



## Fibromatosis of the soleus muscle presenting as pes equinus: A case report

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It is uncommon for soft tissue tumor to present as joint contracture as the first symptom. We report a case of a fibromatosis in soleus muscle presenting as pes equinus. The patient walked with a toe-walking gait pattern and the heel came off the floor about five centimeters. The dorsiflexion of the foot was limited to -50 degrees despite the knee position. A cord-like lesion was palpable from the mid-calf down to insertion of the Achilles tendon. T1- and T2-weighted magnetic resonance images showed a soft tissue lesion with the hypointense signal in her left soleus muscle. Partial resection of the proximal end of the tumor, in combination with adhesiolysis and Achilles tendon lengthening, was performed. Two years after the surgery there was no recurrence of pes equinus, although she experienced mild leg pain after long walks. Pathological assessment revealed the diagnosis as fibromatosis in the soleus muscle.

**Keywords:** pes equinus, fibromatosis, Achilles tendon lengthening, tumor resection

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**P**es equinus is a deformity in which the foot is held in a plantarflexed position. Equinus deformity usually is caused by neurologic disorders, such as hemiplegia and cerebral palsy [1,2], but it also may result from mechanical contracture of the gastrocnemius/soleus muscles due to compartment syndrome [3,4], a burn [5], traumatic fibrosis [6], or an aesthetic procedure [7]. Although very rare, tumors arising in the calf muscles have been reported to cause pes equinus [8-12]. Herein, we report a case of acquired equinus deformity due to fibromatosis of the soleus muscle.

### Case Report

A 40-year-old woman presented with a chief complaint of limited right ankle motion and leg pain after long walks. Her right ankle became gradually plantarflexed and she had been unable to stand on her heel for 2 years. She had no history of trauma or fitness habit in the lower leg. Her symptoms gradually worsened until her presentation at our hospital.

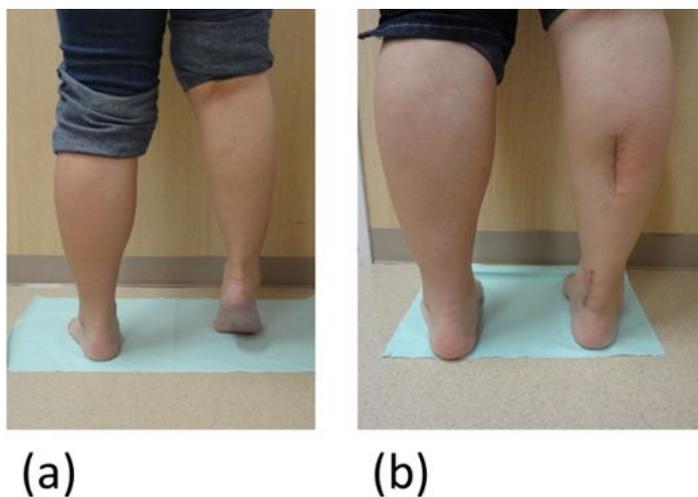
The patient had a toe-walking gait pattern with the right heel elevated off the floor by approximately 5 cm. Range-of-motion examination showed that dorsiflexion of the foot was limited to -50° regardless of the position of the knee (Figure 1a). Sensory loss and limited toe motion were not evident. There was a plantar callosity on the forefoot, and a cord-like lesion was palpable from the mid-calf down to the insertion of the Achilles tendon.

On plain radiography of the lower leg, no calcification was observed. T1- and T2-weighted magnetic resonance imaging showed a soft tissue lesion with a hypointense signal, suggesting highly collagenized tissue, in the right soleus muscle from the proximal one-third of the tibia to the insertion of the Achilles tendon (Figures 2, 3). Sonography revealed that sliding of the Achilles tendon was restricted due to tightness of the cord-like lesion. Diagnostic needle biopsy was performed with the specimen showing fibrous tissue without atypical cells.

