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Case Report

Endoscopic endonasal resection of a *Drechslera hawaiiensis* sphenoid fungal ball

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Received: 03 June 2023
Accepted: 22 August 2023
Published: 08 September 2023

DOI
10.25259/SNI_469_2023

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ABSTRACT

Background: Fungal infections should always be considered in difficult-to-treat paranasal sinus conditions. Sphenoid fungal balls are characterized by the presence of dense fungal masses in the sinus cavity without invasion of surrounding tissues. This case emphasizes the importance of accurate terminology and management and also highlights the involvement of rare pathogens such as *Drechslera hawaiiensis*. Diagnosis is typically based on imaging studies and intraoperative findings. Accurate identification of the pathogen is crucial. Fungal infections of the paranasal sinuses, including fungus balls, can present challenges in diagnosis and treatment. *D. hawaiiensis*, although infrequent, can cause potential life-threatening infections.

Case Description: We present a 26-year-old non-HIV male patient who presented with nasal symptoms and mild headaches. The patient underwent an endoscopic exploration that revealed a soft, grayish lesion with a buttery consistency. Gross total resection was achieved and the lesion was identified as being caused by *D. hawaiiensis*; thus, intravenous antifungal treatment was given.

Conclusion: Endoscopic surgery remains the preferred approach for disease control. Considering alternative treatments and exploring novel approaches are essential in managing complex pathologies in neurosurgical practice.

Keywords: Bipolaris, *Drechslera hawaiiensis*, Endoscopic endonasal resection, Mycotic, Skull base surgery

INTRODUCTION

Sphenoid fungus ball refers to the collection of dense fungal masses in the sinus cavity with no invasion of surrounding tissues. The clinical presentation is non-specific and the diagnosis is based on imaging studies. Surgical treatment, often performed using an endoscopic endonasal approach, is usually curative. Fungi may play a significant role in chronic sinusitis and central nervous system infections, as they can be cultured using sensitive methods in over 95% of patients.[11] Therefore, fungal infection should always be considered when dealing with difficult-to-treat paranasal sinuses conditions. Fungal rhinosinusitis can be categorized into invasive and non-invasive forms based on the presence or absence of microscopic evidence of fungal hyphae within the tissues (mucosa, blood vessels, or bone).[4,6] The non-invasive form, known as extra mucosal, is the most common

and is further divided into allergic fungal sinusitis and fungus ball. This disease is further complicated by its imprecise clinical presentation, which often leads to misdiagnosis and subsequent ineffective medical management. Infection by *Drechslera hawaiiensis* is a rare fungal disease that presents with similar clinical, histologic, and serological features as other fungi, which could lead to an erroneous diagnosis and inaccurate treatment.[8]

It is important to avoid using misleading terms such as aspergilloma, aspergillosis, or mycetoma as there are different pathogens involved, such as *D. hawaiiensis* in this particular case. The latter is a rapidly-growing fungus that produces a grayish colony; the hyphae, conidiophores, and conidia are dematiaceous and are found worldwide in the air, florae, grasslands, grains, rotten food, and soil. When affecting humans, this pathogen can produce subcutaneous or systemic infectious disease.[13] This micro-organism taxonomically belongs to the Class *Zygomycetes* and the Order *Mucorales*, which comprises 14 families, of which four are known to cause diseases in humans. Among these, the *Mucoraceae* family is of utmost medical importance, including the genera *Rhizopus*, *Absidia*, and *Mucor.* Infections caused by this type of fungus typically originate in the nasal cavities and paranasal sinuses, or on the palate, and subsequently spread to the orbit and brain.[5] The majority of *Drechslera* sp. infections are caused by the *Specifera* species, while the *Halodes*, *Longirostrata*, *Curvularia*, and *Hawaiiensis* are less common. *D. hawaiiensis* is infrequent but potentially life-threatening.[2,10] *In vitro* studies of the susceptibility of the isolate to various antifungal agents must be carried out, as amphotericin B, miconazole, clotrimazole, and 5-fluorocytosine are the most commonly used antifungals for *Hawaiiensis* species.[9,25]

