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Case Report

Diagnostic predicament of bilateral ovarian masses with rare and distinct histopathology

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ABSTRACT

Xanthogranulomatous oophoritis is a rare, peculiar, non-neoplastic, chronic inflammatory pathology that can mimic ovarian mass. Affected organ shows replacement of normal tissue with foamy histiocytes, multinucleate giant cells, neutrophils, fibroblasts, plasma cells and areas of necrosis within. Its aetiology is still unknown, and the final diagnosis is usually made through histopathological examination. The present case report describes an unusual presentation of simultaneous occurrence of Xanthogranulomatous oophoritis and fibrothecoma masquerading as bilateral tubo-ovarian masses in a post-menopausal female. Despite initial assumptions and suspicions of tubo-ovarian abscesses, surgical exploration revealed an unexpected pathology. Histopathological evaluation confirmed the diagnosis, emphasising the importance of vigilant and comprehensive assessment in atypical presentations.

Keywords: Xanthogranulomatous oophoritis, Ovarian mass, Fibrothecoma, Histopathology

INTRODUCTION

Xanthogranulomatous inflammation is a non-neoplastic chronic inflammatory condition characterised by extensive replacement of the native tissue with inflammatory cells like foamy macrophages, lymphocytes, plasma cells and neutrophils. As a result, the affected organ has structural destruction and functional failure.[1] Although xanthogranulomatous inflammation is rare, the most common organs this pathology affects include the kidneys and gall bladder.[2] Only a limited number of cases involving the female genital tract have been reported, predominantly with endometrial involvement, and ovarian involvement is somewhat sporadic.[2,3] The exact aetiology of xanthogranulomatous oophoritis is unknown, but it shows a histological appearance similar to other organs.[4] On the other hand, ovarian fibro thecomas are rare sex cord-stromal tumours, accounting for only 1-4.7% of ovarian tumours.[5] They mainly occur during the post-menopausal period, with 90% of cases showing unilateral involvement.[6,7] Although both these entities have been reported independently in literature, the simultaneous occurrence of xanthogranulomatous oophoritis and ovarian fibrothecoma in the same patient has not yet been documented to date, to our knowledge.

CASE REPORT

We present a case of a 50-year-old post-menopausal P4L2 lady who presented to the gynaecology OPD with chief complaints of lower abdominal pain for the past year. There were no other significant clinical complaints. The patient had a known case of type 2 diabetes mellitus on treatment for five years and had a history of pacemaker insertion in 2021. There was a family history of uterine cancer in her mother 15 years ago. On clinical examination, the abdomen was soft and non-tender, with normal cervix and vagina on per-speculum examination. Per vaginal and rectal examination, they revealed a 4×4 cm mass in the right fornix and a 6×6 cm solid mass in the pouch of Douglas. The left fornix and rectal mucosa were free. Routine investigations were within normal limits, including complete blood cell counts, white cell counts, erythrocyte sedimentation rate, and urine routine and

microscopic examinations. Serum amylase and lipase were also within normal ranges.

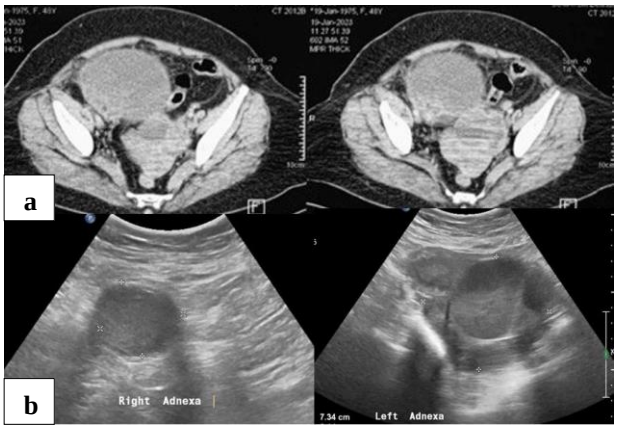

Figure 1: (a) CT scan showing bilateral complex adnexal lesions; right being larger than left; and (b) US abdomen after 6 months showing significant reduction in the size of right adnexal lesion and mild increase in the size of left adnexal lesion with a dilated tubular structure alongside.

