

# Magnetic resonance imaging: evaluation of recurrent eosinophilic granuloma of the ilium

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**A case of eosinophilic granuloma of the ilium is reported, in which the patient was examined by magnetic resonance imaging for post-operative evaluation purposes. The assessment of recurrent musculoskeletal tumour has always been difficult. Radiography, computed tomography, ultrasound and magnetic resonance imaging have been used in the detection of recurrent tumour. Magnetic resonance imaging, combined with contrast enhancement and fat suppression technique, is a sensitive way of detecting recurrent eosinophilic granuloma involving the bony skeleton.**

*HKMJ 1995; 1:348-350*

*Key words: Eosinophilic granuloma; Magnetic resonance imaging*

## Introduction

Eosinophilic granuloma results from granulomatous proliferation of the reticulum cell in the reticulo-endothelial system, and may occur in the skeleton, spleen, liver, thymus, lymph nodes, or skin. Clinical manifestations depend on the tissue type affected and the patient's age, with diffuse, severe disease occurring more frequently in younger children.<sup>1</sup> With bone involvement, plain radiography reveals osteolytic lesions in advanced histiocytosis. The activity of lesion on skeletal scintigraphy varies from increased, to normal, to decreased activity. The ability to detect the lesions of histiocytosis depends on location and size. Radiography and skeletal scintigraphy should probably be used concurrently when defining the distribution of histiocytosis.<sup>2</sup> Computed tomography (CT) or magnetic resonance imaging (MRI) allows better evaluation of a tumour's extraosseous extension and detects any spinal canal encroachment. Magnetic resonance imaging serves the dual role of both lesion identification and staging. It is particularly useful in the

detection or exclusion of a lesion in a child with bone pain, despite negative radiographs. However, the application of MRI for the post-operative evaluation of recurrent disease has not been reported. We describe a case in which MRI detected pathologically documented recurrent histiocytosis involving the ilium.

## Case report

A 30-month-old boy presented with a one-month history of limping gait. There was no preceding account of fever, rash, or weight loss. The child's development had been normal. On examination, the child appeared well with normal neurological findings. The lymph nodes, liver, and spleen were not enlarged. There was a left antalgic gait, but the child denied any local tenderness. The range of movement was preserved on passive examination. His haemoglobin level was 12.4 g/dL, total white blood cell count was  $7.3 \times 10^9/L$ , and platelets  $468 \times 10^9/L$ . The differential count was normal without any blast cells. The erythrocyte sedimentation rate was 47 mm/h. Blood biochemistry revealed the following: calcium, 2.44 mmol/L; inorganic phosphate, 1.48 mmol/L; alkaline phosphatase, 183 U/L; alanine aminotransferase, 13 U/L; aspartate aminotransferase, 32 U/L; total bilirubin, 9  $\mu\text{mol/L}$ . Plain radiograph of the pelvis demonstrated an osteolytic lesion in the left iliac bone and bone scintigraphy showed abnormal uptake at the corresponding site. Non-enhanced MRI showed a well-

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circumscribed mass in the infero-lateral aspect of the left ilium, which was hypointense on T1-weighted (T1W) and hyperintense on T2-weighted (T2W) images. The adjacent muscles were not involved. Open biopsy yielded a piece of bone fragment infiltrated by mononuclear cells which were positive for S100 on immunohistochemical staining. A moderate amount of eosinophils were scattered around the lesion. The diagnosis was compatible with a localised form of Langerhans cell histiocytosis.

The patient was treated by curettage and cadaveric bone chip grafting. There was a gradual and complete clinical recovery. However, an osteolytic lesion persisted on the left iliac bone. At follow up 20 months later, the lytic lesion was more extensive radiographically, even though the child remained asymptomatic. Magnetic resonance imaging scans of the pelvis revealed a lesion with signal intensity predominantly hypointense on T1W images and hyperintense on T2W images. The iliac mass was enhanced together with the adjacent soft tissue after injection of gadolinium-diethylenetriaminepentaacetic acid (Gd-DTPA) (Fig 1). Short T1 inversion recovery (STIR) images demonstrated the lesion particularly well against adjacent darkened structures (Fig 2). A diagnosis of recurrent tumour was made, based on the MRI findings. This was confirmed by subsequent biopsy. As the child was asymptomatic, it was decided that a trial course of chemotherapy with steroid would be used before any surgical intervention was contemplated.

