

An Open-Label Uncontrolled Trial of the Efficacy and Safety of a Hug with Singing and Rocking for Promotion of Relaxation in Pediatric Patients with Severe Motor and Intellectual Disabilities: Study Protocol

ARITOMO AO^{*,**}, OSAMU TOKUNAGA[†], CHIE AITA[‡], TOMOYA SAMURA[§], YUJI SASAKI^{§§},
REIKO MIYASHITA[#] AND TAKESHI MIYANOMAE^{##}

^{*}Department of Child Studies, Tokyo Kasei Gakuin University, Machida 194-0292,

^{**}Clinical Research Center, National Hospital Organization Nagoya Medical Center, Nagoya 460-0001,

[†]Department of Pediatrics, National Hospital Organization Minami Kyoto Hospital, Joyo 610-0113,

[‡]Department of Psychiatry, National Hospital Organization Hizen Psychiatric Center, Kanzaki 842-0192,

[§]Department of Educational Therapy, National Hospital Organization Nara Medical Center, Nara 630-8053,

^{§§}Department of Educational Therapy, National Hospital Organization Higashitokushima Medical Center, Itano 779-0105,

[#]Department of Educational Therapy, National Hospital Organization Kanagawa Hospital, Hadano 257-8585,

^{##}National Hospital Organization Minami Kyoto Hospital, Joyo 610-0113, Japan

Received 22 April 2018, accepted 17 July 2018

J-STAGE advance publication 27 November 2018

Edited by YUSHIRO YAMASHITA

Summary: Children with severe motor and intellectual disabilities (SMID) are continually exposed to stress due to their need to receive daily care. In particular, chronic physical and acute mental stress derived from daily medical care due to unstable health status are issues specific to SMID children. Therefore, it is important to approach these issues for the maintenance of their lives and quality of life. Seventeen children with a SMID-medical care dependent group (SMID-MCDG) score of 25 or more will be enrolled in this study. Intervention by a hug while singing and rocking will be performed once a week for 24 weeks. The practitioner will sing, and slowly rock the child back and forth. Primary endpoint is high-frequency component of heart rate variability by frequency analysis. Secondary endpoints are low-frequency/high-frequency components of heart rate variability by frequency analysis, activity of salivary amylase, the incidence of adverse events, and changes in appearance. Frequency analysis of heart beat changes and salivary amylase activity are used as physiological indexes for assessing response to being held while singing and rocking. In this study, we will examine the efficacy and safety of hugging while singing and rocking as a practice of Ryouiku to promote relaxation in SMID-MCDG children.

Key words severe motor and intellectual disabilities and medical care dependent groups, severe motor and intellectual disabilities, hug, medical care, safety, efficacy

INTRODUCTION

The term “children with severe motor and intellectual disabilities (SMID)” is a classification based on the

degree of disabilities, but not the name of a disease. Suzuki (1995) indicated the presence of children continuously requiring respiratory care, such as mechanical ventilation, tracheotomy, suction, and oxygen ther-

Corresponding Author: Aritomo Ao, Department of Child Studies, Tokyo Kasei Gakuin University, 2600 Aihara-machi, Machida-shi, Tokyo 194-0292, Japan
Tel: +81-42-782-9811, Fax: +81-42-782-9880, E-mail: bluecow73@kasei-gakuin.ac.jp

Abbreviations: BPM, beats per minute; HF, high frequency; IVH, intravenous hyperalimentation; LF, low frequency; MCDG, medical care dependent group; PIMD, profound intellectual and multiple disabilities; sAA, salivary amylase activity; SMID, severe motor and intellectual disabilities.

apy, or parenteral nutrition among children with SMID, and defined a group of children requiring continuous intense medical care over a long period as the SMID-medical care dependent group (MCDG) [1].

Currently, children with SMID are covered by health insurance if their daily medical care meets specific criteria which are reflected in their “SMID-MCDG score”.

According to a survey involving 8 prefectures in Japan in 2008 [2], the number of SMID-MCDG or sub-SMID-MCDG children aged 19 years or younger was approximately 0.3 per 1,000 persons of the same age. The total number of those patients in Japan is estimated to be approximately 7,000, and 70% of them are living at home. It is pointed out that this number has annually increased. In particular, the number of children requiring advanced care, including mechanical ventilation, has relatively increased in children aged 5 years or younger.

