

KAATSU training[®] in a case of patients with periventricular leukomalacia (PVL)

H. Iwashita, T. Morita, Y. Sato, T. Nakajima

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[Objective] The effectiveness of KAATSU training has been reported in wide-ranging fields from sports medicine to rehabilitation, and KAATSU training has been clinically applied. However, there have been only limited reports on pediatric cases. We performed KAATSU training in a pediatric patient with periventricular leukomalacia (PVL) and examined its effectiveness and safety for cerebral palsy. We report here the results of this examination.

[Methods] KAATSU training was performed on a PVL patient on an outpatient basis. This training was performed once a week for 14 weeks and involved mainly three specified motions of the upper and lower limbs. Evaluation was performed using the Gross Motor Function Measure (Gross Motor Function Classification System: GMFM) and videos. In this report, the effectiveness of short-term KAATSU training was compared with that of intensive physical therapy using data in the literature.

[Results] QOL improved due to short-term reduction of muscle tonus and increased acquired movements resulting from KAATSU training.

[Conclusion] The results of this report suggest that KAATSU training can be effective in PVL patients. Further examination is necessary with increased cases and evaluation methods.

Key words: Periventricular leukomalacia, KAATSU training, spastic paralysis, cerebral palsy

Correspondence to:
H. Iwashita,
Komone Acupuncture Clinic,
3-10-18 Mukaihara,
Itabashi-ku, Tokyo, 173-0036,
Japan
iwashita@komone.jp

See end of article for
authors' affiliations

INTRODUCTION

PVL is a disorder reported by Banker et al. in 1962¹⁾. It occurs most commonly in premature infants, mainly those born at less than 32 weeks of gestation. It is a lesion with periventricular necrosis as its major component. Since the corticospinal tracts pass through the periventricular white matter, spastic paraplegia develops that is particularly severe in the lower limbs. The severity of the disability or impairment can vary, including severe impairment such as dragging of feet when walking, inability to sit, inability to walk, and inability to swallow. Intellectual impairment is also seen in many cases. There is currently no effective treatment. We report here a case of PVL in which KAATSU training was effective for spastic quadriplegia. In addition, we compared the effectiveness of short-term KAATSU training with that of intensive physical therapy using data in the literature.

METHODS

1. Ethical consideration

This study was approved by the Research Ethics Committee of the Graduate School of Medicine and Faculty of Medicine at the University of Tokyo (application number: 3088). Informed consent was obtained from the guardian of the patient before participation in this study.

2. Case report

Patient: A boy aged 7 years and 10 months

Diagnosed disease: PVL

Chief complaint: Spastic quadriplegia

Family history: Non-contributory

History of present illness: The patient was born by cesarean section at 28 weeks and 1 day of gestation and had a birth weight of 996g.

The patient and his twin sibling were conceived naturally. His twin sibling was born first and had died at birth.

One- and five-minute Apgar scores were 6 and 7, respectively.

The patient was an extremely low-birth weight infant with apneic events and RDS and was hospitalized in the NICU until 91 weeks after birth.

Age 1 year and 7 months: initiation of physical therapy

Age 2 years and 9 months to 3 years and 10 months: initiation of botulinum toxin therapy (total: 4 times)

Age 4 years and 10 months: bilateral muscle release in the hip

The patient continued rehabilitation therapy but began KAATSU training because muscle tonus increased with the growth of the body and acquisition of movements slowed.

3. Methods

a) Motion conditions

KAATSU belts were placed around the upper arms

in the axillary area and around the upper legs in the thigh area. KAATSU MASTER was used for the training. The upper limb training began with a cuff tightness pressure of 40-45 mmHg and a setting pressure of 80-90 mmHg, and the lower limb training with 55-65 mmHg and 110-120 mmHg, respectively. Since the subject was unable to speak, appropriate pressure was estimated using the coloration of the palms and feet, pulse rate, and facial expression. Pressure was applied and released and this process was repeated three times. Passive ROM training was performed, and then KAATSU training was begun. The training involved the following items: three specified motions of the upper and lower limbs, KAATSU walk, and muscle strength training using light weights. One to two sets of the first item were performed once a week for 14 weeks. Generally, this item was performed 10-20 times for the first set and in an all-out manner for the second set. Careful observations were made of changes in the patient's physical conditions during the training. Care was taken to control the vagal reflex by keeping the patient's head below the level of his heart when the pressure was released.

