International Orthopaedics (SICOT) (2002) 26:61–62
DOI 10.1007/s00264-001-0318-0

CASE REPORT

M. Moran · A. Paul

Octreotide scanning in the detection of a mesenchymal tumour in the pubic symphysis causing hypophosphataemic osteomalacia

Accepted: 18 October 2001 / Published online: 17 January 2002

Abstract Oncogenic hypophosphataemic osteomalacia is a rare condition. The causative tumour is often difficult to locate. Primary tumours have been reported in the head and neck, skeleton, and soft tissue. Octreotide scanning was used in this case and detected a mesenchymal tumour in the pubic symphysis.

Résumé Ostéomalacie hypophosphataemique oncogénétique est une condition rare. La tumeur causative est souvent difficile de localiser. Les tumeurs fondamentales ont été rapportées dans la tête et cou, squelette et tissus doux. Parcourir octreotide a été utilisé dans ce cas et a détecté une tumeur du mésenchyme dans la symphyse pubien.

Introduction

Acquired hypophosphataemia is a rare cause of osteomalacia. It is often associated with mesenchymal tumours, which can be distributed widely throughout the body [2, 5, 8]. Unless these tumours are superficial, they can be difficult to locate. It is important to localise these tumours as resection can lead to a complete resolution of the condition [6]. Octreotide is a somatostatin analogue used in the treatment of some neuroendocrine tumours and acromegaly. It is possible to radiolabel the somatostatin analogue in an attempt to detect tumours that express somatostatin receptors [7].

Case report

A 56-year-old man was diagnosed as having *onco*genic hypophosphataemic osteomalacia following octreotide scanning which localised a primary mesenchymal tumour to the pubic symphysis.

M. Moran (✉) · A. Paul
University Department of Orthopaedic Surgery,
Manchester Royal Infirmary, Oxford Road,
Manchester M13 9WL, UK
e-mail: matt.moran@ukgateway.net
Tel.: +44-161-2761234, Fax: +44-161-2768006

Seventeen years previously this man had suffered bilateral subcapital fractures of the neck of his femur following a road traffic accident. These were treated with Newman pin fixation. Routine biochemistry performed at the time revealed a low serum phosphate at 0.37 (0.7–1.4 mmol/l). Initial serum alkaline phosphotase was mildly raised at 137 (25–100 U/l). Serum calcium was normal at 2.19 (2.15–2.65 mmol/l). He was admitted for further investigation. A bone biopsy was performed from the iliac crest that

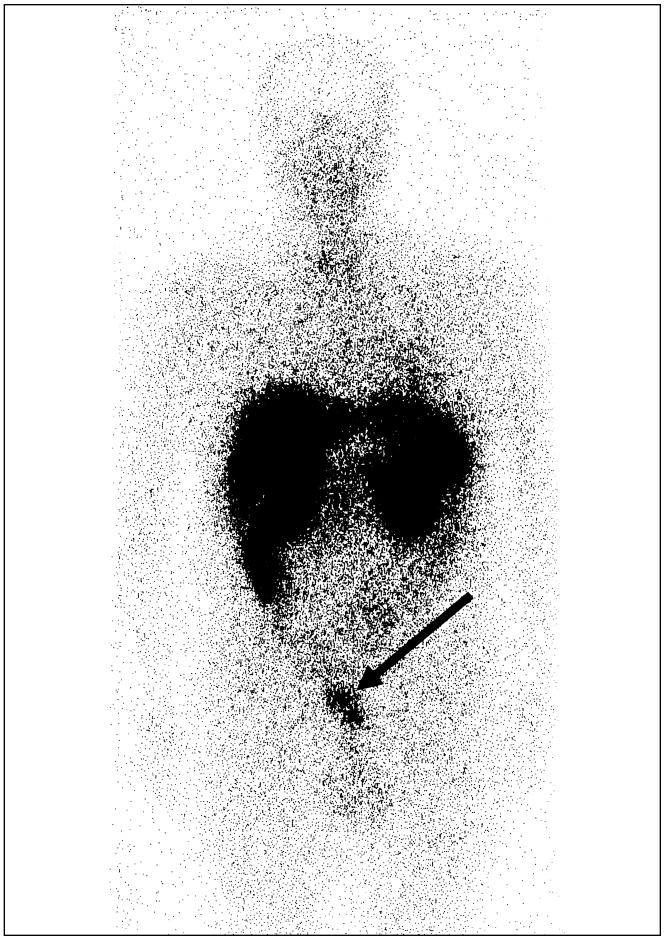

Fig. 1 ^{111}In-labelled pentatreotide image at 24 h showing a hot spot in the pubic symphysis

