

*Chapter 8*

## **CHILD MALTREATMENT ASSOCIATED WITH MULTIPLE BIRTHS IN JAPAN**

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### **ABSTRACT**

The purpose of this chapter is to clarify the impact of multiple births on child maltreatment through a comprehensive literature review and use of nationwide data in Japan. A literature review showed that there have been very few studies on maltreatment in cases of multiple births, with the first survey performed in 1982. The main findings of the literature review were as follows: 1) Families with twins or multiple births might have an elevated risk of child maltreatment. 2) No risk factor specific to the families with multiples was detected. 3) Shaken baby syndrome or inflicted traumatic brain injury might occur more frequently in the case of multiple births. Next, the national annual reports on fatal child maltreatment from July 2003 to March 2013, published by the Ministry of Health, Labor and Welfare of Japan, were used as the initial sources of information. There were 546 cases of fatal maltreatment of children aged 0–17 years during this period, including 15 multiples (all twins) from 14 families. Multiple births were regarded as an exposed group. The relative risks (RRs) and their 95% confidence intervals (CIs) were estimated using the data from the above reports and vital statistics. The RRs of multiple births per individual and per family unit were 1.5 (95% CI 0.9 - 2.5) and 3.0 (95% CI 1.8 - 5.1), respectively. Next, a keyword search was performed to create a full profile for each multiple-birth case. No significant difference between twins and singletons with fatal maltreatment was observed for most characteristics. However, in the case of twins, 0-month victims were rarer, and the number of children per family was larger. One twin died from shaken baby syndrome. The victim's siblings were also maltreated in 6 of the 13 relevant cases, including all 6 co-twins. Premature birth, having a disabled co-twin, delay of growth or development, and parental disfavor tended to be factors of maltreatment when only one twin was maltreated. In conclusion, the literature review and recent Japanese nationwide data suggest that families with multiple births have elevated risk for child maltreatment, and the nonspecific overburden of child rearing might be one possible reason for the higher frequency of child maltreatment for multiples compared to singletons; comparisons between the two twins might be another. Health

providers should be aware that multiple pregnancies/births may place significant stress on families and should provide appropriate support and intervention.

## INTRODUCTION

Multiple birth was recognized as a risk factor for child maltreatment in several studies in the 1980s (Groothuis et al., 1982; Nelson & Martin, 1985; Robarge, Reynolds, & Groothuis, 1982; Tanimura, Matsui, & Kobayashi, 1990). These studies were conducted using hospital data. Population-based data on the relationship between child maltreatment and multiple births is limited. One possible reason is that prospective epidemiologic research on child maltreatment is difficult to conduct due to the underreporting of abuse cases.

It is of little doubt that the current situations surrounding families—for example, family planning, including assisted reproductive technology, child rearing, and maternal/paternal age—are quite different today. Recently, family size has rapidly become smaller, maternal and paternal ages at first childbirth are becoming older, and assisted reproductive technology has spread widely in Japan (Ooki, 2010; Ooki & Hiko, 2012).

In Japan the frequency of child maltreatment in cases of multiple births is around 10-fold higher than among singletons, according to the only hospital-based report done in Japan (Tanimura et al., 1990). With this background, the author performed secondary data analyses using nationwide published data (Ooki, 2013a, 2013b) to estimate the impact of multiple births in cases of fatal child maltreatment (child deaths by maltreatment). The relative risk (RR) of multiple births per individual was around 2.0-3.0, and the RR of multiple births per family unit was around 4.0-5.0 (Ooki, 2013a). It was reaffirmed that families with multiple births had an elevated risk for fatal child maltreatment both per individual and per family unit (Ooki, 2013a). Moreover, it was suggested that the nonspecific overburden of child rearing might be one reason for the higher frequency of child maltreatment for multiples compared to singletons; comparisons between the two twins might be another (Ooki, 2013b).

The purpose of this chapter is to further clarify the impact of multiple births on child maltreatment through a comprehensive literature review and use of updated nationwide data in Japan.

## LITERATURE REVIEW

### Materials and Methods

The comprehensive literature review was performed using PubMed on September 15, 2015. Both keyword search and the secondary snowballing method were used. Regarding the keyword search, papers that contained both (multiples or multiple birth\* or twin\* or triplet\* or plurality) and (child abuse or child maltreatment or infant abuse or infant maltreatment or shaken baby syndrome or SBS or traumatic brain injury or TBI or physical abuse or physical maltreatment or sexual abuse or CSA or neglect) in all fields were extracted and carefully checked. Language was restricted to English. All types of literature—not only original articles but also review articles and comments on previous publications—were included. We focused

only on child maltreatment at the family level and excluded child maltreatment at a sociocultural level, such as negative attitudes toward multiples (for example, neglect for one of the twin pair in some African areas) (Levy, 1964; Okeahialam, 1984). Twin studies that dealt with the effects of maltreatment in childhood on later life (e.g., Jaffee, Caspi, Moffitt, Polo-Tomas, & Taylor, 2007; Mostoufi et al., 2013; Nelson et al., 2002) were also excluded. All literature that showed a potential relationship between child maltreatment and multiple births was selected. Whether or not the researchers paid attention to the status of multiple births as a possible risk factor was not considered when selecting the papers, since the main purpose of this chapter was to get the widest range of useful information on the relationship between child maltreatment and families with multiples.

## RESULTS

Of 626 papers extracted by initial keyword search, 23 met the inclusion criteria (Table 1). Of the 23, there were 21 original articles and 2 comments on 2 different case reports. Six quantitative research papers analyzed hospital data with a variety of sample sizes, and 4 studies analyzed vital statistics/resident registry and/or annual reports of child maltreatment. Case reports totaled 7. No qualitative research, such as interviews of perpetrator mothers, was found to exist.

The first large study that treated the relationship between families with multiples (twins) and child maltreatment was that of Robarge, Reynolds, and Groothuis (1982) and their expanded study (Groothuis et al., 1982). The present author could find no systematic study of child maltreatment of multiple-birth children before these two studies.

