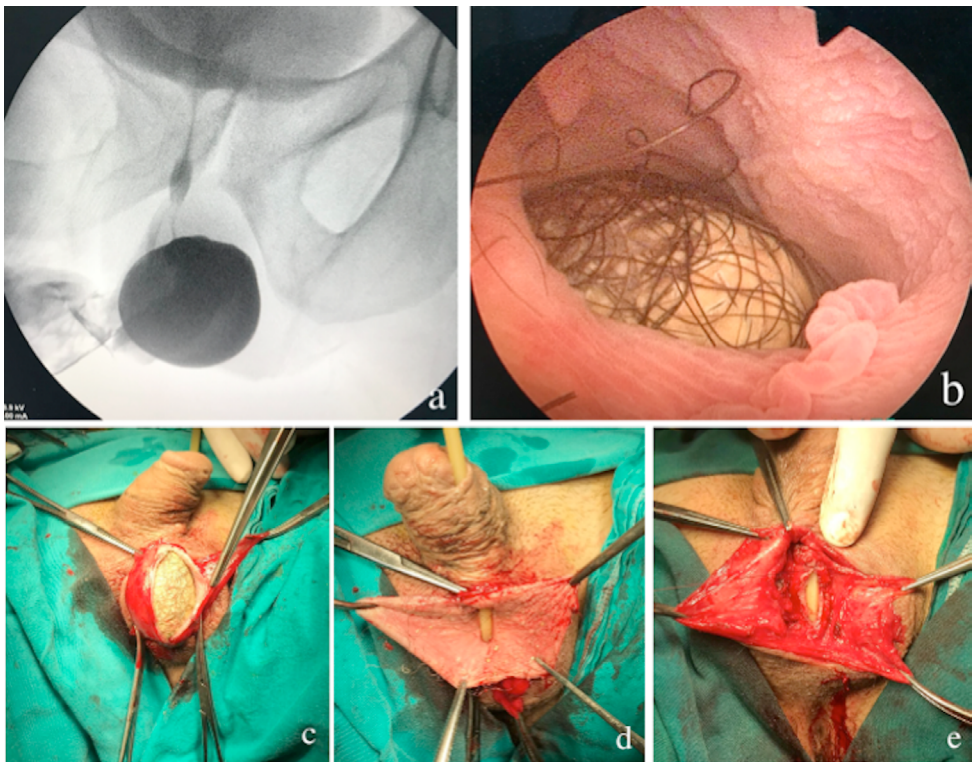

Figure 1. (a): retrograde urethrogram showing a urethral diverticulum and an intradiverticular stone (b): urethroscopy image displaying the hair-bearing urethral stone (c): urethral diverticulum containing the stone in open surgery (d): view of the urethral diverticulum before excision of the excess tissue (e): image after excision of excess tissue, showing the urethra restored to normal calibration

21st postoperative day. After the catheter removal, the patient was instructed to support the surgical site with his finger during urination for three months. At the 6-month follow-up, the patient's semen analysis showed normal sperm parameters and a semen volume of 2.5 ml. The patient experienced no lower urinary tract symptoms, and by the 9th month post-surgery, his wife had become pregnant, indicating the restoration of fertility. Four years after the operation, the patient continues to remain free of any urinary symptoms.

Discussion

The urethra is the least common region for stones within the urinary system. Due to its longer length and anatomical narrowings, urethral stones are more often seen in males. Most urethral stones originate from the kidneys and bladder; however, they can also develop secondary to urethral pathologies like strictures and diverticula [1, 2, 4]. Since the urethra is the terminal part of the urinary system, stones in this region often cause symptomatic obstruction, making large urethral stones relatively rare. Urethral stones generally present with lower urinary tract symptoms such as obstructive complaints, hematuria, recurrent urinary tract infections, and post-void dribbling. However, in cases of stones associated with hairy urethral diverticulum, the situation may differ; these stones can grow significantly over time while remaining asymptomatic. The asymptomatic nature of these stones may be due to the gradual development over the years, leading patients to perceive the minor changes as normal.

Male urethral diverticula are quite rare, with 90% being secondary diverticula [6]. Both primary and secondary types are most commonly found in the penoscrotal region, as in our case [7]. Secondary urethral diverticula are mainly caused by factors that increase intraurethral pressure and lead to fibrosis, scar formation, and necrosis, such as previous surgeries, strictures, infections, and trauma [8]. One significant type of surgery that may result in diverticula is hypospadias repair, where 10–15% of cases develop diverticula as complications, and up to 8% present with a hairy urethra when skin flaps are used [9,10]. In hairy urethral diverticula, stones can grow significantly without causing symptoms [4]. Turbulent flow within the diverticulum leading to stasis and hair within the diverticulum acting as a nidus might play a role in stone development [5].

In the literature, there is only one reported case of a urethral diverticulum causing infertility, and in that case, the diverticulum was congenital [11]. What makes our case even more unique is that the diverticulum in our patient is secondary and, despite containing a giant stone, did not cause any lower urinary tract symptoms that would prompt a urological consultation. Instead, it presented solely as infertility, making this case highly unusual and noteworthy. It was thought that during the expulsion phase of ejaculation, the entire ejaculate could not pass through the diverticulum and the associated stone. The absence of a urethral stricture and an increase in ejaculate volume from 1 ml pre-treatment to 2.5 ml post-treatment supports this hypothesis.

The treatment of urethral stones depends on their size, shape, location, and underlying cause. Small urethral stones are mostly treated using minimally invasive methods like milking, forceps extraction, urethral lithotripsy, or push-back with lithotripsy in the bladder. However, in the case of a urethral diverticulum and associated hairy urethral stone, the treatment becomes more complex. These stones can grow to significant sizes, making endoscopic treatment insufficient. Xie et al. demonstrated successful treatment in 16 patients with hairy urethral stones secondary to hypospadias repair. Their approach included open surgery for stone removal, excision of the excess diverticular tissue, laser epilation of the remaining hairy urethral area, and repair with a buccal mucosal graft if a stricture was present. Additionally, to prevent postoperative fistula formation at the surgical site, they used a technique where the skin incision was made lateral to the stone while the diverticulum containing the stone was incised at the midline [4]. Similarly, we performed an open repair in our patient. However, since there was no stricture, we did not use a buccal mucosal graft, and both the skin incision and diverticulum incision were made at the midline. Moreover, no fistula development was observed in our patient at the 4-year postoperative follow-up.

Hairy urethral diverticula with stone formation is a rare condition, particularly following hypospadias repair. Our case highlights that such stones can be asymptomatic yet cause complications like infertility. Due to the size and complexity of these stones, open surgical intervention is often required. In our patient, successful treatment without postoperative complications, such as fistula formation, was achieved with a tailored surgical approach. This case underscores the importance of considering urethral stones in patients with a history of urethral surgery and atypical symptoms, and it demonstrates the potential for positive outcomes with individualized treatment and follow-up.

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