

*Original Article***Relationship between range of motion of lower extremity and gross motor function in children with cerebral palsy who have walking ability****Satomi Kawarada, RPT, MS,¹ Izumi Kondo, MD, PhD,² Shigeru Sonoda, MD, PhD,³ Eri Yokoyama, RPT, MA,¹ Yuko Tazawa, RPT,¹ Yoshihiko Yabunaka, RPT, MS⁴**¹Aomori Prefectural Asunaro Rehabilitation Center for Children, Aomori, Japan²Medical Examination Section of Functional Recovery, National Center for Geriatrics and Gerontology Hospital, Obu, Aichi, Japan³Fujita Health University Nanakuri Sanatorium, Tsu, Mie, Japan⁴Department of Rehabilitation Science, Osaka Health Science University, Osaka, Japan**ABSTRACT**

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Purpose: The purpose of this study was to investigate the relationship between range of motion of lower extremity and gross motor function in children with cerebral palsy (CP) who have walking ability.

Methods: The subjects were 30 children with CP who were able to walk and were classified as level I – III according to the Gross Motor Function Classification System for children with CP (GMFCS). We measured range of motion (ROM) of lower extremity as follows: 1) hip joint extension in Thomas posture; 2) knee joint extension; and 3) ankle joint dorsiflexion under knee joint extension. At the same time, we evaluated gross motor function at the dimension of standing and the dimension of walking, running and jumping by gross motor function measure (GMFM).

Results: There was a significant difference in ROM of hip joint extension between GMFCS level I and III and also between GMFCS level II and III, and hip joint ROM was closely related to gross motor function. ROM of knee joint extension was more limited in GMFCS level I – II children than in GMFCS level III.

ROM of ankle joint extension did not differ significantly among the 3 GMFCS levels, and the correlation between ankle joint ROM and gross motor function was weak.

Conclusion: The results suggested that lower extremity ROM is one of the factors related to gross motor function in children with CP.

Key words: children with cerebral palsy, range of motion of lower extremity, gross motor function

Introduction

Decreased range of motion (ROM) is one of the most common impairments in children with cerebral palsy (CP). Bell et al. reported that ROM decreased with age and ROM of hip abduction, knee extension, and ankle dorsiflexion decreased significantly around 8 to 12 years of age [1]. It was proposed the reduction in ROM may be a function of the inability of length changes in spastic muscle to keep up with the changes in bone length, and this was supported by in a spastic mouse model [2, 3]. Spasticity typically develops between 6 and 18 months of age in children with CP and it alters the previously normal skeletal anatomy [4]. Kilgour et al. [5] reported that even mild cases, children classified by the Gross Motor Function Classification System (GMFCS) [6] as level I and II, had some restriction in passive ROM of knee extension and ankle dorsiflexion at school age compared to age-matched normal subjects.

On the other hand, in a study in which patients were stratified according to GMFCS, Hanna et al. [7] reported that the loss of gross motor function occurred around 7 years of age for children with moderate and severe CP, classified as GMFCS level III – V. In addition, in another study in which the patients were stratified according to GMFCS, Bartlett et al. [8]

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suggested the possibility that the loss of gross motor function was caused by pain and restriction of ROM for children classified as GMFCS level III – V. Day et al. [9] indicated that mild cases would also have a certain rate of gross motor function loss after adolescence.

We thought that reduced ROM would have some effect on gross motor function even in ambulatory children. The purpose of this study was to determine the relationship between lower extremity ROM and gross motor function in children with CP, and obtain fundamental results for a future longitudinal study. We assumed that the ROM of each joint would affect the gross motor function of each child with CP differently. We performed further analyses in this study to examine this assumption.

Methods

This research was performed at a medical and educational center located in the Aomori prefecture of Japan. Subjects who participated in this study were recruited from this center and were ambulatory children diagnosed with CP. Exclusion criteria included orthopedic surgery within the last 10 years, treatment with botulinum toxin within the past year, and typical dyskinesia. The subjects consisted of 30 children (18 boys and 12 girls) with a mean (SD) age of 9.2 (4.31) (range, 4–8) years, and their GMFCS levels were I (11), II (9), and III (10). Of the 30 children, 4 had diagnosis of hemiplegia and 26 had diplegia. The subjects had no intellectual deficit or mild mental retardation. All of them were able to obey oral instructions. The background information of the subjects is shown in Table 1. The number of subjects who used orthosis was lowest in the GMFCS level I group and AFO was the most popular orthosis used by those in the level III group.

