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 (cyclosporin, 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 (temperature were tumoral markers, ANA, anti RF, CPK, LDH, TSH, tissues polypeptidic neuraminidase were negative except for Widal's serodiagnosis which was positive for 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, PSAa and calcemia. HAmyalgia was A2,11; B 44,51; CW5: BW4. Chest X-ray showed chronic bronchitis; anemia was A2,11; B 44,51; CW5; BW4.

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 patient was visited by 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 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.

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References

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 showed a painless lump in the left 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.
healthy looking young man with a completely normal physical examination and painless, normally palpated temporal artery pulses. Chest x-ray was normal as were his biochemical and serologic profile, including CRP. His ESR was 3 mm, his Hb 13.6 g%, the WBC count 6620/mm³ with 8.5% eosinophils. The biopsy was re-evaluated and the diagnosis of JTA was made (Fig. 1). The patient has remained asymptomatic, since then, performing normally.

Four cases of the disease were reported for the first time in 1975 by Lie et al. (1), who coined the term “juvenile temporal arteritis” for an otherwise asymptomatic disease presenting with a painless lump at the temporal artery region in older children and young adults. The histopathology had revealed intimal proliferation and a lymphohistiocytic infiltrate containing many eosinophils but no giant cells, and some degree of focal disruption of the internal elastic lamina. A fifth case was reported in 1986 (2), the sixth in 1994 (3), two cases of bilateral JTA in 1995 (4) and 1996 (5) each, a ninth case in 1999 (6), and the tenth in 2002 (7), interestingly in an elderly woman who otherwise fulfilled the criteria for the disease. Besides the absence of clinical symptoms and the histopathologic picture characteristic for the disease, there is some question regarding the presence of peripheral eosinophilia. In 6 of the 10 cases so far described, an eosinophil count was not reported (1,3,4). Out of the remaining four, two had impressive peripheral eosinophilia (2,5), one had a normal count (6), and the other had a count just above the upper normal limit (7). Our patient had also an eosinophil count just above normal.

Watanabe et al., who reported the tenth case (7) in an elderly female, suggested that JTA is an accessory form of Kimura disease (8), which is a persistent and recurrent illness with peripheral eosinophilia and histologic findings very similar to JTA involving several arterial regions, albeit most commonly the head.

Two other diseases in which a non-giant cell eosinophilic arteritis of the temporal artery has been reported include the acquired immunodeficiency syndrome (one case) (9) and Buerger’s disease (3 cases) (10). Our patient had no evidence of either of the two conditions. Furthermore, the possibility of Churg-Strauss syndrome, which has been occasionally reported to involve the temporal artery, was easily ruled out in our patient in view of the absence of any typical or serologic finding of this disease.

In conclusion, our patient represents the eleventh case of JTA described in the literature, and as such it is worth reporting, in order to provide as much information as possible for a disease that quite rare, as has already been suggested by several investigators. Furthermore, this report, being the second of this rare disease described in the rheumatologic literature, will sensitize colleagues regarding this unusual temporal vasculitis affecting children and young adults.

Could placental abruption be an antiphospholipid antibody related disorder?

Sirs,

A 35-year-old woman was referred to us at her 12th week of gestation. An unexplained placental abruption occurring at the 22nd week of pregnancy with loss of a normal male fetus had taken place one year before. Antiphospholipid antibodies (aPL) tested according to International Consensus statement (1) repeatedly revealed a medium level of immunoglobulin (Ig) G anticardiolipin antibodies (aCL), while IgM aCL, IgG/IgM anti-β₂-glycoprotein I antibodies (anti-β₂GPI) were absent.

As the patient’s clinical and laboratory data were in accordance with the International Classification Criteria (1), definite APS was considered to be present and anticoagulant prophylaxis was started with nadroparin (Seleparina, Italfarmaco, Italy) at a dosage of 2,850 anti-Xa U self-administered subcutaneously twice a day. Subsequently nadroparin doses were gradually increased to 6,650 U twice daily in order to guarantee an anti-factor Xa level of between 0.1 and 0.6 U/mL over a 24-hour period. By the 12th week of gestation a steady fall in IgG aCL titer was observed, reaching its lowest values at the 22nd, 29th and 32nd weeks, when the most significant decreases in the platelet count were recorded (Fig. 1).

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