MULTIPLE OSTEOSTOMIC SARCOMAS
Report of a Case

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Multiple osteogenic sarcomas occur in elderly persons with Paget's disease of bone, but this form of the disease is a rarity in a young person with no other radiological abnormality.

CASE REPORT

A Hindu boy of fourteen attended hospital in December 1962 complaining of pain and swelling of the left thigh for two months. Three months before he had slipped and hurt himself. He did not have much pain at that time, nor did he have any swelling. A month later he noticed a swelling at the lower end of the left thigh. He attended a hospital where radiographs of the chest and left lower limb showed multicentric areas of dense shadows in the left femur, tibia and patella and multiple metastases in the lungs. The serum alkaline phosphatase was 115 King-Armstrong units. He was admitted to hospital for biopsy which was reported as a parosteal type of fibrosarcoma of bone. He was advised to have the limb amputated but he refused to have this done.

When he was first seen by us he said that the swelling was getting larger. An examination showed an otherwise healthy looking young boy. He could not walk and kept his left knee bent at an angle of about 120 degrees. There was a diffuse uniform swelling of the lowest third of the left thigh with a healed scar over it. The skin over the swelling was glossy but there were no prominent veins. The area was slightly warm and tender, and an effusion was present in the knee. Both active and passive movements of the knee were painful and limited to 30 degrees. Hard lymph nodes were palpable in the left groin above and below the inguinal ligament. No abnormality was detected in any other system of the body.

The radiograph of the left femur showed dense sclerotic of the lower part of the bone (Fig. 1). There were sclerotic areas in the left patella and upper end of the left tibia—appearances similar to those in a patient described by Moseley in 1956 as a case of "sclerosing multicentric osteogenic sarcoma." Multiple sclerotic lesions could be seen in other sites; the upper end of the left femur and both ischial bones (Fig. 2), the upper end of the right femur, the head of the right humerus, the eleventh left rib and the lower end of the right tibia and fibula (Fig. 3). Calcified lymph nodes were seen in the left groin and the left para-aortic region. There were no lesions in the skull or vertebrae. A radiograph of the chest showed multiple calcified sclerotic lesions in the lungs (Fig. 4). The sites of all the sclerotic lesions are shown in Figure 5.
Laboratory investigations gave the following results: haemoglobin 13.4 grammes per cent; packed cell volume 38 per cent; total red blood cells 4 million per cubic millilitre; total white blood cells 5.200 per cubic millilitre; neutrophils 79 per cent; lymphocytes 17 per cent; eosinophils 4 per cent; serum calcium 9.6 milligrams per cent; serum phosphorus 5.8 milligrams per cent; serum protein 7.2 grammes per cent; albumin 3.5 grammes, globulin 3.7 grammes, electrophoresis was normal; serum alkaline phosphatase 250 King-Armstrong units and a week later 280 units; serum acid phosphatase 1.0 unit; sodium 150 milli-equivalents per litre; potassium 4.7 milli-equivalents per litre; thymol flocculation 0; thymol turbidity 2; zinc sulphate turbidity 6; and carbon dioxide combining power 25.8 milli-equivalents per litre. 

**Histology**—Biopsies were taken from the swelling of the lower end of the left thigh, from the lower end of the left tibia and from the lymph nodes in the left groin. The pieces of bone were so sclerotic that they had to be chiselled out. Microscopic examination of the sections from the
tibia showed some bone spicules which appeared normal. The marrow spaces were almost filled with a spongy network of new bone trabeculae with many large pleomorphic osteoblasts covering them. No epithelial cells were seen. Sections from the femur showed new bone similar to that in the piece from the tibia but without old bone mixed with it. The new bone was accompanied by osteoblasts which were not very pleomorphic (Figs. 6 and 7). On the surface of the mass the tumour was not protruding but consisted of a dense layer of very
hyperchromatic somewhat pleomorphic spindle cells with some mitoses. In places the tissue
formed primitive cartilage. Sections from the lymph nodes showed a rim of lymphoid tissue;
the centre was composed of tumour, like that in the femur, with marked bone formation (Fig. 8).

