"Hairy potty"
Ovarian dermoid cyst with fistula to bladder*

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ABSTRACT

Dermoid cysts are usually asymptomatic until complications occur. Spontaneous rupture of a dermoid into an adjacent organ is a rare complication and no such case has been reported in the Philippines.

A 24-year-old primipara consulted for pilimiction. Three years earlier, she had recurrent urinary tract infection and was diagnosed to have a dermoid cyst. Left untreated, the cyst grew in size and urinary symptoms worsened. Ultrasound, CT scan and subsequent laparotomy revealed that the dermoid cyst has penetrated the bladder wall resulting to fistula formation between the dermoid and the urinary bladder. Hair and sebum were seen inside the bladder. A left salpingo-oophorectomy and partial cystectomy of the urinary bladder were done.

The first locally documented case of an ovarian dermoid cyst with fistula to the bladder is presented. A review of literature is made, the predisposing factors, possible cause, diagnosis and management are discussed.

Keywords: dermoid cyst, primipara, pilimiction, fistula, salpingo-oophorectomy, cystectomy

INTRODUCTION

Mature cystic teratomas or dermoid cysts are the most common benign germ cell tumor of the ovary. They may occur at any time during the woman's reproductive years but are commonly seen in young females. Ovarian dermoids are usually asymptomatic and often discovered incidentally. Symptoms are observed in the presence of complications like torsion, infection, hemorrhage, or rupture.[1] Torsion is the most frequent complication while fistula formation between the benign cystic teratoma and surrounding organs such as the urinary bladder, colon, or rectum is one of the least common. When such a communication occurs, the bladder is the most common site.[2] There have been a few reported cases of fistula formation between a dermoid cyst and an adjacent organ in foreign journals. However, in a review of the locally published literature, this is the first documented case of this rare complication.

CASE REPORTS

J.B., a 24-year-old G1P1 (1001), from Legazpi City, was admitted for the first time to our institution last October 6, 2015 with a chief complaint of inability to void.

Her past medical, family and personal history are non-contributory.

Menarche was at age 14. Subsequent menses were at regular monthly intervals, lasting 4-7 days, with 3 moderately soaked napkins per day. No history of dysmenorrhea. Her last normal menstruation was on the 3rd week of September 2015.

She is a Gravida 1 Para 1 (1001). She had an uncomplicated full term spontaneous vaginal delivery last June 6, 2012. Prenatal check-ups were at a health center, no pelvic ultrasound was done.

The history of present illness started 3 years prior to admission when the patient palpated a soft, tender, fist-sized hypogastric mass associated with on and off fever and urinary frequency. These symptoms were noted about six months after her vaginal delivery. She consulted a private doctor and was treated for urinary tract infection. A transvaginal ultrasound showed a dermoid cyst. She was referred to our institution, was advised laparotomy however the patient was lost to follow up.

One year prior to admission, she again experienced urinary frequency and occasional dysuria. She self-medicated with Cefuroxime which relieved her symptoms. No consult was done.

Eight months prior to admission, the mass was noted to have increased in size to approximately 8 cms. Urinary frequency persisted and dysuria became more frequent. She also noticed oil droplets or sebum in her urine, later accompanied by occasional passage of hair or pilimiction. Still, no consult was done.

4 months prior to admission, there was persistence and worsening of her urinary symptoms. She had to pull tufts of hair from her urethra just to be able to void. She

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sought consult in our institution, was prescribed Cefuroxime for her UTI and scheduled for pre-operative evaluation. However, the patient was again lost to follow up.

One month prior to admission, she returned to our institution and was presented at the OB-GYN pre-operative conference. Strands of hair were seen coming out of her urethra (Figure 1). An 8 x 8 cm cystic to doughy mass was palpated anterior to the normal-sized corpus. A repeat transvaginal ultrasound confirmed the presence of a left ovarian dermoid cyst but with a much bigger size of 11.3 x 10.5 x 9.9 cms. The uterus and the right ovary were normal. Hyperechoic linear structures were also seen within the bladder (Figure 2). A fistula to the urinary bladder was entertained so an outpatient referral to the Urology Service was made.

