



Case Report

Psychogenic movement disorder in human T-lymphotropic virus type 1 associated myelopathy



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SUMMARY

Human T-lymphotropic virus type 1 (HTLV-1)-associated myelopathy/tropical spastic paraparesis (HAM/TSP) is a chronic inflammatory disorder of the spinal cord. Acute cases of HAM/TSP and those complicated by movement disorders are rarely reported. Otherwise, psychiatric disturbances are very frequent in infected patients. It can evolve to psychogenic disorders. The case of a 46-year-old woman with acute HAM/TSP complicated by depression and psychogenic movement disorders (chorea of the hands and dystonia-like facial symptoms) is reported. Brain magnetic resonance imaging revealed non-specific small white matter lesions. The involuntary movements arose suddenly and disappeared when the patient was distracted. Two years of psychotherapy and psychiatric follow-up induced complete remission of the symptoms. The association of psychogenic movement disorders and HAM/TSP, increasing the range of neurological manifestations associated with HTLV-1, is related here. Early diagnosis of psychogenic movement disorders is very important to improve the prognosis and treatment of the two conditions, thereby improving the quality of life of HAM/TSP patients and avoiding irreversible sequelae.

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1. Introduction

Human T-lymphotropic virus type 1 (HTLV-1) is a retrovirus that affects the central nervous system, causing a disabling, chronic, and progressive inflammatory disorder of the spinal cord called HTLV-1-associated myelopathy/tropical spastic paraparesis (HAM/TSP).¹ The virus is present worldwide and is endemic in some regions, such as southwestern Japan, Sub-Saharan Africa, South America, and the Caribbean, with outbreaks in the Middle East and Australo-Melanesia.² It is estimated that five to ten million people are infected.² HAM/TSP occurs in less than 5% of infected individuals.^{1,2} HTLV-1 is also associated with psychiatric disorders such as depression and anxiety, and more rarely with movement disorders.^{3,4} There are also some reports of atypical HAM/TSP cases with rapid progression.

Psychogenic movement disorders represent neurological involuntary disturbances that cannot be explained by the presence of organic lesions in the nervous system and are generally associated with emotional alterations (psychogenic).⁵ These show a broad spectrum of manifestations such as tremor, dystonia, chorea, myoclonus, athetosis, ballism, and tics.⁵ To the authors' knowledge, there have been no reports of psychogenic movement disorders in HAM/TSP. A case of HTLV-1-associated myelopathy and depression in a patient who presented psychogenic hand chorea and jaw dystonia is reported here.

2. Case report

A 46-year-old Brazilian woman was admitted to the neuroinfection outpatient clinic of Hospital Universitário Gaffrée e Guinle (HUGG) in April 2008 due to 4 months of progressive weakness in her lower limbs. She became suddenly paraplegic and confined to a wheel chair. Her condition was diagnosed as HAM/TSP and she was treated with steroids without improvement. The patient developed

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severe depression and tried to commit suicide. She was referred for psychiatric follow-up.

In April 2011, the patient presented abrupt onset involuntary oromandibular and upper limb movements. At this time, she was taking haloperidol and chlorpromazine; both were withdrawn. A neurological examination revealed spastic paraplegia with pyramidal signs and involuntary movements of the hands (chorea-like) and jaw (dystonia-like) (Figure 1). The movements disappeared through distractibility maneuvers and sometimes while she was being interviewed. Medical consultations with a psychiatrist and psychotherapy were suggested.

Laboratory analysis showed normal routine hematological, biochemical, liver function, and vitamin B12 tests. Serological tests were negative for antinuclear antibodies, HIV, and syphilis. Anti-HTLV-1 antibodies were detected in serum and cerebrospinal fluid (CSF) by ELISA, confirmed by Western blot and real-time PCR (3.7 copies of pX/100 in peripheral blood mononuclear cells).⁴ Cervical and dorsal magnetic resonance imaging (MRI) revealed spinal cord atrophy. Brain MRI showed unspecific hyperintense signals on T2 and fluid attenuation inversion recovery (FLAIR) images, without contrast enhancement, bilaterally in the periventricular and subcortical regions. After 2 years of psychotherapy, the involuntary movements disappeared (Figure 1).

