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Numerous sites of increased uptake shown on bone scintigraphy in a case of adult T-cell leukemia

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Bone scintigraphy was performed in a 69-year-old male patient with adult T-cell leukemia suffering from right lower limb pain. Numerous sites of increased uptake were seen in the skull, left clavicle, bilateral humeri, bilateral radii and right femur and tibia. Bone radiographs showed multiple osteolytic lesions, most of which corresponded to the abnormal deposits on the bone scans with 740 MBq of ^{99m}Tc-hydroxymethylene diphosphonate. This pattern is rarely reported, but bone involvement of adult T-cell leukemia is not uncommon. Bone involvement was remarkable on the appendicular skeleton when compared with common metastatic bone tumors. Bone scintigraphy may be useful in detecting bone involvement in adult T-cell leukemia.

Key words: adult T-cell leukemia, ^{99m}Tc-hydroxymethylenediphosphonate, bone scintigraphy

INTRODUCTION

ADULT T-cell leukemia caused by human T-cell lymphotropic virus type I (HTLV-I) has a lot of manifestations such as lymphadenopathy, hepatomegaly, splenomegaly, skin lesion and hypercalcemia.[1–3] Although bone involvement is not uncommon,[4–6] bone scintigraphy is rarely reported.[4–7] We report on bone scintigraphy in a case of adult T-cell leukemia.

CASE REPORT

A 69-year-old male patient was admitted to Kagoshima University hospital because of generalized eruptions for 13 months. He had also suffered from a right lower limb pain for two months. A diagnosis of adult T-cell leukemia was made because anti-HTLV-I antibodies were positive in his peripheral blood and monoclonal proviral DNA integrations in leukemic cells obtained from the eruption were demonstrated. Bone scintigraphy was performed with 740 MBq of ^{99m}Tc-hydroxymethylenediphosphonate (^{99m}Tc-HMDP) because of his right lower limb pain. Four hours after the intravenous injection of the tracer, whole body anterior and posterior images were obtained by means of a two-head gamma camera (PRISM 2000, Picker-Shimadzu, Japan). The bone scans showed numerous sites of increased uptake in the skull, left clavicle, ribs, bilateral humeri, bilateral radii and right femur and tibia (Fig. 1). Most of them corresponded to the osteolytic lesions demonstrated in the bone radiographs (Figs. 2 and 3). At the time of bone scintigraphy, the level of serum calcium was 8.8 mg/m*l* (normal value: 8.6–10.2) and that of parathyroid hormone-related protein was 1.1 pg/m*l* (normal value: < 1.1). The serum lactic dehydrogenase (LDH) value was within normal limits and the leukocyte count in the peripheral blood was 3300/μl, with 3% atypical lymphocytes. Bone biopsies of the osteolytic lesions in the right tibia and the right humerus failed to reveal infiltration of atypical leukemic cells. The level of serum calcium rose to 10.6 mg/m*l* 6 months after bone scintigraphy was performed.

DISCUSSION

Takatsuki et al. first described the clinical features of adult T-cell leukemia.[1] Uchiyama et al. report a lot of manifestations of this disease, such as lymphadenopathy, hepatomegaly, splenomegaly and skin lesion.[2] Hypercalcemia is also a common finding.[4,7–9] The frequency of

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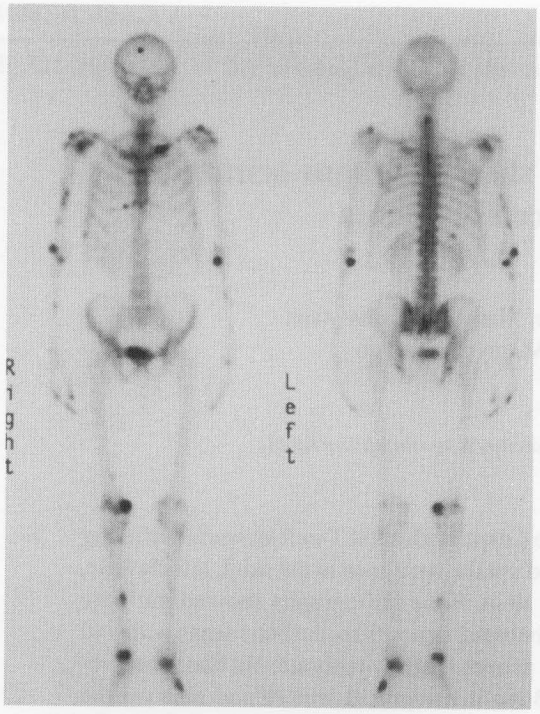


Fig. 1 Whole body bone scans showed numerous sites of increased uptake on the skull, left clavicle, ribs, bilateral humeri, bilateral radii and right femur and tibia.

