**Unusual presentation of trigeminal postherpetic neuralgia as Hemicrania Continua : A case Study**

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**Abstract:**

**Background**: Cranial post-herpetic neuralgia sometimes become refractory to medications. Before increasing the medications detailed history of headache phenotype is very important. **Case report**: A 41-year-old male developed a herpes infection in the right side first division of the trigeminal nerve two years back before presenting to us. He complained of continuous background dull aching headache (VAS 3 to 4) in the right orbito-fronto-temporal region. Besides this he used to experience sharp shooting severe pain (VAS 8 to 9) for one to two hours, occurring two to three times a day every five to six days for the last two years in same distribution. Episodes of exacerbation were associated with cranial autonomic symptoms (CAS), including right-side eye redness, lacrimation, and nasal congestion. His examination revealed nothing additional than allodynia in the same distribution. His MRI brain revealed no significant abnormality. Different medications were tried but in vain. He was started on Indomethacin to which he responded dramatically. **Discussion**: Non responsive cases of PHN form a refractory group and might be subjected to unnecessary medications or interventions. Secondary hemicrania continua have been reported due to different causes other than herpes. So, after ruling out secondary causes indomethacin challenge should be the next step in the diagnostic workup.

**Keywords** – PHN, Post herpetic neuralgia, Cranial Autonomic Symptoms, Secondary TACs, hemicranias continua, secondary hemicranias continua

**BACKGROUND**

The approach to diagnosis and treatment for post-herpetic neuralgia (PHN) is similar to that of neuropathic pain.1 However, different headache phenotypes have been reported after herpes infection showing better prognosis compared to other secondary or primary counterparts. 2,3,4

 International Classification of Headache Disorders (ICHD) 3 does not describe the presence or evolution of different headache phenotypes over time in PHN.5 It also does not mention the course or change in headache phenotype in patients with primary headaches after herpes infection.

In this case, we report the presence of hemicrania continua (HC) phenotype in difficult-to-treat post-herpetic neuralgia in a patient with a primary headache disorder in the same location with excellent response to Indomethacin.

**CASE REPORT**

A 41-year-old male developed a herpes infection in the right side first division of the trigeminal nerve two years back before presenting to us. He was having burning pain with vesicular skin lesions to start with, which were treated by antiviral and gabapentin by a dermatologist. But after crusting and resolution of skin lesions, he developed a change in the character and severity of the headache and was referred to a neurologist.

When he presented to us in the headache clinic to review his history, he described that after the resolution of skin lesions, he developed a continuous background dull aching headache (VAS 3 to 4) in the right orbito-fronto-temporal region. Besides this constant headache, he used to experience sharp shooting severe pain (VAS 8 to 9) for one to two hours, occurring two to three times a day every five to six days for the last two years. Episodes of exacerbations were associated with cranial autonomic symptoms (CAS), including right-side eye redness, lacrimation, and nasal congestion. He used to remain restless during these episodes. In past history, he also described stabbing headache episodes of severe intensity (VAS 8 to 9) lasting for one to two hours in the right orbitofrontal region, occurring in one to two episodes every month for the last ten years. He never consulted for these episodes and used to take oral or injectable painkillers over the counter for these.

 His past records and drug history showed consultations with many doctors. He was treated as a case of post herpetic neuralgia and was on carbamazepine 400 mg, oxcarbazepine 900 mg, gabapentin 300 mg, pregabalin 75 mg, and nortriptyline 10 mg. He also received an amitriptyline short course of steroids in the past for the same. He developed too much sedation without any relief from his pain.

His examination revealed nothing additional than allodynia in the same distribution. His MRI brain revealed no significant abnormality. (Figure:1)

**(Figure:1) His MRI showed a bilateral type one AICA loop 5 mm on the right side and 12 mm on the left side from the root exit point. No altered signal intensity was seen in bilateral cranial nerve VII-VIII complexes.**



He was started on Indomethacin 25 mg three times a day with proton pump inhibitors (PPIs), to which he responded in one day and reported 50 % improvement in one week and 100 % improvement over the next 3 to 4 weeks. He showed aggravation on missing doses. In follow-up, his drug was tapered, and now he is headache free on a 25 mg daily dose for the last three months.

