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**THE USE OF MONOZYGOUS TWIN CASES IN THE STUDY
OF PSYCHOSOMATIC CONCEPTS†**

Workers seriously interested in genetic problems have long deplored emphasis in the clinical literature on case reports concerning disease occurring in both members of a pair of identical twins (concordant). The value of reporting cases of illness appearing in one member of a pair (discordant) is obvious, yet such reports appear only sporadically. Increased attention to the role of the family and society in the genesis of schizophrenia has given new prominence to the study of mental disease in twins. Case reports are now appearing with considerable detail. This, interestingly enough, has been paralleled in medical research by an increased awareness of the significance of twin disease, in part because of the results of organ transplants in identical twins. It is the purpose of this paper to discuss some of the problems in evaluating twin case reports and to emphasize the value of the study of twin cases in the examination of psychosomatic concepts.

The methodology of twin study deserves some comment. At first it might seem that the chief value of case reports of discordant disease in twins would be to lend support to hypotheses concerning the environmental (or at least the "other than genetic") factors in the genesis of disease. As will be shown later, this cannot be accomplished by finding one discordant case. The statistical techniques introduced by Kallman have limited usefulness in establishing the genetic background of disease, but cannot assess the importance of the assorted factors that make up "environment."

In controversies over such issues, the suggestion of study of identical twins reared apart from birth arises. Early studies were not rewarding from the viewpoint of modern personality theory; their chief value has been a negative one: to demonstrate that many human attributes are

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significantly less variable in identical twins. The case material of Newman⁶ and of Burks⁷ suffers from the fault that in most cases the early environmental differences of the separated twins were not great, and where personality differences did exist, the family situation, the characteristics of the parents, or the recall of early experiences, were not sufficiently detailed to allow interpretation. In addition, the tools of projective psychological testing were not used to full advantage.

Price⁸ has questioned whether the assumption that dissimilarity in monozygous twins is as important evidence for the primacy of environmental circumstances as it might first appear. For example, intrauterine disadvantages of circulation or position may be a prenatal cause of differences apparently developing in later life in monozygous twins. Therefore, he has suggested that only monozygous twins with equal (dichoroidal) placental circulations be studied, since in such patients circulatory changes would be less likely to produce differences which might be interpreted as the effect of post-natal environment. Price estimates that dichoroidal circulation is found in 35-40 per cent of all monozygous twins. Such a suggestion has obvious merit, but lack of information about the placenta would sharply limit the twin material available for study.

From a psychoanalytic standpoint, objections can be raised to most twin studies; unless one has an opportunity to examine the subtleties of the developmental years of early childhood in great detail, or to recover the framework of such development by means of therapeutic analysis, the significance of later physiological and psychological data is debatable.

If the limits suggested by Price were followed closely, very little twin case material would be available. It may be that such criticisms invalidate a great deal of work. In any case, monozygous twins reared apart from birth are rare, it is unusual to find them known to have been dichoroidal, and it is difficult to find co-operative pairs to work with, particularly in psychosomatic or psychiatric areas. Such an approach leads to progressively smaller amounts of material and our experience has been that co-operation of the twin pairs remaining available for intensive study depends upon the patients' getting something out of the relationship.

Three cases of comparatively well studied psychosomatic disorders, investigated by teams which included medical and psychological participants, will point up features of twin investigation which have been insufficiently exploited, and will call attention to problems of evaluating twin case reports. These studies have been careful examinations of discordant and dissimilar twins, even though the most ideal criteria for selection could not be met.

CASE I. HYPERTENSION

This set of twins was first studied by Flynn, Kennedy, and Wolf, and reported in 1948 as a case of essential hypertension occurring in one of identical twins.⁸ Their mother had been hypertensive for 30 years. Elsa, the twin later hypertensive, weighed 2½ pounds at birth, her sister Karen weighed 7 pounds. During infancy and childhood Elsa cried more and suffered a series of medical illnesses; at 10 months left lower lobe pneumonia and empyema required thoracotomy, and at 3 years laryngeal diphtheria required repeated intubations which were followed by chronic hoarseness. Elsa implicated this severe hoarseness as a source of her later social inhibition and recalls shyness and frequent anxiety from early childhood. Karen early became the leader and flourished in all respects but one: she frequently lost her temper. Although both were good students, she was always the better. At age 13 the girls separated; Elsa went to a trade school and became a seamstress, Karen went to a conventional high school and became a secretary. On physical examination on entering high school Elsa was found to have a systolic blood pressure of 180 mm. of mercury, but was asymptomatic. Four years later, at graduation from high school, Elsa developed transient symptoms suggestive of hyperthyroidism which were not supported by laboratory studies. Three years later, in 1948, her mother suffered hemiparesis and Elsa's recurrent headaches led to investigation at the New York Hospital, presented in detail in the early paper. Elsa's mother recovered and Elsa again became symptom-free.

