Reprinted from

JOURNAL OF THE HILLSIDE HOSPITAL

Volume IV July, 1955 Number 3

DELUSIONAL REDUPLICATION OF PARTS OF THE BODY AFTER INSULIN COMA THERAPY[1]

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In recent years studies of behavioral changes occurring in altered states of brain function have gone beyond the description and interpretation of isolated phenomena. Emphasis has been placed on such factors as the particular environmental situation in which the behavior occurs, the total constellation or syndrome of associated behavioral changes, and the influence of the premorbid personality. Using these additional methods of study it has been demonstrated that many types of behavior found in brain disease are not defects, such as impairment of perception or memory, but represent forms of adaptation to the stress of illness (16). It has also been shown that various phenomena are not bizarre curiosities or unrelated fragments of neurological dysfunction, but form part of an orderly and meaningful pattern of an altered interaction with the environment.

This report of a single case is presented because of the unusual opportunity it provides to study some of these behavioral changes. First, the patient was in a psychiatric hospital, under observation and in psychotherapy for four months, which made it possible to have an accurate and comprehensive picture of his behavior prior to brain damage. Secondly, the neurological symptoms were of rapid

[1] From the Research Service, Hillside Hospital, Glen Oaks, New York. This investigation was supported in part by grant M-927 from the National Institute of Mental Health of the National Institutes of Health, Public Health Service and by a fellowship of the Dazian Foundation for Medical Research (Dr. Kahn). This paper was read, in part, at a meeting of the New York Society for Clinical Psychiatry at Hillside Hospital, on March 10, 1955.

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onset and actually developed in the presence of the examiners. Finally, it was possible to observe the patient intensively for a prolonged period afterwards, so that the subsequent changes in behavior could be adquately studied.

It is the purpose of this report to evaluate (1) the significance of the alterations in behavior, particularly the delusion of having extra parts of the body, and (2) the implication of the subsequent change in behavior for the understanding of the mechanism of somatic therapies.

Case History

Present Illness

The patient, a 34-year-old man, became acutely ill the night of September 25, 1954. Standing with clenched fists, gritting his teeth and without saying a word, he kept his wife in a corner of their bedroom for hours. The patient's family summoned a psychiatrist who referred him for immediate hospitalization. He received 15 electroshock treatments in a three-week period with some improvement. On transfer to Hillside Hospital, however, the patient was lethargic and failed to answer many questions. He said that he had come to the hospital for such reasons as "stomach disease," "to talk over something with my wife," and "to prepare myself for an examination." He felt he was being watched; that he was influenced by voices coming through the heating system; was being poisoned from a distance; and that there were changes in his body. He said he knew the exact minute when his wife was being unfaithful to him and expressed feelings that the world was coming to an end. There were frequent auditory hallucinations of being called unpleasant and derogatory names.

Past History and Premorbid Personality

This was the patient's first recorded psychotic episode. He is the youngest of five siblings, being the only boy. His father and mother were continually busy running a candy stand, and the patient was cared for primarily by his sisters. As a child he was dependent, demanding, and sought to be the center of attention. He developed a flair for comedy and playing the clown to the extent that he was expected to have a career as a comedian, and on one occasion won second prize in an amateur show. At 12 years of age he became interested in playing drums. This became his sole preoccupation, for he devoted every spare moment to them. The patient suffered episodes of rheumatic fever at 10 years of age and again at 20 and,

at these times, was pampered by his family. During his late adolescence he was withdrawn, spending much of his time at his parents' candy stand. He became overly concerned with his appearance and masculinity, brushed his hair for lengthy periods, exercised to acquire a good build, worried about his chest expansion and, according to his family, stared at his reflection in a mirror "kind of waiting for hair to grow on his chest." He spent much time exercising his left arm because he wanted it to be as strong as his right. He was concerned about a tooth that had not developed fully on the right side of his mouth. To prevent people from noticing this he developed the habit of talking and laughing out of the left side of his mouth, giving the appearance of facial asymmetry.

He never enjoyed or settled down to any work. Besides his efforts as a comedian and drummer, he worked in a pocketbook factory, in a ladies apparel concern, and wrote songs and short stories. He was discharged from one position because he clowned all day long amusing his co-workers. He became a beautician after a friend interested him in it. He wanted to quit this too, but remained at the insistence of his fiancée. He was not ambitious for money, but very much wanted to find a place for himself, needing reassurance about his sense of belonging. Although he comes from a secretive family where each member keeps things to himself, the patient was described as warm and friendly and drawn to the cause of people he considered the underdog or discriminated against. He was sentimental, being quite upset when having to visit someone in a hospital. He was overly sensitive and easily hurt, though not argumentative.

The family reported a change in his behavior during the past six years. He had become more secretive about his friends and activities. He went with his fiancée for more than a year before anyone in the family knew about it. He developed many somatic complaints so that frequent visits were made to his physician, without his family's awareness. Similarly, he told no one that he was in psychiatric treatment, and maintained his secret even though he found it necessary to steal money from his mother's store to pay for this. He also kept secret his difficulty in hearing for many months.

During the war he was drafted into service, became worried about having to go overseas, and sought and received a medical discharge because of rheumatic heart disease. Since 1948 he has been in intermittent psychiatric treatment. This was begun at the persuasion of a friend who was in therapy. The psychiatrist stated that the main effect of the therapy was to get him to look for some

kind of work, although he confirms the patient's inability to hold one job for any length of time. During this period two of his sisters have been under extensive psychiatric treatment for severe phobic reactions.

He has been married for 2½ years, never having dated much previously. His mother believes this the result of his being "pretty much of a home boy" and his fears about his rheumatic heart. His sister, on the other hand, reports that the mother made him feel guilty about leaving her alone in the store. She said he felt the responsibility of helping his mother ever since his father's illness and death, and that he expressed feelings of guilt in leaving her to get married.

The patient met his wife in 1951 at a party, and was married in 1953. During the first two years of their marriage, the patient's wife had two miscarriages. The second one in particular affected the patient. Soon thereafter he complained that there was something wrong with him, that he was not enough of a man, and that there must be a sickness in his body causing his wife to have abortions. He became increasingly depressed, withdrawn and fearful. Later, he became convinced his wife was unfaithful to him and that his wife and brother-in-law were conspiring against him.

