

REPORT ON A PAIR OF MALE MONOZYGOTIC TWINS CONCORDANT FOR SCHIZOPHRENIA

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SUMMARY

A pair of MZ twins with diagnoses of chronic simple schizophrenia has been studied from the genetic, social, medical, and psychiatric standpoint by members of the several professions. Clinical psychologists studied the patients independently and on the basis of psychometric and projective tests found a remarkable similarity in personality profiles. An in-depth family study failed to reveal any evidence of psychiatric disease in other relatives. Consanguinity was absent. The family unit is typical of that described frequently for a schizophrenogenic household, i.e., dominant mother, passive father. Genetic etiologies are discussed in context with occurrence of schizophrenia in MZ twins in this particular family setting.

A schizophrenic disorder is liable to develop in a genetically-predisposed individual. Whether or not the disorder actually develops, or the age at which it develops, as well as the symptomatology and severity of the disorder, are determined by the individual constitutional interactions with factors such as physical and psychological stresses, the family milieu, the social environment, and the cultural milieu.

Twin studies in schizophrenia, in addition to their limited usefulness for genetic analysis, provide a means for evaluating environmental influences by controlling for the genetic variables. Among pairs of MZ twins who are discordant for schizophrenia, the nonschizophrenic twin usually has been the leader of the two, the heavier one at birth, the first-born, the more energetic, and the more self-assertive (Pollin et al. 1966, Stabenau and Pollin 1967, Pollin 1972, Stierlin 1972).

The present paper describes a family study in which a pair of 35-year-old MZ twins, concordant for schizophrenia, were subjected to medical and psychiatric evaluations for a period of more than seventeen years. In addition, psychometric tests were performed independently with each of the cotwins.

We offer this report as a contribution to the literature in developmental psychiatry. We believe that the accumulation of additional data on environmental variables associated with development of characteristic schizophrenic categories, particularly when genetic controls indicate the unlikelihood that sporadic phenocopies are involved, will provide basic clues toward nosological and etiological clarifications.

MATERIAL AND METHODS

The twin subjects of this report have been evaluated in a psychiatric hospital by a psychiatrist, two clinical psychologists, a psychiatric nurse, and an internist-geneticist. The psychological tests include the following: Wechsler-Bellevue Intelligence Scale, Thematic Apperception Test, Bender-Gestalt Test, and Draw a Person Test. Intensive interviews were completed with the two patients, their mother, and their father. Pertinent medical records were obtained from attending physicians and prior to hospitalizations. Blood group antigens, including both major and minor blood groupings, were evaluated. In addition, over fifty photographs of these children from age 3 months to early adulthood were critically evaluated in order to provide additional documentation of zygosity.

CASE REPORTS

PATIENT A

Patient A is a 35-year-old white male who, with his identical twin brother, was referred for in-patient psychiatric evaluation and treatment by their parents, because "both boys stay in their room and communicate only with each other." Each of the twins was severely withdrawn; they both refused to communicate with nurses or physicians beyond mere grunts of "yes" or "no" and avoided eye contact. In each case, the affect was consistently flat and the mood was depressed. Each one denied hallucinatory or delusional activities, and neither of them mentioned any problems. They both seemed to demonstrate complete lack of insight.

Physical examination revealed a 35-year-old white male with an asthenic, ectomorphic habitus, who was completely uncommunicative. Blood pressure was 140/80; the remainder of the vital signs were normal. Examination indicated that the heart, lungs, abdomen, genitalia, neurological system, and integument, were all within normal limits.

Psychological testing revealed the following: Wechsler Adult Intelligence Scale revealed verbal I.Q. of 91, Performance I.Q. of 97, with a Full-scale I.Q. of 93; the Bender-Gestalt Test of Visual Motor Ability showed a nonorganic profile and dynamically suggested a withdrawn person with a poverty of thought who maintains control of strict super-ego defenses. On personality evaluation, the House-Tree-Person Test showed a low energy level and a socially inaccessible person with greater than average fantasy. There were marked feelings of isolation associated with a need for maternal protection. There was repeated evidence of aggressive and suspicious feelings. The Thematic Apperception Test again showed poverty of thought with mere simple description. There was avoidance of any reference of personal initiative or motivation. The Rotter Incomplete Sentence Blank showed statements which were again evasive and denying, with avoidance of any emotional expression or interpersonal contact, and were noteworthy in their lack of affect or feeling.

