A rare vesicovaginal stone case series managed with endoscopic laser lithotripsy and subsequent staged repair of vesicovaginal fistula

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Abstract

Vesicovaginal fistula (VVF) due to various obstetric or gynecological causes is not uncommon but vesicovaginal stone in association with VVF is extremely rare presentation. The symptoms of vesicovaginal stone may be variable from recurrent urinary tract infections to perineal swelling with urinary incontinence. To diagnose stone associated with VVF only pervaginal examination is sufficient without need of any invasive investigations but still it is missed or diagnosed lately. To increase knowledge of this unusual condition we present case series of four such rare cases to outline safety and efficacy of endoscopic laser lithotripsy approach to manage vesicovaginal stone. We have used endoscopic laser lithotripsy method to clear the vesicovaginal stone which is minimally invasive way to provide early recovery by avoiding open surgical approach. Subsequently fistula can be repaired successfully after 6 weeks once bladder and vaginal wall infection and inflammation related to stone subside. To minimize morbidity in our cases we have specially used holmium laser energy to break the stone.

Key words

Vesicovaginal fistula, Pervaginal examination, Endoscopic lithotripsy, Holmium laser, Vesicovaginal stone

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Introduction

Vesicovaginal fistula represents a devastating morbidity[1] in urogynecology in which continuous urine leakage lead to precipitated solutes being constantly washed out from the urinary tract, so primary stone formation is rare.[2]Hence vesico-vaginal stone in association with VVF is relatively a rare presentation.[3,4,5] The risk factors for vesico-vaginal stone(VVS) formation are presence of foreign body(catheter/suture), urinary tract infection[6] and

prolonged duration of disease without treatment in patients of neurogenic bladder,vaginal outlet obstruction or with congenital anomalies of genitourinary tract.[7] Akamaguna et al; found that stones were found only in 7% of VVF cases on radiological evaluation.[8]The presentation of VVS may vary from suprapubic pain,repeated urinary tract infections,pyuria,dysuria,,hematuria and urinary leakage. For diagnosis of VVS only pervaginal examination is required without need of any investigations.[9]

Although very few case reports about stone association with VVF cited in literature, this lacks uniformity in approach due to scarcity of data; So we present case series of four such rare cases to outline safety and efficacy of endoscopic laser lithotripsy approach to manage vesicovaginal stone and for descriptive analysis of etiopathology, clinical and therapeutic aspects of vesicovaginal stone.

Case 1

A 40 year old female presented with continuous urinary leakage pervaginum since last 7 years and now since last 4 months she had suprapubic pain and hematuria for which she had came to hospital. She underwent total abdominal hysterectomy for abnormal uterine bleeding 7 years ago with benign fibroid as histopathology; In post operative phase after catheter removal patient had constant urinary leakage but she has not taken any treatment for same. General examination was unremarkable. Abdominal examination was suggestive of mild tenderness in suprapubic region with healthy scar of previous surgery. Per speculum vaginal examination showed a large stone present in vagina [figure1]along with protrusion into the bladder through indurated margin of VVF without vulval skin excoriation.USG, Pelvic X ray and CT urography were suggestive of vesiovaginal fistula with 4.1cmx4.9cm vesicovaginal stone (1200HU)[figure2]and normal upper tracts. Serum creatinine was normal. Urinalysis showed pyuria with E coli as growth in urine culture. Cystoscopy and Vaginoscopy showed large bladder stone (4 x4cm)extending vaginally through fistulous tract away from to bladder neck and both uretric orifice in supra trigonal region with adequate capacity of bladder.

The VVS was managed in two stages, in the first stage stone was removed under perioperative antibiotics coverage and subsequently fistula was repaired in second stage to prevent future stone recurrence.A gap of 6 weeks was given for resolution of stone related infection and inflammation between two stages of management.We have used holmium laser to break the stone at vesicovaginal junction close to fistula defect followed by laser cystolithotripsy. Intra-vesical portion of stone was separated from the vesicovaginal junction of stone transurethrally by holmium laser high energy settings and broken into large pieces. Then vaginal part of stone was broken into large fragments for easy delivery of all the remaining fragments via wide vaginal opening with additive help of transurethral bladder wash. Finally all residual large fragments of stones were easily extracted transvaginally by gentle manipulation. Sutures were found embedded inside the fragmented stone [figure3]may be cause of stone formation in this case which may be residing there due to attempted repair of bladder injury or vault closure during hysterectomy. Stone

analysis showed crystals of calcium magnesium ammonium phosphate. After six weeks VVF was repaired trans-abdominally by O'Connor method with interpositioning omental flap. Patient voided well after sequential removal of suprapubic and perurethral catheter after 21 days. Because of prolonged history, VVF margins were sent for histopathological analysis,but didn't show any sign of malignancy.Patient is on regular follow-up without any complaints.

