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# **Key Words**

Cheiro-oral-pedal syndrome Medial lemniscus Trigeminal nerve

#### Abstract

The cheiro-oral syndrome is a well-known peculiar sensory disturbance seen around the corner of the mouth and in the palm of the hand on the same side. However, sensory disturbance around the corner of the mouth, in the palm of the hand and in the foot on the same side (cheiro-oral-pedal syndrome) has not been reported until now. We examined 2 cases of cheiro-oral-pedal syndrome; the lesion responsible was confirmed by magnetic resonance imaging to be in the medial lemniscus and in the ventral ascending tract of the trigeminal nerve.

#### Introduction

Cheiro-oral syndrome is a well-known sensory disturbance around the corner of the mouth and in the palm of the hand on the same side. Possible responsible lesions are the parietal lobe [1], thalamus [2–4] and brainstem [5–7]. However, sensory disturbance around the corner of the mouth, in the palm of the hand and in the foot on the same side (cheiro-oral-pedal syndrome) has not been reported until now. We described 2 patients with pontine hemorrhage and midbrain infarct who had the cheirooral-pedal syndrome; the lesion responsible was confirmed by magnetic resonance imaging (MRI) to be in the medial lemniscus and in the ventral secondary ascending tract of the trigeminal nerve.

# **Case Report**

#### Case 1

An 81-year-old female suddenly developed left hemiplogia, headache and dysarthria on March 24, 1990. Diplopia, nausea and vomiting was not observed. She was admitted to our hospital emergently. On admission, her blood pressure was 160/90 mm Hg. She was alert and dysarthria was noted. Her intelligence was normal without disorientation. Eye movement was normal, with no nystagmus. Tongue was deviated slightly to the left. Other cranial nerves were normal. She showed left hemiplegia. Deep tendon reflex was decreased in four extremities. Babinski's and Chaddock's reflexes were not present, but palmomental reflex was elicited on both sides. Paresthesia around the corner of the mouth, in the distal forearm and in the distal lower leg was seen on the left side (fig. 1A). Superficial sense was also decreased in the same area. Deep sense was mildly decreased in the upper and lower extremities on the left side.

Routine blood, urine and CSF examination was normal. An electroencephalogram showed slow alpha activity, and auditory evoked responses showed that the latency of wave III was delayed and the amplitude of wave III was decreased on the right side. Somatosensory evoked response (SEP) stimulated by the median nerve showed normal latency of N9, N11, N13, N21, N35 on both sides, but the amplitudes of N20 and N35 elicited by stimulation of the left median nerve were decreased. SEP stimulated by the posterior tibial nerve showed a normal latency, but the amplitude of N48 elicited by stimulation on the left side was decreased. CT at the onset of the stroke showed a small high-density area in the tegmentum of right pons (fig. 2A). MR1 on June 13, 1990, showed that the lesion was situated in the tegmentum of the right pons (fig. 2B), and no lesion was found in the thalamus, parietal lobe or elsewhere.

Paresthesia in the lower leg disappeared within 2 weeks, but around the corner of the mouth and in the distal forcarm it did not improve.

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# **Cheiro-Oral-Pedal Syndrome**

#### Case 2

A 76-year-old male developed suddenly nausea, blurred vision, dizziness and headache in the morning on July 22, 1990. He could not eat because of nausea. Hemiparesis or diplopia was not seen. He was admitted to our department emergently. On admission, his blood pressure was 140/90 mm Hg. He was alert and his intelligence was normal without disorientation. Eye movement was normal. No cranial palsy was found. No hemiparesis was seen. Deep tendon reflex was slightly decreased in the four extremities. No pathologic reflex was elicited. There was no objective sensory loss, but paresthe-

Fig. 1. Cheiro-oral-pedal syndrome. Paresthesia was seen on the left side in case 1 (A) and on the right side in case 2 (B).

