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CASE REPORT

Serous degeneration of bone marrow mimics spinal tumor

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Abstract
Objective To present a rare case of serous degeneration of bone marrow which resembles primary spinal tumor or bony metastasis to spine.
Summary of background data Serous degeneration of bone marrow or gelatinous marrow transformation is a rare disease characterized by focal marrow hypoplasia, fat atrophy, and accumulation of extracellular mucopolysaccharides abundant in hyaluronic acid. Few literature was reviewed and few clinical case was presented.
Methods Two cases of serous marrow transformation were reported.
Results In the first case, a 29-year-old man suffered from severe left buttock pain. Bone metastasis was impressed in radiology examinations. Percutaneous endoscopic lumbar discectomy was performed along with bone biopsy. In the second case, a 49-year-old man presented lower back pain with radiation to bilateral lower legs. Magnetic resonance imaging revealed a water-like signal lesion in sacrum. Serous marrow transformation was confirmed pathologically in both cases.
Conclusion To the best of our knowledge, a case of serous degeneration of bone marrow resembling malignancy has not been reported in the literature. In this report, two cases demonstrate serous transformation of bone marrow mimics spinal tumor.

Keywords Serous degeneration · Serous atrophy · Gelatinous marrow transformation · Magnetic resonance imaging

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Introduction

Serous degeneration, also known as gelatinous marrow transformation (GMT) [1], is a rare disease characterized by fat cell atrophy, focal marrow hypoplasia and accumulation of extracellular gelatinous material which is identified as hyaluronic acid mucopolysaccharides [2, 3]. Serous degeneration of bone marrow is not specific for a disease, but represents clinical consequence of generalized severe illnesses. Although causes of this transformation remain unclear, serous degeneration has been reported in several chronic diseases, including prolonged starvation, anorexia nervosa [4], malabsorption, alcoholism, malignancy and infectious diseases such as tuberculosis, acquired immune deficiency syndrome

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(AIDS) [5]. Clinically, few report describes serous degeneration in spine.

Case 1

A 29-year-old male, with a history of gout, was referred to our department of Orthopedics to evaluate a severe left buttock pain with radiation to posterior thigh during the last 1 month. He did not present any history of fever, loss of weight, trauma, and previous infection. His physical examination showed low back pain of lumbar spine level, intermittent soreness of left hip and lateral region of left thigh, and numbness of left heel. Laboratory examinations revealed no significant findings including negative human immunodeficiency virus. A radiograph showed narrowing of multiple intervertebral disc spaces (Fig. 1a: lateral view). Magnetic resonance image (MRI) scan revealed herniated disc at L5–S1 level as well as multiple space-occupying lesions at L1, L2, L3 and L4 vertebral bodies with water-like signal intensity [Fig. 1b: T1-weighted, c: T2-weighted, d: short tau inversion recovery (STIR) images]. The tentative diagnosis of herniated intervertebral disc with suspicious multiple myeloma or multiple hemangiomas was made. The patient received percutaneous endoscopic lumbar discectomy (PELD) at L5/S1. Biopsy of the L5 bone lesion was also performed. On microscopy, there were serious degeneration of bone marrow with marked reduction in hematopoietic, fat cells and hypocellular marrow as regard to patient's age (Fig. 1e, f). A diagnosis of serious degeneration of bone marrow was confirmed. After surgical intervention, he regularly followed up for 2 years.

Case 2

A 49-year-old male, a heavy smoker and drinker, with medical history of hypertension and hepatitis B, presented lower back pain with radiation to bilateral lower legs 2 months ago. On physical examination, local tenderness over the lower back region was noted and straight leg raising test (SLRT) was positive. He was managed conservatively for 2 months with painkillers and rest;

