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Case Report
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Xanthogranulomatous Salpingo-Oophoritis – A Rare Case Report

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Abstract

Background: Xanthogranulomatous inflammation is an unusual type of chronic inflammation which causes tissue destruction by inflammatory cells mainly foamy histiocytes. Such inflammation of the female genital tract is rare and limited to the endometrium. Only few cases of xanthogranulomatous inflammation of genital tract have been reported so far in literature.

Case Report: A 32-year-old nulliparous female presented with chief complaints of heavy menstrual bleeding and severe dysmenorrhea for eight years with failed medical management. Per-abdominal examination revealed a suprapubic firm, tender mass of around 14 weeks gravid uterus size. On per vaginal examination, uterus was retroverted with a tender, solid-cystic mass in the right adnexa extending till pouch of Douglas with restricted mobility. Her CA125 (510 U/ml) were raised. Pelvic ultrasound demonstrated adenomyotic uterus and endometriotic cysts in right adnexa with moderate right hydroureteronephrosis which was confirmed on CT urography. She underwent cystoscopic DJ stenting followed by total abdominal hysterectomy with right salpingo-oophorectomy and left salpingectomy. DJ stents were removed on day 7. The postoperative period was uneventful. Histopathological examination of the sent specimen revealed xanthogranulomatous salpingo-oophoritis of right fallopian tube and ovary with endometriotic features and adenomyotic uterus with secretory endometrium.

Conclusion: Xanthogranulomatous inflammation of the tubes and ovaries clinically, radiologically and grossly may mimic ovarian malignancy. Thus, a preoperative diagnosis can prevent radical surgical treatment.

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Introduction

Xanthogranulomatous inflammation is a chronic inflammatory process characterized by massive cellular infiltration of lipid-laden histiocytes along with lymphocytes, plasma cells, and polymorphonuclear leucocytes [1]. This type of inflammation is most commonly seen in kidneys and gall bladder [2]. Other organs affected by this inflammation include the stomach, anorectal area, bones, urinary bladder, testis, and epididymis. Xanthogranulomatous inflammation of the female genital tract is rare and is usually limited to the endometrium. Only a few cases involving both fallopian tube and ovary have been reported [3]. Due to its locally destructive nature and mass-forming capacity due to adhesions, xanthogranulomatous inflammation may sometimes both clinically and radiologically mimic malignancy. Hence, awareness of this entity is important to ensure the proper management of the patients. The aim behind presenting this case is to make the readers aware about this rare entity as one of the differentials.

Case Presentation

A 32-year-old nulliparous female presented to the outpatient department with complaints of heavy menstrual bleeding associated with severe dysmenorrhea for eight months. She was put on hormonal treatment but her symptoms were not relieved. She was a case of primary infertility as well for which she underwent diagnostic lapro-hysteroscopy seven years ago. The findings were suggestive of bilateral tubal blockage and was advised in-vitro fertilization. She also underwent open right-sided salpingostomy with endometriotic cyst fulguration with myomectomy six years ago owing to severe dysmenorrhea then. On examination, she was clinically pale. On per abdominal examination, a suprapubic, firm, tender mass of 14 weeks of gravid uterus was there. On per-speculum examination, cervix and vagina was normal looking. On per vaginal examination, uterus was retroverted with a tender, solid-cystic mass in the right adnexa extending till pouch of Douglas with restricted

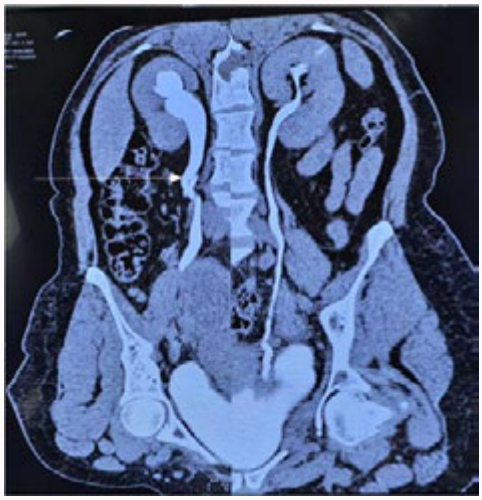

Figure 1: CT Urography showing right adnexal mass compressing right ureter and thereby leading to moderate right sided hydroureteronephrosis.

