

neuropathy [6] or related to progressive destruction of the base of the distal phalanx by the rheumatoid process [1].

Acro-osteolysis has also been reported after trauma, burns, frostbite or neuropathic disorders such as insensitivity to pain and tarsal tunnel syndrome. It has also been observed in systemic diseases such as sarcoidosis, diabetes mellitus, renal diseases and hyperparathyroidism. Vascular disorders (Buerger's and Raynaud's disease), phenitoin or ergot treatment or intoxication with vinyl chloride have also been incriminated.

Idiopathic forms [14] have occasionally been associated with joint erosions [15]. These latter differ from rheumatoid erosions by their rapid progression and their wide distribution. However, clinical manifestations can mimic juvenile chronic arthritis [16].

As no other cause could be disclosed, we hypothesised that our patient presented an idiopathic local form of osteolysis, not related to RA (preserved space of the distal interphalangeal joint), although the extension of the hand bony erosions probably denoted a longer RA evolution than admitted by the patient. Chronic vascular insufficiency of the lower limbs could have contributed to the development of the acro-osteolysis. Clinical examination and biological results were not in favour of any vasculitis or neuropathy related to RA. Occurrence of both phenomena was probably fortuitous.

REFERENCES

- 1 Kemp SS, Dalinka MK, Schumacher HR. Acro-osteolysis. Etiologic and radiological considerations. *JAMA* 1986 ; 255 : 2058-61.
- 2 Dihlmann W. Gelenke-Wirbelverbindungen. Klinische Radiologie einschließlich Computer-tomographie - Diagnose, Differentialdiagnose. 3d ed. Stuttgart: Georg Thieme; 1987. p. 152-4.
- 3 Burgener FA, Korman M. Differential diagnosis in conventional radiology. 2d ed. Stuttgart: Thieme Verlag; 1991. p. 278.
- 4 Dähnert W. Radiology review manual. 2d ed. Baltimore: Williams & Wilkins; 1993. p. 3-4.
- 5 Udell J, Schumacher HR, Kaplan F, Fallon MD. Idiopathic familial acroosteolysis : histomorphometric study of bone and literature review of the Hajdu-Cheney syndrome. *Arthritis Rheum* 1986 ; 29 : 1032-8.
- 6 Resnick D, Niwayama G. Rheumatoid arthritis and the seronegative spondyloarthropathies : radiographic and pathologic concepts. In: Resnick D, Niwayama G, Eds. *Diagnosis of bone and joint disorders*. 2d ed. Philadelphia: W.B. Saunders; 1988. p. 895-953.
- 7 Resnick D, Niwayama G. Osteolysis and chondrolysis. In: Resnick D, Niwayama G, Eds. *Diagnosis of bone and joint disorders*. 2d ed. Philadelphia: W.B. Saunders; 1988. p. 4140-70.
- 8 Bassett LW, Blocka KL, Furst DE, Clements PJ, Gold RH. Skeletal findings in progressive systemic sclerosis (scleroderma). *Am J Roentgenol* 1981 ; 136 : 1121-6.
- 9 Resnick D. Dermatomyositis and polymyositis. In: Resnick D, Niwayama G, Eds. *Diagnosis of bone and joint disorders*. 2d ed. Philadelphia: W.B. Saunders; 1988. p. 1319-31.
- 10 Le Quintrec JL, Chazerain P, Campagne JP, Cerf I, Ziza JM. Acrodermatite continue de Hallopeau avec ostéolyse, précédant de 35 ans un psoriasis commun avec oligo-arthrite. *Rev Rhum* 1992 ; 59 : 844-6.
- 11 Orzheshkovskii VV, Timofeeva NV. Acral osteolysis in Bechterew's disease. *Vopr Revm* 1973 ; 13 : 86-90.
- 12 Peter JB, Pearson CM, Marmor L. Erosive osteoarthritis of the hands. *Arthritis Rheum* 1966 ; 9 : 365-88.
- 13 Rohlfing BM, Basch CM, Genant HK. Acro-osteolysis as the sole skeletal manifestation of rheumatoid vasculitis. *Br J Radiol* 1977 ; 50 : 830-3.
- 14 Bisagni-Faure A, Giraudet-Le Quintrec JS, Job-Deslandre C, Menkes CJ. L'ostéolyse essentielle. Étude de 3 cas et essai de classification. *Ann Méd Interne* 1991 ; 142 : 17-20.
- 15 De Smet AA. Acro-osteolysis occurring in a patient with idiopathic multicentric osteolysis. *Skelet Radiol* 1980 ; 5 : 29-34.
- 16 Costa MM, Santos H, Santos MJ, Medeira A, Da Costa T, De Queiroz V. Idiopathic multicentric osteolysis : a rare disease mimicking juvenile chronic arthritis. *Clin Rheumatol* 1996 ; 15 : 97-8.