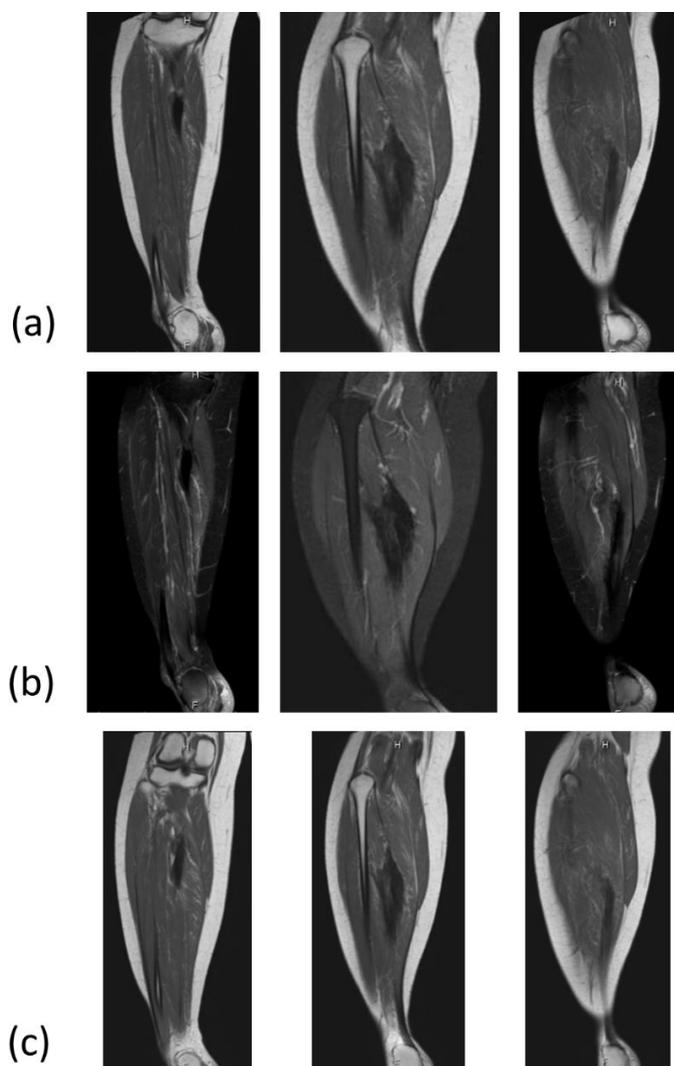
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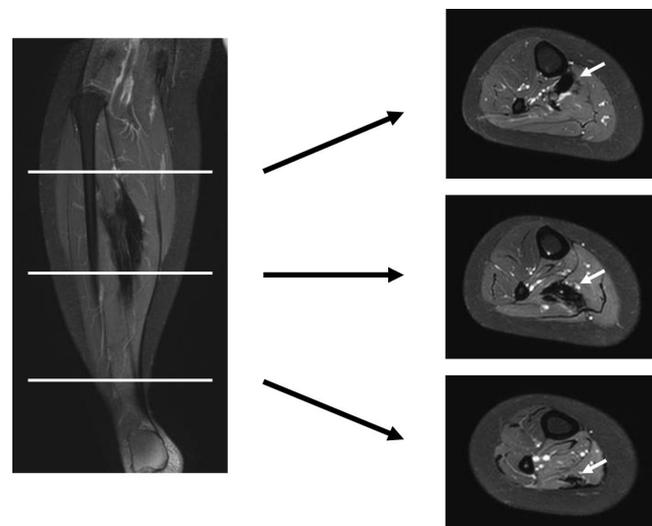
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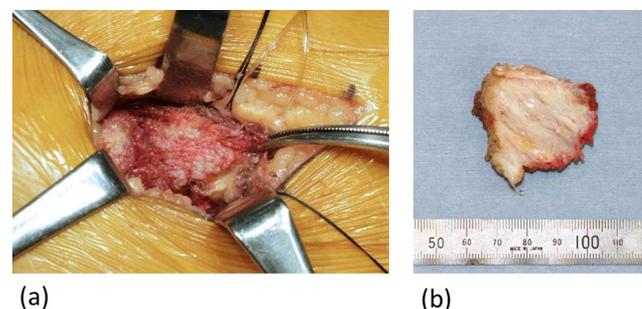
**Figure 1** Appearance of the leg. (a) Preoperative dorsiflexion was limited to  $-50^{\circ}$ . (b) Dorsiflexion 2 years after surgery was  $0^{\circ}$ .



**Figure 2** Series of preoperative findings on magnetic resonance imaging (MRI). Sagittal T1- (a) and T2- (b) weighted MRI revealed a hypointense lesion in the right soleus muscle. (c) The lesion was slightly enhanced on gadolinium-enhanced T1-weighted MRI.