Fig. 2. CT scan and MR1 of case 1 (A, B) and case 2 (C, D). A CT scan in acute stage. B T<sub>1</sub>-weighted MRI in chronic stage (TR = 400 ms, TE = 25 ms). C T<sub>2</sub>-weighted MRI in subacute stage (TR = 2,500 ms, TE = 80 ms). D T<sub>1</sub>-weighted MRI in subacute stage (TR = 400 ms, TE = 25 ms).





2A

2C



2B

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sia around the corner of the mouth and in the lower face, fingers and foot was present on the right side (fig. 1B). No other abnormal CNS signs were detected.

Routine blood, urine and cerebrospinal fluid examination was normal. An electroencephalogram was normal, and auditory evoked responses showed that the latency of wave V was delayed and the amplitude of wave V was decreased on the left side. SEP stimulated by the median nerve showed that latencies of N9, N11, N13, N20 and N35 were normal on both sides, but amplitudes of N21 and N35 elicited by the stimulation of the right median nerve were decreased. SEP stimulated by the posterior tibial nerve showed normal latency, but the amplitude of N48 elicited by stimulation on the right side was decreased. CT at the onset of the stroke showed no abnormality. MRI on the 5 days after onset showed small infarct in the tegmentum of the midbrain on the left side (fig. 2C, D), and no lesion was found in the thalamus and parietal lobe.

Paresthesia around the corner of the mouth and in the lower face and foot disappeared within 3 weeks, but one of the fingers did not improve.

### Discussion

The cheiro-oral syndrome is characterized by paresthesia around the corner of the mouth and in the palm on the same side, and the lesion responsible is considered to be situated in the parietal lobe [1], a limited lesion in the inferior medial portion of the ventral posterolateral nucleus and lateral portion of the ventral posteromedial nucleus [2-4] or brainstem [5-7]. The mechanism of the syndrome is as follows; in the parietal lobe, the sensory areas of the hand and mouth are in very close proximity [8]. In the ventral posteromedial nucleus of the thalamus, the sensory nuclei of the tongue, mouth, cheek and occiput are situated from inside to outside, and in the ventral posterolateral nucleus the sensory nuclei of the hand, upper extremity, trunk and lower extremity are also situated from inside to outside [9]. The nuclei of the hand and the mouth are close together. And in the pons and midbrain, the medial part of the medial lemniscus, which transmits sensory fibers from the upper extremity to the ventral posterolateral nucleus, and the ventral secondary ascending tract of the trigeminal nerve, which transmits sensory fibers from the face to the ventral posteromedial nucleus, are very closely situated in the ventromedial part of the tegmentum [10]. Thus, the cheiro-oral syndrome can be largely explained by the closeness of the sensory fibers from the hand and mouth.

Our 2 cases showed paresthesia in the foot other than paresthesia around the corner of the mouth and in the palm on the same side. Until now, paresthesia in the foot or lower leg has not been reported in the cheiro-oral syndrome. The lesion responsible in our 2 cases was confirmed by MRI and CT to be in the tegmentum in the pons or midbrain on the contralateral side. The classical cheiro-oral syndrome can be largely explained by the anatomical proximity of fibers conveying sensory inputs from mouth and hand. However, this cannot apply to the representation of foot in the sensory pathways because the sensory fibers of the upper extremity, trunk and lower extremity are situated from inside to outside in the medial lemniscus of the pons and midbrain [10]. SEPs showed normal latency and decreased amplitude of N21, N35 and N48 elicited by the stimulation on the disturbed side, which suggests the partial involvement of medial lemniscus. Our cases might be regarded as sensory hemisyndromes in which subjective experience of symptoms was limited to the face, mouth and foot, possibly due to different detection thresholds which could be related to a partial involvement of the sensory pathways.

The disturbance of the ventral secondary tract of the trigeminal nerve and medial lemniscus could cause the cheiro-oral syndrome or the cheiro-oral-pedal syndrome according to the degree of expansion of the lesion.

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