



# Intravenous Lidocain in SUNCT Treatment SUNCT'ta İntravenöz Lidokain Kullanımı

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#### Abstract

SUNCT/SUNA are rare, severe, neuralgiform headaches with high frequency. They are often considered as a primary headache type. It is called episodic or chronic according to the peridiocity. At the time of attack, it can affect the daily life of the patient such that it can keep the patient from oral feeding. Therefore, it is specifically important to relieve the pain of the patient during the headache episode. Here, we present a patient treated with IV lidocaine during an acute episode that gained sustained remission.

Keywords: Headache, SUNCT, lidocain

## Öz

SUNCT/SUNA nadir görülen, şiddetli, nevraljiform karakterde, frekansı yüksek, çoğu zaman primer bir baş ağrısı türüdür. Görülme zamanına göre epizodik ve kronik form olarak ayrılmaktadır. Atak zamanında hastanın günlük yaşamını oral beslenemeyecek kadar etkileyebilmektedir. Bu nedenle özellikle atak döneminde hastanın ağrısının hızla giderilmesi önemlidir. Biz burada günlük yaşamı ağrı nedeniyle oldukça etkilenmiş atak dönemindeki bir hastada intravenöz lidokain ile elde edilen uzamış remisyonu göstermeyi amaçladık.

Anahtar Kelimeler: Baş ağrısı, SUNCT, lidokain

## Introduction

Short-lasting unilateral neuralgiform headache with conjunctival injection and tearing/short-lasting unilateral neuralgiform headache with cranial autonomic features (SUNCT/ SUNA) is a subgroup of trigeminal autonomic cephalalgia. It is a very rare headache type characterized by unilateral, severe, periorbital or temporal pain with short-lasting autonomic features.

Although it is classified as a primary headache, it may rarely occur due to secondary causes. SUNCT/SUNA was thought to be resistant to treatment, but recently published case reports demonstrated satisfactory results that were obtained from acute and preventive therapies.

Here we report a patient with SUNCT who responded to intravenous (IV) lidocaine during the acute attack and is in remission for 8 months with prophylactic treatment.

# **Case Report**

A 77-year-old female presented with a left-sided, very severe, stabbing pain, which was localized at the periorbital and upper maxillary region with duration of 2-4 minutes. The pain was associated with ipsilateral conjunctival injection and tearing. The headache was present for ten years, and started to worsen in 2012. It was learned that the pain relapsed every 2-3 months, lasted for 1 month, and did not have seasonal rhythm. The pain, which appeared about 50 times during the day, was triggered by face washing, eating, and jaw movements.

She had regulated hypertension and diabetes mellitus. Her family history was unremarkable. The patient's blood pressure was 125/80 mmHg, heart rate was 81/min, and body temperature was  $36.1 \, ^{\circ}$ C.

The patient, who presented for the first time 5 months ago, was on carbamazepine 800 mg/day. During this period, the patient

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was treated with indomethacin and her response was evaluated. Indomethacin was increased up to 100 mg/day, but was stopped when no response was obtained. The patient met the diagnostic criteria of the International Headache Institute (IHS) in terms of SUNCT syndrome. Episodic SUNCT syndrome was considered because it relapsed every two to three months and lasted for 1 month. Pregabalin treatment was started and the dosage was increased to 2x300 mg/day. Carbamazepine treatment was gradually reduced and ceased. After a one and a half month painfree period, the pain resumed and the patient was unable to take oral medication on the second visit. Thus, IV lidocaine treatment was planned following hospitalization. The pregabalin treatment was discontinued and gabapentin was started and the dosage was increased to 3200 mg/day. Lamotrigine was added to the treatment with a dose of 200 mg/day because the pain was not completely relieved. Treatment with IV lidocaine was waived as remission was achieved with this treatment.

Two months later, the patient presented for the third time and stated that she could not eat during the last five days due to pain. Topiramate was added to the current treatment and the dosage was increased to 150 mg/day. Gabapentin was decreased to 900 mg/day because of sedation. IV lidocaine (2%) treatment was planned for the severe pain. The patient was taken to the intensive care unit and 1 mg/kg dose was loaded in 15 minutes and infusion was started at 1 mg/kg/hour dosage under continuous monitoring. The dose was increased to 1.5 mg/kg/h due to continued pain. After 30 minutes with this dose, complete painlessness was achieved. The infusion was continued for 48 hours. During this period, the patient had no pain and then was taken to a bed on a ward. Approximately 10 hours later, the patient began to have severe pain again and was re-infused for 24 hours. With a complete response, the patient was discharged with 200 mg/day lamotrigine, 150 mg/day topiramate, and 900 mg/ day gabapentin. On the 5<sup>th</sup> day after discharge, sphenopalatine ganglion blockade was performed.

## Discussion

SUNCT diagnosis is based on clinical features (Table 1). According to the disease course, it is divided into episodic and chronic (1). In terms of pain, it is the shortest and has the highest frequency among trigeminal autonomic cephalalgias. Our patient met the IHS diagnostic criteria for episodic SUNCT syndrome. Rarely, there is a secondary underlying cause, especially posterior fossa lesions (2). Magnetic resonance imaging of the brain, which was performed in order to exclude this condition, was normal.

