Rhinocerebral Mucormycosis in a Type II Diabetic Patient: Case Report

Tip II Diyabetik Hastada Rinoserebral Mukormikozis

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ABSTRACT The purpose of this clinical report is to give information about rhinocerebral mucormycosis which is rarely seen fungal infection to the dental practitioners and represent the prosthetic rehabilitation of an uncommon acquired anterior palatal defect. This case report describes the prosthetic rehabilitation of 63 years old, type II diabetic, male patient who was diagnosed with rhinocerebral mucormycosis. Spontaneous necrotic bone tissue lesions of the premaxillary region developed two months later after the surgical-medical treatment and the anterior palatal defect was a late complication of rhinocerebral mucormycosis. A maxillary removable partial denture was planned for reconstruction of the palatal defect. A round bar attachment and a telescopic crown were used to provide enhanced retention and stability of the prosthesis. A maxillar removable partial denture with bar attachment and a telescopic crown may one of the treatment options to the patient who had an anterior palatal defect which was the result as a late complication of rhinocerebral mucormycosis.

Key Words: Maxillofacial prosthesis; mucormycosis; diabetes complications

ÖZET Bu olgu sunumunun amacı, nadir görülen bir mantar enfeksiyonu olan rinoserebral mukormikozis hakkında dişhekimlerine bilgi vermek ve kazanılmış anterior palatal defektin protetik tedavisini sunmaktır. Bu çalışmada, rinoserebral mukormikozis ile teşhis edilen, 63 yaşındaki tip II diyabetik erkek hastanın protetik tedavisi anlatılmıştır. Hastamızda, cerrahi ve medikal tedavisinden iki ay sonra premaksiller bölgede spontanöz kemik nekrozları gelişmiş ve rinoserebral mukormikozisin geç dönem komplikasyonu olarak anterior palatal bölgede defekt oluşmuştur. Palatal defektin tedavisi için maksiller hareketli bölümlü protez planlanmıştır. Protezin retansiyon ve satabilitesinin sağlanabilmesi için yuvarlak bar tutucu ve teleskop krondan yararlanılmıştır. Rinoserebral mukormikozis sonucu oluşan anterior palatal defektin tedavisinde üst çene hareketli bölümlü protez ile birlikte yuvarlak bar tutucu ve teleskop kron uygulaması bu gibi durumlarda alternatif bir tedavi yaklaşımı olabilir.

Anahtar Kelimeler: Maksillofasiyal protez; mukormikozis; diyabet komplikasyonları

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Diabetes mellitus (DM) is a complex and pernicious syndrome. It is characterized by abnormalities in carbohydrate, lipid and protein metabolism that result either from a profound or an absolute deficiency of insulin, related to autoimmune destruction of the insulin-producing pancreatic β cells (type 1, or insulin dependent DM), or from target-tissue resistance to its cellular metabolic effects, related commonly to obesity (type 2, or non-insuline dependent DM).[1,2]

Hyperglycaemia is the immediate metabolic consequence of DM but, ultimately, there is widespread multisystem damage.[1] In particular microvascular disease (microangiopathy) with capillary basement membrane thickening, macrovascular disease (macroangiopathy) with accelerated arteriosclerosis, neuropathy involving both the somatic and autonomic nervous system, neuromuscular dysfunction, embryopathy, and decreased resistance to infection.[1] DM can have variable, and sometimes, serious effects upon the oral tissues,[3,4] patients with poor glycaemic control being particularly prone to severe and/or recurrent bacterial or fungal infections.[5-8]

Mucormycosis is the third most common invasive fungal infection following aspergillosis and candidiasis.[8-12] Although the vast majority of those affected patients have uncontrolled DM,[7,12]cases also sometimes occur in individuals undergoing transplant-related immunosuppressive therapy,[13] AIDS, renal failure, leukemia,[14,15] and even severe burn patients, all of whom have in common some loss of tissue integrity[16] and compromised systemic immunity.[8,11,12] Very rarely, mucormycosis may develop in an otherwise normal individual.[7,8,11-16]

