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Neurogenic muscle hypertrophy as an uncommon case of the calf enlargement — a case report and literature review

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Abstract

Unilateral limb edema is a common diagnostic problem. Apart from the most common pathologies, i.e. chronic venous insufficiency and lymphedema, differential diagnosis should include unusual causes of asymmetric extremity edema. We presented a case of 41-year-old man with suspicion of lymphedema of the left calf, who reported to a lymphology clinic. We discuss subsequent steps of the diagnostic procedure in case of the calf edema. In our patient, neurogenic muscle hypertrophy was found to be the cause of the calf enlargement. The diagnosis was confirmed by results of ultrasound, magnetic resonance, computed tomography, lymphoscintigraphy and electromyography examinations. Neurogenic muscle hypertrophy is a very rare and unusual cause of the calf enlargement. Nevertheless, it should be taken into account in the differential diagnosis.

Key words: lymphedema, neurogenic muscle hypertrophy, radiculopathy

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Introduction

Unilateral limb edema is a common diagnostic problem, usually consulted by vascular disease specialists. It is usually defined as a palpable thickening of the limb circumference caused by increased amounts of the interstitial fluid [1]; however, this definition does not include less common causes of an increase in limb circumference, e.g. as a result of the overgrowth of anatomical structures (e.g. muscles).

Undoubtedly, one of the most common causes of unilateral edema of the lower extremities is chronic venous insufficiency, which affects almost 50% of women and 40% of men in Poland. A frequently observed cause in everyday practice is also lymphedema, defined as an abnormal accumulation of the interstitial fluid and fibrous-lipid tissue mainly in the subcutaneous tissue due to oncological treatment, trauma, infection or congenital lymphatic system abnormalities. Apart from the two most common causes, the differential diagnosis should take into account other numerous causes of unilateral edema of the lower limb [2], listed in Table 1.

Apart from meticulous physical examination, ultrasound is very helpful in establishing the diagnosis, as it allows visualization of a vast majority of the venous and arterial systems disorders and obtaining information about the location of the edema (suprafascial or subfascial). The choice of further imaging methods (computed tomography, magnetic resonance imaging, lymphoscintigraphy, angiography) in clinical practice depends on the result of physical and ultrasound examinations.

We present a case of a 41-year-old man with neurogenic calf muscle hypertrophy who was originally referred to an angiologist with the suspicion of lymphedema. We discuss subsequent steps of the

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Table 1. Causes of unilateral lower extremity edema, based on [2], modified

Venous	Primary venous disease Post-thrombotic syndrome Iliac vein compression (May-Thurner syndrome) Deep vein thrombosis Superficial thrombophlebitis Klippel-Trénaunay syndrome Venous adventitial cystic disease
Musculosceletal	Ruptured Baker's cyst Ruptured leg muscle Sprain/strain Static foot disorders Fracture
Lymphatic	Lymphedema
Arterial	Intramuscular hematoma Muscular infarction (particularly in diabetes) Vascular malformation Critical limb ischemia (consequence of leg position) Compartment syndrome
Miscellaneous	Infection (bacterial, parasitic) Radiation Mass/tumor Insect/animal bites Complex regional pain syndrome type I Atrophy/hypertrophy Overgrowth syndromes Granulomatous myositis Amyloidosis

diagnostic procedure that enabled the final diagnosis of a rare cause of limb enlargement.

Case report

A 41-year-old man after surgical treatment of the left iliac bone fracture more than a decade earlier, with suspected left leg edema reported to a lymphology clinic. Since the fracture, the patient observed edema and an increase of the calf circumference. He also complained of alternating numbness, particularly of the left foot, for a few years. On physical examination, circumferences of the left thigh and calf were 3–4 cm larger than in contralateral extremities, numerous telangiectasias and small varicose veins were observed in the ankle area. On doppler ultrasound normal arterial flow was confirmed and venous thrombosis was excluded, there were signs of the great saphenous vein insufficiency and its varicose veins, the popliteal vein did not collapse completely in compression test, the subcutaneous tissue hypertrophy were not present in the thighs and calves. Computed tomography (CT) did not reveal any filling defects in the common femoral veins, external iliac veins, internal iliac veins, common iliac veins and in the visible portion of the inferior vena cava.

Post-thrombotic syndrome and lymphovenous edema of the left lower extremity was suspected. Further diagnostic investigations, i.a. lymphoscintigraphy, were scheduled, and the patient was instructed to continue compression therapy and reduce his body weight.