One day prior to admission, she experienced severe hypogastric pain and was unable to void. She was brought to the emergency room and was subsequently admitted by the Department of Surgery with a diagnosis of bladder mass, to consider dermoid cyst.

On admission under the service of Surgery, the patient was conscious, coherent, and not in cardiorespiratory distress with the following vital signs: BP: 120/70 mmHg, CR: 81/minute, RR: 22/minute, Temp: 36.5°C. An indwelling foley catheter was inserted. CBC revealed mild anemia with Hgb of 95 and Hct of 0.29. Urinary tract infection was confirmed with >50 pus cells seen on urinalysis. BUN and creatinine were normal. Whole abdominal CT scan with contrast was requested and cystoscopy with biopsy was planned. Co-Amoxiclav 600 mg IV q12 hours and Paracetamol 300 mg IV q6 hours were started.

On the 3rd hospital day, whole abdominal CT scan with contrast was done which revealed dermoid cyst with contained rupture and suspicious communication with the urinary bladder at the vesical dome (Figure 3). Cystoscopy was deferred and she was referred to the Department of OB-GYN.

On the 5th hospital day, patient was transferred to the OB-GYN ward. She was conscious, coherent, ambulatory, and with normal vital signs. Pertinent physical examination findings centered on the abdomen and the pelvis, there was an 8 x 8 cm doughy mass at the hypogastric area. The mass was slightly tender and with limited mobility. Pelvic examination revealed normal external genitalia and parous vagina. The cervix was firm, closed and smooth, the corpus not enlarged. A slightly tender, cystic to doughy mass measuring 8 x 8 cm was palpated superior and anterior to the uterus. The admitting diagnosis was Gravida 1 Para 1 (1001), Ovarian New Growth, left, most likely a Dermoid Cyst with probable fistula to the bladder. The patient was prepared for an elective pelvic laparotomy to be done together with the surgery residents.

On the 8th hospital day, the patient underwent surgery

Figure 1. Strands of hair coming out of patient's urethra.

Figure 2. Transvaginal ultrasound of the patient showing dermoid cyst and hyperechoic linear structures within the urinary bladder.

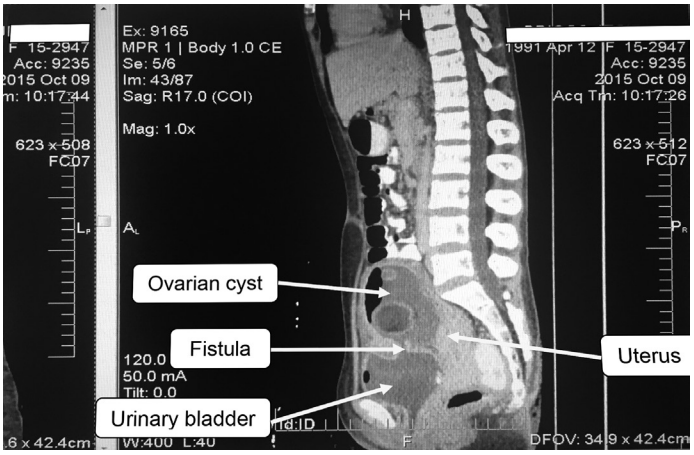

Figure 3. Patient's CT scan with contrast showing communication between the dermoid cyst and the urinary bladder.

under spinal anesthesia. On laparotomy, the left ovary was converted into a cystic to doughy mass measuring 8 x 7 x 4 cm with a smooth, opaque white capsule. The mass was partially twisted on its pedicle and was densely adherent to the urinary bladder (Figure 4). Attempts at adhesiolysis revealed a fistulous connection between

