Acta orthop. scand. *46*, 869–876, 1975

Department of Orthopaedic Surgery and Department of Pathology,
University of Umeå, Umeå, Sweden.

# PRIMARY LIPOSARCOMA OF BONE

SVEN-ERIK LARSSON, RONNY LORENTZON & LENNART BOQUIST

Accepted 13.ii.75

Liposarcoma is not uncommon among soft tissue sarcomas, comprising approximately 8 per cent according to a recent report (the Swedish Cancer Registry, Incidence Report, 1971). In contrast, liposarcomas primarily arising in bone are extremely uncommon despite the fact that the bone marrow is rich in adipose tissue. Catto & Stevens (1963) reviewed the world literature and could find only 15 cases which were interpreted as primary liposarcoma of bone. Only one case, that reported by Dawson (1955), was considered completely convincing. To this can be added reports by Honore et al. (1963), Goldman (1964) and Mastragostino (1965).

The following case is presented because of the rarity of this neoplasm and because it is still a matter of debate whether or not this lesion represents a clinically well defined entity. The finding at primary surgical intervention of a tumour located entirely within bone with no other primary locus and which consisted of undifferentiated lipoblasts indicates that this case represents a genuine primary liposarcoma of bone.

## CASE REPORT

### *Clinical, roentgenological and operative findings*

A 52-year-old female was admitted on January 17, 1958, because of nausea, subfebrility and a weight loss of 8 pounds during the previous 4 months. She complained of steadily increasing pain in the region of the proximal left femur and had a left-sided limp.

Physical examination on admission was normal except for pain on application of local pressure on the major trochanter of the left femur. Motion of the hip joints was normal and Laségue's sign was negative. No tumours were noted on careful examination of the thigh and hip region, or elsewhere, and there was no lymphadenopathy. Routine laboratory examinations of blood and urine were normal.

Roentgenograms of the left hip showed an osteolytic lesion of approximately

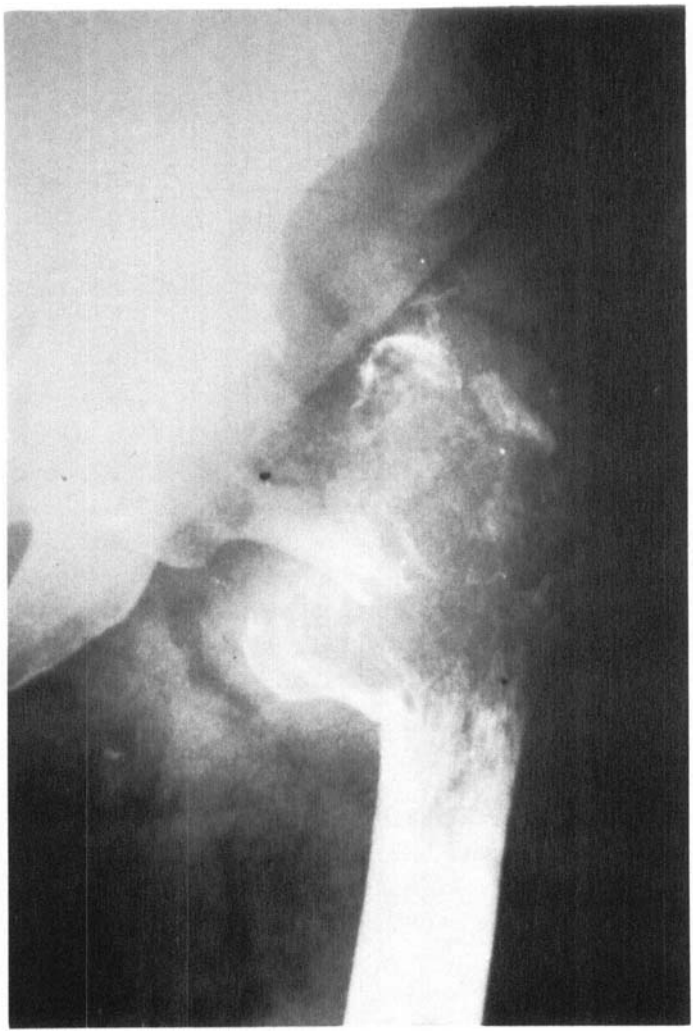

*Figure 1. Roentgenogram of the proximal left femur taken 29 days after admission demonstrating a very rapid progress of osteolytic destructive changes within the proximal femur with a spontaneous intertrochanteric fracture.*

2 inches in diameter in the major trochanter of the femur. The destruction was located entirely within the bone with disappearance of spongious bone and some erosion of the adjacent cortical bone. Roentgenograms of the lungs, stomach and duodenal bulb were normal.