Lymphoscintigraphy revealed normal iliac and inguinal lymph nodes and an increased number of the superficial lymphatic vessels on the thighs and calves bilaterally (Fig. 1). Magnetic resonance imaging (MRI) revealed signs of left calf hypertrophy, particularly of the medial and lateral heads of the gastrocnemius (Fig. 2). No focal abnormalities or post-contrast enhancement were demonstrated. The signal from bone and muscle structures was unchanged. A discrete zone of edematous lesions of the adipose tissue was disclosed just below the inferior pole of the lateral head of the gastrocnemius. In addition, moderate varicose veins of the left calf were observed, particularly those originating from the great saphenous vein. The right calf was unaffected and had normal muscle and bone structures, with no focal lesions and no areas of abnormal post-contrast enhancement. Based on clinical presentation and the diagnostic procedures performed, hypertrophy of the left calf muscle was diagnosed and the patient was referred for further tests.

Electromyography (EMG) revealed reduced amplitudes of motor potentials in the left peroneal and tibial nerves and prolonged F wave latencies. Sensory conduction of the sural nerve and motor conduction of the femoral nerve were within normal limits. The results supported the diagnosis of axonal sciatic nerve injury.

CT of the lumbosacral spine showed slight midline protrusion of the L5/S1 intervertebral disc with no significant compression of nerves, the L4/L5 intervertebral disc protruding towards the back moderately asymmetrically, more on the left side, with thecal sac and left nerve root indentation, and moderate hypertrophic changes of L4/L5 and L5/S1 facet joints. No signs of central or foraminal stenosis were observed.

In our patient, neurogenic muscle hypertrophy was found to be the cause of the calf enlargement. The patient started physiotherapy.

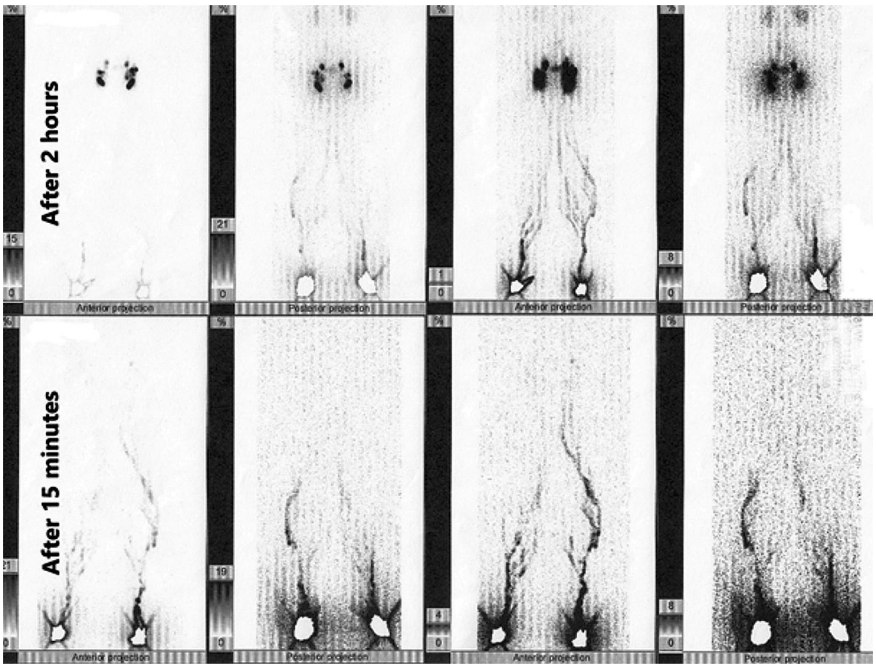

Figure 1. Lymphoscintigraphy. Normal visualization of the iliac and inguinal lymph nodes, increased number of superficial lymph vessels of the thighs and calves bilaterally

