CASE REPORT

Intravenous lidocaine to treat SUNCT syndrome secondary to pituitary tumor. Follow up of a patient for four years. Case report*

Lidocaína endovenosa no tratamento da síndrome SUNCT secundária a tumor de hipófise. Seguimento de uma paciente por um período de quatro anos. Relato de caso

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ABSTRACT

BACKGROUND AND OBJECTIVES: SUNCT syndrome (*short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing syndrome)* is described as unilateral orbitary, supraorbitary or temporal pain, characterized as stabbing or throbbing, severe, disabling, of short duration, varying from 5 to 240 seconds. This is a difficult to treat syndrome, however in our case there has been satisfactory analgesia with intravenous lidocaine.

CASE REPORT: Female patient, 32 years old, with SUNCT syndrome secondary to pituitary tumor (submitted to transsphenoidal hypophysectomy and radio-surgery), started presenting 30 daily pain crises. There has been no analgesic response to surgical procedure. Pharmacological treatment was started with lamotrigine, chlorpromazine and amitriptyline without pain relief. Patient was submitted to intravenous lidocaine infusion cycles with adequate pain relief.

CONCLUSION: In this case of SUNCT syndrome, intravenous lidocaine was able to promote adequate and long-lasting analgesia and may be considered integral part of the treatment of this syndrome.

Keywords: Headache, Lidocaine, Pituitary tumor, SUNCT syndrome, Treatment.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A síndrome SUNCT (*short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing syndrome)* é descrita como dor de localização orbitária, supraorbitária ou temporal unilateral, caracterizada como em pontada ou pulsátil, de forte intensidade, incapacitante, de curta duração, variando entre 5 e 240 segundos. É uma síndrome dolorosa de difícil tratamento, no entanto, no presente caso obteve-se resposta analgésica satisfatória com lidocaína endovenosa.

RELATO DO CASO: Paciente do gênero feminino, 32 anos portadora de síndrome SUNCT secundária a tumor de hipófise (submetida a hipofisectomia transesfenoidal e radiocirurgia), passou a apresentar 30 crises de dor diariamente. Não obteve resposta analgésica com o procedimento cirúrgico. Foi iniciado tratamento farmacológico com lamotrigina, clorpromazina e amitriptilina sem alivio da dor. Submetida a ciclos de infusão de lidocaína venosa obteve adequado alivio da dor.

CONCLUSÃO: Neste caso de síndrome *SUNCT,* a lidocaína endovenosa foi capaz de proporcionar analgesia adequada e duradoura, podendo ser considerada como parte integrante nos pacientes portadores dessa de síndrome.

Descritores*:* Cefaleia, Lidocaína, Neoplasia de hipófise, Síndrome SUNCT, Tratamento.

INTRODUCTION

SUNCT syndrome (short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing syndrome*)* was firstly described by Sjaastad et al.[1]. This syndrome is characterized by orbitary, supraorbitary or unilateral temporal severe, disabling, short duration stabbing or throbbing pain lasting from 5 to 240 seconds. It involves the first division of the trigeminal nerve and is associated to ipsilateral autonomic symptoms, such as tearing and eye redness[1-3]. Autonomic symptoms, in general dramatic, appear immediately after pain onset and, in addition to those already described, there might also be rhinorrhea and nasal obstruction[1]. Although most are primary cases, there are reports of cases secondary to tumors[4], predominantly affecting male patients in the fifth decade of life; however there are reports on females

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and patients aged above 70 years[2,5,6].

A recent review has identified 222 published SUNCT/SUNA cases, most of them of primary origin[2].

As to the origin of pain, it is described that there is activation of ipsilateral hypothalamic lower posterior area[6], as well as of the bilateral hypothalamic area[4], which confirms the hypothesis of the central origin of this headache. A neurovascular origin is also mentioned[2].

The lack of response to clinical and/or surgical treatment is a major feature of this disease. However, some authors have recently shown that intravenous lidocaine has promoted analgesia in patients with SUNCT syndrome[7,8].

This study aimed at reporting a case with clinical diagnosis of SUNCT syndrome secondary to pituitary tumor, which had satisfactory analgesic response with intravenous lidocaine.