Drechslera sp. is commonly found in the soil of tropical climates and hardly causes human infection. When this occurs, it usually affects immunocompromised individuals. These fungi are usually soil saprophytes and plant parasites, possessing a low pathogenic capacity; nevertheless, they are capable of invading the tissues of individuals with impaired resistance when they inhale particles from sources such as vegetables, soil, plants, insect bites, wounds, and minor trauma, causing infection to the host.[1] Collectively, these organisms are referred to as opportunistic fungi and they form pigmented spores. Impairment of resistance mechanisms generally appears in individuals displaying malignancy, diabetes mellitus, radiation therapy, antibiotic treatment, immunosuppressive drug treatment, and steroid use, allowing these pathogens to penetrate the skin and manifest as pustules, abscesses, ulcers, and necrotic areas.[22]

CASE DESCRIPTION

We present a 26-year-old male patient, from a rural location in Baja California, Mexico, with no relevant background, presenting with mild intermittent headaches, nasal congestion, rhinorrhea, labored breathing, and anosmia. However, no cacosmia, visual disturbances, facial deformities, or meningeal signs were present. He was previously misdiagnosed with an esthesioneuroblastoma based on imaging studies. Magnetic resonance imaging (MRI) revealed an isodense lesion that appeared irregular and homogeneous, with peripheral enhancement on T1 weighted image with contrast. Invasion of the ethmoid and sphenoid sinuses with a necrotic center was observed. On T2WI, a highly hypointense appearance of the lesion suggests an infectious etiology [Figure 1]. No signs of bony erosion or invasion within the intracranial compartment were observed. He did not receive any treatment and was referred to our clinic. We decided to perform an endoscopic exploration to biopsy the lesion, and if deemed possible and safe, try gross total resection.

Using a 0° angled rigid endoscope, the right nostril was approached first. The lesion was immediately reached, as it was coming out of the middle meatus which was completely occupied by the tumor. The soft and grayish lesion with a very buttery consistency was easily resected using biopsy rongeurs and ringed curettes. As resection continued, this

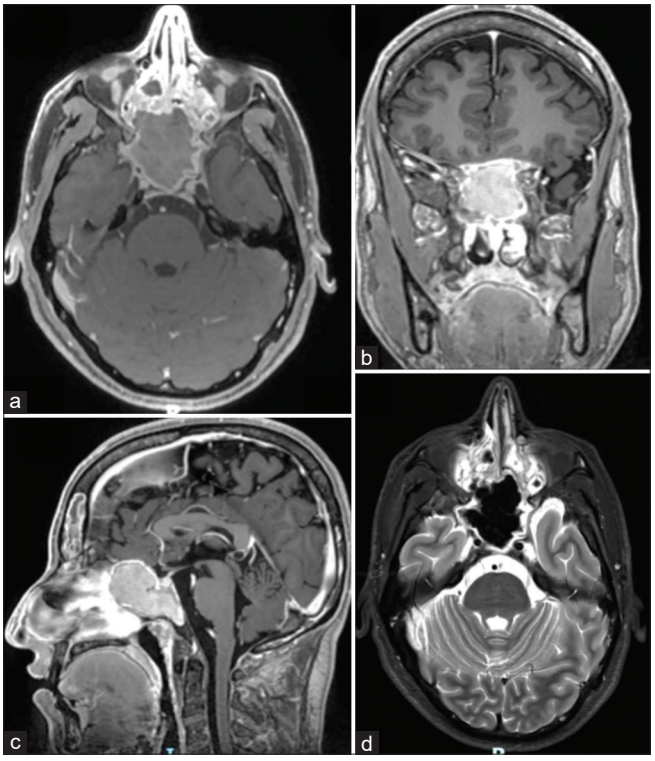


Figure 1: Preoperative T1WI with contrast magnetic resonance imaging: (a) axial, (b) coronal, and (c) sagittal planes in which an isodense irregular lesion can be seen completely invading the sphenoid and ethmoid sinuses. Gadolinium enhancement shows ring enhancement. (d) T2WI sequence revealed a highly hypointense lesion, which is suggestive of an infectious etiology.

