

Pilimiction, A Rare Presentation of Ovarian Teratoma: A Case Report

Feysel Hassen Issack (✉ feyselh@gmail.com)

SPHMMC: St Paul's Hospital Millennium Medical College <https://orcid.org/0000-0002-9066-6919>

Seid Mohammed Hassen

SPHMMC: St Paul's Hospital Millennium Medical College <https://orcid.org/0000-0003-2211-7480>

Seid Kedir Hassen

SPHMMC: St Paul's Hospital Millennium Medical College

Kaleab Habtemichael Gebreselassie

SPHMMC: St Paul's Hospital Millennium Medical College <https://orcid.org/0000-0002-2270-6632>

Ferid Ousman Mummied

SPHMMC: St Paul's Hospital Millennium Medical College <https://orcid.org/0000-0002-4634-4883>

Ibsa Kedir Hassen

SPHMMC: St Paul's Hospital Millennium Medical College <https://orcid.org/0000-0003-1848-9003>

Case Report

Keywords: Pilimiction, Adnexal mass, Bladder teratoma, Trichiuria, Case report

Posted Date: January 5th, 2022

DOI: <https://doi.org/10.21203/rs.3.rs-915549/v2>

License:  This work is licensed under a Creative Commons Attribution 4.0 International License.

[Read Full License](#)

Version of Record: A version of this preprint was published at Research and Reports in Urology on March 1st, 2022. See the published version at <https://doi.org/10.2147/RRU.S356738>.

Abstract

Background: Adnexal teratoma involving the urinary bladder is a very rare condition. Presentation is variable ranging from irritative LUTS (lower urinary tract symptoms) to pilimiction or trichiuria (passage of hair in the urine).

Case presentation: We report a case of a 42-year-old woman who presented with pilimiction and lower abdominal pain. Contrast-enhanced computed tomography scan (CECT) and Cystoscopy were used for the diagnosis. Tumor markers were negative. Right side salpingo-oophorectomy and partial bladder wall excision were performed. Histopathology of the specimen showed features consistent with mature teratoma. The Patient-reported improvement of symptoms in the subsequent follow-up visits.

Conclusion: Pilimiction is a pathognomonic sign of bladder teratomas. Therefore, it is wise to think of this pathology in patients who report the passage of hair through the urine (trichiuria or pilimiction), as in our case.

Cystoscopy and cross-sectional imaging aided in the initial diagnosis. However, a definitive diagnosis was provided by histopathology.

We performed right-side salpingo-oophorectomy and partial cystectomy.

Transurethral resection is associated with a high recurrence rate and is not recommended for secondary bladder teratomas.

Introduction

Teratomas are tumors consisting of three germ layers seen commonly during childhood. Mature teratomas are benign and demonstrate well-differentiated tissues such as sebaceous glands, hair, and teeth. Adnexal teratoma involving the urinary bladder is a very rare condition. The clinical presentation may vary from irritative Lower Urinary Tract Symptoms (LUTS) or urinary retention to pilimiction (passage of hair in the urine). We hereby present a case of mature teratoma of the ovary involving the urinary bladder, which primarily manifested with pilimiction. The condition is rare and there are only a few similar reports in the literature.

Case Presentation

42 years old Para I female patient, was referred to a tertiary hospital after she presented with a passage of hair in the urine and lower abdominal pain for a year. Otherwise, there was no complaint of urinary frequency, urgency, or pain on urination. She did not report fever, foul-smelling vaginal discharge, or pain during coitus. She has not had any known chronic medical illness.

On physical examination, the patient had stable vital signs. There was no remarkable positive physical finding other than an old Pfannenstiel surgical scar. The pelvic exam was negative for any visible genital

lesions, cervical motion tenderness, or palpable adnexal mass.

She underwent a joint evaluation and workup by urology and gynecologic oncology units. On laboratory investigation, CBC (complete blood count) was normal and hematocrit was 40.3%. Urine analysis was remarkable for microscopic hematuria in the range of 3-5 RBC/HPF (Normal \leq 2 RBCs/HPF). But the other parameters of urine analysis were in the normal range. The serum creatinine, blood urea nitrogen (BUN), serum electrolytes, and liver enzymes were also normal. Serologic tests for HIV, hepatitis B, and C as well as treponema were negative.

Abdominopelvic ultrasound examination showed a bizarre shaped tubular echogenic shadow in the lumen of the bladder that looks like a foreign body. Further imaging with a Contrast-enhanced CT scan of the abdomen and pelvis showed a complex heterogeneous mass lesion in the right adnexa measuring 5cm*3.7 cm that has multiple calcifications and fat attenuations inside it. The lesion invaded the right superolateral surface of the bladder with a defect in the bladder mucosa. It was concluded as a right adnexal mass lesion with possible bladder invasion (Fig. 1).

