518-521 *Clinical rheumatology*, 1994, **13**, N° 3

Case Report

Percutaneous Vertebroplasty with Acrylic Cement in the Treatment of a Langerhans Cell Vertebral Histiocytosis

T. CARDON, E. HACHULLA, R.M. FLIPO*, P. CHASTANET**, C. ROSE, X. DEPREZ*, B. DUQUESNOY*, B. DELCAMBRE*, B. DEVULDER

Summary **A 25-year-old man developed multiple eosinophilic granuloma of bone including vertebral and sacral localization. Radiotherapy was initially administered. One year later, a relapse occurred in another vertebrae which was previously irradiated. Percutaneous vertebroplasty was, for the first time to our knowledge performed, in this indication, with a good clinical result with follow-up now for one year. We emphasize that such treatment is permissible only in symptomatic, progressive lesions, with threatened decompensation of spinal stability. This technique should be used only on an adult.**

Key words **Histiocytosis, Vertebroplasty, Acrylic cement.**

INTRODUCTION

Because of its unpredictable course and its sometimes spontaneously favourable outcome, Langerhans histiocytosis raises a difficult therapeutic problem (1). This report concerns the first case to our knowledge, with painful and lytic involvement of L2, treated by vertebral cementoplasty. This management approach resulted in the relief of pain and vertebral consolidation with a one year follow-up.

CASE REPORT

A 25-year-old man was hospitalised in February 1990 for low back pain with left-sided sciatica. Pain had gradually worsened over 2 years, awakening the patient at night despite the use of routine analgesics. Standard X-rays showed two lacunae eroding the cortex, situated in the left wing of the sacrum and within the vertebral body of L3 (Fig. 1). CT scan films showed that the sacral lacuna was the site of a space occupying lesion extending to the epidural space. Histological examination of this lesion showed evidence of an inflammatory granuloma containing numerous CD1 + Langerhans cells, typical of an eosinophilic granuloma. The only other lesion found involved the left middle ear. Radiotherapy (40 grays with an irradiation field extending from L2 to the sacrum) stopped clinical and radiological progression. The ear lesion was treated surgically. A year later, however, the patient was hospitalised because of the recurrence of low back pain, with radiation to the right inguinal fold. Pain was explained by the onset of a collapse of the vertebral body of L2 with osteolysis of the right pedicle (MRI) (Fig. 2). A vertebra plana type collapse of D10 and a mandibular lesion were also detected. In addition, a pulmonary interstitial syndrome suspected on the basis of chest X-ray was confirmed by a high resolution CT scan of the lungs. Bronchoalveolar lavage contained 3% CD1 + cells and respiratory function tests were normal.

Percutaneous methyl-methacrylate vertebroplasty of L2 resulted in a rapid analgesic action (Fig. 3 and Fig. 4). The patient was soon able to stand upright, protected by wearing a corset for 3 months. The various disease sites all remained asymptomatic one year later.

DISCUSSION

Indications for treatment in multiple eosinophilic granulomas and in systemic forms of Langerhans histiocy-

From the Department of Internal Medicine, University of Lille, France; *Department of Rheumatology, University of Lille, France; **Department of Radiology, University of Lille, France.

