

SINGLE CASE STUDY

Capgras Syndrome with Subarachnoid Hemorrhage

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Capgras misidentification syndrome after subarachnoid hemorrhage and the surgical repair of a right middle cerebral artery aneurysm is described. This Capgras delusional belief in an imposter assuming the form of a familiar person may result from disruption of cortical-visual-limbic pathways in the right hemisphere.

Capgras syndrome is the delusional belief in a double of one's self or other people. It is one form of misidentification syndrome in a spectrum of disorders that includes: autoscopy (hallucinatory vision of one's self; Maack and Mullen, 1983), reduplicative paramnesia (reduplication of a familiar place or person, Alexander *et al.*, 1979), and prosopagnosia (misrecognition of familiar faces; Berson, 1984). Putative organic and psychodynamic etiologies have been under critical scrutiny. Organic brain disease has been found in 26% to 42% of patients with Capgras syndrome (Merrin and Silberfarb, 1976).² Right hemispheric and frontal lesions account for most of the organic cases (Wilcox and Waziri, 1983). In contrast, psychodynamic etiologies are emphasized in a review of 133 published cases up to 1983 (Berson, 1983). The psychodynamics of misidentification in Capgras syndrome are described as a psychological means of resolving intense ambivalent feelings by the splitting of affect/identity.

The goal of this case review is to describe a patient with Capgras syndrome in whom a fixed belief of doubles followed an acute right hemisphere injury. This is of interest for three reasons. It shows that a circumscribed delusional belief that discriminates between individuals, without other psychopathology present, can be attributed to a specific neuroanatomical lesion. The neuroanatomical implications are of interest because this case imparts a clinical validity to the cortical-visual-limbic systems described by Bear (1983). Thirdly, the rarity of a circumscribed organic lesion producing Capgras syndrome deserves description in its own right, since the first organic case of Capgras syndrome due to an organic cause was described as recently as 1968 (Gluckman, 1968).

Case Report

Mrs. B. was a 66-year-old right-handed high school graduate. A widowed housewife, the mother of two daughters, one of whom (Mary) lived with her, she enjoyed good medical and psychiatric health in the past. While cleaning her car she felt a sudden headache and telephoned her daughter Mary to ask for help. Mary immediately went to her mother's house and found her mother holding her head and sweating, with a marked red facial flush. She was admitted to the hospital somnolent but arousable. There was a left sixth nerve palsy and bilateral upgoing toes. A computed tomography (CT) scan confirmed the diagnosis of subarachnoid hemorrhage and acute hydrocephalus secondary to subarachnoid and intraventricular hemorrhage. There were thick clots (>1 mm) in the basal frontal interhemispheric fissures and in the right ambient cistern. Diffuse blood was present in the stem of the Sylvian fissure and the Sylvian cisterns bilaterally as well as in the right suprasellar cistern (Figure 1). An anterior communicating artery aneurysm was suspected. However, carotid and vertebral angiography demonstrated a small right middle cerebral artery aneurysm (Figure 2). On the third hospital day, the patient became somnolent and disoriented. A CT scan demonstrated progressive hydrocephalus, and bifrontal ventriculostomies were performed. She improved and became alert, oriented, and conversant. A left hemiparesis developed. This was attributed to vasospasm and improved with treatment with volume and blood pressure elevation. The ventriculostomies were removed 7 days after insertion. Six days later she became more lethargic. The CT scan showed hydrocephalus. A repeat three vessel arteriogram showed only the right middle cerebral artery aneurysm. On the 17th day after her hemorrhage, she underwent a right ventriculovenous shunt and simultaneous clipping of the right middle cerebral artery aneurysm via a pterional approach opening the Sylvian fissure. The systolic blood pressure was kept above 95 mm Hg throughout the procedure. The aneurysm was fully

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² Spier, S. Everything you wanted to know about Capgras syndrome but were too oriented to ask. Psychosomatic Conference, Massachusetts General Hospital, October 14, 1983.

