Case Report

Plasma cell granuloma of the urinary bladder: A pseudotumor - A clinical dilemma

Vinod Priyadarshi, Jitendra Pratap Singh, Debashish Chakrabarty[1], Dilip Kumar Pal

Departments of Urology, and [1]Pathology, IPGMER and SSKM Hospital, Kolkata, West Bengal, India

Abstract Plasma cell granuloma is a rare benign proliferative lesion that often mimics a malignant mass clinically and radiologically and its presentation in the urinary bladder is exceptional. Presuming malignant mass, such lesions often receive radical treatment. We are presenting here one such tumor, which resembled as an urachal tumor and underwent partial cystectomy.

Key Words: Inflammatory pseudotumor, plasma cell granuloma, urinary bladder

Address for correspondence:
Dr. Vinod Priyadarshi, Department of Urology, IPGMER and SSKM Hospital, Kolkata - 700 020, West Bengal, India. E-mail: vinod_priyadarshi@yahoo.com
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INTRODUCTION

Inflammatory pseudotumors of the bladder are rare benign proliferative lesions that resemble a malignant tumor on the clinical, radiological and endoscopic examination and hence, they often receive unnecessary radical treatment.[1] Because the term "inflammatory pseudotumor" is non-specific and the lesions have a variety of histologic presentations, several alternative names such as inflammatory myofibroblastic tumor, plasma cell granuloma (PCG), pseudosarcomatous myofibroblastic proliferation or xanthomatous pseudo tumor are in use, depending upon the predominant inflammatory cell in the lesion.[2] We are presenting here a rare case of PCG, which was present on the anterior bladder wall mimicked an urachal tumor.

CASE REPORT

This was a case report of a 20-year-old married lady, presented with recurrent complains of dysuria and frequency for about 1½ years with occasional lower abdominal pain. Shereported to have undergone laparoscopic bilateral tubal ligation 5 years back. She was subjected to repeated urine examinations in the last 1 year in view of her urinary symptoms. Though the urine cultures were always negative, microscopic urine examination demonstrated hematuria on a few occasions. Her clinical examination, hematological and biochemical studies and plain X-ray abdomen were normal; ultrasonography of abdomen suggested of a projecting mass in the urinary bladder near its dome [Figure 1]. Urine

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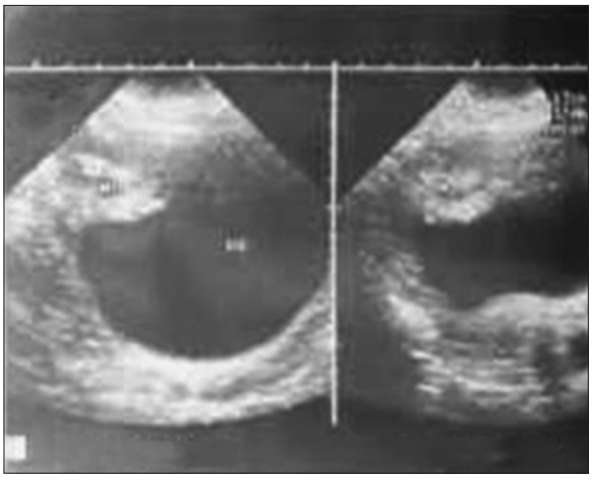

Figure 1: Ultrasonography showing mass protruding into the bladder lumen along anterior bladder wall

cytology was negative for malignant cells. Cystoscopic examination revealed indentation at the dome of bladder; however there was no mucosal growth into the bladder lumen. Contrast enhanced computed tomography (CECT) of the abdomen revealed an enhancing hyperdense mass arising from the anterior wall and dome of the urinary bladder protruding into its lumen and extending anteriorly toward anterior abdominal wall into the linea alba [Figure 2]. Computed tomography guided fine needle aspiration cytology of the mass revealed only inflammatory cells and no malignant cells were seen. Presuming the mass as urachal tumor and inconclusive cytological findings, partial cystectomy with umbliectomy was undertaken [Figure 3]. Post-operative period was uneventful and catheter was removed on the 10th post-operative day.

