Pure somaesthetic alexia: somaesthetic–verbal disconnection for letters

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Summary
We studied a patient who manifested a bilateral reading disorder through the somaesthetic modality, without deficit of elementary tactile sensation or tactile object naming, due to a left parietal infarct. Detailed investigation established the following points. (i) The patient showed normal function on elementary somaesthetic examination, normal function on high level tactile perception, except for minimal impairment of the right hand on the two-point discrimination test, and normal latencies on the somatosensory evoked potential in both hands. (ii) The patient had difficulty in reading letters using any somaesthetic strategy (graphaesthesia, directional joint kinaesthesia and active touch) with either hand. (iii) On a same–different judgement task, the patient’s performance with the right hand was slightly defective on graphaesthesia and active touch, but performance with the left hand was within the normal range for all of the strategies. The patient’s disorder was highly category specific and modality specific, indicating that somaesthetic letter reading can be disrupted not only independent of other high level somaesthetic functions, like object recognition, but also independent of other modes of reading functions, such as visual reading. A lesion involving the intraparietal sulcus, the upper part of the inferior parietal lobule and the adjacent white matter in the left hemisphere may be capable of compromising the pathways for somaesthetic letter reading with both hands.

Keywords: somaesthetic alexia; agraphaesthesia; tactile alexia; tactile aphasia; tactile agnosia

Introduction
Cognitive disorders of stimulus identification that cannot be attributed to elementary sensory deficits, mental deterioration or aphasia have been classified into two major groups. Agnosia is a state of impaired recognition of stimuli presented from a particular sensory mode, which is further divided into apperceptive and associative type. Modality-specific aphasia, e.g. optic aphasia (Freund, 1889; Lhermitte and Beauvois, 1973) and tactile aphasia (Beauvois et al., 1978; Endo et al., 1992), is a state of impaired ability to describe and name objects verbally, despite normal recognition of stimuli. We can also classify these pathological states in terms of material specificity. For instance, in auditory modality, we use the term sound agnosia for a state of impaired recognition of environmental sounds (Spreen et al., 1965; Fujii et al., 1990) and pure word deafness for a state of impaired recognition of spoken language (Hemphill and Stengel, 1940; Albert and Bear, 1974; Saffran et al., 1976). In the visual modality, we use the term prosopagnosia for a deficit of face recognition (Ellis, 1996) and pure alexia for a deficit of written language recognition (Damasio and Damasio, 1983). Regarding the somatosensory domain, however, it is not yet clear whether such a material-specific disorder occurs. To our knowledge, no such case has been reported, although many studies on high level somaesthetic disorders have appeared (Corkin et al., 1970; Roland, 1976; Beauvois et al., 1978; Feinberg et al., 1986; Endo et al., 1992; Caselli, 1993; Reed and Caselli, 1994; Endo et al., 1996; Kim and Choi-Kwon, 1996; Platz, 1996; Reed et al., 1996). The purpose of this article is to report a patient who showed a bilateral reading disorder through the somaesthetic modality without deficit of elementary tactile sensation or tactile object recognition.

Case report
The patient (N.N.) was a 64-year-old right-handed man with a 10th-grade education. He experienced a sudden onset of right-sided weakness and dysarthria on January 24, 1994, and was admitted to a local hospital. These symptoms disappeared in a few days. For further evaluation, he was transferred to the Neurosurgical Service of Sendai National Hospital on February 28, 1994. He had been treated for
long-standing insulin-dependent diabetes which was well controlled. He had never suffered from any central nervous system diseases. The patient gave informed consent to the following examinations.

**Neurological examination**

On admission, the patient was alert and oriented. Visual acuity and visual field on a confrontation test were normal. Both pupils were equal, round and reacted to light. Eye movement was full with normal pursuit and saccadic components. The remaining cranial nerves were also normal. He had no hemiparesis or ataxia. His grasping power was 37 kg in the right hand and 40 kg in the left. Muscle tone of the four limbs was normal. Muscle stretch reflexes were normal and no pathological reflexes were elicited. Sensation of light touch, pain, temperature, vibration and joint motion was normal.

