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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Ying-Yi Yuan*; Production Department Director: *Xu Guo*; Editorial Office Director: *Jin-Lei Wang*.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

August 26, 2023

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INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>



Hemorrhagic Bartholin's cyst in a woman using anti-platelet medication: A case report and review of the literature

Yi-Rong Li, Dah-Ching Ding

Specialty type: Medicine, research and experimental

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C, C, C
Grade D (Fair): 0
Grade E (Poor): 0

P-Reviewer: Di Donato V, Italy; Shao A, China

Received: May 3, 2023

Peer-review started: May 3, 2023

First decision: June 21, 2023

Revised: June 30, 2023

Accepted: August 3, 2023

Article in press: August 3, 2023

Published online: August 26, 2023



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Abstract

BACKGROUND

We report the case of a postmenopausal female with a hemorrhagic Bartholin's cyst who has been using an antiplatelet medication.

CASE SUMMARY

A postmenopausal woman, 84 years of age, had a medical history of hypertension, diabetes mellitus, coronary artery disease (three-vessel disease), chronic kidney disease (stage 3), and dementia. The patient has been taking clopidogrel, an antiplatelet medication, for several years. She presented at our outpatient clinic complaining of painful swelling over her left vulva for several days. A Bartholin's cyst over the left vulva was suspected, and the patient underwent marsupialization under local anesthesia, which was well-tolerated. During the incision procedure, bright-red blood with some blood clots was discharged, and a hemorrhagic Bartholin's cyst was observed. There was no recurrence of the hemorrhagic Bartholin's cyst during the 6-mo subsequent follow-up period.

CONCLUSION

Hemorrhagic Bartholin's cysts rarely occur. We report the case of a postmenopausal female with a hemorrhagic Bartholin's cyst who had been on antiplatelets and was successfully treated with marsupialization. No recurrence was noted during the 6-mo follow-up period. Older females taking antiplatelets should be cautious of bleeding when presenting with a Bartholin's cyst.

Key Words: Hemorrhagic Bartholin's cyst; Anti-platelet; Marsupialization; Menopause; Case report

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Core Tip: We report a case with a hemorrhagic Bartholin's cyst. We updated the information on Bartholin's cyst regarding symptoms, signs, diagnosis, and treatment. Because of the rarity of a hemorrhagic Bartholin's cyst, we provided a strategy to diagnose and treat this kind of Bartholin's cyst.

Citation: Li YR, Ding DC. Hemorrhagic Bartholin's cyst in a woman using anti-platelet medication: A case report and review of the literature. *World J Clin Cases* 2023; 11(24): 5755-5761

URL: <https://www.wjgnet.com/2307-8960/full/v11/i24/5755.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v11.i24.5755>

INTRODUCTION

Bartholin's glands, a pair of peanut-sized mucus-secreting glands, are located at the 4 and 8 o'clock of the vulva[1]. Mucous secretions drained *via* the Bartholin's duct provide lubrication during intercourse and keep the vulva moist. When the drainage duct of the Bartholin's gland is obstructed, a Bartholin's cyst forms[1].

The lifetime risk of developing a Bartholin's cyst is approximately 2%. It is predominantly observed in pre-menopausal women[2]. A Bartholin's cyst is typically small and asymptomatic[3,4]. However, the cyst could become more significant or infected, forming an abscess. In such cases, perivulvar pain (while walking or sitting) and systemic symptoms (such as fever or chills) will be present. A giant cyst can obstruct the urethra and cause recurrent urinary tract infections[3-5].

Various management strategies in both outpatient and inpatient settings have been proposed to treat this disease effectively, including antibiotics, topical silver nitrate application, incision and drainage (I&D), fistulization, destruction, marsupialization, and total resection in refractory cases. Nevertheless, regardless of the management strategy selected, there is still a probability of recurrence[4,6].

Although Bartholin's cyst is the most common lesion of the gland, several other lesions of the gland with either benign or malignant characteristics could be diagnosed[7]. This study reported the case of a postmenopausal female with a hemorrhagic Bartholin's cyst, which was treated successfully with marsupialization. No recurrence was observed during the 6-mo follow-up period.

CASE PRESENTATION

Chief complaints

An 84-year-old postmenopausal female presented to our outpatient clinic with a chief complaint of painful swelling over her left vulva for several days.

History of present illness

She had been taking clopidogrel (an antiplatelet medication, Sanofi Winthrop Ind., Ambares, France) for years. She denied recent fever or chills, trauma to the perineum, or previous surgical history of the perineum.