Course in Hillside Hospital

During his hospitalization he became more withdrawn, and careless in his appearance and in the care of his room. He had little contact with other patients, and was preoccupied with his delusional thoughts and hallucinations.

Physical examination revealed a presystolic and systolic murmur at the apex and a systolic murmur at the base without accentuation of the pulmonary sounds, or signs of enlargement of the heart. His blood serology was negative. There were no neurological findings except for right facial asymmetry and bilaterally diminished hearing. An electroencephalogram shortly after admission showed well-modulated, occasional random 5-7 cps activity, with 90% alpha. It was interpreted as showing minimal abnormality, consistent with drowsiness or a history of recent electroshock therapy.

An amytal test for brain disease (13) was done on November 24, 1954. There was no change in orientation or awareness of illness, but he became more communicative and showed less overt tension during the procedure. Insulin coma treatment was instituted on December 8, 1954, and he had his first coma on December 30. During the course of 18 coma treatments there was no significant change in

behavior until the morning of January 27, 1955. On that day he had his 19th treatment, was given 370 units of insulin, and went into coma for an hour and 50 minutes, comparable to his previous reactions. Following gavage he did not respond in the usual time, and he was given glucose intravenously. He awakened promptly, and was responsive, but a marked right hemiplegia was noted.

He lay in bed with his head and eyes deviated to the left. There was no evidence of aphasia when tested for naming objects. He had a right facial paresis and a right homonymous hemianopia on confrontation. Reflexes were diminished on the right with a positive Babinski, and there was a right hemisensory syndrome with extinction on simultaneous stimulation tests.

He raised his left arm on command but failed to respond when told to move his right arm. When his right arm was raised by the examiner and he was asked to identify it, he looked at it for some moments and said it was a "stranger" and "an intruder." He reported smilingly that there was an extra arm on the right. He was unable to move his right leg on command, and he asked if it were his own. When the examiner walked away from the bed the patient called after him, saying he had left his (the examiner's) arm in the bed and should come and get it. He was ludic (15), euphoric, and loquacious. The patient was oriented for place and recognized the doctors and attendants. The only manifestation of disorientation at this time was a temporal reduplication for one of the examiners, saying that he had seen him sometime in the past.

The patient consistently showed this phenomenon of the reduplicated arm for the next hour. He referred to it as an "extra arm," "a third arm," and "a bootleg arm," or personified it as a "stranger," "this intruder" and "that fellow." When his right arm was shown to him, he denied knowing whose arm it was, asking one of the attendants, "Did you slip me this arm—did you pick this up at an old auction in the neighborhood?" Another time when asked to whom it belonged, he said "*I'm* willing to pay a reward for it, and *you're* asking me point blank." He said he was sure that the arm was not his because "it doesn't extend from my body" and "the dirt under the fingernail is not recognizable." The patient denied any disability of his own right arm, but said he'd never seen the extra arm work. He said, "My arm I can move with a brain impulse; this one I have to move manually since it isn't mine." During the course of questions about weakness in his right arm he said, "If that extra arm belongs to me, then I'm sicker than I thought I was."

The patient did not react to painful stimulation applied to his

right arm or leg. Even with his eyes open and his attention directed to the point of stimulation, he denied perceiving any stimulation on the right arm, saying "You're not fooling me—you're not touching me—you're touching this third arm, that intruder." He correctly identified stimuli applied to the right shoulder, but from the elbow down the touch was displaced to the extra arm. The patient lay with his head and eyes deviated to the left throughout the examination. He had difficulty perceiving any stimulus in the right side of space, a phenomenon which has been termed "spatial inattention." When given phrases to read he ignored the right side, reading only the material on the extreme left. Thus, "GOOD HUMOR ICE CREAM" was read as "GL." When his right arm lay at his side he had trouble finding it. Once, when he was asked to show it to the examiner, he looked over his left side only and said, "I think I've been robbed—where is it?"

A lumbar puncture was done and a clear, colorless fluid obtained. The fluid was under increased pressure even though the patient was relaxed. The pressure was recorded as 300 mm., the total protein was 32 mg. per cent, and there were two white cells per cubic mm.

During the ensuing hours the patient continued to be euphoric and loquacious. He recited long-forgotten lessons and parts of neurological texts whose source was unknown to him. For instance, he gave a complete description of the course of the facial nerve, and described the muscles of the face, calling them by their correct Latin names. He laughed frequently, and recited cryptic remarks as "in the instrument—insulin—instrument—insulin—instrument ward." Disturbances in memory or recall were not elicited. The delusional "extra arm" disappeared.

An electroencephalogram obtained that afternoon was flat in all leads on the left side and showed random 5-7 cps activity, chiefly on the right side. Both alpha and beta were prominent on the right side only. The record was interpreted as showing diffuse dysfunction with left-sided accentuation.

In the afternoon the patient was subdued. The weakness of the leg and arm showed some resolution. His relationship to his therapist was completely changed compared to his previous behavior. There was a complete absence of anger, negativism, withdrawal and depression. He clung to his doctor, shook hands, held him back and did not want to be separated from him. He was pleasantly preoccupied with the morning's episode and joked about it. He was eager to communicate, and even his hearing seemed to have improved.

The next morning the patient was depressed, restless, bewildered,

slow in answering questions and failed to respond when asked about his illness. He spoke in a low voice, at times inaudible. He was disoriented for time of day and was aware of "numbness" in his right arm. He spontaneously asked, "What happened to me—why am I taking all this depletion?" Minimal weakness of the right arm and leg were noted. There was astereognosis in the right hand, but tactile stimuli were correctly localized, and there was no evidence of hemianopia or inattention. When asked about the extra arm the patient was vague and evasive, but did say, "Evidently somebody else was with me and it was their arm I picked up."

That afternoon the patient was given 0.5 gm. amytal sodium. Besides its use as a test for organic brain dysfunction, the drug was given in an attempt to elicit the delusion again (14). At this time there was neither a change in orientation nor a recurrence of the reduplication. There was, however, a marked change in mood and language. The patient became very euphoric and talkative. His speech was characterized by ornate, circumstantial, pedantic, histrionic, and cryptic features, with much use of clichés. For example, asked about the extra arm, he said, "I think it will come looking for me when and if the occasion is propitious, as it were." When asked why he was here, he said, "On the recommendation of the right honorable Dr. Fink, most distinguished doctor on the eastern seaboard Atlantic area whose fame has spread far and wide." Several times he blurted out the cryptic remark—"transference of aggression." When asked what he meant, he said, "If you can't kick your mother-in-law in the head, you try your father-in-law."

