Diabetic Amyotrophy co-occurring with treatment induced neuropathy of diabetes (TIND): a case report

Amiotrofia Diabética co-ocorrendo com neuropatia do diabetes induzida pelo tratamento (TIND): relato de caso

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ABSTRACT
Diabetic amyotrophy is a rare condition in diabetic patients, usually associated with important weight loss. In the present case, we show a rare presentation of diabetic amyotrophy phenotype in the context of treatment induced neuropathy of diabetes neuropathy (TIND) after a strict glycemic control in a patient with long-standing poor controlled diabetes and without related weight loss. Even though it’s an uncommon presentation, because of diabetes high prevalence worldwide the knowledge of its complications is important to the adequate treatment and follow-up to improve patients’ quality of life.

Keywords: diabetic amyotrophy, treatment induced diabetic neuropathy, Bruns-Garland Syndrome, Diabetic lumbosacral radiculoplexus neuropathy, case report.

RESUMO
A amiotrofia diabética é uma condição rara em pacientes diabéticos, geralmente associada a importante perda de peso. No presente caso, mostramos uma rara apresentação do fenótipo de amiotrofia diabética no contexto da neuropatia da neuropatia diabética induzida por tratamento (TIND) após um controle glicêmico rigoroso em um paciente com diabetes mal controlado de longa data e sem perda de peso relacionada. Apesar de ser uma apresentação incomum, devido à alta prevalência do diabetes em todo o mundo, o conhecimento de suas complicações é importante para o tratamento e acompanhamento adequado para melhorar a qualidade de vida dos pacientes.

Palavras-chave: amiotrofia diabética, neuropatia diabética induzida por tratamento, síndrome de Bruns-Garland, Neuropatia radiculoplexa lombossacral diabética, relato de caso.

1 INTRODUCTION
Diabetic amyotrophy, also known as Bruns-Garland syndrome and diabetic lumbosacral radiculoplexus neuropathy, is a rare condition that affects approximately 1% of patients with type 2 diabetes mellitus (DM)1. It is characterized by diffuse pain, weakness and areflexia that typically affects the lower limbs and is asymmetric, usually related to a significant weight loss.
It may occur in a prediabetic state or may follow tight glycemic control in someone with newly diagnosed diabetes\(^2\).

TIND is a recent definition for an iatrogenic condition concerning neurologic complications of insulin therapy. In 1933, a woman was diagnosed with diabetes and insulin therapy was initiated, right after she developed severe pain. This is the first description of TIND in the medical literature\(^3\).

Even though these a rare complications, diabetes affects 463 million people worldwide, and is expected to affect 700 million by 2045, and its neurologic complications are to become more frequent \(^4,5,6\).

The present case shows a rare presentation of diabetic amyotrophy phenotype in the context of treatment induced neuropathy of diabetes neuropathy (TIND), following strict glycemic control in a patient with long-standing poorly controlled diabetes without weight loss.

2 CASE REPORT

A 40-year-old woman, 8 weeks pregnant, previously diagnosed with poorly controlled type 2 diabetes for 12 years using insulin, was admitted for glycemic control. Upon admission, glycated hemoglobin (HbA1c) was 12.1%, ophthalmological evaluation ruled out retinopathy and there was no proteinuria. Gestational ultrasound evidenced caudal regression syndrome, corroborating poor glycemic control. She was discharged after 4 weeks of strict glycemic control, with a new HbA1c of 6%, and rapidly evolved with mild low back pain, burning and tingling sensations in lower limbs, including feet. These symptoms were worse at night and when there was exposure to cold. She progressed with weakness in the lower limbs, worse on the right, with predominant difficulty in climbing stairs and to raise up from chairs. She also presented with axial hyperhidrosis, postprandial fullness, urinary urgency and lipothymia, characteristics of dysautonomia. She was readmitted for diagnostic investigation and pain control, and specialized evaluation by the Neurology service was requested.

On the second hospitalization, the examination showed a body mass index of 27, blood pressure 140/90mmHg supine and standing, and heart rate 82 beats/minute supine and 84 beats/minute standing. There was a slight atrophy of the right leg (FIGURE 1). Strength was preserved in the upper limbs. According to the MRC (Medical Research Council), strength was grade 4 for hip flexion, flexion, extension and adduction of the legs, dorsiflexion of the halluces and eversion of the feet, being worse on the right. Tendi\(n\)ous reflexes were globally hypoactive and ankles reflexes were absent. There was light touch hypoesthesia to the left knee, reduced
vibration sensation to the knees and absent vibration to the ankles. The rest of examination was normal.

