



## Dyspareunia Secondary to a Clitoral Pilonidal Sinus: A Case Report and Literature Review

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### Abstract

**Introduction:** Dyspareunia is a general term used to describe painful sexual intercourse. It can occur due to a variety of reasons ranging from structural problems to psychological concerns. Dyspareunia frequently occurs due to pelvic inflammatory disease, endometriosis, absence of lubrication, and previous trauma. Additionally, eczema, hidradenitis, or other skin diseases of the vulva may cause pain with intercourse. Pilonidal sinus is a common skin disease presenting with hair-nests and sinus tracts occurring in deep skin folds such as the inter-gluteal sulcus.

**Case Presentation:** A 30-year-old dark-haired Asian housewife (gravida 1, para 1) was seen at the general surgery outpatient clinic with a complaint of a foul-smelling thick umbilical discharge as well as dyspareunia. Dyspareunia due to a clitoral pilonidal sinus can be successfully cured with early diagnosis, appropriate antibiotics, and wide surgical excision of the lesion. Delay in diagnosis and treatment is associated with the development of complications such as abscess formation; it also prolongs the time to definitive treatment and affects the prognosis of the disease. In cases of mons pubis swelling, vulvar pain, or unexplained dyspareunia in menstrual aged women, clitoral pilonidal sinus should be kept in mind as a potential cause.

**Conclusion:** Herein, we present the case of a young woman with dyspareunia secondary to a clitoral pilonidal sinus.

**Keywords:** Clitoris, Pilonidal sinus, Dyspareunia

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### Introduction

Dyspareunia is a general term used to describe painful sexual intercourse (1). It can occur due to a variety of reasons ranging from structural problems to psychological concerns. Up to one-third of women describe having painful intercourse at some point in their lives. However, its incidence

and prevalence vary depending on culture, age, outcomes measured, and definition of sexual pain. Dyspareunia frequently occurs due to pelvic inflammatory disease, endometriosis, absence of lubrication, and previous trauma (2). Additionally, eczema, hidradenitis, or other skin diseases of the vulva may cause pain with intercourse.

Pilonidal sinus is a common skin disease presenting

with hair-nests and sinus tracts occurring in deep skin folds such as the inter-gluteal sulcus (3). However, clitoral involvement is extremely rare in patients with pilonidal sinus disease; there are only a few cases described in the literature (4). Therefore, the threshold for suspicion among clinicians is high and its diagnosis and treatment can be delayed in clinical settings.

Herein, we present and discuss the case of a young woman with dyspareunia secondary to a clitoral pilonidal sinus.

### Case Presentation

A 30-year-old dark-haired Asian housewife (gravida 1, para 1) was seen at the general surgery outpatient clinic with a complaint of a foul-smelling thick umbilical discharge as well as dyspareunia. She had these symptoms intermittently for one year. She was prescribed multiple courses of antibiotics and topical ointments by her gynecologist, all of which failed to treat the condition permanently. No underlying gynecologic pathology was found to explain the ongoing dyspareunia. She was referred to the general surgery clinic for the umbilical discharge.

Her past medical history included impaired glucose tolerance that required dietary modifications and regular exercise. However, she admitted not being compliant with such conservative measures. Her past surgical history was notable for the surgical removal of an inter-gluteal pilonidal sinus. Her menses were regular and pain-free with light to medium flow. She had no prior psychiatric disorders.

She had normal vital signs. Her body mass index was calculated as 26.2 kg/m<sup>2</sup>. The physical examination revealed a well-healed surgical scar in the inter-gluteal region together with a foul-smelling umbilical discharge and umbilical swelling. Additionally, there was a small granuloma over the mons pubis, which was swollen and tender. During the physical examination, the patient admitted that the pubic pain and tenderness was more bothersome for her than the umbilical discharge.

### Surgery

Surgery was planned to excise the umbilical pilonidal sinus and the granuloma on the mons pubis. Since her main complaint was severe pain at the granuloma site, we initially focused on the pubic lesion. An elliptical skin incision was made by leaving a 5-mm margin from the edges of the protuberant granuloma. This incision was deepened toward the subcutaneous fat. During the dissection, several hair follicles and sinus tracts were detected. We carefully continued dissecting these sinus tracts and created a hole under the skin. Our dissection ended at the clitoral hood where there were tiny openings of the sinus tracts (Figures 1, 2). These sinuses contained bundles of hair and infected epithelized tracts. Subsequently, the initial incision was extended to



**Figure 1:** Clitoral hood can be seen beneath the subcutaneous tunnel.



**Figure 2:** The granuloma located on the mons pubis and hair containing sinus tracts have been dissected from the initial elliptical incision on mons pubis. This dissection created a subcutaneous tunnel to the clitoral hood. The surgeon's finger is in this tunnel.

a midline incision to achieve complete excision of the sinus tracts (Figure 3). The clitoral body was not primarily involved in the inflammatory process. Therefore, we were able to preserve it by excision and debridement of the adjacent inflamed tissue.

