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Case Report

Onion-skin Hemifacial Dysesthesia Successfully Treated with C2–4 Anterior Cervical Decompression and Fusion: A Case Report

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A 49-year-old man with cervical spondylosis at the C2–4 level presented with onion-skin hemifacial dysesthesia in addition to the right extremities. C2–4 anterior cervical decompression and fusion were performed. Onion-skin hemifacial pain disappeared after surgery. Although we cannot conclude the etiology of the pain was either referred pain or direct injury to the spinal trigeminal nucleus, cervical spondylosis at the middle cervical level has a possibility to present facial pain.

Keywords: onion-skin pattern, facial pain, cervical spondylosis, spinal trigeminal nucleus, cervicogenic headache

Introduction

The spinal trigeminal nucleus (STN) is the longest cranial nerve nucleus, extending caudally from the medulla to the upper cervical segment of the spinal cord.[1–3] The STN has somatotopic arrangement, the central area represented rostrally, and the lateral face caudally.[1–3] Therefore, upper cervical lesion has a potential to cause dysesthesia of face sparing the central area, which is called onion-skin pattern.[1–3] Here, we present a rare case of middle level cervical spondylosis presenting onion-skin hemifacial dysesthesia. We discuss the etiology of facial dysesthesia caused by cervical spondylosis.

Case Report

A 49-year-old man was referred to our hospital because of right facial dysesthesia sparing the central portion. The patient had undergone C3–6 expanding laminoplasty for the cervical ossified posterior longitudinal ligament causing bilateral C8 area dysesthesia and neck pain in another hospital at 35 years old. There was a high-signal intensity area at the C3–4 disc level predominately in the right side. Postoperatively, his neck pain resolved but bilateral dysesthesia in the C8 dermatomal area persisted. Fourteen years later, he woke up in the morning recognizing pain and dullness in his right arm and leg, as well as dysesthesia in his face extending from the lower eyelid to the lower jaw sparing nose and mouth (Fig. 1a). Four days later, he consulted the local hospital where laminoplasty had been performed. At first, stroke was suspected but denied by magnetic resonance imaging (MRI) of the brain. His neurological examination in our hospital showed that the cranial nerve was intact except for the dysesthesia in the face. He had slightly decreased strength in deltoid, biceps, iliopsoas, and hamstrings on the right. Superficial and deep sensations were normal. Deep tendon reflex increased from the biceps to Achilles tendon on both sides. He had no bowel or bladder dysfunction. Cervical spine radiography showed swan neck deformity and instability at the C2–3 level in both flexion and extension views. MRI of the cervical spine showed adequate decompression at the dorsal surface of the spinal cord, but subtle compression on the ventral surface by a soft and hard disc projecting from C3–4 intervertebral disc level (Fig. 2). Intramedullary abnormal signal intensity was evident at the C3–4 disc level, with clearer margin on the right than the left. After wearing the neck collar for 7 days, he reported slight improvement of pain in the arm and the central area sparing dysesthesia was enlarged (Fig. 1b). This improvement suggested that the origin of the pain was located in the neck. Computed tomography (CT) myelography of the cervical spine in extension and flexion positions were performed (Fig. 2). In the flexion position, the spinal cord was compressed from the ventral side at the C3–4 disc level because of C2–3 instability. In the extension position, the spinal cord was decompressed at all cervical levels. We consider that the spinal cord compression at the C3–4 level together with C2–3 instability should be treated for the relief of myelopathy and also hemifacial pain, although the etiology of facial pain was not clear. The patient underwent anterior cervical decompression and fusion (ACDF) at C2–4 level using titanium mesh cage (Pyramesh; Medtronic Sofamor Danek, Memphis, Tennessee, USA) filled with local bones (Fig. 3). Soon after the surgery, his symptoms in the arm, leg, and face disappeared except the numbness in both C8 areas. Postoperative CT shows the C2–4 local kyphotic angle had improved from 25 degrees to 18 degrees in the straight position (Figs. 2, 3).

