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Case Report Inflammatory pseudotumor of fallopian tube: A case report with review of literature

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ABSTRACT

Inflammatory pseudotumor of fallopian tube is an uncommon benign lesion. We report a case of an inflammatory pseudotumor of left fallopian tube in a 29 year-old woman. She presented with pain in lower abdomen, urinary frequency and dysuria. Contrast enhanced computerized tomography revealed a large necrotic mass in left adnexa abutting superior wall of urinary bladder and lower anterior wall of uterus. Left ovary was seen adherent to the posterior part of the mass. Serum CA-125 was within normal limit. The patient underwent tru-cut biopsy and subsequent excision of left adnexal mass. Histopathology revealed an inflammatory pseudotumor. We reviewed the clinical, imaging and pathological features of the inflammatory pseudotumor of fallopian tube and discussed its differential diagnosis.

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1. Introduction

Inflammatory pseudotumor (IPT) of fallopian tube is a rare benign entity of unknown etiology. IPT is a nonneoplastic proliferation of loosely packed fibroblastic spindle cells accompanied by mixed inflammatory cells in a myxoid background. Inflammatory pseudotumor most commonly occurs in lung and orbit but has been reported to involve almost all organs in the body, with fallopian tube being one of the uncommon sites.¹ The term "pseudotumor" was coined by Umiker and Iverson in 1954, owing to its tendency to simulate malignancy both clinically and radiologically.² Once mistaken for cancer, the patient may be subjected to unnecessary cancer management and its deleterious consequences.

2. Case Report

A 29-year old female presented to us with pain in lower abdomen, urinary frequency and dysuria. She had underwent bilateral laparoscopic tubectomy last two years ago. Contrast enhanced computerized tomography showed a large necrotic mass in left adnexa measuring 9cm x 7cm x 6.5cm abutting superior wall of urinary bladder and lower anterior wall of uterus. Left ovary was seen adherent to the posterior part of the mass. Serum CA-125 was within normal limit. Repeated urine cultures were sterile and urine cytology was found to be negative for malignant cells. Computerized tomography guided tru-cut biopsy was done from the mass (Figure 1 A-D) with concern of a possible tumor during which fifteen ml yellow coloured pus was also retrieved. Microbiological examination of the pus revealed presence of Methicillin resistant Staphylococcus aureus. Tru-cut biopsy showed fascicles of fibroblastic proliferation in sclerotic stroma with interspersed chronic inflammatory cells composed of lymphocytes and plasma cells (Figure 2A). Multinucleated foreign body type giant cells (Figure 2B) were also seen. There was no evidence of malignancy. Patient was administered oral antibiotic therapy for one month, but the lesion did not show any radiological resolution. Subsequently, the patient underwent

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laparotomy with gross total excision of the mass. Intraoperative findings revealed a solid cystic organized mass in left adnexa measuring 8cm x 7cm adherent to dome of urinary bladder and lower anterior wall of uterus. Left fallopian tube was incorporated within the mass and was grossly dilated (Figure 2C). Bilateral ovaries and right fallopian tube were free from the mass. Hundred ml yellow colored pus was also drained from the mass which revealed similar microbiological findings as before. Histopathology of the excised mass was similar to that of tru-cut biopsy and in addition also showed presence of incorporated fallopian tube. On immunohistochemistry, the proliferating fibroblastic cells were positive for Vimentin positive (Figure 2D), Smooth muscle actin (SMA) (Figure 2E) and Negative for Pan-cytokeratin, CD34, Desmin, Myogenin, and Anaplastic lymphoma kinase (ALK1) (Figure 2F). Based on histopathology and immunohistochemical findings, the diagnosis was confirmed as inflammatory pseudotumor. Post-operative period was uneventful and patient was discharged on postoperative day seven. Currently, the patient is asymptomatic and is under close follow up to check for possible recurrence.



Fig. 1: Contrast enhanced computerized tomography showed a large necrotic mass (marked with yellow arrow) in left adnexa measuring 9cm x 7cm x 6.5cm abutting superior wall of urinary bladder and lower anterior wall of uterus in coronal view (1A) and sagittal view (1B). Left ovary was seen adherent to the posterior part of the mass (1C, marked with red arrow). Right ovary was separate from mass (1D, marked with orange arrow).