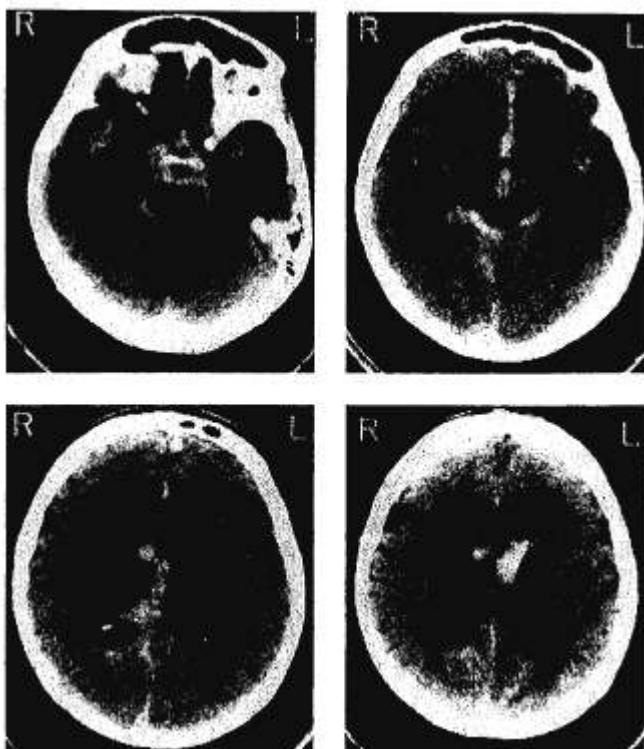


FIG. 1. This CT scan was performed on the day of hemorrhage and demonstrates blood in the basal cisterns, interhemispheric fissure, Sylvian fissures, ambient cisterns, and ventricles.

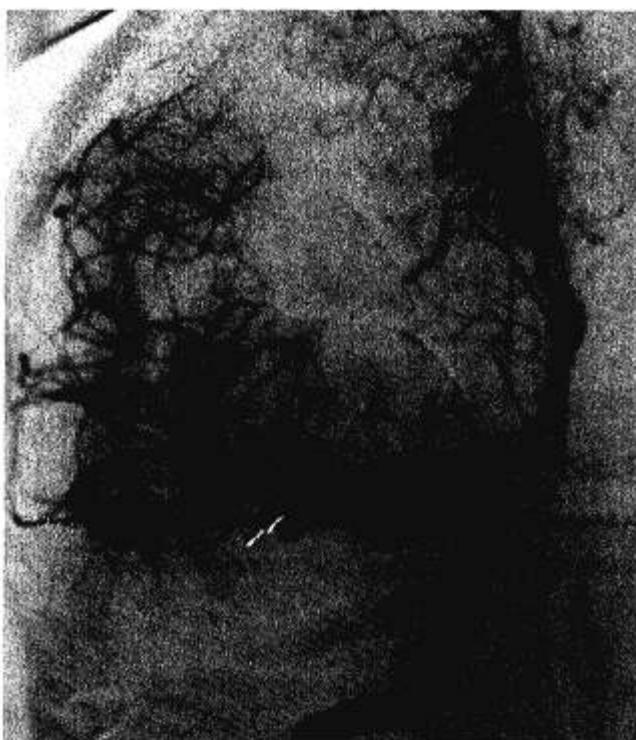


FIG. 2. This right internal carotid arteriogram demonstrates a small middle cerebral artery aneurysm (arrow).

mobilized and all vessels were completely visualized through the operating microscope. A straight Yasargil clip was used to obliterate the aneurysm and tissue adhesive was applied to a weak area of the aneurysmal neck. No vessels were compromised. The ventricular shunt was easily placed into the enlarged ventricles with one insertion. A Hakim valve with a closing pressure of 91 mm Hg was used and the distal catheter was radiologically confirmed to be in the atrium. Post-operatively she became increasingly alert and her left-sided weakness resolved. One observer noted abulia and a positive grasp reflex bilaterally. Physical and mental progress were excellent. She was oriented and conversant and had no focal deficits. A CT scan before discharge showed some persistent ventriculomegaly despite reduction in ventricular size compared with the preoperative CT scan. She was discharged home expecting a full recovery.

Her daughter Mary had taken several months' leave from work to care for her mother, who initially needed help with such tasks as dressing. A few days after discharge Mrs. B. stated that a younger girl had come into the house to masquerade as her daughter Mary. She believed this girl stayed for dinner and was then sent away. This confusion of identities became more persistent. She said, "I know there is a younger girl in the house who looks just like Mary, my younger daughter, but isn't. I can tell that they are different by the sound of their voice even though they look almost the same." At one moment she would speak to the real Mary and tell her about her new sister who looked almost the same as her, and at another moment would actually believe that the real Mary was not who she looked like, but the imposter. When questioned she would try to separate the two Marys by the tone of their voices, their height, or the color of their hair. But none of these differences was identified as stable trait differences. The imposter was always seen within the body of Mary, never as a separate visual hallucination. This confusion of identities remained limited to the daughter with whom she lived. She maintained good reality contact with her own identity as well as that of other people.

The first psychiatric consultation occurred 4 months after her surgery when her daughter was troubled by these strange beliefs and noted her mother's anxiety and tears when she repeatedly told her that there was one Mary, not two. The mental status examination showed a well-dressed, alert, cooperative woman with a smiling but constricted affect. Her belief in the presence of a double for her daughter was unshakable. She could not identify any specific differences between the double and the real daughter. There were no other indices of psychosis, endogenous anxiety, or depression. There was no emotional lability,

inappropriate social behavior, or psychomotor abnormality. There were no localizing neurological signs or frontal release signs. The EEG showed frequent runs of right temporal high-voltage delta and theta slowing.