**Neuroradiological and electrophysiological studies**

An MRI scan of the brain carried out on the 71st post-onset day revealed a small infarct in the postcentral gyrus and a more extensive infarct involving the intraparietal sulcus, the upper part of the inferior parietal lobule, and the adjacent white matter, both in the left hemisphere (Figs 1 and 2). No additional lesions were visualized.

The EEG on the 57th post-onset day was normal. A somatosensory evoked potential study performed on the 64th post-onset day showed normal latencies following median nerve stimulation in both hands (right stimulation: Erb’s point = 10.4 ms, N13 = 13.8 ms, N20 = 20.1 ms; left stimulation: Erb’s point = 10.0 ms, N13 = 13.6 ms, N20 = 20.3 ms).

**Neuropsychological examination**

Neuropsychological examinations were carried out from the 36th to 78th post-onset day. Results were as follows. The patient’s digit span was six forward. Language tests showed no abnormality in his spontaneous speech, auditory comprehension, naming or repetition of sentences. He had no difficulty in reading, but showed mild difficulty in writing with both kanji (Japanese ideogram) and kana (Japanese syllabogram) scripts on the 38th post-onset day, such difficulty not being observed in a later examination. He was evaluated with the Japanese Standard Language Test of Aphasia on the 52nd post-onset day. Auditory comprehension, repetition and visual naming of 20 pictures were perfectly performed. He generated 12 names of animals within 1 min. Reading aloud, reading comprehension, writing to dictation, and writing the names of visually presented stimuli (kanji, kana, and sentences consisting of mixed kanji and kana) were all 100% correct. With single letters of kana, reading aloud, pointing to kana letters from dictation and writing to dictation were perfect (10/10). Examination of graphaesthesia showed that reading of kana letters and Arabic numerals written on either of the patient’s palms was seriously disturbed. In contrast, naming of objects through the tactile modality was perfect in both hands (10/10). He was easily able to name objects from familiar tape-recorded sounds (4/4). Buccofacial and
limb praxis in response to command and in imitation were all performed without difficulty. No clumsiness in manipulating objects or turning pages of a book was observed in either hand (palpatory apraxia; see Yamadori, 1982). Unilateral spatial neglect and sensory extinction to double simultaneous stimuli (visual, tactile and auditory) were absent. He showed mild optic ataxia only when asked to reach a target in the left visual field with the right hand. Right–left orientation and finger recognition were normal. He showed mild constructional disability on the task of copying a cube and difficulty in multiplication and division tasks when two-digit numbers were involved. On the Wechsler Adult Intelligence Scale—Revised performed on the 78th post-onset day, the patient obtained a full IQ of 102, with a verbal IQ of 113 and a performance IQ of 86. He obtained an IQ of 80 on the Kohs Block Test.

To sum up, 5–10 weeks after the onset of right-sided weakness and dysarthria, the patient exhibited no basic neurological deficit, a slight constructional disability, a mild calculation deficit, mild visuomotor ataxia with the right hand in the left visual field, and severe agraphaesthesia in both hands.

**Detailed neuropsychological assessment of agraphaesthesia**

To determine the underlying mechanisms of his bilateral agraphaesthesia which seemed to be unusually isolated, we carried out some additional tests to answer the following three questions. (i) Was his agraphaesthesia really isolated from other tactile impairments? (ii) Did his tactile reading difficulty extend beyond graphaesthesia? (iii) At which level was it impaired, i.e. was it in the somaesthetic domain or in its relation to other cognitive processes?

**Was his agraphaesthesia really isolated?**

We examined his discriminative tactile functions in detail, although bedside neurological examination revealed no gross somatosensory deficit in either hand.

Two-point discrimination was measured on the tip of the second finger and the palm of each hand with an aesthesiometer. Texture discrimination was evaluated with standardized pieces of sandpaper of five different grades (Japan Industrial Standard: 60, 120, 240, 600 and 1000). The patient’s task was to judge whether a pair of stimuli given in succession was same or different in roughness. The 15 pairs included five pairs of identical stimuli and 10 pairs of different stimuli. Size discrimination was evaluated by his ability to differentiate the difference in size of two balls. Four balls differing only in diameter (20, 25, 30 and 35 mm) were prepared. The patient judged whether a pair of balls placed successively in his hand were the same or different in size. Balls were given at ~1-s intervals. Of the 10 pairs presented, four pairs were identical and six pairs were different. Ability of point localization was tested using a drawing of the hand (Fig. 3A). The examiner touched a point on his hand, and the patient was requested either to report the corresponding number in the figure or to point at the corresponding place in the drawing with the opposite hand. Directional cutaneous kinaesthesia (Bender et al., 1982) was tested in two different ways. First, the examiner drew a line on the patient’s palm and asked him either to report the number corresponding to the line or to point to the corresponding line.
Fig. 3 (A) Drawing of the hand used in the point localization task. (B) Drawing of the hand used in the directional cutaneous kinaesthesia task.