At surgical biopsy of the lesion the cortex of the major trochanter was found to be completely intact. After fenestrating the cortical bone, a relatively small, soft lesion was found located entirely within bone with no extraosseous component

whatsoever. On histopathological examination of the curetted specimen no definite diagnosis was reached but lipoidosis and osteomyelitis were considered.

Postoperatively, the patient had progressively increasing, severe pain from the region of the lesion and roentgenograms of the left hip showed local progression of the osteolytic destruction. Roentgenograms of the lumbar spine, sacrum, pelvis and skull were normal.

On February 6, another operation was performed with curettage of the lesion. Bacterial cultures on tissue specimens from the lesion and on blood samples were negative. On histopathological examination, the diagnosis of liposarcoma primarily arising in bone was made.

On February 15, roentgenograms of the left hip showed a spontaneous fracture through the major trochanter (Figure 1) which had occurred despite treatment in tibial traction. Radiotherapy was instituted, and over a period of 25 days a total dose of 4,500 rad was given towards the ventral region of the left hip. In spite of this treatment, roentgenograms showed a further progression of the lesion and no healing of the fracture. The patient's condition went steadily downhill, the neoplastic tissue showed growth through the operation wound and spread of the tumour occurred to the left iliac fossa. On June 26, 1958, approximately 5 months after the first admission, the patient died in pulmonary insufficiency because of widespread metastases in the lungs.

*Necropsy*

At post-mortem examination, tumour tissue was seen to bulge up through the wound in the left trochanter region. The proximal left thigh was completely infiltrated by a necrotic tumour that had invaded the muscles and subcutaneous tissues. The whole proximal part of the femur was completely destroyed by the tumour. The left ilium was intact. Grossly, the tumour was greyish-white, and had a partly soft, partly more firm, fibrous consistency. Within the tumour mass only a few small bone fragments remained of the destroyed proximal femur.

The lungs exhibited miliary metastases and oedema. The liver and the left kidney contained several metastases of varying size. There were also metastases in the retroperitoneal lymph nodes. All the other visceral organs were carefully examined and found to be normal.

*Microscopic findings*

Histopathologic specimens taken at biopsies and at post-mortem examination were re-examined and additional slides were made at the time of re-examination of the case. The following stains were used: Haematoxylin-eosin, van Gieson's stain, Sudan black B, periodic acid Schiff (PAS), Laidlaw's silver impregnation and phosphotungstic-acid haematoxylin (PTAH).

On review of the sections, the diagnosis of liposarcoma of the bone could be substantiated. The primary tumour in bone exhibited considerable polymorphism (Figure 2). It consisted mainly of large cells with vacuolated and empty-looking cytoplasm (Figure 3) with an admixture of small round cells. The nuclei of the large tumour cells were irregular, often hyperchromatic and of varying size (Figure 4). In some nuclei enlarged nucleoli could be discerned. The cell membranes were distinct. Many large tumour cells were rounded, others were irregular. They were

