Urology Case Reports 2 (2014) 181–182

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Oncology

Case of Subcutaneous Leiomyosarcoma of the Scrotum Presenting as a Sebaceous Cyst in a 71-Year-old Man: A Case Report and Review of the Literature☆

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ARTICLE INFO

Article history:
Received 28 July 2014
Accepted 31 July 2014

Keywords:
Leiomyosarcoma of scrotum
Sebaceous cyst
Scrotal lesion
Biopsy

ABSTRACT

Leiomyosarcoma of the scrotum is a rare genital malignancy with approximately 35 reported cases in literature. We present a case of leiomyosarcoma of the scrotum in a 71-year-old man appearing as a sebaceous cyst that later developed ulcerations. However, because the irregular mass developed ulcerations, this should trigger one to consider that lesion is potentially malignant. The pathology report demonstrated malignant spindle cell neoplasm consistent with leiomyosarcoma, which tested positive for desmin and actin stains. On literature review, a study reported a 5-year survival rate of 50%-80%. The clinical features, diagnosis, histopathologic images, and treatment are reviewed.

Introduction

Leiomyosarcoma of the scrotum is viewed as a rare tumor. More than 95% of scrotal sarcomas arise from the spermatic cord, epididymis, or testes. However, sarcomas originating from the scrotal skin or subcutaneous layer are infrequent with about 37 cases reported. The clinical presentation involves a painless, firm, immobile, rubbery mass. We report a case of subcutaneous leiomyosarcoma which presented as a slow-growing scrotal lump initially appearing as sebaceous cyst, and a review of literature.

Case presentation

The patient is a 71-year-old previously healthy male who presents with a chief complaint of a scrotal lump and swelling without any worsening until a week before clinic visit, when the lesion started to bleed and ruptured. The slow-growing lesion was present for 10 years. He reported a slight pain on palpation without any radiation. He had no significant medical history. The skin surface showed a 1.2-cm tan-red exophytic hemorrhagic ulcerated lesion. There was no inguinal lymphadenopathy present.

After examination, the clinical impression was of a 3-cm sebaceous cyst. He was recommended to undergo excision of the lesion within the office setting. The patient was placed in the frog-leg position and was given 1% lidocaine for local anesthesia. An incision was made around the lesion subcutaneously and was removed for histologic evaluation. The pathology report demonstrated malignant spindle cell neoplasm consistent with leiomyosarcoma. The tissue tested positive for desmin and actin stains. Tumor necrosis and mitotic figures were presented to the peripheral and deep histologic margins. The patient later underwent wide excision with the conformation of negative margins.

Discussion

Soft tissue sarcomas comprise 1% of all malignancies and can originate from all types of soft tissue including muscles, tendons, fat, fibrous tissue, synovial tissue, vessels, and nerves. They can arise anywhere where soft tissue is present, with 60% present in the extremities, whereas 30% arise in the trunk, and finally remaining lesions arise in the head and neck. Leiomyosarcomas (tumor of smooth muscle) constitute 10%-20% of soft tissue sarcomas and most often develop in the uterus, gastrointestinal tract, and retroperitoneal region.

Leiomyosarcomas are divided into 2 subcategories depending on the location. Cutaneous leiomyosarcomas arise from the erector spinae muscle of the hair follicle or dartos muscle of the genital

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http://dx.doi.org/10.1016/j.eucr.2014.07.008

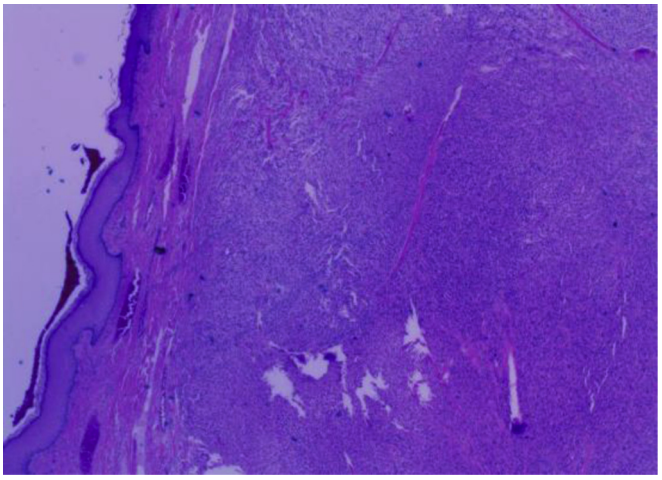

Figure 1. Normal skin is present on the left, whereas the tumor is comprised of a spindle cell proliferation forming rough bundles and fascicles located in the center with abundant brightly eosinophilic cytoplasm.

skin. Subcutaneous leiomyosarcoma is described as a smooth muscle tumor of the skin found in the muscle lining of arterioles and veins in the subcutaneous tissue. Subcutaneous leiomyosarcomas are found in 1%-2% of soft tissue malignancies. Within the adult population, leiomyosarcomas are the most common type, and are most commonly found in the kidney, bladder, or prostate. Cases of scrotal leiomyosarcoma are exceptionally rare, with approximately 37 cases reported to date.