Cystoscopy was also done and revealed a whitish papillary mass with hair like component over the right posterior wall of the bladder near the dome (Fig. 2).

Further workup with tumor markers using serum beta HCG, serum LDH, CA-125, and CEA didn't reveal any clue toward a specific diagnosis.

A joint decision was made to surgically explore the patient. The pelvis was entered through the previous Pfannenstiel incision. The finding was a 4cm*6cm right ovarian mass with solid and cystic areas containing hair, bone, and teeth. Right side salpingo-oophorectomy and partial cystectomy bladder repair were done. The excised mass was sent for histopathological analysis.

Excisional biopsy report showed stratified squamous epithelium, adnexal structures, fatty and bony tissue fragments confirming the diagnosis of mature ovarian teratoma.

On subsequent follow-up visits, the patient reported improvement of symptoms and physical examination was unremarkable.

Discussion

Mature teratomas are the commonest benign tumors of the ovary. They account for 20-50 % of all ovarian tumors and are more prevalent in premenopausal females. In clinical practice, they are characterized by a unilateral involvement, which is often on the right side, although up to 10% of cases can be bilateral.^{3,4} Pathologically, ovarian teratomas are germ cell tumors. The word was first used by Virchow in 1863 and was derived from the Greek 'teras', meaning 'monster'.^{1,2,5}

Mature ovarian teratomas are indolent and asymptomatic tumors. Their diagnosis is often incidental either during a routine pelvic examination or abdominopelvic imaging performed for other indications.

However, some patients may present with symptoms that are often secondary to tumor-related complications. These include acute abdomen, abdominal lump, LUTS, or sepsis. Very rare clinical features such as the passage of hair in the urine (pilimiction), gross hematuria, the passage of hair through the anal orifice, small bowel obstruction, and fistula into the rectum are also reported in the literature.^{5,6,7}

Torsion is the commonest complication of mature ovarian teratoma occurring in 16% of the cases. Other uncommon complications include tumor rupture (1–4%), malignant transformation (1–2%), infection (1%), invasion into adjacent viscera (<1%), and very rarely, autoimmune hemolytic anemia and paraneoplastic syndrome.⁵⁻⁸ Invasion and rupture of the tumor might involve adjacent pelvic and abdominal structures most commonly the urinary bladder. There are also case reports of involvement of the rectum, vagina, small intestine, sigmoid colon, anterior abdominal wall, and peritoneal cavity.⁹

The anatomic proximity of the urinary bladder to the ovaries makes it vulnerable to direct involvement by tumors of ovarian origin. The clinical presentation of this rare occurrence depends on the extent of bladder involvement and the biological nature of the tumor. According to our literature review, superficial involvement of the bladder wall often presents with irritative LUTS such as frequency and urgency. On the other hand, deeper invasion into the bladder lumen by the teratoma manifests itself with urinary tract infection (UTI), hematuria, and LUTS. Many of these features are non-specific and can be easily overlooked. Pilimiction, however, is a rare but pathognomonic feature of full-thickness bladder wall invasion by ovarian teratoma. At times, the hair in the bladder lumen might create an obstructive ball at the bladder outlet and manifest as acute urinary retention.^{1,4,5,9}

Pilimiction was first reported in 1700 by Wallace. Its presence is a specific and diagnostic indicator of ovarian teratoma and fistula formation. Localized ovarian teratomas do not pose a diagnostic difficulty on their own. However, involvement of the urinary bladder is often diagnosed late unless patients present with pilimiction like the case in our patient.

In most similar cases reported so far, the definitive diagnosis was made through the use of cystoscopy, computed tomography (CT) scan, or laparotomy.^{1,5,10} There are also few reports on laparoscopic diagnosis and management of mature teratoma with bladder involvement.¹⁰

Previous reports attributed the pathogenesis of bladder involvement to malignant transformation of the teratoma at some point in time leading to aggressive invasion of adjacent pelvic organs.¹⁷ However, with detailed research of the cases and pathology specimens, it was shown that benign teratomas can also cause fistula formation with nearby structures. Intermittent leakage of tumor contents can lead to the chronic inflammatory process and adhesion formation resulting in fistulation. This is particularly common during tumor necrosis, torsion, and infection. Chronic pressure of the tumor on adjacent organs is also suspected as a possible mechanism for fistula formation.^{9,10}

The urologist has a vital role in the management of such conditions. Joint involvement of a gynecologist and urologist is recommended. Surgical resection of the lesion and ipsilateral fallopian tube together with partial bladder resection is recommended. A malignant transformation should be ruled out with a histopathological examination of the surgical specimen.

