AGGRESSIVE OSTEOBLASTOMA

A CASE PREVIOUSLY REPORTED AS A RECURRENT OSTEOID OSTEOMA

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We review the case of a 58-year-old man with a benign osteoblastic lesion. This originated in the base of the right second metacarpal and eventually involved several adjacent bones, persisting for at least 27 years despite 11 operations. It was originally reported in the literature as a recurrent osteoid osteoma, but we believe it is more properly diagnosed as an aggressive osteoblastoma, since the histological pattern did not change over the years. The lesion has remained locally aggressive with no evidence of malignant characteristics.

There is continuing debate in the literature regarding recurrent aggressive osteoblastic lesions with 'epithelioid' osteoblasts, as to whether these represent benign, locally aggressive lesions (Dorfman and Weiss 1984) or osteosarcomas (Bertoni et al. 1985). The single patient who is the basis for this review has a 30-year clinical history of such a lesion and was first reported in 1970 as a recurring osteoid osteoma (Dunlop, Morton and Elliott 1970).

Since then the patient has had further local recurrences; we have reviewed the pathological material from 11 surgical excisions over 21 years. During this period the pathological process has been unchanged, consisting of a proliferative osteoblastic lesion containing 'epithelioid' osteoblasts.

CASE REPORT

A man, then aged 29 years, first developed pain in 1958, and a ganglion was removed from the dorsum of his right hand in the following year. One year after this, increased density of the second metacarpal was evident (Fig. 1), but a diagnosis of osteoid osteoma was not made until 1965 when radiographs showed an area of radiolucency with at least one central nidus of radiodensity in the base of the right second metacarpal (Fig. 2). An en bloc excision with replacement by an autogenous iliac bone graft was performed. The microscopic appearance (Fig. 3) was considered at the time to be consistent with the clinical and radiological diagnosis of osteoid osteoma, though on review there are some compressed seams of epithelioid osteoblasts.

Pain was relieved for about five months, but then returned, and over the next three years two wider excisions were carried out. The specimen from the third excision showed at least three tumour nodules with well-defined reactive bone trabeculae and surrounding loose stroma looking like granulation tissue (Fig. 4). One year later, in 1969, the patient complained of a tender nodule on the dorsum of the hand. This was excised, and seemed to be attached to the bone by a 'synovial' pedicle. The histological appearance was unchanged; it was considered consistent with the original diagnosis of osteoid osteoma.

Pain and tenderness persisted, but it was felt more in the palm near the base of the thenar eminence. Arteriography showed wide involvement of bone, with an abnormal vascular pattern and pooling of contrast material. The fifth operation, in 1970, was a wide excision of involved bone, including the shafts of the second and third metacarpals, their bases and the distal half of the capitale. The adductor pollicis muscle was partially excised together with additional nodules of apparent tumour. The defect was replaced by a block of iliac bone; and four months later this appeared to be satisfactory. The histology was unchanged.

Pain recurred after only 16 months, but the patient decided to accept this, gaining some relief with aspirin and a lighter job. A further exploration of the involved area in 1974 revealed nothing significant. One year later, despite satisfactory radiographs (see Figure 5), the graft was re-explored and found to be intact. A second incision at the base of the thenar eminence revealed a soft 1 cm mass of tumour tissue in the remaining adductor pollicis, and two months after this another tender nodule was...
Figure 1 – Radiograph in 1960, showing the increased density of the second metacarpal.

Figure 2 – In 1965 there is an area of radiolucency at the base of the second metacarpal with a dense punctate nidus.

Figure 3a – Growth pattern of a typical specimen showing a sclerotic tumour nidus surrounded by a cellular zone with widely separated osteoid trabeculae or foci and absence of osteoid (Haematoxylin and eosin x 50). Figure 3b – The sclerotic nidus, consisting of sheets of osteoid with compressed seams of epithelioid osteoblasts and occasional entrapped osteoblasts (Haematoxylin and eosin x 80).

Low-power view showing a tumour focus surrounded by reactive sclerosis. There are well-defined reactive bone trabeculae surrounded by loose granulation tissue which extends slightly into the surrounding sclerotic margin (Haematoxylin and eosin x 18).
excised from the palm. Both nodules had the familiar histological appearance.

This time there was more lasting relief from pain, but in 1980, four years after the eighth operation, it recurred and increased with an area of exquisite tenderness 2.5 cm in diameter in the palm at the base of the thenar eminence. Radiographs now showed a lytic lesion in the proximal part of the bone graft (Fig. 6) and tomograms showed that the posterior cortex of the graft was intact. This was confirmed surgically, when a 2.5 cm diameter defect was found. The defect was incompletely filled by two types of tissue: one tough and rubbery, the other friable granulation tissue as in previous specimens. The lesion was curetted thoroughly: no bone graft was added.

