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XANTHOGRANULOMATOUS OOPHORITIS: A MASTER DISGUISE!

Histopathology

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ABSTRACT

Introduction- Xanthogranulomatous inflammation is an uncommon cause of chronic destructive process of the affected organ. It is a well-documented histopathological entity in the gall bladder and kidney. However, Xanthogranulomatous inflammation of the female genital tract is unusual and essentially limited to the endometrium. Only a few cases involving the ovary have been reported. **Case Report-** We report a case of 48 years old female who presented with mass per abdomen for 1 month in left lower quadrant, insidious in onset, gradually progressive. Ultrasound of abdomen and pelvis showed a left adnexal large cystic space occupying lesion with low level internal echo with asymmetrically thickened wall –possibly of ovarian origin. Frozen section of the tubo-ovarian mass sent for histopathological studies showed features suggestive of Xanthogranulomatous Oophoritis. **Conclusion -** Clinico- Radiological findings of Xanthogranulomatous oophoritis may mimic malignant ovarian neoplasm and may lead to misdiagnosis. However, the pathologists need to be aware of this entity and should be vigilant before giving a diagnosis on frozen section to prevent a misdiagnosis of malignancy and avoid radical surgery especially in young females.

KEYWORDS

Xanthogranulomatous Oophoritis, Tubo-ovarian mass, Female genital Tract.

INTRODUCTION

Xanthogranulomatous inflammation is an uncommon chronic, non-neoplastic inflammatory process that is characterized by accumulation of lipid laden foamy macrophages intermixed with lymphocytes, giant cells, and plasma cells.[1] It most commonly affects organs like kidney and gall bladder, followed by anorectal area, bone, stomach, and testis.[2] It is unusual in the female genital tract and is essentially limited to the endometrium.[3] The involvement of ovary is uncommon. Only a few cases of this entity are mentioned in the literature and this case is reported in view of its rarity.

CASE REPORT

Here we report a case of 48 years old female, with an obstetric score of P2L2 who presented to Obstetrics and Gynecology OPD with complains of mass per abdomen for 1 month in the left lower quadrant. The mass was insidious in onset and gradually progressive. There were no associated complaints of pain, menstrual irregularities, dysmenorrhoea, fever, burning micturition or weight loss. The patient underwent tubectomy 23 years ago. There was no history of intrauterine contraceptive device usage. She was a known case of hypertension, diabetes mellitus and arthritis and was on medications for the same.

On examination, the vitals were normal. Per abdomen examination revealed a midline mass of 18 weeks gravid uterus size, more towards left side, mobile, firm to cystic in consistency, the lower border could not be made out. On per rectal and per vaginal examination a cystic mass was felt occupying the anterior and left fornices.

Her investigations showed a haemoglobin level of 9 grams/dl with a white blood count of 14.4 x10^3 cells/cumm and absolute neutrophil count of 10.8 x 10^3 cells/cumm. CA125 levels were found to be slightly elevated at 41.9 units/ml. No abnormality detected on urine routine and microscopy and HBA1C was within normal limits. Ultrasound abdomen and pelvis revealed a left adnexal large cystic space occupying lesion with low level internal echo and asymmetrically thickened wall –possibly of ovarian origin.

The patient was prepared for laparotomy with left tubo-ovarian cyst excision with total abdominal hysterectomy and bilateral salpingo-oopherectomy. Intraoperatively, a left tubo-ovarian mass measuring approximately 12 x10 cm found in left adnexa and was sent for frozen sections. Approximately 500 ml of pus was drained from the cyst which was sent for culture and sensitivity. The left tube and ovary not found separately from the tubo ovarian mass and the right ovary was unhealthy with an endometriotic spot. Dense adhesions were noted between uterus and anterior abdominal wall.

On frozen section, the specimen of left tubo-ovarian mass measured 10x9.5x1.5 cm. The wall appeared to be thickened and diffuse yellow speckled areas were noted on the cyst wall (Figure 1).

Figure-1

Microscopic examination revealed replacement and destruction of the normal ovarian stroma by inflammatory exudates comprising of sheets of foamy macrophages, lymphocytes, plasma cells and few neutrophils along with pigment laden macrophages (Figure 2). Areas of necrosis noted. No evidence of malignancy was noted in the sections. Features suggestive of Xanthogranulomatous oophoritis of the left ovary. Subsequently, on routine grossing, H & E sections studied from right ovary, showed evidence of endometrial glands within the normal ovarian stroma consistent with features of endometriosis in the right ovary. Acid fast stain was negative for microorganism. On culture of the pus drained from the left tubo-ovarian mass Escherichia coli was isolated.

