A Report of Three Cases of Vulvovaginal Cyst: Difficult Diagnosis to Successful Treatment with Review of Literature

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Received on: 07 April 2023; Accepted on: 19 May 2023; Published on: xx xx xxxx

Abstract
Aim: To identify different causes of vulvovaginal swelling and step-by-step identification and management options.

Background: Vulvovaginal cysts may start as asymptomatic cysts but may enlarge to big sizes and varied histopathalogy, needing immediate as well as long-term medical attention. We have reviewed three cases of vulvovaginal cysts with unusual presentations.

Case description: The first case presented was a giant dumbbell-shaped Bartholin cyst presenting as a labial cyst. The second case with a provisional diagnosis of Bartholin’s cyst came out to be aggressive angiomyxoma on immune histopathology. The third case presented with pus in the urine, occasionally painful urination, and a vaginal cyst which turned out to be a urethral diverticulum on cystoscopy.

Conclusion: A meticulous clinical history and examination of the patient supported by a few investigations help a gynecologist to formulate a plan of management to deliver the desired results to the patient.

Clinical significance: The most important clinical parameter for the vulvovaginal cyst is the site of occurrence. Later differential diagnosis is made, and investigation and treatment (surgical or medical) are planned according to the suggested approaches and ideas mentioned in the paper. Management protocols can be as straightforward as cystectomy or may require a multiprong, multidisciplinary approach and multiple sitting.

Keywords: Case series, Giant vaginal cyst, Urethral diverticulum, Vulvovaginal cyst.

Journal of South Asian Federation of Obstetrics and Gynaecology (2023): 10.5005/jp-journals-10006-2307

Case Series

A vulvovaginal cyst is a common problem reported in approximately 1 in 200 women, which can adversely affect their quality of life. Diagnosing the cyst right and formulating a management protocol can be challenging for a gynecologist due to varied presentation, symptoms and signs and the diverse causes like embryological, anatomical and dermatological. However, the prevalence of vulvovaginal cysts is underestimated as many are asymptomatic and there is overlap in expertise of anatomy and pathology with urologist, surgeon, and dermatologist. The vaginal and vulval cysts can present very differently as in our cases. One Bartholin cyst came with swelling in the upper part of labia majora, another patient was diagnosed as Bartholin cyst somewhere else; packed and referred to us in view of inoperability and excessive bleeding, was operated on and later diagnosed as malignant angiomyxoma. The last one came with pus through the urethra and anterior vaginal wall swelling later diagnosed as a urethral diverticulum.

Case 1
A 40-year-old P2L2 woman came to the outpatient department (OPD) with chief complaints of perineal swelling which was associated with dyspareunia for 1 year. It was initially small in size (approximately 3 cm × 2 cm) increasing gradually over 1 year to the final size. There were no menstrual, urinary or bowel complaints associated with it. On examination, the swelling was 10 cm × 4 cm occupying the upper part of left labia majora and mons pubis (Fig. 1A). The swelling was fluctuant and mobile such that it could be pushed to the lower part of labia majora and adjacent to the vaginal introitus at the posterolateral wall from its original position and cross fluctuation was positive. There was no associated enlargement of regional lymph nodes. Depending on the location and examination of the swelling, a provisional diagnosis of Bartholin cyst was made. Transvaginal ultrasound (TVS) was done to know the extent of the mass which showed a well-defined elongated cyst extending from the labia majora to the left posterolateral vaginal wall.

Under general anesthesia (GA) and all aseptic precautions incision was given on the most dependent area of the mucocutaneous junction of the left side of the vaginal introitus after pushing the cyst inferiorly (Fig. 1B). Blunt and sharp dissection was done to separate the cyst from the vaginal wall. The cyst came out was in dumbbell shape with the larger part of dumbbell extending into the paravaginal region (Fig. 1C). After deep dissection, the cyst was excised. The dead space was obliterated in layers and hemostasis was achieved. The excised tissue was sent for histopathological examination. On the cut section, greyish material came out. Microscopic examination revealed a cyst – ulcerated and focally lined by columnar epithelium with dense chronic inflammatory
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exudate rich in plasma cells and lymphocytes (Fig. 1D). The features were consistent with Bartholin cyst with superadded inflammation.

In the postoperative period, the patient was discharged the next day. She was comfortable postprocedure. She was followed up in OPD after a week and the incision had healed with no residual swelling or complaints.