Main findings of the literature review were as follows:

- 1) Families with twins or multiple births might have an elevated risk of child maltreatment.
- 2) No risk factor specific to families with multiples was detected.
- 3) Shaken baby syndrome (SBS) or inflicted traumatic brain injury (ITBI) might occur more frequently in the case of multiple births compared to singletons.
- 4) Most studies were performed in the US.
- 5) No quantitative studies of the earlier period used the age of victimized children as an inclusion criterion.
- 6) Eight out of 21 authors overlooked or paid scant attention to the twin/multiple status as a possible risk factor for child maltreatment.

## DISCUSSION

There have been very few studies on maltreatment in cases of multiple births, despite the general recognition that multiple birth increases the risk of child maltreatment. The first survey was performed in 1982, when assisted reproductive technology had not yet been widely spread.

Most quantitative studies to date have dealt with clinically severely maltreated cases. So, it would be useful to perform a questionnaire survey that asks about abusive attitudes of mothers or parents toward their children (for example, whether or not they feel they might have mistreated their children), and to compare the frequency of abusive attitudes of mothers of multiples to that of singletons. Performing detailed interviews of mothers of multiples as to the parenting problems of multiples or abusive attitudes would also be useful to learn the factors related to child maltreatment of multiples.

## SECONDARY DATA ANALYSES USING PUBLISHED DATA

### Materials and Methods

#### *Subjects*

National annual reports on fatal child maltreatment (the first to tenth reports) published from 2005 to 2014 by the Ministry of Health, Labor and Welfare of Japan (in Japanese) were used as the initial sources of information for the present data analyses. All cases of fatal maltreatment of children from 0 to 17 years of age between July 2003 and March 2013 were analyzed. The annual reports tally the cases of fatal child maltreatment (per individual or family unit) according to whether or not the child's death was accompanied by the parent's suicide. Cases of child murder followed by parental suicide were excluded from the present analysis, since the background and risk factors may be different from those in cases of fatal child maltreatment without suicide.

Fatal child maltreatment was defined as child death due to maltreatment. The definitions of maltreatment and parental guardian were based on the Child Abuse Prevention Law of Japan executed in 2000. The types of maltreatment included physical abuse, psychological abuse, neglect, and sexual abuse.

The numbers of women exhibiting any of about 20 physical and mental issues during pregnancy and the perinatal period were surveyed via questionnaire for the local public authorities, and the results were presented in the annual reports. The reported numbers of women with each issue do not necessarily show that that particular issue is a risk factor for fatal child maltreatment, since the frequency of each issue in the unexposed population or general population was not taken into consideration in the report. These data on physical and mental issues were not presented according to the ages of the victims. One limitation of this retrospective questionnaire survey was that there were many missing values among these data.

The number of multiple-birth cases according to calendar year of incidence was the only available information regarding multiples in the national annual reports. The present author performed an exhaustive information search regarding multiple-birth cases using keyword searches in an Internet search engine to create a full profile for each case. Several newspaper stories of each fatal maltreatment case were reviewed as reliable data sources, and the following information was gathered:

- 1) victim's sex/age
- 2) victimizer's age

- 3) relationship between victim and victimizer
- 4) family information
- 5) situation of maltreatment and cause of death
- 6) maltreatment of other children
- 7) background and risk factors
- (8) sentence

## STATISTICAL ANALYSES

### Relative Risks and Their 95% Confidence Intervals for Multiple Births

Multiple births, low birthweight (<2,500 g), and teenage pregnancy were the only variables for potential risk factors, the numbers of which in the general population at birth could be estimated using vital statistics. The author substituted childbirth below the maternal age of 20 in the vital statistics for teenage pregnancy.

The relative risks (RRs) and their 95% confidence intervals (CIs) in cases of fatal child maltreatment related to multiple births were estimated using fatal maltreatment data and vital statistics. The RRs of teenage pregnancy and low birthweight were also calculated to clarify the relative impact of multiple births on fatal child maltreatment. The data on multiple births and low birthweight were presented in all tenth reports, and teenage pregnancy was tracked beginning with the third annual report.

RR was calculated as the ratio of the incidence in the exposed population to that in the unexposed population according to the definition. Multiple births, teenage pregnancy, and low birthweight were regarded as risk factors against singleton births, non-teenage pregnancy, and non-low birthweight, respectively. The analyses were performed using the concept of the birth-year cohort. For example, the incidence in multiple births was calculated as the number of multiple-birth cases with fatal child maltreatment divided by the person-years of the birth-year cohort of the general multiple-birth population in the reported period (between July 2003 and March 2013). The incidence in singletons was calculated in the same manner. There were no data on the number of multiple births, birthweight, or maternal age for children from 1 to 17 years of age in the vital statistics. It was assumed that the percentage of the exposed population in the total general population at birth was constant for children from 1 to 17 years of age. For example, the percentage of multiple births in 2003 was used as the percentage of multiples of 1 year of age in 2004, 2 years of age in 2005, and so on. For the general population data, vital statistics from 1986 to 2013 were used, considering the year of the annual report and the age of the victims. Theoretically, the victims of 17 years of age in the first report (published in 2005) were born in 1986, and the victims of 0 years of age in the tenth report (published in 2014) were born in 2013. The follow-up period of the birth-year cohort was adjusted for the years 2003 and 2013 according to the research period (6 months and 3 months, respectively). The follow-up period of multiple births was distributed from 0.125 years (2013 cohort) to 9.75 years (1996-2002 cohorts) according to the birth year. Then the RR was calculated as the ratio of the incidence in the multiple-birth population to that in the singleton population. The RRs of low birthweight and teenage pregnancy were calculated in the same manner.

Regarding multiple births, all families with at least two live multiple births were recalculated using vital statistics of live births/stillbirths combination. The RR and 95% CI of multiple births were calculated per child unit (multiples as an individual child) and per family unit (families with multiples). When calculating RR per family, the total number of families was adjusted by considering the numbers of families with multiples.

### **Characteristics of Victims and Perpetrators**

Multiples and singletons were compared using the  $\chi^2$  test or Fisher's exact probability, with a threshold significance level of 0.05.