An intra-operative diagnosis of a clitoral pilonidal sinus was attained. Peri-clitoral granulomatous tissue was excised until healthy connective tissue appeared. The resultant tissue defect was closed with minimal tension using interrupted 2/0 Prolene stitches (Figure 3). The specimen was sent to the pathology laboratory for definitive diagnosis.

### Pathology

The histopathological evaluation confirmed the preliminary diagnosis of clitoral pilonidal disease. Pathology sections showed hair-bearing skin with sinus tracts containing clusters of neutrophils, plasma cells, and multinucleated giant cells.





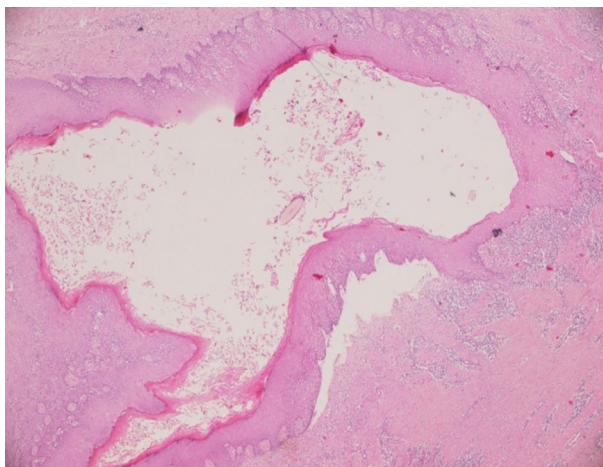
**Figure 3:** The elliptical incision on the mons pubis was extended to the midline horizontally towards the clitoral hood. This incision was closed with interrupted stitches.

(Figures 4, 5). There was also fibrotic tissue with plump fibroblasts and granulation accompanied by ulceration of the overlying epithelium (Figures 4, 5). The tracts rarely had squamous linings surrounded by chronic inflammatory cells. There were bundles of hair attached with mucus. No nuclear atypia was observed.

#### *Outcome and Follow-Up*

The patient was followed at the surgery clinic after the surgery. On postoperative day 3, superficial dehiscence, maceration, and serous discharge were observed at the wound. Two stitches were removed and the wound was left open to heal by secondary intention. A quick recovery was observed during daily dressing changes. On postoperative day 15, the wound was found to have healed completely with minimal scarring.

Six weeks after surgery, the patient was allowed to have sexual intercourse. During the sixth postoperative month, she reported a complete



**Figure 4:** Sinus tract with hair

resolution of the dyspareunia. She denied any pain or discomfort during sexual intercourse and was satisfied with the outcome of the surgery. For the umbilical pilonidal disease, the patient underwent a separate surgery one year after the clitoral procedure. This surgery provided a resolution to her complaint of umbilical discharge. During the umbilical pilonidal disease surgery, the clitoral surgery site was examined for recurrence and was found to be indistinguishable from pre-existing skin with almost no scarring.

#### *Method for Literature Review*

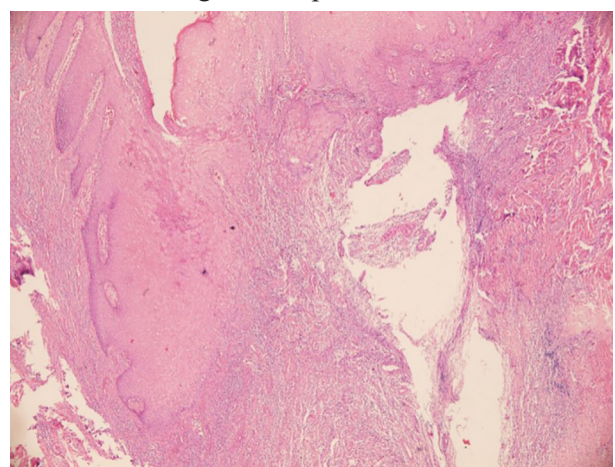
Literature published in English was scanned using Pubmed and Google Scholar. Publications from January 1, 1950, to April 1, 2020, were searched using the terms “clitoral”, “peri-clitoral”, “pilonidal sinus”, and “pilonidal cyst” in various combinations. All reports that included a description of a pilonidal sinus of the clitoris were included.