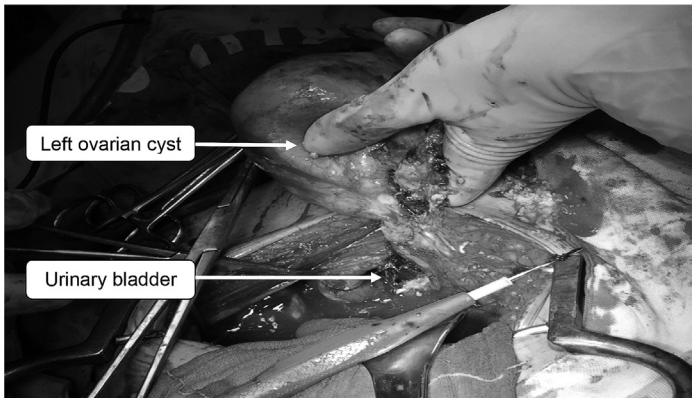

Figure 4. Partially twisted dermoid cyst densely adherent to the urinary bladder. Hair and sebum are seen inside the bladder.

the cyst and the urinary bladder (Figure 5). Tangled mass of hair and sebaceous fluid were seen inside the urinary bladder (Figure 4). The corpus, the right ovary and both fallopian tubes were grossly normal. There was no ascites nor spillage of the contents of the dermoid cyst into the peritoneal cavity. A left salpingo-oophorectomy and partial cystectomy of the urinary bladder were done. Cut section of the mass showed a unilocular cyst containing pockets of sebum and hair. There were no solid areas, no necrosis nor hemorrhages. Her post-operative course was uneventful.

On the 12th hospital day, her 4th post-op day, patient

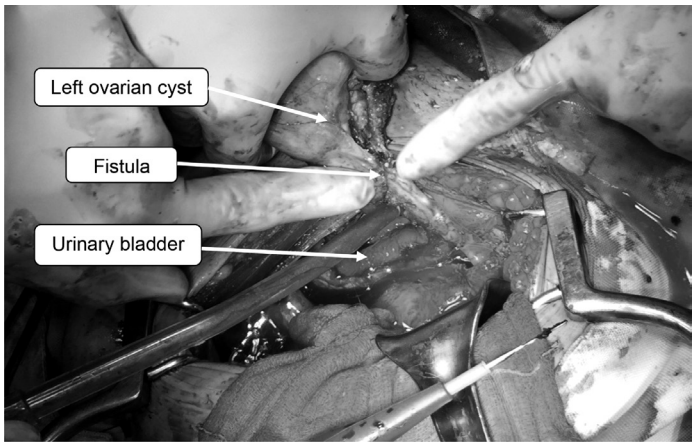

Figure 5. Fistulous connection between dermoid cyst and urinary bladder.

was discharged with the indwelling foley catheter retained for 6 more days. Home medications were Cefuroxime 500 mg BID to complete 7 days, FeSO4 1 tab OD for 1 month and Celecoxib 200 mg BID for pain.

The patient came for follow up on her 10th post-op day. Her urinary symptoms were resolved and the surgical wound was dry and well coaptated. The foley catheter was removed and she was able to void spontaneously.

Final histopathologic result of the specimens submitted revealed a mature cystic teratoma, ovary, left, adherent to the bladder, both with severe acute and chronic inflammation (Figure 6). No diagnostic abnormality recognized, fallopian tube, left.

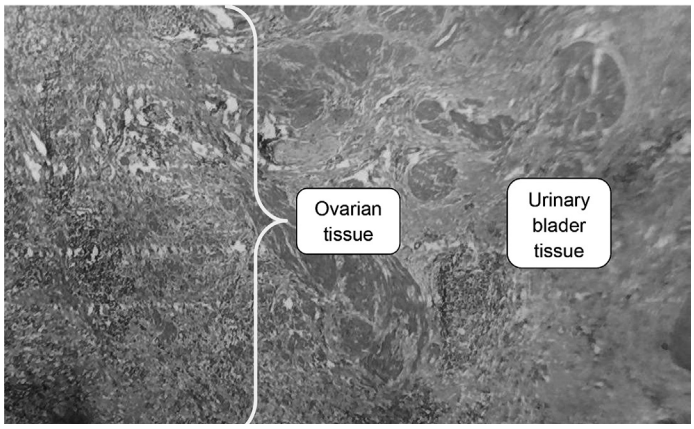

Figure 6. Histopath picture of the specimen submitted showing ovarian and urinary bladder tissues.

