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Case Report

A Case of Bilateral Synchronous Paratesticular Leiomyoma

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Bilateral synchronous paratesticular leiomyoma · Leiomyoma · Histopathology

Abstract
Bilateral synchronous paratesticular leiomyoma (BSPL) is a rare tumor that originates from smooth muscle cells in the paratesticular region. Four BSPL cases have been reported sporadically, starting with the 1991 report by Aus and Boiesen. Herein, we report the case of a 60-year-old male with a bilateral scrotal mass with a maximum size of 7.5 cm. Histological examination revealed oval to spindle-shaped tumor cells with a fascicular growth pattern. Immunohistochemically, the tumor cells were positive for α-smooth muscle actin. The pathological diagnosis was a leiomyoma. Based on the simultaneous bilateral nature of the disease, BSPL was diagnosed. In conclusion, we encountered a rare case of BSPL, and our report may contribute to the understanding of this disease.

Introduction

Paratesticular leiomyoma is a rare tumor that may originate from smooth muscle cells in the paratesticular region. Leiomyomas of the somatic soft tissue arise in the extremities, deep subcutis, or skeletal muscle [1]. It is estimated that approximately 20% of tumors in the scrotum are malignant [2], and the remainder are benign. Benign tumors include adenomatoid tumors, leiomyomas, lipomas, and spermatocele. Leiomyomas of the scrotum have

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been reported sporadically, starting with the 1972 report by Albert and Mininberg [3]. However, bilateral synchronous occurrence of leiomyomas in the paratesticular region is extremely rare, with only four cases reported to date.

Herein, we report an extremely rare case of bilateral synchronous paratesticular leiomyomas (BSPL). This is the fifth report of this tumor worldwide and the largest ever reported.

Case Report

A 60-year-old male without known comorbidities presented to our hospital with a bilateral scrotal mass. He had noticed bilateral testicular swelling for approximately 10 years. The patient had no history of trauma or surgery. Physical examination revealed scrotal heaviness but no scrotal pain. Laboratory data showed satisfactory serum concentrations of alpha-fetoprotein, interleukin-2 receptor, and human chorionic gonadotropin. Contrast-enhanced magnetic resonance imaging scan revealed well-circumscribed tumors of up to 7.5 cm in size in the scrotum with heterogeneous enhancement (Fig. 1a). No other lesions were observed. Based on imaging findings and laboratory data, these tumors were clinically suspected to be adenomatoid or leiomyomas. Inguinal orchiectomy was performed.

The surgically resected specimens were 9.0 × 8.0 × 5.5 cm (right) and 5.0 × 4.0 × 3.8 cm (left) in size. Gross examination revealed a solid gray-white, well-circumscribed, firm mass (Fig. 1b). The cut surface had a white whorled appearance similar to that of raw silk. These tumors were separate from the testes, which appeared brownish in color (Fig. 1c, d). Histological examination showed that the tumors were well demarcated and eosinophilic (Fig. 2a). The tumors were formed by interlacing bundles of smooth muscle cells separated by numerous well-vascularized connective tissue (Fig. 2b). The tumor cells had oval to spindle-shaped nuclei and acidophilic cytoplasm (Fig. 2c). No apparent necrosis or bleeding and mitotic figures were observed (0/10 high-power fields). Immunohistochemically, the tumor cells were positive for α-smooth muscle actin and negative for S-100a, CD34, and c-kit (Fig. 3a–e). The Ki-67 positivity rate was less than 1% (Fig. 3f).

The pathological diagnosis was a leiomyoma. Based on the simultaneous bilateral nature of the disease, BSPL was diagnosed. After resection, the patient was followed-up, and no recurrence or metastasis was observed. The CARE Checklist has been completed by the authors for this case report, attached as supplementary material (for all online suppl. material, see www.karger.com/doi/10.1159/000528821).

Discussion

This report describes a rare case of BSPL. This disease was first documented by Aus and Boiesen, who reported a bilateral leiomyoma of the tunica albuginea [4]. Since their initial report, 4 cases have been reported to date, and this is the fifth [4–7]. Similar to previous reports, in this case, the tumor arising from the paratesticular region was localized without invading the testis or seminal vesicle. In addition, this case involved the largest tumor size among all reported cases.

