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CASE REPORT

Individual Evaluation of the Visual Functions of Children with Severe Motor and Intellectual Disabilities using the Heart Rate Index

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ABSTRACT

Children with severe motor and intellectual disabilities (hereinafter, "children with SMID") produce only slight movements in response to surrounding influences, and it is difficult to evaluate their remaining sensory functions through behavioral observation alone. In this study, we focused on a first-grade Junior high-school student (hereinafter, "Student A") with concurrent visual (light perception only) and hearing (complete deafness) disabilities in addition to severe motor and intellectual disabilities, who was attending a "Special Needs Education School for the Physically Challenged", and we evaluated the student's sensory functions using the heart rate index. As a result, an orienting response, where the presentation of visual light and color stimuli coincides with a transient reduction in heart rate, was found to be highly reproducible, which suggests that the student perceives visual stimuli. Similarly, in response to facial stimuli, Student A's heart rate response to familiar and unfamiliar faces differed, with the heart rate decreasing in response to unfamiliar faces, and increasing in response to familiar faces. Based on this, it seems likely that Student A differentiates faces, and that his heart rate response differs depending on his affinity for the face. While Student A's visual functionality was previously diagnosed by a physician as light perception only, the results of this study suggest that Student A may retain the visual functionality to differentiate and recognize people.

<Keywords>
severe motor and intellectual disabilities (SMID), heart-rate fluctuation, visual function evaluation

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I. Introduction

At Special Needs Education Schools for the Physically Challenged attended by children with motor disabilities of the limbs or trunk, the severity, concurrence, and diversification of disabilities have increased in recent years. In particular, children who have mental or sensory disabilities in addition to severe physical disabilities are referred to as children with severe motor and intellectual disabilities (hereinafter, "children with SMID"). With recent improvements in home teaching programs (Kashiki, Mori & Kumai, 2013) and advances in neonatal care and emergency and critical care, even children with SMID with life-threatening disabilities are now able to live out their daily lives (Ooe & Kawasumi, 2014). Hence, Special Needs Education Schools for the Physically Challenged have many more children with severe and multiple disabilities on their registers than other types of special needs schools in Japan (MEXT, 2015).

Many children with SMID have a severe motor disability. As a result, their voluntary movement is poor, and they produce only slight movements in response to surrounding influences. While teaching staff attempt to provide encouragement with a focus on what they believe to be the remaining sensory functions, it is difficult to correctly determine whether or not movements that seem to be responsive are in fact voluntary. Hence, the question of how to ascertain the remaining sensory function, interest, and concern of children with SMID is a crucial challenge within education for the physically challenged, and a way of objectively evaluating these kinds of sensory function, interest, and concern is needed.

Physiological indices are a means of detecting bodily responses to external influences, to evaluate various sensory functions and psychological states without relying on language or movement. Hence, they have also garnered attention as a method of evaluating the sensory function, interest, and concern of children with SMID. According to a study by Ooba & Era (2002) that examined research trends in the developmental evaluation of children with SMID, physiological indices such as the heart rate and brain waves were used in 29 out of 78 articles (37%). The heart rate in particular is simpler to measure than other methods, imposes little stress on the child, and was used in many prior studies to evaluate the sensory function, interest, and concern of children with SMID (e.g., Ooe, 2012; Kawasumi, Sato, Okazawa, et al., 2008).

In these prior studies, to evaluate sensory functions, interest, and concern, the "orienting response" where the heart rate drops temporarily after presenting a stimulus, and the "expectancy response," where the heart rate increases after presenting a stimulus, were used as indicators. According to Kitajima and colleagues (Kitajima, Koike, Katada, et al., 1993), the orienting response reflects the perception of a stimulus, and is applied to the evaluation of whether or not there is sensory reception of the stimulus. However, the expectancy response is thought to reflect active attention to, for example, the calling of one's name, and is applied to the evaluation of whether or not there is

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interest in or concern about the stimulus. In light of this, since examining the phases of heart rate fluctuation is effective in evaluating the sensory function, interest, and concern of children with SMID, and because measurement is simple, we presume that they can also be applied to scenarios of educating children with SMID.

To date, other evaluations of the remaining sensory functions of children with SMID have been conducted using the heart rate index (e.g. Sasahara, 2010; Kawasumi, Sato, Okazawa, et al., 2008; Mizuta, Katagiri & Ishikawa, 1999). However, many of these prior studies primarily evaluated the tactile, olfactory, and auditory senses, and few have attempted to evaluate visual function (e.g., Sasahara, 2010). In this study, we hence focused on the case of a first-grade Junior high-school student at a Special Needs Education School for the Physically Challenged (hereinafter, "Student A") who had concurrent visual (light perception only) and hearing (complete deafness) disabilities and was diagnosed with severe motor and intellectual disabilities, and we evaluated the student's sensory functions using the heart rate as an indicator.

