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Abnormal sucking behavior in infants as a predictor of developmental delay at 18-month or 3-year of age

(授乳における子どもの行動とその後の発達との関連に関する研究)

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Title: Abnormal sucking behavior in infants as a predictor of developmental delay at 18-month or 3-year of age

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Abstract:

Background: Behaviors in infants may predict not only current developmental status but also later neurodevelopmental outcomes. Sucking is one of the earliest observable behaviors in infants. This study aimed to determine whether abnormal sucking behaviors in infants can predict the developmental outcome at 18 months or 3 years of age.

Methods: A questionnaire asking abnormal sucking behaviors was used to survey mothers of children who visited one of 4 health centers for the routine 18-month- or 3-year-old health check-up in Japan. At the check-up, children were assessed by public health nurses whether he or she passed for age-appropriate developmental milestones.

Results: A total of 472 responses were analyzed. The children were grouped into two groups according to the assessment by public health nurses: in 18-month-old children, 198 were typically developed (TD) and 52 were suspicious of developmental delay (DD); for 3-years-old, 164 were TD and 58 were DD. We found a significant difference in the rate of the lack of smooth suck and rest pattern between TD and DD groups across the two age populations. After controlling provable confounding factors, a logistic regression analysis showed significant association between the developmental delay and lack of smooth suck and rest pattern ($P = 0.004$).

Conclusion: The result suggests that the abnormality in suck and rest pattern of sucking in infants might be a predictor of developmental delay at 18-month- and 3-year-old of ages.

Keywords: Sucking; breast feeding; developmental delay; health check-up; burst-pause pattern

Introduction

Sucking, a precocious motor skill that is important to suckle at the breast or bottle, is one of the earliest observable behaviors in infants. If an infant has carried neurological deficits, certain signs corresponding to the deficits might be found in his or her sucking behaviors. Indeed, mothers of infants with cerebral palsy note that their child has feeding difficulties, such as slow or weak sucking in the neonatal period (Reilly and Skuse 1992). Early sucking problems are important sequelae of neonatal stroke (Barkat-Masih et al., 2010) and present in a consistent proportion of infants with neonatal brain injury (Slattery et al., 2012). Furthermore, accumulating evidence suggests that abnormal sucking performance in preterm infants can predict later neurodevelopmental delay (Sacker et al., 2006; da Costa et al., 2010). For instance, Mizuno and Ueda (2005) examined the expression and sucking pressures of infants with feeding problems and assessed their neurodevelopmental outcome at 18 months of age, and found a significant correlation between feeding performance and neurodevelopmental outcome. Törölä et al. (2012) reported that feeding development of preterm and full-term infants is similar except sucking. These findings suggest that sucking behaviors in infants may predict not only the current developmental status but also later neurodevelopmental outcomes. However, many of experimental studies in this field have included participants from highly selected groups, such as those born preterm or with low birth weights. Therefore, it is not clear how far the results of such studies can be extrapolated to the general population, because it is not unusual that full-term infants with no apparent neurological deficits later show developmental delay.

In this study, we aimed to determine whether abnormal sucking behaviors in infants can predict the developmental outcome at 18 months or 3 years of age. For this purpose, we conducted a survey on mothers of children who were involved in the population-based health check-up program in Japan. It is highly likely that mothers will accurately remember the child's sucking behaviors, because breastfeeding is one of the most intimate social interactions occurring between a mother and her infant.

Methods

Subjects

The study involved parents of children who visited one of the four health centers located in two prefectures in Japan for the 18-month- or 3-year-old health check-up programs during the period from July 2012 to January 2013. The health checkup programs are based upon the Maternal and Child Health Act. They are designed to support parenting activities, promote children's health, and detect diseases and developmental disorders as early as possible, and participate almost 95% of children living in Japan.

