

Adult Langerhans cell histiocytosis involving adjacent vertebra on ^{18}F -FDG PET/CT imaging

Abstract

Langerhans cell histiocytosis (LCH) of the spine involving the intervertebral disk and adjacent vertebra is rare in adults. We report a case of a 56-year-old woman with neck pain and right upper limbs numbness. Fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F -FDG PET/CT) showed an osteolytic bone lesion in the fourth cervical vertebra (C4) with soft-tissue mass, invading the adjacent intervertebral disc and posterior vertebral body of C3, compressing dural sac and cervical spinal cord. The lesion showed increased ^{18}F -FDG uptake with a maximum standardized uptake value (SUVmax) of 6.75. Subsequently, the histopathologic examination confirmed the diagnosis of LCH.

Hell J Nucl Med 2021; 24(3): 269-271

Epub ahead of print: 17 December 2021

Published online: 28 December 2021

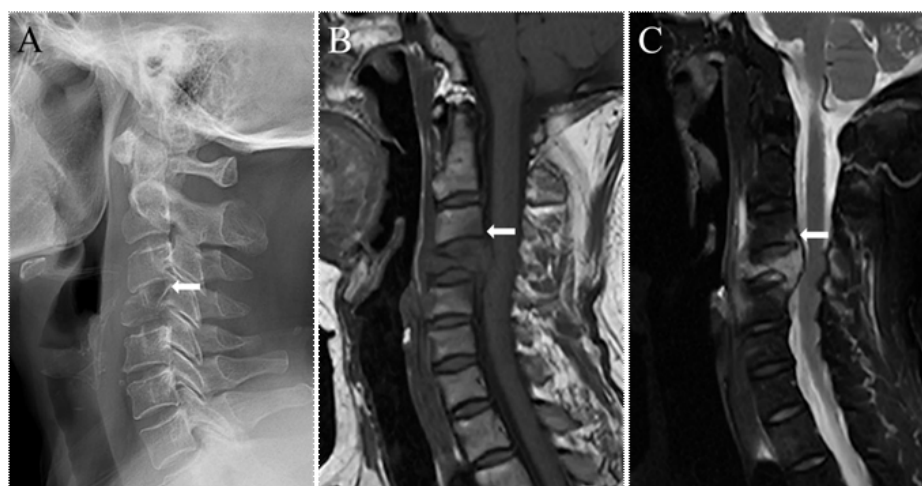


Figure 1. A cervical spine radiograph (A), and magnetic resonance imaging (MRI) of the cervical spine demonstrated the lytic lesion with collapse of the C4 vertebral body. MRIT1WI (B) and T2WI (C) series demonstrated the compression of the dural sac and cervical spinal cord and the focal invasion in the posterior aspect of vertebral body of C3 lesion.

A 56-year-old woman was admitted to our hospital with a one-month history of neck pain and right upper limbs numbness. Laboratory tests were unremarkable. A cervical spine radiograph showed a focal osteolytic lesion with collapse of the C4 vertebral body (A). Cervical MRI further demonstrated the compression of the dural sac and cervical spinal cord and the focal invasion of the adjacent intervertebral disc and the posterior aspect of vertebral body of C3 lesion (Figure 1). A tumor or tumor-like lesion was suspected and the patient underwent ^{18}F -FDG PET/CT scan.

Fluorine-18-FDG PET/CT showed a hypermetabolic lytic lesion with soft-tissue mass (SUVmax= 6.75) in the vertebral body and posterior elements of the C4, causing collapse of C4, invading also the adjacent intervertebral disc and the posterior aspect of vertebral body of C3, compressing the dural sac and cervical spinal cord (Figure 2). The findings of histopathological and immunohistochemical analysis supported a diagnosis of Langerhans cell histiocytosis.

