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Schizophrenia in Kosrae, Micronesia: prevalence, gender ratios, and clinical symptomatology

Merilyne C. Waldo *

*Department of Psychiatry, University of Colorado Health Sciences and Denver Veteran's Administration Medical Center,
4200 East 9th Avenue, Denver, CO 80262, USA*

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Abstract

The utility of genetic isolates for research is in part based on the assumption that the illness of interest is similar across cultures. In this report, we review the data on schizophrenia in Micronesia, a collection of small islands in the western Pacific Ocean. Significant variations in prevalence between the islands have been reported, as have male to female ratios which are strikingly high. We focus on the patients in Kosrae, one of the islands in the Federated States of Micronesia. Twenty-two schizophrenics are identified. We found a prevalence rate of 6.8 per 1000 and a male to female ratio of 6.3:1. Although, in many ways, the patients resemble their Western counterparts, 19 of the patients were episodically mute, especially when untreated or undermedicated. Possible explanations for this unusual symptom are discussed. © 1999 Elsevier Science B.V. All rights reserved.

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1. Introduction

Schizophrenia is a worldwide phenomenon whose presentation is similar in widely diverse cultures, especially when strict criteria are used in establishing diagnoses (Jablensky et al., 1992). The assumption is that this similar presentation reflects a common underlying disease process. Genetic studies of genetic isolates are based on this assumption.

The remote islands of Oceania, including the small islands of Micronesia in the western Pacific Ocean, have several populations that are of potential interest to psychiatric researchers. The islands

typically have populations of 10 000–50 000 individuals, with nearly half the population being under the age of 15. Schizophrenia is commonly found on most of the islands. What has not been established, however, is whether the illness found in Micronesia is identical to that found in most places around the world, or whether the very insularity which makes it a potentially interesting place for genetics research also has led to inbred anomalies.

Previous studies of these populations have raised questions about the nature of schizophrenia found throughout the islands of Micronesia. In 1981, Dale, the psychiatrist for the Trust Territories of the United States, which included all the Micronesian islands, reported significant differ-

* Tel: +1 303 399 8020; e-mail: merilyne.Waldo@uchsc.edu

ences in the prevalence rates of schizophrenia among very similar islands. In the islands of Palau, 7.6 per 1000 persons over the age of 15 had schizophrenia. In Yap, also a western Micronesian island, the prevalence was 9.7 per 1000. There were very low prevalence rates in the eastern islands of Micronesia; for example, in Kosrae the prevalence was 1.2 per 1000, and in the Marshall Islands, the most easterly of the islands, the prevalence rate was only 0.82 per 1000 (Dale, 1981). These compare with the 3.7 ± 2.2 per 1000 prevalence rate for subjects over age 15 in eight large studies worldwide reported by Eaton (1985).

Allan and Hunter, working in the Trust Territories from 1980 to 1983, reported a very strong bias towards male schizophrenics, 12 male referrals to 1 female referral (Allan and Hunter, 1984). Kauders et al. (1982) also noted a male bias of 4:1 on the island of Palau during a 3-month psychiatric rotation there in 1979–1980. These figures are at least double the 2:1 ratio found in developing countries in the WHO 10-country study (Jablensky et al., 1992).

These findings of significant differences between males and females were replicated in the 1988–1990 epidemiological survey by Hezel and Wylie (1992). They found a heavy bias towards males, with few female cases being reported in any of the islands, and no female cases at all in Kosrae, one of the most easterly of the islands. Unlike the previous studies of hospital referrals, they actively sought out cases in the community with the help of key informants, as well as using information from hospital mental health programs. Although their prevalence rates are significantly higher than those reported by Dale in 1981, their data also showed a strong trend towards a lower incidence in the eastern islands as compared with the western islands.

While demographic data are abundant, symptomatic data are available only for the Republic of Palau (Hammond et al., 1983). One of the authors had been part of the team responsible for providing the data reported in the Dale study. They found extensive substance abuse and related violence, as well as substantial affective symptomatology, especially in the male patients. In addition, they report that many of the patients were

originally referred for evaluation following incarceration in the Palauan jail for acts of violence. Palau had the one of the highest incidence rates of schizophrenia in both the Dale and the Hezel and Wylie surveys. However, the male to female ratio of 2.28:1 was the lowest in the Micronesian islands (Hezel and Wylie, 1992). Palau has the most rapidly developing economy in Micronesia.