Measures

We conducted this survey by an anonymous and self-administered questionnaire (**Appendix 1**). Along with a written explanation of the study objective, a questionnaire was mailed to the mothers before the health check-up. Questionnaires were handed to those who consented to the study and collected on-site at the check-up. The questionnaire included items about the demographic information of the child (age,

gender, height and weight at birth, gestational age at birth, and present illness, if any) and of parents (parental ages at birth). The following information with regard to feeding was also included: method of milk feeding (breast, bottle, or their mixture); whether the child had been weaned and, if so, at what age; feeding behaviors from 1- to 4-month-old of age; and feeding behaviors and problems from 4- to 10-month-old of age. These items were based on previous studies describing normal development of milk feeding behaviors in humans [Lucas et al, 1979; Kaye et al, 1980; Drewett et al, 1981; Carvalho et al, 1982; Bowen-Jones et al, 1982; Newman et al, 1990; Narayanan et al, 1991; Qureshi et al, 2002; Masataka, 2003; Maehara et al, 2008; Igarashi et al, 2009; Törölä et al, 2012; Yoshida et al, 2010; Moral et al, 2010], those in non-human primates [Wolf, 1968; Oftedal, 1984; Tanaka et al, 1993], and specific behaviors seen among children with developmental disorders [Gewolb et al, 2001; Neiva et al, 2007; Costa et al, 2010].

At the check-up, children were assessed by public health nurses whether he or she passed for age-appropriate developmental milestones. Milestones consisted of 8 to 10 questions from 4 domains (gross motor, fine motor, communication, and personal/social) adapted from the Enjoji Scale of Infant Analytical Development, which was developed in Japan to evaluate the development of children from birth to approximately 5 years of age [Enjoji, 2009]. This is only a part of the whole check-up procedure to detect various diseases and disorders. Therefore, all the public health nurses participated in this study were trained in mass-screening methods and experienced in screening for developmental disorders.

Statistical analyses

SPSS Statistics ver. 21 and Stata IC ver. 11 were used. The main dependent variables were whether a child passed the screening for developmental delay by public health nurses. Based on the screening, children were divided into two groups: typically developing (TD) children who passed the screening; and children with suspicious of developmental delay (DD), who failed to pass the screening. The independent variables were each of items asking milk-feeding behaviors in the questionnaire. Sex, mother's age, and father's age were used as independent variables as well to control potential confounding factors. Missing values were substituted by the multiple imputation method. Student's *t* test or chi square test was used to evaluate the differences between the two groups of children. $P < 0.05$ was considered to indicate statistical significance. Using logistic regression analyses, we calculated odds ratios and their 95 percent confidence intervals for items likely to associate with developmental delay.

Ethical considerations

This study followed the Ethical Guidelines for Epidemiological Research specified by the Ministry of Education, Culture, Sports, Science and Technology and the Ministry of Health, Labor and Welfare of Japan. The study protocol was approved by the Ethics Committee of Hamamatsu University School of Medicine as a part of "Research on child development (No. 20-82)".

Results

1) Demographics and milk feeding problems in the participants

Of the 800 questionnaires distributed, 557 mothers agreed to participate in the study and all of them returned the questionnaire (response rate was 69.6 %). It is well known that preterm infants demonstrate some sucking difficulties. To eliminate the effect of immaturity, we excluded children born before 36 weeks of gestation, because suck-swallow-respiration coordination has matured by this age [Mizuno & Ueda, 2003]. Complete sets of responses were collected from 472 mothers including 250 mothers of 18-month-old children and 222 mothers of 3-year-old children. Given the probable recollection bias mothers may have, we firstly conducted statistical analyses on each age population separately.

1-1) results of 18-month-old children

The children were grouped into two groups according to the assessment by public health nurses: TD (n = 198) and DD (n = 52) groups. The characteristics and demographic data for 18-month-old children were summarized in **Table 1**. No significant differences were found in male/female ratio, birth height or weight, gestational age, or weaning age between TD and DD groups. Mean of father's, but not mother's, age at birth was significantly higher in DD group than TD group ($t = 2.37$, $P = 0.019$). With regard to milk feeding behaviors, there were significant group differences in three items, the absence of rooting reflex (three in TD group vs five in DD group, $\chi^2 = 8.72$, $P = 0.003$), lack of smooth suck and rest pattern (13 in TD group vs 12 in DD group, $\chi^2 = 12.48$, $P < 0.001$), and keeping a nipple in the mouth (61 in TD group vs 24 in DD group, $\chi^2 = 4.32$, $P = 0.038$). There were no group differences in other items asking milk feeding problems.

