

## Xanthogranulomatous Salpingo - Oopheritis, a Diagnostic Dilemma in Different Age Group: A Case Series

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### Abstract

Xanthogranulomatous inflammation of female genital tract is extremely rare entity. Very few cases have been documented in literature. It is an uncommon cause of chronic destructive process of the normal tissue of the affected organ.

We presented 3 cases of xanthogranulomatous affection of tube and ovary in 27 years, 30 years and 74 years postmenopausal woman with unilateral hydronephrosis without having history of all known predisposing factors like pelvic inflammatory disease (PID), endometriosis, intrauterine contraceptive devices (IUCD). Their clinical examination, radiological evaluation, tumor markers, were likely to get confused with neoplasm in first case and large uterine fibroid mass in second case. Peroperatively It was found tubo-ovarian mass. On histopathological examination it came out with xanthogranulomatous inflammation of tube and ovary. So in this clinical scenario we should keep xanthogranuloma as one of differential diagnosis. It is characterized by the presence of lipid-filled macrophages with admixed lymphocytes, plasma cells, and neutrophils.

**Keywords:** PID; IUCD; Xanthogranuloma.

### Introduction

Xanthogranulomatous salpingo-oophoritis is an uncommon form of chronic inflammation in the genitourinary tract, affect more commonly the kidneys [1]. Other organs affected xanthogranulomatous inflammation involvement are stomach, anorectal area, bone, urinary bladder, testis, and epididymis.

Xanthogranulomatous inflammation of the ovary and fallopian tubes is rare and if found is mostly limited to the endometrium [2]. Only few cases of ovarian and fallopian tube involvement have been reported [2,3]. It is a histopathological diagnosis. On microscopic appearance a large number of lipid containing macrophages replace the tissue with an admixture of lymphocytes, plasma cells, and neutrophils, characteristic of xanthogranulomatous inflammation [4]. Multinucleated giant cells may be present. Xanthogranulomatous lesions of ovary may be confused with neoplasm due to its clinical presentation, radiological appearance, and same gross features.

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In these cases we described xanthogranulomatous inflammation in the female genital tract. We also emphasize the need for awareness of this condition to avoid patient morbidity and extensive surgery due to its clinical features and diagnostic pitfalls.

### Case Report 1

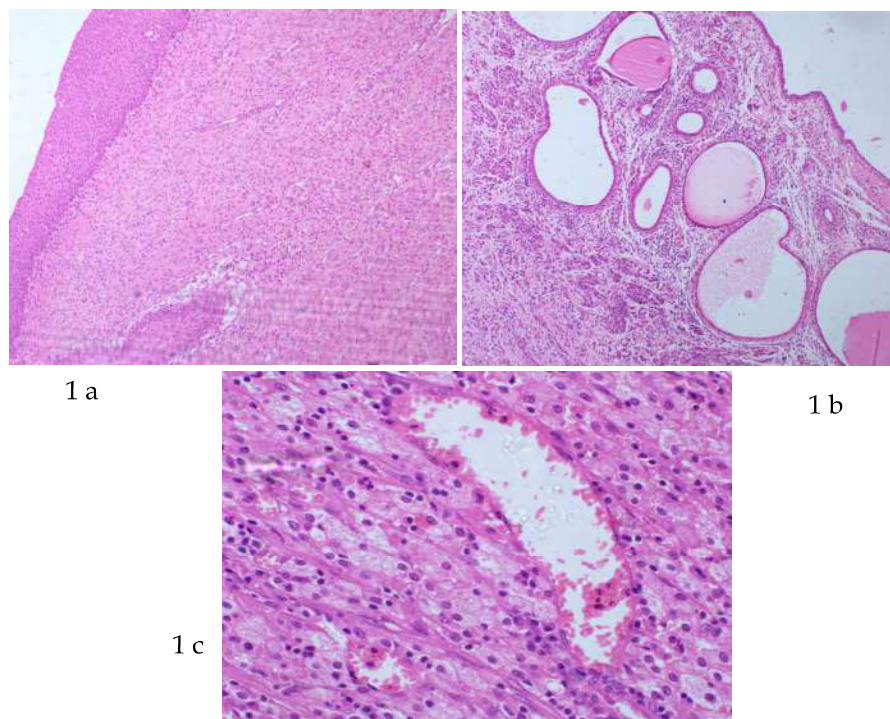
A 27 years old p3003 with history of previous three cesarean sections presented with complains of pain and lump in left iliac fossa region for last 3 months after her last cesarean section. Pain was mild to moderate in intensity without any radiation. It was not associated with food intake, vomiting, hematemesis, malena, loss of appetite, weight loss or altered bowel habits. No h/o urinary complains or menstrual irregularities. On abdominal examination there was a palpable lump in left iliac fossa approx 5 cm in size and mildly tender. On USG ill defined heterogenous echoic complex mass 51x38 mm seen in left iliac region. Uterus and ovaries are normal. CA 125 was 9 IU/ML. On CECT abdomen an oval shaped area 4.9x2.6 cm seen in left iliac fossa showing soft tissue attenuation with small cystic areas continuing left side of uterus. Left ovary is not separately seen. Laparotomy and excision of left ovarian mass was done. Peroperative the left ovary with a cystic lesion adherent to parietes in left iliac fossa was seen.

