target nerve to yield optimal results [8]. All of these factors may provide an advantage when lesioning neural tissue that is irregular in shape, large in size, or in close proximity to neurovascular structures that should not be lesioned. A large sciatic neuroma located in the infrapiriformis foramen provides a relevant example, but this may also apply to more distal neuromas.

Importantly, this case demonstrates the feasibility of treating chronic, refractory PLP by neuroma C-RFA. We suggest further study to evaluate whether the outcome observed in this case might be generalizable to a larger cohort, as well as the attributable effect of C-RFA on pain, physical function, and health-related quality of life in patients with PLP secondary to postamputation neuroma.

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Letter to the Editor

SUNCT Headache Occurring Secondary to a Cerebellopontine Angle Meningioma Reinforces the Hypothesis of Trigeminal Nerve Compression

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Dear Editor,

Short-lasting unilateral neuralgiform headache with conjunctival injection and tearing (SUNCT) is the shortest primary headache included in the trigeminal autonomic cephalalgias (TAC) group of the ICHD-3 [1]. Since its first description, as in many other primary headaches, secondary cases have been reported. We hereby present the first case of SUNCT symptomatic to a cerebellopontine angle meningioma.

A 60-year-old Caucasian woman with no relevant medical history consulted our Headache Unit because of a five-year facial pain. The patient did not relate any trauma or facial manipulation with the pain onset. She described tens of daily pain attacks, lasting 40–50 seconds and without any refractory period between episodes. Episodes tended to aggregate during the daytime, with pain-free periods during the day. The intensity of pain was described as being between 8 and 10 out of

10 on the verbal analog scale (VAS), with a burning and stabbing quality. The pain was described as being in the supralabial area radiating through the zygomatic arch, toward the ipsilateral supraciliary area. During these episodes, she experienced significant ipsilateral hyperemia and ocular itching and tearing. She did not describe rhinorrhea, nasal congestion, changes in facial sweating, pallor, rubor, ptosis, pupillary changes, photophobia, phonophobia, osmophobia, nausea, or vomiting. Light touch over the affected area was able to trigger episodes. Swallowing, speaking, and exposure to cold wind did not trigger the episodes.

When she came to our facility, we considered the possibility of a SUNCT, so magnetic resonance imaging (MRI) with special sequences was requested. Contrast-enhanced MRI revealed an extra-axial tumor, which had an implantation base on the tip of the right temporal bone. It presented intense and homogeneous enhancement after administration of paramagnetic contrast, confirming infiltration into the Meckel cavum and the ipsilateral cavernous sinus. It caused significant compromise on the right trigeminal nerve and right protuberance, without associated edema. Radiological diagnosis was meningioma (Figures 1 and 2).

The patient had been initially diagnosed as having trigeminal neuralgia and was treated with carbamazepine, increasing the dose from 200 mg twice daily (bid) to 400 mg three times daily without improvement. We started lamotrigine, but after increasing the dose from 25 to 50 mg per day, the patient experienced a generalized cutaneous rash, and it was discontinued. Pregabalin was started, and with a 75-mg bid dose, the patient described significant relief, experiencing zero to two episodes per day of mild intensity (2–3/10 in VAS). The patient was referred to neurosurgery and tumor resection was offered, but due to the clinical improvement, she rejected surgery; after 26 months of continuous clinical and radiological follow-up, she has not experienced worsening.

The pathophysiology of SUNCT primary cases has been recently related to neurovascular compression and trigeminal nerve compromise. In focused studies, an aberrant vessel has been found in close association with the fifth cranial nerve in up to 88% of cases in some series. Indeed, ablative procedures and microvascular decompression of the trigeminal nerve have been described with satisfactory results in selected cases [2].

From the first SUNCT case description until today, 69 secondary SUNCT cases have been described. In a recent review, the most frequent cause of symptomatic SUNCT was neurovascular compression (35 cases) mostly located at the cerebellopontine angle, followed by space-occupying lesions (17 cases), infections (10 cases), inflammatory diseases (three cases), and congenital malformations (two cases). Contrary to neurovascular compressions, neoplasm and infection had a wider focus and were mostly located at the preganglionic fibers of the

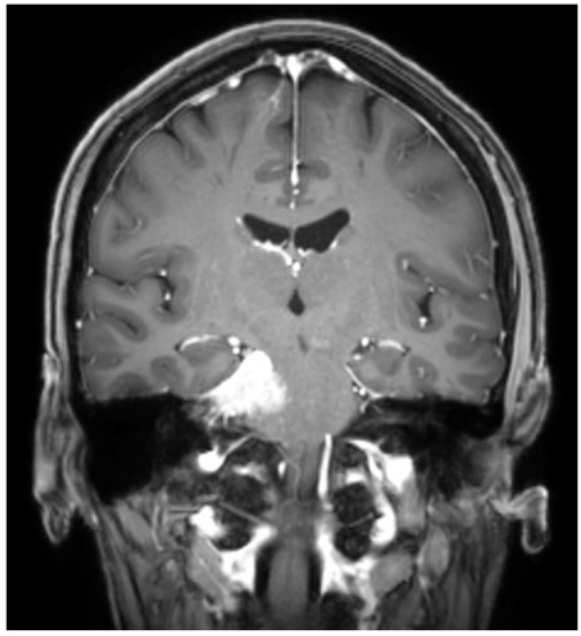

Figure 1. Magnetic resonance imaging coronal plane with contrast. See the contrast-enhanced meningioma located at the right cerebellopontine angle. The size of the tumor is 21.9 × 30.1 × 23 mm (craniocaudal, anteroposterior, and transversal, respectively). See the mass effect compressing the ipsilateral protuberance site and the V cranial nerve root.

