OLGU SUNUMU CASE REPORT

Inflammatory Pseudotumor of the Urinary Bladder in Differential Diagnosis of Pelvic Masses: Case Report

Pelvik Kitlelerin Ayırıcı Tanısında Mesanenin İnflamatuar Psödotümörü

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ABSTRACT Inflammatory pseudotumor of the urinary bladder appears to be an infrequent, benign, non-epithelial tumor which still remains to be a controversial issue with respect to diagnosis and management strategies. Since it is easily misinterpreted as a malignant process, perioperative accurate diagnosis is of profound importance to avoid an extensive disabling surgery primarily in young subjects. From a gynecologic view of point, it should be considered in differential diagnosis of adnexial masses, since it may mimic an adnexial mass which requires a colloborative multidisciplinary approach. A 44-year-old woman suffering from fever and irritative urinary symptoms underwent laparotomy due to a solid adnexial mass. After pathological evaluation, the diagnosis was inflammatory pseudotumor of the urinary bladder. Herein, this rare lesion was discussed with a review of literature regarding the diagnosis and therapy.

Key Words: Urinary bladder, pelvic neoplasms

ÖZET Mesanenin inflamatuar psödotümörü, tanı ve tedavi yaklaşımları açısından hala tartışmalı olan, ender rastlanan, benign bir non-epitelyal tümördür. Malign bir lezyon ile kolaylıkla karıştırılabileceğinden, özellikle genç hastalarda radikal bir cerrahi girişimden kaçınmak için perioperatif tanının doğru konulması son derece önemlidir. Jinekolojik açıdan bakılacak olursa, bu lezyon multidisipliner girişimi gerektiren adneksiyal bir kitleyi taklit edebileceğinden adneksiyal kitlelerin ayırıcı tanısında düşünülmelidir. Kırk dört yaşında, ateş ve irritatif idrar yakınmaları olan bir kadın hastaya solid adneksiyal kitle ön tanısı ile laparotomi uygulandı. Patolojik inceleme sonucu, mesanenin inflamatuar psödotümörü şeklinde rapor geldi. Nadir görülen bu lezyon, tanı ve tedavisi açısından literatür bilgileri eşliğinde tartışıldı.

Anahtar Kelimeler: Mesane, pelvik neoplazm

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Inflammatory pseudotumor (IP) is an infrequent non-epithelial neoplasm of unknown etiology that is suggested to be associated with surgery, trauma or infection in some cases.[1] It has been given a variety of names such as atypical myofibroblastic tumor, plasma cell granuloma, nodular fasciitis, pseudosarcomatous myofibroblastic proliferation

and pseudosarcomatous fibromyxoid tumor pointing out the diagnostic dilemma of both the surgeon and the pathologist.[1]

IP appears unlikely to be a pure inflammatory process but rather a neoplastic lesion since it represents a potential local recurrence and vascular invasion. It is multifocal and IPs originating from a number of tissues such as lung, bone and soft tissue are shown to be clonal and having aberrant karyotypes.[2]

IP seems to be variable in size and appearance, predominates in females and may occur at any age.[1] In spite of histopathological features often mimicking a malignant tumor, clinical course is mostly benign with a slow growth but without metastasis or malignant transformation.

IP is suggested to be a soft tissue mesenchymal tumor of indeterminate or low malignant potential that should be differentiated primarily from a sarcoma with similar clinical and histopathological features.[3-6]

Herein, IP of urinary bladder in a female patient with a preoperative diagnosis of adnexial mass was presented and discussed with a review of literature regarding the differential diagnosis of IP.

CASE

A 44-year-old female patient suffering from fever (39°C), nausea, vomiting, lower abdominal and lumbar pain was admitted to our Department of Gynecology following ultrasonographic (USG) diagnosis of a pelvic mass at the emergency room.

Bimanual gynecologic examination revealed a right adnexial mass, 5-6 cm in size with irregular contours and solid consistency.

Abdominal USG examination was reported to reveal a right kidney increased in size (14 cm) with diminished parenchymal thickness, grade 3 hydronephrosis and an accompanying solid pelvic mass of 6 cm in size.