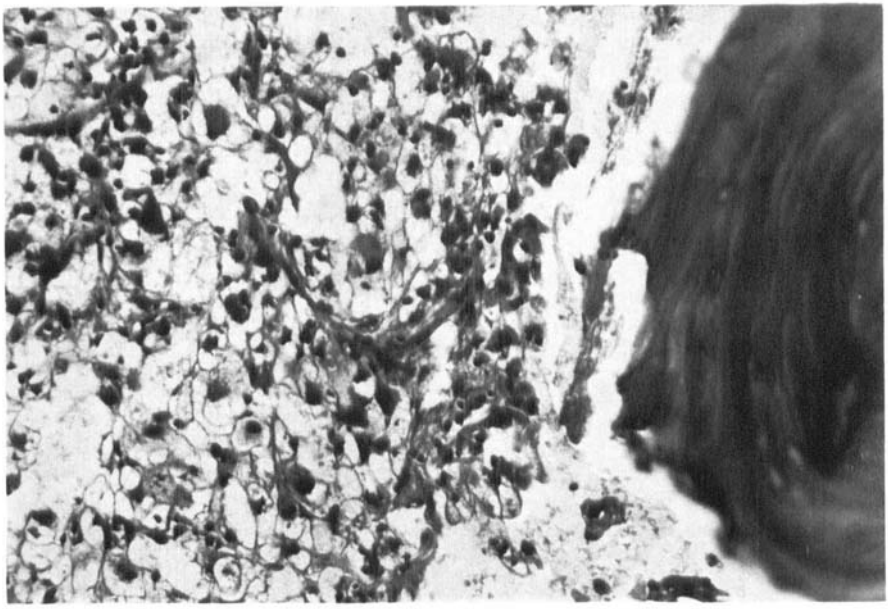

*Figure 2. Photomicrograph of a primary liposarcoma of the bone showing polymorphism of the tumour cells and portion of a bone trabecula. Haematoxylin & Eosin, × 80.*

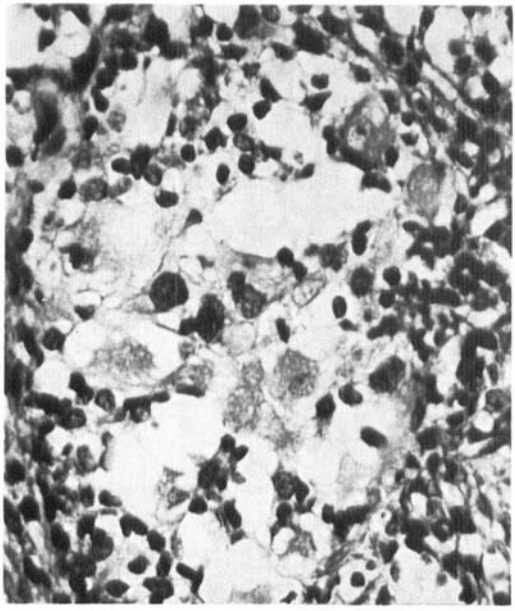

*Figure 3. Liposarcoma of the bone showing large tumour cells with vacuolated cytoplasm and small round cells. Haematoxylin & Eosin, × 100.*

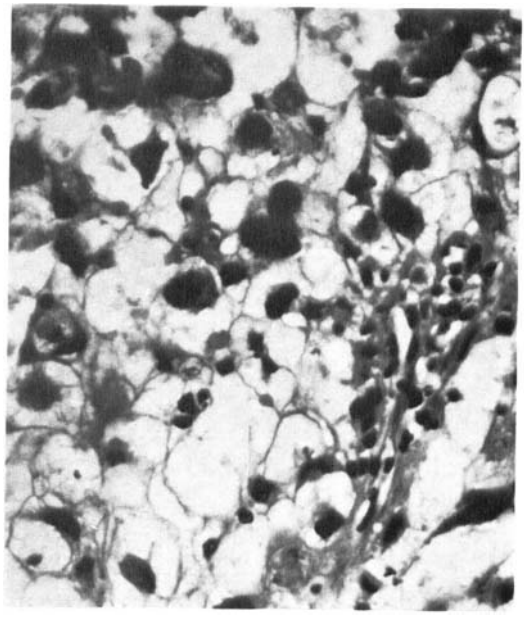

*Figure 4. Another portion of the liposarcoma demonstrating tumour cells with irregular, hyperchromatic nuclei, an empty-looking cytoplasm and distinct cell boundaries. Haematoxylin & Eosin, × 100.*

