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Spontaneous Cortical Intracerebral Hemorrhage Causing Ipsilateral Hand and Oral Sensory Change Without Motor Deficit: A Case Report

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HIGHLIGHTS

- This is a report of a parietal cortex intracerebral hemorrhage with sensory change.
- Cheiro-oral syndrome (COS) was characteristic sensory change of perioral and hand.
- In this case patient is similar to a disease called COS in the past.

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Conflict of Interest
The authors have no potential conflicts of interest to disclose.

ABSTRACT

Intracerebral hemorrhage (ICH) is the second most common stroke subtype associated with high morbidity and mortality rates. Although various brain regions are susceptible to ICH, putaminal hemorrhage is the most common, whereas cortical ICH is less common. Here, we report the case of a 69-year-old man who developed a parietal cortical ICH. The patient developed hypoesthesia and paresthesia in the right upper lip and hand; however, the weakness was not severe. Twenty-five days after the ICH onset, the manual muscle test results were normal, but he had difficulty eating and shaving because of decreased hand dexterity. The rehabilitation focused on improving fine hand motor function and endurance. On the 94th day after ICH onset, paresthesia remained only in the fingertips, and the upper lip sensory change disappeared. Patients with sensory symptoms in the perioral area, hands, and brain lesions were previously referred to as having cheiro-oral syndrome (COS). With the advancement of neuroimaging, the use of this term has decreased, as cerebrovascular events can explain patient symptoms in correlation with neuroanatomy, etiology, and pathogenesis. We report a patient with cortical ICH, also known as COS, which is a stroke syndrome with a good prognosis.

Keywords: Intracerebral hemorrhage; Hypoesthesia; Paresthesia; Parietal lobe

INTRODUCTION

Intracerebral hemorrhage (ICH) accounts for approximately 10%–20% of all patients with stroke and is associated with greater morbidity and mortality than ischemic stroke [1]. Additionally, patients with ICH should be considered critical compared to other neurological conditions because they have disabilities and risk for recurrent stroke, cognitive decline, systemic vascular disorder, etc. The main causes of ICH in adults are hypertension, cerebral amyloid angiopathy, and anticoagulation.

Previous studies have shown that hypertension is the major cause of ICH, and that the incidence of ICH is reduced when blood pressure is controlled and maintained at a lower level. Typically, spontaneous ICH occurs in deep brain structures, including the basal ganglia, thalamus, and pons. The mechanism involves weakening the wall of the small

Author Contributions

Conceptualization: Kim M, Han S, Oh J; Data curation: Shin D, Han S; Formal analysis: Shin D, Oh J; Funding acquisition: Han S; Investigation: Kim M, Oh J; Methodology: Kim M, Oh J; Project administration: Kim M; Resources: Kim M, Han S; Software: Han S; Supervision: Oh J; Visualization: Han S; Writing - original draft: Kim M, Oh J; Writing - review & editing: Kim M, Shin D, Oh J.

cerebral vessel that crosses the deep brain structure. This is due to the rupture of deep vessel microaneurysms, hypertensive cerebral vasculopathy, or cerebral amyloid angiopathy [2]. However, a significant number of ICH cases unrelated to hypertension occur in brain regions, such as the cerebral cortex [3]. We present a patient with focal ICH in the parietal cortex who had unilateral sensory changes without weakness in the perioral region and hands, with no medical history other than new-onset hypertension.

CASE DESCRIPTION

A 69-year-old man with no significant medical history suddenly developed paresthesia and numbness in the right upper lip area and slight weakness in the right upper extremity. The symptoms persisted for one week, and he visited the neurosurgery outpatient department, where he was diagnosed with left parietal ICH based on brain computed tomography (CT) and magnetic resonance imaging (**Fig. 1**). Brain and neck magnetic resonance angiography (MRA) was performed to confirm the absence of arteriovenous malformations (AVM) or carotid stenosis. Blood tests revealed a total cholesterol level of 196 mg/dL and a triglyceride level of 92 mg/dL, which were within the normal range. Amyloid A test results were also within the normal range, 5.2 mg/L. Therefore, the patient was admitted to the neurosurgery department. At that time, he was diagnosed with hypertension and treatment with a calcium channel blocker (amlodipine, 5 mg) was started. Subsequently, the patient's systolic blood pressure remained below 145 mmHg.

