

Chronic Maxillary Sinusitis Caused by an Aspergilloma in the Context of Inadequately Treated Type II Diabetes Mellitus

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Abstract. *Background/Aim:* The prevalence of chronic sinusitis (CS) in Europe is greater than 10%. The causes of CS are diverse. In some cases, dental treatment in the maxilla as well as fungal infection, such as aspergilloma, can lead to CS. Inadequately treated illnesses, such as type II diabetes, are known risk factors for atypical infections. *Case Report:* The present case report describes a 72-year-old female suffering from CS in the maxillary sinus. A few years earlier, the patient received endodontic treatment of a maxillary tooth. For further diagnostics a CT-scan was performed showing an obstructed maxillary sinus on the left due to a polypoid tumor. The patient had been suffering from type II diabetes that had been inadequately treated for several years. The patient was surgically treated with an osteoplasty of the maxillary sinus combined with a supratubinal antrostomy. Histopathological findings revealed an aspergilloma. The surgical therapy was supplemented by antimycotic therapy. In addition, the patient received antidiabetic treatment leading towards stable blood sugar levels. *Conclusion:* Rare entities, such as aspergillomas, can also be the cause of CS. In particular, patients with previous illnesses relevant to the immune system are predisposed for Aspergilloma after dental treatment leading to CS.

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Key Words: Maxillary sinusitis, aspergilloma, diabetes type II, endodontic treatment.

Acute and chronic inflammation of the paranasal sinuses are common. The prevalence of chronic sinusitis (CS) in Europe is more than 10% (1). CS can be differentiated from acute sinusitis (AS) if it persists for more than 12 weeks. CS can also be subdivided by the presence or absence of polyps (2). From a pathophysiological point of view, the acute and the chronic form of sinusitis are two different entities. AS, in particular, is caused by acute viral diseases and/or bacterial infections (3). In rare exceptional cases, fungal diseases can also lead to AS (4). The pathogens lead to swelling of the mucosa in the paranasal sinuses with subsequent ventilation disorders in the area of the osteomeatal complex, particularly as a result of acute rhinitis (5). In the context of CS there are in particular remodeling processes in the area of the mucosa. In CS with polyps, a dysbalance of the T helper-cells in the context of immune modulation with subsequent barrier disorders of the mucosa can be registered (6). However, CS without polyps is characterized by the presence of Th1 cells with the histopathological findings of chronic fibrosis of the mucosa in the paranasal sinus (7).

In some cases, a dental focus and/or dental treatment can also be the cause of CS (8). Unrecognized or inadequately treated diseases, such as type II diabetes, promote an imbalance in the immune system (9). In particular, patients with type II diabetes tend to have atypical causes of infections, including those associated with rhinosinusitis (10). However, up to now there are only a few publications on chronic rhinosinusitis caused by fungal infections, such as aspergilloma, following a dental treatment in the context of in the context of type II diabetes mellitus.

Case Report

A 72-year-old female with type II diabetes mellitus presented with non-specific sinus symptoms that had been present for four years. In particular, the patient felt increased retronasal secretion at night as well as recurrent pain in the area of the left maxillary sinus. Intermittently, there were also olfactory



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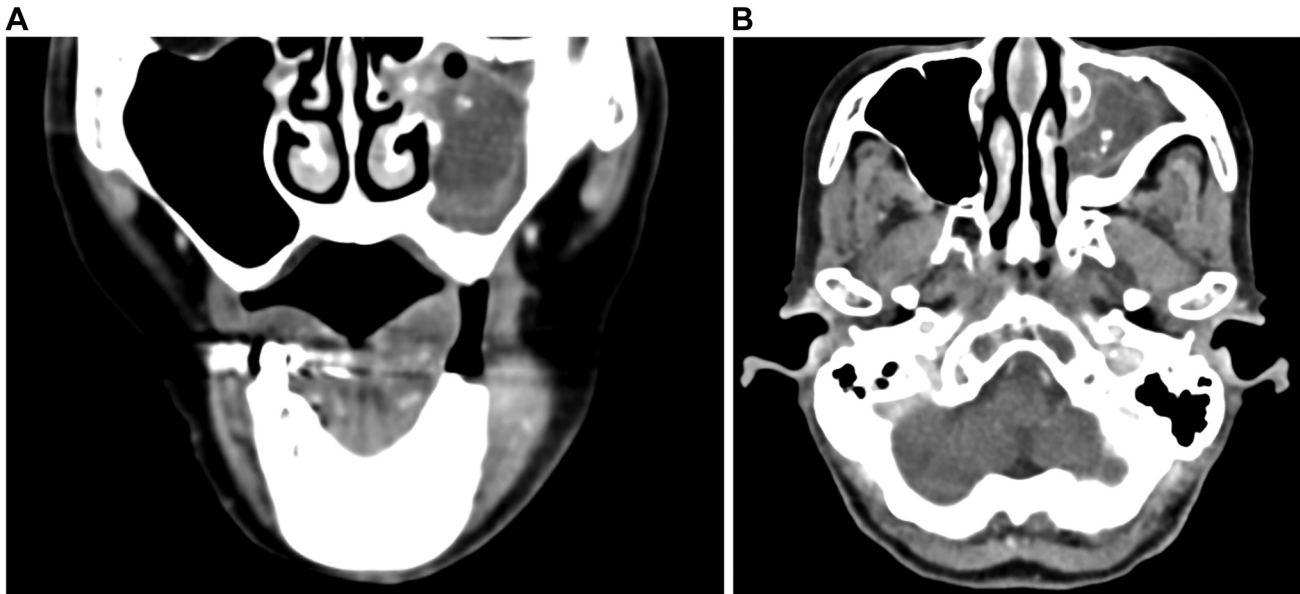