In this report, we look at the patient population of Kosrae, one of the least developed islands. Hezel and Wylie reported a prevalence rate for schizophrenia of 6.4 per 1000 over the age of 15, which is slightly higher than the average prevalence rate of 3.7 ± 2.2 per 1000 worldwide (Eaton, 1985). However, Kosrae is the island with the greatest imbalance in the male to female ratio: it was found that there were no women with schizophrenia on Kosrae, although it was noted indicating that one woman with schizophrenia had lived there some 10 years previously. She had died by suicide. We wanted to look more carefully at the symptomatology of patients in Kosrae and to confirm the male predominance of schizophrenia there.

2. Methods

We compiled a list of known patients of Kosraean origin for diagnostic review. All patients currently identified by the Kosrae Substance Abuse and Mental Health Program were included, whether the individual was in treatment or not. We also sought out all of the individuals from Kosrae who made up the sample from Hezel and Wylie's report. These data were generously made available by the authors, and those individuals who were still alive were included in the list. A small number of reports from the Trust Territory administration, dating back to 1977, were also available, and these individuals were also included for evaluation. Community mental health workers in each of the municipalities were interviewed to identify any other individuals whose behavior was noted to be unusual. Since schizophrenia is known to cluster in families, pedigrees were evaluated. Special attention was paid to any females whose behavior suggested possible inclusion.

In total, 57 individuals were initially identified as (1) being current patients or potential patients in the Substance Abuse and Mental Health Program at Kosrae Hospital, (2) having been included in previous studies, or (3) having been identified by community workers. Except for one schizophrenic patient living in Guam, all individuals were living in Kosrae. Eight individuals with known psychiatric disorders (seven with schizophrenia, one with bipolar disorder), although long-term residents of Kosrae, were from neighboring islands, including a large family of Polynesian origin. Because we were interested in Kosraean symptomatology and incidence, these eight patients are not included in this report.

Hospital and mental health records were reviewed for each of the 49 Kosraeans. All whose records suggested any signs of psychosis at any time or those having chronic mental health problems were interviewed using the SCID-P (Spitzer et al., 1990); diagnoses were made using DSM-IV criteria. Twenty-eight people were interviewed. For 12 people, hospital records, family interviews, and mental health staff were able to provide sufficient information to confirm diagnoses. Nine people on the list were not approached for interviews. Six of these nine had been identified as having psychotic behavior in the community survey of Hezel and Wylie. None had ever been evaluated by mental health workers or been seen at the hospital for mental health reasons. The other three had been evaluated in the late 1970s, but had lived completely ordinary lives since that time. All nine were functional members of society, with no overt signs of distress. Each was personally well known to at least two local collaborators, and their normal functioning and behavior unanimously confirmed in each case. The hospital staff felt very strongly that asking these individuals to submit to a psychiatric interview, or to approach them in any way, would be a very unwelcome invasion of privacy. In small island communities, such as Kosrae, it is virtually impossible to ensure any confidentiality. Even a casual conversation with the mental health doctor is sufficient to raise potentially uncomfortable questions with family and friends. Thus, based on the available information, it was felt that these individuals could reliably be classified as 'normal'.

Four of the 49 patients had clear organic disorders, including two with epilepsy, one with a serious head injury, and one with leprosy. Further diagnostic evaluations of the epileptic patients were not possible, as they were not in adequate treatment for their primary disorders.

All interviews were completed in English, with a Kosraean translator assisting. Family members were included in the interviews whenever possible. Community mental health workers, who personally knew each of the individuals, reviewed and made notes on all individuals who were not personally seen by the author. Diagnoses are summarized in Table 1. Twenty-nine patients of the 49 suspected cases had psychotic disorders. Of the other 20, none had symptoms of psychosis. Nine carried no diagnosis at all.

3. Results

Twenty-two of the patients met DSM-IV (American Psychiatric Association, 1994) criteria for schizophrenia (Tables 1 and 2). Of these, 19 were male and only 3 were female. All had been ill for at least 3 years. This is a male to female ratio of 6.3:1 ($x = 11.64$, 1 df, $p < 0.001$), a remarkably unbalanced ratio, and consistent with earlier reports. When all schizophrenic patients residing in Kosrae are included (as was true of earlier reports), the ratio increases to 7.67:1. This male predominance cannot be accounted for by population statistics, since the male to female ratio in the 1994 census is 1.08:1.00. In addition, special

Table 1
Rediagnosis of previously identified patients using DSM-IV criteria

DSM-IV diagnosis	No. of patients
Psychotic disorders	
Bipolar I and II	5
Psychotic depression	2
Schizophrenia	22
Non-psychotic disorders	
Substance abuse	7
Organic disorders	4
No diagnosis suspected	9

Table 2
Schizophrenic patients in Kosrae

	Male (N=19)	Female (N=3)	Total (N=22)
Age of onset	20.6±4.4	22.7±2.5	20.9±4.0
Away from home at onset of illness	4	1	5
Family history of schizophrenia	18	3	21
Years of education	11.3±1.8	12.0±2.0	11.8±1.8

attempts were made to identify women in the community who might have mild forms of the disorder.