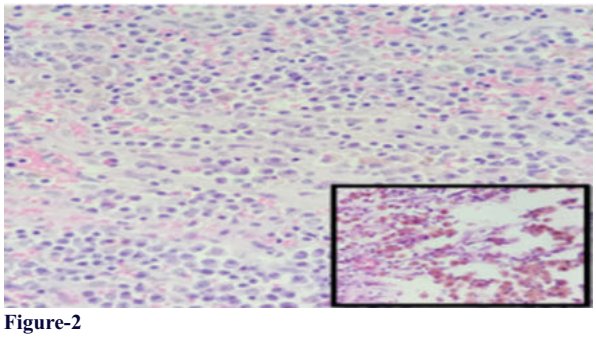

Figure-2

DISCUSSION

Xanthogranulomatous inflammation is a chronic destructive process of the normal tissue and a rare occurrence in the female genital tract. Kunakemakorn et al. (1976) described the first case of Xanthogranulomatous inflammation of uterus, left fallopian tube and ovary in their report of inflammatory pseudotumor in the pelvis in 1976. The youngest case was reported in a 2-year-old by Tanwar H et al (2015).[4] Most frequently it occurs in females of reproductive age group (range: 23 - 72 years), the average age of patients with affected ovaries being 31 years.[5]

The exact pathogenesis of Xanthogranulomatous oophoritis is not very clear and remains a mystery, however, many theories explaining the etiopathogenesis have been postulated, such as theory of infection, endometriosis, intrauterine contraceptive device, inborn errors of lipid metabolism, and drug induced. Amongst all these theories, the most widely accepted theory is that of infection, which is supported by clinical evidence of infection and growth of bacteria such as Escherichia coli, Bacteroides fragilis, and Proteus vulgaris from the affected tissue by culture,[6] which is similar to our case where Escherichia coli was isolated from the pus drained from the tubo-ovarian cyst.

Patients usually presents with symptoms like anorexia, fever, suprapubic pain, adnexal tenderness or a pelvic mass.[7] The gross examination of the affected ovary may show replacement by a solid, yellowish, lobulated well circumscribed mass along with cystic change, sometimes involving the adjacent structures. Microscopically, it is characterized by a massive infiltration by lipid laden histiocytes known as Xanthoma cells and mixed with inflammatory infiltrates (Figure 3). Immunohistochemical stains CD68, CD3 and CD20 are helpful in establishing the diagnosis.[8]

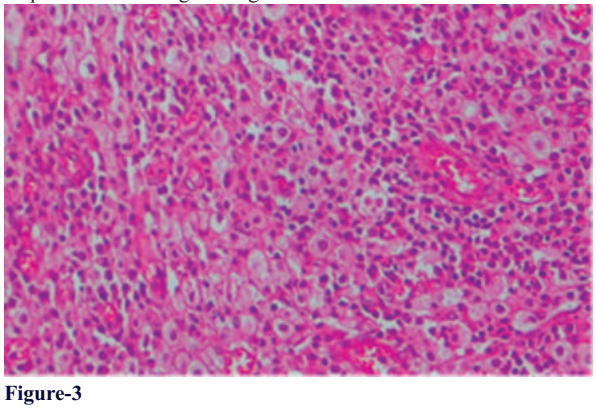

Figure-3

Tumour markers for ovarian malignancy, serum Ca 125 values may be marginally raised.[9] The mass may grow large in size up to 3-7cm and the inflammation when extends to the neighbouring organs; pelvic structures and peritoneum can present as adhesions on radiological examination raising the suspicion of malignancy.[10] Therefore, neoplastic lesions should be ruled out with the help of detailed investigations. Frozen section should be considered in difficult cases in order to avoid misdiagnosis and extensive surgeries in young patients, as was done in our case. Similarly, Chouairy et al (2012) are of the view that Xanthogranulomatous oophoritis may be preoperatively misdiagnosed as an adnexal neoplasm, thus necessitating frozen section to rule out malignancy.[11]

Differential diagnosis to be considered for this entity includes infections like tuberculosis, fungal infections which can be ruled out by culture and special stains for the causative organisms. Malakoplakia is also one of the differential diagnoses which can be differentiated from Xanthogranulomatous inflammation by the presence of basophilic cytoplasmic concentric calcific bodies within histiocytes known as Michaelis-Gutmann bodies. Wather et al (1985) presumed that malakoplakia and Xanthogranulomatous inflammation were identical chronic inflammatory diseases.[12]

The current case was similar to other reported cases in its presentation and histopathological findings as described, favouring a diagnosis of Xanthogranulomatous oophoritis. Treatment of choice for Xanthogranulomatous salpingooophoritis is salpingooophorectomy as was done in this case. Antibiotic therapy has been attempted, but without much success.[13]

CONCLUSION

Xanthogranulomatous oophoritis is a rare condition, which can often pose as a diagnostic challenge for the clinicians, radiologists and the pathologists since the clinico-radiological presentation often mimics malignancy. However, the pathologists need to be aware of this entity and should be vigilant before giving a diagnosis to prevent any misdiagnosis of malignancy. Frozen section can be used in cases of dilemma to rule out malignancy and avoid extensive radical surgeries. A careful histopathological examination aided by immunohistochemistry can provide definitive diagnosis.

CONFLICTS OF INTEREST

None.

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