CASE REPORT

A rare case of painful fibroma arising from tendon sheath of *flexor hallucis longus*

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Abstract

Fibroma of the Tendon Sheath (FTS) is a rare soft tissue tumor commonly found in upper extremities. Usually, the lesions occur in wrists and hands. There are only a few cases of fibroma of the tendon sheath arising from the lower extremities have been reported. This case report describes a rare presentation of painful FTS that arises from *flexor hallucis longus* at the plantar aspect of the forefoot in a middle age lady. To our knowledge there is only one similar case reported previously.

Keywords: Fibroma, Giant Cell Tumor, *Flexor Hallucis Longus*

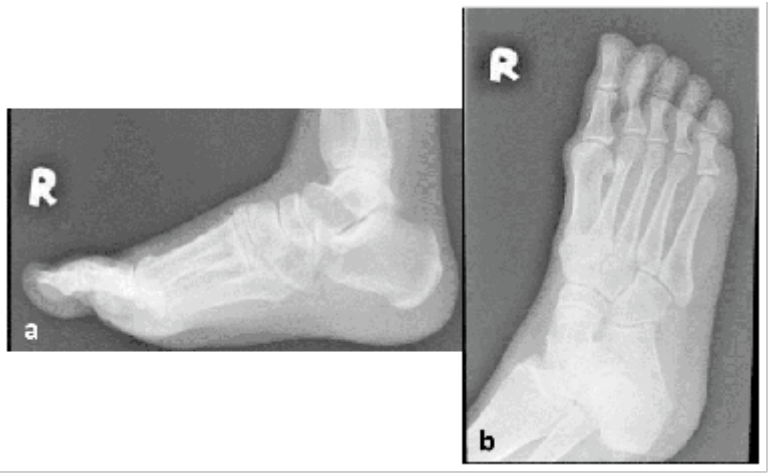
Introduction

Fibromas of the tendon sheath are rarely reported in lower limbs mainly in fingers and hands [1-4]. It commonly occurs among males in middle age group population. The usual presentations are slow growing painless swelling [2-5]. Plain radiograph typically is non remarkable while ultrasounds and Magnetic Resonance Imaging (MRI) are performed in some cases for diagnostic purpose. Excision of the tumor is the treatment of choice with diagnostic confirmation on histo-pathological assessment [1]. Complications such as bony erosions, nerve compressions namely median nerve neuropathy and tendon ruptures were reported although typically these lesions rarely cause serious complications [1]. Histo-logically it shows a hypocellular tumor composed of bland fibroblasts [2-3]. Fibromas have a high rate of recurrence likely a result of incomplete excision of the tumor [3].

