

Look before you leap: A case series of conidiobolomycosis from the Indian subcontinent

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ABSTRACT

Conidiobolomycosis is an extremely rare subcutaneous rhinofacial indolent infection caused by *Conidiobolus coronatus* and *Conidiobolus incongruus*. It is reported mainly from the tropical and subtropical parts of the world, mostly in form of isolated case reports or small case series. Two immunocompetent male patients presented to our center with indolent nasal swelling and features of nasal obstruction. They were treated outside with antibiotics and/or steroids and were referred to us with nonresolving symptoms. Both of them were diagnosed based on the characteristic histopathological findings and were successfully treated with saturated solution of potassium iodide. Conidiobolomycosis is an important differential in patients presenting with centrofacial swelling. Early suspicion and timely referral by the primary care physicians will help in early diagnosis and treatment of this disease.

Keywords: *Conidiobolus coronatus*, *Conidiobolus incongruus*, rhinofacial entomophthoromycosis

Introduction

Conidiobolomycosis is a chronic subcutaneous rhinofacial entomophthoromycosis caused by *Conidiobolus coronatus* and rarely, *Conidiobolus incongruus*.^[1] It is a rare disease reported mainly in the tropics. It is primarily seen in immunocompetent individuals with a significant male preponderance. The diagnostic modality of choice is culture and histopathological demonstration of aseptate hyphae with characteristic Splendore–Hoeppli phenomenon.^[2] The treatment for this disease is a combination of surgery and antifungals (saturated solution of potassium iodide/azoles).^[3] We report two cases of nasal swelling and obstruction in immunocompetent males that were treated prior to presentation with steroids and/or antibiotics but were diagnosed later as conidiobolomycosis.

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Case Reports

Case 1

A 17-year-old male patient, resident of Nepal, with no other comorbidities, presented with slowly progressive painless nasal swelling, ulcer on the left cheek, and features of left nasal obstruction for 1 year. Prior to his presentation at our hospital, he was evaluated extensively. His routine hemogram and biochemical parameters were normal. Magnetic resonance imaging of the paranasal sinuses showed a mass in the left nasal cavity and nasolabial fold [Figure 1]. An incisional biopsy was done outside from the mass that showed features of chronic granulomatous inflammation. With a provisional diagnosis of sarcoidosis, he was started on oral steroids outside for 2 months with partial reduction in size. An excision of the residual mass was done at that time. Steroids were tapered slowly, following that there was an increase in nasal swelling that had infiltrated the cheeks also. The patient was referred to us with nonresolving symptoms. On examination, he was found to have bilateral nasal swelling and an excavated ulcer at

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the junction of nose and the left cheek [Figure 2]. There was a polypoidal mass noted in the left nasal cavity arising from the lateral wall of nose. A biopsy was repeated at this time from the intranasal mass that showed broad aseptate hyphae with Splendore–Hoepli phenomenon on histopathological examination. With a diagnosis of possible conidiobolomycosis, the patient was started on saturated solution of potassium iodide (SSKI) and voriconazole. A follow-up at 5 months showed significant improvement.

Case 2

A 52-year male patient from Bihar with no other comorbidities, presented with nasal discharge and nasal obstruction for 8 months. He also had pain and swelling over the right side of the nose for last 6 months. He was evaluated outside but no diagnosis was made. He was treated with multiple antibiotics prior to the presentation. On examination, the skin overlying the nasal swelling was erythematous and thickened. There was a pink mass noted in the right nasal cavity arising from the lateral wall of nose. His routine hemogram and biochemical parameters were normal. A biopsy done from the mass showed broad aseptate hyphae with no angioinvasion and Splendore–Hoepli phenomenon on histopathological examination [Figure 3]. He was treated postoperatively with itraconazole and saturated solution of potassium iodide. He was doing well at the follow-up done at 6 months.

