CASE REPORT

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An Extensive Invasive Fungal Sinusitis Case: Management and Review of Literature

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ABSTRACT Invasive fungal sinusitis often occurs in patients with immune deficiency. We can group fungal sinusitis under 4 groups. Invasive forms: 1-Acute (Fulminant) 2-Chronic (Indolent). Non-invasive forms are 3-Mycetoma and 4- Allergic fungal sinusitis..Invasive fungal sinusitis is generally seen in immunocompromised patients. Diabetes mellitus and leukemia are the most common underlying pathologies, respectively. Delayed diagnosis and inadequate treatment can be mortal, especially for invasive forms. In this case report, the management of a patient with invasive fungal sinusitis, who was referred to us due to unilateral preseptal edema developing during hospitalization for diabetic ketoacidosis treatment, is presented in the light of the current literature.

Keywords: Diabetes mellitus; paranasal sinus; invasive fungal infection; endoscopic surgical procedure

Recently, technological development in laboratory studies and the increase of immunocompromised patients have increased the incidence of fungal sinusitis and also accelerated the studies on this subject.[1] It has been observed that 70% of cases with acute and chronic fulminant sinusitis are diabetics in ketoacidosis. Patients who receive chemotherapy due to leukemia or lymphoma are the second most common cases.[2]

In this case report, the management of a patient with invasive fungal sinusitis, who was referred to us due to unilateral preseptal edema developing during hospitalization for diabetic ketoacidosis treatment, is presented in the light of the current literature.

CASE REPORT

A 55-year-old female patient was admitted to our clinic with numbness, swelling, and redness on the right side of the face. The patient with type 2 diabetes mellitus was referred to us with a pre-diagnosis of preseptal cellulite upon the onset of complaints while receiving diabetic ketoacidosis treatment in another center. The examination of the patient revealed generalized swelling and redness in the right half of the face. There were swelling and ptosis in the right eyelid. Biochemical results were as; white blood cell (WBC) 15.2 K / uL (4.4-11.3), fasting blood sugar 259 mg/dL (70-115), creatinine 1.68 mg/dL (0.6-1.3), sedimentation 87mm / h (0-12), CRP 23 mg/L (0-5). In the endoscopic view, there was just mucous discharge on the right nasal passage. Magnetic resonance imaging (MRI) showed soft tissue densities with heterogeneous contrast involvement covering the right maxillary sinus, ethmoid sinuses, and frontal recess. No signs of destruction were detected (Figure 1). Paranasal sinus computed tomography (CT) revealed lesions in the right maxillary sinus, ethmoid sinuses, and soft tissue density, which was heterogeneously contrasted in the frontal recess (Figure 2).

The patient underwent endoscopic sinus surgery. During surgery, a pale, gray, fragile mass filling the right middle meatus, ethmoid cells, and maxillary sinus was observed. The mass was removed by opening the maxillary ostium and performing anterior and posterior ethmoidectomy. Insulin, ceftriaxone, metronidazole, enoxaparin, itraconazole were started to the patient.

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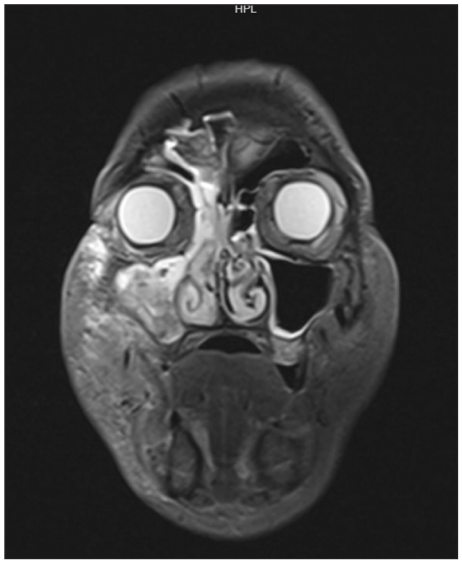

FIGURE 1: Magnetic resonance image before 1st surgery (sequence T2-tirm-cor).

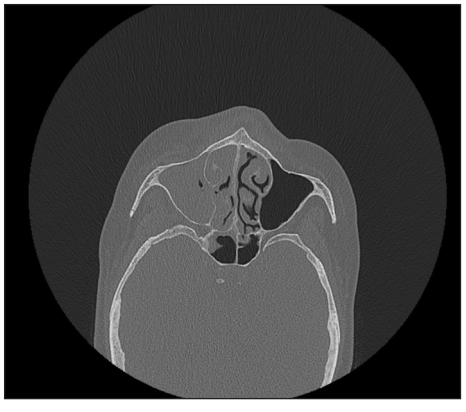

FIGURE 2: Computed tomography image before 1st surgery.

The pathology report revealed mixed inflammatory cell infiltration, and fungal infection compatible with aspergillus. The CRP value on the third postoperative day was 3 mg/L (0-5) and the WBC was 10.2 K/uL (4.4-11.3). Blood sugars were monitored as regulated. However, the patient's complaint of redness and swelling on the face increased. Medical treatment of the patient was completed to 1 week, and upon the increase of complaints, CT and MRI were requested again. CT and MRI showed areas of cortical destruction in the medial, lateral, and superior wall of the maxillary sinus, and soft tissue densities in the ethmoid, frontal and sphenoid sinuses, which were heterogeneously contrasted in the surrounding tissues (Figure 3). The patient was operated again. In addition to endoscopic sinus surgery, Caldwell Luc procedure was applied and necrotic tissues were widely debrided. Fungal structures were present around the infraorbital nerve under the orbital wall and were debrided (Figure 4). Amphotericin B irrigation was applied to all mucosa as much as possible. While reaching the frontal sinus, the dehiscence of the frontoethmoid recess was observed as revealing dura (Figure 5). Postoperative medical treatment was continued intravenously for another 10 days. The pa-