Laboratory findings were as follows:

ESR: 120 mm/hour, CRP: 26 mg/L (<10 mg/L), Ca 12.5:47.7 U/mL (1.9-16.3), Ca 15.3:29 U/mL (6.4-36.5), Ca 19.9:24.3 U/mL (0-33), Urine culture: >100.000 Escherichia coli colonies were present. The remaining laboratory data involving the renal function tests were found to be within normal limits.

She had been taken antibiotics in order to treat the urinary tract infection confirmed by the urine culture (Sulbactam-Ampicillin; Duocid® 1 g 4X2 IV, Gentamycin; Genta® 1X150mg IV). An indwelling nephrostomy catheter was conducted to drain the right side hydronephrosis.

She underwent laparotomy through a median incision. Abdominal exploration revealed a solid mass lesion, 6X6 cm in size, originating from urinary bladder. Urologic surgeons were invited to the operation ward. After cystoscopy and bilateral ureteral catheterization, excision of the mass originating from bladder dome was performed by Urologic surgeon.

Frozen pathologic examination was reported as “Inflammatory pseudotumor of the urinary bladder presenting a mesenchymal proliferative cystic degeneration”.

Postoperative period was uneventful and she was discharged on the 7th postoperative day without any problem.

Final pathological findings were reported as follows: The excised nodular mass was measured to be 6x5x4 cm. Macroscopically, it was well circumscribed but not encapsulated and encased in muscular tissue. On sectioning, the mass was predominantly cystic with translucent material inside and smooth partly hemorrhagic intracystic surface. Solid areas were tan-pink in color.

Microscopically, the lesion was composed of stellate and spindled myofibroblastic cells in myxoid background admixed with inflammatory cells, most of which were lymphocytes (Figure 1). Delicate, thin walled branching blood vessels were al-

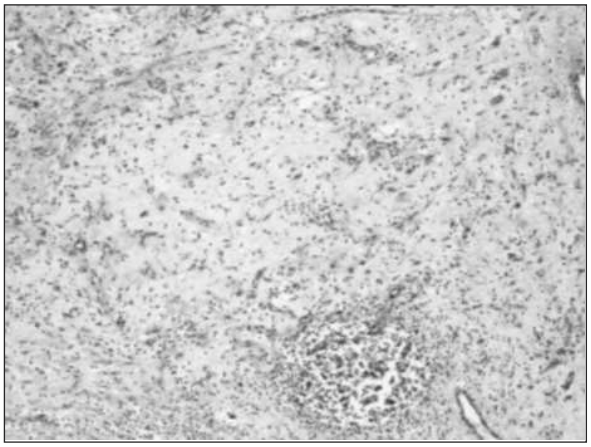

FIGURE 1: Spindle to stellate cells in myxoid stroma with scattered lymphocytes and delicate network of small vessels. (H&E x200)

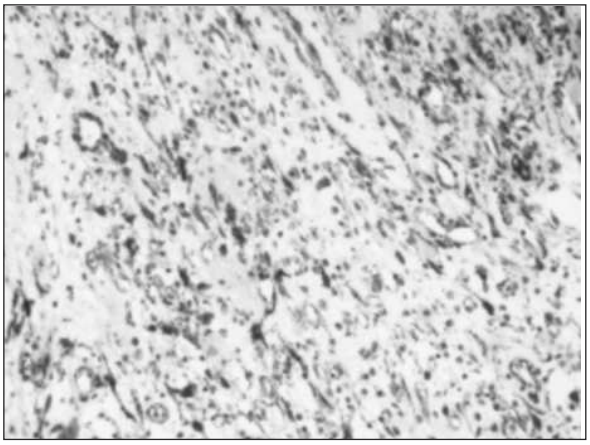

FIGURE 2: Smooth muscle actin positivity in the lesional myofibroblastic cells. (H&E x400)

so present in the myxoid matrix. Myofibroblasts had elongated nuclei but occasional cells with oval nuclei and prominent nucleoli were also noted. The lesion was mostly well-demarcated but invasion into the muscular tissue was focally detected.

Mitotic activity was low, 1 in 10 high power field. No atypical mitoses, hyperchromasia, atypia and necrosis were identified. Lesional cells expressed diffuse smooth muscle actin and desmin (Figure 2). There was no staining with antibodies to cytokeratin, S-100, CD 117 and CD 34. Ki-67 proliferation index was 3%.