3. Discussion

HAM/TSP cases with rapid progression of severe motor deficits have been described in HTLV-1 infection, but not subsequently complicated by movement disorders (chorea and dystonia-like) associated with a psychiatric disturbance (depression).^{1,3} There are occasional reports of movement disorders associated with HTLV-1 infection.^{1,4} Spinocerebellar syndrome has been demonstrated in patients infected by HTLV-1/2, who presented loss of balance, gait

disturbance, trunk instability, and ataxia of the lower limbs, with nystagmus, dysarthria, and postural tremor. A case of HAM/TSP associated with parkinsonian syndrome has also been reported.⁴ The patient case reported here presented with dystonic and choreiform movements, which disappeared after psychiatric treatment. Neuroimaging also did not reveal any basal ganglia lesion, excluding structural conditions. Drugs such as haloperidol and chlorpromazine can cause dystonic movements.⁵ Therefore, considering that the reported case used neuroleptics and antidepressant drugs, a pharmacological etiology was the first hypothesis. However, the movements disappeared through distractibility maneuvers. In addition, after the drugs were suspended, the symptoms did not improve. These facts argue against the movement disorder being induced by drugs.

A psychogenic movement disorder is an involuntary movement that is caused by psychological or psychiatric conditions.⁵ It can affect any part of the body and may result from a somatoform disorder or may be part of a conversion disorder. In contrast to movement disorders associated with biological or structural conditions, psychogenic movement disorders often present a sudden onset with rapid progression. The implementation of motor action with the segment not affected often decreases or eliminates the psychogenic movement disorder,⁵ as observed in the patient presented here. Tremor and dystonia are the most common manifestations. In such cases, the clinical disturbance will improve or disappear with distraction in 86% of cases, as observed in the case patient. The abrupt onset, as reported here, is another frequent characteristic.⁵ Psychogenic movement disorders are difficult to diagnose. There is no single test available to confirm the diagnosis. This is an extremely important issue, mainly in individuals presenting other disorders that can be associated with a wide range of motor disabilities, such as HTLV-1 infection.



Figure 1. Movement disorders in a patient with HAM/TSP. (A) Oromandibular movements with jaw and lip deviation (dystonia-like). (B) Irregular, rapid, and random hand contractions. (C) Normal finger to nose test after remission of the symptoms.

The association of viral infection and mental disturbances has been described previously.³ Previous studies have shown HTLV infection to be associated with depression and anxiety. HAM/TSP patients more frequently have depression and anxiety than HTLV-1 carriers.³ The depression observed in these patients could be associated with the psychological impact of this incurable infection, or may be the consequence of the direct biological effect of the retrovirus infection. In the present case, the acute and severe progression of HAM/TSP causing paraplegia in 4 months contributed to the severe depression, which was characterized by a suicide attempt. The mental disturbance led to the psychogenic movement disorder. In some cases, the precipitating event can be identified.⁵ In this patient, the atypical development of HAM/TSP was the precipitating event.

Although there is no report in the literature on psychogenic movement disorders in HAM/TSP patients, this is a hypothesis to be considered due to the spontaneous improvement of the symptoms and the relationship with depression.⁵ The choreiform and dystonic spasms in the present HAM/TSP patient were triggered by a psychiatric disorder (severe depression with attempted suicide), which is consistent with psychogenic involuntary movements, as well as the abrupt onset of the symptoms, the response to distractibility maneuvers, and the exclusion of an organic cause. This represents the first report of a HAM/TSP case associated with psychogenic movement disorders, broadening the spectrum of neurological abnormalities associated with HTLV-1-infection. Furthermore, the diagnosis of this condition is extremely important in order to avoid unnecessary therapeutic measures. It may result in significant

morbidity and could have a better prognosis if the diagnosis is established early.

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Conflict of interest: No conflict of interest to declare.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.ijid.2015.11.013>.

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