Study on the relationship between sensation of the mandibular position and the oral dysfunctions in the cerebral palsy patients

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Abstract The ability to discriminate the sensation of the mandibular position in the cerebral palsy (CP) patients was studied and the relationship between this ability to discriminate the sensation of the mandibular position and the oral dysfunctions was evaluated. Interdental dimension discrimination tests (IDD test) were performed in order to evaluate the ability to discriminate the sensation of the mandibular position in 18 CP patients and in 15 healthy individuals. Using the test result, points of subjective equality (PSE) and difference limen (DL) were obtained. In addition, the oral dysfunction index was used to evaluate the oral dysfunctions of the CP patients. The results obtained are as follows:

1) PSE in the CP patients was significantly lower than that in the healthy individuals.

2) No significant difference was observed in DL between the CP patients and healthy individuals.

3) A significant correlation was observed between PSE and ODI in the CP patients.

Key words Cerebral palsy, Masticatory muscle, Muscle spindle, Oral dysfunction, Sensation of the mandibular position

Introduction Cerebral palsy (CP) involves central dyskinesia, which is characterized by involuntary movements or the inability to perform smooth movements due to hypermyotonia1–3). In CP patients, functional disabilities not only in their limbs but also in the oral area related to mastication, swallowing and speech caused by uncoordinated movements of muscle groups are observed mainly in the masticatory muscles4–14). Previous studies of the oral functions of CP patients using electromyography (EMG) to examine mastication muscle activities clarified that CP patients have irregular mastication rhythm compared to healthy individuals4,5,9–14).

The uncoordinated mastication muscle activities observed in CP patients are attributed to a dysfunction in the output of information from the central nervous system to the target peripheral organ4,5,9–14). However, there are still many points that remain unclarified.

According to recent psychosensory studies on feedback mechanisms of the muscular sensation of the limbs of CP patients while they performed a movement, it was clarified that CP patients have a characteristic of perceiving muscular sensation that leads to overestimation of their movements15–20). We consider that this abnormal perception characteristic of the muscle sensation is one of the causes of dyskinesia observed in the limbs of CP patients. Therefore, similar to the dyskinesia observed in limbs, oral dysfunctions in CP patients are considered to be related to this abnormal perception characteristic.
in terms of the muscles sensation of masticatory muscles. However, there have been no reports on the perception characteristic of the muscles sensation in the masticatory muscles of CP patients.

Morimoto et al.\textsuperscript{21–23} developed a method which quantitatively evaluates the ability to discriminate the mandibular position sensation with respect to the oral aperture when subjects were instructed to gradually open their mouth using percentage of inaccuracy of response, and the interdental dimension discrimination test (IDD) which qualitatively evaluates the same event using a psychosensory index. Morimoto et al.\textsuperscript{21–23} stated that the score for the ability to discriminate the thickness of sticks determined by the IDD test reflects the characteristic of perception of the muscles sensation of masticatory muscles.

Yamaguchi et al.\textsuperscript{24} studied the ability to discriminate the sensation of mandibular position by CP patients using percentage of inaccuracy of response and reported that CP patients had difficulties in discriminating the thickness of sticks requiring an oral aperture smaller than the standard level compared to healthy individuals. There have been no previous reports on the ability of CP patients to discriminate mandibular position sensation using the psychosensory index.

In this study, we compared the ability of CP patients and healthy individuals to discriminate mandibular position sensation using the IDD test in order to determine the relationship between oral dysfunction and the perception characteristic of the ability of CP patients to discriminate mandibular position sensation in terms of the muscles sensation of masticatory muscles. Furthermore, we studied the relationship between the ability to discriminate mandibular position sensation and oral dysfunctions in CP patients.

**Subject and Methods**

**Subjects**

Eighteen CP patients (10 male and eight female patients; average age, 26.1 years, hereafter referred to as the CP group), who were outpatients of the Department of Dentistry in the General Nursing Center for handicapped children in Fukushima Prefecture and those receiving services from the Kaikoen Social Welfare Service Corporation in Saitama Prefecture, were enrolled in this study as subjects. The Barthel index (BI)\textsuperscript{25–29} of the subjects were relatively high (62.2), indicating that the severity of our subjects’ physical dysfunctions was mild in their daily activities.

