

Japanese Database of Families with Twins and Higher-Order Multiples

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To gain widespread participation, epidemiologic studies of twins from pregnancy through the childhood period are expected to reflect the needs and concerns of families and provide participants with appropriate and useful feedback based on scientific evidence. Our most recent database on families with twins throughout Japan is based on a questionnaire survey conducted from January 2010 to August 2011. Mailed or hand-delivered questionnaires, consisting of over 550 items were used to collect the basic data. The response rate was 40% (956/2,401). This is part of a nation-wide study designed to assess the long-term effect of perinatal conditions on mothers of multiples. Its aim was to study the growth and development of multiples in childhood, and to conduct a genetic epidemiologic study to test the developmental origin of health and disease hypothesis. One of the ultimate aims of this research was to provide evidence-based information on parenting multiples from pregnancy through childhood to families with multiples.

■ **Keywords:** families with multiples, growth and development, developmental origin of health and disease hypothesis, nationwide survey

Since the early 1980s the rate of multiple births has rapidly increased in all developed countries largely due to the widespread use of ovulation induction and multiple embryo transfer (Bryan, 2006). Increasing multiple birth rates have also been observed in Japan. Around 1% of all pregnancies (2% of all live births) are multiples (Ooki, 2011).

Multiple birth is associated with substantial medical, health care, socio-emotional, developmental, educational, and economic consequences for both families and society (Bryan, 2003, 2006; Denton, 2005; Leonard & Denton, 2006; Ooki, 2009; Ooki & Hiko, 2012). The nurturing of multiples entails a higher burden physically, mentally, and economically than that of singletons. Participants in twin studies rightly expect appropriate information from researchers to facilitate the healthy development of their twins. Indeed, the chance to gain useful information for the parenting of multiples is a strong incentive for the parents to participate in such studies. The majority of twin registries throughout the world have been constructed primarily for genetic studies. It appears to be very difficult to achieve a high participation rate and detailed data from families with very young children in Japan, particularly if researchers are focused only on genetic issues, with no feedback for the participants.

Three Aspects of Studies on Multiples

There are several independent fields of research regarding multiples, especially twins. The classic type of research is that which attempts to clarify the relative contributions of genetic and environmental factors on human phenotypes. Twin- and multiple-birth obstetrics is also a long-standing field of research. It is only recently that the special problems and developmental needs of twins and higher-order multiple-birth children and of their families have begun to be recognized as an important research topic (Ooki, 2009).

These different types of studies on twins and higher-order multiples can be linked. The family support practices of families with multiples will become more fruitful if the findings of scientific research concerning twins and multiples in terms of biology, obstetrics, psychology, and human genetics are taken into account, in addition to the research

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on maternal and child care and vice versa. The present project has a strong emphasis on this point.

Strategies for the Collection of Data on Multiples in Japan

Since the frequency of families with multiples is not very high (0.6–1% at most) in Japan, a strategic method of study is crucial if researchers hope to gather sufficient amounts of high-quality data. There are four main types of data included in studies on multiples in Japan (Ooki, 2006). First, vital statistics can be obtained, but it is almost impossible to obtain access to personal information concerning individuals (Kato, 2004). Second, data from large hospitals have been used in the field of obstetrics (Kurosawa et al., 2012). The collection of obstetric data from multiples is relatively easy with the trade-off of selection bias in favor of high-risk infants/mothers. Third, the Basic Resident Registration of municipalities can be used. This registration reflects the whole population of each area, and serves as a possible source for recruitment of families with multiples. However, the cost-effectiveness of this method is extremely low in the case of multiple-birth families. Finally, there is a volunteer-based database of multiples that includes data from mothers belonging to associations for parents of multiples (Ooki & Asaka, 2005). It contains more detailed information on the condition of multiples after birth using vital statistics and hospital data. Although volunteer-based databases may have some selection biases, their cost-effectiveness is very high.

Conceptual Model of the Present Project

The public health problems surrounding multiple birth families cannot be resolved by the efforts of families with multiples alone, even if they create local twins' clubs or groups. A more multidisciplinary collaboration, including specialists from the different domains involved, is essential. The key concept of the present practice is outlined in Figure 1. We are trying to combine research and practice by applying the Evidence-Based Public Health (EBPH) method (Gray, 2008), the Community-Based Participatory Research (CBPR) approach (Israel et al., 2008), and a population approach (Rose, 1994).

The three main components of EBPH are evidence, value, and local needs and resources (Gray, 2008). EBPH requires that decisions about public health practice be based on the best available, current, valid, and relevant evidence. These decisions should be made by those receiving care, informed by the tacit and explicit knowledge of those providing care, within the context of available resources.

