CHAPTER 46

AN INTERESTING RESOLUTION OF AN UNUSUAL HEADACHE

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Case History

A 58-year-old man was sent for review. He had a 5-year history of short-lasting headaches. He reported that he had been having between 3 to 4 to a maximum of 50 to 60 attacks a day of right supraorbital and retro-orbital pain. These attacks lasted 5 to 30 seconds with no trigger factors and no aura symptoms. There was no nausea, photophobia, or phonophobia but his wife volunteered, and he confirmed, tearing of the eyes without redness or nasal congestion. The pain was unaffected by movement. He had never had a significant break from the attacks over the 5 years. There was no effect of wind, swallowing or chewing, and no temporomandibular joint tenderness or clicking. He was on no medications.

His past history was largely unremarkable. There was no significant family history. He was a nonsmoker who drank alcohol occasionally, and this did not trigger attacks.

On examination he was well and in no distress. The fundi were normal and the visual fields full. The eye movements were normal and full and the pupils equal and normal. There was no trigeminal sensory loss, temporal artery tenderness, or facial asymmetry. The jaw opened normally. In the limbs tone and power were equal and normal and the reflexes symmetrical with down-going toes. Gait and coordination were normal. The general examination was normal with a normal blood pressure.

A diagnosis was made and therapy commenced. At review he was completely pain free. He has remained so at least 6 months later.

Questions about This Case

- Are there any other points from the history that could be clarified?
- What, if any, tests would you do?
- What is the provisional diagnosis?
- What treatment would you advise?

Comments on the Questions

1. History taking is a very individual matter and this case is no exception. One could come to a reasonable differential diagnosis with the history as it is. Perhaps the possibility, albeit remote, of giant cell arteritis could have been explored more.
2. The range of tests will always reflect a compromise between how confident one is, how confident the patient is with what you say, and the realities of the medicolegal climate in which one practices. Certainly, a test for electrolytes, a blood count, ESR, and brain imaging seem reasonable as a minimum.
3. The differential diagnosis is relatively limited by the very short-lasting nature of the attacks. I have included some diagnoses in the list which are really not there but interesting to discuss. One possible list would include:
   - SUNCT (short-lasting neuralgiform headache with conjunctival injection and tearing)
   - Paroxysmal hemicrania
   - Chronic cluster headache
   - Chronic sinusitis
   - Giant cell arteritis
   - Trigeminal neuralgia

   It is most unusual to have autonomic features in trigeminal neuralgia and, coupled with the longish attacks (30 seconds) and the lack of any trigger, it is hard to sustain that diagnosis. The other diagnoses are possible; although giant cell arteritis would be very unusual in this age group, it is dangerous to overlook it.
4. Treatment to a certain extent will depend on the diagnosis. The treatment of the secondary problems, such

as giant cell arteritis and sinusitis, is reasonably established. He had been treated with carbamazepine, valproate, and baclofen with the provisional diagnosis of trigeminal neuralgia but these were unhelpful.

Results of Investigations

The patient had normal electrolytes, blood glucose, and blood count. The ESR was not raised and he had had a temporal artery biopsy before initial review. He had had a normal magnetic resonance imaging (MRI) of the brain and no sinus disease.

Case Outcome

I initially thought that this was the history of SUNCT without the C with the differential diagnosis being chronic paroxysmal hemicrania. He was treated with indomethacin 50 mg t.i.d. for a few weeks but this made no difference and was stopped. He saw his dentist who did some work on the right side of an ill-fitting denture and he has been completely well since that time.

Case Discussion

Initially, I had wondered whether this man had SUNCT without the C or a paroxysmal hemicrania. As it turned out he had ill-fitting dentures. It is worthwhile to discuss SUNCT in this case, because the patient presented with the same clinical picture and it is possible that he had SUNCT which went into spontaneous remission, in addition to the more simple explanation that it was fixed entirely by the dental work. The differential diagnosis, after secondary headache has been eliminated, for this type of presentation would include chronic paroxysmal hemicrania and cluster headache which are discussed elsewhere in this volume.

Most patients diagnosed with SUNCT are males. The paroxysms of pain are very short-lasting being usually between 15 seconds and 2 minutes. Patients may have up to 30 episodes an hour although more usually would have 5 to 6 per hour. The occurrence of bouts also may vary and a frequency as low as once or twice in 1 to 4 weeks has been seen in a male patient who, at other times, had up to 20 attacks a day. In another reported case the patient had almost continuous attacks for up to 3 hours. The conjunctival injection seen with SUNCT is often the most prominent of the autonomic features, and tearing also may be obvious. Although these and other autonomic stigmata, such as sweating of the forehead or rhinorrhea, may be seen in many short-lasting headache syndromes, the injection and tearing are an order of magnitude more obvious in SUNCT. Most cases have some associated precipitating factors which may be movements of the neck.

There have been a few reported secondary or associated SUNCT syndromes. The first reported secondary SUNCT syndrome was due to a homolateral cerebellopontine angle arteriovenous malformation diagnosed on MRI. It is notable that two other secondary SUNCT syndromes have both been posterior fossa arteriovenous malformations so that cranial MRI is justified in these patients. The various short-lasting headaches, including their treatments, have recently been reviewed in detail.

Selected Readings

Goadsby PJ, Lipton RB. A review of paroxysmal hemicranias, SUNCT syndrome, and other short-lasting headaches with autonomic features, including new cases. Brain 1997;120: 193–209.

Editorial Comments

Rare and unusual short-lasting headaches are interesting diagnostically and important because of often effective, specific therapeutic interventions. Therefore such cases are even more amazing when their causation is found to be not related to the disorders described. Dr. Goadsby presents such a case and indicates the patient's headaches resolved when "ill-fitting dentures" were repaired! It is so easy to overlook dental causes of facial and head pain. This case also provides a good overview of SUNCT. Time will tell whether Dr. Goadsby's patient remains headache free, let us hope such is the case.