Nagoya J. Med. Sci. **59.** 159 ~ 165, 1996

SOLID VARIANT OF AN ANEURYSMAL BONE CYST (GIANT CELL REPARATIVE GRANULOMA) OF THE 3RD LUMBAR VERTEBRA

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ABSTRACT

A 9-year-old girl with a solid variant of an aneurysmal bone cyst in the 3rd lumbar vertebra showed a good response to low-dose radiation as the only treatment. The solid variant of aneurysmal bone cyst is thought to be a reactive response to intraosseous hemorrhage and is also called giant cell reparative granuloma or giant cell reaction. These lesions in the jaw and the short tubular bones of the hands and feet frequently recur after surgery. Aneurysmal bone cysts of the spine also show a fairly high recurrence rate after incomplete resection or radiation therapy. However, 7 previous cases of the solid variant of aneurysmal bone cyst in the spine and this case did not show recurrence after a mean follow-up period of 45 months. This difference in behaviour suggests that the solid variant should be recognized before surgery as being distinct from conventional aneurysmal bone cysts, especially in the spine.

Key Words: Solid variant of aneurysmal bone cyst, Giant cell reparative granuloma, Lumbar spine, Radiation therapy

INTRODUCTION

In 1953, giant cell reparative granuloma (GCRG) was first reported by Jaffe as a reaction to intra-osseous hemorrhage found exclusively in the jaw bones.[1)] However, the same histology has since been found throughout the skeleton, and is not only called "GCRG"[1–10)] but also "giant cell reaction"[11–14)] and "solid variant of aneurysmal bone cyst (S-ABC)".[15–19)] In this paper, we discuss the case of a 9-year-old girl with S-ABC of the 3rd lumbar vertebra who showed complete recovery after low-dose radiation therapy. In addition, we review the literature on S-ABC of the vertebrae.

CLINICAL FEATURES

A 9-year-old girl complained of backache after playing sports and was referred to our university hospital with the diagnosis of an osteolytic bone tumor in the 3rd lumbar vertebra. Physical examination disclosed a very rigid spine and no neurological abnormalities.

Radiography of the lumbar spine showed an osteolytic lesion in the right half of the 3rd lumbar vertebra with soft tissue extension. The pedicle sign was positive on the right side (Fig. 1). CT demonstrated a large expanded osteolytic lesion with a calcified rim occupying the postero-

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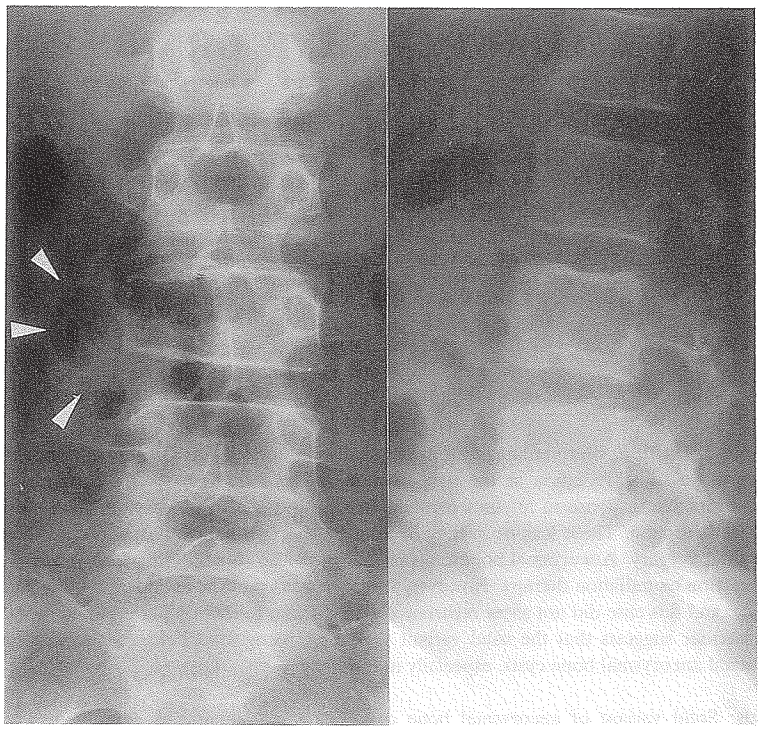

Fig. 1 Radiographs in the antero-posterior and lateral views of the 3rd lumbar vertebra show a geographic osteolytic lesion in the left half of the body. The lateral cortex is thin and ballooned out by a large extraskeletal tumor mass with an eggshell-like calcified rim.

