

Xanthogranulomatous Salpingo-Oophoritis Mimicking an Ovarian Malignancy – A Series of 3 Cases and Review of Literature

Abstract

Background: Xanthogranulomatous salpingo-oophoritis is an uncommon form of chronic inflammation in the genitourinary tract. Its symptoms and radiological findings mimic ovarian malignancy or severe pelvic inflammatory disease.

Case (s): We describe three cases of xanthogranulomatous salpingo-oophoritis diagnosed in our hospital and review literature on clinical features and diagnostic pitfalls of this condition. Our first patient is a 45-year-old lady who presented with abdominal pain and fever. The second patient is a 51-year-old lady who presented similarly. The last patient is a 35-year-old lady who chronic pelvic inflammatory disease. From literature, risk factors include endometriosis, leiomyoma, pelvic inflammatory disease, intrauterine copper device (IUCD) in situ and previous surgery. Most patients had abdominal pain, fever and adnexal mass on examination. Most women were treated based on the working diagnosis of malignancy or severe pelvic inflammatory disease.

Conclusion: Xanthogranulomatous salpingo-oophoritis is a rare condition that is often mistaken for ovarian malignancy clinically and radiologically. Oophorectomy is the recommended treatment but most women are “over treated” with staging laparotomies and hysterectomies that render them infertile. The presence of fever, abdominal pain, identifiable risk factors, MRI characteristics and the use of intra-operative frozen-section may aid in the diagnosis.

Keywords: Xanthogranulomatous salpingo-oophoritis; Xanthogranulomatous inflammation; Xanthogranulomatous oophoritis; Xanthogranulomatous salpingitis; Pelvic inflammatory disease, Ovarian malignancy; Adnexal mass; Ovarian mass

Case Report

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Abbreviations: IUCD: Intrauterine Copper Device; PID: Pelvic Inflammatory Disease

Introduction

Xanthogranulomatous salpingo-oophoritis is an uncommon form of chronic inflammation in the genitourinary tract, described more commonly in the kidneys and gall bladder with rare involvement of the ovary and fallopian tubes. It is a histopathological diagnosis characterized by the presence of foamy histiocytes, chronic inflammatory cells with the presence of lipid-like substances, which can lead to destruction of the organ. In this case series we describe three cases of xanthogranulomatous inflammation in the female genital tract. We also explore the clinical features and diagnostic pitfalls, and emphasize the need for awareness of this condition to avoid patient morbidity and extensive surgery.

Case Presentation

Case 1

Xanthogranulomatous salpingo-oophoritis

Mdm J, a 45-year-old nulliparous woman presented with abdominal pain and subsequently developed fever, chills and rigors. She had a past history of gallstones, appendicectomy and diabetes. On ultrasound pelvis, there was a large multiloculated complex cystic mass with multiple small fibroids. A computed-tomography (CT) scan showed a complex multiloculated pelvic mass with pelvic lymphadenopathy. She had raised total white cell count of 24.76 as well as an elevated C - reactive protein (264.7). The CA-125 was slightly elevated at 41u/mL. 3Blood and urine culture was negative. The patient was treated as for sepsis while investigating the pelvic mass simultaneously. In view of the complex solid cystic nature of the mass, pelvic lymphadenopathy and elevated CA 125 malignancy was suspected. She underwent laparotomy with total hysterectomy, bilateral salpingo-oophorectomy and intra-operative frozen section of the pelvic mass.

Intra-operatively, bilateral tubo-ovarian abscesses were noted. Frozen section findings were that of inflammatory changes with no malignancy. On gross examination, the right tube and ovary appeared as an irregular haemorrhagic mass with its cut section demonstrating dilated tortuous fallopian tube containing

purulent blood stained fluid. There was a leiomyoma adjacent to the tortuous fallopian tube. The left ovary contained a cystic ovarian mass measuring 7.0 x 7.0 x 2.5 cm. On microscopic examination, both tubes showed salpingitis with transmural inflammatory changes. The inflammatory infiltrate extended into the ovaries, and consisted of sheets of foamy histiocytes admixed with numerous plasma cells, lymphoid aggregates, and a few neutrophils. Her post-operative recovery was uneventful.

Case 2

Xanthogranulomatous oophoritis

Mdm M, a 36-year old nulliparous woman presented with menorrhagia. An ultrasound was done which incidentally showed a left complex ovarian cyst measuring 3.2 x 2.7 x 1.6cm as well as a predominantly solid lesion measuring 3.8cm in the left ovary. CA-125 was normal in this patient. Malignancy was a diagnostic consideration in view of the complex nature of the ovarian cyst. The patient was counseled for diagnostic laparoscopy and cystectomy and the possibility of unilateral salpingo-oophorectomy, lymph node dissection and omentectomy.

