CASE REPORT

Successful treatment of SUNCT syndrome with divalproex sodium extended-release

Síndrome SUNCT tratada com sucesso com divalproato de sódio de liberação prolongada

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ABSTRACT

Introduction: Short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) syndrome is a primary rare headache. This syndrome is known to be refractory to pharmacotherapy. Various treatments have been tried for SUNCT syndrome, generally of little benefit. **Objective:** To report a case of SUNCT syndrome who was successfully treated with divalproex sodium extended-release (DVPX ER) and to review the literature about of drugs used in this treatment. **Patient and Method:** The authors report a case of a 61 year-old man, suffering from SUNCT syndrome, who was successfully treated with DVPX ER. We review the literature about several categories of drugs that are used in the treatment to this syndrome. **Conclusion:** SUNCT syndrome is clinically well identified, but it is poorly treated. This is first report of SUNCT syndrome responsive to DVPX ER.

Key words: SUNCT syndrome; headache; divalproex sodium.

RESUMO

Introdução: A síndrome SUNCT (cefaleia de curta duração, unilateral, neuralgiforme com hiperemia conjuntival e lacrimejamento) é um tipo raro de cefaleia e refratária ao tratamento farmacológico. Diversos tratamentos são tentados e, geralmente, de pouco benefício. **Objetivo**: Relatar um caso de síndrome SUNCT tratada com divalproato de sódio de liberação prolongada (DVP ER) e revisar a literatura sobre as drogas utilizadas neste tratamento. **Paciente e Método**: Os autores descrevem um caso de um homem de 61 anos, com síndrome SUNCT que foi tratado com DVP ER. Foi revisada a literatura sobre as diversas drogas que são utilizadas no tratamento desta síndrome. **Conclusão:** A síndrome SUNCT é clinicamente bem identificada, mas é pobremente tratada. Este é o primeiro relato de síndrome SUNCT responsiva ao DVP ER.

Palavras-chave: Síndrome SUNCT; cefaleia; divalproato de sódio.

INTRODUCTION

Short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) syndrome is a primary rare headache, described initially by Sjaastad and colleagues in 1989,[1] and characterized by trigeminal pain accompanied by autonomic manifestations. The pain is strictly unilateral, localized to the orbital region, stabbing or burning in quality, abrupt, of short duration (5 to 240 seconds), frequent (3 to 200 per day), and accompanied by lacrimation, conjunctival injection, and nasal congestion or rhinorrhoea, all ipsilateral to the pain.[2,3] The pain is neuralgiform, so it can be difficult to distinguish from trigeminal neuralgia. The features that distinguish it from other trigeminal autonomic cephalalgias are: very brief duration of attacks that occur very frequently, and the presence of prominent conjunctival injection and lacrimation. SUNCT syndrome is clinically well identified, but it is poorly treated. Several categories of drugs are

used in the treatment to this syndrome, including intravenous lidocaine,[4] anesthetic blockades, verapamil,[5] and anti-epileptic drugs such as gabapentin,[6] lamotrigine,[7] oxcarbamazepine,[8] and topiramate.[9] All are generally of little benefit. Until recently, SUNCT was considered an intractable headache or highly refractory to treatment.[6]

We describe a patient with SUNCT syndrome who was successfully treated with divalproex sodium extended-release (DVPX ER). This drug has not previously been tried in SUNCT syndrome, and to the best of our knowledge this is the first report of SUNCT syndrome responsive to DVPX ER.

CASE

A 61 year-old man with an 11-days history of persistent throbbing headache localized to the orbital region. The headache was always right-sided. Daily pain was of severe intensity. The pain often lasted 5 to 10 seconds and recurred with an average of four to five times per day. The pain was accompanied by autonomic symptoms (conjunctival injection and lacrimation). The headache had no known triggering factors. During this period, the patient used different analgesics and other antiepileptic drug without success. General physical and neurologic examinations were normal. Magnetic resonance imaging was normal. A diagnosis of SUNCT syndrome was made. We started treatment with DVPX ER. Daily dosage was initiated at 500 mg/day, and after 7 days, because of the reoccurrence of occasional attacks, we increased the dosage to 500 mg twice daily. At a dose of 1,000 mg per day (500 mg twice daily) he became pain free. Two months later, DVPX ER was stopped and his pain did not recur in the following 6 months.

DISCUSSION

Our patient's short-lasting headache meets the diagnostic criteria for SUNCT.[10] This syndrome is known to be refractory to pharmacotherapy. Various treatments have been tried for SUNCT syndrome. Rare cases are responsive to lidocaine,[4] anesthetic blockades, verapamil,[5] and anti-epileptic drugs such as gabapentin,[6] lamotrigine,[7] oxcarbamazepine,[8] and topiramate.[9] DVPX ER is an antiepileptic drug used for treating other painful neurologic syndromes such as migraine, cluster headache, and chronic daily posttraumatic headache. DVPX ER stimulates brain GABA synthesis and reduces glutamate levels, one of the excitatory neurotransmitters, by increasing the activity of glutamic acid decarboxylase.

In our case, the initial dose DVPX ER of 500 mg no reduced the number of SUNCT attacks. When we increased the dose to 1,000 mg/day (500 mg twice daily), the attacks progressively reduced until they finally disappeared. To induce a remission, the patient required an average dose of DVPX ER to 1,000 mg/day.

From our experience with this case, DVPX ER should be considered a possible prophylactic treatment for SUNCT syndrome.

REFERENCES

1. Sjaastad O, Saunte C, Salvesen R, Fredriksen TA, Seim A, Roe OD et al. Short-lasting unilateral neuralgiform headache attacks with conjunctival injection, tearing, sweating, and rhinorrea. Cephalalgia. 1989;9:147-56.
2. Pareja JA, Sjaastad O. SUNCT syndrome. A clinical review. Headache. 1997;37:195-202.
3. Cohen AS. Short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing. Cephalalgia. 2007; 27(7):824-32.
4. Matharu MS, Cohen AS, Goadsby PJ. SUNCT syndrome responsive to intravenous lidocaine. Cephalalgia. 2004; 24 (11):985-92.
5. Narbone MC, Gangemi S, Abbate M. A case of SUNCT syndrome responsive to verapamil. Cephalalgia. 2005;25(6):476-8.
6. Hunt CH, Dodick DW, Bosch EP. SUNCT responsive to gabapentin. Headache. 2002;42(6):525-6.
7. D'Andrea G, Granella F, Ghiotto N, Nappi G. Lamotrigine in the treatment of SUNCT syndrome. Neurology. 2002; 57(9):1723-5.
8. Dora B. SUNCT syndrome with dramatic response to oxcarbazepine. Cephalalgia. 2006;26(9):1171-3.
9. Rossi P, Cesarino F, Faroni J, Malpezzi MG, Sandrini G, Nappi G. SUNCT syndrome successfully treated with topiramate: case reports. Cephalalgia. 2003;23(10):998-1000.
10. Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders. Cephalalgia. 2004;24(Suppl 1):1-160.

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