Bilateral triceps surae muscle focal myositis after a cauda equina syndrome☆

Miositis focal de tríceps sural bilateral post-síndrome de cola de caballo

Introduction

Several cases have been reported of neurogenic muscle hypertrophy associated with chronic radiculopathy,[1—4] most of them involving the gastrocnemius and/or soleus muscles in the context of ipsilateral S1 radiculopathy.[1,3,5] The aetiopathogenesis of this entity is unknown.[3,4] A minority of patients with neurogenic muscle hypertrophy present anatomical pathology findings of inflammation of unknown origin.[1,2,4—11] Such cases are described as instances of neurogenic focal myositis,[1,4,5] an infrequent entity presenting with increased volume in the region of the inflamed muscle and frequently with muscle pain.[1] Diagnosis is based on clinical data and complementary test results: moderately elevated serum CPK levels, electromyographic findings of acute denervation in the muscle groups innervated by one peripheral nerve or one nerve root in addition to a pattern of chronic denervation, MRI findings suggestive of inflammation, and anatomical pathology findings indicating inflammatory changes.[1] Corticosteroid treatment, normally at low doses and for short periods of time, has been trialled in patients with focal myositis secondary to nerve lesions.[5] Although response to this treatment varies,[11] it may induce clinical remission and improve neuroimaging findings.[10]

Clinical case

We present the case of a 35-year-old woman who in 2016 underwent emergency surgery due to cauda equina syndrome secondary to L4-L5 spinal disc herniation associated with compression of the L4, L5, and S1 roots bilaterally. After an initial period with bilateral gastrocnemius muscle atrophy, our patient progressively recovered muscle mass in both legs, although residual muscle atrophy was more marked in the right leg. Approximately one year later, she consulted due to a progressive increase in the volume of both calves, associated with bilateral calf muscle pain. The examination revealed bilateral gastrocnemius muscle hypertrophy, predominantly affecting the right leg (Fig. 1), with marked tension upon palpation of the calves, weakness in right toe flexion, painful contracture of the right gastrocnemius muscles upon plantar flexion of the right foot, and inability to toe walk. Muscle strength was otherwise normal (5/5 on the Medical Research Council scale). The Achilles reflexes was absent bilaterally, with the remaining deep tendon reflexes being normal (3/5 symmetrical). A blood analysis revealed a CPK level of 514 U/L,

Figure 1 Posterior and anterior views of our patient's legs (A). T2-weighted MRI scan, coronal (B) and axial planes (C), showing bilateral and predominantly right-sided triceps surae hypertrophy, with increased signal intensity in the right gastrocnemius muscle.

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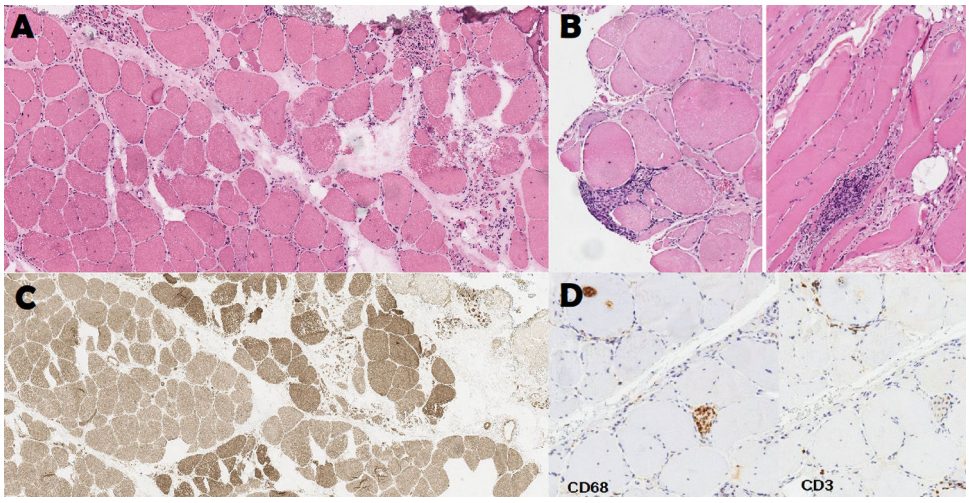

Figure 2 Anatomical pathology study of the right gastrocnemius muscle. (A) Denervation atrophy pattern. Group atrophy. Haematoxylin-eosin stain, 9×. (B) Foci of endomysial inflammatory infiltrate. Haematoxylin-eosin stain, 10×. (C) Fibre type grouping. ATPase pH 9.4 stain, 6×. Macrophages in the endomysium and within fibres. CD3+ lymphocytes in the endomysium and surrounding fibres. Immunohistochemical staining of CD68 and CD3, 10×.

with normal CRP and ESR values. The patient tested negative for autoimmune myositis. An electromyoneurography study revealed fibrillations and fasciculations in the gastrocnemius and tibialis anterior muscles bilaterally, although predominantly in the right leg. Muscle MRI of both legs revealed hypertrophy of the bilateral triceps surae, as compared to the remaining muscles. Hypertrophy was more marked in the right leg, showing increased signal intensity on T2-weighted sequences, suggesting diffuse muscle oedema (Fig. 1). A biopsy study of the right gastrocnemius muscles revealed small endomysial inflammatory foci, mainly composed of CD3+ lymphocytes and macrophages, compatible with focal myositis (Fig. 2). After a month of treatment with oral prednisone dosed at 15 mg/day followed by 2 months with the same treatment on alternate days, the patient reported improvements in pain when walking, and decreases were observed in the volume of the left calf and in muscle tension upon palpation of both legs. CPK levels decreased to 256 U/L.