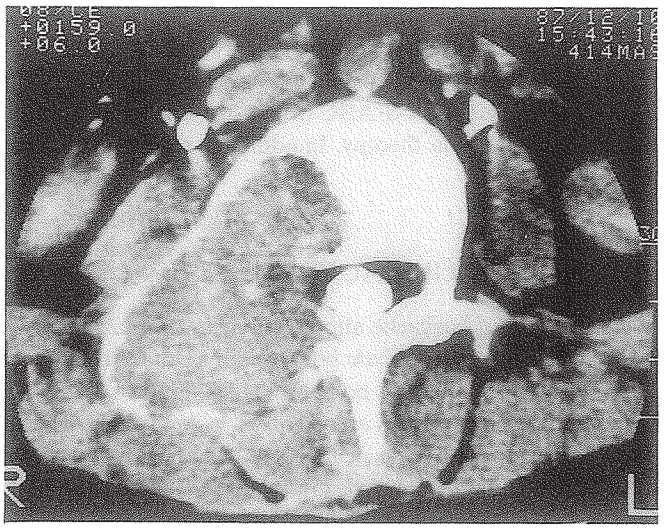

Fig. 2 CT scan showing that the right half of the 3rd lumbar vertebra has been destroyed and that the lesion has progressed through the pedicle to the transverse process. There is a calcified shell around the mass.

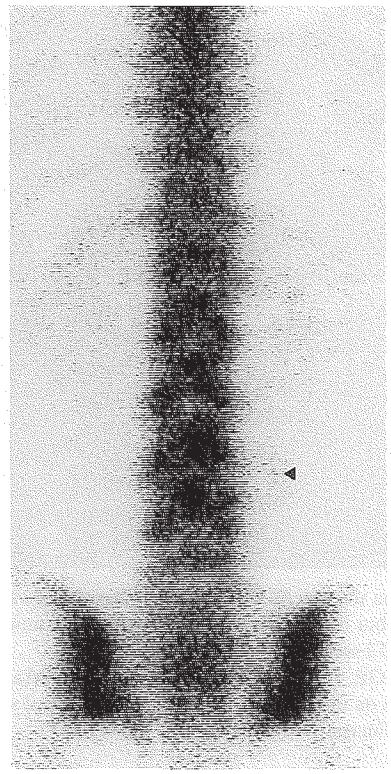

Fig. 3 Posterior view of ^{99m}Tc-methylene diphophonate (^{99m}Tc-MDP) bone scintigraphy reveals weak ring-like accumulation at the border of the lesion.

lateral half of the vertebral body, the pedicle, the transverse process, and the lamina of the 3rd lumbar vertebra on the right side (Fig. 2). ^{99m}Tc-methylene diphophonate (^{99m}Tc-MDP) bone scintigraphy revealed ring-like weak accumulation at the edge of the lesion (Fig. 3). MRI was done before radiation therapy (0.15 T, Toshiba, Tokyo, Japan) and showed a homogeneous iso-intense mass with a normal vertebra on T1-weighted images and a homogeneous bright high signal intense mass on T2-weighted images. No fluid level was demonstrated (Fig. 4-A, B).

Biopsy showed predominant fibroblastic or fibrohistiocytic proliferation along with osteoclast-like giant cell-rich areas. Minor foci of reactive osteoid and fibromyxoid tissue were also detected. However, no large blood-filled space was demonstrated, which was the critical point in differentiating this cyst from an aneurysmal bone cyst (ABC). Mitotic figures were absent (0/10 HPF) (Fig. 5).

From these findings, a diagnosis of S-ABC was made. Low-dose radiation therapy (20 Gy) was initially administered in order to prevent severe bleeding during the operation, but hepatitis prevented us from performing surgery. Enhanced CT was done 3 months after radiation therapy and disclosed reactive bone formation in the lesion. MRI (1.5 T, Signa, GE, USA) 8 months after raditaion therapy showed a nonhomogeneous low signal intense mass on T1-weighted images and a mixed signal intense mass, high to low, on T2-weighted images (Figs. 4-C, D). These findings suggested new bone formation with partial persistence of the lesion, so observation was

