



Case Report

A complicated urethrovaginal fistula following an inappropriate urethral diverticulum management

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ABSTRACT

Objective: Urethral diverticulum is uncommon, therefore appropriate evaluation, preoperative planning and counseling must be done in order to make correct diagnosis and prevent complications.**Materials and methods:** A case of anterior vaginal wall mass was treated elsewhere by a gynecologist as periurethral cyst abscess; incision and drainage were done but a symptom of pus discharge was observed after 2 weeks. Therefore, exploration, cyst wall excision and primary closure were done though histopathological examination surprisingly confirmed the presence of urethral tissue suggestive of diverticulum.**Results:** Subsequently, she developed persistent urinary leakage along with urethrovaginal fistula for which they again performed pervaginal multilayer closure. Patient was later referred to us with recurrent urethrovaginal fistula. We performed posterior urethral fistulectomy with anterior vaginal wall flap and multilayer closure. Three years follow up reveals complete recovery.**Conclusion:** Even urethral diverticulum is a rare condition, should be kept in mind as early diagnosis and management.© 2017 Taiwan Association of Obstetrics & Gynecology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Benign masses of the anterior vaginal wall are uncommon and occur in about 1% of cases [1]. A number of benign anterior vaginal wall masses are known including caruncle, ectopic ureterocele, periurethral cyst (Skene's duct cyst), urethral diverticulum, urethral prolapse, and vaginal wall cyst of embryonic origin (Müllerian and Gartner's duct). The appearance and presenting symptoms appear to be non-specific and non classical which may lead to incorrect and delayed diagnosis. Imaging studies are usually necessary to differentiate them from each other. Appropriate evaluation,

preoperative planning and counseling must be done in order to make correct diagnosis and prevent complication. We report a case of missed urethral diverticulum which was initially suspected and treated as infected periurethral cyst (Skene's duct). We discuss clinical presentation, etiology, investigation modalities and treatment options.

Case report

We report a case of a 64 year, female, P4A2 who had a difficult course of treatment with multiple procedures in a different hospital but later referred to us. The previous course of management is herewith described. Patient who was apparently normal with no medical or surgical history presented to a different hospital with a cystic mass 0.5 cm below urethral meatus at the anterior wall of the vagina measuring 4 × 5 × 6 cm (Fig. 1(a)) associated with increased urinary frequency, pain and tenderness at the anterior vaginal wall.

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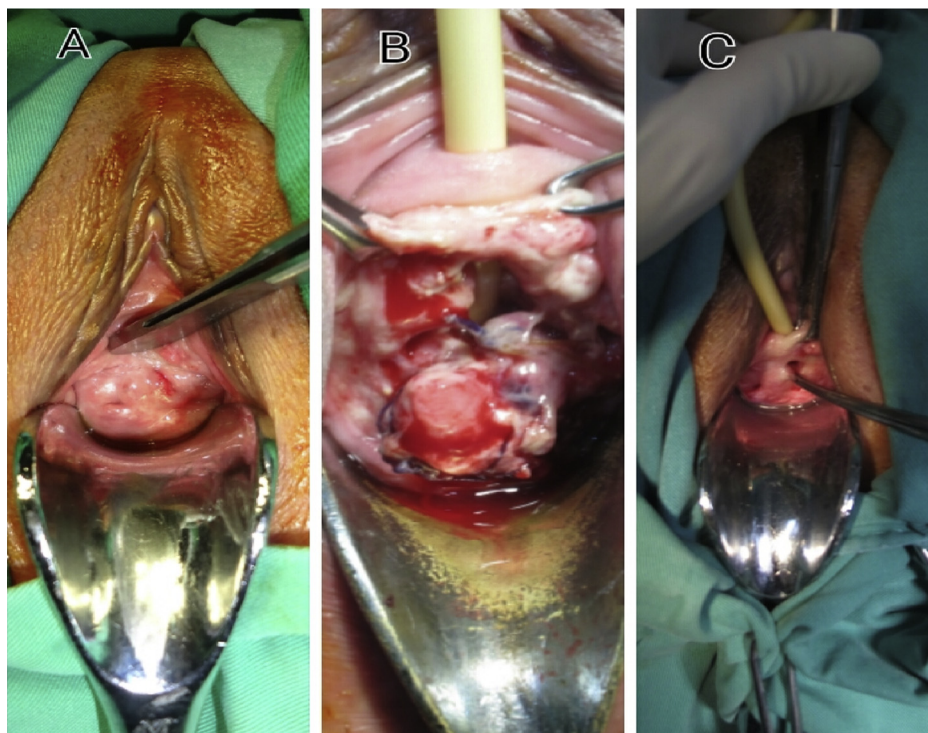


Fig. 1. Intra-operative finding. **a.** Cystic mass at the anterior wall of the vagina; **b.** Urethrovaginal fistula post-diverticulectomy; **c.** Recurrent urethrovaginal fistula.

