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 pseudotumour. 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 undergone 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 fibroblast 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
laparotomy with gross total excision of the mass. Intra-
operative 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 post-
operative 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
tumour, neurofibroma and leiomyoma. However,
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,
Table 1: Description of composite clinical data of patients with adnexal inflammatory pseudotumor.3–5

<table>
<thead>
<tr>
<th>Case</th>
<th>Age and sex</th>
<th>Symptoms</th>
<th>Radiological Findings</th>
<th>Treatment and Intervention</th>
<th>Follow up / Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>53 Female</td>
<td>Fever, bilateral, hydroureteronephrosis and oedema of the left leg.</td>
<td>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.</td>
<td>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.</td>
<td>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.</td>
</tr>
<tr>
<td>2.</td>
<td>18 Female</td>
<td>Abdominal pain and discomfort.</td>
<td>Sonographic evaluation revealed a hyperechogenic mass measuring 32 x 36 mm and located between the right ovary and uterus.</td>
<td>Laparoscopic surgical excision of mass.</td>
<td>No evidence of recurrence at the time of last follow up.</td>
</tr>
<tr>
<td>3.</td>
<td>41 Female</td>
<td>Abdominal pain and abnormal vaginal bleeding.</td>
<td>Sonographic evaluation revealed a circumscribed mass measuring 9cm x 9cm in the left mesosalpinx.</td>
<td>Laparoscopic morcellation of mass.</td>
<td>No evidence of recurrence at the time of last follow up.</td>
</tr>
<tr>
<td>4.</td>
<td>29 Female (Present case)</td>
<td>Pain in lower abdomen, urinary frequency and dysuria.</td>
<td>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.</td>
<td>Laparotomy and surgical excision of mass.</td>
<td>Currently, the patient is asymptomatic and is under close follow up to check for possible recurrence.</td>
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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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References


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