Metastasis of primary testicular leiomyosarcoma to the retroperitoneal space

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ABSTRACT

A 70-year-old man presented with a left scrotal swelling. A computed tomography scan showed an 8-cm left scrotal mass and no metastasis. Radical orchiectomy with high ligation of the spermatic cord was performed. The tumor was classified as a high-grade leiomyosarcoma of the left testis. An intensive follow-up including repeated computed tomography scans was performed. A computed tomography scan 34 months after the surgery showed a retroperitoneal mass in front of the left kidney. Resection of the mass was performed. Microscopically, the mass was metastatic leiomyosarcoma. Intratesticular leiomyosarcoma is rare; only 18 cases have been reported. This is the first case in which leiomyosarcoma metastasized to the retroperitoneal space postoperatively. We herein review the literature and discuss how intratesticular leiomyosarcoma metastasized to the retroperitoneal space in this patient.

Key words: Retroperitoneal metastasis, spermatic cord leiomyosarcoma, spermatic cord sarcoma, testicular leiomyosarcoma, testicular sarcoma

INTRODUCTION

Sarcomas are uncommon tumors, with <8300 new cases reported in the United States in 2001. Sarcomas of the genitourinary tract account for <5% of these cases and <2% of all urological tumors. Approximately 100 paratesticular leiomyosarcomas have been reported in the literature. Adult intratesticular leiomyosarcoma is rare; only 18 cases have been reported. We herein review the literature and discuss how intratesticular leiomyosarcoma metastasized to the retroperitoneal space in this patient.

CASE REPORT

A 70-year-old man presented with a left scrotal swelling. A computed tomography (CT) scan showed an 8-cm left scrotal mass and no metastasis. The levels of human chorionic gonadotropin, alpha-fetoprotein, and lactate dehydrogenase were within normal limits. Radical orchiectomy with high ligation of the spermatic cord was performed. Despite adherence between the tumor and skin, neither tumor invasion nor metastatic nodules were found. The tumor was 4.5 cm in diameter, yellowish-white, solid, and encapsulated. No gross photograph was available, but the tumor was shown to originate from the testis proper with extra-testicular extension to involve paratesticular soft tissues and scrotum. Twenty-three sections were put into cassettes for microscopic evaluation and 110 high-power fields were assessed. Interweaving fascicles of elongated cells were identified. The tumor cells were characterized by moderate to severe cytologic atypia. Mitoses were identified for an average of 4 per 10 high-power fields [Figure 1a]. Atypical mitoses were also detected. No tumor cell necrosis was identified. Immunohistochemical staining showed that the tumor was positive for MIB-1 and SMA, but not for CD68, vimentin, desmin, S-100, CD34, or myoglobin. Although the cell turnover by MIB-1 staining was not quantified by image analysis, it was visually estimated at 10%. The tumor was classified as a high-grade leiomyosarcoma of the left testis (pT1N0). The spermatic cord, epididymis, and tunica vaginalis were not involved. The surgical margins were negative.
An intensive follow-up including repeated CT scans was performed. Although there was no recurrence or metastasis for 29 months, a CT scan 34 months after the surgery showed a retroperitoneal mass in front of the left kidney [Figure 2]. The mass was 7.5 cm in diameter, and no other abnormality was found. Because metastasis of the testicular leiomyosarcoma was suspected, resection of the mass was performed. Five sections were put into cassettes for microscopic evaluation and 10 high-power fields were assessed. An interweaving pattern of elongated cells arranged in fascicles was identified. The tumor cells were characterized by severe cytologic atypia. Mitoses were identified for an average of 10 per 10 high-power fields. Microscopically, the mass was metastatic leiomyosarcoma, and there was no lymphatic drainage invasion [Figure 1b]. An intensive follow-up was restarted, and there has been no recurrence or metastasis for 12 months.

**DISCUSSION**

Adult intratesticular leiomyosarcoma is rare; only 18 cases have been reported. This is the first case in which leiomyosarcoma metastasized to the retroperitoneal space postoperatively. Intratesticular leiomyosarcoma occurs from smooth muscle cells within the testis such as blood vessels, seminiferous tubules, and tunica. It is sometime difficult to distinguish intratesticular leiomyosarcoma from paratesticular leiomyosarcoma. In younger patients, somatic malignancy derived from teratoma should be excluded.

Table 1 summarizes the characteristics of 19 patients with intratesticular leiomyosarcoma. The median age of these patients was 55 years. The mean size of the tumor was 9 cm in diameter. The size was larger in the recurrent group than in the non-recurrent group (mean, 11.5 cm vs. 7.8 cm). The size of the tumor may be a risk factor for recurrence. All underwent radical orchiectomy, and only one patient underwent additional hemiscrotum and pelvic node dissection. Four patients received adjuvant therapy, three received radiation therapy and one received a combination of radiation and chemotherapy. Although radical orchiectomy is thought to be a standard treatment for these patients, it is uncertain which adjuvant therapy is more effective.