Student A's lessons proceeded on the basis of the medical diagnosis that he visually perceives light only. However, according to his homeroom teacher and his guardians, when shown teaching materials and when a guardian's face came close, he was seen to make what appeared to be voluntary responses such as blinking, and was thought to potentially possess a certain level of visual function. Children with cerebral palsy often have concurrent central visual disorder, but according to previous studies, this "rarely means no vision whatsoever," and "relatively good color vision (particularly reds and yellows)" has also been reported (Swift, Davidson & Weems, 2008). There are some children with SMID who attend Special Needs Education Schools for the Physically Challenged who, based on the daily behavioral observations of their caregivers, are "thought to be able to see a little," and it is thought there may be instances where slight visual function remains that cannot be evaluated accurately owing to motor impairment.

Using changes in heart rate as an index to assess Student A's ability to perceive colors and recognize faces, the objective of this study was to investigate whether the visual function of children with SMID can be assessed using heart rate indices.

II. Subjects and Methods

1. Subject

The subject was Student A (male), a first-grade Junior high-school student at a Special Needs Education School for the Physically Challenged. Student A was 12 years of age, and he had been diagnosed by a physician as having severe motor and intellectual disabilities, as well as concurrent visual (light perception only) and hearing (complete deafness) disabilities. Student A was primarily receiving scheduling and instruction in school subjects on an educational program instead of independent activities. He was also

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receiving regular medical care from nurses, guardians, and teachers in accordance with his primary physician's directions, such as artificial respiratory management, oral cavity and nasal cavity sputum aspiration, internal sputum aspiration via endotracheal catheter, and intubated feeding via gastrostomy.

According to Yokochi's classification (Ryouiku Society on Severe Motor and Intellectual Disabilities, 2014), Student A was A1-C (locomotive function: unable to turn over; intellectual development: unable to comprehend language; no significant eyelid movement). We also implemented the MEPA-R (Movement Education and Therapy Program Assessment – Revised) (Kobayashi, 2005), under which the developmental stages of three fields and six domains of a child's motor and sensory fields (posture, mobility, skill), linguistic fields (receptive language and expressive language), and sociality field (interpersonal relations) are evaluated over a 72-month period. The results yielded a development level of 0 years of age in all fields and domains.

2. Measurement device

We used the HOT-1000 manufactured by Hitachi High-Technologies Corporation (Tokyo, Japan) (Figure 1). By placing a headset-shaped holder on the front of the head, this device can detect changes in cerebral blood flow and heart rate to a tenth of a second from the reflection of near infrared light (light with a wavelength of approximately 800 nm) emitted from the scalp. This device also enables measurement from a tablet, and, by tapping the screen at the time of presenting a stimulus, the analyst can record the timing of that presentation. In this study, we used the changes in heart rate measured by the HOT-1000 device as the index of visual function evaluation.

<Figure 1> The heart rate measurement device used in this study.

3. Measurement procedures

The measurements in this study were taken at Student A's home and in a classroom at the Special Needs Education School for the Physically Challenged.

In this study, based on the physician's diagnosis of light perception, to confirm that it was possible to evaluate light perception using the heart rate, we firstly examined changes in the heart rate induced by light stimuli. Furthermore, based on the physician's diagnosis of visual functionality as "light perception only," to evaluate whether there were other remaining visual functions that had previously been considered perceptually impaired, the subject was presented with color perception and facial stimuli, and changes in the heart rate in response to these stimuli were examined (Table 1).

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<Table 1> The evaluation of Student A's sensory functions and objective thereof.

Sensory function	Stimulus	Objective
Light perception	Photo stimuli	To check whether the visual function of light perception, (diagnosed by a physician) could be evaluated with the heart rate index.
Color perception	Color stimuli	To use the heart rate index to evaluate whether there is any remaining visual function of color perception (not diagnosed by a physician).
Facial recognition	Facial stimuli (unfamiliar/ familiar)	To use the heart rate index to evaluate whether there is any remaining visual function related to facial recognition (not diagnosed by a physician).

As the method of presenting the photo stimuli, in this study, we utilized lights created by covering a flashlight with red, blue, yellow, and green-colored cellophane (Figure 2, left). We turned off the lights to darken the room, and presented each of the colored lights on Student A's face from around two meters away for 10 seconds (Figure 2, right). The intervals between shining the lights on his face were around 20 seconds, and when the stimulus was presented, the analyst recorded the timing by tapping the screen of the tablet used for measurement.

<Figure 2> Sensory function evaluation of photo stimuli.
Right: Presenting the light to Student A. Left: The flashlight used for photo stimuli.

After confirming that the visual function of light perception only as diagnosed by a physician could be evaluated by measuring the heart rate changes corresponding to the light perception, we evaluated the student's color vision, which was previously thought to be perceptually impaired. For the color stimuli, we used a picture book called the "Darumasanga" (Kagakui, 2007) that depicts a red character (Daruma), to evaluate visual function related to the perception of colors in a setting where the picture book was read to the subject. The picture book was read to Student A by his former homeroom teacher, who presented the book around 20 cm before Student A's eyes as she read it

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aloud. When the scenes in the picture book changed and the red-colored character appeared, the measuring screen was tapped to record the timing.

Furthermore, to examine Student A's interest in people and his communication potential, we conducted a visual evaluation of his perception and recognition of people's faces, which were previously considered impaired. The facial stimuli were set up so that the faces of a stranger (an unfamiliar face), Student A's former homeroom teacher, and his mother and father would be shown in a sequence at around 20 cm before Student A's eyes. When the face was shown in front of Student A's eyes, the analyst would tap the measuring screen and record the timing.

4. Data processing

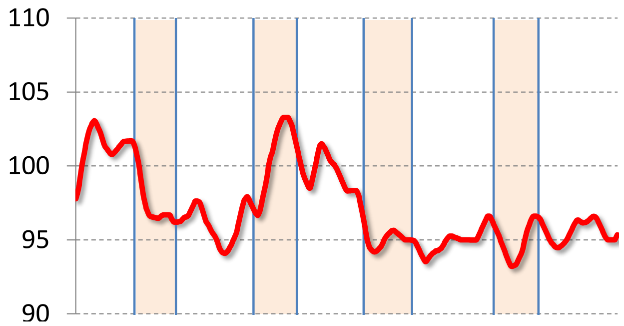
In the analysis of the results, a transient decrease in heart rate immediately after the presentation of each sensory stimuli was determined to be an orienting response, and a transient increase in heart rate immediately after the presentation was determined to be an expectancy response (a continually increasing heart rate after presenting the stimuli), and these responses were further investigated.

To assess the magnitude of heart rate changes elicited by various sensory stimuli, the average heart rates, 3 seconds before and 10 seconds after stimulus presentation, were calculated for each trial. Then, the average heart rate 3 seconds before stimulus presentation was subtracted from the average heart rate 10 seconds after stimulus presentation, to obtain the difference.

III. Results

1. Changes in heart rate associated with the presentation of photo stimuli

The changes in heart rate associated with the presentation of the photo stimuli are shown in Figure 3. When the photo stimulus was presented ten times using the light covered by the colored tape, Student A's heart rate decreased significantly immediately after the presentation 8 out of 10 times (80%).



<Figure 3> Changes in Student A's heart rate associated with the presentation of photo stimuli.(Broken line: Heart rate, Vertical line: Presentation of stimuli)

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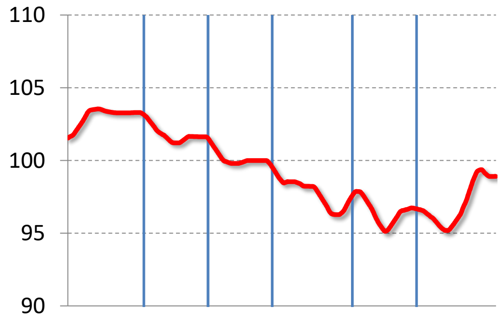
2. Changes in heart rate associated with the presentation of color stimuli

The changes in heart rate during the reading of the picture book are shown in Figure 4. When the book's red character (Daruma) appears, he is repeatedly depicted as falling over, and in this study we measured the change in heart rate while the five scenes in the picture book were read. The result was that when the picture book's scenes changed and the red character was presented, the heart rate was confirmed to decrease significantly 5 out of 5 times (100%) immediately after the character was presented.



<Figure 4> Changes in Student A's heart rate associated with the presentation of color stimuli. (Broken line: Heart rate, Vertical line: Presentation of stimuli)

3. Changes in heart rate associated with the presentation of facial stimuli

As it was suggested that student A perceives the red character, we inferred that he retains more visual functionality than previously thought. Hence, to reveal his potential for communication utilizing visual function, we measured changes in Student A's heart rate when the faces of strangers and familiar people such as his mother, father, and former homeroom teacher appeared before his eyes.

