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ACUTE AXONAL POLYNEUROPATHY WITH PREDOMINANT PROXIMAL INVOLVEMENT

An uncommon neurological complication of bariatric surgery

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ABSTRACT - Bariatric surgery is frequently indicated in the treatment of morbid obesity. Previously unreported complications have been associated to this surgery; among them, neurological complications have gained attention. We report the case of a 25-year-old man submitted to gastric surgery for treatment of morbid obesity who developed, two months after surgery, acute proximal weakness in lower limbs. The electroneuromyography revealed axonal peripheral polyneuropathy with predominant proximal involvement. After treatment with immunoglobulin and vitamin supplementation, rapid clinical and neurophysiologic recovery was observed. We describe the clinical and electroneuromyographic features of this case, stressing the difficulty of initial diagnosis, particularly in the differential diagnosis with Guillain-Barré syndrome. We discuss the importance of nutritional follow-up and the eventual indication of routine vitamin supplementation in these patients.

KEY WORDS: bariatric surgery, polyneuropathy, neurological complications, vitamin supplementation.

Polineuropatia axonal aguda com acometimento proximal predominante: manifestação neurológica incomum de cirurgia bariátrica

RESUMO - A cirurgia bariátrica é freqüentemente indicada no tratamento da obesidade mórbida. Complicações previamente não relatadas têm sido associadas a essa cirurgia; dentre estas, as complicações neurológicas têm recebido destaque. Relatamos o caso de um homem de 25 anos de idade submetido a cirurgia gástrica para tratamento de obesidade mórbida que desenvolveu, dois meses após a cirurgia, fraqueza de predomínio proximal nos membros inferiores, de instalação aguda. A eletroneuromiografia demonstrou polineuropatia periférica axonal nos membros inferiores, de predomínio proximal. Após tratamento com imunoglobulina e suplementação vitamínica, apresentou rápida melhora clínica e neurofisiológica. Descrevemos as características clínicas e eletroneuromiográficas desse caso, destacando a dificuldade diagnóstica inicial, particularmente com relação ao diagnóstico diferencial com síndrome de Guillain-Barré. Discutimos a importância de acompanhamento nutricional e a eventual indicação de suplementação vitamínica de rotina nesses pacientes.

PALAVRAS-CHAVE: cirurgia bariátrica, polineuropatia, complicações neurológicas, suplementação vitamínica.

Gastric surgery is frequently indicated in the treatment of morbid obesity. Neurological complications of gastric surgery for morbid obesity - bariatric surgery (BS) - are increasingly recognized. These include peripheral neuropathy (PN), myotonic syndrome, myelopathy, burning feet syndrome, lumbosacral plexopathy and Wernicke-Korsakoff encephalopathy[1,2]. The most common neurological complication described after BS is PN[1,2]. Among the involvement of peripheral nerves, the distal polyneuropathy is the most common presentation, followed by mononeuropathies, including meralgia paresthetica, and radiculoplexus neuropathy. Guillain-Barré syndrome has also been reported[1,3]. The etiology of peripheral neuropathy after BS is probably multifactorial and different in each subgroup described above. It appears that

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nutritional deficiencies may play the most important role in pathogenesis[1,2,4,5].

The purpose of this paper is to describe an unreported presentation of peripheral polyneuropathy after BS and review the literature concerning peripheral nerves involvement in this particular condition. We report the case of a 25-year-old male who developed an acute symmetrical axonal polyneuropathy with predominant proximal involvement. The initial clinical and electroneuromyografic picture led to difficulties in diagnosis, particularly concerning differential diagnosis with initial Guillain-Barré syndrome.

CASE

A 25-year-old male underwent a successful laparoscopic gastroplasty on January 2005 for morbid obesity (weight 130 kg, Body Mass Index [BMI] 48). After surgery he lost 21 kg within six weeks and tolerated a semi-liquid diet without vomiting. He did not receive vitamin supplementation. About the end of the first month after surgery, he developed paresthesias in medial aspects of both thighs, progressing to involve both feet later on the course.

