Conidiobolomycosis: a diagnostic challenge (case report)

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Conidiobolomycosis: a diagnostic challenge

Introduction

Conidiobolomycosis is a rare chronic granulomatous fungal infection affecting the upper respiratory mucosa and adjacent subcutaneous tissues in immunocompetent male patients[1]. Often, it is challenging to arrive at a timely diagnosis because of a low index of clinical suspicion and difficulty in culturing the fungus[2]. However, it usually responds well to antifungal agents when diagnosed correctly. We present a case of rhino-facial conidiobolomycosis in a patient who was misdiagnosed at various hospitals before seeking treatment at our tertiary-level center. This case report has been reported in line with the SCARE Criteria[3].

Case presentation

Our patient, a 21-year-old previously healthy male student, presented with a progressively enlarging painless nasal swelling extending onto the cheek and obstruction of the right nasal passage since 3 months (Fig. 1). He was referred to our center after failing to respond to treatment at various hospitals. He had no history of trauma, agricultural exposure, diabetes, and risk factors for an immunocompromised status.

Nasal endoscopy revealed a mass arising from the lateral wall of the right nasal cavity that did not bleed to touch. A computed tomography (CT) scan of the paranasal sinuses revealed a lesion in the right nasal cavity that eroded the inferior turbinate and extended into the subcutaneous tissue and deep fascia of the face anteriorly, without intracranial or intraorbital extension (Fig. 2). The cervical lymph nodes were normal in size. We decided to proceed with functional endoscopic sinus surgery (FESS) and optimized the patient preoperatively in line with our hospital protocols, keeping the patient nil-by-mouth 6 hours prior and reviewing blood investigations, which were within normal limits. During FESS (performed by the first author), we excised the right nasal mass and sent it for examination under the review of an infectious disease specialist. A fungal smear from the specimen revealed aseptate hyphae without acute branching (Fig. 3). Histopathologic examination (HPE) confirmed a similar morphology with tissue invasion and the Splendor-Hoppeli phenomenon. The final diagnosis of Conidiobolus coronatum was

Highlights

- Conidiobolomycosis is a rare chronic granulomatous fungal infection.
- It presents with nasal swelling and obstruction in immunocompetent patients.
- The infection usually responds well to Azole antifungals.
- Surgical debridement may be required in cases with obstructive symptoms or bone invasion.

Introduction and importance: Conidiobolomycosis is a rare chronic granulomatous fungal infection affecting the rhino-facial region. It usually occurs in immunocompetent males with agricultural exposure. A high index of suspicion is required to achieve a timely diagnosis in such cases, as the infection usually responds well to early antifungal therapy.

Case presentation: We share a case of this disease occurring in a 21-year-old male presenting with a right nasal mass and external nasal swelling. It could not be diagnosed correctly over 2 months at various hospitals before he visited our center, where we excised the mass via functional endoscopic sinus surgery and identified Conidiobolus coronatum as the causative agent based on histopathologic examination, and MALDI-TOF. The facial deformity resolved after 3 months of therapy with oral Voriconazole.

Clinical discussion: In this report, we discuss the pathogenesis of Conidiobolomycosis, our diagnostic approach, and the use of functional endoscopic sinus surgery, which has not been reported extensively in the existing literature.

Conclusion: In endemic regions, conidiobolomycosis should be considered amongst the differential diagnosis of a nasal mass associated with facial swelling. A multidisciplinary team approach is required to arrive at a timely diagnosis and begin early antifungal therapy.

Keywords: Conidiobolus, Entomophthorales, Fungal infection, Functional endoscopic sinus surgery, Case report

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arrived at by subculturing the isolate and identification with MALDI-TOF. Routine blood investigations were within normal limits.

The patient had an uncomplicated postoperative course and was started on oral Posaconazole initially for 2 weeks considering mucormycosis as a possibility but was switched over to oral Voriconazole for three months after confirming the diagnosis of Conidiobolomycosis. The facial swelling subsided completely after treatment with oral Voriconazole, which the patient adhered to and tolerated well (Fig. 4). A follow-up CT scan of the paranasal sinuses and endoscopic examination after three months and six months did not reveal any residual disease (Fig. 5). The patient was satisfied with the symptomatic and cosmetic improvement and returned to his activities of daily living comfortably.

**Discussion**

*Conidiobolus* is a mold belonging to the order Entomophthoromycota of the class Zygomycetes[4]. *C. coronatus* is the most
frequently isolated species in cases of Conidiobolomycosis in humans, which has been reported mainly in tropical and subtropical countries, including India[5]. The infection is hypothesized to spread by inhalation of spores or trivial trauma to the upper respiratory mucosa [6]. It usually occurs in immunocompetent adult males with agricultural exposure, increasing their risk of coming in contact with spores [7]. We did not suspect Conidiobolomycosis in our patient initially because of the rarity of the disease and the absence of risk factors.

Conidiobolomycosis is characterized by a chronic granulomatous infection beginning in the mucosa over the inferior turbinate, giving rise to a lateral nasal mass that causes nasal obstruction [7]. It spreads via the submucosa to the subcutaneous tissue over the nose, the upper lip, and the cheeks, resulting in a characteristic facial deformity with obliteration of the nasolabial and nasomaxillary creases [8]. It can also spread to the paranasal sinuses via the foramina in the nasal cavities. These features were visible in our patient. In more advanced cases, the infection may spread further to the periorbital region, the pharynx, and the larynx [2]. Systemic dissemination is uncommon. Complications such as nasal obstruction, rhinorrhea, epistaxis, and recurrent bacterial sinusitis may occur and are often difficult to treat, warranting a timely diagnosis and treatment of the infection [4]. The differential diagnoses in these cases are Rhinoscleroma, lymphedema, lymphoma, mucormycosis, and chronic granulomatous aspergillosis [7]. Given the clinical features, fungal smear, and HPE findings, these alternative diagnoses were less likely. Broad, sparsely septate hyphae with an eosinophilic halo around the invading hyphae, known as the Splendore-Hoeppli phenomenon, is a characteristic feature on HPE, as seen in our case [4]. MALDI-TOF, a rapid and sensitive technology in fungal characterization, helped us reach the final diagnosis [8].

Specific protocols for antifungal therapy have not been described because of the rarity of the disease, although successful treatment with a combination of a saturated solution of potassium iodide and azoles has been reported in India [7]. We preferred Voriconazole monotherapy because of the availability of therapeutic drug monitoring at our center and more predictable drug concentrations, to which the patient responded well. The necessity for surgical resection of the nasal mass needs to be tailored for each case and is not usually recommended in uncomplicated cases improving with medical therapy alone [2,9,10]. We resected the mass via FESS to confirm the diagnosis and because it caused significant nasal obstruction. Furthermore, the CT scan revealed that the nasal mass invaded adjacent bony structures, warranting debridement.

Conclusion

Conidiobolomycosis should be suspected amongst the differentials of a nasal mass associated with an anterior facial swelling in an adult male patient. A thorough discussion between a multidisciplinary team at a tertiary care center consisting of an otorhinolaryngologist, a microbiologist, a pathologist, and an infectious disease specialist is required to arrive at a timely diagnosis and decide the line of management, including medical therapy with or without surgery.

Ethical approval

NA.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

V.Bhagia: study design, manuscript preparation, verifying the final draft of the manuscript. V.Bansal: manuscript preparation, verifying the final draft of the manuscript. V.K.: study design, verifying the final draft of the manuscript.

Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

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