Case report

Synovial Sarcoma of the foot enlightening etiology: a case report

Nanze Yu[1], Feng Cheng[2], Luwei Xiao[1], Peijian Tong[3], Changxing Wang[2]

[1] Institution of Orthopedics and Traumatology, Zhejiang Chinese Medical University, Hang Zhou, 310053, P.R.China; [2] Education and Research Institution of Orthopedics and Traumatology of Integrated Traditional Chinese Medicine and Western Medicine, the Second Affiliated Hospital of Zhejiang Chinese Medical University, Hang Zhou, 310005, P.R.China; [3] Department of Orthopedics and Traumatology, the First Affiliated Hospital of Zhejiang Chinese Medical University, Hang Zhou, 310006, P.R.China

Abstract. Synovial sarcoma which mainly affects the periarticular tissues of the extremities in young adults is an uncommon seen soft tissue sarcoma with uncertain causative factors. We report a long-course primary synovial sarcoma of the left foot in a 46-year-old man. The initial symptom occurred when he was 26-year-old. During these 20 years, he underwent two operations, with the diagnosis of synovial cyst and nodular synovitis. This time, a lobulated 7× 5.5 × 6 cm mass was found on computed tomography, and a final diagnosis of synovial sarcoma was made by histological study. The patient underwent a below knee amputation. We report this case aiming to discuss about the potential etiology of synovial sarcoma. Physicians should be aware of the possibility of this malignant mass. (www.actabiomedica.it)

Key words: synovial sarcoma, etiology, case report

Introduction

Synovial sarcoma is a rare malignancy with an annual incidence of 2.57 per 100,000 pts. According to a 1982 to 1997 database of 3400 sarcoma patients from the Memorial Sloan-Kettering cancer center, synovial sarcoma accounts for about 13% of all soft tissue sarcomas in the extremities (1). Unlike the other soft tissue sarcomas, synovial sarcoma is always seen in young patients ageing from 15 to 40, with a slightly male dominance. As it was firstly reported in 1865 (2), synovial sarcoma occurs next to the large proximal joints, while the head and neck region, lung, pleura, mediastinum, cardiac tissue, kidney, retroperitoneum, skin, abdominal wall, chest wall, and bone marrow, etc were also reported as involved (3).

The etiology of synovial sarcoma still remains unclear. We present a case with an extremely prolonged course of synovial sarcoma and discuss about the etiology in the purpose of highlighting the potential occurrence of it.

Case Report

A 46-year-old man presented to our hospital with pain and swelling of the left medial foot. When he was 26-year-old, he had a sudden pain of the left foot with no apparent cause. The pain, always occurring when get cold, could be relieved by socking with hot water. Five years later, the patient went to a local clinic since the pain became severer and occurred more frequently. He was treated with ibuprofen, and the pain relieved. At the age of 43-year-old, he had a surgical excision with a diagnosis of synovial cyst because the symptom couldn't be controlled by drugs.

One year later, as a result of the recurrence of the pain, he underwent another surgical excision, and the pathological report came as nodular synovitis. But the symptom was not eliminated,and it repeatedly occurred after the second operation. It was described that the foot got enlarged and swollen gradually with an unendurable pain while weight-bearing in the last few months before he was admitted to our hospital.

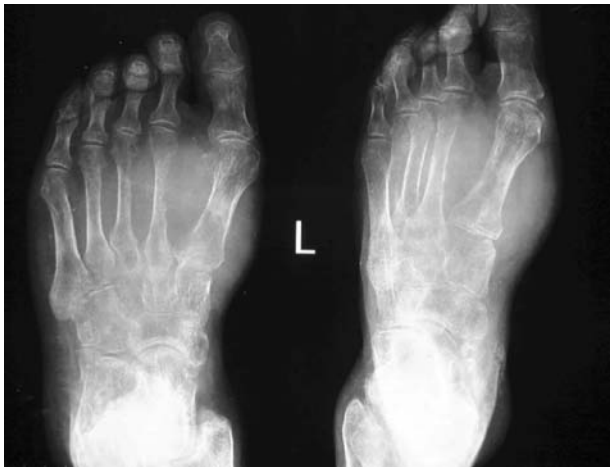

Figure 1. Dorsoplantar and medial oblique radiographs of the left forefoot show bone erosion and periosteal reaction of the medial aspect of the second metatarsal shaft and the proximal medullary canal of the first metatarsal shaft; no obvious calcification in soft tissues.

