

Clinical Quiz

Unilateral muscle hypertrophy and focal myositis following S1 radiculopathy

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Case

A 53-year-old man noticed a progressive swelling of the left calf that lasted for one month. He complained of calf pain without previous trauma or infection. Neurological examination revealed enlargement of the left calf, with a difference in circumference of 10 cm. Strength and sensory examinations were normal. Erythrocyte sedimentation rate and routine laboratory examination were within normal limits. Serum myoglobin level was elevated (103.9 µg/L, normal range 23-72 µg/L), while urin myoglobin level was normal. Serum creatine kinase level was elevated (566 UI/L, normal range 0-177 UI/L). Fibrinogen was mildly elevated (5.4 g/L, normal range 1.8-4.1 g/L). Electromyography of the muscles of the left calf indicated chronic radicular lesion of the S1 segment. Magnetic resonance imaging (MRI) of the lumbar spine showed left-sided disc protrusions of segments L4/L5 and L5/S1. He underwent L4/L5 and L5/S1 lumbosacral laminectomy, partial discectomy, and decompression. Despite the operation, calf swelling and pain remained unchanged.

Computed tomography (CT) of the left leg revealed enlarged muscles of the posterior compartment of the calf and the density of those muscles were markedly decreased (Figure 1a). Whole-body [¹⁸F] fluorodeoxyglucose (FDG) positron emission tomography/computed tomography (PET/CT) showed increased uptake of the ¹⁸F-FDG in the enlarged muscle of the posterior compartment, but standardized uptake value (SUV) was low compared to that of ma-

lignant tumors (Figure 1b). MRI scans of the left calf revealed thickened soleus, tibialis posterior, flexor digitorum longus and of gastrocnemius muscles. Edema of the medial head of the gastrocnemius and soleus muscles appeared as increased signal intensity on T2 weighted images (Figure 1c). Following administration of gadopentate dimeglumine, only the medial head of the gastrocnemius muscle revealed a diffuse, slight enhancement which was compatible with focal myositis (Figure 1 d,e).

Patient underwent biopsy of the medial head of gastrocnemius muscle. Histopathologic examination showed both muscle fiber hypertrophy and atrophy along with inflammatory cell infiltrates (Figure 2).

Despite the remnant of the left-sided disc protrusion of segment L4/L5, described on the postoperative MRI, the patient did not undergo any other surgical procedure. There was not any further progression or reduction in calf enlargement within the one-year follow up period.

Commentary

Neurogenic muscular atrophy is quite frequent in radiculopathy, while progressive hypertrophy in one or more muscle groups of the calf is a rare condition. Enlargement of a muscle may result either from true hypertrophy (increase in the number or the size of muscle fibers) or from pseudohypertrophy (infiltration of the muscle by collagen, fat, parasites, tumor or inflammatory cells)¹.

In our case ultrasound imaging, CT, PET/CT and MRI showed this enlargement to be due to muscle hypertrophy of the posterior compartment. Homogeneously decreased intramuscular echoes in ultrasound imaging, lower density on CT, increased uptake of FDG on PET/CT and increased signal intensity and contrast enhancement on MRI of the medial head of the gastrocnemius muscle were suggestive for muscle edema (pseudohypertrophy). Marked decrease in density of the involved muscles detected on CT (pseudohypertrophy) suggested extensive replacement of the muscles by adipose tissue or edema.

