

— Case Report —

Aseptic Meningoencephalitis Complicated by Retrobulbar Neuritis

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A 25-year-old man was admitted to our hospital for testing and follow-up of aseptic meningoencephalitis. After admission to our hospital, the patient suddenly complained of visual field disorder and a decrease of visual acuity in the right eye. We diagnosed aseptic meningitis complicated by retrobulbar neuritis using MRI. We immediately initiated weekly steroid pulse therapy, and eventually, marked improvement in visual acuity and the visual field disorder was observed without any late effects.

Key words: aseptic meningoencephalitis, retrobulbar neuritis, MRI, steroid pulse therapy

Introduction

Aseptic meningitis is often complicated by encephalitis and myelitis (1, 2). However, retrobulbar neuritis is rare as a complication of aseptic meningoencephalitis. The causative viruses of aseptic meningitis are mumps virus, varicella virus, and others (3). In this article, we described an interesting case in which aseptic meningitis was complicated by retrobulbar neuritis, and which was successfully treated with steroid pulse therapy.

Case Report

A 25-year-old male with fever and headache was admitted to a private hospital on May 9, 1996, in Nagasaki, Japan. Family history and past history revealed nothing

relevant to the disease. In early May 1996, the patient abruptly developed disturbed consciousness (Japan coma scale: 20), and was admitted to the hospital on May 12, 1996. On physical examination, his height was 173 cm, and he weighed 73 kg. His blood pressure was 110/69 mmHg and pulse rate 72/min and regular. He had no anemia or jaundice. Superficial lymph nodes were not palpable. Cardiac sounds were normal. No added sounds were detected in the lung. The abdomen was markedly distended and diffusely tender. Liver and spleen were not palpable. On neurological examination, motor paralysis and elevation of Sandoz's reflex of the left arm were noted. However, there was no sensory paralysis or Foxe's reflex. Motor, sensory and reflex results in the left leg were normal. In the peripheral blood test, the total WBC count was 13500 cells/mm³, and the differential count revealed normal distribution of neutrophils and lymphocytes, with a slight increase (9%) in eosinophils. The CSF taken on admission was turbid, and the total white blood cell count was 1325/3 (Table. 1) with a predominance (81%) of lymphocytes. Protein level was 77.5 mg/dl (normal, <40 mg/dl), and sugar level was 58 mg/dl (normal range, 40-50 mg/dl). In Gram's and AFB stain smear of CSF, neither bacteria nor acid fast bacilli were seen. CSF culture did not grow any bacteria, including *M. tuberculosis*. Polymerase chain reaction (PCR) test for *M. tuberculosis* was negative. The cryptococcal antigen

Table 1. Serial findings of cerebrospinal fluids

	May/12	May/15	May/21	June/5
Appearance	turbid	clear	clear	clear
Cells (/mm ³)	1325/3	828/3	224/3	86/3
(Lymphocyte) (%)	81.1	93	83	95
(Granulocyte) (%)	18.9	7	18	5
Total protein (mg/dl)	77.5	29	29	27
Sugar (mg/dl)	58.0	86	63	58

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test in serum was also negative. Serum tests for Herpes simplex virus type 1 and type 2 antibodies were negative. Although there was motor paralysis of the left arm, brain CT scan and magnetic resonance imaging (MRI) revealed no active or demyelination findings. Also, the number of cells was 1353/3 in this patient's CSF finding. In Merritt's neurology textbook, it is written that the number of cells is 15~250/mm³ in multiple sclerosis (4). Therefore, meningoencephalitis was diagnosed, and the patient was referred and admitted to our hospital on May 27, 1996, for further management. On May 28, in a peripheral blood test, the distribution of eosinophils increased to 10%, and became normal on June 3. In biochemical tests, the CRP was 0.28 mg/dl, and the GPT showed a slight increase (45 IU/l) over the normal value (0-35 IU/l). The clinical course is presented in Fig. 1.

As shown in Fig. 1, the CSF findings and motor paralysis of the left arm improved. However, after admission to our department, a visual field disorder and a decrease in visual acuity suddenly appeared in the right eye on May 29. The patient was referred to the department of ophthalmology of our hospital on the same day, and the visual acuity of the right eye was 0.3. On May 30, body temperature increased to 38.3°C, and the patient complained of pain deep in the orbit. On reexamination by the department of ophthalmology, the visual acuity of the right eye was further decreased to 0.03. The right eye also showed concentric narrowings of the visual field. The left eye was normal. The progressive decrease in visual acuity and the orbital MRI performed at the time of appearance of visual field disorder (May 29) are shown in Fig. 2. The ocular fundus of each

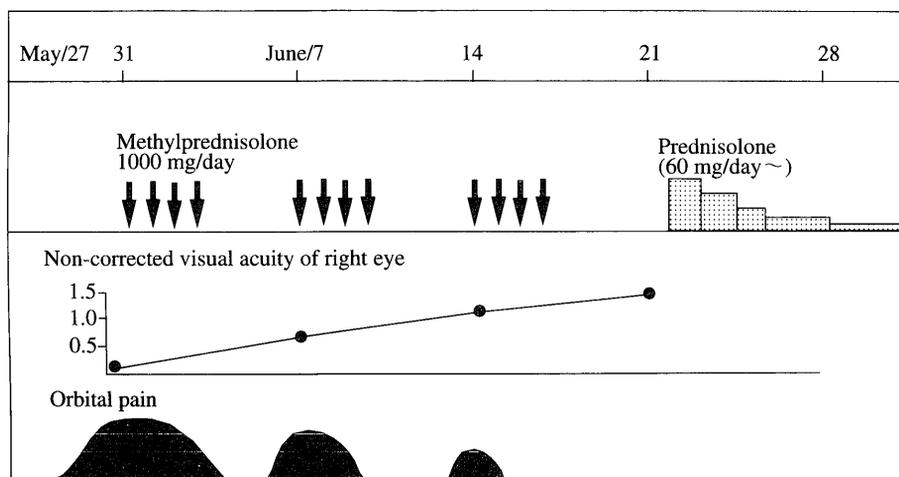


Figure 1. Clinical course showing the effects of corticosteroids on visual acuity and orbital pain.

