Case Report

Recurrent Patent Foramen Ovale-Related Cerebral Infarcts Alternately Causing Bilateral Hand Paresis

Seung-Jae Lee

Department of Neurology, Soonchunhyang University Bucheon Hospital, Bucheon, South Korea

Keywords

Isolated hand palsy · Monoparesis · Hand knob · Stroke · Patent foramen ovale · Embolism

Abstract

Isolated hand paresis is a rare presentation of stroke, which mostly results from a lesion in the cortical hand motor area, a knob-like area within the precentral gyrus. I report the case of a patient who experienced recurrent ischemic stroke alternately involving bilateral hand knob areas, causing isolated hand paresis. There was no abnormal finding on brain and neck magnetic resonance angiography, transthoracic echocardiography, and 48-h Holter monitoring, and there were no abnormal immunologic and coagulation laboratory findings. The only embolic source was found to be a patent foramen ovale, which was proven on transesophageal echocardiography. The patient underwent percutaneous device closure of patent foramen ovale after alternately repeated paresis of both hands despite antiplatelet treatment. This case suggests that ischemic stroke affecting the cortical knob area, albeit extremely rare, may recur due to a patent foramen ovale, and it necessitates complete investigation, including transesophageal echocardiography, to identify possible embolic sources.

© 2017 The Author(s)
Published by S. Karger AG, Basel

Background

Ischemic stroke of the cortical hand motor area causing isolated hand paresis is rare with an estimated prevalence of <1% in the total ischemic stroke population [1, 2]. Its diagnosis is often challenging in real-world clinical practice because it can simulate peripheral neuropathy [3].

Patent foramen ovale (PFO) potentially leading to paradoxical embolism is a common cause of cryptogenic stroke [4]. However, PFO is also a common anatomical variant found in approximately 25% of the general population [4, 5]. Thus, PFO can be incidental or stroke related in patients with cryptogenic stroke [5]. In fact, no benefit has been proven for the device closure of PFO in these patients [6, 7]. Thus, antithrombotic treatment has been recommended until now [8]. In addition, the risk of stroke recurrence is much lower (<1%/year) in cryptogenic stroke patients with PFO compared to the other stroke population [6].

The presented case showed such a rare repeat stroke presentation in which isolated hand paresis alternated between both sides because of recurrent PFO-related stroke involving the cortical hand motor area despite antiplatelet medication, which was eventually treated with device closure of PFO.

Case Presentation

On November 4, 2014, a 51-year-old hypertensive man visited the emergency room with a complaint of sudden right hand weakness. He had no history of drug abuse, heavy alcohol use, and cigarette smoking. In addition, there was no abnormal finding in the screening for common cancer (gastroendoscopy, colonoscopy, and abdominal sonography) performed 3 months before the visit. Initial neurologic examination showed only a subtle decrease (grade IV) in right-hand motor strength without sensory disturbances, tendon reflex abnormality, and proximal muscle weakness (motor score of the National Institutes of Health Stroke Scale = 0). There was no discernible difference in the power between median, ulnar, and radial nerve innervated hand muscles. Routine laboratory tests showed an increased fasting blood sugar level (117 mg/dL), but lipid profiles were normal except for a slightly decreased high-density lipoprotein cholesterol level (43 mg/dL): total cholesterol 180 mg/dL, triglyceride 86 mg/dL, and low-density lipoprotein cholesterol 114 mg/dL. High-sensitivity C-reactive protein level was slightly increased (1.9 mg/dL [range, 0–0.5]). However, all other immunologic and coagulation laboratory tests showed normal values: prothrombin time 12.2 s (range, 9.5–13.5), activated partial thromboplastin time 24.3 s (range, 23–35), fibrinogen 231 mg/dL (range, 170–410), fibrin degradation products 1.5 µg/mL (range, 0–5), D-dimer 0.19 mg/L (range, 0–0.65), anti-thrombin III 80.1% (range, 75–125), anti-nuclear antibody (–), anti-phospholipid antibody immunoglobulin G/immunoglobulin M (–/–), rheumatoid factor 6 IU/mL (range, 0–18), lupus anticoagulant screening (–), protein C 84.9% (range, 72.0–160.0), and protein S 75.9% (range, 69.4–138.3). Diffusion-weighted magnetic resonance imaging (MRI) demonstrated multiple acute infarcts involving the left frontal and right parieto-occipital cortical regions, including the left precentral knob area (Fig. 1a). There was no abnormal finding on intracranial time of flight and extracranial enhanced magnetic resonance angiography (MRA), transthoracic echocardiography, and 48-h Holter monitoring. The patient was treated with aspirin 100 mg, clopidogrel 75 mg, atorvastatin 20 mg, and anti-hypertensive agents. For a detailed evaluation to identi-