The result was that when the stranger's face was shown before Student A's eyes, student A's heart rate immediately decreased significantly 2 out of 2 times (100%) (Figure 5, top left). However, when the face of student A's former homeroom teacher was shown in what was the first occasion of their meeting in around a year, Student A's heart rate immediately increased slightly 2 out of 2 times (100%) (Figure 5, top right). Furthermore, when we measured the change in heart rate on the appearance of Student A's mother and father with whom he has the most regular contact (Figure 5, lower right), his heart rate was confirmed to immediately increase suddenly thereafter 2 out of 2 times (100%). The amount of change at around the time the familiar/unfamiliar faces appeared is summarized in Table 2. Contact with the student was most frequent with his mother and father, followed by his former homeroom teacher, and then the unfamiliar face, and, although his heart rate decreased on seeing the unfamiliar face, with the familiar faces we observed a trend whereby his heart rate increased with the frequency of contact.

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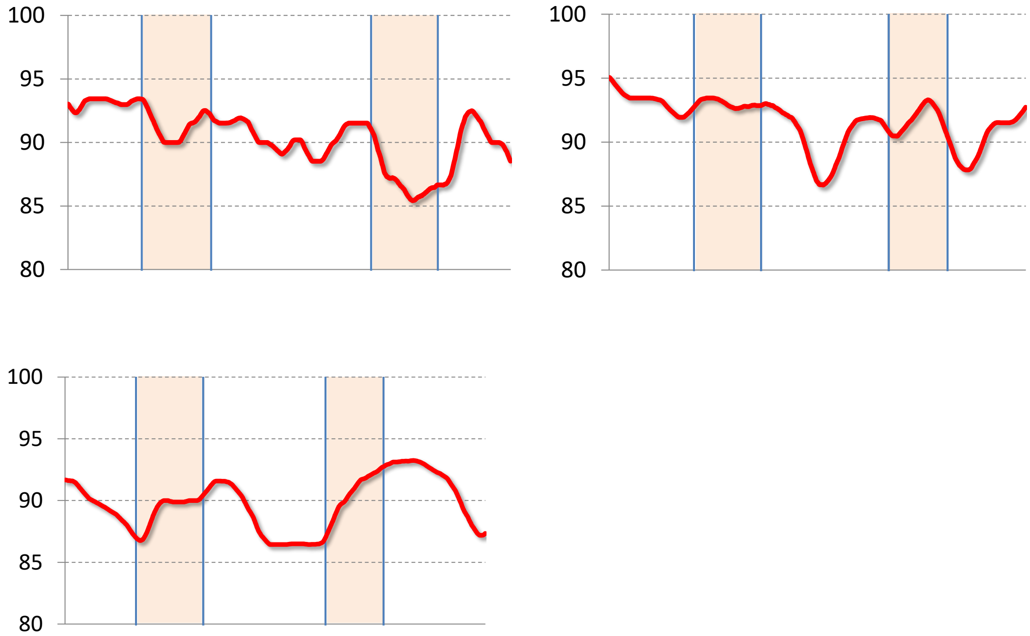

<Figure 5> Changes in Student A's heart rate associated with the presentation of facial stimuli. (Broken line: Heart rate. Vertical line: Start/end of stimuli)

<Table 2> Changes in Student A's heart rate around the time of presentation of the facial stimuli.

		Familiar face	
	Unfamiliar face	Former homeroom teacher	Mother and father
1st	-3.42	0.77	3.38
2nd	-5.72	2.41	6.22

4. Heart Rate Changes After Various Sensory Stimuli

To assess the magnitude of heart rate changes elicited by various sensory stimuli, the differences between the heart rates before and after stimulus presentation were obtained (Table 3). The results revealed that the heart rate among the child subjects decreased by an average of -1.53/min after presentation of light stimuli, and -1.07/min after presentation of color stimuli. Furthermore, regarding face stimuli consisting of strangers' faces, the heart rate after stimulus presentation decreased dramatically, compared to other visual stimuli, by an average of -2.83/min.

On the other hand, regarding familiar faces, after the child subjects were shown their previous homeroom teachers' faces, the average heart rate increased by +0.59/min after presentation of face stimuli. After they were shown their parents' faces, the change was even more remarkable, with the average heart rate increasing by +2.53/min after stimulus presentation.