Twenty-five days after ICH onset, the patient was referred to our department for rehabilitation. He had paresthesia and hypoesthesia on his right hand's palmar surface (**Fig. 2**). Although the manual muscle test results were normal, his finger and hand had decreased dexterity due to hypoesthesia, resulting in decreased fine motor function of his right hand. Therefore, he had difficulty using spoons, chopsticks, and shaving. Additionally, he experienced drooling owing to oral hypoesthesia, which made eating difficult.

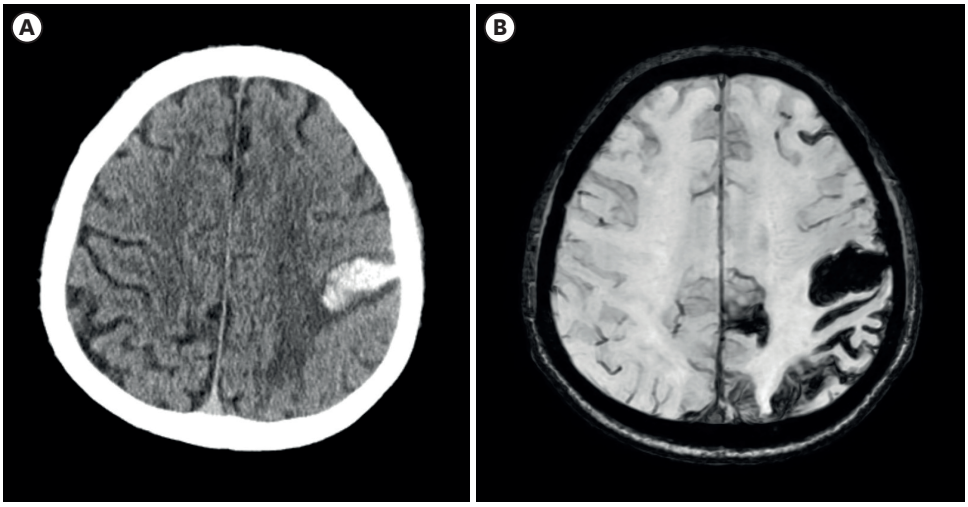

Fig. 1. Initial brain image study. (A) Brain computed tomography shows a hyperdense lesion in the left parietal lobe, suggesting hemorrhage. (B) Brain susceptibility-weighted magnetic resonance imaging shows decreased intensity in the left parietal lobe, suggesting acute hemorrhage.

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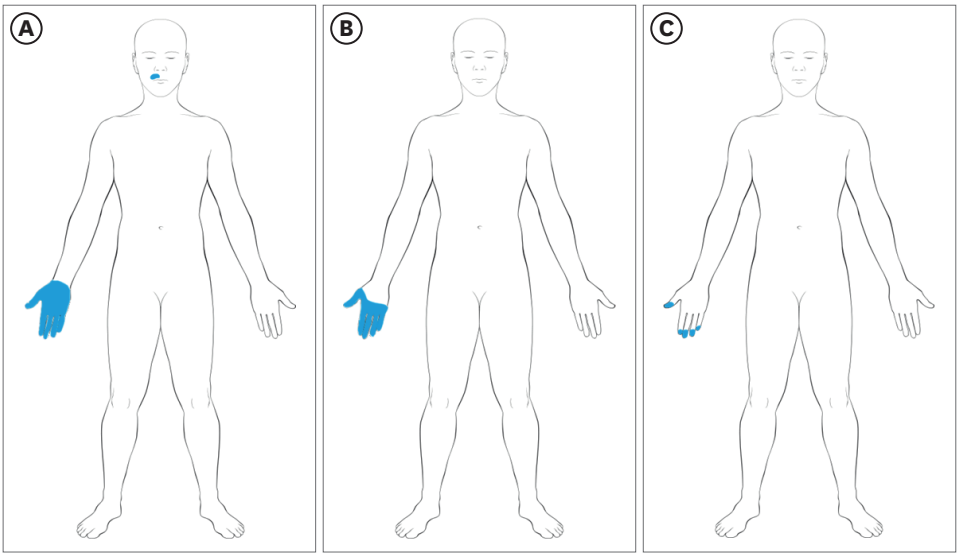

