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SHORT COMMUNICATIONS AND CASE REPORTS

Massive osteoradionecrosis of facial bones and soft tissues

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Summary

Osteoradionecrosis (ORN) is one of the most serious and uncommon complications in head and neck irradiation for cancer. It is defined as a combination of necrotic soft tissue and bone not being able to heal spontaneously, it demonstrates a general resistance to antibiotics and requires conservative surgical management. Even with modern radiation therapy, its incidence is highly unpredictable and varies between 4-30%. We report on a patient with a huge open cavitation in the cheek, communicating with the mouth and extending to contralateral periodontal gingival and temporal fossa. He had been treated with radiation therapy for nasopharyngeal cancer 5 years ago and presented with restriction of the opening of the mouth. Osteonecrosis complicated with osteomyelitis was evident in bilateral mandible and maxillary bones and the temporal bone. The ramus of the mandible and zygomatic arc were resected, subtotal maxillectomy was performed and the defect was repaired by a free double island flap from the scapular and parascapular osteocutaneous latissimus dorsi muscle flap supplied by subscapular artery. To our knowledge, this is the most extensive bone and soft tissue destruction due to radiation reported in the literature.

Key words: 3D imaging, jaw, osteoradionecrosis, radiotherapy, reconstructive surgical procedures

Introduction

ORN is defined as a pathologic process that develops following irradiation of osseous tissue and is characterized by a progressive involvement of extensive areas of soft and bone tissue with a benign chronic mucosal ulceration with exposure of the jaw-bone of more than 3-month duration. The mandible is the most commonly affected bone; other involved bones are the maxilla, temporal bones and the skull base [1-6]. Herein we describe a patient with extensive soft tissue and bone necrosis on his face due to radiotherapy for nasopharyngeal carcinoma. The defect was reconstructed via a free scapular and parascapular osteocutaneous flap combined with latissimus dorsi muscle supplied by subscapular artery.

Case presentation

A 60-year-old man presented to our clinic because of a pronounced restriction of the opening of the mouth. He had not tenderness. Five years ago he was diagnosed with nasopharyngeal carcinoma (T2N2M0) and was treated with external radiation therapy in another health institute with a total dose of 72 Gy in 36 fractions to the primary tumor and 68 Gy in 34 fractions to the neck nodes. Forty-two months following treatment, a local recurrence in the nasopharynx was diagnosed in another institution and the patient was then given cisplatin 100 mg/m^2 and docetaxel 140 mg/m^2 every 3 weeks. After 4 cycles of chemotherapy he was reirradiated to a total dose of 60 Gy in 30 fractions. Details of chemo- and radiotherapy were not precisely known since these therapies were administered elsewhere. The restriction of mouth opening had started one year after the completion of radiotherapy. The distance between the incisal edges of the maxillary and mandibular incisors was 7 mm, the occlusion was abnormal and the oral hygiene bad. On physical examination a large open wound was detected on his right buccal mucosa. There was no history of

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tooth extraction or trauma before or after radiotherapy. He smoked for 20 years, one pack/day, but stopped smoking 17 years ago. Plain radiograph revealed a large cavitation at the right maxillary sinus, with bone resorption on both maxillae and mandible (Figure 1).

Firstly, he was operated on for ankylosis of right temporomandibular joint (TMJ) but the operation failed because of the massive fibrosis and necrosis of the surrounding tissues, as well as of the buccal mucosa. Then, the coronoid process was excised and fibrous bands were released. Culture and biopsy were taken from the necrotic tissues. Biopsy was negative for cancer. Despite daily dressing, the incision line was detached after the operation. *Pseudomonas aeruginosa*, *Stenotraphomonas maltophilia* and *Candida (non-albicans)* were found in the blood and tissue cultures. Piperacillin, tazobactam and amicacin were given to the patient. As actinomycosis was suspected on tissue pathology, imipenem was added.

A free-flap operation was planed to close the wound. Meanwhile the patient developed cough, respiratory distress complaints and massive phlegm secretion from his tracheostomy canule, and lung tuberculosis was diagnosed. The operation was delayed until the infection was resolved. After 6 months, the lesion had progressed with a large skin and subcutaneous soft tissue defect, a huge wound of 6×4 cm on the right cheek was prominent, the right zygomatic arc and the lateral wall of the maxillary sinus were exposed and partial necroses in osseous tissues were observed.