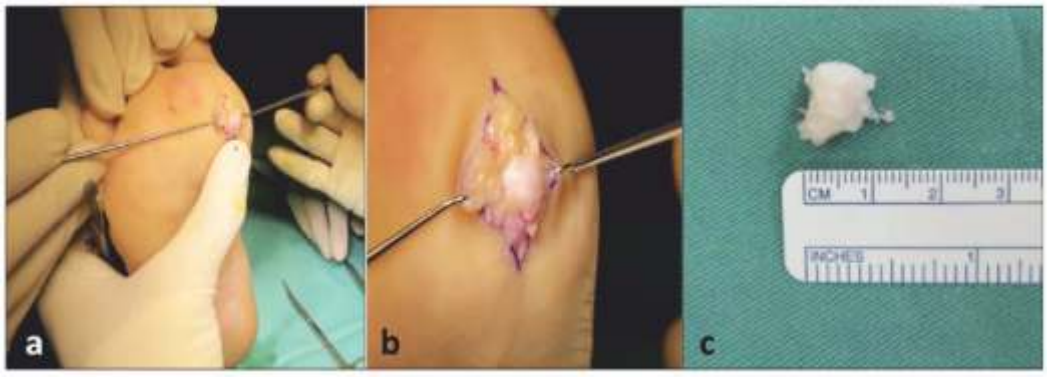
Case Report

A 36-year-old Malay lady presented to us with a single swelling over the sole of the left foot for 3 years in duration. The swelling was 1 × 1 cm in size and did not increase in size over time. It was associated with worsening pain especially during walking. Examination revealed a healthy looking medium built lady with antalgic gait. There was a swelling at the plantar aspect of her left foot, at the first metatarsal head measuring 1 × 1 cm. The swelling was firm in consistency, minimally tender, had well defined margin and it moved with flexion and extension motion of the interphalan-geal joint of the left big toe. There was no evidence of infection. Neurovascular assessment was normal. Blood investigations showed no evidence of infection. Plain radiological assessment showed no bony abnormality (Figure 1). Ultrasonography showed a hypoechoic lesion around plantar aspect of the first metatarsophalangeal joint, located superficial to flexor tendons. Differential diagnosis of ganglion cyst or bursitis were made. No MRI study was performed for our case. The pain did not resolve with analgesics. In view of non-resolving

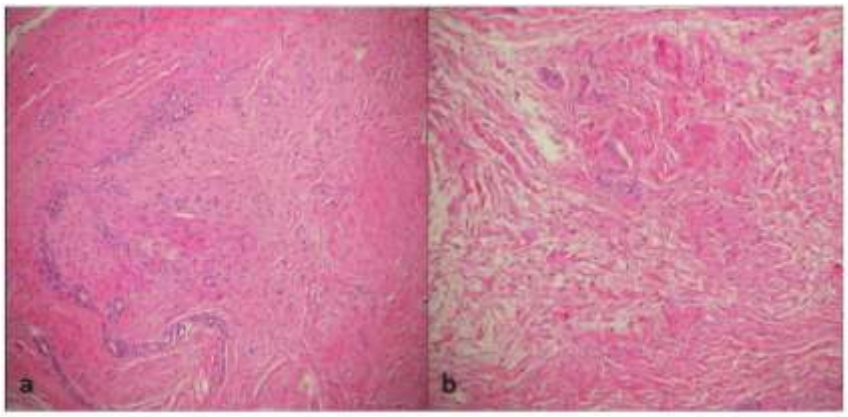
pain we decided for excision of the lesion. We planned for excision biopsy of the swelling performed under general anesthesia. After induction patient was placed in supine position. Intraoperative assessment revealed a firm lesion sized 1 × 1 cm arising at the plantar aspect of the left foot. To be more specific it was located at the 1st metatarsophalangeal head and moved with movement of the interphalangeal joint of the big toe. There was no sign of inflammation over the surrounding skin. A vertical incision was made overlying the swelling. Intraoperatively we noticed a lobulated swelling arising from tendon sheath of *flexor hallucis longus* measuring 12 × 8 × 7 mm. It was firm in consistency and appeared whitish in color. The swelling was removed carefully with preservation of the tendon (Figure 2). Histopathological assessment revealed a fibrocartilaginous tissue composed of bland fibroblastic/ myofibroblastic spindle cells. Blood vessels with scattered adipocytes and nerve bundles are present within the tissue. No atypia or features of malignancy seen. The findings are consistent with fibroma of the tendon sheath (Figure 3). At two years follow up after surgery there is no sign of recurrence.



Figures. 1 (a) and (b): Plain radiograph of the patient showing increased soft tissue shadow at the plantar aspect at the region of metatarsal



Figures. 2 (a) and (b): Intraoperative picture showing the glistening white appearance of the tumor attached to *flexor hallucis longus* tendon



Figures. 3 (a) H&E, 40× and (b) H&E, 100×: Histopathology examination shows abundant bland spindle cells surrounded by collagen fibers with scattered capillaries. There is no cellular atypia or mitotic figures that confirms its benign nature