Langerhans cell histiocytosis is a rare disease that originates from the uncontrolled proliferation and accumulation of Langerhans cells [1], which can affect patients at any age but is more likely to occur in those <15 years of age with prevalence in males [2, 3]. Spinal LCH accounted for <1% of spinal column tumors [4-6] and 6.5%-25% of all skeletal LCH cases, with a predilection for the thoracic spine followed by the lumbar and cervical spine [7]. Typical symptoms are circumscribed tenderness and restricted motion of the affected segments. Adult spinal LCH's most common radiographic features are solitary lytic lesions [8], which usually involved the vertebral body (82%), posterior elements alone (8%) and both anterior and posterior element involvement (10%) [9]. Endplate [10] and epidural [11] involvement have also been reported. However, intervertebral disc and adjacent vertebrae involvement are very rare signs [12, 13]. Preservation of intervertebral disc was considered to be helpful in differentiating LCH from tuberculous or nonspecific vertebral infection [14-17]. Spinal tumors involving adjacent vertebrae include osteosarcoma, chondrosarcoma, chordoma, aneurysmal bone cyst (ABC), giant cell tumor, Ewing sarcoma, lymphoma, myeloma in adults [18-21]. Among them, ABC, giant cell tumors and myeloma may extend through the intervertebral disc involved adjacent vertebrae [22, 23]. Thus, LCH should be listed in the differential diagnosis of lytic lesions involving adjacent vertebral and intervertebral discs, even in middle-aged and elderly patients.

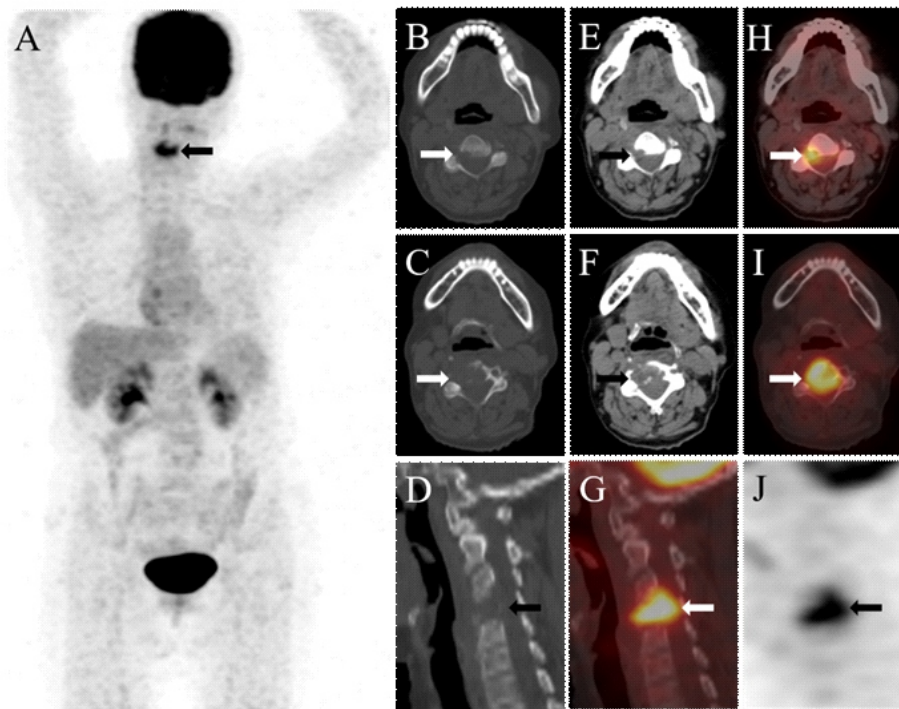


Figure 2. Fluorine-18-FDG PET/CT imaging: Maximum intensity projection (MIP) (A), axial and sagittal CT (B, C, D: bone window and E, F: soft tissue window), positron emission tomography/computed tomography (PET/CT) fusion (G, H, I) and PET (J) images revealed the osteolytic bone destruction of the C4 and the posterior aspect of the vertebral body of C3 with increased ^{18}F -FDG uptake.

Source of funding: The research was supported by the “1.3.5 Project for Disciplines of Excellence, West China Hospital, Sichuan University (ZYGD18016).”