In summary, the diagnostic impression was that of a severely withdrawn person who was incapable of adequate functioning; diagnosis was "chronic simple schizophrenia".

PATIENT B

This 35-year-old male could be described as virtually identical to his twin brother with respect to habitus. His vital signs were normal. The physical findings were completely normal. His affect was essentially the same as that of his brother, already described.

Psychological testing, performed independently by another clinical psychologist, showed the following findings. The patient was willing to cooperate with the examiner though he avoided eye contact and answered only when addressed. He displayed an excessive poverty of ideas, passivity, and concrete thinking. His judgment and insight appeared to be extremely limited. The Wechsler Adult Intelligence Scale showed a Verbal I.Q. of 93, a Performance I.Q. of 96, and a Full-scale I.Q. of 94; the Bender-Gestalt Test of Visual Motor Ability was normal and did not indicate any evidence of organicity. Dynamically, he displayed repeated and obsessive workover, which is a psychotic indicator denoting tension and considerable reality loss. The House-Tree-Person Test finding showed reality loss with some sexual preoccupation and anxiety with feelings of hostility and withdrawal into fantasy. There were paranoid features and findings suggesting a desire to deny and withdraw from unpleasant situations. Figures showed the mother to be a dominant and an aggressive force in the family. In summary, the protocols indicated many passive-aggressive tendencies, feelings of inferiority, and an avoidance of conflict by withdrawal into fantasy. Rotter responses were brief and descriptive and the patient declined to answer most of the statements. The most remarkable findings were denial, poverty of ideas, and a dependent desire to return home; the Thematic Apperception Test exhibited similar findings. The patient tended to be mainly descriptive and refused to involve himself in the relationships pictured. He stated, "That's their story, not mine." There was some evidence of a passive type of hostility such as found when he described the angry feelings of the boy who didn't want to play the violin but gave in and submitted to authority. He tended to picture the mother-figure as compulsively clean and dominant over the family. In summary, the patient presented as an individual who is currently functioning in the average range of intelligence but who emotionally appeared very flat and blunted with extreme poverty of ideas, passivity, and a willingness to withdraw into fantasy. He was shown to possess many covert hostile feelings which he was unable to express outwardly in any acceptable manner. The diagnostic impression was "chronic simple schizophrenia".

Laboratory work on both of the twins can best be summarized collectively since the values were virtually identical. Routine laboratory work, including complete blood counts and urinalyses, were normal; serum cholesterol was 168 mg% for twin A and 171 mg% for twin B; the PBI was 7.1 mcg% for twin A and 7.4 mcg% for twin B; both twins were blood group ORh+, and five minor blood groupings as well as the MN were all identical for each twin.

The cotwins show a remarkable similarity in their physical examinations, psychological testing, laboratory studies, past medical histories, and current modes of adjustment.

INTERVIEW WITH PARENTS

An interview was conducted simultaneously with both of the patients' parents. The mother was the dominant individual in the household, and she appeared to be a martyr type who seemed to take pride in the fact that her sons were always obedient and relished the attention and protection she gave them. She stated that her youngsters had never exhibited any aggressive tendencies with the single exception being immediately prior to their admission to the psychiatric unit when one of her sons told her to "shut up". This behavior was "shocking" and atypical since the rule was that her children had never questioned any type of authority.

The father is a very passive individual who consistently looked toward his wife for ap-

proval and for "permission" to make any statement referable to questions directed toward him by the interviewers. He constantly made self-deprecatory references which included "I am completely to blame for the way my children are today... had I spanked them once in a while, they might have been different... I was overprotective... and if I had it to do over again, I'd treat them differently... I never laid a hand on them in all their lives... I am a total failure..." His wife corroborated this account and then began reinforcing this by stating that she also should assume some of the blame because she believed she was equally overprotective and then repeated essentially the same accounting as given by her husband.

The husband's guilt feelings were clearly evident as he hung his head as if in shame every time his wife made a comment relevant to the early life history of their children and their failure to progress emotionally and psychologically.

