Introduction

Recurrent short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) has been defined by Sjaastad et al. [1] as a distinct and rare clinical entity. Its pathognomonic features characterized by short-lasting unilateral headache along with conjunctival erythema and tearing differ from other unilateral peri- or supraorbital pain syndromes with autonomic symptoms [2–6]. According to the criteria of the International Classification of Headache Disorders (2004) [7], the attacks occur randomly with a frequency of 3–200 per day, and last 5–240 seconds. Potential etiologies and triggers of SUNCT attacks include tumor, ischemic stroke, arteriovenous malformations, hormonal changes, anticancer therapy, and chronic active hepatitis [2, 3, 8–10]. However, there is no report indicating that head traumas lead to an increase in severity or frequency of these episodes. In this study, we report our observations on the worsening of SUNCT episodes after mild frontal head trauma in a case.

Case report

A 50-years-old man (Table 1) was admitted to our headache clinic on 8 Aug 2002 because of recurrent pain in the left peri- and supraorbital regions. His short-lasting pain episodes (<1 minute), accompanied by conjunctival injection, tearing, rhinorrhea, ptosis and periocular sweating, had started about two years earlier (in June 2000) and currently
occurred over 100 times per day. Left peri- or supraorbital pain attacks were precipitated by chewing, stress, and hot or cold food intake and touching or cold breeze on the frontal region.

Pain episodes, initially occurring 15–20 times a day, were described as blunt, pinpricking, burning-like or pressure-like in nature. He had been treated with carbamazepine (CMZ) at another institute and pain was relieved somewhat for three months, but did not completely disappear. Then the frequency of pain attacks reduced from once a day to once a week; CMZ and then oxcarbamazepine (OCZ) were administered for about two months with moderate benefit on the frequency but not on the severity of pain attacks.

In June 2002, when he was taking OCZ, the frequency of attacks exceeded 100 times per day, right after a blunt head trauma at his left frontal region with no loss of consciousness. Because of the frequent pain episodes, he was unable to talk and work. He was hospitalized on 15 Aug 2002 and underwent hematological, serological, biochemical and radiological investigations; magnetic resonance imaging (MRI) and single photon emission computed tomography (SPECT) of the cranial structures did not reveal abnormal

Table 1 Clinical course of the headache

| Institution or period | Pain score* | Frequency | Treatment | Results |
|-----------------------|-------------|-----------|-----------|---------|
| Other institutions (ambulatory) From June 2000 | 4–7 | Random 15–20/day | CMZ, 400 mg/day | Partial recovery for 3 months |
| | 3 | Once per day CMZ, 400 mg/day | OCZ, 600 mg/day | No change |
| Head trauma June 2002 | 8 | 5–10/hour (over 100/day)* | OCZ, 600 mg/day | No change |
| Our department (hospitalized) 15 August 2002 | 2–6 | Over 100/day (5–10/hour) | LTG, 200 mg/day | Frequency reduced to <10/day |
| Our department (ambulatory) 24 September 2002 | 6–9 | None (for 10 days) | Stopped by patient | Rebound |
| | 4 | 2–3/hour | LTG, 200 mg/day | Partial recovery |
| Other institution (hospitalized and ambulatory) 16 October 2002 | 6–8 | No considerable change | Prednisolone, 64 mg/day | No obvious change by time |
| | LTG, 250 mg/day | Gabapentin, 1800 mg/day |
| | TENS |
| | LTG, 250 mg/day | Topiramate, 50 mg/day |
| | Indomethacine 100 mg/day | |
| | 2–4 | <10/day | LTG, 250 mg/day | Partial recovery |
| | 3 | >10/day | Trigeminal block | At first partial recovery; then no obvious benefit |
| | | | LTG (250 mg/day) and (75 mg/day) |
| | 27 May 2003 ND | Complete recovery for 10 days | LTG, 200 mg/day | Complete recovery |
| | 16 June 2003 | >100/day | Amitriptyline, 75 mg/day | Partial recovery |
| | | | LTG, 250 mg/day | (frequency reduced to 10–20/day) |
| | | | TPM, 100 mg/day |
| | | | Methylprednisolone, 64 mg/day |