Multiples were also analyzed as pairs. The percentages and estimated population rate were calculated according to sex combination. The pairwise/probandwise concordance rate of maltreatment regarding twin pairs, including survival cases, was also calculated. In general, the pairwise concordance rate is the probability that both members of a twin pair are affected (maltreated) given that at least one member of the pair is affected. On the other hand, the probandwise concordance rate was the probability that a twin is affected given that his/her co-twin is affected. Only the probandwise concordance rate can be directly compared to the risk rate reported for other familial pairings and to population prevalence figures. Pairwise concordance rates were calculated as  $C/(C + D)$ , and probandwise concordance rates were calculated as  $2 \times C/(2 \times C + D)$ , where C denotes the number of affected concordant pairs and D denotes the number of discordant pairs (McGue, 1992).

## **RESULTS**

### **Relative Risks and Their 95% Confidence Intervals for Multiple Births**

The RRs and their 95% CIs are shown in Table 2. Most RRs were statistically significant regardless of the risk factors. The RRs of multiple births per individual were 1.5 (95% CI 0.9-2.5). The RRs of multiple births per family were 3.0 (95% CI 1.8-5.1). The RR tended to be much lower than the RR of teenage pregnancy, (RR = 12.9, 95% CI 10.1-16.4), but slightly higher than the RR of low birthweight (RR = 1.6, 95% CI 1.2-2.0).

### **Characteristics of Victims and Perpetrators**

There were 546 reported cases of fatal child maltreatment between July 2003 and March 2013. Among these were 15 multiple-birth victims. All 15 multiples were identified through an intensive Internet search. They were all twins, and originated from 14 families.

**Table 1. Literature Review Regarding Child Maltreatment of Multiple Births**

ID	Published year	Author(s)	Country	Multiple births as main topic	Data source	Age of inclusion	Number of multiples in which at least one child was maltreated	Main finding or suggestion regarding the relationship between multiple births and child maltreatment
1	1979	Lee	UK	Yes	case report	-	one twin pair	The mother had shown some evidence of a Munchausen tendency since the twins were taken into care.
2	1982	Robarge et al.	US	Yes	one hospital	not mentioned	38 pairs of twins	The birth of twins appeared to increase significantly the risk of subsequent child abuse.
3	1982	Groothuis et al.	US	Yes	two hospitals	not mentioned	48 pairs of twins	The twin status in and of itself contributed most significantly to subsequent abuse.
4	1982	Nakou et al.	Greece	No	referral to inter-disciplinary team	not mentioned	two pairs of twins	Two pairs of twins (four twins) were included in the 50 abused children.
5	1985	Nelson & Martin	US	Yes	one medical center	not mentioned	10 pairs of twins	An increased risk of child abuse occurs in families of twins.
6	1987	Spaide	US	No	case report	-	one twin pair	Shaken baby syndrome
7	1990	Tanimura et al.	Japan	Yes	nationwide hospital data	not mentioned	22 pairs of twins and one set of triplets	Twins are at high risk of child abuse and one rather than both of a pair of twins was more likely to be abused.
8	1992	Showers	US	No	case report	-	one twin pair	Shaken baby syndrome
9	1994	Hansen	US	Yes	case report	-	one twin pair	Shaken baby syndrome. A routine skeletal survey seemed advisable.
10	1996	Alexander	-	-	(comment on ID 9)	-	-	-
11	1998	Rooks et al.	US	No	case report	-	two pairs of twins	Cervical spine injury (shaken baby syndrome suspected)
12	1998	Becker et al.	Germany	Yes	case report	-	four pairs of twins	Twin status is an independent risk factor for shaken baby syndrome.
13	2000	Miller	-	-	(comment on ID 11)	-	-	-
14	2003	Keenan et al.	US	No	a population-based study	< 2 y	four twins	An increased risk of inflicted traumatic brain injury occurs in multiples.
15	2004	Wu et al.	US	No	statewide registry	< 1 y	1.26% of 4935 multiple births	Plurality was not selected as significant risk factor of infant maltreatment.
16	2006	Dhanani et al.	US	Yes	case report	-	one twin pair	Infralingual laceration. More intensive anticipatory guidance to parents of multiples before the newborn is initially sent home with the family needs to be provided..

**Table 1. (Continued)**

ID	Published year	Author(s)	Country	Multiple births as main topic	Data source	Age of inclusion	Number of multiples in which at least one child was maltreated	Main finding or suggestion regarding the relationship between multiple births and child maltreatment
17	2007	Luke & Brown	US	Yes	vital statistics	< 1 y	47 twins	The highest risk for infant mortality was among the youngest mothers (< 20 years).
18	2008	Talvik et al.	Estonia	No	two hospitals	-	three pairs	Families with twins are at additional risk for shaken baby syndrome.
19	2011	Nambu et al.	Japan	No	online newspaper database	-	one twin pair	The only case with a suspended sentence in their subjects of fatal child maltreatment was that of a mother of twins.
20	2012	Lindberg et al.	US	Yes	20 US child abuse teams	< 10 y	80 twin pairs and 2 sets of triplets	Twins are at higher risk of abusive fractures relative to nontwin contacts.
21	2013	Lang et al.	US	Yes	two hospitals	< 18 y	18 pairs of twins and one set of triplets	Siblings of maltreated multiples often were abused.
22	2013	Ooki	Japan	Yes	nationwide published data	< 18 y	14 pairs of twins	Families with multiples had elevated risk of fatal child maltreatment.
23	2013	Ooki	Japan	Yes	nationwide published data	< 18 y	14 pairs of twins	Non-specific overburden of child rearing might be one possible reason for higher frequency of child maltreatment of multiples.