Conclusion

Though very rare as a presenting symptom, pilimiction is a pathognomonic sign of primary or secondary bladder teratomas. Therefore, it is imperative to consider teratoma involving the urinary bladder in any patient who reports the passage of hairlike particles through the urine as in the case of our patient.

In addition to the clinical history, cystoscopy and cross-sectional imaging aid in the diagnosis of bladder teratoma. Definitive diagnosis is provided by histopathology of the surgical specimen.

The recommended management of secondary bladder teratoma entails complete surgical excision of the primary tumor and part of the involved bladder wall.

Transurethral resection is precluded by the fact that it is associated with a high recurrence rate and not suitable for secondary bladder teratoma like in our patient. For this reason, we opted for open surgical excision of the primary tumor in the right ovary and part of the involved bladder wall.

Abbreviations

BUN: Blood urea nitrogen; CA-125: Carbohydrate antigen-125; CBC: Complete blood count; CEA: Carcinoembryonic antigen; CECT: Contrast-enhanced computed tomography; CT: Computed tomography; HCG: Human chorionic gonadotrophin; HPF: High power field; LDH: Lactate dehydrogenase; LUTS: Lower urinary tract symptoms; RBC: Red blood cells; UTI: Urinary tract infection

Declarations

Ethics approval and consent to participate

No institutional review board approval was required.

Consent for publication

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Data availability

All the generated data are included in this article

Funding

The authors received no funding for this work.

Disclosure

The authors have no competing interests in this work.

Acknowledgments

Not applicable

References

1. Godara R, Karwasra RK, Garg P, Sharma NP. A diagnostic symptom of ovarian dermoid cyst. *Internet J Gynecol Obstet.* 2006;6(1):62-9.
2. Nagamani T, Vani I, Nimmana SP, Kumar YM. Ovarian cystovesical fistula causing pilimiction: an unusual complication of ovarian cyst. *Ann Women Child Health.* 2015 Dec 18;1(1):C1-4.
3. Hamza M, Yasmeen T, Nadeem IA, Fatima N, Fatima S, Huzaifa M. Ovarian dermoid cyst presenting with unusual complaint of hair coming out of the anal orifice-A case report. *JPMA.* 2020 Jan 14;2019.
4. Kizaki Y, Nagai T, Ohara K, Gomi Y, Akahori T, Ono Y, Matsunaga S, Takai Y, Saito M, Baba K, Seki H. Ovarian mature cystic teratoma with fistula formation into the rectum: a case report. *SpringerPlus.* 2016 Dec;5(1):1-6.
5. Tandon A, Gulleria K, Gupta S, Goel S, Bhargava SK, Vaid NB. Mature ovarian dermoid cyst invading the urinary bladder. *Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology.* 2010 Jun;35(6):751-3.
6. Sardesai S, Raghoji V, Dabade R, Shaikh H. Benign Cystic Teratoma of Ovary Perforating into the Urinary Bladder: A Rare Case. *The Journal of Obstetrics and Gynecology of India.* 2012;1(62):54-5.
7. Bhasin SK, Malik SM, Sharma G, Gupta SK. Ovarian dermoid presenting as acute intestinal obstruction: a rare case report and review of literature. *International Surgery Journal.* 2016 Dec 13;2(2):283-5.
8. Guo H, Yin K, Wang Y, Tong X, Yang H, Xia M, Shuang W. Mature cystic ovarian teratoma invading the bladder: A rare case report. *Translational Surgery.* 2018 Jul 1;3(3):62.
9. Naqvi KZ, Abdullah A, Jabeen M, Iqbal F, Edhi M. Ovarian dermoid causing pilimiction. *J Coll Physicians Surg Pak.* 2015 Jan 1;25(1):71-2.
10. Vaishnav A, Sarkar D, Pal DK. Bladder teratoma with pilimiction in a male adolescent. *Urology Annals.* 2020 Jul;12(3):286.

