BILATERAL SUNCT SYNDROME ASSOCIATED TO CHRONIC MAXILLARY SINUS DISEASE

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ABSTRACT - SUNCT syndrome (short lasting unilateral neuralgiform headache with conjuntival injection and tearing) is defined as short attacks of periorbital unilateral pain and accompanied by ipsilateral lacrimation and redness of the same eye. We present an unusual SUNCT case with bilateral pain that started five years ago after an acute maxillary sinus infection that evolved to chronic sinusitis. This association has been described in few SUNCT cases, but its causal role remains uncertain. The patient was a 58 years old man that fulfilled a headache diary that showed the usual circadian pattern, worsening in the morning and afternoon, and responded to treatment with gabapentina. He was submitted to a functional endoscopic sinus surgery and evolved with milder pain. In a review of 21 patients, 5 had a past medical history of sinusitis, but the causal role of this association remained uncertain.

KEY WORDS: SUNCT, bilateral, sinusitis, gabapentina

Síndrome SUNCT de ocorrência bilateral associada a sinusopatia maxilar crônica

RESUMO - A síndrome SUNCT (short lasting unilateral neuralgiform headache with conjuntival injection and tearing) é definida como curtos ataques de dor periorbital unilateral, acompanhada de lacrimejamento e hiperemia conjuntival ipsilateral. Apresentamos um raro caso de SUNCT com dor bilateral com evolução de cinco anos e iniciado após uma infecção de seio maxilar que evoluiu para sinusite crônica. Esta associação foi descrita em poucos casos de SUNCT, porém pouco esclarecida. O paciente era um homem de 58 anos que preencheu um diário de dor que demonstrou o típico padrão circadiano da síndrome, com pioras matinais e vespertinas, e apresentou melhora com uso de gabapentina. Submetido a cirurgia endoscópica funcional em seio maxilar e evoluiu com modulação da dor, sugerindo um potencial efeito benéfico após tratamento da sinusopatia. Na revisão de literatura encontramos 21 casos de SUNCT bilateral, cinco dos quais apresentavam história de sinusite; no entanto, a relação entre as duas entidades permanece ainda incerta.

PALAVRAS-CHAVE: SUNCT, bilateral, sinusite, gabapentina.

CASE

We report on a 58-year-old non white male with a past medical history of bilateral lachrymal duct surgeries and trauma in the left forehead in his infancy, who suffered from periorbital stabbing eye pain lasting 5 to 10 seconds accompanied by conjuntival injection and tearing. The attacks started five years ago after he had an acute maxillary sinus infection presenting with proptosis and lateral deviation of the left eye due to fronto-ethmoidal mucocele, diagnosed by CT scan, requiring surgical drainage of the paranasal sinuses. After this intervention he developed chronic sinusitis, confirmed with CT and MRI scans (Fig 1). A few weeks after the procedure he evolved stabbing eye pain described as “a needle sticking into my eye”, lasting 5 to 10 seconds, accompanied by conjuntival injection and tearing, that could be triggered by cold weather and forceful reading. Although he was not able to count, he referred...
the attacks occurred most frequently in the left eye, occasionally in the right eye and seldom bilateral. He denied nausea, vomiting or any sensitivity to light, sounds or scents. He was instructed to fulfill a headache diary that revealed these attacks in a circadian pattern, worsening in the morning and afternoon (Graphic 1). He reported no pain between attacks.

Before he attended our clinic he was submitted to diverse drug regimens, including carbamazepine, amitriptyline, codeine, tramadol and indomethacin, with no relief. He was started on augmentation doses of gabapentin and evolved with ameliorating of pain attacks and almost complete resolution with 1800mg/daily (Graphic 2), keeping the characteristic circadian pattern.

With the hypothesis that his chronic sinusitis might be generating the attacks, he was submitted to a functional endoscopic sinus surgery (FESS), with removal of great amount of purulent material from his left maxillary sinuses. After surgery he reported change in pain characteristics from paroxysmal stabbing to continuous bifrontal pain, assuming a tightness pattern. All the more we could note that he returned to work and that attacks of pain and redness of the eye were no more observed during clinical appointments, suggesting that the pain was modulated to a milder pattern.

**DISCUSSION**

This patient presented with frequent eye pain attacks that fulfill criteria for possible SUNCT syndrome, as the pain was occasionally bilateral. All the more, his ability in fulfilling the headache diary has contributed to the characterization of attacks frequency and periodicity. The existence of a circadian rhythm in SUNCT, with worsening in the morning and afternoon, has been reported and is supposed to be related to hypothalamus activation during pain attacks.

The treatment of SUNCT pain is, as demonstrated by this case, hard to achieve. Reports on the use of topiramate, lamotrigine and gabapentin have shown to ameliorate or fully relieve patients from attacks. We could not find any randomized control trial comparing these drugs, the choice for gabapentin was own to its ease of access from the patient, rapid titration and adverse effects profile.

Although our patient has reported little relief, the change in pain characteristics and the fact that he has returned to work suggests that FESS and sinus drai-
nagement could have ameliorated the attacks. Based on this sole report it is difficult to establish a connection between SUNCT syndrome and sinus disease, though we hypothesize that the proximity of craniofacial structures and trigeminal nerve endings might be related in precipitating attacks, as also thought on a report of SUNCT attacks triggered by forceful mouth opening. It is speculative that the interventions for treating lachrymal duct disease and head trauma might have caused irritative lesions on nerve terminals in craniofacial structures, but the association to SUNCT attacks remains uncertain as they took place more than 40 years before the development of pain.

In conclusions, SUNCT syndrome remains an infrequent condition, yet an underlying disease must always be excluded. We have demonstrated that the relief from chronic sinus disease can exert influence on SUNCT attacks remain uncertain as they took place more than 40 years before the development of pain.

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Graphic 2. Number of attacks per day after treatment with gabapentin. The peak number of attacks day from day 26 to 30 was attributed to weather changes.