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Bizarre Leiomyoma of the Scrotum

Scrotal leiomyomas with atypical bizarre nuclei are rare, which might be misdiagnosed as malignant tumor. We describe a case of scrotal bizarre leiomyoma in a 65-yr-old man. The tumor was a 1 cm-sized, well circumscribed, oval mass arising from the tunica dartos muscle. Histologically, it was formed by whorling bundles of fusiform cells with occasional atypical, pleomorphic nuclei and pseudoinclusions. Mitosis was not found. Although morphologically atypical, scrotal bizarre leiomyomas take on a biologic behavior not different from that of conventional leiomyoma, they should be distinguished from leiomyosarcoma to avoid unnecessary treatment.

Key Words : *Neoplasms, Muscle tissue; Leiomyoma; Scrotum; Tunica dartos*

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INTRODUCTION

Scrotal leiomyomas are uncommon tumors that arise from the subcutaneous tissue, or dartos muscle (1). The number of reported cases has recently been increasing (2). As in uterine leiomyomas, bizarre nuclei rarely occur in those of the genitourinary tract (3-8). Neither malignant transformation nor recurrences of scrotal bizarre leiomyoma has been reported. Awareness of scrotal leiomyoma with bizarre nuclei is critical not to misdiagnose as leiomyosarcoma because its surgical extent and the follow up are different.

CASE REPORT

A 65-yr-old man was admitted for transurethral resection of known urothelial carcinoma of the bladder. Physical examination revealed an elastic, firm, nontender mass of 1.0 cm in the greatest dimension in the left scrotum, and it was impervious to light. He was not aware of the mass before the physical examination. The serum level of some tumor markers such as alpha fetoprotein and human chorionic gonadotropin-beta was normal. Percutaneous mass excision was performed. It was easily dissected from the tunica dartos.

Grossly, the tumor was a well-circumscribed, 1.0 × 1.0 × 0.8 cm-sized, oval mass that originated from the tunica dartos, which was independent of the testis, epididymis, and funiculus spermaticus. Microscopically, it consisted of interlacing fascicles of spindle-shaped cells, and some of the tumor cells had pleomorphic nuclei and showed focally increased cellularity (Fig. 1). The nuclei were large and multilobulated with hyperchromatic chromatin and macronucleoli (Fig. 2A). Intranuclear cytoplasmic invagination producing eosinophilic pseudoinclusions was frequently observed (Fig. 2B). The tumor cells revealed no mitosis. Immunohistochemically, the tumor cells expressed vimentin (Zymed, San Francisco, CA, U.S.A., 1:50 dilution), desmin (Zymed, 1:50 dilution), and smooth muscle actin (Zymed, 1:100 dilution), but not cytokeratin (AE1/AE3, Zymed, 1:50 dilution), epithelial membrane antigen (Zymed, 1:50 dilution), HMB-45 (Zymed, 1:50 dilution), glial fibrillary acidic protein (Dako, Glostrup, Denmark, 1:350 dilution), or S-100 protein (Zymed, 1:100 dilution). The tumor cells showed a negative reaction for p53 protein (Zymed, 1:100 dilution), and less than 0.1% of the tumor cell nuclei showed Ki-67 labeling index (Zymed, 1:50 dilution). The tumor cells were stained red with Masson-Trichrome.

Resection of the bladder tumor by transurethral cystoscopy revealed noninvasive superficial papillary urothelial carcinoma (G1, Ta). The postoperative course was uneventful for seven months after the operation, and no recurrence has been recognized.

DISCUSSION

Scrotal bizarre leiomyomas have been rarely reported with various names, i.e. symplastic, pleomorphic, bizarre or atypical leiomyoma, and eight such cases could be retrieved in the world literature (3-8). All cases presented with an asymp-

tomatic, pedunculated tumor or ulcerative lesion. Scrotal leiomyomas are rare by themselves, and less than fifty cases have been reported (1, 2, 9). They rarely exhibit pleomorphic nuclear changes analogous to their uterine counterparts. In uterine leiomyoma, symplastic (atypical, bizarre, or pleomorphic) uterine leiomyoma is a term reserved for that with giant cells, nuclear atypism, and minimal mitotic activity (up to 10/10 high-power fields) (10). Eosinophilic cytoplasmic globules as observed in the present case which corresponded well to aggregates of intermediate filaments, are commonly not seen in leiomyomas with bizarre nuclei (5, 6). Atypical cells were suggested to have resulted as a consequence of synthetic progestin treatment like those in uterine leiomyomas. Unfortunately, there is no known relationship of atypical cells to preoperative hormonal treatment even in uterine leiomyomas. Bizarre cells are also regarded as degenerative changes, similar to that of ancient schwannoma, ancient melanocytic nevus, or some dermal tumors (11). In contrast to the smooth muscle tumors of the uterus, the histologic criteria for the diagnosis of scrotal leiomyosarcomas have not yet been established. In the review of Newman and Fletcher, the presence of any mitotic activity was advocated as the criterion of potential malignancy (8). In our opinion, bizarre leiomyoma rather than symplastic or atypical variant is the appropriate term for the scrotal smooth muscle tumors with bizarre nuclei because the former can imply such lesions regardless of mitotic activity.