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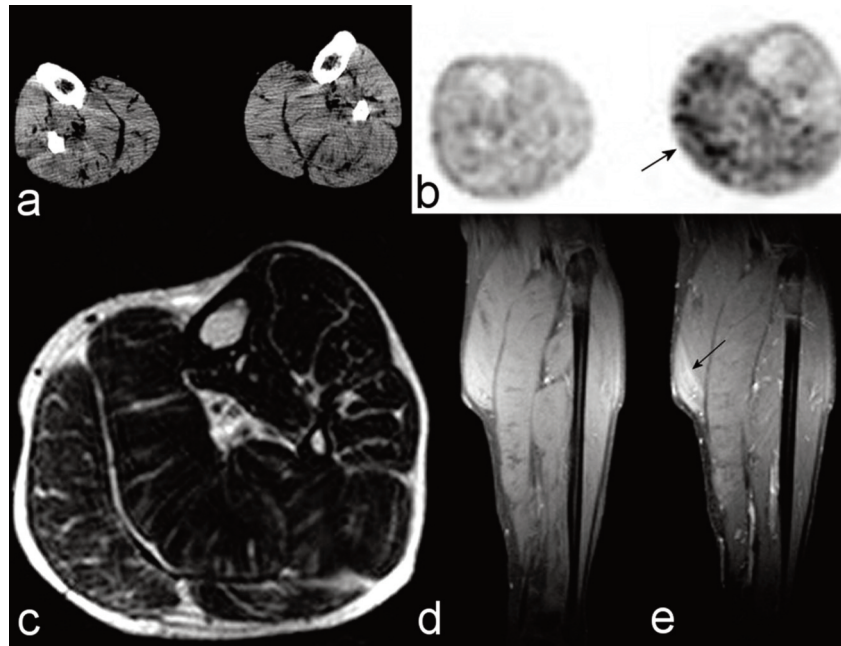


Figure 1. (a) CT of the left calf showed hypertrophy and diffuse-low density of the muscles of the posterior compartment; (b) PET/CT revealed ^{18}F -FDG uptake fusing to hypertrophic muscles, especially to the medial head of the gastrocnemius muscle (arrow); (c) MRI of left calf on axial T2 weighted image (TR 3700 ms, TE 86 ms) shows enlargement of soleus, tibialis posterior, flexor digitorum longus, and of gastrocnemius muscles; T1 weighted images with fat saturation (TR 532 ms, TE 10 ms) before (d) and after (e) iv administration of gadopentate dimeglumine shows slight enhancement of the medial head of the left gastrocnemius muscle (arrow).

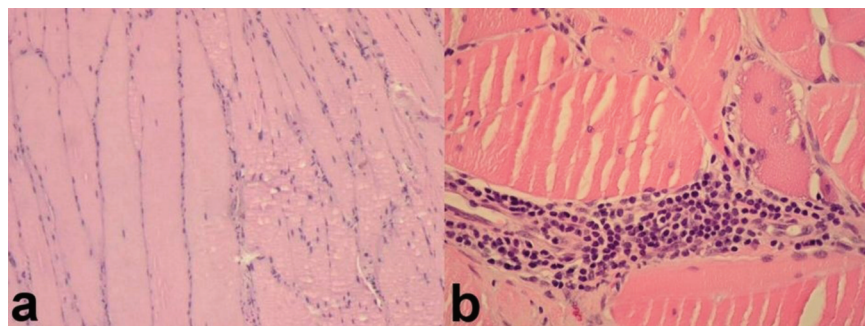


Figure 2. Biopsy of the medial head of the left gastrocnemius muscle showed hypertrophic fibers (a) and focal infiltrates of CD3 positive T lymphocytes and CD20 positive B lymphocytes (b).

PET/CT showed increased uptake of the FDG. The uptake of FDG is a measure of glucose uptake and is used as an indirect parameter of tissue metabolism. FDG uptake in myositis is based on the fact that inflammatory cells use large quantities of glucose through the hexose monophosphate shunt. To the best of our knowledge, this is the first case describing PET/CT finding in focal myositis associated with lumbosacral radiculopathy. Although, PET/CT finding is not specific, it contributes to the overall establishment of the diagnosis.

The MRI examination was consistent with an inflammatory

component in the hypertrophic muscle, compatible with focal myositis. In MRI, muscles with inflammatory edema or fat infiltration appear with increased signal intensity on T2-weighted images. There are two techniques to eliminate the signal of fat: T2-weighted images with fat suppression and short tau inversion recovery (STIR) sequences. Therefore, a combination of T1-weighted images and T2-weighted pulse sequences with fat saturation or STIR, are sufficient for assessing increased fat and/or water content within the muscle tissue. Contrast enhancement improves also the diagnostic ac-

curacy, especially when combined with fat suppression².