History of past illness

She had a history of hypertension, diabetes mellitus, coronary artery disease (CAD) (three-vessel disease), chronic kidney disease (stage 3), and dementia.

Personal and family history

She had 4 children (all with vaginal deliveries). Her family history was unremarkable.

Physical examination

Upon pelvic examination, a flesh-colored cystic nodule measuring approximately 2 cm × 3 cm on the left side of the vaginal vestibule was revealed. A Bartholin's cyst was initially suspected.

Laboratory examinations

There was neither leukocytosis (white blood cell count: 8410/ μ L) nor an abnormal pattern regarding the differential count of the white blood cell. The C-reactive protein level (0.18 mg/dL) was also within normal limits. Prior to marsupialization, no coagulation profiles were checked. On postoperative day 9 (6 d after resuming taking clopidogrel), coagulation profiles were as follows: Platelet count: 193000/ μ L, prothrombin time: 11.6 s, and activated partial thromboplastin time: 27.7 s. All data regarding the coagulation profiles were within normal limits.

Imaging examinations

No image examination was done.

FINAL DIAGNOSIS

A hemorrhagic Bartholin's cyst was made after marsupialization.

TREATMENT

Marsupialization of Bartholin's cyst was performed smoothly. Local anesthesia with 1% xylocaine at the cyst region was performed. Then, an incision was made with a scalpel over the cystic wall to drain the fluid and relieve the pressure. After the incision, bleeding with some blood clots from the cyst was observed. The edges of the incision were then stitched (2-0 Vicryl) to create a small opening or "pouch" that allows the cyst to continue to drain into the vaginal cavity. Compression with gauze was applied to the area after creating the pouch to stop the bleeding, which ceased after several minutes of compression. Stopping the antiplatelet for 3 d was thus suggested. After the procedure, oral antibiotics with Ulex (1st generation of cephalosporin) every 6 h were prescribed for 1 wk. The patient tolerated the procedures well. The postoperative condition of the hemorrhagic Bartholin cyst is illustrated in [Figure 1](#).

OUTCOME AND FOLLOW-UP

No recurrence of the hemorrhagic Bartholin's cyst was observed during the subsequent follow-ups for 6 mo.

DISCUSSION

Bartholin's gland embryologically originates from the urogenital sinus and is supplied by the external pudendal artery [8]. When the Bartholin's duct is obstructed, a Bartholin's cyst forms, which is a chronic inflammatory condition [1]. Based on the concept that chronic inflammation promotes angiogenesis [9,10], Bartholin's cysts would be hypervascularized and congested. Once an additional external force is present, Bartholin's cysts might be prone to hemorrhage. In this case, the patient was bedridden and used a wheelchair. These conditions are thought to result in pressure sores [11]. Similarly, limited mobility would result in the exertion of an extra external force on Bartholin's cyst, which would possibly make the cyst hemorrhagic.

Additionally, the patient has been taking clopidogrel for secondary prevention of CAD. Clopidogrel binds irreversibly to the P2Y₁₂ receptor on platelets and is a common antiplatelet medication for stroke or CAD. Bleeding is a common adverse effect of clopidogrel [12,13]. Reported risk factors for bleeding after clopidogrel use include age \geq 75 years, concurrent use of other antiplatelets or anticoagulants, prolonged use for more than 6 mo, recent trauma or surgery [14-16]. The patient presented in this case was 84-years-old, had already taken clopidogrel for several years before the first outpatient visit, and was thought to be at an increased risk of bleeding events. Based on such conditions, the hemorrhagic Bartholin's cyst might be expected.

A Bartholin's cyst is mainly a clinical diagnosis, which is based on detailed history taking and physical examinations [1, 17]. In our case, the clinical presentation led to the impression of a Bartholin's cyst. Thus, we did not arrange additional imaging examinations. The final diagnosis of a hemorrhagic Bartholin's cyst was made after the surgical intervention. Ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI) are useful tools for diagnosing a Bartholin's cyst [18,19]. For example, on ultrasonography, Bartholin's cyst is a centrally hypoechoic to the anechoic cystic lesion with a clearly defined wall [20]. On CT, the Bartholin's cyst is typically presented as a round, fluid-attenuation, and non-enhancing cystic lesion. If the cyst contains hemorrhagic components, it will become hyperdense on CT [18]. About images on MRI, the Bartholin's cyst is hyperintense on T2-weighted image, while it is hypointense without enhancement on T1-weighted image. For cysts containing hemorrhagic components, the content of the cyst becomes hypointense on T2-weighted images [18,21]. Although Bartholin's cyst is usually diagnosed simply based on clinical clues, there might be cases that are difficult to be diagnosed clinically. For such a condition, clinicians could apply the above imaging examinations.