She underwent diagnostic laparoscopy with left salpingo-oophorectomy. Intra-operatively, the findings were consistent with that of a tubo-ovarian abscess and the uterus and contralateral adnexa was preserved. Histological specimen showed oophoritis of the left ovary with no evidence of malignancy. The patient was treated with intravenous and oral antibiotics. No pathogens were isolated from operative cultures. On histopathological examination of the left ovary and fallopian tube, there was an endometriotic cyst with chronic xanthogranulomatous inflammation and granulation, consistent with resolving abscess. Post-operative recovery was uneventful.

Case 3

Xanthogranulomatous salpingitis

Mdm W, a 48-year-old lady with two previous Caesarean deliveries, was on conservative management of chronic pelvic-inflammatory disease. She had occasional lower abdominal discomfort and dyspareunia. On routine follow-up ultrasonography, there was an interval increase in the size of the left adnexal mass with a complex solid-cystic appearance. CT scan confirmed a solid-cystic complex adnexal lesion with no enlarged lymph nodes. The total white cell count was not raised, but CA-125 was 87.3. She was offered a total hysterectomy with bilateral salpingo-oophorectomy and the possibility of staging operation as malignancy was a consideration. Intra-operatively, a left tubo-ovarian complex with hydrosalpinx was noted. She underwent total hysterectomy and bilateral salpingo-oophorectomy.

Histo-pathological examination revealed chronic xanthogranulomatous salpingitis of left fallopian tube and a haemorrhagic corpus luteum in the left ovary. The right ovary and fallopian tube was unremarkable. She received intravenous antibiotics prior to the operation, which was continued afterwards. No pathogens were isolated from operative cultures but *E. coli* was isolated from blood cultures done prior to operation. Post-operative recovery was uneventful.

Discussion

In terms of clinical and intra-operative findings, this chronic inflammatory condition can mimic serious gynaecological conditions such as ovarian malignancies. It is most commonly found in kidneys and the gallbladder, with rare reports of involvement of female genital tract.

The exact mechanism of this form of inflammation in the female genitourinary tract is unclear. Risk factors described in literature include endometriosis [1], leiomyoma [2], inadequately treated PID [3], IUCD in situ [4], abnormal lipid metabolism, immunosuppression and previous abdominal surgery. Other risk factors include diabetes mellitus type 2, hyperlipidaemia and untreated urinary infections [5]. Various organisms have been implicated but *E. coli* is the most commonly isolated pathogen. A summary of existing literature with details on the proposed risk factors, clinical findings and management has been made in Table 1.

Most patients presented with abdominal pain and fever. Tenderness and presence of adnexal mass was noted on examination in most patients. Ultrasound and CT/MRI findings suggested the presence of complex cystic adnexal masses. Most women were treated based on the working diagnosis of either an ovarian malignancy or severe pelvic inflammatory disease requiring surgical intervention.

All three women described in our case series had different symptoms but imaging findings were similar. They also had similar risk factors such as previous surgery, pelvic inflammatory disease, endometriosis and leiomyoma, as described in other case reports. There are diagnostic pitfalls on many levels when it comes to recognizing xanthogranulomatous salpingo-oophoritis. There is a lack of knowledge of the condition due to its rarity. Radiological findings are usually indistinguishable from that of malignancy. Furthermore, as intra-operative findings usually resemble that in ovarian malignancy, the tendency is for extensive surgery instead. While the recommended treatment for this condition is oophorectomy [6], the lack of diagnostic certainty or awareness has led to surgeries such as total hysterectomy with bilateral salpingo-oophorectomy being performed.

We propose the following considerations in recognizing this condition in clinical practice. In the presence of a suspicious ultrasound scan, the presence of risk factors and clinical and laboratory findings such as raised inflammatory markers may suggest an infective or inflammatory aetiology. MRI findings have also been described in the kidney, gallbladder [7] and two cases of xanthogranulomatous oophoritis. Seung et al. [8] describes features of xanthogranulomatous inflammation as "multiple non enhancing intramural nodules in a thickened wall on T2-weighted images" [8]. The use of intra-operative frozen section is also helpful before proceeding with hysterectomy and contralateral salpingo-oophorectomy.