At physical examination on admission, a tender, circumscribed and little hard mass was found in the medial side of the swelling left foot. The skin was not involved, and no distention of the veins was observed. Toes of the left foot were of normal blood supply and sensation, while impaired activity, and no enlarged local lymph nodes were found. Radiograph revealed bone erosion and periosteal reaction of the medial aspect of the second metatarsal bone and the proximal aspect of the first metatarsal bone (Figure 1). No evident abnormalities were found by chest radiograph. An incision biopsy was performed. MRI of the right foot was performed, and a large lobulated soft tissue mass (7× 5.5 × 6 cm) was identified. It straddled from the first to the forth metatarsals. (Figure 2). The histology was consistent with synovial sarcoma (Figure 3). The patient underwent a below knee amputation of the left foot. Postoperatively, he received adjuvant chemotherapy with adriamycin and ifosfamide. No metastases nor recurrence were found at 6 months after operation, and he will continue to be monitored.

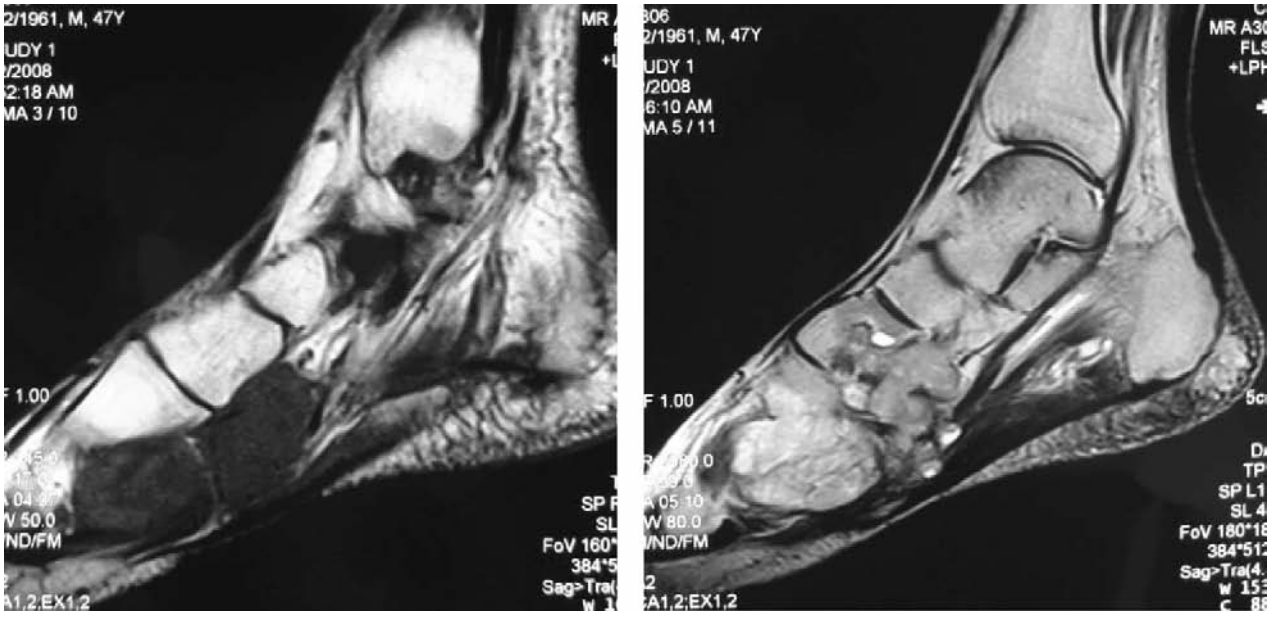

Figure 2. T1-weighted MRI slice shows a low signal intensity mass; T2-weighted MRI slice shows the lobulated mass with high signal intensity and obscure boundary.

