Rates for Specific Birth Defects among Native Hawaiians Compared to Whites, Hawaii 1986-2000

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Abstract

Rates for many birth defects have not been identified for native Hawaiians. Using birth defects registry data, the total major birth defect rate for whites was found to be 4.70% and for native Hawaiians was 4.75%. After adjusting for maternal age, the rates were lower among native Hawaiians for 40 (74.1%) of 54 selected birth defects.

Introduction

A number of studies have reported racial/ethnic differences in risk for specific birth defects. For example, studies have found neural tube defect (NTD) rates to be higher among Hispanics[1,2] and pyloric stenosis among whites.[3,4] However, most studies in the United States that have examined racial/ethnic differences included only one or a few specific birth defects and/or used broad racial/ethnic categories, such as the broad group of Asians and Pacific Islanders. Previous investigations by the authors have demonstrated differences in risk for specific birth defects between Far East Asians (Japanese, Chinese, Korean) and Pacific Islanders (native Hawaiian, Samoan, Guamanian).[5-7]

Few population-based studies in the United States have examined risk for specific birth defects among native Hawaiians, and these studies have tended to focus on one or a few birth defects.[8] Information on rates for a number of specific birth defects among native Hawaiians is important because studies have found native Hawaiians to differ from other racial/ethnic groups in a variety of health factors such as mortality,[9,10] pregnancy outcomes,[11] and selected diseases.[12-14] Moreover, Hawaii State Department of Health (DOH) data (http://www.state.hi.us/health/) indicate that the maternal racial/ethnic group accounting for the highest proportion of births in 2001 in Hawaii was native Hawaiian (27%), while white mothers accounted for 22% of births. The proportion of births with maternal race/ethnicity of native Hawaiian is so high because the native Hawaiian category includes women of mixed-race/ethnicity where only part of their ancestry is native Hawaiian. Of the 63,291 live-births delivered during 1986-2000 to native Hawaiian mothers in Hawaii, 61,440 (97%) of the mothers were listed as part-Hawaiian.

Individuals of native Hawaiian ancestry, no matter what the rest of their ancestry is comprised of, are generally treated as a unique group in Hawaii. Such individuals are subject to a variety of societal and cultural advantages and disadvantages solely because of their Hawaiian ancestry. Thus classification as native Hawaiian may be considered more of a cultural than a genetic distinction. Moreover, native Hawaiians have traditionally been grouped together for evaluation of racial/ethnic differences.

This study sought to identify the rates for a number of specific birth defects among native Hawaiians and to compare the rates to those among whites. In previous investigations, the authors have grouped native Hawaiians with other Pacific Islanders, so this information has not been presented before.

Methods

The Hawaii Birth Defects Program (HBDP) is an active birth defects surveillance system for the entire state.[15] HBDP inclusion criteria consist of all infants and fetuses of any pregnancy outcome (live birth, fetal death, elective termination) at any gestational age that ended in Hawaii with at least one reportable birth defect identified between conception and one year after delivery. HBDP staff identify eligible infants and fetuses and collect information by reviewing medical records at a variety of health care facilities in the state where such infants and fetuses are likely to be diagnosed or treated. Through this multiple source ascertainment system, identification of eligible infants and fetuses where reportable birth defects were diagnosed is considered to be as complete as possible.

For the current investigation, cases were all infants and fetuses in the HBDP delivered during 1986-2000 with one or more confirmed major birth defect where the maternal race/ethnicity was listed as native Hawaiian or white. The HBDP obtains maternal race/ethnicity for cases from birth and fetal death certificates except in those instances where such vital records are not available, in which case the HBDP obtains maternal race/ethnicity from the medical records. For those mothers of mixed race/ethnicity, the HBDP follows DOH guidelines for assigning a single race/ethnicity.

If at least one of a mother's reported races/ethnicities is native Hawaiian, then the mother's race/ethnicity is defined as native Hawaiian. If a mother's race/ethnicity is reported as only white, then the mother's race/ethnicity is defined as white.

In this investigation, there is no overlap between the two racial/ethnic groups studied in that an individual may be assigned to one category or another but not both. Moreover, since race/ethnicity was based on birth certificate data provided by the DOH, where only a single race/ethnicity is mentioned, the researchers were limited to those categories used by the DOH.

