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INTRODUCTION

Endometriosis is a common, benign gynecologic disease affecting 7-10% of the general population. It is defined as the presence of functional endometrial tissue outside of the uterine cavity and is generally associated with pelvic pain, dyspareunia and infertility. The most frequently involved sites are the ovaries (30%) followed by uterosacral and cardinal ligaments (18-24%), the fallopian tubes (20%) and lastly the pelvic peritoneum.

Endometriosis at scar site can be found after cesarean, hysterectomy, amniocentesis, laparoscopic trocar tract, or perineal episiotomy. A classic triad of cyclic pain, perineal mass and previous episiotomy or tear is sufficient to diagnose scar endometriosis. Three typical characteristics of perineal scar endometriosis such as, a past perineal tear or episiotomy during vaginal delivery, a tender nodule or mass at the perineal lesion and progressive and cyclic perineal pain. If these criteria are met, the predictive value of perineal endometriosis is 100%.

Endometriosis in an episiotomy scar is very rare, occurring only in 0.0007% of births. Episiotomy site endometriosis affects between 0.03 and 1.7% of reproductive age women. In an article by Leite et al. of 29,135 deliveries, there were 33 cases of scar endometriosis, 31 were related to cesarean scar and only two to episiotomy with an incidence of 0.01%. In other studies, the incidence of abdominal wall endometriosis after cesarean section was 0.03-1.7% while the incidence of episiotomy region endometriosis after normal birth was 0.06-0.7%. The latent period from delivery to development of symptoms was 2 months to 2 years. Surgical scar endometriosis is seen following vaginal delivery with episiotomy and in abdominal surgery scar following hysterectomy and cesarean section. Perineal trauma, especially perineal tearing or episiotomy scars appear to be more commonly affected if the episiotomy is associated with a vaginal delivery and subsequent uterine curettage. It may also arise from previous dilatation and curettage. Autologous transplantation of vital endometrial cells to an open episiotomy wound during vaginal delivery, especially when manual uterine exploration and postpartum curettage are performed, seems to be the pathogenic mechanism of perineal endometriosis.

RESULTS

Eleven years prior to admission (PTA), a 50 year old, G4P3 (3013), palpated a 1 cm, slightly tender, right vulvar mass. She initially consulted a local clinic and was assessed with episiotomy site infection and was given unrecalled antibiotics for 7 days. Interval history showed gradual increase size of the vulvar mass and severity of vulvar pain noted during menses. She was seen on multiple occasions by different gynecologist and was assessed with vulvar new growth and underwent three vulvar biopsies which showed endometriosis and was given oral contraceptive pills, depo- medroxyprogesterone acetate and also self-medicated with herbal oil.

One month PTA, a whole abdominal ultrasound was requested as part of her employment's annual physical examination, which showed nephrolithiasis at the right upper pole, a solid mass in the inferior pole of the right kidney, bilateral pelvo-calicectasis, subserous myoma measuring 2.1 x 1.9 cm and a right ovarian cyst measuring 2.1 x 1.8 cm. She was then advised to seek consult with an OB-GYN. Upon consult, a 9 cm vulvar mass was noted, a slightly enlarged uterus and bilateral adnexal fullness. Assessment was vulvar mass, probably ectopic endometriosis, rule out malignancy and consider ovarian new growth, bilateral. A transvaginal ultrasound was requested which showed an enlarged uterus measuring 7.12 x 7.0 x 6.48 cm with possible adenomyosis, thickened endometrium, enlarged ovaries with bilateral endometrial cysts. On transperineal ultrasound, it showed right perineal new growth, probably malignant. She was referred to a private Gynecologic oncologist but due to financial constraints, opted to transfer to the outpatient department where she was assessed with ovarian new growth, with a concomitant vulvar mass, probably malignant and endocervical polyp. She was advised peritoneal fluid cytology, Total abdominal hysterectomy with bilateral salpingo-oophorectomy with possible complete surgical staging and vulvar biopsy.

However, while securing clearance for the operation, she experienced burning epigastric pain graded 8/10 and consulted in the Emergency room and was initially seen by the service of Internal medicine. On physical examination, abdomen was soft with direct tenderness on the epigastric area. Initial assessment was Gastroesophageal reflux disease and was given Omeprazole 40mg/IV and was placed on NPO (nothing per orem). Complete blood count showed severe anemia (hemoglobin 85g/L, hematocrit 0.28) and leukocytosis with a WBC count of 26 with neutrophilic predominance of 0.96. Assessment was to consider an intraabdominal infection and was started on Piperacillin-Tazobactam 2g/IV every 6 hours. Urinalysis showed hematuria of 40-50/hpf.

