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Original research

Feasibility and safety of conservative surgery for the treatment of spermatic cord leiomyosarcoma

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HIGHLIGHTS

- How to treat surgically spermatic cord leiomyosarcoma.
- European Mulcentric study, 23 patients enrolled.
- Spermatic cord leiomyosarcoma is a rare disease.
- Conservative treatment of spermatic cord leiomyosarcoma is a therapeutic option.

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ABSTRACT

Objectives: To assess the feasibility and the safety of conservative surgery to treat spermatic cord leiomyosarcoma.
Methods: Patients undergoing inguinoscrotal exploration in 10 different Urological Centers with diagnosis of leiomyosarcoma were enrolled. Preoperative evaluation included physical examination, Scrotal US, Abdominal CT and Scrotal MRI in selected cases. Patients underwent organ sparing surgery or orchiectomy in case of intraoperative FSE was positive for a local infiltration. Data collected were: age, presence of infiltration, length of the lesion, number of lesions, definitive histological outcome, pre and postoperative testosterone level. Follow up was performed with abdomen CT scan and scrotal US.
Results: From January 2007 to December 2013, 23 patients (mean age: 64.7 yrs) were diagnosed with spermatic cord leiomyosarcoma. Each patients underwent scrotal US. 10 patients underwent radical orchiectomy and 13 patients underwent conservative surgery. Mean follow up was 36.5 months. 5 patients (21.7%) developed a recurrent disease, 18 patients (78.3%) had a negative follow up (mean time: 40.8 months). Statistical analysis reveals that there is a significant correlation between number of lesions, length of the lesions and recurrent disease.
Conclusions: Spermatic cord leiomyosarcoma is a rare disease. Conservative surgical treatment of spermatic cord leiomyosarcoma is a feasible therapeutic option for small, single and not infiltrating lesion.

1. Introduction

Primary intrascrotal extra testicular tumors are rare (7–10% of all intrascrotal tumors). Leiomyosarcoma is thought to originate

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from the smooth muscle cells of mesenchymal origin in the wall of cremaster muscle and deferential ducts [1–3]. Low grade leiomyosarcomas have a good prognosis, whereas high grade tumors often develop metastases and have a significant tumor-related mortality [4]. Scrotal ultrasound has a key role in the first identification and description of the lesion; the appearance of these lesions is variable and non-specific. With the exception of liposarcoma, none of the others sarcomas can be differentiated from one another radiologically [2,5]. Frozen section examination (FSE) is necessary [6], but a correct diagnosis is difficult for the predominant lipid component of the lesion [2,3]. These masses are often identified only as mesenchymal tumors, without information about malignancy. Therefore the surgeon discovers that it is a leiomyosarcoma only at the time of the definitive histological report. Spermatic cord leiomyosarcoma is a rare entity and there is no international and validated guideline about its surgical management. Usually intrascrotal extra testicular masses are treated as high grade testicular tumors and a radical inguinal orchiectomy with high ligation of the spermatic cord is performed [1]. However the testicular and scrotal conservative surgery is of great interest in current literature [7,8]. Current literature offers only case reports and literature reviews [9–15]. Purpose of the study is to assess the feasibility and the safety of conservative surgery to treat spermatic cord leiomyosarcoma and to define preoperative criteria to go for a conservative surgery in patients with the evidence of localized disease.