Rhinocerebral mucormycosis is the most common form of infection and has been described classically in patients with diabetic ketoacidosis or poorly controlled DM. Infection starts in the palate or the paranasal sinuses, progresses to the orbit, and, if not diagnosed early, to the brain. If the disease penetrates the mouth, a black, necrotic eschar is often found in the palate, and ischemic, necrotic turbinates may be found in the nose. In addition to the palate, the disease also involves other areas such as the alveolar margin, lips, cheeks, tongue, and mandible, where the ulcer may have a white necrotic-appearing tissue. Ulceration of the palate is due to infection of the nasal fossa and the paranasal sinuses. Orbital dissemination also occurs because of the close proximity of these air cavities, where the disease starts in the form of a sinusitis in most cases.[7,9,10,12]

Rhinocerebral form of mucormycosis is important for dental professionals because the oral sings and symptoms are the first clinical manifestations in early stages of the infection. In this respect, the aim of this clinical report is to give knowledge about this rarely seen fungal infection to the dental practitioners and represent the prosthetic rehabilitation of an uncommon acquired anterior palatal defect (Aramany class VI)[17,18] caused by one of the late complications of rhinocerebral mucormycosis in the type II diabetic patient.

CASE REPORT

A sixty three years old male patient admitted to Başkent University Adana Research and Education Center with the complaints of loss of vision on the left eye and left facial paralysis. The third, fifth, sixth and seventh cranial nerve palsies were detected on the left side of the face during the examination. He also had hypertension and type II diabetes mellitus for six-seven years.

Paranasal sinus tomography of the patient revealed bilateral sinusitis (Figure 1a and b). The endoscopic sinus surgery and the partial resection of the left inferior and middle concha were operated with the suspect of diagnosis of mucormycosis. The histopathologic examination of the surgical specimen was stated as mucormycosis. Intravascular amphotericin B (200 mg/a day) treatment was given for one month. The patient was clinically recovered after amphotericin B treatment and was discharged from the clinic.

The patient was readmitted to the center two months later with the chief complaint of bad taste and malodor in his mouth. The premaxilla was mobile and pus was detected by mild palpation in the intraoral examination of the patient. Paranasal sinus tomography revealed osteomyelitis of the premaxillary region (Figure 2a and b). The patient was consulted with the prosthodontist preoperatively in order to minimize the potential problems during the prosthetic rehabilitation of the patient following surgery. The premaxillary and the surrounding sequestrated bones were extracted. The both mucosal surfaces were sutured primarily. All remaining teeth were intact except the maxillary incisors.

After the surgical treatment, patient's primary concern was the closure of the defect which interfered with his feeding, speaking and aesthetic defi-

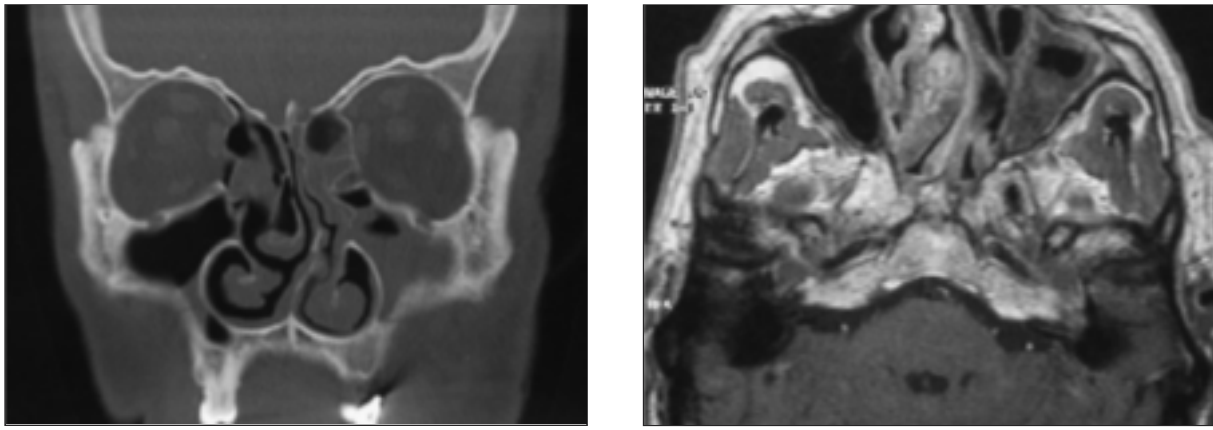

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FIGURE 1a, b: Coronal computed tomography scan **(a)** and contrast enhanced axial T1 weighted magnetic resonance image **(b)** shows diffuse thickening of the mucosa and fluid in the maxillary and ethmoid sinuses.