Omentum was adherent to ovarian mass. On cut section the ovarian mass with solid cystic areas with small amount of caseating material present.

On histopathology- sections from left tube and ovary shows dense mixed inflammatory cell infiltrates composed of sheets of foamy macrophages, plasma cells, lymphocytes, neutrophils and eosinophils. No evidence of malignancy. Diagnosis of left sided xanthogranulomatous salpingo-oophoritis was made (Fig. 1).

### Case Report 2

A 74-year-old multiparous (P4004) post-menopausal woman presented with right flank pain for 2 months and high grade fever on and off for last one month. On USG KUB- in right kidney gross hydronephrosis was present with normal left kidney. TLC-11,200/cm<sup>3</sup>, serum creatinine was 0.63 mg/dl. On urine examination 30-40 WBCs/HPF was present. On urine culture growth of E.Coli was present. On CT Urography Hydro-ureteronephrosis was present till lower ureter. Lower ureter was compressed by large uterine mass of 5.9x4.3x3.8 cm showing calcifications and heterogeneous enhancement with loss of fat planes with ureter. Left kidney and urinary bladder was normal.



**Fig. 1:** Histopathology sections of Cervical section (1a), Endometrial section (1b), Xanthogranulomata at high resolution (1c).

**Table 1:** Summary of existing literature published on salpingo-oophoritis

S. No.	Author	Clinical Entity	Age (Yrs)	Risk factors	Presenting Complaint	Examination Findings	Operation Done
1	Singh UR (1995)	Oophoritis	25	Endometriosis S. typhi infection	Abdominal pain Abdominal mass	Suprapubic mass Enlarged uterus	Partial Oophorectomy
2	Y Gray (2001)	Salpingo-oophoritis	47	PID IUCD in situ Endometriosis	Abdominal pain Loose stools	Fullness in left abdomen	TAH Bilateral SO
3	Furuya M (2002)	Salpingitis	19	Chlamydia cervicitis	Abdominal pain Fever Vaginal discharge	Left adnexal mass	Left SO
4	Furuya M	Salpingitis	49	Leiomyoma Endometriosis	Abdominal pain Fever	-	TAH Bilateral SO
5	Seung Eun Jung (2002)	Oophoritis	48	-	Abdominal pain Fever	Bilateral adnexal mass	TAH Bilateral SO
6	Punia RS (2003)	Salpingo-oophoritis	42	PID	Menorrhagia	Bilateral adnexal masses	Subtotal hysterectomy Bilateral SO
7	Idrees M (2007)	Salpingitis	41	Endometriosis Endometritis IUCD Previous chemotherapy	Abdominal discomfort	Bilateral tender and enlarged adnexa masses	Bilateral SO
8	Altanis S (2007)	Oophoritis Endometritis	84	Diverticulitis Previous chemotherapy	Post menopausal bleed	-	TAH Bilateral SO (could not exclude endometrial mass)
9	Howey JM (2010)	Salpingitis	50	Leiomyoma Previous surgery (appendicectomy) Endometriosis	Abnormal uterine bleeding	Enlarged uterus	TAH Left SO
10	Shukla S (2010)	Oophoritis	42	Endometriosis Fibroids	Abdominal pain	Midline abdominal mass	TAH Bilateral SO Omentectomy
