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Case Report

A rare case of xanthogranulomatous oophoritis

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ABSTRACT

Xantho-granulomatous oophoritis is an uncommon, non-neoplastic, chronic process affecting female genital tract. Its clinical and radiological features mimic ovarian neoplasm. Here we present a case report of 42-year-old female with chronic pelvic pain with adnexal mass with infertility and raised CA 125 levels. Histopathological examination revealed it to be a case of xantho-granuous oophoritis. Xantho-granulomatous oophoritis is very rare disease and only few cases have been reported so far and this case is being reported in view of its rarity.

Keywords: Xantho-granulomatous oophiritis, Ovarian malignancy, Infertility, Endometriosis

INTRODUCTION

Xanthogranulomatous inflammation is an uncommon form of chronic inflammatory process that is destructive to normal histology of involved organs.[1] The organs which are most commonly affected are the kidney and gallbladder, followed by anorectal area, bone, stomach, and testis.[2] In female genital tract, it more commonly involves the endometrium but vagina, cervix, fallopian tube, and ovary can also be affected.[3] The ovarian involvement is rare and is characterized by a massive infiltration of tissues by lipid-laden histiocytes admixed with lymphocytes, plasma cells, and polymorphonuclear leukocytes, multinucleated giant cells.[3] Clinically and radiologically, Xantho-granulomatous inflammation can mimic tumor of the ovary and fallopian tube. Only few cases of xantho-granulomatous inflammation involving ovary have been reported till date.

CASE REPORT

A patient 42 years old female, resident of Bihar with A1 (twin pregnancy) reported to OPD with complaints of on and off pain in abdomen with dysmenorrhea and chronic pelvic pain for 10 years. Patient was married for 14 years and had history of primary infertility. Patient started taking treatment for infertility and tried to conceive for 10 years. Medical management for infertility was initially followed by 6 IUI cycles and 3 IVF cycles to conceive. Patient conceived with twin pregnancy. There was h/o spontaneous abortion of one fetus followed by intrauterine death of second fetus following which pregnancy was terminated by hysterotomy.

Initial serial ultrasound of abdomen and pelvis revealed uterine adenomyosis with b/l adnexal complex cystic lesions with septations and echoes in pouch of douglas involving b/l adnexa s/o endometriosis.

Latest USG report was suggestive of focal adenomyosis in fundal myometrium wall and enlarged bilateral ovaries with hemorrhagic cyst with endometrioma.

MRI revealed uni-cornuate uterus with non-communicating rudimentary right horn with hematometra with B/L ovarian endometriotic cyst with hemato-salpinx with ±endometriosis.

Cytology of peritoneal fluid was done to rule out malignancy which was negative for malignant cells.

Her blood investigations revealed microcytic hypochromic anemia, slightly raised total leukocyte count [14800], CA-125 level was 217 U/mL (normal range 0-35 U/mL).

After clinical and radiological investigations patient and relatives counselled for further medical management for endometriosis and infertility associated with it. Despite of counselling for medical management, patient and relatives were not willing for further medical management due to severe pain and inability to conceive. Decision for total hysterectomy with conservation of ovaries was taken.

Patient was taken up for total laproscopic hysterectomy with bilateral salpingectomy with adhesiolysis in view of inability to conceive with chronic dysmenorrhea. In our case, the patient was operated in suspicion of malignancy as patient presented with adnexal mass with raised CA 125 levels coupled with patients demand for surgical management.

The specimen was sent for histopathological examination. On microscopic examination characteristic dense fibrosis with infiltration of ovarian stroma with sheets and nodules of foamy macrophages, many histiocytes, lymphocytes and neutrophils was seen. The findings were suggestive of xantho-granulomatous inflammation of ovary. Based on these findings and clinical history a diagnosis of xantho-granulomatous oophiritis was made.

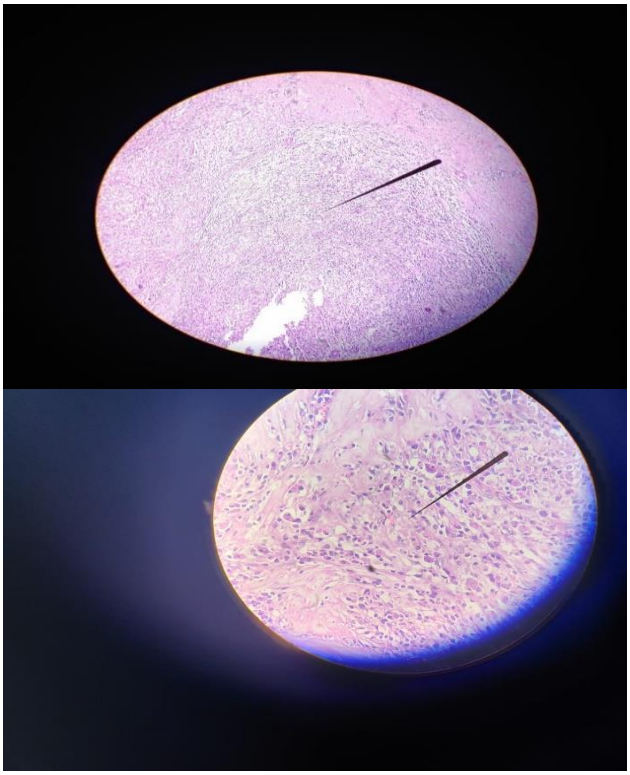

Figure 1: Microscopic appearance of xantho-granulomatous oophirits.

