A Case Report of Atypical Scrotal Leiomyoma

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Key Words

Leiomyoma of the scrotum · Actin · S100

Abstract

Scrotal leiomyoma is a benign tumor derived from the scrotal dartos muscle; it is rare in clinical findings. The cause of the disease is not yet clear. Here, we present the case of a patient with no typical clinical features, but with some typical pathological changes. A middle-aged male patient saw a doctor for paroxysmal itching and burning of the scrotum. The physical examination revealed 3 × 5-cm², ill-defined, pink plaques, an uneven surface, and basal infiltration with pigmentation defects in the center. Pathology results showed dermal spindle cells in nodular hyperplasia. The nucleus was round at both ends, a perinuclear halo could be seen, and staining was positive for actin and negative for S100. Therefore, the diagnosis of scrotal myoma was made.

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Case Report

A 32-year-old man presented with a lump on the right side of his scrotum without any predisposing factors. The lump had been progressively growing in size over the last 3 years. Early on, there were 4–5 soybean-shaped, gray and black, flat wart bands with clustered distribution without exudation and desquamation; however, there was paroxysmal itching or the sensation of ants crawling on the skin. The patient often felt his skin was burning and tingling when the itching was very severe. He self-medicated with Compound Dexamethasone Acetate cream, Xuanshi solution, and other medications for external use. After such treatment, the itching eased but the rashes slowly expanded. After drinking or using hot water to clean the areas, the urticant ache became worse each time. He had used some medicine containing hydrochloric acid 1 year previously. Then, the topical skin developed

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ulcers. After the patient took cephalosporin antibiotics, the wound healed and the warts became flat, but the primary sense still remained. To confirm the diagnosis, he visited the hospital. There was no similar history or genetic diseases in his family.

A physical examination revealed no special abnormalities. A more in-depth examination revealed an approximately $3 \times 5\text{-cm}^2$, pink, ill-defined, hyperplastic plaque with an uneven surface, basal infiltration, sheet depigmentation in the center (fig. 1), and nearly superficial lymph nodes without palpable enlargement.

The skin biopsy showed dermal spindle cells in nodular hyperplasia. The nucleus was round at both ends, a perinuclear halo could be seen, and staining was positive for actin and negative for S100. The combination of HE morphology, immunohistochemistry, and pathology was in line with a diagnosis of scrotal leiomyoma (fig. 2, fig. 3, fig. 4).

Discussion

Leiomyomas are benign soft tissue neoplasms that arise from smooth muscle. In this case, the skin leiomyoma came from the hair erector muscles of the skin, dartos, and blood vessel walls of smooth muscle. This type of cancer is generally divided into 3 subtypes: vascular, genital, and hair leiomyoma. Genital leiomyoma involves the smooth muscle network from the deep dermis such as the scrotal dartos muscle [1]. Cutaneous leiomyomas account for approximately 5% of all leiomyomas, and genital smooth muscle tumors, such as scrotal leiomyoma, have an even lower clinical incidence rate. Europe, the United States, Japan, and some Southeast Asian countries have reported a few cases in recent years [2, 3]. So far, just 3 cases have been reported in our country.

The pathogenesis of the disease is not yet clear, though some scholars have proposed the disease may be related to the male hormone androgen, and experiments have confirmed that the lesions demonstrate androgen receptor expression [4]. Leiomyoma is known to occur in the uterus; however, levels of the female hormones estrogen and progesterone are closely related to scrotal leiomyoma in male endocrine and metabolic disorders, genetic mutation, or immune abnormalities. To confirm this, further study is needed.

The disease often occurs in white males aged 40–60 years [5]. Clinically, it often manifests as solitary tumors showing slow growth [6]. Usually, there are no early symptoms; however, over the course of time, the paroxysmal pain increases. Our patient, who had local lesions, experienced paroxysmal severe itching with burning, tingling, and, as mentioned in the previous literature reports, no pain or bursts of pain. The pain is thought to be due to the smooth muscle contraction, but the itching is difficult to explain. Pathology showed there was no obvious inflammatory cell infiltration.

The clinical manifestations of scrotal leiomyoma may not be characteristic of the disease, and in order to identify it, a variety of differential diagnoses are needed including dermatofibroma, schwannoma, neurofibroma, adnexal tumors, and metastases (table 1). The most important thing is to rely on the pathology-based and differential diagnoses. The histopathological features of the disease are the proliferation of tumor cells with characteristics of smooth muscles cells and actin-positive immunohistochemistry. Therefore, our patient's diagnosis was clear. Most leiomyoma lesions are benign and complete resection of the tumor, such as with Mohs micrographic surgical technique, rather than with radiotherapy treatment, is advised because radiotherapy treatment may induce malignant transformation [7].

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Table 1. Differential diagnoses

Differential diagnosis	Vulnerable population	Clinical manifestation/clinical feature	Histopathological findings
Neurofibroma	Higher prevalence in adults. Cases in children occur less frequently	A single, protruding, rounded, epidermal nodular or fusiform/both hard and soft in quality	S-shaped cell nucleus, thin fibers, rippled wall; S100 and neurofilament immunoreactivity are positive
Neurilemmoma	All ages and both genders can be affected	Often occurs in the limbs, especially on the side of the large nerve flexor/tumors are a scattered soft mass, usually without symptoms	Microscopically, the Schwann cells are arranged bundles or evacuation strips; can have a lobulated or vortex structure. S100 protein, corrugated fiber protein, and myelin basic protein-positive staining
Dermatofibroma	May occur at any age, but more commonly in young patients. Female patients outnumber males	In the upper torso, axilla, upper arm, or neck/brown or reddish skin papules or nodules Fitzpatrick's sign ('concave nest' or dimple sign)	Fasciculated spindle cells are plaque-like hyperplasia in intradermal, collagen fiber hyperplasia; fusiform eosinophilic cytoplasm lightly stained
Dermatofibrosarcoma protuberans	Common in middle-age, with a slight male proclivity	May occur on the chest; limbs common/raised, hardened lumps, on which there are multiple nodules; pink or purple. Lesions gradually increase and integrate, the surface is slightly glossy; slow growth	Spindle cells arranged in wheel- or spiral-shaped pattern, mild atypical changes, CD34 positive
Leiomyosarcoma	Most common in elderly patients, also occurs in the young. Rarely seen in children	The most common location is the retroperitoneal area. Larger tumors are more common in females/hardened nodules occur in the skin can be multilobed, often slightly elevated, can invade the dermis and cause atrophy and ulceration	The tumor is composed of spindle cells staggered into a bundle. Elongated, blunt-ended nuclei. Various cell sizes, a lot of cellular constituent, and nuclear mitosis
Smooth muscle hamartoma	Occurs at birth, in young children and adolescents	Occurs on the waist/in patches, up to a few cm, or can be raised with small follicular papules	Throughout the dermis and subcutaneous tissue there are numerous, scattered, straight and long smooth muscle fiber bundles, with two nearly rounded ends, consistent with the long axis of the muscle fibers and the long axis of the core, which interlace

Fig. 1. Pink, hyperplastic plaques, with an uneven surface, basal infiltration, and ill-defined, sheet depigmentation in the center.

Fig. 2. Spindle tumor cells of nodular hyperplasia, arranged in an interwoven pattern. HE. $\times 100$.

Fig. 3. Classic spindle-shaped tumor cells, abundant cytoplasm, and hyperplasia of fibroblasts in the dermis fibroblasts. HE. ×400.

Fig. 4. Actin-positive immunohistochemistry. ×100.