FINAL DIAGNOSIS:
Gravida 1 Para 1 (1001)
Dermoid Cyst, Left Ovary, with Fistula to the Urinary Bladder

Operative Procedure done:
Exploratory Laparotomy
Left Salpingo-oophorectomy with Partial Cystectomy, Urinary Bladder under SAB

DISCUSSION

Mature cystic teratomas are benign cystic structures that contain mature cell elements from all three germ cell layers. The term dermoid cyst is often used interchangeably with the term mature cystic teratoma. Dermoid is a descriptive term that emphasizes the preponderance of ectodermal tissue. Dermoid cysts contain mostly dermal and epidermal elements like hair, skin, sebaceous glands and sometimes even nails and teeth.[1]

Benign teratomas are slow-growing tumors that may occur from infancy to the postmenopausal years with more than 50% seen in women between the ages of 25 and 50 years. Dermoids are the most common ovarian neoplasm in prepubertal females and are also common in teenagers.[1]

The patient was 24-years-old on admission but was diagnosed to have a dermoid cyst three years earlier, at age 21. Since dermoid cysts are slow-growing tumors, she may have had the cyst for years, possibly even during her prepubertal or teenage years.

About 50 - 60% of dermoids are asymptomatic and are usually discovered incidentally during a routine pelvic examination, during pelvic imaging, or at laparotomy. Symptoms of pain and pelvic pressure are often reported in the presence of complications. Torsion is the most frequent complication (16%) and is commonly seen in young women. Rupture (1-4%) is more common in pregnancy. Infection, hemorrhage, malignant degeneration, and invasion into an adjacent viscera are all unusual complications of dermoids, occurring in less than 1% of patients.[1,2] Fistula formation between a benign cystic ovarian teratoma and an adjacent organ is a rare occurrence with the urinary bladder as the most commonly involved.[2]

There have been a few documented cases of dermoid cyst with fistula to adjacent organs in foreign journals but none locally.

T. Nagamaniet.al. reported a case of a 32-year-old parous woman with straining at micturition, narrow stream, passing hair or pilimiction and turbid urine occasionally. On cystoscopy, hair ball was visualized. CECT demonstrated an ovarian dermoid cyst invading the urinary bladder. TAHBSO and partial cystectomy were done. The diagnosis of fistulous tract between ovarian dermoid with bladder was confirmed.[3] Kaniz Zehra Naqvi et.al. also reported a case of a 25-year-old nulliparous female who presented with lower abdominal pain and pilimiction. Examinations revealed a right sided dermoid which had penetrated the wall of bladder and expelled its contents in the bladder.[4] Rantomalala et al. discussed a case of a right ovarian dermoid cyst which fistulated to the bladder who presented with recurrent urinary infection.[5] Tandon et al. presented a case of a 30-year-old woman with pyuria and dysuria, where ultrasound examination clearly demonstrated an ovarian dermoid cyst invading the urinary bladder. Pathologic evaluation confirmed the diagnosis.[2] Mohammad Mehdi Hosseini et. al. reported a case of a 63-year-old woman with prolonged dysuria and hematuria who had bilateral ovarian dermoid cysts with urinary bladder involvement.[6]

