

Case Report

Fibroma of tendon sheath in the patellar tendon in a Chinese boy

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Abstract: Fibroma of the tendon sheath (FTS) is a benign lesion or a tumor-like reactive lesion arising from the synovium of the tendon sheath. FTS usually occurs in the upper extremities, but rarely in the knee joint. Herein, we report a case of an 8-year-old Chinese boy with no prior trauma in which a FTS was inadvertently found in the infrapatellar region of the left knee. Magnetic resonance imaging (MRI) showed a heterogeneous enhanced intensity in T1-weighted images and mixture signal intensity in T2-weighted images. Histologic examination and immunohistochemical analyses revealed the tumor was FTS. At 1-year of follow-up, the tumor recurred after surgery and surgical treatment was performed again. There was no abnormality during 6 months of additional follow-up. Therefore, careful follow-up is necessary for this and for similar cases.

Keywords: Knee, fibroma of the tendon sheath, child, patellar tendon, neoplasm

Introduction

Fibroma of the tendon sheath (FTS) is a benign lesion, or a tumor-like reactive lesion, arising from the synovium of the tendon sheath [1]. FTS predominantly occurs in the upper extremities, particularly in the fingers, hands, and wrists [2]. It has rarely been reported to occur in the knee joint [3, 4]. So far only 27 cases of FTS have been reported in the knee and only 3 cases involved the patellar tendon [3, 5, 6]. Herein, we present an 8-year-old Chinese boy with FTS originating from the patellar tendon.

Case presentation

In March 2, 2015, an 8-year-old boy with no history of major trauma presented with a 1-year history of a painless mass in the infrapatellar region of the left knee, but recently experienced an increase in growth. He reported no associated fever, chills, night sweats, or weight loss. His family histories were noncontributory. During physical examination, edema, erythema, ulceration and local heat were not noted around his left knee. Flexion and extension in his left knee joint were limited. Laboratory data were unremarkable.

Radiographs of the knee joint showed a soft tissue mass of the inferior patella without bony abnormality. Magnetic resonance imaging (MRI) prior to surgery revealed a 6.0×5.9×4.2 cm, well defined, soft tissue mass to the patellar tendon. On T1-weighted images, the lesion showed low signal intensity in the patellar tendon. On T2-weighted images, the lesion showed a heterogeneous intensity and patchy, striped high signal intensity and striped low signal intensity were seen within the lesion (**Figure 1A-C**), without enhancement of the striped hypo-intensity of the lesion (**Figure 1D**). Clinical and radiological findings suggested a giant cell tumor tendon sheath (GCTTS).

An operation was performed on March 4, 2015. A longitudinal incision was made in the front of the left knee to cut off the skin, subcutaneous tissue and fascia to expose the bulging patellar tendon. It was visible that the masses were mainly in the lower part of the patellar tendon, which wrapped and infiltrated part of the patellar tendon. Dissection was performed along the wall of the patellar tendon to the deep part and the root. It was important during this procedure that the patellar tendon was kept as complete as possible, to avoid damaging important ves-