Fig. 2. Serial follow-ups of paresthetic areas. (A) Twenty-five days after ICH onset. Paresthesia was observed in the entire right hand (palmar surface) and right upper lip. (B) Forty-seven days after ICH onset. The paresthetic area decreased to the right distal hand, and sensory change on the lip disappeared. (C) Ninety-four days after ICH onset. The paresthesia persisted in the right fingertips only.
ICH, intracerebral hemorrhage.

Patient's activities of daily living (ADL) were measured using the Korean-modified Barthel Index Evaluation (K-MBI) with 96 points. Upper extremity function was assessed using a manual function test. His right upper extremity score was 26 points with 3 points for 'carry a cube' and 1 point for 'Peg-board.' His left extremity score was 29 points. In the patient's pinch strength evaluation, lateral pinch was measured equally at 13.5 kg on both hands. However, tip pinch was reduced to 5 kg on the right hand compared to 7.5 kg on the left hand, and 3-point pinch was reduced to 8.5 kg on the right hand compared to 13.5 kg on the left hand due to right hand hypoesthesia. Therefore, a rehabilitation program was conducted comprising occupational and physical therapies, focusing on enhancing endurance and improving fine hand motor function.

The patient was discharged on the 47th day of ICH onset. At that time, the patient's perioral numbness and paresthesia disappeared, and hand's paresthesia decreased, leaving only distal hand paresthesia (**Fig. 2**). When checking ADL, the K-MBI score was 100 points. He could independently use a spoon and shave, but had difficulty using chopsticks.

Ninety-four days after ICH onset, he visited our outpatient department, and his fine motor function had improved. Therefore, the use of chopsticks improved during meals, and the patient was able to eat noodles. Right hand paresthesia improved compared to that before discharge and persisted only in the fingertips (**Fig. 2**). However, fingertip paresthesia persisted even after continuous follow-up. Considering the remaining paresthesia as neuropathic pain, pregabalin 75 mg was administered twice daily, which the patient tolerated well. Brain CT performed approximately 38 weeks after ICH onset confirmed the resolution of the hematoma (**Fig. 3**).

This study was approved by the Institutional Review Board (IRB) of Seoul Medical Center (IRB No. 2023-10-004), and the requirement for informed consent from the patient was waived by the IRB.

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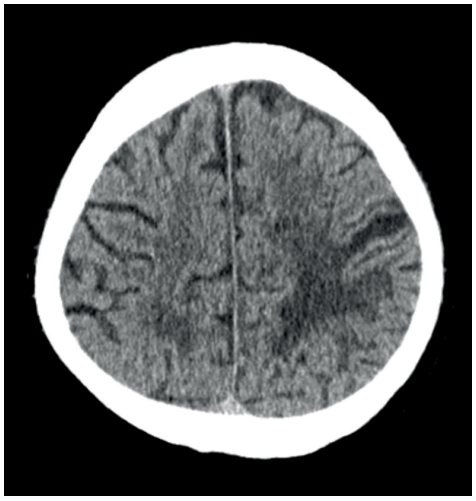

Fig. 3. Follow-up brain image. Thirty-eight weeks after intracerebral hemorrhage onset. Brain computed tomography images showing resolved hemorrhage.

DISCUSSION

Spontaneous ICH accounts for 10%–15% of all strokes worldwide, and it causes severe morbidity and mortality [4]. There are 10–30 patients with spontaneous ICH per 100,000 people each year [4], with a mortality rate of 60% during the first month of onset, and 61%–88% of survivors have residual disabilities [5]. ICH occurs when the blood vessels in the brain parenchyma rupture. Primary ICH refers to ICH without a single clear underlying lesion, and is the most common type of ICH. Secondary ICH is caused by complications from existing lesions such as AVM and tumors [6].

