

## Schizoidness in Twins

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### Introduction

The purpose of this article is to review the deviations found in monozygotic partners of schizophrenic twins in order to examine the concept interpreting schizoid disorders as attenuated or incomplete expressions of schizophrenic heredity ("forme frustes").

The genetic concept of schizoid disorders—designated originally as schizoid psychopathy, or schizoidia (Alanen *et al.*, 1963), currently as schizoid personality, schizophrenic character (Schafer, 1951), or cryptogenic schizophrenia (Brit. Med. J., 1964) assumes that these entities are conditioned by the same specific genotype as full schizophrenic psychosis. Two types of genetic mechanisms can be visualized as underlying a "forme fruste": an imperfect degree of manifestation of full genotype, or an effect of just a part of the genotype, if the hereditary conditioning is a complex one (Gregory, 1960; Planansky, 1955).

Increased occurrence of schizoid phenomena in schizophrenic families (Kallmann, 1938) has been quoted as the main evidence of genotypic communality with full psychosis. However, it is difficult to evaluate the reports claiming increased rates since normative data are not available (Planansky, *unpubl.*). No independent indicator of schizophrenic genotype is available, and thus the definition of schizoid disorder has been inconsistent: some clinicians require evidence of autism, whereas others have been ready to accept any deviant sibling in schizophrenic family as a carrier of the specific genotype. Generally, schizoid pictures would be expected to vary from one carrier to another. A tapered effect of genotype would be theoretically anticipated if a polygenic mode of heredity is involved. All these circumstances contribute to the uncertainty concerning the range of phenotypic expressions of schizophrenic heredity, and hinder their differentiation from schizoid patterns genetically unrelated to schizophrenia.

Much of this uncertainty concerning not only the phenotypic boundaries, but possibly even the existence of the incomplete genetic schizophrenia, can be removed by employing sets of individuals possessing identical genotypes, i. e. monozygotic

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(MZ) twins, one of whom is clearly schizophrenic, and the other non-psychotic. Such pairs are conventionally designated discordant as to schizophrenia.

The success of this approach depends on our ability to identify the "genetic" cases of schizophrenia. Genetic schizophrenia is often equated with process psychosis, the implication being that the latter should comprise deteriorative, malignant psychosis. The reactive, non-genetic psychoses, by the same implication, are usually sought among the mild or transient types. Such a division would be too bold from a genetic view-point, even if the clinicians produced evidence validating the "process-reactive" dichotomy. Severity of the disease process alone will not help us in separation of genetic cases.

Concordance by itself is of little help, since the twins may be concordant either because of common genes, or because of common experiences. The problem of separating "genetic" twin pairs from others has been with us since the beginnings of the psychiatric twin study. Luxenburger (1936, 1940), tested the possibility that there would be less schizophrenia among the siblings of discordant than of concordant MZ pairs, if the discordant pairs consisted of genetic as well as non-genetic cases. Surprisingly, he found the opposite — a very high loading. Similarly, there was no difference in genetic load in the families of his manic depressive twins (Luxenburger, 1933). Rosenthal (1959) examined male pairs in Slater's sample, considered discordant by the investigator, and concluded that there was an almost total absence of schizophrenia in the families of discordant twin pairs. In recent reports (Kringlen, 1964; Tienari, 1963) of well documented discordant pairs, other cases of schizophrenia were found in most families. In view of these conflicting observations, the usefulness of discordant MZ pairs for the separation of phenocopies from truly genetic cases (Allen, 1954) may not apply to psychiatric conditions, where the definition of the trait is notoriously inconsistent.

It appears then that most of the schizophrenics in the pairs considered conventionally as discordant, are of the genetic type — at least they are not less likely genetic than the cases in concordant pairs. It is therefore permissible, for the purpose of this study, to regard their genotypes as specific. Since an unknown proportion of the discordant pairs may still be non-genetic, some of the schizoid patterns may be erroneously attributed to the specific genotype, if we decide to lump all reported pairs in the genetic category.

The examination of schizoid phenomena in twins may help to answer the following questions:

1. Are schizoid deviations in individuals possessing the specific genotype invariably characterized by some of the basic schizophrenic attributes, such as autism, thought disorder, etc., or can the genotype reveal itself also under the guise of normal variations of temperament and character? Only an answer on descriptive level is anticipated. Most likely, a special, direct and prospective study would be required to clarify, whether the schizoid character traits in the cotwin result from incomplete penetration, or whether they are induced reactively, perhaps through identification (Burlingham, 1952; Lowinger, 1963; Rosenthal, 1960).

