Turk J Intensive Care 2022;20:106-109
DOI: 10.4274/tybd.galenos.2022.08831

CASE REPORT / *OLGU SUNUMU*

Mehmet Turan İnal,
Dilek Memiş,
Pervin Hancı,
Gökhan Günbay,
Samet Keleş,
Habibe Tülin Elmaslar Mert,
Rüveyde Garip,
Selis Gülseven Güven,
Emre Hocaoğlu,
Banu Tütüncüler

Mucormycosis in a Patient with Uncontrolled Diabetes Mellitus

Kontrolsüz Diabetes Mellituslu Hastada Mukormikoz

Received/Geliş Tarihi : 21.02.2022
Accepted/Kabul Tarihi : 09.05.2022

Mehmet Turan İnal, Dilek Memiş, Gökhan Günbay, Samet Keleş,
Trakya University Faculty of Medicine, Department of Anesthesiology and Reanimation, Division of Intensive Care, Edirne, Turkey

Pervin Hancı
Trakya University Faculty of Medicine, Department of Chest Diseases, Edirne, Turkey

Habibe Tülin Elmaslar Mert
Trakya University Faculty of Medicine, Department of Infectious Diseases, Edirne, Turkey

Rüveyde Garip
Trakya University Faculty of Medicine, Department of Ophthalmology, Edirne, Turkey

Selis Gülseven Güven
Trakya University Faculty of Medicine, Department of Otorhinolaryngology, Edirne, Turkey

Emre Hocaoğlu
Trakya University Faculty of Medicine, Department of Plastic and Reconstructive Surgery, Edirne, Turkey

Banu Tütüncüler
Trakya University Faculty of Medicine, Department of Neurosurgery, Edirne, Turkey

Mehmet Turan İnal Prof. MD (✉),
Trakya University Faculty of Medicine, Department of Anesthesiology and Reanimation, Division of Intensive Care, Edirne, Turkey

E-mail : mehmetturaninal@yahoo.com
Phone : +90 284 235 76 41
ORCID ID : orcid.org/0000-0001-8462-4299

ABSTRACT Mucormycosis; is a rapidly progressive fungal infection due to filamentous fungi of the mucoraceae family. In this case report, we aimed to present the diagnosis and treatment modalities of a patient who developed rhinoorbital mucormycosis. A 54-year-old patient with a history of hypertension applied to the emergency department with a complaint of wound in the mouth that started four days ago. In the examinations performed here, the patient was diagnosed with diabetic ketoacidosis. In the examination of the patient, it was found that there was a necrotic wound on the left hard palate, a necrotic wound extending from the left inferior turbinate to the nasopharynx, and hyphae in the nasal passage. The patient underwent an aggressive debridement operation on the third day, due to the growth in the fungal culture. In the following clinical examination of the patient, ketone in the urine became negative, and his acidosis status improved. On the same day, the patient was treated with a positive coronavirus disease-2019 (COVID-19) polymerase chain reaction. After 15 days of treatment, the patient died due to COVID-19 pneumonia. Mucormycosis should be doubtful in patients presenting with uncontrolled diabetes mellitus and severe sino-orbital infection. All physicians following diabetic ketoacidosis should be vigilant against this rapidly progressing disease with high mortality.
Keywords: Mucormycosis, diabetic ketoacidosis, mortality

ÖZ Mukormikoz mucoraceae familyasının filamentli mantarlarına bağlı hızla ilerleyen bir mantar enfeksiyonudur. Bu olgu sunumunda rinoorbital mukormikoz gelişen bir hastanın tanı ve tedavi yöntemlerini sunmayı amaçladık. Elli dört yaşında hipertansiyon öyküsü olan hasta, dört gün önce başlayan ağızda yara şikayeti ile acil servise başvurdu. Burada yapılan tetkiklerde hastaya diyabetik ketoasidoz tanısı konuldu. Hastanın muayenesinde sol sert damakta nekrotik yara, sol alt konkadan nazofarenkse kadar uzanan nekrotik yara ve burun pasajında hifa tespit edildi. Üçüncü gün mantar kültüründe üreme olması üzerine hastaya agresif debridman operasyonu uygulandı. Hastanın sonraki klinik muayenesinde idrarda keton negatif çıktı ve asidoz durumu düzeldi. Aynı gün hasta pozitif koronavirüs hastalığı-2019 (COVID-19) polimeraz zincir reaksiyonu ile tedavi edildi. On beş günlük tedaviden sonra hasta COVID-19 pnömonisi nedeniyle eksitus kabul edildi. Kontrolsüz diabetes mellitus ve şiddetli sino-orbital enfeksiyon ile başvuran hastalarda mukormikozdan şüphelenilmelidir. Diyabetik ketoasidoz sonrası tüm hekimler hızla ilerleyen ve mortalitesi yüksek olan bu hastalığa karşı dikkatli olmalıdırlar.
Anahtar Kelimeler: Mukormikoz, diyabetik ketoasidoz, mortalite