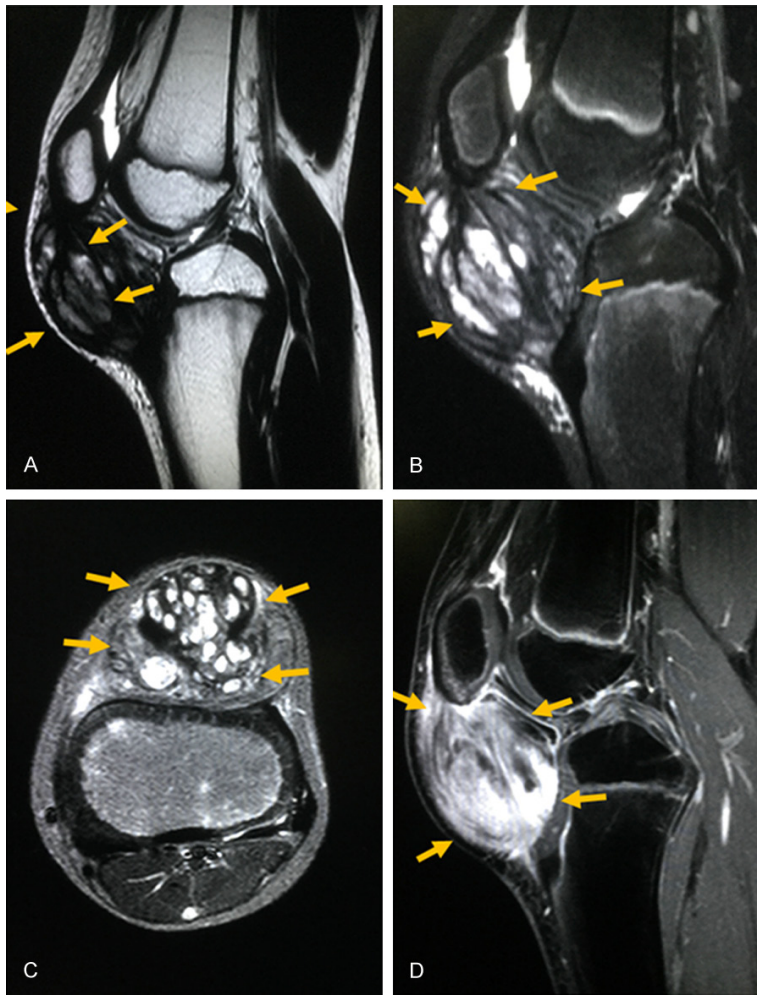


Figure 1. Magnetic resonance (MR) images of the tumor in the left knee. The tumor showed a mixture of low and high signal intensity in sagittal T2-weighted images (A: TR/TE 3276/90), fat-suppressed T2-weighted images (B: TR/TE 3290/80) and axial fat-suppressed T2-weighted images (C: TR/TE 2497/35). The tumor showed a heterogeneous enhancement in fat-suppressed T1-weighted images (D: TR/TE 223/7.5).

sels and nerves. Masses under the patellar tendon were dissected at first, then the patellar tendon was longitudinally incised to isolate and dissect masses of the patellar tendon to the greatest extent. The gross specimen was described as well circumscribed, nodular, gray, grayish pink, and rubbery (**Figure 2A**). Histologic examination showed a lobulated mass, consisting of fibroblast-like spindle cells associated with a dense collagenous stroma, and myxoid areas with cystic degeneration. Bone and cartilage were observed in the tumor. Cellular variants were not obvious. However, a small number of nuclear fissions were seen (**Figure 2B, 2C**). Immunohistochemical analyses of the

tumor cells showed that they were positive for vimentin, KP-1, and negative for S100 protein (**Figure 2D**). These results were diagnostic for FTS. The patient was hospitalized for 6 days after operation. There were no intraoperative or postoperative complications.

At 1-year of follow-up, plain radiographs revealed a painless soft tissue mass in the infrapatellar region of the left knee and confirmed tumor recurrence after surgery. Imaging review was not performed at the surgery. Development of the two legs were similar, and the patient could walk normally. Knee masses occurred once again in the front of the left knee without any obvious causes 5+ months after discharge from hospital. The masses gradually increased without pain and fever, but the activities were limited. The patient was re-hospitalized on February 3, 2016, and MR examination was performed (**Figure 3**). Surgical treatment was performed. Skin and subcutaneous tissue were cut off to expose the tumor. It was visible that the patellar tendon was completely infiltrated.

Moreover, the tumor was hard with septum and adhered to the surrounding tissue. Therefore, the patellar tendon and tumor were dissected as a whole, then the patellar tendon was reconstructed. There was no abnormality by X-ray when the case was reviewed after 1 month (**Figure 4**). There was no abnormality during the 6 months of follow-up. The patient returned to full sporting activities. Further follow-up was scheduled for 2 years owing to the potential for recurrence.

