Doctoral Dissertation

Preventing children from harmful experiences

in early stage of life

（乳幼児を有害な経験から予防するための関連要因についての研究）

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Chapter 1: Introduction

1. CHILDREN'S RIGHTS AND THEIR BASIC NEEDS

Children are the "fortune" of our society.   They have right to live healthily, and be educated and protected.   Children's rights are guaranteed by the Convention on the Rights of the Child (CRC).   The CRC was adopted by the United Nations General Assembly in 1989, and ratified by most countries all over the world.   The CRC set up child protections for the basic human needs of life: development, well-being, and participation.[1]   Protection for children should be a self-evident truth and society should be responsible for it.   Maslow suggested that all human beings have five basic human needs, regardless of culture.   This theory is known as Maslow's Hierarchy of Needs, which describes the following five needs in a hierarchy: physiological needs, safety needs, belongingness and love (social needs), esteem needs, and self-actualization (Figure 1).[2]   The most primitive needs, physiological needs, include needs such as food, water, oxygen, an optimal temperature, and sleeping environment to maintain children's lives.   The second level of needs includes security, protection, and freedom from anxiety or fear.   The third level of needs includes the need to belong to others, and to feel loved by a group.   A group means belonging to a family, peer members, work group, community clubs, and so on. The fourth level, esteem needs, includes self-esteem based on one's accomplishments, status, or appearance.   The highest need is self-actualization, i.e., to accomplish one's achievements with one's potential and unique talent.[3] Maslow explains prepotency on needs; that is, the next level of needs emerge after the first need is fulfilled. In general, basic needs are supposed to be guaranteed step-by-step, from the bottom level to higher-level needs.

Figure 1. Maslow's Hierarchy of Needs
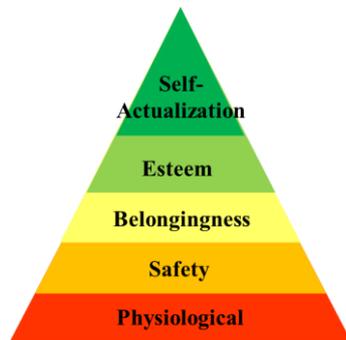
(Maslow AH, *Motivation and Personality,* 1970) [2]

2. HARMFUL EXPERIENCES FOR CHILDREN

However, there are various invasions of children's rights and demands to have basic human needs met. Difficulties that threaten children's rights to live, survive, and safely develop are, for example, child labor and exploitation, limited access to education, lack of access to health care, child maltreatment, involvement of children in armed conflict, child prostitution and pornography, discrimination including bullying, poverty, and others.[4, 5] Among them, unintentional injury is a leading cause of death and disability among children across the world.[6] If safety needs are fulfilled effectively, injuries may be preventable. Preventing child injury is closely connected to improving the situation of child mortality, disability, and the general well-being of children. Besides, child maltreatment takes place in all societies including high-income countries.[1] Child maltreatment may be also preventable if physiological needs, safety needs, belongingness needs, and self-esteem needs are sufficiently protected.

2.1. Unintentional injury

One variety of harmful experience is unintentional injury. Unintentional injury includes transport accidents, falls, accidental drowning or submersion, other accidental threats to breathing, exposure to smoke, fire and flames, and others (i.e., poisoning, etc.). Unintentional injury is a leading cause of mortality in Japan. The infant mortality rate (less than 1 year old) was 9.0/100,000, and 2.9 for children aged 1 to 4 in 2012 as shown in Table 1.[7] Tanaka reported that there were 200–300 excessive infant deaths if the infant fatality rate from unintentional injury is compared to what it is in countries with a lower fatality rate, such as Sweden or England.[8] Therefore, preventing child death due to unintentional injury is an important public concern.

Table 1. Top five causes of child death in Japan, 2012 (Mortality rate per 100,000)

	1	2	3	4	5
0 year	Congenital abnormalities (77.8)	Respiratory disorder (30.2)	Sudden Infant Death Syndrome (13.6)	Unintentional injuries (9.0)	Bleeding disorder (7.8)
1-4	Congenital abnormalities (4.2)	Unintentional injuries (2.9)	Malignant neoplasm (2.4)	Cardiac disease (1.4)	Pneumonia (1.1)
5-9	Unintentional injuries (1.9)	Malignant neoplasm (1.6)	Congenital abnormalities (0.7)	Other neoplasm (0.6)	Pneumonia (0.5)
10-14	Malignant neoplasm (1.9)	Unintentional injuries (1.6)	Suicide (1.3)	Cardiac disease (0.4)	Cerebrovascular disease (0.3)
15-19	Suicide (8.5)	Unintentional injuries (5.7)	Malignant neoplasm (2.8)	Cardiac disease (1.0)	Congenital abnormalities (0.5)

Vital statistics 2012. (Ministry of Health, Labour and Welfare. 2014)[7]

2.2. Child maltreatment

With regard to child maltreatment in Japan, the number of reports to the Child Guidance Center (similar to the Child Protection Service in the USA) is increasing rapidly after the Child Abuse Prevention Act was enforced in 2000 (Figure 2).[9] Child maltreatment includes physical abuse,

psychological abuse, sexual abuse, and neglect.   These are presented in a spectrum in Figure 3.

There are two conceptual axes in child maltreatment: "commission or omission" and "physical or

psychological."   Abused children are harmed or traumatized, physically, psychologically, or both

physically and psychologically.   Common concepts in every type of child abuse and neglect are

that children are unjustly "maltreated," though they are supposed to be protected, loved, and

encouraged to develop.

Figure 2. Number of child maltreatment consultations to the Child Guidance Center
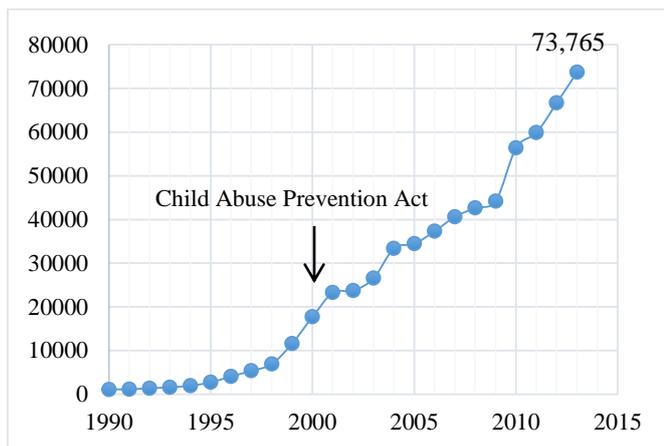
Number of reports to the Child Guidance Center in 2013 (Ministry of Health, Labour and Welfare. 2014) [9]

Figure 3. The spectrum of child maltreatment
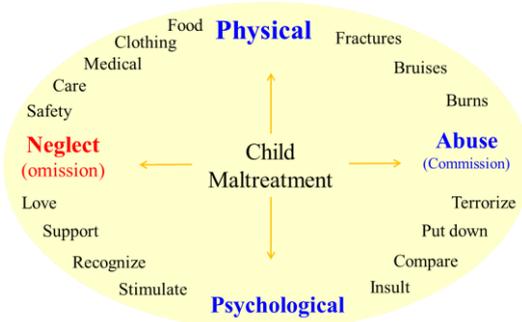
(Kliegman RM, *Nelson Textbook of Pediatrics,* 2011) [10]

3.   RELATION BETWEEN UNINTENTIONAL INJURY AND CHILD MALTREATMENT

In this way, both unintentional injury and child maltreatment are harmful experiences for children in Japan.   In the first place, what kind of relationships may exist between child maltreatment and unintentional injury?

Among the four forms of child maltreatment, neglect is an act of omission in a positive manner or negative manner.   There are two major types of neglect: failure to provide and lack of supervision, according to a coding scheme used in the USA, Modified Maltreatment Classification System (MMCS).[11]   Unintentional injury may partially overlap with the meaning of lack of supervision or failure to provide safety measurements, for example, if a child drowns during an unsupervised bath or is injured by a motor vehicle accident without a child safety seat.   In other words, unintentional injury may exist in close connection with neglect, and to some extent represent parents' neglectful behavior (Figure 4).

Figure 4. The relation between child maltreatment and unintentional injury
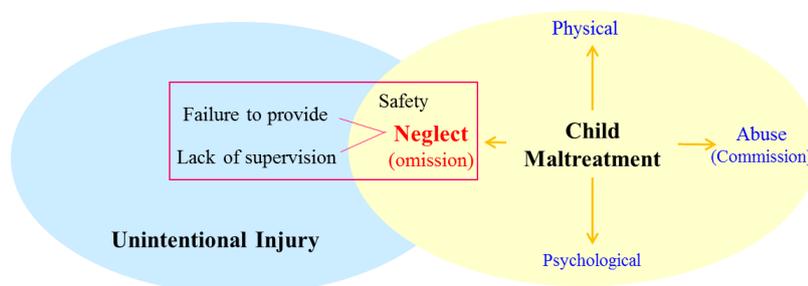
Modified from the spectrum of child maltreatment in Figure 3

Furthermore, Sheets et al.[12] found that 27.5% of abused infants had experienced a previous injury such as bruising, intraoral injury, and others as shown in Figure 5.   A previous minor injury in precruising infants (infants unable to pull to a stand and walk while holding onto something) was defined as a "sentinel injury," namely, one which had been visible before admission due to physical abuse.   Sheets et al. reported that detection and management of sentinel injuries could have the potential to prevent physical abuse.   However, the relationship between neglect and sentinel injuries has not been sufficiently reported so far.   Although minor injury may be a sentinel form of physical abuse, severe injury may result in children's death, as the ultimate outcome of both child abuse and neglect.

One prospective cohort study[13] in California indicated that children with a prior allegation of maltreatment faced a significantly higher risk of death from intentional injury (HR, 95% CI; 5.9, 4.4–7.8) and unintentional injury (HR, 95% CI; 2.0, 1.7–2.4) before the age of five, compared to children without such an allegation.   Comparing types of child maltreatment, children with allegations of physical abuse were significantly more likely to die with intentional injury (HR, 95% CI; 5.2, 3.6–7.6), and less likely to die with unintentional injury (HR, 95% CI; 0.6, 0.39–0.90), compared to children with allegations of neglect[14] as shown in Figure 5.   This study indicated that neglected children tend to die due to unintentional injury compared to physically abused children.

Figure 5. The relation among sentinel injury, child abuse and neglect, and injury death

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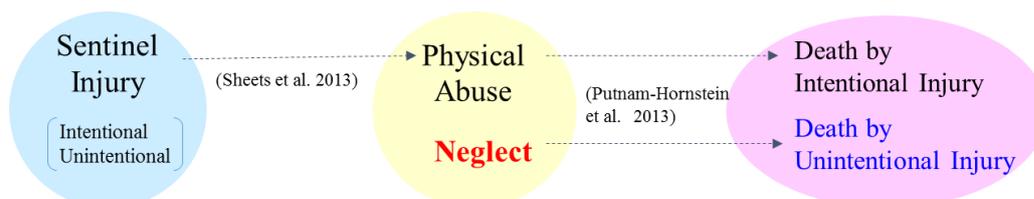
Although the relation between sentinel injury and neglect is not sufficiently understood, the risk factors of unintentional injury and child neglect may be similar. Unintentional injury and child neglect may occur and be exacerbated independently, simultaneously, or successively to each other. Therefore, it is important for health professionals to recognize that unintentional injury and child neglect can occur in similar situations.

4. DEVELOPMENTAL AND LONG-TERM EFFECTS OF UNINTENTIONAL INJURY AND CHILD NEGLECT ON CHILDREN

How do harmful experiences such as unintentional injury and child neglect affect children? Most of a child's life and activities depends on care provided by parents or caregivers. Children develop rapidly, especially as infants and toddlers. The milestones of motor function development among infants and toddlers are, for example, being able to roll over by 6–7 months (96%), being able to pull themselves up to a standing position by 11–12 months (97%), and being able to walk by 15–16 months (93%).[15] This is the reason unintentional injury needs different preventive strategies according to the monthly age and developmental stage of the child. In addition, children's developmental stage matters when considering the effects of child maltreatment as well. Child maltreatment has various effects on the development of the brain,[16] and the infant brain is fragile and damage lasts for a longer period. For example, Bremner et al.[17] reported that hippocampal volume in adult survivors of childhood abuse (physical or sexual) was decreased, and that volume reduction was correlated with the duration of abuse. Teicher et al.[18] found that neglect had the greatest effect on the reduction of the size of the corpus callosum (CC), compared

to physical abuse and sexual abuse.　　Reduced CC size decreases communication between the cortical hemispheres, and decreases right/left cortical integration.[19]　　While neurobiological changes are occurring in the brain, how does neglect affect children's development in cognitive, social, or behavioral ways?　　Neglect in infants and toddlers has a negative impact on their cognitive[20] and language[21] development, and emotionally neglected children are especially likely to show disorganized attachment.[22]　　Therefore, preventing the harmful effects of neglect is very important, particularly in infants and toddlers.

