We report a case in which initially the wrong diagnosis of renal cell carcinoma with bone metastases was made. Nephrectomy and bone biopsy led to the right diagnosis of oncocytoma with transient osteoporosis. This report stresses the importance of pathological investigation and points to oncocytoma in the differential diagnosis of solid renal masses. In addition, the possible relationship between this tumour and migratory osteoporosis, which disappeared after surgery, is described.

**INTRODUCTION**

Oncocytes are transformed epithelial cells rich in mitochondria, probably representing senescent degenerative cellular changes. Renal oncocytoma is a benign tumour of renal tubular origin. Its incidence represents 3 to 10% of all solid renal masses. Most renal oncocytomas are found incidentally and usually follow a benign clinical course. Partial nephrectomy or enucleation has been advocated as curative. Here, we present a case of renal oncocytoma associated with bone lesions, which were due to migratory osteoporosis and muscle weakness.

**CASE REPORT**

A 42-year-old man presented with muscle weakness. For three years, he had been suffering from pain in his arms and legs not associated with trauma. He complained of progressive weakness of his shoulder and upper leg muscles and could not walk alone without help. He was not taking any medication. On physical examination, we saw a patient with atrophic shoulder and upper leg muscles and in the right part of the abdomen a large tumour was palpable. His weight was stable at 68 kg with a length of 1.70 m. Further examination showed no abnormalities. Laboratory examination revealed normal calcium (2.3 mmol/l), normal phosphate (0.8 mmol/l), elevated alkaline phosphatase 132 U/l, normal liver enzymes, and normal creatinine. Haematology showed no abnormalities; the ESR was 14 mm/h. Urine analysis revealed no erythrocyturia or leucocyturia. Electromyography was compatible with myopathy. Computer tomography of the abdomen showed a renal tumour (22 x 13 cm) in the right kidney without invasion in the adjacent structures (figure 1). There was
either a central scar or necrosis inside the tumour. The X-ray of the thorax was normal. Nuclear bone scintigraphy revealed multiple abnormalities, suggesting metastases (figure 2). The diagnosis of renal cell carcinoma with bone metastasis was made. It was decided to perform a nephrectomy; on operation a giant tumour of the right kidney was found (figure 3). Microscopically the diagnosis of oncocytoma was made (figure 4). Histological examination of the biopsy taken of the humerus and sacrum at the site of abnormal uptake on the nuclear bone scintigraphy was compatible with migratory osteoporosis without metastases of the oncocytoma (figure 5). Two months after the operation the patient was able to walk again without help and the muscle weakness had disappeared. Six months after nephrectomy control scintigraphy of the bones showed disappearance of the initial bone lesions.
DISCUSSION

In our patient we found an extremely large renal tumour and on nuclear scintigraphy a picture of bone metastases. Furthermore, there was a three-year history of generalised pain, predominantly in the upper and lower extremities and not associated with trauma. Initially, renal cell carcinoma with bone metastases was diagnosed. Nephrectomy led to the diagnosis of oncocytomas, which are usually discovered incidentally.\(^3\) Sometimes, in a minority of cases, bilateral multifocal renal oncocytoma is found.\(^4\) Because of the benign nature, multicentricity, possible bilaterality and absence of pathognomonic radiographic features, renal oncocytomas should considered in the differential diagnosis of solid renal masses. The fibrotic central scar in a larger mass, as in our case, has been described in oncocytoma and may be the most specific feature (figure 3).\(^5\)

Fine needle biopsy should be considered to avoid radical nephrectomy in selected patients.

In this patient, biopsies of the decalcified ossal lesions of the bone scintigraphy showed characteristics of migratory osteoporosis: oedema, active osteoclastic bone resorption and reactive bone formation in marrow spaces (figure 3).\(^5\)

Directly after nephrectomy all symptoms disappeared and a control bone scintigraphy six months after nephrectomy showed regression of the initial lesions. Probably the pain of the migratory osteoporosis and myopathy had induced the immobility leading to atrophic muscles.

We conclude that it is very likely that the ossal lesions had a pathogenetic relationship with this large oncocytoma in our patient.

Migratory osteoporosis is one of the two subgroups of transient osteoporosis.\(^7\) The other is regional osteoporosis. Transient osteoporosis is a condition of pain mostly occurring in the lower limbs but other locations are described. It consists of monoarticular or oligoarticular pain in young and middle-aged persons associated with temporary rapidly progressing osteopenia. Pain increases with weight-bearing and can last for several months up to years. A significant proportion of patients have recurrences, sometimes in the same joint but mostly at shifting locations in the same or opposite limb. Intervals up to 13 years have been reported.

The aetiology and pathogenesis are poorly understood. Thrombotic aetiology has been suggested based on an abnormality of fibrinolysis seen in patients with transient osteoporosis. Transient osteoporosis has also been described as a variant of reflex sympathetic dystrophy with myopathy and muscle atrophy.\(^4\) The treatment of transient osteoporosis is controversial; most cases are treated conservatively applying joint protection and cyclooxygenase inhibitors.

Furthermore, this case showed that an oncocytoma with paraneoplastic migratory osteoporosis can mimic a renal cell carcinoma with ossal metastases.

