clinical presentation of distichiasis following the transconjunctival approach is being reported.<sup>4</sup>

The transconjunctival approach was popular since 1924 when it was first described by Bourquet for lower eyelid blepharoplasty.<sup>4</sup> Complications reported in various studies include entropion, ectropion, eyelid avulsion, conjunctival pyogenic granuloma, tarsal plate injury, canthal malposition, eyelid retraction, lacrimal injury, corneal abrasion, and lower eyelid buttonhole and laceration.<sup>5</sup> Entropion should be differentiated from distichiasis with malpositioning of eyelid and time of occurrence. Another entity to be noticed and differentiated is trichiasis, a condition in which misdirected lashes are seen in the anterior lamella of the lid.<sup>3</sup>

Treatment of distichiasis is challenging and considerable in number in literature, including the tarsoconjunctival trap door excision<sup>6</sup> or resection of the distal part of the tarsal plate with or without mucous membrane grafting.<sup>1,7</sup> Cryotherapy was advocated as a measure of ablating eyelashes in the previously failed electrolysis treatment or when less than a quarter lid margin was involved.<sup>8</sup> Eyelid division along with cryotherapy to posterior lamella is another approach introduced to prevent depigmentation caused by cryotherapy alone.<sup>6</sup> Epilation and electrolysis can be employed when few accessory cilia are seen, and often requires repeated treatment as lashes regrow within 4 to 6 weeks.

Direct tarsal strip excision offered an effective and safe treatment for distichiasis. As a thin strip of tarsus of 2 mm was excised, regeneration of lid margin occurred without any functional or anatomic deformity. The type of treatment depends on the extent of the lid margin involved and the number of lashes present. Lashes distributed evenly throughout the length of the eyelid require extensive management (Supplementary Digital Content, Fig. 3, http://links.lww.com/SCS/C843).

The introduction of video meibography enables a better understanding of anatomic changes associated with meibomian glands, and the documentation with slit-lamp examination may facilitate to assess the impact of the transconjunctival approach in developing distichiasis.<sup>9</sup> Trauma to tarsus or meibomian gland also triggers to revert to hair-bearing function causing distichiasis.<sup>10</sup> Placing the transconjunctival incision a few millimeters below the tarsal plate along with an atraumatic dissection anterior to the orbital septum,<sup>11</sup> and also avoiding injury to the meibomian glands, this complication can be better prevented.

### CONCLUSIONS

Distichiasis occurred as a complication of the transconjunctival approach is uncommon. It is imperative to understand the complication of this approach before the development of ocular infection, even though the occurrence of the condition is rare. The prudent surgical management for distichiasis should be emphasized according to the extent of lid-margin involvement.

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# OPEN

# Surgical Management of Bilateral Osteoradionecrosis of the Mandible

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**Purpose:** The purpose of this study was to evaluate our 10 years clinical experience in surgical management of patients with bilateral osteoradionecrosis (BORN) of the mandible in head and neck malignancies patients.

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Materials and Methods: The authors reviewed 22 patients with bilateral mandibular bone mineral density changed in image who had failed to respond to conservative treatments. They were treated by radical resection and reconstruction with free flaps immediately or second-stage at our institution between January 2008 and January 2018. Results: Nine patients received immediate bilateral mandibular radical resection. Six bone flaps (4 fibula osteocutaneous [fibular OC], 1 fibular OC + pectoralis major myocutaneous flap [PMMF] and 1 fibular OC + anterolateral thigh flap [ALTF]) and 3 soft flaps (1 PMMF, 1 PMMF + titanium plate and 1 ALTF) were used. Three (33.3%) of these patients complications occurred in the immediate postoperative period, but all patients have an acceptable follow-up outcomes. In remaining 13 patients who only experienced immediate unilateral mandible resection for the first time. Complications occurred in 1 patient (7.7%), and all patients have a good outcome in the immediate postoperative period. In follow-up, 1 patient titanium plate exposed, and 6 patients (46.2%) contralateral mandible ORN developed that underwent radical resection in second time. Three fibular OC, 2 PMMF, and 1 latissimus dorsi myocutaneous flap were used. The overall outcome of our experience with the use of bone or soft tissue transfers in managing BORN of mandible is encouraging.