SUNCT/SUNA was thought to be treatment-resistant until recently. However, the results of the last reported cases were more than satisfactory. We can divide the treatment as acute treatment and preventive treatment (3). In one study, high-dose oxygen and indomethacin injections were tried in acute treatment, but no response was obtained (3). With IV lidocaine, the number of good outcomes in acute treatment is increasing (2,3). Most recently, in 2017, the clinical characteristics and treatment response of 65 patients with SUNCT and 37 with SUNA were evaluated by Cohen (2), and IV lidocaine was found to be 100% effective in acute treatment (3). Similar to the indomethacin response in the diagnosis of paroxysmal hemicrania and hemicrania continua, there are also ideas about the use of IV lidocaine as a diagnostic tool for SUNCT (4). We also administered IV lidocaine to our patient whose daily life activities were severely affected in the last 1 week and found it to be very effective. In the meantime, we arranged the prophylactic treatment. Due to cardiac adverse effects, our patient was continuously monitored during drug administration. No drug adverse effects were observed in our patient receiving a total of 1.5 mg/kg/hour for 3 days. It has been reported in the literature that IV lidocaine may be given at dosages of 1.5-3.5 mg/kg/hour for a maximum of seven days (5). The mechanism of action in SUNCT is unknown (4). Preventive treatment includes lamotrigine, topiramate, gabapentin, and carbamazepine (2,3). Lamotrigine appears to be more effective than others. Trigeminal nerve decompression, deep brain stimulation, and greater occipital

Table 1. Diagnostic criteria for short-lasting unilateral neuralgiform headache attacks

A. At least 20 attacks (fulfilling criteria B, C, D, E)

B. Moderate or severe unilateral head pain, with orbital, supraorbital, temporal and/or other trigeminal distribution, lasting for 1-600 seconds and occurring as single stabs, series of stabs or in a saw-tooth pattern

C. At least one of the following five cranial autonomic symptoms or signs, ipsilateral to the pain:

- 1. Conjunctival injection and/or lacrimation
- 2. Nasal congestion and/or rhinorrhea
- 3. Eyelid edema
- 4. Forehead and facial sweating
- 5. Forehead and facial flushing
- 6. Sensation of fullness in the ear
- 7. Miosis and/or ptosis

D. Attacks occur with a frequency of  $\geq \! 1$  per day for more than half the time

E. Not better accounted for by another ICHD-3 diagnosis

Diagnostic criteria for SUNCT

A. Attacks fulfilling criteria for short-lasting unilateral neuralgiform headache attacks

B. Conjunctival injection and lacrimation

Diagnostic criteria for SUNA

A. Attacks fulfilling criteria for short-lasting unilateral neuralgiform headache attacks

B. Conjunctival injection or lacrimation

**Episodic SUNCT or SUNA** 

Attacks occurring in periods lasting from 1 week to one year, separated by pain-free periods lasting 1 month or more.

### Chronic SUNCT or SUNA

Attacks of SUNCT occurring for more than one year without remission, or with remission periods lasting less than 1 month

SUNCT: Short-lasting unilateral neuralgiform headache with conjunctival injection and tearing, SUNA: Short-lasting unilateral neuralgiform headache attacks with cranial autonomic features, ICHD-3: International Classification of Headache Disorders-3

nerve blockade-stimulation have been reported in selected cases for non-pharmacologic treatment of SUNCT/SUNA. However, blocking of other pericranial nerves and ganglia was not effective (5).

Our patient had severe headache every 2-3 months that lasted for 1 month. The 1.5-month pain-free period post-pregabalin can be attributed to spontaneous remission, not pregabalin. The duration of painlessness with IV lidocaine is variable and has been reported to be as short as the infusion period, and as long as 6 months (2). The patient was treated with lamotrigine 200 mg/day, topiramate 150 mg/day, and gabapentin 900 mg/day following IV lidocaine, and the sphenopalatine ganglion blockage was performed on day 5. The absence of pain for the last 8 months in our patient may be due to the long-term effect of IV lidocaine or the effect of preventive treatments used. In this case, we can say that we acted early for the blockade of sphenopalatine ganglion, and we should have waited for the remission period.

In this letter, we wanted to emphasize the response to IV lidocaine of a patient with SUNCT, whose daily life activities were severely affected, in the view of the literature.

## Ethics

Informed Consent: Consent form was filled out by all participants.

Peer-review: Internally peer-reviewed.

## Authorship Contributions

Surgical and Medical Practices: Ö.K., M.K., L.E.İ., Concept: Ö.K., M.K., L.E.İ., Design: Ö.K., M.K., L.E.İ., Data Collection or Processing: Ö.K., M.K., L.E.İ., Analysis or Interpretation: Ö.K., M.K., L.E.İ., Literature Search: Ö.K., M.K., L.E.İ., Writing: Ö.K., M.K., L.E.İ.

Conflict of Interest: No conflict of interest was declared by the authors.

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