2. The usual information, consisting of current clinical examination, together with some selected retrospective data, and occasionally a brief follow-up observation, may be adequate for judging whether schizoid disorders represent an emergent phase of psychosis, a quiescent state subsequent to an active phase, or whether they are unrelated to the psychotic development.

3. Presence of the schizoid, pre-psychotic personality in the prodromal phases of both twins' illnesses would lend support to a view that schizophrenic psychosis develops as continuation of an early schizoid life adaptation. Absence of a schizoid phase would separate another type of psychosis conceptualized as an autonomous process, independent of, indeed alien to, the original personality structure.

4. The distribution of the intra-pair combinations of clinical features and degrees of severity of the disease could yield cues concerning the dependence of clinical psychosis on heredity. For instance, if severe schizopremia is paired with any clinical type or degree of severity in a more or less random fashion, it will be concluded that the observed psychosis is dependent less on heredity than on other influences. Of course, we would feel more confident about the assigning of respective shares to heredity and environment if we had twins separated from infancy to analyze, but that is a utopian project.

5. Examination of the boundaries of schizophrenic phenotype will be useful for reviewing the applicability of the traditional concordance-discordance schema. In this regard, the notion of the phenotypic continuum will be considered.

A study limited to inspection of the clinical pictures in monozygotic twins cannot aim at a formal analysis of heritability. The latter requires a comprehensive analysis of variances in twins and their families (Kempthorne, 1961), a task that can be hardly achieved in psychiatric conditions. Any generalizations should be qualified by the awareness that the twins' situation is atypical biologically, psychologically, as well as socially, and thus their adaptations may be unusual and atypical, not necessarily applying to ordinary individuals.

This brief review comprises probably all the cases of MZ twins where a schizophrenic twin's partner is either non-psychotic or normal — as far as they were obtained by a specified sampling procedure, and were published with at least partial case histories. All published clinical descriptions of MZ twins were carefully reviewed, and assessment made of the degree of concordance of psychopathology. For comparison, two samples of DZ twins were examined. Admittedly, the samples are not comparable, the information is not complete. In view of the notorious lack of uniformity in the diagnostic usages, and regarding the limited numbers, only cautious generalizations can be made.

### Observations

The variability of the schizophrenic pictures within MZ pairs has been noted since the first modern investigation of psychotic twins by Luxenburger (1928). It was confirmed in the first American study by Rosanoff (1934), and documented

with clinical details by Essen-Möller (1941), and Slater (1953). Essen-Möller, for example, felt that in some pairs each twin could be assigned a different diagnosis. Thus, four typically regressed schizophrenic index cases were found paired with: 1, a borderline schizophrenic case; 2, an intact personality after two transient attacks; 3, a transient post-partum episode; and 4, a recurrent depressive state.

Slater's sample contains more pairs with typically chronic hospital residents than Essen-Möller's sample, but even among these marked intrapair variability is seen. For example, severe, chronic schizophrenia is paired with a brief, early episode, and with an acute post-partum psychosis; a mild, early psychosis is paired with late paranoid schizophrenia; and a typical hebephrenia is paired with late paranoid schizophrenia. In contrast, a marked similarity was found in the illnesses of the recurrent type; in two of the four pairs, the illnesses were of a clearly catatonic type.

### Normal cotwins of schizophrenic index cases

Until recently only a few completely discordant pairs were reported with adequate details. Essen-Möller reviewed the literature up to 1941, and found only 4 MZ pairs where one twin was schizophrenic and the other normal. In Essen-Möller's sample, there is only one pair where the cotwin was normal (Pair No. 8): The index case had a schizo-affective psychosis of recurrent type, the cotwin was never psychotic. No personality traits suggestive of schizoidness were elicited in her history: She was described as always happy and good-natured.

There are several other normal cotwins in the same series of pairs, however their index cases' psychoses were not schizophrenic.

In Slater's sample, normal cotwins were found in seven pairs. Complete information was secured only in one of them (Pair 13). The index case was a deteriorated schizophrenic. Her twin sister had no history of deviant behavior or oddities. No sign of schizoidness was noted when she was seen at 49.

Kringlen's (1964) sample was obtained from 5 Norwegian hospitals. Detailed accounts of 2 concordant and 6 discordant MZ pairs with typically schizophrenic index cases were published. All of the cotwins were clinically normal. Only in one instance (Pair 3) the schizoid label seems to be applicable: The cotwin is described as reticent, having few contacts, none with women. He was often lost in thought, daydreaming, and lived secluded in a small clean cabin, which he built himself. He is described as polite and humble. Mild schizothymic and neurotic traits (according to the investigator's evaluation) were reported in the history of the cotwin of pair 6: he never married, had a tendency toward headaches and sleeplessness, but he never sought medical help. No borderline cases with psychopathology suggestive of schizophrenic process have been reported in Kringlen's sample.

