involving multiple bones, without visceral or lymph-node involvement [6]. Recently, several clinical reviews applying these criteria of PBL have been published [4, 5, 7–12]. There was a slight male predominance and a tendency for senior adults to be affected more often, although PBL can occur at any age. The pelvis or proximal part of the femur is the common site of the tumor. Most cases of PBL are reported to be non-Hodgkin's diffuse large B-cell lymphomas. Stage of disease, age, serum LDH levels, response to chemotherapy, and the use of combined modality therapy are considered to be prognostic factors in PBL. Radiographs and CT images of PBL usually present non-specific findings, mostly various patterns of osteolysis and osteosclerosis coexisting in the bone. As a result, radiological differential diagnosis of PBL from other types of primary bone tumors such as Ewing's sarcoma, osteogenic sarcoma, and chondrosarcoma is often difficult. Therefore, laboratory findings that help differentiate PBL from other common bone tumors are important.

So far, the significance of measuring sIL-2R levels has been rarely reported in clinical reviews of PBL [4, 5, 7–12]. In other primary malignant bone tumors including osteosarcoma, Ewing's sarcoma, chondrosarcoma, and malignant giant cell tumor, median serum levels of sIL-2R are reported not to be significantly elevated compared with those of healthy controls [1, 2]. Recently, Akahane et al. [3] reported that the serum sIL-2R level is valuable for differentiating PBL from other osteolytic malignancies or benign bone lesions that resemble malignant tumor radiographically.

In our two PBL cases, the reduced sIL-2R levels after R-CHOP therapy were linked with the reduction of the tumor size and the osteosclerotic changes in the bone lesions. Moreover, the changes in sIL-2R levels were similar to those in LDH levels known as a prognostic factor of PBL [8]. The fact that sIL-2R levels were associated with the clinical course of PBL means such measurements play an important role in the disease monitoring. Adding to the recently reported results of the diagnostic significance of sIL-2R in PBL, our case study tentatively indicates the clinical significance of sIL-2R levels as a tumor marker in PBL cases.

Disclosure Statement

All authors have no conflict of interest to declare.

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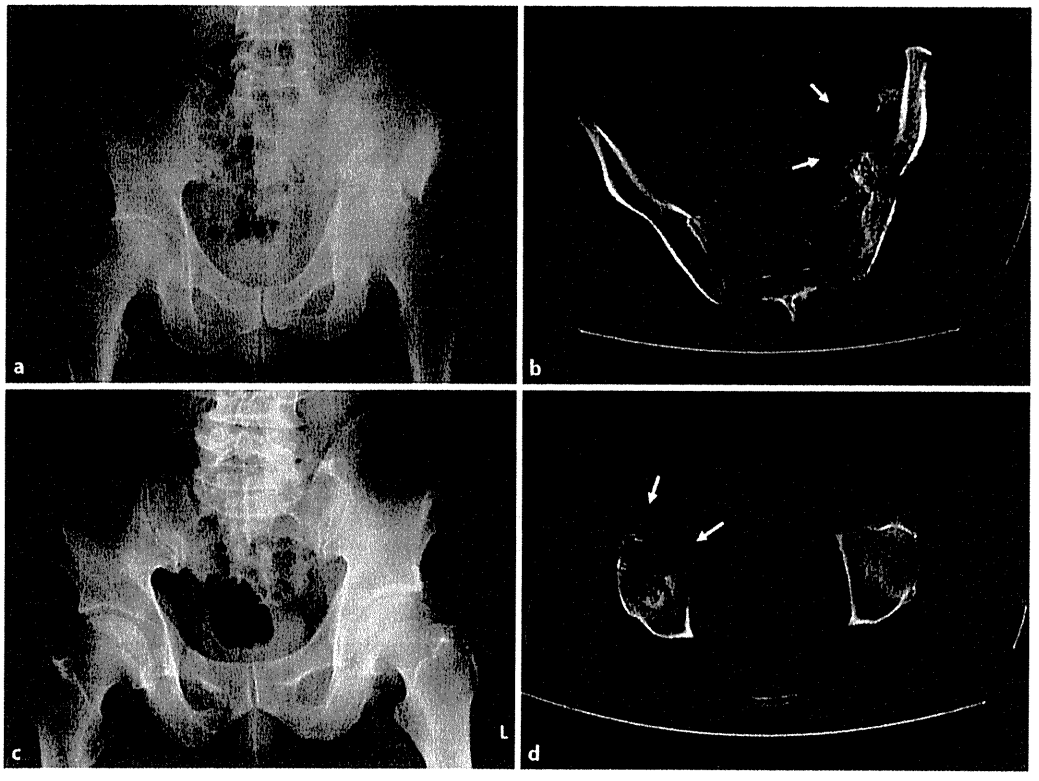

Fig. 1. Radiographs and CT images at hospitalization. **a** Radiograph from June 2007. Osteosclerotic and osteolytic changes in the left iliac wing and a pathological fracture of the left acetabulum were observed in case 1. **b** CT imaging from June 2007. Expanded soft tissue around the left ileum can be recognized (arrows) in case 1. **c** In September 2006, radiography showed an osteolytic change in the right acetabulum in case 2. **d** CT image from September 2006 shows a moth-eaten pattern of destruction of the anterior part of the right acetabulum and expanded soft tissue (arrows) in case 2.

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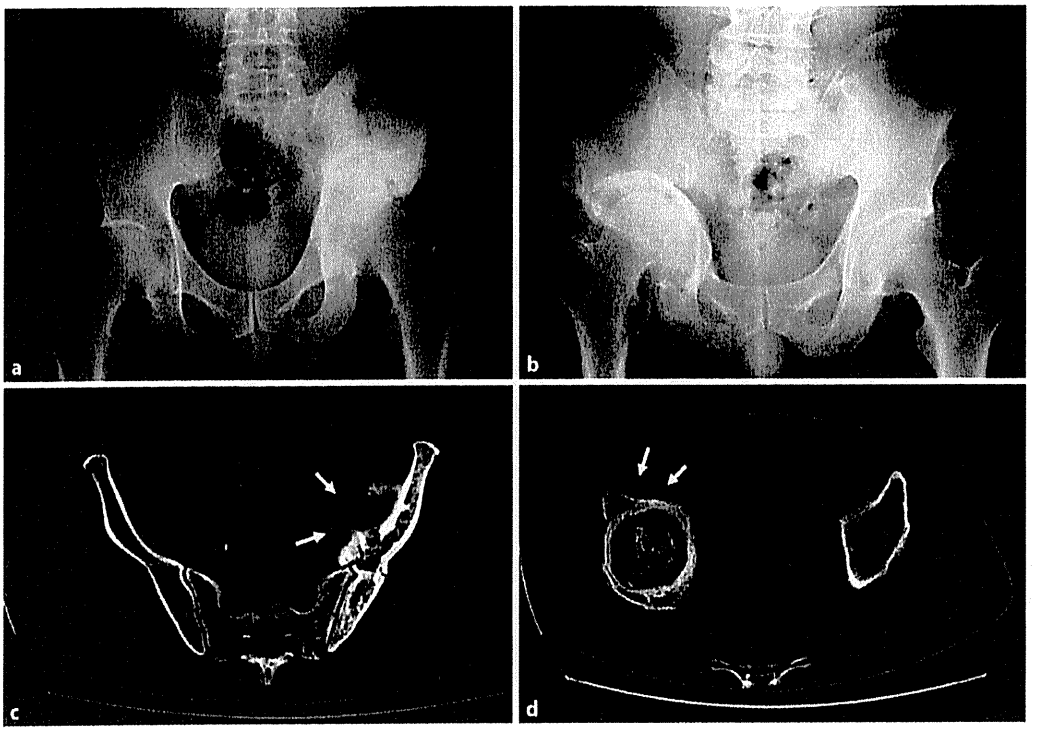

