Extrahepatic metastases from HCC are associated with a poor prognosis, with a mean survival of 7 months and a 1-year survival rate of 24.9%.[3] In 0.8% of patients with a form of internal carcinoma, cutaneous metastasis is the presenting sign. Four percent of cutaneous metastatic lesions occur on the scalp, and 6% on the face.[5] Cutaneous metastases of HCC may appear as rapidly growing nodules on the scalp, chest, or shoulder. They may be single or multiple, firm, painless, nonulcerative, and reddish blue nodules, typically 1 to 2.5 cm.[6] They may present similarly to basal cell carcinoma.

The hemorrhagic nature of metastatic HCC has been widely reported. Metastatic lesions to the skull have been associated with spontaneous epidural hemorrhages.[7] Spontaneous rupture of pleural metastases of HCC has been shown to cause massive hemorrhage, leading to hemothorax.[8] Kamiyoshihara and colleagues[9] reported a case of massive bleeding of biopsied rib tumor that was shown to be metastatic HCC; hemostasis of the biopsy site was achieved only by complete excision of the diseased rib. Cases of postoperative hemorrhage have also been described, including a case of massive hemorrhage after biopsy of a metastatic lesion to the mandible.[10]

The hemorrhagic quality of a metastatic HCC tumor is thought to be due to 2 distinct processes. First, patients with HCC and cirrhosis have declining liver function that affects the regulation of hemostasis on many levels.[11] The fibrotic changes associated with cirrhosis cause a decline in the synthetic function of the liver including a decline in production of coagulation factors such as factors V, VII, IX, X, and XI, as well as prothrombin and anticoagulation proteins such as protein C, protein S, and antithrombin.[4,12] The decreased production of coagulation factors and anticoagulation proteins disturbs the delicate balance of hemostasis and can lead to both hypercoagulable and coagulopathic states.[12] Declining liver function also affects hemostasis by causing a rise in nitric oxide, leading to decreased vascular tone.[13] There is thrombocytopenia due to increased splenic sequestration from splenomegaly. Declining renal function usually accompanies advancing liver failure, causing acquired platelet dysfunction.[12] These pathologic changes can tip the balance to cause a bleeding diathesis.[12]

In addition to the changes in liver function affecting the coagulation cascade, characteristics of the metastatic HCC tumor itself contribute to abnormal hemostasis. Hepatocellular carcinoma and its metastases are typically vascular in nature, with 1 study finding 89.2% of the tumors to be hypervascular. When a metastatic lesion becomes hemorrhagic, it becomes increasingly difficult to attain hemostasis. When standard options fail to control the hemorrhage, radiotherapy has been used to successfully stop the bleeding in a number of cases.[10,14,15]

Although cutaneous metastatic HCC is rare, it should be considered when evaluating a skin lesion in a patient with known cirrhosis or hepatitis. The hemorrhagic nature of metastatic HCC should be appreciated, particularly before any attempted manipulation or resection of the metastatic lesion.

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Unusual Odontogenic Keratocyst of the Maxillary Sinus

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Abstract: An odontogenic keratocyst that eroded into the sinus through the maxillary bone and occupied it, showed replacement of the sinus respiratory epithelium by lesional epithelium, and was associated with fungal rhinosinusitis is presented. A review of the literature disclosed that epithelial replacement has been described in 2 previous case reports, although there is no report on the coexistence of odontogenic keratocyst with fungal rhinosinusitis.

Key Words: Odontogenic keratocyst, maxilla, fungal rhinosinusitis

Odontogenic keratocyst (OKC) is established as a distinct entity because of its specific microscopic features and aggressive behavior that manifests with infiltration of adjacent anatomic

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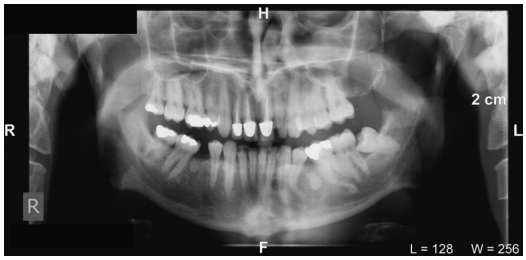

FIGURE 1. Panoramic radiograph shows well-circumscribed radiolucent lesion apical to the first molar tooth.

structures and high recurrence rate. This behavior is emphasized by the introduction of the term *keratocystic odontogenic tumor* in the recent classification of the World Health Organization,[1] although its neoplastic nature is debated. We describe an OKC that eroded into the sinus through the maxillary bone and replaced the sinus respiratory epithelium, while it was associated with fungal rhinosinusitis.

