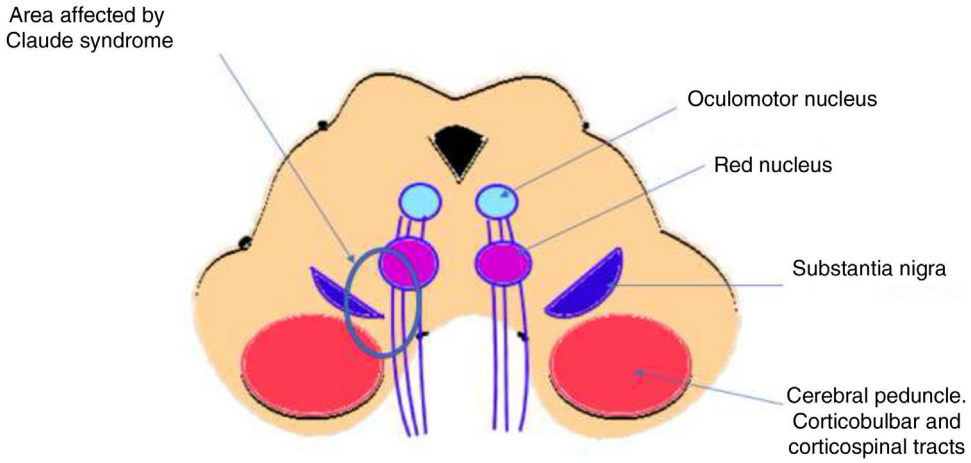

Fig. 2 Claude syndrome is caused by the involvement of the third cranial nerve, red nucleus, and cerebral peduncle.

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Amyotrophic neuralgia of atypical presentation associated with exposure to a hepatitis B vaccine☆

Neuralgia amiotrófica de presentación atípica relacionada con exposición a vacuna frente a hepatitis B

Dear Editor:

Amyotrophic neuralgia is a brachial plexopathy characterised by intense shoulder pain of acute or subacute onset, progressing for hours or days, with subsequent onset of weakness and atrophy of the affected muscles. However, this classic phenotype is only observed in two-thirds of patients.[1] The specific pathophysiological mechanism involved remains unknown. The most probable hypothesis is an immune response developing in patients with a genetic and mechanical predisposition, triggered by a precipitating factor.[2] Reports of cases after vaccination against hepatitis B virus are scarce.[3,4]

We describe the case of a 21-year-old woman reporting distal weakness and progressive loss of muscle mass in the left hand 4 weeks after vaccination with surface antigen against hepatitis B virus. The examination performed in our clinic 2 months after vaccination revealed weakness in the interosseous muscles (3+/5), fourth and fifth finger flexion (4/5), and finger extension (4/5), with amyotrophy of the interosseous muscles and hypothenar eminence of the left hand; all other results were normal, with preserved sensitivity and muscle stretch reflexes (2/4). The electrophysiological study showed decreased compound motor action potential amplitude in the left cubital and radial nerves, with fibrillations, positive waves, and motor unit potentials of increased amplitude and duration in the muscles innervated by both nerves. Results of the sensory nerve conduction study were normal. A cervical 1.5 T MRI scan with neutral and maximal flexion positions obtained sagittal and axial T1- and T2-weighted sequences ruling out Hirayama disease,[5] and no other pathological findings. An MRI scan

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of the brachial plexus showed no lesion. A metabolic study was performed, including determination of vitamin B_{12}, folic acid, and methylmalonic acid levels; total protein test; antinuclear and anti-DNA antibodies; aldolase; TSH; thyroperoxidase and antithyroglobulin antibodies; antiganglioside antibodies; and a blood count; all results were normal. Serology tests for syphilis and *Borrelia* were negative. Final diagnosis was left idiopathic brachial plexopathy. The patient was referred to rehabilitation, with interosseous muscle weakness (4+/5) persisting after 2 years of progression.

Amyotrophic neuralgia is an infrequent disease, with a mean incidence of 1 to 3 cases per 100 000 person-years.[1] Peak incidence occurs between the third and fifth decades of life[2]; the disease is more common in men, with a male-female ratio of 2:1.[6]

Amyotrophic neuralgia may be hereditary (10%) or idiopathic (90%). The latter form may be mediated by genetic susceptibility mechanisms, especially in recurrent forms.[7] Events with potential to induce an autoimmune response are found in 30% to 85% of patients,[2] and include viral and bacterial infections, strenuous exercise, vaccinations, surgery, pregnancy and postpartum, and even psychological stress.[5] In this context, compression and stretching of the brachial plexus due to its anatomical location may locally disrupt the blood-nerve barrier, facilitating passage of proinflammatory agents.[1]

In our patient, the only trigger factor identified was vaccination against hepatitis B virus. Since the patient presented amyotrophy 4 weeks after vaccination, the pathogenic process affecting the brachial plexus would probably have started one week earlier. Only 2 authors have described amyotrophic neuralgia associated with vaccination against the virus.[3,4] In the first report, the vaccine used was recombinant DNA.[3] The other patient was receiving other treatments as human hepatitis B immunoglobulins and chemoprophylaxis for HIV.[4] Therefore, our case is the first to present an association between vaccination with surface antigen against hepatitis B virus and amyotrophic neuralgia with no other associated factors.

The manifestation of amyotrophic neuralgia was atypical in our patient. Pain is not reported in up to 4% of cases; when it does present, the duration varies, with a mean duration of 27.5 days, although it may last less than 24 hours in 5% of cases and more than 60 days in 10%.[5] Pain is usually unilateral, with bilateral involvement in only 25% of cases, usually presenting an asymmetric pattern. The most frequently affected muscles are those innervated by the upper brachial plexus.[2] In our patient, mainly the muscles of the lower trunk were affected, with less marked clinical and electrophysiological symptoms in the muscles innervated by the radial nerve, indicating dysfunction of the posterior column. Predominant lower brachial plexus involvement is infrequent, affecting approximately 7% of female patients and 3% of male patients. Sensory symptoms manifest in 70% of patients. Our patient presented no recurrences, whereas another study reported recurrences in 25% of patients with idiopathic amyotrophic neuralgia, and in up to 75% of patients with the hereditary form.[6]

As many as 80% to 90% of patients present good recovery at 2 to 3 years.[8] However, other studies report that up to 25% of patients are not able to work after 3 years, only 10% achieve complete recovery,[6] and up to 60% may present chronic pain.[9]

Initial treatment aims to control severe pain. High doses of prednisone may be used during the first month to decrease the duration of clinical symptoms and facilitate functional recovery.[10] We did not indicate corticosteroid treatment in our patient due to the delay between symptom onset and diagnosis. After the acute phase and once pain is controlled, patients should receive multidisciplinary rehabilitation, including physical therapy, electrotherapy, and occupational therapy.[1,11]

In conclusion, amyotrophic neuralgia may manifest in an atypical form, presenting distal involvement but no pain. Early diagnosis of these less frequent forms is essential for patients to benefit from corticosteroid treatment. One possible trigger factor is vaccination with surface antigen against hepatitis B virus; therefore, adequate history taking is essential to identifying new cases associated with this vaccine.

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