In addition to effects on child's development, several researches have reported that child maltreatment has long-term effects as well.　　One landmark research is the Adverse Childhood Experiences (ACE) study. [23]　　This study was conducted to assess the associations between adverse experiences in childhood and problems of health and well-being in adulthood.　　ACEs include psychological abuse, physical abuse, sexual abuse, and household dysfunction: violent against of mother, living with household members who were substance abusers, mentally ill or suicidal, or ever prisoned.　　Persons who experienced four or more ACEs not only had higher risks of behaviors (e.g., alcoholism, drug abuse, depression, suicide attempt), but also higher risks of developing diseases in adulthood (e.g., ischemic heart disease, cancer, chronic lung disease).　　ACEs have inerasable effects throughout one's life, and may result in early death (Figure 6).　　In this way, neglect has a negative impact on early development, and infant injuries may lead to disability in later life.　　Therefore, preventing harmful experiences in the early stages of life is very important from the perspective of the whole life.

Figure 6. Potential influences throughout the life span of adverse childhood experiences.
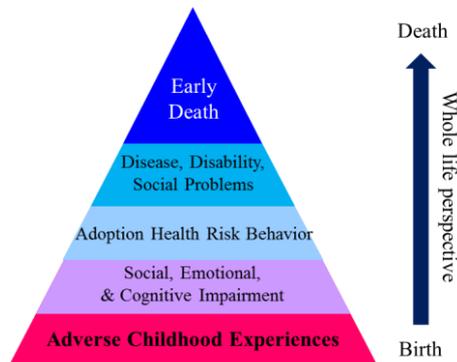
The Adverse Childhood Experiences (ACE) Study. (Felitti VJ, et al. 1998) [23]

5.   RISK FACTORS FOR CHILD MALTREATMENT

How does unintentional injury and child neglect occur?   The occurrence of unintentional injury and child neglect may have similar situations due to lack of supervision, or failure to provide safety.   However, both of them do not occur via a single factor.   Multi-factorial interaction may exist when such harmful experiences are triggered.   There is an "Ecological Model," which explains various risk factors relating to these.   The World Report on violence and health used this ecological model to try to understand the multifaceted nature of violence.[6]   The Ecological Model has four levels to examine factors that influence behavior or increase risks, such as individual factors, relationship factors, community factors, and societal factors.

Considering children's developmental stage and dependence on their parents during the early stage of life, assessing individual and relationship factors in the family may be primary steps to discover preventable factors causing neglect and unintentional injury.   The Ecological Model of risk factors for child maltreatment is shown in Table 2.   According to the literature review,[24]

individual and relationship risk factors of child maltreatment were reported in caregiver's factors, children's factors, and relationship factors including parent-child interactions.

Table 2.　Ecological model for risk factors of child maltreatment

Individual	**Parents or caregivers**: Age, employment status, single parent, self-esteem, child abuse history, drug abuse, alcohol abuse, depression, low self-esteem, anxiety, health problem (physical or mental), psychopathology, lack of self-control, unwanted pregnancy, use of corporal punishment, parenting stress, lack of awareness of child development, socially isolated, involved in criminal activity, poor parenting skills, financial difficulties **Child**: Age, gender, disability, prenatal/neonatal problems, multiple birth, have siblings, crying persistently or unsoothable, personality or temperament (i.e., hyperactivity), externalizing or internalizing behavioral problems, social competence
Relationship	Attachment and bonding, family conflict, family size, family member with health problems, interpersonal violence, socio-economic status of the family, non-biological parent in home, being isolated in the community, lack of social network, breakdown of support from extended family, discrimination against the family, involvement in criminal activity in the community
Community	Tolerance of violence, social inequality in the community, inadequate housing, lack of services, poverty, high unemployment rate, harmful levels of toxin, transient neighborhoods, easily available of alcohol or drug, inadequate policies
Society	Social, economic, health, and education policies that leads to poor living standard, unstable economics, social and culture norms, the existence of child pornography, prostitution, and child labor

Risk factors in child maltreatment listed by the author according to references (Stith[24], WHO[25])

Among the risk factors mentioned above, maternal depression is one factor that impacts on mother-child interactions, parental behavior, and child health and development. In particular, maternal postpartum depression (PPD) has important effects on both mothers' and infants' health. PPD has universal negative effects on maternal functioning and leads to impaired interactions with infants, related to being less affectionate, being withdrawn, intrusive hostile maternal behavior, and impaired interpretation of infant cues.[26, 27] Inadequate caregiving practices are also seen among

mothers with PPD, including discontinuing breastfeeding, placing the infant to sleep in the prone position, and using harsh punishment.[28]   The prevalence of PPD is approximately 13 % in mothers by self-report questionnaire,[29] and is commonly seen in postpartum health care settings.   Although some mothers are suffering from PPD, it is not sufficiently understood how PPD affects infant injury. Therefore, population-based studies may be beneficial for examining the relationship between PPD and unintentional injury.

Although unintentional injury and neglect are similar in that both involve lack of supervision, neglect has much wider content.   Neglect includes any kind of situation in which children's basic needs are not adequately met.   Although comprehensive assessment is needed to assess to what extent basic needs are met for the child, actual conditions of care in neglected children have not been sufficiently examined so far in Japan.   The top priority is to describe children's neglected experiences to assess what kind of care was provided or not provided, and to detect risk factors for individuals and relationships.   The ultimate outcome that child death may have the potential to describe neglected experiences among children has never been revealed.   Thus, utilizing instances of child death will become a precious resource in examining risk factors.

6.   THE OBJECTIVES OF THIS THESIS

The Convention on the Rights of the Child and Maslow's theory insist that the basic needs of the child pile up, so as to achieve self-actualization as shown in Figure 7.   During a child's development, harmful experiences get in the way of children meeting their physiological needs or safety needs.   For example, neglect occurs when physiological needs are not provided for, and

unintentional injury occurs when safety needs are not met.   Child maltreatment has a negative impact on brain development, especially in the early stage of life, and the effects of adverse childhood experiences, including child maltreatment, persist in later life.   Therefore, this study focused on describing actual situations of unintentional injury and child neglect, examining risk factors of these harmful experiences, and suggesting preventive strategies for infants and toddlers.

Study 1 was conducted to examine maternal postpartum depression and infant injury. This study focused on an individual risk factor for mothers, namely depression.   And it enabled examination of the occurrence of infant injury from population-based samples.   **Study 2** was conducted to assess persistent neglect experiences in child fatalities.   This study focused on the worst outcome, namely child death, using autopsy samples, and described persistent neglect experiences.   Throughout these two studies, this thesis aimed to obtain a wide perspective regarding preventing harmful experiences to protect children's health and well-being, to enable them to lead long-lasting meaningful lives.

Figure 7. Relation between study 1 and study 2
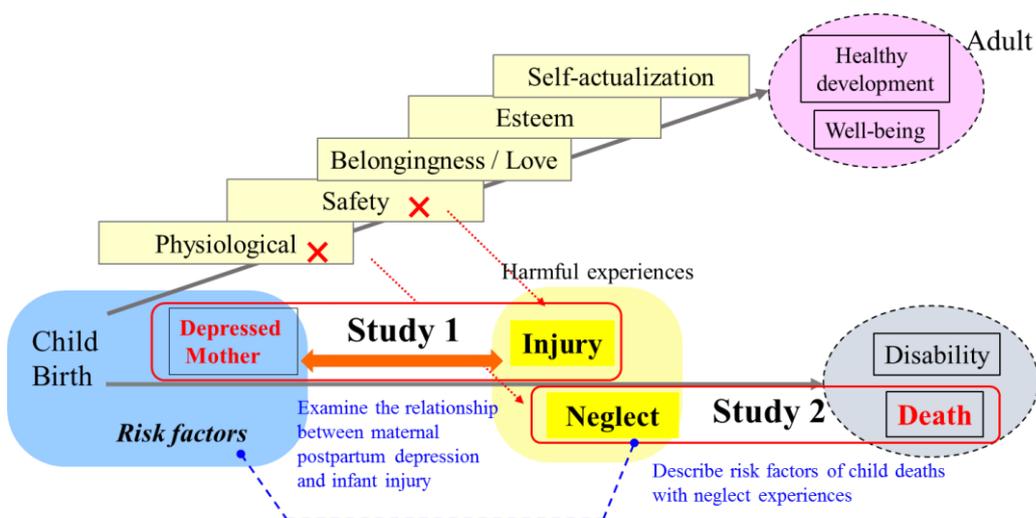
Chapter 2; Study 1

Association between Maternal Postpartum Depression and Unintentional Injuries among 4-months-age infants in Japan

Introduction

In Japan, unintentional injury was the leading cause of death among children.[30]   Although the mortality rate of children aged 1 to 4 years decreased from 6.6 to 3.8 per 100,0000 over 10 years, a reduction of 43%, the mortality rate of children aged 0 to 1 year showed a smaller decrease of 28% [31] from 18.2 to 13.2 per 100,000.[32]   The number of infants under 1 year of age who have been admitted to clinics with unintentional injury is 1,750 times greater than cases of death in the same age group.[8]   Previous studies reported that more than 60% of young children aged 1.5 years experienced unintentional injury, such as falls, near-drowning, or burns within the past 1 year.[33,34] Thus, more interventions are needed to prevent child injury, especially for injuries sustained by infants under 1 year of age.

Risk factors for unintentional injury among children include maternal depression, [35,36] high levels of maternal stress, [37] or highly stressful life events in family.[38]   Further, it was reported that parental supervision and the physical environment of the home have preventive effects on the risk of injury in children under 3 years of age.[39]

However, few studies have reported the association between unintentional injury of infants under 1 year of age and postpartum depression.   In Japan, 95% of infants visit health clinics for routine check-ups at 3-4 months of age.[40]   These routine check-ups provided an opportunity to conduct a questionnaire among participating mothers about unintentional injury among infants and

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postpartum depression.　　The objective of this study is to investigate the association between unintentional injury among 4-month-old infants and maternal postpartum depression.

Materials and methods

Sample

All 54 municipalities in Aichi prefecture, Japan, were invited to participate in this study and 45 municipalities, including Nagoya—the capital city of Aichi prefecture—agreed to be involved. Aichi prefecture has a population of approximately 7.4 million people and 67,913 births were recorded there in 2012.　　The total population of municipalities participating in our study covered 80% of Aichi prefecture's entire population.　　Target subjects of our study were mothers (N=9,707) who were participating in a 3- or 4-month health check-up program between October and November 2012 in the included municipalities.　　An anonymous questionnaire was mailed directly to the women in 34 municipalities before the start of the 3- or 4-month health check-up program, and responses were collected during each health check-up in most municipalities. Eleven municipalities distributed the questionnaire to women directly at the health check-ups, and these participants returned the completed questionnaire to each health center by mail. In total, 6,590 women responded (response rate, 68%).

Our study was approved by the ethics committee of the National Center for Child Health and Development, which determined that it was no longer necessary to obtain consent from participants given that response to the anonymous questionnaire already implied consent to participate in the study.

Questionnaire on unintentional infant injury

The questionnaire assessed types of unintentional infant injury experienced from birth to 4 months via the following response items: 1) accidentally dropped when held by caregiver, or fell from bed (henceforth referred to as 'falls', 2) caught hands or legs in windows or doors, 3) near-drowning in bath tub (henceforth referred to as 'drowning'), 4) animal bites, e.g. bitten by pet, 5) cut fingers with a knife or piece of glass, 6) burned by hot water or iron, 7) foreign body in eyes, nose, or ears, 8) accidental ingestion of drug, cosmetics, or cleaning products 9) motor vehicle accident, or 10) others.   Women selected response items that described injuries experienced by their infant at least once in the time period between births to 4 months of age.

Screening for postpartum depression

We screened women for postpartum depression using the Japanese version of the Edinburgh Postnatal Depression Scale (EPDS).   The EPDS comprises 10 statements including a 4-point Likert scale with a total score range of 0 to 30.   Each statement measures symptoms of depression quantitatively over the past one week.   Widely used internationally for more than 20 years [41], the EPDS has been validated in many languages, including Japanese.[42]   The cut-off point for detecting postpartum depression was 8/9 with 75% sensitivity, 93% specificity and high reliability (Cronbach's α=0.78).   As reported in previous research,[43] Japanese women tend to be less likely to self-report depressive symptoms if the cut-off point of 12/13 is adopted, like in other countries.[44]   We therefore used the cut-off point of 8/9 in this study.