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Reflex sympathetic dystrophy and pregnancy: a case report

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osteomalacia / pregnancy / reflex sympathetic dystrophy syndrome

INTRODUCTION

Reflex sympathetic dystrophy syndrome (RSDS) is a common condition that can escape diagnosis when it occurs during pregnancy. The most common site of involvement in pregnancy is the hip, whereas the extensive form is exceedingly rare. We report a case of extensive RSDS with onset during the second trimester of pregnancy in a 40-year-old patient with an underlying bone disease.

CASE REPORT

A 40-year-old woman with a history of two uneventful pregnancies presented at the fifth month of her third

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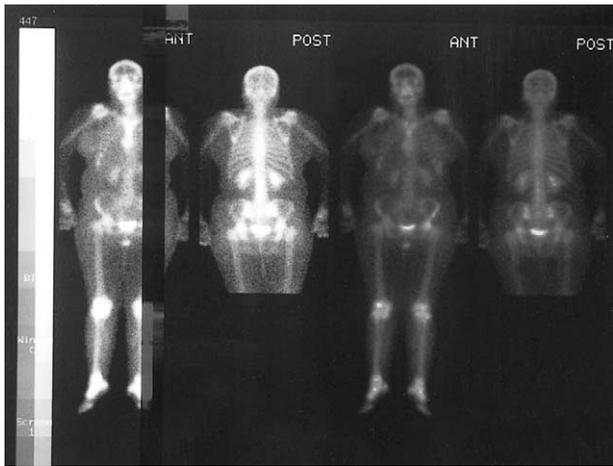


Figure 1. Bone scan: increased uptake in the left hip and in both knees and ankles.

pregnancy with bilateral mixed L5 sciatica, low back pain, and painful functional impairment that made walking and standing difficult. Findings were normal from a magnetic resonance imaging (MRI) scan of the thoracic and lumbar spine. A few weeks later, she started experienced pain in the left hip, with a mixed time pattern. Knee pain occurred within a few days, as well as a flare of pain in the ankles. Examination of the hip after delivery showed mild painful range-of-motion restriction in the above-mentioned joints. Plain radiographs disclosed moderate demineralization in the left hip and both ankles, as well as patchy demineralization in the knees, without joint space alterations. A bone scan showed foci of increased uptake in the left hip and in both knees and ankles (*figure 1*). The diagnosis was extensive RSDS of the lower limbs. Laboratory test results were as follows: erythrocyte sedimentation rate, 20 mm/h; severe hypocalcemia (78 mg/l on one occasion) with hypocalciuria (30, 36, and 39 mg/24 h); serum phosphate level, 37 mg/l; 24-hour phosphate excretion, 900 mg/24 h; serum alkaline phosphatase level, 218 UI/l (normal, < 270 UI/l), and serum 25 (OH)D₃, 11 µg/l (normal, 15 to 30). Osteomalacia related to vitamin D deficiency was suspected after elimination of renal, gastrointestinal, and other causes of osteomalacia.

Absorptiometry measurements showed that bone mineral density at the lumbar spine, femoral neck, and forearm were -2.89, -2.29, -2.75, respectively, indicating trabecular osteoporosis.

Treatment consisted of elimination of weight-bearing, griseofulvin 1.5 g/day for two months, cal-

cium 1 g/day, and vitamin D 4000 IU/day. The clinical course was favorable two months after treatment initiation. Levels of calcium and phosphate returned to normal. After seven months, demineralization was still apparent at the knees and ankles.

DISCUSSION

The frequency of RSDS during pregnancy is probably underestimated as a result of underdiagnosis. Misleading and minimally symptomatic forms usually fail to prompt the imaging studies that could establish the diagnosis. Mean age is 32 years [1, 2]. RSDS can occur during the first pregnancy or subsequent pregnancies and is most common during the third trimester [1-5]. Cases during the second trimester (as in our patient) and postpartal period are rare [5]. The hip is involved in 90% of cases [1, 4, 6, 7], with a predilection for the left side. Our patient had extensive disease, which is exceedingly rare [6, 8, 9]. Risk factors for extensive RSDS include vitamin D deficiency responsible for osteomalacia with microfractures [9-11]; osteoporosis [9]; and low back pain with nerve root pain, which may trigger and perpetuate the reflex arc by stimulating the proprioceptive receptors in the joints [6]. Other factors have been incriminated in the genesis of RSDS of the hip during pregnancy, such as mechanical stress (weight gain and microtrauma related to fetal movements), compression of the pelvic sympathetic nerve and obturator nerve by the uterus, and vascular disorders related to venous stasis by compression of the inferior vena cava.

The diagnosis of RSDS during pregnancy rests on MRI, which was not performed in our patient. The other investigations are contraindicated during pregnancy. Treatment during pregnancy relies mainly on rest and analgesics. After delivery, calcitonin can be given to patients who do not breastfeed. Griseofulvin is rarely used; it was given to our patient because she could not afford more costly drugs. Recovery within a few weeks to a few months after delivery is the rule, even in the absence of treatment. Our patient recovered rapidly with rest and griseofulvin. Radiological abnormalities improve slowly [6]. Recurrences can occur during subsequent pregnancies [1, 2, 9]. Femoral neck fracture is a serious complication that occurs in 14 to 17% of patients with RSDS of the hip [1, 4]. Factors that increase the risk of hip fracture may include the RSDS itself, forced abduction during delivery, and underlying bone abnormalities.

