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Case Report

Pilonidal Cyst of the Penis: Suggestion of a Management Plan Based on Literature Review and a Case Report

Ahmad G. Kanafani, MD[1], Ahmad R. Hammoud, MD[1], Noha A. Bejjani, MD[2], Daniel E. Mahfoud, MD[3], Nazih Youssef, MD[1]

[1]Division of Urology, Department of Surgery, Lebanese American University - Gilbert and Rose-Marie Chagoury School of Medicine, Byblos, Lebanon
[2]Department of Pathology, Lebanese American University - Gilbert and Rose-Marie Chagoury School of Medicine, Byblos, Lebanon
[3]Department of Medical Imaging, Lebanese American University - Gilbert and Rose-Marie Chagoury School of Medicine, Byblos, Lebanon

Corresponding Author: Nazih Y. Youssef, LAU Medical Center- Rizk Hospital, Lebanon, nazih.youssef@lau.edu.lb

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Abstract

Background: Pilonidal sinus rarely affects the penis. This disease is only discussed in case reports, around 26 cases in the literature, and never in textbooks or guidelines. Along with the scarce data in the literature, most of the reported cases were managed differently, and the diagnosis, which could only be confirmed with specimen histopathology, was delayed. Besides the case report of a rare penile disease, the purpose of this paper is to suggest a management plan for suppurative penile lesions to avoid delay in diagnosis and treatment.

Case Report: We encountered a case of penile pilonidal sinus that was initially treated as an abscess based on an ultrasound result. The quick relapse and the suspicious second ultrasound of a hair-like septum in the lesion led to excision and histopathologic confirmation. Penile swelling in patients with a history of Papaverine or any vasoactive drugs injections, trauma, penile prosthesis, diabetes mellitus, intra-abdominal abscess, Tuberculosis, intravenous drug abuse, systemic symptoms, and signs of infection should be managed as an abscess; drainage, appropriate antibiotherapy, and management of risk factors and causes.

Discussion: The usual clinical presentation of pilonidal disease of the penis consists of tender penile swelling, dyspareunia, and purulent discharge, as well as erectile dysfunction which was reported in only one case. In contrast to penile abscess, the presentation usually includes a tender penile swelling, and systemic symptoms such as fever, chills, and night sweats. Physical examination of this patient revealed a non-specific tender, pus-pointing, and pus-containing lesion with mildly erythematous edges, located near the coronal sulcus

Conclusion: Pilonidal cyst of the penis is a rare disease with delayed management in the literature because of misdiagnosis caused by confusion with penile abscess. Though ultrasound could narrow the differential diagnosis, the clinical presentation remains the cornerstone in differentiating between penile abscess and pilonidal sinus.

Keywords: Pilonidal sinus, Penile pilonidal sinus, Penile abscess, Pilonidal cyst of the penis, Pilonidal disease of the penis

Background

Multiple cutaneous pathologies involve the male external genitalia, ranging from neoplastic, infectious, rheumatologic, allergic, immunologic, and dermatologic conditions. Focusing on infectious processes, differential diagnoses remain wide, with a spectrum of organisms causing balanitis, balanoposthitis, cellulitis, intra-cavernosal abscess, folliculitis, and many others. [1] In this article, we report a case of a 21-year-old man presenting with penile swelling that was found to be a pilonidal disease. We performed a literature review of online databases PubMed, SCOPUS and Web of Science using the following search terms: pilonidal disease, pilonidal cyst, pilonidal sinus of the penis, pilonidal sinus of the glans, penile swelling, penile abscess, and penile cysts. The available case series and reports were cited in this paper.