Covariates

Possible covariates were obtained from the questionnaire, including maternal characteristics (age, marital status, employment status, psychiatric history, and the number of people with whom the mother could consult), paternal characteristics (age), infant characteristics (single or multiple birth, birth weight, gestational age, living with siblings), and household characteristics (living with grandmother, grandfather, or other family members, and subjective economic situation).

Statistical analysis

Experience of any type of unintentional infant injury was used as our primary outcome, and two major unintentional injuries—falls and drowning—were used as secondary outcomes. We performed logistic regression analysis to examine the association between postpartum depression and experience of any unintentional injury, falls, and drowning, and adjusted for covariates. We employed STATA SE, version 12 (Stata Corp., College Station, Texas, USA; 2012) for all analyses.

Results

Demographic characteristics of the sample by experience of injury are shown in Table 3. Approximately 10% of infants had experienced any kind of injury at least once since birth to 4 months of age. Parental mean age was 31.4 years (SD=4.8) for mothers, and 33.3 years (SD=5.6) for fathers. Younger parents tended to have experiences of unintentional child injury (both p < 0.001). Half of all children lived with siblings (50.4%), and 12.9% lived with grandparents or other family members. The majority of women were married and cohabited with their partner (98.9%). Most women were not engaged in employment and were homemakers

(78.4%), while 16.7% and 4.9% worked in full-time or part-time jobs, respectively.   Around

12% of women reported difficulties in managing finances, or had an unstable economic situation.

Low birth weight and preterm birth among infants was reported as 8.4% (n=546) and 5.9%

(n=377), respectively.   Of the potential covariates, maternal age, parental age, the number of

people to consult with, subjective economic situation, and living with siblings were associated with

experiences of unintentional injury among infants.

As shown in Table 4, 9.5% of the 6,590 participants (n=623) had an EPDS score of 9 or more.

Women who scored EPDS 9 or above were significantly more likely to have infants who

experienced any kind of injury (16.1%) relative to non-depressed women (9.2%) (p<0.001).

Moreover, the 1.5% of women (n=98) with a psychiatric history were more likely to have a child

who experienced injury (17.3%) than women with no psychiatric history (9.7%) (p=0.012).

Table 5 presents types of unintentional injury.   The number of children who had

experiences any kind of injuries was 647 (9.8% of total infants).   Falls were the most common

(5.6% of total sample), followed by other unintentional injury (2.8%) and drowning (1.2%).

Other unintentional injuries such as accidental ingestion, animal bites, injuries to hands or feet, or

motor vehicle accidents, were rare (<0.5%).   Thus, due to their statistical power, falls and

drowning were selected in the analysis to investigate the association with EPDS.

Table 6 shows bivariate and multivariate logistic regression models for total injury, falls, and

drowning.   Women with an EPDS score of 9 or over were 1.88 times significantly more likely to

have infants who experienced total injury (95% CI: 1.49 to 2.37) in the bivariate model, which

remained significant even after adjusting for potential confounders (OR: 1.59, 95% CI: 1.24 to

2.04). Regarding EPDS association with specific unintentional injury, women with an EPDS score of 9 or more were 1.41 times significantly more likely to have children who had experienced a fall (95% CI: 1.02-1.95), and 1.49 times more likely to experience drowning, although the association is not significant (95% CI: 0.74-3.00) after adjusting for potential covariates.

Of covariates related to mother's characteristics, the maternal status of not working also showed a significant association with lesser experiencing of infant's total injury (OR, 95%CI: 0.78, 0.63-0.97) and fall (OR, 95%CI: 0.70, 0.53-0.93) in the multivariate model.   When mothers had 6-10 people to consult with, there was a significant association with infants' total injury (OR, 95%CI: 0.81, 0.67-0.98) even after adjusting confounders.   However, the significant association did not retain in the case of too many consulting people (more than 10).

Of covariates related to family's characteristics, subjective economic difficulty were significantly more likely to have infants who experienced total injury (OR, 95%CI: 1.73, 1.35-2.21) and fall (OR, 95%CI: 2.20, 1.62-2.97) in the bivariate model, and also in the multivariate model (OR, 95%CI: total injury 1.52, 1.17-1.97, fall 1.97, 1.43-2.72).

Maternal age, living with siblings, living with grandparents or other family, marital status, twin, low birth weight, preterm birth, and maternal psychiatric history had no significant associations with infant's experiences of injury in the multivariate logistic regression.

Discussion

The present study found that postpartum depression was significantly associated with increasing odds of unintentional injuries in 4-month-old infants, particularly for falls. The association remains significant even after adjusting for confounders, including young maternal age,

marital status, social support status, maternal psychiatric history, or economic situation.

Previous studies reported that younger maternal age,[45] single-parent families,[38] lack of social support,[46] and low economic status[47] were associated with unintentional injury among infants and toddlers. Further, maternal depression has been previously linked to occurrences of injury among infants and toddlers.[36] However, to the best of our knowledge, the association between unintentional injury among infants and postpartum depression is underreported. Thus, we added to the literature that mothers with postpartum depression are more likely to have infants who experience unintentional injury, independent from maternal psychiatric history and other potential confounders mentioned above.

Quality of parenting has been reported to be lower among depressed mothers than non-depressed mothers.[48] Mothers who are depressed are less likely to demonstrate child protective behaviors, including the use of specially installed child seats in the car,[49,50] covering electrical plugs [50], and installing smoke alarms in the home.[51] Although we have not assessed the quality of parenting in this study, we found that postpartum depression showed direct association with total unintentional infant injury. Further study is needed to elucidate the mechanism by which parenting quality or the use of child protective devices mediates the association between postpartum depression and unintentional infant injury.

In this study, postpartum depression showed direct association with falls, although the mechanism underlying this association was not revealed clearly. It is reported that the leading cause of falls for infants less than 3 months of age is being accidentally dropped, as defined by E-code (other falls from heights) of the International Classification of Diseases (ICD), or falling from the bed.[52] Thus, although speculative, it is reasonable to assume that mothers with postpartum depression are more likely to accidentally drop infants while holding them. Depressed mothers might be more likely to place infants in an unsafe place, such as on the table or near the

edge of the bed.   Thus, specific fall prevention strategies for depressed mothers, such as education on the correct use of baby slings or installing raised sides to the baby's crib, are needed.

In contrast, we observed that postpartum depression was not significantly associated with drowning.   However, this may be due to the small sample size of drowning cases, for which the point estimate of the odds ratio (OR) was 1.49, which is equivalent to the OR of falls in this study. Fatal unintentional injury due to drowning is much higher in Japan: drowning accounted for 3.8% of fatal unintentional injuries among infants less than 1 year of age in the United States in 2009,[53] compared to 6.0% in Japan.[54]   This difference may be explained by the common daily routine of taking a bath in Japan.   Further, drowning may result in severe neurological sequelae. Therefore, further research is needed to prevent unintentional drowning of infants targeting mothers with postpartum depression especially in Japan.

With regard to maternal employment status, not working mother was a protective factor for infant's injury and fall.   It is still controversial to determine that staying mother at home is better for protecting child injury.   The direction of the effect of maternal employment on child health is inconsistent among previous studies.   Positive impact of maternal employment may be created by increase of family income, health insurance coverage, and maternal self-esteem.   On the other hand, negative impact of maternal employment may lead to less likely to spend time-intensive activities like house-keeping and lower supervision.[55]   Hong et al.[56] reported that the risk of unintentional injury death was 1.3 times higher for children who had a mother working in a manual job, in the children aged 1 to 4.   However, there is a lack of studies to examine the effect of maternal employment on lower age infants' injury like this study samples.   Further research is needed to determine the effect of maternal employment on unintentional injury of infants. Furthermore, working mothers tend to experience infants' injury although they spend less time with their infants at home.   It is also necessary for working mothers to assess their parenting behavior

or home safety, and to examine the mechanism how infant injury occurs at home of working mothers.

We found that having 6-10 people to consult with was a protective factor for unintentional injury in this study (OR, 95%CI: 0.81, 0.67-0.98).   However, the answer of too many people (more than 10) did not show the significant association with injuries, because such mothers might include many people who were not very close to talk usually.   It is important to ask actual number of people whom mothers can consult with, or ask for help easily.   Having several people to consult with for mothers indicate the availability for social support.   Social support has functional contents such as emotional support, informational support, instrumental support and appraisal support[57] as shown in Figure 8.   Consulting with families or friends may provide mothers relief, information and actual help.   In previous research, there was a significant association between maternal social support and the likelihood of children's accidents and injuries.[58]   It is needed for mothers after birth to provide social support through interventions like home-visiting program or peer-mothers support, because mothers received no or minimal social support especially after birth are likely to experience postpartum depression[59] (as shown in Figure 8).

Figure 8. The relation among postpartum depression, low social support, and unintentional injury
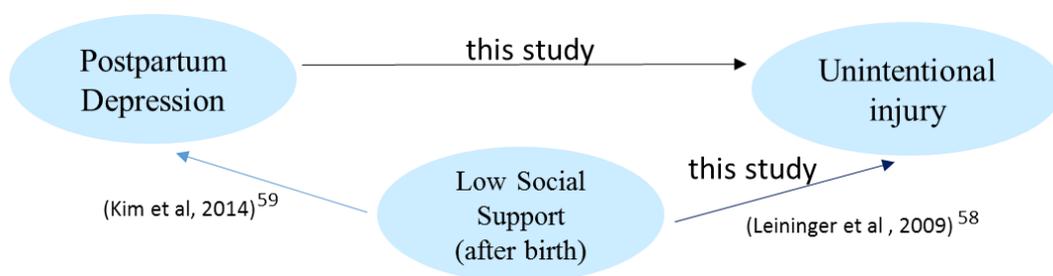
And as another covariate, maternal subjective economic difficulty had a significant association

24

with experiences of infant's injury.   Poverty is a well-known risk factor of child's unintentional injury.[47,60]   Faelker et al.[60] reported that a linear relationship between increasing economic disadvantage and higher risk of child injury within each sex and each age strata, including aged 0 to 4.   Poverty is strongly linked to socio-economic status such as educational level, social class, and types of labour.   Therefore, mothers who felt financial difficulties may have unstable job, less social support, and feel higher stress in childrearing.   It is important to assess the maternal childrearing situation from comprehensive aspects such as maternal mental health, the level of social support, and also financial status of the family.

This study has several limitations.   First, women who did not attend regular health check-ups may have severe postpartum depression, or may have had their child admitted to hospital due to unintentional injury, which underestimates the association between postpartum depression and unintentional infant injury.   Minkovitz[61] reported that children whose mothers had depressive symptoms at 2 to 4 months had decreased to receive age-appropriate well-child visits at 12 months and up-to-date vaccinations at 24 months.   In 2012, the proportion of children who did not participate regular health checkups at 3-4 months old was only 2% in Aichi prefecture.[62]   However, some mothers with postpartum depression might not answer the questionnaire (the response rate of this study was 68%).   The significant association between postpartum depression and unintentional infant injury was found although there was a possibility not to assess mothers with postpartum depression enough.

Secondly, as experiences of unintentional injury were self-reported and recalled from the time period of birth until 4 months of age, social desirability bias and recall bias may exist. Furthermore, we did not examine the severity of injury, i.e., whether the child had been admitted to hospital due to unintentional injury or not.   It is reported that postpartum depressed mothers

25

increased emergency department visits of children, but did not increased hospitalization of them compared to non-depressed mothers in the previous research.[61]   Therefore, it is needed to compare medical records of injury between mothers with depression and without to evaluate actual frequency and severity of infant's injury.

Third, EPDS is a screening tool for postpartum depression, and does not provide formal diagnosis such as structured clinical interview using DSM-IV.   When healthcare providers detect postpartum depression in mothers, it is necessary to consult mothers with psychiatric physicians to make clinical diagnosis and treatments.

Lastly, reverse causation exists because of the study's cross-sectional design.   Experiences of injury among infants may cause maternal depression due to self-blame, or blame from other family members, or even health professionals.   Cox et al.[63] reported that within 5 weeks after child birth was the highest timing of the onset of postpartum depression with EPDS.   Munk-Olsen et al.[64] investigated the diagnosis-specific risks of admission due to mental disorders by ICD-10.   For mothers in their research, the risks of admission were increased during the first 5 months (with the highest risk of 31 to 60 days postpartum) for unipolar disorder, during the first 2 months (with the highest risk of 0 to 30 days postpartum) for bipolar disorder.   According to their findings, postpartum depression may precede infant's injury, however, it is necessary to conduct a prospective research of examining the timing of the onset of postpartum depression and injuries in order to examine a causality between them.

Despite these limitations, the current study sustained the significant association between postpartum depression and unintentional infant injury with a large population-based sample. Further research is needed to detect the mechanism of postpartum depression and unintentional injury, especially falls and drowning.   Further randomization controlled trials targeting mothers with postpartum depression are also needed to investigate the effectiveness of educational

interventions to improve protective behaviors relating to home safety and parental supervision.

