Concurrent Chromomycosis and Aspergillosis of the Maxillary Sinus

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The authors describe a rare case of an infection of both the chromomycosis and aspergillosis of the maxillary sinus in an immunocompetent 72-year-old female who presented with progressive visual loss and dull aching pain of the left eye. Sinuscopy of the left maxillary sinus showed swelling of the mucosa with clay-like materials. Biopsy from the left maxillary sinus showed the typically characteristic morphology of chromomycosis and culture from sinus tissue which yielded Aspergillus. The patient responded to a combination therapy of surgical excision and antifungal agent.

Keywords : *Chromomycosis, Aspergillosis, Sinusitis, Sinus*

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Chromomycosis or chromoblastomycosis is a chronic infection caused by dematiaceous fungi that involve cutaneous and subcutaneous tissue[1-3]. Chromomycosis is characterized by nodules, tumors, plaques, warts, and scaring lesions[2]. The lower extremities are the most frequently infected sites, with lesions occurring less frequently on the shoulders, chest, trunk and face[3]. The etiologic agents of chromomycosis include *Fonsecaea pedrosoi, Fonsecaea compacta, Phialophora verrucosa, Cladosporium carrionii, Botryomyces caespitosus, Rhinocladiella aquaspersa, Exophiala spinifera,* and *Exophiala jeanselmei*[4].

Chromomycosis occurring at a site other than the skin is extremely rare. This disease can rarely occur as mycotic infection in the brain[5-8]. To our knowledge, there is only one case report of chromomycosis of paranasal sinus in the literature[9]. Here, the authors present a case of maxillary sinusitis due to concurrent chromomycosis and aspergillosis.

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Case Report

A 72-year-old woman with underlying hypertension and hyperlipidemia presented with progressive visual loss of the left eye for a month. She gradually developed a dull aching pain and loss of vision in the left eye. The symptoms were not aggravated by eye movement. She had neither fever nor nasal discharge. Her current medications were atenolol and atorvastatin. She denied taking either corticosteroid or herbal medicine.

On physical examination, the vital signs were within normal limit. There was no proptosis, paranasal tenderness, or lymphadenopathy. An ophthalmologic examination showed a visual acuity of hand movement in the left eye and 20/100 in the right eye. Both fundi appeared to be normal discs, vessels and macular. There was positive relatively afferent pupillary defect in the left eye. Other neurological examinations revealed normal findings. The physical examinations of other systems were unremarkable.

CT scan of the brain and orbits revealed an ill defined enhancing soft tissue thickening involving the lateral part of the sphenoid sinus, carvernus sinus, superior orbital fissure, optic canal, inferior orbital

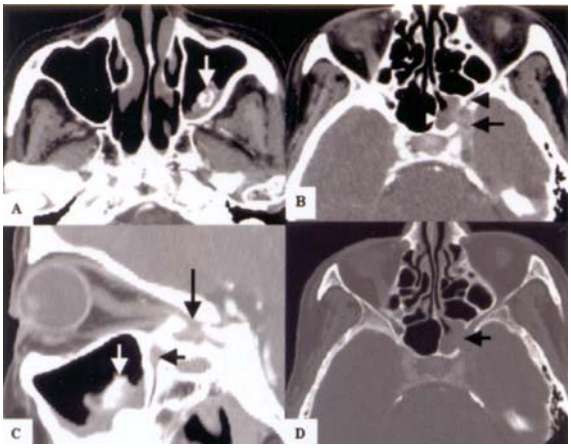

Fig. 1 Hyperdense fungal ball in the left maxillary antrum (white arrow in A and C). Ill-defined enhancing soft tissue lesion occupying the left sphenoid sinus (white arrowhead in B and long black arrow in C), left cavernous sinus with thrombosis (black arrow in C), inferior orbital fissure (black arrowhead in B) and pterygopalatine fossa/fissure (small black arrow in C). Destruction of the lateral bony wall of the left sphenoid sinus (small black arrow in D) is also observed indicating fungal sinusitis

fissure, and pterygopalatine fissure on the left side. The left optic nerve was slightly enlarged. There was also mucosal thickening of the left maxillary sinus with hyperdensed content and sclerotic sinus wall. Fig. 1 shows a hyperdense fungal ball in the left maxillary antrum, an ill-defined enhancing soft tissue lesion occupying the left sphenoid sinus with destruction on the lateral wall. The most likely provisional diagnosis was chronic sinusitis due to fungal infection. Sinuscopy with tissue biopsy of the left maxillary sinus was performed and showed swelling of the mucosa with clay-like materials. Septate hyphae were found from gram stain and Giemsa stain of the sinus tissue.

Histopathology revealed severe acute and chronic inflammation. There were brown, spherical large thick wall cells measuring about 10 m in diameter compatible with chromomycosis (Fig. 2). The special stains (Gomori's methenamine silver and periodic acid-Schiff) were positive. The diagnosis was invasive fungal sinusitis, most likely to be chromomycosis.