A Bartholin's cyst commonly contains clear mucus secreted from the Bartholin's gland; in contrast, a Bartholin's abscess is filled with turbid purulent secretions [22]. In this case, bright-red blood with some blood clots flew out from the cyst, which was unusual to us. Some cases of a hemorrhagic Bartholin's cyst have been reported [19,23-25]. Details of these cases are summarized in [Table 1](#). Two of these cases (27-years-old and 39-years-old) were found to have a hematoma in Bartholin's cyst soon after the Cesarean delivery, which was thought to be a possible postoperative complication [24,25]. The other cases (42-years-old, 46-years-old, and 68-years-old) were presented with straw-colored or dark-stained discharge from Bartholin's cyst during the surgical procedure. The hemorrhagic component of the cyst was confirmed based on the pathological report in these cases [19,23]. The presentation of our case was quite different from those reported previously. Bright-red blood flowing out from the Bartholin's cyst was encountered while making an incision on the cyst. Besides, our case did not undergo a surgical intervention of the perineum before the cyst appeared.

Apart from the above, the hemorrhagic Bartholin's cyst should be differentiated from the endometriosis of the Bartholin's gland [23]. Endometriosis is characterized by the estrogen-dependent proliferation of the endometrial gland and stroma at locations other than the uterine cavity. It generally occurs in females of childbearing age. Patients with endometriosis may develop infertility or cyclic pain [26]. Extrapelvic endometriosis is relatively uncommon compared to

Table 1 Cases of the hemorrhagic Bartholin's cyst

Ref.	Age	Associated history	Location and size	Content of cyst	Treatment
Kallam <i>et al</i> [19], 2017	68	Swelling in the right labial region over the last 2 yr. The lesion was small and painless, and gradually increasing in size (especially over the last 3 mo)	A 10 cm × 7 cm × 3 cm solitary swelling over right labia majora, extending from level of pubic bones down to just above the vulval fourchette inferiorly	Nearly 500 mL of thick straw-colored fluid	Surgical excision
	42	Large swelling near the right introitus for 1.5 yr, and it grew suddenly over the last 6 d. Fever with chills and rigors 5 d prior. History of DM. No history of trauma, weight loss, or loss of appetite	A 23 cm × 11 cm mass at right labia majora; another 6 cm × 6 cm mass at right gluteal region, just below the lesion at right labia majora	Dark stained thick fluid	Surgical excision
Sengül <i>et al</i> [23], 2014	46	A painless mass in the left vulvar region for 3 yr. No dysmenorrhea, dyspareunia or chronic pelvic pain. No history of any other disease or operation	A 6 cm septate cystic lesion on the left Bartholin's gland	Dark brown fluid	Surgical excision of the cyst
Dragojević <i>et al</i> [24], 2012	39	GA 40 wk with twin pregnancy after an IVF. Full term male twins in good condition were delivered by CS. On the 3 rd postoperative day, swelling, pain and regional paresthesia of the right labia majora started. On the 7 th postoperative day, the same pattern of symptoms occurred in the left labia majora, and the symptoms were more severe than the right-side lesion	An 8 cm swelling at right labia majora; another 11 cm swelling at left labia majora	Partly fluid, partly coagulated blood	Surgical removal of the Bartholin's gland and regional reconstruction
Bacalbasa <i>et al</i> [25], 2015	27	GA 39 wk with sustained uterine contraction and quasi-complete cervical dilatation. CS was performed after a negative labor test. On the 3 rd postoperative day, apparition of two tumoral, renitent lesions with vulvar localization were reported, with mass effect on the distal vagina and anal canal. The anterior perineal region was significantly tumefied and very painful when touched	Two pseudotumoral lesions located respectively on both sides of lateral vaginal wall; the right one size 6 cm × 5 cm, the left one size 4 cm × 3 cm	The cystic content in the perineal MRI was heterogenous and hyperintense in T1	Total bilateral resection of Bartholin's glands

DM: Diabetes mellitus; GA: Gestational age; IVF: *In vitro* fertilization; CS: Cesarean section; MRI: Magnetic resonance imaging.



DOI: 10.12998/wjcc.v11.i24.5755 Copyright ©The Author(s) 2023.