Discussion

Facial pain due to cervical lesions is explained by two different theories; referred pain [4,5] and direct influence on the STN.[1,6–13] Cervicogenic headache (CH) is a syndrome characterized by hemifacial pain and headache that is referred from either the bony structures or soft tissues of the neck.[4,5] Pathophysiology of CH is believed the referred pain from an injured cervical disc, ligament, or muscle to the region of the temple-forehead-face.[4,5] Kawabori et al. presented a case of CH due to C5–6 cervical spondylosis.[13] They reviewed other three cases of CH to find which site of cervical spondylosis

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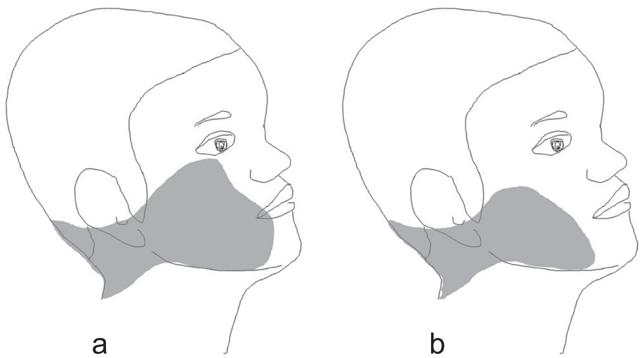

Fig. 1 Schema showing the area of facial dysesthesia, sparing the central portion of the face like onion-skin (a), and the slight improvement of the dysesthesia and enlarged central area sparing dysesthesia after wearing the neck collar (b).

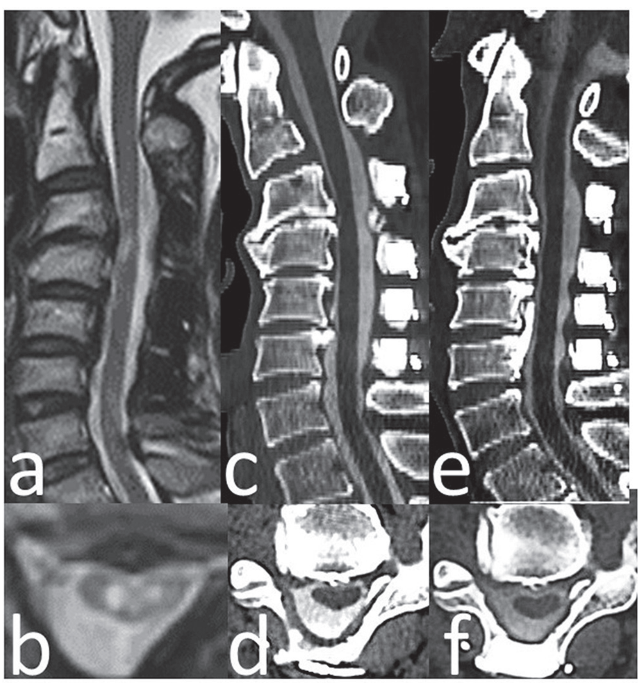

Fig. 2 Sagittal (a) and axial (b) magnetic resonance images at 14 years after expanding laminoplasty showing swan neck deformity and round intramedullary high-signal intensity of the cervical spine with compression myelopathy. Sagittal (c, e) and axial (d, f) computed tomography myelograms showing C2–3 instability. The spinal cord was compressed by the C3–4 soft and hard disc, especially on flexion (c, d) compared to extension (e, f).

