Case Report

Leiomyoma of deep soft tissue mimicking calcific myonecrosis

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Abstract

Leiomyoma of deep soft tissue is an unusual entity reported in less than 60 cases in the English literature. The lesion is now accepted as a rare neoplasm. Leiomyomata of deep soft tissue have been divided into 2 groups, those occurring primarily in women in the retroperitoneum, histologically similar to leiomyomata of the uterus, and those occurring equally in both sexes in the somatic deep soft tissues. Irrespective of location, these lesions can demonstrate calcification, and even less commonly ossification. We report a unique case of a leiomyoma of deep soft tissue that mimicked the clinical and radiographic features of calcific myonecrosis, also a rare mass forming soft-tissue lesion. Clinical and radiographic information are often critical in the diagnosis of soft-tissue lesions; however, this case demonstrates that a lesion can deviate from the standard clinical and radiographic interpretations most commonly attributed to it.

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Introduction

Leiomyoma of deep soft tissue is a rare neoplasm characterized in a series of 36 cases by Billings et al. [1] in 2001. While the existence of this lesion was previously questioned, it is now widely accepted that such neoplasms do exist [1,2]. This neoplasm is thought to arise in 1 of 2 scenarios: those that arise in the pelvic retroperitoneum, are more common in women, and are histologically similar to uterine leiomyomata, and those that arise in the somatic deep soft tissues and present with equal frequency in men and women [1,2]. Regardless of the site of presentation, these tumors are governed by strict diagnostic criteria. Atypia and mitotic activity are absent to minimal, and seen in less than 1 of 50 high-power fields [1]. Often, the somatic lesions demonstrate calcification, most commonly in the form of psamomma bodies. Ossification can also be seen [3]. There is no relationship to trauma. Many of the somatic lesions are identified

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incidentally, while at least some of the retroperitoneal lesions have been identified in women with a history of uterine leiomyomata [1,2]. Herein, we report a unique case of a leiomyoma of deep soft tissue in a female patient, whose clinical and radiographic presentation mimicked that of calcific myonecrosis, demonstrating that a rare lesion can deviate from the clinical and radiologic presentation that has been established.

Case report

A 61-year-old woman presented to the orthopedic clinic with a chief complaint of right buttock and hip pain. She sustained a fall to the area 6 years earlier and appeared to develop a hematoma. According to the patient, there was significant acute pain which resolved. One year before presentation, she experienced an exacerbation of the pain, and 6 months before surgery, the pain had become unbearable, limiting her activities of daily living. On examination, an ill-defined mass in the left thigh was present, was noted to be very firm, and was felt to be calcified. A computed tomography scan of the right hip and proximal femur demonstrated a well-marginated soft-tissue mass. The lesion appeared to be located within the gluteus maximus muscle, in close proximity to the sciatic nerve, measuring 11.4 cm in greatest dimension. The entire lesion was heavily calcified (Fig. 1A). The differential diagnosis at this time included several metabolic disorders and dystrophic calcification secondary to prior trauma.

The patient was seen by the musculoskeletal oncology service. At that time, the patient provided additional pertinent history. Over the past year, the patient noticed that the swelling in the affected area increased, as did the pain and discomfort while seated, standing for prolonged periods, and lying on the affected side. While she denied fever, chills, and night sweats, she did report possible weight loss. Her medical history was significant only for hypertension.

Physical examination was significant for a large firm mass in the center of the right gluteus muscle approximately 15 cm in greatest dimension which was tender to palpation particularly on the medial aspect of the thigh. No erythema, induration, or skin defects were noted. No neuromuscular defects were noted about the extremity. No deficits in range of motion were noted. On review of the axial computed tomography, no nerve impingement was seen (Fig. 1B). The patient returned 1 month later with no relief of symptoms. A complete metabolic workup was performed, and no abnormal results were identified. A plain x-ray demonstrated the same mass, which was essentially unchanged from the prior radiologic examination (Figs 2A and B). At this time, the radiographic images were felt to be most consistent with calcific myonecrosis.

