Adalimumab: Aiding or Alarming in a Pediatric Chronic Recurrent Multifocal Osteomyelitis Patient with Sinusitis?

Alison M. Hawkins, BA[1]; Michael T. Yim, MD, FACS[1,2]; Sarwat Umer, MD[1,3], Torrey Fourrier, MD[1,2]

[1]LSU Health Shreveport School of Medicine, [2]Louisiana State University Health Shreveport Department of Otolaryngology, [3]Louisiana State University Health Shreveport Department of Rheumatology

Abstract

Intro: Chronic recurrent multifocal osteomyelitis (CRMO) is an autoimmune disorder characterized by non-infectious osteolytic lesions and bone inflammation that predominantly presents in long bones of children**.** Rare cases of sinus osteomyelitis have been reported in patients taking adalimumab for CRMO. This case report and literature review describes a case of acute ethmoid sinusitis and osteomyelitis with intracranial extension in a pediatric CRMO patient with recent history of adalimumab use.

Methods: This is a case of sinonasal osteomyelitis presenting with headache and nasal bridge/lacrimal duct abscess observed within a 4-month period of a 13-year-old female discontinuing adalimumab treatment for CRMO. Interventions included functional endoscopic sinus surgery (FESS) and long-term antibiotic therapy as operative cultures grew rare coagulase negative staphylococcus species and pathology results demonstrated viable bone. Outcomes were recorded based on resolution of infection at the time of discharge and clinical improvement monitored with serial examinations and radiologic imaging. A literature review was also performed which found a report of similar cases of CRMO treated with adalimumab in two 11-year-old female patients, both resulting in adalimumab-related sinonasal complications.

Results: The clinical improvement after treatment with FESS, antibiotics, as well as continued avoidance of adalimumab post-surgery, leads us to believe that our patient suffered from immunocompromised sinusitis related to adalimumab use, leading to an advanced presentation of our patient's symptoms. These results are consistent with that of the cases noted in our literature review, as they similarly resolved with sinus surgery and antibiotics. Since cranial CRMO involvement is extremely rare, it is a strong possibility that adalimumab was a culprit in our patient's case presentation.

Conclusion: Adalimumab therapy in the setting of CRMO can be a precipitating factor in the development of neurocranial osteomyelitis and its related complications, and otolaryngologists should be aware of this possibility.

Background

Adalimumab is an anti-tumor necrosis factor (anti-TNF) humanized monoclonal antibody biologic agent that is most-commonly prescribed for rheumatoid arthritis in adults. However, in pediatric populations, adalimumab can be prescribed in conjunction with non-steroidal anti-inflammatory agents (NSAIDs) to treat chronic recurrent multifocal osteomyelitis (CRMO), also known as chronic nonbacterial osteomyelitis (CNO). CRMO is a non-infectious, autoimmune disease that causes inflammation of the bone with a relapsing and remitting presentation [1]. It is typically seen in pediatric females around the age of ten [1]. As CRMO most commonly effects the long bones such as the clavicle or femur, CRMO involvement of the skull is extremely rare [2, 3]. While many patients with CRMO experience adequate relief with an NSAID-only regimen, some require the additional anti-inflammatory benefits of biologics like adalimumab. However, a small number of cases have been recorded relating adalimumab use to the development of acute bacterial sinusitis and intracranial abscesses [4].

Case Presentation & Investigations

An early teenage female who had previously been receiving adalimumab injections for the past 21 months to treat chronic recurrent multifocal osteomyelitis (CRMO) began to experience weight loss, rash, and headaches which persisted for 4 months after discontinuing the medication. After the headaches developed, an MRI brain with and without contrast showed bilateral thickening of the ethmoid sinuses. Two weeks following this MRI, the patient presented as a walk-in at the ophthalmology clinic.

Ophthalmology Clinic

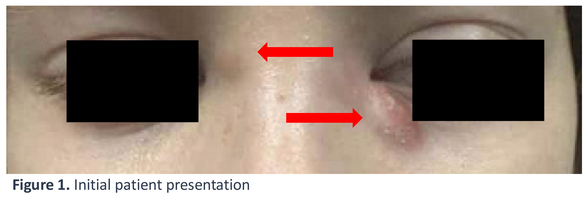
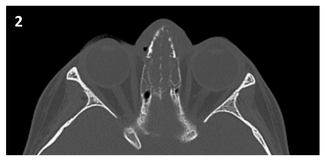


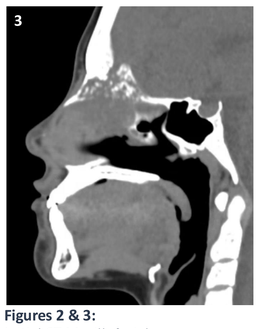
Figure 1. Initial patient presentation

Patient presented with **red, swollen bump** with purulent discharge developing along the left lower eyelid and the right nasal ridge **(Figure 1)**

Emergency Department

CT showing erosive changes in the ethmoid sinuses alongside associated amorphous sinonasal soft tissue with intracranial extension to the cribriform plate





Figures 2 & 3: Initial CT Maxillofacial

ENT Involvement

Started on vancomycin, ceftriaxone, and metronidazole

MRI brain/orbits with turbo inversion recovery magnitude (TIRM) sequencing findings consistent with **ethmoid sinusitis with intracranial extension and osteomyelitis** of the ethmoid sinus, cribriform plate, and frontal bones

