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Hair-bearing epithelium serving as a nidus for intravesical stone formation in a male who underwent exstrophy repair: A case report

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ABSTRACT

Bladder calculi are rare in children; they are typically associated with congenital abnormalities of the bladder, foreign bodies, intestinal mucosa, or infection. There is a high risk of bladder calculi with bladder exstrophy, a rare congenital condition. Primary bladder closure is usually performed in the first days of life, but subsequent bladder calculi are associated with about 15% of bladder exstrophy repairs. We report a case of hair-bearing epithelium serving as a nidus for intravesical stone formation in a 24-year-old male who underwent exstrophy repair as an infant. Our case emphasizes need for close and continued follow-up in patients who receive bladder exstrophy repair as infants. This is the first report in the literature of hair-bearing epithelium serving as the nidus for bladder stones in this unique patient population.

Key Words: Bladder exstrophy; exstrophy–epispadias complex; bladder stone; hair-bearing epithelium.

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Introduction

Bladder exstrophy occurs at a rate of approximately 1: 10,000 to 1: 50,000 live births. Today, bladder exstrophy is treated through two main approaches: modern staged repair of exstrophy (MSRE) and complete restoration of exstrophin (CPRE) [1,2]. The post-operative period is complicated by numerous and variable events such as infection, dehiscence, upper tract dilatation with deterioration, fistulas, stone formation and incontinence [3]. Here, we presented a case of hair-bearing epithelium that served as a

nidus for intravesical stone formation in a male patient who underwent exstrophy repair. Additionally, this case is the first reported incidence of a bladder calculus resulting from hair-bearing epithelium in the scar tissue.

Case report

A 24-year-old male with past history of successful bladder exstrophy repair as an infant was referred to our clinic from an outside hospital with recurrent bladder stones throughout adolescence, starting at age 16. He reported the stones appeared linear and “attached to hairs” [Fig. 1]. Two elliptical shaped stones attached to a hair). Some stones were spontaneously voided per urethra, and others were removed endoscopically. He had undergone three cystolitholopaxies in the past year for bladder stones. He had no other past medical history and was not taking any medications.

Physical exam was unremarkable and revealed a well-healed scar consistent with previous exstrophy repair and normal external male genitalia.



Fig. 1. Two elliptical shaped stones attached to a hair that was voided spontaneously by the patient.

He had been treated for urinary tract infections in the past but his urine and stone cultures were negative for infection at the time of presentation. Stone analysis revealed 20% magnesium ammonium phosphate, 50% calcium phosphate carbonate, and 25% calcium phosphate dehydrate. Computed tomography (CT) showed a linear shaped stone in the bladder without evidence of any renal stones [Fig. 2. Axial slice of a pelvic CT showing a linear shaped calcification in the bladder].



Fig. 2. Axial slice of a pelvic CT showing a linear shaped calcification in the bladder.

Upon endoscopic evaluation, the bladder neck was very narrow and a rigid 22-French cystoscope could not traverse the bladder neck. His urethra

was scarred and tortuous from surgery as a child, but patent, and a flexible 16-French cystoscope was able to be passed through the urethra visualize the bladder. This revealed a well-demarcated scar consistent with a history of bladder exstrophy repair as well as hairs protruding from the midline scar tissue on the anterior bladder wall [Fig. 3. Endoscopic view of a hair protruding from the anterior bladder wall along a midline scar].

The hair follicles and a keratinized line on the anterior, midline bladder wall were ablated using a 365-micron holmium laser fiber [Figure 4. Endoscopic view of laser fiber ablating the keratinized line on the anterior bladder wall]. The patient tolerated the procedure well and upon follow-up cystoscopy three months later there was no evidence of recurrent hairs or stones.



Fig. 3. Endoscopic view of a hair protruding from the anterior bladder wall along a midline scar.



Fig. 4. Endoscopic view of laser fiber ablating the keratinized line on the anterior bladder wall.