On the third day of admission, the patient developed diplopia from left lateral rectus palsy (Fig. 3). A blood test for chemistry showed a plasma glucose of 97 mg/dl, and a creatinine of 0.6 mg/dl. On the sixth day of admission, an operation for left partial medial maxillectomy with left sphenoidotomy was performed. Operative findings revealed swelling of mucosa and clay-like material in the left maxillary sinus. There was polypoid change of mucosa of ethmoid sinus. Sphenoid sinus appeared normal. Culture of sinus tissue from the operation and the pus from sinuscopy yielded *Aspergillus flavus*. Amphotericin-B with a total dosage of 2 grams was administered initially. Then the medication was switched to oral itraconazole 400 mg/day after her neurological symptoms had been alleviated. Two months later, sinuscopy was repeated and revealed only minimal granulation tissue at the sphenoid opening. Histopathology of the sinus tissue from biopsy revealed no organism. Itraconazole was continued for 6 months. The patient had nearly complete recovery from left lateral rectus palsy but visual acuity has been persistent at hand movement.

Discussion

Generally, the frequent invasive fungal sinusitis was aspergillosis and mucormycosis[10,11]. In the present case , histopathological examination of sinus tissue revealed the typical fungal elements that were compatible with chromomycosis. Diagnosis of

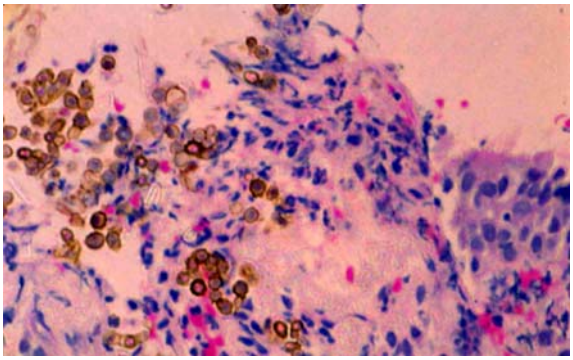

Fig. 2 Chromomycosis. Lesion of the maxillary sinus. Observe the brown, spherical, thick-walled fungal cells. H&E. x400

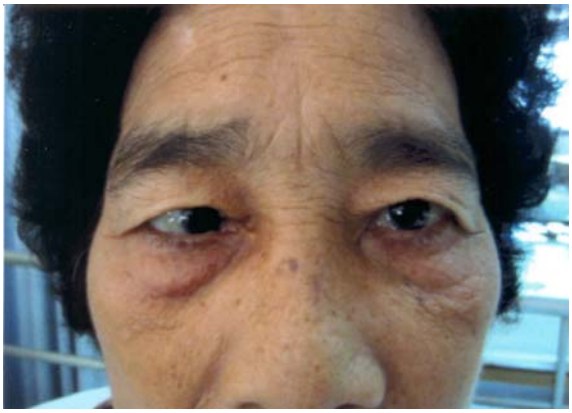

Fig. 3 Left lateral rectus palsy of left eye

chromomycosis is primarily made by morphology, which is sufficient to recognize the disease. The diagnosis should be confirmed by culture on Sabouraud dextrose agar containing chloramphenicol and cycloheximide and kept at 25-30°C for 4-6 weeks[12]. Unfortunately, only *Aspergillus flavus* had been recovered from the tissue culture. Overgrowth of Aspergillus on the media might be explained. The repeated subculture was performed but the results were the same. However, these typical morphological appearances from histopathology are sufficient to diagnose chromomycosis[3,9].

The combination of adequate surgical excision and potent antifungal agents are the cornerstone for treatment of invasive fungal sinusitis [13,14]. In the present case, extensive surgical excision was performed and high dose amphotericin B therapy was given. A previous reported case of sinus chromomycosis also had a good clinical outcome from combined surgical excision and chemotherapy with amphotericin B and 5-fluorocytosine[9]. Antifungal therapy in the presented patient was switched to itraconazole because currently, itraconazole is the promising drug for both chromomycosis and aspergillosis[15-18]. In general, amphotericin B is not recommended for the treatment of chromomycosis, according to the high minimal inhibitory concentration of this fungus[19]. Amphotericin B was chosen for antifungal therapy in the presented patient because of the presence of concurrent aspergillosis. In addition to the extensive surgical excision, the subsequent itraconazole therapy on the present patient may have played an important role to achieve complete recovery of concurrent aspergillosis and chromomycosis of the sinus.

In conclusion, the case that authors reported here demonstrates the concurrent chromomycosis and aspergillosis of the sinus in an immunocompetent host. Chromomycosis can occur as a rare cause of invasive fungal sinusitis. Combined surgical excision and antifungal therapy is recommended for the treatment.

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รายงานผู้ป่วยที่มีไซนัสอักเสบจากเชื้อราโครโมไมโครสิสและแอสเปอจิลโลสิส

วีรวัฒน์ มโนสุทธิ, สมนึก สังฆานุภาพ, พูนพิลาส หงษ์มณี, พัฒนา สรมยุระ, จิรภร เหล่าธรรมทัศน์, วีระพล ประณีตวตกุล

รายงานผู้ป่วยภูมิคุ้มกันปกติที่มีการติดเชื้อราโครโมไมโครสิสและแอสเปอจิลโลสิสที่ไซนัสแมกซิลลารี่ในผู้ป่วยหญิงไทยอายุ 72 ปีที่มีอาการปวดตาซ้ายและมองภาพไม่ชัดเจน การส่องกล้องพบว่าภายในไซนัสแมกซิลลารี่มีลักษณะคล้ายดินโคลน ผลชิ้นเนื้อจากบริเวณดังกล่าวพบลักษณะทางกายวิภาคที่เข้าได้กับเชื้อราโครโมไมโครสิสและผลการเพาะเชื้อราขึ้นเป็นแอสเปอจิลลัส ผู้ป่วยได้รับการรักษาและอาการดีขึ้นด้วยการผ่าตัดและยาต้านเชื้อรา