b) Evaluation

1) GMFM

The GMFM evaluates the motor ability of children with cerebral palsy and involves movements that can be performed by children with normal motor ability by age 5 years. These movements are classified into five dimensions of A: lying and rolling, B: sitting, C: crawling and kneeling, D: standing, and E: walking and running. The GMFM contains 88 items each of which is scored on a 0-3 rating scale and can assess in details the motor ability of young children^{2,3}. The GMFM is effective in the evaluation of short-term changes⁴. It was used to evaluate and compare our patient's motor ability before KAATSU training and

after 14 weeks of training.

2) Video

In cerebral palsy associated with PVL, there is a combination of various factors, including delayed motor development, abnormal postural tone such as hypertonia, and sensory, perception, and cognitive immaturity⁵. Therefore, its mechanism of pathogenesis is difficult to evaluate. Thus, in this report, comprehensive evaluation and examination were performed using evaluation of videos.

RESULTS

1. GMFM results

Table 1 shows the GMFM results of the patient. The GMFM was also evaluated before and after botulinum toxin therapy and before and after bilateral muscle release in the hip. These results are also included in the table. The GMFM dimensions of D: standing and E: walking and running were omitted because the patient was unable to perform these motor functions. When the GMFM results were compared before botulinum toxin therapy and at the third therapy session, the motor functions were markedly improved as shown in Table 1. However, when comparison was made for each injection, there was less improvement. There was almost no improvement at the third therapy session. For bilateral muscle release in the hip, Table 1 shows improvements but the scores were decreased before KAATSU therapy began. That is, bilateral muscle release in the hip has limited effectiveness, and it also places a burden on young children.

For KAATSU training, there was one post-training evaluation at three months after training. Thus, long-term evaluation is necessary. In the short-term evaluation, the scores increased to 51/51 for A: lying and rolling and 36/60 for B: sitting. Therefore, KAATSU training showed effectiveness that was comparable to or better than other treatment methods.

Table 1. GMFM evaluation of KAATSU training, botulinum toxin therapy, and bilateral muscle release in the hip in our patient

	KAATSU training		Botulinum toxin therapy						Bilateral muscle release in the hip	
	At the start	After 3 mo	First		Second		Third		Before surgery	After 1 mo
			Before injection	After 3 mo	Before injection	After 3 mo	Before injection	After 3 mo		
A: Lying and rolling	41/51 (80.0%)	51/51 (100%)	43 (84.3)	43 (84.3)	45 (88.2)	50 (98.0)	50 (98.0)	50 (98.0)	45 (88.2)	51 (100)
B: Sitting	28/60 (46.6)	36/60 (60.0)	11 (18.3)	20 (33.3)	20 (33.3)	26 (43.3)	26 (43.3)	29 (48.3)	20 (33.3)	29 (48.3)
C: Crawling and kneeling	5/42 (11.9)	8/42 (19.0)	0	3 (7.1)	3 (7.1)	7 (16.6)	7 (16.6)	6 (14.2)	3 (7.1)	9 (21.4)

VIDEO EVALUATION

Figure 1 shows images of the patient immediately after beginning KAATSU training. There was severe muscle tonus, and bands were placed at his waist and ankles for immobilization when sitting in the wheelchair (Fig. 1a). Adductor muscle tonus was particularly severe, and three staff members were required to place KAATSU belts on the lower limbs at the first training session. The patient also had difficulty moving the upper limbs only and the lower limbs

only. When the patient performed arm curls, his trunk would bend backward (Fig. 1b). Similarly, when the patient kicked a ball, his trunk would bend backward and he needed to be supported from the back (Fig. 1c). In KAATSU walk, his feet would move forward but his knees would not fully extend, showing equinus deformity (Fig. 1d).