All but one of the patients had an extended family history of first- or second-degree relatives known to have schizophrenia. More than one-third of the patients were the direct descendants of the last king of Kosrae; two others were descendants of his sister. The majority of the others were from two large families. Matrilineal and patrilineal inheritance were equally common in this population.

The age of onset was younger in males than in females (Table 2), and was based on the earliest reports of disordered behavior. The ages must be considered approximations, since many patients were ill for months or sometimes years before being brought for treatment. Most had been in treatment, at least irregularly, since the onset of their disease. Two, however, had received only minimal treatment in the past. The oldest patient had not been treated for many years, despite continuing symptoms and serious personal neglect. Another patient had been in treatment for about 2 months some 20 years previously, and had only recently re-entered treatment when committed by civil action. Five of the patients were living away from their homes, four in school, when they first became ill.

Patients were initially brought to treatment for a variety of reasons. The most common reason was because of withdrawn behavior and mutism (eight patients). The next most common reason was because of violent behavior, including stabbing a girl, threatening to murder family members, and fighting. Four patients were brought in for bizarre behavior, including wandering the streets naked and disrupting church services; another three for

paranoid delusions. No information was available for one patient.

Substance abuse was common in these patients (Table 3). Half of the patients drank alcohol and smoked marijuana. Three of the patients had a serious drinking problem, and met DSM-IV criteria for alcohol dependence. Many of the patients used marijuana whenever it was available. Only one patient met criteria for cannabis dependence. Other drugs are not readily available on Kosrae. Twelve of the patients smoked cigarettes or used chewing tobacco. All 12 reported smoking whenever they could buy or beg a cigarette. The three female patients did not smoke marijuana or cigarettes, and only one had ever used alcohol.

All of the patients had a history of auditory hallucinations. The hallucinations were typical of the sorts recorded elsewhere—usually several voices conversing, often about the individual. Nearly half also had visual hallucinations, including strangers wandering in the village and alien creatures bringing messages from space. In addition, 10 of the patients reported seeing ghosts. Seeing ghosts is a culturally accepted experience,

Table 3
Substance abuse and clinical symptoms in Kosraean schizophrenics

	No	Yes	Unknown
Use of alcohol	11	11	
Use of marijuana	11	11	
Use of nicotine	10	12	
Auditory hallucinations		22	
Visual hallucinations	10	11	1
Sees ghosts	11	10	1
History of violence	13	9	
Paranoid delusions	9	12	1
Elective mutism	3	19	

and was not considered a symptom of psychosis in this sample.

Nine of the patients had a history of violence, and many had spent time imprisoned for this behavior. Agitation and a return of violent behavior was common when patients stopped taking their medications and when using alcohol. It was also common when levels of medication were sub-therapeutic. Twelve of the patients also had paranoid delusions. The delusions were often of the sort seen in Western populations—of people following them, of families having been replaced by aliens or strangers, of being threatened, of communicating with the devil, and of being the spokesperson for world leaders.

The most unusual feature of the schizophrenics in Kosrae was that 19 had episodes of selective mutism. In this population of adult schizophrenics, mutism was not a late symptom in the disorder, but often the earliest sign of breakdown. Nor was it accompanied by any other signs of catatonic behavior, which it usually is in schizophrenia. One man spent most of 20 years mute. Another woman was mute for 3 years. During these times, patients were often quite willing to communicate using facial expression, such as raising eyebrows. The mutism resolved rapidly with the institution of adequate levels of medication, most commonly fluphenazine decanoate, 25 mg every 3 weeks. In the case of the young woman who did not speak for nearly 3 years, 2 mg of haloperidol for 4 days was enough to significantly alleviate this symptom. After 3 months of treatment with 25 mg of depot fluphenazine every 3 weeks, speech was returning to the man who had not spoken in 20 years; however, it was limited and primarily in response to direct questions. Mutism was one of the first signs of impending breakdown and useful in identifying patients who had become non-compliant with treatment.

