

# A Report of Atypical Mycobacterium Sinusitis: Diagnostic Challenges and Treatment Strategies

Travis Weinsheim, DO, PGY-4

Jordan Licata, DO, PGY-2

PCOM Otolaryngology

## Introduction

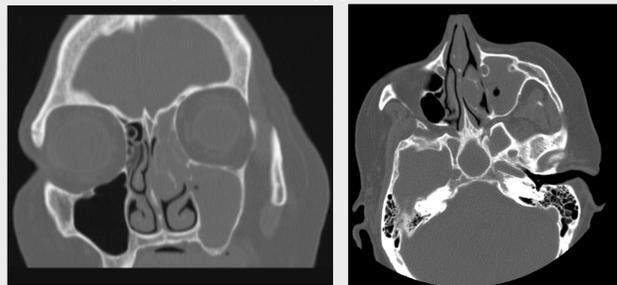
Sinonasal infections by atypical mycobacterium (AM) are not common. First reported in an immunocompetent adult by Spring (1) in 1999, this disease had previously only been seen with AIDS patients. Later studies have reported the incidence of atypical mycobacterium among post-endoscopic sinus cultures as less than 1% (2). Patients with drug-resistant chronic rhino-sinusitis (CRS) are more likely to have AM, suggesting the need for accurate testing in these patients. This was demonstrated by Tichenor (3), who also described the various species of AM that are seen with CRS. Furthermore, there are as yet, no formal guidelines regarding proper treatment of extra-pulmonary AM sinusitis. Here, we present a case report of AM sinusitis and discuss any updates in the literature regarding diagnosis or treatment strategies.

## Case Presentation

### Relevant History

A 65 year old female was seen for evaluation of congestion, epistaxis and facial pain lasting several months. She denied weight loss, hemoptysis or fevers. She had no prior ENT-related surgeries. Her medical history was significant for asthma, hypertension and chronic hepatitis C. She is a former heavy smoker and former IV drug user with associated left arm amputation. She had recently tried OTC nasal steroid sprays and oral antibiotics without relief. Her rhinoscopic exam showed significant crusting, mucopurulent drainage from left middle meatus. She was placed on a combination antibiotic and steroid nasal rinse and radiographic imaging was obtained (CT coronal and axial views seen below)

Figure 1. CT imaging at initial visit



### Course History

- CT imaging at initial visit showed soft tissue in superior left nasal cavity, opacification of left maxillary sinus with a small dehiscence on floor possibly representing introral fistula, obstruction of left osteomaxillary unit and left frontal sinus and outflow tract opacification.
- Lab results including ESR and WBC count were normal.
- Several months later, she underwent FESS, septoplasty and inferior turbinate reduction. Pathology of sinonasal contents revealed chronic inflammation and cultures grew *S. aureus*.
- Patient continued to experience symptoms and was taken back to OR for further debridement and endoscopically directed cultures.
- AFB testing of sinus contents revealed negative staining but culture grew *mycoplasma chelonae*. Blood cultures were also negative for AFB.
- A chest CT was obtained which showed multiple upper lobe infiltrates
- Sputum samples were then collected (in a series of 3 on every other day) and the first sample stained positive for AFB and grew *M. abscessus* on culture. The second sample stained negative for AFB but grew *M. abscessus* on culture. The third sample stained negative for AFB but grew *M. avium complex* on culture.
- Mycobacterium Tuberculosis PCR testing for sinus contents, sputum and blood was negative. A quantiferon test is currently pending.

### Follow-up Course

Patient was referred for evaluation by Pulmonary and Infectious Disease. She is not currently being considered for systemic treatment against AM. She continues to use nasal steroid irrigations and occasional antibiotic rinses. Her symptoms have improved only marginally.

## Discussion

The incidence of extra-pulmonary AM infections has been rising over the years (4). It will become more important to have knowledge surrounding proper testing for these organisms. As seen in our patient, it wasn't until after a second sinus procedure that the diagnosis was made, almost a year after initial presentation. Furthermore, we also demonstrated the variability between culture results. A total of three separate atypical mycobacterium species grew within our patient, *M. chelonae*, *M. abscessus* and *M. avium complex*. A recent study (3) sampled a population of AM sinusitis patients and found ten different species of AM. They also reported the presence of AM in drinking water, which may play role when counseling patients on proper saline nasal irrigation technique. Another interesting finding was the recent discovery of a new organism, *M. abscessus-chelonae* complex or *M. franklinii*, which was found among CRS patients in northeast US (5). This may suggest that there is more interplay and associations between these species than we know. At the least, considerations should be made for the role of obtaining multiple cultures over time in these patients. Solyar (6) demonstrated no significant difference in culture diagnoses of AM sinusitis made in the clinic vs OR; thus, lending credence to office-based procedures.

