MALIGNANT TRANSFORMATION OF OSTEOBLASTOMA

R. MERRYWEATHER, J. H. MIDDLEMISS, N. G. SANERKIN

From the Department of Orthopaedic Surgery, Gloucestershire Royal Hospital, the Departments of Radiodiagnosis and Osteo-articular Pathology, Bristol Royal Infirmary, and the Bristol Bone Tumour Registry

A recurrent spinal tumour confidently diagnosed as osteoblastoma eventually terminated as a sclerosing osteoblastic osteosarcoma which metastasised nine years after the onset of symptoms.

Some cases confidently diagnosed as benign osteoblastoma may eventually have a malignant termination (Mayer 1967, 1968, Schajowicz and Lemos 1970; Lichtenstein 1972; Dorfman 1973; Scranton et al. 1975; Seki et al. 1975; Jackson and Bell 1977). So far only two such cases with metastases have been mentioned in the literature (Lichtenstein 1972; Seki et al. 1975).

We report the case of a patient in whom a vertebral tumour was confidently diagnosed as an osteoblastoma at the time of primary excision and in several recurrences but which subsequently developed into a sclerosing osteoblastic osteosarcoma, causing her death with pulmonary metastases nine years after the onset of symptoms.

CASE REPORT

The patient, a girl aged 16 years when first seen in November 1968, had complained of low backache, sometimes radiating to the left leg, during the previous nine months. There was tenderness over the lower lumbar spine and marked muscle spasm which prevented forward flexion. Radiological examination (Fig. 1) showed enlargement and increased density of the left transverse process of the third lumbar vertebra. The affected transverse process was excised in February 1969. A histological diagnosis of osteoblastoma was made (Fig. 2) and it was noted that the lesional tissue extended to the base of excision, indicating incomplete removal.

In July 1970 there was a painful local recurrence, and radiological examination showed dense bony tissue in the soft tissues at the level of the upper border of the third lumbar vertebra. At an exploratory operation in October 1970 a well-defined bony nodule (1.4 x 1.0 x 0.7 centimetres) was removed. Histologically (Fig. 3) it was essentially

Figure 1—Radiograph, taken in November 1968, showing expansion of the left transverse process of the third lumbar vertebra by an osteosclerotic lesion. Figure 2—Histological detail from the lesion diagnosed as osteoblastoma in 1968, with plump osteoblasts forming broad trabeculae of bone. (Haematoxylin and eosin, x160.) Figure 3—Histological detail of the first recurrence excised in 1970, again diagnosed as osteoblastoma. (Haematoxylin and eosin, x160.)
similar to the lesion removed in 1969, although rather more cellular in parts.

In November 1972 radiography (Fig. 4) showed a large sclerotic recurrence in the left paravertebral region, involving the lamina, pedicles, articular processes and part of the body of the third lumbar vertebra. Although there were no neurological signs of any kind, it was felt that the spinal canal and cord could eventually be involved. Accordingly, at a combined orthopaedic and neurosurgical operation in January 1973, the paravertebral mass was removed together with adjacent muscle, fat and fascia. The left lamina of the second lumbar vertebra, as well as the spinous process and left lamina, articular facets, pedicle, and posterolateral angle and part of the body of the third lumbar vertebra were excised until apparently normal bone was reached. The margins of the wound were treated with diathermy, and the cavity was filled in by a one-piece bone graft from the iliac crest and further packed with bone chips. Histologically the tumour removed in 1973 was not greatly different from the material removed in 1969 and 1970 (Fig. 5). Radiotherapy after operation was considered and rejected because of the proximity of the spinal cord and the kidney.

Subsequent radiological examinations showed recurrence and gradual enlargement of the paravertebral tumour (Fig. 6); the patient complained of severe local pain. In a review of the situation in February 1975, it was again decided not to embark on radiotherapy and further extensive surgery was considered inappropriate except for eventual relief of any intractable pain or for compression of the cauda equina.

In October 1975 a fluctuant area which was noted in the upper part of the old operation scar was thought to be an abscess and was accordingly incised. On incision, thin bloody fluid discharged, revealing a cavity in which an obvious piece of ossified tumour (the size
of a walnut) was lying free, and this was removed. In November 1975
the paravertebral recurrence was excised. Macroscopically this was an
ovoid bony mass (5.5 x 4.0 x 3.7 centimetres) covered by a thin layer of
pink-red velvety granular tissue. The histological appearance of the
tumour, both in the paravertebral mass and the nodule removed from
the cutaneous scar, was now that of a sclerosing osteoblastic
osteosarcoma. The tumour showed poor cellular preservation because
of degenerative changes, and extensive areas appeared partly or
wholly necrotic. An irregular network of tumour bone and osteoid,
mostly arranged as a fine filigree, formed the bulk of the lesion.
Because the pattern of the tumour bone conformed to that of a
well-differentiated osteoblastic osteosarcoma, a careful search was
made for supportive evidence of malignancy, and the tumour was
found to have invaded and permeated numerous veins and arteries
(Fig. 7). Evidence of vascular permeation was looked for in all sections
from the previous operations and none was found.