Leiomyomas arising in the scrotum are benign smooth muscle tumors similar to those of other sites of origin. They can also occur in the epididymis, spermatic cord, dartos, tunica albuginea, and testicular parenchyma [8]. Nevi are the most common benign tumor in the scrotum, and squamous cell carcinomas are the most common malignancy, of which leiomyoma was reported in 10 of the 127 cases of primary scrotal neoplasm [9]. The report of a tumor occurring simultaneously in both testes is very rare, with one report of

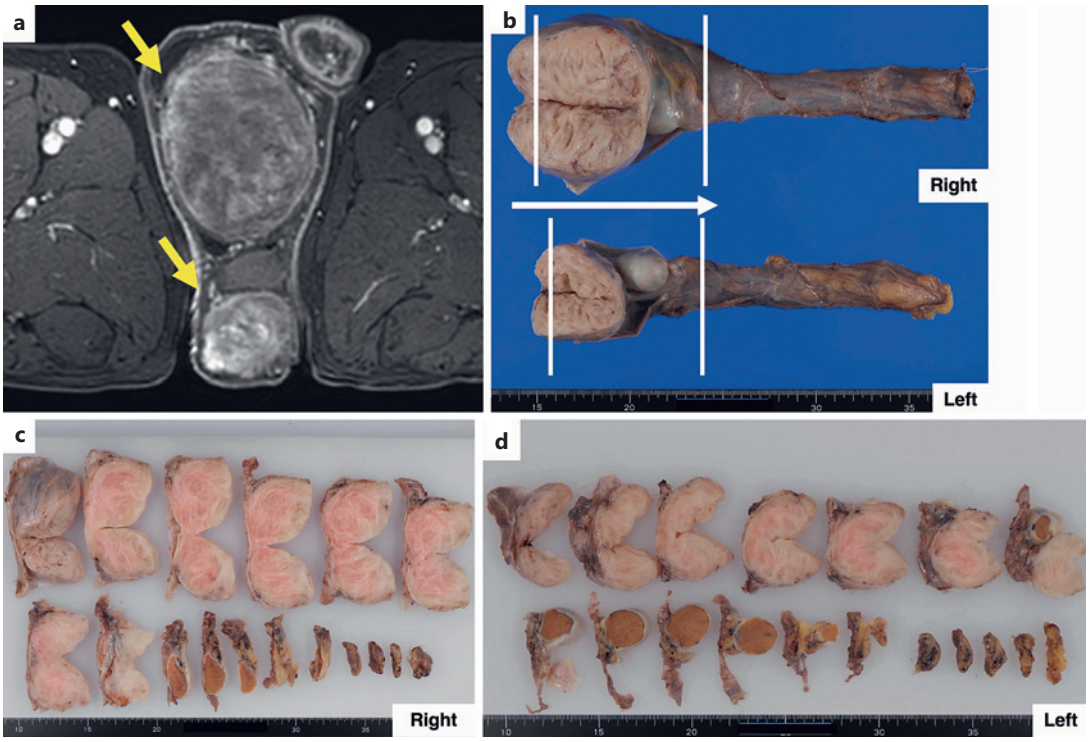

Fig. 1. Radiologic and gross findings. **a** A contrast-enhanced magnetic resonance imaging (MRI) scan showing well-circumscribed tumors up to 7.5 cm in the scrotum (yellow arrows). **b** The surgically resected specimen. The tumors were 9.0 × 8.0 × 5.5 cm (right) and 5.0 × 4.0 × 3.8 cm (left) in size. **c**, **d** Cut surface of the resected specimen. The tumors had a white whorled appearance and separate from the testes.

nonseminomatous germ cell tumors [10], another with liposarcoma [11], and another of metastatic renal clear cell carcinomas occurring in a patient with von Hippel–Lindau [12]. The most common benign tumors in the paratesticular regions contain adenomatoid tumors, leiomyomas, and lipomas, and adenomatoid tumors are the most frequent and leiomyomas are the second most common tumors [2]. In this case, these frequent tumors were suspected clinically. Leiomyomas in uterine corpus can harbor Mediator Complex Subunit 12 mutations, High-mobility group AT-hook (HMGA) 1 and HMGA2 rearrangements, Collagen Type IV (COL4) A5 and COL4A6 deletions, and fumarate hydratase mutations [13]. However, there are no reports of genetic abnormalities in BSPL. The bilateral occurrence of leiomyomas may indicate some genetic abnormality, but this is an issue for further investigation.

It is very challenging to distinguish leiomyomas affecting the testis from those affecting the paratesticular region by clinical examination alone [14]. However, no cases have been reported in which histological differentiation was a problem in previous reports. In general, leiomyomas present as bundles of tumor cells with spindle-shaped nuclei. Immunohistochemistry was positive for smooth muscle markers such as desmin or α-smooth muscle actin. The present case was a morphologically typical leiomyoma, and the immunohistochemistry was positive for α-smooth muscle actin.

In conclusion, we observed an extremely rare case of BSPL. Owing to this disease's rarity, no data on genetic mutations associated with BSPL or its prognosis is available. Therefore, when diagnosing a scrotal mass, attention to the position of the mass in relation to the testis will result in a more accurate diagnosis of BSPL.

