PRIMARY OSTEO-LIPOSARCOMA OF BONE (MALIGNANT MESENCHYMOMA)

Report of a Case

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Although some pathologists have doubted the existence of primary intraosseous liposarcoma and consider it to be either metastatic or an extension from a tumour arising in adjacent soft tissues, a number of acceptable cases have been reported, notably those of Dawson (1955) in the lower end of the left femur of a woman of twenty-eight, and of Catto and Stevens (1963) in the upper end of the right tibia in a girl of sixteen. The latter authors reviewed the world literature and could find only fifteen cases described as primary liposarcoma of bone. They considered that only Dawson's case was completely convincing. To these can be added one of the two cases reported by Mastragostino (1957) and those of Honore, Rogister and Delvigne-VanLancker (1963) and Goldman (1964).

The tumour reported below is unusual because in addition to lipoblastic tissue it produced neoplastic bone and should therefore be regarded as an osteo-liposarcoma. A similar case, arising in the tibia of a youth of seventeen, has been reported by Schajowicz, Cuevillas and Silberman (1966) and described as a malignant mesenchymoma.

CASE REPORT

A boy of fifteen was seen with pain and swelling of the outer side of his right knee which he had had for two months after kicking at a tennis ball. Radiographs showed an expanding lesion of the upper end of the shaft of the right fibula with some periosteal reaction and a pathological fracture (Fig. 1). The differential diagnosis was considered to be between osteomyelitis, Ewing's tumour or osteogenic sarcoma. Biopsy suggested liposarcoma. A chest radiograph was normal at this time and excision biopsy was done.

Pathology—The specimen contained a yellow friable tumour 7.5×5×5 centimetres through the middle of which ran the upper third of the fibula, which showed a pathological fracture below the head. The tumour was soft and infiltrating the surrounding muscles. Several small protrusions of growth could be seen in the medial dissected surface but the joint was not involved. No local or general ossification was detectable.

Histology—The tumour cells were fairly uniform, large and polygonal, or occasionally round or elongated, with a moderate amount of vacuolated granular eosinophilic cytoplasm and large nuclei stippled with chromatin, having prominent nucleoli (Fig. 3). A few giant or bizarre

FIG. 1
Radiograph of right fibula.

forms were seen and mitoses were fairly frequent. Here and there groups of osteoclasts could be seen. There were foci of necrosis though the tumour was well supplied with blood vessels. Frozen sections showed a large amount of isotropic lipid in the cells (Fig. 2).

The diagnosis was in doubt because of some resemblance to clear-celled adenocarcinoma of kidney (Fig. 4), but the kidneys were radiologically normal and further blocks of tissue

FIG. 2
Section of tumour. (Frozen. Oil red O, showing fat, $\times 190$.)

FIG. 3

FIG. 4

Figure 3—Section of tumour. (Haematoxylin and eosin, $\times 190$.) Figure 4—Section of tumour showing resemblance to clear-celled adenocarcinoma of kidney. (Haematoxylin and eosin, $\times 190$.)

were considered to show the features of liposarcoma. In view of the involvement of the dissected surface, indicating incomplete removal of the tumour, the patient was treated by cobalt teletherapy using opposed fields 24×10 centimetres, under general anaesthesia with the limb anoxic, a tourniquet having been applied for thirty minutes at 600 millimetres of mercury beforehand. He had two treatments in eight days with a tissue maximum dose of 9,000 rads, giving a mid-tumour dose of 8,550 rads and minimum primary dose of 7,250 rads.

FIG. 5

FIG. 6

Figure 5—Frozen section of lung showing abundant fat. (Oil red O, $\times 190$.) Figure 6—Section of lung metastasis showing osteoid tissue. (Haematoxylin and eosin, $\times 190$.)

FIG. 7

FIG. 8

Figure 7—Section of muscle metastasis showing osteoid tissue and early ossification. (Haematoxylin and eosin, $\times 190$.) Figure 8—Section of primary tumour showing osteoid tissue and early bone formation. (Haematoxylin and eosin, $\times 190$.)

