Parietal Cheiro-oral Syndrome

Yuzuru Yasuda, Toshiyuki Watanabe and Akira Ogura

Abstract

Cheiro-oral syndrome due to a parietal lesion has been reported in conjunction with a brain tumor, infarction and migraine. Only six reports of cheiro-oral syndrome due to a parietal infarction have been reported to date. We treated a 45-year-old woman with cheiro-oral syndrome due to a parietal infarction. Her sensory disturbance was characterized by paresthesia in the lower face and hand on the left side, and severe involvement of stereognosis and graphesthesia in the left hand. The pathogenesis of parietal cheiro-oral syndrome is discussed.

Key words: paresthesia, threshold, postcentral gyrus, inferior parietal lobule, mouth, hand

Introduction

Cheiro-oral syndrome is a well-known sensory disturbance which affects the unilateral hand and mouth regions. This syndrome was first described by Sittig in 1914 (1) and was attributed to a tumor in the parietal lobe. Although thereafter there were few reports of cheiro-oral syndrome due to parietal lesion (2–4), many cases of cheiro-oral syndrome due to vascular lesions in the thalamus, brainstem and thalamocortical projections have been reported and the pathogenesis of cheiro-oral syndrome in conjunction with these lesions has been discussed. However the pathogenesis of cheiro-oral syndrome in conjunction with a parietal lesion remains unresolved. In the past 15 years, reports of cheiro-oral syndrome due to a parietal infarction have also appeared (5–10). We encountered a patient with cheiro-oral syndrome due to a parietal infarction as detected by magnetic resonance imaging (MRI) and discuss the pathogenesis of parietal cheiro-oral syndrome.

Case Report

A 45-year-old hypertensive woman suddenly developed paresthesia in the left fingers. Hemiparesis, nausea and headache were not noted, and CT was normal. The paresthesia disappeared within 2 weeks. After that she was healthy, but 6 months later, she developed dysarthria, paresthesia in the fingers and slight weakness of the upper extremity on the left side. Headache, nausea and vomiting were absent, and CT was normal. These symptoms gradually subsided in a month. However, she developed paresthesia in the lower face and hand, and slight weakness of the upper extremity on the left side one month later. Headache, diplopia and nausea were absent. As these symptoms did not improve, she was transferred to our department.

Her blood pressure was 162/100 mmHg. She was alert. Her intelligence was normal without disorientation. Eye movement was normal, with no nystagmus. No cranial nerve palsy was noted. She showed slight left hemiparesis. Deep tendon reflexes were slightly exaggerated on the left side. Babinski’s and Chaddock’s reflexes were not elicited. Paresthesia was observed in the lower face and hand on the left side. Objective sensory loss was not present in the face or mouth. Touch, pain, temperature and vibration sensation were normal, but position sensation, stereognosis and graphesthesia (recognition of letters drawn by the examiner on the palm of the hand) were nearly abolished in the left hand.

Routine blood was normal except for hyperlipidemia. Lupus anticoagulant and anticardiolipin antibodies were negative. Urine and cerebrospinal fluid examination were normal. Electrocardiogram and roentgenogram of the chest were normal. M-mode and two-dimensional echocardiographies were normal. An electroencephalogram and auditory evoked responses showed no abnormality. Somatosensory evoked potentials (SEPs) in response to electrical stimulation of the median nerve of the wrist were recorded. Stimulation duration was 0.2 msec and was of sufficient intensity to produce consistent muscle twitches. Recording electrodes were placed over right and left Erb’s point, the seventh cervical vertebra, C3, C4, and Fz. The overall band pass ranged from 5Hz to 2 kHz. Analysis time was 60 msec, and the mean of 1,000 responses was determined. N9, N11 and N13 potentials from both sides showed no abnormality. The latencies of N20 potential from both sides were normal, but the amplitude of N20 on the left side was decreased to 26.8% of the right side. MRI showed a right pre- and postcentral infarction with underlying white matter damage. The infarction was extended posteriorly to underlying white matter of the anterior part of the inferior parietal lobule (Fig.
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Figure 1. MRI showed a right pre- and postcentral infarction (arrows) with underlying white matter damage, and the infarction was extended posteriorly to the underlying white matter of the anterior part of the inferior parietal lobule. A : FLAIR images (6,856/110; inversion time, 1,700), axial view, B: T2-weighted image (2,500/80), sagittal view.

