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Case Report

Pseudoxanthomatous salpingitis masquerading as a tubo-ovarian malignancy in a patient with endometriosis: a rare case report

Nancy Thind[1]*, Moneet Walia[1], Preeti J. John[2], Priya Jindal[1], Devika Khatri[1]

[1]Department of Obstetrics and Gynaecology, Adesh Medical College and Hospital, Kurukshetra, Haryana, India
[2]Department of Pathology, Adesh Medical College and Hospital, Kurukshetra, Haryana, India

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***Correspondence:**
Dr. Nancy Thind,
E-mail: nancythind@yahoo.com

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ABSTRACT

Pseudoxanthomatous salpingitis (PXS), also known as pigmentosis tubae, is an extremely rare benign inflammatory condition of the fallopian tube characterised by the presence of lipofuscin- and hemosiderin-laden macrophages within the lamina propria. Owing to its gross appearance and radiological findings, it may closely mimic tubo-ovarian malignancy, leading to diagnostic dilemma. We report a case of a 40-year-old nulligravida with a history of endometriosis who presented with acute abdomen and was radiologically diagnosed as a complex ovarian mass with markedly raised CA-125 levels. Intraoperatively, a tubo-ovarian mass with chocolate-coloured fluid was noted. Histopathological examination revealed features consistent with pseudoxanthomatous salpingitis associated with ovarian endometriosis. This case highlights the importance of considering PXS in the differential diagnosis of tubo-ovarian masses, especially in patients with endometriosis, to avoid overtreatment. Early recognition of this rare entity can help avoid overtreatment, especially in fertility-desiring patients.

Keywords: Pseudoxanthomatous salpingitis, Hemosiderin, Endometriosis, CA-125, Tubo-ovarian Mass

INTRODUCTION

Pseudoxanthomatous salpingitis also known as pigmentosis tubae, is a rare pathological entity involving the fallopian tube characterized by accumulation OG pigment laden macrophages within the lamina propria.[1,2] It is most commonly associated with endometriosis, chronic pelvic inflammatory disease, or prior pelvic surgery.[2,3] Fewer than 30 cases have been reported in the literature.[2] The condition is characterised histologically by sheets of pigmented macrophages within the lamina propria of the tubal mucosa, leading to expansion and distortion of the plicae.[3,5] Clinically and radiologically, PXS may mimic malignant tubo-ovarian pathology, particularly when CA125 elevated making histopathological examination essential for definitive diagnosis.[4-6] The tubes in this condition is occasionally found to be enlarged and oedematous with mucosa having a dark brown polyploid gross appearance.[2,3] A history of endometriosis, infertility, tubal ligation, endometrial cancer and prior radiation therapy for cervical carcinoma is associated with PXS and can complicate its clinical presentation and management.[1-6] We hereby report a case of Psedoxanthomatous salpingities with Complex ovarian cyst.

CASE REPORT

A 40-year-old nulligravida presented with severe pain in the right lower abdomen for three days. She reported similar episodic pain for the past 10 years and was a known case of endometriosis. Ten years earlier, she had undergone laparoscopic cystectomy for a right ovarian endometriotic cyst. There was no history of weight loss,

loss of appetite, bowel or bladder disturbances. Her menstrual cycles were regular. There was no significant medical or family history. On examination, her vital signs were stable. Abdominal examination revealed tenderness in the right iliac fossa with rebound tenderness. There was no guarding or rigidity. Pelvic examination was unremarkable.

Investigations

Laboratory investigations showed haemoglobin of 12.2 g/dl, total leucocyte count of 6,500/mm³ and platelet count of 274,000/mm³. Liver and renal function tests were within normal limits. CA-125 was markedly elevated at 364 U/ml, while carcinoembryonic antigen (CEA) was within normal range. Pelvic ultrasonography revealed a bilocular heterogeneous cyst measuring 6×6 cm in the right ovary along with fluid-filled dilatation of the left fallopian tube. MRI of the abdomen and pelvis confirmed a right-sided complex ovarian cyst measuring 6.5×6 cm, raising suspicion of a tubo-ovarian mass. Detailed discussion about the management was done with the patient and Informed consent was taken. Patient opted for a fertility sparing surgery and was planned for ovarian cystectomy and proceed. Differential Diagnosis Ovarian malignancy, tubo-ovarian abscess, endometriotic cyst, chronic salpingitis.