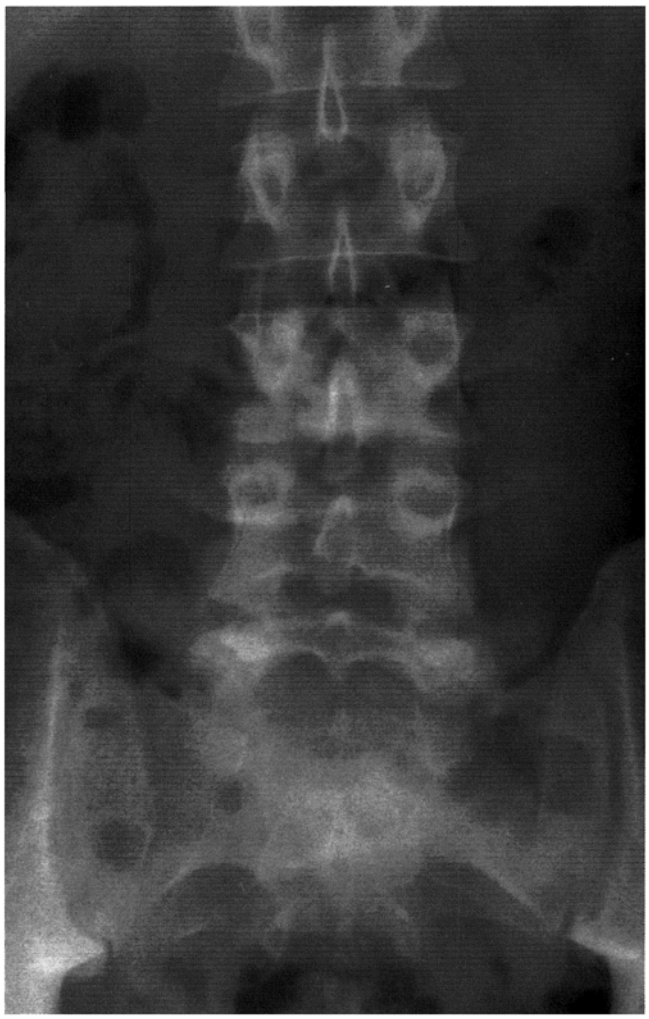


Fig. 1: Slight subsidence of the right portion of the upper plateau of L3 and condensation of the right pedicle. Lytic lesion in the left sacral wing.

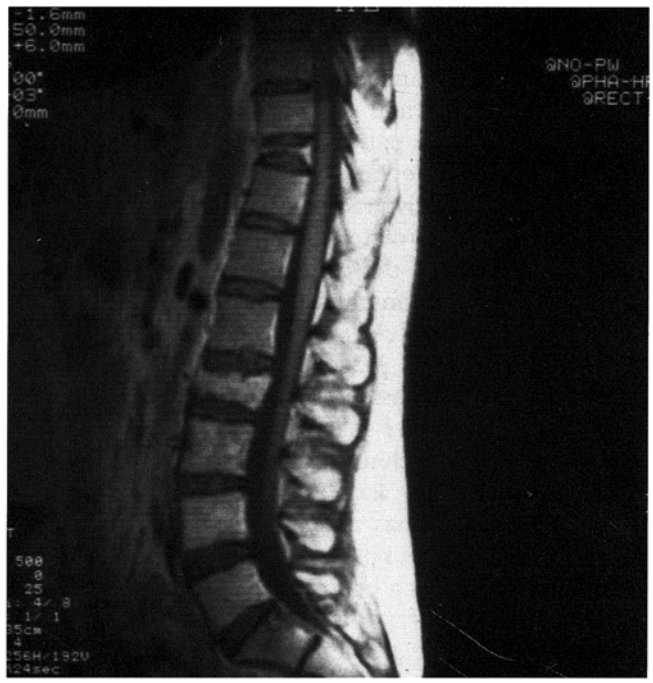
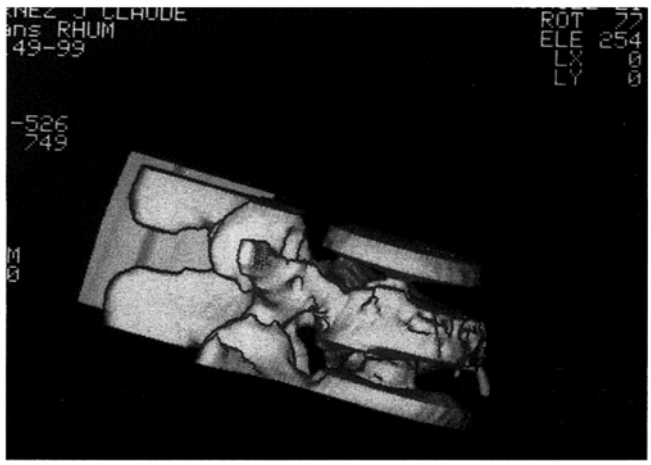


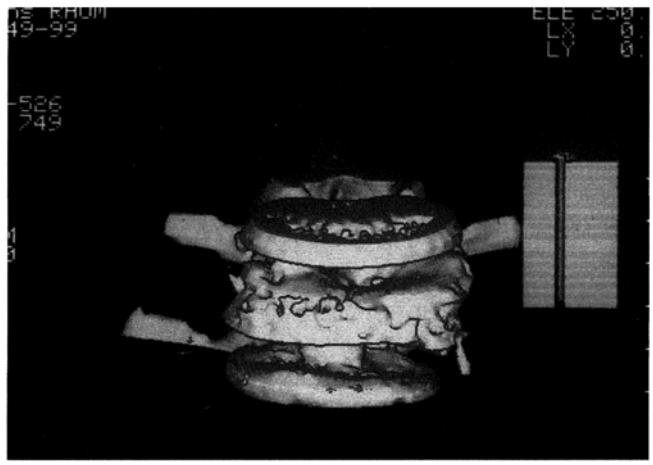
Fig. 2: 2a) MRI, sagittal section, weighted series in T1 Collapse of D10 with vertebra plana appearance Collapse of the body of L2. The hypersignal of the body of the last 4 lumbar vertebrae is related to the appearance of fat, secondary to radiotherapy one year previously.