Fig. 2. Radiographs and CT images after chemotherapy (R-CHOP). **a** In March 2008, radiography showed an increased osteosclerotic change and an obscure fracture line in case 1. **b** In February 2007, radiography showed new bone formation and increased osteosclerosis around the acetabulum as well as intrapelvic migration of the femur head in case 2. **c** In March 2008, CT imaging revealed increased osteosclerosis and the necrotic soft tissue mass around the affected bone lesion (arrows) in case 1. **d** In February 2007, CT imaging revealed the necrotic soft tissue mass around the affected bone lesion (arrows) in case 2.

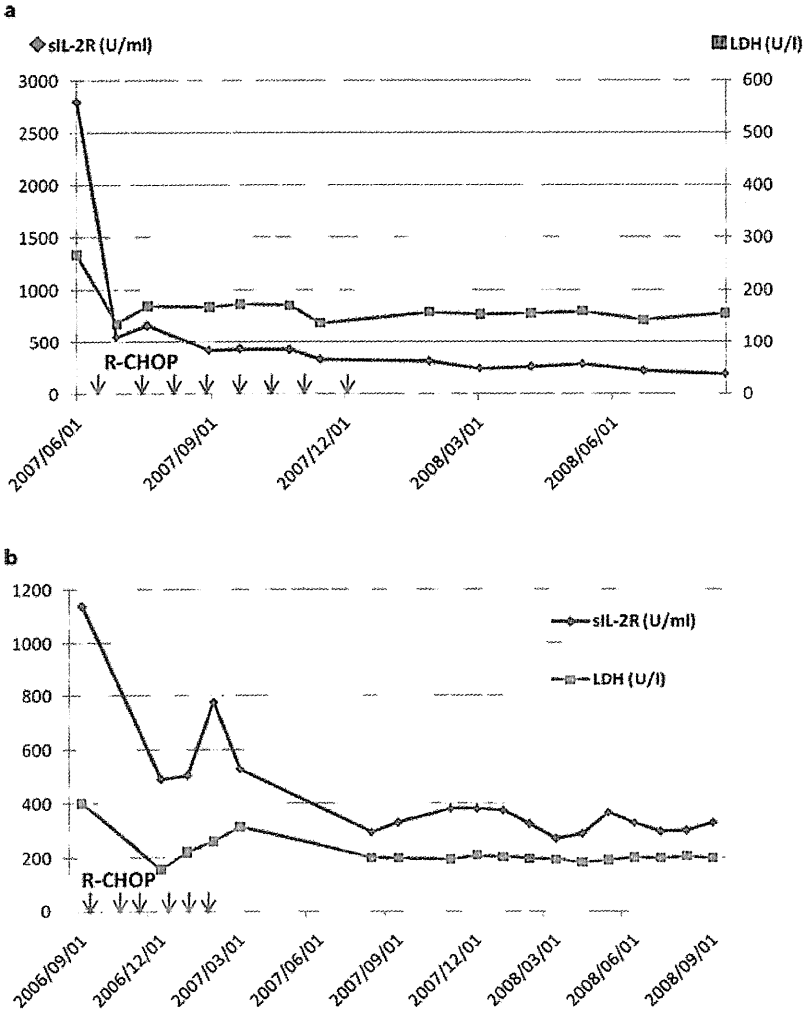

Fig. 3. Serum sIL-2R levels of our two PBL cases normalized after R-CHOP therapy. **a** Case 1, **b** case 2.

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Case Reports in Oncology

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Primary Primitive Neuroectodermal Tumor of the Conus Medullaris in an Elderly Patient: A Case Report and Review of the Literature

H. Shimosawa M. Matsumoto H. Yabe M. Mukai
Y. Toyama H. Morioka

Department of Orthopaedic Surgery, School of Medicine, Keio University, Tokyo, Japan

Key Words
Primitive neuroectodermal tumor · Ewing's sarcoma family of tumors · Conus medullaris · Elderly patient

Abstract
Primary spinal primitive neuroectodermal tumors (PNETs) are very rare conditions. Most of these tumors occur in children and young adults. A 63-year-old man with a primary spinal PNET in the conus medullaris from the L1 to L2 level is presented in this report. The optimal treatment of primary spinal PNETs is yet unknown. Surgical resection, radiation therapy, and chemotherapy have been advocated for the treatment of spinal PNET based on PNETs at other sites. However, the outcome is very poor. There are a few reports of cases with long-term survival and no recurrence. In these patients, en bloc resections were performed.

Introduction

Primitive neuroectodermal tumors (PNETs) are rare malignant neoplasms which occur predominantly in children and young adults. PNETs belong to the Ewing's sarcoma family of tumors and are associated with the same reciprocal chromosomal translocation as the Ewing's sarcoma.

PNETs usually develop in the cerebellum; however, they can also arise from other sites of the central nervous system (CNS) such as the cerebral hemispheres, cortex, pineal gland and brain stem. Primary spinal PNETs are very rare conditions, and up to now,

Hideo Morioka

Department of Orthopaedic Surgery, School of Medicine, Keio University
35 Shinanomachi, Shinjuku-ku, Tokyo 160-8582 (Japan)
Tel. +81 3 5363 3812, Fax +81 3 3353 6597, E-Mail morioka@sc.itc.keio.ac.jp

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only 38 cases have been reported in the literature [1–26]. In this article, we report a rare case of a primary PNET of the conus medullaris in an elderly patient. The clinical, radiological and histological findings are presented and the relevant literature is reviewed.

Case Report

A previously healthy 63-year-old man was admitted to hospital with a 3 months' history of lower back pain projecting into the right leg. Spinal magnetic resonance imaging (MRI) demonstrated a disc hernia at the L2-L3 level and an intradural tumor from the L1 to L2 level (fig. 1a–c). After resection of the disc hernia, an intradural tumor resection was performed. The tumor was resected intralesionally because it was adhered to the spinal cord and nerve roots. Histopathological examination of the tumor specimen revealed a highly cellular, poorly differentiated neoplasm. The tumor was composed of small round cells with scanty cytoplasm and hyperchromatic nuclei. No well-defined Homer Wright and only a few ependymal rosettes were found (fig. 2a). Immunohistochemical staining for neuron-specific enolase (NSE) was strongly positive, as was staining for CD99 (MIC2) (fig. 2b). On the other hand, there was no evidence of significant epithelial differentiation.