One week before hospitalization, at the second month after the surgery, he noticed some weakness mainly in climbing up stairs and some difficulty in getting up from a sitting position. This weakness progressed in days to complete incapacity to get up from a sitting position without assistance, which motivated hospitalization. He also complained of pain in legs. He denied any symptoms on the upper limbs. General physical examination was normal, apart from obesity (BMI=38). There was no clinical or laboratorial sign of malnutrition. Severe weakness at lower limbs was noticed, mainly at proximal sites. Tendon reflexes were normal in the arms, but decreased in the inferior limbs, with absence of Achilles reflexes. Plantar responses were flexor. Hypoesthesia on touch and pin prick in both legs and feet was present. Laboratory investigation was within normal ranges, including complete blood cell count, CK, hepatic enzymes, renal function, total serum proteins, albumin, vitamin B12, folate, serum transferrin and seric iron. The cerebral spinal fluid (CSF) analysis revealed normal results. Vitamins B6 (pyridoxine) and B1 (thiamine) concentrations were under the lower limit of normal ranges according to the laboratory method used (2.9 ng/mL, normal 3.3-26.0 ng/L and <7 nmol/mL, normal 9-44 nmol/L respectively).

On admission, the first electroneuromyography (EMG) revealed sensorimotor polyneuropathy at lower limbs. The amplitudes of both femoral compound muscle action potentials (CMAPs) and sural sensory nerve action potentials (SNAPs) were severely diminished. The H-reflexes from the soleus muscles were absent and F-waves shortest latency was prolonged (32 ms for ulnar nerve and 59 ms for tibial nerve; these values were considered abnormal when compared to those obtained in the subsequent EMG examination, which were 25 ms for ulnar nerve and 45 ms for tibial nerve). On needle electromyogram there were few fibrillations and positive sharp waves at lower limbs (proximal and distal muscles) but rare motor unit action potentials (MUAPs) at thigh muscles. The needle examination of lombosacral paraspinal muscles was normal. Signs of chronic reinnervation were observed in distal muscles of upper and lower limbs.

The patient received adequate doses of intravenous immunoglobulin for five days, as well as vitamin supplementation (B1 and B6). He slowly recovered strength and became able to walk without assistance over the next few weeks, due to improvement of proximal muscle weakness. Following hospital discharge, results of serum antibodies showed no abnormalities: antibodies against gangliosides (GM1, GM1 Asialo and GD1b) and anti-MAG dosage were within normal ranges. He was submitted to a second EMG, four months after the first exam, which confirmed a sensorimotor axonal distal polyneuropathy, with almost recovered femoral CMAPs amplitudes and persistence of decreased sural and superficial peroneal SNAPs amplitudes. The H-reflexes were still absent. The F-wave shortest latency was normal (25 ms and 45 ms for ulnar and tibial nerves respectively). There were few fibrillations and positive sharp waves at proximal and distal lower limbs and distal upper limbs with chronic reinervation MUAPs. As in the first EMG, the examination of lombosacral paraspinal muscles was normal.

DISCUSSION

Bariatric surgery is now widely indicated in the treatment of morbid obesity. The rise of BS brought along with it a variety of previously unreported complications. Among these, neurological complications, although uncommon[2], have recently gained attention[1,2,6]. The most commonly described neurological complications associated to BS are PN and Wernicke-Korsakoff encephalopathy[3,7-12].

Wernicke's encephalopathy is most commonly associated with malnutrition in chronic alcoholism, but it has been described in the literature as a complication of BS, frequently associated with persistent vomiting. It occurs as a result of thiamine deficiency and can be prevented by vitamin supplementation[7,9,10,12].

Peripheral neuropathy is the most commonly described neurological complication of BS[1]. Nutritional deficiencies may play the most important role in the pathogenesis of PN, although each case must be evaluated individually[2,4,7]. It has not been established whether the development of PN after BS for morbid obesity may be attributed to a specific vitamin deficiency[1,2,5,13].

Some authors believe that other factors, such as acute protein-caloric malnutrition secondary to markedly insufficient food intake and/or severe vomiting in combination with vitamin deficiency[7], or a

marked catabolism of fat and/or loss of carnitine[13] may also be involved in the pathogenesis of neurological complications after BS. Rapid weight loss after BS has been associated to vitamins deficiencies (vitamin B12, thiamine and folate)[6]. In the case reported the patient did not have persistent vomiting but had low serum concentration of thiamine and vitamin B6 and normal levels of vitamin B12 and folate. These facts suggest that the rapid weight loss was crucial for the vitamin deficiency in our patient.