At that time support for the role of anger in hypertension seemed demonstrated. The central hypothesis of the authors involved personality differences between hypertensive patients and normals: hypertensive patients were said to be nicer and more compliant but not as elastic and pliable as other people. Their main difficulties were supposed to center around the handling of rage. Hypertensives are chronically angry, unable to recognize their anger and become openly aggressive. A corollary of this hypothesis is that hypertension represents an exaggerated physiological concomitant of rage particularly in susceptible patients. A second corollary is that blood pressure increases in response to threatening situations especially in those that are likely to call forth angry feelings. Elsa and Karen appeared to fulfill the predictions of these hypotheses.

Their mother suffered a second attack of hemiparesis and coma in the spring of 1957, and it was then that Karen's hyperthyroidism was diagnosed. She was continually anxious and lost 20 pounds in weight; a PBI confirmed the diagnosis of hyperthyroidism with a level of 8.9 and she was started on Tapezole.

In September 1957, the twins were admitted to the New Haven Hospital. Karen was a pleasant, co-operative young woman. Her skin was warm and dry. There was a very fine tremor to the outstretched hand. She had a slight stare, but blinked with normal frequency. Extra-ocular movements were normal. There was no lid-lag. Lungs were clear. Her heart was small. P2 was louder than A2. There was a grade I pulmonic systolic murmur. No other murmurs were heard. The thyroid was enlarged about one and a half times normal size, a bit more on the right than on the left; no nodules were palpable. The remainder of the physical examination was within normal limits. X-ray of the chest and heart was unremarkable. A cold-pressor test showed a baseline blood pressure of 110/80; and following a two-minute immersion of her hand in ice water, her blood pressure rose only to 120/94, returning within two minutes to baseline levels of 112/86. Her blood pressure dropped off during sleep to a low at 4:00 a.m. of 90/60 which did not change significantly on her arising in the morning. Blood pepsin was 330 units; NPN was 22 mgm.%; a BEI was 4.2 gammas per cent.

Elsa was a reserved, but not embarrassed young woman. She spoke in a somewhat hoarse voice. There was a gross tremor to her outstretched hands. Extra-ocular movements were normal. A scar of a left pleural drainage was evident and there was diminution of breath sounds at the left base. Her thyroid was palpable bilaterally about one and a half times normal size. Her heart was enlarged to the sixth intercostal space, almost to the anterior axillary line. Rhythm was regular. P2 was louder than A2. There was a grade III presystolic apical murmur, a grade III rumbling diastolic murmur, and a grade III systolic murmur at the apex. At the base a grade III aortic diastolic blow, as well as a grade II aortic systolic murmur, were present. The remainder of the physical examination was within normal limits. A chest film showed left auricular enlargement with some right ventricular enlargement, prominent pulmonary vascular markings, and fluid in the left costophrenic angle. A cold-pressor test showed a baseline blood pressure of 108/80 with a rise to 146/98 after immersion of her hand in ice water for two minutes; her blood pressure returned to 106/80 two minutes after the hand was withdrawn from the water. During sleep her blood pressure fell to 130/70, but rose to 160/70 when she awakened. NPN was 28, blood pepsin was 320 units. A BEI was 5.0 gammas per cent.

Thus, it became evident that while Karen had a documented history of hyperthyroidism at this time at least, Elsa had normal thyroid function, despite the suspicion of hyperthyroidism in the past. Elsa, however, had now developed obvious rheumatic heart disease with mitral stenosis and physiologically significant aortic insufficiency. Some evidence that this might have been the case was present in the original case report where a "faint diastolic blow" was heard along the left sternal border; at the present time at least, it becomes apparent that hypertension in Elsa is much more likely to be the result of her aortic valvular lesion than it is of essential hypertension. However, her response to cold pressor and sleep tests are suggestive of a hyperactive cardiovascular response. The approach to the study was excellent and the case is historically important, yet it cannot be considered a "test case" of essential hypertension. It emphasizes the need for careful co-operation between the psychiatric and medical investigators.