**Conclusions:** An individualized management plan should be given for each patient depending on their own local and general condition. Radical resection followed by vascularized flaps reconstruction is an acceptable and reliable procedure for patients with BORN of the mandible.

Key Words: Bilateral, mandible, osteoradionecrosis, reconstruction

steoradionecrosis (ORN) of the mandible is one of the most serious and dreaded complications of radiotherapy alone or in combination with surgery in head and neck malignancies. The exact incidence of these complications is in fact unknown due to different radiation regimens, fractionations, and delivery methods.<sup>1</sup> In 1984, Marx<sup>2</sup> defined ORN as "an area greater than 1 cm of exposed bone in a field of irradiation that had failed to show any evidence of healing for at least 6 months" and put forward his "hypoxic-hypocellularhypovascular, 3H" etiology theory. However, this definition is not entirely accurate, because ORN can be shown in radiographically without any defects of oral mucosa or facial-neck skin.<sup>3</sup> In 2012, Marx pointed out again that ORN as noninflammatory diseases, caused by a high linear energy transfer. Radiation impairs or kills numerous cell types in the field of radiation including periosteum, jaw bone, and all soft tissue, and result in devitalization of these tissues, which may lead to bone or soft tissue necrosis.<sup>4</sup>

In early and ORN necrosis localized cases, conservative approaches, such as improvement of oral hygiene, use of antibiotics, hyperbaric oxygen, and antifibrotic drugs, may be effective and sufficient.<sup>5–8</sup> However, patients who develop advanced disease with bone exposure, mucosal or/and external skin defects, intractable pain, trismus, and even pathological fracture, conservative treatment may be inadequate. In these circumstances, radical resection of the necrosis involved mandible with nonirradiated, well-vascularized tissue reconstruction is necessitated.<sup>9–11</sup> In recent years, with the development of microvascular techniques to transfer vascularized bone and soft tissues, various methods, such as fibula osteocutaneous (fibula OC), iliac crest, scapula, pectoralis major myocutaneous (LDM) are used for reconstruction

of mandibular defects. Free flap reconstruction of advanced ORN defects can provide acceptable aesthetic and functional results although existence of some complication.

The management of advanced ORN is not an easy job, currently, still remains a difficult and controversial issue. Management of bilateral osteoradionecrosis (BORN) of the mandible, absolutely, is a unique challenge. So the purpose of this study is to review our experience in management of patients BORN of the mandible in head and neck malignancies patients in the past 10 years. We hope that this study can provide some insight to management of BORN of the mandible.

# MATERIALS AND METHODS

We retrospectively reviewed 22 BORN of the mandible patients who received well-vascularized tissue reconstruction in immediately or secondary stage at the Department of oral maxillofacial-Head and Neck Oncology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, China, between January 2008 and January 2018. Data regarding the patient's parameters of primary tumors, radiotherapy, BORN treatment surgical procedures, postoperative complications, therapeutic outcome, and follow-up information were collected from the inpatient and outpatient medical records. The diagnosis of BORN was determined by clinical and radiographic findings. All patients had bilateral mandible bone mineral density changed in panoramic radiographs or computed tomography (CT), with or without mucosal or/and external skin defects. All patients were confirmed by CT scan as BORN of the mandible. All 22 patients received radiotherapy consisted of external beam radiation and linear accelerator (4 MeV). The dose rate/day varied from 1.8 to 2.0 Gy, and 5.0 fractions/week.

The extent of mandibular resection was base on preoperative clinical and image findings, as well as the macroscopic assessment by the time of intro-operation. The macroscopic assessment was determined by removing the necrotic bone until normal bleeding bone appeared. The necrotic surrounding soft tissues were excised together with the necrotic bone. The postoperative bone and soft tissue defects were reconstructed by nonirradiated, well-vascularized bone or/and soft flap. Most of patients underwent tracheotomy to facilitate airway management. Postoperative, all patients received enteral nutrition by a temporary feeding tube for a period of 2 to 3 weeks, until they could swallow fluid food by themselves.

The long-term outcomes were evaluated through follow-up. The criteria for resolution of BORN included absence of pain, no soft tissue defects, and mouth opening improved and bone density stable on radiography during follow-up period.