Introduction

Mucormycosis; it is a rapidly progressive fungal infection due to filamentous fungi of the mucoraceae class of fungi and is frequently seen in people who have diabetes mellitus, corticosteroid use, hematological malignancies such as lymphoma and leukemia, neutropenia, undergoing solid organ/allogeneic stem cell transplant operation, kidney failure, treated with immunosuppressants, cirrhosis, burns, protein energy malnutrition and AIDS (1-3). Depending on organ involvement, mucormycosis can be seen as rhinocerebral, rinoorbital, pulmonary, cutaneous, gastrointestinal or disseminated. The rhinocerebral form is the most common form (1-3). Rhinoorbital infection begins as a result of inhalation of fungal spores and invasion of the nasal mucosa, by invading the arteries, this fungus forms thrombi that reduce blood flow in blood vessels and cause necrosis of hard and soft tissues. Orbital involvement occurs when the fungus infection moves from the paranasal sinuses to the orbital wall. Pain in and around the eye, redness of the eye, decreased vision and proptosis can be seen in patients (1,4,5).

Despite advances in diagnosis and treatment, mucormycosis is still a disease with high mortality (6).

In this case report, we aimed to present the diagnosis and treatment modalities of a patient who developed rhinoorbital mucormycosis.

Case Report

The 54-year-old patient with a history of hypertension applied to the emergency department with a complaint of wound in the mouth that started four days ago. In the examinations performed here, fasting blood sugar was found to be 686 mg/dL and pH 7.30 in arterial blood gas, and ketone was detected in the urine. The patient was diagnosed with diabetic ketoacidosis (DKA) and was hospitalized in the intensive care unit. In the examination of the patient, it was found that there was a necrotic wound on the left hard palate, a necrotic wound extending from the left inferior turbinate to the nasopharynx, and hyphae in the nasal passage. Orbital computed tomography (CT) was performed for the patient whose sample was taken from here, and no pathology was detected in the orbita. Amphotericin B treatment was started on the same day. After one day, the swelling and redness of the left eye increased, and endoscopic debridement was performed. The patient underwent an aggressive debridement operation on the third day, due to the growth in the fungal culture. Maxillectomy, anterior ethmoidectomy, left orbital bone excision, left eye exenteration, and skull base debridement adjacent to the frontal sinus were performed. In the following clinical examination of the patient, ketone in the urine became negative, and his acidosis status improved. On the same day, the patient was started to treated with a positive coronavirus disease-2019 (COVID-19) polymerase chain reaction. The patient was followed up in the intensive care unit for 15 days due to COVID-19 pneumonia. The patient was considered exitus on the 15th day.

Approve was taken from the patient's relatives for this case presentation.

Discussion

In this case report, rhinoorbital mucormycosis in a uncontrolled diabetes mellitus patient was reported.

Although opportunistic fungal infections such as mucormycosis usually occur in immunocompromised individuals, they can also be seen in healthy individuals (5,6). Predisposing factors for mucormycosis are uncontrolled diabetes (especially in patients with ketoacidosis), malignancies such as lymphoma and leukemia, chronic corticosteroid use, immunosuppressive therapy, kidney failure, cirrhosis, burns, previous organ transplant, protein energy malnutrition, and AIDS (1-3). In the study of Yohai et al. (7) on 145 patients, diabetes mellitus was found to be the most common predisposing factor. Gumral et al. (8) also reported that diabetes mellitus was the predisposing factor in 32 of 79 mucormycosis cases and hematological pathologies in 32 of them. The authors also reported that diabetes is newly diagnosed in 16% of patients (9), similarly to our patient. Our patient had uncontrolled diabetes, which is a frequently reported predisposing factor for mucormycosis. Our patient had high blood sugar at the time of admission to the hospital. We think that the patient continued his life with high blood sugar, even though there was no diagnosis of diabetes mellitus in the patient's history.

While the rhinocerebral form is the most common form, the sino-orbital form seen in our patient is seen in only 15% of the cases (2,6). When the fungi are inhaled, the spores turn into hyphae, causing tissue necrosis and thrombosis of blood vessels (2,9). Although the reason why fungi are more common in diabetic patients is not fully explained, fungal microvascular disease has been reported to be

associated with greater tissue destruction and spread in diabetic patients (6). The dysfunction in phagocytic cells due to hyperglycemia and acidosis is an facilitating factor. It has been shown to increase the availability of free iron, which is a requirement for fungal survival, by impairing the binding of iron to transferrin in acidosis, which is also seen in DKA (6). In addition, the fact that some fungal species have a special enzyme system that can increase fungal growth in acidic and hyperglisemic conditions, as in diabetic ketoasidosiz, is also considered as a reason (2).

