

HIRADO: TEMPORAL TRENDS IN INBREEDING AND FERTILITY*

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Until recently, it has been commonplace both to deplore the lack of realism in most, if not all, genetic models used in the study of human populations and to acknowledge that few data existed from which improvements might stem. Interest and energy have, however, begun to focus upon the collection of data which hopefully will serve as bases for the formulation of more insightful and presumably more realistic models. Some investigators have, for example, turned to routinely collected vital and demographic data on the supposition that these observations, if pertinent, would be especially useful because of their numbers, breadth, and depth in time. Thus far, for a variety of reasons, the results have not lived up to expectations. Other investigators have tended to emphasize *ad hoc* censuses. They argue that demographic units are unfortunately more apt to be defined by geographic, political, or socioeconomic considerations than biologic ones, and though this bias can be offset to some degree in specific instances by judicious selection of the units to be studied, an *ad hoc* census of some variety may still be necessary if the routine data are to be fully plumbed. The Hirado Health Survey, to be described shortly, was an effort to combine an *ad hoc* census with routinely collected vital and demographic observations; it was directed, broadly stated, toward the ascertainment of (1) the fertility of the consanguineous marriage, (2) the reproductive performance of the inbred individual, and (3) certain observations on the children born to parents, especially mothers, who are themselves the products of consanguineous marriages. The study itself represents a conscious attempt to incorporate into traditional demographic and census procedures a few readily measurable genetic parameters. Inbreeding is, in Japan, such a parameter, and its relationship to fertility and fecundity may be one of the main avenues through which we can discern and measure the contribution of genetic factors to reproduction.

Numerous considerations, administrative and scientific, entered into the choice of Hirado and the design and implementation of the survey. To mount the kind of demographic study implicit in the objectives just enumerated requires a population of a size sufficient to demonstrate differences in fertility among consanguinity classes, if differences exist, and yet not so large that the population would be inordinately difficult to study. Clearly, the population must be inbreeding at a rate such that a census of persons and households would reveal a substantial number of inbred individuals and consanguineous marriages. Ideally, the population would be sharply delimited geographically, and as static as possible. Moreover, it would seem advantageous in view of the effect of socioeconomic factors upon fertility to select a population as homogeneous in this regard as possible. Finally, if the census observations were to supplement routinely collected data, as proposed, the population to be censused had to correspond to some Japanese administrative unit upon which vital and demo-

graphic data are regularly collected and published. Of the several possible such units, the *shi*, the administrative city, seemed most appropriate. These various considerations along with the desire to develop a study which could be implemented with a relatively small amount of manpower led us to consider island populations. Several of Japan's islands possess this minimal set of requirements, and from among these, Hirado was selected.

Hirado-jima, some 169 kilometers in circuit, lies to the west of Kyushu in the neighborhood of the 33rd parallel. Archaeological evidence attests that the island, or at least portions of it, has been inhabited from the Yayoi era, and possibly earlier. It has played a prominent role in the history of western Japan, first in the defense of Kyushu during Kublai Khan's abortive 13th-century invasion, and subsequently as a commercial center in Japan's pre-Meiji contacts with the Dutch, English, and Portuguese. It has for some eight centuries past been the center of the Matsuura *han*, one of the smaller fiefdoms of north-western Kyushu. In 1954, Hirado-shi was formed from Hirado-jima and a dozen or so small adjacent islands, of which only two, Takashima and Takushima, are inhabited.