Subsequent management of the patient was by palliative
radiotherapy and liberal administration of analgesics. Her condition
slowly deteriorated with massive local regrowth of tumour (Fig. 8). In
January 1977 pulmonary metastases became radiologically demonstrable (Fig. 9) and death ensued three months later. Necropsy was not
performed.

DISCUSSION

The histological diagnosis of osteoblastoma made on the
original tumour in 1969 and the recurrences in 1970 and
1973 was never seriously in doubt and was unanimously
accepted by all the pathologists on the panel of the
Bristol Bone Tumour Registry. Nevertheless, in view of the
repeated recurrence, the opinion of Professor D. C.
Dahlin was sought early in 1975 and he concurred with
the diagnosis of osteoblastoma, noting that, although the
tumour was rather cellular in places, he did not feel it
could be regarded as malignant.

The diagnosis of osteoblastic osteosarcoma made
later in 1975 was challenged by the radiologists on the
panel of the Registry. A very experienced bone
pathologist on the panel was firmly of the opinion that
this lesion must be regarded as a malignant osteoblas-
toma, one step short of osteosarcoma, and not as a
genuine osteosarcoma. In view of the conflict in
histological interpretation, further opinions were sought
from Professor D. C. Dahlin, Professor F. Schajowicz
and Professor H. D. Dorfman. They all regarded this as
an osteoblastic osteosarcoma which had arisen in an
anteecedent osteoblastoma. In particular, Schajowicz
agreed that the 1975 recurrence did not conform to
"malignant osteoblastoma" as described by him
(Schajowicz and Lemos 1976).

The possibility that, on rare occasions, benign
osteoblastoma may undergo sarcomatous change has
been increasingly recognised since Mayer (1967)
described a benign osteoblastoma of the acetabulum
which recurred several times; transformation into an
anaplastic osteosarcoma occurred nine years later and
the patient died from local extension of the tumour into
the pelvis with ureteric obstruction, without any
demonstrable metastases. Scranton et al. (1975), in a
study of osteosarcoma, recorded a possibly similar case
in which a presumed osteoid osteoma of the femur
recurred twice in two years without any change from the
original histological diagnosis of non-malignancy. A
further recurrence in the second year was reported as
osteosarcoma. Over the succeeding years repeated
excisions of recurrent low-grade osteosarcoma ensued, yet the patient remained asymptomatic and free
of metastases 11 years after presentation. Jackson and
Bell (1977) reported a patient with benign osteoblas-
toma of the rib, whose recurrence seven months later
was diagnosed as osteosarcoma and from which the
patient died two and a half years after presentation when
direct extension into the spinal canal caused compres-
sion of the cord. Mention of several cases, without
documentation, has been made by other authors (Mayer

Only two examples of such cases in which metastasis occurred have been recorded in the litera-
ture. One was briefly mentioned, without documenta-
tion, by Lichtenstein (1972); a patient with benign
osteoblastoma of the distal tibia which metastasised to
the lung seven years after operation. A second case was
recorded by Seki and his associates (Seki et al. 1975), in
which a presumed recurrent osteoblastoma of the femur
underwent malignant transformation to osteosarcoma
and multiple metastases developed 10 years after
presentation. It is important to note that there was no
record of radiotherapy in the case reported by Seki et al.
(1975), and in the present case radiotherapy was only
administered in 1976, after osteosarcoma had superv-
ened. The progression to malignancy in these two
metastasising cases therefore cannot be ascribed to
irradiation.

Jackson and Bell (1977) speculated that rarely an
osteoblastoma might either run a locally malignant
course or convert to an osteosarcoma, or a low-grade
variant of osteosarcoma might in its early stages so
exactly simulate benign osteoblastoma that even
experienced bone pathologists could not distinguish them. These speculations apply equally to the present case. The fact remains, however, that the tumour was originally believed to be an osteoblastoma, and this belief had been supported by several experts on subsequent review. The concept that a variety of low-grade osteosarcoma initially might be histologically indistinguishable from benign osteoblastoma must give rise to serious concern.

The relationship of these cases to the condition described by Schajowicz and Lemos (1976) requires to be clarified. In their report of eight cases of “malignant osteoblastoma” they noted that the tumours displayed the characteristics of genuine osteoblastoma but with an aggressive pattern, with more abundant and often plump hyperchromatic nuclei, greater nuclear atypia and numerous giant cells of osteoclastic type. They regarded this type of tumour as the malignant counterpart of osteoblastoma and found it to be only locally aggressive. In none of their cases did they find features expected in genuine osteosarcoma, and none metastasised. It would seem that “malignant osteoblastoma” must be a different condition from those cases of osteoblastoma which undergo transformation into osteosarcoma.

We would like to thank Professor D. C. Dahlin, Professor H. D. Dorfman and Professor F. Schajowicz for their kind cooperation in this case, Mr M. Findlay and Mr A. Wilson for technical and photographic assistance, and Mrs J. N. Nutt for secretarial help.

REFERENCES