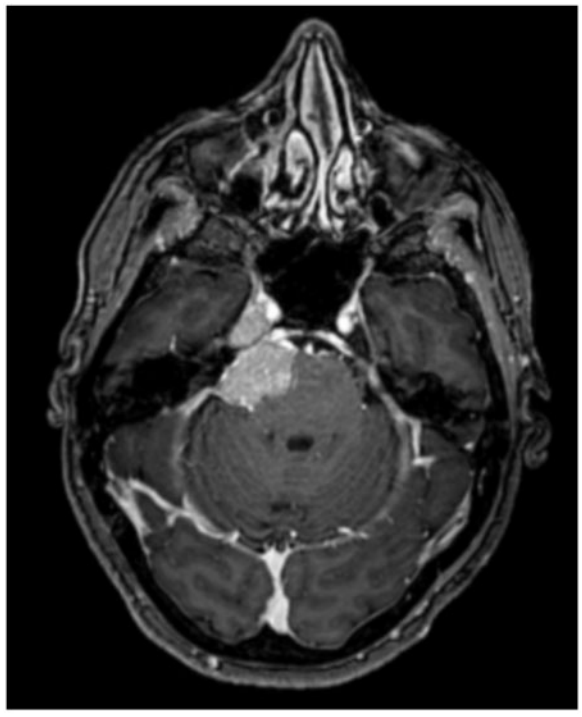

Figure 2. Magnetic resonance imaging axial T1W3DTFE sequence with contrast. The contrast-enhanced meningioma is compressing the ipsilateral trigeminal root and invading the cavernous sinus.

trigeminal nerve. Supporting this idea, there have been nine cases described in which neoplasm removal either by surgical intervention or radiotherapy was an effective treatment. One neoplasm was located in the cavernous sinus and two in the cerebellopontine angle [2, 3].

In our case, the lesion compressed the ipsilateral pons and distorted the trigeminal nerve anatomy, also infiltrating the ipsilateral cavernous sinus. It is not possible to determine which of these was responsible for the syndrome. To date, only one previous case related to a meningioma has been published. Nevertheless, in that case the meningioma was located in the left fronto-temporal lobes, it did not compress the trigeminal root, and the patient did not undergo an intervention due to cardiovascular comorbidity, making it difficult to establish the causality of the finding [4].

SUNCT diagnosis is clinical, and there is no test for it; therefore, it has limitations. Some of its features are shared with other TACs, especially trigeminal neuralgia and short-lasting unilateral neuralgiform headache attacks with cranial autonomic symptoms (SUNA), although for some authors SUNCT is considered to be a subtype of SUNA. Features favoring SUNCT in our patient as opposed to trigeminal neuralgia include (a) the prominent distribution of pain in the ophthalmic division of the trigeminal nerve, which is a rare location for trigeminal neuralgia; (b) the lack of a refractory period; (c) the longer duration of the attacks (40–50 seconds); (d) the stabbing and burning and not electric pain quality; and (e) the lack of response to carbamazepine. Features favoring SUNCT in our patient as opposed to SUNA include presence of both of the following ipsilateral to the pain: conjunctival injection and lacrimation with no other autonomic symptom [1].

Because of the adverse reaction suffered with lamotrigine and the favorable side effect profile of pregabalin and its efficacy in some reported SUNCT cases [5], we settled on symptomatic therapy with the last case. Pregabalin has an inhibitory effect on voltage-gated calcium channels and is used as an antiepileptic drug as well as in the treatment of neuropathic pain. In this case, it was shown that the patient's neuralgiform pain completely responded to it. This case also supports that pregabalin can be considered as a therapeutic option for SUNCT syndrome in selected patients.

We suggest that these MRI findings could explain the causality between meningioma and the trigeminal system. We hypothesize that by compressing the trigeminal nerve, the pathophysiological process of SUNCT gets activated. Nonetheless, we cannot explain why SUNCT-like attacks get provoked and no other types of TACs. Another limitation of this case is that no surgical intervention was performed in our patient, so we cannot demonstrate complete improvement related to lesion removal.

Histologic diagnosis was not done, but radiological diagnosis was highly likely and radiological follow-up did not suggest another cause.

Symptomatic forms of SUNCT are infrequent but not exceptional. Cerebellopontine angle meningioma can cause secondary SUNCT. Brain MRI with special sequences evaluating the trigeminal nerve should be done in every SUNCT patient.

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