Treatment

After counselling, the patient opted for fertility-sparing surgery. She underwent exploratory laparotomy with right ovarian cystectomy.

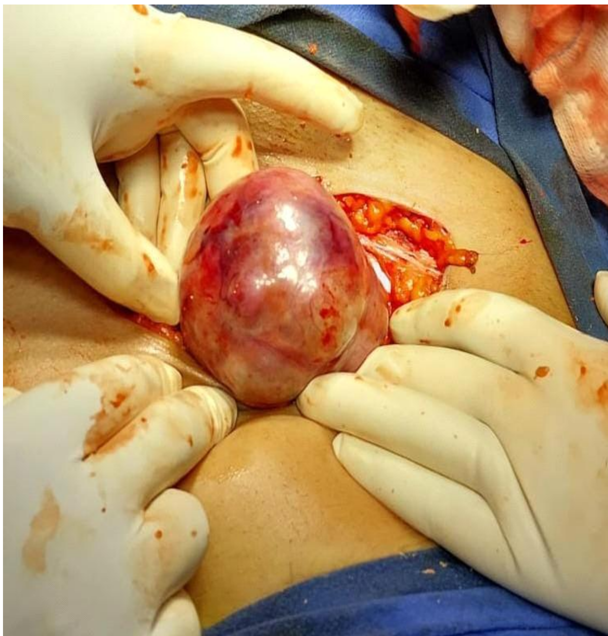

Figure 1: Pre-operative: the right fallopian tube was found to be coiled over the ovarian mass.

Intraoperatively, dense adhesions were noted between the uterus and the complex ovarian mass of size 6x6 cm which was separated by sharp dissection followed by excision of right sided tubo ovarian mass. Figure 1 showing the right fallopian tube was found to be coiled over the ovarian mass. On incision, dark chocolate-coloured fluid was drained. The left adnexa were adherent to the caecum and adhesiolysis was performed. The uterus appeared grossly normal but had restricted mobility. The excised specimen, including the right ovary and fallopian tube, was sent for histopathological examination.

Outcome and follow-up

The postoperative period was uneventful. The patient was started on oral feeds on the same day and discharged on postoperative day four. She was initiated on a gonadotropin-releasing hormone analogue postoperatively. At follow-up after one week, she was asymptomatic and sutures were removed.

Histopathological findings

Microscopic examination revealed expanded and distended tubal plicae with sheets of pigmented histiocytes possessing eosinophilic cytoplasm within the lamina propria, consistent with pseudoxanthomatous salpingitis. The ovarian tissue showed features of endometriosis with many hemosiderin laden macrophages within the wall as depicted in Figure 2.

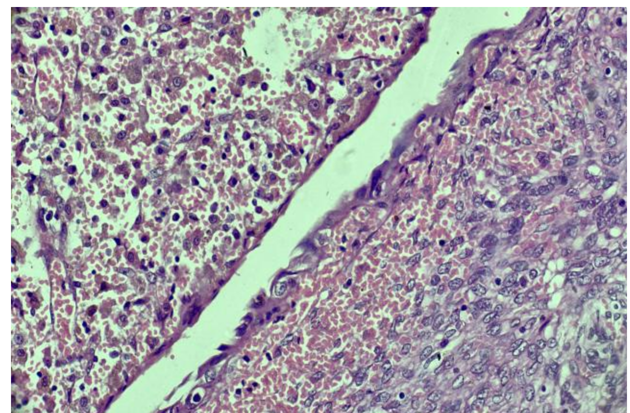

Figure 2: Endometriotic ovarian cyst with attenuated lining epithelium and many hemosiderin laden macrophages within the wall (H and E 400x).

Figure 3 shows pseudoxanthomatous salpingitis-distended plicae of fallopian tube with histiocytes having eosinophilic cytoplasm (H and E 100x). Figure 4 shows histiocytes along with chronic inflammation and pigment laden macrophages within the fallopian tube (H and E 400x).

