Ductal-cutaneous fistula secondary to recurrent Bartholin's cysts: a case report

Danielle M. Ikoma,[1] Sarah A. Shaffer[1]
Keywords: Bartholin's gland, recurrent Bartholin's cysts, fistula

Abstract

Background: Disorders of the Bartholin's duct and gland, including cyst and abscess formation, account for 2% of gynecologic visits annually. An uncommon complication of a Bartholin's duct or gland abscess is fistula formation. Literature has described cases of recto-Bartholin's and recto-vaginal fistulas.

Case: We present a case of fistula development between the perineum and the Bartholin's duct and gland. The patient was successfully managed with fistulectomy and Bartholin's gland excision.

Conclusion: Though fistula formation is a rare complication of Bartholin's duct and gland pathology, investigation is warranted. A ductal-cutaneous fistula is possible in the setting of recurrent cysts located beyond the vaginal introitus. The best method of prevention is appropriate execution of a marsupialization. Complete removal of the fistulous tract and Bartholin's duct and/or gland can result in resolution of symptoms.

[1]Department of Obstetrics and Gynecology, University of Iowa Hospitals and Clinics, Iowa City, Iowa

Introduction

Bartholin's glands, first described by Casper Bartholin in 1677, are bilateral glands located at the base of the labia minora and function to produce mucinous fluid for lubrication at the vaginal introitus.[1-4] The Bartholin's gland structure originates from the urogenital sinus during development and differentiates into three parts: the gland (comprised of mucinous columnar acini), the duct (comprised of transitional epithelium), and the orifice or os (comprised of squamous epithelium).[1-3]

Disorders of the Bartholin's gland and duct comprise 2% of all gynecological visits annually.[5] Pathology results from blockage of the orifice and distal duct, leading to cyst and abscess formation of the duct.[2-5] Bartholin's duct cysts, located at either the 4 or 8 o'clock position relative to the introitus, are generally small (1-3 centimeters), slow

Please cite this paper as: Ikoma DM, Shaffer SA. Ductal-cutaneous fistula secondary to recurrent Bartholin's cysts: a case report. Proc Obstet Gynecol. 2022;11(2): Article 1 [10 p.]. Available from: https://pubs.lib.uiowa.edu Free full text article.

Corresponding author: *Danielle M. Ikoma, Department of Obstetrics and Gynecology, University of Iowa Hospitals and Clinics, 200 Hawkins Drive, Iowa City, Iowa. USA. 52242. Email: danielle-ikoma@uiowa.edu*

Financial Disclosure*: The authors report no conflict of interest.*

growing, asymptomatic, and, therefore, do not require treatment.[1,3,4,6] However, larger cysts may develop leading to symptoms of vulvar pain, dyspareunia, and inability to comfortably perform certain activities such as walking and sitting.[3,4,7] Abscess formation may also occur, leading to more severe symptoms and associated physical exam findings of erythema and edema.[1,3-5]

Cysts and abscesses may intermittently form, then drain spontaneously. In these cases, conservative measures such as warm soaks and frequent rinsing are routinely recommended.[1,3,5] On the other hand, persistent symptoms are typically managed with procedural interventions including: aspiration, incision and drainage (I&D), silver nitrate sclerosis of the duct, fistulation of the duct with Word catheter placement or with marsupialization, fenestration, ablation or excision of the enlarged duct using laser therapy, or surgical excision of the Bartholin's gland. Based on available evidence including a systematic review in 2020, assessing recurrence and healing following treatment of Bartholin duct cysts and abscesses, no superior treatment approach has been conclusively identified; however, it has been suggested that Word catheter insertion be first line treatment.[5,8,9] In addition, aspiration and I&D alone are typically not encouraged due to high rate of recurrence ranging from 13-38%.[3-5] Recurrence rates for other treatment modalities have been reported as 2-17% for Word catheter placement, 3-25% for marsupialization, 8.6% for CO2 laser therapy, and 3.7% for Bartholin's gland excision.[1,3,5] The latter suggests that, in many cases, the gland was not completely resected.

A rare complication of recurrent Bartholin's duct cysts and abscesses is fistula formation.[1] We have identified eight cases previously reported in literature describing fistula development after a history of either chronic or recurrent Bartholin duct and gland abscesses.[10-15] Four cases describe a recto-Bartholin's fistula; however only one of these cases attributes the fistula to pathology arising from the Bartholin's gland and duct.[10,12] The other four cases describe formation of a recto-vaginal fistula that developed after recently being diagnosed with a Bartholin's gland abscess.[11,13-15] We present a case of fistulous tract formation between the perineum and Bartholin's duct and gland.