Pain and tenderness was relieved for two years. It then recurred, though it was adequately relieved at first by 600 mg of aspirin daily in divided doses; but after another year the patient requested a further operation, his tenth. Curettage of the lytic defect, which contained sero-sanguinous fluid and a small amount of rubbery or friable soft tissue, produced gratifying, though incomplete, relief of pain.

Three years after this, in 1987, pain was again an increasing problem, requiring stronger analgesics and interfering with work and leisure. Both radial and ulnar borders of the hand had become exquisitely tender, and the extent of soft tissue involvement seemed to preclude hand-sparing resection. Radiographs showed further extension of the lesion (Fig. 7) and in the hope that curettage might give some relief, the eleventh operation was performed. The lesion was distinctly more vascular and its extension was evident. Histology (Fig. 8) clearly showed the epithelioid osteoblasts which are typical of aggressive osteoblastoma (Dorfman and Weiss 1984). On review, these were present in the original resection material of 1965.

After this eleventh operation pain returned within a matter of months and disability increased. Amputation was offered but rejected by the patient. He was therefore given a course of radiotherapy to the right hand and wrist.
consisting of 5000 CGy central dose in 20 fractions over four weeks, completing his treatment in March 1988. When seen most recently in November 1988, relief of pain had persisted and function remained satisfactory.

DISCUSSION

The initial presentation of this lesion led us to diagnose an osteoid osteoma, and the histological identification of benign osteoblastic tissue seemed to confirm this. The subsequent behaviour forced a reconsideration. The ultimate size, frequent recurrence, the soft tissue involvement, and the aggressive behaviour of the tumour, all raised the question that the diagnosis was more likely to be osteoblastoma or even osteosarcoma. However, the histological characteristics of malignancy were never seen and no evidence of metastatic disease has been detected.

Our case does not appear to be one of osteoid osteoma with multicentric nidus (Glynn and Lichtenstein 1973). Rather, we believe it is best described as an osteoblastoma, the name proposed independently by Jaffe (1956) and Lichtenstein (1956) to designate 'a rather vascular osteoid and bone-forming benign tumour characterised cytologically by the abundant presence of osteoblasts'. Though Mayer (1967) and later Seki et al. (1975) reported malignant transformation of benign osteoblastoma, and Schajowicz and Lemos (1976) described eight cases of malignant osteoblastoma, we do not consider that our case is truly malignant in the absence of distant metastases.

For the same reason, we do not consider our case to be an 'osteosarcoma resembling osteoblastoma' (Bertoni et al. 1985). Instead, the concept of Schajowicz and Lemos (1970) which relates osteoid osteoma with osteoblastoma, and allows some overlap in the diagnosis, helps to explain some features of our case. Borderline osteoblastic lesions are further discussed by Dorfman and Weiss (1984); they propose three categories of benign tumour: osteoid osteoma, osteoblastoma, and aggressive osteoblastoma. This article led us to review all the material from our patient. The basic histological pattern in all instances consisted of one or several dense osteoid niduses surrounded by cellular zones with characteristic large 'epithelioid' osteoblasts. These show an eccentrically positioned nucleus, a prominent nucleolus and abundant clear or finely granular eosinophilic cytoplasm (Fig. 8). In some areas these cells are associated with small osteoid seams; in others they form sheets of cells devoid of osteoid. Occasional mitoses, less than one per 20 high-power fields, could be identified in all specimens, but did not increase in relative numbers over the years. No abnormal mitoses were seen.

In recent specimens the tumour has seemed more like an aneurysmal bone cyst which would be consistent with the radiological appearances in Figure 7. Within cellular areas, small osteoclast-like cells are present in moderate numbers, sometimes in association with osteoid seams, but these cells do not show resorption activity and their multiple nuclei do not have an epithelioid appearance. In some places the sheet-like zones of osteoid exhibit mineralisation and, rarely, necrosis. The basic pattern of dense osteoid with associated cellular zones is separated from the surrounding reactive sclerotic margin by a zone of spindling fibroblasts and vascular stroma resembling granulation tissue. This extends between the non-tumorous trabeculae for 1 or 2 mm and is associated with bone resorption. Otherwise, the spindle cell component is minimal.

Although in our case the tumour has been locally aggressive, with multiple recurrence and extension to adjacent bones and soft tissues, its histological features have remained unchanged over 22 years and there have been no distant metastases. It should therefore, best be classified as an aggressive osteoblastoma (Dorfman and Weiss 1984) rather than a low-grade osteosarcoma.

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REFERENCES


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