Mucormycosis should be diagnosed quickly and treatment should be started quickly. While the survival rate was found to be 76-81% in patients who started the treatment within the first 6 days, the survival rate was reported as 36-42% in the case of starting treatment after 12 days (1). Our patient had a complaint of sores in the mouth that started 4 days ago, but we had no idea about the duration of the high blood sugar since the patient was not diagnosed with diabetes.

Although magnetic resonance imaging is more sensitive than CT in the diagnosis of sinus mucormycosis, both results have been reported to be negative (2,6). Therefore, mucosal biopsy and surgical exploration should be considered in cases with high clinical suspicion. Due to the high mortality of the disease, it is recommended to start treatment as fast as possible (2,6). In our patient the treatment was started quickly. Amphotericin B treatment was started initially.

Control of hyperglycemia and ketoacidosis is important in treatment. Combined surgery and antifungal therapy provides better survival (70%) than surgery (57%) and antifungals alone (61%) (2). In the rhinocerebral form, while the mortality rate was 70% in patients using only antifungals, it decreased to 14% in patients treated with antifungal therapy and surgery (9,10). In our patient, endoscopic debridement was performed one day after hospitalization, but surgical exploration was performed the next day because the necrotic area continued to grow.

Clinical differential diagnosis of the lesion should include chronic granulomatous infection such as squamous cell carcinoma, tuberculosis, syphilis, and other fungal infections (11).

In conclusion, mucormycosis should be considered in patients presenting with uncontrolled diabetes mellitus and severe sino-orbital infection. All physicians following DKA should be vigilant against this rapidly progressing disease with high mortality.

Ethics

Informed Consent: Approve was taken from the patient's relatives for this case presentation.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: M.T.İ., D.M., P.H., G.G., S.K., H.T.E.M., R.G., S.G.G., E.H., B.T., Concept: M.T.İ., G.G., S.K., Design: M.T.İ., Data Collection and/or Processing: G.G., S.K., Analysis and/or Interpretation: M.T.İ., Literature Search: M.T.İ., Writing: M.T.İ.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Karadeniz Uğurlu Ş, Selim S, Kopar A, Songu M. Rhino-orbital Mucormycosis: Clinical Findings and Treatment Outcomes of Four Cases. Turk J Ophthalmol 2015;45:169-74.
2. Roden MM, Zaoutis TE, Buchanan WL, Knudsen TA, Sarkisova TA, Schaufele RL, et al. Epidemiology and outcome of zygomycosis: a review of 929 reported cases. Clin Infect Dis 2005;41:634-53.
3. Tabarsi P, Khalili N, Pourabdollah M, Sharifynia S, Safavi Naeini A, Ghorbani J, et al. Case Report: COVID-19-associated Rhinosinusitis Mucormycosis Caused by Rhizopus arrhizus: A Rare but Potentially Fatal Infection Occurring After Treatment with Corticosteroids. Am J Trop Med Hyg 2021;105:449-53.
4. Leitner C, Hoffmann J, Zerfowski M, Reinert S. Mucormycosis: necrotizing soft tissue lesion of the face. J Oral Maxillofac Surg 2003;61:1354-8.
5. Pogrel MA, Miller CE. A case of maxillary necrosis. J Oral Maxillofac Surg 2003;61:489-93.
6. Saul SR, Aleksic S, Magnotti M. A Patient with Newly Diagnosed Diabetes Presenting with Sino-Orbital Mucormycosis. AACE Clinical Case Rep 2016;2:e41-5.
7. Yohai RA, Bullock JD, Aziz AA, Markert RJ. Survival factors in rhino-orbital-cerebral mucormycosis. Surv Ophthalmol 1994;39:3-22.
8. Gumral R, Yildizoglu U, Saracli MA, Kaptan K, Tosun F, Yildiran ST. A case of rhinoorbital mucormycosis in a leukemic patient with a literature review from Turkey. Mycopathologia 2011;172:397-405.
9. Spellberg B, Edwards J Jr, Ibrahim A. Novel perspectives on mucormycosis: pathophysiology, presentation, and management. Clin Microbiol Rev 2005;18:556-69.
10. Perlroth J, Choi B, Spellberg B. Nosocomial fungal infections: epidemiology, diagnosis, and treatment. Med Mycol 2007;45:321-46.
11. Afroze SN, Korlepara R, Rao GV, Madala J. Mucormycosis in a Diabetic Patient: A Case Report with an Insight into Its Pathophysiology. Contemp Clin Dent 2017;8:662-6.