The passive ROM of the lower extremity was evaluated using a goniometer (R-377; Tiger Medical Instruments, Japan). We measured the ROM of 3 joint movements to determine the restriction of lower extremity ROM: hip joint extension (HE), knee joint

extension (KE), and ankle joint dorsiflexion (ADF). We examined the ROM of these joint movements because we often observe that these are reduced in children with CP in the clinical setting. HE angle while in the Thomas posture, KE angle, and ADF angle with knee extension were measured in a supine position. Two physiotherapists evaluated the ROM of each subject; 1 physiotherapist who treated the subjects moved the joint for the measurement and the second operated the goniometer and recorded the result. We thought that we could measure the maximum ROM when the movement was being provided by the physiotherapist in charge because the child would be relaxed and in an atmosphere similar to that in his/her usual exercise time.

Gross motor function was evaluated using the dimension for standing (dimension D) and the dimension for walking, running, and jumping (dimension E) of Gross Motor Function Measure (GMFM) [10]. The physiotherapist who was in charge of the subject performed the evaluation with GMFM. We decided to measure the ROM and evaluate GMFM around the same time period, and both were performed within 2 weeks. The Medical Ethics Committee of Fujita Health University Nanakuri Sanatorium approved the design of this study.

Spearman's rank correlation coefficient was calculated to define the relationship between GMFM and ROM scores of each joint of the lower extremity. For the comparison of ROM for each GMFCS level, we used the Kruskal-Wallis test and post hoc analysis. A p value of <0.05 was considered statistically significant. All analyses were conducted using PASW Statistics version 17.0 software (SPSS Inc., Chicago, IL, USA).

Results

The results of the ROM measurements and mean scores of GMFM dimension D and E are shown in Table 2. There were no significant age differences among the 3 GMFCS levels ($F(2,27) = 0.541$, $p = 0.558$). The ROM measurements of the lower extremity

Table 1. Subjects' background information

Characteristic	All subjects	GMFCS I	GMFCS II	GMFCS III
N	30	11	9	10
Mean age (SD) (y)	9.27 (4.31)	8.45 (4.46)	9.0 (4.47)	10.4 (4.20)
Age range	4–18	4–17	4–17	6–18
Type of CP	Spastic			
Spasticity distribution	Diplegia (26)	Diplegia (8)	Diplegia (8)	Diplegia (10)
Type (N)	Hemiplegia (4)	Hemiplegia (3)	Hemiplegia (1)	Hemiplegia (0)
Use of Orthosis	AFO (14)	AFO (1)	AFO (4)	AFO (9)
Type (N)	FO (7)	FO (3)	FO (3)	FO (1)
	NO (9)	NO (7)	NO (2)	NO (0)

GMFCS, Gross Motor Function Classification System; CP, cerebral palsy; AFO, Ankle Foot Orthosis; FO, Foot Orthosis

by GMFCS level are shown in Figure 1. There was the significant difference in HE ROM between GMFCS level I and level III and also between GMFCS level II and level III. For GMGCS level III children, there was also a trend that an HE ROM of less than -10 degrees was often observed. For KE ROM, there was no significant difference between GMFCS level I and level II children; however, KE ROM of level III children differed greatly compared to GMFCS level I and level II children. For ADF ROM, although there was no significant differences among the 3 groups, in 9 of the 11 GMFCS level I children, ADF was greater than 0 degrees, while only 1 child in the GMFCS level III group had ADF ROM greater than 0 degrees.