When asked about his arm the patient was evasive, circumstantial and jocular. For example, asked how the extra arm was different from his own arm, he said, "How was it different? For one reason, in the sensitivity of feel. I raised up my left arm and that was all right. When I went to raise up what I thought was my right arm that was all right. But when I went to raise this third arm I did not feel any sensitivity when raising it, lifting it, touching it or otherwise in no manner could I relate it to my corpus—." When asked about weakness in his right arm, he said, "It feels a little less dynamic in its volition, and I'm tempted to believe in its delivery, as it were."

While the delusion was not present at this time, the patient insisted that there had been an extra arm the day before, saying, "I was lying in bed and it came to my aware the presence of another arm in my bed." When he was told that the extra arm was really his own right arm, he said, "Well, I'll tell you. I never argue with

facts. You see if you're surmising that it was, and I were to agree, it would be only for professional courtesy's sake." Or at another time, when the possibility of the extra arm was being questioned by the examiner, the patient said, "I don't think it was mine. It might have been mine, you see, but then I would have to have a comprehensive knowledge of the numerous preponderous volumes of ancient history in associated situations. And then I might be even able to volunteer that extra leg which you spoke of before—and beyond. I might even—be able to extend some photographs of the uterus which I own. If I could have the extra arm, the additional leg, and as I said, other things."

For the next two days he continued to be depressed, spoke slowly in a low voice, and showed no spontaneity. He complained of feeling "depleted." There was no difficulty getting him into a conversation and he would elaborate in a circumstantial way about the pain in his head and the numbness in his hand. He refused to get into conversation about his extra arm, saying, "You're making fun of me." Neurological examination was completely negative.

An electroencephalogram on February 2, 1955 showed a resolution of the asymmetry and abnormality of the previous record. It was similar to that obtained on admission. The patient was given amytal again on February 2 and 9. On both occasions he showed a similar response to that obtained on January 28, with euphoria and changes in language. His attitude, however, toward the extra arm and to the weakness of his right arm was altered. He now said that the extra arm might have been a hallucination due to the drugs he was receiving. He also admitted having had weakness of his right arm, saying, "To the best of my recollection there was a general weakness which might have had a specific attenuating dilemma in the appearance of an arm which might have been, to some degree, in a state of difficulty." On March 2, he was given amytal again. This time his reaction was more like that seen on admission, although he became slightly euphoric and loquacious toward the end.

In the weeks following the eventful insulin coma, there was a change in his clinical behavior. He appeared more sure of himself, and was co-operative and friendly. He started to press for his discharge. He said that there were things to be done which he, and not somebody else, should do, but would not specify these things. His wife visited him and told him she had decided to divorce him and would not accept him back in his home. The patient took this announcement without overt emotion. He was unable to give any reason for his wife's plans, and stated that he forgot to ask her why.

He still wanted to be sent home as soon as possible because, he said, he was ready to take up some kind of business. He felt that he had failed up to now because of reasons unclear to him, but that if he were careful, it would not happen to him again.

This behavior was markedly different from that shown on admission. In contrast to his previous aggressive, uncommunicative and withdrawn behavior, he was co-operative, ingratiating, overanxious to please, and made attempts at socializing with other patients. His hallucinations and delusions were absent. His improvement was maintained, and at the end of March he was discharged to the care of a psychiatrist.

Discussion

The delusion of reduplication of parts of the body has rarely been reported. The earliest reports are by Bechterev (1) in 1926 and Ehrenwald (3) in 1930. In 1935, Schenderov and Gamaleja (9) described six cases and suggested that more might be found if the phenomenon were better known. In these early studies the phenomenon was regarded as a neurologic curiosity and was explained on the basis of sensory disturbances. In the work of Critchley (2), who reported a similar case in 1952, it was considered as a manifestation of disturbance in "body image" due to a parietal lobe lesion of the nondominant hemisphere.

In 1954, four cases were reported by Weinstein et al. (14) demonstrating that the phenomenon could *not* be explained on the basis of sensory impairment, and was not dependent on a focal parietal lesion. They interpreted the delusion as a symbolic phenomenon rather than a sensory or perceptual disturbance, and showed that it occurred only in a setting of diffuse cerebral dysfunction. They indicated that parts of the body were reduplicated which were defective in some way, regardless of whether or not there was any neurological involvement.

It was pointed out that the delusional reduplication of body parts is but one manifestation of reduplicative phenomena. Thus reduplication for time, place and person has also been reported (11). In reduplication for time the patient confabulates that a present experience has also been experienced at a time in the past. For example, a patient identifies members of the staff as old friends or relatives. Reduplication for place is the confabulation that two or more places with the same name and similar attributes exist, when actually there is only one. Thus, a patient may say there are two

hospitals with the same name and same staff, but locate one closer to his home and describes it as a hospital which treats convalescent or minor cases only. In reduplication for person the patient confabulates the existence of two persons when there is actually only one. One woman, for instance, said she had two sons, one named "Bill," and the other "Willie," when actually she had one son named William.

It was shown by Weinstein et al. (14) that reduplication was usually expressed in more than one modality. All patients with delusional reduplication of body parts showed, in addition, reduplication for time, place and/or person. In the present case the patient also expressed temporal reduplication, describing one of the examiners as someone he had known prior to his hospitalization.

The symbolic importance of the various phenomena of reduplication is evident in their motivational character. In the perception of a doctor as an old friend or a relative the patient is reassuring himself that he has less to fear than he would from a total stranger. In reduplication for place the patient who locates the extra hospital near his home or describes it as treating only convalescent or minor cases is minimizing his illness. The patient who confabulated having two sons, while denying her own illness, complained that poor "Willie" was in an accident and was afraid that something terrible had happened to him, thus displacing her concern from herself to the extra person.