2b) MRI, sagittal section, weighted series in T2 (1st echo). More marked hypersignal of the body of L2 related to the recent collapse. No backward displacement of the posterior wall.

tosis remain highly empirical, in particular in adults (2). This can be explained by the rarity of the affection, the heterogeneous nature of its clinical presentations and its unpredictable course, all of which make it difficult to establish prognostic criteria and set up clinical trials (1).

In the present case, there was the risk that the collapse of L2 would progress to vertebra plana with resultant decompensation of spinal statics. Its onset in an area previously irradiated made any surgery difficult and excluded any possibility of radiotherapy. Chemotherapy was felt to be excessive for a non-malignant affection, the course of which is unpredictable, with the possibility of spontaneous regression (3). It would even seem that systemic treatment has little or no influence on the progression of bone lesions (4). Pulmonary involvement did not appear to be aggressive, in particular since





Figs. 4a and 4b: CT scan after cementoplasty. Three-dimensional reconstruction.

it had been possible to persuade the patient to stop smoking.

Vertebroplasty is a novel therapeutic alternative offering the possibility of the rapid relief of pain and increased spinal stability (5). It involves the injection into a damaged vertebral body of acrylic cement in the process of polymerisation, i.e., a mixture of methyl-methacrylate monomer and polymer. The spreads through the lesion, hardens and produces consolidation of the vertebral body. The patient requires only simple local anesthesia or neuroleptanalgesia. The procedure is performed in an operative radiology room. The vertebra is reached via a posterolateral and/or transpedicular approach. Injection of the cement, made highly opaque by the addition of tantalum powder, is monitored on an image intensifier television screen. The instruments used for the puncture procedure are withdrawn as soon as the cement is considered to have hardened sufficiently. The procedure takes one hour on average.

At present this technique is sometimes indicated in primary or secondary vertebral tumours and aggressive vertebral hemangiomas and has even been suggested for fractures related to osteoporosis (5). Cord and radicular compression is a contraindication to the method. Although the long-term outcome is unknown, vertebroplasty was felt to be a potentially less dangerous treatment, in our patient, than chemotherapy with its leutemogenic potential or surgery in an irradiated area. Excellent results, with follow-up now for several years, have been reported in vertebral hemangiomas (5).

Thus, vertebral Langerhans histiocytosis may be a new indication for cementoplasty. Such treatment is justified only in symptomatic, progressive lesions, with threatened decompensation of spinal stability. The technique should not be used in children since the height of the vertebral body may return to normal spontaneously during growth (6).

Fig. 3: Cementoplasty of the body of L2. Right posterolateral approach. Injection of PPMA (polymethylacrylate) Penetration of L2/L3 disc by polymer.

REFERENCES

1. Osband, M.E., Pochedly, C. Histiocytosis X: an overview. Hematology Oncology Clin N Am 1987, 1, 1-7.
2. Greenberger, J.S., Crocker, A.C., Vawte, r.G., Jaffe, N., Cassady, J.R. Results of treatment of 127 patients with systemic histiocytosis (Letterersiwe Syndrome, Schuller-Christian Syndrome and multifocal eosinophilic granuloma). Medicine 1981, 60, 311-338.
3. Sartoris, D.J., Parker, B.R. Histiocytosis X. Rate and pattern of resolution of osseous lesions. Radiology 1984, 152, 679-684.

4. Bollini, G., Jouve, J.L., Gentet, J.C., Jacquemier, M., Bouyala, J.M. Bone lesions in histiocytosis X. J Pediatr Ortho 1991, 11, 469-477.
5. Deramont, H., Galibert, P., Debussche-Depriester, C., Pruvo, J.P., Heleg, A., Hodes, J.E. Percutaneous vertebroplasty with methylmethracrylate : technique, method, results. Scientific exhibits, RNSA Chicago, Radiology 1990, 117 suppl, 352.
6. Ippolito, E., Farsetti, P., Tidisco, C. Vertebra plana. J Bone and Joint Surg 1984, 9, 1364-1368.

Received : 7 June 1993
Accepted : 29 December 1993
Correspondence to : Dr. E. HACHULLA,
Service de Médecine Interne, CHRU, Hôpital Claude Huriez, 59037 Lille, France.