REFERENCES

A young woman with fever and a pericardial effusion

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ABSTRACT

A 19-year-old woman is presented with high-spiking fever, pericardial tamponade and respiratory failure. A diagnosis of adult onset Still’s disease was made. This is a rare inflammatory disease with an unknown aetiology. The diagnosis is made by exclusion and with the help of diagnostic criteria. Treatment with corticosteroids met with a good response.

INTRODUCTION

Fever is one of the most frequent presenting symptoms in medicine. Common causes of fever are infections, neoplasms or drug fever. Non-infectious inflammatory diseases can also cause fever. One of these is adult onset Still’s disease. Although rare, this disease is among the most frequent causes of fever of unknown origin within the group of non-infectious inflammatory diseases.1 In this case report we present a young woman with an unusual presentation of this disease.

CASE REPORT

A 19-year-old woman was admitted because of high-spiking fever of two weeks duration, accompanied by a dry cough, shortness of breath and painful muscles and joints. For some weeks, she had complained of a sore throat. On examination we saw an ill, tachypnoeic patient with a temperature of 41°C, pulse 120 beats/min, blood pressure 100/60 mmHg. There was no rash. Two enlarged lymph nodes were palpated in her neck. Examination of the lungs and heart was within normal limits. Liver and spleen were moderately enlarged. Inspection of the joints showed no abnormalities.

The erythrocyte sedimentation rate was 35 (<12 mm/h), cAMP receptor protein 174 (<10 mg/l), haemoglobin 6.6 (7.2-9.8 mmol/l), mean corpuscular volume 84 (81-96 fl), leucocytes 4.8 (4.0-11.0 x 109/L) (peripheral smear normal), thrombocytes 127 (150-400 x 109/L). Slightly elevated values were obtained for the aspartate aminotransferase (ASAT) at 48 (<45 U/l), alanine aminotransferase (ALAT) at 75 (<45 U/l) and lactate dehydrogenase (LDH) at 668 (<475 U/l). Arterial blood gas (without supplemental oxygen): pO2 7.7 (10-13 kPa), pCO2 4.4 (4.5-6.0 kPa). The chest X-ray showed an enlarged cardiac silhouette and a small infiltrate. The electrocardiogram was normal. Our first suspected diagnosis was an infectious disease, although other diagnoses such as non-infectious inflammatory diseases or (haematological) neoplasms were considered. The patient was treated for a suspected pneumonia with cefuroxim and erythromycin. Cultures of blood and urine were negative. Extensive serology in paired sera for infections caused by Mycoplasma, pneumonine Chlamydia spp., Legionella spp. and adenovirus was negative. Autoimmune tests were negative (antinuclear antibody (ANA), antineutrophil cytoplasmic antigen and rheumatoid factor). Ferritin was 347 (14-150 μg/l). Ultrasound revealed hepatosplenomegaly and a pericardial effusion. Her clinical condition deteriorated, with high swinging fever (figure 1), respiratory failure with bilateral infiltrates and impending pericardial tamponade. She was intubated and received artificial ventilation; just prior to this she was...
given dexamethasone 100 mg iv. The pericardial effusion, which appeared to be a transudate, was drained (250 ml); cultures of this fluid were negative. Within 24 hours her condition improved dramatically and she was extubated. Because adult onset Still’s disease was suspected, we stopped the antibiotics and started treatment with diclofenac: 3 x 50 mg orally per day on day 15. When the fever recurred and liver-enzyme abnormalities increased, prednisone was prescribed in an oral dose of 40 mg daily. Again the improvement was striking (figure 1).

She was discharged in good condition, the laboratory abnormalities normalised. The steroids were tapered successfully over a few months.

**DISCUSSION**

Adult onset Still’s disease is a rare form of seronegative polyarthritis of unknown aetiology. It is characterised by sudden onset of a high-spiking fever, a passing erythematous or salmon-coloured maculopapular rash involving the trunk and extremities (90%), polyarthritis or oligoarthritis (50%) and peripheral lymph node enlargement (50%). Splenomegaly, hepatomegaly, pericarditis and transient pulmonary infiltrates have been described in 30 to 40% of patients. Cardiac tamponade and respiratory failure, as was the case in our patient, is a very rare complication.

Laboratory findings are aspecific with signs of inflammation and elevations of ASAT and ALAT, and LDH. Serum ferritin is markedly elevated in 70% of patients. Characteristically all autoimmune tests are negative. The diagnosis is made by exclusion. Diagnostic criteria are divided into major criteria: fever of at least 39°C lasting for at least one week, arthralgias or arthritis lasting for two weeks, characteristic rash and leucocytosis; and minor criteria: sore throat, lymph node swelling, hepatomegaly or splenomegaly, abnormal liver enzyme tests, negative tests for ANA and rheumatoid factor. To establish the diagnosis a patient should fulfil five of the criteria listed above, as did our patient (two major and all minor criteria). First-line therapy for adult Still’s disease is NSAIDs (about 25% of patients react to NSAIDs). Alternatively corticosteroids (0.5-1 mg/kg/day) can be given. In patients with high fever of longer duration, pericardial effusion and pulmonary infiltrates, in the absence of infectious or autoimmune causes, adult type Still’s disease should be suspected and a trial of corticosteroids is warranted.

**REFERENCES**