A penile abscess is a serious condition, usually reported to be associated with Papaverine or any vasoactive drug injections, trauma, penile prosthesis, diabetes mellitus, history of an intra-abdominal abscess, Tuberculosis, and intravenous drug abuse. [2-9] However, it has been reported to occur spontaneously. [10,11] It usually results from an infectious process with typical causative organisms, namely; Staphylococcus aureus, Streptococci, Bacteroides, and Enterococci. [12] However, cases involving Mycobacterium tuberculosis, Escherichia coli, Klebsiella, Actinomyces, and other anaerobes have been reported. [12] This condition usually presents with systemic symptoms in particular chills, gait disturbances, and night sweats associated with painful penile swelling. [6,9-13] Moreover, the physical examination of patients with penile abscess, which could be located anywhere along the penis, usually reveals a tender erythematous, sometimes necrotic, and ulcerated swelling of the penis associated with signs of systemic infection such as fever and tachycardia. [6,9,10,12,14] The diagnosis is usually established clinically; however, several imaging modalities have been reported to aid in the confirmation of the diagnosis, define the extent of the abscess, and guide the treatment. [9,10,15] The management includes appropriate antibiotherapy and drainage with concomitant management of the risk factors and etiologies. [9,12]

In 1833, Herbert Mayo first discussed pilonidal disease as an inherited condition referring to the Latin term “nest of hairs”. [16] However, it was found to be an acquired chronic inflammatory pathology most commonly located in the sacrococcygeal region and reported to be found in the breast, umbilicus, scalp, and penis. [17] Pilonidal disease is thought to originate from a blocked hair follicle that enlarges, leading to rupture of the pilosebaceous gland, causing edema and inflammation, and resulting in an abscess and sinus formation. [18] The pilonidal disease of the penis is an unusual presentation; it has been reported in the literature and purely in case reports. The risk factors mentioned in the literature encompass a sedentary lifestyle, the presence of hair located on the shaft of the penis, and a high Body Mass Index (BMI). [19] In addition, friction was reported to be a risk factor contributing to the pathophysiology of this condition. This pathology is almost exclusively encountered in an uncircumcised penis. [17]

The most common clinical presentation of pilonidal disease of the penis includes tender penile swelling, dyspareunia with or without purulent discharge, as well as erectile dysfunction, which was reported in only one case.[20] On physical examination, pus-containing swelling is most commonly seen on the coronal sulcus, associated with erythema, inguinal lymphadenopathy, or abnormal ulcerated appearance mimicking penile cancer. [14,17,21,22] In some cases, diagnosis of pilonidal disease of the penis was described to be done clinically; [17] other cases stated using radiographic imaging to aid in the diagnosis, including ultrasonography. [23] However, most of the literature stated that the final confirmation was established by histopathologic examination. [17] Differential diagnosis can be divided into benign lesions such as penile abscess and inclusion cysts or malignant

tumors such as Kaposi's sarcoma and squamous cell carcinoma. [18]

There are no clear guidelines describing the treatment of pilonidal cyst, but there is a consensus for a lesion excision, with several cases reporting performing a circumcision. The purpose of this paper is to report a rare case of penile pilonidal sinus and suggest a management plan for suppurative penile lesions to avoid delay in the diagnosis and treatment.

Case Presentation

This is a case of a previously healthy 21-year-old man, who presented for a mid-distal right lateral penile shaft painful swelling of a few weeks duration and increasing in size during the last few days. The patient is sexually active without any history of sexually transmitted infection. His BMI is 20.5. He denied any recent trauma or recent shaving. He didn't report any dysuria, chills, fever, urethral discharge, or abnormal urine color. On physical examination, the swelling is tender, with a mild bulging pus-containing lesion. (Figure 1)

Figure 1: Penile lesion – Initial presentation

The ultrasound of the penis showed a well-circumscribed cystic lesion containing heterogeneous fluid with internal bands and a thick wall in favor of an abscess. (Figure 2)

Figure 2: B-mode ultrasound shows a cystic lesion measuring 3.8 x 2 cm with low level echoic content related to debris

The patient was started on Amoxicillin-Clavulanic Acid and scheduled for incision and drainage performed in the clinic, leaving a Penrose drain, which was removed on its own two days after the procedure. One month later, a complete resolution of the abscess was seen during a physical assessment. However, six months later, the patient presented with a recurrent lesion at the same location, with increasing size and erythema associated with pain. A second ultrasound showed an interval decrease in the size of the collection, with a serpiginous hyper-echoic line suggestive of hair inclusion. (Figure 3) The patient was started again on a course of Amoxicillin-Clavulanic acid and scheduled one week later for excision and circumcision. However, the patient came to the emergency department earlier after developing chills, severe penile pain associated with discomfort while urinating, and severe phimosis. He was started on Meropenem 1g every 8 hours, and later that night, we noticed a rupture of the penile lesion, with the persistence of the patient's complaints. A swab culture was taken from the ruptured site, which yielded no organism growth after 48 hours of incubation.

Figure 3: B-mode ultrasound showing serpiginous hyperechoic line suggestive of hair inclusion (after relapse)

Circumcision and excision of the penile lesion located at the coronal sulcus were performed under spinal anesthesia. The foreskin and the penile lesion were removed in one block. A small Blake drain was left in the dissection area for drainage. The next day, the Blake drain output was insignificant and was removed with a surgical wound showing a good postoperative result. A dressing was performed daily for the first two days.

The patient was switched to Clindamycin 300 mg every 8 hours and discharged home to continue oral treatment for 10 days in fear of having resistant organisms to Amoxicillin-Clavulanic Acid. Histopathology results revealed the presence of a tortuous narrow fistula tract or sinus, surrounded by granulation tissue enclosing some hair shafts, and by chronic inflammation. Findings are highly consistent with pilonidal sinus. (Figure 4)

The patient was followed up one month and five months postoperatively with good healing of the circumcision and no signs of recurrence.

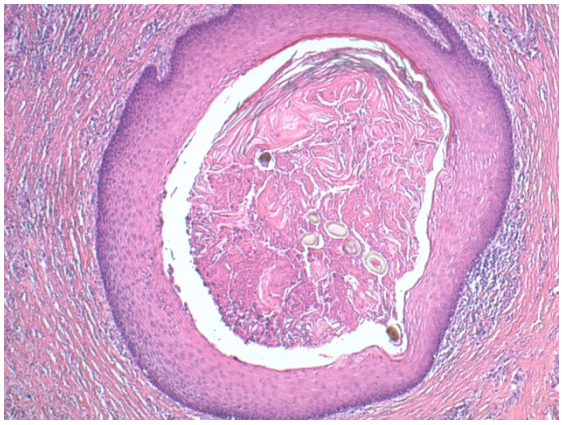

Figure 4: Histopathology – Hair shafts within the pilonidal cyst lined by the squamous epithelium and surrounded by inflamma-tory area

Discussion

The case presented here is that of a 21-year-old man, with a normal BMI, who is maintaining an active lifestyle, with no history of penile hair located anywhere near his lesion, and as such not assimilating with the risk factors stated in the literature for pilonidal disease of the penis, which includes obesity, hirsutism, and a sedentary lifestyle. [19] In addition, the patient denied any Papaverine injection or any vasoactive drugs, trauma, penile prosthesis, diabetes mellitus, history of an intra-abdominal abscess, Tuberculosis, and intravenous drug abuse, which are reported to be associated with penile abscess. [2-9] However, we observe a clinical presentation, similarly described in the literature, in both penile abscess and penile pilonidal disease, which includes a tender penile swelling gradually increasing in size. The usual clinical presentation of pilonidal disease of the penis consists of tender penile swelling, dyspareunia, and purulent discharge, as well as erectile dysfunction which was reported in only one case.[20] In contrast to penile abscess, the presentation usually includes a tender penile swelling, and systemic symptoms such as fever, chills, and night sweats. [6,9,10,11,12] Physical examination of this patient revealed a non-specific tender, pus-pointing, and pus-containing lesion with mildly erythematous edges, located near the coronal sulcus, similar to a reported case by Sagar et Al., which can be either a penile abscess or a pilonidal cyst. [10] No

lymphadenopathy was palpated, and no signs of intra-abdominal abscess were noted. The pilonidal disease presents on physical examination as a swelling that tends to take the appearance of an abscess or cyst, most commonly located on the dorsal aspect of the penis near the coronal sulcus. [17] Associated inguinal lymphadenopathy was reported in 2 cases only. [14,21] Abnormal ulcerated appearance, mimicking penile cancer, was reported in one case. [22]