Table 1: Results of somaesthetic tests

<table>
<thead>
<tr>
<th></th>
<th>Right hand</th>
<th>Left hand</th>
</tr>
</thead>
<tbody>
<tr>
<td>Light touch</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Pain</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Temperature</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Vibratory sense</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Joint sense</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Extinction</td>
<td>Absent</td>
<td>Absent</td>
</tr>
<tr>
<td>Two-point discrimination (mm)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Second finger</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>Palm of hand</td>
<td>&lt;20</td>
<td>&lt;20</td>
</tr>
<tr>
<td>Texture discrimination</td>
<td>15/15</td>
<td>12/15</td>
</tr>
<tr>
<td>Size discrimination</td>
<td>10/10</td>
<td>10/10</td>
</tr>
<tr>
<td>Point localization</td>
<td>9/10</td>
<td>10/10</td>
</tr>
<tr>
<td>Directional cutaneous kinaesthesia</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pointing to visually presented arrow</td>
<td>7/8</td>
<td>7/8</td>
</tr>
<tr>
<td>Matching of two successive trials</td>
<td>16/16</td>
<td>16/16</td>
</tr>
<tr>
<td>Tactile object naming</td>
<td>10/10</td>
<td>10/10</td>
</tr>
</tbody>
</table>

of the drawing (Fig. 3B) with the opposite hand. Secondly, he was required to make a same–different judgement for two successive trials.

Results of somaesthetic examinations, including the tests described above, are summarized in Table 1. On the two-point discrimination test, the patient showed mild laterality, indicating mild discriminatory deficit in the right hand. On the remaining tests, his performance was within normal range.

Did his tactile reading difficulty extend beyond graphaesthesia?

The previous tests clearly showed that the patient’s agraphesthesia could not be traced back to elementary sensory disorders; he showed almost exclusive difficulty in recognizing letters among various stimuli presented tactually. We next attempted to determine whether his difficulty in reading letters and numbers, manifesting as agraphesthesia, was really limited to skin writing, or whether reading difficulties were similarly present when other somaesthetic strategies were used. One can obtain information on letter shape through detection of the direction of joint movements (Schreibendes Lesen or directional joint kinaesthesia) (Bender et al., 1982). Another source of information is voluntary manual exploration of the raised solid shape of a stimulus (active touch) (Critchley, 1953; Gibson, 1962). Thus, we compared the patient’s ability to read three types of letters (kana, kanji and Arabic number) through the three strategies. We also collected control data from eight age-matched subjects (ages ranging from 60 to 68 years).

For graphaesthesia, a symbol was drawn on the palm 5 × 5 cm in size with a dull-tipped pencil and with the speed of ~1–2 s per symbol. For directional joint kinaesthesia, we held the patient’s forefinger and outlined a symbol on a desktop by moving his finger. The symbol was ~7 × 7 cm in size and the speed with which it was outlined was the same as in graphaesthesia. In graphaesthesia and directional joint kinaesthesia, when his response was uncertain, one more trial was given. For active touch, we provided symbols 5 × 5 cm in size (raised 7 mm in height for kana and numbers, and 3 mm for kanji) mounted on a piece of cardboard 10 × 10 cm. The patient was asked to read the symbol by freely touching it. In this task no time constraint was imposed. In all of the tasks, stimuli were provided with normal orientation.

