Brain & Development 43 (2021) 181–185

BRAIN & DEVELOPMENT
Official Journal of the Japanese Society of Child Neurology

www.elsevier.com/locate/braindev

Original article

Shuffling babies and autism spectrum disorder

Yu Okai [a,b], Tomohiko Nakata [a], Kiyokuni Miura [b], Atsuko Ohno [b], Rie Wakako [b], Osamu Takahashi [b], Yuki Maki [a], Masaharu Tanaka [a], Yoko Sakaguchi [a,b], Yuji Ito [a,b], Hiroyuki Yamamoto [a], Hiroyuki Kidokoro [a], Yoshiyuki Takahashi [a], Jun Natsume [a,c,*]

[a] *Department of Pediatrics, Nagoya University Graduate School of Medicine, Nagoya, Japan*
[b] *Toyota Municipal Child Development Center, Toyota, Japan*
[c] *Department of Developmental Disability Medicine, Nagoya University Graduate School of Medicine, Nagoya, Japan*

Received 1 July 2020; received in revised form 31 July 2020; accepted 11 August 2020

Abstract

Background and purpose: Bottom shuffling is a locomotion strategy that precedes independent walking in some infants. Shuffling babies are generally considered to have favorable outcomes. The aim of the present study was to reveal clinical features and neurodevelopmental outcomes of shuffling babies who visited a child developmental center.

Methods: We studied 48 shuffling babies who visited Toyota Municipal Child Development Center from April 2007 to March 2015. We excluded patients with cerebral palsy, Down syndrome, or congenital disorders. In 2018, we retrospectively reviewed the clinical charts of the enrolled children. We investigated family history, neurological findings, and the developmental outcome during the follow-up period.

Results: During the follow-up period, 20 children (42%) were diagnosed with ASD. Gross motor development in infancy was not different between infants with and without ASD. The rate of poor eye contact at the first visit and a delay in the first word speech were significantly higher in infants with ASD than in infants without ASD. A family history of bottom shuffling was significantly less frequent in infants with ASD (10%) than in those without (39%).

Conclusion: Some of bottom shufflers may represent ASD during follow-up. Paying attention to social and cognitive functions in shuffling babies is important.

Keywords: Autism spectrum disorder; Developmental disability; Bottom shuffling

1. Introduction

Bottom shuffling is a locomotion strategy that precedes independent walking in some infants. Bottom shuffling infants move forward in a sitting position by using the buttocks, legs, or arms. The prevalence of shuffling babies has been reported to be 3–9% in various countries [1–4]. Although some infants with cerebral palsy or Down syndrome display bottom shuffling, most non-symptomatic shuffling babies are considered to have normal developmental outcomes, except for mild hypotonia or a delay in independent walking [1,3]. On the other hand, our preliminary study revealed autism spectrum disorder (ASD) in some of shuffling babies (unpublished data). Because of the lack of recent studies of a large number of patients, the long-term

Abbreviations: ASD, autism spectrum disorder; DQ, developmental quotient

* Corresponding author at: Department of Pediatrics, Nagoya University Graduate School of Medicine, 65 Tsurumai-cho, Showa-ku, Nagoya, Aichi 466-8550, Japan.
E-mail address: junnatsu@med.nagoya-u.ac.jp (J. Natsume).

https://doi.org/10.1016/j.braindev.2020.08.007
0387-7604/

developmental outcomes in shuffling babies have not been fully clarified.

The reported prevalence of ASD has increased in the last decade, possibly due to a change in concept and wider recognition [5]. Early diagnosis and intervention may improve long-term social and behavioral outcomes in children with ASD [6]. For early diagnosis of ASD, identification of behavioral markers is important in the setting of medical check-ups of infants. Whether we can identify early signs of ASD in infants with bottom shuffling has not been revealed.

The aim of the present study was to reveal clinical features and neurodevelopmental outcomes of shuffling babies. We investigated the clinical characteristics and developmental outcomes of shuffling babies who visited a public children's rehabilitation center. We hypothesized that many children with ASD is found in shuffling babies, and that early signs suggesting ASD can be identified in infancy.

