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Joint Attention Development in Low-risk Very Low Birth Weight Infants at Around 18 Months of Age

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The purpose of this study was to clarify the developmental characteristics of joint attention in very low birth weight (VLBW) infants with a low risk of complications. Section B of the Checklist for Autism in Toddlers (CHAT) was administered to 31 VLBW and 45 normal birth weight (NBW) infants aged 18–22 months, while the sessions were recorded with a video camera. A semi-structured observation scale was developed to assess infants' joint attention from the video footage, and was shown to be reliable. VLBW, compared to NBW, infants showed significantly poorer skills in 2 of 4 items on responding to joint attention, and in 6 of 10 items on initiating joint attention. VLBW infants need more clues in order to produce joint attention. The difficulty was attributed to insufficient verbal and fine motor function skills. Continuous follow-up evaluation is essential for both high-risk and low-risk VLBW infants and their parents.

INTRODUCTION

Progress in medical treatment has improved the survival rate of very low birth weight (VLBW: < 1,500 g at birth) infants; there is growing concern, however, for their neurodevelopmental outcomes. Studies have shown that children with low birth weight (LBW: < 2,500 g at birth) and/or preterm infants (birth occurring at < 37 weeks of gestation) have an increased risk of developing Autism Spectrum Disorder (ASD),[11,12] which is a neurodevelopmental disorder involving significant social communication and behavioral impairments.[1] Many parents of children with ASD become apprehensive about their child's development before 18 months of age,[10] with one primary concern being an extraordinary lack of eye contact.[1, 6, 8] Early impairment of eye contact or dyadic behaviors is indicative of disability in triadic joint attention behaviors.[6] Joint attention refers to the ability to share attention with another person regarding an object or event of interest[8] and develops between 9 and 18 months of age.[22] It underpins the development of cognitive function, communication capacity, and social skills in infants,[7] thus offering valuable insight into child development and providing important clues for the early detection of neurodevelopmental difficulties.

The absence of joint attention is one of the earliest indications of ASD.[2] Earlier studies have suggested that VLBW and/or preterm infants have different characteristics in the development of joint attention from normal birth weight (NBW: 2,500 - 4,000 g at birth) and/or full-term (birth occurring at 37 - 42 weeks of gestation) infants[5, 23]. Preterm LBW

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infants with a high risk of complications (presence of major neonatal morbidity and cerebral pathology) experience difficulty in developing joint attention skills that most full-term infants acquire by 24 months of age.[14] Meanwhile, few studies have focused on the development of joint attention in VLBW or preterm infants aged 12-24 months, or investigated joint attention behaviors in VLBW or preterm infants with a low risk of complications (absence of major neonatal morbidity and cerebral pathology). Consequently, the developmental progress of joint attention in these infants is currently less than well defined. Therefore, it is important to provide reliable evidence to support the developmental evaluation of joint attention in VLBW infants with a low risk of complications.

The purpose of this study was to clarify the characteristics in the development of joint attention in VLBW infants with a low risk of complications compared to NBW infants. As a means of achieving this goal, we developed a new tool for measuring joint attention in 18-months-old infants.

SUBJECTS AND METHODS

Subjects

The participants consisted of 45 NBW and 31 VLBW infants aged 18-22 months. The VLBW infants were recruited from a support program for VLBW infants and their parents, and the NBW infants were recruited from a hospital with a pediatric outpatient department from June 2013 to July 2014. Infants were evaluated from their clinical records. Inclusion criteria were as follows: (1) birth weight of < 1,500 g and born < 37 gestational weeks for VLBW infants; (2) birth weight of 2,500 - 4,000 g and born between 37 - 41 gestational weeks for NBW infants; and (3) at low risk of neurodevelopmental deficits (no severe chronic lung disease, necrotizing enterocolitis, neonatal sepsis, periventricular leukomalacia and intraventricular hemorrhage above grade II) for VLBW infants. Exclusion criteria were as follows: (1) presence of a congenital or postnatally acquired abnormality; (2) overt intellectual or cognitive problems; (3) exhibiting neurodevelopmental deficits or behavioral problems on routine clinical examination.

