Rare Case of Lasiodiplodia Vitis Invasive Fungal Sinusitis and Role for PCR Testing

Logan F. McColl, MD, MBA[1,2]; Eric Nisenbaum, MD, MSc[1,2]; Kyle VanKoevering, MD[1,2]

[1]Department of Otolaryngology—Head & Neck Surgery, The Ohio State University Wexner Medical Center, Columbus OH, USA
[2]Department of Otolaryngology—Head & Neck Surgery, The James Comprehensive Cancer Center, Columbus, OH, USA

Background

- Invasive fungal sinusitis (IFS) is most commonly due to Aspergillus species and fungi in the Mucorales order.
- These infections are almost exclusively seen in immunocompromised patients.
- Diagnosis is traditionally reliant on tissue staining with Gomori Methenamine Silver (GMS).
- Presented here is an extremely rare case report of IFS due to Lasiodiplodia vitis, requiring universal polymerase chain reaction (PCR) testing for accurate species identification.

Case Description

- 38 y.o. with history of myelofibrosis, 300d post-allogeneic hematopoietic stem cell transplant on tacrolimus and methotrexate.
- Progressive left-sided facial swelling despite multiple courses of antibiotics for presumed sinusitis.
- Found to have severe neutropenia (ANC 0).
- Nasal endoscopy demonstrated left-sided purulence, middle turbinate edema, and intact sensation without lesion(s) or discoloration.
- CT Sinus Without Contrast and MRI Face With/Without Contrast was notable for left greater than right-sided paranasal sinusitis without stigmata of periorbital and/or significant soft tissue abnormalities (Figure 1).
- The patient was taken for endoscopic sinus surgery with left-sided: maxillary antrostomy with endoscopic medial maxillectomy, total ethmoidectomy, and resection of the inferior and middle turbinates.
- Frozen biopsies and bacterial and fungal cultures were all negative.
- Permanent pathology samples with GMS staining demonstrated fungal hyphae in 2 of 6 samples with morphology inconsistent with typical Mucorales or Aspergillus species.
- They were subsequently started on amphotericin B infusions and irrigations.
- The patient underwent an additional four endoscopic sinus surgeries over the following 2.5 months with progressive improvement in tissue edema and eventual GMS clearance.
- No fungal cultures from any of the sinus tissue samples were positive across all surgeries, but GMS was positive in permanent pathologic samples from the first 3 surgeries.
- Universal PCR testing of the patient's tissue samples diagnosed Lasiodiplodia vitis, which has not been reported previously in IFS.[1-3]
- The patient was able to be narrowed to oral voriconazole and voriconazole irrigations with sustained benefit at 6-month follow-up scope examination (Figure 2).
- The patient is currently disease-free and being maintained on oral voriconazole due to persistent severe neutropenia.

