CASE REPORT

Unilateral calf hypertrophy and focal myositis induced by S1 radiculopathy: dramatic response to steroid treatment

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SUMMARY

Unilateral enlargement of the calf in a 57-year-old man with S1 radiculopathy is described in this case report. Short tau inversion recovery-weighted imaging provided evidence of selective hypertrophy and a high signal intensity of the gastrocnemius and soleus. Histopathological analysis of the gastrocnemius revealed an endomysial inflammatory infiltrate and marked denervation lesions. Marked signs of denervation are suggestive of focal myositis secondary to neurogenic damage. The patient was treated with an oral corticosteroid (30 mg/day) and the calf hypertrophy was dramatically reduced within 5 weeks. Our case indicates that steroid therapy should be tried because it may be a potentially treatable disease.

BACKGROUND

Amyotrophy is well known as one of the clinical characteristics of peripheral neuropathy and radiculopathy, whereas muscular hypertrophy secondary to denervation is very rare. Unilateral calf hypertrophy has been reported in some patients with S1 radiculopathy,^{1–7} but the mechanism underlying the development of and appropriate treatment for this syndrome are still unclear.

CASE PRESENTATION

A 57-year-old man with S1 radiculopathy was referred to our clinic with a 1-year history of a swollen right calf. The right calf was more than 3.5 cm greater in circumference than the left calf (figure 1A). Neurological examination revealed weakness of right plantar flexion and reduced right Achilles reflex. C reactive protein was normal and serum creatine kinase was mildly elevated (506 U/L). A needle electromyography of the right

gastrocnemius revealed abnormal spontaneous activity with fibrillation potentials and positive sharp waves. High amplitude motor unit potentials with reduced recruitment were seen during contraction. Results of needle examinations of the right tibialis anterior, tibialis posterior, vastus lateralis and iliopsoas were normal. Normal results were obtained in nerve conduction studies with the exception of an increase in tibial F-wave latencies, indicating proximal slowing of nerve conduction due to S1 radiculopathy. MRI of the lumbar spine revealed spinal stenosis with compression of S1 roots. Short tau inversion recovery (STIR)-weighted MRI displayed selective hypertrophy and high signal intensity of the right gastrocnemius and soleus (figure 2A). Muscle biopsy of the lateral head of the right gastrocnemius revealed mixed myopathic and neurogenic changes, including marked variation in fibre size (hypertrophic and atrophic fibres) with frequent internal nuclei, endomysial inflammatory cell infiltrates, small angular fibres and target fibres corresponding to denervation (figure 3).

TREATMENT

Once the presence of inflammatory changes in his muscle had been confirmed, treatment with prednisolone at 30 mg/day was started.

OUTCOME AND FOLLOW-UP

After the start of the treatment, the calf hypertrophy was dramatically reduced and improvement in the strength of right plantar flexion was seen within 5 weeks (figure 1B). Serum creatine kinase

Figure 1 Unilateral calf hypertrophy (A) and after treatment with prednisone (B).

Figure 2 Short tau inversion recovery images showing hypertrophy and hypersignal of the right gastrocnemius and soleus (A) and after treatment with prednisone (B).

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Figure 3 Muscle biopsy. H&E stain ($\times 200$): endomysial inflammatory infiltrates and frequent internal nuclei (A). Nicotinamide adenine dinucleotide - tetrazolium reductase (NADH-TR) ($\times 200$): target fibres (B), ATPase 10.3 ($\times 100$): small angular fibres (C).

decreased to the normal level. His right leg condition remained stable for 3 months, and prednisolone was gradually tapered to 10 mg/day. STIR-weighted follow-up MRI at 3 months after the treatment showed reduced hypertrophy and a mild decrease in signal intensity (figure 2B).

DISCUSSION

Denervation lesions are sometimes associated with focal myositis.^{1–7} A secondary inflammatory process may occur in denervated muscle and this may lead to muscular hypertrophy. The biopsy showed increased hypertrophic fibres, possibly the result of compensatory work hypertrophy, as another cause of the calf enlargement. The presence of focal muscular hypertrophy may be helpful to search for a denervation lesion in the corresponding territory. In our case, abnormal hypersignal on STIR-weighted MRI suggested the presence of focal myositis before muscle biopsy from the gastrocnemius. In addition, STIR-weighted MRI

was useful for assessing the effectiveness of treatment with a steroid for inflammatory changes.

Although polyarteritis nodosa limited to the calf muscle has a similar MRI profile,⁸ the possibility of such polyarteritis nodosa was ruled out because C reactive protein level was normal and a calf muscle biopsy of the present case revealed no evidence of necrotising vasculitis. Focal myositis of a unilateral calf can occur with an infection,⁹ but the possibility was also excluded because of normal leucocyte level and C reactive protein level.

Since the first report of histologically confirmed focal myositis associated with S1 radiculopathy by Krendel and coworkers, only eight cases have been reported in the literature.^{1–7} Needle electromyography of the gastrocnemius revealed neurogenic abnormalities in all cases, including ours. In contrast to the findings in cases of commoner myositis, typical myogenic abnormalities were found in only one case (11.1%), although focal myositis might have played a dominant role in the pathogenesis. Surgical laminectomies had no effect in reducing calf enlargement.⁷ Six patients, including our patient, were treated with prednisone or prednisolone at a dose of 15–45 mg/day.^{1–3, 5} After the start of the treatment, calf hypertrophy regression was achieved in four cases (66.7%). MRI hypersignal regression was achieved in three of four cases (MRI findings not available in two cases). The clinical features seemed to have been caused by an immune-mediate mechanism because of the dramatic response to steroid therapy. When one is faced with a patient with this syndrome, steroid therapy should be tried because it may be a potentially treatable disease.

Learning points

- ▶ Unilateral calf hypertrophy and focal myositis associated with S1 radiculopathy are rare.
- ▶ Histopathological examination can reveal focal myositis associated with denervation lesions.
- ▶ A steroid can induce a remission clinically and improvement in MRI findings.

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Competing interests None.

Patient consent Obtained.

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