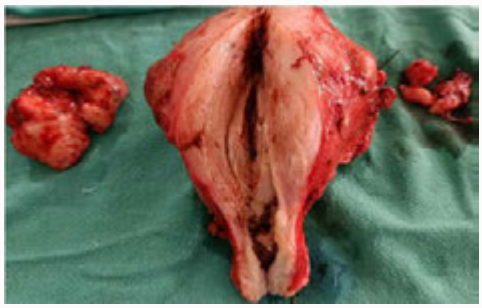

Figure 2: Gross specimen of Total abdominal hysterectomy with right salpingo-oophorectomy and left salpingectomy. Right tubo-ovarian mass of 5 cm × 5 cm observed. The uterus, cervix and left fallopian tube were unremarkable.

mobility. On per-rectal examination, same findings were confirmed with no nodularity felt along uterosacral ligaments. Significant blood investigations showed moderate anemia (hemoglobin = 9.6 gm%, total leucocyte count was 8000/microliter) and raised CA-125 (510 U/mL) with maintained liver and kidney function tests. A pelvic ultrasound showed an adenomyotic uterus with two endometriotic cysts in the right adnexa (4.4 cm × 5.6 cm × 4.6 cm, 3.3 cm × 4.2 cm × 3.4 cm). Magnetic resonance imaging of the whole abdomen showed an adenomyotic uterus with multiple endometriotic cysts in the right adnexa (largest measuring 5 cm), right-sided hydrosalpinx, and right moderate hydronephrosis and hydroureter up to the level of right tubo-ovarian lesion. CT Urography was done and it revealed a large (5×6), thick-walled, complex right adnexal cyst compressing the right ureter thereby causing moderate right hydroureteronephrosis. In view of her symptoms not responding to medical management and imaging findings suggestive of tubo-ovarian lesions causing back pressure changes in the kidney, she was planned for total abdominal hysterectomy with bilateral salpingo-oophorectomy with cystoscopic bilateral DJ stenting. Intra-operatively, multiple endometriotic spots were there on the uterus which was bulky around 14 weeks. Pouch of Douglas was completely obliterated with distorted right sided anatomy. Right ovary was enlarged around 5 cm × 5 cm and was densely adherent to the uterus. Total abdominal hysterectomy with right salpingo-oophorectomy and left salpingectomy was done. Left

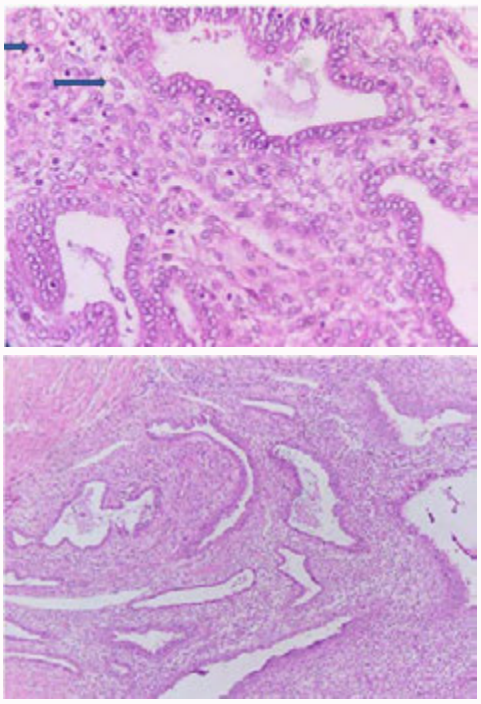

Figure 3: Normal ovarian parenchyma replaced by sheets of Plasma cells and Foamy histiocytes- suggestive of Xanthogranulomatous salpingo-oophoritis.

ovary was normal looking and so was preserved owing to the young age of the patient. DJ stents was removed on postoperative day seven. Her postoperative period was uneventful.