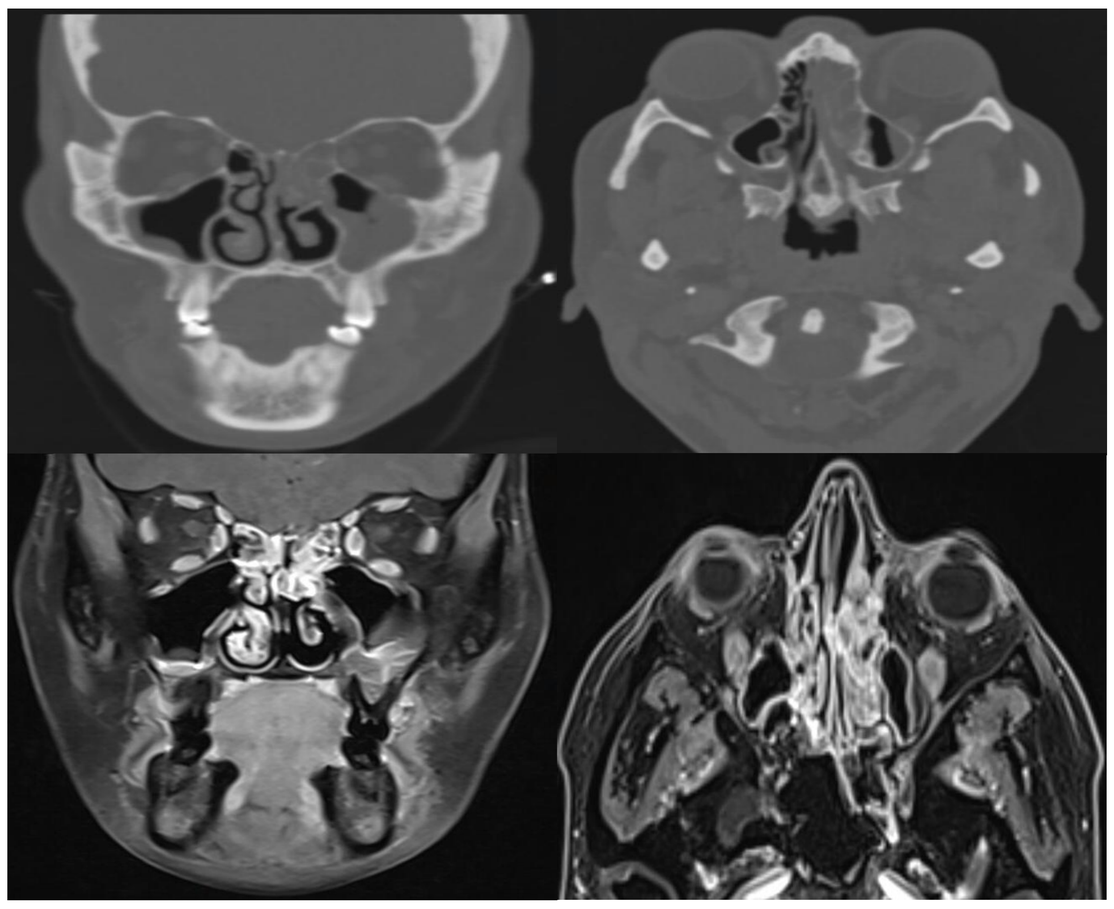

Figure 1. Pre-Operative CT Sinus and MRI Face Scans

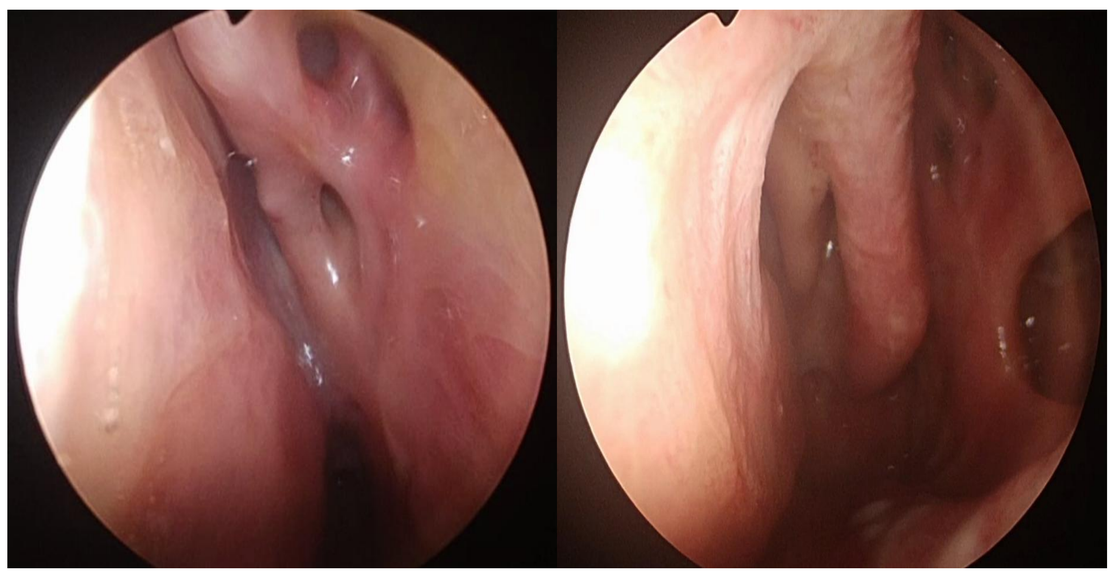

Figure 2. Post-Operative Nasal Endoscopies

Scope still images demonstrating post-surgical changes, including maxillary antrostomies, ethmoidectomies, and posterior septectomy. Resolution of nasal edema, purulence, and crusting was sustained throughout follow-up visits.

Discussion & Conclusion

- This case represents the first documented human infection by Lasiodiplodia vitis, a plant pathogen known for causing causing Botryosphaeria dieback in grapevines.[1-3]
- The use of universal PCR testing in this case allowed for selection of a more narrowed and effective treatment with oral voriconazole instead of intravenous amphotericin B.
- Identification assisted decision-making regarding adjunctive hyperbaric oxygen therapy (HBOT); HBOT was not pursued given Lasiodiplodia vitis would thrive in a high oxygen environment, which is a distinct difference from Aspergillus and Mucor species.
- The successful treatment of Lasiodiplodia vitis with surgical debridement and oral voriconazole adds to the extremely limited literature for the treatment of IFS due to Lasiodiplodia genus and Botryosphaeriaceae family fungi.[4,5]
- This case further highlights the utility of Universal PCR testing in cases of atypical IFS.

References

1. Erami M, Aboutalebian S, Hezaveh SJH, et al. Microbial and clinical epidemiology of invasive fungal rhinosinusitis in hospitalized COVID-19 patients, the divergent causative agents. *Med Mycol*. 2023;61(3):myad020.
2. Chaturantabut S, Kitkumtorn N, Mutirangura A, et al. Identification of pathogens causing invasive fungal rhinosinusitis in surgical biopsies using polymerase chain reaction. *J Laryngol Otol*. 2020;134(7):632-635.
3. Bahethi R, Talmor G, Choudhry H, et al. Chronic invasive fungal rhinosinusitis and granulomatous invasive fungal sinusitis: A systematic review of symptomatology and outcomes. *Am J Otolaryngol*. 2024;45(1):104064.
4. Rodriguez A, Anjan S, Martinez OV, et al. Invasive Rhinosinusitis Caused by Lasiodiplodia theobromae in an Allogeneic Hematopoietic Cell Transplant Recipient Case Report and Review of Literature. Mycopathologia. 2018;183(5):841-845.
5. Maurya AK, Kumari S, Behera G, Bhadade A, Tadepalli K. Rhino sinusitis caused by Lasiodiplodia theobromae in a diabetic patient. Med Mycol Case Rep. 2023;40:22-24. Published 2023 Feb 18.