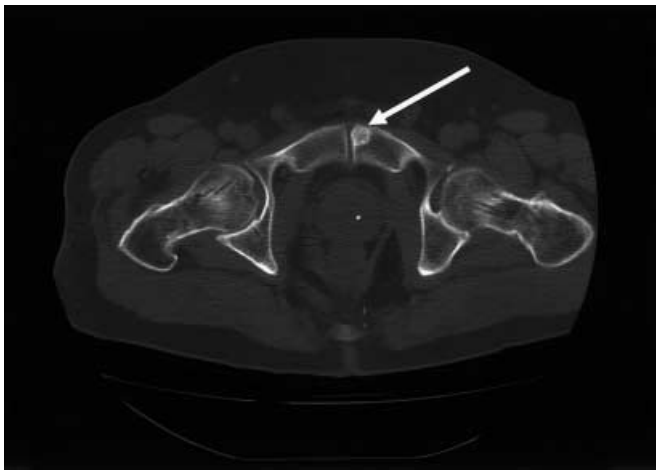

Fig. 2 CT scan of bony pelvis demonstrating a 1 cm lesion with ring calcification

showed a gross excess of osteoid, and a diagnosis of acquired hypophosphataemic osteomalacia was made. A CT scan of the abdomen was normal. A gallium scan did not reveal any abnormal uptake. A technetium-labelled bone scan showed multiple areas of abnormal uptake in the axial skeleton, consistent with a diagnosis of hypophosphataemic osteomalacia. Treatment was initiated with oral calcium and phosphate supplements, as well as calciferol. He was reviewed regularly in outpatients, and improvements were seen in subsequent bone biopsies and in his serum biochemistry. A repeat bone scan performed 6 years after commencing treatment was essentially normal. During the period of follow-up he had not sustained any further fractures. He had, however, undergone coronary artery bypass surgery.

Octreotide scanning recently has become recognised as a means of detecting mesenchymal tumours [7]. This was carried out on our patient in an attempt to localise a potential tumour as the cause of acquired hypophosphataemic osteomalacia. Somatostatin receptor scanning was performed using radiolabelled 111-In-pentatreotide. The scan revealed abnormal increased uptake in the region of the pubic symphysis (Fig. 1). A CT scan showed a 1 cm lesion with ring calcification at the pubic symphysis, typical of a mesenchymal tumour (Fig. 2).

Discussion

Mesenchymal tumours are a common cause of acquired hypophosphataemic osteomalacia. The tumours arise from a wide range of tissues and are usually benign in nature. Approximately half originate from bone [1]. Various imaging modalities have been used to try and find these tumours in the body, ranging from simple CT to total MR skeletal surveys [3]. Octreotide scanning has been used to study lesions, which have been demonstrated by other radiological means [4], and, to a lesser degree, in chasing the location of a primary tumour causing hypophosphataemic osteomalacia. It relies on the expression of somatostatin receptors by mesenchymal tumours. Scanning is commonly performed with ^{111}In-labelled pentatreotide. This allows localisation of the tumour as a target for more detailed anatomical scanning and possible resection. It may even be possible to use this technology therapeutically in radioimmunoguided surgery or labelling of octreotide with a beta-emitting radionuclide [4]. The exact role of somatostatin receptors in oncogenic hypophosphataemic osteomalacia has yet to be elucidated.

This case demonstrates the use of octreotide scanning in the detection of an unknown primary mesenchymal tumour causing oncogenic hypophosphataemic osteomalacia. This technique has been reported only in a few previous cases [4, 5, 9] and never before, to our knowledge, demonstrating a tumour in the pubic symphysis.

Although, as previously stated, these tumours often behave in a benign manner, the long-term sequelae can be serious. Deterioration in renal function and cardiovascular disease are more common in this group of patients. Fractures are common in osteomalacic individuals. Fractures of the neck of femur are known to be common in osteoporotic elderly patients. Although this patient sustained his injuries in a road traffic accident, bilateral subcapital fractured neck of femurs is unusual in a young adult. This case illustrates the importance of investigation for a cause of this type of fracture in a younger patient. In this case a diagnosis of acquired hypophosphataemic osteomalacia was made and appropriate treatment commenced.

References

1. Crouzet J, Mimoune H, Beraneck L, Juan LH (1995) Hypophosphataemic osteomalacia with plantar neurilemoma. A review of the literature (100 cases). Rev Rhum Engl Ed 62:463–466
2. David K, Revesz T, Kratimenos G et al (1996) Oncogenic osteomalacia associated with a meningeal phosphaturic mesenchymal tumour. J Neurosurg 84:288–292
3. Fukumoto S, Takeuchi Y, Nagano A et al (1999) Diagnostic utility of magnetic resonance imaging skeletal survey in oncogenic osteomalacia. Bone 25:375–377
4. Giannakenas C, Kalofonos HP, Apostolopoulos D et al (2000) Scintographic imaging of sarcomatous tumours with [(111) In-DTPA-phe-1]-octreotide. Oncology 58:18–24
5. Gonzalez-Compta X, Manos-Pujol M, Foglia-Fernandez M et al (1998) Oncogenic osteomalacia: case report and review of head and neck associated tumours. J Laryngol Otol 112:389–392
6. McGuire MH, Merenda JT, Etzhorn JR et al (1989) Oncogenic osteomalacia. A case report. Clin Orthop 244:305–308
7. Nguyen BD, Wang EA (1999) Indium-111 pentetreotide scintigraphy of mesenchymal tumour with oncogenic osteomalacia. Clin Nuc Med 24:130–131
8. Prowse M, Brooks PM (1987) Oncogenic hypophosphataemic osteomalacia associated with a giant cell tumour of a tendon sheath. Aust N Z J Med 17:330–332
9. Rhee Y, Lee JD, Shin KH et al (2001) Oncogenic osteomalacia associated with mesenchymal tumour detected by indium-111 octreotide scintography. Clin Endocrinol (Oxf) 54:551–554