CASE REPORT

Langerhans cell histiocytosis: another cause of a fluid–fluid level within an appendicular bony lesion

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ABSTRACT

We report a case of Langerhans cell histiocytosis (LCH) occurring in the pelvis of a 2-year 11-month-old female with fluid–fluid level seen on MRI. Aspiration of the fluid during biopsy showed it to be blood with a few inflammatory cells and eosinophils. Tissue obtained during the biopsy confirmed the diagnosis to be LCH. While fluid–fluid levels have been infrequently encountered in skull lesions due to LCH, they have yet to be reported in lesions of the appendicular skeleton. The aim of this report is to familiarize radiologists with the fact that fluid–fluid levels can occur in LCH of the appendicular skeleton in children.

SUMMARY

We report a case of Langerhans cell histiocytosis (LCH) occurring in the pelvis of a 2-year 11-month-old female with fluid–fluid level seen on MRI. Aspiration of the fluid during biopsy showed it to be blood with a few inflammatory cells and eosinophils. Tissue obtained during the biopsy confirmed the diagnosis to be LCH. While fluid–fluid levels have been infrequently encountered in skull lesions due to LCH, they have yet to be reported in lesions of the appendicular skeleton. The aim of this report is to familiarize radiologists with the fact that fluid–fluid levels can occur in LCH of the appendicular skeleton in children.

CLINICAL PRESENTATION

A 2-year 11-month-old female presented to our hospital with left hip pain. Laboratory tests did not reveal any significant finding and the inflammatory markers were not elevated.

IMAGING FINDINGS

Plain radiographs revealed an osteolytic lesion in the left iliac wing (Figure 1). The lesion was well defined in some areas but also showed cortical destruction with no discernible periosteal reaction. Fat-saturated T2 weighted images showed fluid–fluid level within the lesion (Figure 2a). The non-dependent part of the fluid was similar to water and the dependent portion was low in signal intensity, suggestive of blood products. Fat-saturated T1 weighted MRI showed the lesion to be well defined and isointense to muscle. A faint fluid–fluid level was noted within the lesion (Figure 2b). After the administration of intravenous gadolinium (gadopentetate dimeglumine 0.1 mmol kg⁻¹), the lesion showed peripheral enhancement and an enhancing internal septa was identified within the lesion (Figure 2c). There was also enhancement of the bone surrounding the lesion. The fluid–fluid level within the lesion was unchanged in appearance. The surrounding bone that...
enhanced on the $T_1$ weighted images was high in signal intensity on the fat-saturated $T_2$ weighted images, possibly owing to bony inflammation. Periosteal reaction was seen in both sequences. Another smaller lesion was noted within the right superior pubic ramus, with mild enhancement on post-contrast images. This lesion appeared solid and did not have a fluid–fluid level.

CT-guided aspiration of fluid contents followed by biopsy of the larger left iliac bone lesion was performed. Blood with a few inflammatory cells and eosinophils was aspirated. The biopsy (Figure 3) and immunohistochemistry confirmed it as LCH. A subsequent skeletal survey revealed another lesion in the T11 vertebral body. The patient was treated with intravenous vincristine and oral prednisolone for 6 weeks and is currently well on regular follow-up.

**DISCUSSION**

Fluid–fluid levels are characteristically described in aneurysmal bone cysts but are found in a wide range of bone lesions and are thus a non-specific finding. Fluid–fluid levels have been described in LCH lesions involving the calvarium but not of the appendicular skeleton. This is the first reported case of a fluid–fluid level occurring in a case of LCH of the appendicular skeleton. The occurrence of fluid–fluid level is believed to be due to intratumoral haemorrhage. This case is important as it further highlights the non-specificity of fluid–fluid levels in osteolytic lesions and LCH should still be included in the differential diagnoses of these lesions in children.

**LEARNING POINTS**

1. The aim of this report is to familiarize radiologists with the fact that fluid–fluid levels can occur in LCH of the appendicular skeleton in children.
2. This case also further highlights the non-specificity of fluid–fluid level in osteolytic lesions.

**CONSENT**

Written informed consent was obtained from the patient’s parents for publication of this case report, including accompanying images.
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