Dystonia-like Movement Disorders Ameliorated by Shear Force and Pressure Stimulation - A Case Report of Small Infarction in the Left Posterolateral Thalamus

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Abstract:
Focal dystonia (FD) can develop after thalamic lesions. Abnormal somatic sensations were argued to be responsible for FD. Our patient experienced FD-like movement disorders, agraphesthesia, and a reduced sense of shear force on the skin and pressure to deep tissues of the right upper limb following a small infarction in the left posterolateral thalamus. FD-like symptoms improved while the skin was being pulled or the deep tissue was being pushed in a manner proportional to the strength of muscle contractions. Therefore, the lack of these sensations was suggested to be related to FD-like symptoms.

Key words: focal dystonia, thalamus, shear force, deep pressure sensation

Introduction
Dystonia is defined as a movement disorder (MD) characterized by sustained or intermittent muscle contractions causing abnormal, often repetitive, movements, postures, or both. Dystonic movements are typically patterned, twisting, and may be tremulous. Dystonia is often initiated or worsened by voluntary action and associated with overflow muscle activation (1). Muscle contractions are characterized by an abnormal pattern of muscle activity with co-contraction of the agonist and antagonist muscles (2).

Focal dystonia (FD) refers to dystonia occurring in one region of the body, such as cervical dystonia and writer’s cramp. Recently, it was argued that abnormal somatic sensations might be responsible for FD (3). MDs secondary to stroke are rare, and 37% of them involve the thalamus. In patients with thalamic lesions, the posterolateral thalamus is most commonly involved in MDs following a stroke, and FD is the most common symptom (4). However, few cases of FD as solo manifestation resulting from a small lesion located in the posterolateral thalamus have been reported (5).

We herein report a patient who developed FD-like MDs in the right upper limb after a small infarction in the posterolateral region of the left thalamus. The patient experienced only FD-like MDs and particular abnormality of somatic sensations. The MDs improved while being stimulated to complement the impaired somatosensory information.

Case Report
A 61-year-old right-handed man with 16 years of education and a history of diabetes was hospitalized because of paresthesia in the right upper and lower limbs and mild weakness in the right lower limb. Magnetic resonance imaging on admission revealed a small infarction in the posterolateral region of the left thalamus. The region involved at least part of the ventral posterior complex (VPC) (6) (Fig. 1). The abovementioned symptoms disappeared by the time of the hospital visit; however, MDs (described later) persisted in the right upper limb.

Twenty-five days after the onset of the MD, the patient...
Magnetic resonance images of the brain. Fluid-attenuated inversion recovery images showed a small infarction in the posterolateral region of the left thalamus, which involved at least part of the ventral posterior complex.

Presented to our hospital with a desire to receive rehabilitation therapy. Subsequently, outpatient treatment comprising neurocognitive rehabilitation, several types of drug therapy, and repetitive transcranial magnetic stimulation was administered. However, 2.5 years passed, and the patient did not experience marked improvement in his symptoms. Treatment with botulinum toxin had been performed six times, which was relatively effective. However, its effects lasted only for approximately two months. The patient realized that his symptoms were relatively relieved when an elastic band was wrapped tightly around the right upper arm, and he had been using this strategy.

A neurological examination revealed no abnormalities, except for MDs and abnormal somatic sensations (described later) in the right upper limb. Furthermore, a neuropsychological examination revealed no abnormalities, except for worsening scores on the digit span backward test (Table 1).

Characteristics of MDs

The patient experienced MDs in the right upper limb when performing flexion and extension of the wrist, elbow, and finger joints. When these movements were first performed, the joints moved relatively smoothly. However, with repetition, joint motion became slow and exhibited a start-and-stop pattern, similar to the cogwheel phenomenon. During such movements, strong and persistent contractions of both agonistic and antagonistic muscles were observed and palpable. The symptoms were most severe in the elbow joint. During palmar flexion, the proximal and distal interphalangeal joints of the second to fifth fingers were hyperextended, although the metacarpophalangeal joint was flexed (Fig. 2a). During passive joint movements, neither a movement similar to the cogwheel phenomenon nor muscle rigidity occurred.

Electrodes were placed on the biceps brachii and lateral head of the triceps brachii to record surface electromyograms (SEMGs) when the patient performed flexion and extension of the elbow joint. The SEMGs revealed strong and persistent activity in both muscles (Fig. 3a) compared to the upper left limb (Fig. 3b), even when the patient was instructed to sit on a chair and place his elbow on the table such that he did not need to contract the triceps muscle. These MDs affected the patient’s daily activities requiring repetitive motions, such as washing his hair and using a kitchen knife. The patient reported that “I feel that my right upper arm is always squeezed. When I move it, I experience significant stiffness, and it becomes very difficult to move my elbow after performing repetitive motions of my right
upper arm. In addition, the stiffness spreads to the forearm and hands if further repetitive motions are performed. As a result, I feel it is very difficult to move these areas as well.”