Whereas the above three samples were collected from hospital populations, Tie-nari's (1963) sample is the only one obtained from a full population survey. All male twins born during the 1920-1929 period in Finland were investigated. MZ twins with psychiatric diagnoses were derived from a larger part (70%) of the sample.

Most of the twins were directly examined, and in addition to psychiatric interviews, they were evaluated by a series of the usual psychological tests.

The most striking feature of Tienari's sample is the lack of concordance — none of the cotwins was found to be schizophrenic by the investigator, although the index cases were typical regressed, chronic schizophrenics. Normal cotwins were found in 10 pairs out of 16. In most of these normal cotwins, character or personality features were observed which could be conveniently classified as schizoid. The term "introverted" was used descriptively in about half of the cases.

### **Abnormal, but non-psychotic cotwins**

Essen-Möller found in the literature altogether 11 pairs where the cotwin of a schizophrenic index case was diagnosed schizoid. In his sample there are two such pairs, illustrating the typical problems of diagnostic assessment:

Pair No. 4 (Essen-Möller, 1941). Both twin brothers wandered around the country leading a hobo-like existence, until they were found, after a year's time, destitute, exhausted, and in a psychotic state characterized by anxiety and self-reproaches. Anselm's (index case's) psychosis developed gradually into a typical hebephrenic schizophrenia, whereas Konrad's illness receded after separation from Anselm, leaving behind characterological traces. The latter were called schizoid by the investigator. In the interview, Konrad was found to be absent-minded, dreamy, shy, not fully in affective contact, showing rigid facial expression. His personality was clearly abnormal. The investigator acknowledges that both twins' psychoses belonged in the schizophrenic group; however he does not call Konrad's transient episode a schizophrenia, feeling that it had been induced reactively by Anselm's illness. Konrad's state could probably be interpreted as a schizophrenic character, if the differences between the initial anxiety psychoses were less stressed. In other words, it could be seen as a quiescent stage of the same process which had progressed in chronic schizophrenia in his partner.

It seems that many instances of schizoid personality, schizoid character, or schizoid psychopathy, found in the reports dealing with schizophrenic families, belong in a similar category, namely the arrested phase of schizophrenic process.

In contrast, no psychotic history was obtained in Ida, the cotwin in pair No. 6 (Essen-Möller, 1941). Julia, the index case, was delivered with forceps, and showed asphyxia. She was always weak and sickly, and was slower in school. At 24, she had a typical catatonic illness, and was still in the hospital at 63. Ida's history indicated character traits interpreted as schizoid by the investigator. She is said to have been, at one period of her life, quarrelsome, distrustful, obstinate, and very nervous. Her husband was an alcoholic. She had always been too conscientious and meticulous, even according to her own evaluation, and always worked incessantly. During the interview, she was found normal, warm, and accessible. The question which arises, of course, is as follows: Would we consider a schizoid label were she not known as a cotwin of a schizophrenic patient?

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In Slater's sample we find the following illustrative cases: Pair 291, (Slater, 1953). Dulcie, the index case, was always nervous and worrisome. She became overtly psychotic at 31, and never left hospital. Helen was better in school and always more lively. The cue to her personality can be seen in the following characterization: "They were always close to one another... they drifted apart when Helen got a boyfriend. She met him dancing; but gave him up when she found she preferred her own or her twin's company". When she was seen at 43, she had hypochondriac and neurotic complaints, and her condition was classified as a chronic anxiety state.

Other examples of cotwins who could be classified as schizoid: Cotwin in pair 6 (Slater, 1953) was never hospitalized and led a socially marginal life. Cotwin in pair 23 (Slater, 1953) is described as deeply religious and "a little fanatic". He never married.

In Tienari's (1963) sample, 6 out of 16 non-psychotic cotwins showed a diagnosable abnormality. Somatic complaints were the most frequent ones, but were not described as bizarre. The neurotic-like symptom complexes were of the types usually classified as psychosomatic, hypochondriacal, or neurasthenic, and they occurred in settings of neurotic character rather than as expressions of separate, active neurotic illnesses. The case histories convey an impression of uncomfortable, dysphoric men, some of them introverted, pedantic, rigid, avoiding people, others fatigued, passive, indecisive, anergic. For instance, a physical training instructor (pair 881) felt that "life tasted as if drinking tar," while another cotwin (pair 608), a farmer and lumberman, complained of the "heaviness of life". In the latter case, psychological tests revealed defective intellectual functioning (IQ 61) and poor memory. He was characterized as a primitive, schizoid person, and there was a question of deterioration. His school record was poor, whereas, his twin's (i. e. index case's) achievement in school was excellent. The latter, whose IQ was 72, developed psychosis of a hebephrenic type insidiously, without remission.