Case Description

A 35-year-old G0P0 presented to clinic due to recurrent bilateral Bartholin's duct cysts. She reported four I&D procedures performed over the past 12 months on the right side and marsupialization procedure performed seven years prior on the left. She also noted history of Bartholin's duct abscess on the right two years prior, requiring I&D and antibiotic treatment. However, the patient reported bilateral cysts would often form and resolve spontaneously, without the need for a procedure. Her past medical history was significant for recurrent vaginitis, anxiety, depression, and history of choledochal cyst status post Roux-en-Y hepaticojejunostomy three years prior.

External genital examination at her initial

visit to a specialty Vulvo-Vaginal Disease (VVD) Clinic was notable for a fluctuant, non-tender, non-erythematous, four-centimeter cyst of the left labia located at the level of the posterior fourchette, consistent with Bartholin's duct cyst. Scarring from prior I&D procedures was appreciated on the right perineum, just posterior to the right labia majora. The patient opted for expectant management at that time. Follow-up five months later revealed a persistent left Bartholin duct cyst as previously described, as well as interim development (recurrence, per the patient) of a tender, non-erythematous, fluctuant, three-centimeter cyst located at the level of the posterior right perineum (Figure A). It was recommended she undergo excision of right perineal cyst and marsupialization of the left cyst, which was performed the next day as an in-office procedure under conscious sedation coupled with local anesthetic.

During the procedure, incision of both cysts revealed sticky, gelatinous, straw-colored material consistent with Bartholin's gland fluid. No purulent-appearing or foul-smelling drainage was appreciated. An uncomplicated marsupialization of the left Bartholin's duct cyst was performed. The space within the right-sided cyst was noted to track from the right perineum to the inner right hymen just deep to the ostia of the right Bartholin's duct. This suggested fistula formation between the Bartholin's duct and the perineum. The mucosal lining of the fistula was dissected circumferentially from the perineal opening toward the right lateral wall of the vagina. There was concern, however, that some amount of the mucosal lining remained as the patient was uncomfortable despite pain control measures. The space was then closed with layers of interrupted sutures. Pathology described a benign cyst lined by squamotransitional epithelium with occasional cilia, suggesting involvement of the Bartholin's duct.

Figure A: Exam prior to in-office marsupialization and cyst excision showing a Bartholin's duct cyst at the left labia minora, as well as, a tender, fluctuant, cyst located at the level of the right perineum

A visit one-week after the procedure revealed small openings of the bilateral surgical site incisions, consistent with recent marsupialization procedure on the left and superficial wound dehiscence on the right. Otherwise, the bilateral surgical sites were noted to be healing well overall with no signs of

infection (Figure B). However, at her two-month follow-up visit, there was concern for reformation of the fistula on the right due to development of a tender fluctuant swelling over the prior surgical site. She was subsequently scheduled for complete removal of the abnormal tract in the operating room under general anesthesia which was arranged approximately five months after the prior procedure.

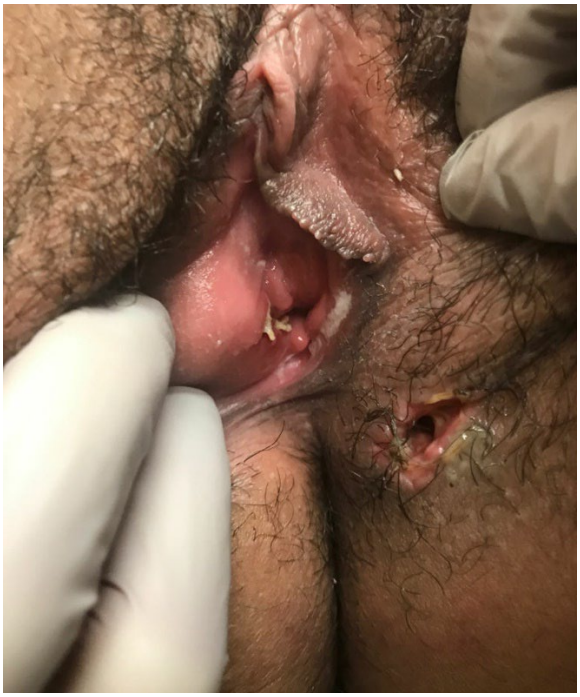

Figure B: One-week status post in-office marsupialization of left Bartholin's gland cyst and excision of right perineal cyst, showing small openings of prior surgical site incisions consistent with marsupialization procedure on the left and superficial wound dehiscence on the right