Conclusion

Maternal postpartum depression was significantly associated with increasing odds of unintentional injuries among 4-month-old infants, especially for falls.   Greater attention should be paid to investigating and detecting maternal postpartum depression when medical professionals encounter infants with unintentional injuries.   Developing a better awareness of parenting behavior, use of safety devices at home, and the amount of social support available to depressed mothers is also needed when managing cases of unintentional child injury.

Table 3. Characteristics of sample (N=6,590)

		Experience of injury from birth to 4 months of age				
		Never (n=5,943; 90.2%)		Once or more (n=647; 9.8%)		p value
Maternal age	(year)	(n)	(%)	(n)	(%)	
	<20	29	0.49	5	0.77	0.001
	20-24	417	7.1	70	10.8	
	25-29	1617	27.4	202	31.3	
	30-34	2225	37.7	215	33.3	
	35-39	1360	23.0	129	20.0	
	40-	261	4.4	25	3.9	
Paternal age	(year)					0.001
	<20	10	0.17	4	0.63	
	20-24	250	4.3	45	7.1	
	25-29	1190	20.3	145	22.8	
	30-34	2023	34.5	214	33.6	
	35-39	1633	27.9	163	25.6	
	40-	757	12.9	66	10.4	
Living with siblings						0.037
	No	2922	49.2	346	53.5	
	Yes	3021	50.8	301	46.5	
Living with grandmother, grandfather or other family members						0.074
	No	5190	87.3	549	84.9	
	Yes	753	12.7	98	15.1	
Marital status						0.171
	Married	5842	98.9	633	98.3	
	Divorced, unmarried, widowed or others	65	1.1	11	1.7	
Low birth weight (<2500g)						0.670
	No	5401	91.6	594	92.1	
	Yes	495	8.4	51	7.9	
Preterm birth (<37 weeks)						0.426
	No	5453	94.0	604	95.3	
	Yes	347	6.0	30	4.7	

Twin						0.175
	No	5848	98.4	632	97.7	
	Yes	95	1.6	15	2.3	
Maternal employment status						0.286
	Full-time	961	16.5	117	18.5	
	Part-time	281	4.8	35	5.5	
	Not working	4590	78.7	481	76.0	
Number of people to consult with						0.014
	0-5	2242	40.4	283	46.6	
	6-10	2334	42.1	217	35.7	
	11-	967	17.4	107	17.6	
Subjective economic situation						<0.001
	Stable	2636	46.5	244	39.9	
	Able to manage	2394	42.2	266	43.5	
	Difficult to manage or unstable	637	11.2	102	16.7	

Table 4.　Maternal postpartum depression among sample

	Experience of injury until 4 month old　(N=6,590)		
	Never (n=5,943; 90.2%)	Once or more (n=647; 9.8%)	p value
EPDS			<0.001
<9	5365　90.8	546　9.2	
9 =>	523　83.9	100　16.1	
Mother's history of mental disease			0.012
No	5862　90.3	630　9.7	
Yes	81　82.7	17　17.3	

Table 5. Unintentional injuries experienced by infants between birth and 4 months of age

Types of injury	Number of injuries	% of total infants (N= 6,590)
Fall	368	5.6
Caught hands or legs in window or door	10	0.2
Drowning	77	1.2
Animal bite	10	0.2
Cuts to fingers	1	0.02
Burns	7	0.1
Foreign body	2	0.03
Accidental ingestion	22	0.3
Motor vehicle accident	9	0.1
Other	182	2.8
Total	688*	10.4

(* multiple answers)

Table 6.   Association between sample characteristics and specific types of injury in children

		Total injury				Fall				Drowning			
		Bivariate		Multivariate		Bivariate		Multivariate		Bivariate		Multivariate	
		OR	95% CI	OR	95% CI	OR	95% CI	OR	95% CI	OR	95% CI	OR	95% CI
EPDS	<9	Reference		Reference		Reference		Reference		Reference		Reference	
	9 =>	**1.88**	**1.49 - 2.37**	**1.59**	**1.24 - 2.04**	**1.78**	**1.32 - 2.39**	**1.41**	**1.02 - 1.95**	1.77	0.95 - 3.29	1.49	0.74 - 3.00
Maternal age (years)	<20	Reference		Reference		Reference		Reference		Reference		Reference	
	20-24	0.97	0.36 -2.60	0.91	0.33 - 2.47	1.43	0.33 - 6.19	1.33	0.30 - 5.87	0.55	0.67 - 4.54	0.51	0.06 - 4.24
	25-29	0.72	0.28 - 1.89	0.71	0.27 - 1.88	1.05	0.25 - 4.43	1.02	0.24 - 4.41	0.53	0.071 - 4.04	0.47	0.06 - 3.65
	30-34	0.56	0.21 - 1.46	0.57	0.21 - 1.52	0.89	0.21 - 3.77	0.91	0.21 - 3.94	0.36	0.047 - 2.70	0.32	0.04 - 2.53
	35-39	0.55	0.21 - 1.45	0.57	0.21 - 1.53	0.82	0.19 - 3.51	0.85	0.19 - 3.69	0.25	0.31 - 1.96	0.25	0.03 - 2.05
	40-	0.56	0.20 - 1.56	0.53	0.19 - 1.53	0.70	0.15 - 3.27	0.67	0.14 - 3.21	0.23	0.021 - 2.63	0.22	0.02 - 2.56
	p for trend	**<0.001**		0.377		**0.004**		**0.019**		**0.008**		**0.024**	
Living with siblings	No	Reference		Reference		Reference		Reference		Reference		Reference	
	Yes	**0.84**	**0.72 - 0.99**	0.87	0.73 - 1.03	0.86	0.69 - 1.06	0.86	0.69 - 1.08	**0.53**	**0.33 - 0.84**	0.64	0.39 - 1.05
Living with grandparents or other family	No	Reference		Reference		Reference		Reference		Reference		Reference	
	Yes	1.23	0.98 - 1.55	1.16	0.92 - 1.47	1.09	0.80 - 1.48	0.99	0.72 - 1.37	0.57	0.25 - 1.31	0.60	0.25 - 1.39
Marital status	Married	Reference		Reference		Reference		Reference		Reference		Reference	
	Divorced or unmarried	1.56	0.82 - 2.97	1.20	0.62 - 2.34	1.00	0.99 - 1.02	1.73	0.79 - 3.79	1.12	0.15 - 8.18	0.96	0.61 - 1.50
Twin	No	Reference		Reference		Reference		Reference		Reference		Reference	
	Yes	1.46	0.84 - 2.53	1.65	0.90 - 3.03	1.52	0.76 - 3.03	1.77	0.81 - 3.85	0.77	0.11 - 5.61	0.74	0.09 - 5.80
Low birth weight	No	Reference		Reference		Reference		Reference		Reference		Reference	

31

	Yes	0.94	0.69 - 1.27	0.88	0.64 - 1.21	0.87	0.58 - 1.29	0.81	0.53 - 1.25	1.3	0.62 - 2.71	1.36	0.63 - 2.92
Preterm birth (<37 weeks)	No	Reference		Reference		Reference		Reference		Reference		Reference	
	Yes	1.00	0.99 - 1.01	1.00	0.99 - 1.01	1.00	0.99 - 1.01	1.00	0.99 - 1.01	1.00	0.99 - 1.02	1.00	0.98 - 1.02
Maternal employment status	Full-time	Reference		Reference		Reference		Reference		Reference		Reference	
	Part-time	1.02	0.69 - 1.53	0.93	0.62 - 1.40	1.37	0.86 - 2.17	1.19	0.74 - 1.92	0.42	0.097 - 1.85	0.48	0.11 - 2.12
	Not working	0.86	0.70 - 1.48	**0.78**	**0.63 - 0.97**	0.79	0.60 - 1.04	**0.70**	**0.53 - 0.93**	0.75	0.43 - 1.32	0.70	0.39 - 1.24
	p for trend	0.135		**0.019**		**0.040**		**0.004**		0.414		0.270	
Number of people to consult with	0-5	Reference		Reference		Reference		Reference		Reference		Reference	
	6-10	**0.74**	**0.61 - 0.89**	**0.81**	**0.67 - 0.98**	**0.70**	**0.55 - 0.89**	0.78	0.61 - 1.00	0.93	0.57 - 1.52	0.95	0.57 - 1.60
	11-	0.88	0.69 - 1.11	0.98	0.77 - 1.25	0.79	0.58 - 1.07	0.87	0.63 - 1.20	0.57	0.26 - 1.23	0.66	0.30 - 1.45
	p for trend	0.054		0.86		**0.028**		0.179		0.89	0.35 - 2.30	0.741	
Subjective economic situation	Stable	Reference		Reference		Reference		Reference		Reference		Reference	
	Average	**1.20**	**1.00 - 1.44**	1.14	0.95 - 1.38	1.24	0.97 - 1.57	1.20	0.93 - 1.53	0.95	0.58 - 1.57	0.92	0.55 - 1.54
	Difficult	**1.73**	**1.35 - 2.21**	**1.52**	**1.17 - 1.97**	**2.20**	**1.62 - 2.97**	**1.97**	**1.43 - 2.72**	1.06	0.51 - 2.23	0.98	0.45 - 2.14
	p for trend	**<0.001**		0.541		**<0.001**		0.843		0.971		0.323	
Maternal psychiatric history	No	Reference		Reference		Reference		Reference		Reference		Reference	
	Yes	**1.95**	**1.15 - 3.32**	1.43	0.81 - 2.51	**1.95**	**1.00 - 3.78**	1.49	0.75 - 2.96	**3.74**	**1.34 - 10.4**	2.33	0.69 - 7.91

Chapter 3; Study 2

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Child deaths with persistent neglected experiences
from Medico-legal documents in Japan

</div>

Introduction

　　The number of child maltreatment reports to the Child Guidance Center has drastically increased every year in Japan.　　Reported cases have increased by more than 5 times in 2012 from 1999 prior to the Child Abuse Prevention Act enforced in 2000.[65,66]　　The proportion of neglect among reported child maltreatment was 28.9%, which was much less than that reported in the USA (78.3%).[66,67]　　The proportion of fatalities due to neglect shows the same tendency had the same tendency: 27% in Japan and 69.9% in the USA among total deaths due to child maltreatment.[67, 68]

　　The reason for this discrepancy may be because it is not possible to assess fatal child maltreatment sufficiently, especially neglect cases in Japan.　　For example, National Police Agency reported 32 children died due to fatal child maltreatment in 2013,[69] but types of child maltreatment were not referred, and those figures were only the cases which the perpetrators have arrested.　　On the other hand, national annual report on fatal child maltreatment has been published by Ministry of Health, labor and Welfare since 2005.　　It selected subjects from death cases reported in newspapers, or cases which municipal authorities could recognize as death due to child maltreatment.　　Therefore, it is possible that some subjects were missed due to being unidentified as child maltreatment, not reported, and not examined adequately.　　Not knowing actual incidence and even definition of child neglect, we have been losing valuable opportunities to prevent successive deaths in the family and also in our society so far.

1   In addition to insufficient reporting system, the numbers of studies on fatal child neglect is

2 also limited in Japan. Although neglect is difficult to define and measure,[70] only seven articles [71]

3 [72] [73] [74] [75] [76] were available from PubMed (by sear.ching with "child neglect" and "autopsy" and

4 "Japan"), and three articles from *Igaku Chuou Zasshi*; Japanese database for medical articles (by

5 searching with "child neglect" and "autopsy" and "death" in Japanese [71, 77] and by hand-searching

6 [78]) as far as we detected. The majority were case reports, and there was neither systematic

7 evaluation nor classification of child fatal neglect so far.

8   A previous research conducted by Welch and Bonner [79] found that children under 2 years old,

9 the youngest children in family, and male infants had a higher risk for fatal child neglect. Use of

10 a holistic view of child neglect and intervention at an early stage may be necessary to reduce the

11 risk factors of neglect and to prevent the worst outcome (death).

12   An innovative approach to access children's death reports could help to identify preventive

13 factors. Every unexpected death notified to police is criminally investigated, and this includes

14 postmortem external examination of corpses and autopsy. Medico-legal documents are a

15 potential source of details of child maltreatment that have not appeared in the previous national

16 reports. The aims of this study were to investigate neglect among child fatalities using

17 medico-legal documents, and to describe characteristics of the socio-familial background and

18 biological data.