**Table 2. Relative Risks and Their 95% Confidence Intervals for Teenage Pregnancy, Low Birthweight, and Multiple Births**

	Fatal child maltreatment <sup>a</sup>	Vital statistics Person-year	RR	95% CI
	N			
<b>Age of pregnancy</b>		(January 2005- March 2013) <sup>b</sup>		
over 20 years of age	393	164,792,117	1	(Reference)
below the age of 20	78	2,536,109	12.9	10.1-16.4
<b>Birthweight</b>		(July 2003-March 2013) <sup>b</sup>		
Non-low birthweight	479	182,904,683	1	(Reference)
Low birthweight	67	16,278,477	1.6	1.2-2.0
<b>Plurality</b>				
Per individual child		(July 2003-March 2013) <sup>b</sup>		
Singleton	531	195,492,217	1	(Reference)
Multiple births	15	3,690,943	1.5	0.9-2.5
<b>Per family</b>				
Families with singleton(s)	495	195,492,217	1	(Reference)
Families with multiples	14	1,845,822	3.0	1.8-5.1

a: Missing values were treated as unexposed cases.

b: Follow-up period is shown in parentheses.

RR, relative risk; CI, confidence interval.

**Table 3. Case Details of Fatal Child Maltreatment for Multiples**

ID	Victim		Victimizer (Age)	Other family members	Employed		Method of maltreatment and cause of death	Maltreatment of other children	Sentence (penal servitude)	Background and risk factors
	Sex	Age			Father	Mother				
1	F	4m	Father (26), Mother (34)	6 children including OS twin pair (two children from former marriage of mother)	No	No	Neglect	Yes (co-twin)	2 years and 6 months for both parents	Premature baby, many children, divorce and second marriage of mother, no support for child rearing from husband
2	F	6y	Father (32), Mother (31)	OS twin pair	Yes	No	Neglect, Physical maltreatment	No	3 years, suspended jail term (5 years) for both parents	Premature baby, cerebral palsy of co-twin, mental disorder of mother, no support for child rearing from husband
3	M	2y	Uncle (36)	aunt, three cousins, MM twin pair	-	-	Physical maltreatment	Unknown	4 years	Non-biological child of victimizer, many children
4	M	1m	Father (24)	mother (26), first-born daughter (2), MM twin pair	No	Unknown	Physical maltreatment	Yes (co-twin)	5 years and 6 months	Low ability of child rearing
5	F	1y11m	Mother (43)	OS twin pair	-	No	Physical maltreatment	No	5 years and 6 months	Impulsiveness, fatherless family.
6	M	8m	Father (29), Mother (21)	first-born daughter (1y 10m), MM twin pair	No	No	Physical maltreatment, Neglect	Yes (co-twin)	father 12 years, mother 8 years	Unexpected twins, teenage pregnancy, low ability to make living
7	M	2y	Mother (29)	first-born son (6y), OS twin pair	Yes	No	Neglect	Yes (co-twin, non-twin elder brother)	6 years	Divorce of victimizer's parents, domestic violence of victimizer's father, divorce of victimizer, low ability of child rearing
8	F	1y	Mother (23)	first-born son (3y), OS twin pair	-	Yes	Physical maltreatment	No	4 years	Teenage childbirth and divorce of victimizer, mental disorder of the victimizer, impulsiveness
9	F	3y	Mother (22)	father (33), OS twin pair, second-born daughter (10m)	Yes	No	Physical maltreatment	No	4 years and 6 months	Teenage pregnancy, delay of language development, child-rearing anxiety
10	F	4m	Mother (27)	grandmother (maternal) (60), father (35), FF twin pair	Yes	Yes	Physical maltreatment	No	3 years, suspended jail term (4 years)	Weight difference between twin pair, no support for child rearing from husband, child rearing anxiety
11	M	1m	Mother (29)	father (29), first born daughter (5y), second born daughter (2y), MM twin pair	Yes	No	Physical maltreatment	Yes (co-twin)	3 years, suspended jail term (5 years)	Fatigue from child rearing.

**Table 3. (Continued)**

ID	Victim		Victimizer (Age)	Other family members	Employed		Method of maltreatment and cause of death	Maltreatment of other children	Sentence (penal servitude)	Background and risk factors
	Sex	Age			Father	Mother				
12 13	M M	7m	Father (24)	mother (21), first born daughter (3y), first born son (2y), MM twin pair	No	Yes	Neglect	Yes (all four children)	No information	Divorce and second marriage of mother, teenage pregnancy, low ability to make living
14	F	6m	Mother (36)	father (34), FF twin pair	Yes	No	Physical maltreatment	No	3 years, suspended jail term (4 years)	Fertility treatment and fetal reduction, fatigue from child rearing, child-rearing anxiety.
15	M	2y	Father (27)	mother (22), MM twin pair, mother and brother of father	Yes	-	Physical maltreatment	No	A decision not to charge	Divorce and second marriage of father, suspect of motor developmental delay and repeated violence for the victim

ID number was assigned per child according to national reports. M, Male; F, Female; OS, opposite-sexed; y, year; m, month.

Table 3 shows the outline of cases (detailed information of ID-1 to ID-14 was reported elsewhere (Ooki, 2013b)). Of the 15 victims, 8 were male and 7 were female; 8 victims were 0 year old. Mean number of children in one family was 3.1 (= 44/14). All except one victimizer (uncle of twins) were the biological father/mother of the twin. The victimizers were mothers only (solitary murderer) in 7 cases, fathers only in 3 cases, and both parents in 3 cases. Five mothers had experienced teenage childbirth of twins. Four or five mothers had experienced divorce in their life course. Four fathers out of 11 did not have a regular occupation. The method of maltreatment was physical maltreatment in 11 cases and neglect in 5 cases (multiple answers allowed). One case (ID-14) was obviously a case of SBS.

