

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

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Case report

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

Posted Date: September 20th, 2021

DOI: <https://doi.org/10.21203/rs.3.rs-915549/v1>

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Abstract

Introduction: 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 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 passage of hair through the urine (trichiuria or pilimiction), as in our case.

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

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 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 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. 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 Contrast enhanced CT scan of 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 right adnexal mass lesion with possible bladder invasion (Fig. 1: CECT of the abdomen and pelvis. A: Coronal reformatted image B: Axial image).

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

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 was 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 (Fig. 3: A, B, &C - scanned pictures of the histopathology specimen).

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% 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 routine pelvic examination or abdomino-pelvic 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 passage of hair in the urine (pilimiction), gross hematuria, 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 rectum, vagina, small intestine, sigmoid colon, anterior abdominal wall, and peritoneal cavity [9].

The anatomic proximity of 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 biologic 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 manifest 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 of 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 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. A 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 histopathological examination of the surgical specimen [1].

Conclusion

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

We performed 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

Carcino-embryonic 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

Acknowledgements

Not applicable

Availability of data and materials

All the generated data are included in this article.

Authors' contributions

SMH and FHI diagnosed the case and conceived the idea. SMH and SKH operated the patient. SKH and IKH were involved in post-operative follow up of the patient. FHI, FOM, KHG, and IKH compiled patient clinical data. FOM and KHG organized the literature review. FHI, KHG, and FOM prepared draft of the manuscript. SMH critically revised and edited the manuscript. FHI was engaged in the correspondence and submission of the article. All the authors read and approved the final manuscript before submission.

Funding

The authors received no funding for writing of this article.

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 publication of the clinical data and will be made available to the editor upon request.

Competing interests

The authors declare that they have no competing interests.

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Tables

Table 1
Important dates in this case

Date	Events and activities	Reports and findings
November 04, 2020	Referred to a tertiary Hospital	
November 04, 2020	Initial surgical OPD visit and evaluation	History: Presented with passage of hair in the urine and lower abdominal pain of 01year duration. Physical examination: stable vital signs. An old Pfannenstiel surgical scar was seen.
November 04, 2020	Initial laboratory investigations report	Complete blood count was normal with Hematocrit of 40.3%, White cell count of 8.6×10^3 . Urinalysis showed urine PH of 6, 3–5 RBC/HPF (normal, ≤ 2 /HPF) and negative for nitrite and leukocyte esterase.
October 02, 2020	Abdomino-pelvic ultrasound report (done before referral)	A bizarre shaped tubular echogenic shadow in the lumen of the bladder that looks like a foreign body.
October 15, 2020	Contrast enhanced CT scan of abdomen and pelvis report (done before referral)	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 supero-lateral surface of the bladder with a defect in the bladder mucosa. It was concluded as right adnexal mass lesion, likely Teratoma, with possible bladder invasion
December 18, 2020	Urology clinic visit and evaluation	Clinical assessment: same Patient was sent for cystoscopy and then linked to gynecology clinic for joint workup.
December 22, 2020	Cystoscopy report	A whitish papillary mass with hair like component over right posterior wall of the bladder near the dome. Some trabeculations were also seen over the right postero-lateral wall.
December 22, 2020	Gynecology OPD visit and evaluation	Clinical assessment: same Worked up with tumor markers (serum beta HCG, serum LDH, CA-125, and CEA) and all were in the normal range. Imaging with chest x-ray. The result was unremarkable. The patient was then linked to gynecology oncology clinic
December 24, 2020	Gynecology oncology clinic visit and evaluation	Clinical assessment: same Admission to ward for preoperative workup and joint surgery with urology was decided.
March 30, 2021	Preoperative work-up	Complete blood count, liver and renal function panels and serum electrolytes were all in the normal range. ECG showed normal findings

