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Chronic rhino-orbito-cerebral mucormycosis: A case report

Dhib I[1], Yaacoub A[1], Bellakhdhar M[2], Ben Messoud NS[3], Mooatamri R [4], Abdelkéfi M[2], Khochtali H [4],Fathallah A[1]
1-Parasitology Mycology laboratory, Farhat Hached hospital, Sousse Tunisia.
2- Department of Otolaryngology Head and Neck surgery, Farhat Hached hospital, Sousse Tunisia.
3- Medicine and oral surgery unit, dental medicine department, Farhat Hached hospital, Sousse Tunisia.
4- Maxillofacial surgery department, Farhat Hached hospital, Sousse Tunisia.

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Introduction

Chronic mucormycosis, a new and emerging clinical entity in immunosuppressed and immunocompetent patients, is a rare presentation that exhibits a challenging diagnosis. We report herein a case of rhinocerebral mucormycosis in a diabetic woman with favorable outcome.

Observation

A 56-year-old diabetic female was admitted on March 2017 to the Otolaryngology Head and Neck surgery department of Farhat Hached hospital for long-lasting left pansinusitis of dental origin and uncontrolled diabetes. The patient was started on levofloxacin for 10 days. Despite antibiotic treatment, the state of the patient worsened and she exhibited left hemiface edema and left half palate swelling. Nasal endoscopy revealed a necrotic appearance of the mucosa with blackish secretions from the nasal cavity. The patient underwent left middle meatotomy, left maxillary sinus debridement, and necrotic tissue biopsy. Histopathological examination of necrotic tissue revealed thick, non-septate perivascular hyphae branching at right angle, which is consistent with mucormycosis. Direct mycological examination showed large and non-septal hyphae (fig.1) but culture was negative. The cerebral and facial computed tomographic scan objectified an aggressive left ethmoido-maxillary sinusitis with significant bone lysis, extension to orbit floor, infra-temporal space, left pterygopalatine fossa and intra-cerebral invasion (fig.2). Amphotericin B treatment was initiated and surgical debridement was performed at the Maxillofacial Surgery Department of Sahloul Hospital. The patient improved on antifungal, surgical treatment and diabetes control. At 2 years follow-up, the outcome was favorable.

Fig.1: Direct mycological examination: mucorale hyphae.

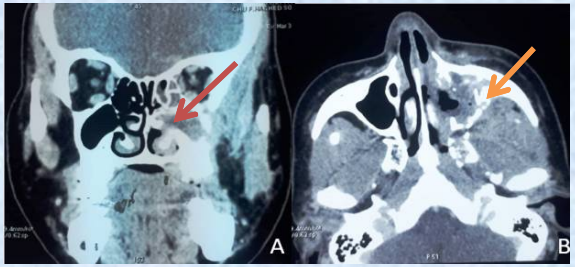

Fig.2:Facial computed tomographic scan (A: sagittal plane, B: axial plane): aggressive left ethmoido-maxillary sinusitis → with significant bone lysis → and extension to infra-temporal space

Discussion and conclusion

Rhinocerebral mucormycosis can be acute or chronic, with the latter having a low frequency (5.6% of rhinocerebral mucormycosis cases) [1]. Chronic or indolent mucormycosis of the paranasal sinuses was described initially in 1964 by Vignale [2]. There is no exact definition of chronicity, which can vary from weeks to months. Classically, it is defined by symptoms that last for more than 4 weeks [1]. In our case, symptoms had lasted 1 month. Cases of indolent mucormycosis are being reported occurring among immunosuppressed and immunocompetent patients [3].

Our patient was diabetic. In the chronic infection, the clinical characteristics are distinguished by chronic evolution, nonspecific symptoms and in some cases absence of frank vascular invasion and necrosis [3]. It is possible that the causative mucorales may show a decreased ability to grow at body temperature, compromising their angioinvasive nature [4]. In the majority of the cases of chronic rhinocerebral mucormycosis, diagnosis was based on histopathology. In case of positive culture, the implicated mucorales were *Mucor ramosissimus*, *Rhizopus nigrans*, *Rhizopus spp*, *Mucor irregularis* and *Apophysomyces variabilis* [1,4,5]. In our case, the causative agent was not identified, but histopathological examination and direct mycological examination of necrotic tissue showed mucorale hyphae.Treatment is similar in both presentations (acute and chronic) including wide surgical debridement of involved tissue and systemic amphotericin B therapy with control of underlying comorbid factors [1,6]. Survival rate in chronic mucormycosis cases (83%) is higher than acute cases (10–35%) [1]. In our case, the patient improved on antifungal, surgical treatment and diabetes control.

In conclusion, chronic rhinocerebral mucormycosis is a rare presentation that requires a high index of suspicion due to atypical presentations.

References

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