



Case report

Vesical calculus associated with vesicovaginal fistula



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ABSTRACT

Vesicovaginal fistula (VVF) is relatively common after radiotherapy, but the formation of a bladder stone after VVF is still rare. The diagnosis is often delayed, and mortality sometimes occurs with delayed diagnosis and treatment, especially in elderly and disabled women. To increase knowledge of this unusual condition—vesical stone associated with VVF—we present a case report, and summarize the published cases to review this topic, focusing on the etiology, symptoms and signs, and recent management strategy. Finally, based on this case report, we emphasize that careful physical examination and the use of less invasive and economic tools, such as plain X-ray are important to investigate this very sick and old lady, because these strategies could help us to make an accurate diagnosis earlier without the need for more aggressive, invasive, and expensive tools.

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Introduction

Vesicovaginal fistula (VVF) represents a devastating morbidity in gynecologic urology, and the majority of cases in the developing world are the result of obstetric trauma. By contrast, in developed countries, pelvic surgery, radiotherapy, and benign and/or malignant pelvic pathology are the leading causes of VVF.¹ The interaction of urinary salt ions, inhibitors, pH, and temperature results in the formation of the microscopic crystals, eventually creating a bladder stone. These tiny stones lead to clinically evident calculi unless there is a concomitance of urinary stasis or mucosal adherence. In cases with VVF, urinary leakage results in the precipitated solutes being continuously flushed out of the urinary tract, so the formation of primary vesical calculus is rare.² The pooling of infected urine or a foreign body (e.g., a suture or urethral catheter)

may be the probable etiologic factor rather than the stasis of the urine in the bladder. As precipitation around a nidus persists, an enlarged calculus may hinder the fistula, increase the amount of stagnant urine inside the bladder, and further enhance encrustation. However, the vesical stone *per se* may foster the erosion of the fistula. Because VVF associated with vesical stones is rare, the diagnosis is often missed and/or delayed. Although the outcome is good in the majority of cases, sepsis sometimes occurs, with resultant mortality, especially in elderly and disabled women. The following case presents an 85-year-old woman with the finding of a 9-cm vesicovaginal stone and sepsis 7 years after diagnosis of VVF.

Case report

An 85-year-old woman, gravida 7, para 6, with cervical cancer, FIGO stage IIb, underwent radiotherapy 15 years ago. She had a more than 10-year history of senile dementia and was bed-ridden for the past 10 years. VVF was diagnosed 7 years ago. During this period, a diaper rather than an indwelling catheter was used to treat her urinary incontinence. This admission was due to 4 days of gross hematuria and fever.

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Physical examination showed acute illness, and vaginal examination revealed a large calculus protruding from the vagina, which obstructed the area of VVF. Pertinent laboratory results showed anemia, leukocytosis, renal function impairment, and poor nutrition. The routine plain film examination of kidneys, ureters, and bladder depicted a large calculus in the region of the bladder. Computed tomography demonstrated a 9-cm vesicle stone occupying almost all the space of the bladder, and the vesicovaginal border was obscured. In addition, hydronephrosis, as well as the appearance of lithiasis in numerous areas, were detected in the bilateral kidneys and ureters. Even though intensive treatment for her sepsis was administered, the patient died on the third day after admission.

Discussion

Although a few reports of VVF accompanied with vesicle stones are available in the literature (Table 1),^{2–8} this is still a rare event. Age seems to be merely a dependent factor in the pathogenesis of VVF. For almost all women of reproductive age, delivery trauma is the predisposing factor of VVF. By contrast, for elderly women, the main causes are surgery and/or radiation. The order of treatment (with surgery and radiation) brings with it differing risks for VVF formation. Nine percent of patients will be complicated with VVF if they receive surgery initially, but only 1.4–5.2% of those, who receive radiation initially, will be complicated with VVF.^{9,10} For women with a history of gynecological cancer, VVF may present as a postoperative or postradiotherapy complication, or be caused by malignant disease. Radiation therapy for women with pelvic cancer can result in primary or secondary fistula formation. Radiation causes the loss of microcirculation in the involved tissue, which results in mucosal atrophy, ulceration, and fistula formation. Women with fistulae after delivery usually develop total incontinence after around 10 days. When radiation therapy is the primary cause of the fistula, the presentation may be delayed from 5 months to 30 years.¹¹ In our case, VVF was diagnosed 8 years later. The interval between the diagnosis of VVF and the detection of vesicle stones is reported to range from 6 months to 8 years. In this case, the interval was 7 years. Clinical features of VVF with vesicle stones can be recurrent urinary tract infections, gross hematuria, pyuria, suprapubic pain, perineal swelling, and/or urinary incontinence. The severity of urinary leakage may be reduced because of the “ball valve” effect caused by a vesicle stone.^{3,12} Examination per vagina is enough for the diagnosis of a bladder stone combined with VVF,