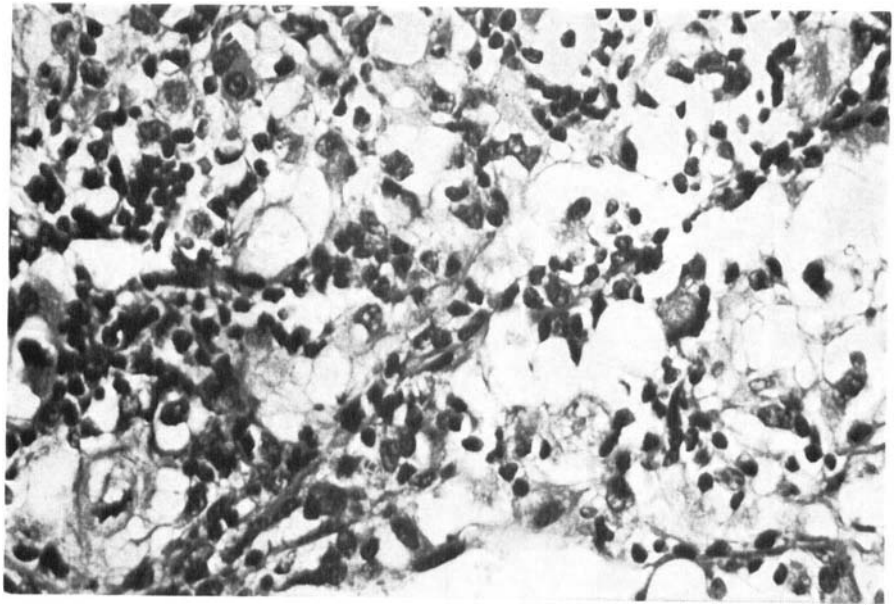

*Figure 5. Another view of the liposarcoma showing a mitotic figure in a large tumour cell (lower left corner) and streaks of small round cells. Haematoxylin & Eosin, × 80.*

strongly positive for fat. Mitotic figures, occasionally of atypical appearance, were seen among the tumour cells.

The general growth pattern of the tumour cells was that of solid sheets with no tendency to acinar arrangement or spinocellular differentiation. A moderately developed and moderately vascularized stroma of connective tissue was observed. Regressive changes, frank necroses and bleedings were seen in the tumour tissue.

The masses of tumour cells infiltrated the areas between the bone trabeculae. The latter were occasionally more or less destroyed and sometimes the bone seemed to have completely disintegrated. The microscopic appearance of the metastases in liver, left kidney, lungs and lymph nodes showed a similar picture.

The histopathological details of the liposarcoma of the bone did not seem to deviate from those seen in liposarcoma primarily localized in the soft tissues.

## DISCUSSION

The above-mentioned liposarcoma was obviously primarily localized in bone. There was no evidence of overgrowth on bone of any liposarcoma arising in the adjacent soft tissues, which can occur in rare instances (Ressel et al. 1966). This was evidenced by X-ray examinations and thorough surgical exploration. Histopathologically, the tumour was strongly positive for fat and was composed of large cells with vacuolated cytoplasm and irregular, hyperchromatic nuclei of varying size and also of small round cells. Because of the variation in the histopathological appearance of these tumours there can be difficulties in the differential diagnosis especially towards other sarcomatous tumours of myxoid, lipomatous and fibrous appearance. In the present case of a primary bone tumour, the microscopic picture was that of a polymorphous liposarcoma corresponding to the mixed category described by Stout (1944). Malignancy was indicated by infiltrative growth and the appearance of metastases.

Clinically, the tumour exhibited a very malignant course. Despite curettage and combined irradiation treatment at a total dose of 4,500 rad, the tumour showed rapid invasive growth with destruction of bone, spread to the iliac fossa and penetration through the operation wound. Approximately 5 months after admission the patient succumbed due to widespread metastases in the lungs, liver and left kidney.

The clinical course in our case corresponds well to that of the patient described by Dawson (1955). This patient was a 28-year-old female with a liposarcoma of mixed cell type in the femur who, despite amputation by disarticulation at the hip joint, died 9 months later due to metastases in the left lung and spine. The first of the two cases reported by Mastragostino (1965) appears also to be a primary lipo-

