Langerhans Cell Histiocytosis in Cervical Vertebra 4 in an Obese Man

Sir,

Langerhans cell histiocytosis (LCH), a rare disease, is characterised by the pathological proliferation and accumulation of Langerhans cells in various tissues. This disease often affects the skeleton, with spinal involvement accounting for 6.5-25%, among which, 11% affects the cervical spine.[1] The clinical manifestations of patients with LCH usually include neck pain, limited mobility, and neurological deficits.[2-4] Osteolytic lesions are the main imaging manifestations. In addition, osteogenic lesions can also be manifested.[2]

Various non-surgical treatments can be used for spinal (LCH), such as activity restriction, non-steroidal anti-inflammatory drugs (NSAIDs), close observation, plaster fixation, steroid hormones, radiotherapy, chemotherapy, etc. These methods are more commonly used, especially in spinal and systemic diseases.[1,2,4,5] For cases of structural instability caused by compression, neurological deficits, anatomical deformities, early lesion clearance surgery, inter-vertebral fixation, and vertebral fusion should be performed.[1,2]

A 32-year male patient was found to have LCH in the fourth cervical vertebra. He was obese and was admitted to the hospital due to neck pain accompanied by limited mobility and treated in the hospital for 16 days.

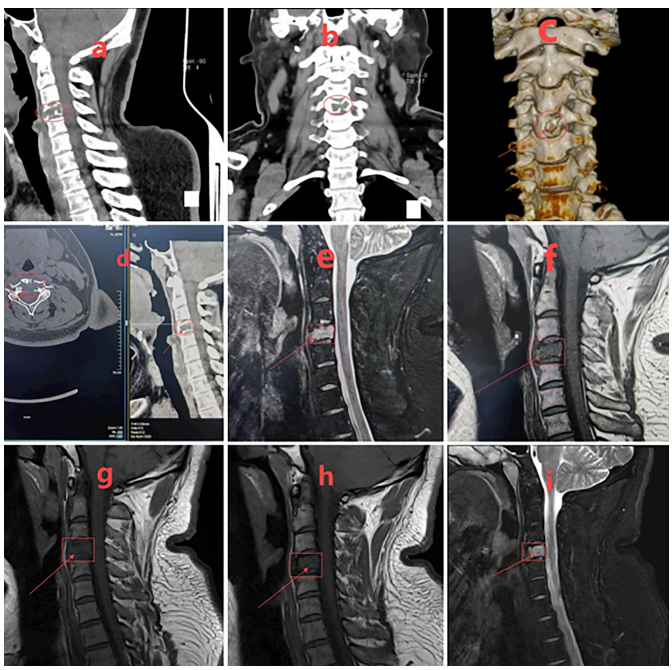

Figure 1: (a-d) Computed tomography (CT) scan and three-dimensional reconstruction suggested destruction and space occupying lesions in the fourth cervical vertebra. (e-i) Magnetic resonance imaging (MRI) examination showed abnormal signals in the fourth cervical vertebra.

The patient complained of neck discomfort without obvious cause five years ago, which was temporarily relieved by oral analgesics. Computed tomography (CT) scan and three-dimensional reconstruction suggested destruction and space-occupying lesions in the fourth cervical vertebra (Figure 1a-d). In order to further confirm it, a magnetic resonance imaging (MRI) examination was carried out. The results showed abnormal signals in the fourth cervical vertebra, which suggested bone destruction (Figure 1e-i).

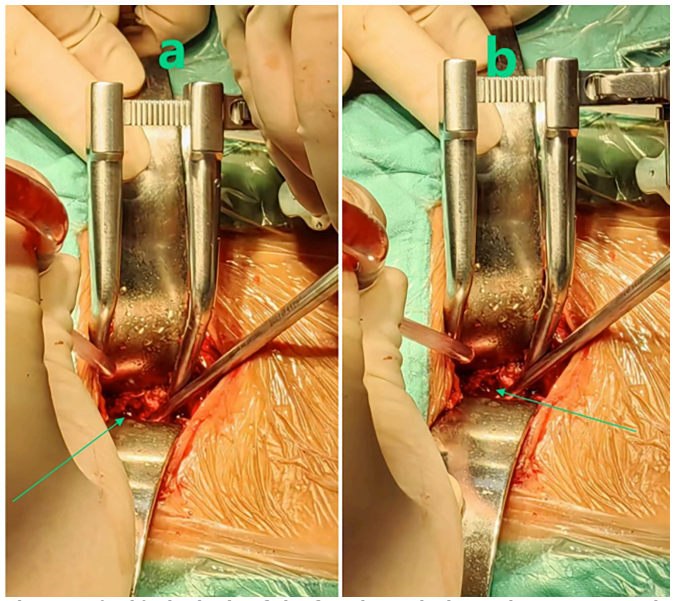

Figure 2: (a, b) The body of the fourth cervical vertebra was severely damaged and had cavity formation, without the invasion of surrounding structures.