Procedures

First, we applied the 4-point scale Japanese version of the M-CHAT[18] to the parents of study subjects. Second, we applied Section B of the Checklist for Autism in Toddlers (CHAT)[2] to our study subjects and their parents according to the Behavior Observation Manual for Early Detection of Children with Developmental Difficulties[21] and the general administration guidelines of the Early Social Communication Scales (ESCS).[16] All 4 observational sessions were recorded by a video camera (Figure 1). Third, we developed the Joint Attention Observation Scale (JAOS), as a tool for use in this research, for the purpose of assessing infants' joint attention behaviors during the course of the video-recorded sessions in CHAT Section B. The JAOS comprises 14 yes/no items that are evaluated by watching the 4 recorded observational sessions, and 3 or 4 items are assessed in each session. The scale items were devised to conform to the criteria of the ESCS[16] in evaluating two components: responding to joint attention (RJA; items 2, 5, 7, and 11) and initiating joint attention (IJA; all other items). RJA is the ability to follow the direction of eye gaze, head turn, or pointing of others, whereas IJA refers to the ability to use direction of gaze and gestures to direct the attention of others.[17] The first author examined the video footage of the sessions of Section B of the CHAT using the JAOS.

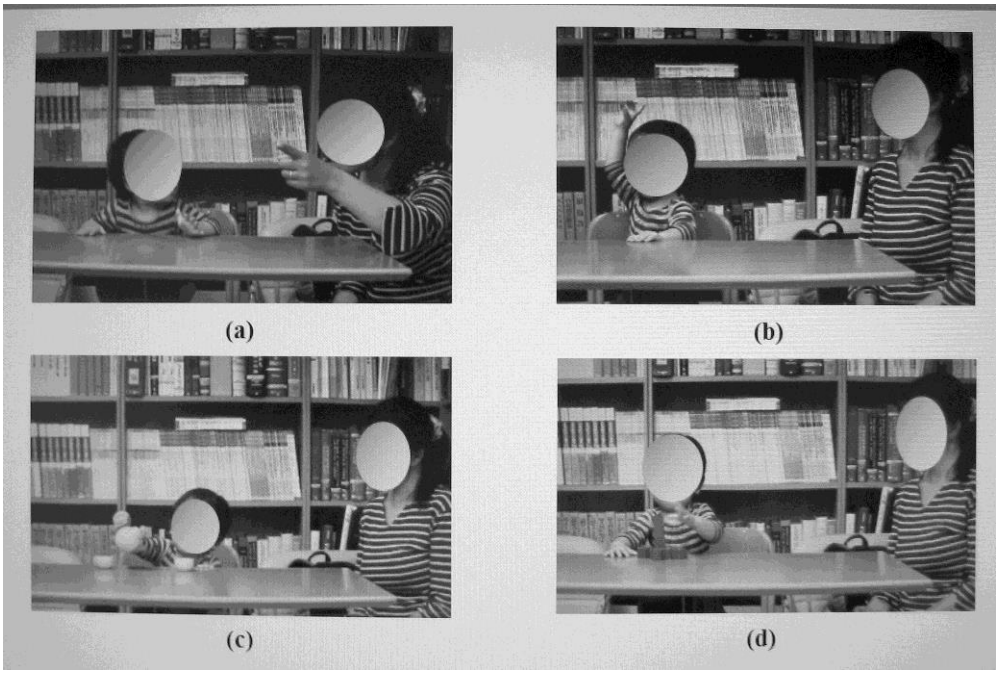

Fig 1. Administration of Section B of the Checklist for Autism in Toddlers[2]
(a) gaze monitoring; (b) protodeclarative pointing; (c) pretend play; and (d) building a tower of blocks.

Data analyses

Given that the JAOS is a new measure, evaluation of its reliability and validity is a vital requirement. As for the reliability, 6 raters (2 public health nurses, 2 special-needs school teachers, a clinical psychologist and a pediatric nurse) participated in the inter-rater reliability scoring trial. First, the author provided a clear explanation of the JAOS along with sample visual images. Next, agreement was examined between raters in regard to the JAOS items. Then, the 6 raters examined 10% of the footage from the sessions both subject groups, which was randomly selected. Finally the reliability of the JAOS was assessed by calculating Fleiss' kappa coefficient. To assess criterion-related validity, we scored JAOS (yes=1, no=0) and 23 items of the Japanese version of the 4-point M-CHAT scale (usually=3, often=2, seldom=1, never=0). Spearman's rank-correlation coefficient was calculated between scores on the JAOS and the M-CHAT items.

Statistical analyses were performed using a chi-square test or Fisher's exact test to evaluate the differences between the VLBW and NBW groups and to compare the results of JAOS evaluation between VLBW and NBW infants.