fy the cardioembolic source, transesophageal echocardiography (TEE) was planned, but he refused the TEE. His symptoms completely disappeared within several weeks.

On August 21, 2016, he revisited the emergency room with a complaint of suddenly developed, mild weakness (grade IV) in the left hand. He was taking clopidogrel 75 mg and atorvastatin 20 mg daily. Similar to the previous attack, there was no change in sensory function and tendon reflex. In addition, no discernible difference was observed in the power between intrinsic hand muscles. Diffusion-weighted MRI showed an acute infarct involving the right middle cerebral artery territory including the right precentral knob area (Fig. 1b). A second head and neck MRA and 48-h Holter monitoring showed no abnormal finding. Then, TEE with bubble test using agitated saline contrast was performed for further investigation of cardioembolic sources. The examination revealed a PFO, demonstrated as a visualization of more than 20 bubbles in the left atrium within 3 beats after the appearance of agitated saline in the right atrium with the Valsalva maneuver (Fig. 1c). However, there was no other high- or medium-risk embolic source, including a plaque in the aortic wall, during the examination. He was considered to have recurrent PFO-related stroke despite antithrombotic medication, and therefore, percutaneous closure of PFO was performed using the 30-mm Amplatzer septal occluder (Fig. 2).

Discussion

This case presents the following peculiar findings: (1) recurrent cortical hand knob infarcts alternately causing isolated paresis of both hands and (2) recurrence of the PFO-related stroke despite antiplatelet treatment. The cortical hand motor area is known to be a precentral knob-like area with an inverted omega shape on the axial brain image [9]. With respect to the stroke involving the “hand knob” area, not only different types of pseudoperipheral palsies have been reported [3, 10], but also functional somatotopy of ulnar-to-radial fingers along the medial-to-lateral axis in the area has been suggested in several studies [11, 12]. Moreover, upper motor neuron signs (brisk deep tendon reflex and Babinski sign) are absent in many cases [3, 10]. Thus, symptoms related to a small lesion in this area can initially be mistaken for those of peripheral neuropathy. Therefore, the clinician should make a decision to perform brain imaging by judging its abrupt nature of onset, concomitant sensory symptom inappropriate for a peripheral lesion, and vascular risk factors in the patients.

The patient had nonlacunar, multiple ischemic lesions in bilateral hemispheres, suggesting an underlying embolic source. This is comparable to previous results which show that most of the cortical hand knob strokes are explained by an embolism from carotid artery disease or heart disease [1, 2]. However, the patient had no evidence of relevant arterial stenosis or a high-risk cardioembolic source on MRA, transthoracic echocardiography, and 48-h Holter monitoring studies. In addition, the lesion distribution in bilateral hemispheres could not be explained by a nonstenotic atherosclerotic arterial segment which may have not been revealed by MRA. Moreover, there was no evidence of coagulopathy or vasculitis in laboratory findings. Hence, he was presumed to have an “embolic stroke of undetermined source” during the first stroke attack [13], and thus, antiplatelet medication was initially prescribed. The TEE performed after stroke recurrence did not show presence of a plaque within the aortic arch wall but rather a PFO as the only cardioembolic source in the patient. Because of recurrence despite antithrombotic medication, percutaneous closure of PFO was performed.

To the best of my knowledge, this case is the first report of recurrent hand knob stroke related to TEE-proven PFO. The possibility of stroke should be suspected in the presence of sudden-onset hand paresis. Once cortical hand knob stroke is diagnosed, a detailed examination should be performed to identify possible embolic sources including PFO.

Statement of Ethics

No ethics approval is needed for a case report. The patient gave informed consent for publication of his clinical details.

Disclosure Statement

The author declares that he has no competing interests.

Funding Sources

This work was supported by the Soonchunhyang University Research Fund.