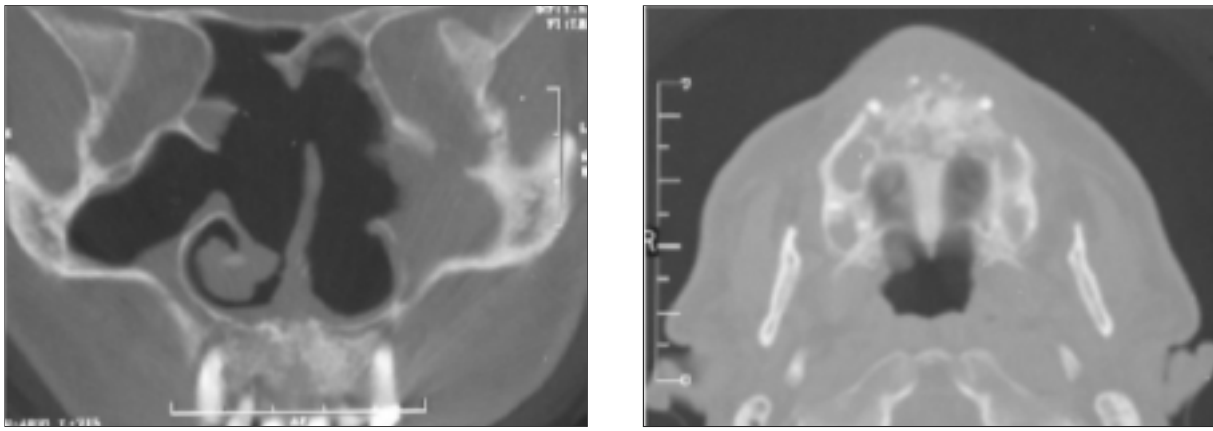

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FIGURE 2a, b: The loss of bone density of the premaxilla, which is appropriate with the osteomyelitis, is seen on the coronal **(a)** and axial **(b)** slices of the paranasal sinus tomography of patient two months after the first surgery. The mucosal thickening of the right and bilateral maxillary is also seen on the coronal slice.

ciency. Clinical examination exhibited that the patient was partial edentulous and had a defect that involved the premaxillary region with a naso-oral opening. The edentulous pattern and premaxillary defect are presented in Figure 3. Patient's oral hygiene was poor and excessive bone and soft tissue loss was observed around the 13 and 23 numbered teeth next to premaxillar resection line. 16, 26 numbered teeth with severe periodontal problems and 13, 23 numbered teeth which were on the resection line were decided to be extracted after the existing fixed partial dentures had removed (Figure 4).

FIGURE 3: Intraoral view of the premaxillar defect.

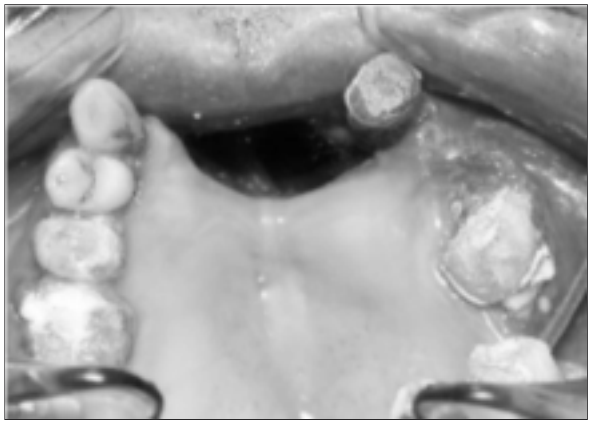

FIGURE 4: Remaining teeth next to premaxillar defect after removing existent fixed partial dentures.