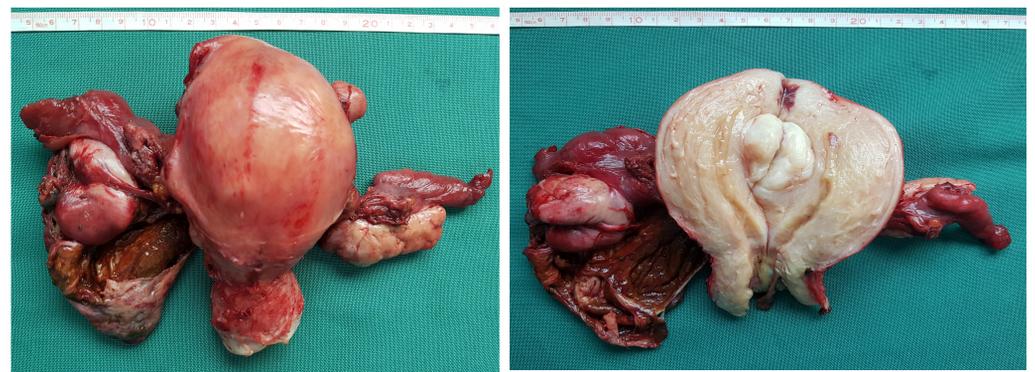
She was referred to gynecology and noted a 9 x 5 cm skin-colored multiloculated, firm, fixed, non-tender mass on the right labia majora topped with blood clots. On speculum exam, cervix was pink, smooth with a 1x1 cm tongue-like polypoid mass protruding from the cervical os. On internal exam, cervix was firm, short with a polypoid mass protruding from the os, uterus was fixed, enlarged to 2 1/2 months size, retroverted, non-tender, right adnexa no mass or tenderness, with left adnexal fullness and no tenderness. On rectovaginal examination, the lower pole of the mass was about 5 cm in diameter. Assessment was right vulvar mass, probably malignant, Ovarian new growth probably endometrial cysts, rule out malignancy and was advised to undergo incision biopsy of the vulvar mass with frozen section; bilateral salpingo-oophorectomy with frozen section followed by total abdominal hysterectomy with possible complete surgical staging.

She was referred to urology and a whole abdominal CT scan was requested which showed a 6.8 x 7.5 cm uterine mass, a hypodense structure component of mass and a renal cyst on the right inferior pole measuring 3.8 x 3.5 x 4.5cm with enhancing component. Urology assessed her with a right renal mass, probably malignant and was advised to undergo partial nephrectomy and was subsequently admitted.

Urology first did Cystoscopy, retrograde pyelography and ureteral stent insertion. On examination of the external genitalia, the vulvar mass, measured 9 x 5 x 3 cm. It was firm and rubbery with vesicular overgrowths and extends about 4 cm deep into the right lateral vaginal wall. We proceeded with incision biopsy of about 1/4 of the vulvar mass (Figure 3), including tumor free margins which was sent for frozen section which showed endometriosis. We then proceeded with hysterectomy and on opening, the omentum had brownish implants over it, there was no free peritoneal fluid noted. Peritoneal washing was done and was collected for possible cytology. On inspection, the uterus was nodularly enlarged to 2 1/2-3 months size. It had a mass at its right superolateral wall which was firm and measured 2 x 1 cm. The lower 2/3 of the uterus had dense adhesions to the rectosigmoid area. The right ovary was cystically enlarged and extends down, deep in to the pelvic cavity while the left ovary was slightly enlarged with endometriotic implants noted over it. Both ovaries were adherent to the lateral wall of the uterus. We then proceeded with adhesiolysis. During adhesiolysis, there was inadvertent rupture of the ovarian cysts, which exuded chocolate brown fluid. We then proceeded with Total abdominal hysterectomy with bilateral salpingo-oophorectomy (Figure 4). The collapsed right ovary measured 10 x 6 cm, its inner lining had smooth areas, but also had thickened to rubbery areas, hence we sent it for frozen section and it was signed out to be an endometrial cyst. On further inspection, there were endometriotic implants deep into the pelvic cavity and in the cul de sac. We proceeded with electrocautery of the implants. The previously collected fluid was discarded.

On cut section of the uterus, the endometrial lining was smooth and thin. There was an intramural myoma at the mid posterior wall measuring 4 x 3.5 x 2.5 cm, which on cut section showed a pearly white whorled cut surface. The myometrium was thickened measuring 3.5 cm with pinpoint hemorrhages typical of adenomyosis. The endocervical lining had a reddish soft polypoid structure that protrudes out into the endocervical os, which was an endocervical polyp. Urology then proceeded with partial nephrectomy, renorrhaphy, hemostasis followed by insertion of JP drain.

Postoperatively, she was given antibiotics, diet progression and was eventually discharged stable. On follow up after two weeks at the Ambulatory care services, her vulvar wound was dry and well coapted as well as her abdominal wound incision. Histopathology of the right vulvar mass showed endometriosis with hormone therapy changes. The uterus had multiple myomas with calcifications, adenomyosis and atrophic endometrium. The cervix had endometriosis, endocervical polyp, nabothian cysts and chronic cervicitis. Both ovaries had endometrial cysts with cystic follicles, while bilateral fallopian tubes had edema and congestion. Subsequent follow up showed a gradual decrease in the size and swelling of the vulvar mass and pelvic exam after 4 weeks were unremarkable. There was a noticeable decrease in the size of the vulvar mass 6 months post-operatively.



Uterus and bilateral ovaries

Cut-section

CONCLUSION

Episiotomy site endometriosis is a rare entity which may be seen in patients presenting with cyclic pain, perineal mass and a history of previous episiotomy. Manual uterine exploration or postpartum curettage may increase the chances of developing episiotomy site endometriosis and thus should be avoided. Wide resection of the lesion including a 1 cm disease free margin is the treatment of choice.

The definitive treatment of extrapelvic endometriosis is by surgery and the diagnosis can be made by histopathological examination of the material. The aim of this surgery is prevention of recurrence of lesions. Surgical removal of the lesion is necessary for the diagnosis and treatment and the complete excision of the lesion together with approximately 1 cm of healthy tissue in order to prevent local recurrence. In some patient, repair of defect with synthetic mesh is necessary and is done by a plastic surgeon. The advantage of surgery includes less risk of recurrence and obtaining tissue pathology to exclude malignancy.



Follow-up after 6 months post-operatively

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Right vulvar mass

Incision biopsy