Prior to the study, the details of this study were fully explained to the subjects themselves, guardians (parents) and caretakers, and their consent was obtained. Approval for the study was given by the President of Colony Ranzango, the Director of Ranzango Hospital and the Manager of the Dental Department.

The criteria for selecting the CP patients were based on those of Yamaguchi et al.\textsuperscript{24}

1) No missing frontal teeth in the upper and lower jaws
2) The position of head and jaw at rest can be maintained
3) Absence of general complications other than CP
4) Not ordinarily taking medications such as muscle relaxants
5) The aim of this study can be understood and informed consent can be obtained

Fifteen healthy adults (10 male and five female patients; average age, 25.1 years old, hereafter referred to as the healthy group) were enrolled as controls.

**Study methods**

1) Ability to discriminate mandibular position

The ability to discriminate mandibular position was measured using the IDD test developed by Morimoto et al.\textsuperscript{21–23}

**Specified movement**

To measure the ability to discriminate mandibular position, the subjects’ Frankfort plane was maintained at a position parallel to the floor and visual information was blocked by eye masks. The measurement procedure was as follows. First, the subjects were instructed to hold a standard stick for 5 sec between the central incisors in their upper and lower jaws without moving. Then, the standard stick was replaced by a test stick, and the subjects were again instructed to hold it at the same position for 5 sec. After the test stick was removed, the subjects were instructed to determine whether the oral aperture for the thickness of the test stick was larger than, the same as or smaller than the thickness of the standard stick. A series of trials were repeatedly performed for each test stick. Nine types of test sticks were randomly assigned to the subjects using a table of random numbers. Stainless-steel standard
and test sticks (manufactured by Tokyo Shizai) were employed in this study. The standard stick was 10.0 mm in diameter. Nine different thicknesses of test sticks, including one with a diameter of 10.0 mm, the same as the standard stick, a group with diameters smaller than that of the standard stick (9.5, 9.0, 8.5 and 8.0 mm) and a group with diameters larger than that of the standard stick (10.5, 11.0, 11.5 and 12.0 mm), were prepared. Assigning the nine types of test sticks was performed in one session; by changing the combination of sticks to be assigned, 10 sessions in total were performed.

**Measurement of the ability to discriminate mandibular position**

As indices for the ability to discriminate mandibular position, the point of subjective equality (PSE) and the difference limen (DL) were employed\(^{30}\). The PSE is an oral aperture which subjects can perceive as being equivalent to the standard oral aperture (10.0 mm) with a probability of 50%. That is, PSE is an index used to evaluate whether or not the standard oral aperture is underestimated or overestimated by subjects. The DL is the range of oral aperture which subjects can perceive as being equivalent to the standard oral aperture (10.0 mm) with a probability of 50%.

**Method of calculating PSE**

The subjects’ scores were totaled according to three choices. From the total scores for choices which evaluated the test sticks as bigger than the standard stick, the maximum thickness which the subjects could perceive as bigger than that of the standard