The parents agreed unanimously that the twins had shown extreme similarity in their behavioral responses since infancy. They preferred each other's company to that of their childhood and later adult peers. There were occasional birthday parties where acquaintances were invited; but these seemed to be rare occurrences and were performed in a perfunctory manner, usually in response to the wishes of their mother. They both graduated from high school and attended a business college for two years. They were both obsessed with their school work, though their grades were about average. They exhibited some degree of artistic ability, though there is some question as to whether there might have been any commercial potential in their artistic ability. The twins shared the same room since childhood. They had always been "close" but during their teens became even closer to each other. There was never any evidence of sibling rivalry, and the only account of any "disagreements" between them concerned very minimal and limited exchanges of ideas and differences in their reactions to rather mundane issues.

Upon the completion of business school, both twins sought employment in the capacity of clerks doing general office work. Significantly, however, in the midst of economic and employment prosperity during the mid and late 1950's, they were both unsuccessful in acquiring jobs. They both "gave up" and have remained sheltered and protected in their home since that time, presumably with the complete blessing and consent from both of the parents.

They were first judged to have a "psychiatric problem" at age 17, though a definitive diagnosis was not entered on their medical records. They spent the past 15 years living in the same room in their parents' home in almost complete lack of communication with individuals other than each other or their parents. There are no other children in the family. Neither twin has ever dated a girl, and their social histories have been severely impoverished. They have not attended any "outside functions" such as movies, sporting events, political rallies, musical recitals, etc., for the past 15 years. Their responses to questions were bland though appropriate. There was no evidence of any hallucinatory behavior. However, they have been completely uncommunicative for a period of at least several months prior to their current hospital admissions.

Their sole objective at the present time is simply to return home to their room where they can resume their past restricted existence. They do not express any hostility toward each other, to the family, or to people caring for them in the hospital.

Past medical histories obtained from the parents revealed that the twins had both been treated for apparent paroxysmal atrial tachycardia which occurred to both of them almost simultaneously at the age of 24 years. There was considerable concern shown by the parents about their cardiac status as evidenced by frequent remarks denoting anxiety and fear least

they engage in any strenuous physical activity such as even running for a bus. Also, each one was treated for apparent collapse of the lung on the same side and, again, almost simultaneously; they also contracted infectious mononucleosis within three months of each other. The remainder of the medical history was essentially noncontributory, though growth and developmental parameters were apparently within normal limits.

There was no evidence for consanguinity in this family and no other relatives were found to have significant psychiatric disease.

DISCUSSION

The etiology of schizophrenia involves interactions of constitutional diatheses and environmental stresses. An individual's constitutional characteristics are the effects of interactions between his genetic make-up and all nongenetic influences from the time of conception. There are two theoretical extremes regarding the relevance of specific genetic factors for different predispositions to schizophrenia. One group consists of the concepts involving general relevance of simple Mendelian genetic factors. i.e., a single dominant autosomal gene (Böök 1953, Slater 1958), a pair of autosomal recessive alleles (Kallmann 1946 and 1953, Hurst 1972), the genes of two autosomal genetic loci (Karlsson 1966). The other group may be represented by Manfred Bleuler's concept of an infinite variety of schizophrenic etiologies involving different combinations of genotypes and life experiences (Bleuler 1968 and 1970). The two theoretical extremes are connected by a series of intermediate and mixed concepts regarding the relevance of genetic differences for different predispositions to schizophrenia (Kaplan 1972). The concept of multiple etiologies (Mitsuda 1967), involving schizophrenic disorders in which the predispositions are associated with simple Mendelian genetic factors, and schizophrenic disorders in which predispositions are not, may be regarded as the 'mixed' concept. The polygenic theories (Ödegård 1963 and 1972, Gottesman and Shields 1967) may be regarded as intermediate concepts, in which different diatheses are associated with cumulative effects of numerous genes at numerous different loci.

Individual differences in predisposition to acquire a schizophrenic disorder in any specific environment are the consequences of individuals being different from each other. The etiological role of biological heredity is involved as a set of potentialities and not a set of already-formed or predetermined characteristics. An individual's genetic make-up only determines his norm or range of reaction, an indefinite but limited assortment of possible manifestations. The more varied the conditions, the more diverse the traits which may develop from any one combination of genes. All of an individual's activities are influenced by the cultural and social systems of his milieu. The fact of cultural involvement, however, is not inconsistent with the relevance of biological functions.

