Clarithromycin in adult-onset Still’s disease. Case report with a 1-year follow-up

Sirs,

The anti-inflammatory action of clarithromycin (CM) is well known (1). A pilot study on the use of CM in RA was published in 2002: 18 RA patients unresponsive to DMARDs were treated with CM for 6 months with a successful result in 10 of them. CM showed a very rapid onset of action and lowered the plasma levels of APL2 and PGE2 (2). These data have been confirmed in other studies in mice and in human leukocytes (3,4). Moreover, other macrolides with immunosuppressive action (cyclomycin, tacrolimus/FK-506, sirolimus/rapamycin) have been used effectively in RA (5-8).

We report the case of a 50-year-old male affected by a violent arthropathy in the shoulders, sternoclavicular regions and knees, with difficulty in walking and performing activities of daily life, hyperpyrexia (temperatures reaching 39°C), headache, and urticaria-like papules in the trunk. WBC/mm³ were as high as 12-16,000, ESR 80 mm, and CRP28 mg/dl (normal value < 5). The initial diagnosis was polymyalgia rheumatica treated with prednisone 50 mg/day with slight clinical and functional improvement. After 16 months he was admitted to the rheumatology unit of a Department of Internal Medicine. His ESR was 120 mm, WBC 18,600/mm³. IgE and IgM were elevated, antiphospholipid antibodies IgG and IgM were present at low titres; complement and CIC were positive; Clq, haemoculture and tests for viral (hepatitis, cytomegalovirus, mononucleosis) or bacteria (Salmonella paratyphi B at a level of 1/200. Also negative were tumoral markers, ANA, anti DNA, AMA, APCA, LAC, cryoglobulins, RF, CPK, LDH, TSH, tissues polypeptidic antigen, NSE, PSAand calcemia. HL-Atypical was A2,11; B 44,51; CW5; BW 4. Chest X-ray showed chronic bronchitis; an X-ray of the vertebral column showed osteoarthrosis; echography of the abdomen, axilllas and urinary tract showed small axillary and inguinal nodes and hepatomegaly; CT scan of the sacroiliac joints showed sclerosis of both joints and rarefaction of the subcortical bone. X-rays of the knees, feet and sterno-clavicular joints, CT scan of the chest and abdomen, total body bone scintigraphy, electroneurography of the lower limbs, biopsy of the bone marrow and ultrasonic bone densitometry were negative. The skin biopsy showed slight derma perivascular lymphocyte infiltrates (chronic dermatitis). The final diagnosis was adult-onset Still’s disease (AOSD), which was treated with methylprednisolone 8 mg t.i.d. added to methotrexate 15 mg/week. Six months later the patient’s HAQ score was 1.625 and fever was often present. ESR was 79 mm; CRP41 mg/dl, WBC 17.800/mm³. Because of the headache the diagnosis was an otorhinolaryngologist whose diagnosis was acute sinusitis which was treated with CM 500 mg t.i.d. for 10 days.

In a short time the fever, headache and arthropathy became less serious (ESR 20 mm, CRP 21.5 mg/dl), but after discontinuing CM treatment the symptoms became serious as before (ESR 52 mm, CRP 49.5 mg/dl). The patient underwent the same treatment again and after a few days showed renewed improvement (ESR 30 mm, CRP 23 mg/dl). A rheumatological consultation on the use of CM in RA was carried out: CM was continued at a dose of 500 mg t.i.d., methotrexate was stopped because ineffective, and methylprednisolone was gradually reduced to 6 mg per day. After 1 year of treatment the patient was able to work every day; the HAQ score was 0; and fever, myalgias and stiffness were present only a few hours per month. WBC three was 14,500/mm³, ESR 50 mm, and CRP 15 mg/dl. No side effect was present.

In conclusion, this case report shows that CM may play a role in AOSD, even in patients unresponsive to high dose steroids and methotrexate. The reason is the anti-inflammatory action of CM. The successful outcome in this case of AOSD and the other successful RA cases in a previous pilot study (2) indicate that CM has a very rapid onset of action; for this reason it could be used in rheumatic patients in association with MP and DMARDs as an initial treatment to achieve a rapid improvement.

G. SAVIOLA¹, MD
L. ABGI ALI¹, MD
S. SHAMS EDWARD², MD
C. DAVOLF, MD
M. TAGLIEtti, MD
1Salvatore Maugeri Foundation IRCCS, Rheumatology and Rehabilitation Unit, Castel Goffredo, Mantua; 2Department of Internal Medicine, Spedali Civili, Brescia, Italy.

Address correspondence to: Gianantonio SAVIOLA MD, Rheumatology and Rehabilitation Unit, Salvatore Maugeri Foundation IRCCS, via Ospeada 36, 46042, Castel Goffredo, Mantua, Italy. E-mail: g.savoli@fsm.it

References
2. SAVIOLA G, ABGI ALI L, ROSSINI et al.: Clarithromycin in rheumatoid arthritis pa-

3. TERAO H, ASANO K, KANAI K et al.: Suppresive activity of macrolide antibiotics on nitric oxide production by lipopolysacchae-
4. MIYAZAKI M, ZAITSU M, HONJO K et al.: Macrolide antibiotics inhibit prostaglandin E2 synthesis and mRNAexpression of pros-
taglandin synthet enzymes in human leuko-
6. FURST DE, SAAG K, FLEISCHMANN MR et al.: Efficacy of tacrolimus in rheumatoid arthritis patients who have been treated un-
successfully with methotrexate: a six-month, double-blind, randomized, dose-ranging stu-
7. KOVARIK JM, BURTIN P: Immunosuppress-
sant in advanced clinical development for or-
8. DROSOS AA: Newer immunosuppressive drugs: their potential role in rheumatoid arth-

Letters to the Editor

Juvenile temporal arteritis: A case report and review of the literature

Sirs,

Juvenile Temporal Arteritis (JTA) is an extremely uncommon vasculitis of unknown etiology affecting older children and young adults. In contrast to the classic form of temporal arteritis, it is not a systemic disease nor does it cause local symptoms at the temporal area. Up until now, ten documented cases have been reported in the literature (1-7). We herein report the eleventh case of this rare entity and review the relevant literature.

Our patient was a 31-year-old male who developed a painless lump in the left temporal area a year ago, which gradually grew to a diameter of about one centimeter. Otherwise, he was completely asymptomatic. He had this lump removed and the biopsy was reported as showing a granulomatous, non-giant cell arteritis with a prominent eosinophilic infiltration. Six months later, while in perfect health, a routine hematol-
ogy, biochemistry and serology (including CRP, RF and HBsAg) were all normal.

While completely asymptomatic, he was re-

documented cases have been reported in the literature (1-7). We herein report the eleventh case of this rare entity and review the relevant liter-

ture.

Our patient was a 31-year-old male who
developed a painless lump in the left temporal area a year ago, which gradually grew to a diameter of about one centimeter. Otherwise, he was completely asymptomatic. He had this lump removed and the biopsy was reported as showing a granulomatous, non-giant cell arteritis with a prominent eosinophilic infiltration. Six months later, while in perfect health, a routine hematology, biochemistry and serology (including CRP, RF and HBsAg) were all normal.

While completely asymptomatic, he was referred to us because of the biopsy report. He denied any symptom compatible with a systemic rheumatic or other disease. He was a