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Aggressive Angiomyxoma of the Prostate Mimicking Benign Prostatic Hyperplasia

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Abstract
Aggressive angiomyxoma (AAM) is a locally invasive soft tissue tumor with a high risk of local recurrence but without metastatic spread. The mesenchymal tumor is relatively site-specific and has a peak incidence in females in their 2nd or 4th decade. Only few cases in males have been reported in the literature. We describe what we think is the first case of an aggressive angiomyxoma arising in the prostate presenting with classical symptoms of benign prostatic hyperplasia.

Key words: aggressive angiomyxoma, prostate, benign prostatic hyperplasia

Introduction

Aggressive angiomyxoma (AAM) is a distinctive, locally invasive soft tissue tumor, that is predominately found in the perineum and pelvis. There is a strong propensity for local recurrence and woman are affected in about 90% of the cases, usually in their 2nd or 4th decade of life [1, 2]. Histopathologically, AAM presents mesenchymal stellate or spindle cells with loose myxoid background of collagen fibers, low mitotic activity and small vessels with thickened walls [3]. To date, only few cases in male patients have been reported. We present what we think is the first case of aggressive angiomyxoma arising from the prostate.

Case Report

A 64-year-old male suffered from ongoing symptoms of prostatic enlargement as frequency, urgency and nocturia. Medical treatment for benign prostatic enlargement did not alter the symptoms. Abdominal and transrectal ultrasound revealed a mildly enlarged but normal prostate (60 ml) with few calcifications and residual urine of about 100 ml. The prostate specific antigen (PSA) level was 1.02 ng/ml; the free to total PSA ratio was 26.4%. The urine test and digital rectal examination were unremarkable. Transurethral resection of the prostate and postoperative recovery were uneventful. The postoperative flow studies showed a maximum flow rate of 14.9 ml/sec without residual urine. Histopathological examination revealed a myxoid tumor with a prominent vascular pattern of dilated capillaries, venules and arteries (Fig. 1A). Immunohistochemistry staining was positive for vimentin, but negative for S100, actin, desmin and CD34 (Fig. 1B). The Ki-67 reaction demonstrated a very low proliferative index of 1%. Magnetic resonance imaging studies were performed after the diagnosis and did not reveal additional tumor in the pelvis or perineum (Fig. 2). We decided to put the patient into a watchful waiting protocol. Four months postoperatively the patient is fine without signs of recurrence.

Discussion

Since its first description in 1983, aggressive angiomyxoma has gained recognition predominately in female patients [2, 3]. Only few cases have been reported in males with origin sites as the scrotum, spermatic cord, inguinal or perianal region, perineum and pelvis. To date, no case arising in the prostate has been reported.

Histologically, typical features include an infiltrative growth pattern, hypocellular myxoid stroma, delicate stromal cells and small and large blood vessels, some with medial hypertrophy. Immunohistochemically, the stromal cells in AAM are positive for vimentin and oestrogen or progesterone receptor and variably positive for desmin, CD34 and alpha smooth muscle actin [2, 4]. Genetic alterations were found particularly on the long arm of chromosome 12 (12q13-15). Strong diffuse nuclear positivity of the candidate gene CDK4 and negative staining for MDM2 tend to improve molecular diagnosis of AAM [4].

The adjective "aggressive" comes from the propensity to recur. Despite its low proliferative index, AAM tends to be infiltrative and highly recurrent. Recurrence may be common (9% to 72%) and may appear as early as several months after surgery or up to 14 years postoperatively [3-5]. Surgery has been the standard-of-care treatment, but can be mutilating with massive blood losses. A period of watchful waiting to assess growth rate seems appropriate in many cases [6]. Few reports describing the positive response to conservative treatment with gonadotropin-releasing hormone agonist in asymptomatic female patients exist [7]. Chemotherapy or radiation therapy are unlikely to help as patients with AAM demonstrate a low proliferative activity [6].