The Census.—In June 1964, a consortium of five universities, namely, Juntendo University, Kyoto Prefectural University of Medicine, Kyushu University, Tokyo Medical and Dental University, and the University of Michigan, initiated a census of Hirado-shi which was completed some three months later. The census takers were either midwives or public health nurses, and occasionally physicians associated with one or another of the previously mentioned educational institutions. All individuals then resident on the island or normally there were enumerated; the basic unit of enumeration was the household. Information was obtained on such items as birth date, birth place, education, occupation, religion, date of marriage (if married), consanguinity, number of persons in the household, the size of the house, diet, income, land holdings, attitudes toward reproduction, and a pregnancy-by-pregnancy listing of the reproductive performances of the husband and wife. The data obtained by interview were regularly compared with existing records of the local *koseki-ka*, the Public Health Office, the Tax Office, and the Agricultural and Fishing Cooperatives. We are obviously deeply indebted to many levels of civil government and many persons without whose uncompromising efforts this study would not have been possible. Brief as this account is, it should be patent that time permits only a limited and preliminary presentation of the data. We have, therefore, elected to emphasize the population and the opportunities it affords to probe some of the biocultural complexities associated with reproduction rather than dwell upon results which are, as yet, tentative.

The Population.—At the time of our census, Hirado was thought to have a population in excess of 40,000 persons (the 1960 Japanese census had revealed 40,881 inhabitants) distributed over some 8,200 households. These individuals resided in 116 *ku* which, in turn, were organized into 60 hamlets, villages, and towns varying in size from a few hundred persons to several thousands. Like most predominantly rural areas of Japan, the population of Hirado is slowly declining; the young find the urban areas more attractive or at least offering more

opportunities, and changing agricultural practices make their presence on the farms no longer essential. These forces, coupled with the active, rural recruitment of employees by some Japanese employers, have accelerated Japan's urbanization, a trend which began before the turn of this century.

In 1960, Hirado's natural rate of increase per 1,000 population was 11.0. This was somewhat greater than the average of Nagasaki, the prefecture of which Hirado is a part, which itself ranked eighth in this respect among the 46 prefectures of Japan. In that same year, Hirado's crude fertility and mortality rates were appreciably higher than the national average. The live birth and death rates were 21.1 and 10.1 per 1,000 population, respectively, as contrasted with 17.2 and 7.6 (for the national figures, see ref. 1). In the immediately preceding five years, the birth rate had fallen by some 6.2 births per 1,000 population whereas in Japan as a whole the decline had been less precipitous, only 2.2 births. Mortality rates in this same interval of time had not changed appreciably either on Hirado or in the nation.

Hirado's population is primarily Buddhist, but it also embraces sizeable groups of Roman Catholics and *Kakure Kirishitan*; the latter are members of a syncretic religion incorporating elements of Buddhism, Catholicism, and Shintoism. While a surprising degree of occupational diversity exists, farming and fishing account for the livelihoods of more than half the island's residents. Relatively more Catholics (62.3%) and *Kakure* (75.5%) are farmers than Buddhists (37.7%), and the latter are, on the average, the more prosperous.

Marriage Practices.—Traditionally, marriages in Japan have been arranged by the parents of the bride and groom, and were destined to serve the social, financial, and other needs of the family and only secondarily the interests of the bride and groom (a more thorough exposition of traditional marriage practices will be found in ref. 2). This is still the pattern of marriages on Hirado. Mate selection is, however, further complicated by religion and propinquity, biologic as well as geographic.

Each of the religious groups to which reference has previously been made is more or less socially isolated from the others, and relatively few mixed marriages occur. In fact, among 10,379 marriages where the spouses reported themselves to be Buddhist, Catholic, *Kakure*, or Shinto, in only 97 marriages were husband and wife of dissimilar religions and of these latter, almost half, 48 to be exact, were Buddhist-Shinto combinations. As judged by Catholic parish records, this social isolation appears to have been the pattern for most of the last century at least. Aside from this proclivity to marry within religious groups, there is a strong endogamous propensity within virtually all villages. Thus, among 10,422 marriages where one or both spouses were still alive and residing on Hirado in 1964 and where the birthplaces of both spouses were known, in 3,901 instances (37.4%) husband and wife were from the same village, and in another 1,494 cases (14.3%) they were from different but adjacent villages. Of the 20,844 individuals involved in these marriages no less than 16,816 were born on the island. There are interesting differences among the religious groups insofar as the choice of spouses is concerned—the *Kakure* are the most endogamous at the village level (78.9%), the Catholics the least (29.9%); but relatively more Buddhists

(20.3%) obtain their spouses from places other than Hirado than do either Catholics (18.5%) or *Kakure* (2.9%).