The delusion of reduplication of parts of the body also is a mechanism facilitating denial of illness. While the patient states that there is nothing wrong with his own body, it is the reduplicated arm or leg which is said to be weak or impaired. In the present instance the patient, who in his premorbid behavior was excessively worried about bodily ailments, was unconcerned about his severe disability. Instead, by denying having any trouble with his arm and saying it was the "extra" arm that didn't work, he was able to maintain his euphoria and jocularity.

While reduplication is shown mainly as a symbolic adaptation to the problem of illness, it may also be a symbolic expression of other wishes, needs and feelings. For example, a patient with intractable pain had been noisy and demanding and had aroused the antagonism of members of the staff. Following a course of electroshock therapy her complaints of pain were gone and her relations with the staff were considerably improved. Along with other changes in orientation, she confabulated that there were two Mount

Sinai Hospitals, the old one where people were mean to her, and the new one where everybody was so nice (12).

The delusional reduplication of parts of the body appears to be related to certain aspects of the premorbid personality. In the present case, and in those reported by Weinstein et al. (14), all the patients demonstrated a special concern with the symbolic importance of physical characteristics. Our patient was concerned with his build, the strength of the muscles of his left arm and the presence of hair on his chest. He was also sensitive about a tooth which had not come out fully and attempted to conceal it. In the earlier study (14) a patient who had developed the delusion of multiple heads following a craniotomy, had been preoccupied with his baldness and had engaged in numerous extramarital affairs to prove that he was capable of attracting women. A patient, who confabulated having three eyes, was very sensitive about a prosthetic eye and would face people directly forward so that divergence of his eye would not be noticed. He was also preoccupied with his build and physical appearance, engaged intensively in Yogi and Judo, and was concerned with his ability to satisfy his wife sexually.

In recent writings on "spatial inattention" (16, 17) it has been indicated that this, too, represents a form of symbolic adaptation. In this case the patient consistently avoided looking to the right side, was unable to find the "extra" arm when it lay at his side, and only read the extreme left part of phrases shown to him. This behavior facilitated the denial of weakness of his right arm as well as helped maintain his delusion of the extra extremity. His personality also showed features that have been described as characteristic of persons with "spatial inattention" (17). He was very secretive, especially in recent years, and seemed overly concerned with the symbolic significance of violence, as shown by his discomfort on visiting hospitals, his fear of going overseas and by an apparent confabulation that he had once been a secret witness to a murder.

The marked ludic behavior shown by the patient during the period of his delusion and during the later amytal tests is also related to his premorbid personality. He was described as having been a good mimic with a flair for clowning. Ludic behavior has also been regarded as a form of adaptive behavior (15) in which the patient acts out a feeling of well-being, implicitly denying his illness.

It is of theoretical interest that the delusional reduplication and "spatial inattention" occurred with right-sided symptoms. Disorders of the "body image" and spatial awareness have been regarded as characteristic of nondominant lesions. In this case, with apparent

dominant hemisphere involvement but without aphasia, it was possible to study these behavioral changes as clearly as with nondominant lesions. One must conclude that in the usual case of a dominant lesion, pathology which is sufficiently extensive for these changes to occur, will also result in an aphasic disorder which masks the phenomena.

The neurologic lesion which was the basis for the reported phenomena and alteration in behavior was a diffuse dysfunction of the cerebrum with accentuation of the left hemisphere. Such disturbances in neurologic function are not uncommon in insulin coma therapy, and are the result of persistent cellular dysfunction despite restoration of the glucose level of the blood. While one may ascribe etiological importance in the production of this patient's hemiplegia to the history of rheumatic fever and the cardiac murmurs, this is not warranted considering the absence of other rheumatic phenomena either before or subsequent to the event. It is more meaningful to regard this incident as one manifestation of the central nervous system damage incurred by prolonged hypoglycemia. Such manifestations include all aspects of nervous system functions including seizures, transient hemiplegia and aphasia, confusional syndrome and Korsakoff psychoses, prolonged coma and death (6). Recent studies of the electroencephalographic changes during prolonged coma (18) and minor neurologic signs following insulin coma treatment (19) amplify the variety of neurologic sequelae of this treatment.

The importance of cerebral damage in the mechanism of somatic therapies has recently been re-emphasized by Weinstein and Kahn (12, 16). They suggest that improvement following somatic therapies is characterized by manifestations of denial in a milieu of altered brain function. A study in this laboratory (5) has supported this hypothesis concerning electroshock therapy. Improved patients showed significantly earlier and more persistent alterations in brain function as measured by the amytal test (13) and by serial electroencephalographic studies (4), and more changes in language and behavior indicative of denial, than did patients who failed to improve.

Recent case reports of the effects of prolonged insulin coma by Revitch (8), Kwalwasser and Caplan (7), Shagass and Rowsell (10), and Yeager et al. (18) point to the behavioral improvement and disappearance of schizophrenic symptoms when cerebral damage supervenes. In this patient, too, there was an immediate, marked and persistent change in behavior concomitant with cerebral damage.

It was possible here to demonstrate not only explicit verbal denial of illness, but other aspects of denial as reduplication, spatial inattention and changes in mood and language. Thus, the observations in his case are consistent with the hypothesis of Weinstein and Kahn. Further studies of the role of premorbid personality in denial, and on the mechanism of somatic therapies are now in progress.

Summary and Conclusion

1. A case history is presented of a 34-year-old man with a four-month history of acute mental illness who was placed on insulin coma therapy. After his 19th coma he developed a right hemiplegia, hemianopia, hemisensory syndrome and "spatial inattention," and became ludic, euphoric and loquacious. At this time he showed delusional reduplication of body parts, expressing the conviction that while there was nothing the matter with his right arm, there was an "extra" arm in his bed which did not belong to him and which did not work.

2. The significance of reduplication is discussed in terms of a symbolic adaptation to illness. This phenomenon, as well as his other changes in behavior such as "spatial inattention" and ludic behavior, is considered to be related to his premorbid personality.

3. The presence of these phenomena in a case with right-sided symptoms is considered with reference to anatomical localization. These findings contradict the traditional view that "body image" disorders and "spatial inattention" depend on a nondominant hemisphere lesion.

4. The subsequent changes in the patient's behavior, in which he showed complete recovery from his illness, is discussed in terms of its implications for the mechanism of action of insulin coma therapy. The results are considered to support the theoretical view that improvement in the somatic therapies is characterized by manifestations of denial in a milieu of altered brain function.