**Table 4. Characteristics of Victims and Perpetrators of Fatal Child Maltreatment**

		Twins (N = 15 from 14 families)		Singletons (N = 531 from 495 families)		Total (N = 546 from 509 families)	Statistic test <sup>b</sup>
		N	%	N	%	N	
Sex of victim (per child)	Male	8	53	274	52	282	n.s.
	Female	7	47	242	46	249	
	Unknown	0	0	15	3	15	
Age of victim (per child)	1 year <=	7	47	289	54	296	n.s.
	<1 year	8	53	232	44	240	
	1-11 months	8	100	121	52	129	p<0.01
	0 month	0	0	111	48	111	
	Missing values	0	0	10	2	10	
Main perpetrator (per child)	Mother	7	47	297	56	304	n.s.
	Father	4	27	83	16	87	
	Mother and father	3	20	37	7	40	
	Others	1	7	98	18	99	
	Missing values	0	0	16	3	16	
Age of mother (per mother years)	≤19	0	0	32	9	32	n.s.
	20-24	5	36	88	24	93	
	25-29	4	29	81	22	85	
	30-34	2	14	59	16	61	
	35-39	1	7	64	17	65	
	≥40	1	7	34	9	35	
	Missing values/no mother	1	7	14	4	15	
	Not reported			123		123	
Age of father (per father years)	≤19	0	0	8	2	8	n.s.
	20-24	2	14	32	9	34	
	25-29	4	29	48	13	52	
	30-34	3	21	54	15	57	
	35-39	1	7	37	10	38	
	≥40	1	7	52	14	53	
	Missing values/no father	3	21	141	38	144	
	Not reported			123		123	
Number of child/children in one family <sup>d</sup>	1	0	0	300	40	300	p<0.01
	2	6	38	250	33	256	
	3	6	38	93	12	99	
	4 or more	4	25	50	7	54	
	Missing values	0	0	58	8	58	
	Not reported			24		24	

**Table 4. (Continued)**

		Twins (N = 15 from 14 families)		Singletons (N = 531 from 495 families)		Total <sup>a</sup> (N = 546 from 509 families)	Statistic test <sup>b</sup>
		N	%	N	%		
Psychological problem of mother <sup>e,f</sup> (per mother, multiple answers allowed)	Child-rearing anxiety	5	36	107	25	112	n.s.
	Impulsiveness	2	14	54	13	56	
	Depressive tendency	2	14	45	11	47	
	Mental disorder	2	14	43	10	45	
	Low ability of child rearing	4	29	114	27	118	
Marriage status of rearer <sup>e</sup> (per family)	No mother	1	7	15	4	16	
	Biological parents	8	57	187	44	195	n.s.
	Divorce	1	7	49	12	50	
	Common-law or second marriage	3	21	66	16	69	
	Others	2	14	75	18	77	
Main method of maltreatment (per child)	Missing values	0	0	46	11	46	
	Not reported			72		72	
	Physical maltreatment	10	67	355	67	365	n.s.
	Neglect	5	33	140	26	145	
	Others	0	0	1	0	1	
Maltreatment for other brothers/sisters <sup>e, g</sup> (per sibling)	Missing values	0	0	35	7	35	
	Yes	9	30	84	28	93	n.s.
	No	15	47	82	27	97	
	Missing values	6	23	138	45	144	
Occupation of mother (per mother)							
	Yes	3	21	94	22	97	n.s.
	No	8	57	204	48	212	
	Missing values	3	21	125	30	128	
Occupation of father (per father)	Not reported			72		72	
	Yes	7	50	152	36	159	n.s.
	No	4	29	33	8	37	
	Missing value/fatherless family	3	21	238	56	241	
	Not reported			72		72	

a: Total number of children was based on the national annual reports on fatal child maltreatment presented by the Ministry of Health, Labour and Welfare of Japan (in Japanese).

b: Missing values were excluded in the statistic tests.

c: Fourth report and after.

d: Second report and after. The number includes parent-child murder-suicide.

e: Third report and after.

f: The denominator was 426 regarding mothers of singletons.

g: Not necessarily fatal maltreatment. Regarding ID 12 and 13, one twin was treated as proband.

Premature birth (ID-10), having a disabled co-twin (ID-2), delay of physical growth (ID-10), motor development delay (ID-15), language development delay (ID-9), and parental disfavor (ID-8) tended to be associated factors in maltreatment in cases when only one twin was maltreated. On the other hand, family dysfunction, namely lack of ability to make a living, including no regular occupation of father (ID-1, ID-4, ID-6, ID-12, ID-13), lack of child-rearing ability of mother/father (ID-7), and unexpected childbirth (ID-6), were observed when both twins were maltreated.

The sentence for the victimizer was identified for 12 families. Four families were given suspended sentences, representing 33% (4/12) of all families and 43% (3/7) of the cases with the mother as solitary murderer.

**Table 5. Characteristics of Twin Pairs**

		Twins (N = 15 from 14 families)		Statistical test
		N	%	
Sex combination of twin pair	Opposite sex	6	43	0.17-0.69
	Same sex	8	57	0.31-0.83
	Male-male	6		
	Female-female	2		
Sex of twins	Male of opposite sex	1	7	0.00-0.19
	Female of opposite sex	5	33	0.09-0.57
	Male of same sex	7	47	0.21-0.72
	Female of same sex	2	13	0.00-0.31
Concordance/ Discordance	Both twins maltreated (C)	6	43	
	One twin maltreated (D)	7	50	
	Missing values	1	7	
	Pairwise concordance rate		46	
	Probandwise concordance rate		63	

Missing values were excluded in the statistical tests.

The results of statistical tests and estimations of individual multiples are shown in Table 4. No significant difference between twins and singletons with fatal maltreatment was observed for most items. However, no 0-month victim was observed among twin victims ( $p < 0.01$ ), which produced statistical significance. Also, the number of children per family was larger in families with twins ( $p < 0.01$ ). No specific feature was observed regarding psychological problems of mothers.

Multiples were also analyzed as pairs. The results are shown in Table 5. The percentage of male victims of opposite-sex pairs was 7% (1/15, 95% CI, 0.00-0.19). Female victims of same-sex pairs was 13% (95% CI, 0.00-0.31). Other siblings of victims in 13 pairs (one unknown: ID3) were also maltreated in 6 cases, including all co-twins of the 6 index twin victims. Thus, the pairwise and probandwise concordance rate of child maltreatment, where index twins were fatal, was 46% (6/13) and 63% (12/19), respectively. The probandwise concordance rate was higher than the prevalence of the total subjects (49%, 93/190, calculated by the value in Table 4).