Figure 1 Post-surgery condition of the hemorrhagic Bartholin's cyst.

pelvic endometriosis[27]. However, endometriosis of the Bartholin's gland is rarer. Only a few cases of endometriosis of the Bartholin's gland have been reported. These cases were all of reproductive age. Cyclic vulvar pain or infertility was complained about in some of these patients[28,29]. Compared with these cases, our patient has been postmenopausal for several decades. The lesion on Bartholin's gland did not appear long before the first outpatient clinic visit. As a result, we could differentiate the previously reported endometriosis of the Bartholin's gland from the hemorrhagic Bartholin's cyst presented in this study.

There is still controversy regarding the best treatment of a Bartholin's cyst[4,6,30]. I&D, fistulization with a Word catheter, marsupialization, and excision of the cyst are all commonly performed procedures[31,32]. I&D is the simplest way to treat Bartholin's cyst, and it has the shortest recovery time due to its simplicity. However, the high recurrence rate is a shortcoming compared to other invasive treatments[3]. Word catheter fistulization is performed by placing the catheter into the incision wound of I&D, and then the balloon is inflated with 2 to 3 mL of saline. To ensure the future patency of the outflow tract, the catheter will remain in place for approximately 4 to 6 wk for the tract to be completely epithelialized[3,4]. Marsupialization is the preferred treatment for recurrent cases. An incision opens the cyst with a

length of the entire cyst wall. After drainage and irrigation, the cyst wall and mucosa are sutured open with absorbable sutures by an interrupted method. Marsupialization could prevent the incision from closing and keep the duct patent for ongoing drainage[3,4]. Excision of the cyst is more commonly suggested in recurrent cases, patients aged over 40-years-old, or patients with lesions larger than 5 cm[4]. A previous study reported that none was superior to the others regarding recurrence rate[6]. Regarding complications following the above interventions, pain, hematoma, fever, scarring, and persistent dyspareunia were all reported. The above interventions generally had a comparable complication rate as the others, except the finding that marsupialization had an increased risk of persistent dyspareunia compared to excision of the cyst. But there was only limited evidence in this systematic review[6]. Among these interventions, fistulization with a Word catheter and marsupialization are the two most commonly applied procedures[31,33]. Previous studies reported that both interventions had comparable recurrence rates[31,33]. Also, a comparable postoperative complication rate was found among these two interventions[31,33]. Intriguingly, a recent retrospective cohort study by Karabük and Ganimi Aygün[34] demonstrated that marsupialization had a significantly lower recurrence rate than fistulization with a Word catheter (8.3% *vs* 18.8%; $P = 0.034$). However, there was no statistical difference regarding comparing the postoperative complication rate among these two groups (marsupialization *vs* fistulization with a Word catheter: 5.3% *vs* 3.1%; $P = 0.495$; all presented with infection)[34].

For recurrent cases, cystectomy or marsupialization may be preferred[3]. However, there is limited evidence to compare these two procedures directly[6]. Because no single therapy has been proven to outweigh others, therapeutic plans should be personalized based on lesion size, presence of symptoms, or recurrence to prevent a future recurrence[6, 34].

In a nutshell, our case demonstrated that marsupialization for a hemorrhagic Bartholin's cyst was feasible. A limitation of this study is that only a single case was presented. Restricted generalizability should be considered. However, further evidence is required to confirm these hypotheses.

In addition to endometriomas of Bartholin's cyst, hemorrhagic Bartholin's cysts are rare events. We report the case of a postmenopausal female on an antiplatelet medication with a hemorrhagic Bartholin's cyst who was successfully treated with marsupialization. No recurrence was observed during the 6-mo follow-up period. Older females taking antiplatelets should be cautious of bleeding when presenting with a Bartholin's cyst.

CONCLUSION

In addition to endometriomas of Bartholin's cyst, hemorrhagic Bartholin's cysts are rare events. We report the case of a postmenopausal female on an antiplatelet with a hemorrhagic Bartholin's cyst who was successfully treated with marsupialization. No recurrence was observed during the 6-mo follow-up period. Older females taking antiplatelets should be cautious of bleeding when presenting with a Bartholin's cyst.

FOOTNOTES

Author contributions: Li YR contributed to the methodology, formal analysis, data curation, writing and original draft preparation of this manuscript; Ding DC contributed to the conceptualization and writing, review and editing of this paper; and all authors have read and agreed to the published version of the manuscript.

Informed consent statement: Written informed consent was obtained from the patient(s) to publish this paper.

Conflict-of-interest statement: All the authors report no relevant conflicts of interest for this article.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Country/Territory of origin: Taiwan

ORCID number: Dah-Ching Ding 0000-0001-5105-068X.

S-Editor: Wang JJ

L-Editor: Filipodia

P-Editor: Wang JJ

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