The clinical presentation of scrotal leiomyosarcoma usually involves a painless scrotal lump present for several months to years with an age presentation of 51 years.[1] Physical examination exhibits firm, rubbery, irregular masses having similar features of a cystic lesion.[2-4] This case presented as a lesion that commonly would be diagnosed as a sebaceous cyst. However, because the irregular mass developed ulcerations, this should trigger one to consider that lesion is potentially malignant.[4]

Wide excision is the recommended treatment for localized leiomyosarcoma of the scrotum.[4] Once diagnosis was confirmed, a wide excision with negative margins was performed to complete treatment. The local recurrence-free interval at 5 years is higher

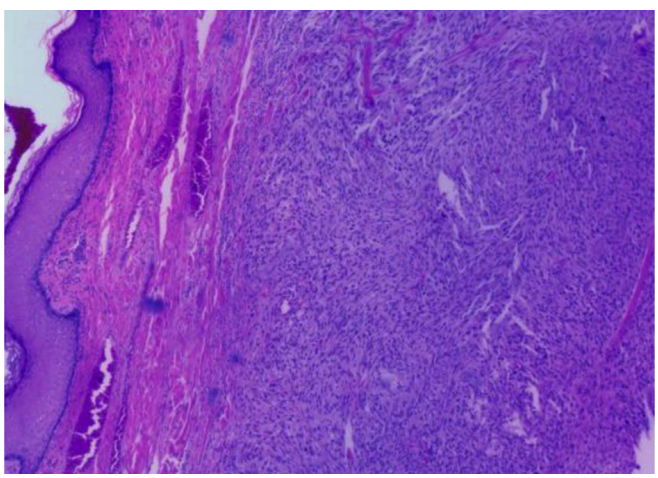

Figure 2. Center of the image demonstrates malignant smooth muscle cells with mitotic activity and nuclear atypia with normal skin on the left

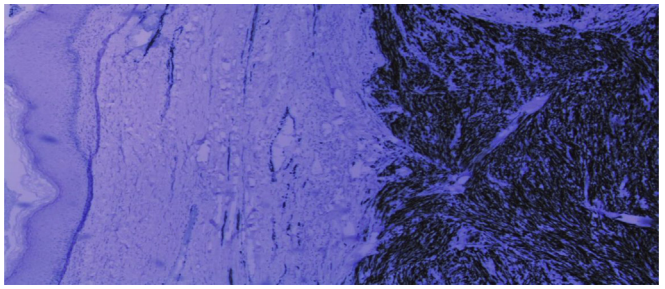

Figure 3. Immunohistochemistry study demonstrates positive tissue staining for desmin on the right which marks the smooth muscle.

with a margin >10 mm (84%) than that of 0 mm (58%) or 1-9 mm (58%).[5] However, for patients who refuse surgery, chemotherapy is a conceivable option. This would include gemcitabine, paclitaxel, and vincristine. In addition, doxorubicin-D, has been used with limited success.

On literature review, long-term follow-up is recommended due reported late local reoccurrence.[3,5] Fagundes et al[6] reported a 5-year survival rate of 50%-80%, perhaps attributing the advances in diagnosis and management of these tumors. Adjuvant therapy can be used to avoid reoccurrence and incomplete resection margin.[7] There have been reported metastases to the soft tissues of the neck[1] and lungs.[8] Literature reported cases of radiotherapy and chemotherapy used as adjuvant therapy, but a universal agreement for their uses is nonexistent. There is, however, increasing agreement that all leiomyosarcomas regardless of grade and histology should receive adjuvant radiotherapy.[9-10] Several studies have reported favorable results with combined surgery and adjuvant radiotherapy with reduced rates of reoccurrence by 10%-20%.[9,10]

Conclusion

High degree of suspicion should arise when a common diagnosis presents atypically. This case demonstrates how a leiomyosarcoma can mimic a common diagnosis—a sebaceous cyst. Atypical findings such as ulceration should alert the clinician to anticipate other diagnosis and institute the appropriate medical management.

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