Case Report

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Temporomandibular Disorder and Disuse Atrophy of the Masticatory Muscles after Surgical Resection of a Schwannoma: A Case Report

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Disuse atrophy involves gradual muscle weakening due to inadequate usage and can cause temporomandibular disorder (TMD). A 45-year old man with TMD symptoms on the left side, who had disuse atrophy of the masticatory muscles on the right side following surgical removal of a trigeminal schwannoma on the right side, first visited the Department of Orofacial Pain and Oral Medicine at Kyung Hee University Dental Hospital with left jaw pain and difficulty in opening mouth and chewing. He had been experiencing difficulties in cognitive function, decrease in visual acuity, impaired speech, and writing deficits after brain surgery. Furthermore, he complained of abnormal occlusion on the right side, which interfered with his ability to chew comfortably and open his mouth effectively. Herein, we describe a contralateral TMD case due to ipsilateral disuse atrophy after brain surgery for a trigeminal schwannoma and our successful treatment with medication, physical therapy, and stabilization splint.

Key Words: Disuse atrophy; Stabilization splint; Temporomandibular disorder; Trigeminal schwannoma

INTRODUCTION

Temporomandibular disorders (TMDs) occur when the dynamic equilibrium and balance between the components of the temporomandibular joint (TMJ) and masticatory system are disrupted, resulting in morphological and functional deformities. The etiology of TMD is complex and multifactorial. Micro- and/or macro-trauma, behavioral factors, occlusal factors, and psychological factors are common etiological factors of TMD.[1)]

There has been a few reported cases of TMD secondary to brain tumor surgery. During brain tumor removal, damage to the trigeminal nerve, which innervates the TMJ, may result in secondary complications, such as sensory abnormalities, paralysis, and TMDs.[2-6)] TMD may also occur due to disuse muscle atrophy, owing to malfunctioning of motor nerves associated with the masticatory muscles.[7-9)] To compensate for muscle atrophy, overuse of the normal and functioning muscles may occur. Atrophy of the mandibular masticatory muscles may result in fatigue of the overused muscles, leading to decrease in the range of motion of the mandible, along with pain during function.[10-12)]

TMD therapies include various conservative modalities, such as medication, physical therapy, and stabilization splints.[13)] Stabilization splints may assist in re-establishing optimal orthopedic stability with reconstruction of the myoclonic reflexes and by maintaining the jaw in the most stable rest position. Thus, stabilization splints may enhance muscle functions and reduce abnormal activity.[14,15)]

In this case report, we described a case of left TMD after right trigeminal schwannoma surgery. The patient experienced sensory and motor neuron dysfunction of the right temporomandibular area. During a 1 year and 9 months follow-up period at the Department of Orofacial pain and Oral

Medicine, the patient was successfully treated with medication, physical therapy, and stabilization splint therapy.

CASE REPORT

A 45-years-old man presented to the Department of Oral Medicine at Kyung Hee University Dental Hospital, complaining of soreness in his left jaw during chewing, and experienced difficulty in opening his mouth. His mouth opening was limited, and the opening pathway showed marked deflection. The patient reported that his TMD symptoms developed one month before he presented at the hospital. The pain intensity was 7/10 on the visual analogue scale (VAS) (0: no pain, 10: maximum pain imaginable). The pain quality was expressed as throbbing, rather than a sharp pain.

The patient's past medical history revealed that a trigeminal schwannoma on the right side was diagnosed and managed with total surgical resection at the neurosurgery department of another hospital 8 years ago. Following surgery, the patient reported cognitive impairment, difficulty in sleeping, decreased vision, impaired speech, and reduced writing capability. In addition, he complained of symptoms in the orofacial region. In particular, his maxillary and mandibular teeth on the right side did not contact during occlusion. Two years ago, the patient completed orthodontic treatment at a local dental clinic to correct the abnormal occlusion. In the last 5 years, he has had hypertension, which has been managed with daily oral medication.

As part of the diagnostic process, various clinical and imaging tests were performed at our hospital. The initial palpation test indicated tenderness of the left TMJ, including the lateral capsule, posterior capsule, TM ligament, and retrodiscal tissue. Vertical mandibular movements were limited. The measurements for comfortable mouth opening and maximum mouth opening (MMO) were 15 mm and 25 mm, respectively. Bilateral movements were also limited to 4 mm on the right side and 3 mm on the left and accompanied by pain during movement to the right side. Furthermore, right posterior openbite of about 2 mm was observed.

Multiple diagnostic images of the TMJ were evaluated. The panoramic view did not demonstrate any changes in the mandibular condyles. However, the cone beam computed tomography analysis demonstrated degenerative changes, including flattening and erosion of the left mandibular condyle. The bone scan showed an increased uptake of the radioactive tracer in the left TMJ joint. On the magnetic resonance imaging (MRI), structural changes were observed on both sides, and the condyle discs on both sides were located comparatively anterior to the respective condyle on opening and closing of the mouth (Fig. 1). The MRI also showed that the volumes of masticatory muscles were smaller on the right than left; in particular, atrophic changes of the lateral pterygoid muscle and masseter muscle were noted. To summarize, the patient was diagnosed with disc displacement without reduction and osteoarthritis of the left TMJ, and disuse atrophy of the right masticatory muscles.

