Transient Capgras Syndrome Secondary to Bilateral Ischemic Stroke: A Case Report.

Manveer Garcha
Lehigh Valley Health Network, Manveer.Garcha@lvhn.org

Keithan Sivakumar MD, MBA
Lehigh Valley Health Network, Keithan.Sivakumar@lvhn.org

Megan Leary MD
Lehigh Valley Health Network, Megan_C.Leary@lvhn.org

Hussam A. Yacoub MD
Lehigh Valley Health Network, hussam_a.yacoub@lvhn.org

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Transient Capgras Syndrome Secondary to Bilateral Ischemic Stroke: A Case Report

Manveer Garcha, BS, MA, Keithan Sivakumar, MD, MBA,† Megan Leary, MD,‡§ and Hussam A. Yacoub, DO, MS‡§*

Abstract: Capgras syndrome is one of a variety of delusional misidentification syndromes that can be associated with acute ischemic stroke, neurodegenerative disease, or metabolic conditions. Most cases reported in the literature are associated with frontal and/or parietal lobe involvement. Transient Capgras syndrome is rare but has been reported. We present a case of transient Capgras syndrome following bilateral cerebral ischemic infarcts in the frontal, parietal, and temporal regions, and involving the right prefrontal cortex. To our knowledge, transient Capgras syndrome with rapid resolution over a period of days is rare.

Key Words: Capgras syndrome, ischemic stroke, temporal lobe stroke

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DMS = delusional misidentification syndrome.

We describe a patient who developed Capgras syndrome 48 hours after presenting with bilateral cerebral hemispheric infarcts. Capgras syndrome is one of a variety of delusional misidentification syndromes (DMSs) that can result from bilateral cerebral infarcts involving the frontal, temporal, and parietal regions, and is characterized by the delusion that someone familiar to the patient has been replaced by an imposter who is physically identical to the known person. Capgras syndrome has been reported in association with acute ischemic stroke as well as other conditions, including metabolic and neurodegenerative disorders. Symptoms of Capgras syndrome rarely present in the acute period after ischemic stroke and are infrequently transient.

CASE REPORT

A right-handed 65-year-old woman with a history of hypertension and rheumatoid arthritis presented with a witnessed generalized tonic-clonic seizure occurring in the setting of recent malaise, diarrhea, nausea, vomiting, and dizziness. On initial evaluation in the emergency department, she was noted to be disoriented and aphasic with a right-sided visual field cut, left-sided neglect, and decreased strength in the left arm. Her National Institutes of Health Stroke Scale score was 4. A computed tomography angiogram revealed a thrombus in the proximal M2 segment of the right middle cerebral artery, as well as signs of a subacute infarction within the confluence of the left middle and posterior cerebral arteries watershed territory. She underwent successful mechanical thrombectomy of the right M2 clot.

A transesophageal echocardiogram revealed a thrombus in the apex of the left ventricle and a left ventricular ejection fraction of 20%, increasing the likelihood of a cardiac source of the cerebral infarcts. The day after initial presentation, magnetic resonance imaging of the brain revealed acute infarcts in the right frontal, insular, and occipital regions, as well as the left parieto-occipital territory (Figure 1). More specifically, the infarct in the right frontal region partially involved the prefrontal cortex, Brodmann areas 44 and 45. After the thrombectomy, the patient's neurologic examination revealed improvement in the aphasia, left-sided weakness, and left hemineglect. Symptoms of mild confusion and disorientation to time persisted. Additional hospital complications included shocked liver, transaminitis, and electrolyte abnormalities requiring repletion.

Forty-eight hours after initial presentation, the patient began to exhibit the belief that her husband was an imposter —someone who looked and sounded exactly like her husband, but was not him. With her eyes closed, the patient was able to recognize her husband's voice. Repeat computed tomography of the head revealed a questionable new infarct in the right temporal lobe. Repeat magnetic resonance imaging of the brain 3 days after initial presentation revealed several new small acute to subacute infarcts in the right frontal, parietal, and, most prominently, right superior temporal lobe (the right temporal new infarct is shown in Figure 2B). The patient was started on anticoagulation at this time, and hypertensive therapy was discontinued.

Over the course of the next several days, the patient remained adamant in the belief that her husband was an imposter, referring to him as "the other Steve, not the real Steve." When questioned, she indicated that the imposter was a close friend, but not her husband. The patient had no difficulty recognizing friends, family members, or

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From the *University of South Florida Health Morsani College of Medicine SELECT Program, †Department of Neurology, and ‡Lehigh Valley Physicians Group–Neurology, Lehigh Valley Health Network, Allentown, Pennsylvania; and §Morsani College of Medicine, University of South Florida, Tampa, Florida.
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Correspondence: Hussam A. Yacoub, DO, MS, Lehigh Valley Physicians Group–Neurology, 1250 S. Cedar Crest Blvd, Suite 405, Allentown, Pennsylvania 18103 (e-mail: hyacoub123@yahoo.com).

