



## Post-stroke short-lasting unilateral neuralgiform headache attacks with autonomic symptoms (SUNA): response to lamotrigine and sphenopalatine ganglion block

Seniha Ozudogru<sup>1</sup> · Sofia Loucao<sup>1</sup>  · Asad Rauf<sup>1</sup> · Melissa Cortez<sup>1</sup>

Received: 10 January 2019 / Accepted: 29 April 2019 / Published online: 11 May 2019  
© Springer-Verlag GmbH Germany, part of Springer Nature 2019

**Keywords** Short-lasting unilateral neuralgiform headache attacks with autonomic symptoms (SUNA) · Sphenopalatine ganglion block (SPG) · Post-stroke headache · Lamotrigine

Dear Editors,

Headache is an often underappreciated complication of stroke, present in up to 38% of cases [1]. Autonomic cephalgias are a recognized post-stroke headache variant [1, 2], though short-lasting unilateral neuralgiform headache attacks with cranial autonomic symptoms (SUNA) are less frequently reported. Furthermore, prior reports describe resolving post-stroke symptoms—either spontaneously or with other anti-epileptics. We report a patient who developed chronic treatment-refractory SUNA following a lateral medullary stroke, which was ultimately responsive to a combination of lamotrigine and sphenopalatine ganglion blocks (SPG). The sphenopalatine ganglion is considered the largest of the parasympathetic ganglia, and plays a key role in the trigemino-autonomic reflex. Thus, SPG blocks have been deployed therapeutically in an attempt to target parasympathetic pathology. SPG has been used in cluster headache, migraine, and hemicrania continua [3]. There have been no reported cases in the literature of the use of SPG blocks in SUNA/SUNCT.

A 52-year-old male with a history of left lateral medullary stroke 3.5 years ago (due to vertebral artery dissection secondary to methamphetamine use) presented to the University of Utah Headache Subspecialty Clinic with chronic recurrent and severe headache, localized over the left eye,

with tearing, ptosis, and facial swelling, which began 6 days following his stroke. Painful attacks lasted 30 s to 10 min, and occurred up to 20 times per day. The patient reported no identifiable triggers and denied a history of headache prior to his cerebrovascular accident. The headache characteristics met the 2018 International Classification of Headache Disorders (version 3) clinical diagnostic criteria for SUNA [4]. Of note, his attacks lacked the necessary erythema required for a diagnosis of short-lasting unilateral neuralgiform headache with conjunctival injection and tearing (SUNCT); furthermore, attacks occurred spontaneously without any refractory period, which differentiates SUNA from trigeminal neuralgia.

The vertigo, dysphagia, and difficulty with coordination in the left upper and lower extremities that were present at the time of his stroke had resolved. MRI brain without contrast from the time of stroke and MRA head and neck shows the left lateral medullary stroke and left vertebral artery filling defect (Fig. 1a, b). Repeated MRI with and without contrast did not produce additional findings.

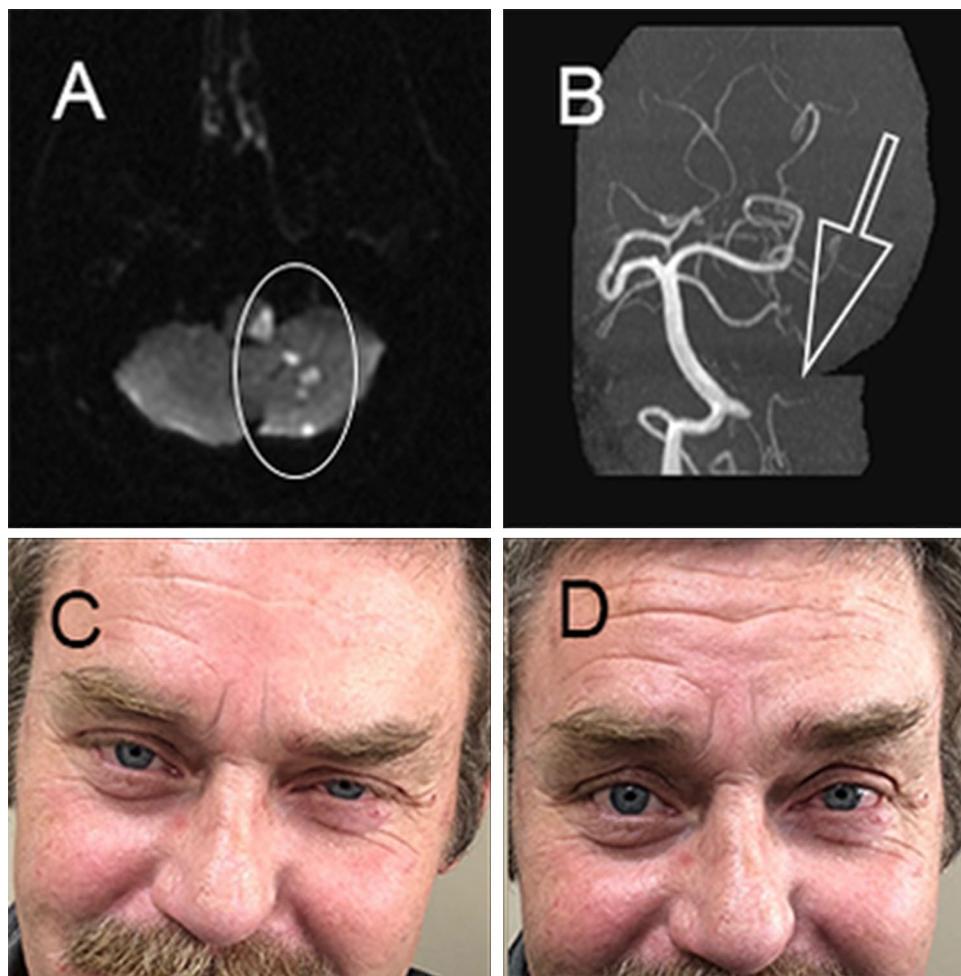
The patient had previously tried numerous medications that had failed to alleviate his headache attacks, including gabapentin 100 mg TID (6-month trial; could not tolerate higher dose), desipramine 50 mg (3-month trial), metoprolol 50 mg (15-month trial), citalopram 20 mg (4-month trial), lisinopril 20 mg (3-month trial), divalproex sodium 500 mg (6-month trial), baclofen 60 mg (3-month trial), cyclobenzaprine, trazodone, sumatriptan, melatonin, labetalol, amitriptyline, duloxetine, and pregabalin (unknown doses and durations). At the initial clinic visit, the patient was started on lamotrigine 25 mg daily. On exam, the patient showed left-sided ptosis, miosis, and swelling (Fig. 1c). At the one-month follow-up visit, the patient reported a decrease in headache attacks from 20 per day to 4–5 per day, though