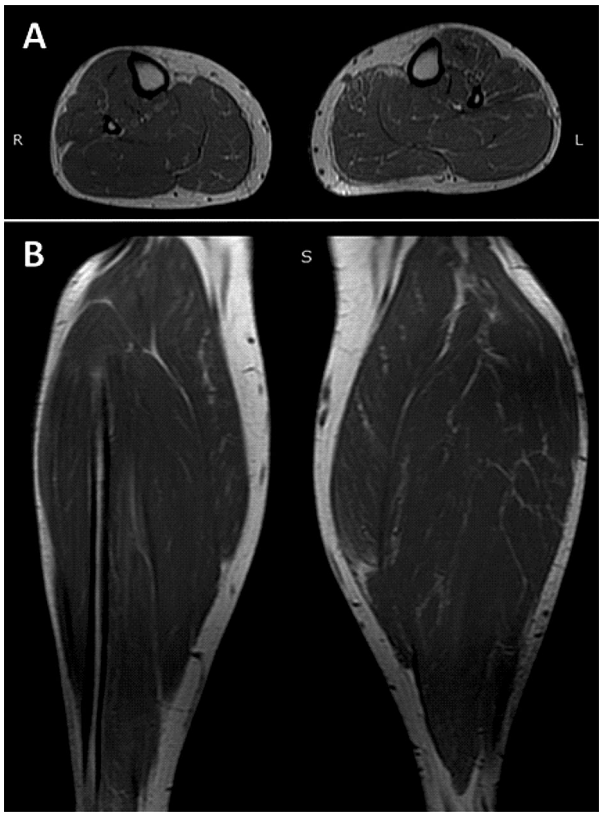

Figure 2. Magnetic resonance imaging (MRI) in transverse (A) and coronal (B) plane. Muscle hypertrophy of the left calf, particularly the medial and lateral head of the gastrocnemius muscle; the skin, subcutaneous tissue, and bones of similar size bilaterally, normal

Discussion

In a patient with unilateral edema of the lower limb, the angiologist first of all would search for pathologies of the venous, arterial and lymphatic system, probably taking into account also the most important traumatic causes. By means of ultrasound examination it is possible to rule out the majority of venous and arterial disorders. Moreover, the thickness of the subcutaneous tissue and its echogeneity may be premises of possible lymphedema, but the results of lymphoscintigraphy is important for establishment of the latter diagnosis (normal lymphoscintigraphy does not exclude lymphedema). In the presented patient, no clinically significant vascular pathologies were found, and the thickness of the skin and subcutaneous tissue was similar on both sides. Therefore, the patient was referred for an MRI, which showed the hypertrophy of the muscles of the calf.

Muscle hypertrophy in neurogenic disorders is unusual finding, as a rule in these conditions is muscle atrophy [3]. Neurogenic muscle hypertrophy, referred to as "denervation hypertrophy", was first described in the 19th century, and is caused mainly by compression of the nerve root, thus most commonly affects the calf muscles (78% of all reported cases) [4], including the gastrocnemius muscle or all muscles of the posterolateral lower leg compartment [5]. In a review of the available literature, comprising less than 30 cases, Zabel et al. [6] showed that neurogenic muscle hypertrophy is usually manifested by painful calf enlargement in males

at the age between 32 and 60, with a previous history of low back pain and sciatica, and that the average time from the onset of the trigger factor to the development of the hypertrophy was 7.3 years. In the vast majority of cases, the cause was a damage to the S1 root [5, 7–10], single cases also reported involvement of the L5 [4] or L4 [11] root or even the cervical spine [12]. In the presented case, at the L4/L5 and L5/S1 levels discopathic lesions were visualized, and the EMG examination confirmed axonal damage to the left sciatic nerve.

The pathomechanism of muscle hypertrophy is not completely clear and is certainly multifactorial. In radiculopathies may occur complex repetitive discharges (CRDs, observed as denervation signs in EMG [7]), which explains some, but not all, cases of calf hypertrophy secondary to L5 or S1 radiculopathy. When carbamazepine or botulinum toxin are administered, positive motor activity is not only reduced or stopped, but also the accompanying muscle hypertrophy is reduced, that additionally supports this hypothesis [13]. In addition to above mechanism, other possible explanations for the pathogenesis of neurogenic hypertrophy have also been proposed: work load — the workload of healthy muscle fibers remaining after damage and stretching and release of growth factors [8]. In addition, attempts have been made to explain the pathomechanism of focal myositis as a continuum of changes induced by chronic stimulation [14]. If chronic stimulation is the trigger for hypertrophy, it is conceivable that as the size of the fiber increases, splitting and then necrosis occur, either because the overgrown fiber exceeds its nutrient supply, or simply because of overstimulation; necrosis is a stimulus that attracts inflammatory cells, especially macrophages [14].

The treatment of neurogenic muscle hypertrophy is not well-established. It can be surgical (microdiscectomy) or conservative, including described as an effective injection of botulinum toxin [4, 8]. Physiotherapy plays an important role as well.

Conclusions

Detailed physical examination remains an essential part of our routine practice, because slowly changing muscle size may go unnoticed by the patient and may be omitted in the medical history [13]. The clinician should be aware that isolated muscle hypertrophy may be a sign of partial damage to peripheral nerves or nerve roots and is likely to occur in any skeletal muscle [11].

Unilateral calf edema should be considered as a rare symptom of lumbosacral radiculopathy. When suspected — history and physical examination, lumbar spine imaging should be performed in conjunction with EMG. Upon confirmation of radiculopathy that correlates with anatomical compression, this rare association should be suspected. However, the remaining non-invasive tests for other most common causes of unilateral calf enlargement, including doppler ultrasound, should not be delayed [15].

Neurogenic muscle hypertrophy is an uncommon, but well-documented phenomenon that physiotherapists should also be aware of, because patients with chronic muscle imbalance due to neuropathic disorders are often referred for a physiological evaluation [16].

Conflict of interest

None.

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