**Intervention for MDs**

Asahi et al. reported that the hanger reflex suppressed the symptoms of cervical dystonia (7). These effects are hypothesized to be induced by shear force on the skin (i.e. a pulling force that is parallel to the skin surface) (7). Based on that report, we applied plastic tape to his extended elbow starting from the posterior surface of the upper arm to the proximal posterior surface of the forearm beyond the joint, which caused the skin over the triceps brachii to be pulled when flexion of the elbow joint was performed (Fig. 2b). The patient was instructed to perform flexion and extension, and his symptoms improved moderately. This effect was also observed on the SEMG (Fig. 3c). However, when plastic tape was applied from the front surface of the upper arm to the proximal part of the forearm (Fig. 2c), no such effect was observed (Fig. 3d). This suggested that the application of shear force stimulation, which is increased following the flexion of the elbow joint, may help alleviate MDs.

Dagostinoa et al. reported a sensory trick for treating writer’s cramp, namely gently grabbing the right wrist with the left hand (8). Accordingly, we wrapped a blood pressure sleeve around the patient’s upper arm (Fig. 2d) to apply pressure and then instructed him to perform flexion and extension. This resulted in remarkable improvement in his symptoms. According to the patient, pressures of 20-30 mmHg were the most effective. Furthermore, he reported that the effect of the blood pressure sleeve was remarkably stronger than that of the elastic band that he had used previously. This suggests that a mild pressure stimulus that is increased during muscle contraction may have been effective in treating MDs in this patient.

To confirm the combined effects of the shear force applied to the skin and pressure stimulus on MDs, we conducted the following examinations:

1. Right arm: No stimulation.
2. Right arm: A piece of elastic tape (14.5 cm long and 7.5 cm wide) was applied to the front surface of the right upper arm by pulling it from the inside to the outside (Fig. 2e). As a result, shear force was applied to the posterior skin of the upper arm, and pressure was applied to the frontal tissues during flexion.
3. Left arm: No stimulation.
4. Right arm: A piece of elastic tape described above was applied to the front surface of the upper arm without pulling. This condition was used as a control for condition 2 (i.e., neither shear force on the skin nor pressure stimulus was applied).

SEMGs were recorded from the biceps brachii and the lateral head of the triceps brachii. The patient was instructed to perform flexion and extension of the elbow in each condition. In condition 2, the findings of SEMGs resembled those of condition 3. However, in condition 4, the findings of SEMGs resembled those of condition 1 (Fig. 3e-h).

The differences observed among conditions 1, 2, and 3 in terms of muscle activity were analyzed quantitatively. The Friedman rank sum test and post hoc Wilcoxon signed-rank exact test were used to compare RMSs among the three conditions. The level of significance was set at p<0.05.

The results revealed differences in the RMS among the different conditions for the biceps brachii (p<0.0001) and triceps brachii (p<0.0001). The RMS for the biceps brachii was higher in condition 1 than in either condition 2 (p<0.0001) or 3 (p<0.0001). However, no marked difference was observed between the RMSs in conditions 2 and 3 (p>0.648). The RMS in the triceps brachii was higher in condi-
Abnormal motion during palmar flexion (a) and interventions for the upper limb (b-e). a: Hyperextension occurred in the proximal and distal interphalangeal joints of the second to fifth fingers, although the wrist and metacarpophalangeal joints were also flexed. The numbers are in chronological order. b: A piece of plastic tape was applied on his extended elbow starting from the posterior surface of the upper arm to the proximal posterior surface of the forearm. c: A piece of plastic tape was applied from the front of the upper arm to the proximal part of the forearm. d: A blood pressure sleeve was wrapped around the patient’s upper arm. e: A piece of elastic tape was applied to the front surface of the right upper arm by pulling it from the inside to the outside.

Since the increase in skin shear force or pressure stimulation during flexion and extension seemed to improve the symptoms, we considered the possibility that these sensations were reduced in the patient. Therefore, we performed a detailed evaluation of the somatic sensation of both the upper limbs in seven areas (anterior and posterior surfaces of the upper arm and forearm, palm, dorsum of the hand, and tip of the middle finger) (Table 2). The results were normal, and there were no evident left-right differences in the thermoceptual thresholds, tactile threshold as measured by the Semmes-Weinstein monofilament test (9), two-point discrimination, or tactile localization. The threshold of pain was higher on the right side than on the left side, but both were within the normal range. In addition, tactile extinction was not observed.