19

20

21 **Methods**

22

23 *Defining Persistent Child Neglect*

24

1　　　　There are several classifications of types of child maltreatment, such as Child Protection

2　　Services (CPS) classification, National Incidence Study (NIS) maltreatment coding system, and

3　　Modified Maltreatment Classification System (MMCS).[80]　　CPS reports are likely to describe

4　　only a primary or single issue, but MMCS assesses multiple subtype co-occurrence.　　Although

5　　NIS is used to examine incidence of child maltreatment nationwide including both CPS records and

6　　non-CPS records, the coding system of MMCS provides a severity weight of caregiver's behavior.[17]

7　　Therefore, we chose MMCS operationally to define neglect experiences of children and to describe

8　　inadequate care before their deaths in the current study.　　MMCS includes two major types of

9　　neglect: "Failure to Provide" and "Lack of Supervision".[70]　　There are five subtypes of failure to

10　　provide and three subtypes of lack of supervision.　　The details of the MMCS coding mechanisms

11　　are available from the website.[11]　　According to MMCS, all documents on children <2 years old

12　　were examined to determine whether any subtypes of neglect existed.　　A child who had at least

13　　one subtype was defined as having an experience of neglect in the present study.

14　　　　In addition, the present study focused on "persistent" neglect experiences.　　According to

15　　guideline for the safeguarding of children in the UK, [81] "Working together to safeguard children",

16　　neglect is defined as the "persistent failure" to meet a child's basic physical and/or psychological

17　　needs.　　A major obstacle to the establishment of a general definition of child neglect is the

18　　problem of threshold, which is establishing what may be considered a minimally adequate levels of

19　　care.[82]　　It is very difficult to pinpoint when exactly the inadequacy of care becomes problematic.[83]

20　　Therefore, the current study selected children who were clearly recorded as having experienced

21　　recurrent incidents of any subtypes of neglect over a certain time until just before their deaths.

22　　There were some cases which had only one episode in the medico-legal documents.　　The current

23　　study did not include such gray-zone cases.　　Persistency means daily occurrence such as dirtiness

24　　of home environment or not taking bath for several days, or incidences of more than one episode

1   such as not receiving any kind of public health service or canceling medical appointments.

2   Persistency of any subtypes of neglect was based on information taken from the medico-legal

3   documents; two co-authors made the judgment of persistence and categorization of MMCS

4   subtypes.

5

6   *Selection of Study Subjects*

7

8   As Figure 9 shows, there were 13,050 postmortem external examinations of all age groups in

9   one prefecture from 2006 to 2011.   Of those, in the 65 cases the age was identified as <2 years.

10   One forensic pathologist carried out the autopsy in 59 cases (90.8%).   The present study

11   precisely examined the documents of 59 autopsies, and evaluated whether each victim had

12   experienced any subtypes of neglect in MMCS.   After detecting 13 children who experiences

13   neglect more than once, persistency was confirmed in six cases, and these then became the study

14   cases.

15

16   *Investigation of Medico-legal Documents*

17

18   The documents were retained in the Department of Legal Medicine in that prefecture.

19   Autopsy case information sheet was developed to obtain baseline uniform information in this

20   department as shown in Figure 10.   The other information was collected through investigations,

21   postmortem external examinations and autopsies.   Examiners in the current study extracted

22   biological data, situation of death, family environment, and the use of maternal and child health

23   services. They noted body lengths, bodyweights, organ weights, and conditions when they died

24   such as home environment, age of family members, family composition, the use of medical, and

1   public health services from pregnancy until child rearing, and so on.

2

3   *Assessment of Weight Loss and Nutritional Status*

4

5   The Waterlow system that assesses the degree of malnutrition[84] was used to evaluate

6   nutritional status according to the weight and length data obtained.   Decreased ratio of weight for

7   length (W/L), represents wasting due to acute malnutrition, and decrease of length for age (L/A)

8   represents stunting due to chronic malnutrition or nutritional growth failure.   We used World

9   Health Organization (WHO) Anthro version 3.2.2[85] to calculate W/L, L/A, and severity of

10   undernutrition.   According to the WHO Anthro manual, body height of children <24 months is

11   described as length in the present study, instead of height, which was used for children aged ≥24

12   months[85].

13

14   *Ethical Consideration*

15

16   This current study was approved by the official ethical review board of University of Tsukuba

17   （document no.807, Sep/13/2013）.

18

19

20

21   **Results**

22

23   *Persistent Neglect*

24

1   Table 7 listed the details of persistent neglect experienced, with MMCS subtype.   Utilizing

2   medico-legal documents, we investigated socio-familial and medical information (Table 8).   In

3   the six identified cases of persistent neglect, the home environment was described as unsanitary and

4   unorganized in three cases.   One child was found in the car. Time until detection varied from 1

5   hour to 6.5 hours.   In the other two cases, the time of the last meal was longer than 12 hours,

6   which was not adequate at 2 months of age.   In case 6 the child had been admitted to hospital

7   with a 1 month history of pneumonia.

8   From family information, both parents were biological parents in six cases.   Two out of six

9   mothers delivered victims at <21 years old.   All victims were youngest child or only child.   In

10   three cases, there were siblings who were 1 year older than victims.   In Case 6, there were twin

11   siblings, and both of them had died before the victim was born.

12   In terms of service use, mothers in case 1, 4, and 6 had fewer perinatal care visits, and also the

13   timing of the first visit was close or passed the possible limit of abortion.   After discharges,

14   mothers in case 2 and 4 refused the neonatal home visit that municipal government conducted for

15   every infant <1 month old.   Children in case 1 and 2 did not receive any vaccines despite being

16   older than 1 month.   In Case 5, we could not obtain the details of health service use from

17   pregnancy until infancy.

18

19   *Biological Data*

20

21   Table 9 lists their body lengths, bodyweights and organ weights.   Reference for body

22   lengths and weights were taken from assessment of physical growth in Japanese children.[86]

23   Reference data of organ weights were drawn from "Reports on medico-legal data from massive

24   investigation performed by the Medico-Legal Society of Japan".[87]   In six cases, all children had

1   lower weight for month age and sex-adjusted average.   The average Z score for bodyweight in

2   six cases was -2.22 (which means 2.22 standard deviations below the mean).   In particular, in

3   case 4, the bodyweight Z score of the victim was -4.0, and most of organs had decreased weights,

4   except for the adrenal glands.   In cases 1 and 2, the thymus had become remarkably involuted

5   compared to other cases.   In Case 6, body length, weight, brain and thymus weights were

6   extremely low because the child had become ventilator dependent around 1 year after

7   hypoxic-ischemic encephalopathy due to asphyxia at 5 months old.

8

9   *Assessment of Nutritional Status*

10

11   Scores for W/L L/A are listed in Table 10, with 3 grades of severity of undernutrition.[88,89,90]

12   Using WHO classification to evaluate nutritious status, z score of W/L and L/A in case 1 were less

13   than -3.0, which represents severe wasting and sever stunting.   Likewise, the child in case 3 had

14   moderate wasting and sever stunting, and the child of case 6 showed severe stunting.

15

16

17   **Discussion**

18   Utilizing Medico-legal documents, we investigated fatal cases with persistent neglect

19   experiences from the two aspects of legal medicine and public health, and described the detailed

20   process and the effects of fatal neglect on children.

21

22   *Socio-family Background and History of Service Uses*

23

24   Multiple-births are noted as one of risk factors for child maltreatment,[91,92] and having several

1   siblings in the family is another risk factor.[93,94]   Although sibling age difference has not been

2   reported as a risk factor, a 1 year difference between siblings was noted in three cases in the present

3   study.   One sibling (case 1; 1 year old) had never received vaccinations and health checkups,

4   and the other sibling (case 3; 1 year old) also had left at home with the victim for several hours.

5   Further research is needed to determine the age and number of siblings most likely to be seen as a

6   burden to parents when raising them together.

7   Moreover, neglect is associated with poverty.   Children of low socioeconomic status were

8   found to be sevenfold more likely to be neglected in US Forth National Incidence Study.[95]   In

9   case 1, even after parents realized their child delayed development, they stated that they had never

10   visited a physician due to financial reasons.   Other parents (case 2) stopped utilizing a daycare

11   center 2 months prior to the death because they could not afford the fee and both parents went out

12   for working while the child was left at home alone.   We could not, however, evaluate the actual

13   financial conditions and parenting attitude of caregivers in more detail.   Collection of

14   information from multi-disciplinary organizations is therefore needed in order to evaluate why and

15   how neglect was occurring.

16   With regard to health service use, three mothers had fewer times of prenatal care and later first

17   visits to obstetrics.   In Japan, 86.9% of mothers visited municipal office and received Mother and

18   Child Health (MCH) handbook by 11 weeks of gestational age after first visit to obstetrics and

19   confirmed pregnancy.[96]   Japanese government recommends 14 prenatal visits at clinics or

20   hospitals. Therefore, later first visit and few number of prenatal care may reflect unexpected

21   pregnancies or failure to abort.   Problems in the prenatal period would be carried over to the

22   child rearing period.   Knowing the timing of the first visit and the number of prenatal care visits

23   is very important for accessing mothers' willingness and conflicts with regard to child rearing.

24   Support for mothers starting in pregnancy may help prevent fatal child neglect.

1

2 *Loss of Bodyweight and Undernutritional Status*

3

4   The extent of weight loss in actual fatal neglect has not been sufficiently recognized in Japan,

5   even though failure to thrive, or FTT is a known indicator of neglect.[97]   As noted, this study

6   confirmed the extent of lower bodyweight.   In case 1 and 3 the children fitted into the definition

7   of FTT given that the Z score of weight for age was less than -2, and their weight loss Z score was

8   -4.0 and -2.7, respectively.   Although the child in case 6 had been diagnosed with FTT on a Z

9   score of -3.7 at 4 months old, it was difficult to evaluate bodyweight at death with regard to

10   nutritional status because of the long period of ventilation dependence.

11   Utilizing the Waterlow system and the WHO classification to evaluate nutritious status, the

12   children in case 1 and 3 had acute and chronic undernutrition.   According to previous studies,

13   children with weight for height below -3 SD based on the WHO standards have more than a nine

14   fold higher risk of death compared to children with weight for height above -1 SD.[98]   It is

15   important for medical professionals who may see neglected children to pay much closer attention to

16   the extent of decrease of bodyweight and nutritional status.

17

18 *Thymus Involution Due to Long-term Child Neglect*

19

20   Autopsy identified thymic involution in case 1, 2 and 6.   The children in cases 1 and 2 had

21   more overlapping subtypes of neglect compared to other four children.   Thymic involution had

22   been correlated with degree and duration of physical abuse and neglect,[99] and immunodeficiency

23   secondary to thymic involution can occur due to long-term child abuse.[73,100]   The child in case 6

24   might have been affected by both chronic maltreatment and the stress of long-term ventilation.

1   But, as seen in case 2, delay in visiting the physician is considered as medical neglect given that the

2   parents failed to take the child to the clinic or hospital even after she had fever, cough, vomiting

3   and fatigue for 1 week. The victim received no vaccination, no regular health check-ups after 1

4   month old, and repeated exacerbation of atopic dermatitis and one occurrence of subcutaneous

5   abscess.   Combination of basic poor skin hygiene, immunosuppression due to thymic involution

6   caused by persistent neglect, and delay of medical care could have led to the child's death, with

7   exacerbation of pneumonia at the end.   To prevent successive deaths from neglect, involution of

8   the thymus should be noted as an important factor in the assessment of the probability of child

9   maltreatment.

10

11   *Collaboration Public Health and Legal Medicine for Prevention of Child Neglect*

12

13   Collaboration public health and legal medicine enabled collection of detailed background

14   information and data on the biological effects of neglect on children in this study.   In previous

15   national reports of fatal child maltreatment, results of reviews were classed together as all types of

16   fatal maltreatment.   Therefore, we focused on only neglected cases, using the MMCS subtype

17   failure to provide and lack of supervision, and the dimension of "persistency".   The present

18   findings, which have not been described in national reports, were related to death situation

19   parameters, including time until detection and time from last meal, age differences between siblings,

20   and biological data from autopsies.

21   Among six victims, previous involvement with Child Guidance Center was found in only case

22   6 at the time of FTT diagnosis.   There was only one child those caregivers were prosecuted and

23   broadcasted in the newspapers, according to the medico-legal documents.   Therefore,

24   medico-legal documents have the potential to review child deaths that had not been reviewed in the

1   existing national child fatal maltreatment reports.

2

3

4   *Limitations and Future Directions*

5

6   There were several limitations in the present study.   Although collected information was

7   multidirectional and meaningful to review, autopsy might not reveal every cause of deaths.

8   Second, if physicians failed to report the case as unnatural death, or failed to understand the

9   importance of postmortem external examination and autopsy for determining the cause of death, the

10   case would never have been recorded and examined by forensic physicians.