Figure 1. (A) CT-scan in a coronal section showing a completely obstructed maxillary sinus on the left due to a polypoid mass. The mass shows ring-shaped enhancement of contrast medium typical of aspergillomas. (B) CT-scan in an axial section showing the extent and the typical contrast agent enhancement of the aspergilloma. CT: Computed tomography.

disorders. Repeated examinations by the outpatient ear, nose and throat doctor (ENT) and conservative therapy with xylometazoline as well as cortisol nasal spray had not resulted in any improvement of the symptoms. The patient also stated that she had had recurrent colds in recent years. In the extended anamnesis, the patient reported problems with the medications concerning her diabetes type II. At the time of the first presentation, a HbA1c (glycated hemoglobin) value of 9% was determined as well as unstable blood sugar levels. An endodontic treatment of tooth 26 was carried out four years ago. In the meantime, however, the tooth had been extracted. There was no treatment in the area of the remaining maxillary teeth. Next to the HbA1c value mentioned above, only a mild form of hypochromic microcytic anemia was found. Due to the pronounced symptoms, a CT scan of the paranasal sinuses was performed. Here a complete obstruction of the sinus maxillaris on the left was shown by a polypoid tumor (Figure 1A and B). The tumor showed ring-shaped enhancement of contrast agent typical of aspergillomas. The surgical therapy consisted of an osteoplasty of the maxillary sinus through an intraoral approach as well as the supratubinal antrostomy on the left. A roundish tumor measuring 3 cm was removed. The histopathological analysis of the tumor revealed complete removal of an aspergilloma, composed of partially concentrically arranged fungal mycelium with characteristic uniform-appearing branching of the fungal hyphae (Figure 2). Invasive fungal elements were not detectable.

Surgical therapy was supplemented by a perioperative antibiotic as well as antimycotic therapy. With regular

rinsing of the maxillary sinus, the patient's symptoms decreased. In addition, type II diabetes could be controlled with various medications towards a stable blood sugar level. The outpatient follow-up revealed no evidence of a recurrence and/or recurring symptoms of sinusitis as well as stable blood sugar levels.

Ethics Approval and Consent to Participate

Written informed consent was obtained from the patient.

Discussion

The symptoms in patients with CS are diverse and often manifest themselves in less pronounced forms compared to AS (5, 6). Consequently, a correct diagnosis is often difficult. Signs of CS are, in particular, increased nocturnal retronasal secretion ("postnasal drip"), recurrent nasal congestion, intermittent olfactory disorders and, in special cases, a feeling of pressure or pain in the area of the affected paranasal sinuses (11). Risk factors for CS are ventilation disorders of the paranasal sinuses, such as those caused by concha bullosa, turbinate hypertrophy and dental infections and/or treatments (8, 12). Therefore attention should be paid to indications of odontogenic causes for CS and a co-assessment by the Department of Oral and Maxillofacial Surgery should be considered. Nasal endoscopy and cross-sectional imaging by computer tomography and/or MRI examination of the paranasal sinuses are of particular importance in the diagnosis

of CS (13). Polypoid mucosal swellings and rare causes of CS, such as aspergilloma, can often be identified here (10, 13). If the findings are unclear, additional laboratory tests and incisional biopsies can offer added value in the diagnosis of CS. Aspergilloma is a localized form of aspergillosis. Aspergillosis manifests itself in particular in the airways after inhalation of the fungus. It can be divided into the form of invasive aspergillosis with partly fulminant courses, allergic aspergillosis and the locally limited non-invasive form in the sense of an aspergilloma (14, 15).