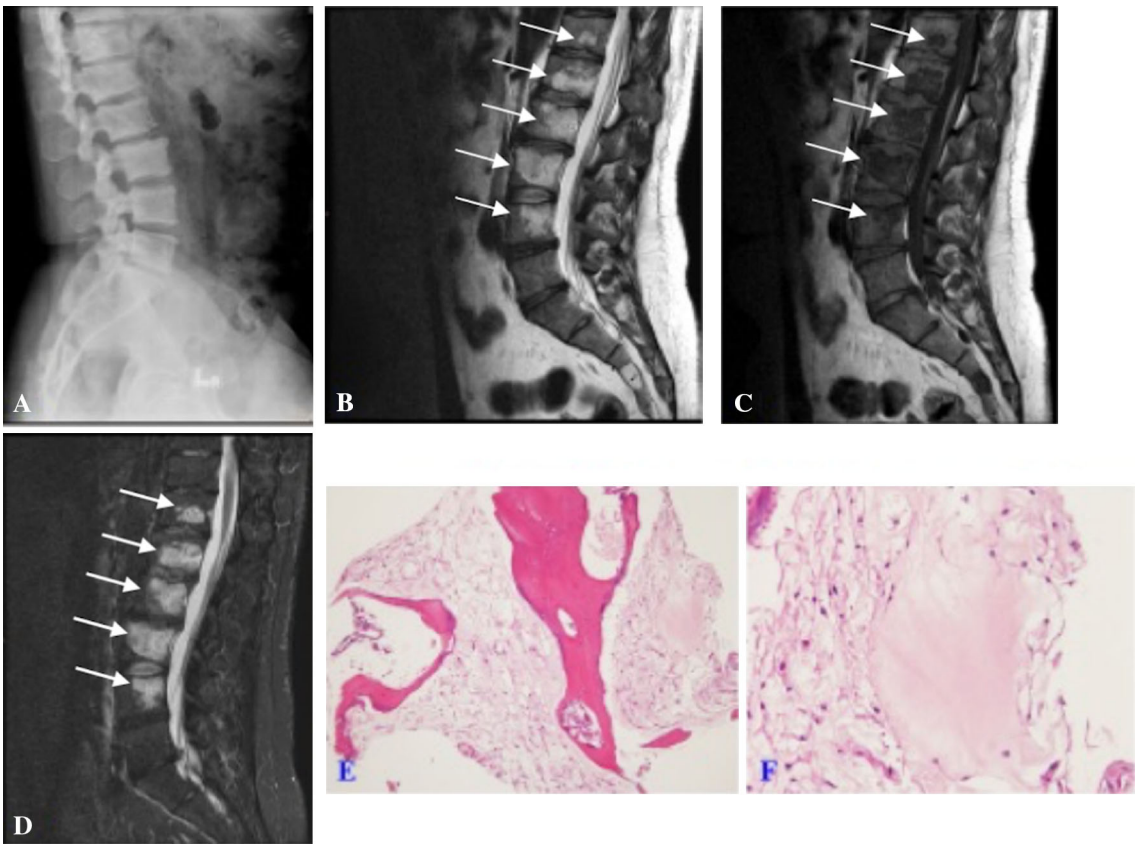

Fig. 1 Radiographic examinations of the 29-year-old man. **a** Radiography shows decreased intervertebral disc height at the L5–S1 level. No osteolytic lesion can be found. MRI examination revealed multiple space-occupying lesions (*arrows*) with low signal intensity in T1-weighted image (**b**), high signal intensity in both T2-weighted (**c**) and short tau inversion recovery (STIR) images (**d**). Loose marrow space with serous degeneration (**e**, **f**)

