



Cleft Lip Nasal Deformity After Mucormycosis Infection: Case Report

Mukormikozis Enfeksiyonu Sonrası Oluşmuş Dudak Yarığı Burnu Deformitesi: Vaka Sunumu

Rhinocerebral Mucormycosis

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Özet

Mukormikozis akut fulminant fungal enfeksiyondur. Mukormikozis çoğunlukla kontrolsüz diyabet [özellikle ketoasidozlu hastalar], lenfoma ve lösemi gibi malignansiler, renal yetmezlik, organ transplantasyonları, uzun dönem kortikosteroid ve immünsüpresif tedavi alımı, yanık, siroz, protein-enerji malnütrüsyonu ve AIDS gibi hastalıklar ile beraber görülse de sağlıklı kişilerde de rastlanabili. 21 yaşında erkek hasta damak yarığı ve burunda şekil bozukluğu şikayeti ile kliniğimize başvurdu. Hastanın hikayesinden, 10 yaşında iken lenfoma tanısı konulduğu, lenfoma tedavisi görülürken damak ve burnunda enfeksiyon geliştiği öğrenildi. Kliniğimizde hasta opere edilerek damak yarığı ve burun deformitesi düzeltildi. Burada mukormikozis enfeksiyonuna bağlı gelişen nasal deformite ve inkomplet damak yarığı olgusu sunulmuştur.

Anahtar Kelimeler

Mukormikozis; Damak Yarığı; Nasal Deformite

Abstract

Mucormycosis is an acute fulminant fungal infection. Mucormycosis usually accompanies uncontrolled diabetes [in particular, patients with ketoacidosis], malignancies like lymphoma or leukemia, renal failure, organ transplantations, long-term corticosteroid or immunosuppressant therapy, and conditions including burns, cirrhosis, protein-energy malnutrition or AIDS, though it also may be seen in healthy individuals. A 21-year-old male patient applied to our clinic with cleft lip and nasal deformity. It was understood from his medical history that he was diagnosed with lymphoma at age 10 and he developed an infection in his palate and nose during the treatment course. His cleft palate and nasal deformity was repaired by surgery in our clinic. Herein we reported a case of nasal deformity and incomplete cleft palate caused by mucormycosis infection.

Keywords

Mucormycosis; Cleft Palate; Nasal Deformity

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Introduction

Zygomycosis, also known as mucormycosis, is an acute fulminant fungal infection originally described by Paultauf in 1885 [1] and caused by the spores of Mucorales belonging to the order of Zygomycetes. These spores exist in the earth, in rotten vegetables, and in the respiratory and gastrointestinal tracts of healthy subjects [2]. The infection is contracted via the inhalation, ingestion, and incubation of the spores [3].

Although mucormycosis mostly occurs in association with conditions such as uncontrolled diabetes [particularly patients with ketoacidosis], malignancies such as lymphoma or leukemia, organ transplantation, long-term corticosteroid or immunosuppressive use, protein-energy malnutrition, and AIDS, it may also occur in healthy individuals [4, 5]. In particular, contact with contaminated water or waste after trauma may lead to the development of mucormycosis [6].

Mucormycosis is subdivided into several types based on its clinical manifestations and the involved anatomical location, such as rhinocebral, pulmonary, cutaneous, gastrointestinal, or disseminated [7]. Of these, rhinocebral mucormycosis is the most frequent form, responsible for one-third of all cases [1]. It is characterized by the progressive fungal invasion of the paranasal sinuses, palate, orbita, and the cranium [2].

In this case report, we describe a case of cleft palate and nasal deformity developing after mucormycosis.

Case Report

A 21-year-old male patient presented to our unit with cleft palate and nasal deformity. Medical history revealed a diagnosis of lymphoma at the age of 10, in conjunction with an infection of the palate and the nose during lymphoma treatment. Physical examination showed the presence of incomplete cleft palate, with left-sided deviation of the nose, wider right nostril, shortening in the left side of the columella, right-sided deviation of the columella base, and more posteriorly located left dome as compared to the right. These nasal deformities resembled those of the cleft lip nasal deformity [Figures 1-2]. An examination of past medical records and discharge notes revealed a histopathologically confirmed diagnosis of mucormycosis, treatment with high dose amphotericin B, and intensive care unit admission, as well as the surgical debridement of the necrotic tissues at the time of palate infection. The patient was operated on in our unit for the correction of the cleft palate nasal deformity.

Discussion

Nasal deformities associated with congenital cleft lip are associated with significant cosmetic and functional problems and are referred to as cleft lip nasal deformity [8]. In this deformity the base of the nose appears longer on the side of the cleft lip, the dome of the cleft side is displaced posteriorly, the base of the columella is deviated toward the non-cleft side, and the nostril on the cleft side is widened and rotates posteriorly. Our patient had similar deformities after contracting rhinocebral mucormycosis.

In rhinocebral mucormycosis, the spores of Mucorales invade the vascular endothelium after inhalation and attach to the internal elastic lamina of the blood vessels. Fungal growth occurs along the elastic lamina of the vasculature, separating lamina



Figure 1. incomplete cleft palate



Figure 2. nasal deformity

from the media. The direct invasion and dissection caused by these fungi result in severe endothelial injury with subsequent thrombus formation and ischemia in the surrounding tissues [9]. Palatal and maxillary necrosis may develop due to the thrombosis caused by spores in the internal maxillary artery or descending palatine arteries [4]. We believe that our patient also probably had impaired circulation in the territories of the descending palatine and internal maxillary arteries, leading to necrosis in the medial wall of the maxillary bone, at the base of the nasal cavity, and in the palate with subsequent deformities closely resembling the cleft lip nasal deformity.

The key components of the management of rhinocebral mucormycosis include early diagnosis, anti-fungal agents, and aggressive debridement. For anti-fungal treatment, parenteral amphotericin B at a dose of 1 to 1.5 mg/kg or oral amphotericin B at a dose of 10 to 15 mg/kg/day may be administered [10]. Our patient had also received treatment with amphotericin B and had undergone surgical debridement of the necrotic tissues of the palate.

Competing interests

The authors declare that they have no competing interests.

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