LETTER TO THE EDITOR

PALINDROMIC RHEUMATISM ASSOCIATED WITH PRIMARY HYPERTROPHIC OSTEOARTHRPOPATHY

Samuel Katsuyuki Shinjo, Mauricio Levy-Neto, Eduardo Ferreira Borba

INTRODUCTION

Hypertrophic osteoarthropathy (HOA) is a syndrome characterized by the triad of finger clubbing, periosteal new bone formation, skin and soft tissue changes, giving an acromegaloïd look.1 It is present in 2 distinct syndromes: primary and secondary. The former is a rare inherited disorder occurring predominantly in males and not associated with any known disease process. The secondary syndrome is acquired and often associated with malignancy, especially lung carcinoma.2-4 Rarely, it can also be associated with nonmalignant disorders, such as chronic liver disease, inflammatory bowel disease, cyanotic heart disease and other forms of shunts (cavernous hemangiomas and arteriovenous fistulae, 3 sarcoidosis, 5 and pulmonary tuberculosis. 8,9 If the underlying disease is correctable, the musculoskeletal manifestations may resolve, suggesting a humoral factor.10 Both types may be associated with peculiar skin changes known as pachyderma, characterized by thickened, furrowed skin. When idiopathic HOA is associated with pachyderma, it is called pachydermoperiostosis.11 In extremely rare instances, it may occur without pachyderma, possibly representing a nosologically distinct form of osteoarthropathy.12

Palindromic rheumatism is a recurrent or relapsing oligoarticular arthritis of short duration with peri- and pararticular tissue inflammation, resulting in no residual clinical or radiographic changes. Hench and Rosenberg first used the term in 1944 to describe 34 patients with recurrent, short-duration polyarthritis.13 In the long term, a substantial proportion of patients with palindromic rheumatism will go on to develop rheumatoid arthritis or other connective tissue diseases (eg, systemic lupus erythematosus, Wegener granulomatosis, or spondyloarthritis), but the determinants of subsequent chronic disease have not yet to been well established.14,15 Positive rheumatoid factor in females and early involvement of the wrist and proximal interphalangeal joints predict subsequent development of rheumatoid arthritis of other connective tissue disease in patients with palindromic rheumatism.16

Besides crystal arthritis, there are several diseases that may be clinically similar to palindromic rheumatism, such as systemic autoinflammatory disorders (periodic fever syndromes), Whipple’s disease, arthritis associated with hyperlipidemia, and intermittent hydrarthrosis.14

In the present work, we report an unusual case of a Brazilian male patient with palindromic rheumatism manifested in association with primary HOA without pachyderma. Disease activity was controlled with diphosphate chloroquine combined with methotrexate, leading to remission of arthritis relapse.

CASE REPORT

A 49-year-old black male civil servant had presented short (hours to 2 days) and intermittent asymmetric migratory oligoarthritis of the feet (metatarsal-phalanges), knees, and hands (wrist, metacarpal-phalanges, and proximal inter-phalanges) during the previous year. Those symptoms were not accompanied by morning stiffness, enthesitis, fever, weight loss, uveitis, intestinal, or genital-urinary alterations. His past history revealed significant digital clubbing since childhood. He reported that his grandfather also had digital clubbing. He was a chronic smoker, but not an abnormal alcohol consumer.

Physical examination showed clubbing and hypertrophic changes in fingers and toes (Figures 1A and 1B). Moreover, arthritis in the left ankle and right second and third metacarpal-phalanges were also observed. No skin thickening, lesions, or cardiopulmonary disease features were detected.

Laboratory investigation showed a persistent 3- to 4-fold increase of erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) levels, along with a normal uric acid level, absence of antinuclear antibodies, and a 1200 IU/mL titer of rheumatoid factor. Hepatic enzymes, blood
cell counts, serum calcium and phosphorus, thyroid hor-
mones, and growth hormone were all within normal ranges.

X-rays of the hands and feet showed no evidence of bone destruction. However, a marked clubbing with mush-
rooming of the tufts was found (Figures 2A and 2B). The chest x-ray was normal while pulmonary computerized to-
mography showed emphysema of the upper lung lobes. Pe-
riostosis in tibiae and fibulae, limited to the diaphysis with a monolayer configuration, were also detected (Figure 3). Echocardiography was normal.

A nonsteroidal anti-inflammatory and an antimalaria
drug (diphasate chloroquine 250 mg/day) were intro-
duced followed by methotrexate (10 mg/week), with effi-
cient control of arthritic crises as well as ESR and CRP levels.

**DISCUSSION**

We reporte an unusual case of a male patient with palindromic rheumatism manifested in association with pri-
mary HOA without pachyderma. Systemic diseases such
as chronic liver disease, inflammatory bowel disease, and
cyanotic heart disease and other forms of shunts, which
could have contributed to characterization as a secondary
HOA type, were not present.

The patient had been asymptomatic until 1 year previ-
ously, when oligoarthritis migratory manifestation com-
enced, without constitutional symptoms. No malignant
manifestations, infectious diseases, hyperlipidemia, or
medicine intake antecedence were found. Regarding con-
nective tissue disease, radiography analysis of the extremi-
ties showed no destruction or erosions; antinuclear antibod-
ies were negative, but titers of rheumatoid factor were el-
evated. Moreover, a persistent elevation of ESR and CRP
levels was observed. Rheumatoid factor is frequently
present in sera of patient with palindromic rheumatism,
with a prevalence of between 30% and 60%, without any
associated rheumatoid disease. The presence of rheuma-
toid factor has been associated with severity of arthritis cri-
ses and a high probability of development of rheumatoid
arthritis during the follow-up of patients.

Palindromic rheumatism can be evaluated by 3 clinical
standards: (a) clinical remission of articular crisis; (b) per-
sistence of recurrent articular crisis; and (c) evolution to
chronic disease such as rheumatoid arthritis (one-third to
one-half of cases). The latency period between the onset
of palindromic rheumatism and the development of rheu-
matoid arthritis is variable, from several weeks to more than
10 years. In case present here, the patient proved posi-
tive for rheumatoid factor, although this was not clinically
suggestive of rheumatoid arthritis, even through radiologi-
ical images.

To date, there is no consensus on the treatment of
palindromic rheumatism. One of the difficulties regarding
the assessment of pharmacological action is the self-lim-
ited crisis of palindromic rheumatism. Moreover, because
of the low incidence of this disease, controlled studies are
difficult to carry out. Nonsteroid anti-inflammatory drugs
and glucocorticosteroid can be used during crises, but their
intake does not decrease the duration or prevent new cri-

![Figure 1](image1.png)

**Figure 1** - Clubbing and hypertrophic changes of fingers and toes, demonstrating marked clubbing and hypertrophic changes (1A, 1B)

![Figure 2](image2.png)

**Figure 2** - Anteroposterior view of radiographs showing fingers and toes with mushrooming of the tufts (2A, 2B)

![Figure 3](image3.png)

**Figure 3** - Periostosis in left tibiae and fibulae, limited to the diaphysis with a monolayer configuration

![Figure 4](image4.png)

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no studies on palindromic rheumatism with leflunomide or methotrexate. In our patient, since administration of non-steroid anti-inflammatory and antimalaria drugs was inefficient in controlling the arthritic crises, we added methotrexate with good results, controlling the crises after 6 months. Moreover, laboratory analysis showed a decrease in ESR, CRP, and rheumatoid factor levels. However, it remains undefined whether this association will retard or stop possible progression to rheumatoid arthritis; this will require a longer follow-up study.

REFERENCES