No history of dysuria or post-void dribbling, no purulent discharge observed from urethral meatus after sub-urethral mass was compressed. Symptoms were started one month back and increased in severity one week prior to admission. Skene's duct abscess was suspected based on initial assessment with perineal ultrasound (Fig. 2). Incision and drainage was done which revealed a sub-urethral mass with about 500 ml of pus oozing after incision. Cefazolin 1 g intravenous 8 hourly and Gentamycin 60 mg intravenous 8 hourly were given for 3 days, the patient was discharged

well at day 2 postoperatively on clindamycin 300 mg capsule 12 hourly for 5 days.

Recurrence of the sub-urethral abscess was noted after 2 weeks. Second drainage and exploration with cyst wall excision was done. Intraoperatively the mass was noted to be extended up to the bladder neck and a primary closure was done. Postoperatively, patient developed persistent gross hematuria with passage of blood clots which led to significant anemia and 4 units of packed red blood cells were transfused. Prolonged catheterization and continuous flushing with normal saline solution were done. Patient was on Cefazolin 1 g intravenous and Gentamycin 60 mg intravenous for 7 day. Hematuria resolved with good urine output and patient was subsequently sent home on cefadroxil 500 mg tablets 12 hourly for 5 days. Histopathological examination (HPE) suggested presence of infected urethral diverticulum showing fibromuscular tissue with chronic inflammation, fibrosis and focal lining of urothelial and squamous epithelial were seen on cyst wall.

A week later she developed urinary leakage per vagina. Patient was referred to urogynecological department. Speculum examination and voiding Cystourethrography showed a 1.0 cm urethrovaginal fistula (UVF) located at mid-urethra (posterior wall of urethra) (Fig. 1(b)). Wound debridement, fistula repair, multilayer closure were performed with intra-operative cystoscopy (Fig. 3) and insertion of a Fr #16 Silicon indwelling Foley, patient was on Cefadroxil 500 mg tablets for 4 days. Later, fistula tract inflammatory tissue developed sub-chronically. She was discharged on 10th post-operative day with the indwelling Foley catheter in situ for a month.

At 4 months postoperatively follow-up visit, patient observed with recurrent urinary leakage. A 0.5 cm urethrovaginal fistula recurred at posterior mid-urethral region. Posterior urethral fistulectomy with anterior vaginal wall flap and multilayer closure were done at 4th week after fistula tract inflammatory tissue developed sub-chronically and intra-operative cystoscopy was done (Fig. 4). During hospitalization Cefazolin 1 g intravenous 8 hourly,



Fig. 2. Perineal ultrasonographic image. An anterior vaginal wall cyst, 4 × 5 × 6 cm left, round, septal mass between the uterus and the bladder.

