



Cheiro-Oral-Pedal Syndrome Secondary to Thalamic Infarction: A Case Report

Ryan D*, Shah P and Le DO S

Department of Neurology, Duke University School of Medicine, USA

Abstract

The Cheiro-Oral-Pedal Syndrome (COPS) is a rare hemisensory syndrome characterized by unilateral sensory disturbances of the corner of the mouth, hand, and foot. It has been described in the setting of neurologic lesions of the brainstem and thalamus that are often vascular in nature. The close proximity of the sensory fibers and sensory nuclei of the mouth, hand, and foot in the medial lemniscus and thalamus explain this possible pattern of hemisensory loss. The possibility of these symptoms being perceived as mild and secondary to peripheral processes places patients at risk of delayed presentation and workup. We present a case of COPS to further raise awareness of a rare presentation of stroke.

Keywords: Cheiro-oral-pedal; Cheiro-oral; All cerebrovascular disease/stroke; Infarction; Other cerebrovascular disease

Introduction

The Cheiro-Oral Syndrome (COS) is a rare, but well described, neurologic syndrome characterized by paresthesia's of the corner of the mouth and hand on the same side [1]. Less has been described about the related COPS, characterized by paresthesia's of the corner of the mouth, hand, and foot on the same side [2]. This distinct pattern was first described in the 1990's [2-4] related to vascular etiologies in the brainstem and thalamus. This pattern can create diagnostic challenges and present delays in diagnosis and treatment considering the perceived nature of these symptoms. We report a case of COPS associated with infarction of the lateral thalamus.

Case Presentation

A 71-year-old male with past history of hypertension, hyperlipidemia, type 2 diabetes mellitus, and prostate cancer presented with paresthesia's of the left lower face, fingers, and toes two days prior to initial presentation. The symptoms initially resolved, but then he had recurrence of symptoms the following day. Prior to presentation, the toes and facial paresthesia's resolved, with persistence of paresthesia's of first two digits of the left hand. He denied other focal neurologic symptoms.

Upon presentation to the emergency department his blood pressure was 131/95. Exam was notable for a well appearing 71-year-old male in no distress. Sensory exam was notable for decreased pinprick in the distal thumb and second digit on the left hand. Mental status, cranial nerve, motor, reflex, coordination, and gait exam were unremarkable. Initial National Institutes of Health Stroke Scale (NIHSS) was 1.

Initial non-contrast Computed Tomography (CT) of the brain showed an age-indeterminate left internal capsule infarct. CT angiogram was without large vessel occlusion or hemodynamically significant stenosis. Initial serum workup was unremarkable. Electrocardiogram showed sinus tachycardia with 2nd degree heart block. Due to suspicion for cerebrovascular etiology, Magnetic Resonance Imaging (MRI) was performed and showed acute infarction in the right lateral thalamus. Transthoracic echocardiography was unremarkable. LDL was 115, and hemoglobin A1c was 6.3%. He was started on aspirin and clopidogrel for 21 days, atorvastatin was increased to 40 mg daily for goal LDL<70, and he was discharged with a 30-day loop monitor with follow up in stroke clinic.

Discussion

COPS is a rare neurologic syndrome characterized by unilateral sensory disturbances of the corner of the mouth, hand, and foot. The first cases described in literature were secondary to midbrain and pontine hemorrhages [2], though cases have been described secondary to thalamic infarction [3] and infarction involving thalamocortical projections in the internal capsule [4]. This

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*Correspondence:

Dylan Ryan, Department of Neurology,
Duke University School of Medicine,
Durham, NC, 27712, USA, Tel:
8609852065;

E-mail: dylan.ryan@duke.edu

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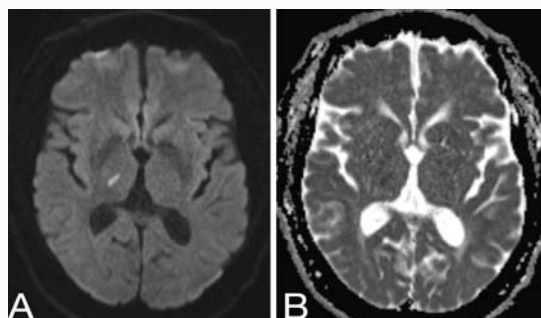


Figure 1: MRI showing area of diffusion restriction on diffusion weighted imaging (A) in the region of the right ventral posterior nucleus of the thalamus with correlate on the apparent diffusion coefficient (B) imaging consistent with acute infarct.

unusual constellation of symptoms can create diagnostic challenges due to the unfamiliarity and can lead to delays in presentation due to the perceived severity of the symptoms involved.

The related, and more thoroughly described, COS, which involves unilateral sensory disturbances of the corner of the mouth and hand, has been thought to be secondary to the close anatomical relationship of the sensory centers of the mouth and fingers in the brainstem, thalamus [3], and thalamocortical projections [5]. It is thought that this presentation occurs secondary to the proximity and associated area topographically correlated with the mouth and digits in the medial lemniscus and thalamus [5]. It has been previously noted through microelectrode mapping techniques in both humans and primates [6,7] that the sensation of the digits accounts for a large proportion of the Ventral Posterior Nucleus of the thalamus (VPN), which accounts for this pattern.

What remains unique in regard to COPS is involvement of the foot on the same side as paresthesia's of the mouth and hand. When considering ascending sensory information, the fibers from the hand and foot are often separated by considerable distance creating an unlikelihood in finding this pattern. Fibers within the medial lemniscus share closer proximity, and as the medial lemniscus rotates

in the pons, the possibility of creating this pattern of presenting symptoms arising due to brainstem lesions. This proximity and the large proportion occupied by sensation of the digits in the VPN also allows for lateral thalamic lesions to present with this unique pattern. As this information travels superiorly this proximity is lost, thus refining possible areas of localization when considering COPS.

In this case, we report a lateral thalamic infarction that presented with COPS in order to raise further awareness about this unique neurologic presentation. The lesion was an infarction of the lateral thalamus in the region of the VPN. Patients presenting with COPS warrant further neurologic workup, especially considering vascular etiologies, to prevent further complications related to the underlying disease process.

References

1. Manning S, King BR, Peffer J, Lescure D. Cheiro-Oral syndrome. *Am J Emerg Med.* 2021;39:151-3.
2. Yasuda Y, Morita T, Okada T, Seko S, Akiguchi I, Kimura J. Cheiro-oral-pedal syndrome. *Eur Neurol.* 1992;32(2):106-8.
3. Yasuda Y, Matsuda I, Akiguchi I, Kameyama M. Cheiro-oral-pedal syndrome in thalamic infarction. *Clin Neurol Neurosurg.* 1993;95(4):311-4.
4. Yasuda Y, Watanabe T, Akiguchi I, Kimura J, Kameyama M. Cheiro-oral-pedal syndrome in the lesion of thalamocortical projections. *Clin Neurol Neurosurg.* 1994;96(2):185-7.
5. Isono O, Kawamura M, Shiota J, Araki S, Hirayama K. Cheiro-oral topography of sensory disturbances due to lesions of thalamocortical projections. *Neurology.* 1993;43(1):51-5.
6. Combarros O, Polo JM, Pascual J, Berciano J. Evidence of somatotopic organization of the sensory thalamus based on infarction in the nucleus ventralis posterior. *Stroke.* 1991;22(11):1445-7.
7. Kaas JH, Nelson RJ, Sur M, Dykes RW, Merzenich MM. The somatotopic organization of the ventroposterior thalamus of the squirrel monkey, *Saimiri sciureus*. *J Comp Neurol.* 1984;226(1):111-40.