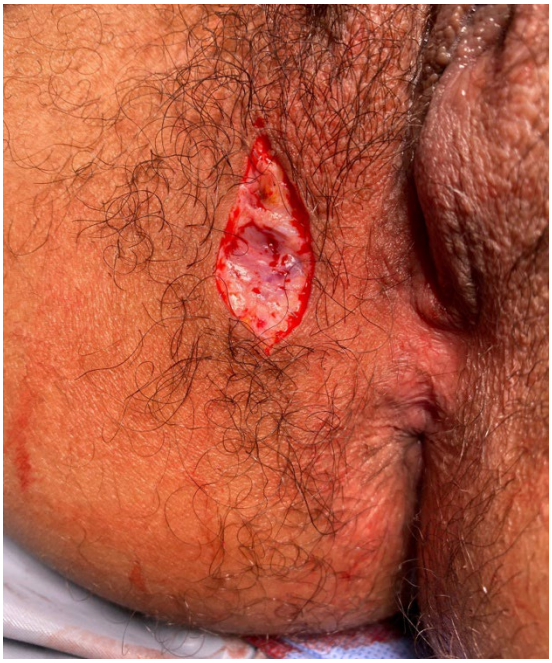

Figure C: Exam under anesthesia in the operative room revealed scar tissue of the right posterior labia majora from the prior in-office procedure, which was removed with an elliptical incision

A cystic structure was identified immediately below the skin and ruptured with return of clear, sticky, non-malodorous fluid. Probing of the cystic structure revealed a tract lined with smooth mucosa that led to an area of palpably fibrous tissue within the region of the right Bartholin's duct, likely representing a ductal-cutaneous fistula (Figure D).

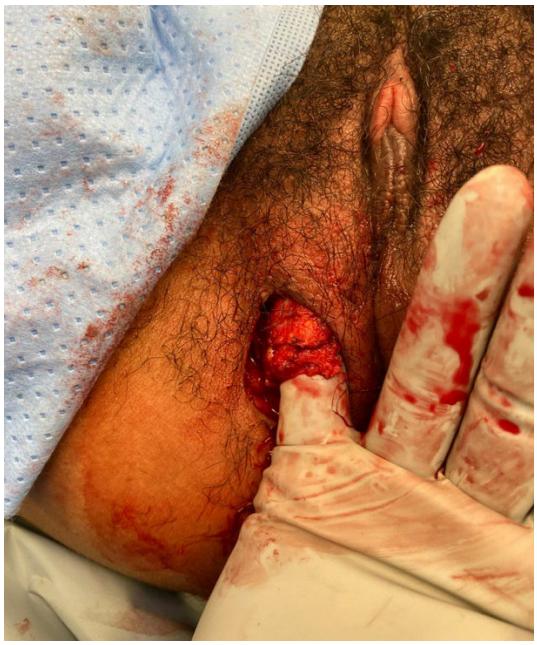

Figure D: Probing of the right perineal cyst revealed a fistulous tract to the Bartholin's duct

Figure E: Traction was applied to the epithelium of the fistulous tract to aid in sharp dissection.

Traction was applied to the mucosal tissue to sharply dissect out the fistulous tract (Figure E) until the entire tract was removed, including tissue thought to be the right Bartholin's gland (Figure F).

Figure F: Defect after complete removal of fistulous tract, including the Bartholin's gland

Transposed subcutaneous tissue was used to fill the deep portion of the defect; a Martius flap was considered, but ultimately not performed. The deep defect was closed with several layers of 3-0 chromic suture in a running fashion, followed by interrupted sutures of 3-0 Vicryl to reapproximate the skin. The procedure was concluded after a repeat marsupialization of the left Bartholin's duct cyst (Figure G). Pathology returned with cyst wall fibrosis, chronic

inflammation, and foreign body giant cell reaction. Further discussion with the pathologist confirmed that glandular tissue was also identified.