2. Methods

2.1. Subjects

We enrolled bottom shuffling infants who visited Toyota Municipal Child Development Center from April 2007 to March 2015. Toyota Municipal Child Development Center covers a population of 480,000 in Toyota and Miyoshi Cities (annual number of births: 4500). One of four pediatric neurologists (K.M., Y.I., Y.S., and Y.O.) diagnosed bottom shuffling in each infant with a clinical examination and parent interviews. Bottom shuffling was defined as moving forward in the sitting position using the buttocks, legs, or arms as a locomotor strategy before walking. We included children not only with a chief complaint of bottom shuffling, but also children who visited for other purposes and in whom bottom shuffling was identified in the histories or examinations. We excluded patients with cerebral palsy, Down syndrome, and seven patients with hydrocephalus, microcephaly, focal cortical dysplasia, white matter lesion of unknown etiology, Klinefelter syndrome, congenital multiple malformation, and congenital heart disease.

2.2. Data collection

Two of the authors (Y.O. and K.M.) reviewed the clinical charts of the enrolled children in 2018. Clinical data including sex, age at the first visit, family history of bottom shuffling, and age at the final visit (up to December 2018) were investigated. Family history was investigated in parents and siblings. Developmental milestones (head control, sitting without support, shuffling, crawling on hands and knees, walking independently, and first word speech) and neurological examination at the first visit (eye contact and muscle tone) were collected. As some of the shuffling babies occasionally crawled on their hands and knees, we investigated occasional crawling in the shufflers. During the follow-up period, diagnosis of ASD was made by child psychiatrists (O.T. and R.W.) according to the Diagnostic and Statistical Manual (DSM) of Mental Disorders published by American Psychiatric Association [7,8]. DSM-IV was used for the diagnosis before April 1, 2017, and DSM-5 after. Follow-up of the psychomotor development was performed until 3 years of age or longer. If follow-up was terminated before the child was 3 years old, two of the authors (Y.O. and K.M.) interviewed the families by phone or letter.

During the follow-up period, we evaluated the developmental quotient (DQ) with the revised version of the Enjoji Developmental Test, which comprises six domains of body movement, hand movement, fundamental habit, human relationship, speech, and language understanding [9]. We used the latest score when children had undergone DQ tests two or more times.

2.3. Statistical analysis

Data were analyzed using SPSS version 24 (IBM Japan Ltd., Tokyo, Japan). We compared the demographics, developmental milestones, occasional crawling on hands and knees, and DQ between children with diagnosis of ASD (ASD group) and those without (non-ASD group) using the Fisher's exact test or Mann-Whitney U test. Significance was established at $p < 0.05$.

2.4. Ethical consideration

This research was conducted after obtaining the approval of the ethics committee at Toyota Municipal Child Development Center. Because of the nature of the retrospective study by clinical chart review, informed consent from the families was not required. For the patients in whom we interviewed the families about the developmental status by phone or letter, written informed consent was obtained from the families.

3. Results

3.1. Clinical characteristics

We enrolled 51 children (25 males) with bottom shuffling. Two children were born prematurely at 35 weeks of gestational age, but no other perinatal complications were seen in these children. The age at the last follow-up ranged from 3 years to 12 years. In 13 children, the follow-up at the center was terminated before the child was 3 years old, and the outcome was obtained with an interview with the families by phone or letter. The families of three children could not be contacted and

these children were excluded from the present study. All children in whom the follow-up was terminated before 3 years old and the interview was conducted with families by phone or letter were not diagnosed with ASD. Among the 48 shuffling babies, 20 (42%) children were diagnosed with ASD during the follow-up period. The age at the first visit ranged from 10 months to 3 years 4 months (median 1 year 7 months) in the 20 children with ASD, and from 7 months to 1 year 8 months (median 1 year 2 months) in the 28 children without ASD. The reason for the first visit was an abnormality of motor development including bottom shuffling in 13 children in the ASD group and 24 in the non-ASD group, mental or language developmental delay in seven in the ASD group and three in the non-ASD group, and strabismus in one in the non-ASD group. The age at the last follow-up ranged from 3 years to 12 years (median 6 years) in the ASD group and from 3 years to 12 years (median 5 years) in the non-ASD group. Clinical characteristics of the ASD and non-ASD groups are shown in Table 1. A family history of bottom shuffling was significantly higher in the non-ASD group (39%) than the ASD group (10%) ($p = 0.046$). At the first visit, poor eye contact was noticed in 12 children (60%) in the ASD group and none in the non-ASD group ($p < 0.001$). The incidence of hypotonia at the first visit was not significantly different between the two groups. No significant difference was found in occasional crawling between the ASD and non-ASD groups.