Author Contributions

As the patient's treating physician, S.-J.L. created the concept and design of this work, acquired and interpreted the data, and wrote and revised the manuscript.

References

- 1 Peters N, Müller-Schunk S, Freilinger T, Düring M, Pfefferkorn T, Dichgans M: Ischemic stroke of the cortical "hand knob" area: stroke mechanisms and prognosis. *J Neurol* 2009;256:1146–1151.
- 2 Castaldo J, Rodgers J, Rae-Grant A, Barbour P, Jenny D: Diagnosis and neuroimaging of acute stroke producing distal arm monoparesis. *J Stroke Cerebrovasc Dis* 2003;12:253–258.
- 3 Phan TG, Evans BA, Huston J: Pseudounilateral palsy from a small infarct of the precentral knob. *Neurology* 2000;54:2185.
- 4 Bang OY, Lee MJ, Ryoo S, Kim SJ, Kim JW: Patent foramen ovale and stroke – current status. *J Stroke* 2015;17:229–237.
- 5 Kent DM, Ruthazer R, Weimar C, Mas JL, Serena J, Homma S, Di Angelantonio E, Di Tullio MR, Lutz JS, Elkind MS, Griffith J, Jaigobin C, Mattle HP, Michel P, Mono ML, Nedeltchev K, Papetti F, Thaler DE: An index to identify stroke-related vs incidental patent foramen ovale in cryptogenic stroke. *Neurology* 2013;81:619–625.
- 6 Dalen JE, Alpert JS: Cryptogenic strokes and patent foramen ovals: what's the right treatment? *Am J Med* 2016;129:1159–1162.
- 7 Li J, Liu J, Liu M, Zhang S, Hao Z, Zhang J, Zhang C: Closure versus medical therapy for preventing recurrent stroke in patients with patent foramen ovale and a history of cryptogenic stroke or transient ischemic attack. *Cochrane Database Syst Rev* 2015;9:CD009938.
- 8 Kernan WN, Ovbiagele B, Black HR, Bravata DM, Chimowitz MI, Ezekowitz MD, Fang MC, Fisher M, Furie KL, Heck DV, Johnston SC, Kasner SE, Kittner SJ, Mitchell PH, Rich MW, Richardson D, Schwamm LH, Wilson JA; American Heart Association Stroke Council, Council on Cardiovascular and Stroke Nursing, Council on Clinical Cardiology, and Council on Peripheral Vascular Disease: Guidelines for the prevention of stroke in patients with stroke and transient ischemic attack: a guideline for healthcare professionals from the American Heart Association/American Stroke Association. *Stroke* 2014;45:2160–2236.

- 9 Yousry TA, Schmid UD, Alkadhi H, Schmidt D, Peraud A, Buettner A, Winkler P: Localization of the motor hand area to a knob on the precentral gyrus. A new landmark. *Brain* 1997;120:141–157.
- 10 Manjaly ZM, Luft AR, Sarikaya H: An unusual cause of pseudomedian nerve palsy. *Case Rep Neurol Med* 2011;2011:474271.
- 11 Kim JS: Predominant involvement of a particular group of fingers due to small, cortical infarction. *Neurology* 2001;56:1677–1682.
- 12 Schieber MH: Somatotopic gradients in the distributed organization of the human primary motor cortex hand area: evidence from small infarcts. *Exp Brain Res* 1999;128:139–148.
- 13 Hart RG, Diener HC, Coutts SB, Easton JD, Granger CB, O'Donnell MJ, Sacco RL, Connolly SJ; Cryptogenic Stroke/ESUS International Working Group: Embolic strokes of undetermined source: the case for a new clinical construct. *Lancet Neurol* 2014;13:429–438.

Fig. 1. **a** Diffusion-weighted image of the first stroke attack, showing multiple infarcts (white arrows) involving the left frontal and the right parieto-occipital cortical regions (including the left precentral knob area). **b** Image of the second stroke attack, showing multiple infarcts (white arrows) in the right middle cerebral artery territory, including the right precentral knob area. **c** The finding of transesophageal echocardiography showing bubbles (white arrow) passing from the right atrium (RA) into the left atrium (LA).

Fig. 2. Chest X-ray (lateral view) showing the Amplatzer septal occluder (white arrow).