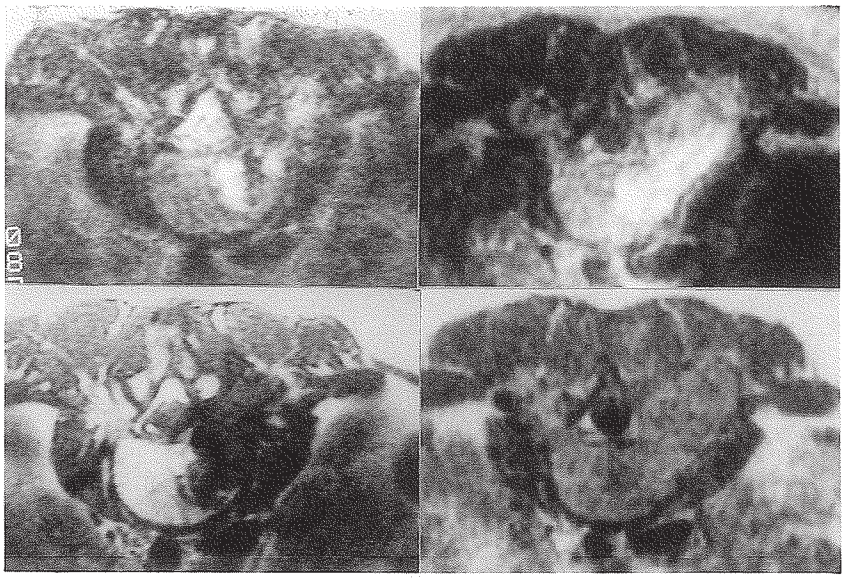

Fig. 4-A,B MRI before radiation therapy (0.15 T, Toshiba, Japan) shows a homogeneous iso-intense mass with a normal vertebra on the T1-weighted image and a homogeneous, bright, high signal intensity mass on the T2-weighted image.

-C,D MRI (1.5 T, Signa, GE, USA) 8 months after radiation therapy shows a nonhomogeneous low signal intensity mass on the T1-weighted image and a mixed signal intensity (high to low) mass on the T2-weighted image.

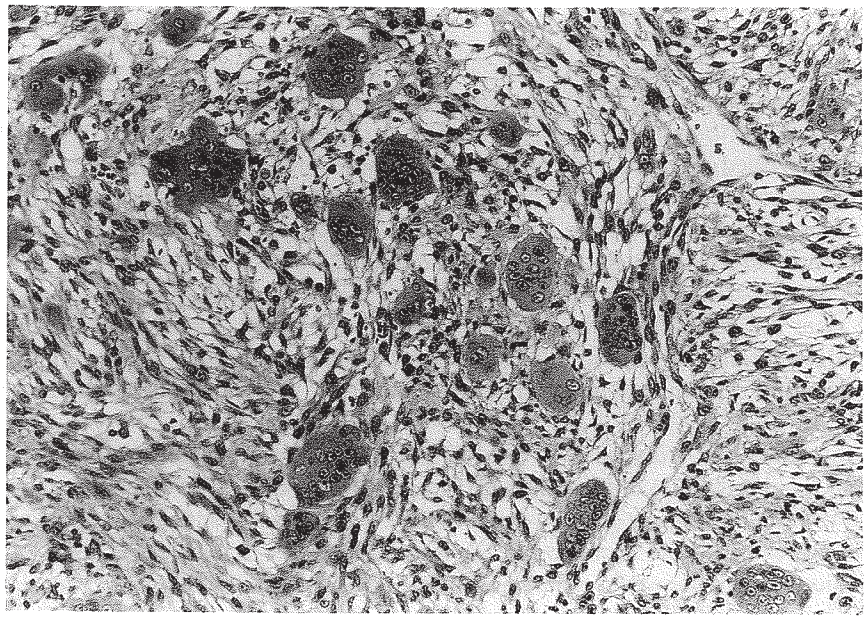

Fig. 5 Biopsy reveals that fibroblastic or fibrohistiocytic proliferation forms the predominant component with minor foci of reactive osteoid and fibromyxoid tissue (X20).