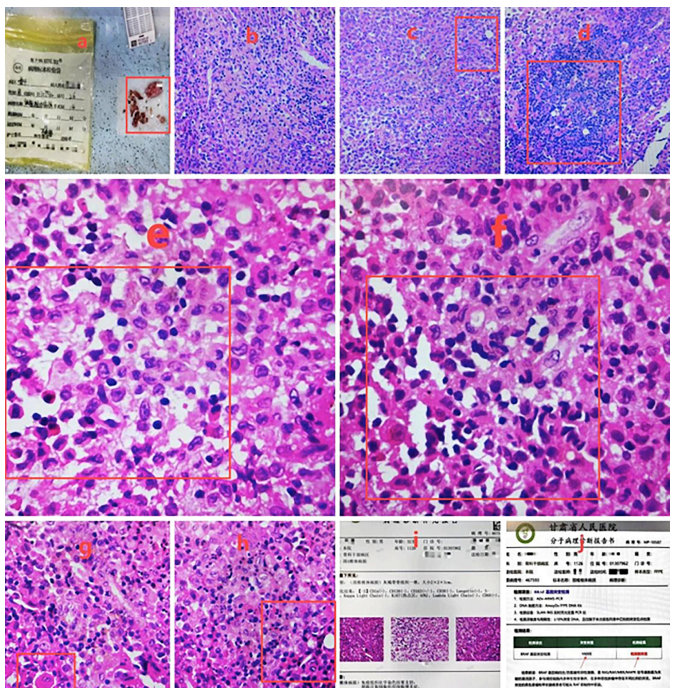

Figure 3: (a-i) A histopathological examination post-surgery yielded a diagnosis of langerhans cell histiocytosis (LCH). (j) Molecular testing for BRAF gene mutations identified the V600E mutation type.

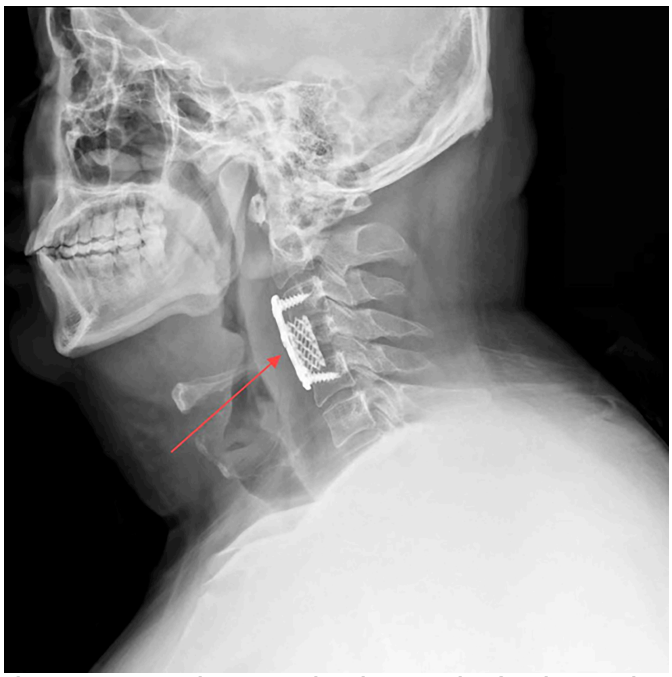

Figure 4: Postoperative x-rays taken three months after the procedure demonstrated proper positioning of the hardware without evidence of new bone destruction.

After admission, the patient completed relevant examinations and tests and underwent surgical treatment under general anaesthesia. The lesion of the fourth cervical vertebra was successfully removed. During the operation, it was observed that the body of the fourth cervical vertebra was severely damaged and had cavity formation, without the invasion of surrounding structures (Figure 2a, b). The necrotic bone was meticulously excised using an ultrasonic osteotome, followed by the removal of residual necrotic tissue with an orthopaedic curette. Afterwards, the surgical site was thoroughly irrigated with saline and hydrogen peroxide solution. Subsequently, a titanium cage was implanted into the defect of the fourth cervical vertebra and secured with a cervical locking plate to ensure the stability of the cervical spine. A histopathological examination post-surgery yielded a diagnosis of LCH (Figure 3a-i). Immunohistochemical staining results were as follows: CD1a (positive), CD138 (negative), D163 (equivocal), CD38 (negative), Langerin (positive), S-100 (positive), Kappa light chain (negative), Ki-67 (hot spot: 40%), Lambda light chain (negative), and CD68 (positive), all of which were consistent with LCH. Molecular testing for *BRAF* gene mutations identified the V600E mutation type (Figure 3j). Postoperative x-rays taken three months after the procedure demonstrated proper positioning of the hardware without evidence of new bone destruction (Figure 4). Three months after surgery, the patient's symptoms gradually disappeared and the cervical spine moved well.

COMPETING INTEREST:
The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:
YZ: Drafted, revised, and edited the manuscript.
ZZ, WX: Conducted data collection, analysis, and interpretation.
CZ: Performed data collection.
All authors approved the final version of the manuscript to be published.

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