Two months after the operation, the patient complained of progressive paresthesia and weakness of the right leg and was referred to our hospital. Spinal MRI demonstrated an intraspinal tumor extending from the Th12 to L2 level. The tumor showed high intensity on both T1- (fig. 3a) and T2-weighted images (fig. 3b). The patient underwent chemotherapy with a protocol for Ewing's sarcoma family of tumors, because the results of the histopathological examination of the tumor led us to reject the diagnoses of lymphoma and poorly differentiated carcinoma and confirmed the diagnosis of PNET (fig. 4). Additionally, he received radiation therapy consisting of 16 Gy to the spinal cord and 14 Gy to the whole brain and spinal cord. After these treatments, his neurologic symptoms completely resolved and the tumor disappeared on MRI (fig. 5a–c).

The patient continued low-dose chemotherapy with etoposide; however, 21 months after the operation, he again experienced paresthesia and weakness of the right leg because of recurrence of the tumor. He was then treated with a multidrug chemotherapy again, but his symptoms did not improve. He died due to progressive paresis 25 months after the operation.

Discussion

Primary spinal PNETs are rare lesions. The majority of spinal PNETs are the result of subarachnoidal spread of tumors in the neuraxis. In 1973, the term PNET was first introduced by Hart and Earle [27] to describe undifferentiated cerebral tumors. Reviewing this term, in 1983, Rorke [28] defined PNETs as 'central nervous system tumors predominantly composed of undifferentiated neuroepithelial cells'. At the same time, they subclassified these tumors on the basis of their cellular differentiation. Finally, in 1993, the World Health Organization (WHO) classification grouped these tumors into the category of embryonal tumors composed of undifferentiated or less differentiated neuroepithelial cells which have the capacity of differentiation to astrocytes, ependymal cells, melanocytes or muscle cells [25].

Histopathologically, PNETs are undifferentiated, small, round-cell tumors with hyperchromatic nuclei and features of neural differentiation, which typically form Homer Wright rosettes. The amount and quality of rosette formation vary substantially; some tumors may only show abortive rosette formation. On immunohistochemical examination, the most useful antibody for the diagnosis of PNET is the monoclonal antibody CD99, directed against the cell surface protein MIC2 whose gene is located on the pseudoautosomal region of the X and Y chromosomes. PNETs often have the same

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reciprocal chromosomal translocation, i.e. t(11;22)(q24;q12), which is the other key to the diagnosis of PNET [20].

PNETs involving the spinal cord are most commonly drop metastases from primary intracranial tumors, which disseminate via the cerebrospinal fluid. Therefore, primary intraspinal PNETs are extremely rare, and to our knowledge, only 38 cases have been reported in the literature so far (table 1) [1–26]. The age of the disease manifestation, including our case, ranged from 0 to 69 years, with an average age of 25.1 years. Our patient is the second oldest of all reported cases so far. In addition, there seems to be a male predominance for these tumors.

The optimal treatment of primary spinal PNETs is unknown because the tumors are very rare. Surgical resection, radiotherapy, and chemotherapy have been advocated for the treatment of spinal PNET based on PNETs at other sites. However, there is no agreement on the radiotherapy schedule, irradiance, and region (spine, brain, or both of them), as well as on the use and regimen of chemotherapy for PNETs. Successful results were reported using combinations of vincristine, doxorubicin, cyclophosphamide, ifosfamide and etoposide in Ewing's sarcoma. Despite multimodal treatment combining surgery, radiotherapy, and chemotherapy, the outcome is very poor. However, there are a few reports of cases with long-term survival and no recurrence. In these patients, en bloc resection was performed [20].

In conclusion, primary spinal PNETs are very rare tumors seldom affecting elderly patients. Therefore, their treatment is not established. In spite of surgery, radiation therapy, and chemotherapy, the outcome is very poor. It seems that the key for long-term survival is early detection and en bloc resection.

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Table 1. Summary of patients with a primary spinal PNET

Patient [ref.]	Age	Sex	Site	Survival period	Recurrence and metastasis
1 [1]	24 years	M	lumbar, cauda equina	10 months	lung
2 [2]	6 months to 10 years	NA	cervical	NA	NA
3 [2]	6 months to 10 years	NA	cervical	NA	NA
4 [2]	6 months to 10 years	NA	thoracic-lumbar	NA	NA
5 [3]	24 years	M	lumbar, intradural, cauda equina	18 months	local recurrence
6 [3]	56 years	M	lumbar, intradural, cauda equina	alive at 36 month	none
7 [3]	39 years	M	lumbar, intradural, cauda equina	30 months	local recurrence
8 [4]	26 years	M	cervical, intradural, extramedullary	10 days	spinal canal, diffuse bone
9 [5]	26 years	F	lumbar-sacral, extradural	alive at 6 months	none
10 [6]	26 years	M	thoracic-lumbar, intramedullary	36 months	between two frontal horns, roof 4th ventricle
11 [6]	15 years	F	thoracic-lumbar, intra- and extramedullary	18 months	local recurrence
12 [7]	7 years	M	thoracic-sacral, intramedullary	20 months	local progression to cervical
13 [8]	16 years	F	lumbar, intramedullary	29 months	brain
14 [9]	47 years	M	lumbar-sacral, cauda equina, intra- and extramedullary	16 months	local progression
15 [10]	3 months	F	thoracic-lumbar, intramedullary	15 days	brain
16 [11]	22 years	F	thoracic-lumbar, intramedullary	alive at 15 months	local recurrence
17 [12]	23 years	F	thoracic, intradural extramedullary	alive at 12 months	none
18 [13]	32 years	M	sacral, cauda equina	29 months	local progression, brain
19 [13]	17 years	M	lumbar, cauda equina	alive at 23 months	none
20 [14]	52 years	M	lumbar-sacral, cauda equina	alive at 12 months	none
21 [15]	5 years	M	thoracic, extradural	alive at 8 months	none
22 [16]	69 years	M	cervical-thoracic, intra- and extramedullary	3 months	none
23 [17]	22 years	F	thoracic, extramedullary	alive at 9 months	local recurrence, brain
24 [18]	49 years	F	lumbar, cauda equina	23 months	diffuse intraspinal progression
25 [18]	29 years	F	thoracic, intramedullary	17 months	multiple intraspinal
26 [19]	26 years	M	cervical, intrameningeal	3 months	local recurrence, diffuse intraspinal
27 [20]	12 years	F	cervical-thoracic, extradural	32 months	local recurrence
28 [20]	10 years	M	cervical-thoracic, extradural	22 months	multiple lung
29 [20]	30 years	F	cervical, extramedullary	14 months	local recurrence
30 [20]	14 years	M	lumbar, extramedullary	alive at 67 months	none
31 [21]	31 years	F	lumbar-sacral, cauda equina	2 months	local recurrence, left frontoparietal
32 [22]	3 years	M	cervical, intramedullary	several days	local progression to brainstem
33 [23]	38 years	M	thoracic, intramedullary	18 months	brain, multiple spinal cord
34 [24]	54 years	F	cervical, intramedullary	NA	none
35 [25]	9 years	F	thoracic-lumbar, extramedullary	alive at 18 months	none
36 [25]	8 years	M	cervical, extradural	alive at 8 months	local recurrence
37 [25]	18 years	M	cervical, intramedullary	alive at 6 months	none
38 [26]	17 years	M	thoracic, intramedullary	alive at 6 months	none

NA = Not available.