*a Determined on a visual analog scale where 0 means no pain and 10 indicates severe pain

*b Immediately after head trauma

ND, not determined; CMZ, carbamazepine; OCZ, oxcarbamazepine; LTG, lamotrigine; TENS, transcutaneous electrical stimulation
findings. Considering the increase of frequency of pain attacks and irresponsiveness to OCZ, lamotrigine (LTG) treatment was given at 200 mg/day. Pain attacks reduced remarkably from over 100 to less than 10 per day. The patient was discharged with partial recovery.

On 24 September 2002, 10 days after the patient had ceased LTG treatment because he felt completely recovered, he was readmitted due to pain episodes recurring 2- to 4-times per hour. LTG was prescribed again but did not lead to complete recovery. Therefore, he visited another institution on 16 October 2002 and was prescribed prednisolone, gabapentin (GBP), LTG and transcutaneous electrical stimulation (TENS) combination therapies with no benefit.

He visited us again on 19 December 2002; LTG and topiramate were given but no improvement was seen. Trigeminal block resulted in partial improvement while he was taking topiramate (75 mg/day) and LTG (250 mg/day), but the frequency was still over 10 times per day. Indomethacine was also given for one week with no benefit and, because of severe gastrointestinal side effects, it was discontinued. On May 27, 2003, when he has been taking LTG 250 mg/day, he visited us again claiming complete recovery for about 10 days; so, LTG dose was attenuated to 200 mg/day. He was readmitted due to intolerable pain in the same region exceeding 100 times/day on 16 June 2003, was placed on methylprednisolone (64 mg/day po), amitriptyline (75 mg/day), topiramate (100 mg/day) and LTG (250 mg/day). Partial recovery in the frequency (10–20 times per day) and severity of attacks (from intolerable into tolerable) was achieved and 9 days later he was discharged.

Discussion

This is a typical case of SUNCT syndrome with recurrent, short-lasting pain episodes on the left frontal and periorbital regions associated with conjunctival injection and tearing and rhinorrhea on the left side. Initially, these episodes occurred with a frequency of 10–20 times/day, lasting less than 1 minute. Interestingly, after left frontal head trauma, the severity of the attacks worsened and the frequency increased. Furthermore, the episodes were not responsive to high dose antiepileptic and antineuralgic treatments. Temporary relief was obtained by antiepileptic and antineuralgic treatments including nerve block, but complete recovery was not achieved.

The usual triggers of pain are chewing, touching, breeze or cold influences on the face, cold or hot drinks, overmovement of facial mimic muscles etc. In this case, the attacks started spontaneously but were triggered by these same stimuli. This case is unique in that the severity and frequency increased right after head trauma. No post-traumatic complication involving cranial structures was seen at MRI and SPECT. Thus, this patient can be considered to have idiopatic SUNCT.

Prolonged (exceeding 1 minute) and refractory attacks have been reported by Matharu et al. [11]. The severity and frequency of SUNCT in our case increased in two periods; the first period started right after the trauma and lasted about 10 weeks, and the second period lasted about two weeks. The durations of both periods were relatively longer than those reported earlier. The first period responded well to LTG but the relief was short-lasting; the second did not. Therefore, methylprednisolone was added, and, the trigeminal nerve was blocked thereafter. Only two short periods with complete recovery not exceeding two weeks could be achieved. LTG, gabapentine and topiramate are the drugs for potential use in SUNCT, and complete relief has been reported for each drug singly or combinations [11–15]. We could not obtain complete recovery although he has been still taking amitriptyline, LTG, topiramate and methylprednisolone. He has been suffering from typical SUNCT attacks at the same location with a frequency at around 10–20/day.

Our observation indicates that head trauma, even mild, should be taken into consideration in the worsening of SUNCT syndrome. The resistance to medication described in the literature is still a major problem for sufferers [11–15].

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