In studying schizophrenia, we are studying secondary products, manifested after the etiological dynamics have occurred. Genetic differences are associated with different predispositions to schizophrenic disorders, but no particular genotype has been

proved to be either necessary or sufficient for development of a schizophrenic disorder. The background of schizophrenia involves genetic differences associated with different dispositions in personality development, and the multitude of human relationships and environmental stresses which interact with the personality idiosyncrasies (Bleuler 1970). There are relatively few published data available which explore, comparatively, specific features of separate mental processes in patients with different forms of schizophrenia and at different stages in development of these forms (i.e., in contrast to comparative descriptions of the characteristics of the already-changed personalities.)

Single-born index cases with the same clinical picture may include a proportion whose conditions have occurred as phenocopies. If, however, one starts with MZ twins as the probands, a separation of the concordant and discordant pairs will in general segregate the genetic cases from the phenocopies (Allen 1954). The occurrence of concordance for schizophrenia in a pair of MZ twins indicates that the etiologies, in the two cotwins, were not sporadic (i.e., as phenocopies), but that they involved interaction between a predisposing genotype and eliciting nongenetic milieu. The principal purpose of the present report has been to provide an intensive retrospective appraisal of the psychological climate in which a pair of MZ twins were reared and who, in turn, developed "chronic simple schizophrenia" at the same time; and who have qualitatively shown almost identical clinical courses and diagnoses. This approach to the problem, while not in any sense as ideal as a *prospective* approach, is nevertheless one that has been infrequently documented in the psychiatric and genetic literature.

On the basis of available history, we find that these patients were reared in an extremely protective environment. We are faced with the problem regarding how much this protective atmosphere was imposed as a result of personality deficiencies in the patients, which might have led to this very type of overprotection from their parents. In addition, we find the oft-quoted family constellation in schizophrenia, comprising a dominant mother and a passive father with the consequent absence of a strong male authority figure in the family. This family is, therefore, well-known for this type of psychodynamic setting. Also, we find a striking similarity in psychological profiles on projective testing of the twins, in addition to the personality characterizations based on histories and psychiatric interviews, which also strongly suggest an almost identical depersonalization process from the standpoint of both qualitative and temporal characteristics. The latter features strongly indicate that, whatever the strengths of the relevant schizophrenogenic environmental forces, they certainly must have been conditioned heavily by a genetic disposition that structured the schizophrenic characteristics even to such detail as the timing of personality decline. On the other hand, one could justifiably argue that there was an unusually strong identification between the twins to the point of their almost copying each other's personality profile with even the inclusion of an environmentally-conditioned time-release factor. It is extremely unlikely, however, that such an event could be copied in such detail as indicated by the results of projective testing with the two cotwins, conducted and evaluated independently by different psychologists.