Neuropsychological evaluation showed there was significant impairment of frontal lobe functions, namely, difficulty establishing, maintaining, and switching conceptual sets, perseveration, and decreased verbal fluency. Ability to plan and reason out the logical significance of details was below expectation for her age.

In view of her misidentification symptoms it is of particular interest that she drew inappropriate conclusions to account for the details she found confusing. Mrs. B. was unable to keep information in the correct temporal sequence. Although she could remember certain facts, she remembered them out of context. She took one design and incorporated it in her recollection of a previous design. Thinking was very concrete. However, constructional abilities were relatively well retained. There were less pronounced impairments in memory for verbal and visuospatial material. Word finding, spelling, reading, and comprehension were mildly impaired.

To summarize, the significant findings on her mental status were concreteness, inflexibility in thinking, perseveration, and impairment of her conceptual thinking. The end result was difficulty in organizing the available information in a meaningful emotional and conceptual manner.

Over the next year, evaluations showed gradual improvement in the patient's delusional thinking. She became less anxious and less preoccupied with her belief in an imposter. Thereafter, she ceased talking about the imposter, and gradually came back to identifying her daughter Mary as a single unique person. The last examination was 15 months after her original surgery. At this time there were no spontaneous complaints expressed by the patient or her daughter. However, when Mrs. B. was questioned she still held the belief that there was an imposter and still tried to identify subtle differences between her real daughter and the imposter. She was not preoccupied with this belief and did not bother to argue with others about it or become anxious. The belief was not preoccupying, but was still there. The rest of her mental status remained normal. She had no gait disorder nor urinary symptoms to suggest normal-pressure hydrocephalus. Her neurological examination was normal. A CT scan 1 year after surgery demonstrated some persistent ventricular dilation and asymmetry, with the right frontal horn being larger than the left, suggesting possible loss of tissue in the right frontal lobe (Figure 3). Neither this nor prior CT scans demonstrated a cortical area of stroke or cortical loss.

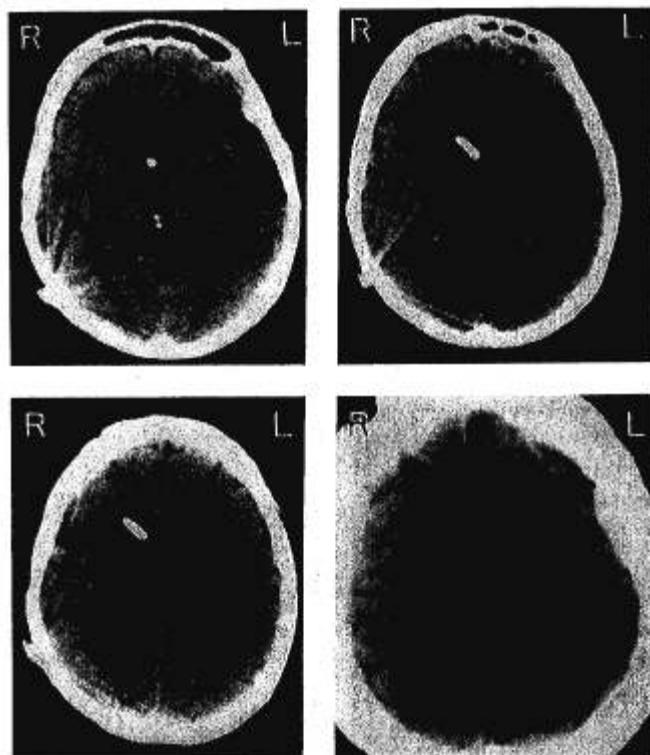


FIG. 3. This CT scan 1 year after surgery shows a right frontal shunt catheter and mild persistent ventriculomegaly with some periventricular low density. No cortical evidence for stroke is present.

Discussion

Mrs. B.'s belief in an imposter masquerading as her daughter conforms to the strict definition of Capgras syndrome as being a fixed delusional belief. There is no misperception or hallucination of another body, ruling out autoscopy, reduplicative paramnesia, and prosopagnosia. There were no parietal signs. Although insight is lost regarding the identity of her daughter, her sensorium was alert and oriented, excluding a generalized state of delirium. The absence of any past psychiatric history excludes chronic psychosis or personality disorder. The presence of a visual hallucination was raised by the patient's claim that there were some varying perceptual differences (e.g., height, color of hair, voice) between her daughter and the imposter and by her occasional separation of the real daughter from the imposter, as described in the history. At those times the real Mary was identified as herself and the imposter as another separate person. This phenomenon plus the vague perceptual differences in the two Marys raises the question of hallucination rather than delusion. Even so, the primary consistent abnormality of her mental status was an intangible belief in a double daughter, that is, a delusional thought rather than a visual hallucination.