The wide variability within the MZ pairs with schizophrenic index cases located in hospitals, was confirmed by Inouye's report (1961). This report is especially significant since the sample was obtained in a non-European population (Japan). The cases were consistently diagnosed and zygosity was determined serologically. No case histories were published. The data were published in statistical form, and are presented in the following table, which has been abstracted from table 4 in Inouye's report.

The category of "Relapsing schizophrenia" was not included in our table. The latter consists of 7 index cases whose illnesses were of catatonic nature and showed marked affective features. Most of the cotwins had also relapsing psychosis.

Inouye's classification of the cotwins as concordant and discordant has been preserved in Tab. 1. His category of "Transient psychotic episodes resembling schizophrenia" has been included in "Transient or chronic mild schizophrenia", on the assumption that the stress is on the psychotic history rather than on the preferred classificatory system.

The index cases are grouped essentially according to the severity of the disease, although clinical criteria may coincide with this division. According to the investi-



**Tab. 1. Clinical variability within MZ pairs with schizophrenic index cases in Inouye's sample**

Index case	Cotwin				
	Concordant			Discordant	
	Chronic progressive schizophrenia	Chronic mild or transient schizophrenia	Neurotic states resembling schizophrenia	Schizoid personality (deviant)	Schizothymic personality (intact)
Chronic progressive schizophrenia	11	3	3	6	—
Mild or transient schizophrenia	—	7	2	10	4

gator's description, the chronic progressive schizophrenia corresponds roughly to the classical *Dementia praecox*, whereas the milder group appears clinically heterogeneous.

Some evidence of sampling effects can be seen in the pairings, as presented in the above table. In the milder group, the psychoses emerged, on the average, at a later age. Thus some of the features, such as the milder course and atypical form, may be a function of the age at onset. The cotwins usually become ill after the index cases in hospital based samples. Accordingly, mild schizophrenia is found among the cotwins of the severely ill index cases, but no severe, deteriorating psychosis is reported among the cotwins of index cases with milder illnesses. These are the features typically found in twin samples collected in psychiatric establishments.

The connection between psychosis and non-psychotic disorders, designated here as "Chronic neurotic states, resembling schizophrenia", is implied by the finding of basic schizophrenic symptoms, such as cognitive distortion, and tendency toward ego-disturbances in the cotwins, according to the investigator.

The category of schizophrenia-like neurotic states probably comprises cases often called "borderline", i. e. cases where the symptoms are not decisive enough for unequivocal psychotic diagnosis. Borderline states have been reported in a recent study of siblings of schizophrenics (Alanen *et al.*, 1963). It is stressed that they are not "...schizoid in the classical sense", but show unmistakable elements of schizophrenic pathology. Those are the types of disorders which evoke an impression that a further progression into a frank psychosis is likely to ensue. Inouye mentions the pseudo-neurotic schizophrenia in this connection. Overlapping with the next category, the deviant schizoid personality, is unavoidable.

### Dizygotic pairs

In all investigations, the primary data on DZ twins are less fully reported than the MZ data. Brief clinical descriptions of DZ pairs are available only in Essen-Möller's and Slater's reports. Ordinary type of schizophrenia is the prevailing type in both series.

In Essen-Möller's sample, all cotwins were free of major deviation. Several of them displayed, during the interview, behavior which was interpreted as neurotic, or revealing peculiarities, best understood in terms of normal personality types. In only one instance was the cotwin called schizoid (pair 49).

According to Slater's tabulations (1953: pp. 256-271), in the 102 DZ pairs with schizophrenic index cases, there were 2 pairs with organically affected cotwins, 5 with affective illness in the cotwin, and 27 with "psychopathy or neurotic illness". In the remaining 68 pairs, the cotwins were normal. Most of the cotwins were old enough to have passed the typical risk period for developing schizophrenia.

Only 3 out of the 27 abnormal cotwins are expressly called schizoid. This lack of use of the term schizoid was deliberate. It was limited to those cases where further accounts are presented elsewhere in the report (Dr. E. Slater, personal communication). In the characterizations of at least 9 other individuals, one finds terms consistent with schizoidness (shy, very morose, miserly, peculiar, always studying something and getting nowhere, keeps to himself, lacking in affection, excessively reserved, rigid, poor affective rapport, etc.). In the remaining cotwins, chronic anxiety states or fairly conspicuous signs of persistent neurotic states are described. In contrast, the 68 cotwins classified as normal, are described in terms of personality characteristics without indications of pathology.