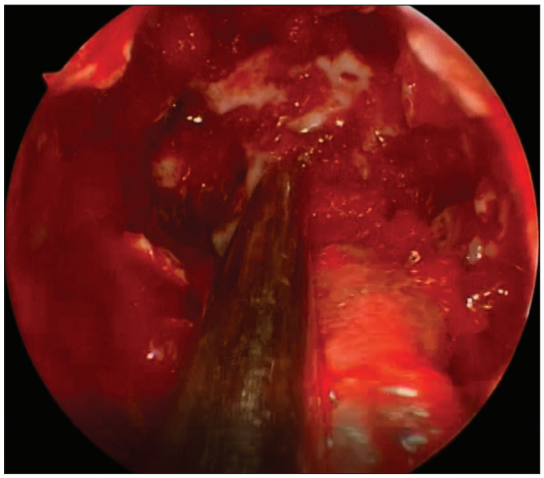
buttery consistency was notorious and this deemed the resection quite simple. Once the middle meatus was deemed completely clean, a right turbinectomy was performed to access the sphenoid and maxillary sinuses. As the lesion had completely destroyed the rostrum sphenoidale, no proper bony landmarks, such as the ostium sphenoidale, were found. Resection of all the reachable portions located within the sphenoid sinus was accomplished through the right nostril first. Subsequently, the left nostril was approached and a left turbinectomy was performed as well. As mentioned, the anterior wall of the sphenoid sinus was completely destroyed as well as the posterior portion of the nasal septum; thus, no posterior septectomy was needed, and access to the sphenoid sinus was quite straightforward. Once gross total resection of the lesion was achieved, bony landmarks within the sphenoid sinus were observed; being the sella turcica and clival recess limited laterally by the parasellar and paraclival segments of the internal carotid artery (ICA), respectively. Neuronavigation was used to confirm the lateral and inferior limits of the lesion located in the clival recess. After resection of the sphenoidal portion of the lesion, bilateral maxillostomies were performed to access the maxillary sinuses. Finally, hemostasis was performed by the placement of gel foam on the surgical site until no active sites of bleeding were observed [Video 1]. No further complications occurred during the procedure, and the patient coursed with an uneventful postoperative course with only mild complaints regarding nasal congestion and nasal scabs, overall, the patient and his family were quite satisfied with the results. Postoperative MRI showed complete resection of the lesion [Figure 2].

The resected material was sent to the microbiology laboratory for fungal studies. Smear preparations with lactophenol cotton blue stain revealed hyphae elements. Cultures on Sabouraud's dextrose agar presented the proliferation of a dematiaceous fungus displaying numerous multi-septate spores. The fungus was determined to be *D. hawaiiensis*, due to its morphological appearance[12] [Figure 3].

DISCUSSION

The utilization of nasal endoscopy and computed tomography scans has increased the frequency of diagnosis for fungal diseases of the paranasal sinuses, including fungus balls.[12] In cases where the patient presents with severe symptoms and standard analgesic treatment fails to mitigate or halt the progression of the disease, surgical intervention may become necessary. Intraoperative observation of a fungal mass displaying pathognomonic features, coupled with definitive documentation of fungal presence through histopathological analysis, smear and culture testing by a specialized microbiology laboratory, lack of evidence of mucosal invasion, and negative serological findings, serve as crucial factors that reinforce the accuracy of the diagnostic process.

A fungus ball is a complex condition that can be categorized into invasive and non-invasive types.[7,17] The clinical symptoms initiate in the nasal fossa and paranasal sinuses and can range from rhinorrhea, nasal obstruction, facial pain, swelling, cacosmia, visual disturbances, sinus



Video 1: Operative video depicting the resection of a sphenoid fungal ball.

