

# Maxillary sinus infection related to *Aspergillus*: case report and recent updates

Pierpaolo Petrelli<sup>1</sup>  
 Fabiola Rinaldi<sup>1\*</sup>  
 Sofia Rastelli<sup>2</sup>  
 Gianluca Botticelli<sup>2</sup>

<sup>1</sup> Department of Innovative Technologies in Medicine & Dentistry, University of Chieti—Pescara ‘Gabriele d’Annunzio’, 66100 Chieti, Italy

<sup>2</sup> Department of Life, Health and Environmental Sciences, University of L’Aquila, 67100 L’Aquila, Italy

## \*Corresponding author:

Fabiola Rinaldi

DDS, Oral Surgery Post-Graduate Student, Department of Innovative Technologies in Medicine & Dentistry, University of Chieti—Pescara ‘Gabriele d’Annunzio’, 66100 Chieti, Italy, fabiola.rinaldi@studenti.unich.it, 08713554220.

## Abstract

**Aspergillus related sinusitis is a fungal infectious disease that, despite its diffusion, is still not well acknowledged to dental operators. Due to the anatomophysiology characteristics of the maxillary sinus, the diagnosis of non-invasive aspergillus sinusitis relies on clinical and imaging signs, and on the medical history of the patient. Here, we present a clinical case describing a fungus ball, which diagnosis resulted particularly difficult, due to the presence of concomitant dental interventions in the same maxillary area. A 50 years-old woman, without any medical issue, came to the private dental practice complaining about nasal discharge from the left side. Dental arch were subjected to several treatments; to better understand the working plan, an orthopantomogram was performed, revealing a radiopacity in the left sinus. The cone-beam computed tomography showed the obstruction of the maxillary sinus, and clarified the presence of mycetoma calculus. The patient underwent surgical Caldwell-Luc intervention and mycetoma sampling; the following in-vitro investigations revealed the presence of *Aspergillus Niger*. The one-year follow-up showed no recurrence of the infection. The reported case is aimed to underline how mycetoma is a benign condition, still unknown to dentists despite its large diffusion. Moreover, the authors want to highlight that an endodontic treatment can offer a favourable environment to mycetoma formation.**

**Keywords:** mycetoma, aspergillosis, sinuses, fungal sinusitis.

## Introduction

*Aspergillus* sinusitis, firstly reported by Katzenstein et al. in 1983, is an infectious disease that, despite its diffusion, still presents a challenge in diagnosis, management, classification, and characterization (1).

The first organs affected by aspergillosis in the case of the Head and Neck region are the nasal cavity and paranasal sinuses.

*Aspergillus* species are more than 185, but *Aspergillus Fumigatus*, *Aspergillus Flavus*, and *Aspergillus Niger* are responsible for over 95 % of all infections (2). Symptoms include anterior and posterior nasal discharge, epistaxis, nasal polyps, nasal obstruction, headache, proptosis, anosmia, and snoring (3). *Aspergillus* genus includes pathogens without keratolytic enzymes and, therefore, cannot actively penetrate undamaged and intact mucus membrane or skin (4).

Diagnosis is based on infiltrative or non-infiltrative fungal hyphae, sinonasal polyposis, and histopathology with a positive fungal culture of the tissue sampled from surgical intervention (5). An early diagnosis, appropriate pre- and post-operative pharmacological steroids and antifungal molecules, an accurate surgical debridement of the polyps, and adequate drainage represent the successful key points of the aspergillosis sinusitis treatment (6).

The anatomical position of the paranasal sinus can eventually lead to the spread of the infection into the cranial cavity, representing a life-threatening complication of the aspergillus sinusitis. In particular, the infection can spread to orbit due to the bone erosion of the fungal polyps. The infection can run into the middle cranial fossa from the superior orbital fissure and optic canal (7).