Figure 2 shows the conditions on the 14th week of KAATSU training. The patient achieved independent sitting and reduced adductor muscle tonus (Fig. 2a).



Figure 1. Photographs taken immediately after KAATSU training began (explanations for Figures 1a, 1b, 1c, and 1d are in the main text)

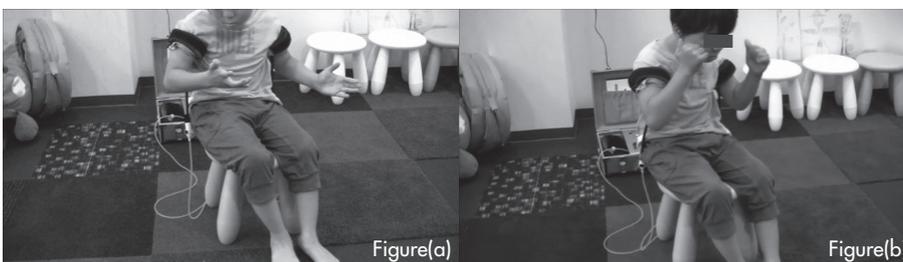


Figure 2. Conditions during KAATSU training on the 14th week (explanations for Figures 2a and 2b are in the main text)

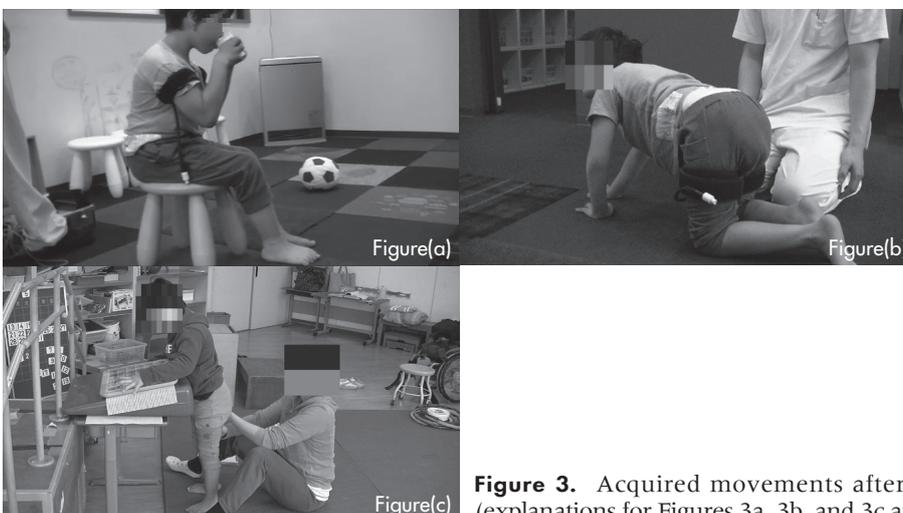


Figure 3. Acquired movements after KAATSU training (on the 14th week) (explanations for Figures 3a, 3b, and 3c are in the main text)

In addition, the patient was able to move the upper limbs only and the lower limbs only. Beginning around the tenth week, he was able to sit on a chair on his own to perform his upper limb training (Fig. 2b).

Figure 3 shows the acquired movements after 14 weeks of KAATSU training. Since paper cups are soft and easily crushed, handling such cups is a difficult act for young children with severe spasticity. Our patient was able to use paper cups after KAATSU training (Fig. 3a). He was previously unable to crawl because his trunk area was weak and he could not maintain the posture. However, he was able to successfully crawl after KAATSU training (Fig. 3b). The changes in the lower limbs were not sufficiently large to be observable on images. However, the patient was able to extend his knees to put his heels on the floor while staff supported him in standing training (Fig. 3c). When comparison was made between the video immediately after training began and that after 14 weeks of training, the results suggest that KAATSU training can help reduce muscle tonus, improve muscle control, and increase muscle strength.