and cystoscopy aids in the confirmation. All the vesicle stones reported in the literature were radiopaque in the kidneys, ureters, and bladder plain film, as was our case. The role of ultrasound or excretory urography is to demonstrate the obstructive uropathy. In a search for possible causes contributing to vesicle stones related to VVF, previous reports have shown a positive correlation with infection.^{2,3} None of these reported patients had an abnormal mineral metabolism or outlet obstruction, and all urinalyses revealed pyuria or bacteriuria.^{2–8} Although many stones are of mixed composition, the majority of stones are struvite. Struvite stones are made of magnesium ammonium phosphate (sometimes called triple phosphate), and arise from chronic infection with urease-producing organisms. Moreover, a foreign body, such as suture stitches or an indwelling catheter, places the patients at risk for urinary infection.¹³

The management of vesical stones combined with VVF can be divided into two parts, including the initial removal of the calculus and the following repair of the fistula.² Staged intervention allows the infection and mucosal edema of the bladder to subside. Removing the stone can be performed via either endoscopy or the open approach.¹⁴ The open approach may be performed through either the suprapubic transvesicle route or the transvaginal route. Dalela et al² recommended removing the stones endoscopically, in order to avoid enlargement of the fistula or injury to the ureter. Although spontaneous healing of fistulae has been reported, the size of the fistulae in the present case was too large to resolve with conservative treatment. In terms of the surgical repair of VVF at the second stage, fistula closure should be individualized and can be attempted transvaginally or transvesically, with or without flap interposition.

In our case, infection might have been the leading cause of the formation of a bladder stone. In fact, this patient initially presented with urinary tract infection and sepsis; therefore, we diagnosed a vesical stone in VVF. Due to the fact of multiparity in the patient herself, a certain degree of genital prolapse related to multiparity might also be a risk factor for stone formation, because a proposed mechanism of stone formation in patients with pelvic organ prolapse is bladder obstruction secondary to urethral kinking.¹³ In addition, the long-term bedridden condition may have caused malnutrition, poor hygiene, and urinary stasis, which could worsen the lithogenesis.

In conclusion, we report this case to emphasize the importance of the early diagnosis of vesical stones in VVF, and highlight the clinical characteristics of this disease because of its rarity. In

Table 1
Literature review of vesical calculus associated with vesicovaginal fistula.

Patient age	Ref.	Causes of VVF	Duration between VVF and stone formation	Predisposing factors of stone formation	Stone analysis	Cystolithotomy	Fistula repair
NA (19 y)	2	Obstructed labor	28.8 mo	Infection	Struvite	Endoscopic (17) Transvesicle (2)	Transvaginal Transvesicle
25 y	3	Obstructed labor	8 mo	Infection	CO, TP, UA	Transvaginal	Transvesicle
42 y	3	Obstructed labor	14 mo	Infection, failed repair	CO, TP, UA	Transvesicle	Transvesicle
32 y	3	Hysterectomy	6 mo	Foreign body (suture)	CO, TP, UA	Transvaginal	O'Conor
26 y	3	Obstetric labor	8 mo	Infection	CO, TP, UA	Transurethral	NA
				Prior repair, catheter			
23 y	3	Obstructed labor	26 mo	Infection, failed repair	CO, TP	Transvesicle	NA
21 y	4	Difficult delivery	2 y	Infection	Struvite	Transvaginal	Transvesicle
35 y	4	Difficult delivery	3 y	Infection, failed repair	TP	Transvaginal	Combined
67 y	5	Hysterectomy, RT	2 y	Retained stent	NA	Endoscopic	Nil
25 y	6	NA	NA	Infection	Struvite, CO	Endoscopic	NA
					Foreign body (suture)		
73 y	7	Hysterectomy, RT	NA	Infection	CP, MAP	Cystolithotomy	NA
58 y	8	Obstructed labor	40 y	Infection	NA	Transvaginal	NA
85 y	a	RT	8 y	Infection	NA	Nil	Nil

CO = calcium oxalate; CP = calcium phosphate; MAP = magnesium ammonium phosphate; NA = no data available; RT = radiotherapy; TP = triphosphate; UA = uric acid.

^a The patient in this report.

addition, careful physical examination and the use of less invasive and economic tools, such as plain X-ray should be emphasized, because these strategies could help us to make an earlier and acute diagnosis of this very sick and old lady. Over-investigation and over-treatment should be avoided.

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