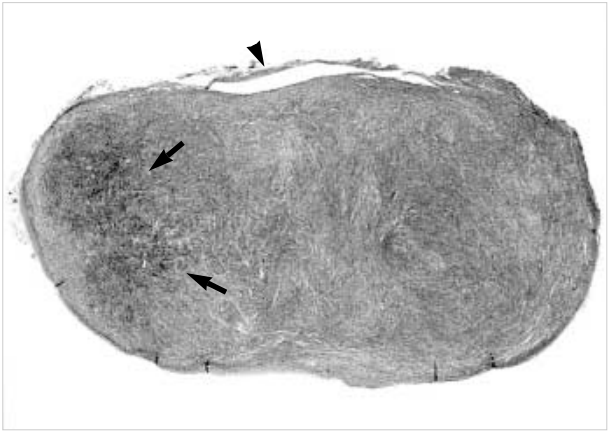

Fig. 1. A well-circumscribed oval mass is contiguous to the tunica dartos (arrowhead). Arrows indicate focal areas with increased cellularity (H&E, original magnification).

The smooth muscle of the tunica dartos is regarded as the cellular origin of scrotal leiomyoma. Clinically, scrotal leiomyoma appears as a small, firm, nontender, slowly growing mass. It may present as a pedunculated lesion with ulceration, needing to be differentiated from squamous carcinoma of the scrotum (12, 13). The painless nature of scrotal leiomyomas corresponds well to the slow growing tumor pushing the nerve trunks outward instead of compressing them. It occasionally presents with a large pedunculated mass or with an ulcerative mass (2).

Simple excision is indicated to achieve cure of scrotal bizarre leiomyoma. Irradiation should be avoided because radiation may induce malignant transformation of the tumor. Long-term urologic follow-up, however, is indicated because of the rarity of scrotal bizarre leiomyomas and lack of evidence whether the lesions are progressive or regressing. It is important that both pathologists and urologists should not equate

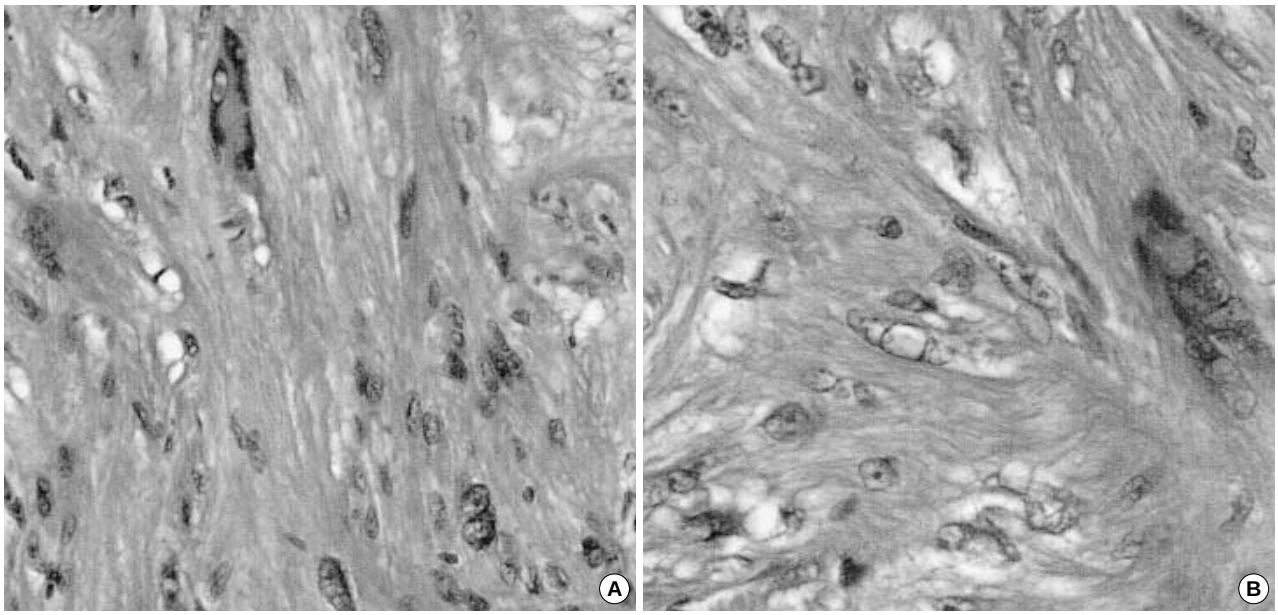

Fig. 2. (A) Tumor cells show enlarged nuclei and multinucleation with fibrillary cytoplasm (H&E, ×200). (B) Intranuclear eosinophilic pseudoinclusions and multinucleated giant cells are seen (H&E, ×400).

bizarre leiomyoma with leiomyosarcoma.

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