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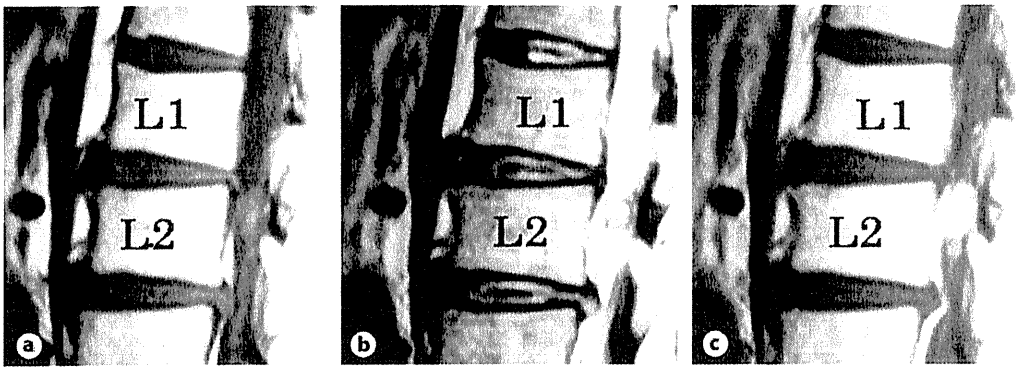

Fig. 1. Spinal MRI showed a disc hernia at the L2-L3 level and a generally isointense intradural tumor with focal high-intensity T1-weighted images at the level of L1-L2 (**a**). On T2-weighted MRI, the tumor demonstrated high intensity with focal low intensity (**b**). The tumor is homogenously enhanced by gadolinium (**c**).

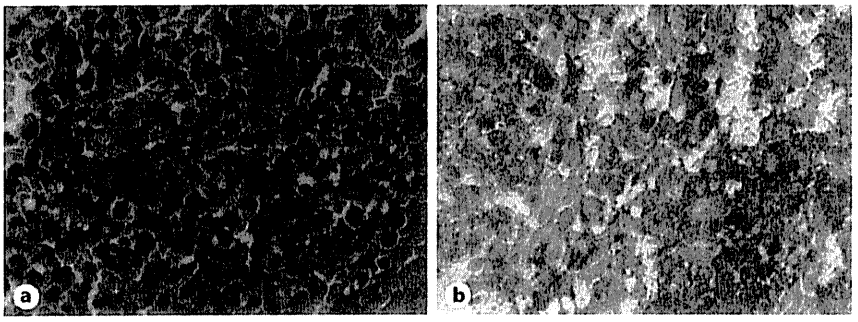

Fig. 2. Histopathological examination of the tumor specimen revealed a highly cellular, poorly differentiated neoplasm. The tumor was composed of small round cells with scanty cytoplasm and hyperchromatic nuclei. No well-defined Homer Wright and only a few ependymal rosettes were found (**a**). Immunohistochemical staining for NSE was strongly positive (pictures not shown), as was staining for CD99 (MIC2) (**b**).

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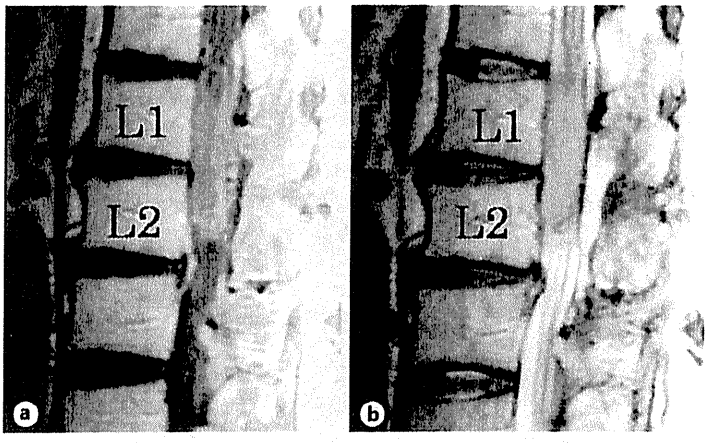

Fig. 3. Spinal MRI demonstrated an intraspinal tumor extending from the Th12 to L2 level. The tumor showed high intensity on T1-weighted image (**a**) as well as on T2-weighted image (**b**).

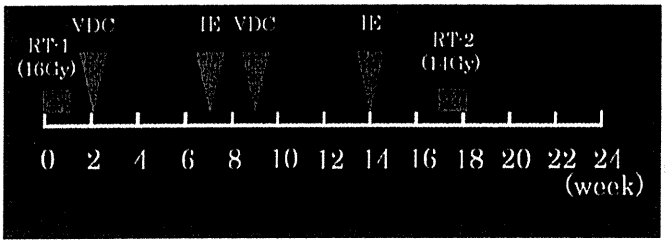

Fig. 4. Protocol of chemotherapy. RT-1 = Local radiation 16 Gy; RT-2 = radiation to brain and spinal cord 14 Gy; VDC = vincristine (1.5 mg/m^2) + doxorubicin (30 mg/m^2) + cyclophosphamide (1,200 mg/m^2); IE = ifosfamide (1.8 g/m^2) + etoposide (100 mg/m^2).

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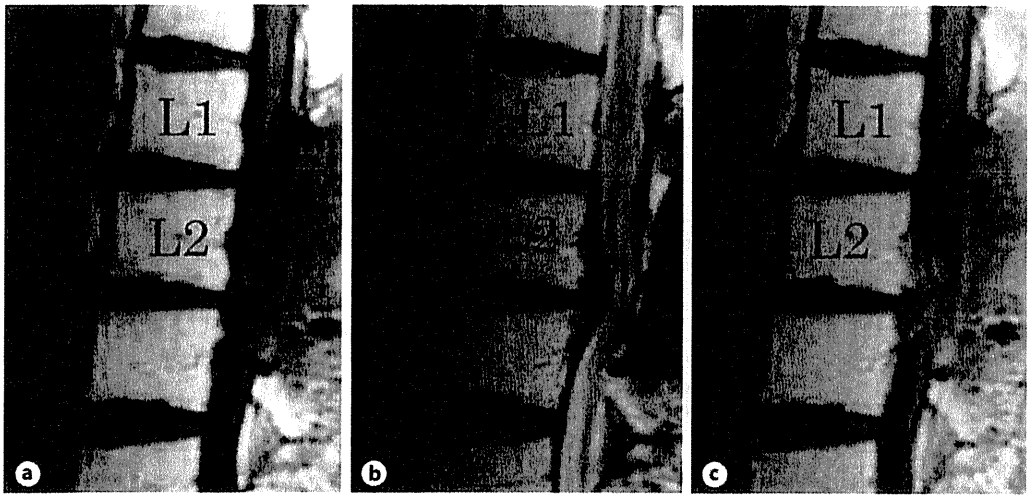

Fig. 5. MRI after chemotherapy and radiation. **a** T1-weighted image. **b** T2-weighted image. **c** T1-weighted image with gadolinium enhancement.