DISCUSSION

When the effectiveness of training in young children is examined, it is most important to determine if the positive results are due to children's development or the effects of training. There are several reports on development that is characteristic of young children with cerebral palsy. In the report by Rosenbaum et al., the motor development curve was until 15 years of age. The children reached 90% of their motor function based on the GMFM by approximately age 5 years, and the motor development curve plateaued by approximately 7 years⁶. In addition, the subjects were determined to have level IV severity of cerebral palsy based on a five-level severity scale of the Gross Motor Function Classification System (GMFCS)⁷. Hanna et al. reported that prognosis differed depending on the GMFCS level and that the muscle strength in level IV children decreased starting around 7 years of age⁸. It

is also speculated from the results of these reports that the subjects were in a state where acquisition of movements was difficult due to his development. In such a state, acquired movements gained in the short term of 14 weeks can be attributed to the effectiveness of KAATSU training.

The effectiveness of short-term KAATSU training was compared with that of intensive physical therapy. Intensive physical therapy increases the GMFM scores compared with conventional physical therapy and is recommended (grade B) in the second edition of the Guidelines for Rehabilitation of Children with Cerebral Palsy⁹. Comparison was made between our patient and other patients with GMFCS level IV (same as our patient) who were reported in the literature^{10,11,12} (as shown in Table 2). In the study of Asakai et al.¹³, the mean GMFM percent score of patients at GMFCS level IV was 28.8% at the first examination, and there was an improvement of 2.1 percentage points after a mean of 1.9 months of intensive physical therapy. In our patient, the mean GMFM percent score was 27.7% at the initial examination, and there was an improvement of 8.1 percentage points after 14 weeks of KAATSU training. Asakai et al. used a short training period with a mean of 1.9 months. The frequency was 5 sessions per week (40 minutes per session) for physical therapy, 5 sessions per week (20 minutes per session) for in-ward training (standing and walking training performed by ward personnel), and a mean of 3 sessions per week for occupational therapy. Other services were provided as needed, including speech therapy and psychological evaluation and counseling. Our patient underwent less frequent and shorter training sessions at one session per week and about 40 minutes per session (including time used to place the device). Asakai et al. found that the effectiveness of intensive training was shown by approximately 7-10 years of age, mainly for gross motor functions in young children with spastic cerebral palsy at GMFCS level IV. The effectiveness was particularly marked by age 6. The mean age of these children was 3 years and 8

Table 2. Effectiveness of intensive physical therapy and KAATSU training in improvement of GMFM

Study		No. of cases	GMFM severity	Mean age	Frequency of training	Training period	Changes in total score
McLaughlin et al.	1998	17	71.3%	7.2 yr	171.8 h/yr	1 yr	4.2%
						2 yr	7.2%
Wright et al.	1998	12	56.5%	4 yr 10 mo	116 min/wk	1 yr	4.4%
Bower E. et al.	2001	28	GMFM III or below	5.5 yr	1 h/day or more	6 mo	5.1%
Asakai et al.	2003	47	GMFM III 52.6%	5 yr 5 mo	40 min/day or more	2.1 mo	3.7%
		55	GMFM IV 28.8%	3 yr 8 mo		1.9 mo	2.1%
Our patient	2013	1	GMFM IV 27.7%	7 yr 10 mo	40 min/wk	14 wk	8.1%

(partly revised from Asakai et al.¹³)

months. Our patient was older at 7 years and 10 months, but there was large improvement of 8.1 percentage points. These results suggest that a certain level of effectiveness can be expected from KAATSU training in young children with cerebral palsy compared with other training methods. In addition, KAATSU training has the advantages of being minimally invasive and easy to perform repeatedly. However, there are many issues, including accuracy in the comparison of our case with cases in other reports because of the small number of cases and non-uniformity of severity, rehabilitation period, and environment. Thus, further studies are necessary.

CONCLUSION

The results of this study suggest that KAATSU training can be an effective treatment method for young children with PVL. Further studies are necessary with increased number of patients and evaluation methods.

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Authors' affiliations

Hiroki Iwashita, Komone Acupuncture Clinic,
Toshihiro Morita, **Toshiaki Nakajima**, Department of Ischemic Circulatory Physiology – KAATSU Training, University of Tokyo,
Yoshiaki Sato, KAATSU International University

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