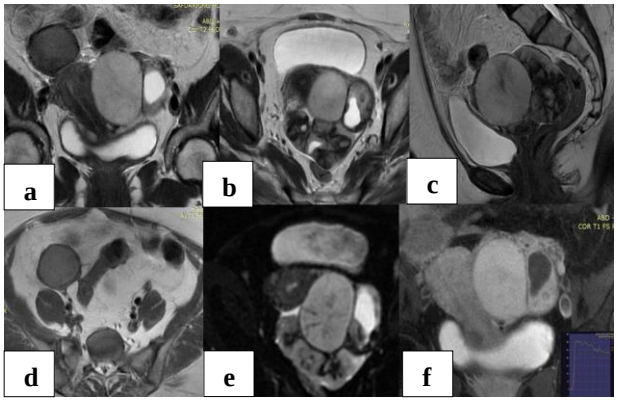

Figure 2: (a-c) MRI T2WI appearing hyperintense with central hypointense scar; (d) right sided lesion appearing hyperintense on T1WI; (e) absence of restricted diffusion; and (f) post contrast scan with heterogeneous enhancement.

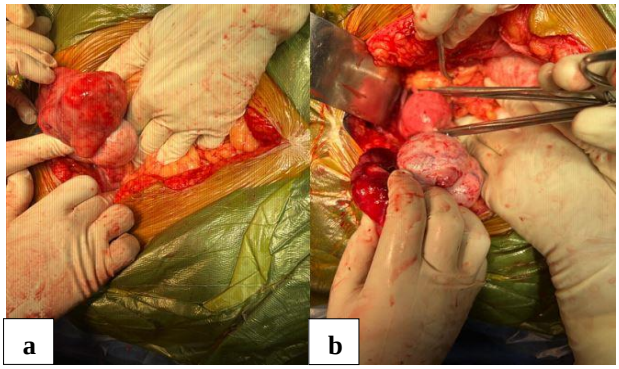

Figure 3 (a and b): Intra-operative findings showed solid enlarged right ovary and solid cystic morphology with exophytic growth involving left ovary.

The patient was then referred to the department of radiodiagnosis. She already had a contrast enhanced computed tomography (CECT) abdomen done six months back in her hometown. The scan revealed a large, well-defined, homogeneous fluid-attenuating abdominopelvic cystic lesion with thick enhancing walls on the right side, measuring 6.6×4.2 cm. No evidence of calcific foci or solid components was seen within. Another minor multiloculated lesion with few enhancing thin internal septations was seen in the left adnexa, measuring 4.6×3.2 cm. The patient was put on antitubercular therapy (ATT), considering the possibility of inflammatory aetiology. However, the clinical symptoms did not resolve.

At present, the transabdominal ultrasound (US) showed a well-defined thick-walled homogeneously hypoechoic right adnexal lesion with ground glass appearance and posterior acoustic enhancement, measuring 3.9×3.5 cm in size. No evidence of any internal vascularity was seen on colour Doppler. The left adnexa showed another similar morphology, a more significant lesion measuring 7.3×8.1 cm, with few thin vascular septae. A dilated tubular structure with incomplete internal septations was seen in the left adnexa - likely a dilated left fallopian tube. Bilateral ovaries were not visualised separately from the lesions. Hence, the right adnexal lesion had considerably reduced size, but the left-sided lesion had progressed. The diagnosis of bilateral endometriomas was considered, with another possibility of tubo ovarian masses (infective aetiology) as a differential diagnosis.

For further characterisation of the lesions, CEMRI of the pelvis was done in August 2023, showing a well-circumscribed oval solid lesion in the left adnexa measuring 8.2×7.3×6.2 cm in size, appearing isointense on T1 images, hyperintense on T2/T2FS images with central hypointense scar. The lesion showed intensely restricted diffusion with no evidence of any susceptibility-weighted artefacts. Heterogeneous enhancement on post-contrast images was seen with a non-enhancing central area. A circumferentially dilated tubular structure surrounding the lesion showed peripheral enhancement and diffusion restriction. The right adnexa showed a well-defined oval T1/T2 hyperintense lesion measuring 4×3.8×3.5 cm, with peripheral enhancement. No evidence of any susceptibility-weighted artefacts or diffusion restriction was seen. Bilateral ovaries were not visualised separately from the lesion. The uterus appeared normal in morphology for age. The above imaging findings diagnosed a bilateral tubo-ovarian abscess (likely tubercular aetiology).

The patient's symptoms did not resolve despite conservative medical management and intake of ATT for eight months. Surprisingly, a rise in serum markers was noted with CA-125 levels 146 U/ml (normal range 0-35 U/ml). Finally, after discussion in a multi-disciplinary tumour board, the patient underwent exploratory laparotomy followed by transabdominal hysterectomy with bilateral salpingo-oophorectomy (TAH+BSO) with

infrasonic omentectomy. Multiple peritoneal biopsies with peritoneal cytology were also done. Intra-operative findings showed a solid enlarged right ovary and solid cystic morphology with a solid exophytic growth involving the left ovary. The uterus and bilateral fallopian tubes were normal.

