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Chondroblastoma of the sacrum

A case report

Death due to a chondroblastoma of the sacrum is uncommon. We report the case of a 48-year-old man with this condition. During a 10-year period, the tumor spread into the retroperitoneal space and was an enormous mass after repeated radiotherapy. The patient died of renal failure. The tumor was histologically benign even at autopsy.

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Benign chondroblastoma is an uncommon cartilaginous bone tumor. It usually involves the epiphysis of a long bone in young adults. The prognosis is considered good. However, several authors (Kahn et al. 1969, Huvos et al. 1977) have emphasized its aggressive nature or sometimes even malignant course. We report a fatal case of benign chondroblastoma in the sacrum.

Case report

A 48-year-old revenue officer was admitted to the National Sagamihara Hospital in 1971 with low back pain of 2 months' duration. The physical examination was normal except for tenderness over the sacrum. Radiographs showed a radiolucent lesion in the lateral half of the upper sacrum adjacent to the right sacroiliac joint with marked peripheral radiopacity (Figure 1).

A biopsy of the lesion was made and histologic examination revealed a very large number of multinucleated giant cells resembling osteoclasts and small, plump mononucleated cells with oval or round nuclei and with pale cytoplasm (Figure 3). The histopathologic diagnosis at that time was giant cell tumor, Grade I. Surgical removal was considered to be too difficult and radiotherapy with 60 Gy was given. The symptoms improved and the tumor appeared to stop growing; radiographically, there was massive calcification. The patient was lost to follow-up 1 year later.

In 1976, he got abdominal pains, varicose veins of the right lower leg, and hemorrhoids.

He was readmitted to the hospital in January 1978 with a 10-cm mass in the lower abdomen. He developed pollakiuria and impaired renal function. An intravenous pyelogram showed no function of the right kidney and hydronephrosis of the left one.

Radiotherapy with 50 Gy was again given with little effect. Renal impairment progressed and nephrostomy was performed to save the left kidney in July 1978. His general condition improved after the operation, but the low back pain increased in the summer of 1979 and he subsequently became unable to walk. He was readmitted in November 1980 with severe pain in the back and lower extremities, which was only controlled by epidural morphine infusion. Laboratory data showed further renal dysfunction. Computed tomography revealed that the tumor almost completely filled the pelvic cavity and had spread into the retroperitoneal space (Figure 2).

Angiography could not discriminate the common iliac vessels because of the developed collateral vessels. The venous drainage from the lower extremities was via collateral circulation at the abdominal wall. The pain was now intractable and the patient was transferred to the University Hospital of Tokyo.

The patient was operated on March 6, 1981. The tumor involved the mesenterium of the right side, and it could not be dissected from the surrounding tissues. Therefore, partial removal of the mass and additional curretage were performed. The tumor in the pelvic cavity showed some fibrotic changes probably due to the radiotherapy. After the operation, the severe pain decreased. He returned to the National Sagamihara Hospital for further radiotherapy. However, his general condition deteriorated again in May 1981 and a colostomy was made in June. Renal function decreased and the patient died a month later.

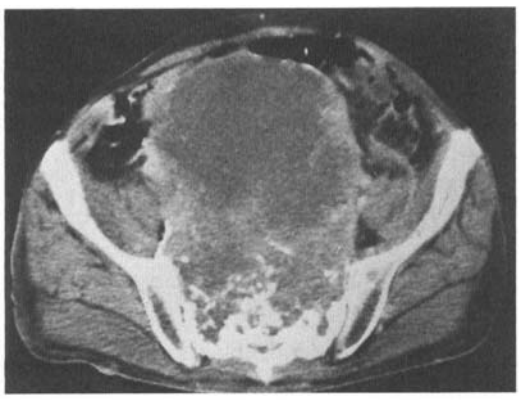

Figure 1. First radiograph, 1971. There is a radiolucent lesion in the sacrum with peripheral sclerosis.

Figure 2. Computed tomography, 1980. Huge intrapelvic expansion of the tumor.

Pathological findings

Autopsy revealed that the tumor originated from the sacral bone and had replaced the upper half of the sacrum and the lower two thirds of the 5th lumbar vertebra. It occupied the entire pelvic cavity and almost the entire right half of the retroperitoneum. It had a well-defined border and its growth was more expansive than invasive.

The tumor compressed both ureters, with bilateral hydronephrosis. Constriction of the large bowel, fibrous adhesion of loops of the small intestine, and partially embedded and narrowed iliac vessels were also found, but no metastases.

The histologic pattern reviewed in the biopsy and found in the operative and autopsy specimens was the same, though the number of the multinucleated giant cells was greatly diminished in the latter. The basic cells were small polyhedral or spherical with clearly defined cell membranes, round or oval nuclei, and clear cytoplasm (Figure 3). Outstanding features were the presence of chondroid areas, which were transitional from the cellular areas, and the presence of spotty calcification. This was more obvious in the newly obtained sample than in the biopsy sample. In special stainings, silver impregnation emphasized the clear cell outlines in the network of reticulin fibrils. The periodic-acid Schiff reaction indicated the presence of glycogen granules in the cytoplasm, and Alcian Blue staining disclosed a chondroid matrix in the intercellular areas as well. There was neither significant cellular atypism nor striking mitotic activity in the tumor cells. These findings supported a cartilaginous origin of the tumor and its benign nature.

Discussion

The differential diagnoses are giant cell tumor, clear cell chondrosarcoma, chondroblastic osteosarcoma, and metastatic deposit from clear cell carcinoma of the kidney or other viscera.