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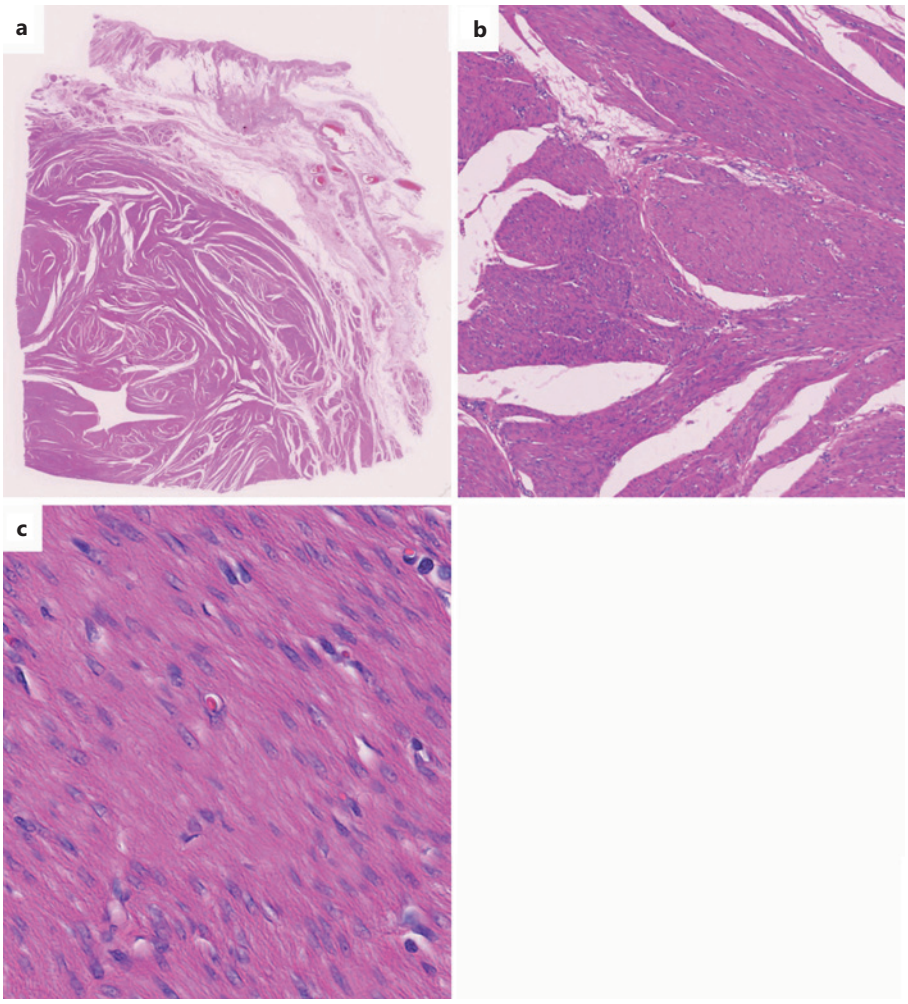

Fig. 2. Histopathological findings. Magnification: ×40 (**a**), ×100 (**b**), and ×400 (**c**). **a** The tumor formed a fascicular growth pattern with no necrosis or bleeding. **b** The tumors were formed by interlacing bundles separated by well-vascularized connective tissue. **c** The tumor cells contained oval to spindle nuclei and acidophilic cytoplasm.

Acknowledgments

The authors would like to thank the patients for allowing us to report their clinical information and data.

Statement of Ethics

This case report was conducted in accordance with the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. This retrospective review of patient data did not require ethical approval in accordance with the national guidelines.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

