RECURRENT BENIGN OSTEOBLASTOMA OF THE
SECOND THORACIC VERTEBRA

A Case Report

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Benign osteoblastoma has been recognised as an entity since the papers of Jaffe (1956) and Lichtenstein (1956). Since then it has been reported by many authors (Dahlin and Johnson 1954; Ackerman and Spjut 1962; Marcove and Alpert 1963; Rosenweig, Pintar, Mikail and Mayman 1963: Lichtenstein and Sawyer 1964; Dahlin 1967; Schajowicz and Lemos 1970). However, very little has been written about progressive lesions. This report concerns a patient with a lesion of the second thoracic vertebra who was followed for five years and developed a recurrence.

CASE REPORT

A married woman, aged twenty-six and in late pregnancy, was transferred to the Neurological Service at Sunnybrook Hospital because of the recent onset of paraplegia. She gave a history of pain down the back of the left leg starting in December 1966, which had been ascribed to the pregnancy. The pain was followed by weakness and then numbness of the legs, first the left and then the right. Between Christmas and New Year she became partially paraplegic, with loss of bowel and bladder function but with sensibility to coarse stimuli retained. Labour was induced, and shortly afterwards bowel and bladder function gradually began to return, but not voluntary movement of the lower limbs.

On examination the patient was found to be alert and cooperative but apprehensive. She could not sit without support. Above the level of T.5 the physical examination was negative, apart from diminished chest expansion because of muscle weakness below that level. Both legs were paralysed and spastic, with marked sustained clonus at the ankles and knees. Both plantar responses were extensor. Apart from some sacral sparing, sensibility to pain,
temperature and light touch was absent up to T.10 and impaired up to the level of T.6. Position and vibration sense were absent up to the anterior superior iliac spines.

Radiographs revealed a destructive lesion of the second thoracic vertebra with collapse of much of the body and absence of the left pedicle (Fig. 1). Paraspinal soft-tissue masses extended mainly to the left of T.2 but also slightly to the right. Two ring-like calcifications were noted in the mass on the left. Myelography showed an extradural block at the level of T.2 (Fig. 2).

First operation—In March 1967 laminectomy was performed from C.7 to T.4. An extradural mass was identified anteriorly, maximal at the T.1 level. It was thought to be malignant. Specimens were taken for biopsy. The dura was opened in order to decompress the cord fully. After this operation the patient soon recovered posterior column sensibility but the motor paralysis remained complete.

Second operation and progress—The biopsy report was benign osteoblastoma. Because no motor recovery had returned, the cord was decompressed anteriorly through a costo-transversectomy approach from the left side two weeks after the first operation. The transverse processes and pedicles of T.2 and T.3 were removed; the tumour was exposed and resected entirely, so far as could be judged macroscopically. After this decompression of the cord, myelography showed passage of the contrast media above T.2, but with an extradural defect extending from T.2 to C.7 (Fig. 2). In the antero-posterior and lateral tomograms there were shadows suggesting calcification in both paraspinal soft-tissue masses.

The patient made an uneventful recovery from this operation and full motor power gradually returned. She remained completely asymptomatic for five years and at yearly reviews the only objective finding was a minor degree of spasticity in the lower limbs.

Later history—After five and a half years, pain in the back of the legs returned, similar to the original pain. On examination a mild Horner’s syndrome was noted on the left side, and possibly an increase in the spasticity of the lower limbs with equivocal extensor plantar responses. Radiographs of the thoracic spine showed a large well-defined mass of uneven density extending into the left upper thorax anteriorly, mainly lateral to the second thoracic vertebra (Fig. 3). Tomography (Fig. 4) showed the main dense lesion to be distinct from the
spine except at the lateral margin of T.2, to which it was connected by a thick pedicle with
the density of bone, spreading out like a mushroom into the thorax.

**Third operation**—For removal of the recurrence a transthoracic approach was selected because
of the radiological appearance of an anterior bony mass and the previous posterior approach.

**FIG. 3**
A radiograph of the chest taken in 1972 showing a large mass in the left upper
chest in relation to the upper thoracic vertebrae.

**FIG. 4**
A frontal tomogram of the lesion showing the attachment of the pedicle to
the body of T.2.

A postero-lateral thoracotomy incision was made from the upper angle of the left scapula, parallel
to the spinous processes, down to the level of C.7. The scapula was then "winged" forward
by dividing the trapezius and rhomboid muscles close to their spinal origins. This allowed
excellent exposure of the upper costo-vertebral articulations. The second, third and fourth
ribs were excised from the mid-axillary line to the bodies of the vertebrae. The pleural cavity
was entered and the anterior aspect of the upper thoracic spine, particularly T.2, was well
displayed. The tumour was a large, hard mass measuring $10 \times 8 \times 5$ centimetres, based on a
wide pedicle extending from the lateral aspect of the second thoracic vertebra (Fig. 5). It was
completely excised together with the anterior half of the second thoracic vertebra. Rib grafts
were used to fill the cavity. The convalescence from this third operation was uneventful.
Photograph taken at the time of thoracotomy in 1972, showing the lesion (arrowed) extending into the left upper pleural cavity. From left to right at the level of the arrow, the structures to be seen are the scapula winged forward, the apex of the lung held retracted by a sponge holder, the lesion itself, and the distorted bed of the second rib. A small triangular segment of the arch of the aorta can be seen about a centimetre down from the arrow.