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May 23, 1955

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This investigation was supported in part by grant M-927 from the National Institute of Mental Health of the National Institutes of Health, Public Health Service and by a fellowship of the Dazian Foundation for Medical Research (Dr. Kahn).

This paper was read, in part, at a meeting of the New York Society of Clinical Psychiatry on March 10, 1955.

Submitted Hillside Journal May 24, 1955

In recent years studies of behavioral changes occurring in altered states of brain function have gone beyond the description and interpretation of isolated phenomena. Emphasis has been placed on such factors as the particular environmental situation in which the behavior occurs, the total constellation or syndrome of associated behavioral changes, and the influence of the premorbid personality. Using these additional methods of study it has been demonstrated that many types of behavior found in brain disease are not defects, such as impairment of perception or memory, but represent forms of adaptation to the stress of illness (16). It has also been shown that various phenomena are not bizarre curiosities or unrelated fragments of neurological dysfunction, but form part of an orderly and meaningful pattern of an altered interaction with the environment.

This report of a single case is presented because of the unusual opportunity it provides to study some of these behavioral changes. First, the patient was in a psychiatric hospital, under observation and in psychotherapy for four months, which made it possible to have an accurate and comprehensive picture of his behavior prior to brain damage. Secondly, the neurological symptoms were of rapid onset and actually developed in the presence of the examiners. Finally, it was possible to observe the patient intensively for a prolonged period afterwards so that the subsequent changes in behavior could be adequately studied.

It is the purpose of this report to evaluate 1) the significance of the alterations in behavior, particularly the delusion of having extra parts of the body and 2) the implication of the subsequent change in behavior for the understanding of the mechanism of somatic therapies.

CASE HISTORY

Present Illness

The patient, a 34-year-old man, became acutely ill the night of September 25, 1954. Standing with clenched fists, gritting his teeth and without saying a word, he kept his wife in a corner of their bedroom for hours. The patient's family summoned a psychiatrist who referred him for immediate hospitalization. He received 15 electroshock treatments in a three week period with some improvement. On transfer to Hillside Hospital, however, the patient was lethargic and failed to answer many questions. He said that he had come to the hospital for such reasons as "stomach disease," "to talk over something with my wife," and "to prepare myself for an examination." He felt he was being watched; that he was influenced by voices coming through the heating system; was being poisoned from a distance; and that there were changes in his body. He said he knew the exact minute when his wife was being unfaithful to him and expressed feelings that the world was coming to an end. There were frequent auditory hallucinations of being called unpleasant and derogatory names.

Past History and Premorbid Personality

This was the patient's first recorded psychotic episode. He is the youngest of five siblings, being the only boy. The father and mother were continually busy running a candy stand and the patient was cared for primarily by his sisters. As a child he was dependent, demanding, and sought to be the center of attention. He developed a flair for comedy and playing the clown to the extent that he was expected to have a career as a comedian, and on one occasion won second prize in an amateur show. At 12 years of age he became interested in playing drums. This became his sole preoccupation, for he devoted every spare

moment to them. The patient suffered episodes of rheumatic fever at 10 years of age and again at 20, and, at these times, was pampered by his family. During his late adolescence he was withdrawn, spending much of his time at his parents' candy stand. He became overly concerned with his appearance and masculinity, brushed his hair for lengthy periods, exercised to acquire a good build, worried about his chest expansion and, according to his family, stared at his reflection in a mirror "kind of waiting for hair to grow on his chest." He spent much time exercising his left arm because he wanted it to be as strong as his right. He was concerned about a tooth that had not developed fully on the right side of his mouth. To prevent people from noticing this he developed the habit of talking and laughing out of the left side of his mouth, giving the appearance of facial asymmetry.

He never enjoyed or settled down to any work. Besides his efforts as a comedian and drummer, he worked in a pocketbook factory, in a ladies apparel concern, and wrote songs and short stories. He was discharged from one position because he clowned all day long amusing his co-workers. He became a beautician after a friend interested him in it. He wanted to quit this too, but remained at the insistence of his fiancee. He was not ambitious for money, but very much wanted to find a place for himself, needing reassurance about his sense of belonging. Although he comes from a secretive family where each member keeps things to himself, the patient was described as warm and friendly and drawn to the cause of people he considered the underdog or discriminated against. He was sentimental, being quite upset when having to visit someone in a hospital. He was overly sensitive and easily hurt, though not argumentative.

The family reports a change in his behavior during the past six years. He had become more secretive about his friends and activities. He went with his fiancee for more than a year before anyone in the family knew about it. He developed many somatic complaints so that frequent visits were made to his physician, without his family's awareness. Similarly, he told no one that he was in psychiatric treatment, and maintained his secret even though he found it necessary to steal money from his mother's store to pay for this. He also kept secret his difficulty in hearing for many months.

During the war he was drafted into service, became worried about having to go overseas, and sought and received a medical discharge because of rheumatic heart disease. Since 1948, he has been in intermittent psychiatric treatment. This was begun at the persuasion of a friend who was in therapy. The psychiatrist stated that the main effect of the therapy was to get him to look for some kind of work, although he confirms the patient's inability to hold one job for any length of time. During this period two of his sisters have been under extensive psychiatric treatment for severe phobic reactions.

He has been married for 2½ years, never having dated much previously. His mother believes this the result of his being "pretty much of a home boy" and his fears about his rheumatic heart. His sister, on the other hand, reports that the mother made him feel guilty about leaving her alone in the store. She said he felt the responsibility of helping his mother ever since his father's illness and death, and that he expressed feelings of guilt in leaving her to get married.

The patient met his wife in 1951 at a party, and was married in 1953. During the first two years of their marriage, the patient's wife had two miscarriages. The second one in particular affected the patient. Soon thereafter

he complained that there was something wrong with him, that he was not enough of a man, and that there must be a sickness in his body causing his wife to have abortions. He became increasingly depressed, withdrawn and fearful. Later, he became convinced his wife was unfaithful to him and that his wife and brother-in-law were conspiring against him.

4. Course in Hillside Hospital

During his hospitalization he became more withdrawn, and careless in his appearance and in the care of his room. He had little contact with other patients, was preoccupied with his delusional thoughts and hallucinations.