11	Yener N (2011)	Salpingitis	41	Secondary infertility	Abdominal pain	Bilateral adnexal tenderness	Bilateral SO
12	Singh R (2011)	Salpingitis	37	-	Abdominal pain Vaginal discharge	Tender right adnexal mass Cervical excitation	-
13	Abeyandara PK (2012)	Salpingo-oophoritis	34	Endometriosis Leiomyoma Primary subfertility Previous surgery	Intestinal obstruction	Distended abdomen	Laparoscopic THBSO Appendicectomy
14	Kalloli M (2012)	Oophoritis	45	PID IUCD	Abdominal pain	Left adnexal mass	TAH Bilateral SO Omentectomy
15	Zhang XS [10] (2012)	Salpingitis	29	-	Abdominal discomfort Fever Vaginal discharge	Left adnexal mass	Left SO
16	Zhang XS	Oophoritis	32	-	Abdominal pain Fever	Left adnexal mass	TAH Bilateral SO
17	D Shilpa [11] (2013)	Oophoritis	18	-	Abdominal pain, fever	Right adnexal mass	Rt SO
18	SM Bindu (2014)	Oophoritis	40	-	Abdominal pain, bleeding per vaginum	Right adnexal mass	TAH Bilateral SO Omentectomy
19	H. Tanwar [12] 2015	Salpingo-Oophoritis	2	-	Abdominal pain, vomiting	Right Adenexal mass	Rt SO
20	Si Ying Pang [13] (2016)	Salpingo-oophoritis	45	Fibroid	Abdominal pain, fever	Right Adenexal mass	TAH with B/L SO with frozen section biopsy
21	Si Ying Pang	Oophoritis	36	Endometriosis	Menorrhagia	Left Adenexal mass	Lt SO
22	Si Ying Pang	Salpingitis	48	Chronic PID	Abdominal pain, Dyspariunia	Left Adenexal mass	TAH with B/L SO

On Transvaginal USG uterus was postmenopausal uterine fundal mass with patchy calcifications and necrosis was present. Diagnosis of uterine fundal/broad ligament fibroid was made. CA-125 was normal. The patient was counseled for total abdominal hysterectomy with b/l salpingo-oophorectomy. In view of Right Hydronephrosis, right percutaneous nephrolithotomy (PCN) drain inserted. It was draining urine around 900 ml/day. On GFR study showed split function of 28% in right kidney.

Right sided 6/26 DJ stenting followed by laparotomy done. On exploration multiple jejunal diverticulams were present. A 5 x 5 cm right ovarian mass was present with thick yellow pus. The ovary was adhered to right pelvic wall and ureter. Ureter was separated from mass. Left ovary, uterus and cervix was grossly normal. Pus was taken for culture and AFB staining. Total abdominal hysterectomy with bilateral salpingo-oophorectomy done. PCN was removed on postoperative day 7. Microscopy and culture for Acid Fast Bacilli done on right PCN fluid, right ovarian cyst pus and urine, which was negative. Patient discharged in stable condition with right DJ stent in situ.

On histopathology- Sections from right tubo-ovarian mass shows dense aggregates of foamy macrophages, giant cells, and lymphocytes along with cholesterol clefts, fibrosis and multiple lymphoid aggregates. Stain for an acid fast bacillus is negative. There is no evidence of necrosis or malignancy.