1-2) results of 3-year-old children

According to the results from screening by public health nurses, the children were divided into TD (n = 164) and DD (n = 58) groups. The characteristics and demographic data for the 3-year-old children were summarized in **Table 2**. We found a significant difference in male/female ratio between TD and DD groups ($\chi^2 = 12.53$, $P < 0.001$), suggesting that more boys than girls failed to pass the screening in this population. No significant group differences were found in other demographics including birth height and weight, gestational age, weaning age, and ages of parents at birth. We found a significant group difference in the lack of smooth suck and rest pattern (nine in TD group vs eight in DD group, $\chi^2 = 4.18$, $P = 0.041$). With regard to milk feeding behaviors, the feeding method before 4-month-old of age significantly differed between the two groups ($\chi^2 = 8.353$, $P = 0.015$), implying that more children of DD group fed mixture of breast and bottle rather than breast alone as compared to TD group. After 4 months old, however, the difference in feeding methods between the two groups did not reach a statistically significant level. There were no group differences in other items asking milk feeding problems.

2) Predictive value of lack of smooth suck and rest pattern for developmental delay

As mentioned above, the lack of smooth suck and rest pattern was the valuable that we found a significant difference between TD and DD groups across different age populations, 18-month- and 3-year-old children. To test the possibility that the lack of smooth suck and rest pattern was associated with other properties of a child or his or her parents, we combined two data sets of 18-month- and 3-year-old children, and divided all the children into two groups: children lacking the smooth suck and rest pattern ($n = 20$) and children with smooth suck and rest pattern ($n = 452$). Then, we compared other items in the questionnaire between the two groups. The following five items in the questionnaire showed significant differences between the two groups with regard to their means and frequency: the birth height, birth weight, gestational age, difficult to maintain suckling posture, and neighborhood observation (all $P < 0.05$). A logistic regression analysis revealed that the lack of smooth suck and rest pattern was significantly associated with above five items (**Table 3**), suggesting that these five items were potential confounding factors on the association between the lack of smooth suck and rest pattern and developmental delay.

Using the combined data set of two age populations, a logistic regression model was built to determine the items that account for the developmental delay as a dependent variable (TD group [$n = 362$] vs DD group [$n = 110$]). Six items mentioned above were entered as independent variables: the lack of smooth suck and rest pattern, birth height, birth weight, gestational age, difficult to maintain suckling posture, and neighborhood observation. **Table 4** showed that the developmental delay was significantly associated with the lack of smooth suck and rest pattern ($P = 0.004$) but not with any of other items entered. The odds ratio for the lack of smooth suck and rest pattern was 4.00 (95% confidence interval: 1.57 – 10.17). This indicated that infants who showed the lack of smooth suck and rest pattern might have 4 times higher risk of developmental delay as compared to infants who showed smooth suck and rest pattern.

Discussion

In this study, we conducted a health center-based survey within 2 prefectures in Japan. Mothers evaluated sucking related behaviors of their children using our questionnaire, and the children were assessed their development at the 18-month- or 3-year-old health check-ups. Our study showed that among behaviors in sucking the lack of smooth suck and rest pattern was significantly associated with developmental delay at both 18-month- and 3-year-old of ages. The result suggests that the abnormality in suck and rest pattern of sucking in infants might be a predictor of developmental delay.

The reason for the association between developmental delay and the lack of smooth suck and rest pattern is unknown from the present study. One possible explanation is that the lack of smooth suck and rest pattern may represent an altered neurodevelopment in the domain of reciprocal social interaction in children who would show developmental delay later. The newborn human infants suck in bursts consisting of four to ten sucks at a rate slightly slower than one per second. The bursts are separated by pauses of roughly the same order of magnitude, 4 – 15 sec, with the nipple remaining in the mouse of the infant, resulting in the suck and rest pattern. The clustering of sucks into bursts separated by pauses is the pattern unique to humans (Wolff, 1968). Mothers and other feeders tend to jiggle their infant or feeding bottle in

response to pauses in the infants' sucking. Kaye (1980) has found that the feeding appears to be an exchange of turns, in which the infants' pause is answered by mother's jiggling and the end of jiggling is answered by the next burst, and suggested that human infants learn to anticipate the turn-taking and to assume their role in it as an active partner during suckling. The association between the burst-pause pattern and non-cry vocalization is also suggested in older infants (Masataka, 2003). These findings suggest that the suck and rest pattern is a component of early parent-infant communication. Therefore, infants carrying vulnerability for developmental delay might show the lack of smooth suck and rest pattern. This speculation warrants a further study in which standardized tools are used to evaluate developmental domains of communication and social interaction in larger population.

