Migraine with Multiple Sensory Auras

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Abstract-

Migraine auras are typically visual in nature but can manifest as disturbances in somatosensory, auditory, and olfactory senses. Reports of multiple sensory auras are rare in the literature, but their existence may offer novel insights into the pathogenesis of this highly common yet complex neurological condition. Here we report a case of multiple sensory auras involving somatosensory, auditory, and olfactory disturbances in a patient with migraine without visual manifestations.

A 45-year-old woman with a 20-year history of migrainous headaches presented with complaints of right-sided facial and hand numbness and paraesthesia. In addition to somatosensory symptoms, she eventually presented with tinnitus, cutaneous allodynia, and phantosmia, each of which was temporally associated with episodes of headache. No abnormalities were detected on NCS, EEG, MRI, and laboratory investigations. Her symptoms were managed by prophylactic medications and acupuncture. The theories of cortical spreading depression, cortical sensitization, and thalamocortical network involvement were discussed as possible explanations for sensory auras in migraine. This case report of migraine with multiple sensory auras spanning somatosensory, auditory, and olfactory modalities offers novel insights into the pathophysiology of migrainous auras.

Acta Neurol Taiwan 2018;27:50-53

INTRODUCTION

Focal neurological symptoms in association with migrainous headache, also known as auras, are most commonly visual in nature. Less prevalent are auditory and olfactory phenomena, of which an increasing number of cases are being reported. Neither auditory nor olfactory symptoms are recognized by the newest guidelines set by the International Classification of Headache Disorders as typical migraine auras[1,2], yet their existence may offer insights into the pathogenesis and diagnosis into this most common primary headache disorder. We report a unique patient with migraine accompanied by auras that span three sensory modalities: somatosensory, auditory and olfactory, without visual manifestations.

CASE REPORT

A 45-year-old woman with a 20-year history of intermittent right-sided headaches associated with

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Received October 1, 2015. Revised November 16, 2015.
Accepted December 21, 2015.

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photophobia and phonophobia presented with complaints of right-sided facial and hand numbness associated with right-sided headache. Her headaches were unilateral, pulsating, occurring twice weekly and often accompanied by nausea and vomiting, although no motor weakness or visual scotomata was reported. Clinically, no focal neurological deficits were noted, and a structural brain MRI with contrast detected no abnormalities. The patient's symptoms prompted a visit to the Emergency Department, after which she was prescribed paracetamol and amitriptyline, given a diagnosis of migraine with sensory aura, and discharged.

In the follow-up clinic visit, the patient was noted to have patchy numbness over the right face, scalp, fourth and fifth finger of the right hand, and the third right toe and inner thigh. These somatosensory symptoms did not display specific patterns of propagation but consistently coincided with multiple episodes of unilateral headache. On examination, sensation to pinprick was reduced in the right V2-V3 distribution, the right fourth finger, anterior aspect of tibia, right toes and inner thigh, without other neurological deficits. The patient was reviewed in clinic three months later, wherein she reported persistent episodic headaches associated with numbness over the right cheek, hand, and toes. Her sensory symptoms were preictal in nature and became worse before each headache episode started. The patient's prophylactic dose of amitriptyline was titrated up to 10 mg daily.

In view of the localized nature of the positive and negative somatosensory symptoms she experienced, laboratory investigations and nerve conduction studies were ordered to exclude other possible causes including neuropathy. Blood tests including a full blood count, electrolyte and renal panel, autoimmune screen for anti-phospholipid syndrome, rheumatoid factor, and vasculitis markers were noted to be within normal limits. Electroencephalogram (EEG) studies interictally did not reveal abnormal waveforms or epileptic discharges. Results from both motor and sensory conduction studies, as well as a sympathetic skin response study did not demonstrate any electrophysiological evidence of motor and sensory neuropathy, or autonomic dysfunction. The patient did not report the existence of visual auras at any point in time. Amitriptyline was titrated up to 15 mg daily, to which the patient responded well, reporting fewer incidences of headache which were then lessened to twice a month on average.

Approximately six months later, the patient complained of moderate intensity headaches (6/10 on the visual analogue scale) which are associated with hearing a constant high-pitched sound. The tinnitus described was not associated with vertigo, but was bilateral and episodic in nature, being worse during a bout of migrainous headache and completely resolving after each headache episode. Based on the available evidence, the patient was advised to undergo acupuncture[3]. Following this, the frequency of headaches was reduced, but each headache remained associated with symptoms of tinnitus and paraesthesia.

The patient continued a therapeutic regime of acupuncture and pharmacological prophylaxis comprising amitriptyline, topiramate and sodium valproate, with some resolution of headache intensity. She still complained of paresthesia as well as decreased sensation of the right fingers and during this period, she also reported cutaneous allodynia of her right third finger. Notably, the patient, who had no known history of psychiatric disorders, currently reported experiencing a "burning smell" that immediately preceded her headaches 1 month later. An otolaryngology work-up to rule out organic causes of her phantosmia, including naso-endoscopy and regular structural MRI of the skull base, did not reveal any abnormalities that may account for this novel symptom. The olfactory hallucinations she experienced spontaneously resolved after each migraine attack. At no point during this period was her sense of taste affected. Psychological and psychiatric evaluation proved negative for somatization disorder. Prophylactic amitriptyline was administered at 15 mg a day, with partial resolution of headache frequency and intensity as well as a mild decrease in sensory auras.