Several pathophysiological mechanisms of calf enlargement following chronic radiculopathy have been previously proposed. In some cases, complex repetitive discharges are responsible for chronic muscle stimulation. Another theory suggests that an increase in the workload imposed on the remaining, innervated muscle fibres is the cause of the enlargement³.

In our case the biopsy of the medial head of gastrocnemius muscle and of the tibia revealed hypertrophy and pseudohypertrophy of the muscle. Elevation of serum creatine kinase and inflammatory cell infiltrates in the muscle biopsy proved the radiological diagnosis of myositis. Focal myositis is a benign inflammatory pseudotumor of voluntary skeletal muscle characterized by the development of an inflammatory cell infiltrates. Several cases of focal myositis have been reported with an accompanying radiculopathy^{4,5}. In these cases it was suggested that focal myositis of neurogenic origin may be secondary to inflammation or that denervation may be the reason of the inflammatory infiltrate⁵.

Preclinical data suggest an immune pathogenesis of neuropathic pain, but clinical evidence for a role of the immune system in the neuropathic pain in patients is less clear. In animal models it has been shown that neuropathic pain is a result of several neurological conditions with a primary immune basis. Injury to the nervous systems results in activation of immune cells; cytokines and chemokines that are released by these immune cells sensitize nociceptive signaling in the peripheral and central nervous systems. The same has been shown in some cohorts of patients with neuropathic pain, although definitive evidence of a spinal glial response to peripheral nerve injury is awaited⁶.

The course of illness is normally benign and it is characterized by slow, spontaneous remission. Oral corticosteroids have

limited effect⁵. The benefits of local botulinum toxin A injections remain to be evaluated in patients with myositis following denervation. In addition, surgical laminectomies had no effect in reducing calf enlargement, as it was reported in other cases of chronic radiculopathy.

This case provides further evidence that chronic radiculopathy could be responsible for the development of neurogenic muscle hypertrophy and focal myositis, and that careful diagnostic evaluation is needed to exclude either the asymmetric presentation of a systemic neuromuscular condition or a tumor.

References

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Questions

1. Which of the following methods may be negative in focal myositis?
 - A. Computed tomography
 - B. Magnetic resonance imaging
 - C. ^{99m}Tc scintigraphy
 - D. Ultrasound

Critique

A large study (Editing remarks: is the study mentioned within the text or within the References? If not it cannot be used in a Question) analyzed the diagnostic utility of ^{99m}Tc scintigraphy in skeletal muscle pathology and only 50% of the patients with biopsy-confirmed inflammatory myopathy had a positive ^{99m}Tc scintigraphy.

The correct answer is C.

2. Which of the following differentiate focal myositis from focal nodular myositis?

- A. Focal myositis involves calf muscles and it presents as the muscle hypertrophy
- B. Intense contrast enhancement is found in focal myositis together with muscle atrophy, while in nodular myositis there is no contrast enhancement of the involved muscle found
- C. Level of muscle enzyme is high in focal nodular myositis, while it is within normal limits in focal myositis

Critique

Focal myositis following neurogenic damage should be differentiated from focal nodular myositis. Unlike nodular myositis which can occur in any muscle and clinically presents as nodular mass-like tumefaction, post-sciatica focal myositis involves calf muscles and it presents as a muscle hypertrophy. The levels of muscle enzymes are high in post-sciatica focal myositis, while they are within the normal limits in focal nodular myositis. On MRI 'nodular' type of lesion and intense, homogeneous, contrast enhancement of the involved muscle is present in nodular focal myositis, while in post-sciatica focal myositis diffuse muscle hypertrophy and moderate contrast enhancement is usually found.