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Editorial Board Member of World Journal of Clinical Cases, Dr. Mukul Vij is Senior Consultant Pathologist and Lab Director at Dr Rela Institute and Medical Center in Chennai, India (since 2018). Having received his MBBS degree from King George Medical College in 2004, Dr. Vij undertook postgraduate training at Sanjay Gandhi Postgraduate Institute of Medical Sciences, receiving his Master’s degree in Pathology in 2008 and his PDCC certificate in Renal Pathology in 2009. After 2 years as senior resident, he became Assistant Professor in the Department of Pathology at Christian Medical College, Vellore (2011), moving on to Global Health City as Consultant Pathologist and then Head of the Pathology Department (2013). (L-Editor: Filipodia)

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RESPONSIBLE EDITORS FOR THIS ISSUE
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Paratesticular liposarcoma: Two case reports

Qi-Gang Zheng, Zhao-Hui Sun, Jia-Jian Chen, Jia-Cheng Li, Xiao-Jun Huang

Abstract

BACKGROUND
Paratesticular liposarcoma accounts for approximately 7% of scrotal tumors. They are rare lesions of the reproductive system with approximately 90% of the lesions originating from the spermatic cord. Surgery, with the goal of complete resection, is the mainstay for treatment of this disease. However, treatment consisting of extended resection to decrease local recurrence remains controversial.

CASE SUMMARY
We report the cases of two patients with paratesticular liposarcomas who were treated with radical testicular tumor resection without adjuvant therapy. Follow-up investigations at 9 mo showed no sign of recurrence.

CONCLUSION
Surgery is the first-line treatment, regardless of whether it is a recurrent or primary tumor. Extended resection carries a higher risk of complications and should not be performed routinely. Preoperative radiotherapy can reduce the local recurrence rate without affecting the overall survival.

Key Words: Case report; Paratesticular liposarcoma; Andrology; Radiotherapy; Surgery; Extended resection

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INTRODUCTION

Liposarcomas account for 5.75% of all tissue subtypes[1]. Atypical liposarcoma/well-differentiated liposarcoma (ALT/WDLPS) is the most common type of liposarcoma. ALT/WDLPS is mostly distributed in the retroperitoneum and deep tissue of the extremities and mediastinum, but rarely in the scrotum[2], with fewer than 200 similar cases reported to date in the English literature[3]. Surgery is currently the most effective treatment for patients with liposarcomas.

A retrospective multi-institutional study of 382 patients (including 106 WDLPSs) showed that the overall local recurrence rate was 49%[4]. Because of the high local recurrence rate, patients have to undergo repeated operations. However, subsequent operations can be disturbed by the scar from previous operations, leading to distortion of the normal anatomy. Hence, the quality and scope of the first operation need to be assessed. During treatment, factors under the surgeon’s control (including tumor integrity and extent of resection) and those reflective of tumor biology (grade and multifocality) affect patient outcomes[5].

We describe two rare cases of paratesticular liposarcomas in which the patients were treated with radical testicular tumor resection. Considering the rarity of cases, we review the literature regarding the application of surgical treatment.

CASE PRESENTATION

Chief complaints

Case 1: A 57-year-old Chinese man was hospitalized in the Urology Department of The Second Affiliated Hospital of Zhejiang Chinese Medical University (Hangzhou, China) because of a painless mass in his left scrotum for 1 year.

Case 2: A 62-year-old man presented with a mass in his left scrotum close to the abdomen, and it had grown slowly over about 30 d.

History of present illness

Case 1: He reported that the mass grew from the size of a soybean to the size of a fist in 1 year.

Case 2: He reported that the mass grew from the size of a soybean to the size of an egg within 20 d.

History of past illness

Case 1: The patient had no history of other illness and had no known allergies.

Case 2: He underwent left radical orchiectomy in our hospital 10 years ago due to the presence of a left scrotal mass. The pathological analysis indicated malignant fibrous histiocytoma. He had no known allergies.

Personal and family history

Case 1: The patient did not smoke or drink, and had no relevant family history.
Case 2: The patient did not smoke or drink, and no relevant family history was reported.

Physical examination
Case 1: The mass was firm with no tenderness, and the testicles were away from the scrotum. The results of the light transmission test were negative.

Case 2: A firm and non-tender mass was found in the left scrotum close to the abdomen and the left testicle was absent.

Laboratory examinations
Case 1: Serum carcinoembryonic antigen (CEA) level was 4.2 ng/mL (normal range: 0-5 ng/mL), and alpha-fetoprotein level was 3.0 ng/mL (normal value 8.0 ng/mL).

Case 2: The serum CEA level was 5.4 ng/mL, and CYFRA21-1 cytokeratin 19 fragment level was 2.49 ng/mL (normal range: 0-2.08 ng/mL).

Imaging examinations
Case 1: Computed tomography (CT) did not show the left testicle; however, there were massive low-density foci (CT value: -80 U) in the left scrotal region; the maximum cross-sectional area was approximately 8.6 cm × 6.8 cm (Figure 1A). Mainly, mature lipid density was noted with spotted nodular calcification (Figure 1B). Initially, we considered this to be a teratoma.

Case 2: Ultrasonography (US) showed a heterogeneous echo mass in the median fat layer of the perineum. CT showed a nodular high-density shadow above the left scrotum in the lower abdomen, approximately 3.2 cm × 2.2 cm in size, and the boundary was unclear.

FINAL DIAGNOSIS
Case 1
Based on the findings of the examination and imaging, a provisional diagnosis of liposarcoma was made.