### Case Report 3

30 years old lady presented with complain of abdominal pain. On USG there were bilateral adenexal lesions with solid component. Right sided lesion was 4.4x5.7 cms with collection in POD, minimal ascitis was present. On pervaginal examination uterus was normal size. In bilateral fornices masses felt (right>left) with restricted mobility. Laparotomy with left salpingo-oophorectomy done in view of bilateral tubo-ovarian mass with high ROMA scoring for CA ovary. In preoperative findings there was dense adhesions of uterus bilateral tubes and ovaries with rectosigmoid junction and surrounding peritoneum and omentum. Left ovary was 6-7 cm but right ovary was not clearly visible due to adhesions but was around 4 cm.

On histopathology- sections from ovary shows fibrosis and dense mixed inflammatory cell

infiltrates comprising of lymphocytes, plasma cells, neutrophils and eosinophils. Sheet of foamy macrophages are seen. Zeihl nelsen stain was negative for Acid fast bacilli. There was no evidence of malignancy. Cytology for malignant and Bact 3D alert test for AFB was negative.

### Discussion

The exact mechanism of this inflammation in the female genitourinary tract is unclear. Risk factors described in literature include infection, endometriosis, [5] leiomyoma, [6] inadequately treated PID, [3] IUCD in-situ, [7] abnormal lipid metabolism, immunosuppression and previous abdominal surgery. Other risk factors include diabetes mellitus type 2, hyperlipidemia, untreated urinary infections, [5] inborn errors of lipid metabolism, and drug induced. Most accepted theory is of infection, supported by clinical evidence of infection and growth of bacteria such as E.coli, Bacteroides fragilis, and Proteus vulgaris from the affected tissue by culture [3].

On radiological investigations xanthogranulomatous salpingo-oophoritis may be confused with malignant ovarian tumor. The clinical presentations of xanthogranulomatous salpingo-oophoritis include fever, abdominal mass, and pain in the abdomen, menorrhagia, anemia, and anorexia. On gynecological examination tender adenexal mass with blood investigations showing elevated ESR and raised white blood cell count. Grossly, the affected ovary is usually enlarged and looks like a tumor with yellowish appearance, with cystic areas with or without pus filled within it.

Xanthogranulomatous salpingo-oophoritis is a histological diagnosis. In addition, immunohistochemical stains are helpful in establishing the diagnosis, such as CD68 in foam cells, CD3 (T lymphocyte marker), CD20 (B lymphocytes marker) [8], Fontana-Masson stain (lipofuscin in foamy macrophages). The makaloplakia is another chronic inflammatory entity of the genitourinary tract. The absence of Michaelis-Gutmann bodies excludes diagnosis of makaloplakia. The leukocyte common antigen (LCA) immunostaining can differentiate between xanthelasma of the fallopian tube and xanthogranulomatous salpingo-oophoritis [9].

A summary of existing literature on salpingo-oophoritis with details on the proposed risk factors, clinical findings and management has been made in Table 1. [10,11]. The youngest case reported was

of age 2 years in existing series [12]. Treatment of choice for xanthogranulomatous salpingo-oophoritis is salpingo-oophorectomy. Medical treatment with antibiotics is not successful in reducing ovarian mass.

### Conclusion

Xanthogranulomatous salpingo-oophoritis is a rare chronic inflammatory condition of the female reproductive tract. It is diagnosed on histopathological findings. xanthogranulomatous salpingo oophoritis as one of the differential diagnoses should be kept in mind while dealing with tubo-ovarian masses in any age group. This condition can be diagnosed with surety with awareness of the condition and proper history, clinical examination, investigations and histopathological correlation. This will help in preventing misdiagnosis and radical surgeries if misdiagnosed as neoplasm of ovary.

*Conflict of interests:* The authors declare that there is no conflict of interests.

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