Bibliography

- Vielgut I, Liegl-Atzwanger B, Bratschitsch G et al. Langerhans-cell histiocytosis of the cervical spine in an adult patient: Case report and review of the literature. *J Orthop* 2017; 14: 264-7.
- Islinger RB, Kuklo TR, Owens BD et al. Langerhans' cell histiocytosis in patients older than 21 years. *Clin Orthop Relat Res* 2000; 379: 231-5.
- Broadbent V, Egeler RM, Nesbit ME Jr. Langerhans cell histiocytosis-clinical and epidemiological aspects. *Br J Cancer Suppl* 1994; 23: S11-S16.
- Zheng W, Wu J, Wu Z et al. Atlantoaxial instability secondary to eosinophilic granuloma of the axis in adults: long-term follow-up in six cases. *Spine J* 2014; 14: 2701-9.
- Reddy PK, Vannemreddy PS, Nanda A. Eosinophilic granuloma of spine in adults: a case report and review of literature. *Spinal Cord* 2000; 38: 766-8.
- Sweasey TA, Dauser RC. Eosinophilic granuloma of the cervicothoracic junction. *Case report. J Neurosurg* 1989; 71: 942-4.
- Huang WD, Yang XH, Wu ZP et al. Langerhans cell histiocytosis of spine: a comparative study of clinical, imaging features, and diagnosis in children, adolescents, and adults. *Spine J* 2013; 13: 1108-17.
- Porn U, Howman-Giles R, Onikul E et al. Langerhans cell histiocytosis of the lumbar spine. *Clin Nucl Med* 2003; 28: 52-3.
- Prasad GL, Divya S. Eosinophilic Granuloma of the Cervical Spine in Adults: A Review. *World Neurosurg* 2019; 125: 301-11.
- Jiang L, Liu ZJ, Liu XG et al. Langerhans cell histiocytosis of the cervical spine: a single Chinese institution experience with thirty cases. *Spine (Phila Pa 1976)* 2010; 35: E8-E15.
- Lim CS, Cho JH. Spinal epidural involvement in adult Langerhans cell histiocytosis (LCH): A case report. *Medicine (Baltimore)* 2020; 99: e18794.
- Özdemir ZM, Kahraman AS, Görmeli CA et al. Langerhans Cell Histiocytosis with Atypical Intervertebral Disc and Sacroiliac Joint Involvement Mimicking Osteoarticular Tuberculosis in an Adult. *Balkan Med J* 2016; 33: 573-7.
- Poulsen JO, Thommesen P. An unusual case of histiocytosis X in the spine. *Acta Orthop Scand* 1976; 47: 59-62.
- Azouz EM, Saigal G, Rodriguez MM et al. Langerhans' cell histiocytosis: pathology, imaging and treatment of skeletal involvement. *Pediatr Radiol* 2005; 35: 103-15.
- Mandegar R, Tang CSW, Pereira EAC et al. Spondylodiscitis following endovascular abdominal aortic aneurysm repair: imaging perspectives from a single centre's experience. *Skeletal Radiol* 2018; 47: 1357-69.
- Bassetti M, Merelli M, Di Gregorio F et al. Higher fluorine-18 fluorodeoxyglucose positron emission tomography (^{18}F -FDG-PET) uptake in tuberculous compared to bacterial spondylodiscitis. *Skeletal Radiol* 2017; 46: 777-83.
- Nakamura J, Yamada K, Mitsugi N et al. A case of SAPHO syndrome with destructive spondylodiscitis suspicious of tuberculous spondylitis. *Mod Rheumatol* 2010; 20: 93-7.
- Rodallec MH, Feydy A, Larousserie F et al. Diagnostic imaging of solitary tumors of the spine: what to do and say. *Radiographics* 2008; 28: 1019-41.
- Orguc S, Arkun R. Primary tumors of the spine. *Semin Musculoskelet Radiol* 2014; 18: 280-99.
- Flemming DJ, Murphey MD, Carmichael BB et al. Primary tumors of the spine. *Semin Musculoskelet Radiol* 2000; 4: 299-320.
- Theodorou DJ, Theodorou SJ, Sartoris DJ. An imaging overview of primary tumors of the spine: Part 1. Benign tumors. *Clin Imaging* 2008; 32: 196-203.
- Parmar HN, Agrawal VA, Shah MS et al. Locally aggressive aneurysmal bone cyst of C4 vertebra treated by total en bloc excision and anterior plus posterior cervical instrumentation. *J Craniovertebr Junction Spine* 2015; 6: 130-3.
- Jiang G, Sun LL, Ye YJ et al. Giant cell tumors of the mobile spine with invasion of adjacent vertebrae: an unusual imaging finding. *BMC Musculoskelet Disord* 2021; 22: 726.

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