Fistula to other organs such as the colon and rectum were also reported. Landmann et. al. has reported a benign ovarian teratoma that fistulated to the rectum which presented with rectal bleeding and a polypoid mass in the rectum.[7] Khanna et. al. reported the case of an 18-year-old woman who had a left teratoma that ruptured into the sigmoid colon and a 39-year-old woman with lower abdominal pain and hematochezia which, upon evaluation, had a mass in the anterior rectal wall from a benign cystic teratoma in her left ovary.[8]

Patients with spontaneous rupture of a dermoid cyst into the bladder may present with complaints such as pilimiction, pyuria, hematuria or passage of other material from a dermoid cyst and at times repeated episodes of urinary retention due to blockage of urethral meatus by hair.[2]

The patient presented with passage of hair in urine which is a pathognomonic sign, recurrent UTI and difficulty in voiding due to blockage of the urethral meatus with clumps of hair.

Various etiologies have been postulated for fistula formation in dermoid cyst. Shiels et. al. reported a case of entero-ovarian dermoid cyst fistula and suspected that a small leak from the cyst caused dense adhesions with the bowel resulting in fistula formation.[9] Peterson et al. found that torsion, trauma, infection, chronic pressure in labor and malignant transformation can cause leakage and fistulation.[10] Chronic leakage thus appears to be a common denominator in non-malignant fistulation with adjacent viscera.[2]

In this case, torsion of the dermoid cyst and the patient's pregnancy and labor may have predisposed her to the fistula formation. Very significant and relevant in the patient's history is the fact that all her urinary symptoms started six months following her vaginal delivery. The patient may have had the dermoid cyst for a long time but it remained asymptomatic until after her pregnancy and delivery. Chronic pressure on the dermoid cyst during her pregnancy and labor could have led to focal ischemic necrosis of the cyst wall which resulted to slow leakage, causing dense adhesions with the bladder, and eventually fistula formation.

A diagnosis of a dermoid cyst is considered when on pelvic examination, a semisolid mass is palpated anterior to the broad ligament. Ultrasound has a more than 95% positive predictive value and a less than 5% false positive rate in detecting dermoid cysts. Most dermoids have a characteristic ultrasound picture: a dense echogenic area within a larger cystic area, a cyst filled with bands of mixed echoes, and an echoic dense cyst.[1]

Fistula formation between a dermoid cyst and the urinary bladder is suspected because of the patient's symptoms and the findings on abdominal and pelvic examination. Diagnosis may be confirmed by ultrasound and CT scan as what happened in this case.

The patient had recurrent urinary tract infection, pilimiction and ended up unable to void. On pelvic examination, a cystic to doughy mass was palpated anterior to the uterus. Transvaginal ultrasound showed the "typical picture" of a dermoid cyst. Also seen were hyperechoic linear structures within the urinary bladder which suggested a fistula formation between the dermoid cyst and the urinary bladder. The whole abdominal CT scan confirmed this pre-operative diagnosis.

As to the management of benign cystic teratomas, cystectomy is the procedure of choice in premenopausal women where normal ovarian tissue is still appreciated. Unilateral oophorectomy or salpingo-oophorectomy is

indicated in patients in whom ovarian tissue cannot be preserved, and when there is involvement of the fallopian tube particularly in large-sized masses.[11]

In this case, a left salpingo-oophorectomy was done because intraoperatively, no remaining normal ovarian tissue was seen. As in previously reported cases, partial cystectomy of the bladder had to be done.

SUMMARY AND CONCLUSION

In summary, an unusual case of a dermoid cyst with fistula to the urinary bladder in a 24-year-old primipara was presented. Rupture or perforation of the contents of the dermoid cyst into an adjacent organ like the urinary bladder may be extremely rare but should be promptly attended to as it could lead to grave consequences. A complete history, a thorough physical examination, a high index of suspicion and the use of appropriate diagnostic modalities like ultrasound and CT scan help in making an early and correct diagnosis of such a case. A rational approach in the management by a multidisciplinary team that includes obstetrician-gynecologists and surgeons would best benefit the patient. Most important too would be the cooperation and compliance of the patient and her family. ■

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