**Objectives**: Cryptic *Aspergillus* species are increasingly recognized as important pathogens in immunocompromised hosts, including recipients of allogeneic hematopoietic cellular transplantations (HCT) and those with hematologic malignancies. Below, we describe a rare case of invasive sinusitis related to *Aspergillus hiratsukae*, previously known as *Neosartorya hiratsukae*, identified via molecular methods. To the best of our knowledge, this report represents the first documented case of invasive sinusitis associated with this species occurring after HCT.

**Materials & Methods:** A 51-year-old woman with a history of chronic myeloid leukemia (CML) associated with a T315I mutation underwent reduced intensity conditioning followed by a matched-related allogeneic HCT. Graft-versus-host disease (GVHD) prophylaxis consisted of post-transplant cyclophosphamide followed by tacrolimus and mycophenolate. Opportunistic infection prophylaxis originally included acyclovir, posaconazole, atovaquone, letermovir, and entecavir for prior exposure to hepatitis B. Her post-transplant course was complicated by acute GVHD of the skin (grade 1) and GI tract (grade IV) successfully managed with corticosteroids and ruxolitinib. Relapsed CML was managed with hydroxyurea and tyrosine kinase inhibitors (TKI) targeting the abnormal BCR:ABL1 protein.

Four months post-transplant, she developed fever, chills, and progressive headaches. A CT of the sinuses demonstrated an acute infiltrative process suggestive of invasive fungal sinusitis, and MRI identified intracranial and intraorbital extension. The working diagnosis was invasive mucormycosis, and she started treatment with liposomal amphotericin and isavuconazole. Right middle turbinate debridement with tissue biopsy showed fungal elements, and culture yielded *Aspergillus* species. A concomitant serum A*spergillus* galactomannan level was 0.15 EIA index (<0.5, negative), and repeat levels remained negative. The patient became asymptomatic and two months later underwent right sphenoid sinusotomy, right frontoethmoidectomy, and right maxillary antrostomy with tissue removal. Cultures yielded no fungal growth and pathology showed inflammation with no fungal elements. The patient experienced mild transaminitis (grade 1) but otherwise tolerated dual antifungal therapy well as she remained asymptomatic.

**Results**: Three months later, the final identification by internal transcribed spacer (ITS) sequencing of the fungal culture revealed *Aspergillus hiratsukae*, susceptible to most antifungals except fluconazole. Repeat MRI revealed partially improved changes and dural and leptomeningeal enhancement. A repeat MRI two weeks later showed improvement of previous changes, and she was subsequently transitioned to monotherapy with isavuconazole, having completed 16 weeks of liposomal amphotericin. The patient remained clinically well and repeat imaging and sinus endoscopy revealed no evidence of residual fungal infection. Isavuconazole was discontinued after the completion of 10 months of therapy. Surveillance imaging showed no evidence of recurrent fungal infection. The patient died two months later as a result of CML progression.

**Conclusions**: Species level identification of *Aspergillus* infections can inform decisions regarding antifungal management. Routine laboratory methods are insufficient for identifying *Aspergillus hiratsukae*, a rare cryptic species that may only be identified using molecular techniques. This case highlights the importance of considering cryptic *Aspergillus* species as a cause of sinusitis in immunocompromised hosts and underscores the importance of molecular diagnostics in securing a diagnosis to guide management.