However, the patient had agraphesthesia on the posterior and anterior surfaces of the right upper arm and on the posterior surface of the right forearm. The sensation of the skin shear force was assessed as follows: a 5-cm-long and 2-cm-wide piece of plastic tape was applied to six areas (anterior and posterior surfaces of the upper arm and forearm, palm, and dorsum of the hand) and pulled to the long side. The threshold felt as the skin was pulled was measured. The results showed that the threshold apparently increased in the posterior surface of the right upper arm when the tape was pulled in the distal, proximal, lateral, or medial directions and in the posterior surface of the forearm when the tape was pulled in the distal direction. No abnormal values were observed in other areas.

To assess pressure sensing, a 1.5-cm-diameter disk was placed on the areas, and forces perpendicular to the skin surface were applied. The threshold at which the patient felt pressure was determined. The results demonstrated that the threshold apparently increased in the posterior and anterior surfaces of the upper arm, posterior surface of the forearm, palm, dorsum of the hand, and middle fingertip of the right limb.

The joint position sense was normal in the joints of the shoulder, elbow, wrist, and metacarpophalangeal and distal interphalangeal joints of the middle finger. The vibratory sensation was normal with no right-left differences in the
acromion process, medial epicondyle of the humerus, styloid process of the ulna, or distal interphalangeal joint of the middle finger. The identification of geometric shapes, materials, and everyday items with either hand was normal.

In summary, there were evident abnormalities in graphesthesias, perception of shear force on the skin, and pressure sensation only in part of the right upper limb. Furthermore, no other abnormal findings were observed.

Discussion

Our patient’s symptoms of MDs were localized to the right upper limb. The symptoms appeared only when the patient performed flexion and extension of the elbow, wrist, or finger joints. During this movement, strong and persistent contractions of both the agonistic and antagonistic muscles were observed, and the SEMGs displayed such contraction
Figure 4. Differences in the root mean squares (RMSs) of the amplitudes on surface electromyograms for the three conditions during flexion and extension of the elbow joint. Box and whisker plots show the distribution of RMS amplitudes. The median is represented as a line located in the middle of each box. The top and bottom of each box represent the 75th and 25th percentiles, respectively. The ends of the whiskers represent the maximum and minimum values. a: Distribution of RMSs on the surface electromyogram recorded for the biceps brachii. b: Distribution of RMSs on the surface electromyogram of the lateral head of the triceps brachii.

patterns. During palmar flexion, the interphalangeal joint exhibited hyperextension, which is an abnormal posture. This indicated that unintended contractions developed in the muscles other than the antagonist. The MDs observed in this patient were thus consistent with the definition of dystonia (1) and its important features (2) in many ways, although twisting or sustained posture were not observed.

An examination of the somatic sensations revealed evident abnormalities only in graphesthesia, sensation of shear force on the skin, and pressure sensation. In the graphesthesia test, the examiner writes a number on the skin using a dull and thin stick. Therefore, shear force is applied to the skin during the test. In the Semmes-Weinstein monofilament test, to assess the sense of touch, sustained pressure is applied to one point on the skin with a thin elastic stick without movement (9). Since the results of this test were unremarkable in the present patient, it was suggested that his pressure sensation on the skin was normal. However, evidently abnormal results were obtained when we examined the pressure sensation with a disk, which affected a wider area. This dissociation may be explained by a loss of pressure sensation in the deep tissues, such as muscles, despite its preservation in the skin. Based on these results, we believe that the abnormalities of the patient’s somatic sensation were restricted to shear force on the skin and pressure to the deep tissues. To our knowledge, this is the first report to reveal an association between partial injury of the VPC in the thalamus and these sensory disorders.

Many types of somatosensory information travel through the VPC contralateral to the stimulated body and then to the primary somatosensory cortex (PSC) ipsilateral to the VPC. PSCs are divided into areas 3a, 3b, 1, and 2, and the types of somatosensory information processed in each area are different. VPC is also divided into several areas, namely the lateral, medial, superior, and inferior regions, and each region has a connection with each area of the PSC with different magnitudes (6) (Fig. 5a). Therefore, the type of somatosensory impairment can vary depending on the location of damage in the VPC. In addition, each part of the VPC has somatotopy as well as PSC (6). Thus far, no study has described how shear force or deep pressure sensations are transmitted within the central nervous system; however, it is presumed that they project to PSC via VPC, as with other somatosensory systems. There is a specific area within the VPC that conveys these sensations, and only the part of the somatotopy of the upper limb representations in that area is damaged; this may explain why our patient had impairments of shear force and deep pressure sensations that occurred only in the upper limb contralateral to the lesion.