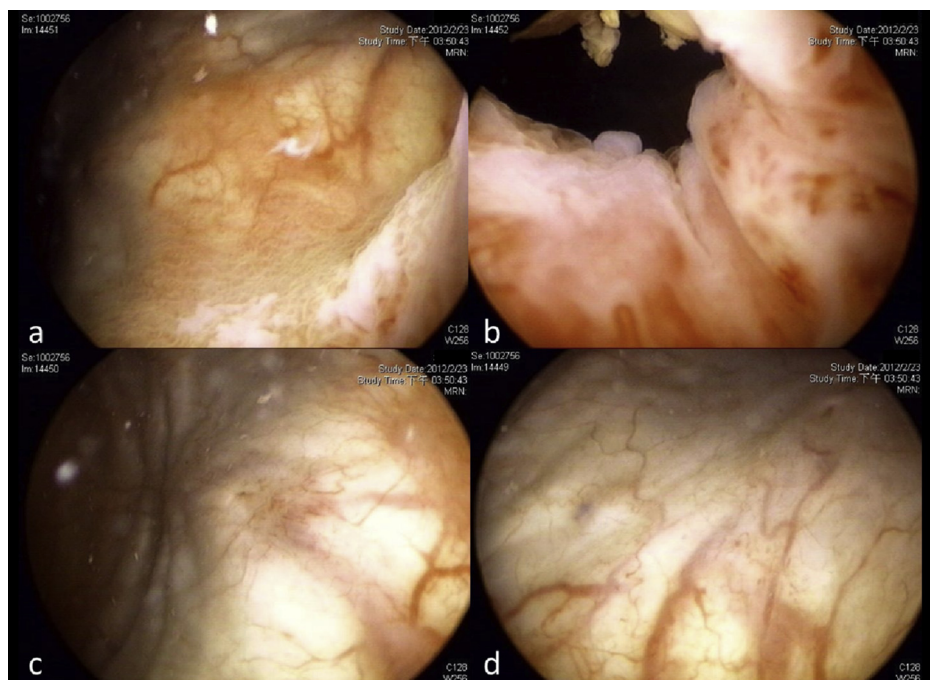


Fig. 3. Intra-operative cystoscopy finding at first fistula repair. **a.** trigon area; **b.** bladder neck and urethra; **c.** right ureter orifice; **d.** left ureter orifice.

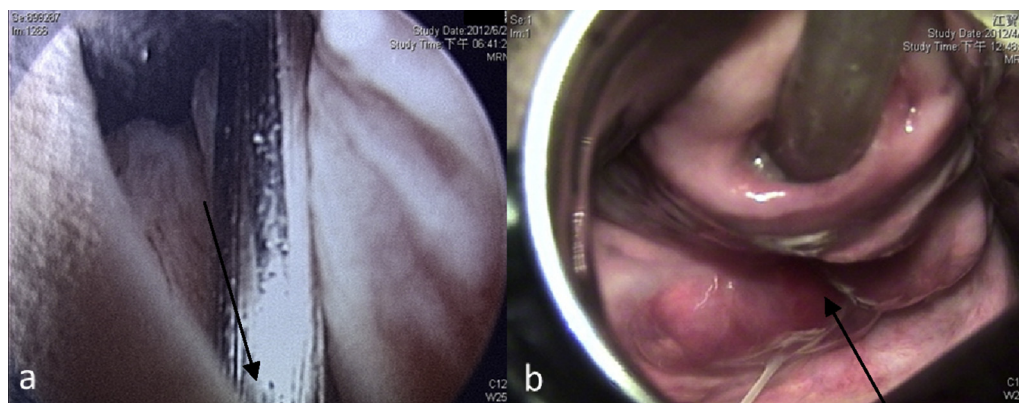


Fig. 4. Intra-operative cystoscopy finding at recurrent fistula repair. **a.** urethrovaginal fistula-urethral view; **b.** urethrovaginal fistula-vaginal view.

Gentamycin 60 mg 8 hourly for 4 days and Ertapenem 1 g intravenous daily for 2 days were given. The patient was discharged on the 4th post-operative day with indwelling Foley's catheter on clindamycin 300 mg 12 hourly for 5 days. Catheter was retained for 4 weeks postoperatively. It has been 3 years since the repair and reconstructive surgery. She is currently asymptomatic and reported no urine leak. The total Urinary Distress Inventory Questionnaire (UDI-6) and Incontinence Impact Questionnaire (IIQ-7) scoring was 3 and 4 at third year postoperatively [2,3]. Long-term follow-up is still recommended.

Discussion

Urethral diverticulum occurs in 0.6%–5% of women with the majority presented at 3rd to 5th decade of their life [4]. Many are difficult to diagnose as seen in our patient. The classic symptoms were dysuria, post-void dribbling and dyspareunia [5]. These are insufficient for proper diagnosis. Beside these, chronic or recurrent urinary tract infections, urinary frequency and/or urgency,