In a study by Coleman et al., wide re-resection after orchiectomy appeared to be beneficial surgical management for spermatic cord sarcoma. Of their 21 patients who underwent wide re-resection, 24% had a positive surgical margin at orchiectomy. None had a positive surgical margin and 29% had residual sarcoma in the specimen after wide re-resection. Our patient had no pathological invasion through the testis and negative surgical margins, but metastasis occurred. Although wide re-resection could not control metastatic disease, it may reduce the positive surgical margin and the local recurrence.

Of the 19 patients whom we have summarized, 5 developed distant disease after orchiectomy.

All the four known grades of tumor were high grade. Histological high grade may be a risk factor for distant metastases. Sites of metastasis included pulmonary (n = 3), discontiguous retroperitoneum (n = 1), and soft tissue and bone (n=1). The median follow-up was 17 months (range, 6–237 months).

Routes of spread for the sarcoma may vary depending on the histological type. Local invasion is the most common route for all histological types of sarcoma. Rhabdomyosarcoma can spread through lymphatic routes, so the risk of retroperitoneal involvement is increased. The other histological types of sarcoma can spread through hematogenous routes.

In our case, both intratesticular and retroperitoneal

![Figure 1](image-url)
Table 1: Case reports of primary intratesticular leiomyosarcoma in the literature

| Case | Author          | Age | Size (cm)* | Tumor grade | Initial Treatment ** | Adjuvant Therapy | Follow-up (month) | Outcome                     |
|------|----------------|-----|------------|-------------|----------------------|------------------|-------------------|---------------------------|
| 1    | Yachia et al.  | 50  | NA         | Unspecified | RO                   |                  | 24                | No evidence of disease    |
| 2    | Catton et al.  | 17  | NA         | High        | RO                   | radiotherapy + chemotherapy | 237               | No evidence of disease    |
| 3    | Hachi et al.   | 53  | NA         | High        | RO                   | radiotherapy     | 12                | Died of lung metastases   |
| 4    | Ali et al.     | 46  | NA         | High        | RO + hemiscrotum + pelvic node dissection | radiotherapy     | 9                 | Died of lung metastases   |
| 5    | Sattary et al. | 49  | NA         | High        | RO                   |                 | 20                | No evidence of disease    |
| 6    | Washecka et al.| 59  | NA         | Low         | RO                   |                 | 53                | No evidence of disease    |
| 7    | Catton et al.  | 47  | NA         | Unspecified | RO                   | radiotherapy + chemotherapy | 72                | No evidence of disease    |
| 8    | Hachi et al.   | 70  | 23         | Unspecified | RO                   |                 | 14                | Died of lung metastases   |
| 9    | Ali et al.     | 65  | 7          | Unspecified | RO                   |                 | 12                | No evidence of disease    |
| 10   | Sattary et al. | 27  | NA         | Low         | RO                   |                 | 30                | No evidence of disease    |
| 11   | Singh et al.   | 26  | NA         | Unspecified | RO                   |                 | unknown           | No evidence of disease    |
| 12   | Takizawa et al.| 76  | 7.4        | Unspecified | RO                   |                 | 12                | No evidence of disease    |
| 13   | Benjamin et al.| 30  | NA         | High        | RO                   |                 | 6                 | No evidence of disease    |
| 14   | Taue et al.    | 56  | 14         | Unspecified | RO                   | radiotherapy     | 24                | No evidence of disease    |
| 15   | Kumar et al.   | 65  | 8.5        | High        | RO                   |                 | 6                 | No evidence of disease    |
| 16   | Mechri et al.  | 73  | 6          | Low         | RO                   |                 | 36                | No evidence of disease    |
| 17   | Raspollini et al.| 77 | 4          | High        | RO                   |                 | 12                | No evidence of disease    |
| 18   | Venkatesh et al.| 55 | 7          | High        | RO                   |                 | 11                | Died of soft tissue and bone metastases |
| 19   | Present case   | 70  | 4.5        | Low         | RO                   |                 | 34                | Retroperitoneal metastasis |

* NA: not available, **RO: radical orchiectomy

Figure 2: A CT scan 34 months after the surgery shows a 7.5-cm retroperitoneal mass in front of the left kidney

Leiomyosarcoma specimens showed no invasion to lymph nodes or evidence of lymphatic routes, and repeated CT scan showed no lymphadenopathy. Primary soft-tissue sarcoma rarely show lymphatic spread. This sarcoma may spread through hematogenous routes to the retroperitoneal space.

In general, CT of the pelvis and palpation of the scrotum and inguinal region have been thought to be essential management tools. CT of the abdomen and chest are also important because 26% of patients, including our patient, developed distant metastasis.

All three patients who had lung metastasis died of disease in the short term (range, 9–14 months). Distant metastasis, especially to the lung, may be a sign of poor prognosis.

However, in the study by Coleman et al., 22% of the patients who either presented with metastatic disease or subsequently developed distant sarcoma after initial treatment had no evidence of disease. All underwent complete surgical resection of metastatic lesions. The median follow-up was 75 months. If possible, we may attempt complete resection of metastatic lesions because it provides a chance to control or cure disease.

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How to cite this article: Komeya M, Sahoda T, Sugiura S, Sawada T, Kitami K, Iemoto Y. Metastasis of primary testicular leiomyosarcoma to the retroperitoneal space. Indian J Urol 2012;28:343-6.

Source of Support: Nil, Conflict of Interest: None declared.

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