Physical examination revealed a presystolic and systolic murmur at the apex and a systolic murmur at the base without accentuation of the pulmonary sounds, nor signs of enlargement of the heart. His blood serology was negative. There were no neurological findings except for right facial asymmetry and bilaterally diminished hearing. An electroencephalogram shortly after admission showed well modulated, occasional random 5-7 cps activity, with 90% alpha. It was interpreted as showing minimal abnormality, consistent with drowsiness or a history of recent electroshock therapy.

An amytal test for brain disease (13) was done on December 24, 1954. There was no change in orientation or awareness of illness, but he became more communicative and showed less overt tension during the procedure. Insulin coma treatment was instituted on December 8, 1954, and he had his first coma on December 30. During the course of 18 coma treatments there was no significant change in behavior until the morning of January 27, 1955. On that day he had his 19th treatment, was given 370 units of insulin, and went into coma for an hour and 50 minutes, comparable to his previous reactions. Following gavage he did not respond in the usual time, and he was given glucose intravenously. He awakened promptly, and was responsive, but a marked right hemiplegia was noted.

He lay in bed with his head and eyes deviated to the left. There was no evidence of aphasia when tested for naming objects. He had a right facial paresis and a right homonymous hemianopia on confrontation. Reflexes were diminished on the right with a positive Babinski, and there was a right hemisensory syndrome with extinction on simultaneous stimulation tests.

He raised his left arm on command but failed to respond when told to move his right arm. When his right arm was raised by the examiner and he was asked to identify it, he looked at it for some moments and said it was a "stranger" and "an intruder." He reported smilingly that there was an extra arm on the right. He was unable to move his right leg on command, and he asked if it were his own. When the examiner walked away from the bed the patient called after him, saying he had left his (the examiner's) arm in the bed and should come and get it. He was ludic (~~31~~ 15), euphoric, and loquacious. The patient was oriented for place and recognized the doctors and attendants. The only manifestation of disorientation at this time was a temporal reduplication for one of the examiners, saying that he had seen him sometime in the past.

The patient consistently showed this phenomenon of the reduplicated arm for the next hour. He referred to it as an "extra arm," "a third arm," and "a bootleg arm," or personified it as a "stranger," "this intruder" and "that fellow." When his right arm was shown to him, he denied knowing whose arm it was, asking one of the attendants, "Did you slip me this arm - did you pick this up at an old auction in the neighborhood?" Another time when asked to whom it belonged, he said "I'm willing to pay a reward for it, and you're asking me point blank." He said he was sure that the arm was not his because "it doesn't extend from my body" and "the dirt under the fingernail is not recognizable." The patient denied any disability of his own right arm, but said he'd never seen the extra arm work. He said "my arm I can move with a brain impulse; this one I have to move manually since it isn't mine." During the course of questions about weakness in his right

arm he said, "If that extra arm belongs to me, then I'm sicker than I thought I was."

The patient did not react to painful stimulation applied to his right arm or leg. Even with his eyes open and his attention directed to the point of stimulation, he denied perceiving any stimulation on the right arm, saying "you're not fooling me - you're not touching me - you're touching this third arm, that intruder." He correctly identified stimuli applied to the right shoulder, but from the elbow down the touch was displaced to the extra arm. The patient lay with his head and eyes deviated to the left throughout the examination. He had difficulty perceiving any stimulus in the right side of space, a phenomenon which has been termed "spatial inattention." When given phrases to read he ignored the right side, reading only the material on the extreme left. Thus, "GOOD HUMOR ICE CREAM" was read as "OL". When his right arm lay at his side he had trouble finding it. Once, when he was asked to show it to the examiner, he looked over his left side only and said, "I think I've been robbed - where is it?"

A lumbar puncture was done and a clear, colorless fluid obtained. The fluid was under increased pressure even though the patient was relaxed. The pressure was recorded as 300 mm., the total protein was 32 mg.%, and there were two white cells per cubic mm..

During the ensuing hours the patient continued to be euphoric and loquacious. He recited long-forgotten lessons and parts of neurological texts whose source was unknown to him. For instance, he gave a complete description of the course of the facial nerve, and described the muscles of the face, calling them by their correct Latin names. He laughed frequently, and recited cryptic remarks as "in the instrument - insulin - instrument - insulin - instrument ward." Disturbances in memory or recall were not elicited. The delusional "extra arm" disappeared.

An electroencephalogram obtained that afternoon was flat in all leads on the left side and showed random 5-7 cps activity, chiefly on the right side. Both alpha and beta were prominent on the right side only. The record was interpreted as showing diffuse dysfunction with left-sided accentuation.

In the afternoon the patient was subdued. The weakness of the leg and arm showed some resolution. His relationship to his therapist was completely changed compared to his previous behavior. There was a complete absence of anger, negativism, withdrawal and depression. He clung to his doctor, shook hands, held him back and did not want to be separated from him. He was pleasantly preoccupied with the morning's episode and joked about it. He was eager to communicate and even his hearing seemed to have improved.

The next morning the patient was depressed, restless, bewildered, slow in answering questions and failed to respond when asked about his illness. He spoke in a low voice, at times inaudible. He was disoriented for time of day and was aware of "numbness" in his right arm. He spontaneously asked, "what happened to me - why am I taking all this depletion?" Minimal weakness of the right arm and leg were noted. There was astereognosis in the right hand but tactile stimuli were correctly localized and there was no evidence of hemianopia or inattention. When asked about the extra arm the patient was vague and evasive, but did say "Evidently somebody else was with me and it was their arm I picked up."

That afternoon the patient was given 0.5 gm amytal sodium. Besides its use as a test for organic brain dysfunction, the drug was given in an attempt to elicit the delusion again (14). At this time there was neither a change in orientation nor a recurrence of the reduplication. There was, however, a marked change in mood and language. The patient became very euphoric and talkative. His speech was characterized by ornate, circumstantial, pedantic, histrionic, and cryptic features, with much use of clichés. For example, asked about the extra arm, he said "I think it will come looking for me when and if the occasion is propitious, as it were." When asked why he was here, he said, "on the re-

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commendation of the right honorable Dr. Fink, most distinguished doctor on the eastern seaboard Atlantic area whose fame has spread far and wide." Several times he blurted out the cryptic remark - "transference of aggression." When asked what he meant, he said, "If you can't kick your mother-in-law in the head, you try your father-in-law."

