

## CASE REPORT

## Multiple Fibromas of Tendon Sheath: Unusual Presentation

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Fibroma of the tendon sheath is an uncommon soft tissue tumor presenting as a solitary, slow-growing, firm, painless, small nodule, which shows strong attachment to the tendon or tendon sheath. It is usually localized on fingers and hand tendons in adults between the age of 20 and 40 years old. This case concerns a 61-year-old man presenting with a 5-year history of multiple cutaneous nodules on both palms and soles. Skin biopsy confirmed fibroma of the tendon sheath. Blood tests showed a high titer of rheumatoid factor and positivity to anti-nuclear antibody. No case of fibroma of the tendon sheath occurring multifocally on both palms and soles has been previously reported. Herein, we report on a very rare case of multiple fibromas of the tendon sheath arising from palms and soles, which supports the pathogenetic hypothesis that this tumor may be a reactive process rather than a true neoplasm. (*Ann Dermatol* 23(S1) S45~S47, 2011)

**-Keywords-**

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### INTRODUCTION

Fibroma of the tendon sheath (FTS) is a rare, benign soft

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tissue tumor<sup>1</sup>. The tumor usually presents as a single, slow-growing, firm, painless, small nodule, which strongly attaches to the tendon or tendon sheath and it is often localized on the hand, particularly on the thumb<sup>1,2</sup>. It usually occurs in males between the age of 20 and 40 years old<sup>1,2</sup>. Histopathologically, it is characterized by a well-demarcated nodule that consists of haphazardly-arranged, fibroblast-like spindle cells embedded in a dense collagenous matrix<sup>2</sup>.

A few cases of FTS have been described in the literature<sup>1-7</sup>. However, multiple FTSs arising concurrently on both the palms and soles has not yet been reported<sup>4,6</sup>. Herein, we report on the unusual presentation of multiple FTSs.

### CASE REPORT

A 61-year-old man presented with approximately a five-year history of multiple scattered non-tender palpable firm indolent subcutaneous nodules on both palms and



**Fig. 1.** Multiple non-movable firm indolent deep seated nodules on the left palm (see arrows).

soles. He recognized the initial skin lesion on his right palmar area after he felt discomfort and pain upon grabbing an object. The lesions have increased in size and number over the last four years on both palms, and, recently, the same multiple lesions appeared on the soles. He complained from time to time of morning stiffness and numbness on his hands and feet. There was no recollection of associated trauma. His family history and past medical history were unremarkable.

Physical examination revealed that the number of nodules was approximately ten, which were located mainly in the center of both palms and soles, and these ranged in size from approximately 0.5 cm to 1.5 cm in diameter (Fig. 1). As the nodules of the soles were deeply located, they were not visible, but palpable. No limitation of hand or finger motion was observed. No joint swelling or other specific skin lesions were found.

Laboratory tests, including blood cell count and blood chemistry, were all within normal ranges. However, a rheumatologic evaluation yielded a positive antinuclear antibody (homogeneous pattern) and positive serum rheumatoid factor with a titer of 5+ (1:16) (positive range 1+ ~5+). Hand X-ray showed no remarkable findings, except for mild osteoarthritic changes in distal interphalangeal joints. For histological diagnosis, a 4 mm punch biopsy was performed from one of the lesions on the left palm. Histopathological findings showed relatively well-demarcated cellular proliferation, including slit-like vascular channels and haphazardly-arranged, spindle-shaped fibroblasts located among the dense collagenous matrix (Fig. 2).

Based on these clinical and histological findings, he was

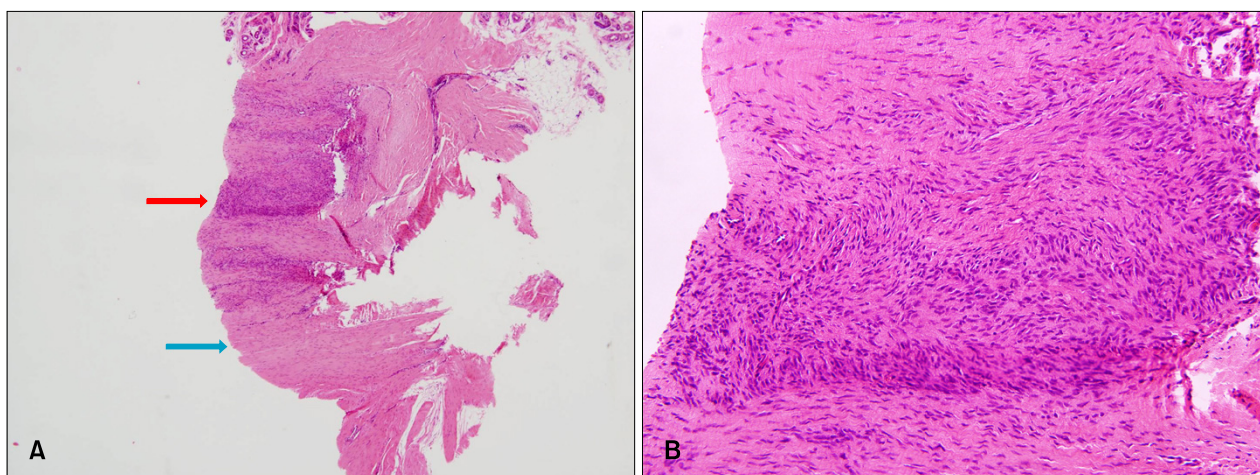
diagnosed as multiple FTSs. The patient did not want to be referred to a hand and foot surgeon for surgical excision. He has been followed up for almost 1 year and his pain and numbness have been treated with oral anti-inflammatory agents (aceclofenac). The number of FTSs has since remained stationary.