Case 2

17 year old female presented to urology OPD with complaints of pain in suprapubic and perineal region with continuous urinary dribbling since 5 years. She was operated with vaginoplasty and right ureteric reimplantation for complete vaginal atresia with right ectopic ureteral opening in 2013. Subsequently she developed VVF possibly due to faulty surgical technique and abnormal vaginal moulding methods. She underwent transvaginal VVF repair in 2014 but it failed again and patient was leaking since then.Her physical examination was normal; local examination revealed brownish yellow colour stone pouting through vagina with foul smell of dribbling urine. There was excoriation of perineal and vulval skin and unhealthy vagina with skin grafts of previous vaginoplasty.USG abdomen showed Right gross Hydronephrosis and gross Hydroureter.Pelvic X ray and CT Urography [figure4] findings were suggestive of vesico-vaginal fistula involving posterior wall of urinary bladder with 5x4cm stone in bladder protruding into vagina through fistulous tract with paper thin parenchyma of right kidney.

Intra operative findings showed large VVF extending into bladder neck and proximal portion of urethra distorting entire trigone, posterior bladder wall and about 1 cm away from left ureteric orifice with vesicovaginal stone(5x4cm) occupying most portion artificial vagina. Unpilated hairs of previous vaginoplasty grafts seen on vaginoscopy [figure5]may be nidus for VVS formation in this case. Right ureteric opening on right side of dome of urinary bladder was completely blind on right Retrogradepyelography, suggestive of right anastomotic site stricture leading to right poorly functioning kidney.We have used holmium laser lithotripsy to break the stone in first stage. Subsequently after VVF repair patient was given three options with 1)bladder neck continence surgery which has low success rate because of weak vaginal and urethral tissues due to previous surgeries or 2)bladder neck closure and mitrofanoff procedure or 3)simple cystectomy with ileal conduit urinary diversion or left permenant ureterostomy with right nephro-uretrectomy but patient was lost to follow up.

Case 3

A 60 year old lady with cervical cancer stage II-a, underwent radiotherapy 12 years ago. She was diagnosed with vesico vaginal Fistula 6 year ago. Now she had presented with episodes of hematuria since 15 days. Physical examination was normal and vaginal examination showed a large stone in the vagina covering most of portion of fistula. Patient had slightly raised serum creatinine 1.38mg/dl.X ray KUB showed a large stone in the region of the

pelvis.USG showed 4x 4cm size vesical calculus with bilateral minimal hydrouretro-nephrosis. CECT abdomen and pelvis with urography showed VVS with thickened of bladder and vaginal wall without any abnormal enhancing areas. Cystovaginoscopy revealed dumbbell shaped stone (4x3cm)with vesical portion extending into vagina through narrow and indurated fistulous tract(1.5x1.5cm) in supratrigonal region away from to bladder neck and orifices with changes of urethrocystovaginitis and no recurrent tumors were seen.

We have used holmium laser lithotripsy to break the stone and biopsy was taken from fistula. Subsequently transabdominal VVF repair with interpositioning well vascularized omental flap was done with negative biopsy, explanation of poor success rate and possibility of future recurrence. However patient didn't choose option of anterior exenteration as complete cure. On follow up CECT pelvis showed no recurrence of tumour and leakage from VVF.Radiotherapy can cause loss of microcirculation in the tissue, which may cause mucosal atrophy, ulceration, and subsequently stasis of urine further lead to stone formation.

