

SHORT COMMUNICATION

Unusual presentation of Bartholin's gland duct cysts: anterior expansions

Roman Rouzier, Madeleine Azarian, Françoise Plantier, Elisabeth Constancis, Bassam Haddad, Bernard-Jean Paniel

The medical records of 32 patients who underwent surgery for a vulvar mucinous cyst located between the urethral meatus and the fourchette were reviewed. Nineteen (59%) patients had been referred to our centre because of cyst recurrence after incision or excision. The majority of the cysts were lined by a single layer of tall columnar cells with zones of squamous metaplasia. Subacute or chronic bartholinitis was demonstrated in 94% of the cases demonstrating that these cysts were anterior expansions of Bartholin's gland duct cysts. No ipsilateral recurrence was observed after cyst and gland excision.

Introduction

Cystic masses of the vulva are relatively uncommon. The cysts located between the urethral meatus and the fourchette are most of the time lined by a mucinous and/or ciliated epithelium. The aetiology of mucinous vulvar cyst remains controversial. Hart has suggested that they are of paramesonephric (Mullerian) origin.¹ Others have reported that glands lined by either mucinous or ciliated epithelia are normal constituents of the vulvar vestibule and develop into cysts when the tract leading to the vulvar surface becomes obstructed.² The mucinous epithelium of these cysts is identical to that of the Bartholin's gland. The Bartholin's glands are situated deeply within the posterior parts of the labia majora. The main duct of each Bartholin's gland opens at the lateral margin of the vagina in the lower half of the vestibule. In case of Bartholin's gland duct obstruction, the gland cannot expand because of its anatomical situation. On the other hand, the duct may become cystic. Most of the time, Bartholin's gland duct cysts expand medially to the gland. Nevertheless, Bartholin's gland duct cysts may expand anteriorly and can be confused with an inguinal hernia or labial mass. The aim of this study was to review our experience with anterior expansions of Bartholin's gland duct cysts.

Methods

The medical records and follow up data of patients who underwent surgery for a Bartholin's gland duct cyst with an anterior expansion were extracted from our vulvar disease database. Cysts with a maximal transverse diameter located above the level of the urethra or below the level of the caudal part of the labia minus were excluded from this study. Subepithelial cysts associated with a delivery-related labia minus tear were also excluded. During the 10-year period from 1993 to 2003, 32 cases of vulvar mucinous cysts located between the meatus and the fourchette were observed in our centre. All records relative to demographics, medical history, presenting symptoms, treatment, pathology and outcome were reviewed. All the patients were offered cyst and Bartholin's gland excision as part of a pilot study and had surgery following informed consent. All the surgical procedures were performed by one surgeon (BJP). The surgical method was uniform during the study period and consisted of a longitudinal incision between the hymen and the labium minus. The cyst was freed medially and laterally from the vagina and from the skin of labia minora, respectively. The upper part of the cyst was identified and clamps were serially applied under the cyst to free it from the underlying tissue. When the lower part of the cyst was reached, the Bartholin's gland was left attached to the cyst and was removed carefully because of its proximity to the rectum. The pathological examination of the specimen was performed using haematoxylin and eosin staining. All the slides were reviewed by the same pathologist (FP).

Departments of Gynecologic Surgery and Pathology, Centre Hospitalier Intercommunal de Créteil, France

Correspondence: Dr R. Rouzier, Department of Breast Medical Oncology, Box 424, University of Texas MD Anderson Cancer Center, 1515 Holcombe Boulevard, Houston, Texas 77030, USA.

Results

Patient characteristics are shown in Table 1. The median size of the cysts was 4 cm (range: 2–7 cm). Vulvar tumefaction was the most frequent motive of consultation but

Table 1. Patient characteristics and histological findings.

Clinical findings	
Median age [range]	29 years [18–49]
Median parity [range]	0 [0–2]
Laterality of the median cyst, <i>n</i> (%)	
Unilateral	26 (81%)
Bilateral	2 (6%)
Unilateral + contralateral classic Bartholin's gland cyst	4 (13%)
Vulvar history	
Typical cyst/abscess of the Bartholin's gland duct, <i>n</i> (%)	19 (59%)
Mean number of previous surgical procedure for these patients	1.7 (1–7)
Previous surgical procedure before referral in our centre	
Incision and/or marsupialisation at least once	10
Cyst excision at least once	10
Cyst and gland excision at least once	4
Median size of the cyst [range]	4 [2–7] cm
Motivation for requesting surgery, <i>n</i> (%)	
Entry dyspareunia	11 (34%)
Vulvar tumefaction	13 (41%)
Discomfort when walking	3 (9%)
No symptoms	7 (22%)
Pathologic findings,* <i>n</i> (%)	
Cyst epithelium	
Columnar cells	30 (91%)
Zones of metaplasia	22/30 (73%)
Total abrasion	3 (9%)
Bartholin's gland	
No abnormality	2 (6%)
Chronic inflammation (inflammatory infiltrate and fibrosis)	21 (64%)
Subacute inflammation (inflammatory infiltrate and oedema)	10 (30%)

* Classic contralateral Bartholin's gland duct cyst were not considered, one patient with bilateral anterior Bartholin's gland cyst underwent a marsupialisation on one side.