Case reports related to ingrown hair follicles accompanied by foreign body reaction and inflammation located at the groin area without the involvement of the clitoral region were excluded.

#### **Discussion**

The pilonidal disease of the inter-gluteal area is a common condition of the skin. Its incidence is about 26 per 100,000 people. Clitoral involvement with pilonidal disease was first described by Thwaite in 1957; since then, only a few cases have been reported in the literature (5). Its incidence has increased relatively during the last decade, probably due to higher rates of reporting and changes in shaving practices.

Initially, pilonidal disease was thought to be congenital in origin; however, this has been disputed when identical acquired lesions were described in barbers' hands (6, 7). There may be an immunological component to the disease as well, creating a tendency to initiate a foreign body-type reaction to hair. In line with this hypothesis, which is based on the presence of an immunological component, once a reaction to



**Figure 5:** Ulceration of the epithelium in the sinus tract and granulation tissue (Hematoxylin Eosin stain, x4)

hair occurs in one part of the body, the likelihood of it being observed in other body parts increases. Our patient demonstrated this clinical picture as she had pilonidal sinus disease in three different anatomical locations; inter-gluteal, umbilical, and clitoral. The reaction to hair in the case of a pilonidal sinus bears similarities with type 4 delayed hypersensitivity, involving plasma cells, giant cells, granuloma formation, and chronic inflammation.

Altogether, 12 clitoral pilonidal sinus cases have been reported in the literature and none of these patients were noted to have dyspareunia as their chief complaint (Table 1). To the best of our knowledge, our case represents the first case reported in the literature where dyspareunia was due to clitoral pilonidal sinus. Most patients discussed in the literature had pain, swelling, or purulent discharge from the mons pubis as the main complaints (4, 8-14).

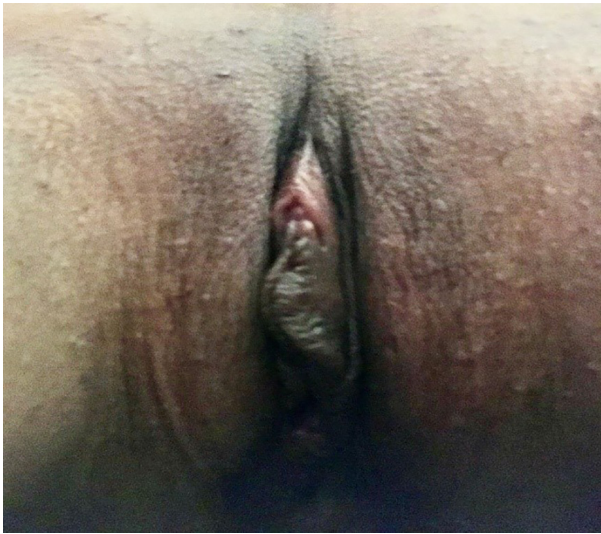
The pilonidal disease of the clitoris is prominent with a granulomatous lesion located at the mons pubis (4, 5, 8). This pubic lesion is connected via sinus tracts to the peri-clitoral area or clitoral shaft (Figure 6). These sinus tracts contain bundles of hair but not hair follicles. Due to recurrent infections and purulent discharge from these sinuses, patients usually undergo either drainage or simple excision procedures followed by multiple courses of antibiotic treatments. However, once the diagnostic certainty is obtained, a wide surgical excision involving the peri-clitoral area can provide the definitive cure (8, 15).

Wide surgical excisions have been performed using several techniques (13, 15). Palmer et al. and Werker et al. utilized tools such as surgical probes or dyes to explore the sinus tracts and determine the width of the excision (8, 11). Some surgeons opted to obliterate the anatomical sulcus to prevent recurrences using