Several studies over the past 20 years have shown that some studies remained consistent during the same period, while others have decreased and others have increased. Considering the temporal trends of ICH risk factors, one explanation for these conflicting results may be that ICH due to hypertension has decreased due to advances in hypertension treatment, whereas the number of patients with ICH due to warfarin use has increased [6]. A large meta-analysis between 1980 and 2006 concluded that the incidence of ICH remained relatively unchanged [7].

Risk factors for ICH include hypertension, smoking, alcohol consumption, diabetes mellitus, cholesterol and lipid levels, cerebral amyloid angiopathy, medication, and genetic risk factors [6]. The patient did not regularly check his blood pressure; therefore, he was unaware that he had high blood pressure. Furthermore, the patient had a history of smoking (45 packs), but had quit smoking 3 years ago. Blood tests revealed no signs of diabetes, and the patient was not on anticoagulants, such as warfarin.

The exact mechanism linking cholesterol and lipids to ICH is not yet clear; however, low cholesterol levels are thought to weaken the endothelial wall. A study investigating the lipid fraction found that ICH risk increased when triglyceride levels were low [8]. In our case, the total cholesterol and triglyceride levels were within the normal range; therefore, ICH was not considered due to lipid problems. In addition, amyloid A levels were within the normal range, which lowered the possibility of ICH caused by cerebral amyloid angiopathy. Additionally,

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brain and neck MRA showed no AVM or carotid artery stenosis. Thus, it was confirmed that ICH was not caused by vascular abnormalities in the brain. Therefore, the patient was diagnosed with cortical ICH caused by uncontrolled hypertension.

The patient exhibited a characteristic decrease in sensation. Paresthesia and hypoesthesia were observed in the ipsilateral perioral region and hand, a suitable area for the patient's brain lesion. Considering the brain homunculus, the sensory representation for the hands, eyes, nose, face, and mouth is continuous; however, the patient showed sensory changes only in the upper lips and hands. Similar cases were first reported by Sir et al. [9] in 1914, in which 3 patients had paresthesia limited to the ipsilateral mouth angle and distal fingers/hand. An autopsy revealed encephalitis, gumma, and vascular malformations in the contralateral postcentral gyrus of the parietal lobe. This characteristic sensory disorder was later referred to as cheiro-oral syndrome (COS).

COS is characterized by the unique distribution of sensory symptoms that are observed in central nervous system diseases. COS is mainly linked to small cerebral infarctions, but can also occur in various diseases such as brain tumors, cerebral hemorrhage, cortical infarction, tuberculoma, subarachnoid hemorrhage, and migraine [10]. Based on previous COS reports, lesions can occur when there is injury to the cerebral cortex, midbrain, pons, thalamus, internal capsule, or corona radiata [11]. Therefore, COS is caused by damage to the area where the sensory fibers of the hand and mouth exist close to any level of the cerebral cortex in the parietal lobe, thalamus, and brainstem [10]. According to previous studies, most patients have thalamic and pons lesions, and few have cerebral cortex lesions [12,13]. In this case, the patient's muscle strength was normal; however, he experienced changes in perioral and hand sensations, and these sensory changes recovered quickly. These symptoms appeared similar to those of COS, which showed a relatively good prognosis in previous studies [13]. Therefore, we believe that the patient likely had COS.

The term COS is not frequently used because rapid advances in intracranial neuroimaging studies have facilitated neuroanatomical correlation and provided better insight into the etiology and pathogenesis of cerebrovascular events. Therefore, it is possible to explain the patient's symptoms related to brain lesions, which seem to have decreased the use of this term. Cortical hemorrhage related to COS is regarded as a stroke syndrome [13].

Our case demonstrates that using neuroimaging studies and other laboratory examinations for diagnosing cortical hemorrhagic lesions improves management of risk factor and early intensive rehabilitation treatment, ultimately improving patient recovery and quality of life.

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