Of possibly greater inherent biologic interest are the kinds and frequencies of consanguineous marriages encountered on Hirado among the three religious groups. These data are presented in Table 1. We note, first, that almost 15 per cent of the marriages are known to be consanguineous. Second, within the religious groups, the frequencies of all consanguineous marriages follow the same pattern as endogamy, that is to say, the highest frequency of consanguineous marriages is among the *Kakure* who are, it will be recalled, the most endogamous at the village level. There are other somewhat more subtle differences among the groups. Thus, for example, the bulk of consanguineous marriages among the Catholics involve second cousins whereas in the other two groups first cousins are the most common. These differences are more apparent in the mean coefficients of inbreeding, which are 0.00484, 0.00150, and 0.00965 for the Buddhists, Catholics, and *Kakure*, respectively. The differences between the Catholics and the other two groups are no less striking if attention is restricted to farmers; the mean coefficients of inbreeding are now 0.00176 for the Catholics and 0.00643 and 0.01042 for the Buddhists and *Kakure*. Considerable vari-

TABLE 1. *Numbers of consanguineous marriages on Hirado by decade of marriage, religious preference of husband, and relationship of spouses.*

Decade	Relationship of Spouses				Unrelated	Total marriages	Proportion consanguineous
	First cousins	1½ cousins	Second cousins	Other			
<i>Buddhist</i>							
1900	18	5	2	3	315	343	0.0816
1910	71	25	25	8	763	892	0.1446
1920	95	23	43	36	1,050	1,247	0.1580
1930	61	26	34	39	1,113	1,273	0.1257
1940	131	47	72	143	1,581	1,974	0.1991
1950	69	29	34	61	1,474	1,667	0.1158
1960	11	9	16	36	620	692	0.1041
Total	456	164	226	326	6,916	8,088	0.1449
<i>Roman Catholic</i>							
1900	0	1	0	2	43	46	0.0652
1910	2	2	4	7	123	138	0.1087
1920	0	2	12	1	164	179	0.0838
1930	2	1	6	1	168	178	0.0562
1940	3	1	19	3	290	316	0.0823
1950	0	3	20	2	245	270	0.0926
1960	0	1	5	0	80	86	0.0698
Total	7	11	66	16	1,113	1,213	0.0824
<i>Kakure</i>							
1900	11	0	1	0	28	40	0.3000
1910	16	2	1	3	84	106	0.2075
1920	9	4	9	6	98	126	0.2222
1930	13	5	8	12	76	114	0.3333
1940	19	7	6	18	109	159	0.3145
1950	15	2	3	21	113	154	0.2662
1960	4	2	4	4	34	48	0.2917
Total	87	22	32	64	542	747	0.2744
Grand total	550	197	324	406	8,571	10,048	0.1470

ability exists between Buddhist villages; one encounters mean coefficients as low as 0.00257 ("Hirado town") and as high as 0.01135 (Takushima).

One naturally wonders whether these differences among the religious groups are of recent origin, that is, whether they may have arisen in the present century. This, however, appears not to be the case. In Table 1 will be found the frequencies of the various kinds of consanguineous marriages by religious group and the decade in which marriage occurred. Note that within all three religious groups there is no evidence that the frequency of marriages involving relatives is on the decline. This has also been observed in other rural areas of Kyushu and Japan,^{3, 4} but there is some evidence that the frequency of consanguineous marriages is declining in urban areas.⁵