When asked about his arm the patient was evasive, circumstantial and jocular. For example, asked how the extra arm was different from his own arm, he said "How was it different? For one reason, in the sensitivity of feel. I raised up my left arm and that was all right. When I went to raise up what I thought was my right arm that was all right. But when I went to raise this third arm I did not feel any sensitivity when raising it, lifting it, touching it or otherwise in no manner could I relate it to my corpus - -." When asked about weakness in his right arm, he said, "It feels a little less dynamic in its volition, and I'm tempted to believe in its delivery, as it were."

While the delusion was not present at this time, the patient insisted that there had been an extra arm the day before, saying, "I was lying in bed and it came to my aware the presence of another arm in my bed." When he was told that the extra arm was really his own right arm, he said "Well, I'll yell you. I never argue with facts. You see if you're surmising that it was, and I were to agree, it would be only for professional courtesy's sake." Or at another time, when the possibility of the extra arm was being questioned by the examiner, the patient said "I don't think it was mine. It might have been mine, you see, but then I would have to have a comprehensive knowledge of the numerous preponderous volumes of ancient history in associated situations. And than I might be ~~ever~~ even able to volunteer that extra leg which you spoke of before - and beyond. I might even --- be able to extend some photographs of the uterus which I own. If I could have the extra arm, the additional leg, and as I said, other things."

For the next two days he continued to be depressed, spoke slowly in a low voice, and showed no spontaneity. He complained of feeling "depleted." There was no difficulty getting him into a conversation and he would elaborate in a circumstantial way about the pain in his head and the numbness in his hand. He refused to get into conversation about his extra arm, saying, "you're making fun of me." Neurological examination was completely negative.

An electroencephalogram on February 2, 1955 showed a resolution of the asymmetry and abnormality of the previous record. It was similar to that obtained on admission. The patient was given amytal again on February 2nd and 9th. On both occasions he showed a similar response to that obtained on January 28th, with euphoria and changes in language. His attitude, however, toward the extra arm and to the weakness of his right arm was altered. He now said that the extra arm might have been an hallucination due to the drugs he was receiving. He also admitted having had weakness of his right arm, saying, "To the best of my recollection there was a general weakness which might have had a specific attenuating dilemma in the appearance of an arm which might have been, to some degree, in a state of difficulty." On March 2nd, he was given amytal again. This time his reaction was more like that seen on admission, although he became slightly euphoric and loquacious toward the end.

In the weeks following the eventful insulin coma, there was a change in his clinical behavior. He appeared more sure of himself, and was cooperative and friendly. He started to press for his discharge. He said that there were things to be done which he, and not somebody else, should do, but would not specify these things. His wife visited him and told him she had decided to divorce him and would not accept him back in his house. The patient took this announcement without overt emotion. He was unable to give any reason for his wife's plans, and stated that he forgot to ask her why. He still wanted to be sent home as soon as possible because , he said, he was ready to take up some

kind of business. He felt that he had failed up to now because of reasons unclear to him, but that if he were careful, it would not happen to him again.

This behavior was markedly different from that shown on admission. In contrast to his previous aggressive, uncommunicative and withdrawn behavior, he was cooperative, ingratiating, over-anxious to please, and made attempts at socializing with other patients. His hallucinations and delusions were absent. His improvement was maintained, and at the end of March he was discharged to the care of a psychiatrist.

DISCUSSION

The delusion of reduplication of parts of the body has rarely been reported. The earliest reports are by Bechterev (1) in 1926 and Ehrenwald (3) in 1930. In 1935, Schenderov and Camaleja (9) described six cases and suggested that more might be found if the phenomenon were better known. In these early studies the phenomenon was regarded as a neurologic curiosity and was explained on the basis of sensory disturbances. In the work of Critchley (2), who reported a similar case in 1952, it was considered as a manifestation of disturbance in "body image" due to a parietal lobe lesion of the non-dominant hemisphere.

In 1958, four cases were reported by Weinstein et.al. (14) demonstrating that the phenomenon could not be explained on the basis of sensory impairment, and was not dependent on a focal parietal lesion. They interpreted the delusion as a symbolic phenomenon rather than a sensory or perceptual disturbance, and showed that it occurred only in a setting of diffuse cerebral dysfunction. They indicated that parts of the body were reduplicated which were defective in some way, regardless of whether or not there was any neurological involvement.

It was pointed out that the delusional reduplication of body parts is but one manifestation of reduplicative phenomena. Thus reduplication for time, place and person has also been reported (11). In reduplication for time the patient confabulates that a present experience has also been experienced at a time in the past. For example, a patient identifies members of the staff as old friends or relatives. Reduplication for place is the confabulation that two or more places with the same name and similar attributes exist, when actually there is only one. Thus, a patient may say there are two hospitals with the same name and same staff, but locate one closer to his home and describes it as a hospital

which treats convalescent or minor cases only. In reduplication for person the patient confabulates the existence of two persons when there is actually only one. One woman, for instance, said she had two sons, one named "Bill," and the other "Willie," when actually she had one son named William.

It was shown by Weinstein et.al. (14) that reduplication was usually expressed in more than one modality. All patients with delusional reduplication of body parts showed, in addition, reduplication for time, place and/or person. In the present case the patient also expressed temporal reduplication, describing one of the examiners as someone he had known prior to his hospitalisation.

The symbolic importance of the various phenomena of reduplication is evident in their motivational character. In the perception of a doctor as an old friend or a relative the patient is reassuring himself that he has less to fear than he would from a total stranger. In reduplication for place the patient who locates the extra hospital near his home or describes it as treating only convalescent or minor cases is minimizing his illness. The patient who confabulated having two sons, while denying her own illness, complained that poor "Willie" was in an accident and was afraid that something terriable had happened to him, thus displacing her concern from herself to the extra person.

The delusion of reduplication of parts of the body also is a mechanism facilitating denial of illness. While the patient states that there is nothing wrong with his own body, it is the reduplicated arm or leg which is said to be weak or impaired. In the present instance the patient, who in his premorbid behavior was excessively worried about bodily ailments, was unconcerned about his severe disability. Instead, by denying having any trouble with his arm and saying it was the "extra" arm that didn't work, he was able to maintain his euphoria and jocularity.