11   Third, the number of subjects was extremely small in the present study.   In these cases, all

12   parents were biological parents and living together with the children.   In previous national report

13   of child maltreatment, house composition had been reported in total maltreatment cases, not

14   divided into only neglected cases.   Did the samples of this study represent typical characteristics

15   of neglected children?   The research targeted to child neglect and familial conditions was limited

16   in Japan.   By searching in *Igaku Chuuou Zasshi* (Japanese medical journal database) with the

17   term of 'child' and 'neglect' and 'family structure' in original articles (on Nov 25th, 2014), 26

18   articles were detected.   However, most of them described risk factors or maternal parenting

19   behavior related to child maltreatment, and did not target neglected cases in particular.   Of them,

20   there was only one article specifically targeted substantiated neglected children.   Abe reported

21   that one third of children who substantiated neglect was living with single biological mother.[101]

22   But, study samples of his research was cases from regional council of countermeasures for Children

23   Requiring Aid (*Youhogojidou taisakuchiiki kyougikai*), whom local municipal governments

24   followed as neglected cases.   In addition to the differences between death cases and living cases

43

1 of neglect, the definition of neglect local government officers may be different from MMCS that

2 this study used.　　Failure to provide is easier to be detected and understood compared to lack of

3 supervision.　　In fact, only two children had lack of supervision in this study although five

4 children had failure to provide.　　Raising awareness that lack of supervision is also a key part of

5 neglect is necessary in order to identify the actual situation of neglect in Japan.　　And then, it is

6 necessary to accumulate various information from both living neglected children and death cases to

7 establish evidences.

8 　　Last, parental responsibility differs on a cultural basis.　　It is recommended to examine what

9 kinds of failure to provide and lack of supervision can be suitable in Japanese society for

10 identifying neglect cases in a timely manner before the neglect becomes fatal.

11

12

13 **Conclusion**

14

15 　　Integrating biological data and information of socio-familial backgrounds obtained from

16 medico-legal documents, we could find child deaths with persistent neglect which might not have

17 been reported in national annual report of child maltreatment.　　The present study enabled to

18 describe several factors of dead children, such as short birth spacing, poor use of maternal and child

19 health services, and biological effects.　　The system of integrating detailed information and

20 evaluating death cases multidisciplinary is needed to identify preventable factors in fatal child

21 maltreatment.

22

23

24 **Acknowledgment**

1

4

1   Figure 9.   Flow chart of study samples

```
┌ ─ ─ ─ ─ ─ ─ ─ ─ ─ ─ ─ ┐
│        13050          │
│   Postmortem external │
│  examinations of all age │
│   between 2006 to 2011 │
└ ─ ─ ─ ─ ─ ┬ ─ ─ ─ ─ ─ ┘
            ┊
            ▼
┌ ─ ─ ─ ─ ─ ─ ─ ─ ─ ─ ─ ┐
│         65            │
│   Postmortem external │
│      examinations     │
│   Under two years old │
└ ─ ─ ─ ─ ─ ┬ ─ ─ ─ ─ ─ ┘
            ┊ ┊ ┄ ┄ ┄ ┄ ┄ ┄ ┄ ┄ ▶ ┌ ─ ─ ─ ─ ─ ─ ─ ─ ─ ┐
            ┊                      │        6          │
            ▼                      │  Autopsy was not  │
    ┌───────────────┐              │    conducted.     │
 *  │      59       │              └ ─ ─ ─ ─ ─ ─ ─ ─ ─ ┘
    │ Autopsy cases │
    │ Under two years old │
    └───────┬───────┘
            │ ──────────────────▶ ┌───────────────────┐
            ▼                     │        46         │
    ┌───────────────┐             │   Not fitted into any │
    │      13       │             │  subtype of neglect in │
    │ Fitted into at least one │  │       MMCS        │
    │ subtypes of neglect in │    └───────────────────┘
    │      MMCS     │
    └───────┬───────┘
            │ ──────────────────▶ ┌───────────────────┐
            ▼                     │        7          │
    ┌───────────────┐             │  Not confirmed    │
    │       6       │             │   persistency     │
    │ "Persistent" fatal neglect │ └───────────────────┘
    │   (study samples) │
    └───────────────┘
```

*The survey started from here.

2

3

4

1   Figure 10. Information sheet (English version as below)

2

3   剖検事例調査票

| 解剖 | 平成　　　　年　　　月　　　日 | 死者 | (□男 □女)　　　　　　　歳 | 所轄署 | |

4

生活	同居者 生活費	□なし(独居) □配偶者 □息子 □嫁 □娘 □娘婿 □孫 □祖父 □祖母 □父 □母 □兄弟姉妹 □他(　　　　　　　　　　　　　　) 　計　　　人暮し □自営・給与等 □年金 □生活保護 □家族の収入 □その他(　　　　　　　)
病歴など		☆ 医療機関、福祉施設、行政からも、できる限り病歴情報を得てください。 ☆ 医療機関が死亡診断書を発行しているときは写しの入手をお願いします。
	情報源 □医療機関 □福祉施設 □行政 □家族・親族 □知人 □その他	通院歴：□あり □なし □不明　女性の場合：経産　　　回 (経妊　　　回) 入院歴：□あり □なし □不明 病　名：□糖尿病　　□脳梗塞　□統合失調症　□癌(部位：　　　　) 　　　　□狭心症　　□脳出血　□うつ病　　　□その他(　　　　　) 　　　　□高血圧　　□B型肝炎　□認知症 　　　　□高脂血症　□C型肝炎　□ｱﾙｺｰﾙ依存症
	手術歴	□あり (内容：　　　　　　　　　　　　　　　　) 　　　　□なし □不明
	歯科受診歴	□あり (最終：　　年　　　月　　) 　　　　　　　□なし □不明
	障害者認定	□あり(事由：　　　　　　) (□1級 □2級 □3級 □4級 □5級 □6級) □なし □不明
	介護認定等	□要支援(□1度 □2度) 　　　　　　　　　　　　　□なし □不明 □要介護(□1度 □2度 □3度 □4度 □5度) 　　　　主な介護者(□配偶者 □息子 □嫁 □娘 □他(　　　　　　　　))
	利用ｻｰﾋﾞｽ	□ﾃﾞｲｻｰﾋﾞｽ/ﾃﾞｲｹｱ □ﾎｰﾑﾍﾙﾊﾟｰ □ｼｮｰﾄｽﾃｲ □入浴(□通所 □訪問) □配食 □訪問看護 □訪問診療または往診 □その他(　　　　　　　　　)
救急隊	臨場 □なし □あり	到着時刻　　　　時　　分 到着時：心拍動(□あり □なし) 呼吸(□あり □なし) 意識(□あり □なし) 処　置：心マッサージ(□あり □なし)，人工呼吸(□あり □なし) 特記事項
医療機関	搬送 □なし □あり	病院搬入時刻　　　　時　　分 搬入時：心拍動(□あり □なし) 呼吸(□あり □なし) 意識(□あり □なし) 処　置：心マッサージ(□あり□なし) 挿管(□あり□なし) 除細動(□あり□なし) 　　　　点滴(□なし □輸液 □輸血) 詳細情報

状況等	(□発見 □加害開始 □事故発生)（＿＿月＿＿日（AM・PM）＿＿時＿＿分頃） 　　　　加害等が継続の場合は終了：＿＿月＿＿日(AM・PM)＿＿時＿＿分 最終生存　（＿＿月＿＿日（AM・PM）＿＿時＿＿分頃） 確認手段(□直接会話 □電話会話 □携帯履歴 □目撃 □新聞・郵便等 □その他) 確認者(□親族(□同居 □別居）□隣人 □知人 □家主・管理人 □ヘルパー □その他(　　　　　　　　　　) 近所付合(□良好 □やや良好 □不良 □なし □不明) 発見者：□家族・親族(□同居 □別居）□隣人 □知人 □通行人 □集金人・配達人 □家主・管理人 □行政職員 □ヘルパー □その他(　　　　　　　　　　　　　)
食事等	最終食事　＿＿時＿＿分頃　　　　　　詳細情報（献立および食材、飲酒量・飲酒継続時間、その他）現 場酒類(□あり □なし) 直前飲酒　＿＿時＿＿分頃 各種習慣 （□飲酒 □喫煙 □服薬 □なし）
備考	

1

2

3　　Figure 10. Information sheet (English version. Translated by the author)

Autopsy	Date (　　　　　　　　)	Case	Sex　(M / F)　　　Age (　　　)	Police station (　　　　　　　　　　　　)
Life	Housemate	□None (Alone) □Spouse □Son □Daughter □Daughter-in-law　□Son-in-law		
		□Grandchild □Grandfather □Grandmother □Father □Mother　□Sibling		
	Cost of living	□Other(　　　　　　　)　　　　　*Total number of people living with is　(　　　　).		
	(multiple choice)	□Independent business/Salary □Pension □Welfare benefit		
		□Income of other family members □Other(　　　　　)		
Medical history	* Please collect information from medical facilities, local municipality, and welfare institutions 　　as far as possible. * Please obtain death certification if medical facility have issued.			
	Source of information 　□Medical facility 　□Municipality 　□Welfare 　　　institution 　□Family member 　□Acquaintance 　□Other(　　　　　)	Regular hospital visits；□Yes □No　□Unknown 　　　　　　　　　　　　*If Woman, number of parity (　　) and gravida (　　) History of hospitalization；□Yes　□No　□Unknown Disease name；□Diabetes　　□Angina　　　　□Hypertension 　　　　　　　□Hyperlipidemia □Cerebral infraction □Hepatitis B 　　　　　　　□Dementia　　□Cerebral hemorrhage □Hepatitis C 　　　　　　　□Schizophrenia □Depression　　□Alcoholism 　　　　　　　□Cancer (region:　　　　)　□Other(　　　　　　)		
	History of operation	□Yes (　　　　　　　　　　　　　　　　　) □No　□Unknown		

48

	Visits to dental clinic	☐Yes (Last time date;) ☐No ☐Unknown
	Certification of the disability	☐Yes (Reason;) ☐No ☐Unknown Certification class;☐1 ☐2 ☐3 ☐4 ☐5

	Certification of long-term care need	☐Support required (☐1 ☐2) ☐No ☐Unknown
		☐Care level (☐1 ☐2 ☐3 ☐4 ☐5)
		       Primary caregiver ☐Spouse ☐Son ☐Daughter
		               ☐Daughter-in-law ☐Other( )

Emergency service	Call-out ☐Yes ☐No	Arrival on the sight; (Time_____) On arrival, heartbeat (☐Yes ☐No), breathing (☐Yes ☐No), consciousness (☐Yes ☐No)

Hospital	Transportation
	☐Yes
	☐No
		Arrival to the hospital; (Time_____)
		On arrival, heartbeat (☐Yes ☐No), breathing (☐Yes ☐No),
		                 consciousness (☐Yes ☐No)
		Treatment; chest compression (☐Yes ☐No), intubation (☐Yes ☐No),
		       defibrillation (☐Yes ☐No), blood transfusion (☐Yes (volume;)☐No),
		       infusion (☐Yes (volume;) ☐No),

Situation of death	☐Found -> ☐Start of trauma or ☐Occurrence of accident Date;(Month Day , (AM/PM) :)
	                     *End of trauma ->Date;(Month Day , (AM/PM) :)
	Last confirmation of living Date;(Month Day , (AM/PM) :)
	  The means of confirmation; ☐Direct conversation ☐Conversation on the telephone
	                        ☐Record of mobile phone ☐Witness ☐Newspaper/Letter, etc ☐Other
	  The person of confirmation; ☐Family member/relatives (☐living with ☐not living with) ☐Neighbors
	                  ☐Acquaintance ☐Landlord/resident manager ☐Helper ☐Other( )
	Neighborly ties; ☐Very good ☐Good ☐Not good ☐None ☐Unknown
	The person of detecting one's death; ☐Family member/relatives (☐living with ☐not living with)
	           ☐Neighbors ☐Acquaintance ☐Passerby ☐Bill collector/deliverer
	           ☐Landlord/resident manager ☐Administrative officer ☐Helper ☐Other( )	

Meal	Last Meal Time; (AM/PM) :)
	Alcohol on-site; ☐Yes ☐No
	Last drinking time; (AM/PM) :)
	Habits;
	  ☐Drinking ☐Smoking
	  ☐Drugs ☐None
		Menu;
		
		Cooking ingredient;
		
		Drinking amount and time;

Remarks		

1

Table 7. Case descriptions with subtypes of Modified Maltreatment Classification System

Case	Age (month)	Sex	Food a)	Clothing	Shelter b)	Medical	Hygiene	Supervision c)	Environment	Substitute care
1	7	M	**Mother had not given milk because a child had vomited. Older sibling had taken over the milk usually.**	wearing clothes	**Room was very dirty and cluttered with wastes of foods, used diapers, clothes, and bottle of formula.**	**A child had never received infant regular checkups and vaccinations.**	**A child didn't take a bath, and diapers were not changed, resulting in diaper rash.**	NA	NA	NA
2	18	F	**Parents did not cook meals, left food which was bought outside, and sometimes it was passed expiry date.**	**The child was not wearing any clothes, only with a diaper.**	**Room was very dirty with wastes of foods, feces, dishes tobacco, and used diapers. The toilet didn't flash.**	**A child had received infant regular checkups only once, and no vaccinations. Parents refused neonatal home visiting. Parents didn't visit hospitals recognizing child's fever, cough and vomiting.**	**A child didn't take a bath, and diapers were not changed for a week, resulting in diaper rash.**	**The caregiver left a child alone at home at night repeatedly.**	NA	NA
3	2	M	(13 hours interval from last meal on the death day because mother was drunken.)	wearing clothes	no problem	no problem	no problem	**The caregiver left the victim and a sibling at home for several hours repeatedly.**	NA	NA

50

4	2	M	(12 hours interval from last meal on the death day because mother felt annoying to care.)	wearing clothes	**Room was littered and piled with many clothes, plastic bags, and cans of formula disorderly.**	**Mother canceled perinatal visit and refused neonatal home visiting.**	no problem	NA	NA	NA
5	6	M	no problem	wearing clothes	(No information of home environment. The child was left inside a cluttered car with blankets, no child seat.)	**Mother canceled medical appointments and refused admission before.**	no problem	NA	NA	(An acquaintance was taking care of him, and left him alone inside a car.)
6	16	F	**A child was placed into infant home at 4 months old due to 'Failure to Thrive' after admission into the hospital.**	wearing clothes	no problem	no problem	no problem	(A child became asphyxia during feeding milk without supervision, in infant home at 5 months old.)	NA	NA

Bold-faced type represents repeated experiences of child neglect.