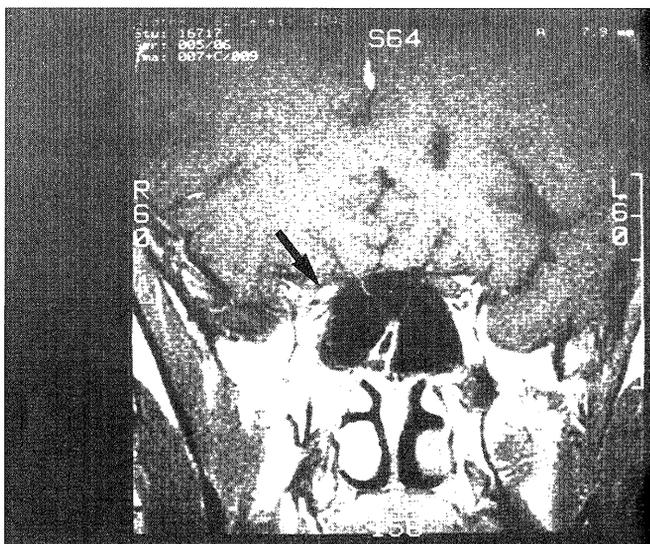


Figure 2. Magnetic resonance imaging (MRI) shows that inflammation was found between the posterior area of the right optic canal, and the anterior area of the optic chiasm as a high signal intensity area.

eye was normal, but the light reflex was weak in the right eye. In MRI, a high-signal area on T1 after Gd's contrast was found between the posterior area of the right optic canal and the anterior area of the optic chiasm. On the basis of these findings, the patient was diagnosed as having retrobulbar neuritis. For treatment of retrobulbar neuritis, weekly steroid pulse therapy was started on May 31. One course of this therapy consisted of dripinfusion of 1000 mg of methylprednisolone for 4 days, followed by no administration of steroid for 3 days. When 3 courses were performed by the middle of June, there was gradual improvement in visual acuity and in the visual field of the right eye, and during the third course, pain in the deep orbit also disappeared. At the time of ophthalmologic examination performed on June 20, the right visual acuity was 1.5, and the right visual field was also almost normal, but the physiological blind spot was somewhat larger. Orbital MRI performed on the same day revealed persistence of inflammation in the posterior area of the right optic canal. However,

clinical symptoms improved markedly, and 60 mg of prednisolone was initiated and reduced gradually on June 20. On July 15, the steroid was withdrawn, and the subsequent course was favorable without any recurrence.

Discussion

We have a question about diagnosing multiple sclerosis and retrobulbar neuritis. Unfortunately, we didn't reserve the patient's cerebrospinal fluids, so we couldn't investigate myelin binding protein, which is important in multiple sclerosis diagnosis. We diagnosed retrobulbar neuritis from the fact that there were no high density areas in the brain CT and from the information in Merritt's textbook.

Reports of aseptic meningoencephalitis complicated by retrobulbar neuritis are rare. Our survey of the Japanese and English literature revealed that only 12 such cases were reported during the 15-year period from 1980 through 1995. In 3 of these reports, the causative viruses were rubella virus, mumps virus and type-1 HSV (5-7). In our case, tests for Coxsackie virus, Echovirus and HSV were negative. Serological tests for rubella & mumps viruses were not determined. Children and young people are mainly affected by retrobulbar neuritis as a complication of aseptic meningitis (8). Postinfective allergy, rather than direct viral attack, is thought to be the mechanism of retrobulbar neuritis as a complication in aseptic meningitis, because the complication occurs in the recovery stage of meningitis in most cases (7). Immune complex is formed in type III allergy, and this may cause autoimmune diseases. In the present case, involvement of postinfective allergy is assumed because retrobulbar neuritis appeared 19 days after the onset of meningitis. There are few reports of other types of neuritis occurring during the recovery stage of meningitis. There are reports on the suspected involvement of postinfective allergy in vestibular neuritis and peripheral neuritis (9-11). A case of Hunt syndrome occurring in the early stage of meningitis has been reported (11). This was thought to be due to direct invasion by varicella zoster virus, which has affinity to the seventh cranial nerve.

The treatment of retrobulbar neuritis has not yet been established. An optic neuritis study group in the US performed pulse therapy (administration of methylprednisolone at a dose of 1000 mg for 3 days) followed by gradual dose reduction over an 11-day period. They reported that recovery of visual acuity was achieved in 62% and improvement in the visual field was observed in 80% of the patients (12). They also reported

the recurrence of retrobulbar neuritis in about 13% of patients 6 to 24 months after the treatment. In Japan, there is only one report, in which 45 mg of prednisolone was administered initially and subsequent gradual dose reduction failed to improve visual acuity, but pulse therapy consisting of 750 mg of methylprednisolone for 3 days resulted in some improvement (13). In the present case, we rapidly diagnosed retrobulbar neuritis using MRI, and immediately initiated weekly pulse therapy, and eventually, marked improvement in visual acuity and visual field disorder were observed without any adverse effects.

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