CASE REPORT

Toe Deformity after Pregnancy due to Fibroma of Tendon Sheath: A Case Report and Review of Literature

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Introduction

Fibroma of the tendon sheath (FTS) is a rare benign soft tissue tumor which mostly occurs in fingers, hands, and wrists¹⁻⁵. This type of tumor is characterized by a slowly growing pathologic feature and made up of scattered fibroblasts in a dense stroma with slit-like vascular channels⁶. Clinically, FTS affects more men than women especially between the ages of 30 and 50 years. In 1979, Chung and Enzinger⁵ presented 138 cases of FTS, out of which only six cases were in the feet. Moreover, FTS occurring in the toe were extremely rare, to the best of our knowledge, no more than five cases have been reported. Most patients with FTS present with mild pain, nodules, even symptoms due to nerve involved or intra-articular compression, whereas, few cases have presented with deformity. Yamamoto *et al.*⁷ reported the first case presenting as a toe deformity due to a fibroma of the tendon-sheath in an 83-year-old man who had a history of trauma. A history of trauma was supposed to be related to occurrence of FTS in 10% of patients. However, here we present a case of FTS developed after pregnancy with no traumatic history. This situation is extremely rare and has not been reported before in the literature.

Case Report

A 26-year-old woman presented with more than one year history of a non-tender palpable indolent subcutaneous mass in the third toe with concomitant short deformity and swelling. There was no history of trauma and it was not until the two months after her pregnancy that she recognized the initial lesion in her third toe without any pain and discomfort. No close attention was given according to her nontypical symptoms and pregnant condition. The nodule increased in size over the course of a year with the deformity becoming more obvious. From time to time, she complained of limita-

tion of motion in the third toe and obvious deformity. Four months after childbirth, the patient consulted an orthopedist and was referred to our hospital.

Physical examination revealed that a mass approximately 2 cm × 2 cm in diameter was located at the plantar basilar part of the third toe in the left foot (Fig. 1). The mass was irregularly shaped, indolent, and painless. The third toe was slightly crisped and obviously swollen with a limited range of motion. No other specific swelling, skin lesions, or limitation of motion were found in the foot and ankle.

The patient underwent laboratory tests including blood cell count, blood chemistry, and hormone test, all of which were within normal ranges. Radiographic examination showed no remarkable abnormal findings except for swelling of the third toe in the left foot (Fig. 2). Ultrasound examination revealed a 10 mm × 7 mm low echogenic area with obscure boundary and no envelope, inside which was no bloodstream signal. The original diagnosis was suspected tendinous synovitis, giant cell tumor of tendon sheath (GCTTS), desmoid tumor, pigmented villonodular synovitis (PVNS), and FTS. In order to work out the correct diagnosis, a biopsy, the gold standard of diagnosis, was taken.

The mass was excised with a marginal margin with no injury to the tendon. On gross examination of pathologic findings, the tumor was rubbery, irregular shaped, ivory white, and well circumscribed, measuring 2.3 cm × 2 cm × 0.8 cm (Fig. 3). It was attached to the tendon sheath. Sections of the tumor showed rubbery and ivory white tissues with no cartilaginous and calcified areas found. Microscopically, the tumor was demarcated and mainly composed of spindle-shaped fibroblast-like cells and rich collagenous stroma with dense eosinophilic collagen and slit-like vascular channels. The spindle-shaped cells formed an interlaced pattern and nucleus mitosis could be seen partly without obvious abnormal cells

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Fig. 1 As can be seen from (A) and (B), clinical presentation of deformity and swelling of the third toe. The toe can be extended passively but not actively.

(Fig. 4). For immunocytochemical phenotyping, tissue sections were stained for vimentin, desmin, S-100, CD68, CD34, Ki67, SMA, NF, and CK. The results were as follows: CK(-), Vim(+), Ki67(<1%+), antiSMA antibody (-), CD34(+), Desmin(-), CD68(-), NF(-), and S100(-).

Based on these clinical and histological findings, the patient was diagnosed with FTS of the third toe in the left foot. Postoperatively, at the two-month follow-up, the patient demonstrated normal range of motion of the toe without any pain. During two years follow-up, no swelling or recurrence of the lesion was noted from the excision.

The patient was informed that data concerning her case would be submitted for publication, and she provided consent. The case report was approved by our hospital.