✉ Seniha Ozudogru  
Seniha.Ozudogru@hsc.utah.edu

✉ Sofia Loucao  
Sofia.Loucao@utah.edu

<sup>1</sup> Department of Neurology, University of Utah, 729 Arapahoe Drive, Salt Lake City, UT 84108, USA

**Fig. 1** MRI and clinical images. **a** MRI brain at the time of the patient's acute stroke shows diffusion changes in the left medulla and left inferior cerebellum that are consistent with acute infarction (circle; diffusion-weighted image). **b** MRA head and neck at the time of the patient's acute stroke shows loss of normal flow in the left V4 segment of the vertebral artery and nonvisualization of the left posterior inferior cerebellar artery (arrow). **c** Pre-sphenopalatine ganglion block (SPG) during asymptomatic episode involving clinically evident left-sided ptosis and miosis. **d** Resolution of left sided ptosis and miosis, post-SPG block



he continued to report significant left-eye and nostril pain. Lamotrigine was then titrated to 100 mg daily over 4 weeks. At his 2-month follow-up, he reported resolution of severe attacks and improved mood stabilization but persistent low-grade left-eye and nostril pain. Thus, we then administered a SPG block (8 cc 4% lidocaine and 2 cc of 4 mg/mL dexamethasone solution; 5 cc was administered bilaterally to each side using a SphenoCath device, Dolor Technologies, SLC, UT, USA), which resulted in immediate relief from residual facial pain and improved craniofacial symmetry (Fig. 1c, d). On subsequent follow-up, the patient reported a further reduction in severe headache attacks with SPG blocks, down from 20 to 2–3 per day. He continues to return for repeat SPG blocks every 4 weeks with ongoing benefit (thus far, the patient has completed five treatment cycles).

Although headaches are a relatively common complication of stroke, the specific occurrence of SUNA is rare [2]. Lamotrigine was shown to be an effective treatment for central post-stroke pain in a previous randomized clinical trial, but specific pain types and stroke localizations were not described [5]. This case adds SPG blocks to the potentially

effective treatment armamentarium for post-stroke pain headache disorders.

**Funding** None.

### Compliance with ethical standards

**Conflict of interest** We have no conflicts of interest to report.

**Informed consent** Written and verbal consent for use of clinical images were obtained from the patient.

### References

1. Altavilla R, Landi D, Altamura C, Bussone G, Maggio P, Corbetta M, Vernieri F (2012) Dorsolateral medullary ischemic infarction causing autonomic dysfunction and headache: a case report. *J Headache Pain* 13(4):327–330. <https://doi.org/10.1007/s10194-012-0427-8>
2. Hassan S, Lagrata S, Levy A, Matharu M, Zrinzo L (2018) Microvascular decompression or neuromodulation in patients

with SUNCT and trigeminal neurovascular conflict? *Cephalgia* 38(2):393–398. <https://doi.org/10.1177/0333102417735847>

3. Robbins MS et al (2016) The sphenopalatine ganglion: anatomy, pathophysiology, and therapeutic targeting in headache. *Headache* 56(2):240–258
4. Headache Classification Committee of the International Headache Society (2013) The International Classification of Headache Disorders, 3rd edition (beta version). *Cephalgia* 33(9):629–808
5. Jin D, Lian YJ, Zhang HF (2016) Secondary SUNCT syndrome caused by dorsolateral medullary infarction. *J Headache Pain* 17:12. <https://doi.org/10.1186/s10194-016-0604-2>