

## Reversible cold-stimulus headache after thalamic hemorrhage: A report of two cases

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### Abstract

Cold-stimulus headache is a primary headache syndrome which is provoked by an external application or ingestion or inhalation of cold stimulus. It has not been reported to occur secondary to another focal structural brain lesion, or as a reversible illness. This is a report of two women who developed cold-stimulus headache on taking ice cold food after the onset of thalamic hemorrhage. The headache was typical of cold-stimulus headache except a relatively long duration of pain lasting half an hour. There was spontaneous remission after a few months. Our patients suggest that cold-stimulus headache can be secondary to thalamic hemorrhage.

### INTRODUCTION

Cold-stimulus headache (CSH) is a primary headache syndrome provoked by an external application (13.11.1 G44.8020) or ingestion or inhalation of cold stimulus (13.11.2 G44.8021).<sup>1</sup> It is previously termed “ice-cream headache” or “icepick headache”. CSH is a short-lasting pain, usually severe, and is induced in susceptible individuals by the passage of cold material (solid, liquid or gaseous) over the palate and/or posterior pharyngeal wall. The reported prevalence of CSH ranges from 31% to 46% in healthy subjects or community based survey<sup>2-3</sup>, 8% in large population study<sup>4</sup>, 37% in subjects from headache-referral clinic<sup>5</sup>, and 27%-93% in migraineurs.<sup>3,6</sup> Till now, CSH has not been reported to occur secondary to a structural focal brain pathology. We report here two women whose CSH occurred after they had thalamic hemorrhage.

### CASE REPORT 1

This was a 44-year-old Taiwanese woman who had an acute onset of right thalamoganglionic hemorrhage (Figure 1), which manifested as left hemiparesis (Grade 3/5) and dysesthesia. After the acute stroke, she had persistent left hemiparesis and thalamic pain syndrome. She denied past history of migraine, muscle contraction headache, exertional headache, craniofacial trauma, or illicit drug usage prior to the cerebral hemorrhage. She previously used to take ice cold snacks such as ice cream (especially strawberry ice cream) and

cold drink, and did not experience any cranio-oro-facial pain. After the cerebral hemorrhage, she initially did not take any ice cold food, as the local culture regarded taking cold food as unhelpful for facilitating recovery from the stroke illness. At the third month after the stroke, she felt well enough to take cold food again. She complained of an abrupt onset of sharp, stabbing and teasing pain over her head after she drank a mouthful of cold water. The pain was mild to moderate in severity, located at the vertex and frontal areas, and subsided spontaneously after



Figure 1. Cranial computer tomography showing a hematoma at the right thalamus and putamen (arrow), compressing the thalamus and the adjacent internal capsule.

half an hour. During the headache attack, there was no ocular dysautonomia, visual disturbance, pain at the tooth, ear canal or other orofacial structures, facial flushing, cheiro-oral paresthesia, or alimentary tract discomfort. There was however a transient exacerbation of numbness over the left upper limb. She had recurrence of similar symptoms on the following weeks, all induced by cold food. She thus decided to consult our Clinic. On examination, her vital signs were stable. Neurological examination revealed spastic hemiparesis (Grade 4/5), hyper-reflexia, positive Hoffmann sign, and paresthesia at left limbs similar to the previous level. She was thought to have CSH, although the duration of pain was relatively long. She declined to repeat the cranial computerized tomography (CT), which was recommended. She did not try ice cold food again after the consultation, until three months later, when she did not experience headache, when she took ice-cream or ice cold drink. In the period between her cerebral hemorrhage and recovery of CSH, she did not experience any migraine, headache or craniofacial pain during the cold weather in winter, or on exposure to wind. She had a total of 15 months follow up after the acute stroke.

## CASE REPORT 2

This was a 57-year-old Taiwanese woman

suffered an acute onset of left thalamoganglionic hemorrhage (Figure 2A) which manifested as right hemiparesis (Grade 3/5) and dysesthesia. She had residual deficits of hemiparesis, thalamic pain syndrome, and dystonia of the right limbs. She denied having migraine, muscle contraction headache, exertional headache, craniofacial trauma, or illicit drug usage prior to the cerebral hemorrhage. She said that she would occasionally take ice cold food such as ice-cream or drink, and never experience any cranio-oro-facial pain. One month after the cerebral hemorrhage, she had a first-ever headache after she had a mouthful of ice-cream. The pain was abrupt in onset, moderate in intensity, sharp, stabbing and teasing in nature, located at the vertex, frontal and retrobulbar region, and subsided spontaneously after half an hour. During the headache attack, there was no ocular dysautonomia, visual disturbance, pain at the tooth, ear canal or other orofacial structures, facial flushing, cheiro-oral paresthesia, or alimentary tract discomfort. She had recurrence of similar headaches over the following three months, which prompted her to seek consultation from our clinic. On examination, her vital signs were stable. Neurological examination revealed spastic hemiparesis (Grade 4/5), hyper-reflexia, positive Hoffmann sign, paresthesia and dystonia at the right limbs similar to the previous level. We thought that she had CSH, although the duration