When the pairs are divided in two groups, one with neurotic, and the other with normal cotwins, there is no striking difference between the diseases of the index cases. The proportions of the clinical types — catatonic, hebephrenic, or paranoid, — are similar in both groups. In both groups, other cases of schizophrenia were found in half of the families. According to Slater's assessment, the disease led to hospital invalidism in nearly all cases. No difference in the outcome was found when the cases were grouped into paranoid and non-paranoid. Clearly, the sample was drawn from long-term patients with severe diseases, and the presence or absence of schizoidness or other abnormalities in cotwins cannot be explained by the recorded attributes of the index cases.

### Discussion

The data reviewed here suggest that in a MZ pair, where the index case is a typical chronic schizophrenic, the cotwin can apparently develop any clinical type of schizophrenic psychosis, or any kind of state diagnosable as schizoid or neurotic character, including non-pathologic personality patterns. Moreover, the cotwin can remain clinically normal throughout the risk period, although at times a schizothymic label may be used; the latter designation describing a temperament attribute. In other words, the clinical pictures which can be seen in an individual possessing the same heredity as a regressed schizophrenic, comprise a graded series of disorders and personality patterns, ranging from a duplication of the psychosis to clinical normality. The measure of gradation is primarily psychopathologic — the degree of personality disintegration — but the assessment is unavoidably contaminated by the observation of social fitness.



In order to understand the meaning of this phenotypic variability range, compatible with identical genotype, we should first consider whether the pairs with non-psychotic and normal cotwins are exceptions — as might be judged from most reports — or whether they are common. Definite answer can be obtained only from the study of distribution in general population, however the reviewed data permit some fairly safe judgments.

Data from two types of sources have been reviewed: four hospital samples and one population sample. In a sample obtained from hospital patients, chronic, severe index cases are unavoidably over-represented (Rosenthal, 1961, 1962). Moreover, pairs with normal or mildly disabled cotwins will be missed, unless the twinship status of all index cases had been checked with the birth register records. This sampling bias will explain the rarity of schizoid and normal cotwins in the hospital samples reviewed here, as contrasted with the population sample where only schizoid and normal cotwins were found. Considering both sources, pairs with non-psychotic and clinically normal cotwins should be quite common. A new facet of the sampling difficulties inherent in psychiatric twin study has been encountered: not in a single pair in well over half of the male Finnish population, in age range of 30-40 years, have both twins been found schizophrenic.

In the aggregate observations, all types of schizophrenia and all degrees of illness in the index cases appear to be paired with non-psychotic or normal cotwins. No trend toward special or restricted types of pairings can be discerned, excepting perhaps the recurrent type. Considering the severity of the disease alone, the most disintegrating Dementia praecox is found quite often in the same pair with clinical normality. Qualitatively, the intra-pair combinations appear to occur more or less at random.

The narrative data show fairly clearly that a considerable proportion of schizoid states in cotwins most likely represent residual states ensuing from an arrested psychotic process. Other instances are suggestive of the pre-psychotic, or perhaps prodromal phase. We are reminded that the pre- and post-psychotic phases of schizophrenia were the original models for the concept of schizoid disorder (Kraepelin, 1915).

Yet the psychopathologic connection with schizophrenic process is not evident in all deviations and personality patterns, described in the reviewed reports. We still do not know, for instance, whether the neurotic states in cotwins are related to the experiential circumstances which triggered the psychosis in the index cases, or whether they should be regarded as a reaction to the twin partner's predicament. We cannot claim such neurotic-like states as schizophrenic phenotypes merely because they occurred in MZ cotwins of schizophrenics. For instance, in a selected series of discordant MZ pairs, studied at the U. S. National Institute of Mental Health, a predominance of obsessive-compulsive symptoms was noted in cotwins (Dr. James R. Stabanau, Personal Communication).

No independent evidence is available to show that schizophrenic psychosis is a maturation of a true psychoneurotic entity, or that schizophrenia may be mellowing into a neurotic state. Clinicians have long considered that a dichotomy exists between neurotic and psychotic disorders; Eysenck (1961) having produced the most

systematic evidence in support of this view. Some separateness of the schizophrenic phenotype is suggested by a study of remission states in schizophrenic patients: no psychometric evidence was found that patients resorted to neurotic defense patterns (Johnston, Mc Neal, 1965). Thus, even in identical twins, the seeming continuum of schizophrenic psychosis-schizoid disorder-neurotic character-normal personality and temperament pattern may consist of two discrete series: true schizoid disorders with underlying schizophrenic pathology, and a heterogeneous remainder, which is psychopathologically separate from the schizophrenic syndrome. This division might reflect the effect of the threshold, blurred by our loose diagnostic usage.