Reproductive Performance.—Some years ago, we presented data which suggested, firstly, that consanguineous unions were less often childless than non-consanguineous ones; secondly, that the average number of liveborn children ever born was greater for the consanguineous than the nonconsanguineous mating; and, finally, that the number of children reaching adulthood differed less between these two types of marriages than might otherwise be expected because of the higher mortality among the inbred offspring.⁶ These data were sparse, and no one of the effects mentioned could be shown to be statistically significant. The study of Hirado was in large part prompted by these findings, for it was presumed that a larger population would either refute or reinforce them. It should be pointed out, perhaps, that there were and remain prior arguments which make these particular findings reasonable. Thus, for example, if immunologic incompatibility contributes either to infertility or to the very early loss of embryos, the burden imposed by such incompatibility would be expected to be less in the consanguineous marriage, because of the genetic correlation between the spouses, than in the nonconsanguineous marriage.

Analysis of reproductive performance is fraught with numerous pitfalls, and complicated by an even greater number of factors, many of a nonbiological nature, which can and do contribute to the size of a given family. Among these, to mention but a few, are education, occupation, income, religion, and age at marriage. These extraneous sources of variation could readily obscure an effect of consanguinity or create a spurious one, but their control is a formidable task. One can attempt to effect this control either through the choice of the comparison groups, or through covariance analysis, or both. We propose the first of these alternatives for this presentation not because it is necessarily the best, but rather because it exposes one of the strengths of the study, namely, the opportunity to extract relatively large groups of individuals or marriages alike with respect to several important concomitant variables. Here, reproductive performances of the consanguineous and nonconsanguineous marriages will be contrasted within fixed decades of marriage for specified occupations and religions. Thus, the contributions of three major concomitant variables—religion, occupation, and years of marriage—can be readily controlled. While this will not necessarily remove all extraneous variation, the two occupational groups which have been selected, namely, farmers and fishermen, are fairly homogeneous with regard to education but less so to income. As previously pointed out,

these two occupations account for the livelihoods of over 50 per cent of Hirado's inhabitants. Attention will be confined to those marriages contracted in the 1910's, 1920's, 1930's, and 1940's. In the first three instances, reproduction would have been completed in the vast majority of families prior to the census; this is not necessarily true, of course, for those marriages in the latter half of the forties. In all, we shall consider four occupational-religious groups, namely, Buddhist farmers, Catholic farmers, *Kakure* farmers, and Buddhist fishermen. The Catholic and *Kakure* fishing groups are too small to warrant analysis; the former consists of only 35 marriages (2 related) and the latter 39 (9 related).

As a preamble to the data, a few remarks directed toward the attitudes of the Hirado *toimin* to reproduction seem appropriate. Many of you will be aware that in 1948 Japan legalized the artificial interruption of a pregnancy if it constituted a threat to the mother's health, worked an undue economic hardship upon the family, or was likely to terminate in a child who would be burdensome to society. The Eugenics Protection Act was frankly designed to control Japan's population growth.⁷ There is no question that this Act effected a dramatic change in Japan's birth rate; live births fell from 33.5 per 1,000 in 1948 to 18.0 a decade later. Acceptance of this means of family limitation has been more widespread in the urban than in the rural areas, but its effects are even now being felt on Hirado. Thus, when asked whether they approved of the artificial interruption of a pregnancy, some 9.6 per cent of Hirado Buddhists asserted they did, 2.8 per cent of the *Kakure* concurred in this opinion, but there was little acceptance among the Catholics, only 0.6 per cent. As might have been anticipated, acceptability was related to the decade of marriage and hence to the ages of the spouses. Among Buddhists, for example, only 4.6 per cent of those couples married in the first decade of this century felt that the artificial interruption of a pregnancy was proper, but 12.2 per cent of those married in the 1950's did. These observations are relevant here, however, only insofar as they may disclose more general attitudes of the individuals married in the 1910's to 1940's toward family limitation; save for the marriages of the forties, few of these couples could have availed themselves of the provisions of the new law. Possibly more closely related to individual reproductive performance than attitude toward artificial interruption of pregnancies is the size of family judged desirable. Here we find that among farmers the Catholics desire the larger families, 4.86 children, on the average, than the *Kakure*, 4.33, or the Buddhists, 4.09. Within Buddhists, the only group where this contrast is possible, there is no difference between farmers (4.09) and fishermen (4.12) in mean desirable family size. Of greater interest still is the fact that within all four groups previously cited, there is no significant difference between related and unrelated marriages in mean desirable family size. There is, then, no conspicuous reason to believe that contrasts within religions and occupations will confound differences in attitudes toward family limitation which might be reflected in actual family sizes.