![Photomicrograph from the lymph node from the left groin, showing cartilaginous bone with some lymphoid tissue at the periphery. (×100.)](image)

**Treatment and progress**—As the disease was generalised amputation was not advised.
Radiotherapy to the left femur was started because of pain. A dose of 2,750 r was given in
ten days using supervoltage irradiation through a single field and the patient felt better.
Radiographs of the chest and affected bones were repeated. The lesion in the left femur did
not show any change but the lungs showed a few more sclerotic shadows. He was then discharged home.

About a month later he came to us again. He was looking much better and happier, as he had no pain in the left thigh, though he could not walk or straighten his knee. Radiographs showed that the lesions at all the sites except the left femur had increased in size. The lesion in the left femur itself had become smaller and more sclerotic. The serum alkaline phosphatase level was 30 King-Armstrong units. As the disease was generalised and the local symptoms were so much better it was decided to treat him with chemotherapy, and he was given intravenous cyclophosphamide (Endoxan) in doses of 200 milligrams daily. Later the dose was increased to 400 milligrams daily. A total dose of 5-6 grammes was given, which caused the white blood cell count to fall to 4,100 per cubic millilitre. He lost a lot of hair from his head but there were no other side reactions. He recovered from the leucopenia during the next two weeks and was then discharged; the swelling of the left thigh was smaller and the alkaline phosphatase level was 28 King-Armstrong units.

When he returned for follow-up again three months later, in July 1963, he could walk with the help of a stick (Fig. 9). Haemoglobin was 12·6 grammes per cent, the white blood cell count 12,000 per cubic millilitre and the serum alkaline phosphatase level had come down to 13 King-Armstrong units.

![The patient four months after treatment with radiotherapy and chemotherapy.](image)
DISCUSSION

Malignant lesions of multicentric origin are often seen in the skin and mucous membranes of the mouth, pharynx, oesophagus, vulva and vagina (Smithers 1960), and sometimes in other organs (Molnar 1959, Willis 1960). Osteogenic sarcoma of multicentric origin often occurs in Paget's disease of the bone (Porretta, Dahlin and Janes 1957), and when bones have been irradiated as in watch-dial painters (Martland 1931). In their study of the literature Porretta and his colleagues found that in 128 cases of osteogenic sarcoma in Paget's disease twenty-seven had occurred at multiple sites. There are, however, few cases of multicentric osteogenic sarcomas occurring in the absence of Paget's disease.

Whether multiple bone lesions are multifocal in origin or result from a single primary tumour is not clear. It seems logical that if the lesions appear simultaneously they are likely to be examples of multicentric osteogenic sarcomas. On the other hand if they appear at intervals and at sites where they were not demonstrable at the time of the first examination, they may be examples of metastases from a primary tumour, though Ackerman (1948) states that late manifestations in themselves do not prove their metastatic origin.

The literature shows instances of multiple osteogenic sarcomas in the absence of Paget's disease: Silverman (1936); Ray and Galstaun (1938); Busso and Schajowicz (1945); Ackerman (1948); Halpert, Russo and Hackney (1949); Finlayson (1953); Mignani (1954); Rowen (1955); Moseley and Bass (1956); Price and Truscott (1957); Lichtenstein (1959); Smithers and Gowling (1961); Wolman (1961); Morse, Reed and Bernstein (1962); Singleton, Rosenberg, Dodd and Dolan (1962). Smithers (1960) commented that in many cases of multiple osteogenic sarcomas multifocal origin is doubtful, for metastases from one bone to the other could account for the condition. Radiological involvement of lymph nodes as in our case is rare.

A raised level of the serum alkaline phosphatase indicative of bone activity is seen in many conditions such as growing children, healing fractures, Paget's disease, rickets and osteogenic sarcoma. Porretta et al. (1957) reported a level of the serum alkaline phosphatase in a case of Paget's disease of 412 King-Armstrong units; and also a level of 71-1 King-Armstrong units in a case of Paget's disease with a sarcoma. Very high values were noted in the case of Price and Truscott (1957) where the highest level was 1,500 King-Armstrong units. In our case the serum alkaline phosphatase value was 285 King-Armstrong units at one time. The interesting thing is that the level went down to 30 after a palliative dose of irradiation to the left femur. This would suggest that the main activity was in this site. It is notable that this level went down still further from 30 to 13 King-Armstrong units after a course of Endoxan.

REFERENCES

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