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CASE REPORT

Leiomyosarcoma of Prostate
A Case Report and literature review

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ABSTRACT
Prostate leiomyosarcoma is an extremely rare and very aggressive neoplasm that represents less than 0.1% of primary malignant tumors of the prostate. We present a patient with primary leiomyosarcoma of prostate and examine the cases reported in the literature to discuss the clinical, diagnostic, and therapeutic aspects of this rare tumor. Median survival was estimated at 17 months (95% CI 20.7–43.7 months) and the actuarial survival rates at 1, 3, and 5 years were 68%, 34%, and 26%, respectively. The only predictors of long-term survival were negative surgical margins and the absence of metastatic disease at presentation. A multidisciplinary approach is necessary for the proper management of this terrible entity.
Surgery with or without chemotherapy seems to be the main therapeutic method for operable leimyosarcomas, but in general, there is no consensus on the best therapeutic approach. Most cases are diagnosed at an advanced stage of the disease. A global multicenter trial is needed to find therapies that would improve the prognosis.

KEYWORDS: Leiomyosarcoma, Prostate, Sarcoma.

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INTRODUCTION

Leiomyosarcoma of the prostate is an extremely rare malignant tumor that represents less than 0.1% of primary malignant tumors of the prostate [1]. It is the most common primary sarcoma of the prostate in adults and comprises 38% to 52% of primary prostate sarcomas [2]. It has a more aggressive clinical course compared to prostatic adenocarcinoma [3]. Its diagnosis is generally made at a late stage, because of the clinical and radiological non-specificity. It is often bulky beyond the prostate gland and invading neighboring organs. His prognosis remains very grim [3].

CASE REPORT

A 39-year-old patient admitted for voiding disorders evolving for 6 months in a context of the deterioration of the general state. On clinical examination, the presence of a hard hypogastric mass, fixed relative to the deep plane. On the digital rectal examination, the prostate was enlarged, nodular, integrating with the bladder base. Pelvic ultrasound showed heterogeneous prostatic hypertrophy. The PSA level was normal at 0.46 ng.

The thoraco-abdomino-pelvic scan revealed an enlarged prostate, heterogeneous after injection of contrast medium, with areas of necrosis, measuring 140/150/100 mm in diameter. There is an infiltration of the surrounding fat and urethral vessels, with upstream bilateral uretero-hydronephrosis. The rectum is pushed back and presacred without a border of separation with the prostate tumor, with the presence of multiple hepatic and pulmonary nodules of secondary appearance (Figure 1).

A prostate biopsy was performed, concluded that a prostatic leiomyosarcoma (Figure 2).

After bladder catheterization for acute retention of urine, adjuvant chemotherapy based on Taxotere / gemcitabine was started combined with analgesic radiotherapy in the pelvis. After three cycles of chemotherapy, the patient then received radiotherapy (60 Gy in 30 fractions). The control scanner showed a slight local improvement, on the other hand, an aggravation of the secondary lesions (Figure 3). The patient died a month later.