Discussion

Bladder exstrophy-epispadias complex is a rare congenital anomaly presenting with protrusion of the urinary bladder through an abdominal wall defect. It occurs in approximately 1 in 35,000 live births with a male predominance of three to one [4]. Repair of bladder exstrophy is often performed during the first 48 hours of life. Surgical repair involves dissecting free the exposed urothelium from the mucocutaneous junction and then approximating the freed urothelial edges to close the bladder [5]. Subsequent bladder calculi in such patients occur at a rate of approximately 15% and are associated with infection, foreign bodies, and urinary stasis [6]. While there are several reported cases in the literature of iatrogenic bladder calculi due to surgical sutures [7] or hair nidus introduced during clean intermittent catheterization [8], this case is the first reported incidence of a bladder calculus resulting from hair-bearing epithelium in the scar tissue. The location of the hair follicles on the anterior bladder wall and occurrence after puberty suggest that the origin was the skin of the suprapubic abdominal wall that was somehow incorporated into the exstrophy repair.

The goals of reconstruction for the epispadias-exstrophy complex include preserving kidney function, creating urinary continence, decreasing urinary tract infections, and creating functionally and cosmetically acceptable external genitalia [7]. Repair of bladder exstrophy begins with closure of the bladder and abdominal wall by either the modern staged repair of exstrophy (MSRE) or complete primary repair of bladder exstrophy (CPRE) [1,2,8]. Irrespective of the type of surgical technique chosen to treat bladder exstrophy/epispadias, certain important principles remain, including radical

mobilization of the posterior vesico-urethral unit from surrounding pelvic tissue; careful and precise suturing of the bladder; bringing the pubic symphysis into close apposition with secure closure of the abdominal wall; and careful attention to post-closure lower extremity and pelvic immobilization [9]. Common complications associated with any type of surgical repair of exstrophy/epispadias include bladder prolapse and dehiscence of primary closure, outlet stenosis, urethral fistula, urethral stricture and superficial wound infections; which often requires reconstruction [3,9]. There are no other reports in the literature of hair-bearing epithelium being incorporated into the bladder wall, however Shnorhavorian *et al.* reported that as they gained experience using CPRE, they made modifications to this technique; specifically performing bladder neck dissections meticulously to exclude adjacent skin and fascia [7].

Bladder calculi in patients with previous exstrophy and or hypospadias repair may be primarily managed with a variety of methods, including laser litholopaxy, lithotrite, and stone crushers. Comparative randomized studies are not available to identify the gold standard method, but a meta-analysis done by Cicone et al showed that transurethral lithotripsy and percutaneous lithotripsy were equally as efficacious [10]; however, in patients with narrow bladder necks due to bladder exstrophy repairs, percutaneous routes may be preferable to avoid further trauma and damage to the bladder neck as it is often their only remaining continence mechanism. But as seen in our patient, the underlying cause must be addressed or stones will recur.

While there are no other reports of hair-bearing epithelium in the bladder leading to calculi formation, there have been reports of urethral

stones forming after the use of hair-bearing donor-site in reconstructive procedures such as urethroplasties and neophalloplasties, or when scrotal skin is unintentionally incorporated into the urethra during hypospadias repair [11-14]. The management differed as Viviano *et al.*, Kampantais *et al.*, and Crain *et al.* recommended urethral depilation as definitive treatment using electrocautery, laser ablation, or thioglycolate solution; while Hiyashi *et al.* recommended periodic self-catheterization to the neourethra to prevent the formation of hairballs [11-14].

Continued follow-up will be required to determine the efficacy of laser ablation for depilation and prevention of recurrent bladder calculi in our patient.

Conclusion

Bladder calculi following exstrophy repair is relatively common, often associated with infection, foreign bodies, and urinary stasis. This is the first report of hair-bearing epithelium serving as the nidus for stone formation. Laser-ablation of the hair follicles was utilized to prevent the recurrence of hairs and bladder calculi.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: All photos were taken with parental consent.

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