Discussion

Fibroma of the tendon sheath is a rare benign soft tissue tumor, and was first defined by

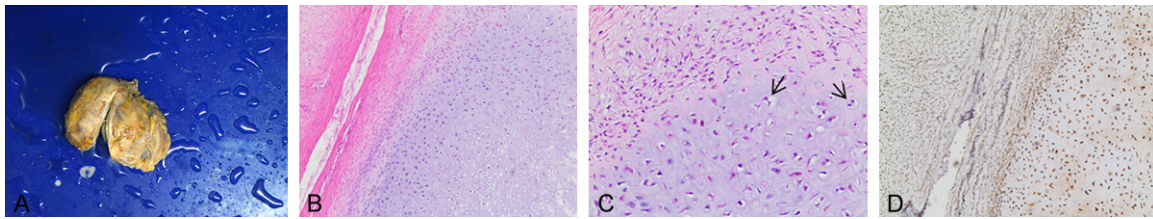


Figure 2. Histological examination of fibroma of the tendon sheath (FTS). (A) The gross specimen was described as well circumscribed, nodular, gray, grayish pink, and rubbery. (B, C) Histological findings of the tumor (B: hematoxylin and eosin staining, $\times 40$; C: Hematoxylin and eosin staining, $\times 100$). Histologic examination showed a lobulated mass, consisting of fibroblast-like spindle cells associated with a dense collagenous stroma, and myxoid areas with cystic degeneration. Bone and cartilage were observed in the tumor. Cellular variants were not obvious and a small number of nuclear fissions were observed (black arrows). (D) Immunohistochemical analyses of the tumor cells showed that there was positive staining for vimentin.

