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CASE STUDY

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Facial sensory disturbance associated with cervical compressive myelopathy

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General Note
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ABSTRACT

Cervical myelopathies do not usually cause facial sensory disturbance. We encountered, however, 2 patients developed facial sensory disturbance associated with cervical compressive myelopathy. We compared neurological findings and MRI findings of these patients. Patient 1 gradually developed tingling sensations of his left hand, difficulty in walking, urinary frequency, urinary incontinence, difficulty in urination, sensory disturbance of the left hand, and sweating abnormality. Patient 1 exhibited hyperalgesia of his left face, weakness of the left intrinsic hand muscle, clumsiness of the bilateral hands, hyporeflexia of the bilateral upper extremities, hyperreflexia of the left lower extremity, sensory level at C7, and autonomic disturbance. MRI revealed C5-6 intervertebral level myelopathy. Patient 2 gradually developed tingling sensations of her left occipital region, left hand, left sole of

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foot, and left side of the mouth. Patient 2 exhibited lively deep tendon reflexes, and slightly decreased vibration senses. MRI revealed C6-7 intervertebral level myelopathy. The facial sensory disturbance of these patients seems to be associated with cervical compressive myelopathy. The spinal trigeminal nucleus projects to C5-C8 levels in the rat. Furthermore, cervical spinal cord projects to the spinal trigeminal nucleus even from C7 to T1 in the rat. The facial sensory disturbance of these patients may be associated with stimulative lesions of projections from the spinal trigeminal nuclei to the cervical spinal cords or projections from the cervical spinal cords to the spinal trigeminal nuclei.

Keywords: Cervical myelopathy, Facial sensory disturbance, Projection, Spinal trigeminal nucleus

Abbreviations: Sp5O -the spinal trigeminal subnucleus oralis.

1. INTRODUCTION

Previously, cervical myleopathy patients present with symptoms not in faces but in extremities and trunks. Only the impairment of vibration and temperature sensibility in the trigeminal skin area in patients with chronic symptoms after soft-tissue injury of the cervical spine is reported (Knibestöl et al., 1990). Upper cervical myelopathy patients presented with cranial nerve symptoms such as facial sensory disturbance are also reported (Chang, 1995; Matsubara et al., 1990). The spinal trigeminal nucleus plays a crucial role in craniofacial nociceptive transmission in human (Sessle, 2000). Only the caudal part of the nucleus is concerned with the conduction of pain and temperature, whereas the oral part must have other functions (Olzewski, 1950).

The localization and function of the spinal trigeminal nucleus and tract are studied, and it is localized to C2 or C3 (Chudler et al., 1991; Honda et al., 2008; Nakae et al., 2008; Noma et al., 2008; Shibuta et al., 2012; Shigenaga et al, 1986; Shigenaga et al., 1988; Suzuki et al, 2008). However, projections from the spinal trigeminal subnucleus oralis to the spinal cord distribute at C5-C8 levels in the rat (Devoize et al., 2010). Furthermore, cervical spinal afferent fibers even from C7 to T1 project to the spinal trigeminal complex in the medulla oblongata in the rat (Phelan et al., 1991). Small component projections via the dorsal funiculus ascend to terminate in the trigeminal nuclear complex in amphibians (Muňoz et al., 1997).

We encountered 2 patients presented with facial sensory disturbance associated with cervical compressive myelopathy. We describe clinical characteristics and discuss the association between facial sensory disturbance and cervical compressive myelopathy.

2. MATERIALS AND METHODS

Patient 1

A 58-year-old man came to the hospital because of tingling sensations of his left hand and difficulty in walking. He was well until approximately 2 years earlier, when he developed unsteadiness and impotence. When he tossed and turned his head, he felt dizziness and floating sensations. Approximately 1 year earlier, he developed unsteadiness twice. He was diagnosed as cervical myelopathy. The doctor recommended undergoing operation. However, he did not undergo operation. Three months earlier, urinary frequency and incontinence and difficulty in urination developed. He felt tingling sensations of his left hand and difficulty in walking because his left leg was crippled, and he stumbled. He felt dizziness when he tossed and turned his head leftward in the bed. When his left hand touched hot water, he was unable to feel how hot the water was, and 2 seconds later, a burning sensation spread to his left shoulder. This burning sensation continued after his hand put into cold water. He was unable to identify button holes by his left hand. He sweated in the head but did not sweat in the body.

At 8 years of age, he admitted to the hospital because of a renal disease. At 13 years of age, he underwent an operation of appendicitis.

His younger brother developed a liver disease.

He had neither drunk nor smoked. He had dealt with real estate.