Contents with parentheses (   ) was one time occurrence and not confirmed persistency.

NA (not available) is unobtainable information. 'No problem' is obtained and confirmed information by the documents.

The details of MMCS coding mechanism is available from the website at http://www.unc.edu/depts/sph/longscan/.

Supplemental explanations were added operationally in this study for implementing the coding system precisely and adjusting Japanese situations.

a) Providing inadequate food (e.g. expired food) was including 'Failure to provide food'.

b) As an indicator of 'Failure to provide shelter', the dirtiness was defined that more than 30 % of area in the floor was covered or cluttered with objects including unsanitary wastes or harmful things (e.g. used diapers, or tobacco).

c) 'Lack of supervision' was leaving a child alone at home more than three hours, repeatedly, or leaving a child alone inside a car.

Table 8. Socio-familial and medical information obtained from Medico-legal documents

	Case 1 (7 months, M)	Case 2 (18 months, F)	Case 3 (2 months, M)	Case 4 (2 months, M)	Case 5 (6 months, M)	Case 6 (16 months, F)
Situation of death						
Cause of death	unclear	**pneumonia**	unclear	unclear	unclear	pneumonia, multiple organ failure
Home environment	**dirty**	**dirty**	clean	**unorganized**	**found at car**	**admitted into hospital**
Time until detection	1.5 hours	6.2 hours	4.5 hours	6.5 hours	1 hour	-
Time from last meal	4 hours	6.2 hours	**13 hours**	**12 hours**	3 hours	-
Last meal before death	**Almost no milk (Given milk was taken by a sibling.)**	**3 pieces of sushi (expiry date was yesterday)**	milk 110ml	milk 20-30ml	milk 300ml	-
Family information						
Age of father (year)	21	20	35	38	53	36
Age of mother (year)	18	22	30	32	31	34
Age of mother at delivery (year)	**17**	**20**	29	31	31	33
Number of siblings	1	0	1	1	3	2 (twin)
Age of siblings (year)	1	―	1	1	9, 8, and 3	**Both of them died at 1.3 and 2.3 years old**
Age-difference between siblings	**13 months**	―	**more than 10 months**	**17 months**	more than 2.5 years	6.5 years

Use of maternal and child health services

	1	2	3	4	5	6
Weight at birth	2486 g	2998 g	2214 g	2412 g	2400 g (approx.)	2466
Number of prenatal care	**2**	12	13	**2**	NA	NA
First visit of prenatal care (gestational week)	**28**	9	8	**19**	NA	**around 7 months**
Number of infant regular health check-ups	**None**	**Once (at only one month age.)**	Once (at one month age.)	Once (at one month age.)	NA	1, 3 months
Neonatal home visit	NA	**Refused**	NA	**Refused**	NA	NA
Vaccination	**None**	**None**	(None)*	(None)*	NA	NA

NA; not available   * Vaccination starts around two months old in Japan.

Table 9. Biological data obtained from Medico-legal documents

case	1	mea	SD	z	2	mean	SD	z	3	mea	SD	z	4	mea	SD	z	5	mea	SD	z	6	mean	SD	z

	n		score				score		n		score		n		score		n		score				score
Height (cm)	61	69.2	2.4	-3.4	80	79.4	2.7	0.2	55	57.9	2.2	-1.3	57	57.9	2.2	-0.4	67	67.8	2.4	-0.3	61	77.5	2.6	-6.3
Bodyweight (kg)	4.6	8.2	0.92	-4.0	9	9.9	1.0	-0.9	3.7	5.5	0.67	-2.7	4.8	5.5	0.67	-1.0	7.5	8.0	0.9	-0.6	6.1	10.1	0.97	-4.1
Organ weight (g)																								
Brain	830	934	57	-1.8	930	1022	103	-0.9	640	616	59	0.4	510	616	59	-1.8	800	842	80	-0.5	490	1022	103	-5.2
Heart	26	42	5.7	-2.8	45	49	8.3	-0.5	30	31	5.2	-0.2	35	31	5.2	0.8	45	36	2.2	4.1	40	49	8.3	-1.1
Lung (lt)	45	67	12.8	-1.7	75	78	22.4	-0.1	45	51	7.3	-0.8	55	51	7.3	0.5	65	59	10	0.6	35	78	22.4	-1.9
Lung (rt)	60	82	17.2	-1.3	85	92	27.5	-0.3	50	61	11.4	-1.0	50	61	11.4	-1.0	80	72	12	0.7	65	92	27.5	-1.0
Liver	260	327	30.0	-2.2	420	392	62.0	0.5	160	210	35.0	-1.4	170	210	35	-1.1	290	269	31	0.7	180	392	62.0	-3.4
Spleen	15	34	13.7	-1.4	45	37	13.1	0.6	15	41	11.2	-2.3	15	41	11.2	-2.3	40	15	29	0.9	25	37	13.1	-0.9
Pancreas	10	14	3.7	-1.1	11	21	9.3	-1.1	4	6	1.9	-1.1	6	6	1.9	0.0	10	14	12	-0.3	5	21	9.3	-1.7
Kidney (lt)	20	27	4.0	-1.8	27	32	7.1	-0.7	20	22	4.0	-0.5	20	22	4	-0.5	40	26	5.3	2.6	25	32	7.1	-1.0
Kidney (rt)	20	30	5.1	-2.0	28	32	6.5	-0.6	15	21	4.3	-1.4	20	21	4.3	-0.2	40	25	4.9	3.1	25	32	6.5	-1.1
Adrenal gland (lt)	2	1.9	0.92	0.1	2	1.9	0.53	0.2	1	2.4	0.68	-2.1	2	2.4	0.68	-0.6	1	1.8	0.6	-1.3	2	1.9	0.53	0.2
Adrenal gland (rt)	2	2.1	0.95	-0.1	1.5	1.6	0.58	-0.2	1	2.1	0.63	-1.7	2	2.1	0.63	-0.2	1	1.7	0.56	-1.3	2	1.6	0.58	0.7
Thymus	10	37	12.3	-2.2	9	25.3	9.4	-1.3	25	30.7	10.7	-0.5	30	30.7	10.7	-0.1	40	37	10.6	0.3	5	25.3	9.4	-2.2

Reference was matched month-age and sex on each case.

* Reference of Case 2 was 12 months old girl, because the reference data was shown in monthly figure during 0-11 month age and yearly

after 1 year old on each sex.

Table 10. Nutritional status

Severity of undernutrition

case	1	2	3	4	5	6
Weight for height (Z score)	-3.34	-1.31	-2.49	-0.78	-0.04	-0.05
Height for age (Z score)	-3.98	-0.35	-3.09	-1.85	-0.71	-6.33
Wasting (acute)	**Severe**	Mild	**Moderate**	Normal	Normal	Normal
Stunting (chronic)	**Severe**	Normal	**Severe**	Mild	Normal	**Severe**

WHO classification of child undernutrition

Severe	Moderate	Mild
Z score < -3	$-3 \leqq$ Z score < -2	$-2 \leqq$ Z score < -1

Chapter 4: Discussion

This thesis discusses the risk factors of unintentional injury and child neglect in infants and toddlers.   Study 1 focused on associations between maternal postpartum depression (PPD) and unintentional injury, and study 2 described persistent neglect experiences from child fatalities. Both studies were aimed at identifying preventable factors and strategies for avoiding harmful experiences.

In study 1, maternal postpartum depression was commonly seen as a health problem faced by mothers after birth, and was detected in 9.5% of mothers.   Mothers with postpartum depression were more likely to have infants who experience unintentional injury, after adjusting from maternal psychiatric history and other potential confounders such as young maternal age, lack of social support, and financial difficulties.   Although maternal postpartum depression is often paid attention to as one of the risk factors for child abuse, this study revealed that it can also be one of the risk factors of unintentional infant injury.

The advantage of study 1 is that it was a population-based cross-sectional study.   Samples represented around 70% of mothers and infants in one prefecture because the proportion of attendance at regular infant check-ups at 3–4 months was 98%[62] and the response rate was 68%. Infant check-ups are a primary and precious opportunity for public health professionals to assess the health conditions of both mothers and infants.   In this primary setting like infant check-ups, assessment for every mother and child plays an important role in supporting their ability to start and maintain a healthy life.

Study 2 described child fatalities in which experiences of persistent neglect.   These children were represented as ultimate outcomes.   Persistence did not refer to one-time occurrence.   In

six cases, "beginning" or "mild omission" levels of neglect had existed prior to death in six cases, although unintentional injuries were not detected from medico-legal documents.   The advantage of study 2 was a detailed description of child deaths which had not been examined sufficiently with medico-legal documents which were very difficult for researchers to utilize for reviewing so far. And as far as searching previous researches, it was the first study to evaluate persistent neglect experiences using MMCS in Japan.   Through this process, death situations could disclose how these children did not receive adequate supervision, were not provided enough care, and that this represented how they had not been loved and protected.   In study 2, the presence of close-age siblings, insufficient use of public health, and loss of weight or under-nutritional status were detected. Two studies suggested preventive strategies from primary aspects and ultimate aspects.

There are several approaches to take regarding prevention for unintentional injury and neglect. Macmillan et al.[102] addressed the framework for prevention of child maltreatment and associated impairment as shown in Figure 11.   The preventive strategies include, for example, prevention before occurrence, including a universal approach and a targeted approach, prevention of recurrence, and prevention of impairment.

Figure 11.   Framework for prevention of child maltreatment and associated impairment
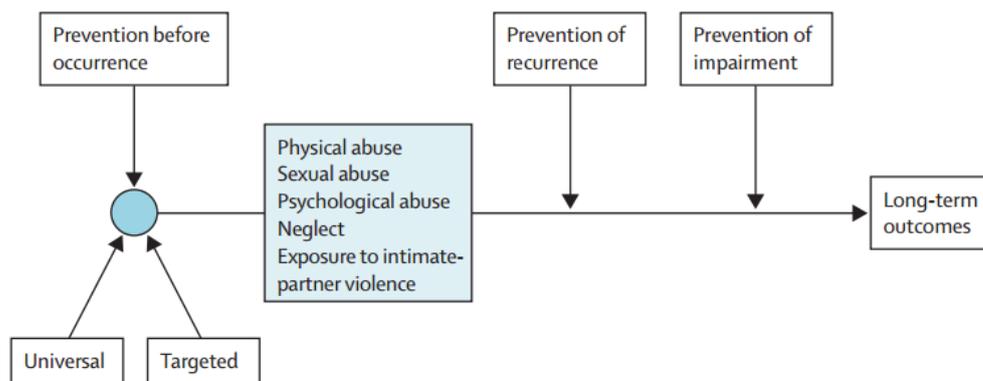
(Macmillan et al. 2009) [102]

Several preventive strategies can be found out from study 1 and 2 as shown in Figure 12.