3. Discussion

Inflammatory pseudotumor is a rare, benign lesion of unknown etiology that usually presents with variable and nonspecific imaging features giving impression of a benign or malignant neoplasm. Inflammatory pseudotumors, most commonly arise in the lung and orbit, although they may also develop in various organs, including those in the abdomen and pelvis.¹

Important differentials diagnosis to be considered on histopathology are spindle cell carcinoma, inflammatory myofibroblastic tumor, solitary fibrous



Fig. 2: Histopathology revealed fascicles of fibroblastic proliferation in scleroticstroma with interspersed chronic inflammatory cells composed of lymphocytes and plasma cells (2A, H&E, 200x). Multinucleated foreign body type giant cells were also seen (2B, H&E, 400x). Fallopian tube incorporated within excision biopsy (2C, H&E, 100x). On immunohistochemistry (IHC), proliferating fibroblasts were immunopositive for Vimentin (2D, IHC, 400x) and Smooth muscle actin (2E, IHC, 400x).

neurofibroma and leiomyoma. However, tumour, immunohistochemistry (IHC) can help in differentiating spindle cell carcinomas, IMFT and leiomyoma from inflammatory pseudotumors. On IHC, the spindle cells of inflammatory pseudotumors are negative for cytokeratin, usually stain positive for vimentin, and variably positive for Smooth muscle actin, whereas the spindle cell carcinomas are immunopositive for cytokeratin and epithelial membrane antigen. IMFT is immunopositive for ALK whereas inflammatory pseudotumor is ALK negative. Leiomyoma is immunopositive for desmin whereas inflammatory pseudotumor is desmin negative.

Only three cases of inflammatory pseudotumor have been reported in adnexa. One was an infiltrative type involving the uterus, parametria, and mostly the left adnexa that was successfully treated with antibiotics, with total regression of the lesion.³ The other two cases presented as circumscribed mass located between ovary and uterus, which were surgically removed.^{4,5} The composite clinical data of these previous cases and including our case have been summarized in Table 1.

The cause and pathogenesis of inflammatory pseudotumor still remain controversial. It is thought as a reactive inflammatory process secondary to surgery, trauma, or infection.⁶ In the present case, tubal ligation may be the offending cause of such a lesion. A subset of inflammatory pseudotumors appears to be associated with a variety of infectious agents including Epstein-Barr virus,

Case s	Age and sex	Symptoms	Radiological Findings	Treatment and Intervention	Follow up / Outcome
1.	53 Female	Fever, bilateral, hydroureteronephrosis and oedema of the left leg.	Computed tomography (CT) revealed a 13x10.5 cm mass in the pelvis, mostly at the place of the left adnexa, uterus and both parametria. Mass was also involving the surrounding tissues and producing bilateral hydroureteronephrosis.	Laparotomy was performed but no radical operation could be performed due to extensive involvement of surrounding structures by the mass. Antibiotic therapy was given for one month.	Follow-up CT, 4 and 8 months after laparotomy showed local regression of IPT. The last follow-up CT, 20 months after laparotomy, revealed no evidence of tumor.
2.	18 Female	Abdominal pain and discomfort.	Sonographic evaluation revealed a hyperechogenic mass measuring 32 x 36 mm and located between the right ovary and uterus.	Laparoscopic surgical excision of mass.	No evidence of recurrence at the time of last follow up.
3.	41 Female	Abdominal pain and abnormal vaginal bleeding.	Sonographic evaluation revealed a circumscribed mass measuring 9cm x 9cm in the left mesosalpinx.	Laparoscopic morcellation of mass.	No evidence of recurrence at the time of last follow up.
4.	29 Female (Present case)	Pain in lower abdomen, urinary frequency and dysuria.	Contrast enhanced CT showed a large necrotic mass in left adnexa measuring 9cm x 7cm x 6.5cm abutting superior wall of urinary bladder and lower anterior wall of uterus. Left ovary was seen adherent to the posterior part of the mass.	Laparotomy and surgical excision of mass.	Currently, the patient is asymptomatic and is under close follow up to check for possible recurrence.

Table 1: Description of composite clinical data of patients with adnexal inflammatory pseudotumor. ³⁻⁵

Actinomyces, Pseudomonas species and mycoplasma.⁷ There is no consensus on definite treatment of inflammatory pseudotumor of fallopian tube. However, as these lesions have a benign course, total resection of tumor and preservation of normal tissues at surgical margins remains the best curative treatment. One case of resolution by antibiotic therapy have been documented, with no reports of either the local recurrence or distant metastasis.

4. Conclusion

Inflammatory pseudotumor should be considered as a differential diagnosis in a female patient with adnexal mass, especially with a history of prior pelvic surgery. It is imperative to correctly diagnose inflammatory pseudotumor to avoid unnecessary radical surgery or chemoradiotherapy.

5. Conflicts of Interest

All contributing authors declare no conflicts of interest.

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None.

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