A chest radiograph taken two months later showed metastases. From then on the patient's general condition deteriorated and the lung metastases quickly increased in size and number. He suffered from gross dyspnoea and occasional haemoptyses and died after five months.

Necropsy—This was performed eleven hours after death. The main findings were those of radionecrosis and residual tumour in the original site; massive lung metastases (some of which were yellow and fatty whilst others were necrotic and haemorrhagic); a single small deposit in the ileal mucosa; a metastasis in the right adrenal gland; and small metastases in the left rectus abdominis, vastus medialis and psoas muscles and in the right psoas and teres major muscles. Some muscle deposits showed gritty foci thought to be due to calcification or ossification. No soft part primary tumour was found and none in other bones or viscera.

Histology—Many of the metastases showed necrosis, particularly in the lungs. Where they were well preserved the appearances resembled those of the primary growth, though in some places there was much more anaplasia, with bizarre and giant tumour cells. Plentiful lipid was seen in non-necrotic tumour cells, well shown in the adrenal glands and lungs (Fig. 5). In addition, a number of them, especially in the muscles, showed osteoid tissue and/or bone formation (Figs. 6 and 7), and in places a little cartilage. This bone and cartilage was initially regarded as reactive but on more critical examination and subsequent review of the sections by the Bone Tumour Panel of the British Empire Cancer Campaign and Pathological Society of Great Britain and Ireland, it was considered to be neoplastic and produced by modified tumour cells which closely resembled osteoblasts. The original tissue was re-examined and further blocks made: in a few places microscopic foci of unquestionable neoplastic osteoid tissue and bone were detected (Fig. 8), these areas having a histological structure identical with that of osteosarcoma. The bulk of the tumour bone was of intramembranous type but an occasional focus of cartilage was found.

DISCUSSION

To be accepted as primary, the tumour must be unequivocally arising in the interior of the bone and there must be no other source of liposarcoma. Histologically the tumour cells must have the appearance of lipoblasts. In the present case the radiological appearances were those of an intraosseous tumour and this was confirmed by examination of the excised specimen. The growth was manifestly arising in the upper end of the shaft and surrounded by a concentric mass invading the neighbouring muscles. The medullary cavity of the fractured ends was full of growth. We have seen a number of soft part liposarcomas in close proximity to bone and none resembles the present case in this respect. Reszel, Soule and Coventry (1966) in a review of 222 cases of liposarcoma involving the limb girdles and extremities encountered none destroying bone. Histologically the tumour was composed of rather uniform lipoblastic cells containing large or small drops of fat. No myxoid or spindle-celled foci were found. There was a delicate reticulin network surrounding small groups and individual cells. The more vacuolated parts had a superficial resemblance to clear-celled carcinoma of kidney: other parts were of a more hibernomatous appearance, though in a few places the tumour cells were smaller and lacking in fat. The number of mitoses and poor differentiation suggested high malignancy and the patient survived only nine months after diagnosis and excision.

The cases of Dawson (1955) and Catto and Stevens (1963) also were fatal within a few months. The patients of Honore *et al.* (1963) and Goldman (1964) were followed up for only a few months before publication of their papers but personal communications revealed that Goldman's patient was still alive and free from recurrence or metastases three years later, whereas that of Honore *et al.* died within a year of operation with metastases to the lungs. Mastragostino's patient (1957) was alive and well four years after excision of a tumour in the right upper fibular shaft (in a man of thirty).

The present tumour is particularly interesting because of its dual nature, and one can only explain this by invoking the concept of the intermutability and totipotentiality of mesenchyme (Willis 1960). In retrospect it would appear that amputation might have been preferable to radiotherapy because the latter failed to eradicate the remains of the primary growth, though it is extremely doubtful if the ultimate fatal outcome would have been influenced.

SUMMARY

1. A case of primary intraosseous liposarcoma is described which was producing tumour bone (osteo-liposarcoma: malignant mesenchymoma) in the right fibula of a boy of fifteen.
2. Death occurred from pulmonary metastases nine months after excision and cobalt teletherapy.
3. Only one similar case has been reported.

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