3.2. Developmental milestones

Developmental milestones in the ASD and non-ASD groups are shown in Fig. 1. In the two children born prematurely at 35 weeks of gestational age, the developmental milestones were evaluated according to the corrected age. No significant differences were seen in the age of gross motor development (head control, rolling, sitting without support, shuffling, and walking independently) between the ASD group and the non-ASD group. Regarding language development, the age of first word speech was 11–28 months (median 15 months) in the ASD group and 9–24 months (median 12 months) in the non-ASD group. One child in the ASD group did not develop word speech at the last follow-up period and was excluded from the statistical analysis. The age at the first word speech was significantly delayed in the ASD group ($p = 0.025$).

3.3. Dq

DQ was evaluated in 34 children (19 children in the ASD group and 15 in the non-ASD group). The age at the evaluation of DQ ranged from 12 months to 63 months (median 38.5 months). The total DQ of all domains was not significantly different between children with and without ASD (Table 1). Among the six domains of DQ, fundamental habit and human relationship were significantly lower in the ASD group ($p = 0.010$ and 0.008, respectively).

4. Discussion

We evaluated clinical features and developmental outcomes in 48 shuffling babies. During the follow-up period, 42% of the shuffling babies were diagnosed with ASD. The rate of poor eye contact at the first visit and a delay in language development were significantly higher in the children with ASD, and a family history of bottom shuffling was less frequent in children with ASD.

Bottom shuffling is observed in several disorders. Some infants with cerebral palsy show bottom shuffling [1,3]. Diplegia or hemiplegia may affect the locomotor strategies in infancy. Infants with Down syndrome also

Table 1
Characteristics of shuffling babies.

	With ASD n = 20	Without ASD n = 28	p value
Male	12 (60)	12 (43)	0.380
Family history	2 (10)	11 (39)	0.046
Poor eye contact at first visit	12 (60)	0 (0)	<0.001
Hypotonia at first visit	17 (85)	23 (82)	1.000
Occasional crawling	12 (60)	16 (54)	1.000
Developmental quotient range (median)	n = 19	n = 15	
Total	18–96 (81)	47–115 (87)	0.181
Body movement	28–100 (74)	35–106 (73)	0.704
Hand movement	15–110 (86.5)	46–123 (91)	0.205
Fundamental habit	18–92 (84)	56–117 (93)	0.010
Human relationships	16–92 (73.5)	62–118 (85)	0.008
Speech	15–137 (83)	27–123 (88)	0.627
Language understanding	18–137 (101)	56–123 (89)	0.274

Data represent the number (%) or range (median).
ASD; autism spectrum disorder.

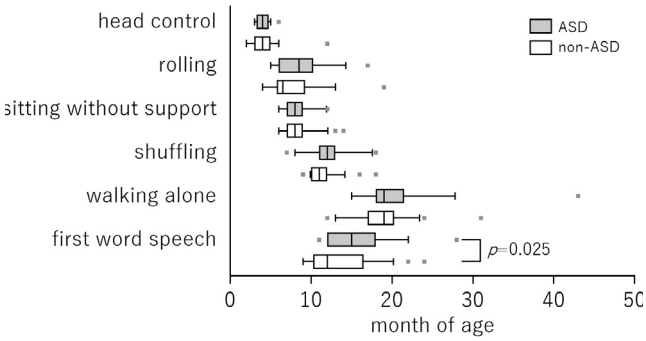

Fig. 1. Box-and-whisker plots of gross motor development and first word speech. Gray boxes show the autism spectrum disorder (ASD) group, and white boxes show the non-ASD group. No differences were seen in gross motor development. The mean month of first word speech was significantly delayed in the ASD group.

