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Secondary SUNCT syndrome caused by viral meningitis

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Sirs: The syndrome of short-lasting unilateral neuralgiform headaches with conjunctival injection and tearing (SUNCT) is classified as belonging to trigeminal autonomic cephalalgias according to the International Classification of Headache Disorders, 2nd edition (Table 1) [1].

In 1991, Bussone first reported a case of secondary SUNCT syndrome, which was caused by an arteriovenous malformation at the cerebellopontine angle. Since then, secondary SUNCT syndrome has been reported following cavernous hemangioma of the brainstem [2], vertebral basilar artery malformation [3], and pituitary gland prolactinoma [4–6]; in most cases, the lesions were in the posterior fossa and the pituitary glands. We report a rare case of secondary SUNCT syndrome caused by viral meningitis.

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Case report

A 49-year-old male had been complaining of general fatigue since July 31, 2007. On August 3, he developed a headache in the forehead bilaterally, and he had a slight fever on the next day. At 3:00 a.m. on August 3, a stabbing pain lasting less than 3 seconds suddenly occurred on the right side of the forehead, cheek, and jaw. He felt pain while he was awake. The pain attacks occurred at least once every two or three minutes, and at most once every 10 seconds; attacks occurred with a frequency from 100 to 200 times per day. During the attacks, he showed dacryorrhea, swelling of the eyelids of the right eye, and rhinorrhea. The patient was not able to keep still because of his severe, stabbing headache. During the daytime of August 6, the pain moved to the right occipital and right upper auricular regions. The symptoms persisted, and he was admitted on August 7. His past history and family history were not contributory.

The patient reported a stabbing headache in the right side of the forehead, cheek, and jaw, as well as in the upper auricular and occipital regions. On physical examination, he had a slight fever of 37.3 °C, but no conjunctival afflux, dacryorrhea, or rhinorrhea. His consciousness was clear, and he had no neck stiffness. He had mild hypoesthesia in the area of the first division of the right trigeminal nerve.

Routine laboratory examinations were normal except for a low uric acid level. Examination of the cerebrospinal fluid showed that the cell count was increased to 406/m^3 (all mononuclear cells), the protein level was increased to 110 mg/dl, and the glucose level was slightly decreased to 59 mg/dl (the blood glucose level was 153 mg/dl). Brain MRI and MRA revealed no abnormalities. On the thermogram (Fig. 1), the skin temperature was higher around the right orbital region than around the left orbital region, indicating that skin blood flow was increased on the right side due to decreased right sympathetic nerve function.

This patient was diagnosed as having viral meningitis, and it was decided to treat only the headache, since spontaneous improvement of the meningitis was expected. Sumatriptan was subcutaneously injected twice a day during 3 days after admission. It was dramatically effective for relieving the headache, and the effect lasted for 12 hours.

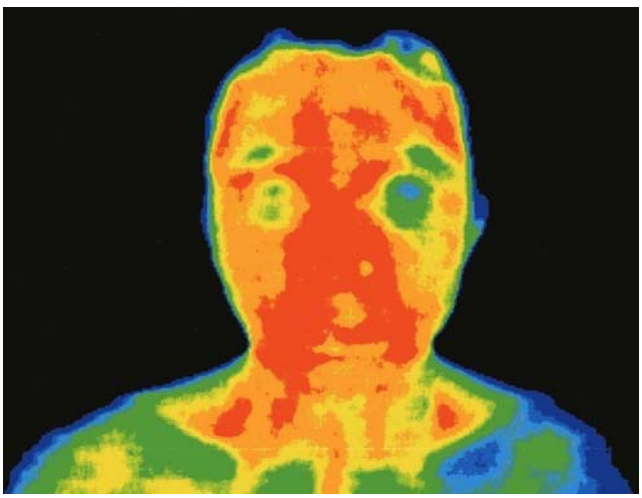

Fig. 1 The thermogram shows that the skin temperature was higher around the right orbital region than around the left side, suggesting decreased right sympathetic nerve function

Headache attacks completely disappeared on the fourth hospital day. Loxoprofen, indomethacin, and pure oxygen were ineffective for his headache. The second CSF examination was performed on August 28. The cell count had decreased to 23/mm^3, the protein level had decreased to 39 mg/dl, and the glucose level had increased to 60 mg/dl (the blood glucose level was 96 mg/dl). Pain and hypoesthesia in the area of the right first division of the trigeminal nerve disappeared with improvement of the meningitis.

The patient had not experienced a further attack of headache without any preventive drug until the time of discharge.

Discussion

The headache in this case was considered to be induced by viral meningitis. Nevertheless, the headache in this case fulfilled items B-D in the diagnostic criteria of SUNCT (Table 1) [1]; thus, this case could be considered to be secondary SUNCT syndrome. Only one similar case of meningoencephalitis [8] has been previously reported. Differential diagnoses included cluster headache and paroxysmal hemicranias, both of which were subtype of autonomic cephalalgias. In this case, the duration of headache was less than three seconds, and such a short duration did not meet the diagnostic criteria of cluster headache. Although paroxysmal hemicranias is completely prevented by indomethacin, which could not prevented headache attacks at all in this case.

Table 1 The International Headache Society 2004 diagnostic criteria of SUNCT

A	At least 20 attacks fulfilling criteria B–D
B	Attacks of unilateral orbital, supraorbital or temporal stabbing or pulsating pain lasting 5–240 seconds
C	Pain is accompanied by ipsilateral conjunctival injection and lacrimation
D	Attacks occur with a frequency from 3 to 200 per day
E	Not attributed to another disorder

The pathophysiology of the headache in this case could be as follows. First, the viral infection raised the excitability of the trigeminal nerve, especially the first division, resulting in pain around the forehead. This excitatory impulse arrived at the caudal nucleus of the trigeminal nerve to activate the superior salivary nucleus of the facial nerve. As a result, the function of parasympathetic fibers traveling with the facial nerve was activated to induce clinical signs such as dacryorrhea and rhinorrhea.

The administration of sumatriptan dramatically relieved the headache. We think that sumatriptan worked by acting on the serotonin receptor of nucleus tractus spinalis and terminating of the trigeminal nerve. Based on the thermogram results, the patient showed sympathetic hypofunction on the face. This phenomenon can be explained by compression of sympathetic nerves with perivascular inflammation due to focal vasodilation of the internal carotid artery or stimulation of parasympathetic nerves [9].

In patients suspected of having SUNCT, possible underlying diseases, such as viral meningitis, should first be ruled out.

Conflict of interest The authors declare no conflict of interest.

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