Follow-up. It is interesting that the nonhypertensive, more independent twin has recently developed hyperthyroidism under circumstances that would support some of the specificity hypotheses associated with that illness. A few details will be presented. Elsa went to night school, became a secretary herself and took a job in the clothing industry, "by chance" a block from Karen. Neither has married. Karen continues to be the dominant twin, leading them into various "broadening" cultural pursuits.

Elsa was quick to agree to a follow-up study; Karen, however, didn't want any part of it at first, but was eventually won over and quickly became the active participant doing most of the talking, writing, and making decisions about arrangements. She had changed her mind because a few months before she had started to be sleepless, restless, and easily upset. This followed the second episode of coma and hemiparesis of the mother and at this time her hyperthyroidism was diagnosed. Subsequent

blood iodine studies have revealed both twins to be within normal limits while Karen was on medication.

An attractive question here involves the choice of illness. Was Karen's hyperthyroidism a reaction to the stress of a threat similar to that which had previously evoked hypertensive symptoms in Elsa? Such an hypothesis sheds no special light on stress specificity. However, the difference in character organization, with Elsa possessing hypertensive characteristics and Karen hyperthyroid ones, supports the observation that personality differences are intimately, if not casually, related to the choice of illness.

Some of the differences in the twins are obvious. Karen is more impulsive, more easily upset, more likely to get mad and "blow up." Elsa rarely experiences this kind of feeling and the family, chiefly Karen, almost automatically tends to impose on her under most circumstances.

Karen dominates Elsa to an extraordinary extent. Even in the testing and examining situation of the study, Karen not only took over the role of making all the arrangements, she also tried to dominate the study by being protective, motherly, more interested, more warm and engaging than Elsa. It was as if in the last ten years Karen had taken over the responsibility of what was going to happen to both of them. Elsa seems to have accepted this as her fate and to be resigned to it.

This speculation is supported by what seems to have happened at the time their mother had the second episode of hemiparesis. Elsa, who was concerned, but not severely upset, developed no symptoms. Ten years before, she had developed headaches when her mother was ill: then, she and her sister were still actively competing with each other. Since then, Elsa seems to have consolidated her passive role in relation to her sister and has changed her job and her way of life in order to become a closer companion to her sister. Karen, on the other hand, has also given up competing with Elsa, at the cost of assuming responsibility for her. Both girls still live at home and the ties with the parents are not yet broken, but it would seem that the loss of the mother now would be more of a blow to Karen than to Elsa.

Elsa's fantasy life appears to be richer and more troubled. Her ability to enjoy her inner experiences is relatively blocked by anxiety and by anxiety-induced restriction of functioning. Karen has a more controlled inner life and uses repression more successfully. On the surface, Karen appears to function more smoothly, independently, and vivaciously, her responses on the psychological tests have a wholesome juvenile quality, while Elsa's have the quality of an adult neurotic patient. The intellectual inhibition is striking. Elsa obtained a verbal I.Q. of 113 compared to 120

for Karen, a performance I.Q. of 121 compared to 133, and a total I.Q. of 117 compared to 128.

Karen seems to fill the clinical descriptions of hyperthyroid patients. Such patients have lively, active, and genuinely appealing qualities. Early achievement of apparent maturity, self-sufficiency, and protectiveness of others is prominent. A strong sense of responsibility founded as a defense against inner dependency and fear of death is also typical.^{4,5}

CASE II. DISSIMILAR ASTHMA IN MONOZYGOUS TWINS

Crede reported asthma of markedly differing severity in identical 26-year-old men.² There were evident personality differences, which included separate ways of regarding the parents. The better adjusted twin, "healthier" except for his asthma, a kind, passive, pleasant person, was preferred by the parents and seemed to be making a continuous effort to please them. His asthma was much worse than that of his brother and was improved only during separation from the family while he was in the army. He was less able to be emotional, to cry or to complain, or to vent hostility than his brother. B had these characteristics to such a degree that people actually disliked him. As a consequence of this, B's adjustment in all spheres was considerably poorer than A's, but he had minimal asthma.