Among the four groups, if childless marriages are summed over all four decades, the frequency of these unions among related and unrelated marriages is found to be 9.66 per cent (unrelated) as contrasted with 5.54 per cent (related) for

TABLE 2. *Some estimates of the reproductive performance of four religious and occupational groups on Hirado.*

Decade	Total families	Total live births	Mean live births	Variances in live births	Total deaths*	Proportion dying*	Mean survivors
<i>Buddhist farmers</i>							
Spouses unrelated to one another:							
1910	355	1,984	5.59	10.80	391	0.1970	4.49
1920	452	2,604	5.76	10.76	443	0.1701	4.77
1930	371	1,775	4.78	6.99	279	0.1571	4.03
1940	509	1,888	3.71	3.79	191	0.1011	3.34
Spouses related to one another:							
1910	77	505	6.56	10.64	123	0.2436	4.96
1920	89	536	6.02	8.54	109	0.2034	4.80
1930	62	360	5.81	7.68	81	0.2250	4.50
1940	102	392	3.84	2.90	38	0.0969	3.47
<i>Catholic farmers</i>							
Spouses unrelated to one another:							
1910	87	702	8.07	7.51	115	0.1638	6.75
1920	119	846	7.11	11.05	117	0.1382	6.13
1930	96	620	6.46	9.24	74	0.1193	5.69
1940	173	988	5.71	6.16	87	0.0880	5.21
Spouses related to one another:							
1910	6	36	6.00	7.36	7	0.1944	4.83
1920	11	95	8.64	3.66	13	0.1368	7.45
1930	5	36	7.20	3.70	6	0.1667	6.00
1940	20	114	5.70	7.30	17	0.1491	4.85
<i>Kakure farmers</i>							
Spouses unrelated to one another:							
1910	65	337	5.18	10.12	58	0.1721	4.29
1920	77	441	5.73	8.89	82	0.1859	4.66
1930	63	297	4.71	6.85	43	0.1447	4.03
1940	80	344	4.30	4.21	54	0.1569	3.63
Spouses related to one another:							
1910	15	73	4.87	10.57	20	0.2740	3.53
1920	18	120	6.67	11.73	25	0.2083	5.28
1930	22	117	5.32	4.60	19	0.1624	4.45
1940	26	110	4.23	2.80	6	0.0545	4.00
<i>Buddhist fishermen</i>							
Spouses unrelated to one another:							
1910	89	566	6.36	13.07	151	0.2667	4.66
1920	142	868	6.11	11.22	152	0.1751	5.04
1930	143	704	4.92	9.56	111	0.1576	4.15
1940	265	1,029	3.88	4.70	105	0.1020	3.49
Spouses related to one another:							
1910	14	80	5.71	12.50	18	0.2250	4.42
1920	25	192	7.68	9.42	43	0.2240	5.96
1930	20	105	5.25	5.68	18	0.1714	4.35
1940	64	294	4.59	3.02	23	0.0782	4.23

* Only nonaccidental deaths prior to age 21 are included.