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CASE STUDY

Anaplastic Transformation of Follicular Thyroid Carcinoma in a Metastatic Skeletal Lesion Presenting with Paraneoplastic Leukocytosis

Robert Nakayama,[1,2] Keisuke Horiuchi,[1,2] Michiro Susa,[1] Seiichi Hosaka,[1] Yuichiro Hayashi,[3] Kaori Kameyama,[3] Yoshihisa Suzuki,[4] Hiroo Yabe,[1] Yoshiaki Toyama,[1] and Hideo Morioka[1]

Background: Anaplastic transformation of differentiated thyroid carcinoma (DTC) is a rare event with a poor clinical outcome. It usually occurs in the primary site or in regional lymph nodes, but rarely in distant metastatic lesions.

Summary: A 55-year-old woman with persistent pain in the left hip joint visited our hospital. She had a history of DTC that had been surgically removed 12 years earlier. Clinical images showed a tumorous mass in the left pelvis, indicative of bone metastasis. The patient underwent surgery to remove the tumor and remained stable until local recurrence was found 5 weeks after the surgery. The patient subsequently underwent radiation therapy; however, she died of respiratory failure due to lung metastases 2 months after the surgery for the recurrent lesion. The surgical specimens were diagnosed as anaplastic thyroid carcinoma, indicating that anaplastic transformation of thyroid follicular carcinoma occurred in the metastatic skeletal lesion. In addition, the patient had an unusually high white blood cell count throughout the course. Based on elevated serum granulocyte colony-stimulating factor (G-CSF) levels and positive immunostaining for G-CSF in the surgical specimens, the patient was diagnosed with paraneoplastic leukocytosis.

Conclusion: To our knowledge, this is the first case of anaplastic transformation of DTC arising in a metastatic bone lesion described in the literature. In addition, the present case also exhibited severe leukocytosis accompanied by elevated serum G-CSF levels. Clinicians should be aware of the possibility of this occurring in their patients with DTC, as this development calls for a rapid change from observational follow-up to aggressive treatment.

Introduction

DIFFERENTIATED THYROID CARCINOMA (DTC), which includes both papillary carcinoma and follicular carcinoma, accounts for more than 90% of all thyroid malignancies (1,2). DTC is characterized by an indolent clinical course with a relatively favorable prognosis. Even those patients diagnosed with metastatic tumors may still experience long-term survival (3–5); therefore, surgeries for metastatic lesions are often warranted to improve quality of life. In contrast, anaplastic thyroid carcinoma (ATC) is a highly aggressive neoplasm with a poor prognosis (6–10), accounting for less than 3% of all thyroid carcinomas (1,2). ATC can arise *de novo* or, more commonly, through anaplastic transformation (or dedifferentiation) from preexisting DTC (11,12), which almost exclusively occurs in the thyroid gland *in situ* or in the regional lymph nodes (8,13). Anaplastic transformation in extrathyroid sites, on the other hand, is exceptionally uncommon, and only a few cases have been reported in the literature (14–18).

We herein present a very rare case of anaplastic transformation of metastatic follicular thyroid carcinoma in a metastatic skeletal lesion, which, to our knowledge, is the first case reported in the literature. The present case is also unique in that it was accompanied by paraneoplastic leukocytosis, which is known to herald a poor clinical outcome in a variety of cancers. Since anaplastic transformation forebodes an extremely poor prognosis, clinicians seeing patients with a history of DTC should be aware of this condition, because it often heralds a progressively deteriorating clinical course.

Patient

A 55-year-old woman was referred to our service with a 3-month history of left hip pain and gait disturbance. She had a history of thyroid cancer that had been treated with total

Departments of [1]Orthopedic Surgery, [4]Anti-aging Orthopedic Research and [2]Pathology, School of Medicine, Keio University, Tokyo, Japan.
[3]Department of Orthopedics, Kyosai Tachikawa Hospital, Tokyo, Japan.

thyroidectomy at the age of 43 years. She subsequently developed multiple skeletal metastases and underwent spinal cord decompression and posterior spinal fusion for a pathological fracture in the thoracic vertebrae at the age of 51 years. The surgical specimens were histologically diagnosed as well-differentiated follicular thyroid carcinoma with no findings suggestive of anaplastic transformation. Radioactive iodine (^{131}I) therapy was administered thrice after the surgery.

At presentation, X-rays of the pelvis revealed an osteolytic lesion in the left periacetabular region (Fig. 1a). Magnetic resonance imaging of the pelvis showed a bone tumor located in the left anterior inferior iliac spine extending to the superior ramus of the left pubis (Fig. 1c, d). The tumor was infiltrating into the surrounding soft tissue, and there was extensive edema around the lesion. Technetium^{99m} bone scintigraphy showed strong uptake in the same region and faint accumulation in the skull, ribs, and spinal column. Chest X-ray showed no apparent metastatic nodules in the lung (Fig. 1b). In addition, there was a marked increase in the white blood cell (WBC) count (29,800/μL), despite the fact that the patient showed no sign of infection. To alleviate the pain and disability caused by the tumor in the hip, extensive curettage was performed. The postoperative course was uneventful, and she was discharged from the hospital 2 weeks after the surgery.

On gross observation, the tumor was tan-whitish in color with areas of extensive coagulative necrosis and infiltration into the surrounding soft tissues. Histological analysis revealed that the tumor cells were composed of anaplastic cells containing eosinophilic cytoplasm and hyperchromatic nuclei with prominent nucleoli. Follicular arrangements seen in the previous surgical specimens were totally lost. Moreover, an overt infiltration of neutrophils and lymphocytes was observed. Immunohistochemical analysis revealed that the tumor cells were positive for pankeratin (AE1/AE3) and vimentin and negative for thyroglobulin and thyroid transcription factor (TTF-1). The MIB-1 labeling index reached ~10%. Based on these findings, a diagnosis of anaplastic transformation of DTC in the metastatic bone lesion was made (Fig. 2a, b).

The patient returned to our hospital with a complaint of worsening hip pain and swelling around the surgical wound site 3 weeks after her discharge. Pelvic computed tomography revealed an expanding mass with peripheral enhancement around the left acetabulum (Fig. 3a). Laboratory test results revealed a sharp increase in the WBC count (35,300/μL). Given these findings, infection at the surgical site was initially suspected, and emergency debridement was performed. However, there was no apparent abscess formation, but a tan-whitish necrotic mass reminiscent of the previous tumor. The bacterial culture showed negative results, and the surgical specimens were diagnosed as anaplastic carcinoma. At this point, the levels of hematopoietic cytokines in the serum were examined to investigate the cause of the increased granulopoiesis. The serum levels of granulocyte colony-stimulating factor (G-CSF) were highly upregulated (73.7 pg/mL; normal range, <18.1 pg/mL, intra-assay coefficient of variation; 2.75%–4.53%, interassay coefficient of variation; 10.4%–13.2%). In addition, immunostaining of the surgical specimens revealed positive staining for G-CSF (Fig. 2c). Since there were no findings indicative of infection, the patient was diagnosed with local recurrence and paraneoplastic leukocytosis caused by aberrant production of G-CSF from the tumor cells.