The patient underwent resection of the lesion. The submitted specimen was an 11-cm ovoid, well-circumscribed mass with a tan fibroconnective tissue capsule. The specimen was bisected, revealing calcified lobulations with focal areas of tan to white whorled surfaces (Fig. 3). Routine hematoxylin and eosin sections revealed diffuse area of dystrophic calcification and hyalinization (Fig. 4A) with adjacent areas of mature appearing smooth muscle cells (elongated nuclei with blunt ends) with abundant eosinophilic cytoplasm. No atypia or mitoses were identified. Numerous psammoma bodies were identified within the neoplastic smooth muscle areas (Fig. 4B). The smooth muscle component was confirmed with smooth muscle actin and desmin staining. The diagnosis of leiomyoma of deep soft tissue was rendered. At last clinical follow-up (2 months after surgical excision), the patient was pain free and had returned to her prior level of function.

Discussion

Leiomyomata, benign smooth muscle tumors, are seen most commonly in the uterus and skin. They are rarely identified in
Leiomyomata of the deep soft tissue of the somatic type have been described previously in case reports [3–16] and a series of 36 consultation cases reported by Billings et al. [1] in 2001. Billings et al. established the 2 histologic types, somatic soft tissue and retroperitoneal [1]. Because leiomyosarcomas of the deep soft tissue are far more common than leiomyomata of deep soft tissue, strict diagnostic criteria must be observed [2]. These criteria include absence of atypia, no to minimal mitotic activity, and absence of necrosis. Leiomyoma of deep soft tissue is amenable to surgical excision, and most cases do not recur even in the presence of positive margins [1]. The histologic differential diagnosis includes soft-tissue lesions that are known to occur in the deep soft tissue and include leiomyosarcoma and schwannoma. S-100 immunohistochemical staining is sufficient to exclude a schwannoma as positivity would support the neural differentiation of the lesion. Smooth muscle actin or desmin staining can be added to support or refute the presence of smooth muscle, as was done in our case. The histologic characteristics of leiomyosarcoma, including mitoses, atypia, and necrosis, are generally enough to differentiate this lesion from leiomyoma of deep soft tissue.

Leiomyomata of the deep soft tissue are known to calcify, and even ossify [1,3], but none in the literature have a history of preceding trauma. In fact, the history given by the patient in combination with the radiographic findings in this case would lead to a suspicion for calcific myonecrosis. Calcific myonecrosis is often associated with a traumatic event, usually years before presentation [17]. This lesion is as rare as leiomyoma of deep soft tissue. It was originally described by Gallie and Thomson [18] more than a half century ago and is characterized by the replacement of skeletal muscle in one or more compartments with a fusiform mass demonstrating dystrophic calcification and in some cases central liquefaction [17,19].

Typically, radiographs of calcific myonecrosis demonstrate a fusiform mass within skeletal muscle that is heavily mineralized. Histologically, the lesions demonstrate infarcted skeletal muscle with cholesterol clefts, hemosiderin deposition, and extensive dystrophic calcification [20]. No smooth muscle component is identified as part of calcific myonecrosis.

This leads to an interesting diagnostic quandary. The lesion in this case is clearly one of smooth muscle, and yet, clinically and radiologically, it had all the features of calcific myonecrosis. It is possible, and most likely, that the lesion was present at the time of the trauma in 2004. In our case, it
appears that a leiomyoma of deep soft tissue may have undergone the changes of calcific myonecrosis without the above described histologic features of calcific myonecrosis. It seems then, that leiomyomata of deep soft tissue may undergo secondary changes which can make the clinical and radiologic diagnosis difficult to entertain, if not impossible. In conclusion, leiomyoma of deep soft tissue should be considered in the differential diagnosis of any deep soft-tissue lesion that is extensively calcified despite a history of trauma sustained to the area. Leiomyoma of deep soft tissue can undergo changes similar to that of calcific myonecrosis. Pathologists should be aware of this possibility when confronted with such a lesion in the diagnostic setting.

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