Classification of paranasal sinus Aspergillosis includes invasive (acute fulminant, chronic invasive, granulomatous invasive) and non-invasive (fungus ball and allergic fungal rhinosinusitis) forms, which differ in pathophysiology and clinical signs (8).

Due to the anatomical structure of the maxillary sinus and the physiology of Schneider’s Membrane, diagnosis of non-invasive aspergillus sinusitis relies on clinical and imaging signs and the medical history of the patient.

Here we present a case of fungus ball, which diagnosis resulted particularly tricky due to the presence of concomitant dental interventions in the maxillary area.

## Case report

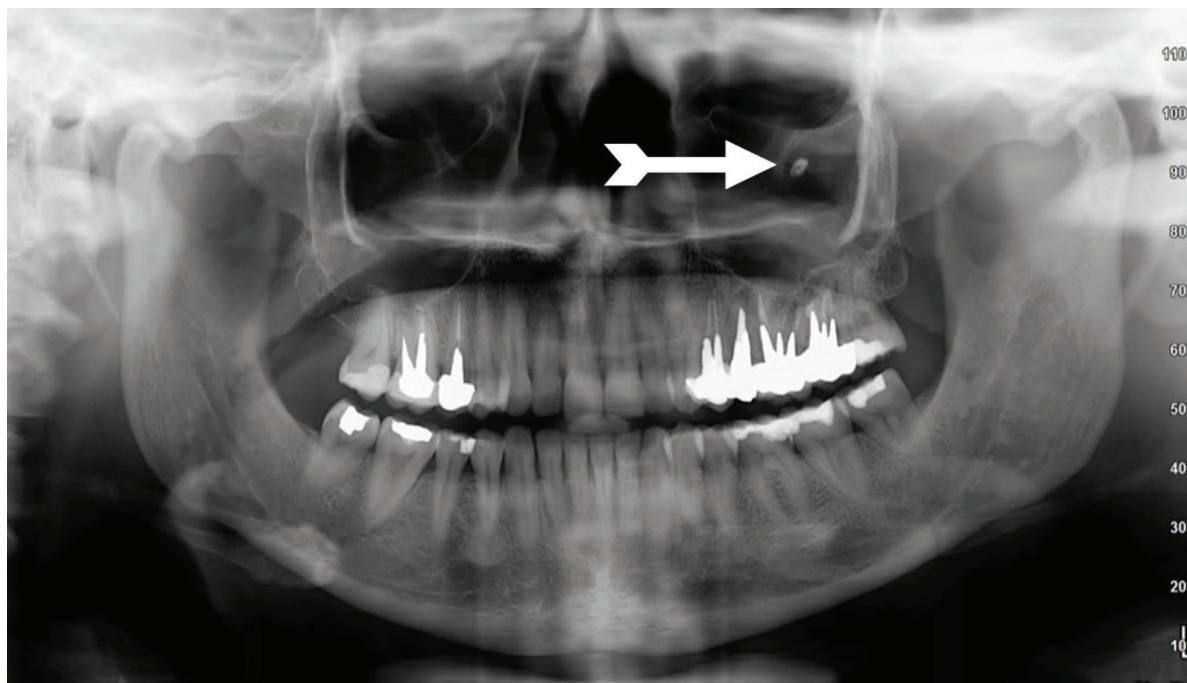
50 years - old woman, resulted with no pathological condition in her medical history, came to the private dental practice complaining about nasal discharge from the left side. Beyond the nasal discharges, no other symptoms were referred.