Ethical Approval

This study was approved by the Ethics Committee of Kobe University Graduate School of Health Sciences in accordance with the World Medical Association Declaration of Helsinki (approval date: October 17, 2011). Parents of the infants were informed of the details of the research. Written informed consent was obtained from all parents.

RESULTS

Subjects

Table I presents the characteristics of the VLBW and NBW infants, and shows that there was no significant difference in the chronological age of the NBW and corrected age of the VLBW infants. Similarly, there was no significant difference between the groups regarding gender or birth order.

Table I. Characteristics of very low birth weight and normal birth weight infants

	VLBW infant (n = 31)		NBW infant (n = 45)	
Birth weight (mean±SD)	989±313 g		3057±384 g	
1,000-1,500g at birth	15	(48%)		
<1,000g at birth	16	(52%)		
Chronological age (mean±SD)	21.5±1.3 months		19.1±1.1 months	
Corrected age (mean±SD)	19.1±1.3 months			
Gestational age (mean±SD)	28.4±3.0 weeks		39.1±1.3 weeks	
Preterm	16	(52%)		
Extremely preterm	15	(48%)		
Small for gestational age	11	(35%)		
Gender				
Male	16	(52%)	27	(60%)
Female	15	(48%)	18	(40%)
Birth order				
First born	23	(74%)	39	(87%)
Subsequent born	8	(26%)	6	(13%)
Maternal age at birth (mean±SD)	34.5±4.8 years			
20-24	1	(3%)		
25-29	3	(10%)		
30-34	9	(29%)		
35-39	13	(42%)		
40-44	5	(16%)		

Extremely preterm: birth occurring at <28 weeks of gestation; NBW: normal birth weight (weighing 2,500 to < 4,000g at birth); preterm: birth between 28-36 weeks of gestation; SD: standard deviation; small for gestational age: weighing <10th percentile for gestational age at birth; VLBW: very low birth weight (weighing <1,500g at birth).

Reliability and validity of the JAOS

The reliability results for individual JAOS items are shown in Table II. Among the 6 raters involved in the reliability scoring trial, the average Fleiss' kappa coefficient across the 14 JAOS items was .83 (range: .72-1.0).

Table II. Reliability results for individual Joint Attention Observation Scale items, using Fleiss' kappa coefficient

	Inter-rater reliability
I. Gaze monitoring	
1. Points at the object before the parent points at it	.87
2. Looks at the object after the parent points at it	.77
3. Looks at the object and then switches gaze to the parent	.80
II. Protodeclarative pointing	
4. Finds and then points at the light	.85
5. Looks at the light after tester looks at it	.80
6. Looks at the light and then switches gaze to tester	.85
III. Pretend play	
7. Looks at tester → tester gives toys → watches toys	.77
8. Tries to share attention with the tester before pretending	.72
9. Pretends or plays with toys and then switches gaze to the tester	1.00
10. Pretends or plays with toys and then switches gaze to the parent	.79
IV. Building a tower of blocks	
11. Looks at tester → tester gives blocks → watches blocks	.79
12. Tries to share attention with the tester before building blocks	.78
13. Builds or plays with blocks and then switches gaze to the tester	1.00
14. Builds or plays with blocks and then switches gaze to the parent	.87

The criterion-related validity was examined using Spearman's rank-correlation coefficient between the score of JAOS and the M-CHAT items. As indicated in Table III, the JAOS score was fairly correlated with the total score of M-CHAT. Among 23 M-CHAT items, 8 items were associated with the JAOS score and 5 items were moderately correlated with the JAOS score. Among 4 M-CHAT items evaluating joint attention (item 7, 9, 15, and 17), 3 items were relevant to the JAOS score. There was no definite correlation between item 17 and the JAOS score.

Table III. Relationships between the total score of Joint Attention Observation Scale and M-CHAT items

	Total score of JAOS
1. Enjoys being swung, bounced on your knee, etc.	.166
2. Shows interest in other children	.037
3. Likes climbing on things such as stairs	.146
4. Enjoys playing peek-a-boo/hide-and-seek	.085
5. Play pretend, for example, to talk on the phone	.157
6. Uses index finger to point, to ask for something	.457**
7. Uses index finger to point, to indicate interest in something	.379**