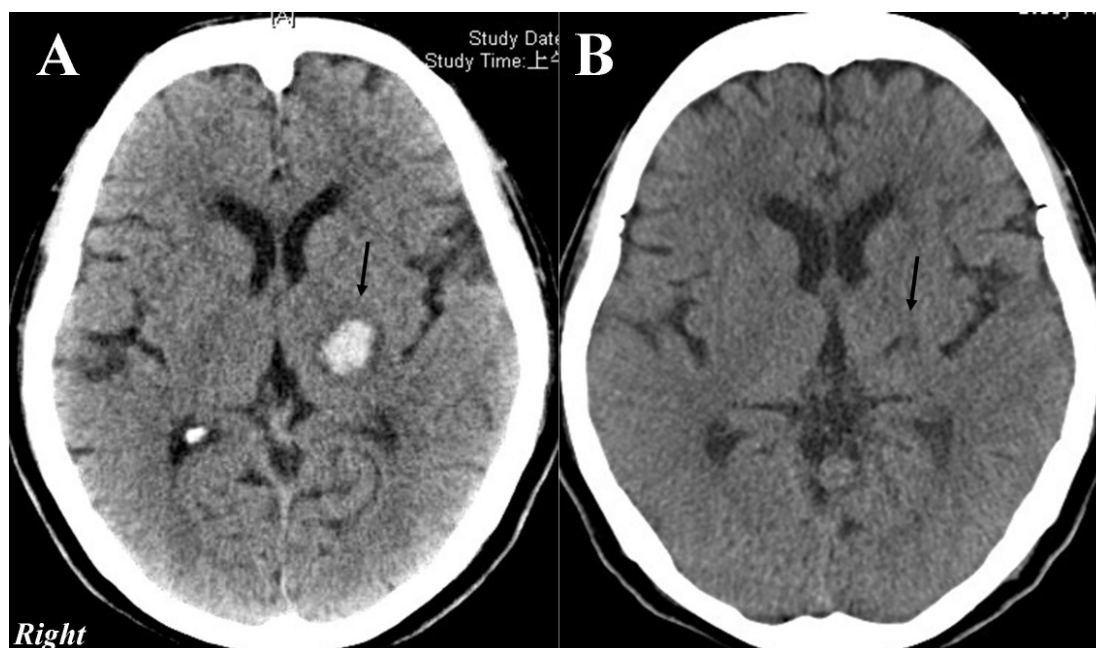


Figure 2. Cranial computer tomography showing a hematoma at the left thalamus and putamen (arrow) (A), and resolution of the hematoma 4 months later (arrow) (B).

of pain was longer than usual. Cranial CT revealed a residual hypodense lesion from the previous thalamic hemorrhage (Figure 2B). The CSH progressively declined in pain intensity during the next two months, without any active treatment. Between the onset of cerebral hemorrhage and recovery of CSH, she did not have migraine, headache or craniofacial pain during the cold weather in winter, or upon exposure to wind. The total follow up from acute stroke was one year.

## DISCUSSION

New-onset headache may occur following stroke<sup>7</sup> or cerebral concussion.<sup>8</sup> They are mostly migraine or tension-type headache. Complications from delayed cerebral hemorrhage, increased intracranial pressure, acute hydrocephalus, arterial dissection may also cause new-onset headache. Our patients did not have clinical or radiological evidence of these complications to explain their headache.

We believe that both our patients have CSH. This is based on the typical description of their pain, and the close temporal relationship of their headache to the ingestion of ice cold food. The main unusual feature of our patients as compared to the classical CSH is the longer duration of pain of our patients, lasting up to half an hour. CSH typically begins a few seconds after a rapid ingestion of cold foods or beverages. It peaks in 30 to 60 seconds<sup>3</sup>, and recedes 10 to 20 seconds later.<sup>3</sup> The pain may rarely persists for two to five minutes. We believe that the pathology in thalamus, which caused the CSH by modulating the pain response, have also modulated the time control, resulting in the prolonged duration of the pain.

Both of our patients developed CSH after thalamic hemorrhage. Their CSH was in remission after a few months. CSH has been reported to be more common among patients with past history of head injury, especially in females.<sup>2</sup> However, there was no previous report of CSH arising from focal brain damage. Our cases thus demonstrate that CSH can be secondary to a focal structural pathology, in particular from thalamic hemorrhage. The onset of their CSH was one to 3 month after the cerebral hemorrhage, although the CSH could have developed earlier, as both our patients did not take any ice cold food in sufficient amount immediately after their cerebral hemorrhage, due to local cultural taboo. There may thus be an underestimation of the true incidence of secondary CSH, due to lack of awareness, as

well as change of food habit after acute illness as in our culture. In fact, it is not just abstinence from cold food, but a slow ingestion of the food that could ameliorate the pain attack in CSH<sup>9</sup>, thus reducing its manifestation.

After the cerebral hemorrhage, our patients did not experience migraine, ear symptom, muscle contraction headache or other orofacial pain in response to various environmental triggers. CSH is usually associated with migraine<sup>2-3,5-6</sup>, suggesting some shared common mechanism. In contrast to migraine, CSH is more common in male adolescence<sup>2</sup>, and is induced by cold but not other thermal stimulus. Therefore, CSH is likely to involve mechanisms separate from migraine.

Besides the 'doctrine of specific nervous energies' or 'labeled lines' postulation, the modality-specific peripheral sensory neurons expressing multiple transduction molecules currently frames the specific sensory response model in human.<sup>10</sup> CSH is specifically provoked by an application of cold stimulus to the oropharyngeal area. In recent years, three subtypes of sensory receptors, which interacted with each another, are found to contribute to the cold signal transduction<sup>11</sup> that is conveyed to the thalamic nuclei for modulation. In our patients, the hemorrhagic stroke presumably caused an alteration of thalamic modulation of cold transduction, resulted in the development of CSH. An involvement of the transduction signal of cold but not other thermal modality changes as in our patients also supports the concept of modality-specificity of sensory modulation. On the other hand, a reversible recovery of CSH implicates a dynamic neural activity for cold signal integration.

In conclusion, we reported two patients with CSH from thalamic hemorrhage. Thus, CSH may occur secondary to a focal structural brain lesion, in particular, thalamic hemorrhage. The secondary CSH may have longer pain duration, and reversible with spontaneous remission.

## DISCLOSURE

Conflict of Interest: None

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