External beam radiation therapy at a dose of 50 Gy (25 fr) to the left pelvic region was subsequently initiated, and there

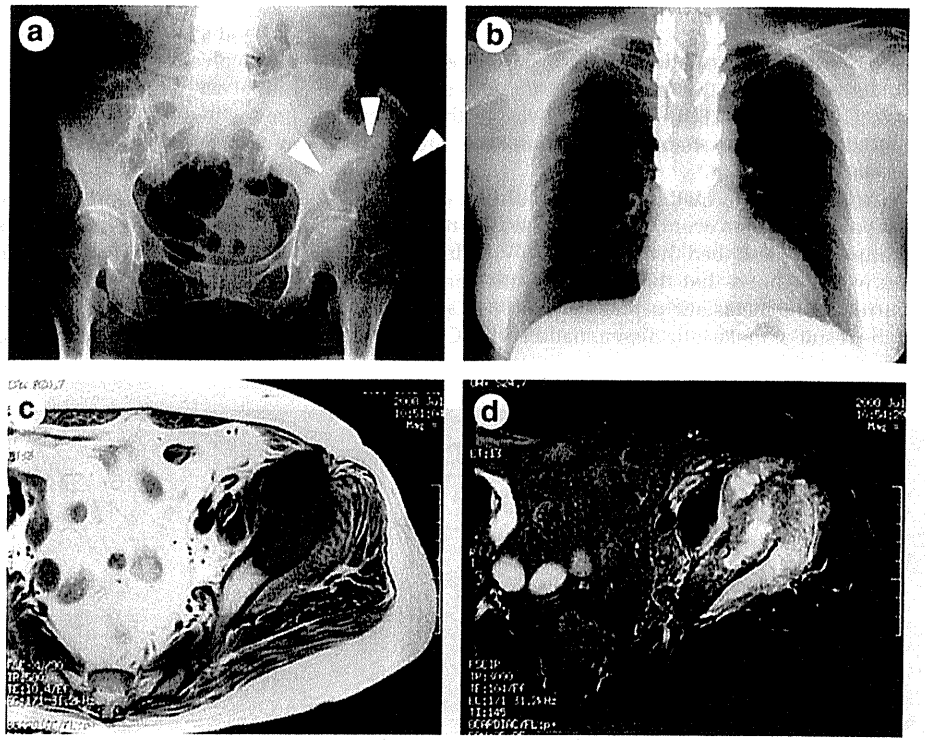

FIG. 1. **(a)** A plain anteroposterior radiograph of the pelvis showing an osteolytic lesion in the left periacetabular region (arrowheads). **(b)** A plain chest X-ray at presentation showing no visible metastatic nodules in the lung. **(c)** An axial T1-weighted magnetic resonance image of the pelvis revealing a bone tumor in the left periacetabular region. **(d)** A short-tau inversion recovery image showing extensive edema in the surrounding soft tissue.

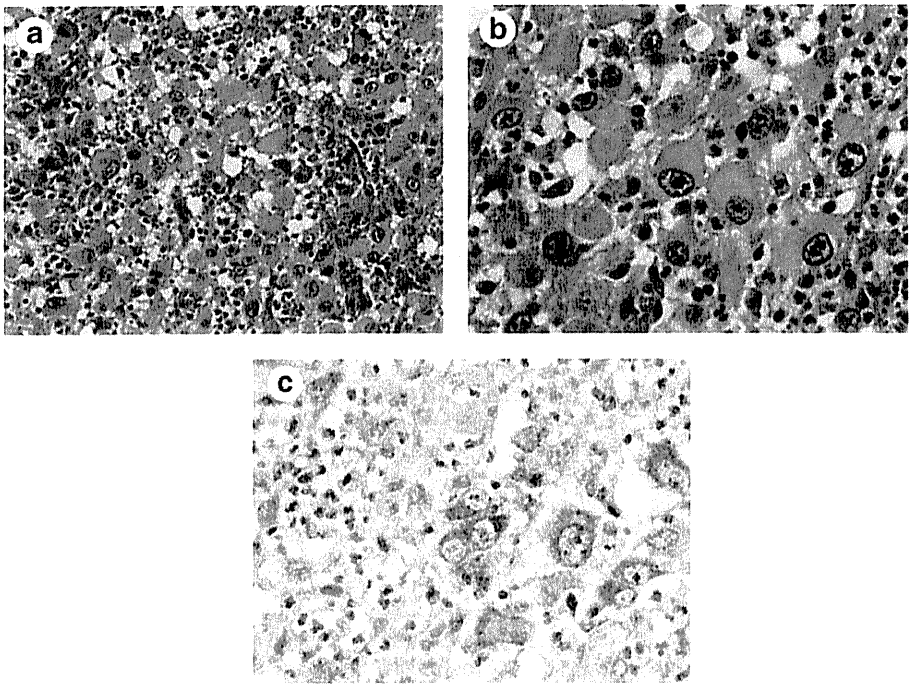

FIG. 2. **(a)** Hematoxylin-eosin staining of the surgical specimen. Low-power field. **(b)** High-power field. **(c)** Positive immunostaining for granulocyte colony-stimulating factor in the tumor cells.

was a temporal resolution in the symptoms and leukocytosis (Fig. 4). However, around the same time, the patient developed severe pleural effusion due to rapidly enlarging pulmonary tumors suspected to be metastases from the pelvic lesion (Fig. 3b). Her general condition deteriorated soon after, and she died 14 weeks after the initial pelvic surgery.

Discussion

Anaplastic transformation of DTC is a rare event that usually occurs in the thyroid gland *in situ* or in surrounding cervical lymph nodes (8,13). To our knowledge, this is the first report that describes anaplastic transformation arising in a metastatic bone lesion.

Anaplastic transformation of DTC in distant extrathyroid sites is an uncommon event, and only a few case reports in the literature have described this condition (14–18). In the present case, we concluded that the anaplastic transformation had occurred in the metastatic pelvic bone lesion *in situ*, but was not the result of metastatic dissemination of ATC to the bone for the following reasons: (i) in our patient, there was no metastatic lesion in the cervical lymph nodes, and the thyroid had been surgically removed 12 years before the onset of the pelvic lesion; (ii) the metastatic lesions in the lung and spine showed no significant change in size and remained clinically stable; and (iii) the tumor in the left pelvic bone was the first lesion that showed a rapid increase in size throughout the course.