The left side maxillary teeth presented several treat-

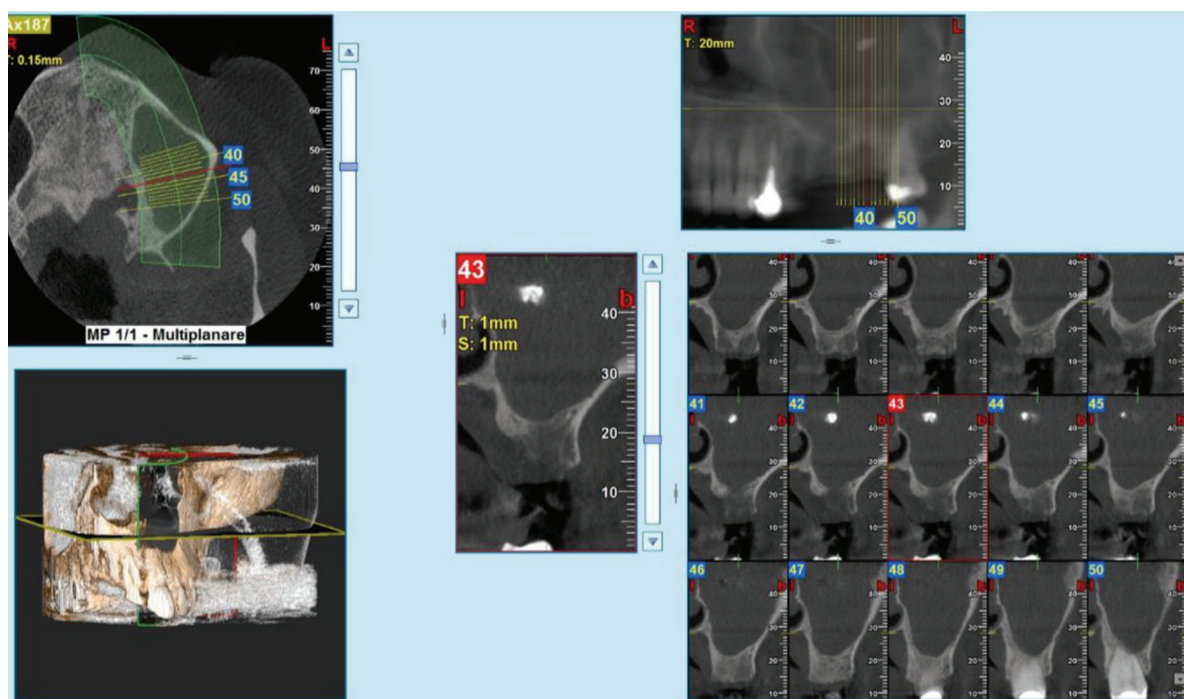
ments, so an orthopantomogram (OPG) was taken to overview the situation.

The OPG revealed a radiopacity in the left sinus, well demarked and defined but unusual and inconsistent with the dental situation, so any endodontic material and dental fixture were excluded (Figure 1).

Successively, the cone-beam computed tomography (CBCT) showed the thickening and obstruction of the maxillary sinus and clarified the presence of mycetoma calculus (Figure 2). Therefore, the patient was referred to the local maxilla-facial department for surgical treatment. Surgical treatment included a Caldwell-Luc inter-



**Figure 1.** Initial OPG. On the left side, it is possible to appreciate the sinus opacity as well as the presence of a material strongly hyperdense, indicated by the arrow. The teeth of the correspondent arches present fixed prosthesis and the presence of several endodontic treatments.



**Figure 2.** CBCT reconstruction. Cross sections confirmed the complete sinus obstruction and the presence of an hyperdense material.



**Figure 3.** One Year follow-up. Teeth have been extracted due to inadequacy to support the crowns and substituted with fixed prosthesis-implant supported. There was no recurrence of the lesion.

vention with the sinus revision and mycetoma sampling for culture, which revealed the presence of *Aspergillus niger*.

The OPG performed at a one-year follow-up showed no recurrence of the infection (Figure 3).

Successively the patient revealed she worked in the countryside, and the source of infection was probably the hay.

## Discussion

The anatomy of the maxillary sinus, which is a cavity, and the histology of the Schneider membrane, make clinical signs of infectious, iatrogenic, or endogenous disease quite similar and challenging for the specialists of the maxillofacial area (dentists, ENTs, maxillofacial surgeons).