Date	Events and activities	Reports and findings
March 31, 2021	Operated	The pelvis entered through the previous Pfannenstiel incision. There was a 4cm*6cm right ovarian mass with solid and cystic areas containing hair, bone and teeth. It has dense adherence to dome of the bladder. Upon gentle dissection off the bladder wall, it was seen that it has a direct communication with the bladder lumen. Right side salpingo-oophorectomy and partial cystectomy was done. The excised mass was sent for histo-pathological analysis
March 31, 2021 to April 06, 2021	Post-operative course	On post op period patient had stable vital signs. The Foley catheter was kept for 5 days. Subsequently, the urine started clearing. She was discharged after 07 days of hospital stay.
April 15, 2021	Excisional biopsy report	The excised biopsy specimen showed a stratified squamous epithelium, adnexal structures, and fatty and bony tissue fragments with a conclusion as Mature Teratoma.
April 21, 2021	Follow up visit to urology and gynecology oncology clinic	The patient reported improvement of symptoms and physical examinations were unremarkable

Figures

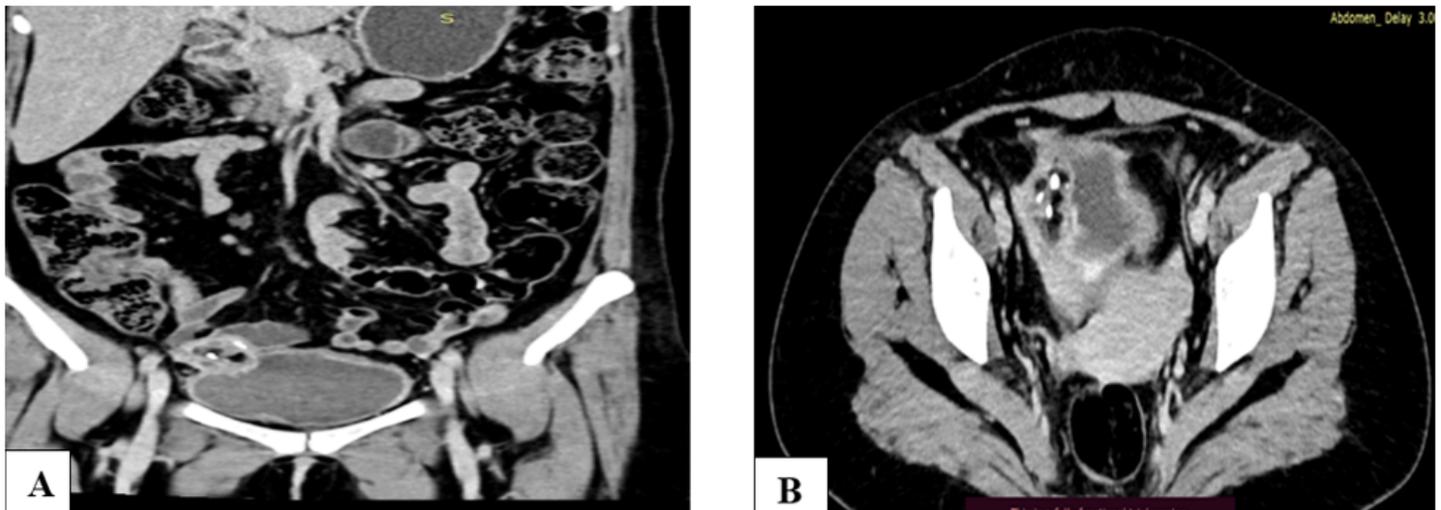


Figure 1

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

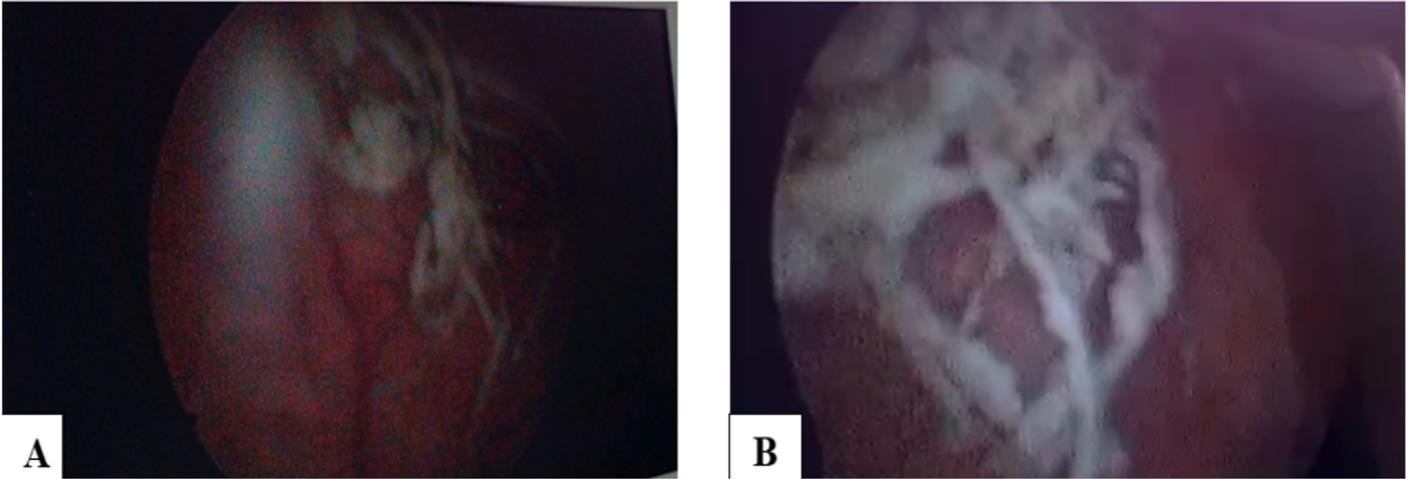


Figure 2

Cystoscopy. A: Far view B: Near view

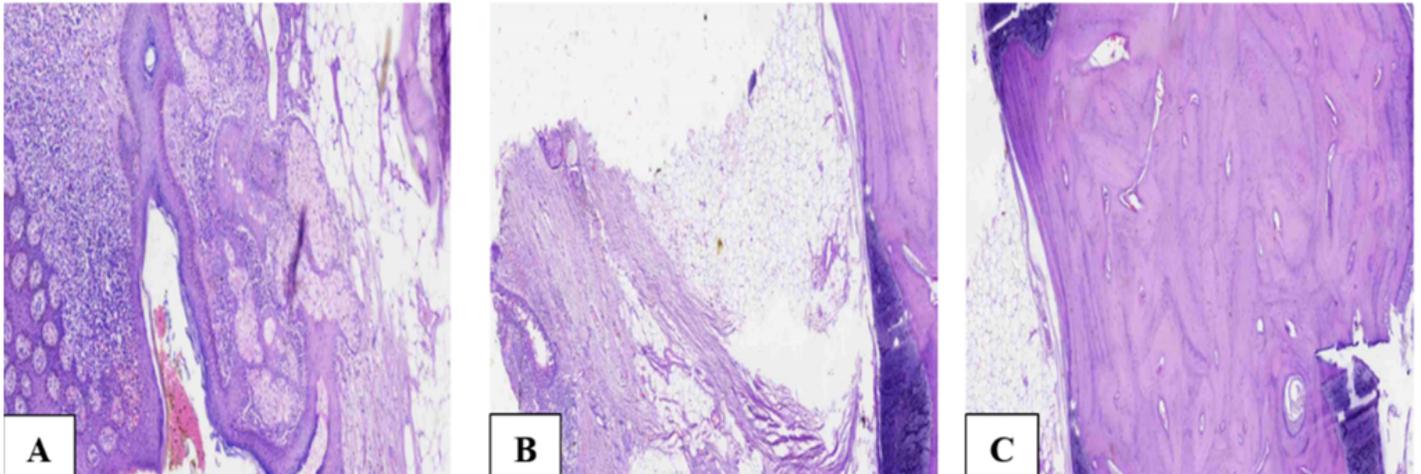


Figure 3

A, B, &C - scanned pictures of the histopathology specimen

Supplementary Files

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