The final histopathological report of the right ovary showed large areas of necrosis with sheets of foamy histiocytes, moderate lymphoplasmacytic infiltrates and numerous multinucleated giant cells and hemosiderin-laden macrophages – suggestive of xanthogranulomatous oophoritis. However, surprisingly, the final histopathological report of the left tubo-ovarian mass showed tumour cells arranged in intersecting fascicles and a storiform pattern with immune-histochemistry (IHC) stain positive for WT-1- suggestive of fibrothecoma, left ovary.

DISCUSSION

Xanthogranulomatous inflammation of the female genital tract, particularly ovarian tissue, is an exceedingly rare pathology, and the exact etiopathogenesis of its occurrence is unknown. Various past hypotheses have been proposed in the literature, including persistent genital infections, endometriosis, intra-uterine devices, inborn errors of lipid metabolism in macrophages and ineffective antibiotic therapy along with a combination of these factors.[8] Studies have reported infection with causative organisms such as *Escherichia coli*, *Staphylococcus aureus*, *Bacteroides fragilis*, *Proteus* and *Actinomyces* as a probable cause. Glove dusting powder and uterine artery embolization have also been proposed in etiopathogenesis.[9] It mainly affects females in the reproductive age group between 23-72 years.[10] Patients usually present with features of chronic pelvic inflammatory disease and tubo-ovarian mass, because of which it is commonly mistaken for malignancy or genital tuberculosis.[11]

A similar presentation was also seen in our patient, who had long-standing lower abdominal pain, which was not resolving despite conservative management with the course of antibiotics. Perplexing radiological imaging findings with normal serum markers initially lead to a misdiagnosis of likely tubercular aetiology. However, no clinical improvement was seen even on initiating ATT. Another unusual feature in our case was the increasing size of the left adnexal lesion, which showed internal vascularity. Initially thought to be an infective aetiology, persistent clinical complaints with an increase in the size of the mass lesion and new onset mildly increased serum CA-125 levels were puzzling. Despite sincere attempts to reach a pre-operative diagnosis, the definite diagnosis could only be made on histopathological examination, parallel with previous xanthogranulomatous oophoritis reported in the literature by various authors.[12-14] On the other hand, to our surprise the left adnexal lesion was a simultaneous ovarian fibrothecoma. Fibrothecomas are a rare type of sex cord-stromal tumour seen in perimenopausal females that typically presents with non-specific signs and symptoms secondary to mass effect causing chronic pelvic pain and metrorrhagia. Rarely, they may present with instances of hormonal production, cause encasement of organs or related to Meigs syndrome.[15] Most fibro thecomas/fibromas are solid masses with oval/lobulated appearance that might cast acoustic shadows due to fibrous tissue.[16]

However, as in our case, this is not always true, and atypical imaging findings can misdiagnose them as tubo-ovarian abscesses or malignancies. Like other solid ovarian tumours, surgery is the mainstay of management for confirmation of diagnosis, as in our case. The patient underwent surgical exploratory laparotomy, which revealed the definitive diagnosis.

Unclear etiopathogenesis of xanthogranulomatous oophoritis with numerable proposed hypotheses and simultaneous occurrence of ovarian fibrothecoma might suggest a possible role of common risk factors predisposing the ovary to both inflammatory and neoplastic processes. Alternatively, chronic inflammation within the ovary due to fibrothecoma may have triggered secondary xanthogranulomatous inflammatory changes in the contralateral ovary.

To our knowledge, this is the first case in literature with simultaneous occurrence of xanthogranulomatous oophoritis and ovarian fibrothecoma in same patient. However, a similar exceedingly uncommon case of mucinous cystadenoma of the ovary with concurrent xanthogranulomatous oophoritis has recently been reported in the literature by Nouh et al.[17]

CONCLUSION

Xanthogranulomatous oophoritis and ovarian fibrothecomas are both uncommon entities. The simultaneous occurrence of these rare pathologies in the same patient makes this case unique and noteworthy. This case highlights the need for further research on such associations and possible common etiological factors for the concurrent occurrence of ovarian inflammatory and neoplastic pathologies. It also underscores the importance of a comprehensive and multi-disciplinary approach, including clinicians, radiologists and pathologists, for unravelling complex ovarian diseases, which often mimic common pathologies like tubo-ovarian abscesses. Documentation and extensive dissemination of such rare associations and cases are crucial to expand the existing literature and knowledge of clinicians and radiologists so that these can be thought of or kept in a list of rare differentials when challenged with such atypical ovarian pathologies.

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