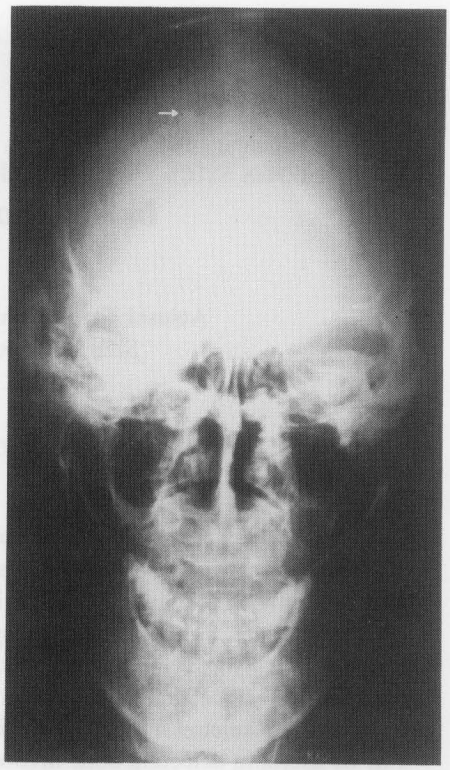


Fig. 2 The radiograph of the skull showed a round and well-defined osteolytic lesion on the parietal bone (arrow). Marginal sclerosis was not seen.

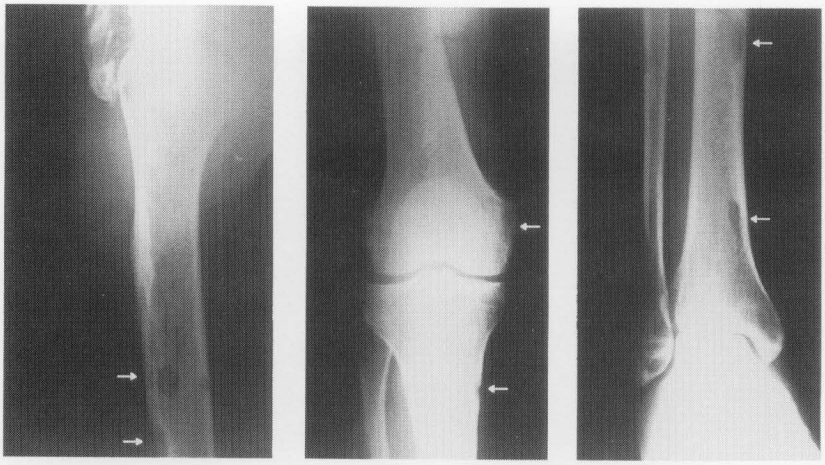
bone involvement is reported in 17–36% of cases.[7,8]

Some authors reported bone scintigraphy in adult T-cell leukemia.[4–7] Hoshi et al. performed bone scintigraphy in 26 cases and reported that abnormal findings were hot spots including one or several lesions and that there were none with diffuse abnormal uptake in the skeleton.[5] The positive rate was reported to be 27% of the cases (7/26).[5] George et al. reported that the radionuclide bone scan was normal on the skull with multiple osteolytic lesions and that large osteolytic areas affecting the proximal tibiae and fibulae showed no increased uptake on radionuclide bone scans except at the site of a pathological fracture.[4] Ohuchida et al. reported that a bone scan did not show accumulation in the skull radiography of which revealed multiple osteolytic lesions and diffuse bone demineralization.[6] Bunn et al. reported that radionuclide bone scans showed a generalized increased uptake of the tracer that was symmetrically prominent in the joints and the skull in all patients who were scanned.[7] In our case, most of the sites of increased uptake on the bone scans were consistent with the osteolytic lesions on the bone radiographs. Hoshi et al. reported this pattern in adult T-cell leukemia,[5] but it seems to be uncommon.

Regarding the mechanism of bone involvement in adult T-cell leukemia, some investigators suggest that the actions of substances stimulating osteoclastic bone resorption may affect bone involvement in adult T-cell leukemia.[5,6,8,10] Motokura et al. reported that leukemic cells produced parathyroid hormone-related protein which stimulates osteoclastic bone resorption and might be an important factor in the development of hypercalcemia,[11] but Bunn et al. reported that one bone biopsy sample showed a malignant T-cell infiltrate,[7] and an osteoclastic mechanism may explain the bone involvement in adult T-cell leukemia in most cases.

