Visceral leishmaniasis in a patient with systemic juvenile arthritis treated by IL-1RA agonist (Anakinra)

Sirs,

The treatment of patients with severe systemic idiopathic juvenile arthritis (sJIA) is likely to improve thanks to the advent of new biotherapies. Tumour necrosis factor (TNF) blockade by TNF receptor analog (Etanercept) was demonstrated to be effective in approximately fifty percent of sJIA patients having polyarthritus but with restriction of no systemic symptoms (1). More recently, Verbsky et al. and Pascual et al. reported their experience with recombiant IL1-RA agonist Anakinra, in a few patients with sJIA, showing early remission of both systemic symptoms and arthritis (2, 3). In spite of apparent high benefit for sJIA patients, anti-cytokine therapies may induce serious and unpredictable side effects. We report a case of visceral leishmaniasis (VL) in a patient with sJIA while receiving treatment by Anakinra.

This 9 year-old girl, living in the southeast of France, had sJIA since the age of 4 years old. In spite of high dose steroids, she developed severe polyarthritis, unresponsive to methotrexate and cyclosporine. In addition, she developed three episodes of MAS in which one was under treatment by Etanercept. In November 2004, her arthritis was very active with thirty-four affected joints. She also had rash, splenomegaly, and enlarged lymphnodes (8). Cytopenia, hypergammaglobulinemia and hypertriglyceridemia are common reflecting various degrees of MAS (9). These features are shared with sJIA patients in whom macrophages present high level of activation mediated by proinflammatory cytokines especially IL-6, IL-1p and TNF (10). SJA patients have episodes of MAS triggered by viral infection and medications. Cytokine blockade induces experimental macrophage tolerance to intracellular opportunists and is the main complication in patients with sJIA because it mimics the disease and the life style of this opportunistic. VL is endemic in southern France where the reservoirs are chronically infected dogs. VL is endemic in southern France where the reservoirs are chronically infected dogs. In spite of apparent high benefit for sJIA patients, anti-cytokine therapies may induce serious and unpredictable side effects. We report a case of visceral leishmaniasis (VL) in a patient with sJIA while receiving treatment by Anakinra.