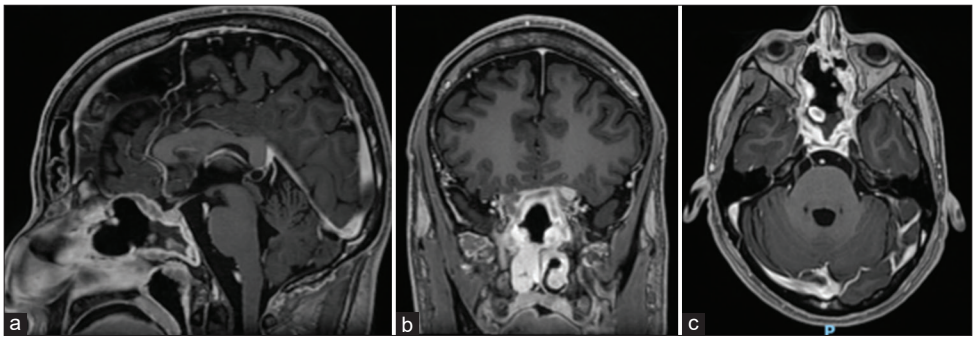


Figure 2: Postoperative T1WI with contrast magnetic resonance imaging: (a) sagittal, (b) coronal, and (c) axial planes revealed gross total resection of the lesion.

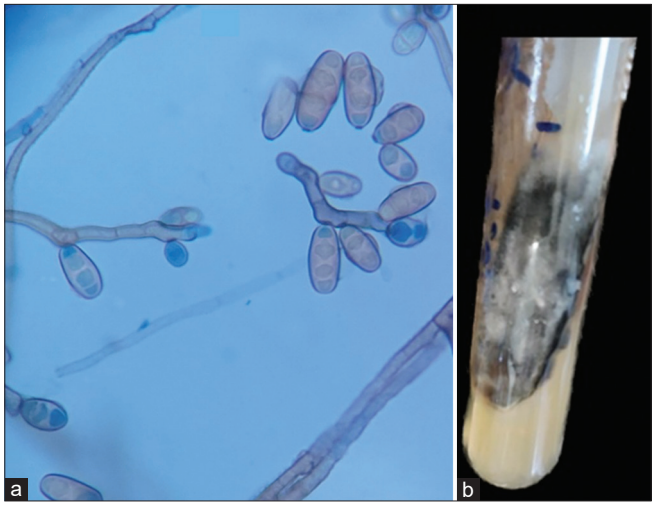


Figure 3: (a) Micromorphology of the filamentous fungus *Drechslera hawaiiensis* using lactophenol cotton blue stain. Hyaline fungi can acquire the blue hue from lactophenol, whereas dematiaceous fungi will exhibit a brown discoloration caused by the mold's pigment production. (b) Macroscopic sample of the thick, greasy, and dark material extracted from the antrum. There was an abundance of dark brown mucous which possessed a smooth and buttery texture, along with buttery consistency and grayish color.

fullness, and bone destruction, gradually evolving from months to years. These symptoms typically do not improve with standard antimicrobial treatment. Involvement of fungal infection in the central nervous system is a serious complication that requires prompt assessment and treatment, especially when complications such as erosion of the cranial base, intracranial expansion, focal epilepsy, and orbital penetration leading to amaurosis are present.[8,20] Radiological evidence of sinus opacification, characterized by heterogeneous opacification, metal-dense spots, and pseudo-tumoral appearance with bone lysis and heterogeneity, is nonspecific.[21] Proper diagnosis involves imaging studies along with additional criteria: the presence of mucopurulent cheesy or clay-like materials within the sinus, a dense conglomeration of hyphae separate from the sinus mucosa, and nonspecific markers of chronic inflammation such as lymphocytes, plasma cells, and eosinophils in the affected tissue. A pathognomonic finding that suggests the presence of fungi is mucinous tissue containing abundant eosinophilic granulocytes, indicative of an allergic process.[23] According to deShazo *et al*., criteria for fungal infections, a thick, greasy, and darkish material originating from the antrum, with copious secretion of dark brown mucus are highly specific.[5,15] Sabouraud agar is considered the standard culture for identifying different types of fungi. *Aspergillus* sp. is the primary cause of colonization within the paranasal sinuses, and due to its high prevalence, diagnostic commercial tests are available; nevertheless, it is crucial to differentiate between various types of fungi, even those with low prevalence rates. While these fungi may be rare, it is important not to overlook them, which is why conducting cultures is always necessary to accurately identify the true pathogen responsible for the disease.