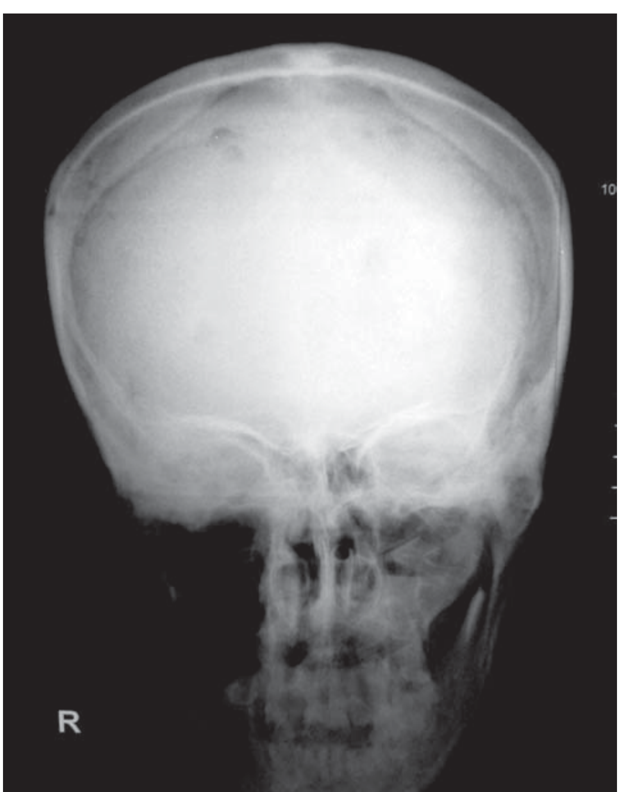


Figure 1. Anteroposterior radiograph shows a large lytic area in the right maxillary and mandibular region.

Peripheral facial paralysis was also present. A computed tomography (CT) scan demonstrated a large cavity beginning at the inferior border of the right zygoma, invading the temporal muscle and extending to the masticator space. The cavity was communicating with the mouth through the erosion of the lateral wall of the maxilla. Necrosis was also present in the periodontoid soft tissue of the left mandible, extending to the contralateral temporal muscle. There were many lytic areas in the anterior and lateral walls of the right maxillary sinus, posterior wall of the left maxillary sinus, left pterygopalatine plate and nasal crest. Right mandibular head and body had cortical irregularities, fragmentations and lytic areas, associated with medullary soft tissue and air bubbles indicating osteomyelitis. The right temporal part of the temporal bone had some subcortical lytic areas, as well as air was detected in the TMJ secondary to surgery. Magnetic resonance imaging (MRI) showed destruction of soft tissues without tumor recurrence (Figure 2). CT scan with 3D-volume rendering images was carried out to evaluate the extent of the necrosis before orofacial procedure (Figure 3). Subtotal resection of the right mandibular ramus, resection of the zygomatic arch, and subtotal resection of the maxilla with preservation of the infraorbital rim were performed. A double skin island of scapular-parascapular osteocutaneous free flap combined with latissimus dorsi was used to reconstruct the defect of resection (Figure 4).

Discussion

The pathophysiology of ORN is simply a radiation-induced wound healing defect [2]. Histopathological findings are vascularization and revascularization of the mandible which indicate that ORN is an ischemic necrosis caused by radiation-induced obliteration of the inferior alveolar artery and branches of this artery, whereas revascularization from branches of the facial artery is disturbed by radiation-induced vascular disease and periosteal damage [3]. The reduction in the vascularization induced by radiation makes the bone much more susceptible to infection, so that it can develop infected ORN spontaneously or following surgical procedures [4,5]. Finally destruction of bone marrow and reduction or complete loss of bone vitality occurs. The high content of calcium of the bones results in higher absorption of radiation than the surrounding soft tissues,

