CASE STUDY

Ethmoid-Orbital Mycetoma Caused by Bipolaris sp. Case Report

Micotema etmoidal-orbitario por Bipolaris sp. Presentación de caso

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In February 2013, a male patient aged 44 presented at the surgery with a bilateral nasal obstruction, thick nasal discharge and anosmia, with no ophthalmologic symptoms. He mentioned previous episodes of bronchospasms caused by taking aspirin prescribed as prophylaxis for ischaemic heart disease. The nasal cavity endoscopy study findings showed severe bilateral nasal polyposis with abundant, thick endonasal secretion.

Paraclinical tests: X-rays showed significant opacification of all paranasal sinuses with imaging of calcium intensity at right ethmoid recess level, which had eroded the ethmoidal sinus and had invaded the orbital cavity, causing a slight displacement of the medial rectus muscle (Fig. 1A and B). The CT and MRI scans showed no signs of infiltration or spread of this inside the orbital contents (Fig. 1c and D).

Findings from laboratory tests showed an increase in the number of eosinophils in peripheral blood of 600 per mm³ and high IgE levels of 980 ng/ml.

Diagnoses of inflammatory disease of the upper respiratory tracts, induced by salicylates (Samter and Beers triad, Fernand Widal syndrome) and ethmoid mycetoma with intraorbital compromise were suggested.

The patient received topical nasal steroids (Triamcinolone Acetonide) and bronchial steroids (Budesonide/Formoterol), fluidisers (Carbocisteine), anti-leukotrienes (Montelukast) and antibiotic therapy (clarithromycin).

The patient was scheduled for elective endoscopic resection of the nasal polyposis in the outpatient department. During the right endoscopic ethmoidectomy, we extracted abundant detritus of dark grey necrotic material, along with the resected polypoid mucosa. Part of the thick discharge suctioned along with the polypoid tissue and fungal material was referred for histopathological and microbiological testing respectively.

During the right ethmoidectomy, ocular “bouncing” indicated a major defective area in the external ethmoid wall (lamina papyracea), 1 cm in diameter, without infiltration or rupture of the fibrotic membrane surrounding the intraorbital contents. Subsequent to endoscopic resection, the sinus cavities were carefully irrigated with a saline solution and carboxymethylcellulose dressings were applied (Sinu-Knit® Nasal Dressing, ArthroCare ENT) between the nasal conchae and the lateral nasal wall of each side, to prevent post-operative middle meatal adhesions.

The histopathological study findings showed the presence of polypoidal fragments which presented a mucous epithelium with a stroma infiltrated with numerous eosinophils, lymphocytes, plasmatic cells, mastocytes and neutrophils and Charcot–Leyden crystals (Fig. 2A). Grocott’s Methanamine Silver stain showed up multiple fungal spores with septated structures and yeast infected elements inside the polypoid epithelium stroma.
Figure 1  (A and B) Coronal and axial CT scans which show significant opacification of the ethmoid and maxillary sinuses, with imaging of regular appearance and calcium intensity, which led to the erosion of a part of the right ethmoidal sinus and had invaded the orbital cavity causing a slight displacement of the medial rectus.  (C and D) Coronal and axial MRI imaging in T2 which show opacification of the ethmoid maxillary sinuses with variable type changes.  The variable changes in the paranasal sinuses affected in this case relate to the presence of inflammatory tissues with discharges of high glycoproteins concentrations and low concentration of free water (allergic mucine).  In the right ethmoid recess there is a hypointense image which correlates with that present in the CT scan.  The low signal capture in T2 or “empty signal” is due to the high concentration of different iron and manganese metals concentrated by fungal microorganisms.  This low signalling imaging is regular and no infiltration signs of repercussion are noted on intraorbital content level.

Figure 2  (A) Histologic cross-section (haematoxyline-eosin) imaging in in it in which the infiltrated respiratory epithelium stroma with numerous eosinophils, lymphocytes, plasmatic cells, mastocytes and neutrophils with Charcot-Leyden crystals is appreciated.  (B) Grocott’s Methanamine Silver stain with septated structures and yeast infected black stained corpuscles.  (C) Culture in Sabouraud agar inhibiting agent showing a growth of colonies with black velvety and dark grey spores.
A culture of the necrotic material referred to as mycetoma was made in Sabouraud agar with dextrose and chloramphenicol, showing a growth of colonies with black velvety and dark grey spores. Findings under the microscope were consistent with *Bipolaris spicifera*.

Post-operative progress of the patient was satisfactory. The patient did not receive anti-fungal treatment but from week 3 after surgery he began to receive immunotherapy. Up to the 6th post-operative month the patient did not present recurrent polyposis, or bronchial asthma episodes and recovered some olfactory function which had been impaired prior to surgery.

**Discussion**

Due to their ubiquitous nature, fungal spores are continuously inhaled and come into contact with the upper respiratory system’s mucosa. The reason why fungus may be found in the upper respiratory system of immunocompetent patients without causing any disease whilst in other patients the opposite occurs, is unclear. However, several authors have shown that in allergic fungal rhinosinusitis (AFRS) the individual becomes hypersensitive to the fungus.

Marple reviewed the different physiopathological theories in AFRS, showing that the eosinophilic response of the host to the presence of fungus inside the nose and paranasal sinuses is what leads to clinical presentations of disease (nasal polyposis, formation of expansive mucocelle, allergic fungal mucine, etc.). In this case, activation would stem from the inflammatory cascade and be accompanied almost simultaneously by other factors such as: IgE mediated sensitivity (atopy), HLA T cell specific receptor expression, exposure to specific fungi and aberration of the local defence mechanisms of the mucosa.

According to Fokkens et al., there are 5 known forms of fungal disease which affects the nose and the paranasal sinuses: acute invasive AFRS (including rhinocerebral mucormycosis), chronic invasive RHA, invasive granulomatous AFRS, mycetoma (fungal ball) and non-invasive AFRS.

The *dematiaceous* fungi (black yeasts) belonging to the *Bipolaris* sp. genre and its taxonomy was established in 1986 by McGinnis et al., after studying multiple cultures. del Palacio et al. states that in several works published previously, many cultures processed by infections produced by *Bipolaris* sp., had been reported as displaying growth of a "contaminating fungus", and as a result many infections from this fungus and other *dematiaceous* species were not recognized.

This fungus has been pointed out in literature6,9 as the aetiological agent of mycotic infections in children and adults infected by the human immunodeficiency virus and in atopic immunocompetent patients with nasal polyps who do not respond to conventional medical treatment. Several authors state that *Bipolaris* sp. is an opportunistic pathogen, characteristic of warm, wet climates and is frequently related to immunocompetent patients with mycetomas and AFRS.

**Conflict of Interest**

The authors have no conflicts of interest to declare.

**References**