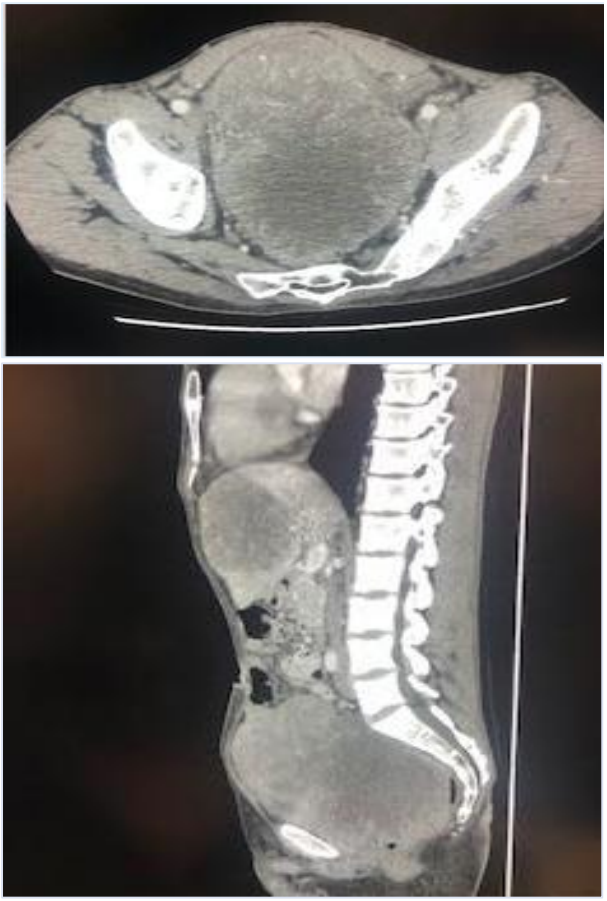

Figure 1: Abdominopelvic CT scan showing the prostate tumor invading the bladder. (Cross / sagittal section).

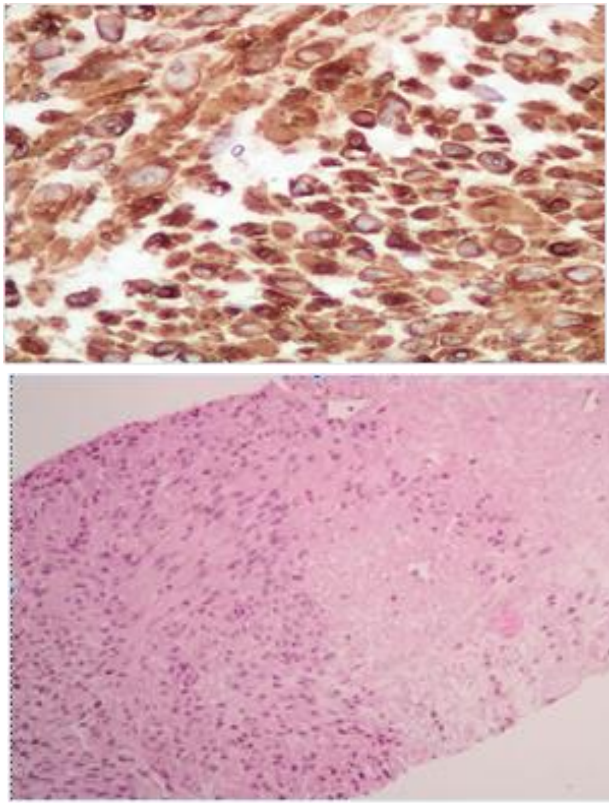

Figure 2: anatomopathological examination showing a partially necrotic fusocellular tumor proliferation with positive, strong, and diffuse immunostaining of anti-desmine AC tumor cells in favor of prostatic leiomyosarcoma.

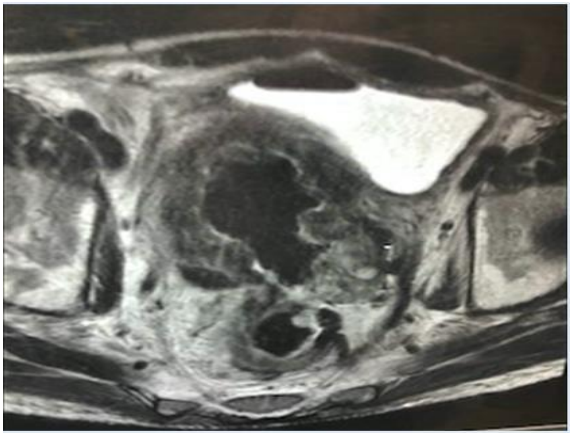

Figure 3: Abdominopelvic CT scan after chemotherapy showing partial radiological improvement.

DISCUSSION

Primary prostate leiomyosarcoma (PLSOP) is the most common sarcoma affecting the prostate, although it is generally a rare tumor [5]. PLSOPs cause an obstruction and tend to involve neighboring organs [5]. The etiology of PLSOP is unknown or poorly understood [5].

