Concurrence of Psoriasis and Relapsing Polychondritis: A Case Report

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ABSTRACT:

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Relapsing polychondritis is a rare, autoimmune disorder affecting multiple organs primarily the cartilaginous tissues. Several diseases have been reported to be associated with relapsing polychondritis including autoimmune diseases, vascular diseases, hematologic diseases, and skin diseases.

We herein report a case of a 64-year-old Thai female patient with a ten-year history of chronic plaque psoriasis treated with topical corticosteroid and a one-year history of psoriatic arthritis treated with methotrexate and sulfasalazine, who developed bilateral painful, and erythematous swollen ears for 2 months. The clinical and histopathological findings confirmed the diagnosis of relapsing polychondritis. The patient was successfully treated with azathioprine.

Key words: Relapsing polychondritis, psoriasis, psoriatic arthritis

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Introduction

Relapsing polychondritis (RP) is an uncommon autoimmune connective tissue disease affecting cartilaginous and proteoglycan-rich tissues. Auricular and nasal cartilages are the most commonly affected, but it can also damage respiratory cartilage, joints, eyes, kidneys, and the cardiovascular and nervous systems[2].

Psoriasis is a common skin disease with well-defined erythematous scaly plaques as a characteristic feature. Patients with psoriasis also have increased risk of developing metabolic syndrome, arthralgia, and destructive arthritis, which can significantly affect their quality of life[3].

Case report

A 64-year-old woman with diagnosed psoriasis vulgaris for ten years later developed psoriatic arthritis (PsA) on her hands and feet for one year. She had been prescribed topical corticosteroid for her chronic plaque psoriasis, methotrexate 10 mg weekly and sulfasalazine 2,000 mg daily for her PsA. Her psoriasis was well-controlled with PASI score around 2 to 3. She also had hypertension and chronic kidney disease in which she took manidipine, enalapril, and atenolol to control her blood pressure.

The patient revisited the dermatology clinic with new complaint of bilateral painful ears for two months. Physical examination revealed mild erythema, swelling, and tenderness on both of her ear pinnae, limited to cartilaginous parts, sparing the earlobes. (Figure 1) She denied history of any previous ear trauma.

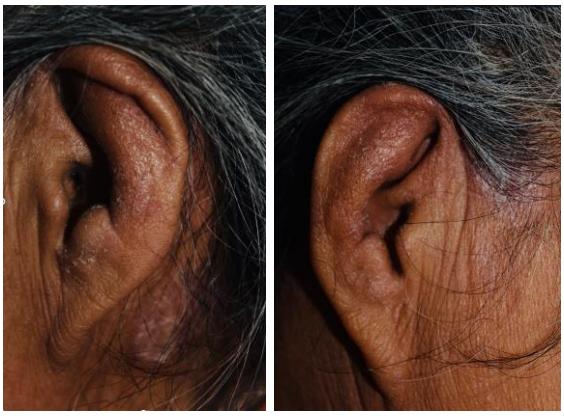

Figure 1 Mild erythema, swelling of both ears, confined to cartilaginous parts of the ears, sparing the earlobes

The biopsy was performed from the erythematous area of her left ear including the skin and the underlying cartilage. The histopathology showed normal epidermis and dermis, lymphocytic infiltration of cartilage with foci of chondrocyte destruction, loss of chondrocyte architecture, and presence of eosinophilic hue (Figure 2), compatible with chondritis. Thus, she received a diagnosis of RP using the Damiani and Levine criteria, including one clinical criterion (bilateral auricular chondritis) and the biopsy confirmation of inflamed cartilage.

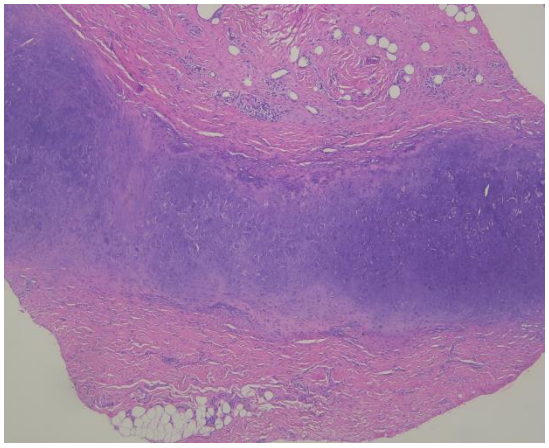

Figure 2 Histopathology shows lymphocytic infiltration of the cartilage with foci of chondrocyte destruction, loss of lacunar architecture, and the presence of eosinophilic hue (H&E, objective X10)

Discussion

RP was first reported in 1923 by Jack Wartenhorst as "polychondropathia"[4], but its name was later changed to RP due to its episodic course of the disease[5]. The etiology is not fully understood, but the circulating antibodies against collagen type II, IX, XI, matrillin-1 and cartilage oligometric matrix protein (COMP) are believed to be involved[1].