Neurological examination exhibited as follows: weakness of the left abductor pollicis brevis and abductor digiti minimi; clumsiness of the bilateral hands at rapid alternative movement; areflexia or hyporeflexia of the bilateral biceps and brachioradialis (finger jerks at the left biceps and brachioradialis reflexes), and hyperreflexia of left lower extremity; hyperalgesia of left side of the face (Figure 1a) and left side of the body, hyperesthesia of left side of the body; sensory level at C7 (Figure 1b); and left side dominant vibration disturbance; and autonomic disturbance: urinary disturbance, impotence, ahidorosis of the body and

FINGER JERKS: Finger jerks are jerky flex movement of fingers when we examine biceps and/or brachioradial reflexes. Usually they are observed when biceps and/or brachioradial reflex increased, and they are interpreted as a sign of increased tendon reflex. However, finger jerks are observed with absent biceps and/or brachioradial reflex. They are interpreted as a segmental sign of C6 gray matter and represented C6 spinal cord segment lesions. When finger jerks associated with absent brachioradial reflex, they have been called "inverted brachioradial reflex". Therefore, finger jerks sometimes used as a diagnostic marker of C6 segment myelopathy as well as phenomenon associated with increased deep tendon reflexes which represent corticospinal tract lesions.

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Hyperhidrosis:
Hyperhidrosis means increased sweating. Myelopathy patients with ahidorosis sometimes show compensately hyperhidrosis above the levels of their meylopathy.

compensatory hyperhidrosis of the head.

Patient 2

A 64-year-old woman came to the hospital because of tingling sensations of left side of her head, hand, leg, and left side of her mouth. She was well until approximately 2 years earlier, when she developed a tingling sensation of the left occipital region. Recently, the tingling sensation became continuous and she felt it usually. One month earlier, she developed a tingling sensation of her left hand when she used her personal computer. A few weeks earlier, she developed a tingling sensation of her left sole of foot. One week earlier, she developed an abnormal sensation of left side of her mouth. The tingling sensations were spontaneous abnormal sensations. She stumbled on her left foot when she went upstairs.

Twenty years earlier, she developed a cervical intervertebral disc hernia. Conservative treatment improved her symptoms.

Her mother died of a cerebral thrombosis at 76 years of age. Her younger sister died of a subarachnoid haemorrhage at 46 years of age.

She had neither drunk nor smoked. She taught education in a university.

Neurological examination exhibited as follows: lively deep tendon reflexes particularly increased bilateral brachioradialis and triceps reflexes; and slight decreased vibration senses of the bilateral wrists.

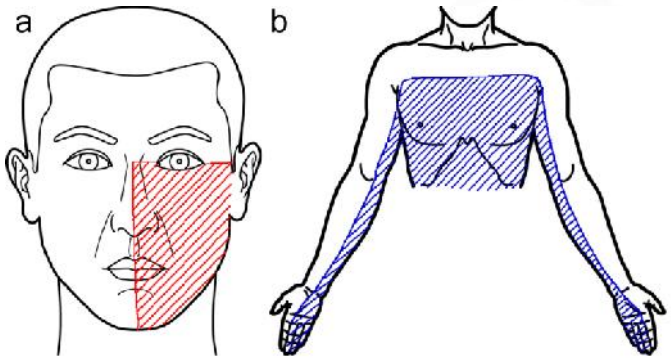

Figure 1 The distribution of the sensory disturbance examined by pinprick sensation in patient 1.
(a) The hyperalgesia in left V2 and V3 regions of the face.
(b) The C7 sensory level.

Methods

We examined these patients' magnetic resonance images (MRI) of cervical spinal cords and brains. We compared neurological findings and MRI findings. All participants or next-to-kin gave their informed consent to the study, which was equivalent to be approved by the institutional review board or ethics committee (Ethics Committee of Tokyo Metropolitan Ohtsuka Hospital, Tokyo, Japan).

Myelomalacia:
Myelomalacia is one of MRI findings of myelopathy. Myelomalacia represents T2 high intensity in the spinal cord and it represents pathological change such as tissue edema.

3. RESULTS

Cervical spinal cord MRI of patient 1 revealed C5-6 and mild C4-5 intervertebral level myelopathy with C5-6 level myelomalacia (Figures 2a, b, c). Brain MRI of patient 1 revealed normal findings without apparent infarct (Figure 2d). Patient 1 presented with hyperalgesia of left side of the face (Figure 1a).