## DISCUSSION

Chung and Enzinger first defined FTS as an entity in 1979<sup>11</sup>. This rare tumor has been reported mainly in the orthopedic field and generally occurred as a solitary nodule on the fingers, feet, elbows, and knees, and, rarely, intra-articular areas<sup>1-7</sup>.

This case may be very unique in that FTS occurred multifocally and concurrently on the palms and soles. The patient complained of rheumatic symptoms, including morning stiffness, and his blood test showed a high titer of rheumatoid factor and a positive result for anti-nuclear antibody. However, X-ray findings and other physical examination did not fit into the Revised Criteria for Rheumatic Arthritis by the American College<sup>8</sup>. He has shown only mild osteoarthritic changes in distal interphalangeal joints of both hands, but not in proximal interphalangeal or metacarpophalangeal joints, which were the favorite involved site in rheumatoid arthritis. Joint damage in rheumatoid arthritis usually occurs within the first 2 years<sup>9</sup>. Although he presented with 5 years of long standing history, the patient did not show any definite rheumatoid arthritic pathophysiological findings in X-rays, except for serum positivity and morning stiffness.

The pathogenesis of FTS has not been clearly established



**Fig. 2.** (A) The relative well-demarcated tumor composed of hypocellular hyalinized collagenous areas (blue arrow) and hypercellular densely arranged collagenous areas in the dermis (red arrow) (H&E, ×40). (B) Scattered spindle cells are embedded in a dense collagenous matrix with a slit-like vascular space (H&E, ×100).

with regard to whether the origin is a neoplasm or reactive fibrosing process. Dal Cin et al.<sup>10</sup> reported that the presence of clonal chromosomal abnormality characterized by a t(2:11)(q31-32;q12) in ten out of 20 karyotyped cells suggested that this proliferation is not a reactive fibrosing process, but a neoplasm. Others have found that the right hand was more frequently affected than the left, and most cases occurred in the palm of hand and in the plantar region of the foot<sup>11</sup>. This finding suggests that the origin of FTS may be a reactive process by trauma, stimulation, or inflammation. This case also favored the reactive pathogenesis in formation of FTS. Skin lesions developed on palms and soles and are consistently affected by prolonged pressure and motion. Seropositivity for rheumatoid factor may indicate prolonged inflammation, which could be associated with multiplicity in this patient. His symptoms of morning stiffness and numbness showed moderate improvement with administration of oral anti-inflammatory agents. Since we placed him on oral anti-inflammatory agents, the number and size of FTSs have been maintained. From these findings, sustained inflammation and stimulation may play an important role in FTS and its multiplicity.

The majority of patients with FTS are between the ages of 20 and 40 years and the male: female ratio has been described as 1.5~3:1<sup>7,11</sup>. Most patients do not complain of any symptoms. However, 31% of cases present with tenderness and mild pain due to compression of nerves underlying FTS<sup>11</sup>. Numbness and morning stiffness were observed in this case also. Although his symptoms were controlled by oral anti-inflammatory agents, the possibility of compression of nerve on palms and soles cannot be excluded. Therefore, even though it is practically difficult to excise out all FTSs, removal of the tumor is necessary, which provokes pain. Surgery for local excision should be performed carefully, because the recurrence rate is 24% and all of the cases are in the hands and finger<sup>7</sup>.

Differential diagnosis should be made with an epidermal cyst, mucinous cyst, neuroma, leiomyoma, nodular fasciitis, and giant cell tumor of the tendon sheath (GCTTS)<sup>12</sup>. In particular, clinical features of GCTTS are similar to those of FTS. However, FTS is distinguished from GCTTS by histopathologic features, which include the fact that GCTTS are less hyalinized and more cellular, and with histiocytes and monocytes as well as multinucleated giant cells, foam cells, and hemosiderin-laden macrophages<sup>10,12</sup>. Regarding multiple nodules on the palmar area, Dupuytren's contracture should be considered as a differential diagnosis. It is the best known multiple palmar fibroma-

tosis<sup>13</sup>. Clinical manifestation usually showed flexural contracture of the hand, particularly the ring and little finger area. This patient did not show any limitation of finer or hand movement; therefore, diagnosis of Dupuytren's contracture was easily ruled out in the clinical setting.

We herein report on a very rare case of multiple FTSs on the palms and soles with a highly elevated serum rheumatoid factor. This case implies that FTS may not be a true neoplasm but a reactive process provoked by sustained inflammation and stimulation. FTS should be considered in clinical differential diagnosis of multiple nodules on the palm and/or the sole.

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