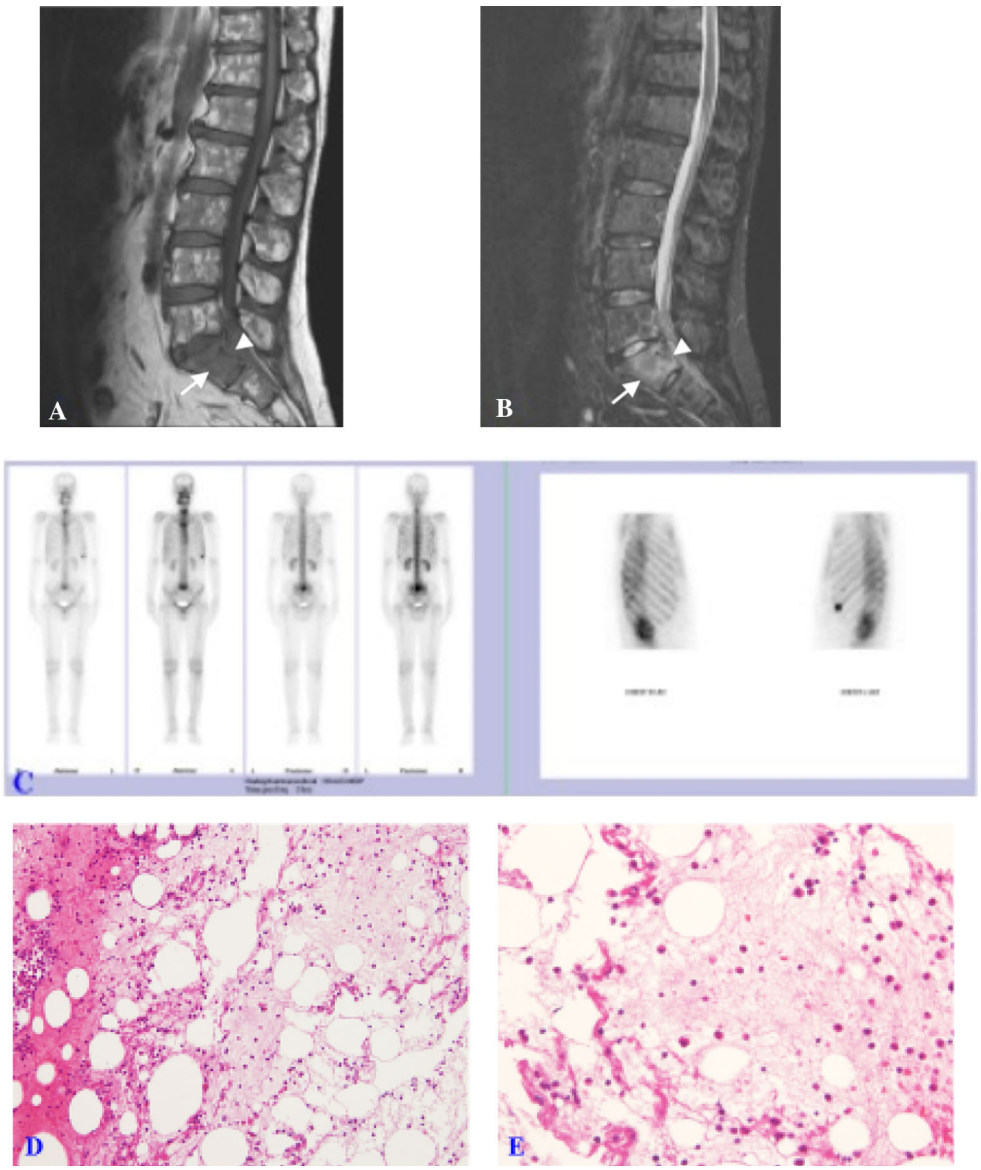

Fig. 2 MRI examination of the 49-year-old man. One amorphous space-occupying lesion (*arrows*) with low signal intensity in T1-weighted image (**a**) and high signal intensity in short tau inversion recovery (STIR) images (**b**) is noted at the superior sacrum. Cortical break-down is noted at the posterior vertebral body (*arrowheads*). **c** Bone scan. Hemorrhage with degeneration change in the loose marrow space (**d**, **e**)

however, there was no symptomatic improvement. MRI scan revealed amorphous lesion with heterogeneous high T2 and low T1 signal lesion located in upper sacrum with cortical destruction at the posterior border of the S1 body (Fig. 2a: T1-weighted, b: STIR images). Bone scan also demonstrated hot focus at S1/S2 (Fig. 2c), favoring malignant bone metastasis, primary bone tumor or osteomyelitis. Bone biopsy was thereby performed. Microscopically, the specimen shows fragmented trabecular bone with no malignancy in hematoxylin and eosin (H&E) sections. Focal hemorrhage with fibrin substance change are also seen. This degeneration change in the loose marrow further confirms no malignancy (Fig. 2d, e). A diagnosis of serous degeneration was made. After discharge, he did not complain for 1 year.