Figure G: Final result after primary closure of the right fistulectomy and Bartholin's gland excision, and marsupialization of the left Bartholin's cyst.

Follow up at the patient's one-month postoperative visit revealed a well-healed, three-centimeter scar of the right perineum and healing wound of the left vaginal introitus, consistent with recent marsupialization (Figure H).

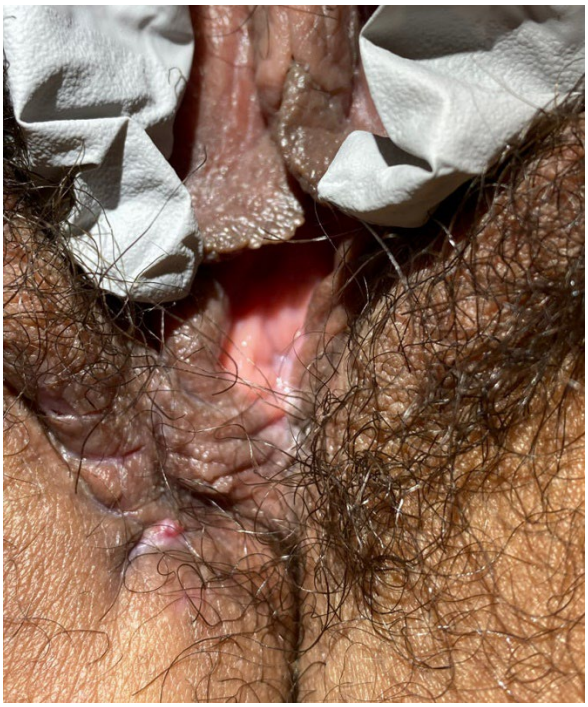

Figure H: One-month status post fistulectomy, right Bartholin's gland excision, and marsupialization of left Bartholin's cyst showing well-healed incisions and resolution of symptoms.

Discussion

Bartholin's duct cysts and abscesses are conditions of the Bartholin's gland that result from obstruction of the ostia which serves to drain the duct.[1-5] A rare complication from these disorders is fistulous tract formation, as published in eight prior case reports describing recto-Bartholin's and recto-vaginal fistulas.[1,10-15] However, our case report demonstrates a fistula may also form between the Bartholin's duct and the surface of the body, albeit an aberrant location, to allow drainage of the Bartholin's gland secretions. Risk factors for fistula formation in the pelvis

include obstetric complications, vaginal trauma, recent pelvic surgery, inflammatory bowel disease, radiation, and cancer.[13,15] Bartholin's duct abscesses are an extremely rare risk factor for fistula formation, but conceivable due to localized inflammation, pressure upon mucosal structures with basic activities like walking, sitting, and shifting positions in the presence of a swollen abscess, and alteration of tissue integrity with recurrent lesions.[1,12-15] It is unknown whether recurrent Bartholin's duct cysts without abscess formation increase the risk of developing a fistula, although our case suggests this etiology.

There are a number of vulvar lesions that may mimic a Bartholin's gland cyst or abscess. These include cystic lesions such as: epidermal inclusion cysts, mucinous cysts of the vestibule, cysts of the canal of Nuck, Skene's duct cysts, papillary hidradenomas, and hematomas.[3,4,7,16,17] Epidermal inclusion cysts are common asymptomatic cysts that arise from obstructed hair follicles leading to a small 2-5mm papule.[3,4,16,17] Mucinous cysts of vestibule arise from the minor vestibular glands and can range in size from millimeters to several centimeters.[3,16,17] Cysts of the canal of Nuck are usually located at the superior aspect of the labia minora.[16] They are composed of the process vaginalis, a herniating peritoneal sac that accompanies the round ligament and often resembles an inguinal hernia.[16,17] Unlike epidural inclusion cysts and mucinous cysts of the vestibule, they can exceed over 5 cm in size.[4,16,17] Skene's glands are similarly located superior to the vaginal introitus but are found medially at the right and left of the urethral meatus.[16] They secrete a small amount of mucous material and can lead to cyst formation if blockage of the duct occurs.[16,17] Papillary hidradenomas are benign, cystic, papillary tumors thought to arise from the anogenital glands.[4,18] They are often slow growing solitary lesions and have a tendency to ulcerate.[4,18] Hematomas are often preceded by trauma such as straddle injuries or abuse.[3] Treatment is not warranted for the above-mentioned cystic lesions if they are asymptomatic or small in size.[16,17] However, if infection, irritation, or increasing size is noted, treatment is usually necessary.[3,16-18]