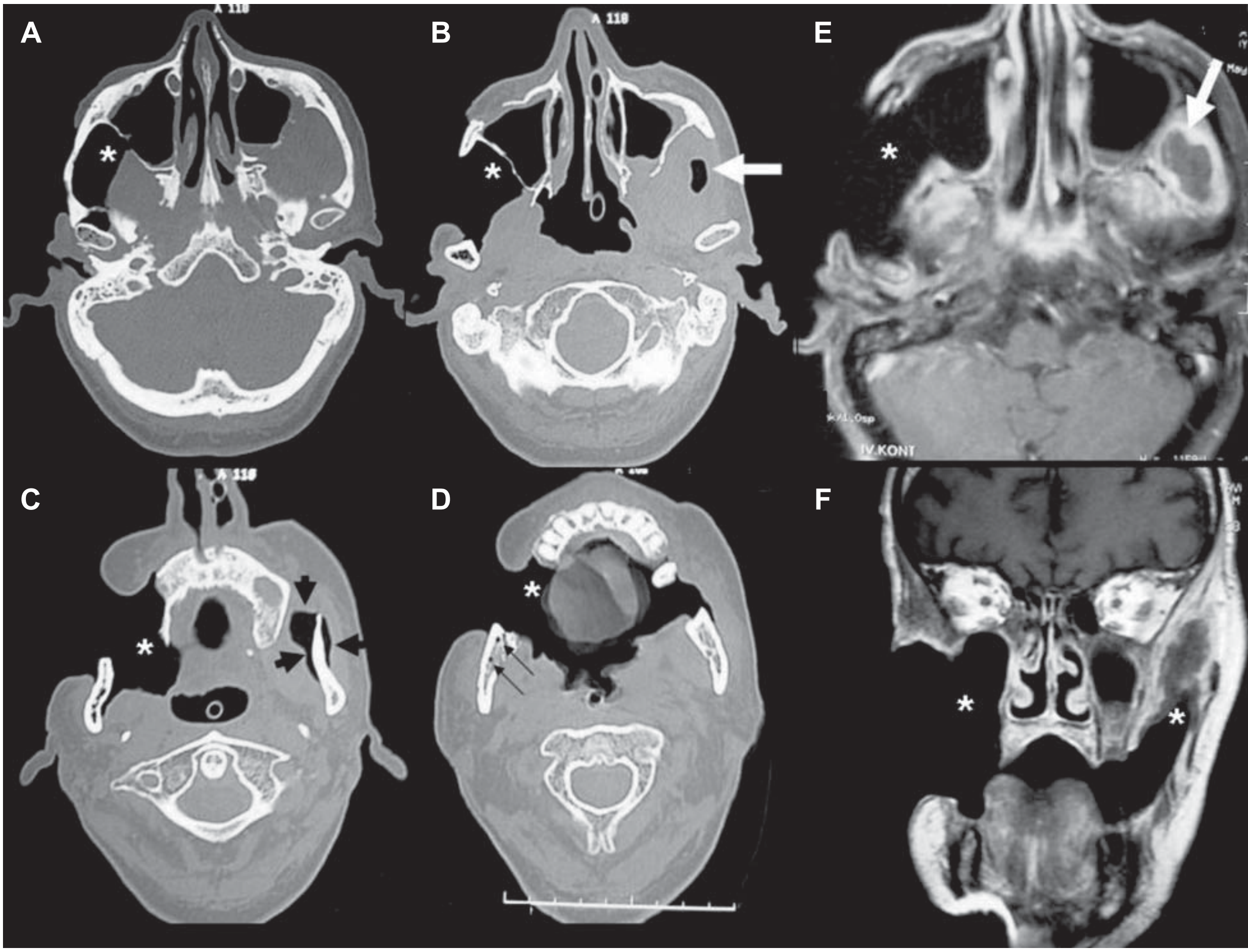


Figure 2. Axial CT **(A-D)** and contrast enhanced T1-weighted axial and coronal MRI **(E, F)** images: The cavity (asterisk) begins at the inferior border of the right zygoma, invades the temporal muscle and extends to the masticator space. The cavity communicates with the mouth through the erosion of the lateral wall of the maxilla. Necrosis is also present in the periodontoid soft tissue of the left mandible (black arrows). Lytic areas are seen in the anterior and lateral walls of the right maxillary sinus, posterior wall of the left maxillary sinus and the pterygopalatine plate. The right temporal part of the temporal bone has some subcortical lytic areas (thin black arrows in **C**). Contrast-enhanced MRI scans **(E, F)** demonstrates the continuum of the cavity with the oral cavity, contralateral periodontoid and temporal soft tissues. The left temporal muscle shows peripheral enhancement (white arrow).