Discussion

This is the first case report describing serous degeneration of bone marrow which mimics spinal tumor in lumbar spine. For both of our cases, secondary malignant neoplasm of bone marrow was first impressed by radiograph. Pathological evidence, however, reveals no

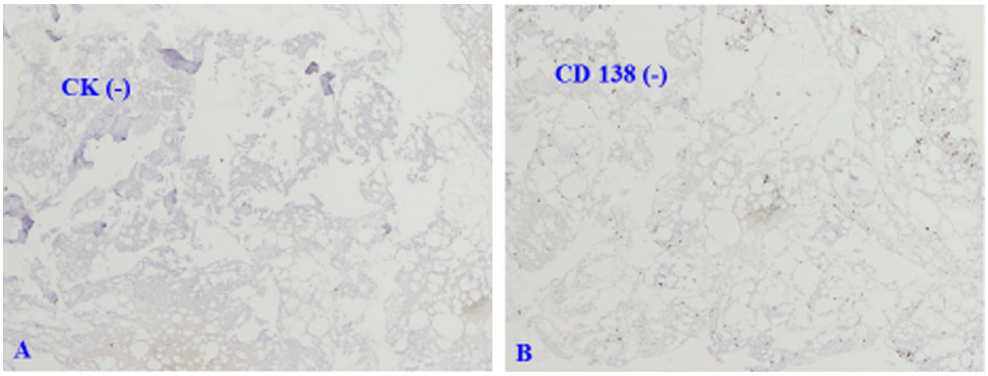

Fig. 3 Immunohistochemical stain was negative for cytokeratin (CK), and specific increased in plasma cell by immunostaining for CD138

malignancy but a picture of serous degeneration of bone marrow.

Serous degeneration of bone marrow is a pattern of injury and develops in different pathologic conditions [3]. It is most often associated with cachexia secondary to chronic diseases like tuberculosis, malignancy [6, 7], leukemia post chemotherapy, acute febrile illnesses, systemic lupus erythematosus, chronic renal failure, severe hypothyroidism and states of chronic nutritional deficiency such as starvation, and anorexia nervosa [4]. However, these symptoms did not occur in our cases. The etiology of our cases is idiopathic. Additionally, although Bohm [3] found there was a slight peak of GMT change in the age group of 20- to 29-year-olds, serous degeneration is seldom seen in spine in young population. Notably, the ages of our two cases are 29 and 49 years old, respectively. This 29-year-old man was the youngest patient who was diagnosed as serous degeneration and was first reported. It may be hardly diagnosed if no pathological evidence.

The serous degeneration usually starts in peripheral skeleton and eventually reaches the axial skeleton [8]. These pathologic changes can be recognized on MR as fat-poor marrow, which emits water-like signal on all pulse sequences, i.e., low on T1W and high on T2W and STIR sequences [9–11]. No enhancement could be identified after administration of gadolinium contrast medium, which helps to distinguish it from malignant or infectious diseases.

The primary pathological finding in the bone marrow is stromal edema with associated micro-vesicular change. The bone marrow is hypocellular in these regions due to loss of normal hematopoietic elements. It is important to recognize that the subcortical bone marrow is normally hypocellular and occasionally demonstrates edema. Immunohistochemically, the specimen shows negative for cytokeratin (AE1/AE3) and not specific increased immunostaining for CD138 (Fig. 3a: cytokeratin, b: CD138), indicating that there is no malignancy in both cases.

Additionally, based on the primary pathological finding in the affected bone marrow, the pattern is stromal edema with micro-vesicular change. The bone marrow is hypocellular in these regions due to loss of normal hematopoietic elements. Since diffusion weighted imaging (DWI) and its apparent diffusion coefficient (ADC) signal was proved to correlate inversely with cellularity in some researches, it implied that DWI may be helpful to differentiate serous transformation from malignant tumors with high cellularity. However, it lacks enough evidence to prove the hypothesis in this report.

In conclusion, these two cases demonstrate serous transformation of bone marrow in lumbar spine. A comprehensive understanding of the disease entity with careful interpretation of MR findings is critical to prevent the misdiagnosis of it as malignant or infectious diseases.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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