Case 4

A 14 year old girl with myelodysplastic disease and paraplegia referred to urology for urinary incontinence and recurrent urinary tract infections since last 3 years. She was bedridden since last 7 years. Physical examination showed tachycardia with cachexia. Vaginal examination showed a large stone coming from inflamed wall of vagina through VVF with perineal skin excoriation and continuous leakage of dirty urine with bedsore and neuropathic ulcer on foot. Xray, USG and CT KUB were suggestive of VVF with 7x5cm vesicovaginal stone and bilateral moderate hydroureteronephrosis with thick walled bladder.Serum creatinine was raised (1.86mg/dl) and Urinalysis showed pyuria,bacteriuria with E coli in urine culture. With adeqaute perioperative antibiotic coverage cystoscopy and vaginoscopy showed large bladder stone (7x5 cm)extending into vagina through inflamed fistulous tract in supra trigonal region away from to bladder neck and laterally placed uretric orifices with extremely trabeculated and inflamed bladder full of turbid urine.

We have used holmium laser to break the stone at vesicovaginal junction close to fistula defect followed by laser cystolithotripsy. Stone analysis showed crystals of magnesium ammonium phosphate. After six weeks trans-abdominal VVF repair with omental interposition flap was done with negative VVF margins. Postoperative phase was uneventful and patient was started on clean intermittent catheterization(CIC)1 month after removal of per urethral catheter. Patient is on regular follow-up with CIC and anticholinergics having no complaints. Etiology of stone formation may be prolonged urinary incontinence, neuropathic urethrovesical dysfunction, recumbent posture of long duration, lack of medical or rehabilitation care, vaginal outlet obstruction lead to vaginal pooling of urine, urinary stasis in bladder and subsequent urinary infection causative for encrustation.

Discussion

Halban in 1900 reported first patient of vaginal stone with vaginal cystocele.[10] There are only few reports of vesicovaginal stones associated with VVF available in the literature. Patients with VVS are associated with wide variety of clinical features likewise dysuria, pyuria,hematuria, storage lower urinary tract symptoms, suprapubic pain, vaginal outlet obstruction, perineal swelling, dyspareunia, urinary incontinence and/or retention.[11] The degree of urinary incontinence may be decreased due to ball valve effect caused by part of stone lying inside bladder.[3,12]

The theory postulated behind the formation of VVS arise from stasis of urine through fistula tract in obstructed vagina.[13]Subsequently this stagnant urine with infection may enhance stone encrustation.[14] Another theory may be that precipitation near the nidus persists and lead to enlargement of stone which decreases the drainage through fistula. That enhance the stasis of urine in bladder which promotes stone formation. However such a vesical stone may rarely erode through fistula wall.[15,16]

As VVF association with stone is rare, its diagnosis is routinely delayed and/or missed. Dalela *et al.*[2] found that most of patients are disadvantaged from poor socio-economic classes lead to gap of an average of 28.8 months after the onset of the fistula before the patients consultation to doctor, such patients came to hospital when the disease has become painful and hard to manage. All the VVS reported in the literature were radiopaque in the plain X ray film, which was similar in our cases. USG or CT urography was used to rule out obstructive uropathy and to know detailed anatomy or any associated anomaly. We have noticed pyuria , bacteriuria and no abnormal metabolism in urine analysis of all our cases, similar to other reported cases in literature. [2,3,4,17,18,19] Although many stones in our cases were of mixed composition, the majority of stones were struvite, produced in chronic infection. Risk factors for such urine infection may be a foreign body, such as suture or catheter. [20]

All VVS should be managed in two stages, with interval of 6 to 12 weeks for resolution of stone related infection and inflammation. The stone removal could be done with open or endoscopic techniques.[21] In review of literature, most of cases stone was managed by transvesical suprapubic open cystolithotomy or by transvaginal route with chisel and hammer or forceps.[2,9,19,22,23,24] Dalela et al[2] suggested stone could be removed endoscopic cystolithotripsy which can prevent unnecessary trauma to the ureter and extension of the fistula tract by forceful traction applied during open methods of stone retrieval.