Histopathology of the mass suggested the presence of plenty of pleomorphic plasma cells with typical eccentric nuclei infiltrating into the bladder wall in the midst of other inflammatory cells, abundant spindle cells without cellular atypia mitotic figures and malignant cell component, which was suggestive of PCG [Figure 4]. Immunocytochemistry was positive for vimentin and actin with the presence of all three immunoglobulins IgG, IgA and IgM while it was negative for cytokeratin, desmin and epithelial membrane antigen.

Patient has been kept in close surveillance; she is symptom free and without any evidence of recurrence after 2 years of follow-up.

DISCUSSION

PCG is a benign inflammatory mass of unknown etiology, which comprises of mixed cell infiltrate, predominantly of polyclonal mature plasma cell with few histiocytes and lymphoid cells along with variable fibrous tissue component within or surrounding the infiltrate.[3] It was first described in 1973 by Bahadori and Liebow.[4] The lung is the most common site of occurrence, although it may occur in any organ. Occurrence of PCG within the urinary bladder is extremely rare, however it is the most common site in the urogenital tract.[2,5]

Inflammatory pseudotumor of the urinary bladder was first reported by Roth in 1980, when he described an ulcerated bladder lesion as reactive pseudosarcomatous response.[6] However, first description of PCG in the urinary bladder was made by Jufe *et al*. in 1984.[7] There are only a few case reports in literature so far and the exact incidence is unclear because of the various nomenclatures i.e. inflammatory myofibroblastic tumor, inflammatory pseudotumor, pseudosarcomatous myofibroblastic proliferation, inflammatory myofibroblastic proliferation, inflammatory myofibrohistiocytic proliferation, xanthomatous pseudotumor are in use to describe this entity.[2,4,8] However, contrary to previous reports, in a recent review Young suggested the term "Inflammatory myofibroblastic tumor" should be used as a separate entity, that is neoplastic and typically occur in children.[9]

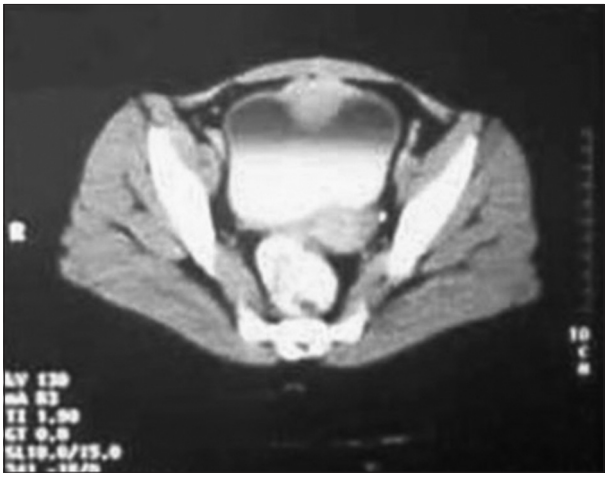

Figure 2: Contrast enhanced computed tomography showing anterior wall bladder mass