sarcoma of the bone. This patient was a 30-year-old man with a lesion of the proximal end of the fibula which was resected. Histopathological examination showed a liposarcoma of mixed cell type. The patient was still alive after 4 years, with no evidence of recurrence. As in our case there was severe local pain at the onset of tumour growth. Another case was convincingly reported by Catto & Stevens (1963). This patient was a 16-year-old girl with a tumour of the upper end of the tibia with the microscopical appearance of a liposarcoma of mixed cell type. Despite mid-thigh amputation this patient died 9 months after admission, with pulmonary and liver metastases. Honore et al. (1963) reported on a 36-year-old female with a liposarcoma of mixed cell type in the distal femur causing severe pain. The patient was alive with no signs of any metastases at the time of the report; i.e. 17 months after the onset of the first symptoms and 10 months after surgical treatment (exarticulation at the hip joint) had been instituted. Goldman (1964) reported a case of a liposarcoma of mixed cell type with primary localization to the mid-portion of the ulna in a 33-year-old man. This patient was alive without any evidence of metastases 3½ years after a supracondylar amputation of the humerus. Severe local pains over the tumour were noted in this patient also. Except for these reports of convincingly described cases there are reports of probable but not proven cases which are summarized in the reviews by Catto & Stevens (1963) and Schwartz et al. (1970). In addition, two cases of primary bone tumours composed of both lipoblastic and osteogenic neoplastic tissue were described by Schajowicz et al. (1966) and Ross & Hadfield (1968). Although there seem to be rare cases of primary bone tumours of two different types of mesenchymatic origin, our case obviously represents a genuine primary liposarcoma of the bone.

Microscopically, the cases of primary liposarcoma referred to above were of mixed cell type presenting a marked polymorphism. Both clinically and histopathologically these tumours showed all the characteristics of a sarcomatous neoplasm with a high potential of malignancy. Radical resection when feasible, or amputation in cases with more peripheral localization, is to be recommended as well as irradiation treatment. Although rare, this neoplasm appears to constitute a clinically well defined entity. Examining all cases of this tumour recorded by the Swedish Cancer Registry between the years 1958–1968 we have been unable to find another case with a proven diagnosis other than the one presented here.

SUMMARY

A case is presented of a rare primary liposarcoma of bone localized to the major trochanter of the left femur of a 52-year-old female. Despite combined treatment with curettage and irradiation with a total dose of 4,500 rad the neoplasm showed rapid invasive growth with destruction of the bone, spread to the iliac fossa and outgrowth through the operation wound. Approximately 5 months after admission the patient succumbed due to widespread metastases in the lungs, liver and left kidney. The histopathology, clinical course and treatment of this rare neoplasm of the bone are discussed.

ACKNOWLEDGEMENT

This work was supported by grant no. 711-B74-02X from the Swedish Cancer Society.

REFERENCES

Catto, M. & Stevens, J. (1963) Liposarcoma of bone. *J. Path. Bact.* **86**, 248–253.

Dawson, E. K. (1955) Liposarcoma of bone. *J. Path. Bact.* **70**, 513.

Goldman, R. L. (1964) Primary liposarcoma of bone. Report of a case. *Amer. J. clin. Path.* **42**, 503–508.

Honore, D., Rogister, G. & Delvigne-VanLancker, M. A. (1963) Apropos d'un cas de liposarcome intraosseux. *Acta chir. belg.* **62**, 887–895.

Mastragostino, S. (1965) Tumours lipoblastici primitivi dello scheletro. *Chir. Organi. Mov.* **44**, 18–36.

National Board of Health and Welfare, The Cancer Registry, (Ed.) *Cancer incidence in Sweden 1959–1965*, p. 64, Stockholm 1971.

Ressel, P. A., Soule, E. H. & Coventry, M. B. (1966) Liposarcoma of the extremities and limb girdles. A study of two hundred twenty-two cases. *J. Bone Jt Surg.* **48**-A, 229–244.

Ross, C. F. & Hadfield, G. (1968) Primary osteo-liposarcoma of bone (malignant mesenchymoma). *J. Bone Jt Surg.* **50**-B, 639–643.

Schajowitz, F., Cuevillas, A. R. & Silberman, F. S. (1966) Primary malignant mesenchymoma of bone. *Cancer (Philad.)* **19**, 1423–1428.

Schwartz, A., Shuster, M. & Becker, S. M. (1970) Liposarcoma of bone. Report of a case and review of the literature. *J. Bone Jt Surg.* **52**-A, 171–177.

Stout, A. P. (1944) Liposarcoma—malignant tumor of lipoblasts. *Ann. Surg.* **119**, 86–107.

*Key words:* bone; neoplasm; liposarcoma

Correspondence to:

Dr. S.-E. Larsson, M.D.
Department of Orthopaedic Surgery
University of Umeå
901 85 Umeå, Sweden