We have used a novel method with holmium laser to break the stone at vesicovaginal junction close to fistula defect followed by laser lithotripsy in bladder and vagina to avoid causing any undue tissue dissection. Finally all residual large fragments of stones were easily extracted transvaginally by gentle manipulation. Use of high energy laser was the most suitable way to deal huge stone burden in presence of inflamed tissues.[25] Such a method of endoscopic laser lithotripsy to break the stone with use of a small size sheath was advantageous and specific in our case series. This can drastically reduce morbidity of the first stage by avoiding conventional method of stone removal, which may lead to fast recovery. Due to long standing stone and recurrent infections in such cases bladder wall was chronically inflamed and frigile so utilization of laser energy precisely was our target.

In most of published series, VVF was repaired after some interval in staged manner. In the second stage, fistula can be closed by transabdominal or tranvaginal route with or without interpositioning flap. In our all cases VVF was

repaired after six weeks tranvesically through transperitoneal route by open O'Connors method with omental flap interposition because all VVF were large and complex. VVF was repaired by experienced team of surgeons with a sufficient infrastructure [15] following principles of Couvelaire of adequate visualization, dissection, approximation of the margins and urine drainage to decrease the risk of surgical failure.[26]

Conclusion

The patients of prolonged untreated VVF are prone to form vesicovaginal stone which increases overall morbidity. Every VVF patient should be carefully evaluated with through pelvic examination before getting done routine investigations. This approach will be helpful to make quick and accurate clinical diagnosis of VVF associated with stone. Xray KUB is noninvasive and cost effective option for confirmation of diagnosis by avoiding over investigation. Endoscopic method to clear the vesicovaginal stone with laser technology is minimally invasive,less morbid and provide early recovery by avoiding open surgical approach. Once malignancy is ruled out, fistula can be repaired successfully after bladder and vaginal wall infection and inflammation related to stone subside.

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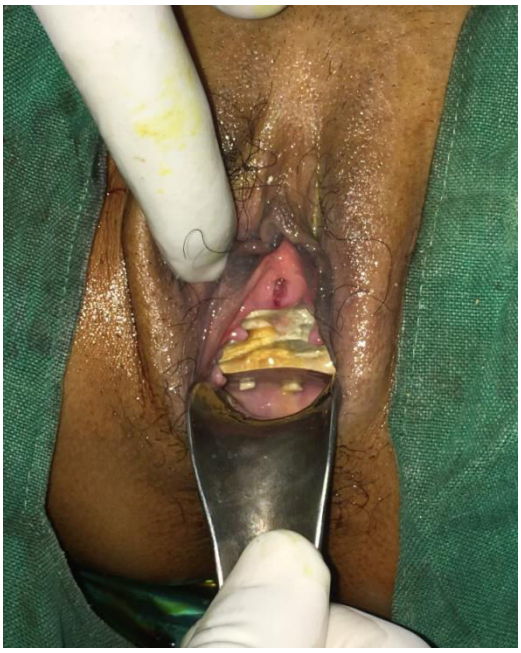

FIGURE1: Pervaginal examination showing vaginal portion of vesicovaginal stone.