DISCUSSION

Inflammatory pseudotumor has been reported to arise from a variety of tissues involving stomach, pancreas, liver, spinal cord meninges, uterus and retroperitoneum.[7] IP of the urinary bladder is an infrequent pathologic entity originating from the submucosal stroma, first described by Roth in 1980.[8]

Although etiopathogenesis is explained by a localized inflammatory response to surgery or instrumentation, malignancy or infection such as chronic cysytitis, in most of the subjects the nature of the lesion remains obscure.[7] Prior inflammation, hysterectomy for leiomyoma, pyelonephritis, ruptured diverticulitis of sigmoid colon, transurethral bladder resection, indwelling catheter, cystitis are reported to be associated with IP of the bladder.[4] Our patient's medical history did not reveal such etiologic factors.

It may be clinically presented with hematuria, abdominal pain, irritative voiding symptoms or as an asymptomatic mass.

It occurs at any age with a strong female predominance (75%).

Since both histopathological and clinical features appear to be greatly variable and complicated, it is difficult to make the correct diagnosis and classification of those lesions which makes it a controversial issue for both the surgeon and the pathologist. An infiltration of a variable number and type of mononuclear cells consisting of lymphocytes, plasma cells, eosinophils, foamy histiocytes overlying a background of fibrous tissue is demonstrated. This leads to different terms regarding the nomenclature of those lesions such as inflammatory pseudotumor, inflammatory myofibroblastic tumor, plasma cell granuloma, fibrous xanthoma and pseudosarcoma. It may be easily misinterpreted as a malignant process pri-

marily a sarcoma due to similar clinical presentation and histology.[9]

Immunohistochemical analysis may aid in the accurate diagnosis. It usually demonstrates positive immunostaining for vimentin and actin while occasionally positive immunostaining for desmin. Immunoreactivity for myoglobin, keratins, S-100, CD 34 is usually determined to be negative although aberrant expression of cytokeratins may be occasionally demonstrated.[3]

In our patient, lesional cells expressed diffuse smooth muscle actin and desmin. There was no staining with antibodies to cytokeratin, S-100, CD 117 and CD 34. Ki-67 proliferation index was 3%.

IP of the bladder generally behaves as a benign lesion and is not prone to metastasis. On the other hand, demonstration of potential infiltrative local growth, vascular invasion, malignant transformation, local recurrence, development of multifocal noncontagious tumors and proven metastasis in some cases with acquired clonal chromosomal abnormalities in cell cultures pointed out a probable neoplastic origin.[1] Due to this potential for malignant spread and progressive organ damage, early diagnosis and treatment seem to be mandatory. Conservative excision of the tumor by transurethral access or partial cystectomy is adequately curative with rare recurrences. Avoiding an erronous diagnosis leading to unnecessary agressive management with its attendant complications is extremely critical.[3] Keeping in mind that most of those cases are young, every effort should be made to establish early diagnosis in those cases in whom the prevention of extensive disabling surgery is of profound importance. In spite of a few cases in literature reported to be treated with long term antibiotics followed by spontaneous regression, surgical resection is recommended to prevent recurrences.

As a conclusion, IP of the bladder remains to be a problematic entity with respect to accurate diagnosis due to diverse clinical and histological presentations and the following management strategies. Prompt diagnosis and treatment are of utmost importance to prevent a misdiagnosis of a malignant process such as sarcoma that requires extensive surgery with potentially devastating complications. Simple excision of the lesion is adequately curative in almost all cases. Close follow-up is absolutely recommended in order to manage potential local recurrences or metastasis.

Additionally, from a gynecologic view of point, since the presenting patient was assessed as a subject with an adnexial mass preoperatively, it should be emphasized that those unusual lesions may mimic adnexial masses preoperatively and the masses originating from the urinary bladder should be considered in the differential diagnosis since a perioperative colloborative multidisciplinary approach may be required. Preoperative evaluation with Intravenous pyelography (IVP), cystoscopy and ureteral catheterization should be indicated in differential diagnosis and management of adnexial masses with hydronephrosis.

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