<table>
<thead>
<tr>
<th>Evaluated items</th>
<th>Evaluated scores 1</th>
<th>Evaluated scores 2</th>
<th>Evaluated scores 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Posture Unable to maintain one’s posture</td>
<td>Movable, Malpositioned</td>
<td>Stable</td>
<td></td>
</tr>
<tr>
<td>Deformation in the head Presence</td>
<td>Absence</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Symmetry of the face Asymmetry</td>
<td>Symmetry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Presence or absence of mouth breathing Presence</td>
<td>Absence</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Language Unable to speak</td>
<td>Difficulty in listening</td>
<td>Able to speak normally</td>
<td></td>
</tr>
<tr>
<td>Maintaining one’s jaw at rest Unable to follow instructions</td>
<td>Able to perform</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Movement of jaws Unable to follow instructions</td>
<td>Able to perform</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Symmetrical lips Asymmetry</td>
<td>Symmetry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Closure of lips Unable to follow instructions</td>
<td>Immediately open one’s mouth after closing</td>
<td>Able to perform</td>
<td></td>
</tr>
<tr>
<td>Lip protrusion Unable to follow instructions</td>
<td>Time consuming due to involvement of other actions</td>
<td>Able to perform</td>
<td></td>
</tr>
<tr>
<td>Tongue protrusion Unable to follow instructions</td>
<td>Time consuming due to involvement of other actions</td>
<td>Able to perform</td>
<td></td>
</tr>
<tr>
<td>Elevation of tongue Unable to follow instructions</td>
<td>Time consuming due to involvement of other actions</td>
<td>Able to perform</td>
<td></td>
</tr>
<tr>
<td>Swallowing Unable to follow instructions</td>
<td>Time consuming due to involvement of other actions</td>
<td>Able to perform</td>
<td></td>
</tr>
<tr>
<td>Movement of lips during swallowing Presence</td>
<td>Absence</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bruxism Presence</td>
<td>Absence</td>
<td></td>
<td></td>
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<tr>
<td>Slavering Presence</td>
<td>Absence</td>
<td></td>
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</tbody>
</table>
stick was obtained. This thickness was defined as the upper stimulus limen (UL). From the total scores for choices which evaluated the test sticks as smaller than the standard stick, the minimum thickness which the subjects could perceive as smaller than that of the standard stick was obtained. This thickness was defined as the lower stimulus limen (LL). The PSE was calculated by dividing the sum of UL and LL by two (the average).

**Method of calculating DL**

Similar to the method of calculating the PSE, UL and LL were first obtained, then DL was calculated by dividing the difference between UL and LL by two (the average).

**Evaluation of oral dysfunction**

To evaluate oral dysfunction, we employed the oral dysfunction index (ODI) developed by Watanabe. Table 1 shows the standards for determining ODI. In the ODI, points are allotted for each item depending on the ability that determines the state of oral dysfunction. The total score, which is the sum of scores of each item, was used to evaluate the oral dysfunction. The total score ranged from 25 to 39 points. The lower the total score, the more severe the oral dysfunction.

**Statistical analyses**

The Student $t$-test was employed to compare PSE and DL between the CP and healthy groups. Spearman’s rank correlation coefficient was employed to determine the correlation between PSE and ODI in the CP group.

**Results**

**The ability to discriminate mandibular position**

Figure 1 shows the PSE of one subject. Table 2 shows the mean $\pm$ standard deviation of PSE and DL in the CP and healthy groups, and results of the significance test of the mean values.

PSE in the CP group was $9.99 \pm 0.25$ mm, which was significantly lower than that in the healthy group, $10.23 \pm 0.15$ mm ($P<0.01$). DL in the CP group was $0.41 \pm 0.18$ mm, while DL in the healthy

<table>
<thead>
<tr>
<th></th>
<th>CP group</th>
<th>Healthy group</th>
<th>The significance test</th>
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<tbody>
<tr>
<td>PSE</td>
<td>$9.99 \pm 0.25$</td>
<td>$10.23 \pm 0.15$</td>
<td>**</td>
</tr>
<tr>
<td>DL</td>
<td>$0.41 \pm 0.18$</td>
<td>$0.36 \pm 0.18$</td>
<td>**</td>
</tr>
</tbody>
</table>

mean $\pm$ s.d. **: $P<0.01$
group was $0.36 \pm 0.18$ mm. No significant difference in the DL was observed between the two groups ($P>0.05$).

### Table 3 ODI in the CP group

<table>
<thead>
<tr>
<th>number of cases</th>
<th>ODI</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>39</td>
</tr>
<tr>
<td>2</td>
<td>25</td>
</tr>
<tr>
<td>3</td>
<td>30</td>
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<tr>
<td>4</td>
<td>29</td>
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<td>5</td>
<td>39</td>
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<td>6</td>
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<td>31</td>
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<td>16</td>
<td>39</td>
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<tr>
<td>17</td>
<td>27</td>
</tr>
<tr>
<td>18</td>
<td>36</td>
</tr>
</tbody>
</table>

- mean ± s.d. 32.4 ± 5.0
- max 39
- min 25

### Oral dysfunction

Table 3 shows results of ODI in the CP group. The mean ODI in the CP group was 32.4 ± 5.0, with 25 being the minimum and 39 the maximum.

Figure 2 shows the correlation coefficient of the difference between PSE and the standard oral aperture, and ODI (and their scatter diagram). The correlation coefficient of the difference between PSE and the standard oral aperture, and ODI was 0.60, indicating a significant correlation ($P<0.01$).