Cervical spinal cord MRI of patient 2 revealed C6-7 intervertebral level myelopathy (Figures 3a, b). Brain MRI of patient 2 revealed normal findings without apparent infarct (Figure 3c). Patient 2 complained of abnormal sensation of left side around the mouth.

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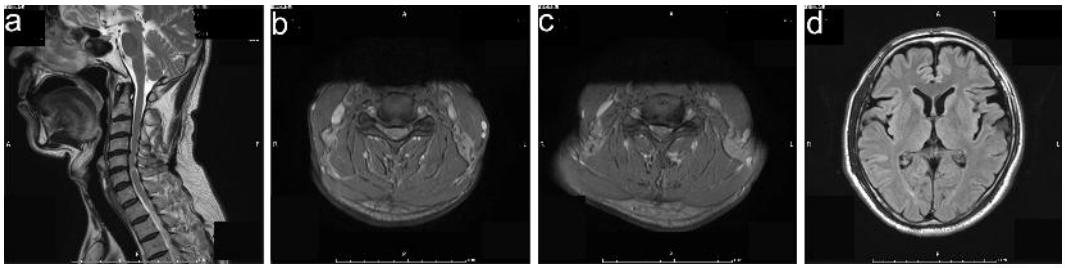

Figure 2 Magnetic resonance images (MRI) of patient 1
(a) T2 weighted sagittal section of the cervical spinal cord. Compressive myelopathy at C5-6 intervertebral level and mild compressive myelopathy at C4-5 intervertebral level. The spinal cord at C5-6 intervertebral level showing myelomalacia.
(b) T2 weighted axial section of the cervical spinal cord at C4-5 intervertebral level. The spinal cord showing deformity and high intensity.
(c) T2 weighted axial section of the cervical spinal cord at C5-6 intervertebral level. The spinal cord showing deformity and high intensity.
(d) Fluid attenuated inversion recovery (FLAIR) cerebral MRI. We were unable to identify any abnormal finding such as cerebral infarct.

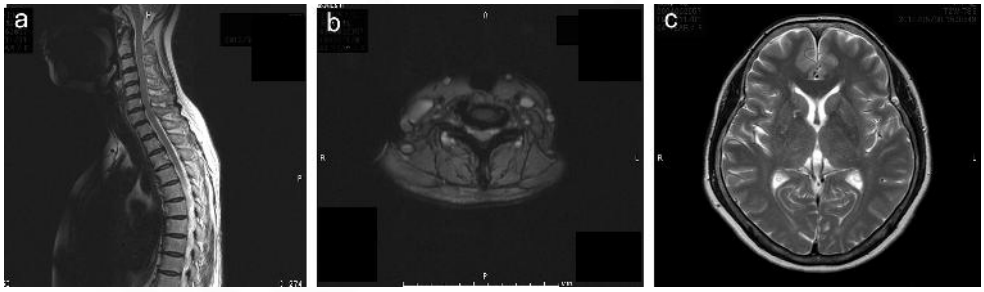

Figure 3 MRI of patient 2
(a) T2 weighted sagittal section of the cervical spinal cord. Compressive myelopathy at C6-7 intervertebral level.
(b) T2 weighted axial section of the cervical spinal cord at C6-7 intervertebral level. The spinal cord showing deformity.
(c) T2 weighted cerebral MRI. We were unable to identify any abnormal findings such as cerebral infarct.

4. DISCUSSION

This study described two patients presented with facial sensory disturbance associated with cervical compressive myelopathy. Patient 1 presented with hyperalgesia of the left face, weakness of the left intrinsic hand muscle, clumsiness at bilateral rapid alternative movements, hyporeflexia of bilateral biceps and brachioradialis, hyperreflexia of left lower extremity, sensory level at C7, and autonomic disturbance. MRI revealed C5-6 (and mild C4-5) intervertebral level myelopathy. Patient 2 presented with tingling sensations of her left head, hand, leg, and left side of the mouth, lively deep tendon reflexes particularly increased bilateral brachioradialis and triceps reflexes, and slightly decreased vibration senses of bilateral wrist. MRI revealed C6-7 intervertebral level myelopathy.

Previously, upper cervical myelopathy patients presented with facial sensory disturbance are reported (Chang, 1995; Matsubara et al., 1990). The burning sensation in the face associated with C1 myelopathy suggests some pathologic condition residing in the spinal trigeminal nucleus (Chang, 1995). Hypalgesia around mouth associated with C1-C2 tumor is reported (Matsubara et al., 1990). These findings are explained by the localization of spinal trigeminal nucleus and tract. The impairment of vibration and temperature sensibility in the trigeminal skin area in patients with chronic symptoms after soft-tissue injury of the cervical spine indicates damage to the central trigeminal system in upper spinal cord segments (Knibestöl et al., 1990). However, there may be additional cerebral

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injuries and the authors consider the brainstem and upper spinal cord as the probable site of injury. Therefore, excluding upper cervical cord lesions, this seems to be the first report of facial sensory disturbance associated with cervical compressive myelopathy.