Following healing period, periodontal therapies were performed. Then construction of the maxillofacial prosthesis was planned to supply his feeding, to help his speech and to support upper lip for improving his appearance. Therefore a removable partial denture with a bulb was used to close the naso-oral opening, round bar attachment (MP clip; Metalor Dental AG, Neuchatel Switzerland) and telescopic crown (Adapta deep drawing system, Bego, Bremen, Germany) was preferred to improve the retention and stability (Figure 5, 6). Co-cr-mo alloy (Wironit; Bego, Bremen, Germany) for casting removable partial denture framework and Ni-cr alloy (Metaplus VK, AZ & PARTNER AG, Konstanz, Germany) for casting telescopic crown was chosen.

While establishing occlusal relationships, occlusion was created with artificial incisors and canine teeth out of contact to prevent the masticatory forces which would be destructive for supporting structure of the anterior defect surrounding area. Furthermore late contacts of canine in eccentric positions were produced (Figure 7). Thus, it would also help to provide preventing mesial tipping movement of Kennedy Class IV design removable partial denture and enhancing stability. Ceramometal restorations (IPS d.SIGN Opaquer; Ivoclar Vivadent) and primer coping were cemented zinc polycarboxylate cement the day before the initial placement, of partial denture. The adjustment of bearing surfaces and occlusion was obtained and then the patient was advised to have a subsequent recall appointment for evaluation of oral tissues structures.

DISCUSSION

Prosthetic rehabilitation may be often the best solution because the patients with mucormycosis usually also have an underlying systemic disease and the general health conditions of the patients are poor, consequently surgical options may not be possible. Therefore, maxillofacial prosthetic rehabilitation is of paramount importance for these cases. Schmidt et al stated that the combination of zygomaticus and standard endosseous implants can be used to reconstruct patients after extensive resection of the maxilla and in their study they pre-

FIGURE 5: Cemented ceramometal crowns with round bar and primer coping which a play major role in improving retention, stability and support of the denture.

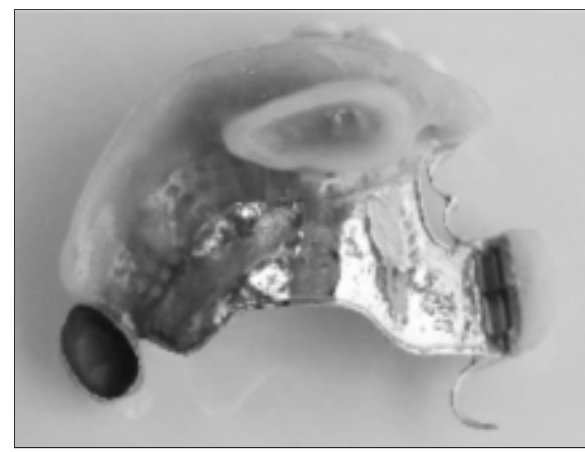

FIGURE 6: The inner surface view of complete maxillofacial prosthesis.

FIGURE 7: Completed prosthetic rehabilitation.

sented prosthetic rehabilitation of a patient with mucormycosis.[19] Nevertheless, the results are not always satisfactory and there are irreversible sequels with irreparable functional damage after the surgical debridement of necrotic tissue. Whereas in some cases debridement may involve orbital nucleation, some part of the face or almost entire maxilla.[7,14,16] These conditions may make both prosthetic rehabilitation and surgical procedures difficult. Thus, early recognition of mucormycosis is necessary to control the spread of infection, which can lead to high morbidity and mortality.

In 1997, an estimated 124 million population worldwide were living with diabetes. By the year 2010, the number of the people with diabetes worldwide is projected 221 million, and in certain regions of the world (for example Asia, Africa), diabetes rates could rise twofold or threefold.[2,3] Therefore, dentists will frequently encounter DM patients in their offices and they must be familiar with the oral signs and symptoms of the disease and should maintain a high level of suspicion in patients with DM. All patients diagnosed with diabetes must be identified by history. The medical treatment that has been applied to DM patient such as medications, regimen, exercises and degree of glycemic control over and above any systemic complications resulting from diabetes must be precisely established and the dentist must also organize recall appointments more frequently. Correspondingly any dental patient with cardial sings and symptoms of diabetes (that is, polydipsia, polyuria, polyphagia, weight loss, weakness), or who present with an oral manifestations (for instance, xerostomia, burning mouth syndrome, lichen planus, gingivitis/periodontitis or fungal infections), should be referred to a medical consultant for diagnosis and treatment.

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