Etiological factors of SMID are primarily classified into acquired factors such as sequelae after central nervous system injury due to encephalitis or brain damage and aging-related severe status in degenerative disease, and many factors such as severe asphyxia and neonatal diseases. Especially, the number of children with SMID related to congenital or neonatal factors has increased recently. In these children, serious disorders of brain function remain, leading to prolonged conscious disturbance. “Ryouiku” and education for such children is difficult. “Ryouiku” is educational therapy under medical care, and is provided in institutions of the Medico-Social Welfare Service for SMID. In practice, including at schools for special needs education, it is difficult to assess such children’s emotions or responses to actions [3].

On the other hand, outside of Japan the term “profound intellectual and multiple disabilities (PIMD)” is used as an entity similar to “SMID-MCDG”. Although unlike Japan no criteria have been established for medical care, children with PIMD are defined as having both severe intellectual disability and motor dysfunction [4]. In addition, they have a wide range of health disorders such as severe sensory disturbance, chronic pulmonary infection, or gastroesophageal reflux [5,6]. Furthermore, there are marked individual differences in the cognitive function and communication capacity among children with PIMD. It is difficult to assess individual needs or abilities, and no effective tool for assessment has been established to date [7].

Thus, it is difficult to evaluate children’s conditions or responses due to the severity of disabilities involving cognitive function in the SMID-MCDG or PIMD

group, making supportive intervention more difficult.

In response to these problems, there has recently been an attempt in the academic field of education for children with disabilities to use physiological indexes for assessment. A study using the acceleration/deceleration responses of heart beat as an index reported deceleration response as positive evidence for orienting responses to stimuli [8,9]. Another study using a frequency analysis of heart beat changes investigated the effects of educational intervention, regarding the power spectrum of a high-frequency component as an index of the parasympathetic nerves and the power spectrum calculated by dividing a low-frequency component by a high-frequency component as an index of sympathetic nerve response [10,11]. Another study measured cerebral blood flow by near infrared spectroscopy, and indicated changes in cerebral blood flow after stimulus presentation as evidence for the recognition of stimuli [12,13]. Furthermore, a study using salivary amylase activity (sAA) as an index investigated the state of comfort/discomfort based on changes in the value after various actions [14–16]. An increase in sAA reflects eustress, and its decrease reflects distress. These physiological indexes are used as indices of autonomic nervous system activity or stress, and are interpreted as primarily reflecting the emotional responses of children with disabilities.

However, since all of these were case studies involving a small number of children (1 to 2 children), and educational intervention methods were exploratory, neither an effective method of Ryouiku nor the efficacy of assessment by physiological indexes has been demonstrated.

Few studies have scientifically demonstrated the relaxation effect of a hug, but recent studies have shown that the heart rate of an infant transported while being hugged is reduced significantly in only a few seconds (i.e., the parasympathetic nerve enters a significantly-relaxed state). Also, it was shown that children in a group who were hugged while being transported rarely moved or cried and the heart rates were lower than those in the group hugged while sitting [17]. Therefore, for the purpose of temporary release from chronic stress, we will perform intervention by a hug in hopes of achieving a relaxation effect. Because it is impossible to hug and transport children with a ventilator, we will incorporate movement by rocking. Also, practitioners will sing to alleviate the unnaturalness of rocking silently and to make it easier for them to mark the tempo of rocking. In this study, we will examine the efficacy and safety of hugging while singing and rocking as a practice of Ryouiku to promote relaxation

in 17 SMID-MCDG children. Frequency analysis of heart beat changes and sAA as physiological indexes will be used to assess response.

METHODS

Participants

The number of children was set at 17, at which level exploratory assessment is possible. Among children with SMID, those requiring continuous intense medical care will be enrolled in this study. Those with an SMID-MCDG (Table 1) score of 10 to 24 for medical care items for a period ≥ 6 months are regarded as sub-SMID-MCDG, and those with a score of ≥ 25 are regarded as SMID-MCDG.

Inclusion criteria

- 1) Multiple and severely handicapped children with SMID-MCDG scores of 25 or more.
- 2) Children with tracheotomy or who need mechanical ventilation.

- 3) Children aged 17 years or younger.
- 4) Children with body weight lower than 20 kg.
- 5) Children whose consent for participation in this study is provided by the representative.

Exclusion criteria

- 1) Children recognized as inappropriate by the principal investigator or other investigators.
- 2) Children who suffered multiple and severe handicaps from traumas such as traffic injury, etc.
- 3) Children who need sustained continuous suction.
- 4) Children with a history of bone fracture within 3 years before the time of registration.

Study treatment plan

The study procedures are shown below. The examination/observation schedule is shown in Table 2.

The study treatment will be conducted by a team consisting of several health care professionals: a practitioner, who will directly encourage disabled children, an assistant responsible for heart-rate and sAA meas-

TABLE 1.
Score of SMID-MCDG

Evaluation item		Score
I	Motor function: Sitting position	
	Respirator management	10 points
	Endotracheal intubation, tracheotomy	8 points
	Nasopharyngeal airway	5 points
	O ₂ inhalation or SpO ₂ of $\leq 90\%$ at a percentage of $\geq 10\%$	5 points
	Frequent suction (once per hour or more)	8 points
	(or ≥ 6 times/day)	(3 points)
	Continuous use of a nebulizer (or ≥ 6 times/day)	3 points
	IVH	10 points
II	Total oral assistance	3 points
	Tube feeding (including nasogastric/ gastrostomy feeding)	5 points
	Intestinal fistula/enteral nutrition	8 points
	Use of a continuous infusion pump (on intestinal fistula/enteral nutrition)	3 points
	Continuous dialysis (including peritoneal dialysis)	10 points
	Regular urethral catheterization (≥ 3 times/day), artificial anus (each)	5 points
	Postural change (total assistance)(≥ 6 times/day)	3 points
	Sweat-related clothes changing and postural correction (≥ 3 times/day) in the presence of hypertonia, which is not relieved by surgery/drug therapy	3 points

<Judgement>

SMID-MCDG: Sitting position can be maintained at best as Motor function in I, and total score in II is more than 24 points.

Sub-SMID-MCDG: Sitting position can be maintained at best as Motor function in I, and total score in II is more than 9 points, and less than 25 points.

IVH, intravenous hyperalimentation.

TABLE 2.
Examination/observation schedule

Timing		Observation period			
Timing of execution (reference: first day of the test strategy)		The following examinations/observations will be conducted at 7-day intervals for 24 weeks (A 6-day prolongation is permissible).			
Point		Before the start of a hug	Swinging by a hug	Immediately after the completion of a hug	Resting after completion
Examination item (Clinical parameter)	Heart rate (HF component, LF/HF component)	●	●		●
	Salivary amylase activity	●		●	
Observation item	Presence or absence of medical treatment	●	●	●	●
	Monitoring of adverse events	<----->			

HF, high frequency; LF, low frequency.

urement, and a physician/nurse responsible for medical observation. Study treatment is done in principle in the morning. If it is difficult to implement in the morning, it will be carried out at the same time each time. The following procedures will be performed once a week for 24 weeks:

- (1) Before the start of the study treatment, oral/intranasal suction and excretion treatment will be conducted. If suction is performed, children will be rested for at least 3 minutes.
- (2) Initially, sAA will be measured. The sAA are measured using a salivary amylase monitor (NIPRO, Osaka). Inserting a dedicated tip under the tongue for about 30 seconds and collecting saliva. Subsequently, the heart rate will be determined for 5 minutes. During measurement, attention must be paid so that each child may not be touched for purposes other than measurement. When saying something, the content must be limited to matters regarding measurement.
- (3) While touching the child's body, the practitioner will say "Let me give you a hug from now". To slowly reduce myotonia or hypersensitivity to being touched, the trunk and limbs will be touched over a period of 5 minutes. Simultaneously, singing or saying something is permissible.
- (4) The practitioner will hug the child while paying attention to the child's state or any attached medi-

cal instruments. A transverse hug will be given so that the upper body may sit slightly upright. The target angle is 45 degrees, and attention must be paid so that the child does not dislike the posture (through negative expressions or marked myotonia). The head should be placed on the practitioner's right. However, if the state of scoliosis or attachment of medical instruments affects the stability and safety of the hug, a hug on the contralateral side is permissible.

◆ For a child using mechanical ventilation, a hug with singing and rocking will be given in accordance with the following procedures:

- i. A physician or nurse temporarily removes the mechanical ventilation.
- ii. The physician or nurse supplies gas using an Ambu bag.
- iii. Under gas supply with the Ambu bag, the practitioner hugs the child.
- iv. When the hugging position becomes stable, the mechanical ventilation is attached again. At this point, to prevent the junction's becoming disconnected due to upper-body movement, adequate room should be established between the corrugated tube and junction.
- v. The physician or nurse should confirm the adequacy of mechanical ventilation under direct vision. The following points must be

checked: the presence or absence of a tear/distortion of the corrugated tube, junction loosening, water retention in the circuit/water trap, and occlusion of an expiratory valve, as well as cuff pressure.

(5) According to the following procedures, intervention by a hug with singing and rocking will be conducted, and the heart rate will be measured for 5 minutes:

- i. The electrode pad of a heart rate monitor will be attached to the bilateral forearm of the child.
- ii. In a hugging posture, the child will be informed of the start of singing/rocking. Simultaneously, the child's body will be touched. Although the site of touching is not designated, the same site (and the same site as described for Procedure (3)) should be touched every time.
- iii. After announcing the start of singing/rocking, the heart-rate measurement will be started. The heart rate monitor will be set to automatically stop after 5 minutes.
- iv. In a hug posture, the child's upper body will be slowly rocked up and down. The target heart rate is 45 to 50 beats per minute (BPM) on a metronome. However, the favorite tempo may differ among children, and the speed should be regulated in accordance with each child's appearance or expression.
The practitioner will sing Japanese children's songs matching the tempo of the rocking. The first song is "Kintaro" (only the first verse). The second song is "Urashima Taro" (until the second verse). The third song is "Momotaro" (until the third verse). When singing the second or third song, the child will always be informed of the start of singing/rocking according to the procedure described in "ii". Approximately 1-minute intervals should be established between two songs. A natural relationship must be built by saying things such as: "Do you feel good?" or "You look so happy!".
- v. After the completion of the rocking connected with the 3 songs, the practitioner will say something to the child, and touch the child's body to convey the completion of their activity. Although the site of touching is not designated, the same site (and the same site as described for Procedure (3)) should be touched every time.

(6) Immediately after the hugging intervention with singing and rocking, sAA will be measured.

(7) While paying attention to the child's state or attached medical instruments, the practitioner will lay the child on the bed. At this point, the most stable position must be prepared using a cushion. Subsequently, the heart rate will be measured for 5 minutes. If the electrode pad of the heart rate monitor deviates while laying the child on the bed, it should be reattached after positioning.

◆ Children wearing mechanical ventilation should be laid on the bed as stated previously (the study procedures (4) i-v in Study treatment plan).

Endpoints

The primary endpoint is the high-frequency (HF) components of heart rate variability as measured by frequency analysis. The HF component appears due to activation of the parasympathetic nerve, and the LF component increases as the HF component decreases. Accordingly, research using the HF component as an index showing the activity of the parasympathetic nerve, and using the ratio of the low-frequency (LF) and HF components as an indicator of sympathetic nerve activity is the mainstream practice currently [18]. In a study in which 21 adults with physical and mental health listened to music with 1/f fluctuation, the LF/HF components significantly decreased (i.e., sympathetic nervous activity subsided) after listening, and it was shown that this result correlated with the subjects' subjective evaluation of listening to music [19]. In this study, in order to assess the effect of stress relief in targeted children by a hug, we will adopt the HF component reflecting parasympathetic nerve activity as the endpoint.

Secondary endpoints are the LF/HF components by frequency analysis of heart rate variability, activity value of salivary amylase, the incidence rate of adverse events, appearance change (in facial expression, body movements).

Statistical analysis

Analysis of the primary endpoint

With respect to the HF component, summary statistics will be calculated at every point on each day of the study treatment. To examine changes after the start of the study treatment in Week 1 during the 24-week period, the paired t-test will be used, and the 95% confidence interval will be estimated.

Analysis of the secondary endpoints

With respect to LF/HF components and salivary

amylase, summary statistics will be calculated at every point on each day of the study treatment. To examine changes after the start of the study treatment in Week 1 during the 24-week period, the paired t-test will be conducted, and the 95% confidence interval will be estimated. Furthermore, the incidences of adverse events will be calculated.

DISCUSSION

The results obtained from this study will provide suggestions on the efficacy of a new support model for children with SMID who are biased toward medical care, and to provide useful proposals for support to improve their quality of life. The approach in this study is a proposal for individual support that could be applied in many children with SMID-MCDG, not only those who participate in this study, but also those who are treated at institutions of Medico-Social Welfare Service and at home.

DECLARATION

Competing interests

The authors declare that they have no competing interests.

Funding

Not applicable.

Authors' contributions

AA devised/planned this study, and finally approved the protocol. He is the study director. OT, CA, TS, YS, RM, and TM are responsible for study planning, summarization, and safety information management.

Ethics approval and consent to participate

This trial was approved by the National Hospital Organization's central review board for clinical trials (approval number: H28-0902002). The study has been registered in University Hospital Medical Information Network Clinical Trial Registry (UMIN000025855) on Feb. 27, 2017.

Prior to this study, the principal investigator or investigators should obtain written informed consent based on the patient's or their legal representative's free will after sufficiently explaining the purpose/contents of this study to patients or their legal representatives using an explanatory document. A person in parental authority for each patient is regarded as a legal representative. However, if such a person is absent, a

relative within the third degree or guardian of a minor is regarded as a legal representative. In patients admitted involuntarily due to abuse, a guardian of the minor is regarded as a legal representative.

REFERENCES

1. Michiko Y, Yasuyuki S. Entity and nursing of children with severe motor and intellectual disabilities. Supervised by Masahiko Egusa, 2nd version of the manual for nursing of severe motor and intellectual disabilities. Ishiyaku Pub, Tokyo. Inc. 2005; 158-164 (In Japanese).
2. Tateo S, Naoto K, Hidetaka T, Takakuni T, Isao T, et al. Current status and problems of medical care for children with severe motor and intellectual disabilities: A questionnaire survey involving 8 prefectures in Japan. J Jpn Pediatr Soc. 2008; 112:94-101 (In Japanese).
3. Yoshikazu N, Ryuichi K. Current Status of Education for Children With Profound and Multiple Disabilities Who Need Intensive Medical Care: A Nationwide Survey of Special Education Schools. Jpn J Spec Educ. 2013; 51(2):115-124.
4. Nakken H, and Vlaskamp C. A need for a taxonomy for profound intellectual and multiple disabilities. J Policy Pract Intellect Disabil. 2007; 4:83-87.
5. Arvio, M. & Sillanp, M. Prevalence, etiology and comorbidity of severe and profound intellectual disability in Finland. J Intellect Disabil Res. 2003; 47:108-112.
6. Van Splunder, J., Stilma, J. S., Bernsen, R. M. D., & Evenhuis, H. M. Prevalence of visual impairment in adults with intellectual disabilities in the Netherlands: Crosssectional study. Eye. 2006; 20:1004-1010.
7. Vlaskamp, C. & Cuppen-Fontaine, H. Reliability of assessing the sensory perception of children with profound intellectual and multiple disabilities: A case study. Child Care Health Dev. 2007; 33:547-551.
8. Toshihiko K, Kenji M, Keiko T, Kazuko O, Kiyoshi O. Characteristics of transient heart rate responses and persistent heart beat changes in children with severe motor and intellectual disabilities: Review in daily practice of educational therapy. Iryo-no-Hiroba. 2004; 44(4):34-38 (In Japanese).
9. Hirokata O, Munenobu Y. Longitudinal analysis of the responses of children with severe motor and intellectual disabilities to practitioners' actions. J Jpn Soc Sev Mot Intellect Disabil. 2005; 32(2):211 (In Japanese).
10. Yasuyo Y, Tatsuya K. Evaluation of Footbath Effect: Spectral Analysis of Heart Beat. Tottori Univ J Fac Reg Sci. 2006; 2 (3): 343351.
11. Yasuyo Y, Tatsuya K. The efficacy of hot-packed warm baths on limbs of severely physically and mentally handicapped children. Tottori Univ J Fac Reg Sci. 2009; 5(3):219-225.
12. Hirokata O. Examination of the responses of children with severe motor and intellectual disabilities to practitioners' actions: Introduction of assessment using near infrared spectroscopy (NIRS). J Sev Mot Intellect Disabil. 2008; 33:223 (In Japanese).
13. Daisuke H, Takamichi T, Kotaro T, Hiroyuki I, Hideo S. Near-infrared spectroscopic study of the meaning of indi-

- vidual responses made by persons with profound multiple disabilities. *Jpn J Dev Disabil.* 2008; 30(5):388-398 (In Japanese).
14. Kazunori T, Maiko W, Mieko O, Masaki Y. Correlation of Salivary Amylase Activity With Eustress in Patients With Severe Motor and Intellectual Disabilities. *Jpn J Spec Educ.* 2008; 45(6):447-457.
 15. Takeshi K. An attempt to evaluate salivary amylase activity in children with severe motor and intellectual disabilities (1st report)-Relationship with eustress-. *J Sev Mot Intellect Disabil.* 2008; 33:113-119 (In Japanese).
 16. Aritomo A. An assessment with salivary amylase activity in educational therapy intervention in children with severe motor and intellectual disabilities. *J Sev Mot Intellect Disabil.* 2015; 40(3):393-400 (In Japanese).
 17. Esposito G, Yoshida S, Ohnishi R, Tsuneoka Y, Rostagno M, et al. Infant calming responses during maternal carrying in humans and mice. *Current Biology,* 2013; 23(9):739-745.
 18. Junichiro H. An analysis of autonomic nervous functions by heart rate variation, In: cardiovascular diseases and autonomic functions, Hiroshi I, Igaku-Shoin Ltd, Tokyo, 1996:71-109.
 19. Yoshimitsu S. 1/f Fluctuation Characteristics of Heart Rate Variability in Amyotrophic Lateral Sclerosis Patients using Artificial Respirators. *Kawasaki Medical Welfare Journal.* 2008; 18(1):271-275.