While reduplication is shown mainly as a symbolic adaptation to the problem of illness, it may also be a symbolic expression of other wishes, needs and feelings. For example, a patient with intractable pain had been noisy and demanding and had aroused the antagonism of members of the staff. Following a course of electroshock therapy her complaints of pain were gone and her relations with the staff were considerably improved. Along with other changes in orientation, she confabulated that there were two Mount Sinai Hospitals, the old one where people were mean to her, and the new one where everybody was so nice (12).

The delusional reduplication of parts of the body, appears to be related to certain aspects of the premorbid personality. In the present case, and in those reported by Weinstein et.al. (14), all the patients demonstrated a special concern with the symbolic importance of physical characteristics. Our patient was concerned with his build, the strength of the muscles of his left arm and the presence of hair on his chest. He was also sensitive about a tooth which had not come out fully and attempted to conceal it. In the earlier study (14) a patient who had developed the delusion of multiple heads following a craniotomy, had been preoccupied with his baldness and had engaged in numerous extramarital affairs to prove that he was capable of attracting women. A patient, who confabulated having three eyes, was very sensitive about a prosthetic eye and would face people directly forward so that divergence of his eye would not be noticed. He was also preoccupied with his build and physical appearance, engaged intensively in Yogi and Judo, and was concerned with his ability to satisfy his wife sexually.

In recent writings on "spatial inattention" (16, 17) it has been indicated that this, too, represents a form of symbolic adaptation. In this case the patient consistently avoided looking to the right side, was unable to find the

" extra" arm when it lay at his side, and only read the extreme left part of phrases shown to him. This behavior facilitated the denial of weakness of his right arm as well as helped maintain his delusion of the extra extremity. His personality also showed features that have been described as characteristic of persons with "spatial inattention" (17). He was very secretive, especially in recent years, and seemed overly concerned with the symbolic significance of violence, as shown by his discomfort on visiting hospitals, his fear of going overseas and by an apparent confabulation that he had once been a secret witness to a murder.

The marked ludic behavior shown by the patient during the period of his delusion and during the later asytal tests is also related to his premorbid personality. He was described as having been a good mimic with a flair for clowning. Ludic behavior has also been regarded as a form of adaptive behavior (15) in which the patient acts out a feeling of well-being, implicity denying his illness.

It is of theoretical interest that the delusional reduplication and "spatial inattention" occurred with right-sided symptoms. Disorders of the "body image" and spatial awareness have been regarded as characteristic of non-dominant lesions. In this case, with apparent dominant hemisphere involvement but without aphasia, it was possible to study these behavioral changes as clearly as with non-dominant lesions. One must conclude that in the usual case of a dominant lesion, pathology which is sufficiently extensive for these changes to occur will also result in an aphasic disorder which masks the phenomena.

The neurologic lesion which was the basis for the reported phenomena and alteration in behavior was a diffuse dysfunction of the cerebrum with accentuation of the left hemisphere. Such disturbances in neurologic

function are not uncommon in insulin coma therapy, and are the result of persistent cellular dysfunction despite restoration of the glucose level of the blood. While one may ascribe etiological importance in the production of this patient's hemiplegia to the history of rheumatic fever and the cardiac murmurs, this is not warranted considering the absence of other rheumatic phenomena either before or subsequent to the event. It is more meaningful to ascribe this incident as one manifestation of the central nervous system damage incurred by prolonged hypoglycemia. Such manifestations include all aspects of nervous system functions including seizures, transient hemiplegia and aphasia, confusional syndrome and Korsakoff psychoses, prolonged coma and death (6). Recent studies of the electroencephalographic changes during prolonged coma (18) and minor neurologic signs following insulin coma treatment (19), amplify the variety of neurologic sequellae of this treatment.

The importance of cerebral damage in the mechanism of somatic therapies has recently been re-emphasized by Weinstein and Kahn (12,16). They suggest that improvement following somatic therapies is characterized by manifestations of denial in a milieu of altered brain function. A study in this laboratory (5) has supported this hypothesis concerning electroshock therapy. Improved patients showed significantly earlier and more persistent alterations in brain function as measured by the amytal test (13) and by serial electroencephalographic studies (4); and more changes in language and behavior indicative of denial, than did patients who failed to improve.

Recent case reports of the effects of prolonged insulin coma by Revitch (8), Kwalwasser and Caplan (7), Shagass and Rowsell (10), and Yeager et.al (18) point to the behavioral improvement and disappearance of schizophrenic symptoms when cerebral damage supervenes. In this patient, too, there was an immediate, marked and persistent change in behavior concomitant with cerebral

damage. It was possible here to demonstrate not only explicit verbal denial of illness, but other aspects of denial as reduplication, spatial inattention and changes in mood and language. Thus, the observations in his case are consistent with the hypothesis of Weinstein and Kahn. Further studies of the role of premorbid personality in denial, and on the mechanism of somatic therapies are now in progress.

SUMMARY AND CONCLUSION

1. A case history is presented of a 34-year-old man with a four month history of acute mental illness who was placed on insulin coma therapy. After his 19th coma he developed a right hemiplegia, hemianopia, hemisensory syndrome and "spatial inattention," and became ludic, euphoric and loquacious. At this time he showed delusional reduplication of body parts, expressing the conviction that while there was nothing the matter with his right arm, there was an "extra" arm in his bed which did not belong to him and which did not work.

2. The significance of reduplication is discussed in terms of a symbolic adaptation to illness. This phenomenon, as well as his other changes in behavior such as "spatial inattention" and ludic behavior, is considered to be related to his premorbid personality.

3. The presence of these phenomena in a case with right-sided symptoms is considered with reference to anatomical localization. These findings contradict the traditional view that "body image" disorders and "spatial inattention" depend on a non-dominant hemisphere lesion.

4. The subsequent changes in the patients behavior, in which he showed complete recovery from his illness, is discussed in terms of its implications for the mechanism of action of insulin coma therapy. The results are considered to support the theoretical view that improvement in the somatic therapies is characterized by manifestations of denial in a milieu of altered brain function.

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