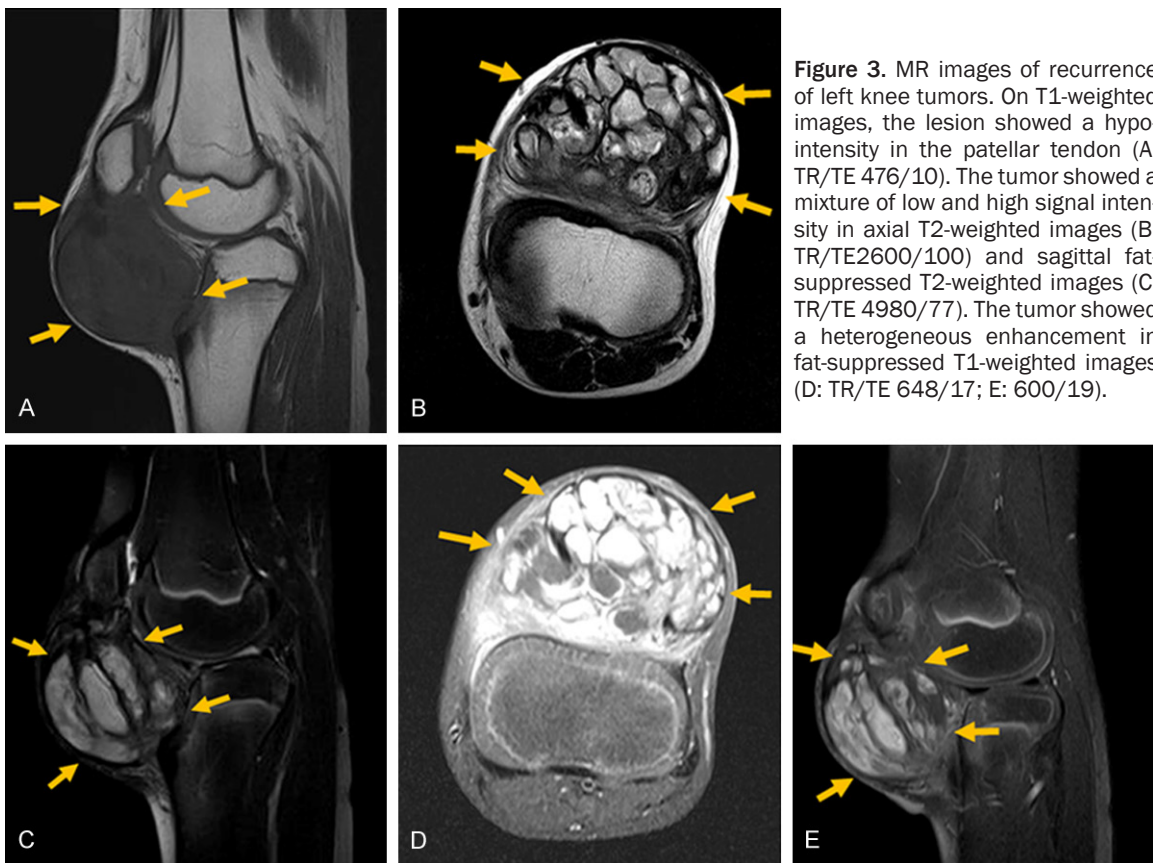


Figure 3. MR images of recurrence of left knee tumors. On T1-weighted images, the lesion showed a hypo-intensity in the patellar tendon (A: TR/TE 476/10). The tumor showed a mixture of low and high signal intensity in axial T2-weighted images (B: TR/TE 2600/100) and sagittal fat-suppressed T2-weighted images (C: TR/TE 4980/77). The tumor showed a heterogeneous enhancement in fat-suppressed T1-weighted images (D: TR/TE 648/17; E: 600/19).

Geschickter and Copeland in 1936, while the 138 cases Chung and Enzinger [2] reported in 1979, remain the world's largest series on tendon sheath fibromas and serve as the foundation for much of our clinical and pathological understanding of these tumors. However, it is unclear whether FTS is a reactive hyperplasia or neoplasm, with some individual cases reporting an identical chromosome abnormality characterized by $t(2;11)(q31-q32;q12)$, and the clonally occurring chromosome abnormality

found in this fibroma of tendon sheath suggests that this proliferation is neoplastic and not a reactive fibrosing process [7].

FTS typically develops in young adult men, with men outnumbering women by a ratio of 1.5-3:1, with a peak incidence at 20-50 years of age, and a history of trauma related to the development of the lesion noted in 9% of the cases [2, 6, 8]. The typical symptoms in almost all cases are that of a painless slowly growing



Figure 4. X-ray examination of the left knee. A: After operation. B: 1-month after operation.

mass, although a few may be accompanied by pain or tenderness. FTS often attaches to the tendon or tendon sheath, has clear borders, is lobulated or nodular, solid, rubbery, tough and resilient, the texture is plane and uniformly gray.

Radiography reveals a soft tissue mass without bony abnormality. MRI has greatly increased the capacity for showing detailed pathological changes, and has become an indispensable tool for diagnosis and follow-up testing of FTS. MRI findings have been previously reported, with the majority of the lesions showing T1-weighted images with a low signal, and the

surrounding muscle signal equal or slightly lower, and a few showing a slightly higher signal, and mixed low and high signal in T2-weighted images. Pinar et al. [1] reported that differences in the amounts of hyalinization and the number of proliferating fibroblasts might generate variations in T2-weighted MRI findings. More hyalinized or sclerosed forms of FTS will tend to show lower intensities on T2-weighted images, whereas a more cellular variant will have a higher T2 signal. Enhancement after contrast also varies, with some cases reporting no enhancement, some reporting mild to significant homogeneous enhancement, and others reporting peripheral enhancement, the edge enhancement is more obvious than the central enhancement [1, 8]. However, in this case the tumor showed a mixture of low and high signal intensity in T2-weighted images, T1-weighted images, and a heterogeneous enhancement.

Differential diagnoses include GCTTS, as suggested in this case, and pigmented villonodular synovitis (PVNS). GCTTS mostly develops in young adults, is the most common

benign synovial tumor of the hand and wrist, and represents a localized manifestation of pigmented villonodular synovitis. GCTTS is usually iso-intense on T1-weighted images and inhomogeneously hyper-intense on T2-weighted images [9]. PVNS is often accompanied by knee joint swelling pain and limited activities it growth within the joint more often is characterized by a villous structure. Hemosiderin deposition in the lesion may be manifested as hypo-intense areas on T2-weighted images.

Treatment of FTS is by local excision, but the recurrence rate can be as high as 24% [2]. Due to the adherence of lesions to surrounding ten-

dons and tendon sheaths, and as it is a benign tumor, to preserve function, lesions are not completely removed, which is likely to be one reason for the high recurrence rate. In the case presented here we were aware that if the range of the operation was too large, this may, in an 8-year old child, adversely affect the development and function of the knee. On the other hand, if the range of the operation was too small, there was likely to be a high chance of recurrence. Therefore, careful follow-up is necessary for this case and for similar cases.

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Disclosure of conflict of interest

None.

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