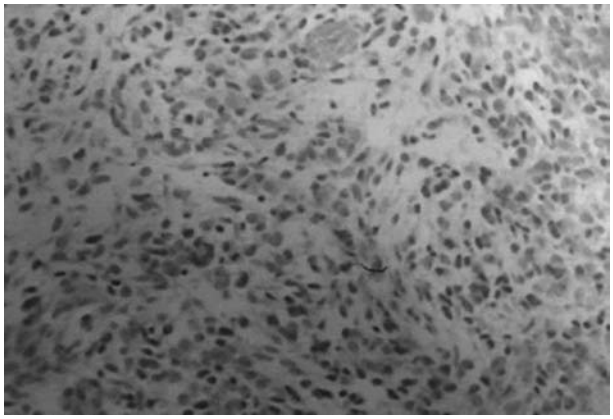

Figure 3. Macroscopic photograph shows the sarcoma

Discussion

Synovial sarcoma is a relatively form of soft tissue sarcomas. Though called "synovial sarcoma", it is not derived from synovial tissues, and seldom occurs intra-articularly. Immunochemistry and ultrastructural researches reveal that synovial sarcoma derives from as yet unknown multipotent stem cells capable of differentiating into mesenchymal and/or epithelial structures and lack of synovial differentiation (4). Synovial sarcoma may be histologically classified into 4 types as biphasic, monophasic spindle-cell, monophasic epithelioid, and poorly differentiated types. The biphasic and monophasic spindle-cell types are dominant, and the poorly differentiated type shows worst prognosis.

It has been clearly demonstrated from cytogenetic and molecular genetic aspects that translocation of t(X;18) is exclusively associated with synovial sarcoma (4), while the etiology still remains unknown.

Most patients had an experience of trauma before the diagnosis of synovial sarcoma was made, though the connection between them has not been definitely identified. Sisir et al. (5) reported that trauma is rather a coincidence and not a causative factor, even though the lesions are more often found in the lower extremities, and trauma probably causes hemorrhage in the lesion, making it larger and therefore more noticeable to the patient. In a retrospective study (6), 71.4% (10/14) of all patients had a history of trauma, including torsional injuries and blunt trauma. The time from trauma to tumor diagnosis was between 3 and 9 months. In our case, though the patient denied trauma history, there were surgical excisions 1 and 3 years before admission which was in accordance with the clinical history. Thus, a possibility of trauma reasoned tumor can not be denied.

Synovial sarcoma always presents in adolescents or adults younger than 40-year-old. Most commonly, it starts with a slowly enlarging deep-seated painful mass. In our case, the initial onset age (26 yrs.) was in a high incidence age group, though no definite diagnosis was then made. Local recurrence of synovial sarcoma is common, usually within 2 years from surgery, but it can be delayed for up to 10 years (3). Our patient had a surgical excision 3 years before admission, which may be considered as a local resection of preexisting tumor tissue. No routine chemotherapy or postoperative radiotherapy were administered, and the symptom recurred in a year which was in accordance with the reported clinical history. Thus, a possibility of prolonged existing tumor may not be denied.

As far as we know, no case of synovial sarcoma accompanied by a synovial cyst or nodular synovitis history has been observed, nor a case of canceration of synovial cyst or nodular synovitis. In our case, the patient showed a course as long as 20 years during which the cyst or inflammation may persistently exist as a physical or inflammatory stimulation. Though no relationship may be identified, a possibility of prolonged existing stimulation as a causative factor can not be denied.

Synovial sarcoma presents an unfavorable prognosis, the estimated 10-year survival of patients younger than 20 years is about 90%, whereas it is only 25% in patients older than 40 years (1). Because of its rare occurrence, insidious position and atypical symptom, misdiagnosis and delayed diagnosis are not uncommon. Synovial sarcoma of foot diagnosed as acute gouty arthritis (3), Morton's neuroma (7), trapped nerve and tarsal tunnel syndrome (8) were reported. A definite diagnosis relies on biopsy: it is important to highlight the possibility of synovial sarcoma, especially in patients with a history of trauma, operation, other synovial lesions, etc. An early diagnosis and treatment may result in improved prognosis.

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Correspondence: Nanze Yu
E-mail: yunanze@gmail.com
Tel. +086 13967191400