Considering the diagnostic hypothesis of diabetic amyotrophy induced by strict glycemic control, further investigations were carried out. Cerebrospinal fluid showed high protein levels of 115 mg/dL (normal 15-45 mg/dL), without pleocytosis. Computed tomography (CT) of the lumbar spine and pelvis were performed to rule out compressive causes, showing only lumbar degenerative features. Electroneuromyography showed positive sharp waves and fibrillation potentials in the lumbar paravertebral musculature, and motor units of increased amplitude and neurogenic recruitment in the vastus medialis, vastus lateralis and rectus femoris muscles bilaterally, in addition to findings compatible with sensory-motor polyneuropathy with axonal and distal predominance affecting the four limbs. Laboratory tests including autoimmunity panel, vitamin B12 level, thyroid function and infectious serologies (HIV, syphilis, viral hepatitis) were all negative. A new ophthalmological evaluation showed proliferative diabetic retinopathy. Proteinuria of 278 mg was found in 24h urine sample, both findings typically described in the context of TIND.

Considering the clinical history and complementary exams, the diagnosis of diabetic amyotrophy after strict glycemic control was made, associated with findings suggestive of TIND. The patient was discharged with optimized pain treatment and intensive motor physical therapy, and was reevaluated after 10 weeks, showing significant improvement of weakness and sensory complains.

3 DISCUSSION

Diabetic amyotrophy was first reported in 1890, and even after 131 years of scientific development, there is still no established definition about the pathophysiology of this condition, that typically affects men aged over 50 years with type 2 diabetes mellitus. Proposed mechanisms include ischemic injury, metabolic disfunction and inflammation.

The striking clinical presentation is acute onset severe pain in the hip and thigh, followed by asymmetric leg weakness, that can become symmetrical, and leg atrophy. Typically, pain and weakness begin in the proximal region of the lower limb and can progress distally and to contralateral limb. There is often significant associated weight loss, in addition to sensory changes such as paresthesia and allodynia. CSF shows elevated protein with normal cellularity. Neurophysiological studies show a reduction in the amplitudes of sensory and motor action potentials, with possible prolongation of F wave latencies and a slight reduction in motor conduction velocities, and electromyography typically shows denervation including
paravertebral and thoracic muscles\textsuperscript{7}. Treatment should focus on adequate glycemic control, pain control and physical therapy in the most severe cases\textsuperscript{8}.

Our case report describes diabetic amyotrophy in a young female patient (<50 years), with long-standing poorly controlled diabetes and no weight loss, that evolved immediately after a strict glycemic control.

Glycemic control with a 2-point drop of HbA1c in 3 months leads to a 10\% risk of TIND, while a 5-point drop in 3 months presents a risk of over 90\%\textsuperscript{4}. Classically, TIND manifests as small fiber neuropathy, with predominantly painful and autonomic findings, and evidence of microvascular damage (retinopathy, proteinuria)\textsuperscript{4}. Our patient had a significant drop in HbA1c, from 12.1\% to 6\%, reflecting a reduction of 6.1 points in 1 month, in addition to rapid development of proliferative diabetic retinopathy and proteinuria (although pregnant, she didn’t have it one month before glycemic drop), findings suggestive of concomitant autonomic dysfunction (orthostatic intolerance, postprandial fullness, axial hyperhidrosis and urinary urgency) and acute neuropathic pain in lower limbs with allodynia, all features expected in TIND. Such data suggests that diabetic amyotrophy may be a phenotype that occurs in the context of TIND.

4 CONCLUSION

To our best knowledge, this is the first case report of diabetic amyotrophy following strict glycemic control and concomitant with clinical findings of TIND, in a patient with long-standing poorly controlled type 2 diabetes, without precipitating weight loss. Diabetic amyotrophy clinical presentation possibly being triggered by aggressive treatment of diabetes is essential for the correct diagnosis and clinical management, both of which are essential to improve the quality of life of these patients.
REFERENCES


ANNEXES

FIGURE 1 – Pictures showing the patient on the second hospitalization, presented with a slight atrophy of the right leg at inspection.