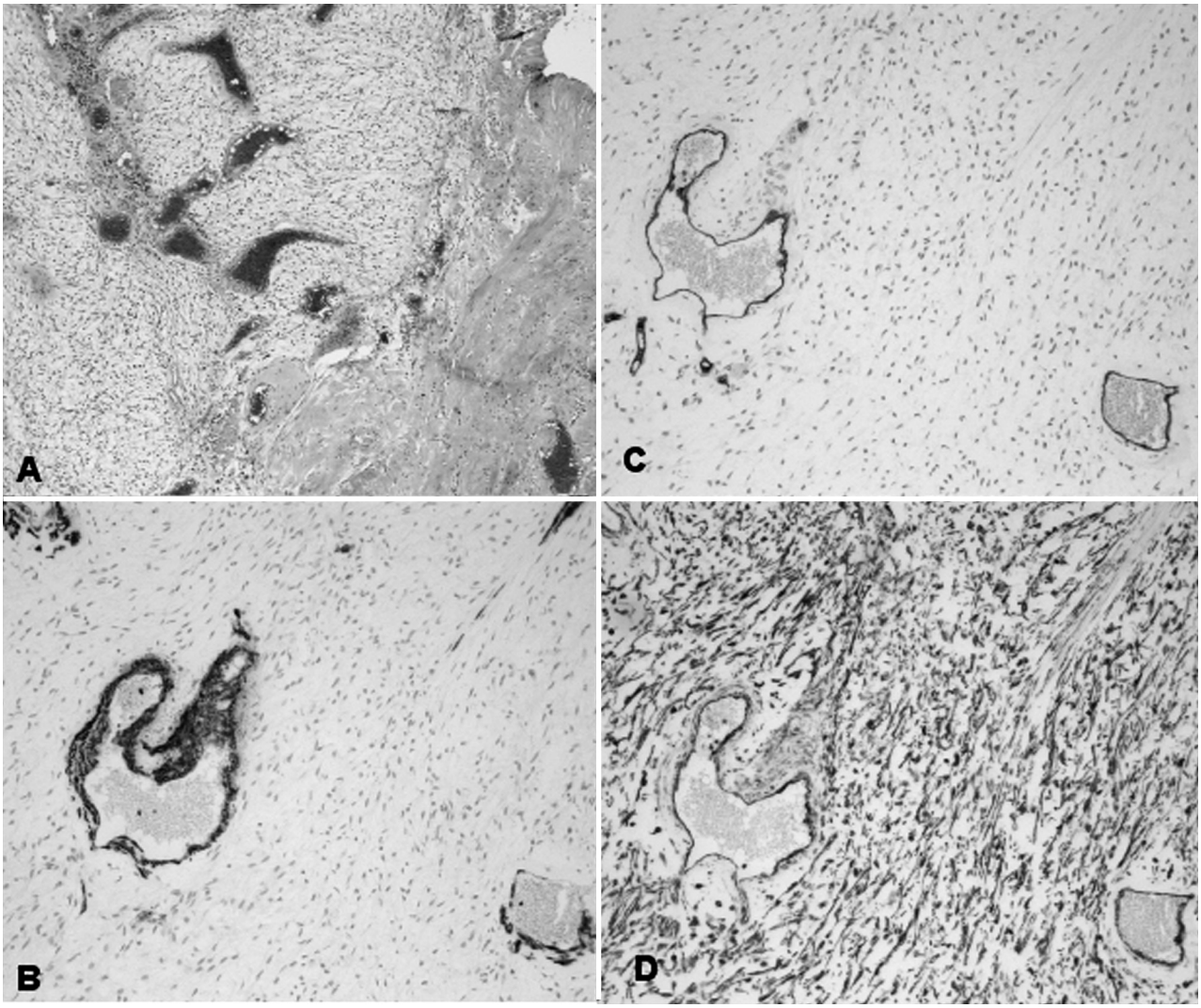

Fig. 1. A: Spindle cells in myxoid stroma with multiple blood vessels (hematoxylin and eosin). B: Positive staining for actin in stromal cells, C: Positive Staining for CD34 in stromal cells, D: Positive staining for vimentin in stromal cells.

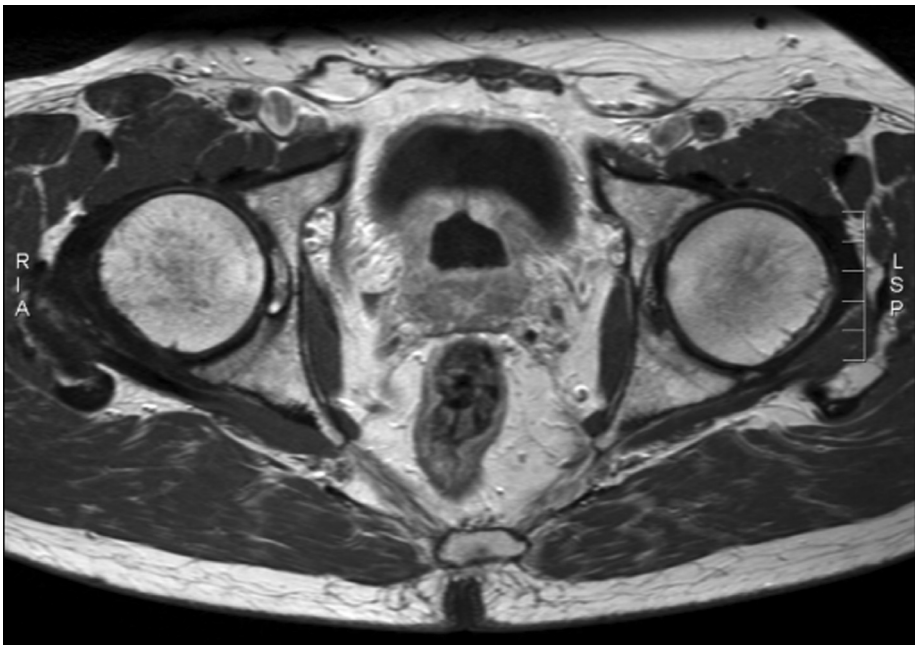

Fig. 2. Magnetic resonance imaging after transurethral resection of the prostate not showing further tumor.

Recurrence may be due to incomplete resection but patients with negative margins are as likely to have recurrence as those with positive margins [5, 8]. In spite of the high recurrence rate no distant metastases have been reported [2, 4].

Computerized tomography (CT) or MRI studies can detect the local extension and possible infiltration of other pelvic organs. CT scan demonstrates a well-defined moderate enhancement mass that is iso- or hypoattentuated relative to the muscle [9]. On T1-weighted MR images, AAM is isodense when compared to muscle, but after contrast material administration there may be moderately enhancement. On T2-weighted images, the tumor is described as having distinctive internal architecture composed of a swirled or layered appearance [10]. In our case, no preoperative imaging was done, but postoperatively MR imaging was able to rule out any tumor in the pelvis or perineum.

Problems in the differential diagnosis arise from other forms of soft tissue tumors since AAM resemble benign tumors with a low risk of recurrence (e.g. myxoma, myxoid lipoma or neurofibroma) or malignant tumors with metastatic potential (e.g. myxoid liposarcoma, myxofibrosarcoma or embryonal rhabdomyxosarcoma) [11].

All patients treated for AAM need close long-term follow-up, regardless of the primary treatment, to assess for recurrence.

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