Sacroiliitis as a manifestation of Hodgkin’s disease in young females

Sirs,

Uncommon symptoms can impede the correct diagnosis of Hodgkin’s lymphoma (HL); a good example is skeletal involvement at the disease onset that can mimic a rheumatic disorder.

Case report no. 1. A 14-year-old female had low back pain with incomplete right sciatica. Her laboratory parameters were: WBC 11.400/mm³, ESR 62/68/74 mm, CRP 2.5/5.6/4.8 mg/dl (<0.5 mg/dl); RF 34/32 i.u. (normal value < 20), and cryoglobulins present in traces. HLA-B27 was absent. Echography of kidneys and urinary tract was normal. X-ray of the pelvis showed bilateral sacroiliitis with erosions on the right side (Fig. 1). Treatment with ketoprofen 320 mg per day had satisfactory results.

Twelve months later the patient experienced a recurrence with hyperthermia to 39°C, asthenia, 6 kg weight loss, and right supraventricular adenopathy. ESR was 95 mm, CRP 5.0 mg/dl, and RF 32 i.u.; ANA was present at low titres. Needle biopsy showed a stage IV Hodgkin’s lymphoma, subtype - mixed cellularity. TC scan confirmed involvement of the mediastinum, pulmonary parenchyma and lien. A 99Tc whole body bone scan showed increased uptake in the right iliium and near the right sacroiliac joint, and little uptake in the left sacroiliac joint. Case report no. 2. An obese 16-year-old female had atypical right coxalgia with incomplete sciatica, evening fever, Raynaud’s phenomenon, telangiectasia, and morning stiffness in the hands. X-ray of the right hip was negative. Haemoglobin was 10.2 g/L, ESR 90/880 mm, CRP weakly positive, and RF weakly positive. Treatment with low dose steroids and piroxicam was gradually tapered as symptoms remitted.

Five months later the patient developed acute pain at the left sacroiliac joint, fever, arthralgia and myalgia. ANA was 1:80 speckled, ESR 66/79 mm, CRP 3.6 mg/dl, RF weakly positive, and gammaglobulins 25.7%. HLA B27 was absent.

X-ray of the pelvis and Tc99 whole body bone scan were negative; a CAT scan of the lumbar spine showed disk protrusion at L3-L4 and L4-L5 with displacement of the peridural membrane and compression of the dural sac; echography of the hips and pelvis were negative. A Tc99 whole body bone scan showed increased uptake in both sacroiliac joints, which was marked in the left synovial tract (Fig. 2). CT of the sacroiliac joints confirmed fine erosions on the left side. The patient was treated with deflazacort, indomethacin and sulfasalazine 3 gm/day until she developed a left supraventricular lymphopathy. Needle biopsy one year after the onset of the disease showed a Hodgkin’s lymphoma (stage IV A), subtype: nodular sclerosis-mixed cellularity. A further whole body bone scan with Ga67 185MBq and SPET of the chest demonstrated involvement of the neck and lumbar and right iliac regions.

Pauciarticular JIA (juvenile rheumatoid arthritis) occurs in females less than 6 years of age. Juvenile enthesitis-related arthritis occurs in boys usually over 8 years of age; it is HLA B27 positive and involves both sacroiliac joints only after several years. Psoriatic arthritis is associated with psoriasis, affecting females throughout childhood and associated in 40% with HLA-B27 positive sacroiliitis.

ANA are present in 40-75% of children with pauciarticular onset JIA, especially in females and in association with chronic anterior uveitis. Acute anterior uveitis is often associated with HLA B27 positive juvenile enthesis-related arthritis. RF may be positive only in a few cases of polyarticular JIA.

In both of our cases the patients were female, over 14 years of age, without psoriasis, and with ANA and RF at borderline titres. They had unilateral sacroiliitis without involvement of the eyes, and the antigen HLA B27 was absent. HLAB27 negative unilateral sacroiliitis could be vital diagnostic clue to the presence of a Hodgkin’s lymphoma.

G. SAVIOLA1 F. DESIATI2
L. ABDI ALI3 E. LUPI4
C. TRENANT4 I. PONTIKAKI4
L.D. NOTARANGELO5 V. GERLONI4

1Rheumatology and Rehabilitation Unit and 2Radiology Department, Salvatore Maugeri Foundation IRCCS, Castel Goffredo, Mantua; 3Pediatrics Department, University of Brescia; 4Centre for Pediatric Rheumatology, University of Milan, Italy.

Address reprint requests to: Gianantonio Saviola MD, Rheumatology and Rehabilitation Unit, Salvatore Maugeri Foundation IRCCS, via Ospedale no. 36, 46042 Castel Goffredo, Mantua, Italy. E-mail guasiola@fsm.it

References