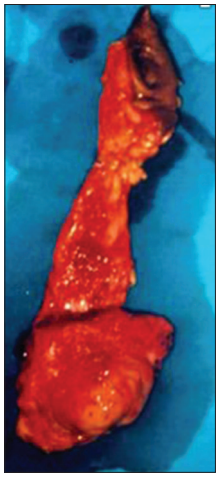

Figure 3: Partial cystectomy with umbilectomy specimen

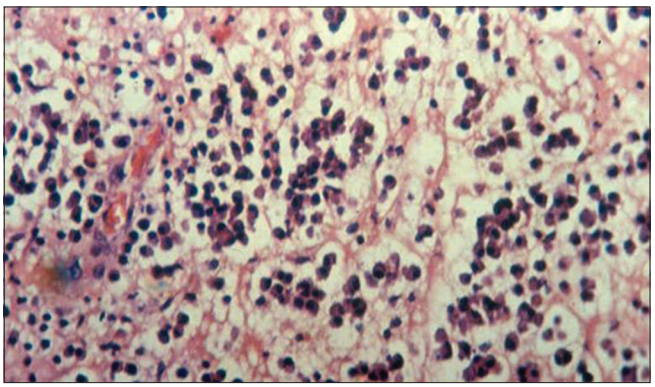

Figure 4: Histopathology of the mass showing plenty of pleomorphic plasma cells with typical eccentric nuclei

Though this lesion may occur at any age, it is typically seen in young adults (average age 28 years) and is rare in children. Women appear to be twice at risk as compared to men.[2] The size of the lesion is quite variable and has been reported to be as large as 9 cm, but most do not exceed 6 cm.[10] Painless gross hematuria from exophytic and ulcerated lesions is the most common initial manifestation and may result in anemia. Other symptoms include frequency of urination and dysuria and rarely feature of urinary tract obstruction. The lesion may be found at any site in the bladder. However, the trigonal involvement has not been reported except for secondary trigonal invasion from lesions developing on the posterior wall of the bladder.[2] The tumor appears as a polypoid intraluminal mass or a submucosal mass with or without extension into the perivesical fat. Some lesions demonstrate extensive sclerosis and infiltrating margins.[2,10]

The cause and pathogenesis of inflammatory pseudotumor still remain controversial. It is thought as a reactive inflammatory process secondary to surgery, trauma, or infection.[2] 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, Actinomyces, Pseudomonas species and mycoplasma.[5] Histologically, they are characterized by the presence of acute and chronic inflammatory cells with a variable fibrous response which is variably dominated by differentiated vimentin positive myofibroblastic spindle cells. The inflammatory component consisting mainly of lymphocytes and plasma cells which invariably show non neoplastic polyclonal kappa and lambda immunoglobulin light chains on immunocytochemical analysis.[2,3,7] This differentiates these lesions from plasmacytomas, that consist of diffuse sheets of neoplastic, variably differentiated, monoclonal plasma cells in the midst of mitotic activity and amyloid deposition.[4,11]

The imaging characteristics inflammatory pseudotumors vary widely and are nonspecific; most present with large masses mimicking malignant lesions. Sonographic findings show a variable pattern of echogenicity, with ill-defined or well-defined margins. Prominent vascularity may be shown with color or power Doppler sonography. CECT may show homo or heterogeneity and hypo-, iso-, or hyperdensity. Magnetic resonance imaging shows a hypointense lesion on T1- and T2-weighted images, but marked gadolinium enhancement. As such, definite radiologic differentiation from malignancy is not clearly possible.[2]

On endoscopic examination too, these lesion are mostly broad based, singular and appear malignant.[10] They are submucosal or deeply seated many a times and are not amenable for transurethral resection or biopsy.[2,12] similar to the present case. Such cases present as clinical dilemma and image guided histological biopsy (ideally core biopsy) remains the last resort.

There is no consensus on the best treatment modality due to the rarity of the disease. Though it is a benign lesion, most author prefer complete resection, especially in the form of partial cystectomy or transurethral resection, considering the difficulty to differentiate these lesions from malignant masses.[1,2,13] A more conservative approach for such lesions, using tamoxifen or high dose corticosteroid with gradual tapering, is reported to be equally effective provided definitive preoperative diagnosis can be made.[8,13-15] Long term antibiotic therapy may aid in resolution of mass especially if the primary insult is infective.[1] A medical therapy should be offered wherever a complete excision is not amenable.[13,15] However, recurrence is rare after complete excision.[1,10]

CONCLUSION

Although a rare condition, PCG can occur anywhere in the urinary tract and especially in the bladder. Awareness of PCG and its varied presentations on the part of treating clinicians may avoid unwarranted radical surgery.

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