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Disuse atrophy of masticatory muscles after intracranial trigeminal schwannoma resection: A case report and review of literature

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ABSTRACT

INTRODUCTION: Temporomandibular disorders (TMD) are diseases of the temporomandibular joint and masticatory muscles, and are often difficult to be diagnosed because they have various symptoms, pathological conditions and causes.
PRESENTATION OF CASE: Herein, we report a 78-year-old male referred to our hospital with a diagnosis of TMD and presenting with facial asymmetry, marked deviation to the right side on vertical mandibular movement and complaints of abnormal perception at the right oral and buccal region. Past medical history revealed that he had undergone a right intracranial trigeminal schwannoma resection 9 years prior. Computed tomography (CT) showed disuse atrophy of the right side of 4 masticatory muscles and 2 suprahyoid muscles controlled by the motor component of the mandibular division (V3) of the trigeminal nerve (TGN). Together with the neurosurgeon, we confirmed that there was no recurrence of the tumor and explained to the patient that the oral and maxillofacial symptoms are after-effects of the operation, and we provided oral hygiene instructions and coordinated cleaning of the inside of the oral cavity.
DISCUSSION: Although it is difficult to compare treatment methods from case to case, we believe that in our case, the patient's understanding of the cause of his discomfort contributed significantly to the improvement of his quality of life.
CONCLUSION: We experienced a case of masticatory muscle disuse atrophy during long-term follow-up after resection of intracranial trigeminal schwannoma. Further studies are needed to develop the diagnostic and therapeutic protocols for disuse atrophy.

1. Introduction

Temporomandibular disorders (TMD) are diseases of the temporomandibular joint and masticatory muscles, and are often difficult to be diagnosed because they have various symptoms, pathological conditions and causes. The morphologic and functional characteristics of masticatory muscles are maintained by sensory and motor innervation from the trigeminal nerve (TGN) [1]. Hypertrophy, atrophy and fat infiltration of masticatory muscles can be related to physiologic processes such as activity and disuse [2]. Disuse atrophy is a muscle wasting or a decrease in muscle volume due to the muscle no longer being used [3]. Since the masticatory muscles are skeletal muscles, they are influenced by such physiological processes. The trigeminal nerve is the largest of the twelve cranial nerves. The sensory function of the TGN is to provide tactile, proprioceptive, and nociceptive afferents from the face and oral cavity while its motor function activates the movement of eight bilateral sets of muscles, including the four muscles of mastication: the masseter, the temporal muscle, and the medial and lateral pterygoids [4]. The other four muscles are the tensor veli palatini, the mylohyoid, the anterior belly of the digastric and the tensor tympani, all of which are controlled by the motor component of the mandibular division (V3) of the TGN [1,5]. Damage to the TGN by conditions such as a tumor, demyelination, multiple sclerosis or compression can lead to masticatory muscle dysfunction that eventually leads to TMD [6,7]. The clinical

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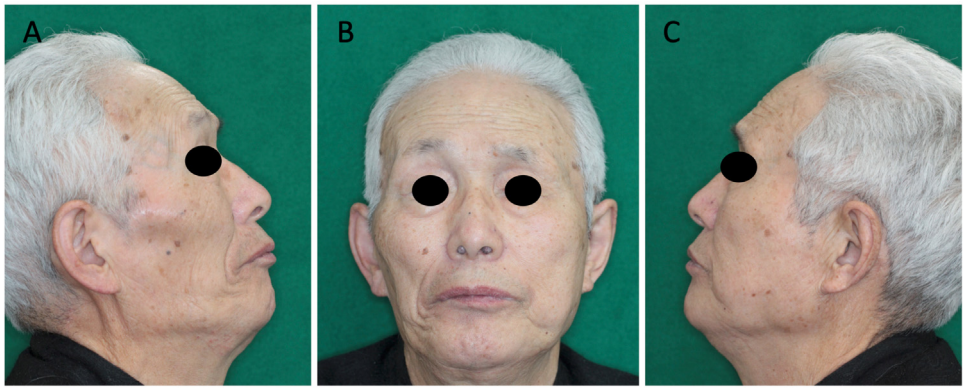

Fig. 1. Extraoral photograph at first visit. Right lateral view (A), frontal view (B) and right lateral view (C) show asymmetry. Invagination was shown under the zygomatic bone.