The random combination of full psychosis with various types of schizophrenia, true schizoid states, as well as clinical normality, calls for interpretation consistent with genetic conditioning. The concept of fluctuating psychotic processes will be plausible without requiring auxiliary hypotheses. The concept of psychotic process implies that time is the variable relevant to the description of any schizophrenic or schizoid state: The process emerges, proceeds, ceases to proceed, retreats, or even submerges below the level of clinical recognition. It is logical to visualize these fluctuations as reflecting changes in the concomitant physiological states. For instance, fluctuations in the condition of the capillaries were observed to coincide with changes in clinical state (Maricq, 1964). This notion of fluctuating process implies that schizoid, prepsychotic or arrested states may be encountered at any stage of schizophrenic development.

Assuming that the combinations of the clinical pictures within the pairs occur more or less at random, and taking the absence of clinical abnormality as an extreme case of the age differences at the onset of the clinical disease, the following variables can be considered: age at the onset, rate of progression, clinical form, and severity of the illness. The inter-play of these variables is consistent with a model of a disease process subject to a threshold for its clinical appearance (Edwards, 1960). A switch mechanism could be visualized as bringing about the threshold effect: To illustrate, a cotwin of a regressed schizophrenic could remain irreversibly spared of abnormality, if the switch event did not occur at the critical point. Levels of metabolic substances at or above which certain processes originate or deviate, events upsetting the sequence of the individual development, or even learning situations occurring at a specific time ("traumas") could trigger the genotypically conditioned process.

We have been accustomed to the schizophrenia-normality dichotomy, creating a semantic threshold. The twin observations help us to appreciate that a phenomenal continuum exists, reflecting the seemingly gradual emergence of the psychotic process. Even if all schizoid phenomena are actually prodromal stages of full schizophrenia, it still may be practical and accurate to preserve the term schizoid for everyday clinical usage.

Evidence of a separate type of schizophrenia, ushered in by a schizoid phase, is still inconclusive. According to Essen-Möller's interpretation of his findings, the schizophrenic component occurs in both twins, if it is at all present: Even if the cotwin's illness is of a borderline character, with mild or no gross schizophrenic symptoms, the schizoid admixture can be found. These conclusions were based on a very small

number of cases. In other narrative reports ample data can be found, suggesting concordant occurrence of the schizoid phase or features. However, no consistent, direct and prospective observations are available.

The recurrent, agitated type apparently lacking the schizoid prodromal stage appears to be suggestively defined through the concordance, reported in Slater's and Inouye's investigations. However, we do not know whether the full concordance is a rule. Patient's stay in hospital is usually brief, and thus only spotty observations are available. This type should be studied together with cases of periodic affective states.

A quantitative connotation is so much a part of the schizoid concept that the notion of reduced severity may be inseparable from the qualitative definition. This quantitative aspect, determined largely by the degree of social severity of the disease, has bearing on evaluation of psychopathology, ascertainment of schizoid subjects, and on statistical expression of intrapair similarity.

A schizoid individual, or a quiet schizophrenic outside of the hospital, is not necessarily less "sick", either globally or in different dimensions of his psychopathology, than a schizophrenic on a chronic ward. Impressionistic assessments based unavoidably on practical, social considerations, are inadequate for the establishing of the point separating deviations characterized by the basic schizophrenic attributes, from non-specific disturbances and normal personality variants. Specified components of schizophrenic psychopathology will have to be measured.

An undetermined proportion of schizoid and other crypto-schizophrenic twins remains outside of the samples because they are not socially disruptive. Their intrapair variability is not observed. Bleuler (1917, 1950) stressed there must be many more cases of latent schizophrenia, schizophrenic psychopathy, and neuroses masking schizophrenia than cases of diagnosed psychosis. In very mild deviations, the differentiation between concordant and discordant relationships would be difficult and inconsistent, since it depends on assessment of social impairment. If these mild cases are assessed randomly as concordant or discordant, the concordance rates originally obtained with ordinary schizophrenic index cases should not be affected by inclusion of schizoid index cases in the sample. Since the thoroughness and consistency of ascertainment influences significantly the computation of the concordance rates (Allen, 1955), the method employed in collecting the sample will be especially important.

Selection of index cases for high severity of the studied disturbance, with random combinations of the severity degrees, may overstress the intrapair differences: pairs with concordantly mild disturbances will not be included. Intra-pair similarity in the population, expressed through correlation techniques, may then be higher than the value describing the registered sample, taken from the same population. This technical problem was recognized in dealing with twins from an official register of alcoholics (Kaj, 1960). In the twin studies of psychological traits, employing student's populations or other ordinary samples, this technical problem does not arise (Cattell *et al.*, 1955; Gottesman, 1963; Vanderberg, 1962).