CLINICAL REPORT

A 38-year-old man presented with intermittent pain, swelling, and a feeling of "fullness and pressure" on the left side of his face that he had first noticed about 1 year ago and got worse during scuba diving. His medical history was significant for chronic rhinosinusitis that had repeatedly been treated with administration of antibiotics and local corticosteroids. He was otherwise healthy and not in any other kind of medication.

Clinical examination revealed a hard, nonfluctuant, and slightly tender swelling covered by normal mucosa in the maxillary vestibule, distant to the first molar tooth. Regional lymph nodes were not palpable. According to the referring dentist, the associated teeth were vital. Panoramic radiograph showed a well-circumscribed radiolucent lesion apical to the first molar tooth (Fig. 1).

A provisional diagnosis of OKC was made, and an incisional biopsy was performed under local anesthesia. Intraoperatively, the cystic cavity was found to contain a whitish material. Microscopic examination of 5-μm-thick, formalin-fixed, and paraffin-embedded tissue sections showed connective tissue lined by a uniformly thin parakeratinized epithelium without rete ridges that had a corrugated parakeratinized surface and a basal layer composed of columnar cells with reverse nuclear polarity (Fig. 2). Focal separation of the epithelium from the connective tissue was seen. The connective tissue was mildly infiltrated by inflammatory cells, predominantly lymphoplasmacytes. The diagnosis was OKC.

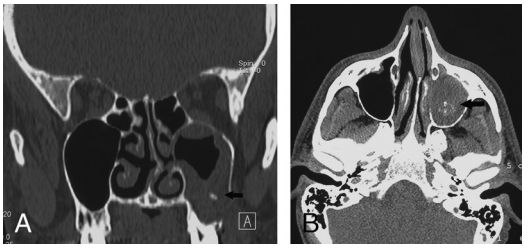

FIGURE 3. A, Computed tomography scan, coronal view, reveals diffuse radiopacity mainly toward the floor of the left maxillary sinus, bone resorption at the anterolateral wall of the maxilla close to the alveolar process, and microcalcifications (arrows). B, Computed tomography scan, sagittal view, reveals diffuse radiopacity mainly toward the floor of the left maxillary sinus and microcalcifications (arrows).

A computed tomography scan revealed a diffuse radiopacity mainly toward the floor of the left maxillary sinus. Bone resorption was evident at the anterolateral wall of the maxilla close to the alveolar process (Fig. 3). Radiopacity of the sinus with microcalcifications or "metallic dense" spots were interpreted as "consistent with aspergillomas."

The cyst was enucleated, the sinus cleaned through a Caldwell-Luc approach, and a drain was placed through a rhinoantrostomy. The patient was administered amoxicillin 500 mg plus clavulanic acid 125 mg, 3 times daily, for 5 days. The drain was removed after 72 hours; his postoperative recovery was uneventful, and 19 months after operation, he is free of disease.

Macroscopically, the cyst measured about 3.5 × 2 × 1 cm. Microscopic examination confirmed the diagnosis of OKC. Focally, the epithelium showed increased cellularity and drop-shaped rete ridges, whereas the cells in the basal third had large and hyperchromatic nuclei (Fig. 4). Mitoses were not identified. In addition, there was a focus of abrupt transition of the cystic epithelium to respiratory pseudostratified columnar epithelium that gave the impression of active replacement or "pushing" (Fig. 5). The connective tissue of the cystic wall was vascular, edematous, with foci of calcifications, cholesterol crystals, and mild lymphoplasmacytic infiltration, whereas no granulomatous

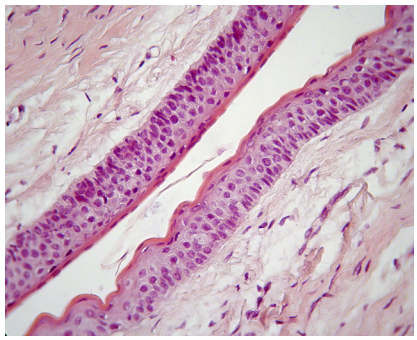

FIGURE 2. Connective tissue lined by a uniformly thin parakeratinized epithelium without rete ridges. Notice the corrugated parakeratinized surface and a basal layer composed of columnar cells with reverse nuclear polarity (hematoxylin-eosin stain, original magnification ×400).