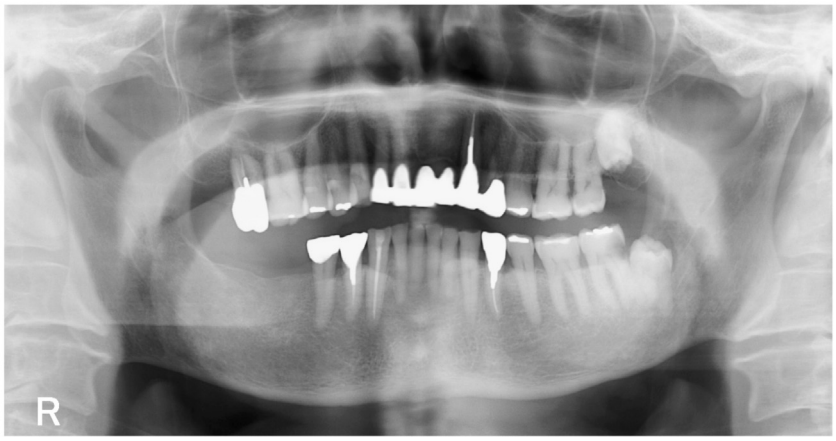

Fig. 2. Panoramic X-ray at first visit.
The right mandibular condyle situated anterior gliding, whereas the left mandibular condyle was contained in the mandibular fossa.

presentation varies depending on the site of involvement, i.e., whether it's intracranial or extracranial. Lesions that involve the brainstem rarely present with isolated TGN deficits, owing to the compact distribution of neural structures in this region. Cranial nerve deficits involving III, IV, VI, VII and XII are usually associated [8]. Lesions involving the cisternal segment or Meckel's cave typically present with trigeminal neuralgia, facial weakness (VII CN) and/or hearing loss (VIII CN). Cavernous sinus involvement usually presents with ocular manifestations, cranial nerve III, IV, V1, V2, and VI deficits. Peripheral segment involvement of the terminal branches of V1, V2, and V3 is most commonly involved in perineural spread of head and neck malignancies, which is most frequently seen along the maxillary division. Pain, paraesthesia, or analgesia in the distribution of the involved trigeminal nerve are typical, with weakness in chewing, mouth opening and deviation of the jaw as common motor symptoms. Specific sensory symptoms can include absent corneal or oculocardiac reflexes (V1); absent jaw jerk reflex and numb chin syndrome (V3) are also seen [9]. Trigeminal schwannoma is a rare benign tumor originating from the Schwann cells of the trigeminal nerve sheath [10]. This tumor can arise from the cisternal segment, trigeminal ganglion in Meckel's cave or from one of the three branches of the nerve. Clinically, it usually presents with facial pain, numbness and paraesthesia in the distribution of one or all the divisions of the trigeminal nerve, depending on the location of the tumor [9,11]. Here we present a case of disuse atrophy of the masticatory muscles caused by the resection of a right intracranial trigeminal schwannoma, who was introduced to our hospital under the diagnosis of TMD with complaint of abnormal perception.

2. Case presentation

A 78-year-old male patient was referred to our hospital in April 2019 complaining of discomfort in his right jaw under the diagnosis of right TMD. Specifically, the patient complained of a swelling-like feeling at the upper and lower right gingiva and constant coldness in his right cheek that he had endured for 1 year before consultation. In addition, he complained of food impaction in the area of the upper right posterior vestibule that caused him discomfort. He has gotten occlusal adjustment of upper right first molar by the previous clinic as it was impinging on the lower gingiva because of over-eruption; however, the patient didn't feel any improvement. His extraoral findings showed facial asymmetry (Fig. 1A–C), and invagination under the zygomatic bone was recognized (Fig. 1A and B). Temporomandibular joint (TMJ) examination showed normal mouth opening (52 mm) but marked deviation to the right side on vertical mandibular movement. Lateral mandibular movements were within the normal range; however, limited movement to the left side was observed. There was no tenderness or pain at the right gingiva, lower lip or mental region upon palpation. Panoramic X-ray showed the right mandibular condyle situated anterior gliding, whereas the left mandibular condyle was contained in the mandibular fossa (Fig. 2).

Past medical history of the patient revealed subtotal resection of a right brain tumor diagnosed as trigeminal schwannoma 9 years prior. In April 2011, the patient was admitted to the emergency department because of complete left hemiplegia and disturbance of consciousness, which gradually progressed until he was in a coma. Examination revealed a gigantic right intracranial trigeminal

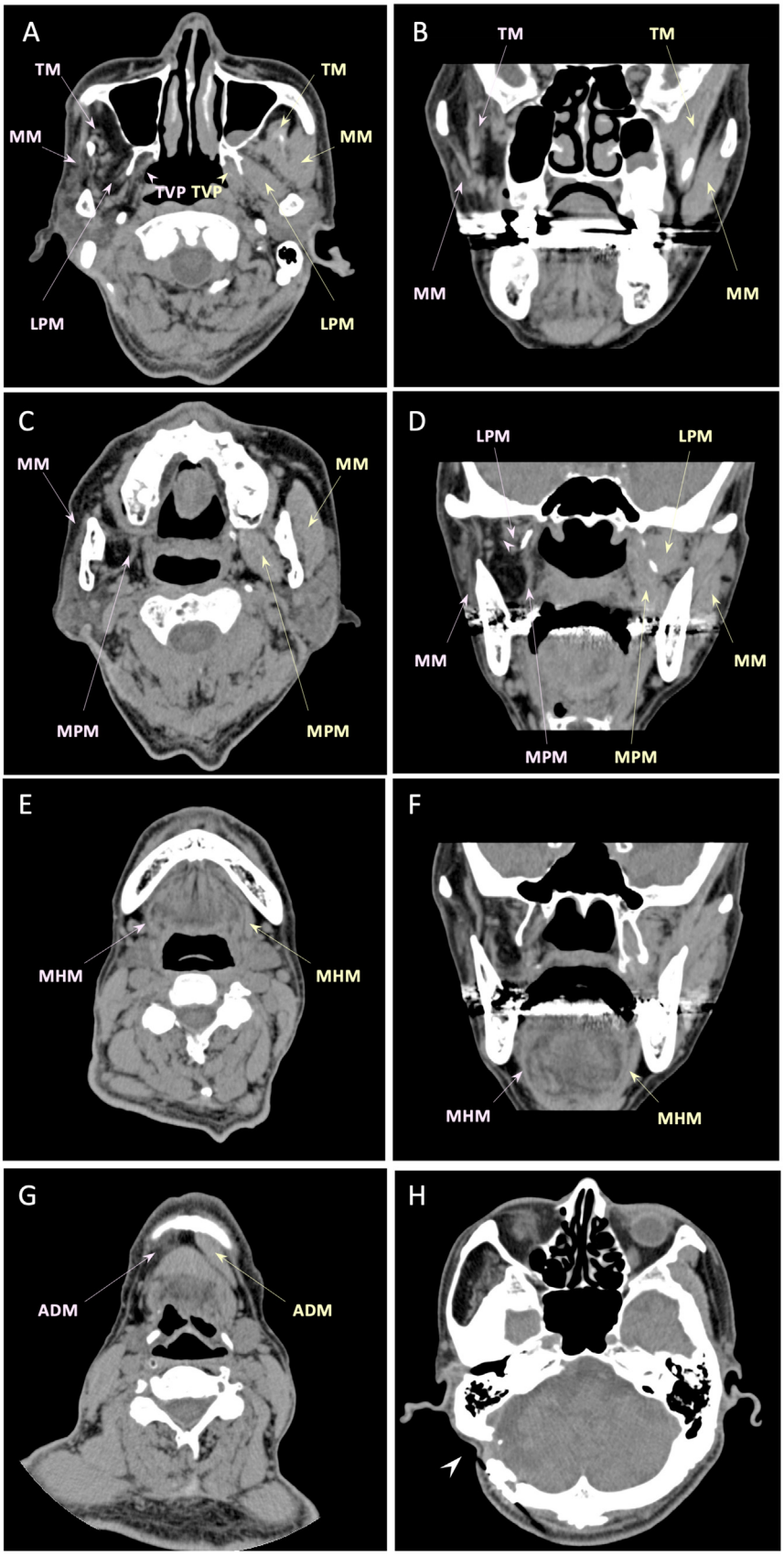

Fig. 3. Computed tomography (CT) of the maxillofacial regions showing atrophy of masticatory muscles of right side. A) Axial view at the level of mandibular condyle and coronoid process. B) Coronal view at the level of coronoid process. C) Axial view at the level of the mandibular foramen. D) Coronal view at the level anterior to the pterygoid plate. E) Axial view at the level of submandibular space. F) Coronal view at the level of the pterygoid plate. G) Axial view at the level of submental space. (H) Axial view showing bone defect site of intracranial tumor resection through right occipital/suboccipital craniotomy (arrowhead). TM, temporalis muscle; MM, masseter muscle; LPM, lateral pterygoid muscle; MPM, medial pterygoid muscle; TVP, tensor veli palatini muscle; MHM, mylohyoid muscle; ADM, anterior belly of the digastric muscle.

Table 1
Characteristics of 9 cases with disuse atrophy in masticatory muscles.

Author	Year	Age	Gender	Initial symptoms	Cause	Muscles involved	Trigeminal symptoms	Additional symptoms
Luyk et al. [15]	1991	42	Female	Preauricular pain	Intracranial meningioma	Masseter Temporalis Lateral pterygoid Medial pterygoid	Hypoesthesia Decreased corneal reflex Absent jaw jerk reflex	Orofacial pain TMJ click
Imauchi et al. [16]	2002	75	Female	None	Masticatory habit (functioning left side for 40 years)	Masseter Temporalis Lateral pterygoid Medial pterygoid	None	None
Chiba et al. [17]	2012	70	Female	Dull pain in submandibular region	Pure trigeminal motor neuropathy	Masseter Temporalis Mylohyoid Lateral pterygoid Medial pterygoid Tensor veli palatini Anterior belly of digastric	None	TMJ discomfort Jaw deviation
Madasamy et al. [20]	2012	8	Female	Facial deformity	Parry Romberg syndrome	Masseter	None	None
Chou et al. [21]	2014	58	Male	Progressive paraesthesia Continuous pain at temporal and anterior auricular area	Trigeminal Schwannoma	Masseter	Trigeminal neuralgia	None
Wilson et al. [18]	2015	29	Female	None	Pure trigeminal motor neuropathy	Masseter Temporalis Lateral pterygoid Medial pterygoid	None	None
Kämppi et al. [19]	2018	57	Male	Muscle pain	Pure trigeminal motor neuropathy	Masseter Temporalis Lateral pterygoid Medial pterygoid	None	None
Yeon et al. [22]	2018	45	Male	Soreness in jaw Trismus	Trigeminal Schwannoma	Masseter Temporalis Lateral pterygoid Medial pterygoid	Hypoesthesia	TMD Jaw deviation
Present Case	2020	74	Male	Discomfort in gingiva	Trigeminal schwannoma	Masseter Temporalis Mylohyoid Lateral pterygoid Medial pterygoid Tensor veli palatini Anterior belly of digastric	Paresthesia	Jaw deviation

schwannoma. Because 12 days of medical treatment did not relieve the coma, intracranial tumor resection was performed through a right occipital/suboccipital craniotomy. Although almost 90% of the tumor was resected, the portion attached to the brainstem could not be removed. The patient's condition improved gradually after the operation, and he was discharged in October 2011.

Computed tomography (CT) for the TMJ examination at the first visit showed atrophy of masticatory muscles at the right-side maxillofacial region (Fig. 3A–G). The temporalis muscle (TM), masseter muscle (MM), lateral pterygoid muscle (LPM), medial pterygoid muscle (MPM) and tensor veli palatini muscle (TVP) showed atrophy or absence on the axial view at the level of the mandibular condyle and coronoid process (Fig. 3A), coronal view at the level of the coronoid process (Fig. 3B), axial view at the level of the mandibular foramen (Fig. 3C) and coronal view at the level of anterior of the pterygoid plate (Fig. 3D). The mylohyoid muscle (MHM) was atrophied on the axial view at the level of submandibular space (Fig. 3E) and the coronal view at the level of the pterygoid plate (Fig. 3F). The anterior belly of the digastric muscle (ADM) was also absent from the axial view at the level of submental space (Fig. 3G). An axial view in Fig. 3H shows the bone defect site of the intracranial tumor resection through the right occipital/suboccipital craniotomy (arrowhead).

The final diagnosis was disuse atrophy of masticatory muscles caused by the resection of the right intracranial trigeminal schwannoma. We contacted the attending neurosurgeon and requested a follow-up on the cranial neoplasm by magnetic resonance imaging (MRI), 9 years after the initial surgery, and confirmed that there was no recurrence of the tumor. Together with the neurosurgeon, we explained to the patient that the oral and maxillofacial symptoms are after-effects of the operation, and we provided oral hygiene instructions and coordinated cleaning of the inside of the oral cavity every 1 month at the primary care dental clinic. Understanding the cause of his medical condition allows the patient to better manage his symptoms, and his quality of life seems to have improved. Currently, the patient has had no recurrence or exacerbation of the former discomfort (swollen right upper and lower gingiva, numbness, and constant cold sensation in the right cheek), actively participates in oral hygiene maintenance, and is able to eat more comfortably than before.

3. Discussion

Determining the cause of unilateral facial asymmetry is quite challenging. The clinician has to rule out all possible causes and maintain a high level of suspicion to reach a definitive diagnosis. We present a case of subtotal resection of an intracranial trigeminal schwannoma, which is known for its complex functional and sensory post-operative complications. The functional complications include atrophy of the muscles of mastication and chewing disturbance while the sensory complication include facial dysesthesia, paresthesia and hyperalgesia along the distribution of the TGN or its individual branches [12]. In our case, the patient experienced a condition of paresthesia in his right cheek and gingiva after surgery because of damage to the TGN resulting in sensory impairment of the orofacial region. In addition, the patient had a chronic problem with food impaction in his right-side cheek, which could be attributed to loss of muscle tone and therefore, loss of the muscles' cleansing effects. TMJ movements are controlled by certain groups of muscles, for instance, the muscles of mastication and some of supra-hyoid muscles (mylohyoid and anterior belly of digastric muscles). All these muscles are innervated by the mandibular division of the TGN [13]. Consequently, damage to this branch will lead to disturbance in mandibular movements, such as the mandibular deviation during opening found in our case. Furthermore, the morphologic or functional characteristics of masticatory muscles are continuously altered by physiological changes and/or pathological processes [14]. Thus, trigeminal nerve damage may be associated with disuse atrophy of the masticatory muscle.

Our review of the English literature in PubMed revealed 8 case reports of masticatory muscle atrophy, and we present the char ac ter is tics of each case and its condition along with those of our case in Table 1. We found only one other published case report about masticatory muscle atrophy following resection of trigeminal schwannoma, which emphasized the rarity of this condition. The other 7 reports investigated masticatory muscle atrophy due to different reasons. Luyk et al. reported masticatory muscle atrophy associated with facial pain secondary to intracranial meningioma [15]. Imauchi et al. described a similar case; however, the reason behind the atrophy was a masticatory habit [16]. The case was discovered incidentally. Three authors explained that pure trigeminal neuropathy can lead to masticatory muscle atrophy. Pure trigeminal motor neuropathy is characterized by trigeminal motor paralysis without trigeminal sensory disturbances and without the involvement of the other cranial nerves. Because long-standing trigeminal motor denervation is irreversible, no effective therapy is available for those patients. Chiba et al. described unilateral atrophy of the masticatory muscles and osteonecrosis of the mandibular ramus due to pure trigeminal motor neuropathy, attributing the ramus defect to wasting and weakness of the left jaw-closing muscles [17]. Wilson et al. and Kaamppi et al. demonstrated that pure trigeminal motor neuropathy resulted in focal atrophy of the masticatory muscles [18,19]. Madasamy et al. reported a case of 8-year old child with progressive hemifacial atrophy due to a rare condition called Parry Romberg syndrome [20]. This syndrome is characterized by slowly progressive atrophy involving one side of the face. The disease is self-limiting, and has no definite cure. Chou et al. described a case of solitary masseter muscle atrophy combined with atypical trigeminal neuralgia because of trigeminal schwannoma [21]. Yeon et al. reported a case very similar to ours, with TMD and disuse atrophy of the masticatory muscles after surgical resection of a trigeminal schwannoma [22]. The patient was treated by medications, physiotherapy and a stabilization splint, and achieved a satisfying result, and the authors concluded that a stabilization splint was more effective than other previous treatments.

Trigeminal motor neuropathy may be suspected from symptoms, but electromyography is mandatory diagnostic tool for confirmation of paresis and muscle atrophy. Chiba et al. mentioned in their case report that electromyography was valuable for showing the weak contractions of the affected muscles [17]. In our case, electromyography wasn't requested, however, we will consider it in our future cases for establishing the diagnostic protocol in the future. Although it is difficult to compare treatment methods from case to case, we believe that in our case, the patient's understanding of the cause of his discomfort contributed significantly to the improvement of his quality of life. This report has been reported in line with the SCARE 2018 criteria [23].

4. Conclusion

Unilateral atrophy of masticatory muscles should prompt the clinician to think of an intracranial tumor and ask for thorough imaging of the head and maxillofacial region, in particular brain MRI, to rule out any cerebral cause. In addition, neurological examination is crucial in such cases. A few rare cases of masticatory muscle disuse atrophy after intracranial trigeminal schwannoma have been reported, suggesting that further studies to develop a diagnostic and a therapeutic protocol should be performed.

Declaration of Competing Interest

None to declare.

Funding

None to declare.

Ethical approval

Ethical approval was exempted by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. Identifiable patient information has been removed.

Author contribution

Nagwan Elsayed: Writing of original draft.
Tsuyoshi Shimo: Conceptualising and writing of the paper.
Masayasu Tashiro: Examination of X-ray and CT data.
Eiji Nakayama: Supervision of analysis of the CT data.
Hiroki Nagayasu: Contributed in critical reading.

Registration of research studies

NA.

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