Caution should be exercised when immunocompetent individuals exhibit symptoms suggestive of fungal diseases, with risk factors including exposure to the fungus, particularly in endemic areas. Numerous conditions such as esthesioneuroblastoma, sinusitis, chordoma, tension headache, vascular headache, foreign bodies, brain abscess, epidural abscess, meningitis, and subdural abscess should be considered as part of the differential diagnosis based on imaging studies.[19] Obtaining a comprehensive medical history is crucial for an accurate diagnosis and optimal outcome. While endoscopic sinus surgery is the preferred treatment option, there is limited objective data on overall benefit and patient satisfaction, forcing further investigation.

The patient's prior misdiagnosis several months before the consultation proved to be a significant distractor, leading to a chain reaction of colleagues presuming that esthesioneuroblastoma or a neoplasm was the sole possibility. This presumption could have delayed an earlier intervention and developed an unfavorable outcome. The patient's rural background suggests that fungal soil may be prevalent in that region. Since the complete surgical removal of infected tissue may not be feasible, topical corticosteroid aerosols should be used under antimycotic coverage to prevent recurrence.[7] Neoplastic pathologies can imitate this fungus, given its similarity in imaging studies. Analyzing factors surrounding the individual and maintaining close monitoring of such pathogens are necessary to avoid further complications.

The patient did not experience any sequelae following the surgery. To consider unusual and infrequent alternative diagnoses, such as invasive fungal pathology with an uncommon germ is necessary to achieve a successful medical diagnosis. Therefore, it is necessary to implement diligent monitoring, including the potential for recurrent surgery to extract infected tissues, as well as providing comprehensive postoperative care at the local level. It is critical to always consider a rare pathogen in any sinonasal pathology, particularly when it concerns the anterior skull base. Failure to make a prompt diagnosis may result in a poor outcome, while, when an adequate diagnosis and medical management are performed, good outcomes are usually achieved [Table 1].

Table 1: Reported cases of fungal sinusitis/encephalitis* by *Drechslera hawaiiensis*.

Study	Age (years)	Outcome
Present case	26	Currently under surveillance after surgery and antifungal therapy. Good outcome at the last follow-up
Álvarez[2]	19	Resolution after 4 surgeries and antifungal therapy (7 years follow-up after last intervention, asymptomatic)
Fryen *et al*.[7]	20	Resolution after surgery and antifungal therapy (6 months follow-up, with improvement of initial symptoms)
Fuste *et al*.[8]*	31	Death 11 days after admission for antibiotic therapy
Young *et al*.[25]	15	Resolution after 4 surgeries and antifungal therapy (1 year follow-up, with improvement of initial symptoms)
Morton *et al*.[16]*	18	Resolution after surgery and antifungal therapy
Washburn *et al*.[24]	31	Recurrence after three surgeries and antifungal therapy (17 months follow-up, stable with medical treatment)
Maskin *et al*.[14]	24	Resolution after three surgeries and antifungal therapy (16 months follow-up after last intervention, stable with bilateral amaurosis)
Castelnuovo *et al*.[3]	39	Resolution after surgery and antifungal therapy (2 years follow-up, asymptomatic)
Salcedo *et al*.[18]	10	Resolution after surgery and antifungal therapy (23 months follow-up, with disease-free CT)

CT: Computed tomography, *: Patients with fungal encephalitis

CONCLUSION

Fungal pathogens have the potential to imitate neoplastic processes, and detecting them may require multiple analyses as they may not be easily isolated in a single test or detected through histology. *D. hawaiiensis* is an infrequent pathogen but potentially life-threatening when not identified correctly. Fungus balls can result in secondary mycotic manifestations, which can be fatal if not treated effectively. Management of these infections usually requires local surgical excision and prolonged courses of antifungal agents to prevent recurrence.

Acknowledgment

We, the authors, appreciate the valuable contribution of Dr. Edith Sánchez Paredes from the UNAM School of Medicine Microbiology Department, who was responsible for the microbiological identification of the fungus, and kindly provided the microphotograph depicting this microorganism.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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How to cite this article: Lara-Olivas JA, Sangrador-Deitos MV, Marian-Magaña R, Padilla-Leal KE, Soto-Hernández JL, Gómez-Amador JL. Endoscopic endonasal resection of a *Drechslera hawaiiensis* sphenoid fungal ball. Surg Neurol Int 2023;14:321.