In order to determine the threshold at which sensory changes and sensory inhibition occurred, the brushing motion direction, two-point discrimination, and a static touch test were performed. The results of these tests

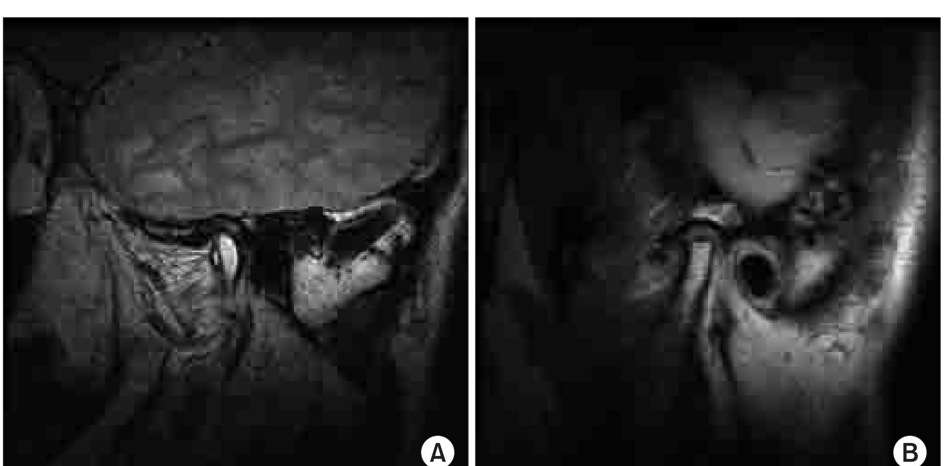

Fig. 1. Magnetic resonance imaging images of temporomandibular joints: (A) right and (B) left, sagittal T2 weighted images in closed mouth position.

revealed hypoesthesia on the right maxillary (V2) and mandibular (V3) divisions of the trigeminal nerve (Fig. 2). Electromyography (EMG) was performed under three conditions: resting, clenching, and chewing. The left side was used as the reference. In all conditions, the right masticatory muscles, e.g. temporalis and masseter, showed a lower EMG level than the left masticatory muscles. In resting condition, the EMG values of the right masseter and temporalis muscles were 75.5% and 76.6%, respectively. In the cotton roll clenching and chewing tests, the values of the right masseter and temporalis were lower than those of the left side (Table 1). The Epworth sleepiness scale, Pittsburgh sleep quality index, STOP-BANG questionnaire, and Symptom Checklist 90 Revised were used to evaluate sleep disturbances and psychological status. The scores were all within the normal range.

During the first visit, the patient was shown how to maintain a stable jaw position, and analgesics and anti-inflammatory drugs were prescribed as part of the initial treatment plan. During the two-month follow-up period, 50% reduction in pain was observed with the use of medications and physical therapy. However, discomfort due to the posterior openbite on the right side persisted. The patient complained that the force of the right masticatory muscle was weaker than that of the left, and that touch sensation on the right facial region was less sensitive compared to the preoperative level of 60%-70%.

Two months after the initial visit, the patient was advised to wear a stabilization splint every night. Pain in the left jaw improved following the use of the splint for one and a half months. The VAS score decreased from 7 to 2. Subsequently, masticatory muscle pain disappeared, and the

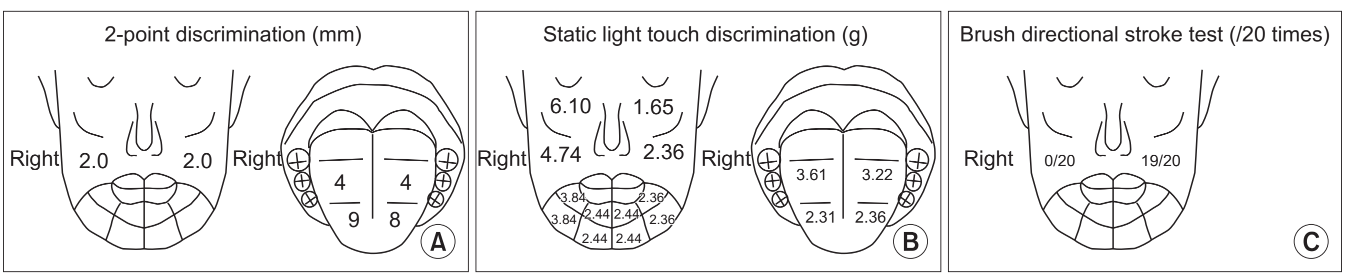

Fig. 2. Results of the quantitative sensory tests: (A) two-point discrimination test, (B) static light touch discrimination test, and (C) directional discrimination test (brush directional stroke test). Tests B and C showed that sensitivity of the right side in the V1, V2, and V3 regions (maxilla and mandible) was lower than that of the left. Tests A and C did not check the chin area.