8. Plays properly with toys	.287*
9. Brings objects over to parents to show them something	.306**
10. Looks at the parent in the eye for more than a second or two	.228*
11. Seems oversensitive to noise	.135
12. Smiles in response to parent's face or his/her smile	.245*
13. Imitates parent	.332**
14. Responds to his/her name when parent calls	.146
15. Looks at a toy across the room if the parent points at it	.283*
16. Able to walk	.309**
17. Looks at things the parents are looking at	.127
18. Makes unusual finger movements near his/her face	.297**
19. Tries to attract parent's attention to his/her own activity	.271*
20. Wonders if the child has hearing problem	.173
21. Understands what people say	.380**
22. Stares at nothing or wanders with no purpose	.295**
23. Looks at parent's face to check his/her reaction	.100
Total score of M-CHAT	.468**

* p<.05, ** p<.01 (Spearman's rank-correlation coefficient)

JAOS evaluation for VLBW and NBW infants

Differences between VLBW and NBW infants in terms of scores on the JAOS are shown in Table IV. Significant differences between the two groups were observed in RJA items 7 and 11. However, no significant differences were found for items 2 and 5. Significant differences between the two groups were also observed in IJA items 4, 6, 9, 12, 13, and 14. However, there were no significant differences for items 1, 3, 8, and 10.

Table IV. Comparison between very low birth weight and normal birth weight infants on the Joint Attention Observation Scale[a]

	VLBW Infant (n = 31)		NBW Infant (n = 45)			
	Yes	No	Yes	No	P	
I. Gaze monitoring						
1. Points at the object before the parent points at it *(IJA: indication of interest)*	1	30	8	37	.053	
2. Looks at the object after the parent points at it *(RJA with two clues)*	29	2	37	8	.137	
3. Looks at the object and then switches gaze to the parent *(IJA: social referencing)*	2	29	4	41	.527	
II. Protodeclarative pointing						
4. Finds and then points at the light *(IJA: showing)*	6	25	19	26	.037	*

5. Looks at the light after the tester looks at it *(RJA with two clues)*	6	25	16	29	.126	
6. Looks at the light and then switches gaze to the tester *(IJA: social referencing)*	11	20	28	17	.022	*
III. Pretend play						
7. Looks at the tester → tester gives toys → watches toys *(RJA with a clue)*	25	6	45	0	.003	**
8. Tries to share attention with the tester before engaging in pretend play *(IJA: social referencing)*	19	12	35	10	.119	
9. Pretends or plays with toys and then switches gaze to the tester *(IJA: social referencing)*	26	5	44	1	.038	*
10. Pretends or plays with toys and then switches gaze to the parent *(IJA: social referencing)*	5	26	6	39	.491	
IV. Building a tower of blocks						
11. Looks at tester → tester gives blocks → watches blocks *(RJA with a clue)*	25	6	45	0	.003	**
12. Tries to share attention with the tester before building blocks *(IJA: social referencing)*	17	14	36	9	.019	*
13. Builds or plays with blocks and then switches gaze to the tester *(IJA: social referencing)*	24	7	45	0	.001	**
14. Builds or plays with blocks and then switches gaze to the parent *(IJA: social referencing)*	5	26	17	28	.041	*

IJA: initiating joint attention; NBW: normal birth weight; RJA: responding to joint attention; VLBW: very low birth weight.

[a] Chi-square test or Fisher's exact test: * $p < .05$; ** $p < .01$.

DISCUSSION

The aim of the present study was to clarify the developmental characteristics of joint attention in VLBW infants with a low risk of complications. To achieve the aim, we developed the JAOS for assessing infants' joint attention behaviors from the video footage collected when administering Section B of the CHAT. We obtained an average kappa coefficient of .83, indicating strong inter-rater reliability for the JAOS.[13] As for the criterion-related validity, the JAOS score was correlated with the total score and 3 out of 4 joint attention related items of the M-CHAT.

We found no differences in the 2 RJA items (items 2 and 5). However, significant differences were shown in the other 2 items (items 7 and 11). Pointing and head-and-eye orientation provide clues for the establishment of joint attention,[7] and this is thought to be a factor that affects infants' RJA behavior. The items that showed significant differences between both groups consisted of only one clue, i.e., the change in the tester's eye orientation, for the RJA to be established. Earlier studies show that infants should be able to produce RJA without clear clues as they grow up. Although infants at 9 months of age have been found to be unable of producing the RJA with clues involving only the eyes, about 50% of the infants at 12 and 14 months of age could do so.[15] Likewise, 42% of the infants at 18 months of age were found to successfully produce the RJA when the clue was only a change in the tester's eye orientation.[4] Our findings indicate that VLBW infants require more pointing or head and eye orientation in order to produce RJA compared to NBW infants.