Misidentification syndromes have been recognized

since the 1800s as possible sequelae to temporal lobe epilepsy, confusion, severe stress, anoxia, narcolepsy, parietal lesions, ecstasy, and hypnagogic twilight states (Critchley, 1979). Organic causes for Capgras misidentification syndrome have been increasingly identified in up to 42% of cases.³ Nondominant hemisphere lesions, particularly the right frontoparietal area, have been the most common organic findings. The Capgras syndrome itself has been seen in both left and right temporoparietal lesions begging the question of any specific anatomical site being unique to the syndrome³ (Levine and Grek, 1984).

The first study of the association between a focal brain lesion and delusional thinking was reported by Levine and Grek (1984). They found that delusion after right hemispheric stroke depends on an interaction between a focal lesion and a more diffuse brain atrophy. They concluded that the superimposition of a new right hemispheric lesion on a diffusely atrophied brain seems to cause delusions that can be expressed verbally because the left hemisphere is relatively preserved. This case supports Levine and Grek's contention. The strengths of the case are that there is no past psychiatric history and that we did not have to rely solely on radiographical and psychological inference as to the localization of the lesion.

Why might an individual with a right hemispheric lesion develop an illusion of doubles to selected people? The connection between misidentification of selected familiar people and right hemispheric lesions lies with the neuroanatomical pathways connecting the frontal cortex, limbic system, temporoparietal lobes and sensory cortex (visual in this case). The temporoparietal cortex receives projections from all the sensory association areas and projects widely to frontal cortex and limbic structures. There are two main cortical visuolimbic systems—dorsal and ventral (Bear, 1983). The dorsal (parietofrontal) system connects the dorsolateral frontal cortex with the cingulate gyrus, the parietal lobule, and the visual cortex. It is responsible for the surveillance of the environment and arousal to affectively relevant stimuli. Abulia results from a lesion to this system. The ventral (temporofrontal) system connects the orbital frontal cortex with the amygdala, hippocampus, and visual cortex. It is responsible for sequential temporal relationships and storage of visual emotional associations. This system is responsible for weighing alternative drive responses. A lesion in this system may result in a reflexive release of emotion to environmental stimuli without consideration of learned consequences. In Mrs. B.'s case, the lesion may involve the dorsal

system, as suggested by the constriction of affect. Clinically, the ventral system may also be impaired in that the patient does suffer from abnormalities in ordering temporal relationships and storage of visual emotional associations. This is suggested by Bear's model of the neurology of emotion (Bear, 1983).

These clinical findings and neuroanatomical pathways lead us to propose that Capgras syndrome may result from disruption of the sensory-temporoparietal limbic system. The patient's inability to maintain attention and to select the affectively important incoming stimuli results in a subcortical confusion. There is limbic anxiety, poor memory, and affective rigidity. The resultant feeling is one of "sentiments d'étrangete" as described by Capgras in 1923.

The question arises concerning the ill understood awareness of changes in thinking and feelings results in the attribution of confusion about the identity of a particular person. The hypothesis is that the patient experienced a sense of loss of something very important to her, in this case her intact cognitive functioning. She felt badly that she had lost something extremely valuable. Loss of clear thinking is a major stress to an elderly person. She identified the loss as something she knew was important to her, namely her daughter. This daughter was chosen because, in the context of the forced regression of the illness, there was ambivalence toward the person on whom she was forced to be dependent. In other words, the loss was displaced. This mechanism is different from dissociation, projection, splitting, paranoia, and autism as seen in typical functional psychosis.

Conclusion

This case report demonstrates that the specific delusional belief of Capgras may result from right frontal hemispheric pathology in the context of more diffuse cognitive impairment without having to invoke any functional psychological cause. However, the psychological mechanism of displacement is operative and explains why familiar individuals become the focus of confusion. A person with a cerebral lesion can discriminate among individuals. Disorders of belief (delusion) are not the exclusive realm of functional psychosis. Organic brain disorders are subject to the same heterogeneity of expression as functional psychotic syndromes. Capgras syndrome may be the clinical manifestation of disruption of cortical-visuo-limbic brain pathways. The evaluation of the patient with misidentification syndrome should include a thorough neurological, neuropsychological, and psychiatric evaluation.

³Joseph, A. B., and Kucharski, A.: Neuropsychiatric evaluation of 25 patients with misidentification. Unpublished manuscript.

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