Independent psychological testing supported the clinical appraisal in this pair. A follow-up recently obtained from Dr. Crede reveals that twin A, after psychotherapy, now also has minimal asthmatic difficulty, but that B probably has had a brief psychotic episode and continues to have considerable social difficulty.

The special importance of this case, well recognized by the original investigators is the necessity of fitting case material into an holistic frame; that is, one should not be deceived into seeing differences between twins simply as a matter of totally "better" or "worse" background situations.

CASE III. DUODENAL ULCER IN ONE OF IDENTICAL TWINS

A case of peptic ulcer in one of identical twins, has been published;³ here the physiological parameters such as blood pepsin and gastric intubation studies were the same, but differences in character structure and the degree of external distress were quite evident. The subjects are strikingly similar in background and appearance. They make the same initial impression; passive, shy, dependent, anxious, semi-skilled workers. The ulcer twin began to have symptoms during a near-psychotic breakdown in his wife when she was having an affair with another man and threatened to kill their children. The patient's difficulty with his wife, though somewhat diminished, has persisted since the onset of illness and so have the ulcer symptoms. In contrast, his brother is married to a motherly woman who dependably manages family affairs. Both patients have high blood pepsin levels, but show on interview and psychological testing, modest but real differences in their responses to stressful circumstances. Although superficially similar, the ulcer twin is more shut off, resentful, bitter, and less capable of having satisfying relationships. Blind analysis of projective test results highlighted certain personality differences and correctly identified the ulcer twin.

This case study supports the hypothesis that the level of blood pepsin is genetically determined and that the occurrence of ulcer depends upon the presence of gastric hyper-

secretion, plus a special combination of circumstances. We found support for Alexander's conception of a type of passive-dependent personality who develops peptic ulcer in the presence of circumstances which threaten the nurturance of these needs.

DISCUSSION

Of considerable value from the dynamic point of view is the study by Kent of 19 pairs of twins seen in a child guidance agency.⁷ Her findings suggest that the superficial environment of the family may appear to be remarkably similar for each one of the twin pair: there is no separation of the twins, the family is intact, the twins are physically identical, go to the same school, wear the same clothes, and yet these are coarse similarities. Individual twins may live in a markedly different intra-familial environment; the different concept of the parents about each twin (for example, identification of separate twins with other family members by the parents), and the division of roles between the twins, especially as regards dominance, competitiveness, and leadership, form two obvious bases for such differences.

The study of illness in monozygous twins offers more than information about the genetic determinants of disease. The shaping of development, normal and pathological, is illuminated by such studies. For the development of any character trait, symptom, or illness, many factors are usually involved; there are now hundreds of physiological and psychological studies related to developmental shifts and changes. Careful study of single cases or small groups of cases with physiological and psychological measurements can be used to test hypotheses concerning various factors in illness.

Certain pertinent issues deserve further comment. The problem of susceptibility to illness and the identity of the physiological response in twins are relatively unexplored areas. Physiological studies have shown close similarities within twin pairs, with respect to onset of puberty and level of blood pressure, gastric secretion, blood pepsin, electrocardiogram, and electroencephalogram, and point in the direction of identical patterns. In general, basic physiological processes are more narrowly defined in monozygous twins. The problems of susceptibility to illness based on integrated psychosomatic concepts is much more difficult to answer as it depends in part on the development of differences including personality de-differentiation. While there is a natural tendency for monozygous twins to develop along similar lines, a tendency reinforced in part by identification with each other and in part by a lack of distinction between the twins by the parents, other factors tend to result in personality differences.

Through careful accumulation of discordant and dissimilar twin cases, it may be possible to check the relevance of infantile development, character structure, and environmental stress in the development of illness. "Psychosomatic" illness offers an especially intriguing laboratory for such investigation because of the availability of measurable physiological data.

The three cases presented do not by themselves solve the central problem of character predisposition vs. "stress" as the source of triggering mechanisms in disease. Knowledge that an illness has genetic determinants does not reduce the importance of looking for other factors that may play a role. If an intimate relation between a general line of development and an illness exists in one of identical twins, it must still be noted that the original source difference can be intrauterine, but not genetic.

Still, even this criticism of discordant cases may not be well founded; if it can be established that the twins studied are not greatly different from other twin pairs except in the one pattern of development under scrutiny, if this pattern can be shown to be crucial for the symptom, character trait, or illness which develops, such a study of twin cases will continue to be of value in determining the genesis of disease.

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