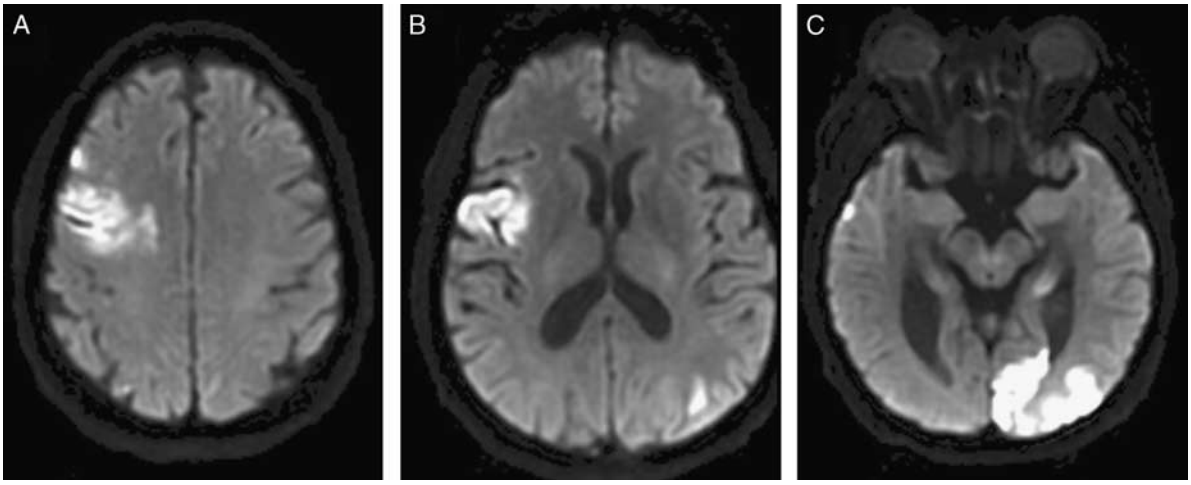

FIGURE 1. Axial magnetic resonance diffusion-weighted imaging showing areas of restricted diffusion in the (**A**) right frontal and (**B**) insular regions as well as the (**C**) left occipital region, indicating acute stroke.

members of the care team. Health care providers could not convince her that "Steve" was indeed her husband. The delusional symptoms persisted for 3 days but then gradually began to resolve. Ten days after the initial event, the patient seemed free of the delusion and was discharged to an inpatient rehabilitation facility.

DISCUSSION

Common to the various DMSs is a fixed false belief that a person, place, or object is someone or something other than what he, she, or it appears to be. One particularly fascinating and uncommon example of DMSs is the Capgras delusion. Originally described by Capgras and Reboul-Lachaux in 1923, the classic presentation of Capgras syndrome is characterized by the delusion that someone familiar to the patient has been replaced by an imposter who is physically identical to the known person. Our patient believed her husband's "doppelganger" (look-alike) to be a close friend, a relatively fortunate circumstance, as classically the perceived imposter in Capgras syndrome is ascribed a malevolent intent.

Multiple hypotheses have been put forth regarding the underlying pathophysiology of Capgras syndrome. Capgras (Capgras and Reboul-Lachaux, 1923) proposed that it resulted from lesions in areas of the brain responsible for familiarity, while others have posited dysfunction of the pathways involved in facial recognition (Young et al, 1991, 1993) or a disconnection between areas of face recognition in the temporal lobe and the limbic system (Hirstein and Ramachandran, 1997). In a comprehensive review of cases of DMS, investigators found that 57% of patients with delusions