Buddhist farmers, 4.84 per cent with 6.00 per cent for Catholic farmers, 8.07 per cent with 6.09 per cent for *Kakure* farmers, and 10.49 per cent with 4.91 per cent for Buddhist fishermen. Of these four contrasts, the first and the last, i.e., the two occupational comparisons within the Buddhists, are statistically significant, one at the 5 per cent level and the other at the 1. Both contrasts, it will be noted, indicate childless marriages to be approximately half as frequent if the spouses are related than if they are not related. In this respect, then, the Hirado data support those from Kuroshima mentioned earlier. It should be noted that sociologic, as well as biologic, explanations can be adduced for this finding. Thus, for example, we know that consanguineous marriages in Japan more frequently involve first than subsequent sons, and that upon first sons a heavy responsibility to and for the family exists. It is conceivable that this added responsibility is reflected in reproductive performance, and specifically in a compulsion to have more children, on the average, to ensure perpetuation of the family. This thesis can and will be tested.

Table 2 presents some estimates of the reproductive performances of individuals belonging to the four religious-occupational groups. It should be noted that these data are restricted to live births and to live born children dying prior to their twenty-first birthdays. Most of the offspring of the marriages contracted in the 1910's and 1920's will, of course, have passed through this period of risk of death, but not those offspring of the marriages of the thirties and forties. With regard to the former children, their cumulative mortality rates agree closely with the *Third to Sixth Japanese Life Tables*, those derived from these years. Be this as it may, we note that among Buddhist farmers in every decade more liveborn children have been born to consanguineous marriages, on the average, than nonconsanguineous ones. But as on Kuroshima the proportion of liveborn children dying is greater among the inbred than the non-inbred children, and the "net" reproductive advantage is less accordingly. This same tendency is to be observed in the three other groups although the Catholics provide relatively little information, so few are the consanguineous marriages. Of the 16 possible comparisons of pairs of means, the only ones to emerge significant involve comparisons where the mean number of inbred, liveborn children exceeded the mean number of noninbred and liveborn offspring. No obvious differences related to consanguinity emerge from inspection of the variances in numbers of live births; however, it is worth noting that the weighted average, over decades, of the variances in number of live births to consanguineous marriages is smaller than the comparable statistic for nonconsanguineous marriages. This suggests that the differences in means are not ascribable to a few "exceptional" marriages but reflect a more general reproductive pattern. If these differences are accepted at face value, and if, in truth, consanguineous marriages are on the decline, there arise interesting implications with respect to the maintenance of genetic variability in populations such as Hirado. Not the least of these is a continuing erosion of the genetic variance.

What interpretation, now, is to be placed on these data? What insight do they afford into the genetic control of fertility and fecundity? Do they support the notion that the increased mortality commonly associated with inbreeding

may, in human populations, be largely or wholly offset by conscious replacement of the deceased children? What are their implications with respect to the estimation of the genetic burden of disease and disability resident in human populations? These and numerous other questions come to mind. However, pursuit of answers is dampened somewhat by the uncertainties which still must be attached to the findings themselves. There remains the possibility that systematic differences exist between consanguineously and nonconsanguineously married spouses. More can be done to establish whether this is or is not true. Thus, one can attempt to remove the residual variability in income, for example, by covariance analysis, and we propose to do this. There remain almost again as many marriages of individuals in other occupations to be examined. And, finally, definitive analysis will seek to determine whether family size varies directly with the degree of relationship between the spouses. Mortality is known to vary so.

At a time when many thoughtful citizens are concerned that man's numbers will outstrip his resources, it seems that any information which enhances our understanding of the genetic component of reproduction will contribute to the evaluation of the consequences of population control. It would appear self-evident, however, that the proper evaluation of this component will require all of the skill that demographers and population biologists can muster, so numerous are the possible sources of obfuscation. Multidisciplinary or at least multiple-variable approaches must hold the field until such time as the interrelationships between the biologic, cultural, and socioeconomic domains are more clearly exposed than now is the case. *Ad hoc* censuses, such as the one here described, can contribute mightily to this latter task.

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