In our case, bone biopsies of the osteolytic lesions in the right tibia and the right humerus showed no evidence of infiltration of atypical leukemic cells, nor was the result of bone scintigraphy suggestive of metabolic bone disease. But, slight metabolic change might have been latent when the bone scintigraphy was performed because hypercalcemia 6 months after that may be mainly due to the progress of the disease. It needs further investigation to explain this phenomenon.

The sites of metastatic bone tumors are usually seen in the axial skeleton.[12] In adult T-cell leukemia, George et al. reported that lytic bone lesions were identified in both axial and appendicular skeletons.[4] Involvement of the appendicular skeleton was remarkable in our case. The distribution of bone lesions in adult T-cell leukemia seems to be different from that of common metastatic bone tumors. This may be due to the production of substances stimulating osteoclastic bone resorption by leukemic cells.[10,11] Further studies are needed to clarify this.



(a) (b) (c)

Fig. 3 Radiographs of the right humerus (a), right knee (b) and left leg (c) showed multiple osteolytic lesions without sclerotic rim (arrows).

CONCLUSION

We reported a case of adult T-cell leukemia showing numerous sites of increased uptake on bone scintigraphy without hypercalcemia. Bone involvement was remarkable on the appendicular skeleton when compared with common metastatic bone tumors. Bone scintigraphy may be useful in detecting bone involvement in adult T-cell leukemia.

REFERENCES

1. Takatsuki K, Uchiyama T, Sagawa K, Yodoi J. Adult T-cell leukemia in Japan. *In* Topics in Haematology, Seno S, Takaku F, Irino S (eds.), Amsterdam, Excerpta Medica, pp. 73–77, 1977.
2. Uchiyama T, Yodoi J, Sagawa K, Takatsuki K, Uchino H. Adult T-cell leukemia: clinical and hematologic features of 16 cases. *Blood* 50: 481–492, 1977.
3. Kawano F, Yamaguchi K, Nishimura H, Tsuda H, Takatsuki K. Variation in the clinical courses of adult T-cell leukemia. *Cancer* 55: 851–856, 1985.
4. George CD, Wilson AG, Philpott NJ, Bevan DH. The radiological features of adult T-cell leukemia/lymphoma. *Clin Radiol* 49: 83–88, 1994.
5. Hoshi H, Nagamachi S, Jinnouchi S, Ohnishi T, Shigemi F, Watanabe K. Bone scintigraphy as a prognostic factor in patients with adult T-cell leukemia-lymphoma. *Clin Nucl Med* 19: 992–995, 1994.
6. Ohuchida T, Nishitani H, Kamikawaji N, Niho Y, Ooiwa T, Matsuura K. "Adult T-cell leukemia/lymphoma" with bone demineralization. *Skeletal Radiol* 14: 194–197, 1985.
7. Bunn PA, Schechter GP, Jaffe E, Blayney D, Young RC, Matthews MJ, et al. Clinical course of retrovirus-associated adult T-cell lymphoma in the United States. *N Engl J Med* 309: 257–264, 1983.
8. Levine PH, Jaffe ES, Manns A, Murphy EL, Clark J, Blattner WA. Human T-cell lymphotropic virus type I and adult T-cell leukemia/lymphoma outside Japan and the Caribbean basin. *Yale Journal of Biology and Medicine* 61: 215–222, 1988.
9. Kiyokawa T, Yamaguchi K, Takeya M, Takahashi K, Watanabe T, Matsumoto T, et al. Hypercalcemia and osteoclast proliferation in adult T-cell leukemia. *Cancer* 59: 1187–1191, 1987.
10. Fukumoto S, Matsumoto T, Ikeda K, Yamashita T, Watanabe T, Yamaguchi K, et al. Clinical evaluation of calcium metabolism in adult T-cell leukemia. *Arch Intern Med* 148: 921–925, 1988.
11. Motokura T, Fukumoto S, Matsumoto T, Takahashi S, Fujita A, Yamashita T, et al. Parathyroid hormone-related protein in adult T-cell leukemia-lymphoma. *Ann Intern Med* 111: 484–488, 1989.
12. Rogers LF. Malignant bone tumors. *In* Paul and Juhl's Essentials of Radiologic Imaging, Juhl JH, Crummy AB (eds.), 6th ed., Philadelphia, J.B. Lippincott Co., pp. 164–185, 1993.