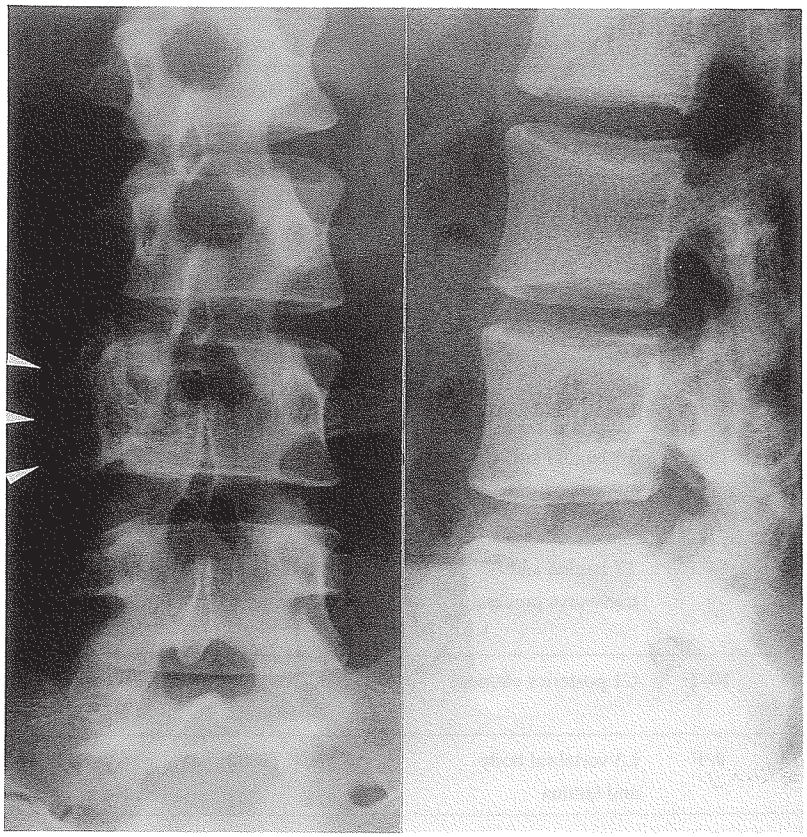

Fig. 6 After follow-up for 6 years and 8 months, X-rays of the 3rd lumbar vertebra and extraskeletal tumor mass show a density very similar to that of the normal vertebra, suggesting that the osteolytic lesion has healed completely.

continued. Within 1 year after radiation therapy, the lesion was completely filled with new bone and the osteolytic lesion area disappeared without shrinkage at the site. A radiograph taken 6 years and 8 months after radiation therapy demonstrated complete healing and mature bone formation (Fig. 6). The patient had no limitation of motion and no pain.

DISCUSSION

In 1953, Jaffe described GCRG as a reactive response to intraosseous hemorrhage found exclusively in the jaw.[1] GCRG has a similar histology to aneurysmal bone cysts, except for the absence of large blood-filled spaces.[16,17] Following Jaffe's paper, several authors have classified the same histological lesion in other parts of the skeleton as extragnathic GCRG,[2–10] giant cell reaction[11–14] and S-ABC.[15–19]

There have only been 7 previous reports of S-ABC (or GCRG) in the spine: three cases by Sanerkin,[15] one by Vergel De Dios,[17] two by Oda[16] and one by Bertoni.[19] Six of these cases and our case are listed in Table 1 (the case of Vergel De Dios was excluded as the exact location

Table 1. Clinical features of the solid variant of aneurysmal bone cyst in spine.

Case	Author	Age/Sex	Site and complication	Treatment	Follow-up
1	Sanerkin	7/M	L4 lamina	Laminectomy	6 years No recurrence
2	Sanerkin	6/F	T2 neural arch Extra-dural cord compression	I.L.R. (90%) Laminectomy R.T. (1500 rad)	1 year No recurrence complete recovery
3	Sanerkin	13/M	T7 lamina Extra-dural cord compression	I.L.R. (subtotal) Laminectomy R.T. (1500 rad)	6 months No recurrence Nearly complete recovery
4	Oda	17/F	T1 lamina and spinous process	En bloc resection (subtotal) Laminectomy	1 year No recurrence
5	Oda	16/F	T7 lamina and transverse process	Curettage Bone graft R.T. (5000 rad)	8 years 3 months No recurrence
6	Bertoni	10/F	C1 posterior element	Curettage Bone graft	3 years 2 months No recurrence
7	Sato	9/F	L3 vertebral body and lamina	R.T. (2000 rad)	6 years 8 months No recurrence

I.L.R. — Intralesional resection
R.T. — Radiation therapy

was not given). Among the seven listed cases, there was one lesion in the cervical spine, four in the thoracic spine and two in the lumbar spine. In all cases, the lesion was mainly situated in the posterior elements. Four cases were treated surgically (laminectory in one, subtotal resection in one and curettage with bone graft in two), but the surgical margins were inadequate because complete resection of this type of spinal lesion proved difficult. Radiation therapy was selected for three cases and two of them had intralesional resection before radiation. These seven patients showed good healing of the lesion and there were no differences in outcome between surgery alone, surgery and low-dose radiation, and radiation therapy without surgery. Two of the seven patients had symptoms of cord compression, but all seven showed nearly complete recovery after treatment.

Several authors have reported relatively high local recurrence rates of 33%,[5] 36%,[7] 40%[14] and 50%[2] for GCRG or giant cell reaction in the jaws and the short tubular bones of the hands and feet. A large series of ABC in the spine also showed a relatively high recurrence rate, 70% following incomplete resection,[20] as well as 50% after radiation therapy,[21] 22% after radiation, and 0.6% after partial resection and radiation.[22] However, there was no local recurrence after a mean follow-up period of 45 months in all eight spinal cases of S-ABC, indicating a good response to surgical intervention or low-dose radiation therapy.The natural course of S-ABC

(GCRG) in the spine might be benign unless pathological fracture or cord compression occurs. Before starting treatment, S-ABC should be diagnosed by CT and MRI, which does not show any fluid-filled space or fluid level. After confirming a diagnosis of S-ABC, partial resection can be applied as one method of treatment, including curettage and bone grafting. However, low-dose radiation therapy (under 20 Gy), or both surgery and radiation may be used, since radiotherapy not exceeding 20 Gy is relatively safe.[22] If radiation is the choice, an interval of at least 3 months before subsequent surgical treatment is recommended to assess the efficacy of radiation.

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