Table 2 summarizes the results. His performance on all three tasks was below the lowest control score, except for performance on directional joint kinaesthesia and active touch for numbers with the left hand. Regarding the hand employed, he responded more correctly with the left hand than with the right hand. The results demonstrate that the patient had difficulty in reading letters through all somaesthetic strategies...
Table 2 Results of somaesthetic reading (number of correct responses out of 10 trials)

<table>
<thead>
<tr>
<th></th>
<th>Kana</th>
<th>Kanji</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>R</td>
<td>L</td>
</tr>
<tr>
<td>Graphaesthesia</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patient N.N.</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>Control mean</td>
<td>9.3</td>
<td>10</td>
</tr>
<tr>
<td>Control range</td>
<td>8–10</td>
<td>10</td>
</tr>
<tr>
<td>Directional joint kinaesthesia</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patient N.N.</td>
<td>5</td>
<td>7</td>
</tr>
<tr>
<td>Control mean</td>
<td>9.5</td>
<td>9.8</td>
</tr>
<tr>
<td>Control range</td>
<td>8–10</td>
<td>9–10</td>
</tr>
<tr>
<td>Active touch</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patient N.N.</td>
<td>2</td>
<td>5</td>
</tr>
<tr>
<td>Control mean</td>
<td>8.7</td>
<td>8.7</td>
</tr>
<tr>
<td>Control range</td>
<td>7–10</td>
<td>6–10</td>
</tr>
</tbody>
</table>

R = right hand; L = left hand.

The level of impairment

It became clear that the patient had selective difficulty, in both hands, in reading linguistic symbols confined within the somatokinaesthetic domain. The next question was whether this difficulty in symbol identification was at the perceptual level or at the associative level. To test this, we presented a pair of shapes and asked the patient to judge whether they were the same or different. In addition to the linguistic materials, we prepared 20 pairs of nonsense figures (5 × 5 cm in size with 3 mm in height) consisting of 10 pairs of identical shapes and 10 pairs of different shapes. The stimuli (pairs of shapes) were again presented at intervals of ~1 s through three methods (graphaesthesia, directional joint kinaesthesia, and active touch). The order of presentation of pairs was randomized. For graphaesthesia and directional joint kinaesthesia, when the patient’s response was uncertain, one more trial was given. In active touch, we imposed no time limits for each stimulus, but prohibited him from exploring the first stimulus after exploring the second stimulus. We collected control data for nonsense figures from five subjects who had participated in somaesthetic reading tests.

Table 3 summarizes the results. In the performance for the nonsense figures, the patient’s judgement, when he used his right hand, was defective on graphaesthesia and active touch but not on directional joint kinaesthesia. His performance with the left hand was within normal range for all of the strategies. His performance for kana, kanji, and numbers showed a tendency similar to that for nonsense figures except for kanji on graphaesthesia.

Discussion

Our patient showed difficulty in naming letters but not in naming objects through tactile presentation. His difficulty in naming letters was demonstrated not only with graphaesthetic strategy but also with kinaesthetic and active touch strategies. In the visual domain, he had no difficulty in reading kana and kanji letters or Arabic numerals. Words and sentences were read flawlessly. For single letters of kana, reading aloud and pointing at kana letters from dictation were all correct, indicating that non-lexical as well as lexical visual–verbal and auditory–visual routes were preserved. Thus, the patient’s reading disorder could not be supramodal. He showed neither mental deterioration nor aphasia. Critchley (1953) used the term tactile alexia for a disturbance in which there is a loss of faculty of recognition, with the eyes shut, of letters or figures drawn upon the skin. The present patient’s deficits, however, were not confined to difficulty in recognizing tactually written letters. Therefore, pure somaesthetic (or somatokinaesthetic) alexia may be more proper to describe this patient’s disorder.

The patient showed normal function on elementary somaesthetic examination and showed normal latencies on the somatosensory evoked potential in both hands. We found asymmetry on the two-point discrimination task, indicating that he had a slight somaesthetic deficit in the right hand. However, taking his age into account, his performance with the right hand on the two-point discrimination task was within normal range according to data reported by other researchers (Stevens, 1992; Kim and Choi-Kwon, 1996). The results for somaesthetic matching tasks in skin writing and active manipulation also showed slight deficits in the right hand. These facts may explain part of the deficits of the right hand performance. However, this mild somaesthetic disorder observed in the right hand cannot account for the considerable and disproportional reading impairment of the right hand, and of course it cannot account for his reading impairment of the left hand.