Figures

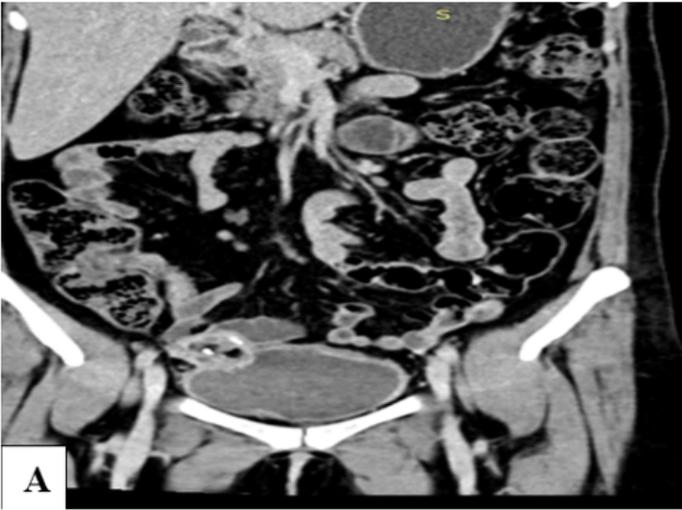


Figure 1

CECT of the abdomen and pelvis. A: Coronal reformatted B: Axial image

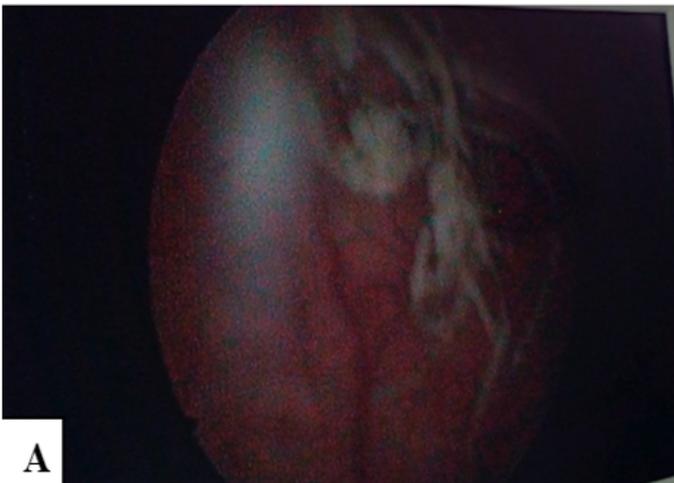


Figure 2

Cystoscopy. A: Far view B: Near view

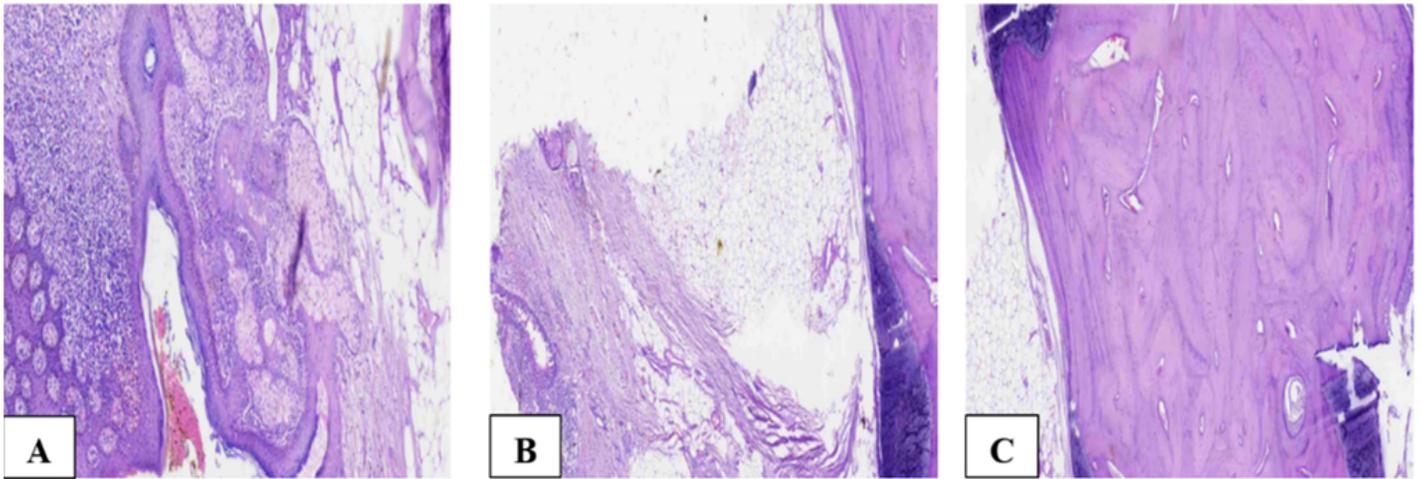


Figure 3

Scanned pictures of the histopathology specimen. A, B, &C

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [CAREchecklistEnglishFilled.pdf](#)