## RESULTS

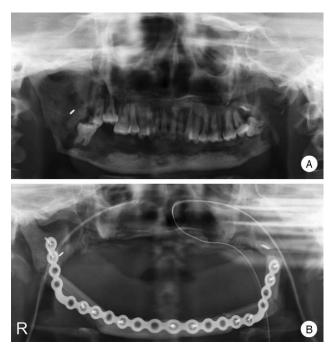
The patients (13 male, 9 female) ranged in age from 31 to 76 years (mean, 51.5 years). The most frequent primary tumor was nasopharyngeal carcinoma (59.1%), followed by oral carcinoma (22.7%). Four of 5 oral carcinoma patients were treated with a combination of surgical procedures and radiation therapy (with/ without chemotherapy). The remainder of oropharyngeal and nasopharyngeal carcinoma patients were treated with radiotherapy (with/ without chemotherapy) alone. And the total radiation dosage ranged from 60 to 158 Gy (mean, 100 Gy; medial, 110 Gy). Of the 22 patients, primary hypertension was found in 4 patients, coronary disease in 1 patient, and diabetes in 1 patient. The interval between the termination radiation of primary tumor and clinical diagnosis of ORN ranged from 3 months to 16 years (mean, 5 years 4 months; medial, 4 years). The LENT-SOMA scores ranged from 1 to 3 (mean, 2.45; medial, 3). The VAS scores ranged from 1 to 8 (mean, 5; medial, 6). The detailed information was shown in Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C884.

The most common clinical findings included pain, limitation of mouth opening, and soft tissue defects (mucosal or/and external skin). Not all patients' soft tissue defects occurred on the bilateral side of mandibular area, 9 cases occurred in unilateral side (Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C885). Of the 22 patients, 9 received immediate bilateral mandibular radical resection (Fig. 1) and 13 received immediate unilateral mandibular resection. In bilateral resection groups, most patients (66.7%, 6/9) postoperative mandibular defects were reestablished mandibular continuity successfully by fibula OC, and 2 of these patients used composite flaps (fibula OC + PMMF, fibula OC + ALT). In addition, soft tissue flaps were used to restore intraoral lining or external skin defects in another 3 patients: 1 PMMF, 1 PMMF + titanium plate, and 1 ALT. Complications occurred in 3 (33.3%) of these 9 patients in the immediate postoperative period. Fistula and dehiscence of the partial mucosal suture line occurred in 1 composite flap (fibular OC + PMMF), and this problem was resolved with local wound care at postoperative day 23. Titanium plate was exposed in a patient (PMMF + titanium plate) due to local infection at postoperative day 11, second operation, removing the Titanium plate and local wound care, and was done and healed. Neck infection which let to fistula occurred in a fibular OC patient, and also resolved with local wound care. No donor site complications were noted. The mean length of hospital stay was 19 days (range, 13-33 days).

Of the 13 immediately unilateral mandible resection patients, 9 patients (69.2%, 9/13) postoperative defects were reconstructed with microvascular bone transfers consisting of fibula OC (7), iliac crest bone only (1), iliac crest bone plus muscle and skin (1). The rest 4 patients (30.8%, 4/13) were repaired with soft tissue flaps using PMMF (3) and PMMF + titanium plate (1). Neck infection occurred in 1 fibula OC transfer patient in the immediate post-operative period. This patient infection was resolved early with local wound care and reasonably antibiotics support. None of this patients occurred donor site complications. All patients initially received postoperative enteral tube feeding. The mean length of hospital stay was 15 days (range, 9-19 days).

The mean follow-up was 1 year and 2 months (range, 5-36 months). There was base on clinical and radiographic findings, no infection, no bone exposed, and no ORN or tumor recurrence in all bilateral mandibular resection patients (100%, 9/9). Obviously, the bone flap reconstructed groups had better speaking intelligibility, food oral intake, and facial symmetry than soft flaps one. In the unilateral mandibular resection groups, 6 (46.2%) patients have satisfactory cosmetic and functional results and contralateral mandible ORN stable in the follow-up period (Figs. 2 and 3). It is to be regretted in 7 patients, of which 1 titanium plate exposed, and 6 (46.2%) contralateral mandible ORN developed including soft tissue defects (n = 3) and pathological fracture (n = 3) that required second-stage operative intervention (Fig. 4). So 3 fibula OC, 2 PMMF, and 1 LDM flap were used for reconstruction of postoperative defects. No patients died from recurrent tumor or distant metastasis. The detailed management information for BORN of the mandible was shown in Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C885.