The spinal trigeminal nucleus and tract have localized to C2 or C3 (Shigenaga et al., 1986; Shigenaga et al., 1988; Chudler et al., 1991; Honda et al., 2008; Nakae et al., 2008; Noma et al., 2008; Shibuta et al., 2012; Suzuki et al., 2008). However, dorsal part of the spinal trigeminal subnucleus oralis (Sp5O) projects to the dorsal horn at C1 level, to the dorso-medial motor nuclei at C3-C5 levels, whereas the ventral part of the Sp5O projects to the ventro-medial motor nuclei at C1-C4 levels, and to the dorso-medial motor nuclei at C5-C8 levels in the rat (Devoize et al., 2010). Furthermore, cervical spinal cord projects to spinal trigeminal complex even from C7 to T1 in the rat (Phelan et al., 1991). The anterograde transport of horseradish peroxidase (HRP) is used to examine the distribution and morphology of spinal afferent fibers terminating in the rat spinal trigeminal complex (Phelan et al., 1991). Small unilateral injections of HRP in the dorsal and ventral horns are centered at levels corresponding to C7-T1. A small number of anterogradely labeled fibers course within the ipsilateral spinocerebellar tract, insular trigeminal-cuneatus lateralis nucleus, and dorsolateral region of trigeminal nucleus interpolaris (Phelan et al., 1991). Small component projections via the dorsal funiculus ascend to terminate in the trigeminal nuclear complex in amphibians (Muňoz et al., 1997). Injury of projections from the spinal trigeminal nucleus to the cervical spinal cord or injury of projections from the cervical spinal cord to the spinal trigeminal nucleus may cause facial sensory disturbance.

Spinal trigeminal nucleus: Spinal trigeminal nucleus receives facial pain and temperature sensation afferent fibers. The spinal trigeminal nucleus and tract have localized from medulla oblongata to C2 or C3.

The possible explanations of present patients' facial sensory disturbance are as follows: 1) injury of axons or dendrites from the spinal trigeminal nucleus in the cervical cord, 2) injury of axons or dendrites from the spinal gray matter to the spinal trigeminal nucleus, 3) the spinal trigeminal nucleus or tract in human localized to the middle or lower cervical level, 4) compression of the middle or lower cervical cord affect the spinal trigeminal nucleus or tract, 5) occult cerebral lesions such as microinfarcts unable to be identified by MRI. Since facial sensory disturbance of present patients developed gradually and sequentially with other cervical myelopathy symptoms, occult microinfarcts seem to be difficult to explain the facial sensory disturbance of these patients. MRI revealed cervical compressive myelopathies without any brain lesions. Although somatosensory evoked potential (SEP), particularly trigeminal SEP data are unfortunately unavailable, facial sensory disturbance of these two patients seems to be caused by cervical compressive myelopathy.

Patient 1 presented with hyperalgesia of the face and patient 2 complained of abnormal sensation of the face. Therefore, these findings seem to be caused by stimulation of sensory system. The compression of the cervical spinal cord may have stimulative effects on the projections from the spinal trigeminal nuclei to the cervical spinal cords or on the projections from the cervical spinal cords to the spinal trigeminal nuclei.

5. CONCLUSION

We described 2 patients developed facial sensory disturbance associated with cervical compressive myelopathy. These may be associated with stimulative lesions of projections from the spinal trigeminal nuclei to the cervical spinal cords or stimulative lesions of projections from the cervical spinal cords to the spinal trigeminal nuclei.

SUMMARY OF RESEARCH

1.We encountered two patients presented with facial sensory disturbance associated with cervical compressive myelopathy.
2.Cervical compressive myelopathy may cause facial sensory disturbance.
3.The possible mechanisms are stimulative lesions of projections from the spinal trigeminal nuclei to the cervical spinal cords or stimulative lesions of projections from the cervical spinal cord to the spinal trigeminal nuclei.

FUTURE ISSUES

I would like many neurologists to pay attention to facial sensory symptoms associated with cervical compressive myelopathy. This phenomenon seems to be rare, however, publication of this manuscript may notice it. Accumulation of this rare symptom and/or signs and anatomical localizations of associated lesions will make the relationship of this symptom and/or signs and background lesions more accurate.

DISCLOSURE STATEMENT

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