### Discussion

#### Subjects

CP patients suffer from central dyskinesia resulting from a nonprogressive disease of the brain present from fetal to postnatal stages\(^1\)\(^-\)\(^3\). Lesions are found in the pyramidal or extrapyramidal system of CP patients exhibiting various forms of dyskinesia\(^1\)\(^-\)\(^3\). Generally, CP patients are classified based on the form of dyskinesia of the limbs: the spastic type is caused by disorders in the pyramidal system and the athetoid type by disorders in the extrapyramidal system\(^1\)\(^-\)\(^3\). (However, there are intermediate types of CP which make classification of CP difficult. Therefore, in this study, we did not focus on the type of CP.

![Fig. 2 Correlation between the difference between PSE and standard oral aperture and ODI in the CP group](image-url)
BI is the subjective indication of the activities in daily living (ADL) of those with physical dysfunctions\textsuperscript{26–29}. The average BI of our subjects in the CP group was 62.2, which was relatively high, indicating that the severity of physical dysfunction was mild.

**Method**

**Discrimination ability**

Methods of evaluating human sensory perception using the psychosensory index are broadly separated into a method using the stimulus limen and a method using the difference limen\textsuperscript{30,32,33}. A method using the stimulus limen determines the boundary between being able and unable to discriminate. A method using the difference limen evaluates the limit of perceiving the difference between two or more stimuli. The ability to perceive such a difference, that is, the ability to discriminate, is presented by indices such as PSE and DL.

PSE and DL are employed as indices of perception characteristics for sensations such as brightness and hue for vision, loudness and pitch of sound, quality and richness of taste and proprioception in the psychosensory and ergonomic fields\textsuperscript{31}. In the field of dentistry, PSE and DL are employed for the evaluation of the ability to discriminate mandibular position\textsuperscript{21–23}, and for visual evaluation of the color of an artificial crown; they are also used for evaluation of the sweetness of sugars\textsuperscript{34}.

The IDD test evaluates the ability to discriminate mandibular position using PSE and DL as indices, and evaluates the ability to discriminate differences between two or more stimuli\textsuperscript{21–23}. In this study, a certain oral aperture was obtained by making the subjects hold the standard or test sticks between the central incisors in the upper and lower jaws as stimuli.

Yamaguchi \textit{et al.}\textsuperscript{23} studied the range of the oral apertures which CP patients could the ability of discriminate using the IDD test. They reported that CP patients correctly discriminated the thickness of samples up to the maximum oral aperture of 12.0 mm in diameter and the minimum oral aperture of 8.0 mm in diameter, the same as the range for healthy individuals. Therefore, they confirmed that the range of oral apertures (12.0 to 8.0 mm in diameter) for the healthy group could be applied to CP patients.

**Mandibular position sensation**

Sensory receptors for discriminating mandibular position sensation are considered to be associated with the periodontal ligament, mandibular joints, tendons, and the muscle spindle of the occlusal muscle\textsuperscript{35}.

Broekhuijen \textit{et al.}\textsuperscript{36} administered local anesthesia to subjects in various parts of the oral cavity including the mandibular joints and periodontal ligament, and reported no changes in the mandibular position sensation. Morimoto \textit{et al.}\textsuperscript{21} studied the mandibular position sensation in subjects whose mandibular joints were injured or pathologically calcified and reported that no differences were observed when compared to the healthy group. On the other hand, Morimoto \textit{et al.}\textsuperscript{22,32} studied the mandibular position sensation in patients with the Duchenne type of myodystrophy whose muscular tissue was damaged and in those for whom either side of masticatory muscles had been surgically removed. They reported that the ability of these patients to perceive mandibular position sensation was inferior compared to that of the healthy group. Based on the aforementioned reports, we considered that the mandibular position sensation is strongly affected by the muscular sensation stemming from the muscle spindle rather than from the sensory receptors of the mandibular joint or periodontal ligament.

