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Relapsing Polychondritis in a Patient with Longstanding Ankylosing Spondylitis

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Introduction

Relapsing polychondritis (RPC) is a rare autoimmune condition of unknown etiology that is characterized by recurrent episodic inflammation that leads to destruction of cartilaginous structures. RPC has been associated with autoimmune disorders such as rheumatoid arthritis, systemic lupus erythematosus, and Sjogren's syndrome, but rarely ankylosing spondylitis. We describe a young male with ankylosing spondylitis who developed RPC after ear trauma.

Case Description

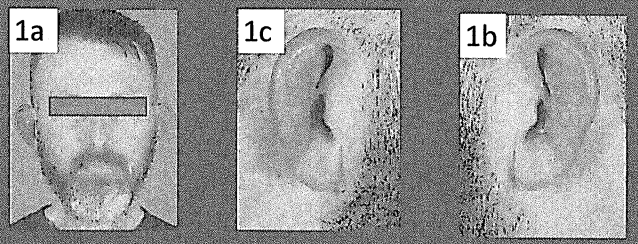
- This is a case of a 49 year old male with ankylosing spondylitis (AS) diagnosed at age 22, manifested by periodic right knee swelling, back pain, and ankylosis of bilateral sacroiliac joints. He was in remission for the last 11 years on etanercept, indomethacin, and sulfasalazine except for episcleritis in May 2017 that resolved after increasing indomethacin.
- In August 2017, had a vasovagal episode resulting in right shoulder fracture and right ear trauma. On follow up with otorhinolaryngology for hearing loss and right ear trauma he was noted to have bilateral auricular swelling consistent with relapsing polychondritis.
- He was treated with oral corticosteroids. Pulmonary function tests were normal. Given no evidence of life or organ threatening disease, etanercept was changed to adalimumab as it is more effective in episcleritis and sulfasalazine was replaced with dapsone.
- Upon taper of prednisone to below 20mg, auricular swelling relapsed, therefore methotrexate was started enabling a taper below 20mg.

Clinical Data

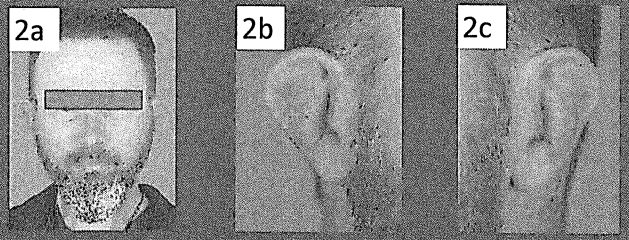
Test	Result	Normal Values
ESR	66	4-27 mm/hr
C-Reactive Protein	3.10 mg/dL	<0.03 mg/dL
G6PD activity	18.1 U/ g Hgb	7-20.5 U/g Hgb
WBC	10.6	3.4-9.8 x 10^3
Hemoglobin	10.7	11-16 g/dL
Platelets	495	124-362x 10^3
Creatinine	0.62	0.6-1.0 mg/dL
Pulmonary function tests	normal	normal
Audiometry	Sensorineural hearing loss	normal

Clinical Images

Pre-treatment



Post-treatment



Axial Imaging

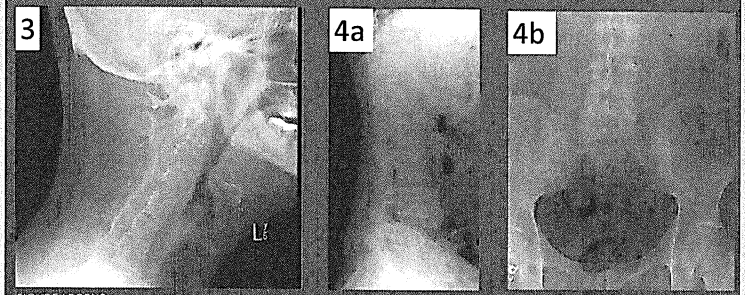


FIGURE LEGEND:

Figure 3: C-Spine 2006: straightening of the cervical spine, squaring of the vertebral bodies, and syndesmophytes.

Figure 4a: L and S spine 2006: ankylosis of bilateral SI joints.

Figure 4b: squaring of lumbar vertebral bodies.

Classification Criteria

1986 Diagnostic Criteria for Relapsing Polychondritis

Major Criteria	Minor Criteria
Proven inflammatory episode of ear cartilage	**Eye inflammation**
Proven inflammatory episodes of nose cartilage	Hearing Loss
Proven inflammatory episodes of layrngotracheal cartilage	Vestibular dysfunction

****Diagnosis is made by two major or one major plus two minor criteria**

Discussion

- Relapsing polychondritis is a multi-organ disease that presents with episodic inflammation of cartilaginous structures. Although its etiology is unknown, approximately 50 % of cases report antecedent cartilage injury.
- The pathogenesis is presumed to be autoimmune in nature and is associated with class II major histocompatibility (MHC) markers. In our case, the patient had AS, a Class I MHC associated disease, that had been significantly immunosuppressed for over the 11 years prior to development of RPC.
- The underlying cause of RPC most likely involves innate immunity. That such a condition may complicate both Class I and II MHC associated disorders of acquired immunity, implies a complex interplay between the innate and acquired arms of the immune system.

Conclusion

- This case differs from prior case reports of ankylosing spondylitis in association with relapsing polychondritis where the relapsing polychondritis occurred soon after initiation of anti TNF therapy with resolution upon its discontinuation.
- In our case the disease developed after 11 years of immunosuppressive therapy, suggesting that it is not drug induced, but likely due to different pathogenetic pathways.

References

[redacted]

I have no financial disclosures. * The view(s) expressed herein are those of the author(s) and do not reflect the official policy or position of Brooke Army Medical Center, the U.S. Army Medical Department, the U.S. Army Office of the Surgeon General, the Department of the Air Force, the Department of the Army or the Department of Defense or the U.S. Government.