In these 22 patients, 4 methods were used, including bone flap repair, bone flap plus soft tissue flap, soft tissue flap, soft tissue flap plus titanium plate repair. Among them, 12 (54.5%) cases were repaired with bone flap, and 3 cases were repaired with bilateral fibula by second-stages. Four (18.2%) cases were repaired by combination of bone flap and soft tissue flap, of which 2 cases were repaired with soft tissue flap by second-stages; 4 (18.2%) cases were treated with soft tissue flap alone, 2 of them were treated with soft



**FIGURE 1.** Preoperative (A) and postoperative (B) panoramic radiograph (clinical presentation 1). The figure illustration of bilateral segmental defects reconstructed with a single fibular free flap.

tissue flap by second-stages, and the other 2 (9.1%) were treated with soft tissue flap plus titanium plate. The detailed management information for BORN of the mandible was shown in Supplementary Digital Content, Table 3, http://links.lww.com/SCS/C886.

#### DISCUSSION

Bilateral osteoradionecrosis of the mandible is a very serious but not common complication of radiotherapy for head and neck



**FIGURE 2.** Preoperative photograph and panoramic radiograph (clinical presentation 6). The figure illustration of bilateral mandible bone mineral density changed in panoramic radiograph (C), and with right (A) or without left (B) external skin defect.



**FIGURE 3.** Postoperative photograph and panoramic radiograph (clinical presentation 6). The figure illustration of unilateral resection defects reconstructed with a fibular free flap. There was a satisfactory result in operation area (A and C) and no evidence show that the contralateral mandible ORN developed (B and C) in the follow-up period. ORN, osteoradionecrosis.



**FIGURE 4.** Postoperative panoramic radiograph (clinical presentation 21). The figure illustration of unilateral resection defects reconstructed with a fibular free flap in the first time (right), and second-stage operative intervention was given when the contralateral mandible ORN developed (left). ORN, osteoradionecrosis.

malignancies. With the development of irradiation techniques and oral health, such complications are increasingly rare in recent years. But once it occurs, the management will be the headache jobs for oral and maxillofacial surgeon. In our series, all (100%) patients received bilateral mandibular areas irradiation and the total radiation dosage ranged from 60 to 158 Gy. Indubitably, the higher radiation dosage delivery and bilateral mandibular areas irradiation must be the key risk factors for this complication occurred. Not all patients with BORN of the mandible have the same serious necrosis of bone or soft tissues in bilateral mandibular bone mineral density changed in panoramic radiographs image or CT scan), 9 combined bilateral soft tissues but 13 only have unilateral soft tissue defect does not necessarily represent the severity of bone necrosis, but soft tissue defects must be considered in the treatment plan.<sup>10,12</sup>

No article has been reported on the management of BORN of the mandible. Several factors including general condition and expectation of the patient, location and size of bony and soft tissue defects, local recipient vessels condition, donor sites and primary tumor recurrence, etc must be taken into account before making treatment plan. Undoubtedly, BORN of the mandible, conservative treatment including oral hygiene improvement, antibiotics, hyperbaric oxygen may be inadequate and radical resection is imminent. According to the ORN resection consensus, surgery must excise the necrotic bone until normal bleeding bone appeared, at the same time including surrounding necrotic soft tissues.<sup>13–16</sup> Resection is an easy job, but how to repair the resected mandibular defect is a unique challenge. It is important to choose a bone flap that can be restored the continuity of the mandible or a soft flap that can be resurfaced the intraoral lining and extraoral skin. In 9 immediate bilateral resection patients, we used 6 fibula flaps, of which 4 were osteocutaneous and 2 were composite flaps. None of the fibula flap lost, only 1 fibular OC and 1 composite flap fistula and dehiscence occurred in the immediate postoperative period. In 13 immediate unilateral resection patients, we used 9 bone free flaps, of which 7 fibula OC flaps and 2 iliac crests. Neck infection occurred in 1 fibula OC transfer patient in the immediate postoperative period. Because of long bone length (maximum, 25-27 cm) and long vascular pedicle (maximum, 12-15 cm) can be obtained, increased the incidence of donor site complications.<sup>15</sup> We agree with others that mandibular ORN postoperative bone defect reconstruction with a fibula free flap can provide adequate cosmetic and functional results, especially the defect involves the anterior arch.<sup>10,13,15,17</sup> In our experience, for the bilateral mandibular defects reconstruction, single fibular free flap is the first and unique-choice. Jacobson et al<sup>18</sup> also reported that using a single fibular free flap reconstruction of BORN of the mandible. In contrast, for the unilateral mandibular defect reconstruction, fibular free flap is an optimal candidate. It can provide adequate length, height, and strength of bone for dentures or implants. If fibular flap unsuitable, other flaps such as iliac crest or scapula can be used.<sup>10</sup>

