

DOI: <http://dx.doi.org/10.18203/2320-1770.ijrcog20204334>

Case Report

Xanthogranulomatous salpingo-oophoritis: a case report and review of literature

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Received: 11 August 2020

Accepted: 15 September 2020

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ABSTRACT

Xanthogranulomatous inflammation of ovary is a rare disease that is characterized by presence of large number of lipid laden macrophages with an admixture of neutrophils, lymphocytes, plasma cells and multinucleated giant cells. It is misdiagnosed as ovarian tumour that leads to extensive surgery including hysterectomy. In this report we describe a case of Xanthogranulomatous salpingo-oophoritis along with review of literature.

Keywords: Oophoritis, Salpingitis, Xanthogranulomatous inflammation.

INTRODUCTION

Xanthogranulomatous inflammation is a chronic inflammation that destroys the affected organs. Inflammation is commonly described in kidney, gallbladder, stomach, urinary bladder, bones, testis and epididymis.^{1,2} Xanthogranulomatous inflammation of the female genital tract is rare and mainly limited to ovary, fallopian tube and endometrium.^{3,4,5} Clinically patients present with fever, pain and fullness in lower abdomen. In this report we describe a case of unilateral xanthogranulomatous salpingo-oophoritis, which presented as tubo-ovarian mass.

CASE REPORT

A 25 year old female (P2L2) presented with fullness of lower abdomen and abdominal pain for 10 days duration. Her history revealed that she is having in situ intrauterine contraceptive device (IUCD). The ultrasonography of abdomen and pelvis showed a well defined hypoechoic solid mass in left adnexa measuring 7x5 cm, which was on the abdominopelvic MRI showed hyperintense irregular, solid tuboovarian mass. The left ovary was indiscernible from the mass, while right ovary and fallopian tube was normal and uterus showed in situ

intrauterine contraceptive device in lower segment. Hematological investigation showed haemoglobin of 8.0 gm%, total leucocyte count of 12,000/mm³, ESR 26 mm/hr, liver function tests and renal function tests were within normal limits. On exploratory laparotomy the left tuboovarian mass was found to be adherent to urinary bladder, anterior wall of uterus and bowel, resected tuboovarian mass was received for histopathological evaluation. The specimen of left tubo-ovarian mass measured 6x4.8x2.5 cm and weighed 25 gm. The external surface of it was irregular and showed attached fallopian tube. The cut surface of tubo-ovarian mass was solid and was yellowish in colour and showed areas of necrosis (Figure 1A). Microscopic examination revealed diffuse infiltration of ovarian tissue by sheets of foamy histiocytes with abundant lipid laden vacuoles in their cytoplasm and inflammatory cells comprising of lymphocytes, plasma cells and polymorphs (Figure 1B). Histology of fallopian tube also showed presence of xanthogranulomatous inflammation. On Immunohistochemistry foamy macrophages showed diffuse cytoplasmic positivity for CD 68 (Figure 1C), confirming a diagnosis of xanthogranulomatous salpingo-oophoritis. Periodic acid Schiff stain, gram stain and acid-fast stains were negative for microorganism. Bacterial culture of tubo-ovarian mass was not done as specimen was already fixed in formalin.

Table 1: Year of publication, country and operation performed in xanthogranulomatous salpingo-oophoritis.

Ref No.	Country & year of publication	Age	Operation performed	NOC	Ref No.	Country & year of publication	Age	Operation performed	NOC
3	USA, 1976	26	THBSO	1	25	India, 2014	32	Left SO	1
4	USA, 1977	24	Right SO	1			30	THBSO	
6	USA, 1984	26	THBSO	1	26	India, 2014	31	THBSO, colectomy	2
7	India, 1995	25	Ooph.	1	27	India, 2015	2	Right SO	1
8	Tunisia, 1999	34	THBSO	1	28	USA, 2015	45	Left SO	1
9	India, 1999	25	THBSO	1			36	Left SO	
1	USA, 2001	47	THBSO	1	29	Singapore, 2016	45	THBSO	3
10	Korea, 2001	48	THBSO	1			48	THBSO	
11	India, 2003	42	Left SO	1	30	India, 2016	45	THBSO	1
12	India, 2007	18	Right SO	1	31	India, 2016	44	THBSO	1
13	Europe, 2007	84	THBSO	1	32	Pakistan, 2017	47	THBSO	1
14	India, 2009	25	THBSO	1	33	India, 2017	14	Right SO	1
15	India, 2010	42	THBSO	1	34	India, 2017	17	Pelvic mass with B/L SO	2
16	India, 2011	28	Bilateral SO	1			38	Left SO	
17	China, 2011	32	THBSO	1	35	India, 2017	21	Left SO	2
18	India, 2012	35	THBSO	1			43	THBSO	
2	India, 2012	45	THBSO	1	36	India, 2017	20	Right Ooph.	1
19	Sri Lanka, 2012	34	THBSO	1	37	India, 2018	24	Right SO	1
20	India, 2013	61	THBSO	1			17	Partial oophorectomy	
21	India, 2013	61	THBSO	1	38	India, 2018	24	Ovarian cystectomy	3
22	India, 2013	28	Ovarian cystectomy	1			25	Partial ooph. & appendectomy	
23	India, 2013	18	Right SO	1	39	India, 2019	60	THBSO with omental	1
24	India, 2014	40	THBSO	1	40	India, 2019	35	THBSO	1