Case 2

A 28-year-old married P1L1 female came with a swelling progressively increasing in size on the right labia majora associated with heaviness at the perineum for 6 months. Local examination showed a well-circumscribed, 12 cm x 6 cm x 6 cm mass soft to firm in consistency arising from the right posterolateral vaginal wall (Fig. 2A). Considering the diagnosis of a large Bartholin cyst patient was planned for an excisional biopsy. While dissecting, the mass was found to be very vascular and reaching up to the pouch of Douglas (POD), so the procedure was abandoned and the patient was referred to our hospital.

At our center, the patient was investigated with TVS confirming the mass with very high vascularity, and magnetic resonance imaging (MRI) showed hyperintense T2 signal and hypointense T1 signals of well-defined mass extending cephalically till POD, and no lymphadenopathy was seen. After obtaining informed written consent, a diagnostic laparoscopy was done to see the extent of the mass and to rule out any iatrogenic injury at the previous center. A wide local excision of the mass which was infiltrating into paravaginal, pararectal, and ischiorectal areas of the right side of the vagina was done under GA in collaboration with general surgeon (Fig. 2B). Histopathology was suggestive of aggressive angiomyxoma (Fig. 2C). Immunohistochemistry was consistent with it and was positive for estrogen, progesterone receptors, desmin and smooth muscle actin (SMA) (patchy immunoreactive). Her postoperative recovery was uneventful. The patient was given three doses of gonadotropin-releasing hormone agonist (injection, leuprolide 3.75 mg) once a month. No recurrence has been reported so far on follow-up visits of 1.5 years. The last MRI done (one year postoperative), 2 months back is normal and the patient is still under regular surveillance.

Case 3

Our third case of P2L2 patient came to us with pain during urination and pus per urethra. On examination per speculum, she had a tense 10 cm x 5 cm x 6 cm mass arising from the anterior wall of the vagina completely covered with vaginal wall with no punctum
visible (Fig. 3A). On pressing the cyst, the pus used to come through the urethra in small amounts. She was taken up for examination under anesthesia after preoperative workup. Preoperative cystoscopy was done and 5–6 small orifices (Fig. 3B) were seen on the posterior wall of the urethra throughout its length, bladder mucosa, and ureters were normal. Pus was drained through the vaginal surface with a small incision and a urethral catheter was placed in situ for persistent drainage of urine and giving time for the urethral defects to close spontaneously. The patient was discharged on oral antibiotics and was reassessed every week. After one month of conservative treatment, the patient was replanned for exploration. Cystoscopy revealed closure of all defects of the urethra communicating with the vaginal cyst. Vaginal cyst marsupialization/cystectomy with reconstruction was planned as the cyst has again grown to the same size as earlier. The anterior surface of the cyst wall was densely adherent to the urethra and the posterior surface of the bladder due to the chronic and infected nature of the cyst. Its position and nature were taking the diagnosis toward the urethral diverticulum with recurrent infections leading to various fibrotic bands in between. While creating planes preoperatively, the bladder had a serosa injury which was immediately repaired and later on, cystoscopy and methylene blue test confirmed the integrity of the bladder. A total of 90% of the cyst was removed and the remaining 10% of the cyst wall near the bladder and proximal part of the urethra was marsupialized. The histopathological finding of the specimen showed ulcerated lining epithelium with acute inflammatory cells infiltrates and granulation tissue (Fig. 3C). The patient was discharged and then on the follow-up, she was found to be asymptomatic.

The principles of transvaginal urethral diverticulectomy include complete removal of diverticulum wall, watertight closure of urethra, multi-layered and non-overlapping closure of surrounding tissue with absorbable suture and preservation or creation of integrity of the bladder. A total of 90% of the cyst was removed and the remaining 10% of the cyst wall near the bladder and proximal part of the urethra was marsupialized. The histopathological finding of the specimen showed ulcerated lining epithelium with acute inflammatory cells infiltrates and granulation tissue (Fig. 3C). The patient was discharged and then on the follow-up, she was found to be asymptomatic.

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**Discussion**

Vulvovaginal areas are contributed embryologically by paramesonephric ducts, urogenital sinuses, and mesonephric ducts. Skene’s and Bartholin’s glands are formed from the urogenital sinus; the mesonephric duct can persist as Gartner’s duct and non-obliteration of the canal of Nuck can also be a potential site for cyst formation along the inguinal canal and labia. Along with the embryologically prone sites vulvovaginal skin, mucosa and connective tissue are alternative possible sites for cysts due to friction during intercourse and changing activity of glands over puberty, reproductive, perimenopausal and menopausal age.2,3