DISCUSSION

Xantho-granulomatous inflammation is an uncommon, non-neoplastic, chronic process in which there is destruction of affected organ by massive cellular infiltration of foamy histiocytes admixed with multinucleated giant cells, plasma cells, fibroblasts, neutrophils, and foci of necrosis.[4,5] Xantho-granulomatous oophiritis is rare and unusual form of chronic oophiritis. It is also called as ovarian fibroxanthoma. Xantho-granulomatous inflammation of female genital tract usually involves endometrium. But it can also affect fallopian tubes and ovaries. Kunakemakorn in 1976 was the first to report a case of xantho-granulomatous inflammation of serosa of the uterus, left fallopian tube and ovary. It presents as mass in pelvic cavity which can be misdiagnosed as ovarian malignancy.[6] The inflammation can involve surrounding organs and peritoneum leading to adhesion which further supports the diagnosis of malignancy. The clinical, radiological and macroscopic features of xantho-granulomatous oophiritis is often confused with ovarian malignancy. The exact etiopathogenesis of disease is unknown. However, it can be associated with infection, inappropriate antibiotic use, long standing pelvic inflammatory disease, endometriosis, intrauterine contraceptive device, uterine leiomyoma. Microorganisms such as *Escherichia coli*, *Proteus* spp., *Staphylococcus aureus*, *Bacteriodes fragilis*, *Salmonella typhi*, *Actinomyces*, *Streptococcus (Enterococcus) faecalis*, viridans *Streptococci*, *Torulopsis (Candida) glabrata*, and group B *Streptococci* are identified from the affected tissue.[7]

Clinical features include long standing history of pelvic inflammatory disease with symptoms such as anorexia, fever, suprapubic pain, menorrhagia, or vaginal bleeding, adnexal tenderness and a pelvic mass.[8,9] It most commonly affects the females of reproductive age group. Differential diagnosis can be neoplastic and non-neoplastic. Most important differential diagnosis is Malakoplakia which is non neoplastic and can be differentiated from xantho-granulomatous oophoritis by presence of the cytoplasmic concentric calcific bodies (Michaelis Guttmann bodies) in malakoplakia. Neoplastic conditions such as lymphoma or leukaemia, malignant small cell tumour and sclerosing stromal tumour, clear renal cell carcinoma may come under the differential diagnosis. Treatment of choice for xantho-granulomatous oophoritis in oophorectomy. In this case we performed total abdominal hysterectomy with bilateral salpingo oophorectomy.

CONCLUSION

Xantho-granulomatous oophoritis is a rare entity and has clinical presentation and radiological features similar to that of common benign and malignant adnexal disease and hence requires a high index of suspicion for diagnosis. Radical surgery can be avoided in long standing cases of pelvic inflammatory diseases, endometriosis, patient with intrauterine copper devices with close follow up of such patients and early diagnosis of xantho-granulomatous oophoritis. In this case, prolonged severe unbearable pain along with infertility leading to inability to conceive took emotional toll on patient and led to the decision of total abdominal hysterectomy.

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REFERENCES

1. Jung SE, Lee JM, Lee KY, Han KT, Hahn ST. Xanthogranulomatous Oophoritis: MR Imaging findings with Pathologic Correlation. AJR. 2002;178:749-51.
2. Bhatnagar K, Narang V, Garg B, Sood N. Xanthogranulomatous Oophoritis: A Rare Case Report. Iran J Pathol. 2018;13(3):372-6.
3. Bindu SM, Mahajan MS. Xanthogranuloma- tous oophoritis: A case report with review of literature. Int J Health Allied Sci. 2014;3:187-9.
4. Manandhar T, Rajbhandari S, Thakur A, Bhandari S, Dhakal S. Xanthogranulomatous Salpingo-Oophoritis Presenting as an Ovarian Malignancy. Cureus. 2021;13(2):e13363.
5. Reid J, Qiao JH. Xanthogranulomatous oophoritis. PathologyOutlines.com website. Available at: https://www.pathologyoutlines.com/topic/ovarynontumorxanthogranoophoritis.html. Accessed on 10 Nov, 2022.
6. Gami N, Mundhra R, Guleria K, Arora VK, Garg S. Recurrent pyometra and xantho-granulomatous salpingitis: A rare pathologic association in a postmenopausal lady. J Mid-life Heal. 2014;5:156.
7. Gray Y, Libbey NP. Xanthogranulomatous salpingitis and oophoritis: a case report and review of the literature. Arch Pathol Lab Med. 2001;125:260-3.
8. Khan B, Aziz AB, Ahmed R. Case of Xantho-granulomatous Oophritis. J Ayub Med Coll Abbottabad. 2017;29:162-4.
9. Karigoudar MH, Kushtagi AV, Karigoudar RM, Sirasagi A. Xanthogranulomatous oophoritis-A Rare inflammatory lesion. J Krishna Inst Med Sci Univ. 2013;2:111-5.

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