Case Report

Prepubic inflammatory liposarcoma presenting as urosepsis

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ABSTARCT

Inflammatory well-differentiated liposarcoma is a rare soft-tissue tumor which is predominantly retroperitoneal in origin. We report a 72-year-old male without co-morbidities with suspected urosepsis and an obstructing ureteric calculus. Despite adequate diversion and broad-spectrum antimicrobials, the leukocytosis persisted. Further imaging revealed a locally infiltrating prepubic mass which was suspicious of a lymphoid malignancy and was found to be a high-grade liposarcoma on biopsy. He underwent open wide local resection of the tumor and the histopathology revealed an inflammatory well-differentiated liposarcoma with areas of neutrophilic abscess and necrosis. A dramatic response to the surgical resection was seen and the leukocytosis resolved within a few days.

INTRODUCTION

Inflammatory well-differentiated liposarcoma is a rare variant of soft-tissue sarcoma characterized by the presence of dense lymphocytic infiltration in the adipocytic component. This entity was first described in 1997 in case series describing unusual variants of liposarcomas which mimic lymphoid disorders on histopathology.[1,2] Cases reported in the literature are predominantly retroperitoneal in location,[3] with a few reports in the paratesticular,[4,5] scrotal,[6] or thoracic[7,8] location. Inflammatory liposarcoma in the prepubic location has never been reported. Herein, we report a case where this tumor eluded clinical detection while mimicking sepsis or a lymphoproliferative disorder.

CASE REPORT

A 72-year-old male presented with high-grade fever for 1 month and a high leukocyte count of 44,000/μl. The patient was obese (body mass index of 36.4) but had no other co-morbidities. During the evaluation of the fever, a 15-mm calculus was found impacted in the right upper ureter, and a percutaneous nephrostomy was placed for upper urinary tract diversion. Despite the broad-spectrum antibiotics and adequate diversion, the leukocytosis (42,000/μl) persisted. Hematology consultation was sought with a suspicion of lymphoproliferative disorder, but the peripheral blood smear showed only leukemoid reaction. The past history of a right orchidectomy, 20 years back, probably for an infective etiology and the clinical finding of an innocuous pre-pubic swelling, which was firm and non-tender, suggested further diagnostic radiological evaluation. A pelvic magnetic resonance imaging was obtained, which revealed an enhancing soft-tissue mass infiltrating the left pubic bone, adductor brevis, and the inferior rectus abdominis muscles, with the right spermatic cord structures appearing inseparable from the mass as shown in Figure 1. The mass abutted the dorsal surface of the penile shaft without infiltrating the Buck's fascia or the corpora cavernosa.

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A biopsy revealed spindle cells with foamy vacuolated to eosinophilic cytoplasm, multiple nuclei, and abundance of mixed inflammatory infiltrate, suggestive of a high-grade liposarcoma. An open wide local excision of the mass, including an anterior pubectomy was performed [Figure 2] and the histopathology showed spindle cells arranged in short intersecting fascicles and in focal storiform pattern, admixed with dense lymphoplasmacytic infiltrate, along with multiple neutrophilic abscesses and areas of necrosis, compatible with the diagnosis of inflammatory atypical lipomatous tumor/well-differentiated liposarcoma, FNCLCC Grade 1. Nuclear positivity for MDM2 on immunohistochemistry confirmed the diagnosis [Figure 3]. All the resected margins were free from tumor. The patient had an uneventful convalescence, and the leukocyte counts returned to the normal range within a few days. Our multidisciplinary

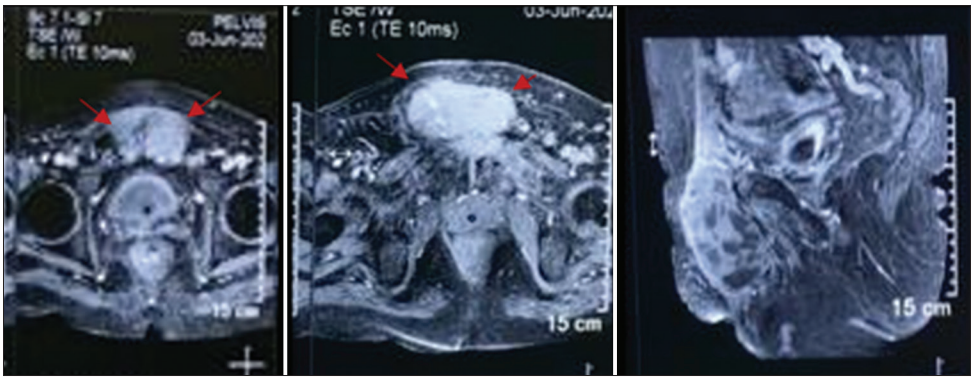

Figure 1: MRI pelvis showing an enhancing soft-tissue mass infiltrating into the pubic bone and adjacent muscles. MRI: Magnetic resonance imaging. Red arrows show locally infiltrating nature of the tumor