Histological appearances of the first biopsy specimen in 1967. Large pieces of osteoid lie in cellular connective tissue. There are many giant cells. (Haematoxylin and eosin, × 80.)
Another view of the first biopsy, showing trabeculae of woven bone in similar connective tissue. (Haematoxylin and eosin, ×32.)

Histological appearance of the recurrent tumour resected in 1972, showing numerous thin, incompletely mineralised trabeculae and broad trabeculae of woven bone with irregular cement lines. No cellular connective tissue is seen. (Haematoxylin and eosin, ×16.)
PATHOLOGY

Numerous small fragments of brownish red tissue submitted from both operations in 1967 showed histological appearances typical of benign osteoblastoma, with marked variations in different regions. In some places the resemblance to giant-cell tumour was complete; in most of the tissue broad osteoid seams lay in cellular stromal tissue in which osteoblastic giant cells were prominent (Fig. 6); in other places thick trabeculae of woven bone lay deep within the lesion (Fig. 7). The tumour had broken through the cortex at several sites, giving rise to a slight reactive bone formation.

Piecemeal removal of the tumour in 1972 again produced numerous small fragments of gritty tissue, some yellow, some greyish pink. The histological appearances had changed markedly. Much of the tissue consisted of broad trabeculae of mature woven bone, often with prominent, irregular cement lines, adjacent to foci of much less mature woven bone in which osteoblastic and resorptive activity could be seen (Fig. 8). The stroma consisted of scanty loose connective tissue with infrequent cells, none of which resembled the earlier stromal cells. Two other types of tissue were present—large foci of dense fibrous tissue, and foci of necrotic osteoid which was acellular and quite loose in structure. Foamy histiocytes were present in both these tissues.

DISCUSSION

Most reports on benign osteoblastoma indicate that simple curettage or excision of the tumour results in cure (Lichtenstein 1956, Ackerman and Spjut 1962, Lichtenstein and Sawyer 1964, Schajowicz and Lemos 1970). Radiation therapy has been used alone or in conjunction with surgery (Giannestris and Diamond 1958, Ackerman and Spjut 1962, Rosensweig and his colleagues 1963, Lichtenstein and Sawyer 1964, Schajowicz and Lemos 1970). Only one recurrence has been described in detail (Lichtenstein 1956, Lichtenstein and Sawyer 1964).

Pochacewsky, Yen and Sherman (1960) reviewed thirty cases from the literature and four more cases in an attempt to define the roentgenologic features of benign osteoblastoma. They found the purely lytic lesion to be very uncommon and recognised only one case. They describe the characteristic radiological features of benign osteoblastoma as a predominantly osteolytic expanding lesion with evidence of some bone formation or varying degree of calcification. Penetration of the cortex and a soft-tissue mass is common, and the mass is usually outlined by a rim of calcification. After radiation the lesion tends to become more densely calcified. Of lesions involving the vertebra, the neural arch was the commonest site, and they said that only when the tumour reached "considerable" size was the vertebral body involved.

This case presented many radiological features of the so-called characteristic case described by Pochacewsky and his colleagues. For example, there was a soft-tissue mass with penetration of the cortex, involvement of the pedicle and a mainly osteolytic lesion with calcification within the tumour mass. The marked involvement of the vertebral body associated with bilateral masses suggests that the site of origin in this case was the body of the vertebra. The importance of tomography in showing areas of calcification within the lesion, not seen in plain views, was evident in this case and can be of value in differential diagnosis. Tomography was also important in the pre-operative assessment of this recurrent lesion, demonstrating no bony lesion within the spinal canal and the attachment of the main lesion to the body of T.2 by a lateral pedicle.

The histological appearances of the recurrence reported here obviously differ from those of the only previous case, of which unfortunately there is no published photographic record (Lichtenstein 1956). Dahlin and Jaffe both regard extensive calcification as an indication of maturation of the lesion, though neither author supports this view with tissue obtained on more than one occasion from any patient. Several authors have reported extensive calcification after radiotherapy (Golding and Sissons 1954, Kirkpatrick and Murray 1955, Jaffe 1956), but
there are no histological studies to determine whether this is due to reactive bone around the lesion or to neoplastic bone within it. Marcove and Alpert (1963) found that peripheral sclerosis was sometimes caused by tumour bone, but they do not appear to have obtained serial specimens.

There is no doubt here that extensive new bone formation had occurred within the recurrence. Indeed, the differences in histological appearance of the two specimens are sufficient to raise the question whether the two lesions are related. The only alternative diagnosis that may be considered seriously is that the second lesion represents ossification of a haematoma formed after the second operation. The absence of blood pigments, of cholesterol crystals, and of any evidence of organisation, and the presence of considerable quantities of osteoid, are all against that diagnosis. The appearances are indeed those which could be expected from increasing maturation of a benign osteoblastoma.

**SUMMARY**

1. A case is reported of a benign osteoblastoma of the body of the second thoracic vertebra causing paraplegia in a woman aged twenty-six.
2. The tumour was resected, apparently entirely, through a costo-transversectomy approach, and the paraplegia resolved almost completely.
3. Five and a half years later symptoms recurred, due to a recurrence in the form of a large, partly calcified tumour in the left upper thorax which was resected in toto via a transpleural approach.
4. The considerable histological differences between the original tumour and the recurrence are discussed.

**REFERENCES**


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