show shuffling. Approximately 11% of children with Down syndrome show shuffling [10]. Hypotonia and unstable postural maintenance may trigger bottom shuffling in children with Down syndrome. Except for these neurological or congenital disorders, infants with bottom shuffling are generally considered to have good long-term developmental outcomes [11]. In contrast, our study revealed many children with ASD in shuffling babies. With widening recognition of ASD, the prevalence of ASD has risen in the last decade [5]. Most reports of shuffling babies are from the 1970′s to 1980′s, and diagnosing ASD in the 70′s to 80′s may have been difficult [1,3]. In the report by Bottos et al. in 1989, the rate of speech deficit in the control group with bottom shuffling was higher than that of children with other locomotor strategies [3]. The speech deficit in the report may suggest underlying ASD in infants with shuffling. Our results indicate that many children with ASD are found in bottom shufflers, even in non-syndromic infants with bottom shuffling, and that follow-up for social and behavioral development is important in shuffling babies.

Early diagnosis and appropriate intervention are important for the management of children with ASD [6]. In the present study, we revealed a higher rate of poor eye contact, a delay in the first speech, and a less frequent family history of bottom shuffling in the ASD group. These characteristics can be clues to diagnose ASD in the early stage. On the other hand, the rate of hypotonia at the first visit and motor developmental milestones were not different between infants with and without ASD. Although pediatricians tend to focus on motor development and muscle tone in the examination of shuffling babies, they should also pay attention to social and language skills.

The reasons for bottom shuffling in infants are still unknown and may be multifactorial. A high incidence of a family history in the non-ASD group suggests a genetic predisposition, although causal genetic mutations or variants have not been reported. Family-related factors such as overprotective or ambivalent parental attitudes are related to locomotor strategies including bottom shuffling [3]. Hypotonia has been observed in shuffling babies and is thought to be one causative factor of bottom shuffling [1,12]. Our study also showed a high rate of hypotonia in shuffling babies. Several studies have reported low muscle tone in infants with ASD [13]. From these observations, low muscle tone may be a cause of bottom shuffling in infants with ASD. Second, children with ASD, attention-deficit/hyperactivity disorder, or intellectual disability often have characteristic trajectories of motor development [14–17]. Reported characteristics of motor development in ASD include clumsy, dyskinetic, dystonic, parkinsonian, ataxic, or asymmetric postures or motions; abnormal general movement; and persistent primitive reflex [16]. Bottom shuffling may be a characteristic trajectory of the motor development in ASD. A third possible mechanism of bottom shuffling in ASD may be hypersensitivity of sensations. Children with ASD often have sensory abnormalities, such as tactile hypersensitivity or hyposensitivity [7]. Crawling causes sensory stimulation of the hands and knees, and infants with ASD may use this characteristic strategy of locomotion, bottom shuffling, to avoid the sensory stimulation.

Our current study has several limitations. First, this study was conducted in a single developmental care center. Although this center is the only facility for children with developmental problems in the two cities, a high prevalence of ASD may be biased by the population of children visiting the center. In many infants with bottom shuffling that are identified at regular checkups in public health centers, caregivers may not need to worry too much about ASD. Although the prevalence rate of ASD may be abnormally high, the present study provides important information suggesting that children with ASD are included among shuffling babies. Second, this study was performed retrospectively, and the timing of evaluation of DQ and developmental outcomes was not standardized. Prospective cohort studies are needed to confirm the accurate prevalence of ASD and outcomes of shuffling babies.

In conclusion, some bottom shufflers may represent ASD during follow-up. Although pediatricians tend to focus on motor skills in shuffling babies, paying more attention to social and cognitive functions such as eye contact and language development is also important. A large-scale population-based prospective study is needed to reveal the neurodevelopmental outcome of bottom shufflers.

Contributors' statements

Yu Okai drafted the initial manuscript and approved the final version of the manuscript for submission.

Yu Okai and Kiyokuni Miura reviewed clinical charts of the enrolled children and conducted the statistical analysis.