REFERENCES

- 1 Brocq O, Simon E, Bongain A, Gillet JY, Euller-Ziegler L. Fracture du col fémoral compliquant une algodystrophie au cours de la grossesse. *Press Méd* 1999 ; 28 : 1165-6.
- 2 Gouin F, Maulaz D, Aillet G, Pietu G, Passuti N, Bainvel JV. Fracture du col du fémur compliquant une algodystrophie de hanche au cours de la grossesse. *Rev Chir Orthop* 1992 ; 78 : 45-50.
- 3 Favier T, Begue T, Lelirzin R, Pidhorz L. L'algodystrophie de hanche au cours de la grossesse. *Press Méd* 1993 ; 22 : 270.
- 4 Poncelet C, Perdu M, Levy-Weil F, Philippe HJ, Nisand I. Reflex sympathetic dystrophy in pregnancy: nine cases and review of the literature. *Eur J Obstet Gynecol Reprod Biol* 1997 ; 86: 55-63 [abstract].
- 5 Doury P. L'algodystrophie de la grossesse et du post-partum. *Sem Hôp Paris* 1996 ; 72 : 117-24.
- 6 Hamidou M, Lassoued S, Fournie B, Fournie A. Algodystrophie de hanche et grossesse. *J Gynecol Obstet Biol Reprod* 1990 ; 19 : 324-6.
- 7 Gétin Y, Bucas JP, Houlné P. Forme compliquée d'algodystrophie de hanche et grossesse. *Rev Fr Gynécol Obstét* 1983 ; 78 : 477-9.
- 8 Bocquet B, Cedoz ME, Doux-Girard MF, Philipot R, Frobert JL, Chandet M. Algodystrophie extensive des membres inférieurs au cours de la grossesse. *Press Méd* 1993 ; 22 : 1104-5.
- 9 Billey T, Dromer C, Pagès M, Caulier M, Lassoued S, Fournié B. Fracture spontanée du col fémoral au cours d'une algodystrophie de hanche pendant la grossesse. *Rev Rhum Mal Ostéoartic* 1992 ; 59 : 494-6.
- 10 Dumolard A, Gaudin P, Juvin R, Asquier C, Phélip X. Algodystrophie extensive des membres inférieurs révélatrice d'une ostéomalacie par syndrome de Fanconi. *Rev Rhum (Ed Fr)* 1997 ; 64ème année : 601-2.
- 11 Juchet H, Ollier S, Nicodeme R, Arlet P. Algodystrophie et ostéomalacie. *Press Méd* 1993 ; 22 : 1282.

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Focussing of extracorporeal shock wave therapy (ESWT) in the treatment of calcifying tendinitis

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calcifying tendinitis / extracorporeal shock wave / focussing / ESWT

J.E. Charrin and E.R. Noel reported results from 32 patients with calcifying tendinitis which were treated with ESWT under ultrasonic guidance [1]. Patients received $2 - 3 \times 2000$ impulses of 0.32 mJ/mm^2 without local anaesthetics in an open trial. The authors found the results (subjective improvement 55% and Constant score not statistically changed after 24 weeks) less favourable than those reported before [2]. As a main reason for this they named the ultrasonic guidance that was used.

Charrin and Noel [1] stated to be the first to use ultrasonic guidance and a dedicated extracorporeal lithotripter for the treatment of calcifying tendinitis, but in 1992 Dahmen [3] already recommended ultrasonic guidance using an experimental lithotripter. While good success rates were reported from studies if the shock waves were focussed using fluoroscopy [2], the vast majority of patients in daily clinical practise is treated without focussing or by employing ultrasonographic focussing.

We can confirm the findings of Charrin and Noel regarding the importance of exact focussing of ESWT based upon a prospective randomised study with independent observer. Technical specifications of the device we used and the fluoroscopic focusing procedure have been published recently [4]. Fifty patients were included in this study. One group received 2×2000 impulses ($ED_{+} 0.35 \text{ mJ/mm}^2$ measured with a PVDF-hydrophone) under local anaesthesia focused on the origin of the supraspinatus tendon. Patients in the second group received ESWT focused to the calcified area. Statistical analyses of the results showed a significant superiority of ESWT focused to the calcified area in all observed parameters after one year. Interestingly there was no significant difference in the resorption rate of the deposits between both groups although the clinical results were statistically different. Improvement in Constant score and pain was comparable to the natural history of the disease [5] and to Charrin and Noel [1] when ESWT was focussed on the insertion of the tendon.

We do agree with Charrin and Noel [1] that fluoroscopic control is more precise in identifying a calcific deposit and focussing the shock waves. We could demonstrate a significant and clinically important effect of ESWT when it was exactly focussed under fluoroscopic

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