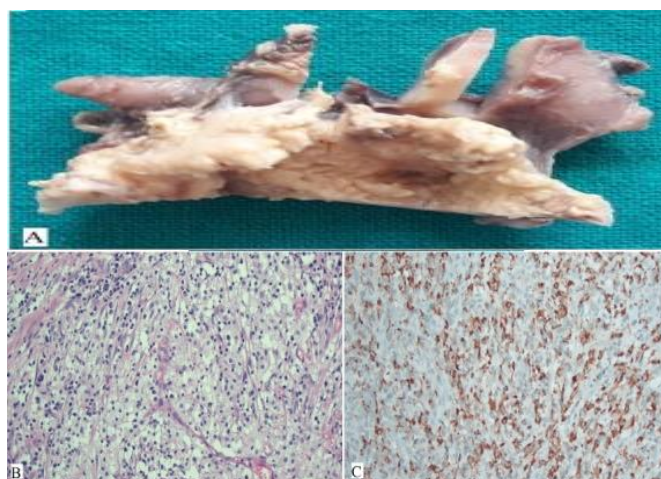


Figure 1: (A) Cut section of tubo-ovarian mass shows yellowish necrotic appearance and attached fallopian tube, (B) histology shows diffuse proliferation of foamy histiocytes and inflammatory cells. (H&E 400), (C) shows CD 68 cytoplasmic positivity in foamy macrophages (IHC).

DISCUSSION

The first case of xanthogranulomatous salpingo-oophoritis was described in 1976, since then only 46 cases of this entity were published in literature.^{1-4,6-40} The age, name of country, year of publication and treatment given in these 46 cases is shown in (Table 1). In the 46 published reports 12 cases are from developed countries; which comprised of 5 cases from USA, 3 from Singapore and one case each from Korea, Europe, China, and Tunisia.^{1,3,4,6,8,10,13,17,28,29} Out of 34 cases reported from developing countries, 32 are described in Indian literature and one case each is documented from Pakistan and Sri Lanka.^{2,7,9,11,12,14-16,18-20-27,30,31-40} The age range of these cases varied from 2-84 years and the most frequent sign and symptoms were lower abdominal or suprapubic pain, fever, menorrhagia, or vaginal bleeding and adnexal tenderness and a pelvic mass.^{1-4,6-40} The risk factors for xanthogranulomatous salpingo-oophoritis is pelvic inflammatory disease, endometriosis, IUCD, antibiotic treatment and cervical stenosis.^{1-4,6,7,10,11,13-15,18,29,32,38} The gross examination of the affected ovary may show replacement by a well circumscribed, solid, yellowish, lobulated mass along with cystic change.^{1,2,16-18}

Microscopically, it is characterized by a massive infiltration by lipid laden histiocytes known as Xanthoma cells and mixed with inflammatory infiltrates of the affected tissues.^{1,2,16,18} Immunohistochemical stains CD68, CD3 and CD20 are helpful in establishing the diagnosis. Although pathogenesis of vacuolated macrophages is not clear, many authors believe it to be a secondary degenerative change representing the accumulation of different substances ingested by histiocytes. The proposed causes of vacuolated macrophages are abnormality in lipid metabolism and ineffective clearance of bacteria by phagocytes.^{1,2} Bleeding and obstruction may predispose to infection, producing tissue necrosis followed by the release of cholesterol and other lipids that is phagocytized by macrophages.¹ In several reports' bacteria including *Actinomyces*, *Staphylococcus aureus*, *Streptococcus faecalis*, *Escherichia coli* were grown on culture of tubo-ovarian abscess. It is thought that the colonization of the IUCD in the endometrial cavity is followed by the shedding of bacteria through the lumen of the fallopian tube.² Alternatively ovarian surface is exposed to bacteria at the time of ovulation, the corpus luteum becomes infected, producing an ovarian abscess. Oophorectomy is the most recommended treatment for xanthogranulomatous oophoritis, however intra-operatively these lesions usually mimic ovarian malignancy and hence call for the tendency towards extensive surgery, including total hysterectomy. In 26 out of 46 reported cases hysterectomy with bilateral salpingo-oophorectomy was performed.^{1-3,6,8-10,13-15,17-21,24,26,29-32,35,39,40}

The differential diagnosis of xanthogranulomatous salpingitis and oophoritis includes both non-neoplastic and neoplastic conditions. The non-neoplastic conditions are mainly infections such as tuberculosis and fungal infections and malakoplakia that need to be ruled out by cultural studies and special stains such as periodic acid Schiff stain, gram stain and acid-fast stains.

CONCLUSION

High prevalence of xanthogranulomatous salpingo-oophoritis is seen in Indian women. Frozen section study of tubo-ovarian mass may be useful in establishing accurate diagnosis, so that unnecessary hysterectomy can be avoided.

ACKNOWLEDGMENTS

Dr. Sonali Lanjewar, Department of Pathology, Methodist University Hospital, University of Tennessee, Health Science Center, Memphis, USA.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Patel KA, Chothani KP, Patel B, Lanjewar DN. Xanthogranulomatous salpingo-oophoritis: a case report and review of literature. *Int J Reprod Contracept Obstet Gynecol* 2020;9:4316-9.