Xanthogranulomatous salpingo-oophoritis is a histological diagnosis. Findings include foamy histiocytes and chronic inflammatory cells such as plasma cells, lymphocytes and multinucleated giant cells, with the presence of lipid-like substances. In our patients, there was also distortion of the

fallopian tubes and destruction of tubal lining. In addition, immunohistochemical stains are helpful in establishing the diagnosis, such as CD68 in foam cells, CD3 (T lymphocyte marker), CD20 (B lymphocytes marker) [6], Fontana-Masson stain (lipofuscin in foamy macrophages). The absence of Michaelis-Gutmann bodies excludes diagnosis of makaloplakia,

another chronic inflammatory entity of the genitourinary tract. The presence of inflammatory cells can be made prominent by performing leukocyte common antigen (LCA) immunostaining. This can also help differentiate between xanthelasma of the fallopian tube and xanthogranulomatous salpingo-oophoritis [1,9].

Table 1: Summary of cases from available literature.

Author	Clinical Entity	Age	Risk Factors	Presenting Complaint	Examination Findings	Operation Done	Organism Isolated
Abeysundara PK	Salpingo-oophoritis Appendicitis	34	Endometriosis Leiomyoma Primary subfertility Previous surgery	Intestinal obstruction	Distended abdomen	Laparoscopic THBSO Appendectomy	<i>Pseudomonas</i>
Punia RS	Salpingo-oophoritis	42	PID	Menorrhagia	Bilateral adnexal masses	Subtotal hysterectomy Bilateral SO	<i>Staph aureus</i>
Y Gray	Salpingo-oophoritis	47	PID IUCD in situ Endometriosis	Abdominal pain Loose stools	Fullness in left abdomen	TAH Bilateral SO	<i>E. coli</i>
Singh R	Salpingitis Appendicitis	37	-	Abdominal pain Vaginal discharge	Tender right adnexal mass Cervical excitation	-	<i>Enterobius vermicularis</i>
Howey JM	Salpingitis	50	Leiomyoma Previous surgery (appendectomy) Endometriosis	Abnormal uterine bleeding	Enlarged uterus	TAH Left SO	-
Kalloli M	Oophoritis	45	PID IUCD	Abdominal pain	Left adnexal mass	TAH Bilateral SO Omentectomy	-
Yener N	Salpingitis	41	Secondary infertility	Abdominal pain	Bilateral adnexal tenderness	Bilateral SO	-
Furuya M	Salpingitis	19	Chlamydia cervicitis	Abdominal pain Fever Vaginal discharge	Left adnexal mass	Left SO	<i>N. gonorrhoea (vaginal discharge)</i>
Furuya M	Salpingitis	49	Leiomyoma Endometriosis	Abdominal pain Fever	-	TAH Bilateral SO	<i>Bacteroides fragilis (vaginal discharge)</i>
Idrees M	Salpingitis	41	Endometriosis Endometritis IUCD Previous chemotherapy	Abdominal discomfort	Bilateral tender and enlarged adnexa masses	Bilateral SO	-
Altanis S	Oophoritis Endometritis	84	Diverticulitis Previous chemotherapy	Post menopausal bleed	-	TAH Bilateral SO (could not exclude endometrial mass)	-
Seung Eun Jung	Oophoritis	48	-	Abdominal pain Fever	-	TAH Bilateral SO	-
Singh UR	Oophoritis	25	Endometriosis <i>S. typhi</i> infection	Abdominal pain Abdominal mass	Suprapubic mass Enlarged uterus	Partial Oophorectomy	<i>S. typhi</i>
Shukla S	Oophoritis	42	Endometriosis Fibroids Primary infertility	Abdominal pain	Midline abdominal mass	TAH Bilateral SO Omentectomy	<i>E. coli</i>
Naik M	Oophoritis	25	-	Abdominal pain Fever	-	-	-
Zhang XS	Salpingitis	29	-	Abdominal discomfort Fever Vaginal discharge	Left adnexal mass	Left SO	-
Zhang XS	Oophoritis	32	-	Abdominal pain Fever	Left adnexal mass	TAH Bilateral SO	-

TAH: Total Abdominal Hysterectomy; TLH: Total Laparoscopic Hysterectomy; SO: Salpingo-Oophorectomy

The presence of endometriosis and leiomyoma should be commented on, as this may provide aetiological insight on the pathogenesis. We also recommend performing stains for gram stain, fungi and acid-fast bacilli as well as sending intra-operative cultures, blood or urine cultures such that appropriate antibiotics can be administered if pathogens are isolated.

Conclusion

Xanthogranulomatous salpingo-oophoritis is a rare chronic inflammatory condition of the female reproductive tract diagnosed on histo-pathological findings. With awareness of the condition and appropriate history, clinical examination, investigations and eventual histopathological correlation, the certainty of the diagnosis can be increased and as a result, the appropriate treatment and surgery can be administered.

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