Discussion

Fibroma of the tendon sheath was initially described by Copeland and Geschickter⁸ as either “reactive fibrosis” or “fibrotic neoplasm”, and is still unclear. However, it is generally accepted that this tumor is a fibrous mass attached to the tendon sheath characterized by slow-growing feature. FTS mainly occurred in the fingers, hands, and wrists, accounting for 75% to 82% of the locations of FTS².

To our knowledge, FTS rarely occurs in the foot, no more than 20 cases having been reported. Even the largest series reported in 1979 by Chung and Enzinger⁵ includes only six cases occurring in the foot, among which was only one case in the toe, and five in other regions of the foot. In 1985, Hashimoto *et al.*³ also reported one case in the toe and two in other

Fig. 2 (A) and (B), respectively show X-ray films of anteroposterior position and oblique position showed no remarkable abnormalities of the left foot, except for swelling signs of the soft tissues in the third toe.

Fig. 3 (A) showed an ivory white soft tissue tumor was adherent to the tendon sheath. After excision the entity measured 2.3 cm × 2 cm × 0.8 cm as shown in (B).

regions of the foot. In 1987, Sarma *et al.*⁹ reported a case in the foot region and the toe was not included. Lourie *et al.*⁶ reported a case eroding the third metatarsal bone of the foot in 1992. In 1993, Chi and Oda¹⁰ presented the first case of FTS with ulceration originating in the foot without toe involvement. In 2001, Yamamoto *et al.*⁷ presented a case of toe deformity due to FTS. In 2012, Kim *et al.*¹¹ reported a case of fibroma of the flexor hallucis longus tendon sheath. To date, only four cases in the toe have been reported in the literature. Our case also presented with toe deformity as reported by Yamamoto, however, the occurrence was in a 26-year-old woman after pregnancy without traumatic history.

As for the etiology of FTS, an association with trauma accounts for 10% of the cases reported¹. The latest study showed that tumorous changes caused by chromosomal abnormality of 2:11 translocation may induce FTS¹². In our case, the etiology is still unclear. Histopathologically, fibroma of the tendon sheath usually presents with spindle- or stellate-shaped cells, slit-like blood vessels in densely collagenized stroma. The cells are with small, fusiform nucleus even in fission. Many areas are of increased cellularity, formed in an interlaced pattern while some regions of this tumor are hypocellular with variable cellularity present.

Differential diagnosis must be made since FTS resembles many other lesions. FTS is often confused with GCTTS which is one of the most common soft tissue tumors in the foot and ankle¹³, they both arise in similar locations and own similar appearances and radiographic signs. However, microscopically, FTS is hypocellular with slit-like vessels in a dense stroma, while GCTTS is more cellular with multiple nucleated giant cells, foamy histiocytes, and hemosiderin¹⁴. Nodular fasciitis also resembles FTS; however, it is poorly circumscribed and often shows characteristic findings such as extravasation of erythrocytes and running of many immature capillaries in a parallel direction in myxoid areas. Also it grows rapidly and usually reaches to its full size within 3 to 6 weeks¹⁵, while FTS is usually slowly growing. Benign fibrous histiocytoma macroscopically resembles FTS while it has a distinct storiform pattern histologically⁵ and is often accompanied by clusters of foamy histiocytes and siderophages. Neurofibroma may be confused with FTS with myxoid areas; however, S-100 protein immunohistochemistry can be helpful for the differential diagnosis. Schwann cells in neurofibroma have S-100 protein immunoreaction products in the nuclei and cytoplasm¹⁶, while FTS is negative for S-100 protein. Highly similar desmoplastic fibroblastoma could be distinguished from FTS by the

Fig. 4 (A) (HE ×40) showed hypercellular densely collagenous areas are presented in this tumor. Spindle cells are embedded in a dense collagenized stroma with slit-like vessels as shown in (B) (HE ×100).

appearance of slit-like vessels. Pigmented villonodular synovitis consists of a large amount of foamy, histiocytic cells¹⁷ with hemosiderin present, which are absent in FTS.

The recurrence rate of FTS which was reported to be 24% in the series of Chung and Enzinger⁵, and was supposed to have high relation to the extent of excision¹⁸. However, the recurrence of FTS in the toe had not been reported in the

literature. In our case, the patient was followed up for two years and there was no recurrence. The patient's toe appeared normal without limitation of motion.

FTS occurring in the toe is extremely rare and can sometimes be presented as toe deformity. Pathologic analysis is typically required for an accurate diagnosis. We presented a rare case of FTS occurring in a woman's toe after pregnancy.

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