Indeed, cysts can expand until they occupy the entire volume of the sinus (9); an iatrogenic disease caused by the presence of materials irritating the sinus mucosa presents with nasal discharge and unilateral sinusitis (10). In contrast, infectious diseases cause the thickening of the membrane, nasal discharge, and usually present as bilateral (11).

Recently, fungal rhinosinusitis incidence increased due to the spread of different pathogens, the aging of the population, and the presence of comorbidities and chronic diseases, such as diabetes and immunological diseases requiring long-term steroid therapy immunosuppressive treatments (12).

In a study by Loidolt et al. (13), approximately the frequency of fungal rhinosinusitis was 10% of all patients who underwent surgical treatment due to chronic sinusitis.

Innovation in medical technology, including advancements in radiological imaging and diagnostic endoscopy, allowed to define the disease classification, diagnosis, and management (8).

The mycetoma or fungal ball consists of a slow but continuous deposition of purulent non-invasive fungal calculi in the paranasal cavities (14). It can present more often unilateral, which is the main difference with allergic fungal sinus, even though cases of more cavities have been reported (15). Mycetoma generally occurs in adults, with a preference for the female gender.

Mycetoma's localization usually is more frequent in the maxillary sinus and more rarely (5%) in the sphenoid sinus (16). Patients suffering from mycetoma do not present a compromised immune system nor significant alterations in the levels of immunoglobulin or IgG subclasses (17). However, the slow development of the disease, the quality of the symptoms, and the characteristics of not-invasiveness, make the diagnosis of mycetoma late. Berry et al. reported 29% of patients receive the diagnosis within a year from the symptom onset (18). Indeed, the quality of the symptoms, which include headache, cough, and facial pain, are similar to chronic bacterial rhinosinusitis; therefore, instrumental diagnosis is fundamental for a differential diagnosis (19).

Risks factors associated with the mycetoma are represented by systemic diseases which can play a key role in the onset of the fungal ball, such as anatomical variants (deviation of the nasal septum), Wegener's granulomatosis, asthma, lymphoproliferative diseases, and diabetes (18). In addition, the previous root canal treatments are the dental conditions that can predispose to mycetoma (4).



The instrumental diagnostic tool of the first level for mycetoma diagnosis is the orthopantomogram (OPG), which allows appreciating the opacification of the maxillary sinus as well as focal radiopacity (50% of cases) pathognomonic signs of mycetoma (20).

Computed tomography (CT) imaging represents the second level of radiological exam. Its accuracy allows to gather information about the nature and the dimensions of the lesion and plan the surgical approach. CT signs of mycetoma are heterogeneous material in the involved sinus, sinus obliteration, and the presence of spot strongly radiopacity (21). The sensitivity and specificity of CT in the presence of these findings were calculated respectively in 62% and 99% (22).

In the reported case, the patient did present the symptoms of chronic rhinosinusitis, with purulent nasal discharge, and previous endodontic treatments in the maxillary sinus involved. Therefore, the instrumental diagnostic tools, OPG and CBCT, allowed performing the diagnosis of mycetoma, confirmed by the subsequent histological exam.

The treatment of mycetoma is surgical, consisting of a Caldwell-Luc intervention, as in our case, using endoscopic techniques<sup>5</sup>. The outcome depends on continuous follow-up and patient compliance. In our case, being the infection's source, the patient was prescribed to stay away from the countryside.

Garofalo et al. in 2016 reported how the pure endoscopic technique might be hard and difficult to perform given the usual position of the fungal ball, in the anterior recess, proposing the gauze technique as safer and easier surgical treatment for the mycetoma (23).

However, in the study, it is reported how the difficult position of the lesion, and therefore a difficult removal, might lead to the relapse (23).

The reported case offered an occasion to underline how mycetoma is a benign condition still unknown to dentists despite its diffusion and how accurate should be an endodontic treatment which can offer an environment favorable to mycetoma formation.

## Conflict of Interest

None.

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