CASE REPORT

Female patient, 32 years old, submitted to transsphenoidal hypophysectomy to treat pituitary macroadenoma. Since tumor was only partially removed, treatment was completed with radio-surgery. Before surgical procedure, patient reported severe stabbing right periorbital headache, visual analog scale (VAS=10), with more than 30 daily crises. Symptoms were triggered by physical effort and were associated to ipsilateral conjunctival hyperemia and tearing. Diagnosis was SUNCT syndrome secondary to pituitary tumor. After surgical procedure there has been total pain relief with recurrence 30 days later. Indomethacin (150mg/day) was started with total pain relief (VAS=0); however, patient presented hematemesis which has forced the withdrawal of the non-steroid anti-inflammatory drug. Lamotrigine (100mg/day), chlorpromazine (40mg/day) and amitriptyline (50 mg/day) were then administered for 3 months without improvement in pain intensity (VAS=10) or frequency (30 crises/day). Amitriptyline and chlorpromazine were maintained and lithium carbonate in increasing doses was added. However, three months after this therapy there was no pain relief. Lithium carbonate was withdrawn and intravenous lidocaine was started in fixed dose of 240mg (3mg/kg) continuous infusion for two hours. In the first day of infusion there has been total pain relief (VAS=0); however five days later crises have returned, however with lower intensity (VAS=5-7) and frequency (10 crises/day). Given the outcome, we decided to weekly repeat intravenous lidocaine (240mg) for a period of 10 weeks.

Patient started to be evaluated before and after each lidocaine administration and progressive and sustained improvement was observed since pain crises were induced only by effort and pain was moderate (VAS=5). At the end of the 7th week, patient was asymptomatic even with physical efforts, remaining like that for six months. After this period pain has returned however with lower frequency and intensity and patient was once more treated with intravenous lidocaine for 10 weeks. Patient was submitted to six intravenous lidocaine cycles for four years, with irregular intervals, with satisfactory analgesic results.

DISCUSSION

SUNCT syndrome secondary to brain tumors is an uncommon disease and there is still no consensus for its management. In this case, transsphenoidal hypophysectomy to treat pituitary macroadenoma has resulted in transient pain improvement. This fact contrasts with the report of other author who has observed pain relief with the surgical procedure[4].

There are different hypotheses to explain pituitary tumor-induced pain. It is believed that the mechanical action resulting from the presence of the tumor and the consequent stretching of structures involved in pain processing may be responsible for the symptoms. This hypothesis explains pain in the presence of macroadenomas. Another possible explanation would be the invasion of cavernous sinuses causing ipsilateral pain. Still another hypothesis suggests that pain would be caused by humoral mechanism; however this is only applied to patients with hormone-producing tumors[2,3].

In our case, it was suggested that the presence of the tumor could be considered the triggering and maintaining SUNCT syndrome factor, explained by previous exuberant symptoms and transient relief when tumor mass was decreased. Similarly, pain recurrence 30 days after surgical procedure reinforces the mechanical genesis by the presence of tumor[5]. On the other hand, satisfactory analgesic response with indomethacin suggests the possibility of tumor inflammatory reaction[5].

The decision to introduce lidocaine to treat this patient was due to its analgesic and anti-inflammatory action. This drug acts on voltage-dependent sodium channels resulting in reversible anesthetic action[7,8]. As soon as the local analgesic action is developed, electric excitability threshold gradually increases, action potential peak decreases, neuronal impulse conduction increases and conduction safety factor is decreased, promoting action potential propagation reduction and also nervous conduction failure[9].

Lidocaine affinity for sodium channels is higher in open and inactivated states. In addition, lidocaine is usage-dependent blocker, being that the higher the frequency of neuronal stimulation, the more ionized lidocaine molecules have access to action sites, generating better blockade and analgesia[9]. In SUNCT syndrome, the analgesic effect resulting from the already described mechanism has variable duration and may be extended to approximately three to six weeks[7,10].

With regard to intravenous lidocaine dose for analgesic purposes there is still no defined consensus. Fixed dose of 240mg lidocaine used in this case was based on analgesic results reported by previous studies[8,10-12], and lidocaine-induced analgesia was satisfactory and long-lasting, consolidating its indication as part of the multimodal SUNCT syndrome management.

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