At this time, it is also not clearly understood whether there are diagnostic or prognostic differences between the various AM species that reside in the sinonasal or respiratory tract. It seems more likely that the various species that make up AM have a large amount of overlap but further research is needed.

Our patient's clinical presentation was similar to other reports in the literature however she had radiographic evidence of bony erosion within the sinus cavity. This has not been clearly stated in the literature but Hicks (7) presented a patient with AM sinusitis similar to our own with CT imaging showing bony erosion in the sinuses. Further evidence is needed to confirm these findings as related.

Treatment of AM sinusitis remains unclear although most would advocate for surgical excision first and foremost. Long-term antibiotic therapy against AM has also been described without a clear consensus on type, duration or route although macrolide treatment up to 6 months or longer has been seen (8).

## Conclusions

Sinonasal infections associated with AM are not very common although their incidence has been increasing recently. The lack of formal guidelines regarding diagnosis and treatment can only lead to worse outcomes in patient care. Recognizing this rare entity is the first step and influenced our decision to present this case report. As demonstrated in the literature and from our report, there is a need to establish a diagnostic criterion for AM sinusitis which would take into account obtaining consecutive cultures in addition to clinical signs and radiographic findings. Lastly, the paucity of studies aimed at specifically treating AM sinusitis leaves much room for future studies.

There are several limitations in this study; 1) as a case report we are inherently limited by sample size, 2) our patient was unable to follow-up in the months following her second debridement due to closures related to Covid-19, 3) we were unable to obtain endoscopic photos.

This case report will hopefully increase the knowledge base to drive further research into this rare disease process.

## References

1. Spring PM, Miller RH. Initial report of primary sinusitis caused by an atypical pathogen (Mycobacterium chelonae) in an immunocompetent adult. *Ear Nose Throat J*. 1999;78(5):359-364.
2. Suh JD, Ramakrishnan VR, Tajudeen B, Reger C, Kennedy DW, Chiu AG. Identification and treatment of nontuberculous Mycobacterium sinusitis. *Am J Rhinol Allergy*. 2011;25(6):421-424. doi:10.2500/ajra.2011.25.36773. Journal Article, *Name of Journal*
3. Tichenor WS, Thurlow J, McNulty S, Brown-Elliott BA, Wallace RJ Jr, Falkinham JO 3rd. Nontuberculous Mycobacteria in household plumbing as possible cause of chronic rhinosinusitis. *Emerg Infect Dis*. 2012;18(10):1612-1617. doi:10.3201/eid1810.1201644
4. Piersimoni, C., & Scarparo, C. (2009). Extrapulmonary infections associated with nontuberculous mycobacteria in immunocompetent persons. *Emerging infectious diseases*, 15(9), 1351-1544. <https://doi.org/10.3201/eid1509.0812595>. Journal Article, *Name of Journal*
5. Simmon, K. E., Brown-Elliott, B. A., Ridge, P. G., Durtschi, J. D., Mann, L. B., Slechte, E. S., Steigerwalt, A. G., Moser, B. D., Whitney, A. M., Brown, J. M., Voelkerding, K. V., McGowan, K. L., Reilly, A. F., Kim, T. J., Butler, W. R., Edelstein, P. H., Wallace, R. J., Jr, & Petti, C. A. (2011). Mycobacterium chelonae-abscessus complex associated with sinopulmonary disease, Northeastern USA. *Emerging infectious diseases*, 17(9), 1692-1700. <https://doi.org/10.3201/eid1709.101667>
6. Solyar A, Lee AS, Przybyszewski B, Lanza DC. Atypical mycobacterium detection in refractory chronic rhinosinusitis. *Otolaryngol Head Neck Surg*. 2012;146(6):1012-1016. doi:10.1177/0194599812437333
7. Hicks MD, Karempelis PS, Janus SC. Mycobacterium Chelonae Sinusitis in an Immunocompetent Adult. *JAMA Otolaryngol Head Neck Surg*. 2016;142(8):805-806. doi:10.1001/jamaoto.2016.1865
8. Mullin D, Jothi S, Healy D. Mycobacterium chelonae infections involving the head and neck. *Ann Otol Rhinol Laryngol*. 2009;118(10):714-720.