## DISCUSSION

### Relative Risks and Their 95% Confidence Intervals for Multiple Births

The present data showed that families with multiple births had increased risk of fatal child maltreatment. The RR, however, was not higher than the RR of teenage pregnancy. The results also showed that the RR of multiples per individual, namely of being a child member

of a multiple birth, showed marginal significance and was not largely different from the RR of low birthweight.

Regarding the work of Robarge et al. (1982) and Groothuis et al. (1982), their research interest was not necessarily twins as a risk factor for child maltreatment, but the stressful situation associated with the birth of twins due to the increase in family members, inadequate spacing of children, and rearing more than one infant at a time. Although their questionnaire survey for mothers was hospital-based, their results suggested that the proportion of child maltreatment in families with twins was higher than in families with singletons. The noteworthy finding was that the twins themselves were not necessarily abused, but rather the siblings of twins were. This means that having twin children can result in a reduction of the time and energy that the mother has for meaningful relationships with the father and other siblings within the family unit (Robarge et al., 1982). The present study design could not verify their findings, since no data were available as to whether victimized singletons are siblings of multiple births.

On the other hand, Nelson and Martin (1985) reported that of 310 registered abused/neglected children, 16 (5.2%) were twins, which was about 2.5-fold higher than the approximated general percentage of twins (2%). They concluded that twins themselves were also at high risk, supporting the result of Nakou, Adam, Stathacopoulou, and Agathonos (1982). The data of Nakou et al. (1982) showed that 4 out of 50 registered abused children were twins, although the authors paid no attention to this finding. It is not surprising that multiples themselves are at high risk, since multiples had many general risk factors for child maltreatment, for example, low birthweight, prematurity, birth defects, neonatal complications, and so on. According to the nationwide hospital-based data provided in 1986 by Tanimura et al. (1990) of 231 children subjected to abuse or neglect, 23 (10.0%) were products of multiple births (22 were twins). They compared this percentage to that of twin deliveries (number of mothers) in the general Japanese population (0.6%). They should have compared the percentage with that of live multiple births, since their research interest was the risk of being abused as a twin, not the risk of abuse occurring in families with twins. According to the vital statistics, the percentage of multiple live births among total live births in 1986 was 1.4%. The percentage of twins in the maltreated population was thus around 7-fold (10.0/1.4) higher than in the general population.

It is important to note that the ratio of the percentage of specific factors in child maltreatment cases to the percentage in the general population—for example, the percentage of multiple births in child maltreatment cases divided by the percentage of multiple births in the general population at birth—does not yield the correct estimation of RR. This method gives an alternative underestimation of RR, since this method did not consider the percentage of the singleton (unexposed) population and the age of the subjects, although the degree of underestimation seemed not to be fatal. This method has been used several times in studies of the child maltreatment of twins (Nelson & Martin, 1985; Tanimura et al., 1990; Talvik, Alexander, & Talvik, 2008).

Using the data presented by Luke and Brown (2007), the percentages of total maltreatment deaths before 1 year of age among singletons and multiple births from 1995-2000 in the US were recalculated as 0.0232% (4,325/18,636,575) and 0.0607% (47/77,460), respectively, which produced an RR of 2.62 with 95% CI 1.96-3.49 per child. This value is slightly higher than the present result, but not higher than that estimated by Tanimura et al. (1990), although the age distribution of the victims was very different. The difference among

the present data, the data of Luke and Brown (2007), and the data of Tanimura et al. (1990) was that the former two data sets corresponded to fatal child maltreatment, i.e., child deaths, and the latter corresponded to survivors of maltreatment admitted to the hospital. The higher proportion of twins in the data of Tanimura et al. (1990), however, was not rationally explained by this difference in the data. One possible explanation is that multiples in general might be admitted into the hospital compared to singletons due to reasons other than child maltreatment, and thus they were apt to be over-ascertained. More research should be performed on multiple-birth status among the survivors of child maltreatment.

Most previous clinical studies focused on multiple births per child. This is not necessarily appropriate from the public health or preventive medical point of view, because most difficulties in child rearing related to multiple births were due to the rearing of more than one child of the same age at the same time in the same family (Bryan, 2003; Ellison et al., 2005; Ooki, 2009, 2010; Ooki & Hiko, 2012; Pinborg, Loft, Schmidt, & Andersen, 2003). For example, the comparison of two infants (twins) consisting of one low-birthweight twin and one non-low-birthweight twin sometimes is a source of stress for mothers. These anxieties or feelings of stress may not be induced if rearing only one low-birthweight singleton. If multiple births were treated as individual births, the associated risk of rearing two or more children of the same age at the same time in the same family would be underestimated. The rapid increase of iatrogenic multiple births is now a public health concern, one that goes beyond purely obstetric problems (Ooki, 2011a, 2011b; Ooki & Hiko, 2012). Nevertheless, this serious situation is rarely recognized not only among child support members, but also among professionals in the field of parent and child health and even in families with multiples themselves (Ooki, 2009, 2011a, 2011b; Ooki & Hiko, 2012).

According to recent vital statistics, the total fertility rate tended to decrease and fell to below 2 over a long period in Japan (1.4 in 2014). This suggests that the risk of having at least one maltreated baby in one family may become higher in families with multiples, which have at least two children, than in families with singletons.

The present results also showed that teenage pregnancy was a significant risk factor for fatal child maltreatment. Luke and Brown (2007), using US vital statistics, showed an increased risk of infant maltreatment deaths among healthy, full-term singletons among those born to mothers aged 20 and younger. The pattern for twins was similar and more noticeable, with elevated risks among younger mothers.

Most of the limitations of this chapter could be attributed to the data collection system itself. Although this chapter was based on the annual reports of nationwide survey, the data gathering was far from comprehensive. The high percentage of missing values of all three risk factors showed the difficulties of gathering data on child maltreatment. The present RR should be interpreted as the general tendency of these three risk factors.

Many of the problems that occur during pregnancy and the perinatal periods are associated with one another. For example, multiple births are associated with many perinatal problems, such as low birthweight, Caesarean section, neonatal asphyxia, impending abortion/threatened premature delivery, and pregnancy hypertension. For instance, about 70% of multiples are low birthweight in Japan (Ooki, 2010). Being a member of a multiple could be considered an additional risk factor for low birthweight. The present aggregation of data cannot permit multivariate analyses restricting the confounding factors.