REFERENCES

- Allen G. 1954. Discussion. Proceedings of the Conference on Problems and Methods in Human Genetics. *Am. J. Hum. Genet.*, 6: 162-164.
- Bleuler M. 1968. A 23-year longitudinal study of 208 schizophrenics and impressions in regard to the nature of schizophrenia. In D. Rosenthal and S.S. Kety (Eds.): *The Transmission of Schizophrenia*. Pergamon Press, Oxford.
- Bleuler M. 1970. Some results of research in schizophrenia. *Behav. Sci.*, 15: 211-219.
- Böök J.A. 1953. A genetic and neuropsychiatric investigation of a North-Swedish population, with special regard to schizophrenia and mental deficiency. *Acta Genet. (Basel)*, 4: 1-100, 133-139, and 345-414.
- Gottesman I.I., Shields J. 1967. A polygenic theory of schizophrenia. *Proc. Natl. Acad. Sci. USA*, 58: 199-205.
- Hurst L.A. 1972. Hypothesis of a single-locus recessive genotype for schizophrenia. In A.R. Kaplan (Ed.): *Genetic Factors in "Schizophrenia"*. Charles C. Thomas, Springfield.
- Kallmann F.J. 1946. The genetic theory of schizophrenia: an analysis of 691 schizophrenic twin index families. *Am. J. Psychiatry*, 103: 309-322, 1946.
- Kallmann F.J. 1953. *Heredity in Health and Mental Disorder*. Norton, New York.
- Kaplan A.R. 1972. Introduction; and, Conclusion: genetics and schizophrenia. In A.R. Kaplan (Ed.): *Genetic Factors in "Schizophrenia"*. Charles C. Thomas, Springfield.
- Karlsson J. 1966. *The Biologic Basis of Schizophrenia*. Charles C. Thomas, Springfield.
- Mitsuda H. 1967. *Clinical Genetics in Psychiatry*. Igaku Shoin, Tokyo.
- Ödegård Ö. 1963. The psychiatric disease entities in the light of a genetic investigation. *Acta Psychiatr. Scand.*, Suppl. 169: 94-104.
- Ödegård Ö. 1972. The multifactorial theory of inheritance of predisposition to schizophrenia. In A.R. Kaplan (Ed.): *Genetic Factors in "Schizophrenia"*. Charles C. Thomas, Springfield.
- Pollin W. 1972. A new approach to the use of twin study data, in studies of the pathogenesis of schizophrenia and neurosis. In A.R. Kaplan (Ed.): *Genetic Factors in "Schizophrenia"*. Charles C. Thomas, Springfield.
- Pollin W., Stabenau J.R., Mosher L., Tupin J. 1966. Life history differences in identical twins discordant for schizophrenia. *Am. J. Orthopsychiatry*, 36: 492-509.
- Pollin W., Stabenau J.R. 1968. Biological, psychological and historical differences in a series of monozygotic twins discordant for schizophrenia. In D. Rosenthal and S.S. Kety (Eds.): *The Transmission of Schizophrenia*. Pergamon Press, Oxford.
- Slater E. 1958. The monogenic theory of schizophrenia. *Acta Genet. (Basel)*, 8: 50-56.
- Stabenau J.R., Pollin W. 1967. Early characteristics of monozygotic twins discordant for schizophrenia. *Arch. Gen. Psychiatry*, 17: 723-734.
- Stierlin H. 1972. The impact of relational vicissitudes on the life course of one schizophrenic quadruplet. In A.R. Kaplan (Ed.): *Genetic Factors in "Schizophrenia"*. Charles C. Thomas, Springfield.

RIASSUNTO

Una coppia di gemelli MZ con diagnosi di schizofrenia cronica semplice è stata studiata, dai rispettivi specialisti, dai punti di vista genetico, sociale, medico e psichiatrico. Psicologi clinici hanno studiato i pazienti indipendentemente e, in base a test psicometrici e proiettivi, hanno rilevato una notevole somiglianza dei profili della personalità. Ad un approfondito studio familiare non sono risultati altri casi d'interesse psichiatrico, né è risultata consanguineità. La famiglia è rappresentativa del tipo frequentemente descritto come famiglia schizofrenica, cioè madre dominante e padre passivo. Viene discussa l'eziologia genetica della schizofrenia.

RÉSUMÉ

Un couple de jumeaux MZ avec diagnostic de schizophrénie chronique simple a été étudié aux points de vue génétique, social, médical et psychiatrique. Des psychologues cliniques ont étudié les patients indépendamment et, sur la base de tests psychométriques et projectifs, ont trouvé une remarquable ressemblance dans les

profils de la personnalité. L'étude familiale n'a pas pu révéler d'autres cas d'intérêt psychiatrique ni de consanguinité. La famille représente le type fréquemment décrit comme famille schizophrénogénique, c'est-à-dire mère dominante et père passif. L'étiologie génétique de la schizophrénie est discutée.

ZUSAMMENFASSUNG

Ein EZ-Paar mit einfacher chronischer Schizophrenie wurde aus genetischen, soziologischen, medizinischen, und psychiatrischen Gesichtspunkte untersucht. Psychologische Kliniker untersuchten die einzeln durchgeführten psychomotorischen und Projektionstests, und stellten eine erhebliche Ähnlichkeit im Persönlichkeitsprofil fest. Eine gründliche Familienuntersuchung ergab keine weiteren psychiatrischen Fälle sowie auch keine Blutsverwandtschaft. Die Familie gehört zu dem oft als schizophrénogen bezeichneten Typ mit dominierender Mutter und passivem Vater. Anschliessend Diskussion über die Ätiologie der Schizophrenie.

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