Tomohiko Nakata, Kiyokuni Miura, Atsuko Ohno, Rie Wakako, Osamu Takahashi, Yuki Maki, Masaharu Tanaka, Yoko Sakaguchi, Yuji Ito, Hiroyuki Yamamoto, Hiroyuki Kidokoro, Yoshiyuki Takahashi, and Jun Natsume critically reviewed and revised the manuscript.

All authors approved the final version of the manuscript as submitted and agree to be accountable for all aspects of the work.

Conflict of interest and disclosure

Jun Natsume is affiliated with the endowed department from Aichi prefectural government (Department of Developmental Disability Medicine). No other COIs to disclose.

Acknowledgements

We thank Dr. Toru Kurokawa from Seiai Rehabilitation Hospital, Dr. Yasunari Sakai from the Department of Pediatrics at Kyushu University, and Dr. Ryutaro Kira from the Department of Pediatric Neurology at Fukuoka Children's Hospital for providing the English version of the revised Enjoji Developmental Test. We wish to thank all participants and families of the study.

References

[1] Robson P. Shuffling, hitching, scooting or sliding: some observations in 30 otherwise normal children. Dev Med Child Neurol 1970;12:608–17.
[2] Narazaki O, Narazaki A. An epidemiological study on shuffling babies at a health check-up at 18 months of age (In Japanese). No To Hattatsu (Tokyo) 1986;18:484–9.
[3] Bottos M, Dalla Barba B, Stefani D, Pettenà G, Tonin C, D'Este A. Locomotor strategies preceding independent walking: prospective study of neurological and language development in 424 cases. Dev Med Child Neurol 1989;31:25–34.
[4] Størvold GV, Aarethun K, Bratberg GH. Age for onset of walking and prewalking strategies. Early Hum Dev 2013;89:655–9.
[5] Christensen DL, Maenner MJ, Bilder D, Constantino JN, Daniels J, Durkin MS, et al. Prevalence and characteristics of autism spectrum disorder among children aged 4 years - early autism and developmental disabilities monitoring network, seven sites, United States, 2010, 2012, and 2014. MMWR Surveill Summ 2019;68:1–19.
[6] Reichow B, Hume K, Barton EE, Boyd BA. Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). Cochrane Database Syst Rev 2018;5:CD009260.
[7] American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed: Washington, DC; 2013.
[8] American Psychiatric Association. DSM-IV: Diagnostic and statistical manual of mental disorders. Washington, DC: American Psychiatric Association; 1994.
[9] Enjoji M, Yanai N. Analytic test for development in infancy and childhood. Paediatria Japonica 1961;4:2–6.
[10] Saito K, Watanabe Y. The early rehabilitation in children with Down. Paying special attention to shuffling and comorbidities of developmental disorder (In Japanese). No To Hattatsu (Tokyo) 2016;48:122–6.
[11] Bellman M, Byrne O, Sege R. Developmental assessment of children. BMJ 2013;346 e8687.
[12] Robson P. Prewalking locomotor movements and their use in predicting standing and walking. Child Care Health Dev 1984;10:317–30.
[13] Serdarevic F, Ghassabian A, van Batenburg-Eddes T, White T, Blanken LME, Jaddoe VWV, et al. Infant muscle tone and childhood autistic traits: a longitudinal study in the general population. Autism Res 2017;10:757–68.
[14] Pan CY, Tsai CL, Chu CH. Fundamental movement skills in children diagnosed with autism spectrum disorders and attention deficit hyperactivity disorder. J Autism Dev Disord 2009;39:1694–705.
[15] Sacrey LR, Zwaigenbaum L, Bryson S, Brian J, Smith IM, Raza S, et al. Developmental trajectories of adaptive behavior in autism spectrum disorder: a high-risk sibling cohort. J Child Psychol Psychiatry 2019;60:697–706.
[16] Setoh P, Marschik PB, Einspieler C, Esposito G. Autism spectrum disorder and early motor abnormalities: Connected or coincidental companions?. Res Dev Disabil 2017;60:13–5.
[17] Liao HF, Lee SC, Lien IN, Chen CJ, Soong WT, Tseng CC. Locomotor strategies before independent walking: prospective study of 50 mentally retarded children. J Formos Med Assoc 1992;91:334–41.