Discussion

Fibroma of the Tendon Sheath (FTS) is a rare benign soft tissue tumor. It is usually found in the upper extremities namely in fingers, hand and wrists. It rarely occurs in the lower extremities. [1-4]. The largest case series were by Chung and Erzinger in 1979 who mentioned 98% of their 138 subjects occurred in those locations [2-5]. Man has more predominance towards these tumors and it generally occurs in the middle age group [1]. This benign tumor is a slow growing mass. Typically, it presents as painless mass and rarely causes problem to patients [2-5]. Our patient presented with a solitary swelling that gradually increases in size. However, since it was associated with pain especially during walking, she decided to seek treatment. Some patients do have previous incidence of repetitive trauma to the swelling site. [1]. She denied any previous injury. Lu *et al*. in 2016 reported 38% of his subjects has neuro-vascular bundle involvement requiring decompression intraoperative. Due to the rarity of this tumor and its unusual location in this patient diagnosis of FTS was not initially included. Other differential diagnoses that should be considered are Giant Cell Tumor of Tendon Sheath (GCTTS), nodular fasciitis, neurofibroma, leiomyoma, scar tissue and fibrous histiocytoma [6]. FTS and giant cell tumor have a close resemblance in the site of origin [6]. Furthermore, the gross appearance of the mass is usually being closest to giant cell tumor of the tendon sheath making both of them difficult to be differentiated macroscopically [7].

Plain radiograph has no role for diagnosis. It is performed to visualize and soft tissue swelling or bony involvement. As FTS is basically a benign tumor there is no remarkable findings expected to be seen from a plain radiograph. However, there are cases reported where a large mass compresses surrounding fat and muscles and presence of erosive bony changes which are rarely described [5]. Ultrasound is suggested for its noninvasive convenience especially for superficial mass [7]. MRI is preferred when the tumor is large and deeply seated or to demonstrate any involvement

of neurovascular structures. This modality can assist in performing safe resection range and selecting most suitable incision. MRI is also preferred in detecting recurrence in the early stages [7]. Typically, FTS will show low signal or slightly high signal intensity on T2 weighted images.

Macroscopically, FTS is described as a well-defined solitary nodule that appears as pearl white or grey colored. It lies in close proximity or adhered to the neighboring tendon or tendon sheath. Most of the masses are less than 2 cm in size [7]. Difficulty in distinguishing FTS from other benign lesions macroscopically have been reported. Thus, histopathologic examination which occasionally requires immunohistochemical assistance might be necessary. Differentiating FTS from GCTTS is a challenge in a deeply seated masses as both lesions may share similar size, location and gross appearance [7].

Histopathological assessment will reveal its benign nature. Typically, it is composed of benign proliferation of fibroblasts surrounded by collagen fibers [1-3]. They have also been described as small lobules with slit like vascular channels consists of hyalinized collagen fibers and spindle cells [2, 7]. These features are shared with GCTTS microscopically. However, the latter often has presence of histiocytes, monocytes as well as multinucleated giant cells, foam cells and hemosiderin-laden macrophages [1].

Excision has always been the treatment of choice. Nevertheless, local recurrences ranging between 10-45% have been reported [5]. Ciati *et al*. in 2009 mentioned high incidence of local recurrences at 24% and those cases were lesions located at the hands and fingers. It was suggested that the recurrence depends on the accuracy of the surgery [1, 3]. Such a high recurrence rate is due to the difficulty in complete resection of the tumor as it is closely adhered to the tendons, tendon sheath and nerves [7]. A malignant transformation has never been described [1, 6, 8].

Conclusion

FTS is a rare benign soft tissue tumor commonly occurs in the wrists and hands. Swelling in the foot should trigger the clinicians to think of FTS as one of the differential diagnoses. Most patients will present with painless swelling although 30% of the patients might have painful swellings. Clinically, it is difficult to distinguish them from giant cell tumor of the tendon sheath. Imaging modalities were not conclusive. Imaging can help in distinguishing low grade, benign and malignant swelling and not definitive for diagnosis. Excision is the treatment of choice. Histopathological assessment in FTS will show benign proliferation of fibroblasts. Absence of giant and multinucleated cells differentiate FTS from GCTTS. Recurrence is a common complication, usually due to incomplete removal. As a conclusion fibroma of the tendon sheath of *flexor hallucis longus* should be included as the differential diagnosis when a patient presents with a painful mass in the plantar site of the forefoot.

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