The prevalence rate for schizophrenia in Kosrae is 6.8 per 1000 individuals over the age of 15 (4.0 per 1000 of the total population). This figure is based on the extended population of patients, including those schizophrenics of non-Kosraean origin. We have calculated it this way to have figures comparable to earlier reports, as none have adjusted for possible inclusion of patients origi-

nally from other islands. This incidence rate is significantly higher than the 1.2 per 1000 reported by Dale (1981), and very similar to the 6.4 per 1000 reported by Hezel and Wylie (1992). When we include in this figure the additional eight people with affective psychosis, in order to compare it directly with Hezel and Wylie's population of individuals with *psychotic* disorder, the prevalence rate increases to 8.7 per 1000. These are conservative figures, since it remains possible that some patients remain hidden by families. However, virtually all individuals living on the island are personally known, or related to, someone in the mental health program, and thus it is unlikely that any patients with serious disorders would escape detection.

4. Discussion

The results of this survey confirm earlier reports of a strikingly high morbidity risk for males as compared to the females in Kosrae. There is, to date, no adequate explanation why Micronesian males are at so much higher risk than females for psychotic illnesses. Previous suggestions of increased male vulnerability and stress due to rapid shifting of cultural roles (Hezel and Wylie, 1992) do not take into account the fact that the ratios in Kosrae, one of the less developed Micronesian islands, are significantly higher than the ratios, for example, found in Palau, where westernization is proceeding at a far more rapid rate. Kosrae continues to have limited access to the West, with no television or radio. Regular airline service began only about 20 years ago, and is still only several days a week. Only recently have videos been available on the island. Communication by computer via the internet had been available for less than 1 year when the survey was completed (Spring of 1997). Traditional culture was replaced some 100 years ago by missionaries, and the new tradition is strong and guides much of life in Kosrae. The church is very important in private and public life. For example, no planes are allowed to land in Kosrae on Sundays, in order not to break the Sabbath.

It remains true that substance abuse is an almost exclusively masculine problem (Micronesian Seminar, 1997. Alcohol and drug use in the Federated States of Micronesia. Unpublished manuscript), and it is possible that the aggravated symptomatology and aggression that accompany the use of alcohol may contribute to the identification of male patients. However, more than half of our population was first brought for evaluation because of withdrawn behavior and selective mutism. It is possible that these are culturally more accepted behaviors in women and that it is only when other more florid symptoms emerge that women are identified as ill. The reverse may also be true—that males who withdraw and become quiet without reason are culturally unusual.

The high incidence of selective mutism is an unusual and unexpected finding in this population. Mutism is now very rare in developed countries, and is usually accompanied by other catatonic symptoms in adults (Jablensky et al., 1992). It was not uncommon in schizophrenic patients before the widespread use of psychotropic medication, and was found to be the primary clinical correlate of the length of untreated illness in patients (Waddington et al., 1995). It has also been associated with cognitive dysfunction and deteriorating function (Waddington et al., 1987), and with paranoia (Altshuler et al., 1986). In the patients in Kosrae, however, it is often one of the initial signs of illness. This symptom, though occasionally encountered, is not common in other Micronesian islands (personal observation).

The literature on mutism suggests several forms, which may stem from very different causes. In schizophrenics, it is sometimes seen as the most severe form of poverty of speech, and thus a significant negative symptom (Iager et al., 1985; Andreasen and Flaum, 1991; Fenton and McGlashan, 1992). It is also commonly associated with catatonia, itself now rarely reported. Of 22 cases of mutism reported by Altshuler et al. (1986), only one paranoid schizophrenic male, age 35 with no previous episodes of mutism, did not have accompanying signs of catatonia. In children and adolescents, however, it is often seen as a severe form of anxiety, especially social anxiety (Black

and Uhde, 1992; Dummit et al., 1997). The patients in our population do not otherwise have the usual symptoms of severe negative schizophrenia, they did not exhibit severe paranoia, nor were there any other signs of catatonia. In addition, the often rapid and complete resolution of their mutism with psychotropic medication is not common for negative symptoms. Except for a single report of a mute and anxious adolescent developing schizophrenia (Eldar et al., 1985), there do not seem to be reports of other populations with this early and intermittent form of mutism associated with schizophrenia.

It is possible that this symptom is, in part, culturally influenced. In Kosrae, silence plays a significant role in the society as a response to anger and disagreement. Otherwise-normal individuals will often remove themselves for days at a time and refuse to speak, thus adopting selective mutism as an alternative to more aggressive forms of communication (Williams, personal communication, 1997). However, none of the patients whose mutism resolved with medication were known to resort to the use of silence for communication purposes.

Despite these unusual features of schizophrenia, the patients in Kosrae resemble their Western counterparts in most ways. The many symptoms of florid psychosis as well as negative symptoms are similar, and the content of their delusions often identical. They respond well to the same medications and to similar treatment regimes as in the West. Further study of the population, especially the women, may help to shed light on the differences.

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