CBPR is a research approach that emphasizes collaborative partnerships between community members, health care providers and researchers to generate knowledge and solve local problems (Israel et al., 2008). Each partner contributes unique strengths and knowledge to improve the health and

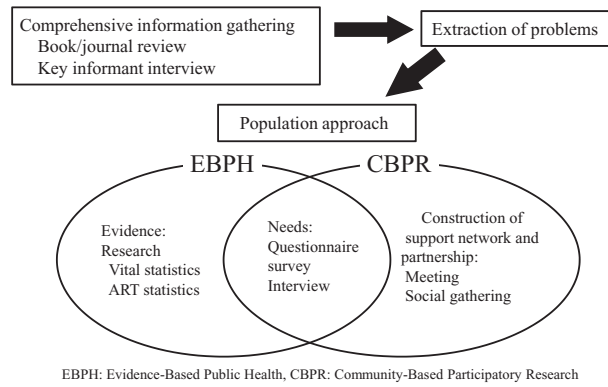


FIGURE 1
Conceptual framework of the project.

well being of community members. The researchers must know what issues the community members are willing to work on, and they must understand the related attitudes, values, beliefs, and customs.

A large database of twins, recruited from pregnancy to adulthood, was started in 1987 (Ooki & Asaka, 2005, 2006). The primary purpose of this database was genetic epidemiologic twin research. However, it turned out that the data were also a useful resource for the provision of information on many features specific to twins — for example, information on their growth and development — and that this information could be fed back to families with multiples and to health professionals. The CBPR approach seemed very suitable for constructing a large volunteer-based database of families with multiples in childhood.

Construction of Prefectural-Level Human Network: Ishikawa Support Network for Multiple Birth Families

Japan consists of 47 prefectures, the basic unit of local government, and 1,742 (as of April 2012) municipalities. National government policies for the health of mothers and children are planned and administered by the Ministry of Health, Labor and Welfare. At present, most of the functions of maternal and child health administration have been transferred from the prefectural level to the municipal level. Typically, a single prefecture has several public health centers, which serve several municipalities within their catchment area. Given this background, the construction of a database of families with multiples in childhood began in 2004 at the prefectural level in Ishikawa prefecture. The detailed strategy and methods for constructing this network have been reported elsewhere (Ooki, 2006, 2009; Ooki & Hiko, 2012).

A human network to support multiple birth families was organized alongside the demographic research and questionnaire surveys using the CBPR approach. This network was constructed with the help of the relationships between

families with multiples, support groups for child rearing, governmental and medical institutions, and universities. The Health and Welfare Bureau of Ishikawa Prefecture provided assistance in a positive way: several intensive meetings were held for the purpose of exchanging information among members of associations for the parents of multiples, medical staff, public health nurses, midwives, and twin researchers. Workshops, round-table discussion, and other events were held periodically in both the local and central districts. Moreover, past research results on multiples were written up in a way that was understandable to mothers of multiples and provided in fact sheets, brochures, and leaflets presented to participants in workshops, family support events, and similar settings. Finally, the Ishikawa Support Network for Multiple Birth Families was founded in July 2005.

This network comprises a wide range of core members, including leaders of the twin mothers' clubs, maternal and child health professionals and researchers. Its aims are to hold workshops, family support events, and specialized parenting classes for families with multiples, to facilitate the exchange of information, skills and discussion on maternal and child health policies, and to promote research on multiple births in general. The goals of the project are to contribute to the development of welfare programs for multiple birth families as well as to coordinate research useful for both human genetics and maternal and child health. The well-established and sophisticated strategies used to recruit twin families into the registry have been very useful for the construction of the human network and information distribution in this program.

The Japan Multiple Births Association (JAMBA)

By constructing a support network at the prefectural level, continuity of support, an expansion of cooperation, a wide range of information exchange, and responsiveness to the distinct characteristics of the region became possible. Other prefectures have followed suit by constructing the same kind of support network. Although the method varies according to the situation in a given prefecture regarding human resources and governmental/medical institutions, the idea of a population-based approach is constant. This strategy is welcomed by the many participants and other involved parties. Finally, the JAMBA was established on February 22, 2010 (the date 2/22 chosen to symbolically reflect twins), in celebration of families with multiples (Ooki & Hiko, 2012). This association became a member of The International Council of Multiple Birth Organizations on June 2010. One of the main missions of JAMBA is to promote research on multiple births for evidence-based support of families. The foundation of a Japanese database of families with multiple births was thus developed.

Outline of Japanese Database of Families with Multiple Births in Childhood

The author has constructed a database of families with multiple births, consisting of 951 mothers of twins and higher-order multiples from several associations for parents of multiples throughout Japan (Ooki & Asaka, 2005). Continuous data have been gathered in diverse ways from 2001 to 2012.

The most recent database consists of about 2,400 families with multiples, who were recruited from January 2010 to August 2011 during collaborative research with multiple-births families throughout Japan. This is part of a nationwide study designed to assess the long-term effect of perinatal conditions of mothers parenting multiples on the growth and development of multiples in childhood and using genetic epidemiologic twin studies to test the developmental origin of health and disease hypothesis. One of the aims of this research was to provide evidence-based information on parenting multiples from pregnancy through childhood to families with multiples.