According to the study of Wu et al. (2004) of 15 perinatal and sociodemographic variables studied, 11 were found to be significantly related to infant maltreatment. Five

factors had adjusted RRs of 2 or greater: mother smoked during pregnancy (RR 2.8); more than two siblings (RR 2.7); medical beneficiary (RR 2.1); unmarried status (RR 2.0); and low-birthweight infant (RR 2.0). Although the RR of multiple births per individual was 1.5, the effect of plurality was not statistically significant, likely due to the small percentage of multiple births (2.6% of all subjects) compared to other risk factors.

According to the report by Schnitzer, Covington, Wirtz, Verhoek-Oftedahl, and Palusci (2008), no single data source was adequate to provide thorough surveillance of fatal child maltreatment, but combining just two sources substantially increased case ascertainment. Unfortunately, most record linkage, including that between birth records and child maltreatment, was almost impossible in Japan. The assumption that the percentage of the exposed population in the general population was constant for children from birth to 17 years of age, which was made in the calculation of RR, was not necessarily appropriate. The percentage of the exposed group might gradually decrease with age, since the children in the exposed group would die more frequently compared to the children in the unexposed group because of reasons other than child maltreatment, especially at an earlier age. This seemed, however, to have little effect on the present results, since fatal child maltreatment is very rare, and the mortality rate of children themselves is extremely low in Japan.

## Characteristics of Victims and Perpetrators

### *Risk Factor of Maltreatment as Individual Multiples or Families with Multiples*

Most of the child and parental factors related to fatal maltreatment did not differ between cases with twin and singleton victims. The only two differences that emerged were that 0-month victims were rarer and the number of children per family was larger in multiple-birth cases than singleton cases. The mean number of children per family was significantly larger for the multiple-child subset compared to total reported child fatal maltreatment cases (Table 4) or the Japanese general population (mean number is 1.4 according to 2014 vital statistics). In the case of families with multiples, parent(s) must rear at least two children of the same age at the same time. It was pointed out decades ago that close spacing of children might be a significant risk factor for subsequent child maltreatment (Robarge et al., 1982). The annual reports by the Ministry of Health, Labor and Welfare of Japan also pointed out that having more than one child, including multiples, is one of the risk factors for child maltreatment, although the evidence was not shown by the Ministry. The present results support this point.

In the case of ID-14 in Table 3, the mother, aged 36, was primipara and had used assisted reproductive technology and fetal reduction. Recently, assisted reproductive technology has become one of the typical contexts of families with multiples in Japan (Ooki, 2011a, 2011b). Given this fact, candidates for assisted reproductive technology should receive pre-treatment counseling, informing them of the frequency of multiple births and helping them assess their readiness for multiple-birth child rearing.

It was suggested that in the case of families with multiples, physical, mental, and social overburden for child rearing (Bryan, Denton, & Hallett, 1997; Denton, 2005; Ooki, 2009; Ooki & Hiko, 2012) in general rather than specific factors to multiples might be one of the risk factors for child maltreatment.

### ***Risk Factor of Maltreatment as a Twin Pair***

Previous hospital-based (as opposed to population-based) studies have found that in twin pairs, both twins tend to be maltreated on one level or another (mild, moderate, and severe). Both twins were found to be maltreated in 3 pairs out of 5 pairs (Groothuis et al., 1982) and in 6 out of 10 pairs (Nelson & Martin, 1985). This tendency, however, was not specific to multiples, since about half of the siblings of index singletons were also maltreated, as shown in Tables 3 and 5.

On the other hand, according to Tanimura et al. (1990), only 4 pairs (both twins) were maltreated out of 21 pairs (19%), indicating that one rather than both of a pair of twins was more likely to be maltreated in Japan. The present results did not support this indication. The difference between the study in this chapter and that by Tanimura et al. (1990) was that the former study included at least one fatal case in all 14 pairs.

Tanimura et al. (1990) also reported that whether one or both twins were maltreated was influenced by different underlying factors. Maltreatment of both twins occurred when the maltreating parent had psychosocial problems or when the family was beset by serious socioeconomic difficulties. When only one twin in a pair was maltreated, only one member, whether he/she was maltreated, seemed to have problems relating to child rearing. The present result was in good accordance with this finding.

One specific feature related to the rearing of twins is the comparison of growth and development of two children of the same age in the same family. This comparison sometimes imparts complicated stresses, anxieties, irritation, and a sense of guilt to parents, especially the mother (Bryan et al., 1997; Denton, 2005; Ooki & Hiko, 2012). In these cases, the delayed child tends to be the one who is maltreated. On the other hand, in the case of ID-2, the normal twin with a disabled co-twin was maltreated, because the victim could not live up to the mother's over-expectations. Thus, in these cases the mental condition of the mother was a complicating factor.

There have been no vital statistics showing the precise percentage of opposite-sex pairs among total twin pairs. An estimated one-third of all twin pairs in Japan are opposite-sex, since two-thirds of all pairs are estimated to be dizygotic (Ooki, 2011a, 2011b). The approximate percentage of male twins or female twins of opposite-sex pairs in the general twin population is 17% (1/6), and percentages of male twins and female twins of same-sex pairs in the general twin population are both equal to 33% (1/3). The present percentage of female victims of same-sex pairs ( $2/15 = 13\%$ , 95% CI 0.00-0.31) seemed low in relation to the approximate percentages in the general twin population (33%); the reasons for these low percentages are unclear.

### ***Case Reports and Method of Maltreatment***

Few case reports are available that describe the methods used to abuse multiples (Dhanani, Nield, & Ogershok, 2006). Of them, shaken baby syndrome (Becker, Liersch, Tautz, Schlueter, & Andle, 1998; Hansen, 1994; Showers, 1992; Spaide, 1987; Talvik et al., 2008), cervical spine injury (Rooks et al., 1998), and Munchausen syndrome (Lee, 1979) have been reported.

