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Langerhans cell histiocytosis presenting spinal compression injury following vertebral fracture.

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INTRODUCTION:

We report a rare case of multiple spinal Langerhans cell histiocytosis (LCH) with neurological deficits following vertebral fracture.

CASE:

15-year-old boy had developing low back pain 2 months ago. He went to nearby orthopedic clinic and was seen conservatively. He could hardly walk with sensory disturbance of lower limbs 1 week ago. His neurological symptom was deteriorating rapidly. He was referred to our hospital by ambulance. He suffered from Frankel C neurological deficit of both lower leg with bladder and rectal disturbance. Plain X ray revealed vertebra plana of T4 due to compression fracture. CT showed osteolytic lesions in the T3-5 vertebra. MRI demonstrated extraskelatal tumor anterior to T3-5 vertebra and posterior protrusion of T4 which developed spinal cord compression. He underwent emergency laminectomy of T2-5, and partial tumor resection followed by posterior fixation using instrumentation of T1-7. His neurological symptom dramatically improved after the operation. Histological diagnosis was confirmed as LCH. Systemic examination revealed no other LCH except thoracic spine. As a result, he did not have lesion except for thoracic vertebra. He left our hospital 4 weeks after the operation. Systemic chemotherapy (VBL, PSL, 6-MP, MTX) was given. The patient shows no neurological symptom and no limitation of daily life, and imaging assessment demonstrates complete remission 1 year postoperatively.

DISCUSSION:

LCH is a comparatively rare tumor and the annual incidence is reported at approximately 6 per million children per year. Multiple spinal LCH has been reported in only 55 cases in the literature review published in 2011, demonstrating that about 30% showed neurological symptoms. However, few had neurological deficits which required emergency surgical decompression as we reported here. Although the use of chemotherapy to treat LCH is still controversial, chemotherapy is commonly advocated for multiple spinal LCH. Radiation should not be used as first choice, especially in children, because of secondary malignancy and growth arrest.

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