dyspareunia was also reported by 34% of patients. The seven asymptomatic patients were offered surgery because of history of Bartholin's gland duct abscess. Six patients had bilateral disease: two patients with bilateral anterior cyst and four with classic contralateral Bartholin's gland duct cyst. Nineteen (59%) patients had undergone at least one surgical procedure for a Bartholin's gland duct cyst or abscess (incision/marsupialisation, excision, cyst and probably incomplete gland removal). In most of these cases, the origin of the cyst had not been suspected and had led sometimes to imaging such ultrasonography or MRI. The surgical procedures planned in our department were cyst and Bartholin's gland excision for all patients but one patient with bilateral disease refused to undergo bilateral gland excision and had a marsupialisation for the contralateral cyst. Mean operative time was 60 min (range: 35–110). There was no surgical complications; no patients required blood transfusion. Histological findings are shown in Table 1. Pathologic examination of the Bartholin's glands revealed a chronic

bartholinitis in 21 cases, subacute inflammation in 10 cases and no abnormalities of the Bartholin's gland in only 2 cases. Functional results were satisfactory in 27 cases but five patients reported dyspareunia lasting more than three months. None of these patient underwent additional surgery. Treatment included psychological counselling and support, topical anesthetic and for some patients tricyclic medications. Only one patient had a dyspareunia for more than one year; however, this latter patient had a contralateral recurrence. The dyspareunia resolved after a pregnancy and a vaginal delivery. With a median follow up of 36 months, no ipsilateral recurrences were observed after gland excision suggesting that cyst and Bartholin's gland excision provides a definitive treatment contrary to marsupialisation or simple cyst excision. Two patients developed contralateral Bartholin's gland cyst.

Discussion

In this study, we report that mucinous cysts located between the urethra and the fourchette may be Bartholin's gland duct cysts and that cyst and gland excision provides a definitive treatment.

Vulvar cysts located between the urethra and the fourchette may be separated in two entities. The first one is constituted by small cysts located in the labia minus, principally on its medial side, resulting from a delivery tear, or in the vestibule resulting from a previous episiotomy. The second entity comprises vulvar cysts more deeply located and topped with labia minora. This clinical difference is important because our study concerns only the second entity and our conclusion may not be extrapolated to the first group. Pathological examination cannot help to separate the two sorts of vulvar cysts: almost all the cysts in our series displayed a cylindrical epithelium and frequently squamous metaplasia. The histology of vulvar cysts has been studied to determine their embryological origin and some authors^{1,3} advocated that they could be of Mullerian while others supported a Wolffian origin, especially when located close to the clitoris or the paraurethral region.⁴ Friedrich and Wilkinson have reported a series of 20 vulvar cysts which were stained by haematoxylin and eosin, periodic acid–Schiff reagent, alcian blue and other histochemical reagents.⁵ They supported the theory that mucinous vulvar cysts arise from the urogenital sinus. Robboy *et al.*² confirmed this hypothesis and reported that glands lined by mucinous or ciliated epithelia were encountered in 9 of 19 vulvas that were examined at autopsy thus demonstrating that these glands are normal constituents of the vulvar vestibule. Concerning the embryologic origin of mucinous vulvar cyst, Mullerian origin can definitively be refuted as patients with Mullerian duct aplasia have a normal vulva. Wolffian origin of mucinous cyst has been excluded because histochemical studies of mucinous cyst of the vulva showed positive Alcian blue and Mayer's

mucicarmine staining while normal, benign and malignant structures of mesonephric origin did not.⁵ In our database, 32 cases of cyst located between the urethra and the fourchette corresponded to Bartholin's gland duct cyst while pathology revealed a different diagnosis in only three cases, meaning that anterior expansion of Bartholin's gland duct cyst should probably be considered as the most frequent diagnosis for vulvar cysts located between the urethra and the fourchette.

Nineteen patients in our series had already been operated on before referral to our centre. In all cases, the pathological examination of specimen obtained at the surgical procedure performed in our centre revealed a remnant of gland. Some form of Bartholin's gland pathology was evident in all except two cases indicating an association between the cysts and Bartholin's glands. Other supporting evidence of the association is the success of radical surgery particularly in those where prior surgery was conservative and failed.

Conservative management of Bartholin's gland duct cyst includes marsupialisation, fistulisation, aspiration, incision and drainage, use of CO₂ laser and silver nitrate insertion. Although cystic enlargement of Bartholin's gland duct is a very common condition, few data are available concerning long term outcome after treatment. The recurrence rates vary from 0% to 24%.⁶ Nineteen (59%) patients in this series had previous surgery. Excision of the cyst and the gland was indicated because these individuals had repeated episodes of recurrence after prior conservative therapy. Five patients reported dyspareunia after cyst and gland

excision in this series and one could argue that patients were over-treated. Actually, treatment of Bartholin's gland duct cyst with an anterior expansion requires extensive dissection resulting in scarring, which may by itself result in a temporary dyspareunia. As all patients had cyst and gland removal in our series, any comparison in terms of outcome between cyst and gland removal and conservative treatment is not possible. Nonetheless, we demonstrated that cyst and Bartholin's gland excision precludes recurrence and may be recommended after failure of conservative treatment.

References

1. Hart WR. Paramesonephric mucinous cysts of the vulva. *Am J Obstet Gynecol* 1970;**107**:1079–1084.
2. Robboy SJ, Ross JS, Prat J, Keh PC, Welch WR. Urogenital sinus origin of mucinous and ciliated cysts of the vulva. *Obstet Gynecol* 1978;**51**:347–351.
3. Janovski NA. Dysontogenetic cyst of the vulva. Report of a case with reference to etiologic classification of vulvar cysts. *Obstet Gynecol* 1962;**20**:227–231.
4. Novak ER, Woodruff JD. *Novak's Gynecologic and Obstetric Pathology, 5th edition*. Philadelphia: W B Saunders Co, 1962.
5. Friedrich Jr EG, Wilkinson EJ. Mucous cysts of the vulvar vestibule. *Obstet Gynecol* 1973;**42**:407–414.
6. Hill DA, Lense JJ. Office management of Bartholin gland cysts and abscesses. *Am Fam Physician* 1998;**57**:1611–1616, 1619–1620.

Accepted 5 December 2004