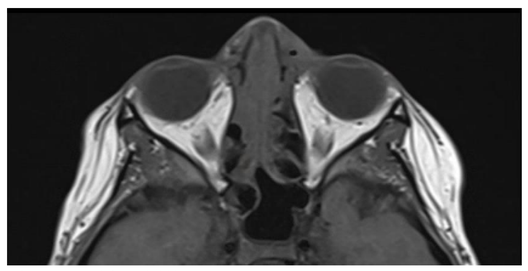


Figure 4: MRI Orbits 10/2024

Underwent **bilateral anterior functional endoscopic sinus surgery (FESS**) in which mild mucopurulence of the nasopharynx and significant obstructive polypoid edema were found throughout the middle meatus, septum, middle turbinates, and bilateral ethmoids

Bilateral partial resections of the middle turbinate, uncinectomy, maxillary antrostomy, and anterior ethmoidectomy.

Discharge Home

Post-Operation

DISCHARGE:
- Continued to improve clinically
- Discharged with a peripherally inserted central catheter (PICC) line → 4 weeks of IV ceftriaxone and PO metronidazole q8 hours

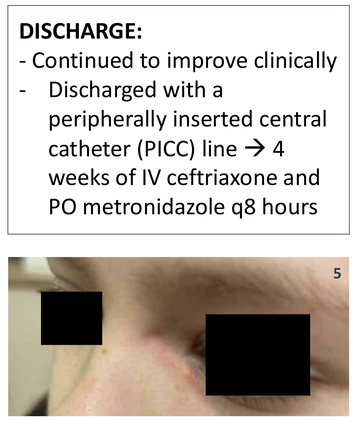
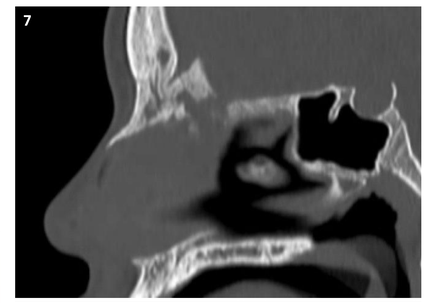


Figure 5 & 6: 1-month and 8-month follow-up images



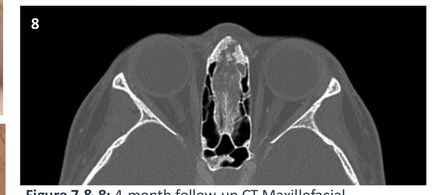


Figure 7 & 8: 4-month follow-up CT Maxillofacial

FOLLOW UP:
- No longer taking adalimumab
- No repeat events

Discussion

One 2018 publication from Odense University Hospital in Denmark describes two 11-year-old female patients taking adalimumab for CRMO that both developed acute sinusitis and severe intracranial complications [4]. Both females in this report underwent functional endoscopic sinus surgery (FESS) with antibiotic treatment and discontinuation of adalimumab. Both patients experienced regression of abscesses and did not experience recurrence in the follow-up period while remaining off adalimumab [4].

These cases greatly resemble that of our 13-year-old female patient, emphasizing the importance of exploring adalimumab and its potential affiliation to intracranial complications in children with CRMO. The clinical improvement of our patient after treatment with FESS, antibiotics, as well as continued avoidance of adalimumab post-surgery, leads us to believe that our patient suffered from immunocompromised sinusitis related to adalimumab use, leading to an advanced presentation of our patient's symptoms. These results are consistent with that of the cases noted in our literature review, as they similarly resolved with sinus surgery and antibiotics. Since cranial CRMO involvement is extremely rare, it is a strong possibility that adalimumab was a culprit in our patient's case presentation.

Conclusions

- Adalimumab use in pediatrics could be a potential cause of acute sinusitis and intracranial complications
- FESS, antibiotic treatment, and adalimumab discontinuation offer a promising treatment plan for patients with acute osteolytic sinusitis related to recent adalimumab use
- Physicians should take caution and consider possible sinugenic complications when prescribing adalimumab in pediatric populations with CRMO

Contact

Alison Hawkins, MS4
LSU Health Shreveport SOM
1501 Kings Hwy, Shreveport, LA 71103
amh004@lsuhs.edu

References

1. Chronic recurrent multifocal osteomyelitis (CRMO). American College of Rheumatology. February 2023. Accessed September 27, 2025. https://rheumatology.org/patients/chronic-recurrent-multifocal-osteomyelitis-crmo
2. Zhao DY, McCann L, Hahn G, Hedrich CM. Chronic nonbacterial osteomyelitis (CNO) and chronic recurrent multifocal osteomyelitis (CRMO). *Journal of Translational Autoimmunity*. 2021;4:100095. doi:10.1016/j.jtauto.2021.100095
3. Watanabe T, Ono H, Morimoto Y, et al. Skull involvement in a pediatric case of chronic recurrent multifocal osteomyelitis. *Nagoya J Med Sci*. 2015;77(3):493-500
4. Kofoed MS, Fisker N, Christensen AE, Kjeldsen AD. Sinogenic intracranial complications: is adalimumab a culprit?. *BMJ Case Rep*. 2018;2018:bcr2017221449. Published 2018 Jan 17. doi:10.1136/bcr-2017-221449