Abusive head trauma is a form of child physical abuse that involves inflicted injury to the brain and its associated structures. Colloquially called shaken baby syndrome (SBS), abusive head trauma is the most common cause of serious or fatal brain injuries in children aged 2 years and younger (Hinds, Shalaby-Rana, Jackson, & Khademian, 2015). Becker et al. (1998)

reported 5 twins severely affected by SBS, from among 4 pairs of twins. According to their discussion, twin status could influence the quality of some psychological aspects of the victimizing parent(s); a related factor promoting the formation of a stressful situation and familial dysfunction was the parents' social isolation resulting from the increased demands of time and energy involved in caring for twins. According to the computerized medical literature research by Becker et al. (1998), two case reports (Showers, 1992; Spaide, 1987) on twins with SBS were found, but the authors attached no special value to the victims' status as multiples. According to Rooks et al., (1998), the likely mechanism of the cervical spine injury of their two cases of twins was severe hyperflexion compression insult from violent shaking. Although these reports suggested that the prevalence of SBS in maltreated twins/multiples might be higher than that of singletons, no epidemiologic evidence was reported.

According to the population-based study of inflicted traumatic brain injury (ITBI) in young children by Keenan et al. (2003), relative to the general population, products of multiple births incurred an increased risk of ITBI. More recently, Talvik et al. (2008) studied the relationship between SBS/ITBI and the typical pattern of babies' crying behavior in two tertiary centers for children in Estonia. In their report, 4 children out of 26 cases of SBS/ITBI in Estonia were twins (15.4%), a much higher percentage compared to the general twin frequency in the country (2.1%). They found that the time curve of crying (minutes crying per day according to age in weeks) was similar to the curve of the incidence of SBS/ITBI cases (number of SBS/ITBI children according to age in weeks), which both reached their peak at 6 or 7 weeks. They concluded that families with twins are at additional risk for SBS/ITBI and that parental complaints of excessive crying by their infants should be taken as a signal that they need counseling.

In this chapter, one fatality (7% = 1/14) was caused by SBS. The sixth annual reports, which represented all fatal maltreatment of children from 0 to 17 years of age between April 2008 and March 2009 in Japan, presented the total number of deaths from SBS. According to this report, SBS as a cause of death in maltreatment cases under 3 years of age was 1% (1/86). Compared with the result of this chapter, this figure suggests a higher prevalence of SBS in fatal child maltreatment of multiples. Further study is needed on this relationship, as well as further research on the crying patterns of multiples in general.

According to the recent epidemiologic study by Lindberg et al. (2012), who examined physically abused children with serious injuries, twins were at substantially increased risk of abusive fracture relative to non-twin victims, although this study did not present information on the pairwise concordance/discordance of twins.

Recently, Lang, Cox, and Flores (2013) analyzed children's hospital data retrospectively and found that multiple-gestation-birth maltreated children were significantly more likely than single-birth maltreated children to have abdominal trauma (13% vs. 1%, respectively;  $p < 0.01$ ) and fractures (83% vs. 39%;  $p < 0.01$ ) and to be injured at a younger mean age (12.8 months vs. 34.8 months;  $p < 0.01$ ). The mean age of child death in the present sample was also significantly younger as to multiples than singletons (16.9 months vs. 33.2 months;  $p < 0.01$ ); however, this result should be interpreted with caution because the range of age at child death was very small for multiples compared to singletons. Multiple births ranged in age from 1 month to 6 years regarding present data and from 0 to 7 years in Lang et al. (2013). On the other hand, singletons ranged in age from 0 months to 17 years regarding present data and from 0 to 16 years in Lang et al. (2013). Mean age might not be the best statistic to estimate the average age at child maltreatment, since the age distribution has positive skewness,

especially in the case of singletons. For the sake of reference, the median age of child death in the present sample was 8 months in multiples and 18 months in singletons.

### ***Sentencing for Fatal Maltreatment***

According to the Japanese study by Nambu et al. (2011), the sentences handed down by the court clearly tended to be more lenient for female offenders of fatal child maltreatment. They noted that the only case with a suspended sentence out of 24 cases of fatal child maltreatment from January 2008 to December 2009 was that of a mother of twins (this case is ID-10 of this chapter). This may be due to the popular belief that men are partly responsible for any misconduct committed by their wives (Nambu et al., 2011). The present result suggested that this tendency might be especially noticeable in the cases of mother as solitary murderer of a multiple, possibly due to consideration of the severe conditions such mothers face.

## **LIMITATIONS**

The limitations of this chapter can be attributed to the data collection itself. First, although all 15 victims counted in the annual reports were identified, the author could not ascertain directly whether these cases were indeed the ones represented in the annual reports. Second, although the present dataset was from a multiyear nationwide survey, it still did not have sufficiently high statistical power to detect the statistical significance. The present results might be strongly influenced by chance factors.

## **CONCLUSION**

The results of the comprehensive literature review consistently showed that families with multiples have elevated risk of child maltreatment. Child maltreatment has a wide spectrum of phenomena, from parents' general abusive attitude to their children to fatal child maltreatment. The abusive attitude of mothers or parents of families with multiples has not been sufficiently studied. More research, both population-based and hospital-based quantitative studies and qualitative studies with detailed interviews, is needed.

Recent Japanese nationwide data showed that families with multiple births had elevated risk for fatal child maltreatment, but this risk was not as high as previously thought. Multiple births should be considered a risk factor for child maltreatment, not only per individual child but also per family unit. Moreover, in the case of child maltreatment for multiple births, the nonspecific overburden of child rearing might be one possible risk factor and parental comparisons between twins might be another. Among fatal maltreatment cases, 0-month victims were rarer and the number of children per family was larger.

Health care providers should be aware that multiple pregnancies/births may place significant stress on a family (Ooki, 2009; Ooki & Hiko, 2012), and these situations might make families with multiples a potential high-risk group of child maltreatment. In this light, health care providers should offer appropriate support and intervention beginning with pregnancy.

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## Conflict of Interests

The author declares no conflict of interests.

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