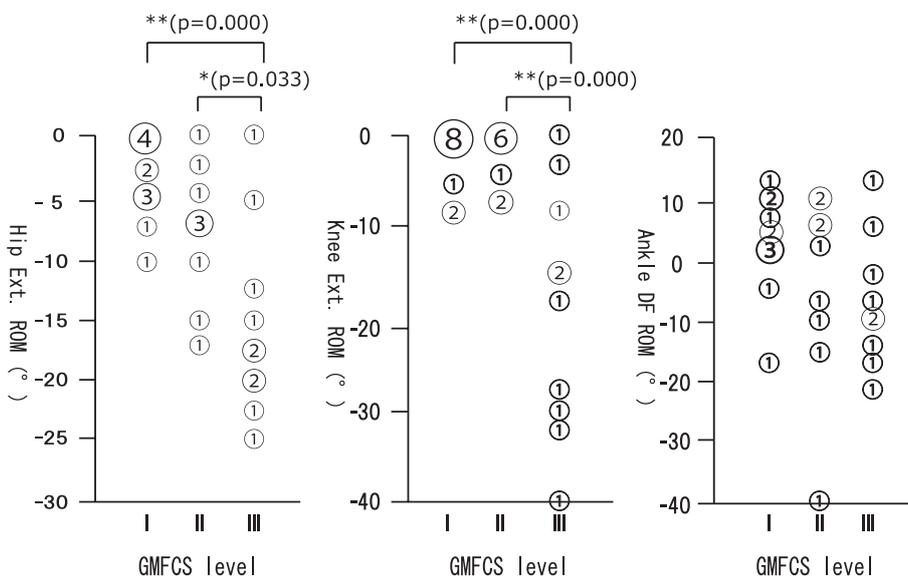
Regarding the relationships between ROM and gross motor function, there were significant correlations between HE ROM and GMFM dimension D ($r_s = 0.680, p = 0.000$) and dimension E ($r_s = 0.710, p = 0.000$) (Fig. 2). There were also significant correlations between KE ROM and GMFM dimension D ($r_s = 0.559, p = 0.001$) and dimension E ($r_s = 0.530, p = 0.003$)

(Fig. 3). However, there were also significant, although somewhat lower, correlations between ADF ROM and GMFM dimension D ($r_s = 0.393, p = 0.032$) and dimension E ($r_s = 0.414, p = 0.023$) (Fig. 4). In the group with ADF ROM greater than 0 degrees, the more the ROM increased, the more the GMFM score increased, while in the group with a ADF ROM less than 0 degrees, the more the ROM decreased, the more the GMFM score decreased. However, there was no significant correlation between ADF ROM and GMFM dimension D ($r_s = 0.218, p = 0.417$) or dimension E ($r_s = 0.170, p = 0.381$) in the group with an ADF ROM greater than 0 degrees, and there also was no significant correlation between ADF ROM and GMFM dimension D ($r_s = -0.364, p = 0.201$) and dimension E ($r_s = -0.231, p = 0.362$) in the group with an ADF ROM of less than 0 degrees (Fig. 5).

Regarding the relationship between age and the ROM of each joint, there were significant correlations with the ROM of HE ($r_s = -0.367, p = 0.023$), KE ($r_s = -0.530, p = 0.063$), and ADF ($r_s = -0.439, p = 0.015$), (Fig. 6).

Table 2. ROM measurements and scores of GMFM dimension D and E

	Group			P value (by GMFCS level)	F-value
	GMFCS I	GMFCS II	GMFCS III		
Hip Ext. ROM (°)	-3.41 (3.40)	-8.06 (5.56)	-15.5 (7.80)	0.000	11.468
Knee Ext. ROM (°)	-1.59 (3.02)	-1.39 (2.20)	-18.75 (13.40)	0.000	15.345
Ankle D/F (KE) ROM (°)	3.18 (8.37)	-4.44 (16.05)	-7.00 (10.53)	0.139	2.128
GMFM D (%)	94.6 (3.88)	79.0 (13.61)	45.0 (20.26)	0.000	33.705
GMFM E (%)	89.7 (8.36)	69.9 (14.09)	28.6 (14.74)	0.000	64.019



**Significant at $p < 0.01$, *significant at $p < 0.05$.

Figure 1. Range of motion (ROM) of lower extremity according to Gross Motor Function Classification System (GMFCS) level.

The number in each circle is the number of subjects. There was a significant difference in hip extension ROM between GMFCS level I and level III and also between GMFCS level II and level III.