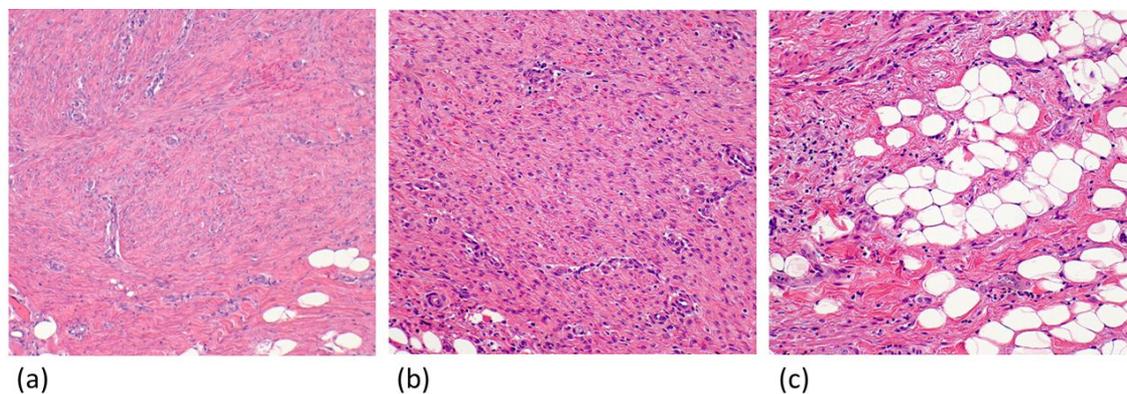


**Figure 3** Coronal T1-weighted magnetic resonance imaging revealed a lesion from the proximal one-third of the tibia to the insertion of the Achilles tendon (arrows).



**Figure 4** Intraoperative gross findings of the lesion. (a) The boundary of the lesion from normal muscle was not clear. (b) The cut surface of the lesion was composed of light-colored fibrous material.

We diagnosed the present case as pes equinus due to a fibrotic lesion in the soleus muscle, suggesting possible fibromatosis. Surgery was planned for 2 purposes: (1) to make a final histologic diagnosis using a large sample and (2) to correct the equinus deformity. Surgery was performed under general anesthesia with the patient in the prone position. First, a posterior incision was made in the proximal one-third of the calf over the proximal gastrocnemius muscle. Once visualized, the tumor had a light-colored gross appearance and no obvious capsule; thus, the boundary of the lesion from normal muscle was not clear. The proximal end of the lesion, which was strongly adhered to the posterior periosteum of the tibia, was resected intralesionally ( $30 \times 30 \times 20$  mm) (Figure 4a). Then, the soleus muscle containing residual tumor was released from the ventral soft tissue as far as possible. At this point, dorsiflexion improved from  $-50^{\circ}$  to  $-20^{\circ}$ . Subsequently, a posteromedial incision was made and the Achilles tendon was visualized.



**Figure 5** Histopathologic findings of the lesion. **(a-b)** Fibro-collagenous tissue with a proliferation of spindle-shaped cells was seen (hematoxylin and eosin stain, magnification  $\times 40$  and  $\times 100$ ). **(c)** Tumor infiltration into adjacent adipose tissue was seen (hematoxylin and eosin stain, magnification  $\times 40$  and  $\times 100$ ).

The distal end of the tumor was palpable in front of the musculotendinous junction and was tightly adhered to the Achilles tendon. Manual adhesiolysis was performed as proximal as possible. Then, the Achilles tendon was gently lengthened by approximately 2-cm by using a sliding lengthening technique. Final dorsiflexion improved to  $0^\circ$ .

The resected tumor was elastic, hard, and had a tendon-like appearance with poorly defined margins. The cut surface was composed of light-colored fibrous material (Figure 4b). The section showed fibrocollagenous tissue that was poorly circumscribed with a proliferation of spindle-shaped cells and infiltration to surrounding soft tissue structures. Mitotic figures were rarely seen. Pathologic examination revealed that the tumor was compatible with fibromatosis (Figure 5). To determine the subtype of fibromatosis, immunohistochemical analysis was performed. The spindle-shaped cells tested positive for beta-catenin (clone, 14/beta-catenin; dilution, 1:200; BD Biosciences, San Jose, CA) in the cytoplasm and for alpha-smooth muscle actin (clone, 1A4; dilution, 1:5000; SIGMA-ALDRICH, St. Louis, MO) in the nucleus. These results are inconsistent with deep fibromatosis (desmoid tumor), which shows positive staining of beta-catenin in the nucleus [13]. Therefore, we could not determine the subtype of fibromatosis.