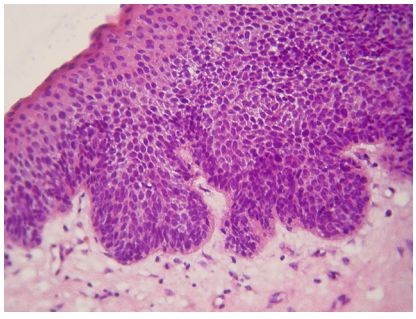

FIGURE 4. Odontogenic keratocyst epithelium shows increased cellularity, drop-shaped rete ridges, and cells with hyperchromatic nuclei in the basal compartment (hematoxylin-eosin stain, original magnification ×400).

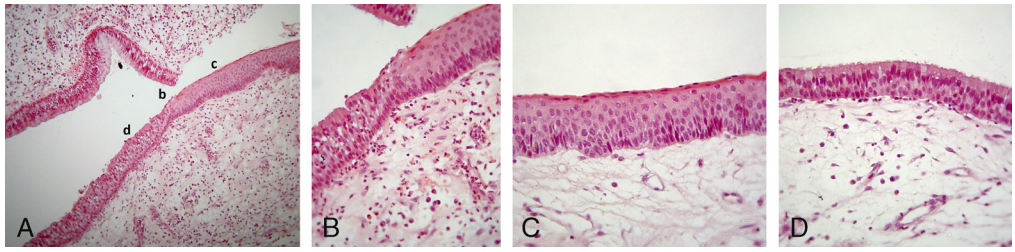

FIGURE 5. Abrupt transition of OKC epithelium to respiratory epithelium: (A) low-power view, (B) area of penetration, (C) OKC epithelium, (D) respiratory epithelium (hematoxylin-eosin stain, original magnification ×100 [A], ×400 [B–D]).

reaction, fibrosis, or necrotic foci were evident. Periodic acid-Schiff stain revealed fungal hyphae and spores in the mucosal connective tissue, close to but not infiltrating vessels (Fig. 6). Final diagnosis was OKC with yeast.

A complete blood count was not suggestive of hematologic disease or diabetes mellitus, and an HIV test was negative.

DISCUSSION

Sinus involvement by OKCs is estimated to occur in less than 1% of the cases,[2] but replacement of respiratory epithelium with OKC epithelium is unusual. Yamazaki et al[3] described a case of an OKC in a 39-year-old woman that involved an impacted maxillary premolar, expanded to the maxillary sinus, and finally reached the inferior nasal meatus. Replacement of the OKC lining by respiratory epithelium was attributed to the proximity of the cyst to the nasal cavity. Abrupt change of OKC lining to respiratory epithelium, as seen in our patient, was reported by Vencio et al[4] in a 27-year-old woman. The cyst was associated with an impacted right second maxillary molar and also invaded the sinus floor.

This microscopic finding is reminiscent of the extension of intraepithelial neoplasia of the oral cavity[5,6] and other carcinomas[7] along adjacent ductal epithelium basement membrane. "Ductal or glandular involvement" is considered an important pathway of spread of carcinomas. In our case, we assume that the OKC epithelium "crept" on the sinus wall replacing the normal respiratory epithelium and extended to involve most of the antral cavity. Thus, we agree with Vencio et al[4] in that replacement is consistent with the infiltrative behavior of the OKC reflected in its classification as a neoplasm. Dysplastic features, as seen in the present case, are occasionally described in OKCs, but have not been associated with an aggressive growth.

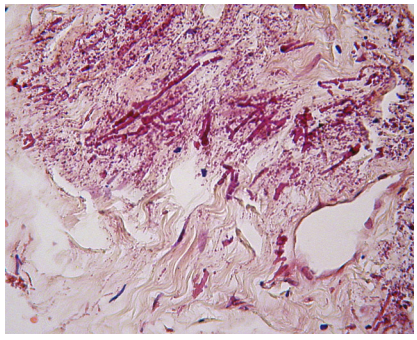

FIGURE 6. Fungal hyphae and spores in the mucosal connective tissue wall (periodic acid-Schiff stain, original magnification ×400).

Our patient had a history of chronic rhinosinusitis that is the most common form of rhinosinusitis, estimated to affect about 20% of the population.[8] Radiological evidence of sinus opacification with calcifications was consistent with fungus balls or aspergillomas, defined as noninvasive accumulations of dense conglomeration of fungal hyphae in 1 sinus cavity. Histological evidence, however, of spores and hyphae in the mucosal connective tissue is diagnostic of an invasive fungal rhinosinusitis, in particular chronic invasive fungal rhinosinusitis. This form is usually associated with *Aspergillus fumigatus* and may appear in patients under corticosteroid treatment who are subtly immunocompromised.