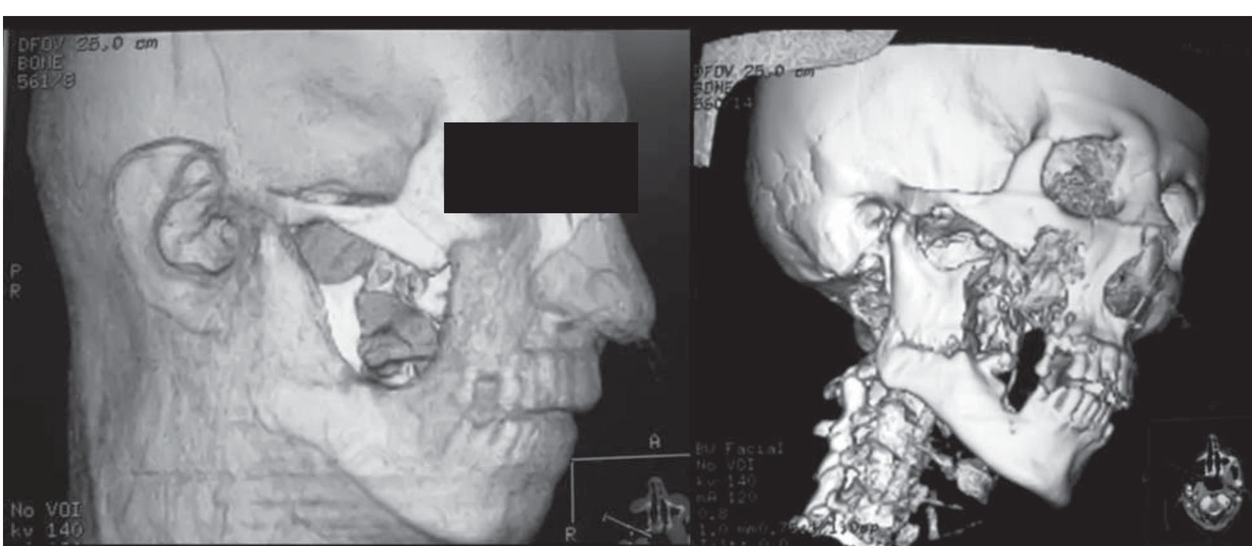
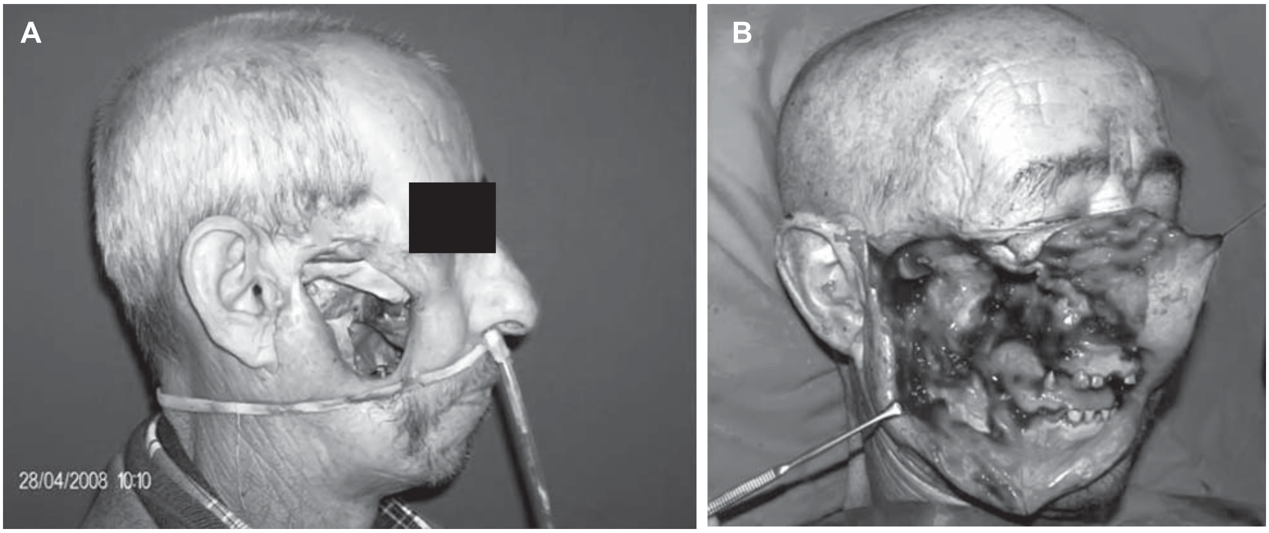


Figure 3. Three dimensional volumetric computed tomography with soft tissue and bone algorithms of the maxillofacial region.