Case 2
Based on the above findings, a provisional diagnosis of liposarcoma was made.

TREATMENT
Case 1
He underwent radical resection of the left testicular tumor on the 4th d after admission. The surgeon dissociated the entire spermatic cord up to the inner ring at the epididymis muscle. The spermatic cord was severed at the inner ring and sutured separately. The entire tumor and left testis, outside of the sheath, were resected. The tumor was a 14 cm × 8 cm × 6 cm, soft, gray-yellow mass (Figure 1C and D) with its capsule was intact and close to the testis. Pathological analysis showed that the tumor was composed of relatively mature adipocytes, single vesicular adipoblasts in the focal area, fibrous tissue with mucus deformation in the stroma (Figure 2A and B), and an intact tumor capsule. Immunohistochemical analysis revealed S100+, cluster of differentiation 34 (CD34), CD34+, vimentin-positive (Vim+), smooth muscle actin-positive (SMA+), CD68, and Ki67 < 1%, which supported the diagnosis of ALT/WDLPS.

Case 2
The patient underwent resection of the left inguinal tumor. The surgeon separated the tumor from the scrotum by 1 cm for R0 resection, and then resected it. The tumor measured about 7 cm × 5.5 cm × 2.5 cm, and we found a 3.5 cm × 3 cm × 2.5 cm gray-white mass in the middle with a clear boundary. Pathological analysis indicated that the tumor was an atypical liposarcoma (Figure 2C and D). Immunohistochemical analysis showed CD34+, S-100+, SMA-, VIM-, actin-negative (Act-), CD68 partial+,
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Figure 1 Computed tomography images. A: Computed tomography scan showing spotted and nodular calcification; B: The maximum cross-sectional area was about 8.6 cm × 6.8 cm; C: The giant tumor was a gray-yellow mass, measuring 14 cm × 8 cm × 6 cm, and its capsule was intact and close to the testis; D: The cut plane of the tumor was gray-yellow.

and Ki67 30%+, supporting the diagnosis of ALT/WDLPS.

OUTCOME AND FOLLOW-UP

Case 1
The patient was discharged 4 d after the operation without drugs or adjuvant therapy. At 9 mo postoperatively, there was no signs of recurrence.

Case 2
The patient showed good postoperative recovery without any adjuvant therapy. His symptoms were relieved 9 mo postoperatively, and there were no signs of recurrence.

DISCUSSION

To date, surgeons believe that extended resection should be performed for liposarcomas to reduce the possibility of local recurrence. In a French retrospective multiinstitutional study of 315 patients with retroperitoneal sarcomas who underwent complete gross tumor resection (R0/R1), 250 (79%) patients underwent compartmental organ resections including 120 (38%) patients who underwent compartmental complete resection. In the multivariate analysis, compartmental complete resection was associated with an improved 3-year cumulative incidence of local recurrence (10% compared with 47% after simple resection and 52% after non-compartmental organ resection). On the basis of these results, Bonvalot et al. recommended routine compartmental resection, regardless of histology. Nevertheless, this view is now being questioned. A retrospective analysis of 76 patients after complete resection (R0/R1) showed that 38 (46%) underwent organ resection; however, only 6 (7%) patients had
organ invasion\cite{6}. The data demonstrated that organ resection was not associated with overall survival (OS) or disease-free survival (DFS), whereas organ preservation was associated with a reduced risk of postoperative complications\cite{6}. When analyzing relapsed patients undergoing secondary surgery, the same conclusion was reached\cite{6}.

If we only focus on the rate of local recurrence but not on patients’ postoperative survival time, this is a typical “analysis paralysis” situation. Patients undergoing extended resection have a higher risk of complications than those not undergoing extended resection. Bonvalot et al\cite{4} reported that 16% of patients who underwent extended surgery have complications, and half of them require a second operation. The above situation can be aggravated by malnutrition\cite{8}. Therefore, conservative resection is undoubtedly more suitable for patients with poor physical condition, such as elderly men. Thus, for the two patients described herein, considering their ages, only gross marginal negative resection (R1) was performed.

In the past, pathologists thought that chemotherapy and radiotherapy were limited with regard to the biological characteristics of liposarcomas. In a recent retrospective analysis of patients with liposarcomas who were mainly treated with an anthracycline-containing regimen, the objective response rate was 12%; additionally, there was no correlation between chemotherapy and OS or DFS; however, radiotherapy can reduce the local recurrence rate\cite{9}. An analysis of 261 patients with liposarcomas showed that local failure-free survival was significantly improved if adjuvant radiotherapy was performed preoperatively instead of simple surgery (hazard ration [HR] = 0.42, 95% confidence interval [CI]: 0.21-0.86, $P < 0.05$\cite{10}. When performing radiotherapy for the same type of tumor, the required field of vision and dose are smaller preoperatively than postoperatively, and this modality is associated with fewer complications. Additionally, there is no evidence that radiotherapy has an effect on OS\cite{10}. Hence, surgeons should pay more attention to the positive effect of radiotherapy before performing surgery for liposarcomas; however, the two patients discussed herein did not undergo radiotherapy before surgery.

**CONCLUSION**

Surgery is the first choice of treatment, regardless of primary or recurrent liposarcoma. However, extended resection is not beneficial for OS and should not be performed...
routinely. We recommend selective resection of compartmental organs only if there is a clinical suspicion of invasion at the time of surgical resection. Radiotherapy can create a balance between the local recurrence rate and OS. Preoperative radiotherapy is a less harmful than extended resection to reduce the burden of marginal viable tumor.

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