**Discussion**

In ICHD-3, Trigeminal post-herpetic neuralgia (PHN) is kept under neuropathies and facial pains and treated primarily with medications for neuropathy or neuralgia. Its diagnosis needs a history of herpes infection followed by pain onset in the same area of more than three months.It has been noted that pain may have burning and itching, but patients diagnosed with PHN may not be responsive to multiple medications for neuropathic pain. 5

So, like in our case, a detailed history of the exact phenotype is of great help in such situations. Clear history revealed that our patient had probable trigeminal autonomic cephalalgia (ICHD- 3.5) phenotype headache for the last ten years and developed features of HC after the resolution of herpetic lesions. He was diagnosed as trigeminal PHN with secondary HC (13.1.2.2 and 3.4.2) for his recent headache episodes. This was further supported by a complete response to Indomethacin.

To the best of our knowledge, only three case reports have been published with such observations. Kürşad et al. reported two cases of PHN presenting as short-lasting neuralgiform headaches with conjunctival injection and tearing (SUNCT) responding well to pregabalin and gabapentin.2 Thomas et al. also reported two cases presenting as SUNCT who reacted well to lamotrigine and pregabalin.3 In 2018, Prakash et al. said two cases had hemicrania continua-like presentation in PHN and complete response to Indomethacin.4 Our patient presented with a hemicrania continua phenotype similar to this study, but unlike others, he had features of probable TACs for ten years (before herpes infection).

Irritable nociceptor and deafferentation models are proposed to explain pain in post-herpetic neuralgia.1 Patient of primary hemicrania continua has shown activation of the contralateral posterior hypothalamus and ipsilateral dorsal rostral pons.6 How this herpetic infection (peripheral nerve damage) leads to a new phenotype or conversion of already existing probable TACs in hemicrania continua is unknown to us. We also speculate the possibility of a change in primary headache phenotype after herpes infection in our case. To the best of our knowledge, no such observations have been reported to date.

Such non responsive cases of PHN form a refractory group and might be subjected to unnecessary medications or interventions. Secondary hemicrania continua have been reported due to different causes other than herpes.7 So, after ruling out secondary causes indomethacin challenge should be the next step in the diagnostic workup.

The presence of CAS and non-responsiveness to medications in PHN cases should raise suspicion of the development of secondary TACs.

**Conclusion:** In refractory cases of post herpetic neuralgia history should be reviewed for transformation to other headache phenotypes commonly to trigeminal autonomic cephalalgias.

**Declaration of conflicting interests**

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**Clinical Implications:**

* Refractory cases of PHN can transform to other TACs
* Determining headache phenotypes is helpful in treating patients of refractory PHN.

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Response to reviewers comments –

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| S.No.  | Reviewers comments  | Answers  |  Page/section/Para  |
| 1. | The patient has hemicrania continua, however, the pain is mild. There can be few other possibilities for the pain as well (esp. when pain is milder), were they looked for? | Thanks for the comment. Patient was having mild continuous pain with severe episodes of short lasting headache and with presence of cranial autonomic symptoms was qualifying for Hemicrania continua as per ICHD 3. It was further confirmed by good response to indomethacin which is one of the diagnostic criteria for the diagnosis. Other possibilities including TTH, neuropathic pain etc. were considered and he has received treatment for them already in last two years. | Case report section , para 2 |
| 2. | The patient did not get improvement with gabapentin/carbamezepine, but relieved on indomethacin. Please elaborate | Thanks for the comment. This headache phenotype( Hemicrania continua with Paroxysmal Hemicrania) is one of the indomethacin responsive headaches. Hence indomethacin responsiveness has been included as one of the diagnostic criteria in ICHD 3 for these headaches. Exact mechanisms still has not been clear for this particular response. | Case report section , para 3 |