Abstract

A 30-year-old man with bronchial asthma complained of horizontal diplopia. Partial oculomotor nerve palsy with restrictions of elevation and adduction, and mydriasis was observed in the left eye. Cranial magnetic resonance imaging demonstrated an infarct lesion in the territory of the left superior median mesencephalic branch of the posterior cerebral artery. Based on bronchial asthma, hypereosinophilia, mononeuropathy multiplex, pulmonary eosinophilia and positive perinuclear antineutrophil cytoplasmic antibody in the serum, the patient was diagnosed as having Churg-Strauss syndrome. This is the first case of oculomotor nerve palsy due to midbrain infarction associated with Churg-Strauss syndrome.

Case Report

A 30-year-old man with a 20-year history of bronchial asthma complained of horizontal diplopia on April 2003. He was admitted to Nihon University Itabashi Hospital in July 2003. Corrected visual acuity was 1.2 in both eyes. Funduscopic findings did not demonstrate any abnormalities. Two prisms of esotropia were observed in the primary position. Slight restrictions of elevation and adduction were detected in the left eye (Fig. 1). Pupil diameter was 3.5 mm in the right eye and 4.5 mm in the left eye under light conditions. Both light and near reflexes were sluggish in the left eye. Palpebral fissure was 10 mm in each eye. On neurological examination, there were no other abnormalities except for left partial oculomotor nerve palsy and hypesthesia in the territory of bilateral sural nerves. Complete blood counts demonstrated a white blood cell count of 31,900/mm$^3$ with marked hypereosinophilia (66%), an elevated erythrocyte sedimentation rate at 55 mm in the first hour, and elevation of CRP at 2.7 mg/dl. The serum perinuclear antineutrophil cytoplasmic antibody (p-ANCA) was positive at 324 enzyme-linked immunosorbent assay units. Computed tomography demonstrated several massive lesions in the right superior lobes of the lung. Pulmonary eosinophilia was confirmed based on biopsy of the lesion by bronchoscopy. Cranial magnetic resonance imaging (MRI) showed an infarct lesion in the territory of the left superior median mesencephalic branch of the posterior cerebral artery (PCA) (Fig. 2). The patient fulfilled 4 of 6 diagnostic criteria proposed for CSS by the American College of Rheumatology in 1990 (1), because bronchial asthma, hypereosinophilia, mononeuropathy multiplex and pulmonary eosinophilia were detected. Furthermore, based on the elevated serum titer of p-ANCA, the patient was diagnosed as having CSS (8).

Key words: allergic granulomatous angiitis, Churg-Strauss syndrome, magnetic resonance imaging, midbrain infarction, oculomotor nerve, perinuclear antineutrophil cytoplasmic antibody (p-ANCA)
Third Nerve and Churg-Strauss Syndrome

Figure 1. Hess chart. Slight restrictions of elevation and adduction were detected in the left eye.

Figure 2. Cranial magnetic resonance imaging on T2-weighted image (A: Axial image, B: Enlargement of A). An infarct lesion in the territory of the left superior median mesencephalic branch of the posterior cerebral artery was demonstrated (arrow).
Methylprednisolone 1,000 mg/day×3 days was administered, followed by prednisolone 80 mg/day×14 days. Left oculomotor nerve palsy, hypereosinophilia, pulmonary eosinophilia, and bilateral sural nerve involvements improved. However, the ischemic lesion in the midbrain remained unchanged. The patient remained asymptomatic under prednisolone at 15 mg/day.

**Discussion**

In CSS, various central nervous system involvements such as cerebrovascular diseases, cranial nerve palsies, disturbance of consciousness, convulsion and encephalopathy have been observed with a frequency of 6.2–27% (3, 9–11). Cerebrovascular diseases were previously reported to be a complication in 3.1–6.4% (2, 3, 5). Sehgal et al (5) noted that cerebral infarction was observed in 3 of 47 patients with CSS. In addition, cranial nerve palsies showed an incidence of 3.1–16% (2–4), and the most common symptom was ischemic optic neuropathy (4, 12–15). Previous reports have stated that oculomotor (2, 6, 7), trochlear (13,14), trigeminal (16), abducens (2), facial (2, 4, 17), vagus (17) and hypoglossal (17) nerves were involved in CSS patients. Among these, only 3 cases of oculomotor nerve palsy were reported (2, 6, 7). In one of these 3 cases, peripheral ischemia of the oculomotor nerve was suspected (7). In the other 2 cases, neither the pathogenesis of oculomotor nerve palsy nor the neuroimaging findings was described (2, 6). In our patient, MRI demonstrated an infarct lesion in the territory of the superior median mesencephalic branch of the PCA. This is the first case of oculomotor nerve palsy due to midbrain infarction associated with CSS. We considered that infarction was induced by angiitis.

**References**