Fig. 5b describes the role of sensory information in the pathophysiology of FD as explained by Conte et al. (10). If there is a problem with the somatosensory input (indicated by the upward arrow) or the subcortical structures that process the input, such as the thalamus, cerebellum, and superior colliculus, the somatosensory information transmitted to the PSC would be wrong. Thus, the modulation of the activity of the primary motor cortex and basal ganglia cannot be accurately performed, leading to an abnormality in the motor output and thereby resulting in FD. Fig. 5c shows our hypothesis of the patient’s symptoms based on the model by Conte et al. FD in the patient presumably occurred since the shear force and deep pressure sensations cannot be accu-
Table 2. Results of Somatosensory Examinations.

<table>
<thead>
<tr>
<th></th>
<th>Right upper limb</th>
<th>Left upper limb</th>
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<td>AUA</td>
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<tr>
<td><strong>Temperature</strong></td>
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<tr>
<td>Threshold (°C)†</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Warmth</td>
<td>37</td>
<td>38</td>
</tr>
<tr>
<td>Cold</td>
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<tr>
<td>Noxious hot</td>
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<td>45</td>
</tr>
<tr>
<td>Noxious cold</td>
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<td><strong>Tactile</strong></td>
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<tr>
<td>Pressure on skin</td>
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<tr>
<td>(threshold)‡</td>
<td>3.61</td>
<td>3.61</td>
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<tr>
<td>Two-point discrimination (mm)§</td>
<td>6</td>
<td>9</td>
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<tr>
<td>Tactile localization (distance, [mm])¶</td>
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<td>15</td>
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<tr>
<td>Pain (threshold, load on pin [g]; range: 1–20)†</td>
<td>6</td>
<td>6</td>
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<tr>
<td><strong>Tactile extinction</strong></td>
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<td>-</td>
</tr>
<tr>
<td><strong>Graphesthesia</strong></td>
<td></td>
<td></td>
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<tr>
<td>(numbers from 0 to 9; max: 10)¶</td>
<td>2</td>
<td>2</td>
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<tr>
<td><strong>Shear force on skin</strong> (threshold, [N/cm²])</td>
<td></td>
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<tr>
<td>Distal direction</td>
<td>0.20</td>
<td>1.80</td>
</tr>
<tr>
<td>Proximal direction</td>
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<tr>
<td>Lateral direction</td>
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<td>1.20</td>
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<tr>
<td>Medial direction</td>
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<td>0.80</td>
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<tr>
<td>Deep pressure sensation (threshold, [N/cm²])</td>
<td>3.40</td>
<td>2.82</td>
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<tr>
<td><strong>Joint position sense</strong></td>
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<td>(number of correct answers; max: 10)a</td>
<td>10</td>
<td>10</td>
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<tr>
<td>Vibratory sense</td>
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<tr>
<td>(left-right differences)</td>
<td>-</td>
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<tr>
<td>Identification of geometric shapes (number of correct answers; max: 6)b</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Material identification (max: 6)c</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Identification of everyday items (number of correct answers, max: 6)d</td>
<td>6</td>
<td>6</td>
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Abnormal values have been indicated in bold.


†Temperature changed at ~0.1 °C/sec. ‡Semmes–Weinstein Monofilament (range: 1.65–6.65). §The patient closed his eyes, and the examiner touched various points on his right or left upper limb with a pencil tip for approximately 0.5 seconds. Then, the patient was asked to open his eyes and indicate the points by touching the same locations using a finger of the other arm immediately. ¶The examiner wrote a digit using a thin dull stick. The patient was asked to answer which digit was written. aJoints were moved through approximately 50% and 10% of the joint range of motion on the left and right side, respectively. bFrom a group of shapes consisting of a sphere, circular cone, cylinder, cube, triangular prism, and hexagonal column, the patient received one shape to handle in a location shielded from the patient’s view. Subsequently, all the shapes were shown to the patient and the patient was asked which shape he had handled. cVelvet, sponge, vinyl, wood, sandpaper, and rough cloth were used. dBall-point pen, scissors, spoon, clothes peg, golf ball, and stapler were used.