Mailed or hand-delivered questionnaires, consisting of over 550 question items with free-description answers, were used to collect the basic data. These items include questions about pregnancy, circumstances of child rearing, especially the physical, mental, and social condition of mothers and fathers, and the physical growth and motor and language development of multiples. The response rate was 40% (956/2,401). Most medical data in all our databases were obtained from *The Maternal and Child Health Handbook*, which is presented by the Ministry of Health, Labor and Welfare to all pregnant women in Japan. The growth data of children based on mass examinations are usually recorded in this handbook, and it serves as a valuable source of health information for pregnant women, as it contains detailed medical records on pregnancy and delivery, as well as on childcare, for children up to 6 years old. The handbook also presents the growth standards of weight and height/recumbent length and motor and language developmental milestones every 10 years; for example, 1980, 1990, 2000, and 2010. The author advised the mothers to refer to these records when completing the questionnaire. Although this method seemed to be the most effective way to collect large amounts of data on twins after birth, it did not produce perfect longitudinal data. No information on detailed chorionicity could be gathered. The zygosity of twin pairs was determined by questionnaire (Ooki & Asaka, 2004).

These questionnaire surveys are now in progress. Regarding ethical issues, all the mothers in the maternal associations cooperated voluntarily in this research, mainly through the presidents of their associations.

Basic characteristics of the Japanese Database of Families with Multiple Births are shown according to two age groups in Table 1

TABLE 1
Basic Characteristics of Japanese Database of Families with Multiple Births in Childhood

			0–6 years group (N = 461 pairs)		6–18 years group (N = 320 pairs)		
Sex of twin individuals							
	Male		430	46.6%	324	50.6%	
	Female		492	53.4%	316	49.4%	
Birth year of twin pairs							
	Mean ± SD		2006 ± 2		1998 ± 4		
	Median		2006		1998		
	Range		2003–2011		1991–2005		
Age							
	0 year		36	7.8%	6 years ^a	14	4.4%
	1 year		77	16.7%	7–8 years	70	21.9%
	2 years		72	15.6%	9–10 years	53	16.6%
	3 years		92	20.0%	11–12 years	58	18.1%
	4 years		82	17.8%	13–14 years	44	13.8%
	5 years		61	13.2%	15–16 years	48	15.0%
	6 years		41	8.9%	17–18 years	33	10.3%
Zygosity and sex combination							
	Monozygotic	Male–Male	59	12.8%	65	20.3%	
		Female–Female	70	15.2%	69	21.6%	
	Dizygotic	Male–Male	69	15.0%	49	15.3%	
		Female–Female	79	17.1%	43	13.4%	
		Opposite-Sexed	142	30.8%	74	23.1%	
	Unclassified	Male–Male	10	2.2%	11	3.4%	
		Female–Female	14	3.0%	7	2.2%	
	Insufficient information	Male–Male	6	1.3%	0	0.0%	
		Female–Female	12	2.6%	2	0.6%	
Family construction							
		Nuclear family	383	83.1%	257	80.3%	
		Three-generation family	65	14.1%	55	17.2%	
		Others	4	0.9%	5	1.6%	
		Missing	9	2.0%	3	0.9%	
Siblings							
	Multipara	Both elder and younger	8	1.7%	9	2.8%	
		Elder	128	27.8%	88	27.5%	
	Primipara	Younger	38	8.2%	49	15.3%	
		Only multiples	282	61.2%	171	53.4%	
		Missing	5	1.1%	3	0.9%	
Maternal age at twins birth (year)							
	Mean ± SD		33.1 ± 4.0		32.0 ± 4.1		
	Median (range)		33 (22–44)		32 (21–43)		
	Missing		10		5		
Paternal age at twins birth (year)							
	Mean ± SD		35.5 ± 5.6		34.4 ± 4.9		
	Median (range)		35 (24–67)		34 (21–49)		
	Missing		20		10		
Gestational age (weeks)							
	Mean ± SD		36.3 ± 2.1		36.2 ± 2.5		
	Median (range)		37 (25–41)		37 (26–41)		
	Missing		8		6		
Method of pregnancy							
		Spontaneous	226	49.0%	196	61.3%	
		iatrogenic					
		Ovulation-stimulation	70	15.2%	48	15.0%	
		ART	158	34.3%	70	21.9%	
		Missing	7	1.5%	6	1.9%	
Neonatal condition (twin individuals)							
		Healthy	847	91.9%	547	85.5%	
		Hyposthenia (not so healthy)	53	5.7%	40	6.3%	
		Neonatal asphyxia	14	1.5%	35	5.5%	
		Missing	8	0.9%	18	2.8%	

Note: Children aged over 18 years old (n = 158) and 12 sets of triplet were excluded from the calculation.
^aSchool children.

Conclusion

Most data on multiples after birth can only be obtained from volunteer families. Epidemiologic studies are expected to be performed based on community networks of multiple families. Likewise, they are expected to reflect their real needs and social concerns, with appropriate and useful feedback based on scientific evidence. If research that actively involves the multiple birth families concerned and other community members is performed with the CBPR approach, both families and researchers can benefit, and good relationships will be developed. The concept of the community support net-

work seems to be one effective means both for collecting data and providing support for multiple-birth families. Using this mechanism, evidence-based research on multiple-birth families can be performed, and its results can be easily disseminated.

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