Cheiro-Oral Syndrome Due to Lesions in the Corona Radiata

Tsuyoshi Omae, MD; Takashi Tsuchiya, MD; and Takenori Yamaguchi, MD

Background and Purpose: We describe three patients with cheiro-oral syndrome caused by a small lesion in the corona radiata confirmed by high-resolution magnetic resonance imaging.

Case Descriptions: Case 1: A 56-year-old hypertensive man who developed hypesthesia and paresthesia in the left perioral area and hand was found to have a small hematoma just lateral to the right internal capsule. Case 2: A 67-year-old man noticed hypesthesia around the left mouth angle and thumb and index finger. Magnetic resonance imaging revealed a lesion in the right corona radiata. Case 3: A 45-year-old hypertensive man developed numbness in his perioral region and left hand that later spread to his shoulder. Magnetic resonance imaging revealed a recent small infarct in the lower lateral aspect of the right corona radiata.

Conclusions: A small lesion in the corona radiata can cause cheiro-oral syndrome, whose pathogenetic mechanism in such patients may be explained by the somatotopical location or by the differing vulnerability of the neuropils in the corona radiata.

KEY WORDS • hypesthesia • magnetic resonance imaging

Cheiro-oral syndrome is a unilateral sensory disturbance limited to the hand or fingers and the ipsilateral perioral region. This syndrome was initially attributed to a lesion in the contralateral postcentral gyrus of the parietal lobe by Sittig. Later, this unusual distribution of sensory abnormalities was related to thalamic lesions. Recently, cases associated with brain stem lesions have been reported. We report three patients with a cheiro-oral syndrome caused by small lesions in the contralateral corona radiata, confirmed by high-resolution magnetic resonance imaging (MRI).

Case Reports

Case 1

A 56-year-old hypertensive man noticed numbness and tingling in his left hand when he awoke in the morning. The numbness later extended to the left perioral region. On admission to our hospital on day 2 after onset, blood pressure was 176/86 mm Hg with a regular pulse rate of 72 beats per minute. He was alert and well oriented. The cranial nerves were intact except for perioral numbness on the left side. Neither dysarthria nor weakness of the extremities was present. The only neurological signs were hypesthesia and paresthesia in the perioral area and hand on the left side. Computed tomographic (CT) scan revealed a small hematoma around the posterior limb of the right internal capsule. Magnetic resonance imaging with a 1.5-T superconducting unit (Magnetom H15, Siemens) performed on day 124 showed a small lesion at the lateral site of the right internal capsule (Figure 1).

Case 2

A 67-year-old man with a history of hypertension, diabetes mellitus, and liver cirrhosis developed dysarthria and weakness of the left hand in the afternoon. He was admitted to our hospital 4 days after onset. Blood pressure was 166/92 mm Hg. Upon neurological examination, he was found to be fully alert, and the higher brain functions were normal. He had dysarthria and slight left supranuclear facial palsy and slight weakness of the left arm. Cerebellar function was normal. The hypesthesia was located around the left mouth angle and thumb and index finger. On admission, CT scan revealed a hypodense area in the right corona radiata. High-resolution MRI on day 17 displayed a lesion in the right corona radiata. The lesion was clearly enhanced with gadolinium diethylene triamine pentaacetic acid on T1-weighted image (Figure 2).

Case 3

A 45-year-old hypertensive man suddenly developed numbness in his left hand, followed by ipsilateral perioral numbness. The symptoms disappeared about 10 minutes later. After admission to a local hospital, the same symptoms occurred again but with the numbness spreading to his left shoulder. Dysarthria and weakness of the left upper extremity also developed; however, CT scan showed no abnormality. Although the symptoms and signs had disappeared 3 days after admission, he was referred to our hospital for further examination on day 11. Blood pressure was 140/90 mm Hg, and no neurological abnormality was found. High-resolution
FIGURE 1. Case 1. Left: Computed tomographic scan of patient on day 2 reveals small hematoma in right internal capsule. Middle: Magnetic resonance imaging scan performed on day 124. Axial section, T2-weighted image (repetition time [TR] 2,300 msec, echo time [TE] 90 msec) reveals old cerebral hemorrhage (white arrow). Right: T1-weighted image (TR 40, TE 11) of coronal section demonstrates lesion in lateral aspect of higher portion of internal capsule (black arrow).

FIGURE 2. Case 2. Left: Magnetic resonance imaging scan of patient on day 17. T2-weighted image (repetition time [TR] 2,500, echo time [TE] 90) shows hyperintense area in right corona radiata. Middle: T1-weighted image (TR 700, TE 25) before gadolinium diethylenetriaminepentaacetic acid (Gd-DTPA) injection demonstrates hypointense area (white arrow). Right: T1-weighted image (TR 700, TE 25) after Gd-DTPA injection shows faint enhancement (white arrow).

FIGURE 3. Case 3. Magnetic resonance imaging scan of patient on day 16. Left and middle: T2-weighted (repetition time [TR] 2,400, echo time [TE] 90) and T1-weighted (TR 600, TE 17) images, respectively, demonstrate ischemic area in lateral aspect of right internal capsule. Right: T1-weighted image (TR 600, TE 17) after gadolinium diethylenetriaminepentaacetic acid injection indicates that lesion is recent.
MRI revealed a recent small infarct at the lower lateral aspect of the right corona radiata (Figure 3).

Discussion

The mechanism causing the peculiar distribution of sensory impairment in cheiro-oral syndrome resulting from a single lesion has been explained as a close somatotopical location in the postcentral gyrus of the parietal lobe, thalamic sensory nuclei, and medial lemniscus of the brain stem. The somatotopical localization of the hand and the mouth is close in the thalamic nuclei and medial lemniscus of the brain stem. However, the cortical topography of these regions was found by electrophysiological and anatomic studies to be separated by that of the upper portion of the face. This finding makes a simple anatomic explanation of this syndrome based on a single parietal lesion difficult. In the case of the somatic motor cortex, the cortical areas representing these regions were reported to be more sensitive to electrical stimulation than other regions. The areas in the somatosensory cortex are also thought to be so sensitive to stimuli that sensory impairment restricted to the hand and the corner of the mouth can be produced by a single lesion.

In one case of cheiro-oral syndrome in which Kinoshita et al. reported a lesion in the corona radiata, there was no discussion of the pathogenetic mechanism. Although Bogousslavsky et al. recently reported cases of this syndrome due to opercular infarct involving the underlying white matter, their discussion focused mainly on the rarity of cortical cheiro-oral syndrome caused by stroke and on the anatomic consideration.

In an experimental neuropathologic study, Jones and Powell demonstrated antegrade axonal degeneration resulting from a lesion in the ventroposterior nucleus of the thalamus in a rhesus monkey. They also found that fibers from the medial part of the ventral posterolateral nucleus and the lateral part of the ventral postero medial nucleus run together and project to the somatosensory cortex. This study indicates the possibility that the thalamocortical fibers from the hand are adjacent to those from the mouth in the corona radiata. Thus, a single small lesion can cause cheiro-oral syndrome in such a region. On the other hand, if the fibers from the hand and mouth are more vulnerable than other fibers, as is speculated to be the case in the cortex, a single lesion in the corona radiata can cause cheiro-oral syndrome.

References

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