FAMILIAL ACCUMULATION OF CONSANGUINEOUS MARRIAGES¹

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Summary Distribution of consanguineous marriages among spouses, husband's parents and wife's parents was analyzed based upon data on 20,563 families in six populations in Japan. There were highly significant correlations of presence or absence of consanguinity among the three couples in most populations. That is, products of consanguineous marriages tend to marry each other and a product of consanguineous marriage tend to marry a relative. Such familial accumulation of consanguineous marriages may lead to a higher inbreeding level in the offspring of consanguineous unions than that estimated from parental relationship only. Moreover, maternal inbreeding effects may overlap the consanguinity effects. Therefore, the effects of consanguinity may be overestimated, if familial accumulation of consanguineous marriages is ignored.

INTRODUCTION

Most studies on the effects on consanguineous marriages in man have been carried out with no regard to consanguinity in the previous generations. Though kindreds with two or more related couples are occasionally encountered, few studies have clarified whether or not such observed familial accumulation of consanguineous marriages are coincidental. The present communication deals with the results of analyses of data on consanguinity in Japanese populations which reveals non-random distribution of consanguineous marriages among families.

MATERIALS

Data on consanguineous marriages in spouses, husband's parents and wife's parents were obtained from following reports.

1) A study of the effects of inbreeding on fertility (Tanaka et al., 1964).

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The materials were composed of all couples cohabiting for five years or more and residing in two adjacent rural communities in the southern district of Fukuoka Prefecture. One, denoted as K population, was surveyed by Furusho, and the other denoted as H population, by Yanase and collaborators.

2) A study of isolated population (Yanase, 1966).

Data on two isolated populations, Hs in Fukuoka Prefecture and Hi in Tokyo, were cited from this report. The samples of H and Hs came from the same source but were analyzed individually, since they were not entirely identical; *i. e.* H sample was composed of the couples cohabiting for five years or more, and Hs was composed of parents of children born during the years 1943–1963. Yanase divided non-consanguineous couples into a native, endogamous group and a non-native, exogamous group. Most following analyses were carried out under this classification.

3) A study on the effects of maternal inbreeding on prenatal deaths (Tanaka et al., 1967).

The materials had been obtained by Furusho and his colleagues in a community in the central district of Kumamoto Prefecture (denoted as U population).

4) A study of the effects of parental consanguinity and inbreeding on stillbirths and prereproductive mortality (Schull *et al.*, 1970).

The data were collected from all couples residing in Hirado Island in Nagasaki Prefecture.

5) A study of consanguinity in Fukuoka city (Nakamoto, 1969).

The data were composed of all consanguineous couples whose wives were 30–50 years of age, residing in the central district of Fukuoka city, and non-consanguineous couples matched in age and certain other variables with the consanguineous ones.

Thus, data on 20,563 families in total were analyzed from various angles.

RESULTS

The distributions of consanguineous marriages in spouses, husband's parents and wife's parents are listed in Table 1.

1) Correlation of consanguineous marriages between spouses and their parents.

Each sample devided into two groups according to whether the spouses are related or not, and the rates of consanguineous marriages in husband's parents and in wife's parents were compared between the two groups (Table 2). The consanguinity rates in husband's parents and in wife's parents are higher in the group of related spouses than in that of not-related spouses in all but Hi populations. The difference are significant in 10 of the 14 comparisons, mostly at high levels.

Next, the rate of consanguineous marriages between spouses were compared among four groups by consanguinity between husband's parents and between wife's parents (Table 3). In all but Hi populations, the rate is the lowest in group (0)-(0), in which husband's parents and wife's parents are both unrelated, followed by (+)- (0) and (0)-(+), and the highest in (+)-(+), in which the rate is thrice or more as high as that in (0)-(0), and the differences are highly significant.

In all the populations the rate of consanguineous marriages between spouses is higher in group (0)-(+) than in group (+)-(0).

2) Correlation of consanguinity between husband's parents and wife's parents.

Families were devided into two groups according to whether spouses are related or not, then in each group the observed frequencies of four combinations (0)-(0), (+)-(0), (0)-(+), and (+)-(+) by consanguinity in husband's parents and wife's parents were compared with the frequencies expected upon the supposition that consanguinity between husband's parents are independent from that between wife's parents, using consanguinity rates set out in Table 2. All the populations except Hi show a similar trend, *i.e.* the observed frequencies is higher than the expected in groups (0)-(0) and (+)-(+), while the observed is lower than the expected in groups (+)-(0)

Popula-	Spouses, related (+)	Hust	Total			
tion	or not (0)	(0)-(0)	(+)-(0)	(0)-(+)	(+)-(+)	•
	(+)	81	10	13	15	119
К	(0)	340	25	31	12	408
	Total	421	35	44	27	527
	(+)	158	27	26	17	228
н	(0)	764	82	51	11	908
	Total	922	109	77	28	1,136
<u></u>	(+)	122	25	20	14	181
Hs	(n) ∫ N	365	64	52	12	493
	(⁽⁾⁾ (Q	355	39	5	0	399
	Total	842	128	77	26	1,073
	(+)	131	18	23	2	174
Hi	$(\mathbf{n}) \in \mathbf{N}$	336	46	46	13	441
	(⁽⁰⁾) Q	202	29	3	1	235
	Total	669	93	72	16	850
	(+)	96	19	13	8	136
U	(0)	569	95	53	11	728
	Total	665	114	66	19	864
	(+)	1,124	153	173	48	1,498
Hirado	(0)	7,433	624	586	83	8,726
	Total	8,557	777	759	131	10,224
	(+)	2,282	117	131	11	2,541
Fukuoka	(0)	3,066	115	160	7	3,348
	Total	5,348	232	291	18	5,889

 Table 1. Distribution of consanguineous marriages among spouses, husband's parents and wife's parents.

N and Q are respectively the native, endogamous group and non-native, exogamous group.

Popula-	Consanguineous	· • •	When spouses are			
tion	marriage between	related	not-related	total	χ^2	ϕ
K	Husband's parents	0.2101	0.0907	0.1176	12.65***	0.1549
	Wife's parents	0.2353	0.1054	0.1347	13.34***	0.1591
	Spouses		· · · · · ·	0.2258	·	
	Husband's parents	0.1930	0.1024	0.1206	14.09***	0.1114
Н	Wife's parents	0.1886	0.0683	0.0924	31.45***	0.1664
	Spouses	·		0.2007		
	Husband's parents	0.2155	0.1289	0.1435	9.17**	0.0924
			(N 0.1543)			
			\Q 0.0977/			
Hs	Wife's parents	0.1878	0.0774	0.0960	21.17***	0.1405
			(N 0.1298)			
			Q 0.0125/			
	Spouses		•	0.1687		—
	Husband's parents	0.1149	0.1317	0.1282	0.35	-0.0202
			(N 0.1338)			
			Q 0.1277/			
Hi	Wife's parents	0.1437	0.0932	0.1035	3.80	0.0669
			(N 0.1338)			
			\Q 0.0170/			
	Spouses	·		0.2047		
	Husband's parents	0.1985	0.1456	0.1539	2.46	0.0534
U	Wife's parents	0.1544	0.0879	0.0984	5.71*	0.0813
	Spouses		<u> </u>	0.1574		
	Husband's parents	0.1342	0.0810	0.0888	44.64***	0.0661
Hirado	Wife's parents	0.1475	0.0767	0.0871	80.78***	0.0889
	Spouses			0.1465		
	Husband's parents	0.0504	0.0364	0.0425	6.90**	0.0342
Fukuoka	Wife's parents	0.0559	0.0499	0.0525	1.05	0.0133
	Spouses			0.4351		

 Table 2. Rate of consanguineous marriage and correlation of consanguinity between spouses and husband's parents or wife's parents.

Significance level of difference between the related and non-related groups is shown by (5%), (1%) and (1%).

and (0)-(+) (Table 4). χ^2 -test revealed that consanguinity in husband's parents and that in wife's parents were significantly correlated in most populations (Table 5).

3) Coefficient of parentage in spouses in groups classified by parental consanguinity.

Mean coefficients of parentage in spouses were compared among four groups classified by presence or absence of consanguinity between husband's parents and between wife's parents (Table 6). The values in groups (+)-(0), (0)-(+) and (+)-(+), are larger than that in group (0)-(0). Mean coefficient in related spouses was in the order (0)-(0)<(+)-(0)<(0)-(+)<(+)-(+) in H population, whereas the

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Table 3.	Consanguinity rate in spouses by consanguinity
	between husband's parents and between
	wife's parents.

Population	Husband's parents and wife's parents related(+) or not(0)					
-	(0)-(0)	(+)-(0)	(0)-(+)	(+)-(+)		
ĸ	0.1924	0.2857	0.2955	0.5556***		
H	0.1714	0.2477*	0.3377***	0.6071***		
Hs	0.1449	0.1953	0.2597**	0.5385***		
Hi	0.1958	0.1935	0.3194**	0.1250		
U	0.1444	0.1667	0.1970	0.4211***		
Hirado	0.1314	0.1969***	0.2279***	0.3664***		

Significance level of difference between (0)-(0) group and other groups is shown by * (5%), ** (1%) and *** (0.1%).

Table 4.	Observed and expected frequencies of four groups c	lassified
	by consanguinity between husband's parents and	
	between wife's parents.	

Mahar I Andreas	Spouses,		Husban	id's parents and wife's parents, related or not						
Popula-	related(+)	(0)	-(0)	(+)	(+)-(0)		(0)-(+)		(+)-(+)	
tion	or not (0)	Obs.	Exp.	Obs.	Exp.	Obs.	Exp.	Obs.	Exp.	
	(+)	.681	.604	.084	.161	.109	.186	.126	.050	
K	(0)	.833	.814	.061	.081	.076	.096	.029	.010	
	Total	.799	.764	.066	.102	.084	.119	.051	.016	
	(+)	.693	.655	.118	.157	.114	.152	.075	.036	
Н	(0)	.841	.836	.090	.095	.056	.061	.012	.007	
	Total	.812	.798	.096	.110	.068	.081	.025	.011	
	(+)	.674	.637	.138	.175	.111	.147	.077	.041	
Hs	(\mathbf{N})	.740	.736	.130	.134	.106	.110	.024	.020	
		.890	.891	.098	.097	.013	.011		.001	
	Total	.785	.774	.119	.130	.072	.082	.024	.014	
	(+)	.753	.758	.103	.098	.132	.127	.012	.002	
Hi	$(\mathbf{n}) \in \mathbf{N}$.762	.750	.104	.116	.104	.116	.030	.018	
	(0) i Q	.837	.857	.123	.126	.013	.015	.004	.002	
	Total	.779	.782	.114	.115	.088	.090	.020	.013	
	(+)	.706	.678	.140	.168	.096	.124	.059	.031	
U	(0)	.782	.779	.131	.133	.073	.075	.015	.013	
	Total	.770	.763	.132	.139	.076	.083	.022	.015	
	(+)	.750	.738	.102	.114	.116	.128	.032	.020	
Hirado	(0)	.852	.849	.072	.075	.067	.071	.010	.006	
	Total	.837	.832	.076	.081	.074	.079	.013	.008	
······································	(+)	.898	.897	.046	.048	.052	.053	.004	.003	
Fukuoka	(0)	.916	.916	.034	.035	.048	.048	.002	.002	
1	Total	.908	.907	.039	.040	.049	.050	.003	.002	

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Consanguinity		Correlation of consanguinity between				
Popula-	between	husband's parents and wife's parents				
tion	spouses	χ ² -value	\$\phi_coefficient			
	(+)	23.40***				
K	(0)	20.69***				
	Total	54.52***	0.3217			
	(+)	13.94***	· · · · · · · · · · · · · · · · · · ·			
н	(0)	4.07*				
	Total	23.28***	0.1431			
	(+)	9.54**	······································			
Hs		0.63				
	(0) (Q	0.55				
	Total	10.99**	0.1012			
	(+)	0.35				
Hi		4.40*				
		0.55				
	Total	2.52	0.0545			
	(+)	5.19*				
U	(0)	0.39				
	Total	3.51	0.0637			
	(+)	15.38***	· · · · · · · · · · · · · · · · · · ·			
Hirado	(0)	18.03***				
	Total	41.06***	0.0634			
	(+)	2.31	······································			
Fukuoka	(0)	0.15				
	Total	2.00	0.0184			

Table 5. Correlation of consanguinity between husband's parents and wife's parents.

Significance level is shown by (5%), (1%) and (0.1%).

	wife's pare	ents.			
Population	Hu	Total			
	(0)-(0)	(+)-(0)	(0)-(+)	(+)-(+)	-
K	0.0103	0.0165	0.0140	0.0288	0.0119
	(0.0534)	(0.0578)	(0.0475)	(0.0518)	(0.0530)
Н	0.0080	0.0119	0.0172	0.0378	0,0097
	(0.0467)	(0.0480)	(0.0509)	(0.0623)	(0.0485)

Table 6. Mean coefficient of parentage in spouses in groups classified by consanguinity between husband's parents and between wife's parents.

Mean coefficients in related spouses are shown in parentheses.

coefficients in (0)-(0) and (+)-(0) were higher than those in (0)-(+) and (+)-(+) in K population.

DISCUSSION

The present analyses indicate that there are significant correlations of presence or absence of consanguinity among spouses, husband's parents and wife's parents.

Schull *et al.* (1970) stated that the frequency of consanguineous marriages were randomly distributed among spouses, husband's parents and wife's parents in Hirado, contrary to our results obtained using different methods. According to them, the frequency of consanguineous marriages in spouses, husband's parents and wife's parents were 14.65% (excluding those of unknown relationship), 8.89% and 8.66%respectively. And in 131 (1.28%) of 10,264 marriages husband's parents and wife's parents were both related, in 374 (3.66%) of 10,224 marriages spouses and at least one of their sets of parents were related, and these two numbers deviated insignificantly from those expected if consanguinity of the three marriages were independent of each other. Upon this supposition, however, the number of families in which husband's parents and wife's parents are both related is expected to be

 $(0.0889 \times 0.0866) \times 10,264 = 79.02 \ (0.77\%),$

which differs from the observed numbers 131 (1.28%) significantly at the 0.1% level (t=5.91). Number of families, in which the spouses and at least one parental couple are consanguineous, is expected to be

 $\{1-(1-0.0889) \times (1-0.0866)\} \times 0.1465 \times 10,224 = 251.51 \ (2.46\%),\$ which differs from the observed number 374 (3.66%) significantly at the 0.1% level (t=7.83). Thus, correlation of consanguinity among the three marriages is reconfirmed in Hirado data using the procedure of Schull *et al.*

Yanase (1966) mentioned that the consanguinity rate between wife's parents in the group of related spouses was significantly higher than that in Q group (nonnative, unrelated), but not higher than that in N group (native, unrelated). Our analysis revealed a significant correlation of consanguinity between wife's parents with that between spouses in Q group as mentioned by Yanase, and a positive, but insignificant correlation in N group. Consanguinity rate of husband's parents was higher in the group of related spouses than that in unrelated ones in both N and Q groups in Hs population (Table 2). And consanguinity between husband's parents was correlated with that between wife's parents significantly at the 1% level in the group of related spouses in Hs and at the 5% level in N group in Hi population (Table 5).

Munakata (1955) reported that $11.76 \pm 2.92\%$ of offspring born to related parents and $5.39 \pm 1.90\%$ of offspring born to unrelated parents were married to relatives in a mountainous district in Fukushima Prefecture. This difference is significant at the 5% level and, thus, consanguineous marriages tended to recurrent in successive generations.

Geographical and/or social isolation may be at least one of main causes for such accumulation of consanguinity. Of the populations studied in the present report,

K, H, Hs and Hi were geographically isolated mountainous villages, each of which was composed of a number of subpopulations more or less isolated from each other. Hirado population was separated into three religious groups (Buddhists, Catholics and Kakure-kirishitan), which were rather strictly isolated from each other with a few exceptions.

The trend of accumulation of consanguineous marriages was observed also in the two remaining populations of which the sample from Fukuoka was an urban population and U population was composed of several rural communities adjacently scattered in a plain with no apparent isolation within each population. Therefore, the accumulation of consanguinity may not be explicable by isolation only. Some explanations may be plausible such as: some families prefer to marry relatives in order to keep from dispersion of properties, and others have difficulties to find mates among unrelated persons owing to family history of hereditary diseases, syphilis, leprosy, etc.

Familial accumulation of consanguinity may have following effects:

1) Increase in inbreeding coefficient.

Consanguineous marriages in successive generations increase inbreeding coefficient in the offspring.

2) Joint effects with maternal inbreeding.

Adverse effects on fertility, mortality and morbidity of inbreeding in successive generations have been recognized in various experimental animals. These are attributed to single or joint effects of consanguinity and maternal inbreeding. Tanaka and others (1964, 1967, 1972, 1977) pointed out possibilities of such maternal inbreeding effects in man. If consanguineous marriages are contracted in successive generations, the effects of consanguineous marriages of a spouses and those of wife's parents overlap each other.

Thus, the accumulation of consanguinity may give overestimates of inbreeding effects, if consanguineous marriages in the previous generations are ignored.

REFERENCES

- Munakata, T. 1955. Studies on consanguineous marriages in a mountainous district in Fukushima Prefecture, Japan (in Japanese). Fukushima Med. J. 5: 131–136.
- Nakamoto, N. 1969. A study of consanguinity in Fukuoka, an urban population of large size mating patterns and degree of inbreeding (in Japanese). *Fukuoka Acta Medica* 60: 184–209.
- Schull, W.J., Nagano, H., Yamamoto, M. and Komatsu, I. 1970. The effects of parental consanguinity and inbreeding in Hirado, Japan. I. Stillbirths and pre-reproductive mortality. Am. J. Hum. Genet. 22: 239-262.
- Tanaka, K., Yanase, T. and Furusho, T. 1964. Effects of inbreeding on fertility in man. *Proc. Jap. Acad.* 40: 852–856.
- Tanaka, K., Ohtsuki, N. and Furusho, T. 1967. Effects of maternal inbreeding on prenatal deaths in man. Proc. Jap. Acad. 43: 801-806.
- Tanaka, K. 1972. Possible effects of maternal inbreeding on congenital abnormalities and mental retardation in man. Jap. J. Hum. Genet. 16: 170-181.

- Tanaka, K. 1977. Genetic effects of maternal inbreeding in man on congenital abnormality, mental defect, infertility, and prenatal death. Jap. J. Hum. Genet. 22: 55-72.
- Yanase, T. 1966. The study of isolated populations (in Japanese). Jap. J. Hum. Genet. 11: 125 -161.