## Discussion

Eosinophilic granuloma, with Letterer-Siwe disease



**Fig 1.** Gd-DTPA-enhanced T1-weighted (TR 468/15) axial image showing contrast enhancement of the recurrent tumour (arrow) at the left ilium and the adjacent muscles

and Hand-Schuller-Christian syndrome, comprise the disease entity known as Langerhans cell histiocytosis. These lesions are often well-defined and localised on MRI, and less commonly, more diffuse. The signal intensity is decreased on T1W images and increased on T2W images at the site of histiocytic proliferation.<sup>3,4</sup> On fat suppression (STIR or ChemSat) images there is definite increased signal compared with the surrounding marrow.<sup>4</sup> Similarly, in spinal lesions, diffuse infiltration of the entire vertebral body—with or without vertebral collapse—is seen as decreased signal intensity on T1W images and increased signal intensity on T2W images.<sup>5</sup> The lesion typically shows contrast enhancement with Gd-DTPA.

The evaluation of recurrent musculoskeletal tumour has always posed a diagnostic problem. Although plain radiography is conventionally used in clinical follow up, CT and MRI have often been employed for the detection of recurrent tumour. However, MRI has been found to be more sensitive than CT in detecting recurrent musculoskeletal tumour.<sup>6</sup> In the patient with a metallic implant, MRI may allow the diagnosis of recurrent tumour when CT is non-diagnostic. Currently, MRI has replaced CT in the detection of recurrent musculoskeletal neoplasms. Although ultrasonography and MRI are characterised by similar sensitivity and specificity, ultrasonography may be more difficult to interpret in the early post-operative period. Hence, recognition of the post-operative appearance on MRI is valuable when interpreting MR images obtained after musculoskeletal surgery. Following operative removal or complete response of tumour to radiation or chemotherapy, early fibrosis and granulation tissue present in the tumour bed may cause difficulties in the post-



**Fig 2.** Gd-DTPA-enhanced STIR (TR 1500/20 TI 150) image in the coronal plane showing a bright signal intensity lesion at the left ilium

therapeutic evaluation for recurrent disease. Low signal intensity on T2W images after treatment has a 96% sensitivity for indicating an absence of active neoplasm. When increased signal intensity is obtained, the sensitivity for the detection of residual active tumour is 70%.<sup>7</sup> As the time gap between therapeutic intervention and the MRI examination increases, a hyperintense space-occupying lesion is likely to represent recurrent tumour. Organised fibrous scar tissue—unlike tumour lesion—should appear hypointense on both T1W and T2W images.<sup>8</sup> The presence of mass effect on surrounding tissues is also suggestive of recurrent tumour. Infiltration of soft tissue and destruction of bone beyond the structural changes that occurred before therapy are strongly indicative of recurrent tumour. However, in the patient who has undergone radiation therapy alone, lesions with high signal intensity on T2W images may represent both recurrent tumour and radiation-induced tissue changes.<sup>7</sup> Areas of low to intermediate signal intensity on T1W images without signal increases on T2W images and without nodular configuration typically represent post-surgical, post-chemotherapy or post-radiation therapy changes.

The situation is more complicated when paediatric patients with bone lesions are treated by curettage and bone-chip allografts. These allografts comprise cancellous bone harvested from cadaver sites which have been ground into different-sized fragments. These serve as a template for new bone formation and promote healing after curettage. Scans of these bone-chip allografts (T1W and T2W) have been described.<sup>9</sup> The most common finding is either homogeneous intermediate or heterogenous intermediate signal intensity on T1W images (with flecks of hyperintense fatty marrow). On T2W images, speckled increased signal intensity is often seen. The contrast agent Gd-DTPA has also been used to differentiate scar from tumour. Enhancing lesions represent tumour or organising scar or granulation tissue. However, well-organised scar tissue should not be enhanced on T1W sequences after Gd-DTPA injection.<sup>10</sup> Hence, if one finds an enhanc-

ing soft tissue mass lesion at the operative site with the typical MRI findings of the original tumour, replacing the bone-chip allograft signals, a diagnosis of recurrent tumour can be made with considerable confidence. This case with such findings was confirmed pathologically as having recurrent histiocytosis. Magnetic resonance imaging is a useful modality in the post-operative imaging of paediatric musculoskeletal tumour. When used with contrast enhancement and a fat suppression technique it is a sensitive way of detecting recurrent eosinophilic granuloma involving the bony skeleton.

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