Discussion

To date, only six reports of cheiro-oral syndrome due to definite parietal infarction have been reported (5–10). Table 1 indicates the cases of cheiro-oral syndrome due to a parietal infarction. We identified three causative lesions in parietal cheiro-oral syndrome.

The first causative lesion, as revealed in the cases of Tagawa et al (5) and Isobe et al (10), is restricted to the postcentral gyrus (PCG in Table 1). The sensory dysfunction is characterized by the preservation of stereognosis and graphesthesia. Involvement of the rostral postcentral gyrus may cause decreased position and discriminative touch sense, and that of the more caudal part of the postcentral gyrus may cause decreased pain and temperature sense.

The second causative lesion is in the postcentral gyrus and the anterior part of the inferior parietal lobule (PCG+APIPL in Table 1), as described by Bogousslavsky et al (6), Mrabet et al (7) and Matsuoka et al (8). The sensory dysfunction is characterized by severe involvement of stereognosis and graphesthesia. In this group, pain, temperature, or position sense can be involved according to the extent of involvement of the postcentral gyrus. As it has been reported that the anterior part of the inferior parietal lobule plays a crucial role in tactile object recognition (11), the involvement of this area causes astereognosis. The causative lesion in the case reported by Matsuoka et al (8) was confined to the cerebral cortex in postcentral gyrus and the anterior part of the inferior parietal lobule. However, the causative lesion in the cases of Bogousslavsky et al (6) and Mrabet et al (7) was not confined to the cerebral cortex in the postcentral gyrus and was extended to the underlying white matter of the postcentral gyrus and the anterior part of the inferior parietal lobule, as in the present case.

The third causative lesion is restricted to the base of the central sulcus (BASE in Table 1), as in the case reported by Shiga et al (9), and the sensory disturbance is characterized by the absence of objective sensory loss. The primary sensory area in the cerebral cortex is thought to be in Brodmann’s area 3, 1, and 2. Brodmann’s area 3b is positioned in the anterior wall of the postcentral gyrus, but Brodmann’s area 3a is in the bottom of the central sulcus and partially in the posterior wall of the precentral gyrus. The origin of N20 potential in somatosensory evoked potentials (SEPs) to median nerve stimulation is thought to be in Brodmann’s area 3b (12, 13). The case of Shiga et al (9) showed normal N20 potential which indicates that Brodmann’s area 3b was not involved.

In a patient with parietal cheiro-oral syndrome, the reason paresthesia appears only in the hand and mouth regions has been examined. Chin, nose and face areas exist between the hand and mouth areas in the postcentral parietal cortex of
Table 1. Clinical Findings of Cheiro-oral Syndrome Following a Parietal Infarction

<table>
<thead>
<tr>
<th>Author</th>
<th>Age/Sex</th>
<th>Causative lesion</th>
<th>Topography</th>
<th>Paresis</th>
<th>Superficial sensation</th>
<th>Vibration sense</th>
<th>Position sense</th>
<th>Stereognosis</th>
<th>Graphesthesia</th>
<th>Paresis</th>
<th>SEP</th>
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<tbody>
<tr>
<td>Tagawa</td>
<td>51/M</td>
<td>PCG</td>
<td>O-A</td>
<td>D</td>
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<td>Bogousslavsky</td>
<td>63/M</td>
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<td>Present case</td>
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<td>ND</td>
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Macaca fascicularis with multi-unit microelectrode recordings (14). Because Sutherling et al reported a similarity of cortical sensory organization of the fingers between humans and other primates (15), almost the same somatotopy, namely the existence of the chin, nose and face area between the hand and mouth areas, seems to exist in humans. To date, the classical cheiro-oral syndrome has been explained by the closeness of the sensory nuclei or fibers from the hand and mouth at any level in the parietal lobe, thalamus, brainstem and thalamicortical projections. However, the precise mechanism remains unclear.

The present case showed normal latency and decreased amplitude of N20 potential, elicited by stimulation on the disturbed side, which indicates the partial involvement of Brodmann’s area 3b (12, 13). Recently, Yasuda et al reported that paresthesia recovery occurs in the following sequence: thorax, foot, mouth and then hand, in patients with cerebral infarction or hemorrhage who showed paresthesia in the thorax, foot, mouth or hand region (16). This suggests that the detection threshold, from highest to lowest, occurs in the following sequence: thorax, foot, mouth, and hand. Likewise, the detection threshold of the chin, nose and face may be higher than the mouth or hand; therefore, paresthesia appears only in the mouth and hand region in parietal cheiro-oral syndrome.

References