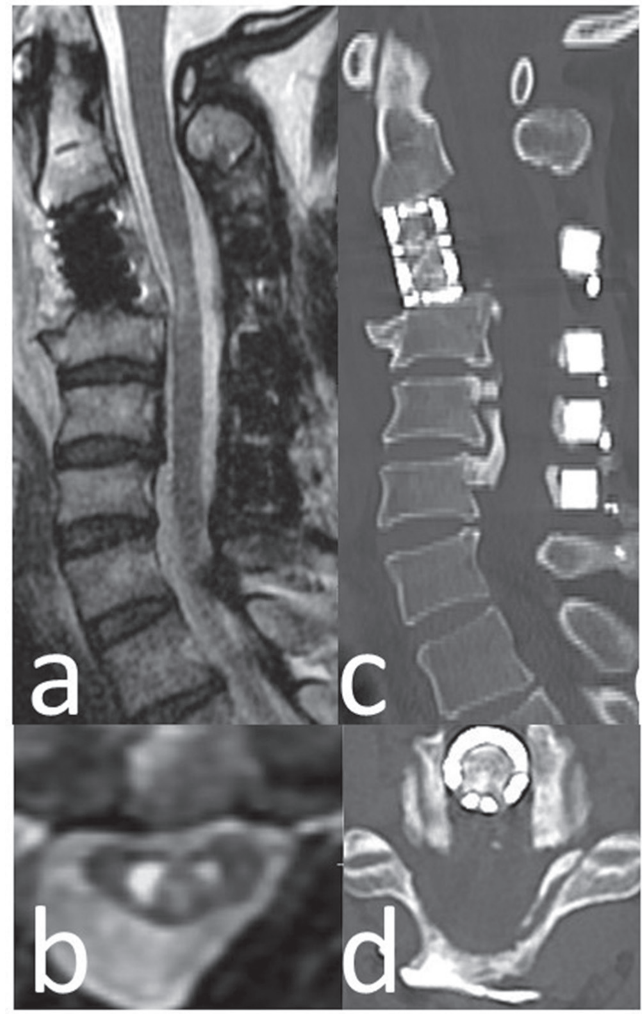

Fig. 3 a, b: Postoperative magnetic resonance images showing adequate decompression of the spinal cord at the C3–4 level. c, d: Postoperative computed tomography showing C3 vertebrectomy and fusion with mesh cages filled with local bone. The C2–4 local kyphotic angle had improved from 25 degrees to 18 degrees in the straight position.

were lower than C4–5 disc level. This functional convergence of cervical and trigeminal sensory pathways allows the bidirectional referral of painful sensations between the cervical and trigeminal sensory receptive fields of the face and head.[4,5,13]

The other mechanism is the direct injury to the STN.[1,6–13] The STN is the longest nerve nucleus and extends caudally from the medulla to the upper cervical segment of the spinal cord.[1–3] Cytoarchitecturally, the STN is divided into three portions, subnucleus oralis, subnucleus interpolaris, and subnucleus caudalis, which are located along the longitudinal axis of the brain stem.[1,3] The central area of the face is represented by the rostral portion of the STN, and the more outer areas of the face are represented by the caudal portion of the STN.[1,3] Therefore, lesions invading the subnucleus caudalis cause onion-skin pattern sensory impairment.[1,6–13] There are conflicts about the location of the subnucleus caudalis, C2,[6,10] C3,[7,11] and C4[1,3] spinal segment. Taren et al. studied the anatomic pathways related to pain in face and neck using monkey and man.[1] He reported that degeneration in the descending tract of V following retrogasserian rhizotomy in man and monkey confirms that trigeminal nerve are represented as far caudally as C4. This experiment, we consider, is the most invasive but reliable. There are some cases of onion-skin pattern sensory impairment involved intraaxial lesions, and all the lesions were beyond the C4 spinal level, such as cerebral infarction (medulla),[7] syringomyelia (cervicomedullary junction to T_{12}),[8] demyelination,[9] and spinal cord injury caused by needle entry (medulla to C7).[11] Extramedullary lesions also affect the STN, such as atlanto-axial dislocation,[10] retroodontoid mass,[6] presenting onion-skin dysesthesia.

In this case, spinal cord at the C3–4 disc level was compressed from anteriorly because of ventral cord shifting due to C2–3 instability. MRI shows an abnormal signal intensity in the spinal cord at the C3–4 disc level predominantly in the right side, but this abnormality was already evident 14 years ago. From a viewpoint of neuronal density or hemodynamics, spinal cord at the C3–4 disc level especially in the right side was considered to be more fragile than the other side. Therefore, progressing cervical spondylosis brought about new symptoms including myelopathy only in the right side. The surgical result was good, but pathophysiology of the hemifacial pain is not clear. If the facial pain was due to CH, C2–4 spinal stabilization might have affected either C2–3–4 facet joint or C3, 4 nerve root. Another possibility is that the decompression of the spinal cord at the C3–4 disc level (C5 spinal segment[2)]) might affect the STN if not directly. Because the central area of the face was spared from pain at preoperative state like onion-skin pattern, and the spared area spread after wearing the neck collar. This spread of spared area resembles the case Chang reported, burning sensation presenting a typical onion-skin pattern after falling retreated from the center towards the periphery of the face after a few days of conservative therapy.[6] The spinal cord compression at C3–4 disc level with C2–3 instability might affect the STN, if indirectly, bringing about spinal cord edema or ischemia spreading craniocaudally.

Conclusion

Onion-skin hemifacial pain disappeared post C2–4 ACDF. Although we cannot conclude the etiology of the pain was either CH or direct injury to the STN, cervical spondylosis at the middle cervical level has a possibility to present facial pain.

Conflicts of Interest Disclosure

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices in the article. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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