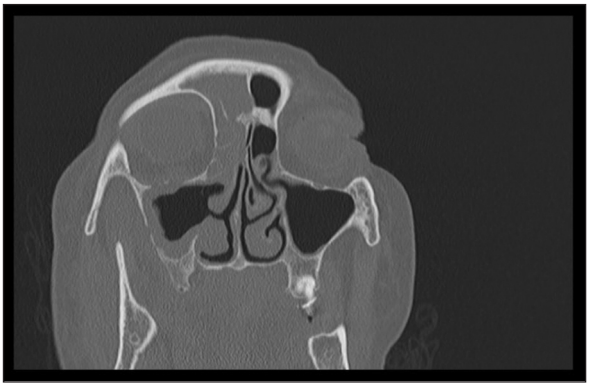

FIGURE 3: Computed tomography image before 2nd surgery.

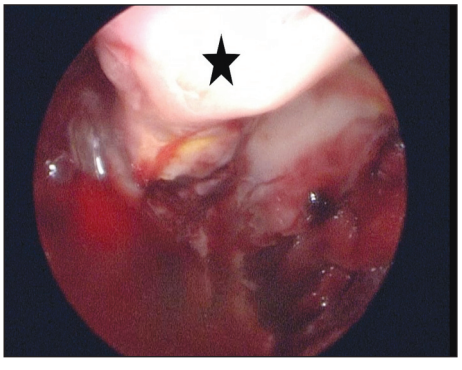

FIGURE 4: Intraoperative image of the right maxillary sinus. Star: Infraorbital nevre.

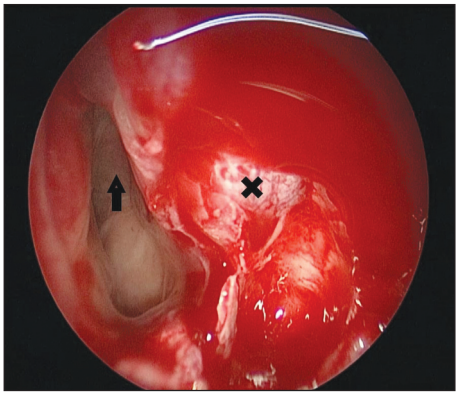

FIGURE 5: Intraoperative images of right frontoethmoid recess. X: Dura Arrow: Right frontal sinus.

tient's clinic improved significantly and she was discharged with oral itraconazole 200mg 2x1 treatment. Informed consent was obtained from patient included before surgery.In the postoperative first month control, no pathology was detected in the nasal endoscopy, except for minimal crusting in the ethmoid cavity. No recurrence was observed in the one-year follow-up of the patient.

DISCUSSION

3 consecutive months duration of inflammation of paranasal sinuses and nasal mucosa is termed as chronic rhinosinusitis.[3] This inflammation may be due to microbes (bacteria and fungi) or allergic and non-allergic causes.

For fungal rhinosinusitis; the invasive forms are acute fulminant, chronic, and granulomatous invasive fungal rhinosinusitis. The non-invasive forms are fungus ball and allergic fungal rhinosinusitis.[4] Aspergillus the most common causative species followed by candida and mucor.[5,6]

Chronic fungal rhinosinusitis is an emerging pathology occurring generally in immunosuppressed patients like diabetics and patients on corticosteroid therapy.Clinically, a patient having chronic fungal rhinosinusitis may have all the symptoms of chronic rhinosinusitis.[7] The delay in the diagnosis may cause the disease to spread to vital structures like the cranial system.[8-10]

Immunomodulation,extensive surgical debridement, and antifungal therapy are the trivet of invasion management, as they are considered to be the most important factors for mortality.[11] In our case, we performed both endoscopic sinus surgery and Caldwell- Luc approach to eradicate the invasion.

Concerning chronic fungal rhinosinusitis, a recent study reported that azole derivatives (voriconazole or itraconazole) were consistently better than amphotericin B. The duration of oral voriconazole administration is still debatable; typically, it ranges from 6 to 18 months.[12-14] We gave itraconazole 200 mg to our patient intravenously. Additionally, we irrigated maxillary sinüs with amphotericin B solution perioperatively. This irrigation procedure was first applied as far as we searched the literature.

Briefly, chronic invasive fungal sinusitis is a challenging, stubborn, morbid, and possibly fatal disease. A careful history along with radiological findings and microbiological diagnosis may be helpful in such types of infections. Early diagnosis and preventing causing reasons as diabetes are important elements of invasion management. Also, extensive surgery and antifungal drugs are crucial for controlling these destructive infections.

Ethical Approval

All procedures performed in this study were in accordance with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent

Informed consent was obtained from individual participant included in the study before surgery.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Erkan Yıldız, Selçuk Kuzu;* ***Design:*** *Selçuk Kuzu, Çağlar Günebakan;* ***Control/Supervision:*** *Çağlar Günebakan, Şahin Ulu;* ***Data Collection and/or Processing:*** *Şahin Ulu, Abdülkadir Bucak;* ***Analysis and/or Interpretation:*** *Abdülkadir Bucak, Erkan Yıldız;* ***Literature Review:*** *Selçuk Kuzu, Çağlar Günebakan;* ***Writing the Article:*** *Erkan Yıldız, Selçuk Kuzu;* ***Critical Review:*** *Abdülkadir Bucak;* ***References and Fundings:*** *Selçuk Kuzu;* ***Materials:****Çağlar Günebakan.*

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