Discussion

We present the first published case of bilateral denervating focal myositis. The lack of systemic symptoms or involvement of any other muscle group, together with the history of cauda equina syndrome, led us to rule out an underlying systemic process and suggested 2 concomitant processes of neurogenic focal myositis. Although MRI did not reveal muscle oedema in the left triceps surae, and the biopsy study revealed pathological findings in the right gastrocnemius muscle, the patient presented bilateral symptoms and response to corticosteroid therapy was more marked on the left side. The fact that such an infrequent process presented bilaterally and was circumscribed to the triceps surae muscles in a patient with lesions to several nerve roots suggests that some individuals and certain muscles may have a predisposition for this condition. As in previously reported cases, our patient responded to corticosteroid therapy, which improved pain and muscle tension and decreased CPK levels.

Conflicts of interest

The authors have no conflicts of interest to declare.

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The diagnosis process of Collet-Sicard syndrome caused by skull base fracture: A case report

El proceso de diagnóstico del síndrome de Collet-Sicard por fractura de la base del cráneo: reporte de un caso

Sr. Editor:

Collet-Sicard syndrome caused by trauma is rare.[1] Vocal cord paralysis (VCP), difficulty swallowing, loss of taste in the posterior third of the tongue, tongue muscle and sternocleidomastoid muscle atrophy and paralysis are the typical clinical manifestations.[2] The patient complains to the doctor only about hoarseness and difficulty swallowing. They will ignore the change in taste and muscle. So some doctors misdiagnose it as simple VCP.

The female patient is 35 years old. The patient was unconscious when she fell from a height of 9 meters on June 24, 2020. The Glasgow Coma Scale score was 8/15 (E2, V1, M5). Computed tomography (CT) showed that occipital fractures on both sides and fractures of left transverse process of atlas. She underwent tracheal intubation twice because she removed the tube by herself. Her consciousness was clear and removed the tube on June 26. She was dysphonic, difficulty swallowing and given a nasogastric tube after swallowing assessment. Neurotrophic therapy was applied when the damage of laryngeal nerve was considered. She accepted swallowing rehabilitation and acupuncture treatment. The electronic fiber laryngoscope (EFL) showed complete bilateral vocal cord paralysis on July 23 (Fig. 1A). The hoarseness of the patient began to improve significantly on July 29. We found the tongue of patient was tilted to the left and the posterior third of the tongue had decreased taste on July 30. The EFL indicated that the right vocal cord was normal, and the left vocal cord was still completely paralyzed on August 21 (Fig. 1B). The patient passed the swallowing assessment successfully and the nasogastric tube was removed on August 28. One month later, the patient still had a hoarse voice, decreased taste, and a crooked tongue. The difficulty swallowing was disappeared. The EFL still showed complete paralysis of the left vocal cord (Fig. 1C).

The most common cause of Collet-Sicard syndrome is tumor metastasis of skull base, followed by vascular disease and trauma.[3] Collet-Sicard syndrome caused by skull base fracture had only been reported 8 times in English literature. The cranial nerves IX—XII near the jugular foramen are damaged when the skull base fractured.[3] Some scholars speculated that the pathogenesis was nerve edema or bone fragments directly compressing the nerve.[4,5] Since hoarseness and difficulty swallowing are the main symptoms, it is easy for doctors to misdiagnose it as simple VCP. Symptoms of VCP are hoarseness, difficulty swallowing and difficulty breathing.[6] Both tracheal intubation and trauma can cause VCP. Arytenoid dislocation or recurrent laryngeal nerve damage is the main cause of VCP after tracheal intubation.[7] EFL can be used to observe arytenoid cartilage to determine whether it is dislocated. In this case, the patient received tracheal intubations twice in a coma. So the tracheal intubation was first considered as the cause of VCP. The anesthesiologist reported that the tracheal intubations went smoothly. The EFL did not indicate arytenoid dislocation. Therefore, the anesthesiologist disagreed with the speculation that tracheal intubation caused VCP. Trauma can also lead to VCP. CT showed the left transverse process of atlas was fractured (Fig. 1D). But the transverse and longitudinal diameters of the atlas are large, the atlas fractures rarely cause VCP. VCP is possible exist when the atlas has a Jefferson fracture.[8,9] In this case, the degree of atlas fractures was mild. So it was not the responsible of this disease. We were confused until we discovered the tongue of patient was tilted to the left (Fig. 1G). And then, we tested the patient's sense of taste. The patient told us that she lost the taste in the posterior third of the tongue. It was a typical clinical manifestation of cranial nerve IX injury. Both cranial nerve IX and X are damaged at the same time, there will be hoarseness and difficulty swallowing. Cranial nerve XII injury will cause atrophy of the ipsilateral tongue muscles, and the tongue will be deflected to the affected side. There was no clinical manifestation of cranial nerve XI damage in this patient. We suspected that the cranial nerve XI damage might be mild, or the course of the disease might be short and had not been manifest. CT indicated that bilateral skull base fractures (Fig. 1E and F). Collet-Sicard syndrome was diagnosed successfully.

In conclusion, when patients with head trauma have hoarseness and difficulty swallowing, CT examination of the skull base must be performed. The doctor must check the