**ODI**

Oral function is effected through coordination between masticatory muscles, anterior and posterior cervical muscles to maintain the head position, facial muscles, and internal and external muscles of the tongue\textsuperscript{37}. When movements of these various muscle groups are uncoordinated, various forms of oral dysfunction occur. As a method of quantifying symptoms of oral dysfunctions, ODI has been employed to evaluate the severity of oral dysfunction\textsuperscript{22} and the effect of food-taking training\textsuperscript{4,10,38} in patients with psychosomatic disorders including CP patients.

Sensory information regarding mandibular position sensation is considered to influence the feedback mechanisms of oral function. Therefore, we considered that oral dysfunction in CP patients can be attributed to a muscular perception characteristic that is specific to the sensation of mandibular position. In this study, the correlation coefficient
was employed to determine the relationship between the ability to discriminate mandibular position sensation and oral dysfunction.

**Results**

**PSE and DL**

The mean PSE in the healthy group was 10.23 mm, similar to 10.24 mm reported by Morimoto et al.\(^\text{22}\). The mean PSE in the CP group was 9.99 mm, which was significantly lower than that in the healthy group of 10.23 mm (Table 2). Accordingly, the ability to discriminate mandibular position sensation of the CP group has the perception characteristic of overestimating the thickness of samples compared to the healthy group. Suzuki et al.\(^\text{39}\) studied the ability to discriminate the muscular sensation of the limbs of CP patients and reported that CP patients have the perception characteristic of evaluating small changes in muscular tension as large changes compared to a healthy group. As a cause for this phenomenon, Suzuki et al.\(^\text{39}\) assumed that γ-motor neurons that regulate the muscle spindle in CP patients were excited excessively compared to those of healthy individuals, resulting in an increase in the firing frequency of afferent impulses. That is, since the mandibular position sensation is considered to be related to perception characteristics of masticatory muscle sensation similar to that of limbs\(^\text{21,22,35,40–43}\), a decrease in PSE of the mandibular position sensation in CP patients reflects excessive excitement of γ-motor neurons that regulate the muscle spindle in the masticatory muscles.

DL in the CP group was 0.41 mm, while that in the healthy group was 0.36 mm. No significant difference in DL between the two groups was observed (Table 2), indicating that the range of limen level that the CP patients were able to discriminate was the same as that in the healthy group. Accordingly, it was clarified that PSE strongly reflects the muscular perception characteristics in terms of the ability to discriminate mandibular position sensation in CP patients, compared to DL. Thus, we decided to employ PSE as an index of the ability to discriminate mandibular position sensation in order to study the relationship between the PSE and ODI in CP patients.

**Oral dysfunction**

The mean ODI in the CP group was 32.4 points, and ranged from 25 to 39 points, which was higher than the previously reported ranges\(^\text{38,44,45}\). We considered that the severity of oral dysfunction in our subjects was mild.

**Correlation between the difference between PSE and standard oral aperture and ODI**

A significantly positive correlation was observed between PSE and ODI in the CP group, indicating that the lower the PSE, which represents the ability to discriminate mandibular position sensation in the CP group, the more severe the oral dysfunction.

It is found that the CP patients have the perception characteristic of overestimating the ability to discriminate of the muscular sensation in their limbs compared to healthy individuals, and this overestimation makes coordination of limb movements difficult\(^\text{15–20}\). The same can be said for oral dysfunction. Similar to the limbs, CP patients have the perception characteristic of overestimating the sensation in the masticatory muscles, and therefore, CP patients have more difficulty in performing intended smooth movements compared to healthy individuals.

Results of this study indicate that one of the causes of dyskinesia in the oral area of CP patients is that the feedback of sensory information from masticatory muscles regarding their activities is not well controlled.

**Conclusions**

To study the ability to discriminate mandibular position sensation in CP patients, PSE and DL were obtained from 18 CP patients and 15 healthy individuals as the control group using the IDD test. The correlation between the ability to discriminate mandibular position sensation and oral dysfunction in the CP patients was investigated. The following conclusions were obtained.

1. PSE in the CP group was significantly lower than that in the healthy group.
2. No significant difference was observed in the DL between the CP patients and healthy individuals.
3. A significant correlation was observed between PSE and ODI in the CP group.

Based on these results, one of the causes of dyskinesia in the oral area of CP patients is that the feedback of sensory information from masticatory muscles regarding their activities is not well controlled.
References