The ideal nonirradiated, well-vascularized tissue for the replacement of the missing mandible is vascularized bone flap. However, not all patients are optimal candidates, patients with the poor general condition, venerable age, lower life expectation, or posterior mandibular defect, reconstruction with vascularized soft tissue flap must be considered. In our study, we used 6 PMMF (2 + titanium plate) and 1 ALT flap in the first time reconstruction. The 2 titanium plate used patients, 1 titanium plate exposure in a short time another postoperative 13 months. So we believe that it is unwise to application titanium plant combined with a soft tissue flap for reconstruction of mandibular defect in radiotherapy patients. We agree with the others that the well-vascularized soft tissue can provide an expedient means for primary wound healing, while minimizing morbidities associated with more complex reconstruction.<sup>19,20</sup>

With a mean follow-up after reconstruction 1 year and 2 months, all bilateral mandibular resection patients experienced complete healing and resolution of symptoms. However, in the unilateral resection group, 6 patients ORN stable, only 6 patients who contralateral mandible ORN developed that require second-stage operative intervention. So 3 fibula OC, 2 PMMF, and 1 LDM flap are used in the second-stage reconstruction. Nevertheless, the overall outcome of our experience with the use of bone or soft tissue transfers in managing BORN of mandible is encouraging. The bone flap reconstructed groups had better speaking intelligibility, food oral intake, and facial symmetry than soft flaps one.

In summary, for BORN patients who with bilateral bone and soft defect in the same time, we suggest that do bilateral resection and reconstruction with vascularized bone or combine flap and first choice is fibula OC. In contrast, do unilateral resection which soft tissue defect occurred in the first stage, second-stage operation intervention only when the other side ORN developed. Bone flaps are first choice for reconstruction of BORN postoperative defect, especially fibula OC. For patients who are in poor local and general condition, or have a posterior mandibular defect, reconstruction with tissues flaps may be a good choice.

In conclusion, the principle management of BORN of the mandible is making an individualized plan for each patient depending on their own local and general condition. Well- vascularized bone or soft tissue flaps transfer for reconstruction of BORN of the mandible can obtain a good wound healing or acceptable aesthetic and functional results.

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# Cerebellum Tumor Presenting Itself With Positional Vertigo and Benign Paroxysmal Positional Vertigo

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**Abstract:** The purpose of this case is to describe the positional vertigo observed in a patient diagnosed with cerebellar arteriovenous malformation, pay attention to the importance of medical history taking and physical examination in vertigo patients.

A 51-year-old patient went to the Ear, Nose, and Throat clinic with a complaint of vertigo. His vertigo was like peripheral vertigo at the beginning. Dizziness was triggered by head movements. He experienced tinnitus in the left ear during vertigo attacks. The patient also had neck pain. In physical examination, natural bilateral tympanic membrane and facial examination were observed. Other physical examinations were normal. In the positional vertigo tests, the right Dix-Hall Pike test was positive and a downbeating geotropic nystagmus was found. The patient was treated with canalith repositioning maneuver (Epley maneuver). Oral medical treatment started and after 4 days, the patient reported that his gait balance was disturbed and his neck pain continued. After that magnetic resonance imaging was requested. Magnetic resonance imaging was consistent with cerebellar arteriovenous malformation. The patient was consulted to the neurology service.

Cerebellar arteriovenous malformation had features like to peripheral vertigo, and the correct diagnosis is made due to suspected headache and other neurological symptoms.

**Key Words:** Arteriovenous malformation, cerebellum, medical history taking, positional vertigo, tumor

**P** ositioning nystagmus is defined as the nystagmus generated by a change in head position with respect to gravity. It is classified

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Informed consent was obtained from the participants included in the study. The authors report no conflicts of interest.

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