J Headache Pain (2004) 5:147–150
DOI 10.1007/s10194-004-0084-7

BRIEF REPORT

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Worsening of SUNCT syndrome after frontal head trauma

Received: 16 October 2003
Accepted in revised form: 13 May 2004

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Abstract SUNCT syndrome is a rare condition characterized by a short-lasting periorbital pain associated with autonomic symptoms. In this paper, a 50-year-old man with SUNCT syndrome is presented. Preexisting SUNCT syndrome in this patient worsened following mild frontal head trauma. Despite various medication regimes and trigeminal nerve block, complete recovery was not achieved at the end of one year follow-up period.

Key words SUNCT syndrome • Head trauma

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 pathogonomonic 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 periorbital sweating, had started about two years earlier (in June 2000) and currently

Table 1 Clinical course of the headache

Institution or period	Pain score[a]	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 Once a week OCZ, 300–600 mg/day		Partial recovery
Head trauma June 2002	8	5–10/hour (over 100/day)[b]	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 considerble change	Prednisolone, 64 mg/day Gabupentin, 1800 mg/day LTG, 250 mg/day TENS	No obvious change by time
Our department (hospitalized and ambulatory) 19 December 2002	2–4	<10/day	LTG, 250 mg/day Topiramate, 50 mg/day Indomethacine 100 mg/day	Partial recovery
	3	>10/day	Trigeminal block LTG (250 mg/day) and (75 mg/day)	At first partial recovery; then no obvious benefit
27 May 2003 16 June 2003	ND 5–8	Complete recovery for 10 days >100/day	LTG, 200 mg/day Amitriptyline, 75 mg/day LTG, 250 mg/day TPM, 100 mg/day Methylprednisolone, 64 mg/day	Complete recovery Partial recovery (frequency reduced to 10–20/day)

[a]Determined on a visual analog scale where 0 means no pain and 10 indicates severe pain
[b]Immediatly after head trauma
ND, not determined; *CMZ*, carbamazepine; *OCZ*, oxcarbamazepine; *LTG*, lamotrigine; *TENS*, transcutaneous electrical stimulation

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

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 fascial 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 descrbed in the literature is still a major problem for sufferers [11–15].

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