hematuria, bloody urethral discharge, urinary incontinence, urinary retention, pelvic or urethral pain and vaginal mass should be included as possible diagnostic criteria to avoid misdiagnosis and improper management [5,6]. A high level of suspicion and appropriate imaging are the essential steps in establishing the proper diagnosis [7]. Urethral diverticulum usually located ventrally over the middle portion of the urethra corresponding to the area of anterior vaginal wall which is 2–3 cm inside the introitus. The etiology of urethral diverticulum is not well established. However trauma of childbirth is one of the possible predisposing factors. A repeated inflammation and obstruction of periurethral as well as urethral glands resulted in cyst/abscess formation, these eventually rupture into the urethral lumen and remain as an outpouching which epithelialize to become a true diverticula [8]. Urethra can be efficiently inspected using urethrocytoscopy where the ostia of diverticulum can be visualized. However sometimes it's difficult to visualize the ostia particularly when diverticula collapsed. It may be possible to identify the ostium content by compressing the diverticulum during urethroscopy [9]. Transperineal or

transvaginal ultrasound is safe, inexpensive and rapid real time information regarding diverticulum as well as surrounding structures [7], additionally other pathological diagnostic approach like leiomyoma can planning without distorting a normal be done. Magnetic Resonant Imaging (MRI), this imaging study has emerged as the criterion standard in diagnostic imaging for urethral diverticula, as it reveals the extent and location of the diverticula. In patients with strongly suspect symptoms, MRI seems to demonstrate the diverticulum with the highest sensitivity and specificity. Furthermore, planar technology allows the exact ostium to be identified prospectively in many cases and can give the operating surgeon a “roadmap” from which to guide the operative intervention [10]. Other investigation modalities like voiding cystourethrogram (VCUG) and double balloon urethrogram (DBU) has been described with varying degree of sensitivity and specificity. In other study, MRI had a higher sensitivity for detecting diverticula and a much higher negative predictive rate than double balloon urethrogram (DBU) [11]. Of note is that a timely diagnosis is of paramount importance: a delay in UD diagnosis >12 months was reported to be associated with a high risk of post-operative complications [12]. Surgical management is not indicated in patients with minimal to no symptoms. Additionally, in patients with a small diverticulum, excision of the diverticulum is not necessary and can be very difficult, with increased risk of urethral trauma. For patients with mild symptoms, antibiotics and in some cases anticholinergic therapy can provide relief [13].

Diverticulectomy and marsupialization of the diverticulum sac into the vagina by incising the urethrovaginal septum are the surgical treatment options [8]. A care should be taken in any procedure as vagina is rich of microvasculature and nerves supply. Complete excision of urethral diverticulum with full opening is given a cure rate of about 70% [14]. In our case, a complete excision of diverticulum was done. Though, she developed complication of urethrovaginal fistula. High risk of complication may occur due to delayed repair, diverticular size >4 cm and complex configuration of diverticula [15]. The common complications are urinary incontinence (15%), urethrovaginal fistula (8%), urethral stricture (5%) and recurrent urethral diverticulum (10%). To avoid such complications, meticulous surgical technique, good hemostasis, avoidance of infection, preservation of a good vascularity, anterior vaginal wall flap and multilayer closure should be employed.

A suspected urethral diverticulum should be evaluated as its size, possible content and surrounding anatomical structures. The evaluation not only using cystourethroscopy but should include cystourethrography, MRI and intravenous urography depend on the case. Differential diagnosis of an anterior vaginal wall mass always should include urethral diverticulum. In such cases, compression of the mass may express urine with purulent material from external meatus. Proper evaluation is mandatory for successful management of diverticulum specially encountering a failed surgery. We believed that due to improper diagnosis and malpractice management like not using a Foleys catheter before and during surgery and disorientation of the anatomy led to massive bleeding and organ injury requiring multiple procedures which could have been prevented. In conclusion, an accurate history taking and a careful clinical awareness with high index of suspicion are required in all patients with remarkable vaginal mass along with anterior vaginal wall cyst, should be critically diagnosed for urethral diverticulum to

avoid misdiagnosis and complications. Therefore, an excellent surgical outcome can be achieved with meticulous preoperative assessment and proper surgical technique.

Consent

Written informed consent was obtained from the patient for publication of this Case report and the accompanying image and use of the images.

Authors' contribution

TS Lo: Protocol/Project development, Data collection, Data analysis, Manuscript editing.

SB Jaili: Data analysis, Manuscript writing and Manuscript editing.

R Ibrahim: Data analysis; Manuscript editing.

Financial disclaimer/Conflict of interest

None.

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