Another important clinical feature of the present case is the involvement of paraneoplastic leukocytosis. Although we cannot rule out the possibility that the increase in G-CSF production was derived from inflammatory reaction and/or metastatic lesions other than the pelvic tumor, positive immunostaining for G-CSF in the pelvic tumor cells indicates the pelvic lesion as the major source of G-CSF in the present case. Paraneoplastic leukocytosis is often seen in patients with lung, gastrointestinal, genitourinary, or head and neck cancers (19–22). This condition is manifested by an abnormally high WBC count often caused by aberrant production of hematopoietic cytokines, including granulocyte-macrophage

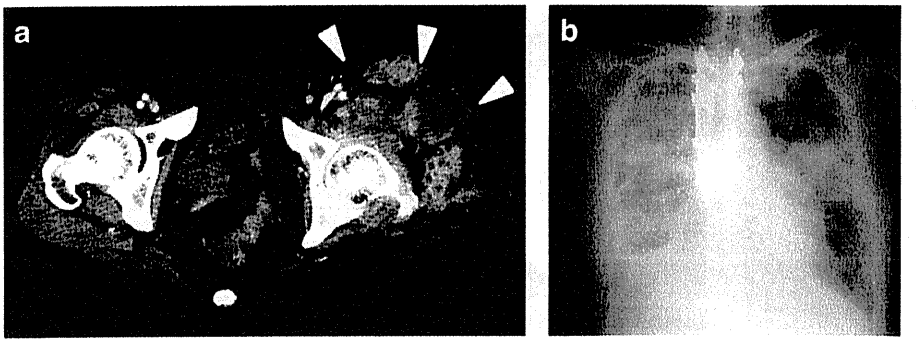

FIG. 3. **(a)** An enhanced computed tomography image of the pelvis 5 weeks after the initial curettage. **(b)** A plain chest X-ray taken 11 weeks after the initial curettage showing multiple metastatic tumors and pleural effusion in the lung.

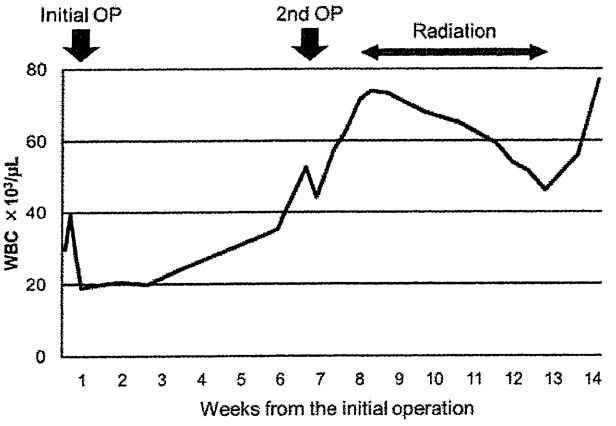

FIG. 4. Time course of the WBC count during the clinical course. Temporal remission of leukocytosis was achieved after each treatment; however, the WBC count continuously rose throughout the course and reached 77,000/μL at the time of death. OP, operation; WBC, white blood cell.

CSF, G-CSF, interleukin 3, and interleukin 6, from the tumor cells (23–27). Paraneoplastic leukocytosis rarely occurs in thyroid tumors, and there are only a few case reports in the literature (28–35). Of note, this phenomenon is seen more frequently in patients with ATC than in those with DTC (35), indicating that aberrant production of cytokines tends to occur when the tumor cells undergo anaplastic transformation. Furthermore, in a multivariate analysis by Sugitani *et al.*, leukocytosis (WBC count of > 10,000/μL) was found to be an independent prognostic factor for patients with ATC in addition to the duration of complaints (< 1 month), tumor size (> 5 cm), and presence of distant metastatic lesions (36). However, their study did not clarify whether the leukocytosis was caused by abnormal production of hematopoietic cytokines from the tumor cells or derived secondary to the inflammatory reaction. Therefore, further studies are warranted to elucidate the cause of leukocytosis in patients with ATC and to determine how leukocytosis affects the biological behavior of the tumor.

In summary, we report a case of anaplastic transformation of follicular carcinoma in a metastatic skeletal lesion, which was accompanied by severe paraneoplastic leukocytosis. Although this event is not common, clinicians should be aware of the possibility of anaplastic transformation in metastatic foci, because occurrence of this event could drastically affect the treatment modality of patients diagnosed with DTC who would have been expected to have a relatively indolent clinical course and longer prognosis.

Disclosure Statement

The authors declare that no competing financial interests exist.

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Address correspondence to:
Robert Nakayama, M.D., Ph.D.
Kelo University School of Medicine
Department of Orthopaedic Surgery
35 Shinanomachi, Shinjuku-ku
Tokyo 160-8582
Japan

E-mail: robert@a2.keio.jp

第 42 巻 第 2 号

骨盤褐色腫を合併した原発性副甲状腺機能亢進症の1例*

吉田進二** 森岡秀夫 鈴木禎寿
西本和正 保坂聖一 矢部啓夫
戸山芳昭

[Key words：brown tumor（褐色腫），pelvis（骨盤），hyperparathyroidism（副甲状腺機能亢進症）]

褐色腫は副甲状腺機能亢進症の約1%に合併する骨病変の1つである[4]．今回われわれは，骨盤褐色腫を合併した原発性副甲状腺機能亢進症の1例を経験したので，若干の文献的考察を加えて報告する．

症 例

患者：52歳，女性．

主訴：左殿部痛．

既往歴・家族歴：特記すべきことなし．

現病歴：2001年頃より両殿部痛が出現し，近医で坐骨神経痛と診断されて保存療法を施行されたが，左殿部痛が残存した．2002年11月に精査のために行ったMRI検査で骨盤腫瘍が疑われたため，同年12月に当科を紹介され受診した．

初診時所見：左殿部に圧痛を認めたが，その他に明らかな異常所見を認めなかった．

単純X線：右腸骨には仙腸関節から臼蓋部にかけて，左腸骨には仙腸関節付近に辺縁硬化像を伴う嚢腫様病変を認めた（図1）．

MRI：右腸骨臼蓋部および左腸骨部仙腸関節近傍にそれぞれ長径約7 cm大の腫瘍性病変を認めた．腫瘍は周囲に膨隆し，内部はT1強調画像で低信号，T2強調画像で高信号を示した．また，左側の病変は内部にT2強調画像でfluid-fluid levelの形成を認めた（図2）．

骨シンチグラフィー：両側腸骨部に腫瘍辺縁優位の集積亢進と内部の低下，またその他の骨盤部，頭蓋骨・脊椎などにも異常集積を認めた（図3）．

一般血液生化学検査所見：アルカリホスファターゼ（ALP）が1,659IU/lと異常に上昇していた以外に明らかな異常所見は認めなかった．

以上の所見より，転移性骨腫瘍や何らかの原因により生じた多発性骨病変を疑い，2002年12月に病理組織診断確定のため左腸骨骨腫瘍の生検術を施行した．

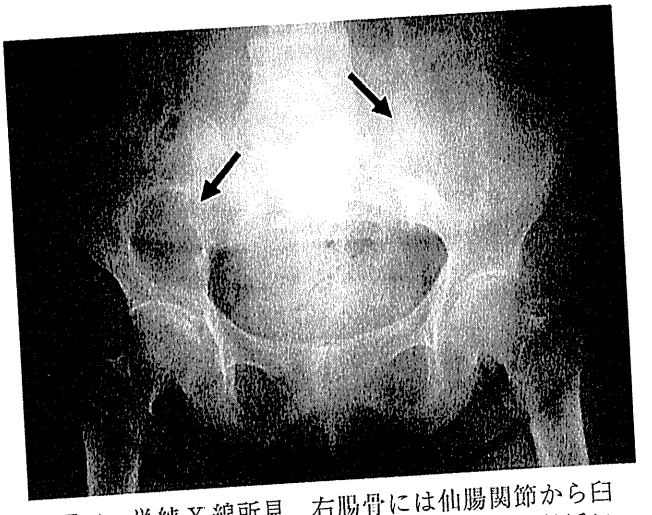

図1 単純X線所見．右腸骨には仙腸関節から臼蓋部にかけて，左腸骨には仙腸関節付近に辺縁硬化像を伴う嚢腫様病変を認める（左右ともに矢印で示した）．

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* A case of primary hyperparathyroidism with brown tumor in the pelvis.