Compared to the physical examination of a penile abscess that usually reveals tachycardia, a tender erythematous, sometimes necrotic and ulcerated swelling of the penis with no precise location described, signs of an intra-abdominal abscess that might predispose to penile abscess might be present. [6,9,12,14,17] Our initial diagnosis was a penile abscess since it was described similarly in the literature, [10] and the first radiologic investigation favored this diagnosis. The literature reports the use of imaging, aiding in the diagnosis and treatment, most notably an ultrasound of the penis, where a penile abscess can be described, and strands of hair can be seen in the case of pilonidal disease. [23] Yamada et al. stated the use of MRI when an ultrasound was yielding inconclusive results. [15] As per most penile abscess cases in the literature, we initially drained the abscess and gave a course of antibiotics: Amoxicillin-Clavulanic Acid, which is the conventional treatment of a penile abscess. However, we experienced a recurrence of the same presentation a few months later with new radiologic evidence of hair-like strands on ultrasound, similar to one case that reported a recurrence of pilonidal disease after drainage only. [24] Accordingly, the decision was made to excise and circumcise the patient for definitive treatment, which was successful. In the literature, 15 cases reported performing a circumcision as a definitive treatment for this pathology. Unlike no other reported cases, we placed a drain intra-operatively to prevent postoperative collection, which was removed the next day with minimal output overnight. Unlike our case, the growth of bacterial organisms was stated in six cases, with the most common organisms cultured: Actinomyces, A. Israelii, Candida albicans, and Staphylococcus aureus. [14,21,24-27] Actinomyces israelii, although a rare finding with pilonidal sinus was reported in four cases in the literature. [21,24] On histologic examination, sulfur granules are seen, which are characteristic of this infection. [21] Surgical treatment is similar; however postoperative antibiotics, such as Penicillin, are essential to prevent the recurrence of the infection. [21,27]

Histopathology examination remains necessary for confirmation of the diagnosis, as most cases were diagnosed post-operatively. To note that in many instances the hair found in the sinus was of different color from the patient's natural hair color. [21]

Our patient was followed up one month and five months after his surgery, respectively, without any reports of sequelae or complications. The patient reported no urinary symptoms, normal erection, and ejaculation. Post-op complications of penile abscess drainage and debridement vary in the literature from sequelae of chordae and penile curvature to erectile dysfunction and recurrence, sometimes as severe as requiring a penectomy as per Ehara et al. [13,28-30] In comparison to the pilonidal cyst of the penis where no post-op sequelae have been reported to date.

In reference to the reviewed literature, our patient, who is uncircumcised, initially presented with a painful penile swelling at the coronal sulcus with a history of friction. Though the presentation is more in favor of a pilonidal cyst, we managed the patient as if he had an abscess without any pathognomonic symptom or sign, however with ultrasound findings in favor of the latter. Since the clinical presentation did not complement the ultrasound findings, we could have avoided delaying the appropriate management by requesting an MRI. As for the drain placement after the excision of the pilonidal cyst, the output was insignificant, and its need is questionable. Moreover, drain placement after pilonidal cyst excision was

not mentioned in the reviewed literature; therefore, we don't recommend it. Additionally, the choice of Clindamycin post-excision was based on the rapid progression of the infectious process while using Amoxicillin-Clavulanic Acid to treat possible resistant organisms that did not grow on the culture medium. We believe that the choice and duration of antibiotic treatment should be based on culture results and clinical status.

Conclusion

After reflecting on this case and reviewing the literature in regard to the pilonidal cyst of the penis, we recommend careful history-taking to differentiate between the risk factors and clinical presentation of a penile abscess and pilonidal cyst. Performing an ultrasound of the penis, as an attempt to define the pathology presented is helpful but can sometimes be misleading. Performing an MRI might help the diagnosis when encountering doubtful clinical presentations and inconclusive ultrasound findings. In addition, taking a pus swab culture can direct antibiotic therapy. If the swelling is highly indicative of a pilonidal cyst of the penis, we recommend excising the lesion while circumcising the patient, instead of incising and draining, to prevent recurrence. Histopathologic examination is always warranted to confirm the diagnosis of the pilonidal sinus of the penis.

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