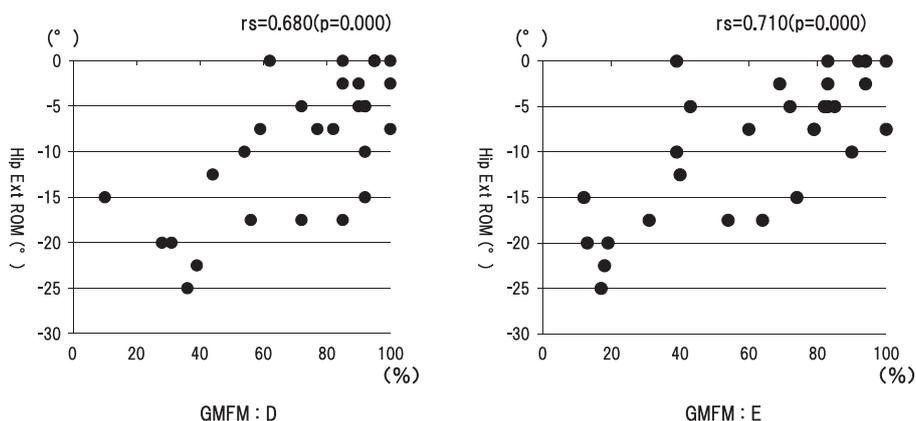


Figure 2. Relationships between hip extension ROM and Gross Motor Function Measure (GMFM) dimension D and E. There were significant correlations between hip extension ROM and GMFM dimension D and E.

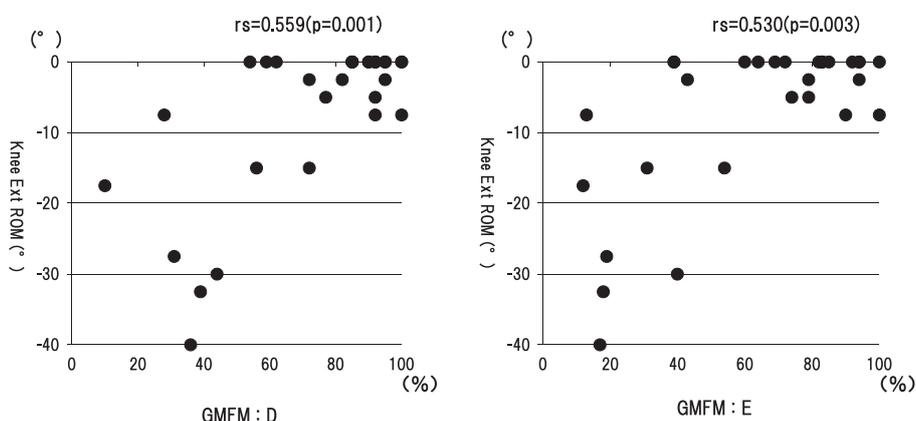


Figure 3. Relationships between knee extension ROM and GMFM dimension D and E. There were significant correlations between knee extension ROM and both GMFM dimension D and E.

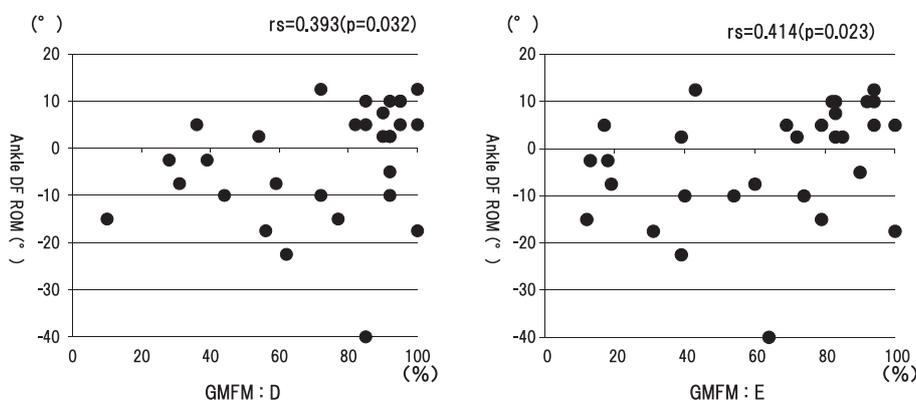


Figure 4. Relationships between ankle dorsiflexion ROM and GMFM dimension D and E. There were somewhat lower correlations between ankle dorsiflexion ROM and GMFM dimension D and E, compared to the correlations with hip extension and knee extension ROM.