Aspergilloma occurs as a non-invasive form of aspergillosis and is rarely localized in the area of the paranasal sinuses. Although fungal sinusitis is rare, accounting for up to 9% of all sinusitis, *Aspergillus fumigatus*, with up to 90%, is the most common type of fungus with a preferred localization in the maxillary sinus (16, 17). Fungal infections often present as opportunistic infections in immunocompromised patients (18). It is generally known that systemic diseases, such as type II diabetes, affect the immune system and consequently promote opportunistic infections (10). However, direct relationships between aspergilloma-associated sinusitis and inadequately controlled type II diabetes mellitus have rarely been described. Other risk factors that promote the development and maintenance of chronic sinusitis of the maxillary sinus are dental treatment, even in immunocompetent patients (19). Consequently, dental treatment, such as endodontic treatment, in the area of the maxillary teeth with accompanying type II diabetes poses a high risk of developing aspergilloma of the maxillary sinus.

In the context of radiological imaging, aspergillomas often show a typical ring-shaped uptake of contrast agent with subsequent swelling of the mucosa in the area of the maxillary sinus (4, 13). Affected paranasal sinuses show a hypodense rim with hyperdense parts in the middle, which can often be attributed to the storage of heavy metals and/or overpressed root filling material in the CT-scan (4).

Depending on the etiology, there are both conservative and surgical therapies for the treatment of CS. Conservative treatment includes local nasal rinses, treatment with cortisol-containing nasal sprays and, in individual cases, long-term antibiotic therapy (20). Patients with polypoid CS can also receive immunomodulatory therapies (21). If there is no improvement in symptoms through conservative measures, functional endoscopic sinus surgery is the therapy of choice (20).

However, the treatment of aspergilloma as the cause of chronic maxillary sinusitis consists of the immediate surgical removal of the aspergilloma and the accompanying infected mucosal parts in the context of an osteoplastic maxillary sinus operation (14, 17). In addition to endonasal approaches, an intraoral approach *via* the anterior wall of the maxillary sinus is particularly suitable. In order to ensure

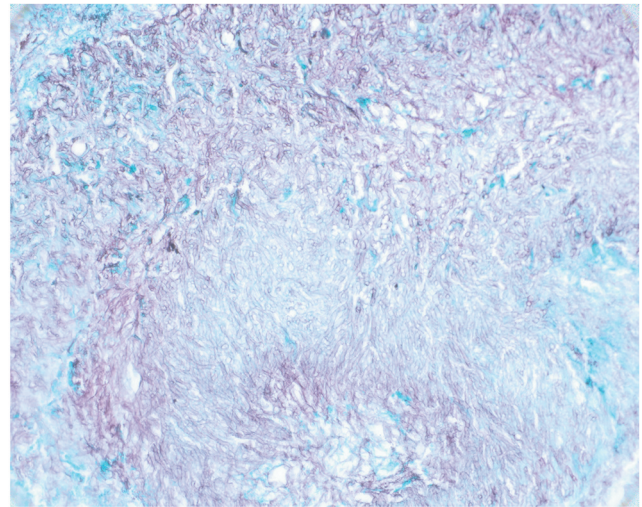


Figure 2. Grocott's methenamine stain of the excised specimen. Massively assembled, partially concentrically arranged fungal mycelium, with hyphae appearing to branch at accurate 45-degree angles within the mucosa can be observed (magnification $\times 400$).

adequate ventilation, the osteoplastic maxillary sinus operation should be supplemented with a supratubinal antrostomy including drainage of the affected maxillary sinus. The definitive diagnosis of aspergilloma is made by microbiological and/or histopathological examinations. Surgical therapy should be supplemented with antibiotic and antimycotic therapy (22). In order to prevent recurrences of aspergilloma, it is particularly necessary to adequately control diseases that influence the immune system, such as an underlying diabetic disease (9).

Currently, there is no precise information on recurrence rates for chronic sinusitis due to aspergilloma in type II diabetes patients. However, the recurrence rate in patients with chronic sinusitis, regardless of etiology, is up to 60% (23). Regular clinical follow-up including radiological imaging are the key elements in follow-up care for aspergilloma of the maxillary sinus in order to diagnose possible recurrences (24).

Conclusion

This case report underlines that rare entities, such as aspergillomas, can also be the cause of CS. In particular, patients with previous illnesses relevant to the immune system, such as type II diabetes mellitus, are predisposed to fungal infections, such as Aspergilloma, causing prolonged courses of CS. To the best of our knowledge this case report is one of a few showing the direct connection between an underlying diabetic disease and CS in the context of an aspergilloma following dental treatment.

Conflicts of Interest

The Authors have no relevant financial or non-financial interests to disclose.

Authors' Contributions

KOH, FB, and FD treated the patient and revised the article. FD, and FB researched the scientific literature. AZK provided histopathological findings. FD and FB wrote the article. All Authors gave final approval for publication.

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Received February 8, 2023

Revised February 23, 2023

Accepted March 6, 2023