VIRGINIA DERMATOLOGY SOCIETY

2021 Annual Spring Virtual Meeting | Abstract Submission

Relapsing polychondritis in pregnancy managed with azathioprine

Sarah Simpson, MD; Jane Gay, BA; Patrick Rush, DO; Alex Holliday, MD; Philip Wakefield, MD
Carilion Clinic and Virginia Tech Carilion School of Medicine

A 42-year-old female, eight weeks pregnant following in vitro fertilization, presented with a two-month history of tender, swollen, and red ears. Her symptoms abated after a one-week prednisone course, but they rapidly recurred. An extensive review of systems was negative. Examination demonstrated erythema, edema, and tenderness of her bilateral auricles with lobe sparing (Figure 1). Nasal cartilage and trachea appeared normal. Biopsy revealed fragmented hyalin cartilage with neutrophilic infiltrate, dermal hemorrhage, and deep dermal fibrosis, consistent with relapsing polychondritis (RP). The presence of anti-collagen type II antibodies supported the diagnosis. Prednisone was restarted but could not be tapered below 20 milligrams daily. At 23 weeks' gestation, rheumatology initiated azathioprine allowing discontinuation of prednisone in 8 weeks. She achieved complete clinical remission on azathioprine monotherapy (Figure 2) and delivered a healthy full-term infant.

Relapsing polychondritis is a rare immune-mediated multisystem inflammatory disease affecting cartilaginous structures. Associated autoimmune diseases occur in 30 percent of adult cases.[1] RP in pregnancy is rare. Case reports and series suggest pregnancy outcomes are often favorable,[2,3] although there is one reported case of neonatal RP.[4] Treatments include NSAIDs and glucocorticoids for limited disease.[5] Resistant cases may require colchicine, dapsone, methotrexate, azathioprine, or TNF alpha inhibitors.[1,5] During pregnancy, low-dose steroids are safe when necessary,[6] but there is scant literature describing ideal management options for resistant disease in pregnancy. Azathioprine has been safely used to manage lupus during pregnancy.[7] Our case supports azathioprine may be safe and effective for resistant RP in pregnancy.

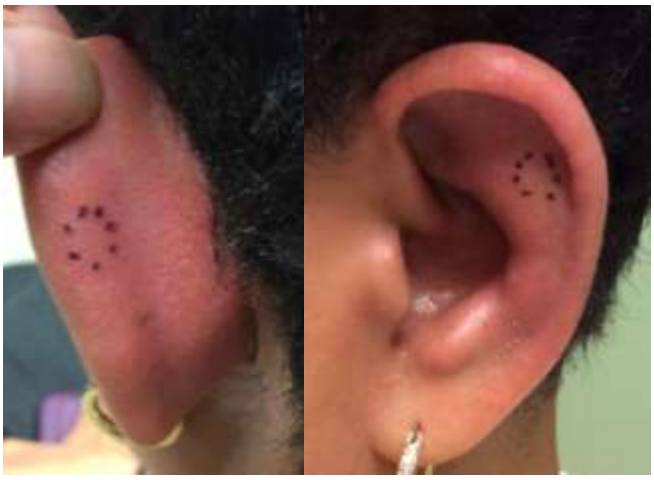

Figure 1: Photograph of left ear and planned biopsy site

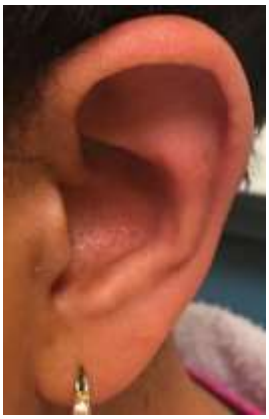

Figure 2: Photograph of left ear taken on azathioprine monotherapy.

References:

1. Fonseca AR, de Oliveira SK, Rodrigues MC, Aymoré IL, Domingues RC, Sztajnbok FR. Relapsing polychondritis in childhood: three case reports, comparison with adulthood disease and literature review. *Rheumatology International*. 2012;33(7):1873-1878. doi:10.1007/s00296-011-2336-6
2. Krakow D, Greenspoon JS, Firestein GS. Relapsing Polychondritis and Pregnancy: Normal Fetal Outcome Despite Maternal Flares. *JCR: Journal of Clinical Rheumatology*. 1996;2(2):118-119. doi:10.1097/00124743-199604000-00013
3. Papo T, Wechsler B, Bletry O, Piette A-M, Godeau P, Piette J-C. Pregnancy in relapsing polychondritis. Twenty-five pregnancies in eleven patients. *Arthritis & Rheumatism*. 1997;40(7):1245-1249. doi:10.1002/art.1780400708
4. Arundell FW, Haserick. JR. Familial chronic atrophic polychondritis. *Archives of Dermatology*. 1960;82:439-440.
5. Rednic S, Damian L, Talarico R, et al. Relapsing polychondritis: state of the art on clinical practice guidelines. *RMD Open*. 2018;4(Suppl 1):e000788. Published 2018 Oct 18. doi:10.1136/rmdopen-2018-000788
6. Bandoli G, Palmsten K, Forbess Smith CJ, Chambers CD. A Review of Systemic Corticosteroid Use in Pregnancy and the Risk of Select Pregnancy and Birth Outcomes. *Rheumatic Disease Clinics of North America*. 2017;43(3):489-502. doi:10.1016/j.rdc.2017.04.013
7. Fava A, Petri M. Systemic lupus erythematosus: Diagnosis and clinical management. *Journal of Autoimmunity*. 2019;96:1-13. doi:10.1016/j.jaut.2018.11.001