The apparent random combinations relate to clinical pictures, not to the presence and absence of the disease process. In contrast to the MZ pairs, the line separating

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the concordant from discordant pairs in DZ series appears to be sharper: the cotwins are either psychotic, or normal, as in Essen-Möller's series. In Slater's sample, aside from the normal DZ co-twins, there is a proportion of cotwins with definite deviations, but as far as can be judged from the terse descriptions, they are neurotic or other minor deviations without the schizophrenic flavor. A more than ordinary load of neurotic and similar states would be expected in view of the generally reported background of minor disturbances in schizophrenic families. Yet a precise differentiation from true schizoid states is necessary, otherwise the difference between MZ and DZ discordances might be blurred, as seen from the following illustration. In Slater's DZ series, there are 10 concordant and 102 discordant pairs. The discordant pairs include 7 with an affective or organic state, and 27 with a neurotic or psychopathic cotwin. If these 27 pairs were added to the 10 concordants (employing a broad definition of schizoidia, as found at times in older writings), the simplified concordance rate would be  $37:105 = 35\%$  ( $SE \pm 9.46$ ). Similarly calculated MZ concordance would be 65%.

In the DZ pairs, the proportions of normal cotwins are much too high to be interpreted exclusively as instances of a below-the-threshold process, masked by normality, as is presumably the case in normal MZ cotwins. Comparison should be made within individual sample where ascertainment was consistent for both categories. Thus in Essen-Möller's sample, obtained predominantly from consecutive admissions, checked for twinship in birth registers, the proportion of normal cotwins is conspicuously larger in DZ than in MZ pairs (21:4 in DZ as against 4:7 in MZ).

Substantially higher concordance rates in MZ than in DZ pairs, reported in all studies except Tienari's, have been the mainstay of the genetic theory of schizophrenia, complementing other evidence of family concentration. Thus far, evidence of family concentration has been based on the occurrence of full schizophrenia in families and twin pairs, selected through severely ill, schizophrenic, index cases: schizoid cases were noted only incidentally. It is unlikely that the high ratio of MZ to DZ concordances would be abolished by counting schizoid cotwins as full psychoses in hospital samples. We can only guess how the MZ:DZ concordances ratio would be affected in population sample where all subjects are screened, if the diagnostic threshold were lowered. Some of the pairs "schizoidia-schizophrenia" would be already collected in hospital samples. Pairings "schizoidia-schizoidia" might be under-represented in a population sample, being collected occasionally as discordant because of inconsistent assessment of severity. Some of the pairings "schizoid-normal" would be missed as normal. It is difficult to visualize, in advance, the relative magnitudes of schizophrenia rates in all categories of relatives, with the extended definition of the schizophrenic phenotype. It is unlikely that the new rates would parallel neatly the rates of the twin-family investigations, as are known, for instance, from Kallmann's work (1946).

These observations suggest that further inquiry on the clinical level in twins and their families should be focused on the study of those forms of schizoid adaptation and neurotic states which are not obviously referable to an actual psychotic phase, with

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the aim of separating the specific deviations from the diffuse and heterogeneous background of minor disturbances. Most urgently needed information concerns the basic Bleulerian components of the schizophrenic mode of life, as recently reformulated by Meehl (1962). Psychological study of the clinically normal twins should clarify the sub-clinical limits of Bleulerian specificity. Considering the nearly limitless difficulties and pitfalls inherent to the twin study alone, the first task still is to find a feasible technique which would secure the desired primary data.

Finally, concordance-discordance should be evaluated on two different levels: the conventional, all-or-none type of evaluation in terms of the concordant-discordant dichotomy for the purpose of comprehensive statements, not requiring precision. It implies an arbitrary definition of the presence of schizophrenic disease. The latter could be established, for instance, by at least one of the basic schizophrenic attributes. A quantitatively expressed degree of similarity between the twin partners in the individual pairs, measuring separate dimensions as well as describing a summarized estimate along phenotypic continuum. In such measurements, the line between psychopathological phenomena and normal range would be blurred. Empirical cut-off points would have to be introduced.

In conclusion, the reviewed data show that in identical genotypes, the schizoid disorder may develop as phenotypic equivalent of any type of schizophrenic psychosis, representing thus an expression of schizophrenic heredity. In most instances the true schizoid disorder is nothing but one of the developmental phases of schizophrenia, and should not be viewed as an incomplete or attenuated form of full psychosis. Presumably, in clinically non-deviant personalities, components of schizophrenic pathology could be identified through psychological study, and thus could be considered expressions of the specific genotype. The available twin data deal with schizoid phenomena on clinical level, and do not permit to include schizophrenia as a theoretical construct in the verified expressions of the specific genotype.

