

CASE REPORT

Female Genital Tract Affected by Xanthogranulomatous Inflammation: A Series of Four Cases and Review of Literature

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ABSTRACT

Objective: Xanthogranulomatous inflammation is a form of chronic inflammation that rarely affects the female genital tract. Endometrium is most commonly involved in the genital tract; ovary and fallopian tube is rarely affected. The purpose of this case series is to make clinicians familiar about this rare condition.

Case description: Here we are presenting a series of four cases, three of xanthogranulomatous oophoritis and one of xanthogranulomatous salpingitis each. One of our cases appeared as carcinoma ovary and other as metastasis of carcinoma cervix to ovary.

Conclusion: Xanthogranulomatous inflammation of the female genital tract is an uncommon entity, clinical and radiological features of this entity may mimic ovarian tumor, and hence it must be considered in the differential diagnosis of ovarian mass. Histopathological examination is essential for its diagnosis. Awareness of this entity is vital to avoid an incorrect and overdiagnosis of malignancy.

Keywords: Inflammation, Oophoritis, Salpingitis, Xanthogranulomatous.

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INTRODUCTION

Xanthogranulomatous inflammation is a form of chronic inflammation characterized by an abundant clustering of lipid-laden histiocytes admixed with other inflammatory cells. These are nonspecific but characteristic to xanthogranulomatous inflammation. This inflammatory response destroys normal tissues. This type of inflammation is most commonly seen in kidneys and gallbladder. But can be seen in other organ systems such as pancreas, appendix, eyes, female genital tracts, colon, and urachus. Genital tract involvement is rare with this condition.¹ Kunakemakorn was the first to report inflammatory pseudotumor in the pelvis.² Here we are presenting a series of 4 cases, 3 of xanthogranulomatous oophoritis and 1 of xanthogranulomatous salpingitis each. One of our cases appeared as metastasis of carcinoma cervix to ovary. This inflammation has been seen rarely in association with carcinoma endometrium but association with carcinoma cervix is rarer.

CASE DESCRIPTION

Case 1

A 24-year-old P₂L₂ female was presented to us with complaints of dull aching, progressively increasing pain in lower abdomen for the last 3 years. She also complained of dyspareunia, burning micturition, and increased frequency of micturition for the last 3 months. She was having heavy menstrual bleeding for last 3 years. Her gynecological examination showed discharge per vaginum, normal-sized uterus with elongated tender mass felt attached to the right side of the uterus. Her transvaginal sonography showed a normal-sized uterus with a 6.6 × 4.5 cm well-circumscribed mass just adjacent to the right lateral wall of uterus, left adnexa was normal. Antibiotics were given with suspicion of PID. However, symptoms did not resolve; so we planned for laparoscopy. Intraoperative right ovary was enlarged (4 × 5 cm) and densely adherent to posterior uterine wall, forming a tuboovarian mass, 20–30 mL thick purulent material was drained from it. Left tube and ovary was normal. Ovarian abscess drainage and removal of right ovary was done as

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much as possible; the entire right ovary could not be removed as it was densely adherent to uterus. Her post-operative course was uneventful. Final histopathology report showed dense infiltration of histiocytes, plasma cells, and lymphocytes. No granulation or infective organism was seen. Overall features were consistent with xanthogranulomatous oophoritis. Patient improved symptomatically after the surgery and following with us in good health.

Case 2

64-year-old P₃L₃ postmenopausal female presented with complaints of postmenopausal bleeding and foul smelling discharge per vaginum for the last 3 months. Gynecological examination showed no visible growth on cervix or vagina; uterus was postmenopausal size with free bilateral fornix and parametrium. Pap smear was done, which showed highly atypical cells suggestive of malignancy. Colposcopy-guided cervical biopsy showed moderately differentiated squamous cell carcinoma. Clinical diagnosis of carcinoma cervix stage 1a was made. Exploratory laparotomy with the plan of type III radical hysterectomy was done under GA. A friable growth of 3 × 3 cm was there in right ovary extending on sigmoid colon. Right ovary, fallopian tube, and friable mass over sigmoid colon was removed and sent for histopathological evaluation. Radical hysterectomy

was abandoned as carcinoma cervix was inoperable owing to suspected ovarian metastasis. Histopathological evaluation showed xanthogranulomatous oophoritis of the right ovary.

Case 3

A 39-year-old P₂L₁ female presented with pain in lower abdomen for last 10 months. She had average menstrual flow with dysmenorrhea. Her pervaginal examination showed uterus of 10 weeks size with restricted mobility and vague tender mass was palpable in both fornices. She received antibiotics for PID but did not relieve. Imaging showed a right-sided multiloculated complex cystic tuboovarian (TO) mass of size 8 × 6 × 8 cm with omental thickening with ascites (Fig. 1A). CA125 was raised (191 IU/mL). Laparotomy was planned with suspicion of carcinoma ovary, intraoperatively multiple mesenteric cysts extruding straw colored fluid was there in abdomen. Right-sided tuboovarian mass with matted intestine densely adherent to TO mass and uterus was seen, which were removed. Pouch of Douglas was obliterated. Hysterectomy with bilateral salpingoophorectomy and omental biopsy was done (Fig. 1B). Histopathology evaluation showed xanthogranulomatous oophoritis of right ovary with unremarkable left ovary and uterus (Figs 1C and D).