The most common clinical manifestations of RP are auricular chondritis, followed by nasal chondritis. Patients typically present redness, swelling, and tenderness on one or both ears confined to the cartilaginous parts. The clinical course is chronic-relapsing and recurrent. Repeated and long-standing inflammation subsequently destroy cartilage tissues. As a result, the ears may become thickened due to fibrosis. Less common findings include nasal chondritis, costochondritis, arthritis, and ocular inflammation. Some manifestations are uncommon but can be fatal including chondritis of the airways, cardiac valve pathology, aortic aneurysm and renal failure[2]. Cutaneous manifestations are non-specific, and the most common findings are erythema nodosum, purpura, and aphthosis[2].

The histopathology of the early stage reveals breakdown of the normal lacunar structure of the cartilage, neutrophillic infiltrates, followed by lymphoplasmacytic infiltrates. In the late stage, the cartilage will be replaced by granulation tissue and fibrosis[6]. The biopsy specimen of our patient was compatible with early stage of RP.

Presently, no standard treatment has been established for RP. Various treatments have been described only in case reports. The aim of treatment is to control inflammation and to prevent multi-organ damage. In mild cases, non-steroidal anti-inflammatory drugs (NSAIDs), dapsone and colchicine are treatments of choice. However, in severe cases, systemic corticosteroids and immunosuppressive agents should be considered[2]. The pathogenesis of RP is not well understood but the role of several cytokines such as tumor necrosis factor (TNF)-α,

interleukin-1 and interleukin-6 have been described[7]. Case reports indicated RP responded well to anti-TNFα such as adalimumab, infliximab and etanercept[8,9]. The use of tocilizumab (anti-IL-6) and anakinra (anti-IL-1) also showed promising results but the numbers of cases are small[10].

RP has been found to be associated with several diseases including autoimmune, hematologic and skin diseases[1]. The coexistence of psoriasis and RP are rare. Only ten cases of RP coexisting with psoriasis have been described[8,11]. Most cases of RP with psoriasis had milder clinical RP than usual RP[8]. The patients in the previous case reports received different medications including prednisolone 30 mg/day (3 cases), NSAIDs (2 cases), eternercept (2 cases), adalimumab (1 case), infliximab (1 case), and one case showed spontaneous remission[8]. Only three case reports of patients presenting RP with psoriasis and PsA were found in the literature[8]. Those three patients had psoriasis with PsA eleven years, nine years and one year, respectively before developing RP[8]. However, no causal relationship between these two diseases has been discovered[8]. Some hypotheses suggested that RP may be related to PsA[9] because serum (COMP) levels are elevated in both PsA and RP[8].

Our patient was further evaluated for other organ involvement. Her ear examination and audiogram were normal. Her eye examination revealed no signs of ocular inflammation. Her echocardiogram was unremarkable without any valvular pathology. Considering the potential association between RP and autoimmune diseases, we performed a blood test for autoimmune panel. The results showed positive ANA (1:64) fine speckled, low positive rheumatoid factor at 10.40 (0-14), whereas anti-dsDNA, ANCA, anti-MPO, and anti-PR3 were all negative.

Our patient developed RP, despite taking 10 mg/week of methotrexate and 1000 mg/day of sulfasalazine. Therefore, the rheumatologist decided to add azathioprine 50 mg/day as another immunosuppressive drug. The patient responded well and the lesions on her ears subsided five months after starting azathioprine. She continues taking this dosage of all three immunosuppressive medications until now, which has been 15 months. She does not experience any side effects of the treatment.

Our long-term management plan for this patient is to taper down the immunosuppressive agents to the lowest effective maintenance dose. In case the diseases relapse or the patient develops complications from immunosuppressives, we may consider using TNF-α antagonist as our next treatment of choice. It has been proven to be effective in both psoriasis and RP[9].

In conclusion, we described a case of RP occurring concomitantly with psoriasis vulgaris and PsA. Our patient achieved remission by administering azathioprine, in combination with methotrexate and sulfasalazine without any complications associated with immunosuppressive therapy.

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