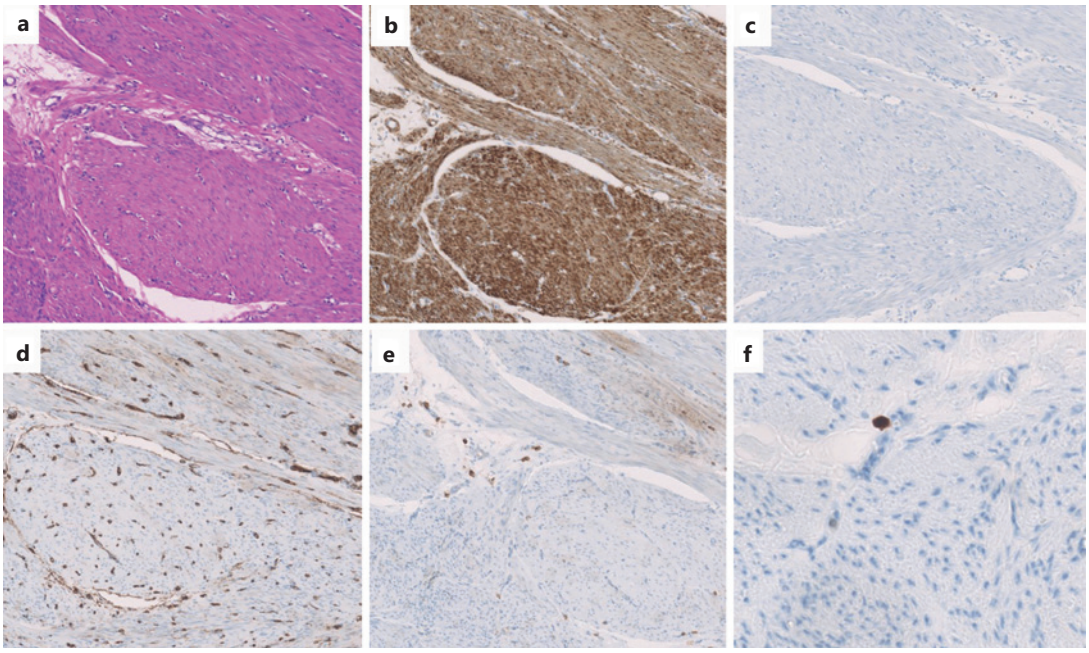

Fig. 3. Immunohistochemical findings. **a** HE. **b** αSMA. **c** S100a. **d** CD34. **e** c-kit. **f** Ki-67. Magnification: (**a**–**f**) ×400.

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Author Contributions

Akira Ishikawa: Diagnosis and preparation of the manuscript. Jun Shibata, Ryosuke Nobuhiro, Go Kobayashi, and Yoichi Saito: Management of tissue specimens. Hiroyuki Nose: Management of this case. Naohiro Uraoka and Naohide Oue: Diagnosis and correction of the manuscript.

Data Availability Statement

All the data in this study are included in this article. Further inquiries can be directed to the corresponding authors.

References

1 Billings SD, Folpe AL, Weiss SW. Do leiomyomas of deep soft tissue exist? An analysis of highly differentiated smooth muscle tumors of deep soft tissue supporting two distinct subtypes. Am J Surg Pathol. 2001 Sep;25(9):1134–42.
2 Lioe TF, Biggart JD. Tumours of the spermatic cord and paratesticular tissue. A clinicopathological study. Br J Urol. 1993 May;71(5):600–6.
3 Albert PS, Mininberg DT. Leiomyoma of the tunica albuginea. J Urol. 1972 May;107(5):869–71.

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4 Aus G, Boiesen PT. Bilateral leiomyoma of the tunica albuginea. Case report. Scand J Urol Nephrol. 1991;25(1):79–80.
5 Lia-Beng T, Wei-Wuang H, Biing-Rorn C, Chia-Chun T. Bilateral synchronous leiomyomas of the testicular tunica albuginea. A case report and review of the literature. Int Urol Nephrol. 1996;28(4):549–52.
6 Wang AX, Feng SL, Chang JW. Leiomyomas of the bilateral tunica albuginea of testes: a case report. Int J Clin Exp Pathol. 2015;8(8):9703–5.
7 Fernandez A, Krishnamoorthy S, Muralitharan S, Johnson T, Ramanan V. Bilateral synchronous paratesticular leiomyoma: a rare entity. J Clin Diagn Res. 2017 Apr;11(4):PD05–PD06.
8 Leonhardt WC, Gooding GA. Sonography of epididymal leiomyoma. Urology. 1993 Mar;41(3):262–4.
9 Siegal GP, Gaffey TA. Solitary leiomyomas arising from the tunica dartos scroti. J Urol. 1976 Jul;116(1):69–71.
10 Heidenreich A, Moul JW, Srivastava S, Engelmann UH. Synchronous bilateral testicular tumour: non-seminomatous germ cell tumours and contralateral benign tumours. Scand J Urol Nephrol. 1997 Aug;31(4):389–92.
11 Raza M, Vinay HG, Ali M, Siddesh G. Bilateral paratesticular liposarcoma - a rare case report. J Surg Tech Case Rep. 2014 Jan;6(1):15–7.
12 Moradi A, Farhoumand D, Bouzari B, Shakiba B. Simultaneous bilateral testicular metastases from renal clear cell carcinoma: a rare presentation in von hippel-lindau disease. J Kidney Cancer VHL. 2022;9(2):19–21.
13 Mehine M, Makinen N, Heinonen HR, Aaltonen LA, Vahteristo P. Genomics of uterine leiomyomas: insights from high-throughput sequencing. Fertil Steril. 2014 Sep;102(3):621–9.
14 Tammela TL, Karttunen TJ, Makarainen HP, Hellstrom PA, Mattila SI, Kontturi MJ. Intrascrotal adenomatoid tumors. J Urol. 1991 Jul;146(1):61–5.