Discussion

Conidiobolomycosis is a fairly uncommon disease with isolated reports from the tropical and subtropical zones of Africa, Asia, and America.^[4] Isolated case reports from the Indian subcontinent has been reported mainly from the states of West Bengal, Punjab, Tamil Nadu, and Delhi.^[5–8] It is a chronic inflammatory granulomatous disease that involves the nasal mucosa and presents as nasal obstruction and swelling of the nose (intranasal and/or extranasal). The possible mechanism of acquisition of this disease is inhalation of spores from environmental sources.^[9] It is seen in immunocompetent individuals and no known risk factors are described. It has a slow indolent course and progresses over the course of months to cause firm subcutaneous nodules and starts becoming attached to the subcutaneous tissues. They usually have a clear line of demarcation from surrounding structures. It may rarely invade the orbit and the brain. Imaging may help in delineating the extent of the disease.^[9]

The common differential diagnosis for conidiobolomycosis includes the following: mucocutaneous tubercular and nontubercular mycobacterial infections, chromoblastomycosis, sporotrichosis, rhinosporidiosis, nocardiosis, squamous cell carcinoma, and cutaneous T-cell lymphoma. Due to the benign and indolent nature of the disease, these patients initially present to the primary care physicians. The diagnosis is often missed initially due to the rarity of the disease and lack of awareness among the treating primary care physicians. They are often

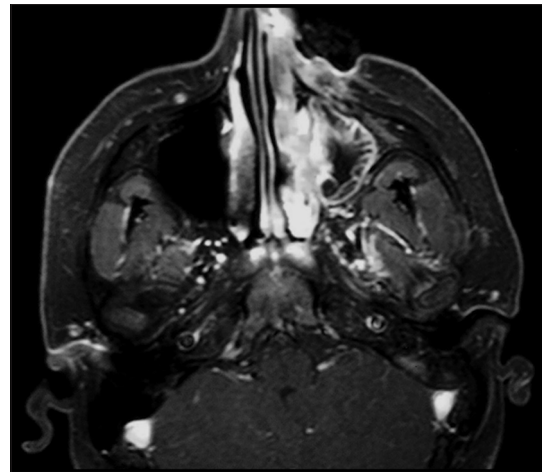


Figure 1: Postgadolinium T1-weighted images showing nodular enhancing soft tissue infiltrating the skin overlying the nose, causing ulceration. In addition, there is invasion of the left nasal cavity and maxillary sinus



Figure 2: Nasal swelling and ulceration of the left cheek

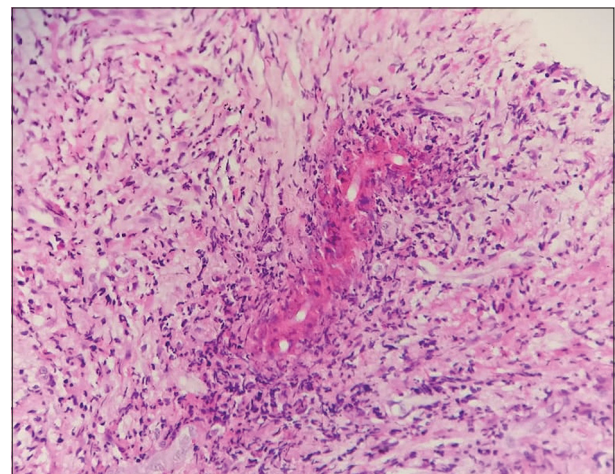


Figure 3: Histopathological section shows subcutaneous dense inflammatory infiltrate comprising eosinophils, lymphocytes, and occasional giant cell. There are few broad aseptate hyphae showing Splendore–Hoepli phenomenon

treated with multiple courses of antibiotics and/or steroids before they are referred to otorhinolaryngologists/infectious disease specialists.

Conidiobolomycosis should be kept in differentials in patients presenting with painless centofacial swelling and symptoms of nasal obstruction. It can be easily diagnosed with timely biopsy and histopathology/culture. The presence of aseptate hyphae and Splendore–Hoeppli phenomenon helps in clinching the diagnosis in most cases.^[4] It can be treated successfully with azoles or SSKI with or without surgery.^[9]

There is a need to create awareness about this rarely diagnosed but prevalent fungal disease among the primary care physicians, as they are very often the first health-care contact. Early suspicion and prompt referral by the primary care physician will help in early diagnosis, early initiation of treatment, and better outcomes.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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