Prostate leiomyosarcoma has been reported in children and adults ranging from 2.5 years to 80 years of age [6].

PLSOP patients tend to have clinical signs that can clinically mimic the symptoms of benign prostatic hypertrophy: pollakiuria, poor urine flow, urgency, hematuria, and acute urine retention.

Leiomyosarcoma can also present as an exophytic hypogastric mass of the prostate infiltrating the rectum, a perineal mass, or by metastases to the liver and lungs [7]. Most cases are diagnosed at an advanced stage of the disease [7].

Digital rectal examination may show an enlarged prostate that may appear benign or a hard prostate mass that has spread to the capsule, rectum, pelvic sidewall, perineum, and seminal vesicle or involving the base of the bladder [7,8].

Serum PSA can be normal or high and does not allow the monitoring of leiomyosarcoma of the prostate after treatment [8].

Abdominopelvic ultrasound would reveal hydronephrosis; thickening of the bladder wall or infiltration of the base of the bladder [9]. Transrectal prostate ultrasound is a useful technique to better assess the characteristics of the prostate. It can show heterogeneous hypoechogenic lesions in the prostate, an invasion of the capsule or an extension in the rectum, the pelvic sidewall, the seminal vesicle, or the ejaculatory duct [9,10].

Echo-guided transrectal biopsy tends to be the usual approach to obtain samples of the prostate lesion for the histological examination which would establish the diagnosis of leiomyosarcoma of the prostate [7, 9, 10].

Transurethral resection of the prostate for symptoms of the lower urinary tract or retention of urine may lead to the accidental diagnosis of leiomyosarcoma during the histological examination of the resection coupons [1].

Thoracoabdomino-pelvic computed tomography or magnetic resonance imaging (MRI) can be used to assess

the characteristics of the prostate lesion, its location, and its local extent. They can indicate whether or not there is lymphadenopathy or metastases in the abdomen or the thorax, and also allow the patient to be evaluated and followed up after treatment. Bone scintigraphy makes it possible to search for bone metastases [1].

Therapeutic management has not yet been codified and there is no consensus on the best approach. Several protocols have been described in the literature [10,11], radical prostatectomy alone or with chemotherapy or radiotherapy; Chemotherapy and radiotherapy; Chemotherapy alone [10]; Radiotherapy alone [11].

Therapeutic combinations include surgery, pre- or post-operative radiotherapy, and neoadjuvant or adjuvant chemotherapy [12].

Radical retro-pubic prostatectomy is a good curative option for prostatic sarcoma [2].

Among the different chemotherapy regimens, an anthracycline-based combination (doxorubicin or epirubicin) with alkylating agents (cyclophosphamide, ifosfamide) and vinca alkaloids (vinblastine or vincristine) or a platinum-based combination was used with mixed results. [2,10,11]

The 5-year survival of prostatic leiomyosarcoma is very poor, ranging from 0% to 60%. [2,3]

The prognosis for prostatic leiomyosarcomas is often poor. Survival is very variable according to the series, it is on average less than 10% at 5 years [11]. It is relatively better in patients with complete curative resection and weak mitotic activity on biopsy. [13]

CONCLUSION

Adult prostatic leiomyosarcoma is a rare tumor, often metastatic when diagnosed, with a normal PSA level. CT and especially MRI play an important role in the assessment of extension and post-therapeutic monitoring, but only the anatomopathological examination, supplemented by an immunohistochemical study can confirm the diagnosis. Their therapeutic management is not codified at present and their prognosis remains very grim and can only be improved with a multidisciplinary approach and an early diagnosis, allowing to carry out a complete radical surgery, the only effective therapy.

AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors. Indeed, all the authors have actively participated in the redaction, the revision of the manuscript, and provided approval for this final revised version.

COMPETING INTERESTS

The authors declare no competing interests with this case.

PATIENT CONSENT

Written informed consent was obtained from the patient for publication of this case report.

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