Figure 1 depicts time course of symptom onset, frequency of headaches, and treatment modalities in a line diagram.

DISCUSSION

The pathophysiology of migraine auras is largely unknown[4]. A leading theory, cortical spreading depression (CSD), stipulates a general dysfunction throughout the neocortex, which may account for disturbances in

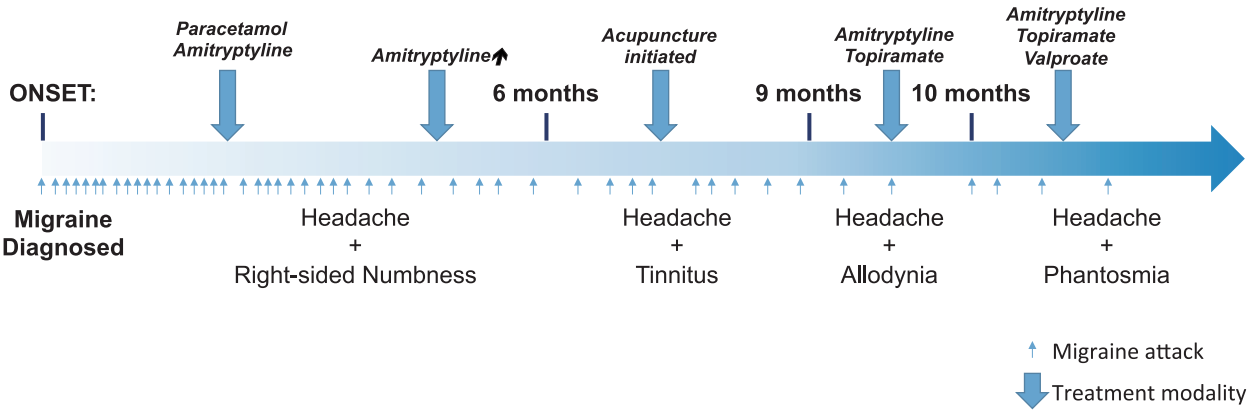

Figure 1. Line diagram depicting time course, frequency of headaches, and treatment modalities in a line diagram.

numerous sensory modalities[5]. Most migraine auras involve visual and somatosensory symptoms, but it is now known that auditory and olfactory symptoms are also possible[6-10]. Here we present a case of a patient who unusually exhibited positive and negative somatosensory symptoms, as well as auditory symptoms of tinnitus and most interestingly, olfactory hallucinations. Given the lack of typical visual disturbances and the persistence of sensory symptoms, epilepsy must be excluded as a possible alternative diagnosis. One weakness of the methodology presented is the use of routine EEG which was performed interictally, thereby not being able to exclude the possibility of ictal epileptiform discharges or slow potential shifts caused by spreading depression. In addition, the EEG was performed without Direct Current analysis which may have been able to detect CSD itself. Nevertheless, the close temporal association between headaches and sensory symptoms exhibited in this patient was consistent with established diagnostic criteria for migraine[11]. To this end, we tailored treatment modalities for this patient to include migraine prophylactic medications and acupuncture, which conferred a significant reduction in headache intensity and sensory aura occurrence, further strengthening the diagnosis of migraine.

Auditory hallucinations are rarely seen in classical cases of migraine. The patient described has episodes of bilateral tinnitus which spontaneously resolved as her headache subsided, thereby fulfilling the temporal criteria of migraine auras. Similarly, olfactory hallucinations are also uncommon in migraine patients, with a reported prevalence in one center of 0.66%[12]. In the absence of physical signs or neuroimaging abnormalitoes, the phantosmia experienced by the patient we described fulfill the three criteria of typical migraine auras, namely reversibility, duration of less than one hour, and gradual onset[1].

It is thought that cutaneous allodynia and tinnitus may both be manifestations of cortical sensitization [13]. Disturbances in smell and hearing may reflect neurophysiological changes that arise from a common origin, possibly the temporal lobe, although the patient reported here did not experience gustatory hallucinations[14]. This unique presentation illustrates the protean manifestations of the CSD phenomenon: since migrainous auras are known to be related to changes in cortical excitability, it is possible that CSD may be simultaneously prominent in the parietal and temporal cortices during the multisensory auras experienced in this patient. Recent research has suggested the involvement of a thalamocortical network as being important in migraine physiology[15]. As the thalamus is an important relay center for sensory input from different modalities, it can be postulated that this patient may have underlying abnormalities in the thalamocortical circuitry. In all, this patient highlights the possibility of dynamic functional

involvement in multiple cortical regions manifested as migrainous sensory auras. Further research is warranted to address these theoretical possibilities as they have important implications in discerning the pathophysiology and treatment of migraine with aura.

AUTHOR CONTRIBUTIONS

LL examined, treated, and counselled the patient. NO and LL participated in data collection and coordination and helped to draft the manuscript. All authors read and approved the final manuscript.

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