There are some limitations to our study. This is a survey for mothers of infants and relied solely on their recall. We assessed parents only for limited properties such as age. Therefore, it is unknown whether either of parents might have developmental disorder or its trait. Because the developmental delay is judged by health care nurses in the mass-screening setting, it is difficult to test the association of the sucking problems to a specific domain of development. As a result, our data present here should be regarded as preliminary, although the sample size was effective to detect significance.

In conclusion, our preliminary study suggests that the abnormality in suck and rest pattern of sucking in infants might be a predictor of developmental delay at 18-month- and 3-year-old of ages. In order to reach a firm conclusion, prospective cohort studies would be required.

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Table 1. Demographic data and milk feeding behaviors in 18-month-old children

Item	TD (n=198)	DD (n=52)	P value
Demographic data			
Sex, male/female	89/109	29/23	0.16
Birth height, cm	48.7 [2.2]	48.0 [2.4]	0.54
Birth weight, g	3034.4 [405.3]	2938.9 [542.2]	0.24
Gestational age at birth, weeks	39.0 [1.37]	38.3 [2.8]	0.09
Mother's age at birth, years	30.6 [4.9]	31.4 [4.9]	0.33
Father's age at birth, years	31.9 [5.6]	33.9 [5.3]	0.019 *
Weaning age, months	12.6 [2.9]	13.0 [2.5]	0.77
Feeding behaviors before 4-month-old of age			
Feeding method			0.77
Breast only	109	26	
Bottle only	9	2	
Mixture of breast and bottle	80	24	
Difficult to maintain suckling posture	28	13	0.06
Drink milk not well	6	1	0.67
No rooting reflex	3	5	0.003 **
Feeding behaviors from 4- to 10-month-old of age			
Feeding method			0.88
Breast only	116	29	
Bottle only	10	3	
Mixture of breast and bottle	72	20	
Number of feedings per day	7.5 [2.4]	7.4 [2.4]	0.81
Duration of each feeding, min	14.7 [6.7]	15.1 [8.2]	0.73
Nocturnal feeding	166	44	0.89
Abnormal sucking behaviors			
No signs on hunger or full stomach	9	2	0.87
Lasting breast tension after suckling	29	8	0.89
Playing with nipples without drinking	24	10	0.18
Arrhythmic sucking	75	20	0.94
Lack of smooth suck and rest pattern	13	12	< 0.001 **
Feeding irregularly	142	30	0.05
Biting a nipple or breast	52	15	0.71
Keeping a nipple in the mouth	61	24	0.038 *
Observation of surroundings	156	44	0.35

Data are shown in mean and [SD]. Note a significance: * $p < 0.05$; * $p < 0.01$.

Abbreviations: TD, typically developing; DD, suspicious of developmental delay; SD, standard deviation.

Table 2. Demographic data and milk feeding behaviors in 3-year-old children

Item	TD (n=164)	DD (n=58)	P value
Demographic data			
Sex, male/female	66/98	39/19	< 0.001
Birth height, cm	48.4 [2.2]	48.7 [2.6]	0.46
Birth weight, g	3049.7 [396.5]	2995.0 [378.4]	0.36
Gestational age at birth, weeks	39.0 [1.3]	38.6 [1.5]	0.071
Mother's age at birth, years	30.6 [5.0]	31.6 [4.9]	0.16
Father's age at birth, years	32.1 [5.9]	33.0 [6.0]	0.35
Weaning age, months	17.5 [8.1]	17.1 [8.0]	0.71
Feeding behaviors before 4-month-old of age			
Feeding method			0.015
Breast only	93	22	
Bottle only	0	1	
Mixture of breast and bottle	71	35	
Difficult to maintain suckling posture	21	5	0.39
Drink milk not well	8	0	0.09
No rooting reflex	5	0	0.18
Feeding behaviors from 4- to 10-month-old of age			
Feeding method			0.05
Breast only	96	24	
Bottle only	8	6	
Mixture of breast and bottle	60	28	
Number of feedings per day	7.5 [2.3]	7.0 [2.1]	0.44
Duration of each feeding, min	15.2 [7.4]	15.5 [6.9]	0.67
Nocturnal feeding	130	34	0.39
Abnormal sucking behaviors			
No signs on hunger or full stomach	8	2	0.65
Lasting breast tension after suckling	29	8	0.49
Playing with nipples without drinking	31	7	0.24
Arrhythmic sucking	58	22	0.73
Lack of smooth suck and rest pattern	9	8	0.041
Feeding irregularly	100	42	0.12
Biting a nipple or breast	53	11	0.06
Keeping a nipple in the mouth	70	17	0.07
Observation of surroundings	135	45	0.43

Data are shown in mean and [SD]. Note a significance: * $p < 0.05$; * $p < 0.01$.