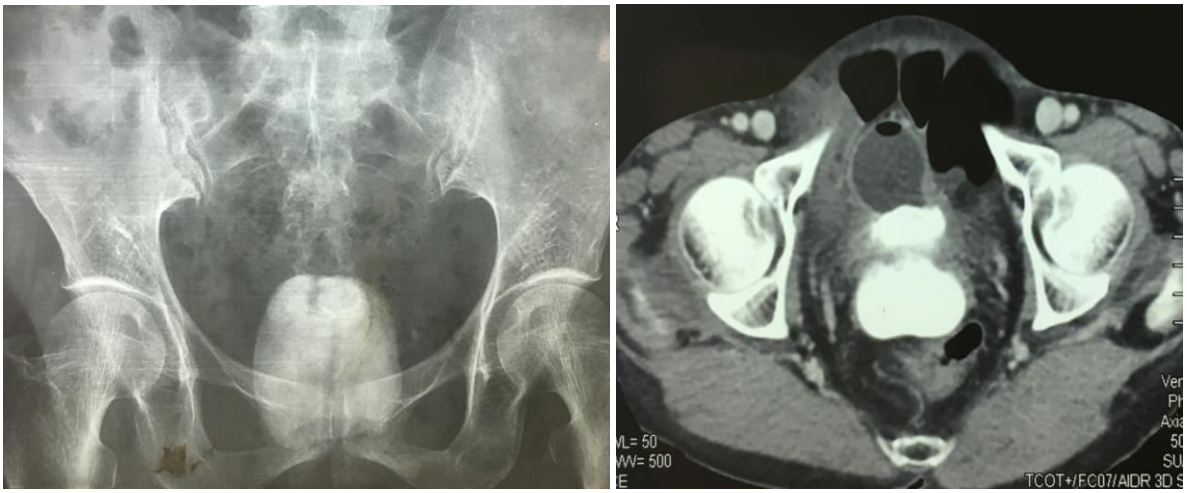

FIGURE2: Pelvic x-ray film showing large radio opaque shadow lying low in pelvic area and CT scan of pelvis showing of vesiovaginal fistula containing vesicovaginal stone.

FIGURE3: Suture embeded inside the vesicovaginal stone on vaginoscopy and same after stone removal.

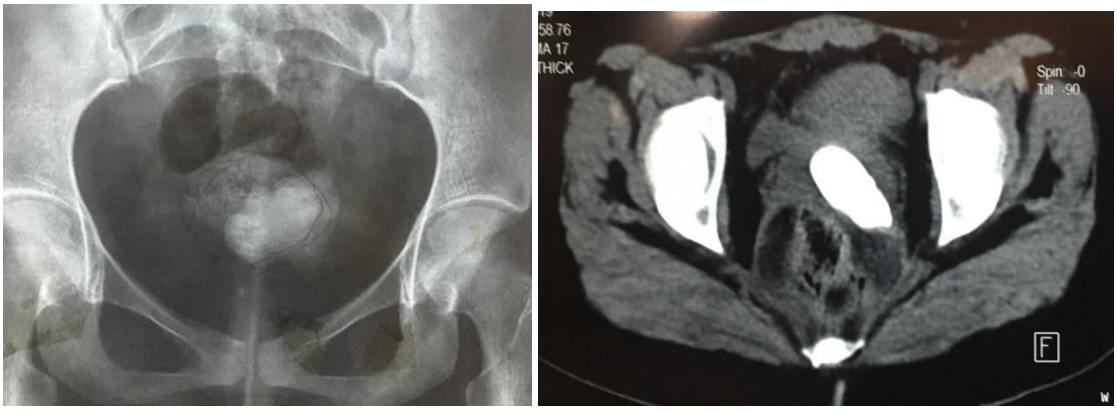

FIGURE4: Pelvic x-ray film showing double shadow of stone in pelvic area and CT scan of pelvis showing vesicovaginal stone .

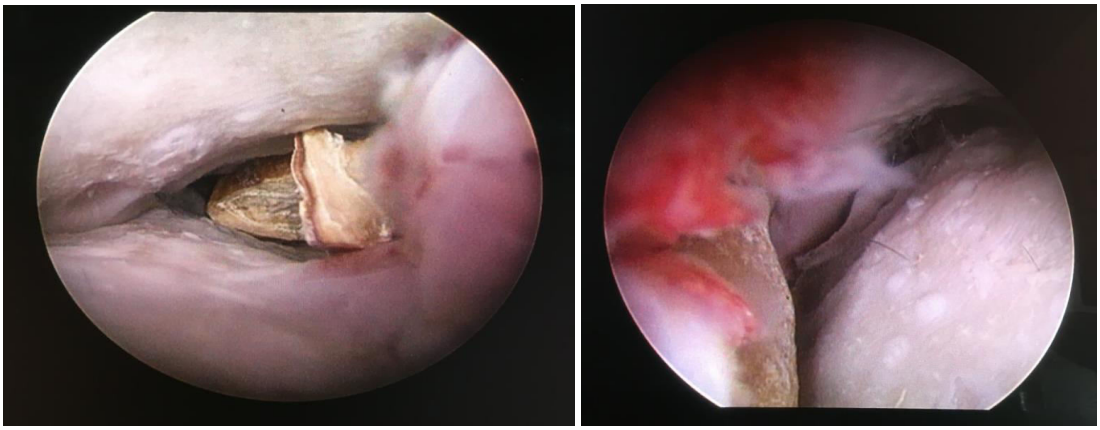

FIGURE5: Vaginoscopy view showing vaginal portion of stone with hairs on graft and fistulous communication of bladder.