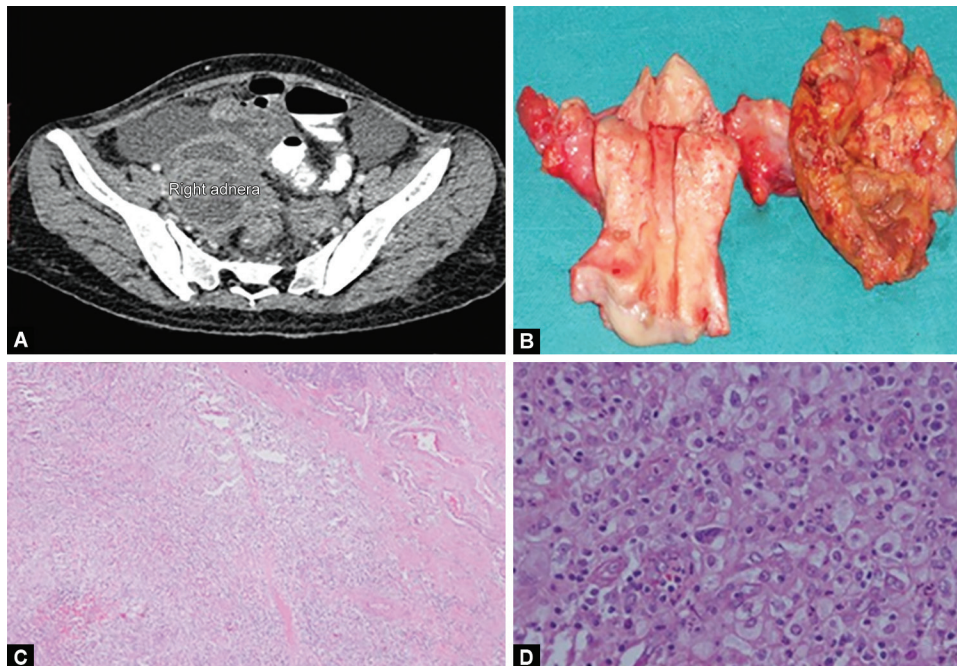
Case 4

36-year-old nulliparous female presented with primary infertility of 10 years. She had average menstrual flow without dysmenorrhea. Patient received complete antitubercular treatment for genital tuberculosis 8 years back. Ultrasonography showed bilateral TO masses. She received doxycycline for PID. Laparoscopy and tubal clipping or salpingectomy was planned in view of hysterosalpingogram showing bilateral hydrosalpinx with tubal block. Intraoperative picture showed bilateral hydrosalpinx with TO masses around 4 × 4 cm. Bilateral salpingectomy was done. Histopathological evaluation showed xanthogranulomatous

salpingitis of the right fallopian tube with unremarkable left fallopian tube.

DISCUSSION

Genital tract involvement of xanthogranulomatous inflammation is a rare entity. When genital tract is involved, endometrium is the most commonly affected part, ovarian and fallopian tube involvement is rare.^{3,4} Involvement of cervix has also been reported.^{5,6} Inoue et al. reported involvement of parametrium with this inflammation without endometritis or salpingitis in a 68-year-old lady, this was first reported case of this type.⁷ Exact etiology and pathogenesis is not very clear but pelvic inflammatory disease (PID), abscess, necrosis, and hemorrhages play a role in eliciting this inflammatory response so far reported in the literature. PID is the most frequent association, infectious agent isolated was *Escherichia coli* in most of the cases reported.^{3,8} Other organisms isolated were *Klebsiella*,⁹ *Bacteroides fragilis* and *Streptococcus mitis*.¹⁰ Uterine artery embolization as a causative factor has also been reported.⁹ Tai et al. reported xanthogranulomatous inflammation of both fallopian tube and urinary bladder simultaneously in a 52-year female.¹¹ Xanthogranulomatous oophoritis is mostly seen in reproductive-age females but can be seen in any age group. The average age is 38.5 years (range 23–72).¹ Though can occur in any age group, the youngest reported case was of a 2-year-old female child who presented with a mass in the abdomen.¹² Patients were present with pain in the lower abdomen, fever, discharge per vaginum, and sometimes anorexia. Symptoms can be there for days^{12,13} or for years.¹⁴ Examination identifies adnexal mass. Laboratory investigations show raised markers of inflammation (e.g., erythrocyte sedimentation rate and total leukocyte count). Contrast-enhanced computed tomography shows thick-walled complex solid–cystic lesions and variably enhancing solid areas, which appear hypo intense on T₁ weighted MRI image.¹⁵ Intraoperatively



Figs 1A to D: (A) Computed tomography image showing right adnexal mass; (B) Cut section of the specimen showing right tuboovarian mass; (C) Dense infiltration of histiocytes, plasma cells, and lymphocytes (10× view); (D) Dense infiltration of histiocytes, plasma cells, and lymphocytes (40× view)

ovary appears replaced by an yellowish brown mass, adherent to adjacent organs, sometimes containing purulent material inside. Both gross and radiological examination may mimic malignancy as in two of our cases. Only histopathological examination of tissue can differentiate it; so it plays an important role in diagnosing this entity and hence guides further management. They can also be confused with tubercular TO masses, which also may have similar presentation; again histopathology (HPE) can only differentiate these two. Xanthogranulomatous inflammation on HPE will show inflammatory exudates comprising sheets of foamy macrophages, lymphocytes, plasma cells, and some neutrophils, while tubercular TO masses will show granuloma with central caseating necrosis surrounded by epithelioid histiocytes. It is an inflammatory process. Removal of mass is preferred modality of treatment as it responds poorly to medical management by antibiotics as seen in three of our cases. Misdiagnosed as ovarian malignancy can lead to radical surgery as in the third case presented here. So keeping this diagnosis as differential will prevent unnecessary surgery especially in young patients.

CONCLUSION

Xanthogranulomatous inflammation of the female genital tract is an uncommon entity, clinical and radiological features of this entity may mimic an ovarian tumor, and hence it must be considered in the differential diagnosis of ovarian mass. Histopathological examination is essential for its diagnosis. Awareness of this entity is vital to avoid an incorrect and overdiagnosis of malignancy.

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