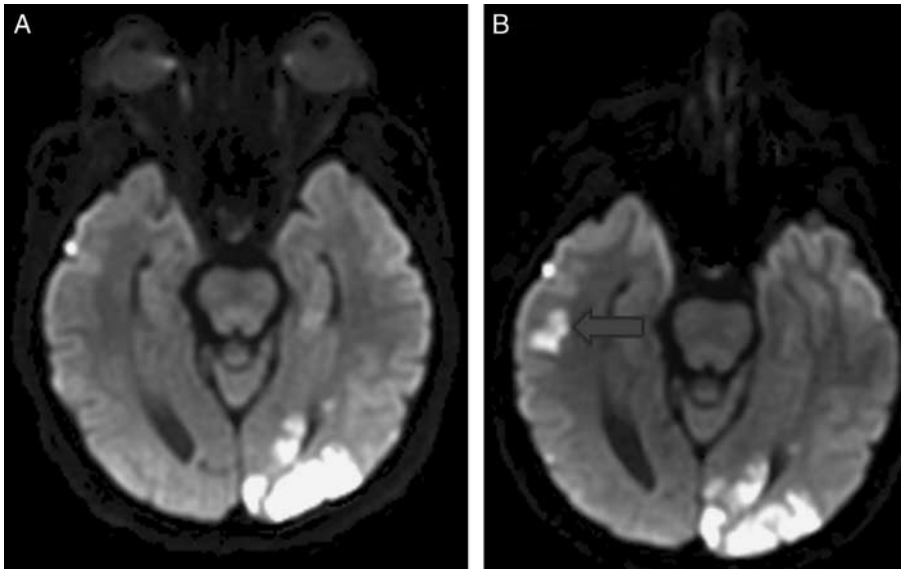

FIGURE 2. Axial magnetic resonance diffusion-weighted imaging showing a new area of restricted diffusion in the right temporal region (**B**, arrow) when compared with the initial study (**A**).

of hypofamiliarity had lesions in the right hemisphere, 11% had lesions in the left hemisphere, and 32% had bilateral hemispheric lesions (Darby and Prasad, 2016). The majority of lesions were in the frontal and parietal lobes, with the temporal and occipital lobes, basal ganglia, and thalamus being less commonly involved.

Several authors have sought to expand on the theory that DMSs result from lesions within the brain connectome. A study examining Capgras syndrome found evidence of impairment in the inferior fronto-occipital fasciculus, which connects core face-processing areas with frontal face-recognition areas responsible for emotional response to familiar faces (Bobes et al, 2016). Another connectivity study examining lesion-induced Capgras syndrome found defects in areas of the brain functionally connected to the left retrosplenial cortex, associated with familiarity, and the right frontal cortex, associated with expectation violation (Darby et al, 2017). These disconnection models help explain how single lesions can disrupt multiple functional regions, as posited by the two-factor model, in which a single factor triggers the delusion and an additional factor disrupts the processing of the delusion (Coltheart, 2010).

There is generally a delay in the presentation of Capgras delusion after a cerebral insult, although there are rare cases of immediate presentation (Darby and Prasad, 2016). The duration of delusions varies widely, with a median of 42 days before complete resolution of symptoms (Darby and Prasad, 2016).

It has been suggested that the typical delay in presentation and resolution is a result of maladaptive neuroplasticity (Darby and Prasad, 2016). The relatively close temporal association between the onset of the delusion and the discovery of a new right-sided temporal infarct, and the rapid resolution of symptoms in our case, support the notion that maladaptive plasticity does not necessarily occur in all cases of Capgras syndrome. Initial imaging studies of our patient revealed partial involvement of the right prefrontal cortex, Brodmann areas 44 and 45. Repeat magnetic resonance imaging of the brain several days after initial presentation revealed an infarct involving the right temporal region. Both right temporal region (Darby and Prasad, 2016) and right prefrontal cortex have been theorized to be associated with delusions. Lesion-induced Capgras syndrome can be time-delayed; therefore, in the case of our patient, it is difficult to ascertain whether the initial frontal or new temporal lesion was the main culprit.

Capgras syndrome has been observed in a number of medical conditions, including stroke, traumatic brain injury, tumor, neurodegenerative diseases such as Alzheimer and Parkinson disease, infection, vitamin B_{12} deficiency, hepatic encephalopathy, diabetes, and hypothyroidism. Resolution of symptoms of Capgras syndrome due to ischemic stroke varies. Approximately 50% of patients show recovery over a period of several days to weeks, 20% show some improvement, and 30% exhibit no change (Kaplan and Saddock, 2008). There are case reports of successful treatment with mirtazapine (Spiegel et al, 2008) as well as pimozide (Passer and Warnock, 1991). Overall, as reported in the current literature, Capgras symptomatology is typically persistent, resolving slowly over time, if at all. To our knowledge, spontaneous resolution of stroke-related Capgras syndrome over several days, as occurred in our patient, has rarely been reported.

Six months after initial presentation, the patient was contacted for follow-up. She reported that within 4 weeks of her discharge from the hospital, she started to recognize her husband and no longer had the delusion that he was an imposter. Resolution of symptoms, the majority of which was seen 10 days from initial presentation, was not complete, as she continued to have reservations. Other family members, however, reassured the patient of her husband's identity. The patient reported that resolution of symptoms was complete 7 weeks after her initial presentation.

Symptoms of Capgras syndrome rarely present in the acute period following ischemic stroke and are infrequently transient. This case report adds to the limited body of literature on transient Capgras syndrome. Clinicians should be aware of this possible behavioral complication of acute ischemic stroke.

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