case of microsporidiosis manifested as mutiple intracranial lesions separated in space and time, and neurological and radiological findings were improved with albendazole administration. A 33-year-old man presented with headache, fever, and dysphasia. His consciousness was clear. Neurological examination revealed acalculia, agraphia, and homonymous hemianopsia. He had a past history of febrile convulsive seizures of unknown cause until 14-years-old, but no history of immunodeficiency. T1-weighted magnetic resonance (MR) imaging showed a hypointense lesion with a hyperintense part, and ring-like enhancement with gadolinium-diethylenetriaminepenta-acetic acid (Gd-DTPA), in the left temporal lobe. T2-weighted and diffusion-weighted MR imaging showed the lesion surrounded by moderate hyperintense areas. He underwent gross total resection of the lesion. Histological examination demonstrated intracellular clusters of small basophilic spore-like bodies in the astrocytes, suggestive of microsporidia-infected astrocytes. However, immunohistochemical, polymerase chain reaction, and serological analyses failed to confirm the definitive diagnosis of microsporidiosis, so that he received no further treatment. Three years later, he presented with sensory disturbance in the left side of his face and left cerebellar ataxia, followed by fever, abnormal sensation in the left side of his face, and aggravated ataxia of the left upper and lower extremities on day 10 after admission. T1-weighted MR imaging with Gd-DTPA showed an enhanced lesion with irregular margin in the left cerebellar peduncle. T2-weighted MR imaging showed a diffuse hyperintense region around the lesion. Cerebrospinal fluid culture, serological analysis for autoimmune disease, and thoracic, abdominal, and pelvic computed tomography and 18Ffluorodeoxyglucose-positron emission tomography detected no abnormalities such as cancers or other lesions in the extracranial organs. No definitive diagnosis was obtained, but recurrence of microsporidiosis was the most probable cause. Administration of albendazole (600 mg/day) was started on day 15, because of rapid neurological and radiological deterioration. This treatment resulted in clinical improvement and disappearance of the lesion on MR imaging after daily administration for 4 weeks. He was discharged on foot with moderate sensory disturbance in the left side of the face and ataxia. Based on the clinical course and negative findings, the final diagnosis was microsporidiosis. This case suggests that microsporidiosis in the central nervous system can persist even in immunocompetent patients without involvement of any other organs, and that albendazole administration is likely to be effective ¹⁾.

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