After surgery, the diseased limb was set in a splint with  $-15^\circ$  dorsiflexion. Two days after surgery, active ankle dorsiflexion within patient tolerance and passive ankle dorsiflexion up to  $-15^\circ$  were started. The patient was allowed to bear full weight with a short leg brace four weeks after surgery. Two years after surgery, there was no recurrence of pes equinus (Figure 1b), though she experienced mild leg pain after long walks.

Range-of-motion examination showed dorsiflexion of  $0^\circ$  and plantarflexion of  $50^\circ$ . Follow-up magnetic resonance imaging revealed no obvious progression of the residual tumor.

## Discussion

Soft tissue tumors arising in the calf muscles are a rare cause of acquired equinus deformity; however, several cases of hemangioma and hamartoma have been reported [8-12]. To our knowledge, this is the first case of fibromatosis of the soleus muscle presenting as pes equinus.

Fibromatosis is characterized by spindle-shaped fibrous cells with rare mitoses accompanied by abundant collagenous material [14]. Musculoskeletal fibromatosis is divided into 2 major types: superficial and deep. Superficial fibromatosis includes palmar fibromatosis (Dupuytren's disease) and plantar fibromatosis (Ledderhose disease). Dupuytren's disease is characterized by flexion contracture of the digits due to cord-like expansion of the aponeurotic slips. Ledderhose disease is a relatively rare foot disorder in which a small nodule grows large enough to cause plantar contracture [15, 16]. In contrast to superficial fibromatosis, deep fibromatosis, also known as a desmoid tumor, grows more rapidly and is more aggressive [17]. Nuclear beta-catenin expression helps to distinguish desmoid tumors from other benign lesions [13]. In the present case, the location of the lesion and the intraoperative findings suggested a desmoid tumor. However, there was no nuclear beta-catenin expression, which is critical for diagnosing a desmoid tumor. Therefore, we could not determine the subtype of fibromatosis conclusively.

Operative management of pes equinus should be considered depending on the severity and pathophysiology of the case. When treating ischemic contracture, excision of the muscle infarction presenting as a cord-like induration is recommended, in combination with myolysis and tenolysis [18]. Achilles tendon lengthening can be performed additionally, utilizing a Z-lengthening or fractional lengthening technique [3, 18]. Excision of the fibrotic lesion combined with Achilles tendon lengthening has been reported for treatment of equinus deformity due to traumatic contracture of the gastrocnemius muscle caused by blunt injury or an aesthetic calf volume reduction procedure [6, 7]. With regard to tumor-induced equinus deformity, Klemme et al, reported that open Achilles tendon lengthening provided acceptable results for treatment of equinus deformity caused by hemangioma of the calf muscle [10]. In contrast, Nakamura et al, reported a case of pes equinus due to intramuscular hemangioma, which recurred four times after Achilles tendon lengthening. They suggested that hemangioma excision was necessary to prevent recurrence of equinus deformity [8]. In the current case, dorsiflexion improved from  $-50^{\circ}$  to  $-20^{\circ}$  after partial tumor resection and adhesiolysis, which further improved to  $0^{\circ}$  after Achilles tendon lengthening.

The necessity for total tumor resection has been controversial due to the high incidence of recurrence even after wide resection [19]. A local recurrence rate of 72% has been reported in cases of desmoid tumors with positive margins [19]. Furthermore, the recurrence rate at 5 years was reported to be 21% in limited fasciectomy groups [20]. Recently, several investigators reported that minimally invasive procedures are useful because fibromatosis is not curable [20, 21]. Considering the possibility of recurrence and functional damage, we performed only partial tumor resection in the present case. Although neither enlargement of the lesion nor recurrence of equinus deformity was observed 2 years after surgery, further careful follow-up should be performed in the future.

In conclusion, we described a case of fibromatosis of the soleus muscle presenting as pes equinus. We must be aware that soft tissue tumors can be a cause of equinus deformity. Partial tumor resection combined with adhesiolysis and Achilles tendon lengthening were effective to treat equinus deformity in this case.

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