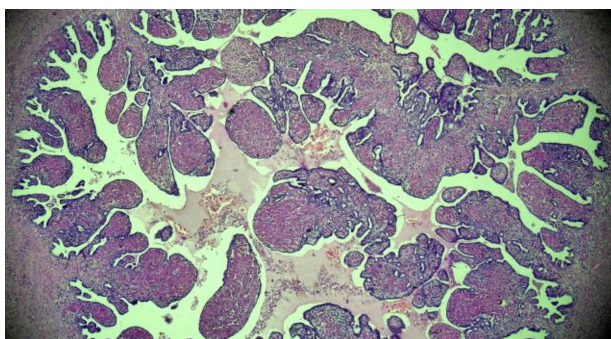

Figure 3: Pseudoxanthomatous salpingitis-distended plicae of fallopian tube with histiocytes having eosinophilic cytoplasm (H and E 100x).

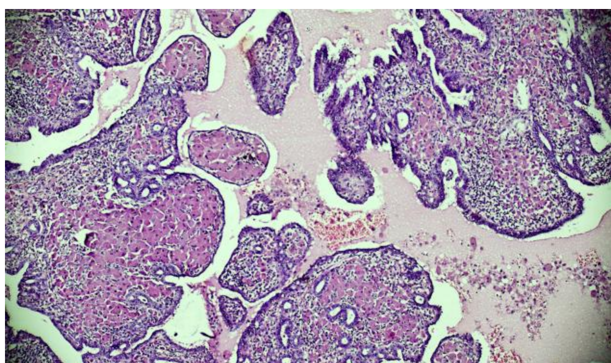

Figure 4: Histiocytes along with chronic inflammation and pigment laden macrophages within the fallopian tube (H and E 400x).

DISCUSSION

Pseudoxanthomatous salpingitis (PXS) is an extremely rare benign inflammatory condition of the fallopian tube, first described several decades ago and reported in fewer than 30 cases in the literature.[1] It is characterised histologically by the accumulation of hemosiderin- and lipofuscin-laden macrophages within the lamina propria of the tubal mucosa, resulting in expansion and distortion of the plicae.[1-3]

Most reported cases demonstrate a strong association with ovarian endometriosis.[1-4] The proposed pathogenesis involves repeated cyclical haemorrhage from adjacent endometriotic lesions, leading to breakdown of blood products and subsequent phagocytosis by macrophages within the tubal mucosa.[2-4] Chronic inflammation and oxidative stress are believed to contribute to the accumulation of pigment within histiocytes. This explains why PXS is frequently identified in patients with long-standing endometriosis, as seen in our case. Clinically and radiologically, PXS poses a diagnostic challenge. Several published reports describe its presentation as a tubo-ovarian mass, often with elevated CA-125 levels, thereby closely mimicking ovarian malignancy.[1-4] Elevated CA-125 in such cases is attributed to peritoneal irritation and underlying endometriosis rather than neoplastic pathology. Imaging findings are non-specific and may show complex adnexal masses or hydrosalpinx, making preoperative diagnosis difficult. Histopathological examination remains the gold standard for diagnosis. The key distinguishing feature is the presence of sheets of pigmented histiocytes in the lamina propria without cytological atypia or stromal invasion, thereby differentiating it from malignant or premalignant lesions.[2-5] Awareness of this entity is essential for both pathologists and clinicians to avoid misinterpretation and overtreatment. From a surgical perspective, most cases are diagnosed incidentally following excision performed for suspected malignancy or complicated endometriosis.[1] Fertility-preserving surgery is appropriate when malignancy is excluded, particularly in reproductive-age women. Our case further supports the literature in highlighting that PXS should be considered in the differential diagnosis of tubo-ovarian masses in patients with known endometriosis. In summary, pseudoxanthomatous salpingitis is a rare but important benign condition that may clinically and radiologically simulate malignancy. Recognition of its association with endometriosis and reliance on histopathological confirmation are crucial to ensure appropriate management

CONCLUSION

Pseudoxanthomatous salpingitis is a rare benign condition of the fallopian tube that can closely mimic tubo-ovarian malignancy, particularly in patients with underlying endometriosis and elevated CA-125 levels. Histopathological examination is essential for definitive diagnosis. Awareness of this entity can help avoid overtreatment, especially in fertility-desiring patients.

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