Once a cyst is seen, the assessment includes attention to its location, size, numbers, consistency, tenderness, color, surface and its temperature, secretions, mobility, cross fluctuation, and impulse positivity test. Location is the first guide toward diagnosis. Along the anterior wall is the urethral diverticulum, at 1 and 11 O’clock is Gartner’s gland cyst and more lateral along the anterior wall is Skene’s gland cyst. Similarly, a posterior wall cyst can be a perianal abscess and at 5 and 7 O’clock is Bartholin’s cyst. Benign mullerian cysts can occur anywhere in the vaginal wall. The superficial cyst on the labia minora is a mucus cyst; on the labia, majora is an epidermal inclusion cyst, and in between the two lies Hidradenoma papilliform. Aggressive angiomyxoma is a soft tissue neoplasm that usually arises in the perineum mimicking a Bartholin cyst. Aggressive angiomyxomas are rare mesenchymal tumors that occur predominantly in females of reproductive age, with peak incidence typically in the fourth to fifth decades of life.4 In our first case of Bartholin cyst, the swelling was located on labia majora and mons but cross fluctuation was till the posterior wall of the vagina. Case 3 was an anterior wall cyst with pus coming through the urethra on pressing making us think of the urethral diverticulum and investigate accordingly.

Tenderness and a history of pain and fever directed toward an established infection in the cyst. Culture and sensitivity testing of the secretions in such cases must be done to guide toward the right call for antibiotics. Vulval and vaginal abscesses are usually polymicrobial, with *Staphylococcus aureus* and *Escherichia coli* being the most predominant organisms.5 Uncommonly, *Neisseria gonorrhoea* and *Chlamydia trachomatis* may also be implicated.

Most of the cyst are diagnosed on the basis of symptoms and position. Bartholin cysts usually need no further evaluation except in cases with huge sizes. Transvaginal sonography is a first-line investigation that may need to be supplemented with computed tomography (CT), MRI, and cystourethrogram in a few
cases. Postexcision histopathology is a must which may need to be supplemented with immunohistochemistry.

Management of vulvovaginal cysts depends on the age and symptoms of the patient, the risk of recurrence and malignant conversions, and the patient’s acceptability of treatment options. Recurrence is reported with all types of vulvovaginal cysts but only Bartholin, Benign Mullerian, and Gartner cysts have a tendency of malignant transformation which makes a gynecologist have a preferential bend toward surgical excision.

Bartholin cyst can be managed conservatively with yearly surveillance (if the patient is less than 40 years of age and the cyst is small in size), incision and drainage, fistulation with word catheter, marsupialization, excision or CO₂ laser vaporization. Usually, Bartholin’s gland cyst is round or ovoid, and irregular shape (such as dumbbell shape) is uncommon. This might lead to a diagnostic dilemma when physicians encounter an irregular (not round or ovoid) shape of a cystic mass in the corresponding region. The present case is unique in that, it was very large and was in a dumbbell shape.

Management is guided by the cyst dimension, symptoms, patient’s age, and history of recurrence. Perineal USG can help us find the connection between the two dumbbell areas, to know the solid and cystic nature, and study planes and Doppler flow for risk stratification for malignancy.

Urethral diverticulum diagnosis may need voiding cystourethrogram and MRI. It is treated by surgical method with or without a prior antibiotics course as per the infective status of the patient. Surgical options can be marsupialization, endoscopic transurethral incision, or urethral diverticulectomy and reconstruction followed by prolonged urethral catheterization for continuous drainage of urine and giving time for urethral epithelization. Urogynecology/urologist expert opinion should be sought as the surgical repair may be associated with complications like fistula formation, recurrence, and incontinence. Urethroscopy is a great advantage in the era of endoscopy with the help of which the number and exact site of defect/outpouching of urethra can be seen and followed later on if required.

Aggressive angiomyxoma is a soft tissue neoplasm with the term aggressive emphasizing its local infiltrative tendency and high risk of recurrence. The tumor is well known for local recurrences (so the term malignant). It is difficult to make a correct diagnosis preoperatively and the chances of misdiagnosis are between 70 and 100%. An appropriate management using a multidisciplinary approach with wide local excision, surveillance, and gonadotropin hormone-releasing hormone (GnRH) agonists are crucial to manage such patients.

**Conclusion**

In summary, based on our reviews and observations, the most important clinical parameter for vulvovaginal cyst is the site of occurrence. Later differential diagnosis is made, and investigation and treatment (surgical or medical) are planned according to the suggested approaches and ideas mentioned in the paper. Management protocols can be as straightforward as cystectomy or may require a multiprong, multidisciplinary approach and multiple sitting.

**References**