

Case Report

Relapsing polychondritis: a case report

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ABSTRACT

Relapsing polychondritis (RP) is an immune-mediated systemic disease characterized by recurrent attacks of inflammation of cartilage and proteoglycan-rich tissues, resulting in progressive anatomical deformation and functional impairment in the affected areas. It may for a long time cause unspecific signs, which may cause a significant delay in establishing the diagnosis and initiating the right treatment. We present the case of a 49 year old male who came to our clinic with a two month history of worsening pain, swelling, and erythema of his left ear auricle. He reported tenderness and warmth over the pinna, along with fever and malaise. The patient was diagnosed with RP and started steroids which are the first choice of therapy. The aim of this case is to emphasize the critical need for timely identification of RP in order to start the appropriate treatment and prevent future complications.

Keywords: Autoimmune, Auricle, Corticosteroids, Relapsing polychondritis

INTRODUCTION

Relapsing polychondritis (RP), is a rare autoimmune disease characterized by inflammation and destruction of cartilage. The most affected areas are the ear cartilage (pinna), the nose cartilage and the respiratory tract. It can also affect the eyes, heart, and blood vessels, but these are very rare findings.¹ In an autoimmune disease, antibodies or cells produced by the body attack the body's own tissues (in this case cartilaginous tissue), leading to inflammation and damage. The etiology of RP is not fully known. An autoimmune reaction against type II collagen (which is mostly found in cartilage and sclera) is thought to be the main cause.² The annual incidence has been reported as 3.5 cases per million and though it can occur at any age, the most common age is 40 to 50 years. This pathology shows equal rates in both genders.³ Symptoms can include ear pain, swelling, redness, hearing loss, and tinnitus. Some patients present nasal congestion and epistaxis or even ocular symptoms like conjunctivitis or scleritis. Clinical manifestations vary from auricular chondritis, nose deformity to life-threatening systemic features. Red swollen pinna is found

in 90% of RP cases, while other clinical manifestations are not as typical. RP poses a diagnostic challenge due to the fact that it's a very rare condition, has atypical clinical manifestations, and there aren't precise diagnostic criteria.^{4,5}

CASE REPORT

A 49-year-old male presented to our clinic with a two month history of worsening pain, swelling, and erythema of his left ear auricle. The patient reported tenderness and warmth over the pinna, along with fever and malaise.

On examination, the left auricle appeared erythematous, edematous, and tender to palpation. There was no involvement of the ear lobe. Otoscopic examination revealed intact tympanic membrane, with no evidence of middle ear pathology. The patient has used antibiotics before being admitted to the hospital, but with no improvement.

His blood tests during his initial presentation showed a normal PCR, elevated WBC (14.6 K/ul), elevated

Lymphocytes (5.8 K/ul), elevated monocytes (1.4 K/ul). CT-scan didn't gave extra information, except what was already assessed. He reported that during his treatment with Levofloxacin, he experienced a rash all over the body, and a burning sensation mostly on his knees, so the treatment was changed to ceftriaxone and amikacin. After the change in treatment the patient didn't experience rash anymore, but during the night the burning sensation on his knees persisted. The patient had not responded to antibiotic treatment during his hospitalisation. He had been evaluated by the rheumatologist and during the consultation he presented pain and increased sensitivity when palpated the laryngeal and tracheal cartilages. Other test like antinuclear antibodies, antiphospholipid antibodies, antineutrophil cytoplasmic antibodies (ANCA), complement C3 and C4 levels, rheumatoid factor were taken and they were all negative. No ocular disorders were determined in the ophthalmological consultations.

The patient was diagnosed with RP and treatment was started of 40 mg/day methylprednisolone (MP) and diclofenac 75 mg/day for pain relief. Therapy was applied for 5 days. The patient complaints improved and he was discharged home on a methylprednisolone taper for 20 days (the first 10 days 16 mg/day, and the next 10 days 12 mg/day) and also naproxen 550 mg/day for pain relief. From the moment that the patient was diagnosed and treated for RP, his symptoms improved and he has remained stable.



Figure 1: Left auricle appeared erythematous, edematous, and tender to palpation.

DISCUSSION

Although RP is rare, the disease's multiple clinical presentations and recurrent episodic nature can cause significant diagnostic delays and are often overlooked by physicians. Auricular chondritis is the most common presentation of RP, that leads to acute, erythematous,

tender inflammation of the external ear with sparing of the lobule.^{5,6}

Diagnosis is primarily made clinically using one of three accepted clinical criteria established by McAdam et al and revised by Damiani et al and Michet et al.⁷⁻⁹

McAdam et al created the first set of diagnostic criteria.⁷ In order to establish the diagnosis, at least three symptoms such as bilateral auricular chondritis, nasal chondritis, nonerosive seronegative inflammatory polyarthritis, ocular inflammation, tracheal or laryngeal cartilages' inflammation, and audio-vestibular dysfunction had to be present.

Damiani et al introduced the modified McAdam's criteria, which included histological changes and response to treatment.^{7,8,10} The diagnosis could be made three-way: either based on McAdam's criteria with no histological tests necessary; in the presence of at least one McAdam's sign accompanied by typical histologic changes; or in the presence of chondritis in at least two different sites and a good treatment response.

Michet et al proposed the most recent set of criteria, once more based solely on symptoms.^{9,10} In order to meet the criteria, a patient has to present either with cartilage inflammation in at least two locations, or with a singular-site chondritis accompanied by at least two of the following symptoms: seronegative arthritis, ocular inflammation, hearing loss, and/or vestibular dysfunction.

Our patient fulfilled the criteria proposed by Damiani et al for RP (auricular chondritis, tracheal or laryngeal cartilages' inflammation) with good response to corticosteroids.⁶ Since our patient improved with corticosteroids, an invasive biopsy, with associated morbidity, was ultimately not required.

CONCLUSION

RP is an uncommon, but progressive autoimmune disease which can lead to fatal complications. As RP is a clinical diagnosis, increased awareness of the disease presentation and clinical characteristics may increase disease recognition and improve treatment outcomes.

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