## Delayed tongue necrosis simultaneous with bilateral osteoradionecrosis of the jaw secondary to head and neck irradiation

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We present a rare case of massive tongue necrosis occurring simultaneously with bilateral osteoradionecrosis (ORN) of the jaw in a patient with a history of treatment, including surgery and postoperative radiotherapy, for a retromolar trigone carcinoma 8 years earlier. There is a distinct possibility that the extractions and administration of local anesthesia with a vasoconstrictor contributed to the onset of ORN; together, these events may have influenced the blood supply to the tongue. A glossectomy was performed after hyperbaric oxygen therapy. One month after the surgical procedure, the patient responded satisfactorily to the treatment, showing significant improvement in speech and oral food intake, as well as significant decrease in lingual pain. Although the simultaneous occurrence of these oral complications is rare, the practitioner must be aware of the factors that instigate ORN and compromise vasculature as well as the clinical signs of tongue necrosis. Additionally, the possibility of tongue necrosis secondary to irradiation of the head and neck should be taken into consideration when an irradiated patient undergoes tooth extractions under local anesthesia with agents containing epinephrine. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016; e1:e1-e5)

Tongue necrosis is a rare oral complication associated with irradiation of the head and neck region. This condition may attributed to a wide range of causes, including malignant tumors, use of a vasoconstrictor, severe hemorrhage, embolism, abscess of the floor of the mouth, syphilis, giant cell arteritis, chemotherapy, and previous head and neck radiotherapy.<sup>1-5</sup> The tongue has a rich vascular supply, consisting mainly of the bilateral lingual, pharyngeal, and facial arteries, in addition to collateral circulation. Generally, when tongue necrosis is observed, it commonly affects one side of the tongue, mainly the tip or the anterior half.<sup>1-5</sup> The base of the tongue is never involved. Few cases of bilateral tongue necrosis have been reported in the literature.<sup>1-5</sup> There are no reports of tongue necrosis occurring simultaneously with osteoradionecrosis (ORN) of the jaw.

Primary tumors of the tongue are common, whereas metastatic tumors rarely occur at this site. Squamous cell carcinoma (SCC) is the most frequent primary malignant neoplasm occurring at this site.<sup>6</sup> Treatment for tongue cancer usually involves surgery to remove the cancer.<sup>6</sup> Chemotherapy, radiation therapy, and targeted drug therapy may also be recommended,

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depending on other factors, including the type and stage of cancer, possible side effects, and overall health of the patient. Radical neck dissection with ligation of the external carotid artery in patients receiving radiotherapy of the head and neck region has been reported as a cause of unilateral tongue necrosis.<sup>6</sup>

A search of texts published until 2016 and included in the MEDLINE database was conducted using specific terms: "lingual necrosis," "lingual infarction," "tongue necrosis," "tongue infarction," "giant cell arteritis and tongue necrosis," "lingual ischemia," and "head and neck radiotherapy and lingual necrosis." Among the search results, we found only three cases of tongue necrosis occurring after head and neck radiotherapy.<sup>5-7</sup> Therefore, the purpose of this report was to delineate a case of massive tongue necrosis occurring simultaneously with bilateral ORN of the jaw in a patient with a history of surgery and postoperative radiotherapy for SCC of retromolar trigone.

#### **CASE REPORT**

A 56-year-old male was referred by his oncologist physician for dysphagia caused by a painful, swollen, and discolored tongue. The patient had a medical history of treatment for SCC (T2 N2 M0) of the right retromolar trigone area with a combination of surgery (tumor resection/modified radical neck dissection) and postoperative fractionated radiotherapy (5 fractions/week; 2.0 Gy/fraction; 34 sessions) 8 years earlier.

To obtain more information about the surgery, we contacted the head and neck surgeon, who informed us that the patient had received right radical neck dissection (lymph nodes levels II to IV) without ligation of the external carotid artery. A total of 68 Gy had been applied to the oral cavity and

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#### ORAL MEDICINE

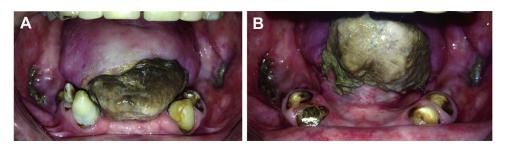


Fig. 1. A and **B**, Brownish-black discoloration, edema, and tenderness involving the anterior third of the tongue bilaterally upon initial evaluation with movement limitation that compromised the patient's speech and deglutition.