Abbreviations: TD, typically developing; DD, suspicious of developmental delay; SD, standard deviation.

Table 3. Result of logistic-regression analysis on relationships between "Lack of smooth suck and rest pattern" and other items

	Odds ratio	SE	z score	P value	95% CI
Birth height	3.76	2.15	2.32	0.020 *	1.23 - 11.54
Birth weight	2.97	1.46	2.22	0.027 *	1.13 - 7.8
Gestational age at birth	2.83	1.33	2.22	0.026 *	1.13 - 7.12
Difficult to maintain suckling posture	0.36	0.17	-2.15	0.032 *	0.14 - 0.91
Observation of surroundings	2.52	1.13	2.05	0.040 *	1.04 - 6.11

Note a significance: * $p < 0.05$. Abbreviations: CI, confidence interval; SE, standard error.

Table 4. Result of logistic-regression analysis on relationships between developmental delay and questionnaire items

	Odds ratio	SE	z score	P value	95% CI
Lack of smooth suck and rest pattern	4.00	1.90	2.91	0.004 **	1.57 - 10.17
Birth height	0.53	0.39	-0.84	0.40	0.12 - 2.26
Birth weight	1.94	1.09	1.19	0.23	0.65 - 5.84
Gestational age at birth	1.62	0.72	1.1	0.27	0.68 - 3.87
Difficult to maintain suckling posture	0.58	0.23	-1.34	0.18	0.26 - 1.28
Observation of surroundings	0.57	0.25	-1.23	0.22	0.23 - 1.38

Note a significance: ** $p < 0.01$. Abbreviations: CI, confidence interval; SE, standard error.

Appendix 1. Contents of the questionnaire

Category	Survey Contents [anser]
Demographic data	• Gender of child [male or female] • Perinatal situation : height [cm] • Perinatal situation : weight [g] • Perinatal situation : fetus week number [weeks] • Mother's age at birth [years old] • Father's age at birth [years old] • Does a child have a disease under treatment now? [yes or no] • Weaning period [months or ongoing]
Feeding behaviors before 4-month-old of age	• Feeding method [Breast only or Bottle only or Mixture of breast and bottle] • During lactation, do you often had a hard time to embrace a child, and to support it (including not only the mother's milk but also the artificial milk)? [yes or no] • Did the child suckling mother's milk or baby formula well? [yes or no] • When you brought your nipple close to cheeks of a child, did your child able to hold nipple in his/her mouth well (including the feeding bottle)? [yes or no]
Feeding behaviors from 4- to 10-month-old of age	• Feeding method [Breast only or Bottle only or Mixture of breast and bottle] • In the case of combination [ratio of mother's milk and baby formula] • The suckling number of times during one day [times] • The time that one times suckling takes [min] • Do you often feed milk in night? [yes or no]
Abnormal sucking behaviors	• In the case of hunger and a full stomach, a child cried, and did you show signal? [yes or no] • Does a stitch of the breast often continued in spite of what I nursed? [yes or no] • Did the child not often sucking mother's milk with only a nipple in its mouth? [yes or no] • When mother's milk became hard to flow at the time of the nursing, a child greatly moved a mouth, and do you often quickened rhythm? [yes or no] • Was burst-pause(taking a rest when I suck mother's milk, and beginning to breathe it again) often smooth? [yes or no] • Did you maintain the same in time to do lactation? [yes or no] • A child bit a nipple, and I strongly breathed it and did it, and were you often hurt on the breast? [yes or no] • Did a child not often separate it with a nipple in its mouth? [yes or no] • During the feeding, does child often looked around to see the face of mother in the circumference? [yes or no]
Entry by the public health nurse	• The passage of the screening [pass or failure] • State of the child at the health check-up [nothing peculiar or hyperactive or unresponsive or unable to distinguish]