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Perichondritis:When relapsing polychondritis should be considered?

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Abstract

Aim : Relapsing polychondritis (RP) is a rare inflammatory disease affecting multiple systems, characterized by recurrent inflammation of cartilaginous structures and proteoglycan-rich organs. We present two cases admitted to our department.

Case 1: A 71-year-old male with a history of gastropathy and gastroesophageal reflux presented with persistent pain and redness of the left pinna despite prolonged antibiotic treatment. He had a previous diagnosis of sensorineural hearing loss and tinnitus for two years, along with similar symptoms in the contralateral ear and the distal part of his nose in preceding years. After investigations, RP was suspected and treated with corticosteroids, resulting in favorable outcomes.

Case 2: A 45-year-old male with Crohn's disease and abdominal tuberculosis history presented with intense pain and redness of the right pinna. Initial treatment with antibiotics showed slight improvement in pain. Past medical records revealed previous episodes of nose redness and pain, knee arthralgia, and left eye redness resolved spontaneously prior to the auricular symptoms. High CRP levels initially decreased without normalization. The patient was admitted and treated with corticosteroids with remarkable improvement, and further investigations for RP were initiated.

Conclusion: Thorough examination of medical history, particularly previous occurrences of RP manifestations, aids in diagnosis. RP's etiology remains unknown, underscoring the importance of considering it in clinical practice

Discussion

-RP, a chronic condition marked by periods of exacerbation and remission, lacks a precise etiology. However, it is widely viewed as an immune-mediated disorder, given its well-documented association with other rheumatic and autoimmune diseases.

-Relapsing polychondritis occurs most frequently between the ages of 40 and 60 years.
-One case of RP associated to Crohn's disease was reported in littérature (2)

-In 1976, McAdam et al.created the first set of diagnostic criteria → 1979, Damiani et Levine: modified McAdam's criteria (histological changes and response to treatment) →1986, Michet et al.:the most recent set of criteria (based solely on symptoms).(3)

-The ear is the most frequently affected organ, typically with inflammation of the pinna, often leading to a cauliflower deformity, and characteristic sparing of the ear lobule.(2)

-The nose is another target organ, in which the loss of septal cartilage may result in a "saddle nose" appearance.(2)

-Eye involvement manifests as episcleritis, conjunctivitis, or iritis. (2)

-Arthritis or joint pain: frequently observed.

-Respiratory symptoms: a mild cough → severe manifestations (suffocation due to edematous tracheitis or life-threatening bronchial constriction).

-Aortic insufficiency, dissecting thoracic aneurysms, or systemic vasculitis→fatal outcomes

-Minor chondritis in the nose or ears :non-steroidal anti-inflammatory drugs, glucocorticoids, colchicine, or dapsone. (1)
-In more severe cases/reduce reliance on glucocorticoids: conventional disease-modifying antirheumatic drugs (cDMARDs) (methotrexate, azathioprine, cyclosporin, leflunomide, mycophenolate mofetil, cyclophosphamide..) (1)

1st observation

A 71-year-old male with a medical history of gastropathy of the antrum and a gastroesophageal reflux.
-Symtoms: Pain and redness of the left pinna sparing the tragus and the lobule non improved with broad-spectrum antibiotics, targeting staphylococcus with good cartilage distribution.
-Medical record: sensorineural hearing loss and tinnitus progressing over 2 years;
Similar symptomatology of the controlateral ear 2 years ago;
pain and redness of the distal part of the nose 1 year ago

⟹ Clinical suspicion of RP

-Treatment: corticosteroids at a dose of 0.5mg/kg/day, totaling 10 days.
-Great improvement
-Investigations: Thoracic CT-scan; PFTs; TTE and ophthalmologic examination (all normal)

Before treatment

After treatment

2nd Observation

A 45-year-old male with a medical history of Crohn disease and abdominal tuberculosis
-Symptoms: Pain and redness of the right pinna sparing the tragus and the lobule and precedeed by a right knee arthralgia and redness of the left eye
-Medical record: pain and redness of the distal part of the nose 4 months ago resolving spontaneously
-Put on antibiotics with slight clinical improvement (pain)
-CRP was high at first than decreased without being negative

⟹ Clinical suspicion of RP

-Treatment: corticosteroids at a dose of 0.5mg/kg/day totaling 10 days
-We completed the investigations like the other patient
-The improvement was spectacular

Before treatment

After treatment

Conclusion

The diagnosis of RP should be considered when perichondritis does not improve with antibiotic treatment. Indeed,a thorough analysis of the entire medical history with specific questions about the occurrence of the manifestations of the disease in the past helps to establish the diagnosis.

References

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