The histopathological examination revealed the specimen of uterus with cervix of 12 cm × 11 cm × 9.5 cm. Sections from the endomyometrium showed secretory endometrium with adenomyosis while sections from the cervix show ecto-endocervicitis with nabothian cyst. Sections examined from right fallopian tube shows features of xanthogranulomatous salpingitis with endometriosis and paratubal cyst. Sections examined from right ovary show xanthogranulomatous oophoritis with endometriotic cyst and areas of endometriosis. Surface inclusion cyst and cystic follicle also identified. Sections of left fallopian tube show unremarkable histology.

Discussion

Xanthogranulomatous inflammation of the female genital tract is a very rare diagnosis. Kunakemakorn et al. described the first case of xanthogranulomatous inflammation of the uterine serosa, left fallopian tube, and ovary in their report of inflammatory pseudotumor of the pelvis way back in 1976 [4]. The age range of these cases varied from 2 to 84 years with the youngest documented case of a 2-year-old who got operated for mass abdomen described by H Tanwar et al. [3]. The exact etiology of this inflammation is unknown but the possible risk factors include endometriosis, intrauterine contraceptive device, long-standing pelvic inflammatory disease and abnormal lipid metabolism. Persistent infection with microorganisms like Bacteroides fragilis, *Escherichia coli*, *Proteus vulgaris*, and *Salmonella typhi* can also be causative in the pathogenesis of xanthogranulomatous oophoritis [5]. Another presumptive etiology is chronic infection leading to tissue necrosis. As a result, there is a continuous release of cholesterol and other lipids from the dead cells which is ineffectively phagocytosed by macrophages leading to a xanthomatous process. In our case,

there can be a possibility of un treated pelvic inflammatory disease which led to damaged bilateral tubes causing infertility. She also had adenomyosis and endometriosis with history of endometriotic cystectomy done in the past. Both these together were the elicitable risk factors in our case.

Clinical features associated with xanthogranulomatous salpingo-oophoritis include pain in the abdomen, abdominal mass, fever, menorrhagia, anorexia, and anemia. Xanthogranulomatous changes in adnexal structures grossly, clinically, and radiologically may mimic ovarian neoplasm [6-8]. Hence, gynecologists need to be aware of this entity. Grossly the affected ovary is usually enlarged, replaced by a solid yellow tumor-like nodular mass that may even be cystic due to liquefactive necrosis. It tends to adhere to the surrounding organs and pelvic peritoneum leading to their destruction and further arousing suspicion of malignancy. Microscopically, the cellular architecture is replaced by sheets of lipid-laden macrophages or foamy histiocytes admixed with lymphocytes, plasma cells, and neutrophils. Histopathological examination along with immunohistochemistry enables us to clinch the diagnosis. Immunohistochemistry can help establish the diagnosis with CD8 (foam cell positive), CD3 (T lymphocyte marker), CD20 (B-lymphocyte marker), and κ and λ (both positive in polyclonal B-lymphocytes) [9].

Both neoplastic and non-neoplastic conditions can be considered in differential diagnoses. The non-neoplastic conditions include tuberculosis (differentiated by AFB staining), fungal infections (differentiated by PAS staining), and malakoplakia (differentiated by Michaelis Guttman bodies in the cytoplasm). The neoplastic conditions include lymphoma, leukemia, malignant small-cell tumors and sclerosing stromal tumors [5].

The treatment of choice for xanthogranulomatous salpingo-oophoritis is salpingo-oophorectomy. Since this lesion is found to mimic ovarian neoplasm, there is a tendency towards extensive surgery including total hysterectomy. Keval et al. have reported that in 26 out of 46 reported cases, total hysterectomy with bilateral salpingo-oophorectomy was performed [10]. A preoperative biopsy or frozen section can be planned to avoid such radical surgeries, especially in those patients with relevant history and clinical symptoms where there is a low clinical suspicion of ovarian malignancy.

Conclusion

Xanthogranulomatous salpingo-oophoritis is a rare entity and a preoperative diagnostic challenge for clinicians. A final diagnosis can be made only after a histopathological examination. A high index of suspicion for those females with risk factors for the pathology and low clinical suspicion of malignancy can prevent radical surgery. Authors suggest, on table frozen section in such cases.

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