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Case Report

A benign illusion: elevated CA 19-9 and bilateral xanthogranulomatous salpingitis hiding behind pelvic endometriosis

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ABSTRACT

Xanthogranulomatous salpingitis (XGS) is a rare, benign, chronic inflammatory tubal condition that frequently mimics advanced malignancy, and its concurrent presentation with endometriosis is exceptionally rare. We present the case of an asymptomatic 44-year-old female who was found to have a complex tubo-ovarian mass and an elevated CA 19-9 during a routine solid organ donor evaluation. Suspecting malignancy, diagnostic laparoscopy was performed. Crucially, intraoperative frozen section analysis revealed benign pathology, prompting a fertility-sparing surgical approach involving left mass excision, right salpingectomy, and endometriosis resection. Final histopathology unexpectedly confirmed bilateral XGS with concurrent pelvic endometriosis. This case highlights that XGS can develop entirely silently alongside endometriosis, likely driven by chronic hemorrhage rather than bacterial infection. Intraoperative pathological evaluation is imperative to prevent the radical oncological overtreatment of complex, adherent adnexal masses.

Keywords: Xanthogranulomatous salpingitis, Pelvic endometriosis, Asymptomatic adnexal mass, Tubo-ovarian mass, Intraoperative frozen section

INTRODUCTION

Xanthogranulomatous salpingitis (XGS) is an exceptionally rare, destructive form of chronic granulomatous inflammation affecting the female reproductive tract.¹ Histologically, it is defined by the complete effacement of normal tubal architecture, which is replaced by a dense inflammatory infiltrate predominantly composed of lipid-laden foamy macrophages (xanthoma cells), admixed with plasma cells, lymphocytes, and neutrophils.¹ While xanthogranulomatous inflammation is well-documented in the gallbladder and kidneys, its occurrence in the fallopian tubes and ovaries remains a clinical rarity.^{2,3}

In clinical practice, XGS is notorious for masquerading as an advanced pelvic malignancy.² Patients classically present with severe lower abdominal pain, fever, altered bowel habits, and a complex tubo-ovarian mass on radiological imaging.^{1,2} This alarming presentation frequently prompts extensive, irreversible oncological surgeries before a benign histopathological diagnosis is finally established.^{2,3} The precise etiology of XGS remains a subject of debate, though it is most often considered a late sequela of inadequately treated pelvic inflammatory disease (PID).⁴ However, other localized sources of persistent inflammation and tissue necrosis-such as uterine leiomyomas and pelvic endometriosis-have been implicated in its pathogenesis. It is theorized that the recurrent, cyclic hemorrhage characteristic of

endometriosis results in a continuous release of cholesterol and lipids from necrotic erythrocytes, which are subsequently phagocytosed by macrophages, thereby initiating the xanthogranulomatous cascade.^{3,5}

Despite this theoretical pathophysiological link, documented cases demonstrating the co-occurrence of XGS and endometriosis remain remarkably sparse.^{2,5} Furthermore, nearly all reported cases of XGS are highly symptomatic.^{2,4} Herein, we present a unique case of bilateral XGS arising alongside pelvic endometriosis in a completely asymptomatic female. Discovered incidentally and complicated by an elevated tumor marker, this case highlights the critical importance of retaining benign inflammatory etiologies in the differential diagnosis of complex adnexal masses.

CASE REPORT

A 44-year-old woman presented to our institution for routine medical clearance prior to a prospective living solid organ donation. She was entirely asymptomatic. A comprehensive clinical history revealed no episodes of pelvic pain, dysmenorrhea, abnormal uterine bleeding, fever, or unexplained weight loss. Her general physical and systemic examinations were unremarkable.

During her preoperative donor evaluation, routine laboratory results were largely within normal limits, aside from mild anemia (hemoglobin 10.4 g/dL). However, tumor marker analysis revealed a notably elevated Carbohydrate Antigen 19-9 (CA 19-9) level of 96.25 U/mL, which is approximately two-and-a-half times the upper limit of normal.

A routine screening transvaginal ultrasound subsequently identified an unexpected, complex left tubo-ovarian mass measuring roughly 6 x 5 cm. (Insert Figure 1 here: Ultrasound imaging of the adnexal mass - Placeholder). The pairing of a complex adnexal mass with a significantly elevated CA 19-9 level naturally raised our clinical suspicion for an underlying neoplastic process, such as a mucinous ovarian tumor. To definitively rule out malignancy and safely clear her for organ donation, surgical exploration was mandated.

The frozen section returned a reassuring diagnosis of a benign, severe inflammatory process, with no evidence of cellular atypia or malignancy. Guided by this intraoperative pathological assessment, the team proceeded with a measured, organ-sparing surgical approach: a laparoscopic excision of the left tubo-ovarian mass, a right salpingectomy, and careful excision of the posterior uterine endometriotic lesions.

The definitive postoperative histopathology report yielded an unexpected diagnosis. Microscopic examination showed extensive tubal infiltration by sheets of lipid-laden foamy macrophages (xanthoma cells), admixed with dense chronic inflammatory cells. Figure 2 illustrates the

microscopic slide showing foamy macrophages – Placeholder.



