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Inflammatory Pseudotumor Presenting as a Mesosalpingeal Mass

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Abstract

We describe a case in which the clinical and pathological features of a mesosalpingeal mass led to a diagnosis of inflammatory pseudotumor, an unusual tumor found in a rare location. The differential diagnosis is rather broad and includes lesions ranging from Castleman disease to an immunoglobulin G4 (IgG4)-related fibrosclerosing tumor. The patient is alive and well at last follow-up.

Case report

A 41-year-old woman (G2P1) presented with a 5-month history of abdominal pain and abnormal vaginal bleeding. Clinical and ultrasound examination revealed a 9-cm mass in the left mesosalpinx, which was consistent with a leiomyoma. Intraoperative examination revealed a nodule attached to the left mesosalpinx, with no connection to the uterine body. The nodule was adherent to the peritoneal surface of the mesosalpinx. No other lesions, including retroperitoneal masses, were detected. The patient underwent laparoscopic morcellation of the nodule. Multiple tissue fragments of white color and soft consistency, ranging from 10-80 mm in size, were sent to the Pathology Department for analysis (Figure 1).

Microscopic examination revealed a proliferation of spindle cells with a fascicular and vaguely storiform architecture. The spindle cells resembled fibroblasts without nuclear atypia, rather than smooth muscle cells, and did not demonstrate a granulation tissue-type appearance. Mitotic figures were rare (less than 1 per 10 high-power fields [HPF]). Only sparse collagen was noted. A prominent vascular proliferation was identified throughout the lesion, with some vasocentric inflammatory cells obliterating vessels in some areas. The prominent inflammatory infiltrate consisted of diffusely distributed lymphocytes and plasma cells, as well as aggregates of inflammatory cells with rare lymphoid follicles exhibiting a “lollipop” appearance. No tumor necrosis was detected (Figures 2A and 2B).

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An immunohistochemical examination of the spindle cell component was negative for actin and desmin (excluding a peculiar leiomyoma or an inflammatory myofibroblastic tumor – a rare benign mesenchymal tumor with unknown etiology), for CD34 and CD31 (excluding a benign vascular tumor), for STAT6 (excluding solitary fibrous tumor), and for CD21 and CD35 (excluding a follicular dendritic cell neoplasm) (Table 1). Neither kappa nor lambda restriction was noted with chromogenic in situ hybridization, excluding a plasmacytoma (Figures 3A-C). However, the CD21 and CD35 markers highlighted the architecture of the follicles, suggesting a stroma-rich and vascular-rich Castleman disease, characterized by lymphoid and vascular hyperplasia of the lymph nodes but also found in the mediastinum and retroperitoneum (1). A noted consultant and co-author of the previously referenced paper (1) (See Acknowledgment) reviewed the current case and could not substantiate a diagnosis of vascular-rich Castleman disease. Idiopathic retroperitoneal fibrosis was also a diagnostic consideration. This disease usually arises close to the aortic bifurcation and typically features a more collagenized background than seen here. Furthermore, this historical entity has been largely replaced by the recognition of fibroinflammatory lesions related to immunoglobulin 4 (IgG4).

The inflammatory infiltrate showed reactivity for IgG4, but the number of plasma cells that stained positive for IgG4 was less than 50 per 10 HPF (Figure 4). The IgG4 serum level was not examined preoperatively, but the serum IgG4 level was within normal limits (0.6400 g/L) after surgery. The lesion did not fulfill published criteria for IgG4-related disease, particularly because the serum IgG4 level did not exceed 135 mg/dL (2). IgG4-related disease is a rare lesion with autoimmune etiology, associated with the development of fibrotic tumor-like lesions that can affect multiple organs (most commonly involving the pancreas but also the head and neck, thoracic and hepatobiliary systems, and retroperitoneum. Such proliferations (3-5) had not been reported in the gynecological tract until recently when an IgG4-related lesion involving ovary was reported (6). Unlike the example described here, a prominent eosinophilic infiltrate was described. Our final diagnosis was consistent with inflammatory pseudotumor of the mesosalpinx. Given this unusual diagnosis and the history of morcellation, a CT scan was performed recently, which was reported as within normal limits.

Discussion

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

Only two cases of inflammatory pseudotumor have been reported in the pelvis thus far. 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 (7). The second reported inflammatory pseudotumor was similar to ours, represented by a circumscribed mass located between the right ovary and uterus, which was surgically removed (8).

For the current case, surgical excision led to the entire removal of the lesion, and no other systemic corticosteroid or antibiotic treatment was indicated. Inflammatory pseudotumor, although occasionally sharing some histological features with IgG4 fibroinflammatory disease and a vascular proliferation associated with Castleman disease, appears to be a distinct clinicopathologic entity, reactive rather than neoplastic, in which conservative treatment is indicated.

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Figure 1.
Macroscopy: multiple tissue fragments of white color and soft consistency

Figure 2.

(A) Low-power microscopic appearance highlighting the vasculature, diffusely distributed and aggregated inflammatory infiltrates with a lollipop-like lymphoid follicle. (B) High-power microscopic appearance demonstrating bland spindle cells and scattered inflammatory cells

Figure 3.
CD21 (A) and CD35 (B) negative in the spindle cells but highlighting the architecture of the follicles within the lesion. The inflammatory infiltrate showed reactivity for IgG4 (C); however, the number of plasma cells that stained positive for IgG4 was less than 50 per 10 high power fields

Table 1**Immunohistochemical markers and methodology**

Antibody	CLONE	VENDOR	Ref #	DILUTION	RETRIEVAL
CD 21	1F8	DAKO	M0784	1:500	Protease 1 - 12'
CD 31	JC70	Roche	760-4378	N/A	CC1 16
CD34	QBEN10	Roche	790-2927	N/A	N/A
CD 35	Ber-MAC-DRC	DAKO	M0846	1:50	Protease 1 - 12'
DESMIN	DE-R-11	Roche	760-2513	N/A	CC1 24'
IgG4	HP6025	BINDING SITE	MC011	1:2000	CC1 24'
KAPPA-ISH	Kappa DNP Probe (495-524)	Roche	760-1201	N/A	ISH-Protease 2 - 4'
	Kappa DNP Probe (538-567)		760-1202		
	Kappa DNP Probe (597-626)		760-1203		
	Kappa DNP Probe (637-666)		760-1204		
LAMBDA-ISH	Lambda DNP Probe (664-693)	Roche	760-1205	N/A	ISH-Protease 2 - 4'
	Lambda DNP Probe (704-733)		760-1206		
	Lambda DNP Probe (768-797)		760-1207		
	Lambda DNP Probe (826-855)		760-1208		
SMA	1A4	Cell Marque	202M-96	1:100	N/A

Note: The Roche Benchmark Ultra instrument was used