Figure 12.   Possible preventive approaches based on study 1 and study 2
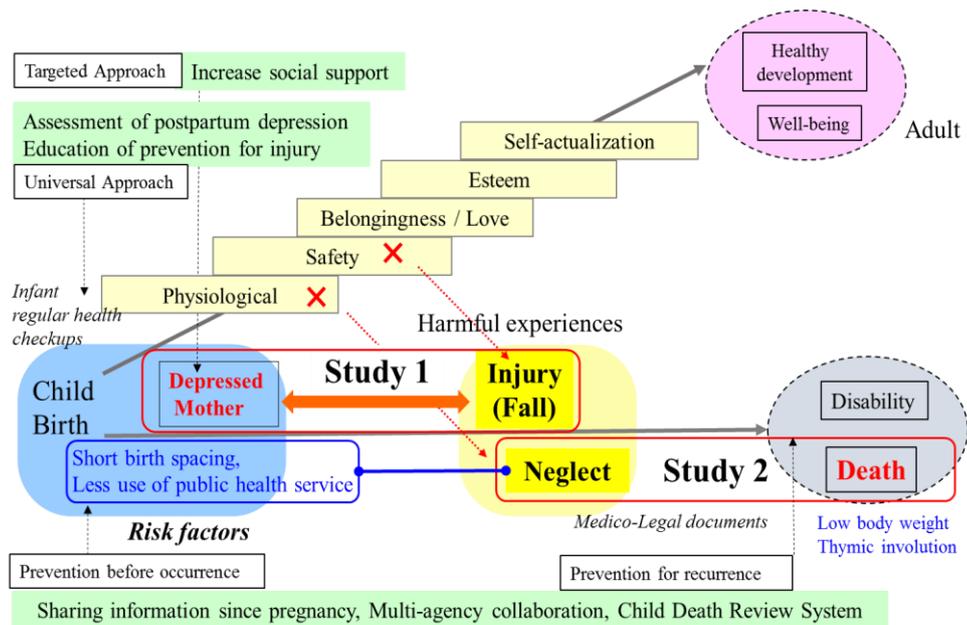
1) Universal preventive approach

Unintentional injury or child neglect occurs when physiological needs or safety needs are not met.   These experiences are harmful for children.   To prevent them before they occur, one possible opportunity is the primary health care setting, such as regular infant health check-ups. Assessing mothers and infants at regular infant health check-ups in study 1 acts as a universal preventive approach (as shown in Figures 11 and 12), as it evaluates all mothers for postpartum depression.

In addition, home visiting program can be also a universal approach to detect depressed mothers. Neonatal home visits and infant family home visit services (Hello Babies Services; *Konnichiwa Akacyan*) are provided to all families from the local municipal government in Japan.

For example, in Fukuoka city, Yoshida et al.[103,104] developed a community-based maternal mental health program to comprehensively support depressed mothers with a multidisciplinary team. Public health nurses or midwives were trained with the Child-Rearing Support Manual, and visited homes to assess maternal mental health using three self-report questionnaires, the Edinburgh Postnatal Depression Scale (EPDS), The Bonding Scale Toward Baby (*Akacyann no Kimochi Shitumonnhyou*) and a checklist of risk factors regarding postpartum depression and infant abuse. According to the assessment of these questionnaires, mothers suspected to be depressed are recommended to visit psychiatric physicians in some cases. Suzumiya[105] reported that the proportion of mothers scoring 9+ on the EPDS had decreased gradually from 13% to 8% in Fukuoka city during the five years after the program was introduced. The average day of the first home visit was 55.2 ± 42.6 days after birth to assess the EPDS score, and an average number of visits was 3.2 ± 1.7. The consulting rate to medical facilities was 0.4–0.5% of mothers during the five years, therefore, there was a stigma or hesitation to visit psychiatric facilities. Further research is needed to reveal the reason for the decreasing prevalence of detection of PPD. However, visiting programs have continued to include one-quarter of mothers with a high EPDS score after the conclusion of the study period (18–23 months after first visits to each child). Child-rearing conditions are affected by maternal mental status, familial characteristics, social support, and financial situation. Therefore, assessing maternal PPD with EPDS may not be enough, because there may be false negatives appearing in attempts to detect it, and various assessment tools are needed to evaluate how mothers need support for child-rearing.

2) Targeted preventive approach

Furthermore, <u>home visiting</u> can be one aspect of a targeted preventive approach (as shown in <u>Figures 11 and 12</u>) as a way not only to assess risk factors related to the health of mother and child,

but also to promote safe child-rearing methods for mothers to avoid further child injuries. In previous research, a lower level of social support is associated with unintentional injury,[58] and mothers are more likely to experience PPD if they received no or minimal social support.[59] Besides, lack of social support is also associated with less safety in the home environment.[46] To take a targeted approach to mothers with PPD, home visiting by health professionals such as nurses are one strategy to increase social support. Home visiting improves mother-infant interactions and decreases the severity of PPD,[26, 106] reduces psychological distress, improves perceived social support[107] and improves the home environment.[108] However, Fujiwara et al.[109] reported that receiving one or two visits by public health nurses resulted in no substantial reduction in maternal stress and no increase in social trust in Japan. Visiting only a few times might not have a significant enough effect on the reduction of maternal stress in this study. Infant family home visit services (such as the Hello Babies Services; *Konnichiwa Akacyan*) promotes visits to all households with infants of less than four months in age in Japan. Community volunteers can visit, instead of public health nurses or midwives. Training programs for such community volunteers is also important along with training programs for health professionals that Yoshida has started in Fukuoka[103,104]. In addition, it is necessary to examine what frequency of home visitation is most suitable for mothers and children to decrease the actual incidence of injury in Japan, and not only for detecting high-risk families.

In addition to prevention for maternal PPD and infant injuries, home visiting has another effect as a preventative mechanism for child maltreatment, which was reported by several randomized controlled trials (RCT) and systematic reviews.[110-115] Hahn et al.[110] reported that home visitation programs showed a 40% reduction of child maltreatment as the median effect size from 21 studies, programs delivered by nurses demonstrated more favorable effects compared to programs delivered by paraprofessionals, and beneficial effects were consistently evident when

programs by paraprofessionals were conducted over two years. There are several aspects of home visits which result in beneficial outcomes, such as program objectives, the theory (mechanism) of change through the program, selecting a targeted population due to their needs, and program components and activities. Segal et al.[111] mentioned that a combination of underlying theory, program components, and targeting specific populations was critical to maximize the effects of the program. Moreover, Dalziel et al.[116] evaluated 33 home visiting programs to assess cost-effectiveness (i.e., cost per case of child maltreatment avoided), and seven "cost-saving" programs were found to target high-risk populations (i.e., first-time mothers, low-income families), employed mostly health professionals as visitors, and used a comprehensive approach involving more than merely home visitation. There are two nation-wide home visiting programs in Japan, neonatal home visiting and infant family home visit services, both targeting infants and parents universally. Targeted home visiting to preterm babies, or families at high risk of child maltreatment are also conducted by health care centers, however, programs vary among communities, and do not have a unified strategy, evaluation system, and lack theory-based approaches. Therefore, it is necessary to consider how to design efficient home visiting programs to decrease child maltreatment, including how to select target populations and visitors, find an appropriate intensity and duration, and determine what kind of theory should be applied in the program.

3) Approach to prevention of recurrence

To prevent recurrence of harmful experiences and death, another possible opportunity is for the system to learn from fatal cases. Assessing risk factors for fatal child neglect from medico-legal documents in study 2 can be a preventive approach for recurrence (as shown in Figure 11 and 12).

In several countries, the Child Death Review (CDR or Child Fatality Review) system has been developed to understand how and why children die due to external causes, including child maltreatment and unintentional injuries. CDR teams use the findings to take action that can prevent other deaths, and improve the health and safety of children.[117] The CDR is beneficial to prevent not only child maltreatment death, but also deaths arising from unintentional injury, because CDR usually targets all deaths due to external causes. In the US, the CDR system has been established since the 1970s, and all states have introduced it since 2012. The US National Child Death Review Case Reporting System (NCDR-CRS) has also collected data across states, and evaluated findings and actions taken after CDR. The entries of NCDR-CRS via the Internet is very high (approximately 1,700 elements),[118] compared to Japanese death certification (less than 40 elements). Palusci et al.[119] reported 2,285 fatalities due to child maltreatment among 49,947 child deaths from 2005 to 2009 in the NCDR-CRS, and one-half of fatal maltreatment cases had neglect identified. As important insight from their research, of the 2,285 maltreatment deaths, 1,020 cases had recommended or planned actions or strategies, and 109 had actually been implemented by July, 2012.

It is said that "The death of a child is a community responsibility," in the first line of the operating principles of a program manual for the Child Death Review used in the US.[117] How do the Japanese consider their responsibilities toward preventable children's deaths? The national annual report of child maltreatment in Japan has a limitation to sample collection as described in study 2. Study 2 tried to reveal background information of neglected children, although the collection of data was very limited; the source of information comprised medico-legal documents in one prefecture. Fujita[120] reported wide differences in the autopsy rate among infant deaths due to Sudden Infant Death Syndrome (SIDS) and suffocation, from less than 10% to over 60% in some prefectures. SIDS should be diagnosed by autopsy, therefore, abuse cases might be

diagnosed as SIDS or suffocation without either an autopsy or child death review.   It is urgent to establish CDR procedures in Japan with a multidisciplinary professional team to make the most of the information that can be provided by children's deaths about possible abuse and neglect.   A CDR should be conducted both nationally and locally to find preventive strategies.   When local municipals of health care centers conduct local CDRs, it is necessary to support the person in charge of the children because they may face enormous levels of psychological distress through blaming themselves, and through being blamed by community members and also by the media. Staff and also citizens should be aware of the importance of learning lessons from child death, without resorting to blame.

4) Approach to prevention before occurrence

Interagency collaboration is important to conduct CDR with multidisciplinary professionals, and also important to assess high-risk families and support them before injuries or child maltreatment occur.   Some families have had problems since the pregnancy period, as shown in study 2.   The prevention before occurrence approach can be thought to comprise sharing and utilizing information regarding risk factors from the perinatal period to the child-rearing period. The regional council of countermeasures for Children Requiring Aid (*Youhogojidou taisakuchiiki kyougikai*) plays an important role in the community in taking preventive actions both before occurrence and recurrence of child maltreatment, including informing pregnant women who are at high risk of child maltreatment (*Tokutei ninpu*).   There is little research available to evaluate how this regional council may work effectively, or prevent recurrences of child maltreatment in the long term.   Establishing a database of substantiated child maltreatment cases is necessary for evaluating the effects of support for children and their families, and following up on the consequences for these children.

Tanaka[121] estimated the costs of economic loss through death due to unintentional injury in childhood by using a life-time cost calculation from Gross National Product of 2000. It was estimated at ￥290 billion ($2.9 billion). Wada[122] estimated the direct cost of child maltreatment (i.e., costs of child social welfare services, administrative costs) and indirect cost of child maltreatment (i.e., death-related costs, medical costs). It amounted to approximately ￥1.6 trillion ($16 billion) in 2012. This social cost of child maltreatment for only the year 2012 is almost equal to the total amount of damages of ￥1.9 trillion caused by the 2011 Tohoku earthquake and tsunami in the Fukushima Prefecture. We must recognize that these costs do not merely represent a financial burden on society. Moreover, they represent the extent to which children are likely to die prematurely, suffer disability, or be placed under child protection. Losing children or increasing the number of children with disabilities indicates a huge loss: one of precious children who had possibilities to grow, work, and create in order to aid our society. Every agency and professional needs to be aware of the community's responsibility to protect children from any harmful experiences. A move toward better collaboration between agencies, and a vertically-segmented administrative system should be avoided, and sharing detailed information should be understood as important. We ought not to hide case information under the banner of personal information protection. Citizens also need to be aware that everyone has to play a role in the community's responsibility to prevent child deaths, and not just take the attitude of criticizing offenders.

Aside from the above-mentioned preventive approaches, other multidirectional strategies are needed to prevent various other factors which this thesis has not sufficiently evaluated. Preventive strategies are expected to target children in the early stages of life, in order to prevent consequences that might be experienced later in life.

Chapter 5; Conclusion

Both neglect and unintentional injury can occur in daily continuous care when children's basic needs such as physiological needs and safety are violated.　According to study 1 in infant health check-ups as primary health care setting, unintentional infant's injury were associated with maternal postpartum depression, and there is a need to support for mothers in order to prevent infant injury.　From medico-legal documents in study 2, death cases with persistent neglect could be detected which never had been reviewed in national reports of child maltreatment, and there were several findings of risk factors to learn lessons for the prevention.　Preventive approaches should be holistic view to assess risk factors in whole family from primary health care setting, to share information at multidisciplinary meetings, and to define preventive measurements from reviewing child deaths.　Every agency, professional, and also citizen needs to be aware of the responsibility to protect children from any harmful experiences, as it is said that "The death of a child is a community responsibility".　Toward better children's healthy development and well-being in later life, preventing harmful experiences based on evidences is necessary for early stage of life through wide range of approaches.

Chapter 6; Acknowledgement

Chapter 7; References

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