Figure 1: Gross image of left fallopian tube showing luminal yellowish discoloration,

The patient underwent a diagnostic laparoscopy. Intraoperatively, the surgical team visualized the left-sided tubo-ovarian mass, alongside dense pelvic adhesions and distinct, clear endometriotic deposits scattered across the posterior uterine surface. Recognizing the macroscopic overlap between severe endometriosis, inflammatory masses, and malignancy, the surgical team opted to send a tissue sample of the fallopian tube for immediate intraoperative frozen section analysis.

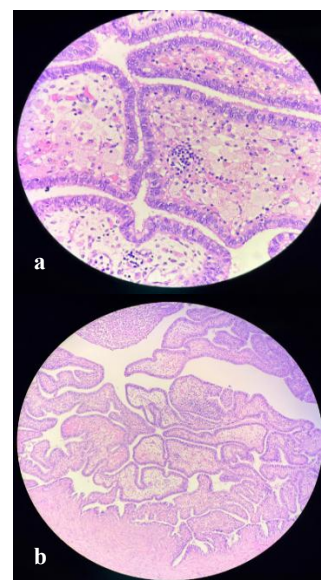


Figure 2: (a) H&E. 400X higher magnification showing distended pliae with foamy histiocytes and inflammatory cells; (b) H&E100X. Section show expanded lamina with presence of foamy histiocytes, congested vessels and lymphocytes.

Surprisingly, these classic features of XGS were present bilaterally in both fallopian tubes. Additionally, the tissue

excised from the left ovary and posterior uterus confirmed the presence of typical endometriotic cysts and stroma. The patient's postoperative recovery was uneventful, and she was discharged without complications

DISCUSSION

This case represents a highly unusual presentation of bilateral xanthogranulomatous salpingitis (XGS) developing concurrently with pelvic endometriosis. By reporting this unique clinical scenario, we aim to highlight several critical learning points for gynecologists, general surgeons, and radiologists facing complex adnexal masses.

XGS can develop silently

While XGS is classically defined by a severe, symptomatic clinical picture mimicking chronic PID or advanced malignancy (featuring pain, fever, and leukocytosis), our findings definitively demonstrate that this intense inflammatory process can develop entirely silently.^{2,4} The incidental discovery of this densely adherent mass during a routine preoperative workup challenges the assumption that severe granulomatous tubal inflammation must be accompanied by systemic or localized symptoms.

Endometriosis as a primary trigger

Although the bilateral XGS developed as a distinct pathological entity separate from the left-sided endometriotic cyst, their spatial relationship and the presence of dense posterior uterine adhesions strongly suggest a shared inflammatory microenvironment. Pelvic endometriosis generates a localized state of chronic inflammation, anatomical distortion, and recurrent tissue hemorrhage.^{3,5} This case supports the hypothesis that the continuous release of blood products (erythrocytes, lipids, and cholesterol) into the pelvic milieu provides the necessary substrate for macrophage recruitment and their subsequent transformation into pathognomonic foamy xanthoma cells, even in the absence of an active bacterial infection.³

The deception of tumor markers

The significant elevation of CA 19-9 (96.25 U/ml) added a profound layer of diagnostic complexity to this case. While CA 19-9 is traditionally associated with gastrointestinal and mucinous ovarian malignancies, it is well-documented that severe endometriosis and profound pelvic inflammation can falsely elevate this marker. Clinicians must learn that an elevated CA 19-9 in the presence of a complex pelvic mass does not definitively equate to malignancy and must be interpreted within the broader clinical context of potential inflammatory or hemorrhagic conditions.

The imperative of intraoperative pathology

Perhaps the most vital takeaway from this case is the absolute necessity of utilizing intraoperative frozen section analysis. Radiologically and macroscopically, a densely adherent tubal mass coexisting with uterine cysts and an elevated tumor marker easily mimics an advanced, invasive pelvic malignancy.² Had our surgical team relied solely on the preoperative suspicion of cancer, this young, asymptomatic patient might have been subjected to an irreversible and unnecessary radical hysterectomy with bilateral salpingo-oophorectomy. The judicious use of frozen sections allowed for a measured, organ-sparing approach that resolved the pathology while preserving the patient's anatomical integrity.

Ultimately, clinicians and surgeons must retain rare, benign inflammatory entities like XGS in the differential diagnosis of complex, adherent adnexal masses, particularly when associated with signs of endometriosis.

CONCLUSIONS

This study significantly advances our understanding of xanthogranulomatous salpingitis by proving that this profound inflammatory process can manifest entirely asymptotically and bilaterally, challenging the traditional diagnostic paradigm that relies on clinical signs of pelvic infection. By establishing a concurrent histopathological link between XGS and pelvic endometriosis, it provides compelling clinical evidence that recurrent, localized hemorrhage-rather than active bacterial infection-can serve as the primary catalyst for xanthogranulomatous transformation. Furthermore, this case highlights a critical clinical pitfall: the deceptive elevation of CA 19-9 in benign inflammatory states. Recognizing these silent, endometriosis-driven presentations and mandating the use of intraoperative frozen section analysis will directly improve patient care by averting irreversible, radical oncological resections in women presenting with complex but ultimately benign adnexal masses.

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