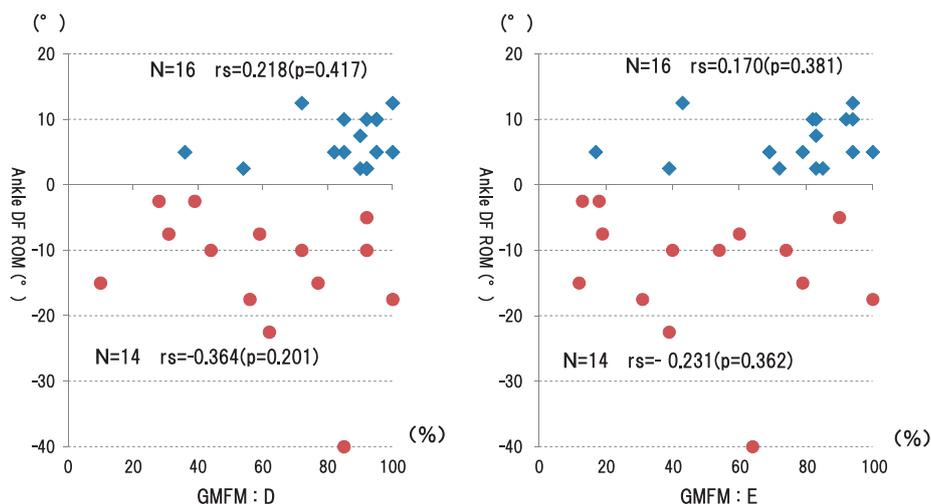


Figure 5. Relationships between ankle dorsiflexion (ADF) ROM and GMFM dimension D and E in the groups whose ADF was greater than 0 degrees and less than 0 degrees. For the ADF ROM in the group whose ADF was greater than 0 degrees, the more the ROM increased, the more the GMFM score increased, while in the group whose ADF was less than 0 degrees, the more the ROM decreased, the more the GMFM score decreased. However, there was no significant correlation between ADF ROM and GMFM dimension D and E in either group.

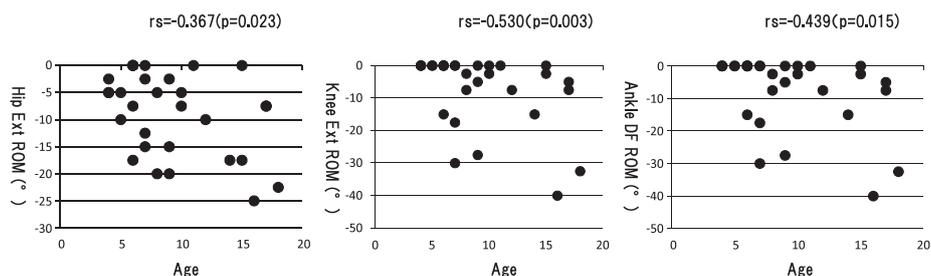


Figure 6. Relationship between age and the ROM of each joint. There were significant correlations between age and the ROM of hip extension, knee extension, and ankle dorsiflexion.

Discussion

Although ROM limitation is a common impairment observed in children with CP, it is not known when it occurs, during what period it progresses the most, and whether there are differences in the location and degree of progression with reference to the severity and the level of spasticity. However, it has been suggested that the limitation of ROM in children with CP worsens with age [1], and the results of the current study indicate the same trend.

A study on the deterioration of walking ability in children with CP reported that, even in children who could walk, more than 40% experienced a reduction in gait ability after adulthood [11, 12]. In an investigation of adult CP patients who were older than 20 years of age performed by Minato et al. [13], it was suggested that there were close relationships among deformity, contracture, and severity as evaluated by GMFCS. They also reported that patients whose gross motor

function was reduced compared with their best period were often observed in the group of patients whose deformity and contracture had progressed to a certain level.

In the results of this study of children with CP who could walk, there was a trend of higher gross motor function when the limitation in HE and KE ROM was less. In general, it suggested that the course of gross motor function in children with CP is affected by numerous impairments such as muscle strength, selective motor control, and spasticity of the hamstrings [14]. Although, we do not disagree with the idea that limitation of ROM is one of the factors affecting the development course of gross motor function, a longitudinal study would be necessary to corroborate this.