### Summary

Discordant schizophrenic monozygotic twin pairs were examined in order to clarify the view interpreting schizoid disorders as attenuated or incomplete expressions of schizophrenic heredity. The aggregate observations indicate that typical schizophrenia can be paired with all types and degrees of schizophrenic or schizoid illness, including clinical normality. These intra-pair combinations seem to occur more or less at random. The reviewed schizoid states showing evidence of schizophrenic psychopathology represent one of the developmental phases of schizophrenic process. Yet the psychopathologic connection with schizophrenia is not evident in all deviations and non-deviant personality patterns. Thus the seeming continuum of schizophrenic psychosis — schizoid disorder — neurotic character — normal personality may consist of two discrete series: one with underlying schizophrenic psychopathology, and the other independent of the psychotic process. This division might reflect a threshold effect, dependent upon a switch mechanism.

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#### RIASSUNTO

Allo scopo di chiarire il punto di vista, secondo cui i disturbi schizoidi sarebbero una espressione attenuata o incompleta di un'eredità schizofrenica, sono state esaminate coppie discordanti di gemelli monozigotici schizofrenici. Le osservazioni raccolte indicano che la schizofrenia tipica può accompagnarsi a tutti i tipi e gradi di malattia schizofrenica, o schizoide, compresa la normalità clinica. Tali combinazioni intra-coppia sembrano verificarsi più o meno a caso. Gli stati schizoidi riesaminati, indicanti una psicopatologia schizofrenica, rappresentano una delle fasi di sviluppo del processo schizofrenico. Ciononostante, il legame psicopatologico con la schizofrenia non risulta evidente in tutti i tipi, devianti e non-devianti, della personalità. Perciò, l'apparente *continuum*, costituito da psicosi schizofrenica — disordine schizoide — carattere neurotico — personalità normale, è probabile che consista di due serie diverse: una con un substrato di psicopatologia schizofrenica, e l'altra indipendente dal processo psicopatologico specifico. È probabile che tale divisione rifletta un effetto-soglia, attivato da eventi specifici.

#### RÉSUMÉ

L'A. a examiné des couples discordants de jumeaux MZ schizophrènes, afin de clarifier le point de vue interprétant les désordres schizoïdes comme l'expression atténuée ou incomplète d'une hérédité spécifique. Les observations obtenues montrent que la schizophrénie typique peut s'associer à tous les types et degrés de la maladie schizophrénique ou schizoïde, ainsi qu'à la normalité clinique. Ces combinaisons intracouples semblent survenir au hasard. Les formes schizoïdes réexaminées, montrant une psychopathologie schizophrénique, représentent l'une des phases évolutives du processus schizophrénique. Malgré tout, la connection psychopathologique avec la schizophrénie n'est pas évidente dans toutes les formes de déviation et non-déviation de la personnalité. Ainsi, il est possible que les formes, apparemment sans interruption, de psychose schizophrénique — désordre schizoïde — caractère névrotique — personnalité normale, soient constituées de deux séries séparées: l'une montrant un certain degré de psychopathologie schizophrénique, et l'autre indépendante du processus psychotique spécifique. Cette division peut présenter un effet de seuil, causé par des processus spécifiques.

### ZUSAMMENFASSUNG

Diskordante schizophrene EZ Zwillinge wurden untersucht um klarzustellen, ob schizoide Abnormalitäten als mitigierte oder unvollständige Ausdrucksformen schizophrener Vererbung anzusehen sind.

Summarische Beobachtungen deuten darauf hin, dass typische Schizophrenie in ein und demselben Paar mit allen Formen und jeder Intensität von schizophrener oder schizoider Erkrankung, einschliesslich klinischer Normalform vorkommen kann. Diese paarweisen Kombinationen scheinen sich mehr oder weniger zufallsmässig zu ereignen. Die beschriebenen schizoiden Zustände, die Merkmale schizoider Psychopathologie aufzeigen, sind als eine der Entwicklungsphasen des schizophrenen Prozesses aufzufassen.

Jedoch die psychopathologische Verbindung mit Schizophrenie ist nicht in allen Zuständen, ob normal oder abnormal, nachzuweisen. Daher kann das anscheinende Kontinuum von schizophrener Psychose, — schizoider Abweichung — neurotischem Charakter — normaler Persönlichkeit, aus zwei separaten Serien bestehen: Eine ist basiert auf schizophrener Psychopathologie, und eine ist unabhängig vom psychotischen Prozess. Diese Zweiteilung kann einem Schwelleneffekt entsprechen, der auf einem Schaltungsmechanismus beruht.