The patient’s somaesthetic reading difficulty was observed through three different modes of stimulus presentation. It should be noted that he showed impaired reading of kana and kanji letters, even when he was using the directional joint kinaesthesia method, through which he could normally
match two successive figures, kana and kanji letters. This finding further supports the view that the patient’s disability in somaesthetic reading cannot be attributed to somaesthetic dysfunction itself. In other words, his disorder was not one of perception, but rather of association.

The results of the present study clearly show that a disorder in the somaesthetic naming of letters can be observed independently of a disorder in the tactile naming of objects. Bender et al. (1982) stated that astereognosia is strongly correlated with agraphaesthesia, but that examples of dissociation in both directions occur. Unfortunately, they did not provide concrete data to prove their point.

We speculate that there may be different intrahemispheric and interhemispheric pathways for letter naming and object naming. Figure 4 shows posited routes for somaesthetic reading and tactile object naming, and a probable locus, in the present patient, of impaired letter naming with both hands.

As was shown by the MRI scan, our patient had a small infarct in the left postcentral gyrus and a more extensive infarct in the intraparietal sulcus, the upper part of the left inferior parietal lobule and the adjacent subcortical region. The small infarct in the left postcentral gyrus may explain his very mild sensory deficit (in two-point discrimination) in the right hand, but is not satisfactory to explain the somaesthetic alexia of the bilateral hand. Bilateral somaesthetic alexia is probably a result of the posterior lesion which effectively prevented somaesthetic letter information from reaching the language area ipsilaterally and contralaterally. The damaged area (in the intraparietal sulcus and the upper part of the inferior parietal lobe and the adjacent subcortical region) might normally be involved in information transfer related to somaesthetic–graphemic decoding and might serve as a relay station for both hemispheres. Somaesthetic information about the letters received by the left postcentral gyrus would be sent to the adjacent somatosensory association cortices for further discriminative analysis without identifying the letters by name. It would then be sent to the left somatosensory association cortices through the corpus callosum (there are interhemispheric connections between homologue areas in the two hemispheres). For somaesthetic reading, this information would be sent to the left angular gyrus. Disconnection of the left somatosensory association cortices from the left angular gyrus could explain the patient’s impairment in somaesthetic reading with his left hand. As for object naming with the left hand, a different pathway might have been available, e.g. the pathway from the anterior part of the inferior parietal lobule of the right hemisphere, including secondary somatosensory areas, to the homologous area in the left hemisphere, and then, to the intact language area.

This anatomical explanation of dissociation between verbal
Pure somaesthetic alexia

Fig. 4 Posited routes for somaesthetic reading, tactile object naming, and disordered route in the present patient. ‘XX’ marks the pathway which could be blocked to cause somaesthetic alexia in both hands. The cortical areas involved are: (1) primary sensory area; (2) somatosensory association area; and (3), (4) and (5) other posterior association cortices.

letter identification and object identification necessarily remains speculative, but the difference of quality of the two types of stimuli, i.e. one requires essentially two-dimensional analysis and the other three-dimensional one, may account for the processing difference at the later stage of somatosensory analysis.

Regarding pure (visual) alexia, Damasio and Damasio (1983) demonstrated, on the basis of anatomical–functional study, that the crucial anatomic correlate was a lesion of the paraventricular white matter of the left occipital lobe. Compared with their data, our patient’s extensive lesion was situated higher and more rostrally. Hence it is quite probable that our patient’s lesion compromised the pathway for reading letters in the somatosensory domain but not in the visual domain.

Finally, a note of caution is required as to the bilaterality of the sensory disturbance after unilateral brain damage.

There have been several studies demonstrating a bilateral sensory deficit, especially a discriminative sensory one, after occurrence of a unilateral cerebral lesion (Carmon and Benton, 1969; Corkin et al., 1970; Carmon, 1971; Fontenot and Benton, 1971; Caselli, 1991b; Kim and Choi-Kwon, 1996). However, interpretations must be cautious if the response mode is verbal, because the deficits may not reflect an assumed discriminative sensory dysfunction but merely a tactile–verbal disconnection, as shown in our patient with bilateral somatosensory alexia and in patients with bilateral tactile aphasia or tactile anomia.

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References


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