Using as denominators information provided by the DOH Office of Health Status Monitoring as derived from birth certificates, the authors calculated the rates for 54 specific birth defects for native Hawaiians and for whites separately. The authors chose the specific birth defects because they were relatively common, easily identifiable, and/or influenced morbidity and mortality. A case with more than one birth defect was included in all appropriate birth defects categories. Relative risk, defined as the ratio of the rate among native Hawaiians to the rate among whites (comparison population), and associated 95% confidence intervals (CIs) were calculated for each birth defect category using Poisson probability. In the majority of population-based birth defects studies in the United States that examined race/ethnicity, the most frequently reported race/ethnicity was white. Thus for consistency native Hawaiians were compared to whites in this study. In order to simplify the analysis and avoid confusion, comparisons with other racial/ethnic groups were not made.

The maternal age distribution differed between whites and native Hawaiians, with native Hawaiians generally giving birth at a younger age (Table 1). As a result, the authors also calculated relative risks after adjusting for maternal age via direct standardization using whites as the standard population. The authors examined other variables that might be associated with birth defect risk such as plurality and infant sex but they did not find these variables to differ greatly between the two racial/ethnic groups and thus did not control for these variables.

Table 1.— Total number of live births to white and native Hawaiian mothers in Hawaii during 1986-2000 by maternal age group

Maternal age (yrs)	white number (%)	native Hawaiian number (%)
≤19	3,905 (5.4)	12,049 (19.0)
20-24	19,310 (26.7)	20,514 (32.4)
25-29	21,479 (29.7)	16,453 (26.0)
30-34	17,061 (23.6)	9,478 (15.0)
35-39	8,576 (11.8)	3,828 (6.0)
≥40	2,074 (2.9)	947 (1.5)
Unknown	11 (0.0)	22 (0.0)
Total	72,416	63,291

Results

During 1986-2000, the HBDP identified 3,404 cases with major birth defects delivered among 72,416 live births with white mothers, resulting in a rate of 4.70%. During the same time period, there were 3,004 cases of major birth defects delivered among 63,291 live births with native Hawaiian mothers, resulting in a rate of 4.75%. The resulting crude relative risk was 1.01 (95% CI 0.96-1.06) and relative risk adjusted for maternal age was 0.87 (95% CI 0.84-0.90).

Table 2 presents the rates for specific birth defects among whites and native Hawaiians. The crude rate was lower among native Hawaiians for 24 (44.4%) birth defects and higher among native Hawaiians for 30 (55.6%) birth defects. The crude rate was significantly higher among native Hawaiians for 7 (13.0%) of the birth defects and significantly lower for 7 (13.0%) of the birth defects. However, there was no clear pattern to those birth defects demonstrating the higher or lower crude rates among native Hawaiians.

After adjusting for differences in maternal age distribution between the racial/ethnic groups, the relative risks indicated decreased risk among native Hawaiians for 40 (74.1%) of the birth defects. In particular, native Hawaiians had lower risks of NTDs, a number of cardiovascular defects such as conotruncal defects and septal defects, and oral clefts. However, only two of the birth defects (Ebstein's anomaly and trisomy 13) demonstrated relative risks below 0.50; none of the relative risks were greater than 1.47. The only significant differences observed were lower risks among native Hawaiians for two birth defects (ventricular septal defect and trisomy 21).

Discussion

This investigation examined the risk of a large number of specific birth defects among native Hawaiians. Although review of the literature discovered other studies that provided rates for selected birth defects among native Hawaiians, the authors were unable to identify any studies that provided rates for a large variety of birth defects among this racial/ethnic group.

While native Hawaiians may comprise a large proportion of the population of Hawaii, such is not expected to be the situation for other states. However, this study is important because it demonstrates differences in birth defects rates among populations of different race/ethnicity. Such populations are likely to differ with respect to genetics, behaviors, and exposures. Information on differences in birth defects risk between such populations may be useful in eliciting the causes of birth defects.

Table 2.— Rates per 10,000 live births of specific birth defects with white and native Hawaiian mothers, Hawaii, 1986-2000						
Diagnosis	white		native Hawaiian		crude RR1	adjusted RR2
	no.	crude rate	no.	crude rate		
Anencephaly	25	3.45	26	4.11	1.19	0.96
Spina bifida	37	5.11	29	4.58	0.90	0.82
Encephalocele	15	2.07	12	1.90	0.92	0.79
Holoprosencephaly	3	0.41	5	0.79	1.91	1.22
Hydrocephaly	75	10.36	80	12.64	1.22	0.91
Microcephaly	61	8.42	79	12.48	1.48*	0.83
Anophthalmia/microphthalmia	27	3.73	24	3.79	1.02	0.85
Cataract	10	1.38	8	1.26	0.92	1.32
Glaucoma	0	0.00	1	0.16	-	-
Anotia/microtia	5	0.69	16	2.53	3.66*	0.77
Truncus arteriosus	6	0.83	3	0.47	0.57	0.60
Transposition of great arteries	29	4.00	37	5.85	1.46	0.90
Tetralogy of Fallot	17	2.35	36	5.69	2.42*	0.88
Single ventricle	5	0.69	6	0.95	1.37	1.05
Ventricular septal defect	300	41.43	287	45.35	1.09	0.83*
Atrial septal defect	130	17.95	145	22.91	1.28	0.87
Endocardial cushion defect	21	2.90	19	3.00	1.04	0.83
Pulmonary valve atresia/stenosis	42	5.80	74	11.69	2.02*	0.92
Tricuspid valve atresia/stenosis	9	1.24	19	3.00	2.42*	1.06
Ebstein's anomaly	6	0.83	1	0.16	0.19	0.45
Aortic valve stenosis	11	1.52	6	0.95	0.62	0.94
Hypoplastic left heart syndrome	14	1.93	13	2.05	1.06	1.08
Coarctation of aorta	25	3.45	10	1.58	0.46*	0.70
Interrupted aortic arch	4	0.55	1	0.16	0.29	0.77
Anomalous pulmonary venous return	3	0.41	8	1.26	3.05	1.06
Choanal atresia/stenosis	11	1.52	6	0.95	0.62	0.70
Cleft palate	43	5.94	66	10.43	1.76*	0.81
Cleft lip with/without cleft palate	73	10.08	80	12.64	1.25	0.88
Esophageal atresia and/or tracheoesophageal fistula	21	2.90	14	2.21	0.76	0.84
Pyloric stenosis	123	16.99	35	5.53	0.33*	0.98
Small intestinal atresia/stenosis	17	2.35	22	3.48	1.48	0.92
Anal, rectal, and large intestinal atresia/stenosis	34	4.70	37	5.85	1.25	0.84
Hirschsprung's disease	13	1.80	11	1.74	0.97	1.24
Biliary atresia	6	0.83	7	1.11	1.33	0.88
Malrotation of intestines	15	2.07	23	3.63	1.75	1.03
Hypospadias and epispadias	232	32.04	152	24.02	0.75*	0.90
Renal agenesis/hypoplasia	39	5.39	35	5.53	1.03	0.97
Cystic kidney	32	4.42	27	4.27	0.97	0.83
Obstructive genitourinary defect	120	16.57	87	13.75	0.83	0.87
Bladder exstrophy	2	0.28	1	0.16	0.57	1.06
Persistent cloaca	1	0.14	1	0.16	1.14	1.06
Congenital hip dislocation	100	13.81	47	7.43	0.54*	0.82
Polydactyly	94	12.98	115	18.17	1.40*	0.87
Syndactyly	93	12.84	68	10.74	0.84	0.92

Diagnosis	white		native Hawaiian		crude RR1	adjusted RR2
	no.	crude rate	no.	crude rate		
Reduction deformity of upper limbs	20	2.76	30	4.74	1.72	1.04
Reduction deformity of lower limbs	7	0.97	12	1.90	1.96	1.02
Craniosynostosis	57	7.87	29	4.58	0.58*	0.76
Diaphragmatic hernia	25	3.45	16	2.53	0.73	0.76
Omphalocele	22	3.04	12	1.90	0.62	0.91
Gastroschisis	22	3.04	24	3.79	1.25	1.47
Situs inversus	6	0.83	10	1.58	1.91	0.72
Trisomy 21	123	16.99	65	10.27	0.60*	0.77*
Trisomy 13	19	2.62	7	1.11	0.42	0.49
Trisomy 18	37	5.11	17	2.69	0.53*	0.72

Cases with more than one eligible birth defect are included in all relevant categories.
[1]Crude relative risk, ratio of the rate among native Hawaiians to whites (comparison population).
[2]Adjusted relative risk, ratio of the rate among native Hawaiians to whites (comparison population), adjusted for maternal age.
*95% confidence interval does not include 1.00, i.e., the relative risk is statistically significant.

The United States is becoming more racially diverse, with various racial/ethnic groups beginning to comprise a higher proportion of the total population. However, much of the information on birth defects in the United States has been derived from predominantly white populations, and when racial/ethnic differences were analyzed, the focus was generally on broad racial/ethnic groups. Few studies have provided birth defects rates for particular subgroups.[16] Such information may become of greater importance as these subgroups increase in number.

Individuals of mixed race/ethnicity are not unique to Hawaii. In Census 2000, 2.4% of the population in the United States reported more than one race (http://www.census.gov/prod/2001pubs/c2kbr01-6.pdf). The proportion of the population reporting more than one race varied by geographic area, being highest in the West and lowest in the Midwest. The state with the highest rate of people reporting more than one race was Hawaii (21%), followed by Alaska (5.4%), California (4.7%), and Oklahoma (4.5%). States with over one-half million of their population reporting more than one race were California, New York, and Texas.

Researchers have performed a number of studies of race/ethnicity where individuals were assigned to a single racial/ethnic group. However, the racial/ethnic groups in these studies could also be considered heterogeneic. For example, in Census 2000 2.5% of the individuals who specified their race as white also reported another race, and 4.8% of individuals who specified their race as African-American also reported another race. Hispanic ethnicity also can be considered a heterogeneic category, considering that it includes individuals of varying degrees of ancestry from a wide geographic area including the Carribean and Central and South America. Even grouping together individuals who report themselves solely as white blurs distinctions between individuals from different parts of Europe. Thus bias may also exist in these studies. As identification of mixed race/ethnicity becomes more prominent in the future, researchers will have to recognize the potential for bias when assigning such individuals to a single racial/ethnic group, invent methods for evaluating racial/ethnic differences that factors in mixed race/ethnicity, or cease performing analyses of race/ethnicity.

The total rate of any major birth defect was significantly lower for native Hawaiians than for whites after adjusting for differences in maternal age distribution. Adjusted relative risks indicated decreased risk among native Hawaiians for 74% of the more than fifty specific birth defects studied, including neural tube defects, many cardiovascular defects, and oral clefts. However, for only two of the birth defects did the adjusted relative risks indicate significantly decreased risk among native Hawaiians.

Potential explanations for the dissimilarities between whites and native Hawaiians with respect to rates for specific birth defects include racial/ethnic differences in diagnosis of birth defects or ascertainment of cases even if a diagnosis was made. For example, if a particular racial/ethnic group were associated with lower socioeconomic status, such a racial/ethnic group might have less access to health care and diagnostic procedures. Differences in diagnosis might be expected to most affect those birth defects that are internal and less likely to impact morbidity or mortality. For example, several studies that have noted secular increases in rates of comparatively minor heart defects have associated the changes with increased use of diagnostic procedures such as echocardiography.[17,18] Other investigations have suggested racial/ethnic differences in access to and use of prenatal diagnostic procedures to account for racial/ethnic differences observed for birth defects

among live births.[19,20] However, the present study included all pregnancy outcomes, so preferential access to or use of prenatal diagnostic procedures by one racial/ethnic group over the other would be expected to have limited impact on any observed differences in birth defect rate. In addition, racial/ethnic differences were observed with respect to birth defects that are fairly easy to diagnose. The lack of consistent patterns in the differences in crude birth defects rates between whites and native Hawaiians, i.e., whites had higher rates for some birth defects and native Hawaiians had higher rates for other birth defects, would also suggest that the racial/ethnic differences in birth defects observed were not greatly influenced by diagnosis and ascertainment biases, at least not for all of the defects.

Aside from ascertainment or diagnostic factors, any differences between whites and native Hawaiians in birth defects risk would likely be due to genetics or environment or some combination of the two. Genetic differences between native Hawaiians and other racial/ethnic groups have not been extensively studied. Native Hawaiians might differ from whites with respect to environmental factors such as diet, exposure to potential teratogens such as pollutants, and cultural practices. An investigation reported native Hawaiians more likely than whites to be overweight, physically inactive, and smokers, factors that may be associated with birth defects risk.[21]

In this study, there is potential for bias associated with inclusion of mothers of mixed race/ethnicity. Another limitation of this investigation is the relatively small number of cases, particularly among certain specific birth defects categories. However, several statistically significant differences in birth defect rates between whites and native Hawaiians were identified. It must be pointed out that among a large number of analyses for statistical significance, particularly involving small numbers of cases, a certain proportion of statistically significant differences might be expected to occur by chance. If the assumption is made that one in every twenty analyses will lead to a statistically significant result, then significant differences would be expected in 2.7 of the 54 birth defects categories examined in this study solely by chance. This investigation reported two significant differences.

The observation in this study that birth defects rates varied between two different racial/ethnic groups in Hawaii may be useful in generating hypotheses regarding the causes of these birth defects. Moreover, this study provides information on birth defects among native Hawaiians; such data may be useful for health care providers when planning services or assessing risk for this racial/ethnic group.

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