CASE REPORT
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Facial Diplegia in *Plasmodium vivax* Malaria

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Background Facial diplegia has diverse etiologies, including viral and bacterial infections such as diphtheria, syphilis and Lyme disease, and also protozoal infection in very rarely cases.

Case Report A 20-year-old male patient was admitted to our hospital due to bilateral weakness of the upper and lower facial muscles. Examination revealed that the patient had a facial diplegia of the peripheral type. A peripheral blood smear demonstrated the presence of the asexual trophozoite stage of *Plasmodium vivax* with ring-form trophozoites, which led to a diagnosis of malaria. A serum work-up revealed increased IgG titers of antibodies to myelin-associated glycoprotein and ganglioside GD1b. The patient was administered antimalarial treatment, 1 week after which he showed signs of recovery. To our knowledge, this is the first case of facial diplegia after malaria infection, providing evidence that the mechanism underlying the condition is related to immune-mediated disease.

Conclusions Facial diplegia can manifest after *P. vivax* infection.

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Key Words facial diplegia, malaria.

Introduction

Facial diplegia can be induced by inflammatory, infective, traumatic, and infiltrative causes. Keane found that benign and self-limited syndromes were responsible for one-half of cases.[1] One of the most common underlying causes is Guillain-Barré syndrome (GBS), which itself is attributable to nondescript respiratory or gastrointestinal infections, especially from viruses. Serologic reports have found other types of bacterial infections, including mycoplasma pneumonia, Lyme disease, and *Campylobacter jejuni*.[2] Protozoal infection has also been suggested as a cause of GBS, but the incidence is reportedly very rare. We report herein facial diplegia in a male patient with malaria.

Case Report

A 20-year-old male patient was admitted to our hospital in July 2008, due to bilateral weakness of the upper and lower facial muscles associated with facial palsy. He reported a high fever and headache that had lasted for 3 weeks. Four days before admission, he had experienced difficulty closing his eyelids and the next day felt facial weakness. On admission, his temperature was 38.3℃. Neurological examination revealed his pupils to be isocoric with prompt reflexes to both direct and indirect light; his extraocular movements were full and there was no nystagmus. Bilateral facial weakness of the peripheral type was noted: the patient was unable to close his eyes tightly, smile, blow, or whistle. He did not complain of hyperacusis and his gag reflex was normal. Further examination revealed normal power in all four limbs. The patient's deep tendon reflexes were normal and his sensory functions were not impaired.

A peripheral blood smear demonstrated the presence of the asexual trophozoite stage of the protozoal parasite *Plasmodium vivax*, with ring-form trophozoites. The patient's hemoglobin was 13.8 g/dL and his white cell count was 3.92×10^3/μL, comprising 59.7% neutrophils. Serum levels of glucose, sodium, and potassium were 116 mg/dL, 137 mM, and 4.7 mM/L, respectively. The patient's cerebrospinal fluid (CSF) was clear in appearance, cells were absent, and levels of protein and glucose were 34.7 mg/dL and 66 mg/dL, respectively. Serologic tests for leptospiral infection, Lyme disease, herpes simplex, varicella zoster, and Epstein-Barr virus were negative in the serum and CSF. His IgG index was 0.03 and his erythrocyte sedimentation rate was 38 mm/h. High titers of antibodies to

myelin-associated glycoprotein (MAG) were found to be 1,200 BTU (normal value, <1,000 BTU). Titers of ganglioside GM1 IgG and IgM were not increased, but anti-GD1b IgG titers were increased to 1,180 BTU and anti-GD1b IgM titers were within the normal range.

Brain MRI with contrast revealed enhancement of the right facial nerve. Ultrasound evaluation of the abdomen revealed splenomegaly. Nerve conduction studies of the extremities produced normal results. Sensory evoked potentials and visual evoked potentials were normal, and there were no observable R1 or R2 responses in the blink reflex test. On facial EMG, denervation potentials founded in the bilateral orbicularis oculi, orbicularis oris, and frontalis muscles. Normal EMG recordings were observed in the bilateral masseter muscles. Antimalarial therapy with hydroxychloroquine sulfate was commenced, and the patient showed signs of recovery after 1 week.

Discussion

Bilateral facial weakness is present in less than 1% of facial palsy patients, with facial paralysis resulting from a wide range of congenital abnormalities, cranial trauma, and infections. In the present case, facial diplegia developed suddenly after fever. All laboratory data, including serologic tests, were normal except for the anti-MAG and anti-GD1b titers in CSF. These results suggest that the cause of the facial diplegia is related to an immune-mediated condition, such as GBS.

GBS is a form of peripheral neuropathy that is commonly triggered by infections and is related to immune-mediated mechanisms. A large number of infectious agents that mediate GBS have been identified, but protozoal parasitic infection has been reported in only a limited number of patients with *Leishmania donovani, P. falciparum,* and *P. vivax* malaria.[3-6] To our knowledge, only 22 cases of GBS after malarial infection have been reported, and this is the first case of facial diplegia after a malarial infection.

The mechanisms underlying the malaria-induced development of GBS are unknown. The malaria parasite may damage the peripheral nerves by vascular occlusion, thus causing anoxic stagnation in the vasa nervosum, which leads to temporary demyelination and recovery after disappearance of the parasitemia and establishment of normal blood flow in the vasa nervosum. Immune-mediated damage is believed to be the cause of GBS. In malaria, asexual-stage infections are accompanied by the release of cytokines and other immunological mediators.[7] In our case, immunologic studies revealed that anti-MAG and anti-GD1b titers were elevated, possibly indicating that the development of GBS after malaria infection is an immune-mediated disease rather than the result of direct damage by parasites.[8] Drug-induced polyneuropathy is unlikely, since paralysis occurred in our case before commencing treatment.

P. falciparum malaria is associated with a poorer outcome compared with other types of malarial infection.[5,6,9] In other reports, almost half of patients developed respiratory and bulbar paralysis, which ultimately leads to death. These findings are worse than those usually encountered in GBS.[1] However, several *P. vivax* malaria patients, including our case, have survived and did not develop bulbar involvement.[10] The relevant patient population is too small to allow us to draw a reliable conclusion, but we can assume that *P. vivax* malaria can be complicated by GBS, which has a low incidence of bulbar affection and a better clinical outcome than *P. falciparum* malaria.

Conflicts of Interest

The authors have no financial conflicts of interest.

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