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<Table 3> Changes in Heart Rate (/min) Before and After Stimulus Presentation During Each Trial (/min)

		1st trial	2nd trial	3rd trial	4th trial	5th trial	AVERAGE
Photo stimuli		-4.11	+2.73	-2.82	-1.93	—	-1.53
Color stimuli		-1.47	-1.47	-1.59	-0.26	-0.54	-1.07
Facial stimuli	Unfamiliar	-2.21	-3.46	—	—	—	-2.83
	Familiar (teacher)	+0.90	+0.29	—	—	—	+0.59
	Familiar (parents)	+1.13	+3.92	—	—	—	+2.53

IV. Discussion

In this study, the visual function of Student A, who is visually impaired as well as mentally and physically disabled, was assessed using changes in heart rate as an index, in order to investigate whether the visual function of children with SMID can be assessed using heart rate indices. The transient reduction in Student A's heart rate that was associated with the presentation of the photo stimuli had a high reproducibility of 8 out of 10 times, which, also based on the timing of the start of the change, was likely to be an orienting response. This suggests that Student A is capable of perceiving photo stimuli. The physician also diagnosed Student A as having a concurrent visual impairment of light perception only and it seems likely that his remaining visual functions are evaluable using the heart rate as an indicator.

It was previously thought that Student A's color perception was impaired, based on the physician's diagnosis of light perception only. However, the daily observations of the homeroom teacher and his guardians also reported a slight eyelid response to characters containing color. Hence, considering that other visual functions may remain, a change in heart rate was also detected while a picture book depicting a red Daruma character was read. As a result, when the red character was presented before the student's eyes, his heart rate decreased immediately afterwards, and this heart rate decrease that seems likely to be an orienting response was confirmed to be highly reproducible. Therefore, it seems likely that Student A perceived the red character, and detected the changes in the scenes. This suggests that Student A retains greater visual function than expected, and we thus hoped that this remaining function, which was difficult to evaluate with prior methods, could be demonstrated using the heart rate.

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Accordingly, in order to examine Student A's potential for interpersonal communication, in an attempt to confirm whether or not he is capable of perceiving and recognizing faces – the foundation of communication – we also examined the changes in heart rate when Student A saw an unfamiliar face and the faces of his former homeroom teacher, mother, and father that he knew well. When he was shown the face of a stranger, his heart rate decreased, and the amplitude change was more significant than with the photo or phono stimuli. Furthermore, the heart rate decrease was also of high reproducibility, and is thus likely to be a response of perception and of orientation towards the stranger's face.

So how does the heart rate change with a face that Student A knows well? In this study we detected the heart rate when Student A was shown the face of his former home room teacher, who had interacted with him daily at school until a year earlier, and when Student A was shown the faces of his father and mother who he sees every day. When the faces of the former homeroom teacher and the student's parents appeared before Student A's eyes, his heart rate increased significantly, and the phases of the change differed from the changes when other sensory stimuli or the stranger's face were presented. The increase in heart rate when Student A saw faces he knew well likely correlates with affinity, and the heart rate increased significantly more when seeing the face of his mother and father who he sees every day, than when seeing the face of his former home room teacher, who he had not seen in a year.

A heart rate increase of this kind immediately after Student A saw a very familiar face is likely to be an expectancy response. According to Kitajima and colleagues (Kitajima, Koike, Katada, et al., 1993), expectancy responses can be considered to reflect active attention towards a stimulus, and they point out that expectancy responses appear when a pleasant emotion arises in particular. In this study, the heart rate increase that was viewed as an orienting response from Student A was limited to when he saw the faces of his mother and father and former homeroom teacher with whom he had interacted on a daily basis, and based on this, since good relationships had been created through daily interactions with Student A, it was inferred that an expectancy response appeared that coincided with a pleasant emotion upon seeing the faces of his mother and father and former homeroom teacher.

As stated above, although comprised of only one case, this study suggests that it may be possible to assess the remaining visual function of children with SMID using heart rate indices. For many children with SMID, response movements are very weak due to the severe movement disability, and even in cases where very little visual function remains, it is extremely difficult to assess visual function by observing behavior alone. The results of this study provide new insight into the assessment of visual function for children with SMID using heart rate indices, by which very little prior research has been done. In the future, it will be necessary to conduct additional visual assessments with

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other children with SMID using heart rate indices, and to further verify the usefulness of this method.

Depending on the presence or absence of remaining sensory functions and the interest or concern on the part of the student, the method of presenting teaching materials and communication support differ greatly. The results of this study suggested that even for children with SMID such as Student A, who have severe motor impairments and for whom evaluation based on behavioral responses is problematic, it may be possible to utilize physiological indices to evaluate interest and concerns through their remaining sensory functions and pleasant emotions. Based on these findings, we suggest it might be beneficial to use physiological indices appropriately in a school education setting in order to understand the actual condition of children with SMID and evaluate their learning. We hope that the fragment of this student's hidden reality that was revealed in this study can serve as a helpful clue in providing individually tailored and effective educational assistance.

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