No other case of OKC associated with fungal rhinosinusitis and fungus balls was found in the literature. In our patient, the root canal therapies in the central incisors and right lateral incisor were not close to the sinus; thus, a role for the heavy metals from root filling materials, in particular, zinc from zinc oxide–containing materials, do not seem probable. The blockage of the ostium by the OKC, however, may have resulted in the obstruction of sinus ventilation and the creation of anaerobic conditions that allowed germination of fungi, which probably became invasive because of the local immunosuppression caused by the prolonged corticosteroid treatment.[9,10]

As our patient was proved to be immunocompetent, the complete surgical removal of the cyst and the sinus drainage and aeration achieved with the rhinoantrostomy were considered curative, both for the OKC and the fungal rhinosinusitis.[11]

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Complex Midfacial Reconstruction With an Implant-Supported Framework

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Abstract: This clinical report describes the treatment of a patient with osseointegrated extraoral implants supporting a framework retainer and acrylic resin mesostructures and a large silicone midfacial prosthesis. A metal framework was used to splint the implants together and provided satisfactory retention for the facial prosthesis. A 2-piece prosthesis that composed of an obturator and facial prosthesis was fabricated. Cosmetic improvements as well as the ability to speak, swallow, and, to a lesser degree, chew, were achieved for this patient.

Key Words: Extraoral implant, facial prosthesis, obturator, facial defect, framework

Patients with extensive maxillofacial defects are confronted with functional limitations of vision, speech, mastication, deglutition, and, perhaps most devastating, the psychologic impact that such defects have on the quality of life.[1] The goals of prosthetic rehabilitation for these patients include separation of oral and nasal cavities to allow adequate deglutition and articulation, possible support of the orbital contents to prevent enophthalmos and diplopia, and support the soft tissue to restore the midfacial contour and an acceptable aesthetic result.[2] Adequate retention and acceptable cosmetic results are required for successful prosthetic rehabilitation of a patient with a large facial defect. Osseointegrated implants can provide support and retention using the remaining bones.[3–8] In midfacial and oculofacial defects, fabrication of the retention component is often complicated by the orientation of the abutments, the interabutment distance, and the remaining anatomic structures.[9] Considerable attention has been given to framework design for the extraoral implant–supported prostheses[8,9]; however, reports on the attachment design of the larger combination maxillofacial prostheses are few.

This clinical report describes the treatment of a patient with osseointegrated implants supporting a framework retainer acrylic resin mesostructures and a large silicone facial prosthesis.

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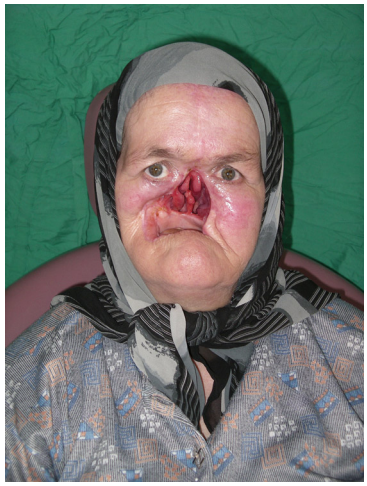

FIGURE 1. Initial facial appearance of the patient.

CLINICAL REPORT

A 55-year-old woman was referred to our clinic for the rehabilitation of a large midfacial defect. History revealed that the patient had been operated on for basal cell carcinoma that occurred in her nasal skin in 1994. During the next 11 years, she had underwent 2 operations for the penetrating basal cell carcinoma. The residual defect included total loss of the midface bilaterally (Fig. 1). The upper lip, alveolar bone, and hard palate were totally gone except for a small fragment of the tuberosity on the left side. Also, right oral commissure, nose to frontal bone, and parts of the cheeks bilaterally were absent. There was a significant impairment of deglutition and speech, which exhibited on articulation resonance disorder with moderate hypernasality. The patient was able to swallow liquids and pureed solids. Radiotherapy was performed, and the patient received 6000 cGy of external beam irradiation in 30 sessions for 6 weeks. Hyperbaric oxygen treatment was not applied.

After 2 years from radiotherapy, patient came to our clinic for treatment. She was examined to place extraoral implants with

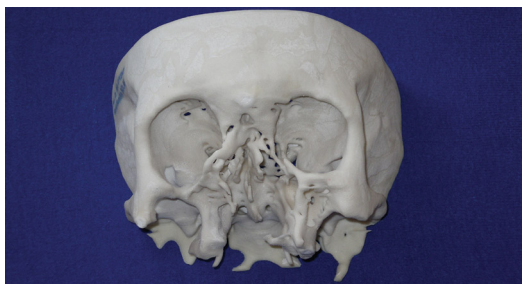

FIGURE 2. Bone model of patient.