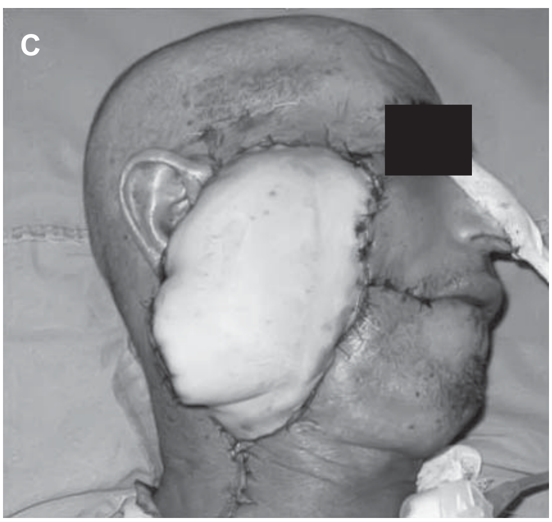


Figure 4. A. Preoperative photograph of the patient shows a large soft tissue defect on his right cheek. Arcus zygomaticus, ramus of the mandibula and right maxillary bones are exposed. **B.** Intraoperative view shows the large defect. **C.** The defect is reconstructed by the scapular-parascapular osteocutaneous free flap combined with latissimus dorsi muscle.

subsequently secondary radiation produces increased damage of the tissues [6].

Seventy-five percent of the cases with infection had surface organisms with candida species and streptococci predominating. The amount of necrotic tissue present and the poor vascularity of the surrounding tissue make high antibiotic levels in the affected tissue difficult to achieve. Finally, if ORN cases are infected, the condition can become rapidly progressive, destructive and difficult to treat [5].

Trauma is another entity that has been strongly implicated in the pathogenesis of ORN. Common causes of trauma to the bone or overlying soft tissue include fractures, post-irradiation dental extractions, ill-fitting dental prostheses and even coarse food [1]. However, such tissue may break down when injured or even become necrotic in the absence of trauma [2].

The anatomic differences of the mandible and the maxilla may be a factor in irradiation effect differences. The mandible contains more compact bone and, because of the scattering of the rays, the osteocytes of the mandible may receive more irradiation. Its superficial location in the path of external radiation predisposes to radiation injury. In addition, the blood supply of the mandible is composed of one vessel with the inferior alveolar artery encased in bone. There is not much room for expansion when inflammation and edema occur in the mandible. On the other hand the blood supply in the maxilla is more diffuse; many more anastomoses tend to nourish any bone whose blood supply is diminished. The most vulnerable part of the mandible is the buccal cortex of the premolar, molar, and retromolar regions [4,5-8]. Other involved bones include the maxilla and rarely the skull base, the sphenoid and the temporal bones [1].

The patient's dental status is another significant risk factor for development of osteonecrosis [1]. Patients who are dentulous are at greater risk compared with edentulous patients or those who had no tooth extractions. The reason is that, due to the high calcium content, teeth in the line of radiation can act as secondary sources of radiation to adjacent bone, as can metallic restorations within them [1,9]. Fortunately, in many cases, devitalized bone fragments will be sequestered and lesions will spontaneously heal. However, when ORN is progressive, it can lead to intolerable pain, fracture and necessitate jaw resection [8-10]. Despite such massive necrosis, the absence of tenderness suggests radiation damage in maxillary and mandibular nerves in our patient. The risk for developing a spontaneous ORN is somewhat unpredictable and related to the aggressiveness of radiotherapy, radiation dosage and delivery, fractionation, elapsed time since radiation, the proximity of the tumor to the mandible, tumor size, volume of mandible within the radiation field, and use of bilateral opposed portals. ORN has been rarely seen with doses below 5000 cGy [1]. Individual patient sensitivity should also be considered in risk factors. In our patient the development of a late damage of radiation was not a surprising finding. The fraction size was 2 Gy. In patients receiving radical doses of radiotherapy for nasopharyngeal carcinoma, the fractional dose should be similar or equal to the conventional dose of 2 Gy, if logistically feasible. At all times, pre- and post-irradiation oral and dental care forms an important aspect of the management plan. This may help prevent the rare complication of extensive maxillary ORN, particularly for patients receiving more than one course of radiotherapy for nasopharyngeal carcinoma.

The benefits of adjuvant hyperbaric oxygen therapy is still controversial in reconstructive surgery [11,12]. When the soft and hard tissue defect is small, the fibula-free osteocutaneous flap transfer has a high success rate and low perioperative complication rate, making it an ideal choice for reconstruction of the bony defect [13-16]. In cases with large soft-tissue requirements the scapular area is the choice of the donor site. However, the management and placement of large amount of soft and bone tissues in an appropiate position is extremely difficult. Even if the flap remains viable and well-perfused, a persistent wound healing deficiency might prevent complete healing between host connective tissues and the transfer flap [12].

This case is a representetative and educational one, demonstrating the cascade damage of the radiation which was reconstructed by scapular-parascapular osteocutaneous free flap.

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