FD-like symptoms observed during flexion and extension of the elbow improved in the present patient when a shear...
Figure 5. Schematic illustration of the normal sensorimotor processing and alterations in focal dystonia. a: Projection from the ventral posterior complex of the thalamus to the primary somatosensory cortex [Padberg et al. (2009) (6)]. Light gray area indicates the ventral posterior complex (VPC) of thalamus. Projections from the ventral posterior lateral nucleus to areas 3a, 3b, 1, and 2 in the primary somatosensory cortex (PSC) are indicated with gray arrows, and projections from the ventral posterior superior nucleus to those areas are indicated with black arrows. The thickness of the arrows reflects the magnitude of the connections. L: ventral posterior lateral nucleus. I: ventral inferior lateral nucleus. S: ventral posterior superior nucleus. M: ventral posterior medial nucleus. b: Overview of the normal sensorimotor processing [Conte et al. (2019) (10)]. 1: Somatosensory input, such as temperature, pain, tactile, vibratory, and joint position sensations. 2: Unconscious proprioceptive input. c: Possible overview of the abnormal sensorimotor processing in the focal dystonia of the patient. 1: Somatosensory input, such as temperature, pain, tactile, vibratory, joint position sensations. 2: Unconscious proprioceptive input. 3: Hypothetical inputs of shear force and deep pressure. Light-gray area: VPC of thalamus. Black ellipse: lesion of the patient
force stimulus was applied to the skin covering the posterior surface of the upper arm (i.e. over the triceps brachii). When a finger receives a force that mimics the shear force applied to the skin when the fingers are moved, the sensation of finger movement is elicited (11, 12). Consequently, it was considered that not only the information from the muscle spindles but also the information on the skin shear force is important for regulating muscle contractions (11). Our patient experienced a decrease in the shear force sensation in the skin of the posterior surface of the upper arm, which may have resulted from inadequate transmission of information on the contractions of the triceps brachii to the central nervous system. Using our method, we ensured that shear force was applied to the skin during contractions of the triceps brachii. Such an application may compensate for the inadequate transmission of information, which may enable the central nervous system to produce appropriate inhibition.

FD-like symptoms during flexion and extension of the elbow improved following pressure stimulus application to an area where the pressure sensations in the deep tissues deteriorated, specifically around the upper arm or the anterior surface of the upper arm. The patient voluntarily used a band, the pressure of which was strong but stable. However, the use of weak stimulation that varied according to the contraction/relaxation of the muscles was effective for the patient. The stimulation was provided by a blood pressure sleeve with pressure settings of 20-30 mmHg and a piece of elastic tape that was placed on the biceps brachii with tension. Recent studies have demonstrated the existence of fibers that transmit nonpainful pressure sensations from the deep tissue (13, 14). Pressure to the muscles from other tissues, such as the skin, increases during muscle contraction. Such increased pressure information is also transmitted to the central nervous system of the fibers and may be used to inhibit excessive contraction of the agonist muscles and contraction of the antagonist muscles. Our patient’s FD-like symptoms improved following the application of a pressure stimulus that was increased in a manner proportional to muscle contraction. This may represent the compensation for inadequate information, and such compensation may enable the central nervous system to produce appropriate inhibition.

Several limitations associated with the present study warrant mention. First, the somatosensory test is dependent on the patient’s subjectivity, and electrophysiological evidence is not available to support this result. Further studies are required to confirm whether or not abnormal results are observed in several types of electrophysiological tests, such as ordinary somatosensory-evoked and pinprick-evoked brain potentials (15), and in the amplitude changes observed in 10-Hz electroencephalographic oscillations (16). Second, to confirm a decrease in the responses to these stimuli, a method needs to be developed to detect brain potential elicited by shear force on the skin and pressure stimulation of deep tissues. In addition, our results were obtained from a single patient. Whether or not FD or FD-like symptoms resulting from localized brain injury are generally associated with abnormal somatosensory abnormalities remains unknown. Further studies should include more patients with lesions in various areas with such MDs.

The contribution of sensorimotor integration to the pathophysiological background of idiopathic FD has been discussed repeatedly (3). However, sensory examinations have revealed abnormalities only in the tactile temporal discrimination (17), tactile spatial discrimination (18), and joint position sense (19). Such abnormal findings are not severe; thus, none of the abnormalities are sufficiently strong to explain FD. For idiopathic FD, assessing skin shear force sensation and deep tissue pressure sensation and performing the same intervention may be meaningful if abnormalities are observed, as in the present case.

The authors state that they have no Conflict of Interest (COI).

Ethics statement
The patient provided his written informed consent after receiving a detailed description of the study. This study was approved by the Ethical Committee of Yamagata Prefectural University of Health Science and conducted in accordance with the Declaration of Helsinki.

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References

11. Edin BB, Johansson N. Skin strain patterns provide kinaesthetic information to the human central nervous system. J Physiol 487:


