Possible association between mandibular repositioning device for sleep apnea and osseous lytic lesion with fracture of the coronoid process of the mandible: A case report

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Mandibular repositioning devices (MRDs) increase the patency of the upper airway by repositioning the mandible forward, resulting in displacement of the oropharyngeal tissues preventing upper airway collapsibility. Mandibular anterior repositioning is counteracted by muscle force from the temporalis muscle. A 39-year-old man had an osteolytic lesion with fracture of the coronoid process of the mandible secondary to wearing a MRD for sleep apnea. Continuous stress generated on the coronoid process temporalis muscle resulted in osteolysis and fracture of the coronoid process on the patient’s right side, resulting in swelling and limited ability to open his mouth. The patient was managed with intravenous antibiotics to control the osteomyelitis and surgical debridement, with removal of the coronoid process of the mandible. It is unclear why the fracture only occurred on the right side. Pathologic fracture of the coronoid process due to chronic stress and secondary osteomyelitis is a rare severe complication of treatment for obstructive sleep apnea syndrome with a MRD. (Quintessence Int 2016;47:141–145; doi: 10.3290/j.qi.a34805)

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Obstructive sleep apnea syndrome (OSAS) is a disturbance of normal sleep patterns with associated daytime sleepiness. It is a common disorder that affects people of all ages and is characterized by episodes of upper airway obstruction during sleep, which result in a reduced blood oxygen saturation, loud snoring, breathing interruptions, and arousal due to gasping or choking following obstructive respiratory events.1

OSAS is caused by blockage within the upper airway, resulting in repeated increases in resistance to airflow. The airway obstruction causes blood oxygen (oxyhemoglobin) desaturation, and carbon dioxide accumulation and the drop in oxygen levels disturbs sleep causing sleep fragmentation. In long-standing
cases of the syndrome, cardiovascular, metabolic, and occupational hazards may occur. Various etiological factors have been attributed to OSAS. Among them are craniofacial anomalies like retrognathism of the maxilla and mandible, increased lower facial height, elongated soft palate, enlarged tongue, decreased posterior airway space, and inferior positioned hyoid bone. Other anatomical factors are presence of a high-arched palate, enlarged palatine tonsils, and enlarged uvula.

Various surgical and nonsurgical management techniques are aimed at improving ventilation. One of the nonsurgical management techniques is the use of oral-dental appliances to counteract the obstruction of the upper airway. A commonly used appliance is the mandibular repositioning device (MRD). The rationale for using a MRD is to increase the patency of the oronasopharynx. This can be achieved by repositioning the mandible forward, with resultant rearrangement of the oropharyngeal tissues thereby preventing upper airway collapsibility.

Various minor and relatively severe side effects associated with the use of MRDs have been reported in the literature. These side effects are often mild and resolve with adjustment of the device. Some of these side effects are dental malocclusion, temporomandibular joint (TMJ) pain, myofascial pain, tooth pain, salivation, TMJ sounds, dry mouth, and gingival irritation.

The coronoid process is a relatively weak part of the mandible. It is deeply located within muscular coverage, which makes it less prone to fracture. However, it can be fractured as a result of direct blunt trauma, penetrating trauma, and sudden muscular contraction. Its incidence of fracture among facial bone ranges from 0.6% to 4.7%.

Information about fractures of the coronoid process of the mandible after wearing a MRD for sleep apnea does not exist in the literature. Here we present a case with an osteolytic lesion with fracture of the coronoid process of the mandible secondary to wearing a MRD for sleep apnea.

**CASE REPORT**

A 39-year-old man was admitted with swelling in his right cheek and limited ability to open his mouth. The patient (apnea-hypopnea index 32.7/h sleep) had a history of wearing a custom-made, titratable duobloc type of MRD with vertical dimension of 7.1 mm every night for the previous 4 months for mandibular advancement. The device was set in the maximal comfortable protrusion after which the patient titrated an additional 1 mm in about 4 weeks. After 4 months he experienced pain in the mandibular angle region, and an additional intraoral radiograph showed a well-delineated radiolucency in the right angular corner of the mandible, considered to be a coincidental finding. Two weeks thereafter pain concentrated in the right parotid region a few weeks after wearing the MRD while eating and an active craniomandibular disorder was excluded although the patient was advised to stop using the MRD until resolution of the pain. One week later symptoms subsided but then returned 4 weeks later. Thereafter, he experienced progressive swelling in the right parotid region for 10 days with limited ability to open his mouth, which caused him to stop wearing the device.

Medical history showed that the patient had no musculoskeletal injuries nor history of head trauma. There was no history of osteoporosis or bony metabolic disease. The patient was not a bruxist and he gave no history of myalgia.

The panoramic radiograph and cone beam computed tomography (CBCT) at presentation showed an osteolytic lesion with fracture of the coronoid process of the mandible (Fig 1), which was absent before wearing the MRD (Fig 2). Clinically, the mouth opening was restricted to 16 mm, without deviation of the opening path during excursion. The right TMJ was painful upon palpation, with swelling in the right pre-auricular region. Intraoral examination showed swelling of the right cheek, but no dental problems and no intraoral abscess formation.

The MRI scan showed abscess formation on the right peri-auricular region between the temporalis and masseter muscles.

Frozen sections from the right coronoid process showed a fragment of connective tissue and muscle with a focal area of Malpighian epithelium with strong
inflammatory cells and granulation tissue. This is in line with osteomyelitis with focal abscess formation in the muscle.

A CT scan of the head revealed an extensive osteolytic lesion with signs of bone fracture and accompanying soft tissue swelling around the right coronoid process of the mandible (Fig 3).

The coronal sections further illustrate the extent of the osteolytic process and also show the fracture of the coronoid process with the top pulled out. The fracture of the coronoid process with an extensive zone of bone loss could be seen as the cause of the significant trismus.

The patient was admitted and placed on intravenous antibiotics and analgesics. Intravenous antibiotics were given to control the osteomyelitis; thereafter, surgical debridement with removal of the coronoid process of the mandible was done. At the 3-week postoperative follow-up visit, the patient was doing well with little or no pain. A panoramic radiograph was taken to check the healing process of the osteomyelitis after debridement. Intraorally, there was good wound healing and no signs of infection or dehiscence. The active mouth opening was 38 mm.

DISCUSSION

A MRD produces favorable results within a short time and is an alternative treatment when upper airway surgery is not indicated. The device pushes the tongue and jaw forward thereby increasing the airway and improving airflow. Most oral appliances are designed to maintain the mandible and tongue in a protruded posture during sleep, thereby preventing upper airway occlusion. The MRD also reduces the probability that soft tissue will collapse and narrow the airway during breathing. A MRD is a noninvasive treatment for OSAS and better tolerated by patients and less cumbersome than continuous positive airway pressure (Fig 4).

The MRD forces the mandible forward and, in the process, the coronoid process also moves anteriorly. The anterior movement of the mandible is counteracted by muscle force from the temporalis muscle,
which retracts the protruded mandible. The retraction force from the temporalis muscle is concentrated at the point of insertion on the coronoid process of the mandible (Fig 5).

The coronoid process of the mandible is a thin, triangular eminence, which is flattened from side to side. It is composed of dense cortical bone with little or no spongy bone. The continuous tension on the temporalis muscle creates stress on the coronoid process. This stress leads to osteolysis of the coronoid process and, finally, fractures. Thus a stress fracture with secondary infection and irritation of the surrounding muscles results in trismus. We have no explanation for why the lesion originated only on the right side of this case. No pathologic changes were detected on the left side of the patient.

The causative factors of non-traumatic fracture are unknown. However several factors may act alone or in combination to increase the risk of bone fracture. Such factors are: bone mechanical properties, the loading force, and the property of the bone itself.11 Though the role of microdamage in increasing the fracture is not clear, the existence of microdamage in bone has been known to reduce bone elastic properties during loading.11

A continuous high magnitude stress produces positive conditions for microdamage, which can result in stress fracture. Presence of bone microdamage due to overload and accumulation of stress in a small bone area such as coronoid process can lead to stress fracture of the bone. Other possible causes of osteolytic lesions of the mandible are central giant cell tumors of the bone, giant cell granuloma, aneurysmal bone cyst, and brown tumors of hyperparathyroidism. However, these conditions rarely affect the coronoid process of the mandible.12

This observed complication is very rare and, to our knowledge, such a severe complication like fracture of
the coronoid process secondary to a MRD has not been previously published in the literature.

CONCLUSION

Pathologic fracture of the coronoid process due to chronic stress and secondary osteomyelitis is a rare but severe complication of treatment for OSAS with a MRD.

REFERENCES