**Table 1:** Clinical data of the previously-reported clitoral pilonidal sinus cases

Author, Year	Age	Presenting Symptom	Comorbidity	Time to Definitive Treatment	Surgery	Peri-clitoral Disease	Skin Closure	Recurrence
Thwaite, 1957	17	Pain, Swelling	None	11 months	Excision	Present	Primary	None
Palmer, 1957	29	Pain, Swelling, Discharge	None	13 months	Midline incision from Mons Pubis to tip of clitoris with help of a probe. Wide excision deepened down to the fascia	Present	Primary	None
Betson, 1962	29	Pain, Swelling	None	4 months	Wedge shaped block excision including the clitoris and upper vulva	Present	Primary	None
Radman, 1972	22	Swelling	None	36 months	Excision of the sinus tracts	Present	Primary	None
Werker, 1990	23	Suppurated Infection on Mons Veneris	None	12 months	Bonney's Blue injection to fistula tract, followed by excision	Present	Primary	None
Baker, 2008	30	Peri-clitoral Abscess	None	24 months	Wide local excision extended around the hood of clitoris removing the prepuce	Present	Primary	None
Kafali, 2008	30	Pain, Swelling	None	48 months	Asymmetric excision and closure using Karydak's procedure	Present	Primary	None
Maor-Sagie, 2010	8	Pain	Premature Birth	3 months	Excision including a sinus tract leading to the Mons Pubis, clitoris was preserved and left minor labium reconstructed	Present	Primary	None
Kanis, 2014	33	Pain, Swelling, Drainage	None	9 months	The entire sinus tract excised with cystic bed Required a second procedure for clitoral hood revision	Present	Left open	None
Kushwa-ha, 2015	63	Peri-clitoral Bleeding and Discharge	Bone Marrow Transplant	1 month	Surgical excision	Present	Primary	None
Stenson, 2018	29	Recurrent Clitoral abscess	None	36 months	Surgical excision	Present	Primary	None
Stenson, 2018	27	Peri-Clitoral Pain	None	N/A	Marsupialization	Present	Left open	N/A





**Figure 6:** Appearance of the clitoris one year after the peri-clitoral excision of the pilonidal sinus

the Karydakís flap technique (13). Others performed similar surgeries by making a horizontal skin incision from the mons pubis to the clitoris and excised the sinus tracts (4-6, 8-16). In some cases, the surgeons had to include the clitoral body in the specimen due to its involvement by the pilonidal sinus disease (9, 12, 15). However, excision of the clitoral body was found to be associated with sexual dysfunction (9, 12). In our case, we were able to preserve the clitoral body; the patient's sexual function was restored completely in the postoperative period.

Our literature review revealed that skin closure after the surgical excision has been performed using similar techniques; all but one excision site was left open (15). Kanis et al. left the surgical site open to prevent a concurrent infection (15). However, they had to reconstruct the separated clitoral hood afterward in a second surgical intervention. Likewise, our patient experienced wound dehiscence probably due to the tension at the skin edges. After the removal of three stitches, the wound was left open to allow secondary healing. Fortunately, our patient's wound healed well and no additional surgery was required. No recurrence was observed after the definitive surgical treatment in the previously reported cases.

As shown in Table 1, the time to diagnosis and definitive treatment for clitoral pilonidal disease have both been relatively long; several cases reported gaps of more than two years with intermittent

symptoms of purulent drainage and tenderness (10, 12, 13, 16). Hence, clitoral pilonidal disease is challenging to diagnose and the granuloma on the mons pubis can be confused with other skin conditions such as Skene duct cysts, sebaceous abscesses, or cellulitis (13-15).

This difficulty in diagnosis can partially be attributed to the lack of knowledge and scarcity of reports in the literature. Additionally, its apprehensive location and recurrent nature might make it difficult for the patient to address the disease at a doctor's appointment. For clinicians, knowing how to spot certain risk factors for pilonidal disease is important for early diagnosis. These risk factors include chronic trauma, irritation, and frequent shaving using improper techniques. Additionally, obesity, immobility, family history, as well as a personal history of the disease should be considered.

Clitoral pilonidal disease mostly occurs in menstrual-aged women (7-15). However, two cases did not comply with this generalization. One of them was the case of a pre-pubertal girl reported by Maor-Sagie et al., while the other was a 63-year-old woman discussed by Kushwaha et al. (4, 14). A common trait can be recognized in these two cases when studied closely: both had a remarkable past medical history. The eight-year-old girl was prematurely born and the elderly lady had multiple comorbidities including Castleman's disease treated with bone marrow transplantation. The other patients reported in the literature did not have any significant past medical or surgical history.

## Conclusion

Dyspareunia due to clitoral pilonidal sinus can successfully be cured with early diagnosis, appropriate antibiotics, and wide surgical excision of the lesion. Delay in diagnosis and treatment is associated with the development of complications such as abscess formation; it also prolongs the time to definitive treatment and affects the prognosis of the disease. In cases of mons pubis swelling, vulvar pain, and unexplained dyspareunia in menstrual aged women, a clitoral pilonidal sinus should be kept in mind as a potential cause.

**Conflicts of Interests:** None declared.

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