** YOSHIDA Shinji, MORIOKA Hideo, SUZUKI Yoshihisa, NISHIMOTO Kazumasa, HOSAKA Seiichi, YABE Hiroo, & TOYAMA Yoshiaki
慶應義塾大学医学部整形外科学教室

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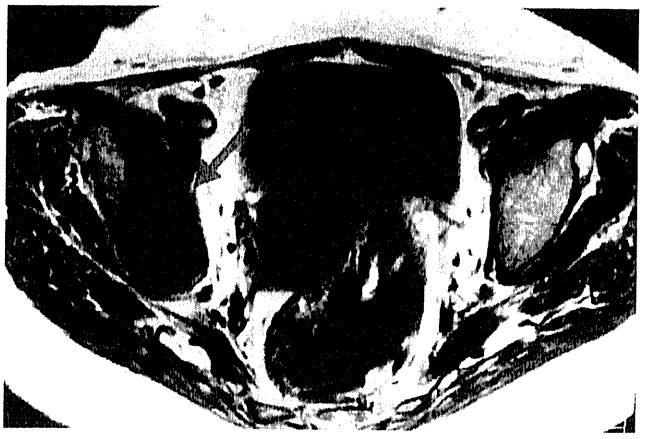
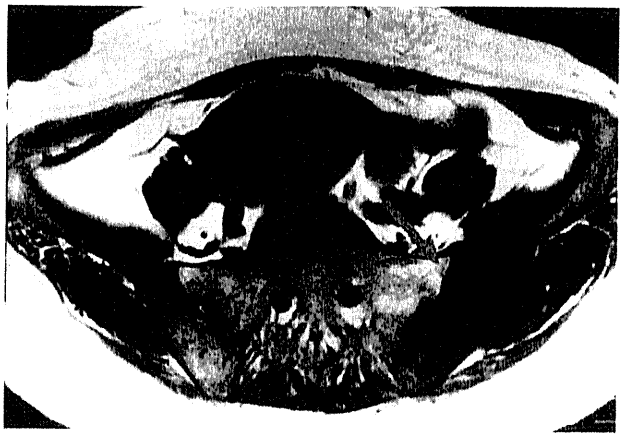
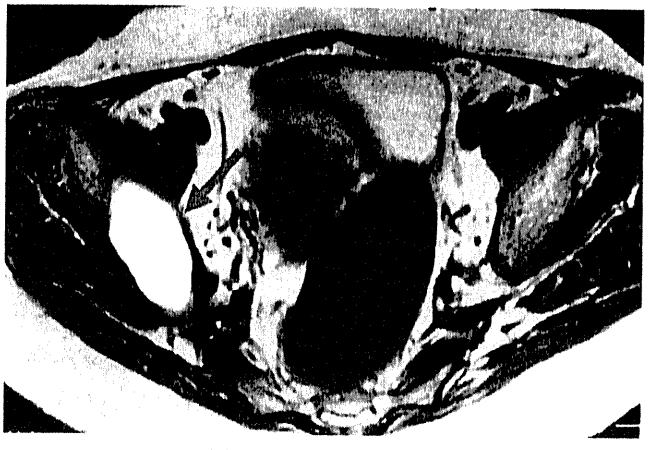
（受稿 2010年11月5日）



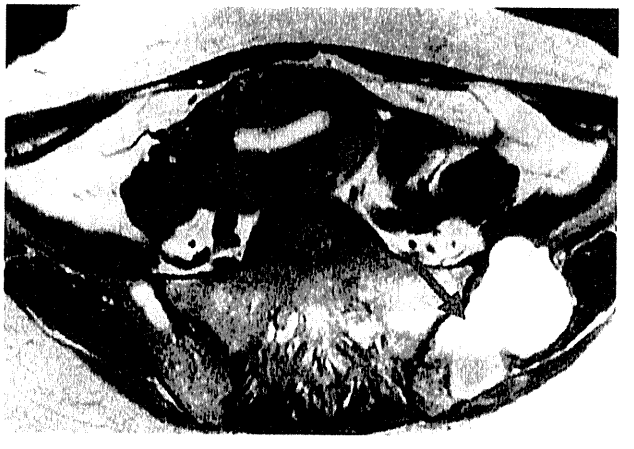
(a) T1WI (右腸骨)



(b) T1W1 (左腸骨)

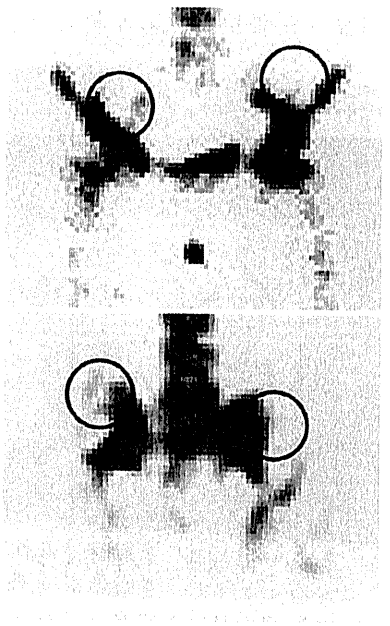


(c) T2WI (右腸骨)

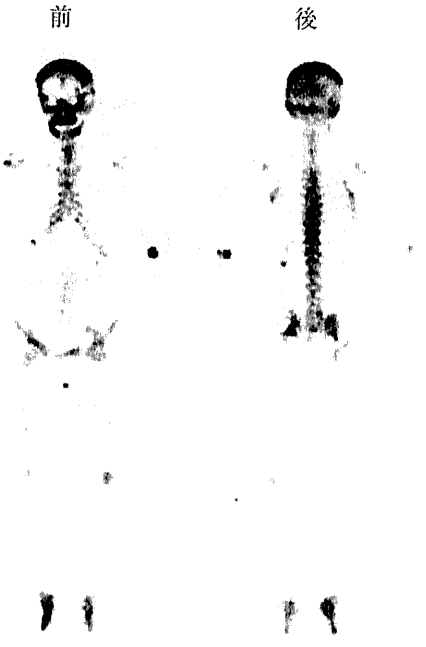


(d) T2W1 (fluid-fluid level)

図 2 MRI. 右腸骨臼蓋部および左腸骨部仙腸関節近傍にそれぞれ長径約 7 cm 大の腫瘍性病変を認める. 腫瘍は周囲に膨隆し, 内部は T1 強調画像で低信号, T2 強調画像で高信号を呈する. また, 左側の病変は内部に T2 強調画像で fluid-fluid level の形成を認める (左右ともに矢印で示した).



(a) 両側腸骨部



(b) 全身像

図 3 骨シンチグラフィー. 両側腸骨部に腫瘍辺縁優位の集積亢進と内部の低下 (左右ともに丸印で示した)(a), またその他の骨盤部, 頭蓋骨・脊椎などにも異常集積を認める (b).