The current results did not show any differences in IJA between the groups, as evaluated in the gaze monitoring session. Contrary to gaze monitoring, protodeclarative pointing is a

self-motivated gesture of the index finger to indicate to others an object of interest.[2] The only clue for the behavior is the tester's question, "Where's the light?" or "Show me the light." Even if the infants successfully pointed at the light, they may still have felt uncertain about their behavior; as such, many infants turned their gazes to their parents to check their reactions. Additionally, previous research suggested that preterm infants have difficulty in sustaining their attention regardless of their risk status.[24] These views may support the differences in the outcomes of items 3 and 6 as well as the significant between-group difference for item 6. Items 3, 6, and all of the IJA items in the pretend play session and block building session are pertinent to social referencing. Significant differences were observed in all of the IJA items in the block building session, whereas only item 9 exhibited a significant difference between the groups during the pretend play session. Social referencing comprises looking at the facial expressions of others for help in deciding what to do.[25] A factor that may affect the production of social referencing is the nature of the sessions. Block building requires fine motor function than pretend play does. The higher the infants build blocks, the more social referencing behaviors occur, because infants try to receive praise from the tester and their parents each time they accomplish the build-up. Unfortunately, LBW and/or preterm infants are significantly poorer in motor development [20] and, thus, built fewer towers with blocks compared to NBW infants. Consequently, more significant differences were observed in IJA items in the session for building a tower of blocks than in the pretend play session. Overall, the present study indicates that VLBW infants lack sufficient verbal and fine motor function to match the level of IJA produced by NBW infants.

Visual and cognitive impairment is not just confined to VLBW and/or preterm infants who have a high risk of complications. Preterm VLBW preschoolers without neonatal brain disorders and with normal cerebral ultrasound findings showed poor spatial attention accuracy and a higher incidence of stereopsis impairment.[23] Low-risk LBW preschoolers at 5 years of age showed lower scores in visual motor ability and visual perception skills compared to NBW preschoolers.[5] VLBW infants in the current study were not diagnosed with congenital, neurological, developmental, or cranial nerve disorders, but their joint attention skills were significantly lower than those in NBW infants. A conceivable explanation for this result is related to fetal brain development. Caravale et al[5] indicated that both growth and networking of the brain are completed during the last 6 weeks of gestation, and that the brain weighs only half of the term weight at 30 weeks of gestation. Cerebral growth and maturation prior to term are crucial aspects for the prognosis of VLBW infants.[3]

Although the explicit causal relationships among joint attention skills and brain prematurity have not been established, problems in joint attention have the potential to become increasingly obvious as infants mature. The current study shows 2 characteristics in the development of joint attention in VLBW infants with a low risk of complications compared to NBW infants: VLBW infants need more clues so as to produce RJA, and inadequate verbal and fine motor function lead to an insufficient development of IJA in VLBW infants. These results support the previous studies' findings that VLBW infants have different characteristics in the development of joint attention from NBW infants.

As previously mentioned in introduction, joint attention development is impaired in the children with ASD and LBW infants have an increased risk of developing ASD. The findings that VLBW infants showed poor skills in joint attention indicate the need of follow-up assessment from the perspective of early detection of ASD. Sayeur et al[19] suggested that clinical follow-up after the first year of life is vital for all children born preterm, and it has been suggested that LBW children should ideally be followed to early school age so as to

measure more subtle problems.[9] Therefore, continued examinations and follow-up care are essential for both high-risk and low-risk VLBW infants and their parents.

The present study has some limitations. Our study's generalizability is limited by the small sample size and restricted data collection facilities. Further research is, thus, needed to examine the association between medical treatment and joint attention skills in VLBW infants. However, the sample size did not stand up to close examination in that regard. Second, we did not assess infants' DQs. Joint attention plays a key part in shaping infants' cognition. However, it is difficult to evaluate joint attention from the value of DQs. Given that joint attention is a major factor in determining ASD, further studies are needed to examine the possibility to assess joint attention from the viewpoints of DQs. Third, we did not collect data of maternal ages of NBW infants. In the future, taking both study groups' maternal age into account would be valuable in assessing the association between maternal age and joint attention development.

In conclusion, our study demonstrated significant differences in both RJA and IJA skills between VLBW infants with a low risk of complications and NBW infants at 18-22 months of age. Our findings suggest that continuous follow-up evaluation is essential for both high-risk and low-risk VLBW infants and their parents.

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