Table 1. EMG score of each masticatory muscle during initial examination

Muscle	Rest position	Posterior clenching		Gum chewing	
		Right	Left	Right	Left
Masseter					
Right	0.74 (76)	7.8 (9)	7.1 (10)	9.3 (50)	3.7 (17)
Left (reference)	0.98 (100)	88.7 (100)	74.0 (100)	18.8 (100)	22.4 (100)
Temporalis					
Right	1.37 (76)	5.8 (8)	5.1 (7)	5.4 (24)	5.9 (25)
Left (reference)	1.81 (100)	70.1 (100)	74.9 (100)	22.3 (100)	23.7 (100)
SCM					
Right	0.91 (108)	8.4 (68)	8.6 (72)	8.6 (151)	6.0 (97)
Left (reference)	0.84 (100)	12.4 (100)	12.0 (100)	5.7 (100)	6.2 (100)

EMG, electromyography; SCM, sternocleidomastoid muscle.
Values are presented as number (%).
Rest position; when the patient is sitting on a chair and resting, EMG scores of masticatory muscles including masseter, temporalis, and SCM; posterior clenching: EMG measurements of the muscles on the same side (right or left) when clenching a cotton roll posteriorly; gum chewing: EMG measurements with chewing gum on the same side as the muscles being tested (right or left).
When the EMG scores of muscles on the left were taken as a reference, the right EMG scores were significantly lower in the neck muscles, including masseter, temporalis, and SCM.

patient was able to chew on the right side. Partial improvement in his mouth opening was also observed. The patient was followed-up at intervals of 1 to 2 months for 8 months. Following the initial conservative treatment (drug therapy, stabilization splint, physical therapy), the patient's mouth opening and pain intensity improved significantly. His MMO increased to 37 mm, and the VAS score decreased to 1. After one year, MMO increased to 40 mm. The left TMJ was painless, although mild stiffness was detected upon stimulation. Osteoarthritis in the left TMJ was still evident in the follow-up bone scan taken at 1 year and 9 months after the initial visit. Activity values decreased (right: 59 → 49; left: 80 → 74). In the follow-up MRI, significant improvement was observed in the irregularity of the cortical line and erosion of condyle. The patient has been using a stabilization splint for a period of 2 years, and his TMD symptoms have significantly improved.

DISCUSSION

Jaw closing involves the temporal, masseter, and medial pterygoid muscles. Jaw opening utilizes the lateral pterygoid muscle. These muscles are considered as masticatory muscles, and they are innervated by the mandibular branch of the trigeminal nerve.[16] An intact trigeminal nerve is critical because this nerve is responsible for muscle functions, such as chewing and speaking, as well as the sensation of the orofacial area. In addition, the morphologic or functional characteristics of masticatory muscles are continuously altered by physiological changes, such as perioral habit and/or pathological processes, such as disuse related to nerve damage.[17] Thus, trigeminal nerve damage may be associated with disuse atrophy of the masticatory muscle, which ultimately impairs the individual's quality of life.

Disuse atrophy refers to a type of muscle atrophy that occurs when the muscles are no longer used as usual. A lack of energy intake, unbalanced nutritional status, and inactivity of muscles all lead to a reduction in muscle mass, which is associated with disuse atrophy and loss of function. Loss of muscle mass occurs in common diseases including cancers[18] and neurodegenerative disorders.[19] Masticatory muscle is not only crucial for mastication but also important in speech, and aesthetic reasons. An understanding of the mechanisms of disuse atrophy is crucial for designing countermeasures and/or rehabilitation protocols.[20]

Schwannoma is a benign tumor of the nerve sheath from the Schwann cells. Schwannomas commonly occurred in the vagus and the cervical sympathetic chain, whereas trigeminal schwannomas are rare.[21] We presented a case of contralateral TMD after surgical removal of a trigeminal schwannoma. This brain tumor is usually detected as a large mass because of painless undisturbed growth, leading to diagnostic delay.[22] Treatment depends on the size, location, and stage of the mass, but surgical excision is usually performed.[23] Malignant transformation is rare in schwannoma, and the prognosis after complete excision is remarkable.[24] However, facial dysesthesia, paresthesia, and hyperalgesia are frequently observed. Chewing disturbance after surgical treatment has also been reported. According to Ramina et al.[25], trigeminal hypoesthesia and facial pain are common, affecting up to 53% of patients after surgery for their trigeminal schwannoma. This may be due to nerve damage resulting from the surgical approach.

The stabilization splint is a removable hard acrylic splint that provides a temporary ideal occlusion.[26] Providing an ideal occlusion by the use of a stabilization splint reduces neuromuscular imbalance. The masticatory muscles can be relaxed over several visits until a consistent jaw relationship is reached. Previous clinical studies have demonstrated the efficacy of stabilization splint in the treatment of myofascial pain.[27,28] The patient in our study had significantly improved symptoms after TMD treatment with a stabilization splint. It was more effective than other previous treatment. The mechanisms of action of stabilization splint therapy include biofeedback, visual feedback, relaxation of the masticatory muscle, and resolution of the neuromuscular imbalance. Only a few cases of TMD after postoperative trigeminal nerve injury have been reported in the literature. Therefore, in order to clarify our findings, future multi-centered studies with larger sample sizes and longer follow-ups are needed.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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