Historically, chondroblastoma has been mentioned as a "variety of giant cell tumor originating in connection with an absorption of misplaced islands of cartilage" by Kolodny (1927) and as a "calcified giant-cell tumor" by Ewing (1928). Codman (1931) separated the tumor, now generally considered to be chondroblastoma, from giant cell tumor of bone under the name of "epiphyseal chondromatous giant cell tumor". Jaffe and Lichtenstein (1942) established the new entity of benign chondroblastoma as a tumor of cartilaginous origin and not a variant of giant cell tumor. Chondroblastoma and giant cell tumor may have common radiographic features, but the initial radiographs in our case showed scattered radiopaque deposits that are quite unusual in giant cell tumors. A decisive point is that our case had an obvious chondroid matrix and round-shaped chondroblastic cells with clearly defined cell membranes. The pattern of the basic mononuclear cells were fairly uniform without malignant signs. Neither osteoid formation nor ossification was detected in spite of the spotty calcification. Thus, chondrosarcoma and chondroblastic osteosarcoma were

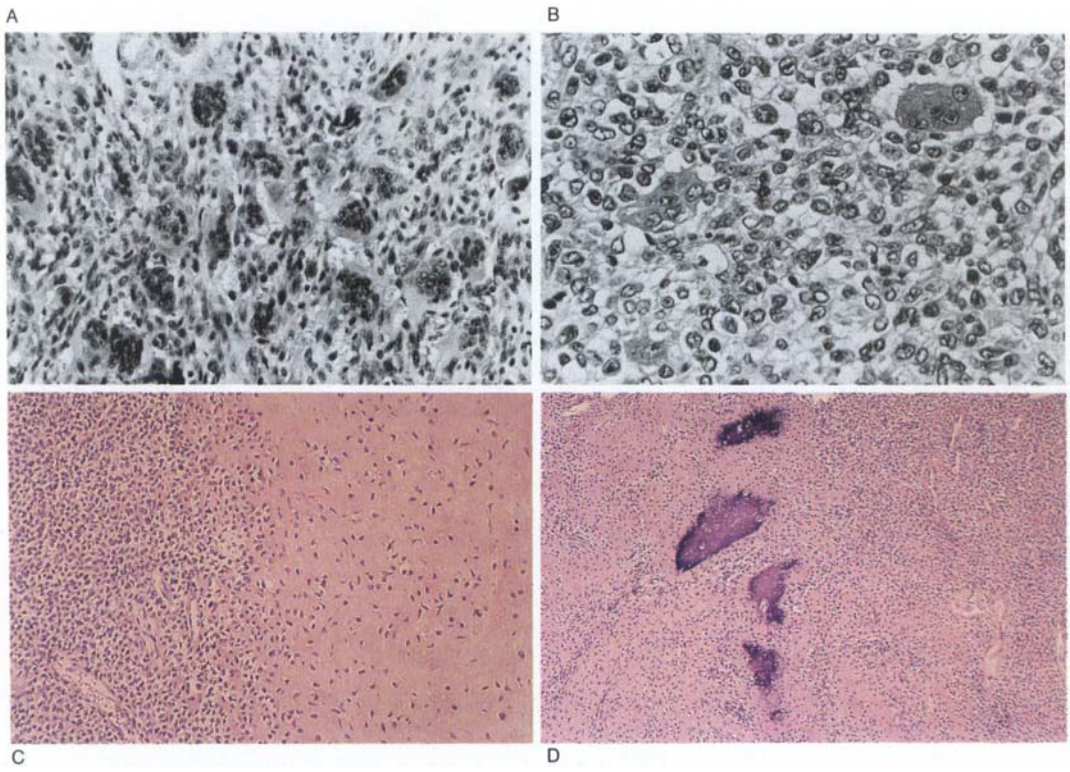

Figure 3. Photomicrographs of tumor specimens.
A. The original biopsy. The cellular pattern is uniform and consists of small cells with round nuclei and multinucleated giant cells. (Hematoxylin-eosin × 150).
B, C. The resected specimens. The structure is essentially similar to that of the biopsy (B, Hematoxylin-eosin × 200) with chondroid matrix adjacent to the cellular area (C, Hematoxylin-eosin × 40).
D. The autopsy material. Spotty calcification and calcific deposits distinctly outline the individual cells. (Hematoxylin-eosin × 25).

ruled out. The possibility of renal cell carcinoma was easily denied because of no carcinomatous changes in the kidneys.

The occurrence of chondroblastoma in the sacrum is extremely rare. The 458 cases reported by Huvos and Marcove (1973), which included 125 cases of Dahlin and Ivins (1972) and 182 by Salzer et al. (1968), included no case with chondroblastoma in the sacrum. McBryde and Goldner (1970), who collected 150 tumors from the English literature, found no sacral case. Feldman (1977) reviewed 691 cases reported in the world literature and found none of sacral origin. To our knowledge, there is 1 case in Ackerman's 100 cases (Ackerman & Spjut 1962), 1 case in Schajowicz's (1978) 115 cases, and 1 case reported by De Souza Andrade et al. (1974). These cases, however, are not separately described.

There are several reports on chondroblastoma with high local recurrence rate, aggressiveness, and even metastatic growth (Huvos 1979, Mirra 1980, Schajowicz 1981). They are sometimes called "malignant" chondroblastoma. Some of these cases have developed self-limited, "benign" lung metastases that have been regarded as iatrogenic implantation.

Wirman et al. (1979) described a case with scapular chondroblastoma that continued to grow slowly in spite of moderate radiosensitivity and had two clinical recurrences. After 34 years the patient finally had contracted pulmonary metastases despite benign histology. Dahlin (1978) also commented on a case with a similar clinical course. Continuous growth may be the natural history of untreated chondroblastoma.

In our case, the possibility of malignant change as being radiation-induced sarcoma was denied; the histologic pattern of the tumor even at autopsy was benign.

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