

SUNCT (Short-lasting unilateral neuralgiform headache attack with conjunctival injection and tearing) associated with pituitary lesion^{☆,☆☆}



Cefalea tipo SUNCT (*Short-lasting unilateral neuralgiform headache attack with conjunctival injection and tearing*) asociada a lesión hipofisaria

Dear Editor:

SUNCT (short-lasting unilateral neuralgiform headache attack with conjunctival injection and tearing) syndrome was first defined by Sjaastad et al. in 1978.¹ Since then, several studies on this topic have been published.^{2,3} The International Classification of Headache Disorders currently places it in the trigeminal autonomic cephalalgias subgroup.⁴ Although it is considered a primary headache in most cases, other cases are secondary to structural abnormalities in the posterior fossa, and especially in the hypophyseal region.^{5,6} More controversial is whether or not pharmacological agents may trigger this entity. For example, dopaminergic agents may exert a bidirectional influence. We describe one case of SUNCT syndrome secondary to consumption of ergoline dopamine agonist in a patient with hyperprolactinemia due to pituitary microadenoma.

Our patient was a 24-year-old man with a diagnosis of hyperprolactinemia secondary to right-sided pituitary microprolactinoma without cavernous sinus invasion. The patient was in treatment with cabergoline. After treatment onset, he experienced intense and pulsatile headache episodes. Pain was located in the right periorbital region and accompanied by conjunctival injection, tearing, and ipsilateral rhinorrhoea. Episodes lasted no more than 3 minutes, but they progressively grew more frequent until the patient was experiencing episodes every 30 minutes daily. Headache met criteria for SUNCT syndrome. Doctors suspended treatment with cabergoline and began lamotrigine dosed at 50 mg/12 h. A new MRI revealed a pituitary microadenoma with no changes with respect to a previous MRI (Figs. 1 and 2). Fifteen days after changing treatment, the patient showed clear signs of improvement; by 3 months, he had fewer than 5 episodes per month. Since prolactin levels remained high, doctors began treatment with quinagolide (non-ergoline dopaminergic agent); this reverted prolactin levels to normal but did not exacerbate the headaches.

The literature describes patients with SUNCT syndrome with pituitary micro or macroadenomas, both functional and non-functional.^{7–12} Although the pathophysiological



Figure 1 Sagittal section, MRI T1-weighted sequence before headache.



Figure 2 Sagittal section, T1-weighted MRI sequence with contrast after headache.

relationship has yet to be fully established, the most plausible hypothesis for most authors is possible mechanical excitation of the trigeminal nerve due to a local compressive effect at the level of the cavernous sinus. However, this does not explain how microadenomas would cause this syndrome. Another hypothesis points to the potential neurohumoral alteration caused by these tumours. In line with this hypothesis, there are published descriptions of cases of exacerbation or onset of SUNCT after starting treatment with dopaminergic agonists,^{9–12} as in the case we present in this article. However, there are also cases in which starting treatment improved a prior SUNCT-type headache.⁷ Therefore, both effects are possible, but the pathophysiological mechanisms are unknown in both situations. In our case, the time between treatment onset and pain onset, the absence of prior symptoms of SUNCT syndrome despite presence of microprolactinoma, absence of clinical and radiological changes in the underlying pituitary disease, and resolution of pain after discontinuing the drug

[☆] Please cite this article as: Pulido Fontes L, Mayor Gómez S. Cefalea tipo SUNCT (*Short-lasting unilateral neuralgiform headache attack with conjunctival injection and tearing*) asociada a lesión hipofisaria. *Neurología*. 2015;30:458–459.

^{☆☆} Presented as poster format in the headache section at the 64th Annual Meeting of the Spanish Society of Neurology.

all support the idea that the ergoline-dopaminergic agonist is an important triggering factor for headache. Starting treatment with another agonist did not result in exacerbation of the headache for reasons that remain unclear, but this could be due to the non-ergoline nature of the second drug. This situation is also described in one published case.⁹

Therefore, and despite the neurophysiological questions that remain unanswered, awareness of these cases of secondary SUNCT syndrome is important. Doctors should also consider any intercurrent medications that could significantly promote the development of headache, as in our case.

References

1. Sjaastad O, Russel D, Horvan I, Brune U. Multiple neuralgiform, unilateral headache attacks associated with conjunctival injection and appearing in cluster. A nosological problem. *Proc Scand Migr Soc.* 1978;31.
2. Pareja JA, Sjaastad O. SUNCT syndrome: a clinical review. *Headache.* 1997;37:195–202.
3. Pareja JA, Caminero A, Sjaastad O. SUNCT syndrome: diagnosis and treatment. *CSN Drugs.* 2002;16:373–83.
4. International Headache Society. The International Classification of Headache Disorders. 3rd ed (beta version). *Cephalalgia.* 2013;33:629–808.
5. Caminero AB, Mateos V. El hipotálamo en el SUNCT: similitudes y diferencias con las otras cefaleas trigeminoautonómicas. *Rev Neurol.* 2009;49:313–20.
6. Levy MJ, Matharu MS, Meeran K, Powell M, Goadsby PJ. The clinical characteristics of headache in patients with pituitary tumors. *Brain.* 2005;128:1921–30.
7. Matharu MS, Levy MJ, Merry RT, Goadsby PJ. SUNCT syndrome secondary to prolactinoma. *J Neurol Neurosurg Psychiatr.* 2003;74:1590–2.
8. Rocha-Filho PAS, Galvao ACR, Teixeira MJ, Rabello GD, Fortin I, Calderano M, et al. SUNCT syndrome associated with pituitary tumors. *Arq Neuropsiquiatr.* 2006;64:507–10.
9. Massiou H, Launay JM, Levy C, El Amrani M, Emperauger B, Bousser MG. SUNCT syndrome in two patients with prolactinomas and bromocriptine-induced attacks. *Neurology.* 2002;58:1698–9.
10. Larner AJ. Headache induced by dopamine agonists prescribed for prolactinoma: think SUNCT? *Int J Clin Pract.* 2006;60:360–1.
11. Levy JM, Matharu MS, Goadsby J. Prolactinomas, dopamine agonists and headache: two case reports. *Eur J Neurol.* 2003;10:169–73.
12. Jiménez Caballero PE. SUNCT syndrome in patient with prolactinoma and cabergoline-induced attacks. *Cephalalgia.* 2007;27:76–8.

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