Differential diagnosis should also include solid lesions including fibromas, lipomas, leiomyomas, and squamous cell carcinoma.[3,4,16,17] Fibromas are a benign solid tumor of the vulva, normally arising from the deep fibrous tissue of the introitus, round ligament or perineal body.[19] Lipomas are benign, slow-growing lesions usually seen on the labia majora and contain mostly mature fat cells and connective tissue.[3,16,17] Similarly, leiomyomas of the vulva are benign smooth muscle tumors that most often arise on the labia majora.[4,16] Consideration for excision of lipomas, fibromas, and leiomyomas include pain, rapid growth, or cosmetic indications.[3,16] Squamous cell carcinoma of the vulva may present as a lump or ulcerated lesion, usually with a long-standing history of vulvar symptoms.[20] Any suspicious vulvar lesion should be biopsied, regardless of size.[3,20]

Management for symptomatic Bartholin's duct cysts and abscesses ranges from expectant management to

surgical procedures including aspiration, I&D, silver nitrate sclerosis of the duct, fistualization of the duct with Word catheter placement or marsupialization, carbon dioxide laser therapy (e.g., fenestration or duct ablation), and surgical excision of the Bartholin's gland. Antibiotic therapy is not routinely recommended and is only indicated in the setting of cellulitis of the region.[7,21] However, it is commonly added to treatment regimens in setting of infected cysts (abscesses), pregnancy, immune-suppressed or immune-compromised state, or as an adjunct to surgical management, although an unnecessary one.[1,3,5,7,21,22] Cultures of Bartholin's gland abscesses often return as polymicrobial with predominance of aerobic etiologies.[5,7] Currently there is lack of evidence to suggest a superior treatment method to reduce risk of recurrence and complications.[5,8] However, a recent literature review recommends insertion of a Word catheter be first line treatment due to ability to perform the procedure in an in-office setting, avoid general anesthesia, and reduce overall resource utilization.[9]

Management of a fistula that develops as a result of Bartholin's gland and duct pathology will require consideration of the type of fistula (i.e., the structures that improperly communicate), prior treatment history, co-morbid conditions, and patient preference. In general, fistulectomy by a variety of surgical approaches, with or without removal of the Bartholin's gland, was the treatment of choice in our review of the literature and led to complete resolution of symptoms in 6 out of 7 cases [10-15]. In this report, we describe fistulectomy and Bartholin's gland excision followed by primary closure. Pathologic evaluation was used to confirm excision of the Bartholin's gland as well as the absence of malignancy. At time of composition of this case report, our patient denied recurrence of symptoms 12 months after the second procedure described.

The strength of this report is the ability to describe surgical management of a rare complication of Bartholin's gland cysts in a step-wise fashion with the aid of illustrations. We acknowledge, however, that due to the rare nature of fistula formation secondary to recurrent Bartholin's gland cysts, there remains limited data on prevention, standard of care, and post-treatment management.[8,9] In general, benefits of surgical excision of the fistulous tract and Bartholin's gland duct include decreased risk of recurrence.[10-15] This approach, conversely, carries some disadvantages such as the requirement for deep sedation in an operating room setting, increased resource needs and costs, and risks of post-operative complications such as transient increase in pain, wound dehiscence, infection, and recurrence.[9]

Conclusion

Our manuscript illustrates that management of Bartholin's gland cysts with standard procedures such as I&D and marsupialization can still lead to recurrence, abscess formation and, in rare cases, fistula formation. Recurrent vulvar cysts with or without spontaneous drainage occurring beyond the 4 or 8 o'clock position around the vaginal introitus, as typically seen in Bartholin's gland and duct conditions, warrant investigation into a possible ductal-

cutaneous fistula. Complete removal of the fistulous tract and abnormal Bartholin's duct, with or without the gland, in an operating room has been shown in a small number of reports to result in complete resolution of symptoms. This surgical management also seems to prevent recurrence of not only the fistula, but the original Bartholin's duct cyst or abscess. Pathological analysis is necessary to confirm removal of desired tissue, especially if removal of the Bartholin's gland is a surgical goal.

Conflict of interest statement: The authors declare no conflict of interest.
Funding: This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.
Approval: Written permission from the patient described in this case report was obtained.

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