2. Methods

Patients who underwent inguinoscrotal exploration in 10 different European Academic Medical Centers with definitive diagnosis of leiomyosarcoma as postoperative histological outcome were prospectively enrolled. This prospective study got the approval of the Ethical Committees of the different Centers. Central review of the histology was done by a senior pathologist to ensure the definitive diagnosis. Preoperative evaluation included physical examination, Chest X-ray, serum tumor markers (alpha fetoprotein, beta human subunit of chorionic gonadotropin, lactate dehydrogenase), testosterone level, Scrotal US, Abdominal CT scan and Scrotal MRI only in centers where this test was feasible. Patients were informed about the planned surgical procedure and signed a consent form. The procedure was performed by the same Surgeon for each participating center. The Surgeon identified for each center is a Consultant Urologist with particular expertise in testicular malignancy and in testicular sparing surgery. Each patients underwent an inguinal access to the testis. Once the spermatic cord was identified it was clamped to enable the Surgeon to safely manipulate the spermatic cord and the neoplastic lesion(s). Once the lesion was about to be excised a separate FSE of the spermatic cord lesion margins, whose length was 5 mm, was performed. In case of FSE was positive for a local infiltration an orchifuniculectomy with an high ligation of the spermatic cord was performed. If no infiltration was detected an organ sparing surgery (OSS) was the treatment of choice. Local infiltration was assessed during the FSE looking at the section margins separetly excised and at margins of the lesion excised. Data collected in the two groups were: age, presence of infiltration, length of the lesion, number of lesions, definitive histological outcome, pre and postoperative testosterone level at 6 months. Follow up was performed with abdomen CT scan to assess nodal involvement and scrotal US to assess local recurrence every 3 months for the first 12 months and then every 6 months. Data were summarized using descriptive statistics. Uni/multivariate logistic regression models were used to describe the probability of recurrence as a function of the following variables: patient's age, presence of infiltration, length of the lesion, number of lesions, and definitive histological outcome. A Wald Chi square test was used to assess p value.

3. Results

23 patients were enrolled in 10 Urological Division in the period between January 2007 and December 2013. Each patient had a confirmed postoperative histology report of a Leiomyosarcoma. Enrolled patients mean age is 64.7 years (SD 5.9) (Table 1). The two groups considered (OSS vs Radical Orchiectomy) appeared homogenous from a demographic point of view as the mean age is 62.8 years (SD 5.1) for OSS group and 66.1 years (SD 6.7) for Radical Orchiectomy group. Each patient's clinical history was similar as anyone underwent a Urological assessment because of a rapidly growing scrotal mass. In all patients, physical examination reveals the presence of a palpable mass. 5 patients had more than one extra testicular scrotal lesion. 18 patients had a solitary lesion at the scrotal US, 3 patients had two lesions while 2 patients had three lesions. Each patient had a preoperative CT scan which was negative for nodal involvement for any of them. Negative were also the preoperative serum markers in all the patients. 17 patients underwent preoperative scrotal MRI and none of them had a mismatch compared to the scrotal US findings. The mean size of the extra testicular lesion was 17.6 mm (range 8–50 mm. SD 10.4). The bigger ones were found to be in that cases in which there was more than one solitary lesion. 10 patients (43.4%) underwent orchifuniculectomy and 13 patients (56.6%) had an OSS. Table 2 underlines the surgical outcomes. Intraoperative frozen-section results were: 8 malignant tumors, 1 benign tumor and 14 mesenchymal neoplasia. The review of the definitive pathological report confirmed the diagnosis of leiomyosarcoma for all cases. Mean follow up was 36.5 months (SD 25.3). 5 patients (21.7%) developed a local recurrent disease discovered clinically and by scrotal ultrasound. This 5 patients had orchiectomy before and they were surgically re-treated for the local recurrence and then sent for oncological referral. 18 patients (78.3%) had no recurrence during their follow up period (mean time 38.3 months). None of the 13 patients treated with OSS relapsed or died with no evidence of local recurrent disease. Statistical analysis reveals that there is a significant correlation between number of lesions, size of the lesions and recurrence (Table 3). There is a significant reduction of postoperative testosterone levels in patients treated with radical orchiectomy but no patients reported associated symptoms.

4. Discussion

This series on Spermatic Cord Leiomyosarcoma represents one of the largest available in the Literature and may provide an insight on the clinical behavior of the disease and the efficacy of surgical treatments provided.

Primary intrascrotal extra testicular tumors are rare (7–10% of all intrascrotal tumors). More than 75% of these lesions arise from the spermatic cord. Mesenchymal tumors can be benign or malignant [1–3]. Among the malignant tumors, 20% has a leiomyosarcoma

Table 1
Preoperative patients features.

	Pts charateristics
Number	23
Age yrs (range, SD)	64.7 (56–75, 5.9)
Scrotal US findings	18 singular lesion 3 two lesions 2 three lesions
N° MRI performed (%)	17 (73.9%)
N° positive serum markers	0 (0%)
N° pts with normal preoperative testosterone	21 (91.3%)