Regarding lower extremity ROM according to GMFCS level in this study, HE and KE ROM decreased significantly in GMFCS level III children. This was partly because the study subjects, who were school

age children at GMFCS level III, used a wheelchair for long distance or rapid locomotion more often than children at GMFCS level I or II; therefore, they probably did not have much experience of leg extension in daily life and had lower leg muscle weakness [15]. In addition, selective motor control of the lower extremity might also affect the reduction in ROM. It was suggested that selective motor control ability decreased with the severity of gross motor function, and for GMFCS level III children in particular, there was a large range in selective motor control ability, and there were some children whose ability was much more reduced than GMFCS level I and II children [14].

There was no significant difference in ADF ROM among the 3 GMFCS levels, and it also had only a weak relationship with gross motor function. However, when separating the groups according to whether or not ADF ROM was greater than 0 degrees, there were opposite relationships between ROM and gross motor function. It was affected by the typical posture observed during the gait of children with CP. There are several postures observed during gait, such as, crouch, equinus foot with knee hyperextension, and equinus foot with flexion deformity of knee. The ankle joint position differed according to these positions. In children who kept standing or walked in the equinus position, limitation of ankle dorsiflexion was likely to be worse as the child's activity level increased/decreased. In contrast, in children who crouched more, hip and knee joint flexion tendency increased with increased height and weight, and other unfavorable factors, and as a result, the ankle joint might become more dorsiflexed. For these reasons, we attempted to examine the relationship by separating the groups according to whether or not ADF was greater than 0 degrees. Abel et al. [16] reported that children with higher motor function had less limitation of hip and knee joint ROM; however, ankle joint ROM tended to be larger in patients with lower function.

Therefore, for the ROM exercise of the ankle joint, although the ROM exercise would be effective when the limitation of ankle dorsiflexion induced by the equinus posture reduced gross motor function, whose ankle was dorsiflexed with flexion of hip and knee joint as the result of crouching posture during standing and walking, it would be better not to excessively enlarge the ankle joint ROM.

Stretching, brace therapy, and orthopedic surgery were used in the treatment for limitation of lower extremity ROM in children with CP. While prolonged muscle stretching is effective in reducing the spasticity and enlarging the ROM, evidence for improvement in ROM with manual stretching is limited [17]. Although brace therapy was reported to be effective in a prospective study, its effectiveness for the prevention of muscle shortness and deformity has not been determined [18, 19]. Muscle release surgery for the

lower extremity improves ROM [20, 21]; however, long-term results have not yet been reported [22]. In summary, regarding treatment for halting the progression of ROM limitation, there are no conclusive guidelines.

Recently, it was suggested that in children with gait ability, muscle strength of the lower extremity is related to gross motor function [15]. Muscle strength exercise performed with specific methods was reported to be useful, and its effectiveness was confirmed by RCT [23]. Gage [24] suggested that muscle shortness or deformity of the muscle skeletal system changes the length of the joint lever arm and thus reduced joint torque. Therefore, since reduction of joint ROM reduces muscle strength, it is necessary to maintain the ROM; however, currently there are no findings about the influence of limitation of ROM on muscle strength exercise.

The limiting factors for interpreting the results of this study were the insufficiency of statistical power due to the paucity of subjects ($n = 30$) and the inability to detect the direct influence of ROM on gross motor function since it was not a longitudinal study. In addition, although this study included 4 children with hemiplegia, we used the average ROM of both legs as the measurement and did not discriminate these children from those with diplegia. While we used the average ROM of both legs as the measurement, GMFM, which was used in this study as the index of gross motor function, included items where the ability of each leg is considered separately, such as, "standing while holding on to a large bench with 1 hand, lift the right or left foot for 3 seconds," and for children with hemiplegia, 1 of these items was often possible. Therefore, in the future, when we perform the analysis with a larger sample size, we should examine the data separating children with hemiplegia from children with diplegia. Moreover, the physiotherapist in charge participated in the evaluation of GMFM and ROM, and this could affect the child's performance at assessment. In fact, the participation of physiotherapist in charge was used because we hoped that the children would give their best performance.

In conclusion, although this was a cross sectional study, the relationship between HE and KE ROM and gross motor function was demonstrated. In future studies, to establish effective treatment for the prevention of ROM limitation, it will be necessary to investigate the long-term effect of ROM limitation within a longitudinal study, and to study whether the effect of muscle strength exercise is influenced by the presence of ROM limitation.

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