56 Gy to the entire chain of cervical nodes. The patient completed treatment for head and neck cancer and attended all follow-up appointments. He had remained disease-free since the end of his treatment in 2008. His medical history was negative for vasoconstrictor use, syphilis, cardiac arrest, or embolization. The patient was not taking any medication.

An initial oral examination revealed edema, tenderness, and bilateral, brownish-black discoloration involving the anterior third of the tongue, with movement limitation that compromised his speech and deglutition. This tongue tissue area was necrotic, without bleeding, and sharply demarcated. The mucosa of the posterior two-thirds of the tongue had a normal appearance. In addition to this tongue alteration, bilateral exposure of necrotic bone of the body of the mandible was detected during oral examination (Figures 1A and 1B). The patient had a dental history of bilateral multiple exodontia (first and second right and left molars), which had been performed by his dentist 12 months earlier. According to the dentist, the reason for dental extractions was caries caused by radiation. The surgical procedure had been performed with the patient under local anesthesia (three cartridges, lidocaine 2% with epinephrine 1:100,000), and there were no complications. According to the dentist, the surgical wound did not heal adequately, and soft tissue breakdown and bilateral necrotic bone exposure occurred 20 days after the surgery. The patient denied symptoms of fever, arthralgia, neurologic alterations, weight loss, visual disturbances, jaw claudication, or temporal tenderness.

Hematologic examination results were within normal parameters (white blood cells: 3.8 kU/L, hemoglobin: 14.4 g/dL, hematocrit: 42.8%, platelet: 250 kU/L). The erythrocyte sedimentation rate and cross-reactive protein were within normal limit (15 mm/hr and 2.5 mg/L, respectively). Panoramic radiograph demonstrated irregular diffuse osteolysis affecting alveolar bone on both sides of the mandible's body and teeth alveoli with delayed remodeling (Figure 2). Because there was a strong diagnostic hypothesis of tongue necrosis as a consequence of prior radiotherapy for head and neck cancer, a tongue biopsy was performed. The specimen revealed soft tissue necrosis without signs of malignancy. In light of the diagnosis, Horton syndrome (temporal arteritis) was excluded because claudication of the legs, localized headache, abnormal temporal artery with tenderness, and decreased pulse were not present, and the erythrocyte sedimentation rate was not higher than 50 mm/hr. A carotid Doppler examination of the neck arteries yielded normal findings. The patient refused to undergo a superficial



Fig. 2. Panoramic radiograph demonstrating irregular diffuse osteolysis affecting the alveolar bone of the mandible bilaterally.

temporal artery biopsy. With regard to the clinical and radiologic findings of the bilateral bone exposure in the mandible, we considered the hypothesis of ORN of the jaw.

The patient's initial management started with hyperbaric oxygen therapy (30 sessions: 20 sessions before surgery and 10 sessions after surgery; 2.4 ATA; 90-minute session in a monoplace chamber). Subsequently, a glossectomy was performed with the patient under general anesthesia, resulting in secondary wound healing (Figure 3). Bilateral marginal bone resections were performed to treat ORN on both sides of the mandible. Microscopic examination of the tongue specimen revealed fibrous proliferation with extensive areas of hyalinization associated with obliterated blood vessel lumen. Microscopic examination of the bone specimens revealed necrotic bone associated with a nonspecific chronic inflammatory process and the presence of colonies of filamentous bacteria. These investigations confirmed the diagnosis of tongue necrosis and ORN caused by radiation therapy for head and neck cancer.

One month after the surgical procedure, the patient responded satisfactorily to treatment with significant improvement in speech and oral food intake and decreased lingual pain. The tongue lesion and ORN of the jaw healed completely over a 6-month follow-up period (Figures 4A-4C).

#### DISCUSSION

Tongue necrosis is generally rare and may be associated with a wide range of pathologies. Differential diagnosis in tongue necrosis should include malignancies (carcinoma, sarcomas, lymphomas, metastatic tumors); Download English Version:

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