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Case report

Facial Diplegia Revealing Lyme Borreliosis

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Abstract

Introduction: Diagnosis of neuroborreliosis may be difficult. Neuroborreliosis mainly results in lymphocytic meningitis and in meningoradiculitis. ***Case report:*** We report the case of a patient who developed a sudden facial diplegia, revealing neuroborreliosis proved by positive blood and cerebrospinal fluid serology. The patient had no previous history of tick bite and migrans erythema. The patient was given ceftriaxone therapy (2 g/day for 21 days), leading to resolution of all clinical symptoms. ***Conclusion***: Our report underscores that neuroborreliosis should be considered in patients exhibiting facial diplegia. Thus, Lyme serology should be performed systematically in these patients. Altogether, early management is crucial, before the onset of neurological manifestations at late stage, leading to disabling sequelae despite antibiotic therapy.

Keywords: Facial diplegia, Lyme Borreliosis, Lyme serology, neurological complications.

INTRODUCTION

Lyme Borreliosis is a zoonosis transmitted by ticks, linked to a spirochete: Borrelia burgdorferi. After inoculation of the pathogen, lyme borreliosis progresses in three stages. The primary stage is determined by the migrant erythema associated with polyadenopathies, evolving spontaneously in a favourable manner in 3 to 4 weeks [1, 2]. The secondary phase is characterized by the appearance of multi-systemic attacks, particularly joint and cardiac, due to the dissemination of the infection by hematogenic route [1, 2]. Finally, the tertiary stage is defined by the chronicity of the different localizations of infection [1-3] Neurological attacks of lyme borreliosis are mainly represented by lymphocytic meningitis and meningoradiculitis during the secondary phase [1, 2]. We report the original observation of a patient who presented a facial diplegia revealing Lyme borreliosis.

OBSERVATIONS

One patient, aged 29, with no medical or surgical history, was hospitalized for a peripheral facial graduation with a sudden onset; the rest of the neurological examination was normal, including no meningeal syndrome.

The clinical examination was also normal (figure-1). Biological tests showed: sedimentation rate: 12 mm at first hour, C-reactive protein: 0. 97mg/L, hemoglobin: 15. 1 g/dL, leukocytes: 9. 45 G/L (neutrophil polynuclear: 5. 99 G/L); blood ionogram and liver tests were normal. Blood cultures and cytobacteriological examination of urine were negative. Chest x-ray was normal. Emergency nuclear magnetic resonance (NMR) imaging of the brain was normal. Viral serologies (urian virus, rubella, cytomegalovirus, Epstein-Barr virus, adenovirus, Myxo-virus influenzae and parainfluenzae, human immunodeficiency virus) as well as syphilitic serology (TPHA, VDRL) were negative. In contrast, Lyme serology was highly positive for IgG at 220 IU, but not IgM (< 0. 8), indicating seroconversion. The immunologicalassessment (antinuclear antibody test, neutrophil anticytoplasm antibodies, cryoglobulin, cryofibrinogen) wasnegative. Lumbar puncture showed hyperproteinorachy (0. 55 g/L) with normoglycorachy (0. 65 mmol/L); cytological analysis showed 300leukocytes/mm3 (95% lymphocytes). Direct examination and cultures of cerebrospinal fluid were negative; Lyme serology in cerebrospinal fluid objective IgG at 1445 IU with a high index of intra thecal synthesis of specific immunoglobulins at 23. In the end, the diagnosis of neuroborreliosis revealed by a facial diplegia was retained. Parenteral antibiotic therapy with ceftriaxone was introduced at a daily dose of 2 grams for 21 days, allowing complete regression of all clinical manifestations (figure-2).

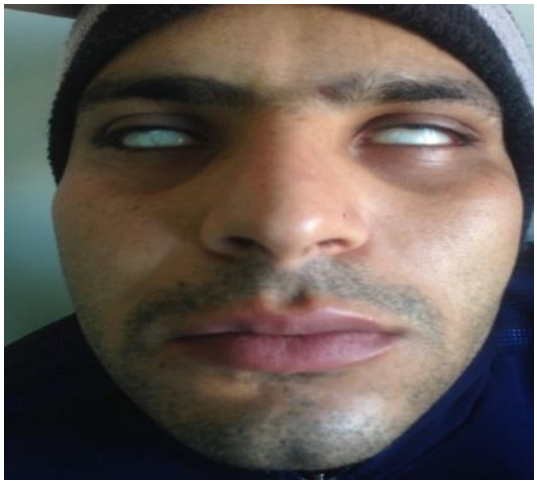

Fig-1: Before Treatment

Fig-2: Final Result

DISCUSSION

The prevalence of neurological complications of lyme borreliosis was assessed differently according to the series, being evaluated between 10 and 68% [4-6]. If they have been described at all stages, their mode of presentation is polymorphic, depending on the iroccurrence in the course of the disease [4-6].

Thus, in the primary stage of the disease, neurological manifestations generally result in headaches of varying intensity related to lymphocytic meningitis [1, 2, 4-6].