A recent controlled study of PN after BS identified risk factors for development of PN, more often observed in these patients than in obese patients submitted to cholecystectomy[1]. Of the 435 patients who had BS, 16% developed PN, compared to only 3% in the cholecystectomy group ($p<0.001$). Most important risk factors included rate and absolute amount of weight loss, prolonged gastrointestinal symptoms, not having a nutritional specialized follow-up after BS, postoperative surgical complications and jejunoileal bypass. There were no differences in the serum concentrations of vitamin B12 and folate between the two groups. Other vitamin levels were not routinely measured. In our patient, the excessively rapid weight loss in addition to the fact that the patient was not having a vitamin supplementation probably contributed to the development of PN.

Different clinical patterns of PN after BS are described in the literature. A review of case reports identified 60 patients with PN after BS. Among these, the most common presentation was peripheral polyneuropathy (67%), followed by mononeuropathies (30%), mainly meralgia paresthetica[3]. However, peroneal palsy has also been reported after weight loss[14].

Thaisetthawatkul et al. described three distinct clinical patterns of PN after BS: sensory-predominant polyneuropathy, mononeuropathy and radiculoplexus neuropathy. Electrophysiological studies of patients with clinical diagnosis of peripheral polyneuropathy showed large fiber involvement and no evidence of demyelination. A minority of patients with normal EMG had clinically small-fiber neuropathies. All patients had symmetric sensory symptoms and signs and some had distal motor weakness involving hands and feet. The mononeuropathy group had mainly asymmetric involvement at common sites of entrapment (median neuropathy at wrist in most of the patients). In the radiculoplexus neuropathy group, the symptoms began asymmetrically and remained unilateral in almost all patients. EMG demonstrated axonal pattern involving the roots, plexus and nerves[1].

Our patient had acute onset of prominent weakness in proximal muscles of lower limbs and clinical symptoms and signs of distal peripheral polyneuropathy. The markedly decreased amplitude of both femoral CAMPs and prolonged F-waves latencies, demonstrated by EMG, raised the possibility of conduction block due to demyelination, involving proximal nerve segments, which might suggest the hypothesis of initial Guillain-Barré syndrome. However, further clinical course was not compatible with this diagnosis. The distal sensory-predominant polyneuropathy consistent with axonal disorder, also demonstrated by EMG, has been clearly described in the literature[2,12].

A similar case was reported by Nascimento et al. They described a patient submitted to BS for morbid obesity which developed acute and severe onset of lower limb proximal weakness and pain. This patient had thermo-algesic hypoesthesia with T10 sensory level. The thoraco-lumbar spine magnetic resonance was normal. The authors concluded that the clinical and EMG findings were compatible with an acute neuronopathy or sensorimotor axonopathy and a possible myeloradiculoneuropathy. The patient was treated with vitamins supplementation and methylprednisolone, and gradually recovered strength[15].

Feit et al. described two cases of ataxia, chorea and polyneuropathy after BS and extensive demyelination of nerve fibers has been seen at autopsy in one case[13]. Ishibashi et al. described three cases of acute axonal polyneuropathy associated to Wernicke-Korsakoff syndrome in which neuropathic symptoms and signs rapidly improved after thiamine supplementation. These three cases followed gastric surgery other than BS. Sural nerve biopsies in two patients revealed mild axonal degeneration with scattered myelin ovoid formation. The authors proposed three different mechanisms for the axonal dysfunction: distal conduction block due to demyelination, distal axonal regeneration and physiological conduction failure on the axolemma[16].

In our case, after treatment with adequate vitamin supplementation (B1 and B6 vitamin) and immunoglobulin, the patient rapidly recovered the proximal muscle strength. Repeated EMG after four months, showing almost normal amplitude for bilateral femoral CAMPs and normal F-waves latencies, suggests that there was proximal nerve involvement, rapidly reversible. Clinical improvement occurred earlier than expected considering a "pure" axonal motor degeneration. This improvement of proximal motor

function could be, in part, attributable to other pathologic features as conduction block due to demyelination or conduction failure on the axolemma, such as postulated by Ishibashi et al.[16]. In such scenario, t reatment with immunoglobulin could have contributed to improvement, although it is not possible to confirm this hypothesis.

In conclusion, this patient had acute axonal sensorimotor polyneuropathy with predominant proximal involvement as a complication of BS. This uncommon neurological complication may be due to rapid weight loss and vitamin deficiency. Initial clinical and neurophysiologic characteristics may pose difficulties to correct diagnosis. We emphasize that adequate nutritional orientation after surgery may diminish this specific neurological complication. Routine vitamin supplementation might be useful in these patients, although this remain to be established.

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