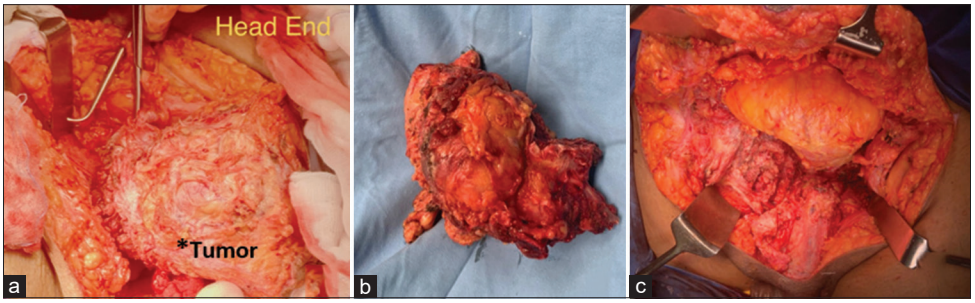

Figure 2: (a) Intraoperative picture showing a locally infiltrating soft-tissue mass (b) Resected specimen with negative surgical margins (c) Tumor bed after complete resection. *Tumor

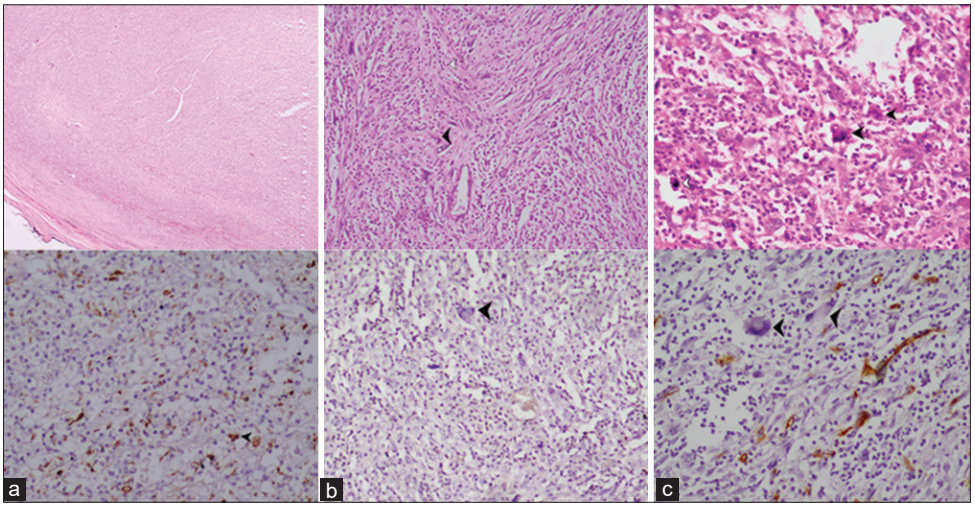

Figure 3: Histopathological evaluation (a) H and E staining: Well-circumscribed tumor compressing the surrounding tissue (a: H and E, ×20); large atypical spindle cells (black arrowhead) in vague fascicles, infiltrated by dense mixed inflammatory cells (white arrowhead) (b: H and E, ×100); large multinucleated tumor cells (black arrowheads) and atypical mitosis (white arrowhead) (c: H and E, × 200) (b) IHC markers: Atypical spindle cells (arrowhead) showing nuclear positivity for MDM2 on immunohistochemistry (a;IHC,x200); tumor cells showing negativity for ALK (b;IHC,x200) and CD34 (c;IHC,x200) on immunohistochemistry

tumor board decided against adjuvant radiotherapy in view of the well-differentiated low-grade pathology. In the absence specific guidelines, the patient has been advised a follow-up schedule similar to other soft-tissue sarcomas, with 3 monthly visits for the first 3 years, 6 monthly for the next 2 years, and annually thereafter. After 6 months of clinical follow-up, the patient is free of recurrence and a right ureterolithotomy is planned for the ureteric stone.

DISCUSSION

Inflammatory liposarcoma is a rare subtype of soft-tissue sarcoma comprising 2% of all the liposarcomas. In the majority of the cases, the diagnosis is purely made on the pathological examination and is a challenge for the pathologist. The clinical features are mostly nonspecific or are related to the mass effect of the tumor and a presentation with fever is extremely rare.[9]

Our patient was admitted with the clinical picture suggestive of sepsis and the search for an underlying focus ultimately led to the diagnosis of a soft-tissue mass in the prepubic location. The etiology of this peculiar presentation was explained by the presence of extensive lymphoplasmacytic infiltrate, including eosinophils and foamy histiocytes, and multiple neutrophilic abscesses with areas of necrosis as seen on the histopathological evaluation of the tumor.

The existing literature on inflammatory liposarcoma is sparse and comprises of two case series and individual case reports. Retroperitoneum is the most commonly described location similar to the other variants of liposarcoma.[1,2] The prepubic location of the tumor and the close proximity to the penile shaft, in our patient, posed a unique surgical challenge and required combined efforts with the orthopedics team for an oncologically safe wide excision of the tumor.

The pathological distinction of an inflammatory well-differentiated liposarcoma from the other lymphoproliferative disorders such as lymphomas, Castleman's disease, sclerosing mesenteritis, and idiopathic retroperitoneal fibrosis is crucial and failure to do so can have grave consequences on the choice of the treatment and the prognosis. Fine-needle aspiration biopsy may miss the lipomatous part of the tumor and can lead to misdiagnosis.[2,3] Immunohistochemistry for MDM2 expression is highly specific for well-differentiated liposarcomas and can be a useful adjunct in the difficult cases.[10]

Inflammatory well-differentiated liposarcoma does not metastasize in the absence of dedifferentiation but is notorious for multiple recurrences owing to its infiltrating growth pattern.[11] Close surveillance is essential, and adjuvant radiotherapy may be offered to those with high-grade tumors or local recurrences.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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