Then, neurological manifestations canappear, as in our patient, during the secondary stage of the disease. In our observation, no tick bites or migrant erythema skin lesions were found during questioning; these data support the results of a previous study involving 25 patients with lyme borreliosis complicated by neurological manifestations, which detected tick bites in 44% of cases [6]. At this stage, neurologic manifestations of lyme borreliosis are observed in 10-15% of patients [4-6]. They are essentially represented, on the one hand, by lymphocytic meningitis, usually asymptomatic (88-100% of cases) [5, 6], and on the other hand, by sensory meningoradiculitis, which are observed in 67 to 83% of neuroborreliosis cases [6-8]. Meningoradiculitis develops on average 3 months after the tick bite; they occurmainly in the lower limbs (in the metameric zone of the initial bite), resulting in tables of cruralgia or hyperalgic sciatica, with a nocturnal recrudescence often responsible for total insomnia [2, 5, 6]. Peripheral neuropathies constitute, in terms of frequency, the 2nd neurological complication during lyme borreliosis (38. 2% of cases); they are often located in the lower limbs (appearing on average 5 months after the tick bite) [2, 5, 6]. While sensory neuropathies are the most common (80%), sensory motor impairments have been reported in the form of multineuritis (10%) or proximal and distal polyneuritis (10%); these neuropathies may become complicated by motor sequelae despite antibiotic therapy [2, 5, 9, 10]. Otherneurological localizations are possible, such as cranial nerve damage (30% of cases) associated with a meningeal reaction [2, 5, 9, 10]. All cranial nerves may be affected.

In a series including 131 patients with neurologic manifestations complicating Lyme disease, there were: peripheral facial paralysis (70%),

oculomotor nerve paralysis (20%), but also cochleovestibular nerve, (7. 5%), trigeminal (22. 5%) or glossopharyngeal (2. 5%) [5]. During neuroborreliosis, facial paralysis is more Common in children than adults [1, 2, 5]. In two studies, facial paralysis was reported in 7% to 15% of adult patients with lyme borreliosis [6, 11]. If facial paralysis is mostly unilateral, it can also be bilateral, as in our patient [5].

In fact, if facial depligia are rare, our observation has the interest of emphasizing that neuroborreliosis should be systematically sought insubjects with a facial diplegia. In our observation, the cerebrospinal fluid analysis showed a lymphocytic cellular reaction and hyperproteinorachy. Djukic *et al*., [12] recently noted that patients with neuroborreliosis, compared to subjects with viral meningitis, had a higher level of lactates in the cerebrospinal fluid; this assay was performed in our patient (2.76mmol/l).

Diagnosis of facial graduation with lyme borreliosis was confirmed by positive Lyme serology in cerebrospinal fluid (associated with intra-thecal synthesis of IgG immunoglobulins) and blood. The search for a specific synthesis of IgG, IgA or IgM antibodies against Borrelia burdorferi is the diagnostic test of choice in neuroborreliosis, the positivity of which may precede that of blood serology [13-15]; the sensitivity and specificity of this test have been estimated at 75% and 97% respectively [16]. However, the specific Polymerase Chain Reaction (PCR) detection method is not recommended in practice, as it is very insensitive during neuroborreliosis, its sensitivity having been evaluated at 5% incerebrospinal fluid [17]. Brain MRI can visualize hypersignal staking contrast after injection of gadolinium at the facial nerve on T2-weighted sequences [18, 19]. In our patient, brain MRI was normal, probably due to its early onset during the first 24 hours following the onset of facial graduation. The objective of antibiotic treatment is the complete eradication of Borrelia burgdorferi, in order to avoid aggravation of symptoms and progressionto a tertiary phase [1, 2]. 3rd generation cephalosporins are the antibiotics of choice to spread across the blood-brain barrier and within the cerebrospinal fluid. According to the latest recommendations of the Société de pathologie infectieuse française, the preferred molecule is ceftriaxone at a daily dose of 2 grams for 21 to 28 days; in the case of penicillin allergy, oral treatment with doxycycline may be proposed, the efficacy of which can be super imposed on that of 3rd generation cephalosporins [2, 23, 24].

In all cases, when neuroborreliosis is suspected, antibiotic therapy should be administered as a matter of urgency before serological results are received. The prognosis for recovery from secondary phase neurological damage of lyme borreliosis is generally good under treatment. Thus, our patient showed a complete regression of facial graduation during the two weeks following the initiation of ceftriaxone therapy.

However, in a recent series, Skogman *et al*., [25] found that 13% ofpatients had sequelae of facial paralysis. In 2013, in a series of 50 patients with neurroborreliosis, Eikeland *et al*., [26] reported that the following factors were predictive of sequelae: delay in antibiotic therapy, multi-systemic involvement, lack of complete regression of clinical manifestations after 4 months of treatment; in contrast, cerebrospinal fluid abnormalities we're not a prognostic factor in these patients. Finally, the central neurological complications of lyme borreliosis in the tertiary phase are burdened with significant morbidity, and in particular sequelae despite appropriate antibiotic therapy.

CONCLUSION

A neurological picture of facial graduation must give rise to suspicion of lyme borreliosis, including in patients who have not presented, as in our observation, migrant erythema or tick bites. These data emphasize that Lyme serology should be routinely performed in these patients. In fact, therapeutic management is essential before neurological complications occur in the tertiary phase of lyme borreliosis, which are responsible for significant sequelae despite antibiotic therapy.

Competing Interests

The authors declare no competing interest.

Authors' Contributions

All authors made substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data; drafting the article or revising it critically for important intellectual content; and final approval of the version to be published.

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