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SPONTANEOUS REGRESSION OF A MALIGNANT PRIMARY BONE TUMOUR

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A histologically confirmed malignant, primary bone tumour in the pelvis, presumably an osteosarcoma, underwent spontaneous regression. The large tumour was inoperable and gave rise to severe pain as well as difficulty in walking. After 2 years of progression, with increasing destruction of the pelvic bones, the clinical and radiological condition improved spontaneously, and at present the patient is alive, almost symptom-free, after 6 years follow-up.

Key words: osteosarcoma; chondroblastoma, bone tumour, spontaneous regression

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The literature contains very few reports of apparently spontaneous regression of histologically confirmed malignant tumours. Considering that malignant primary bone tumours make up only 0.5 per cent of all malignant tumours, cases of spontaneous remission in this group of tumours are of course extremely rare. To our knowledge, only eight are on record (Everson & Cole 1966).

CASE REPORT

A 36-year-old man was first seen in February 1971. Since 1968 he had been suffering from increasing pain on weight-bearing in the region of the left hip, occasionally radiating to the leg. His general condition was good, and except for mild atrophy of the muscles of the left thigh and a limp, physical examination showed no abnormalities. Radiography disclosed a widespread osteolytic lesion in the left half of the pelvis (Figure 1). Chest radiography was normal. ESR was 100 mm and alkaline phosphatase was slightly elevated. Open biopsy was done at the anterior demarcation of the tumour in the iliac bone. It showed – as did subsequent revisions of the same preparations – benign chondroblastoma (Figure 2a). In view of the clinical and radiological findings, however, this diagnosis did not seem convincing. Therefore, an exploratory operation through a wide posterior approach was performed in March 1971. In the iliac bone there was a tumour about $12 \times 8 \times 8$ cm, anteriorly involving the superior ramus of the ischium and posteriorly the sacro-iliac joint and the lateral mass of the sacral bone. Ample tumour tissue was excised for histological examination which showed – as did subsequent histological revisions of these preparations – a malignant, primary bone tumour, presumably a highly differentiated chondroblastic osteosarcoma, although the possibility of malignant transformation of a chondroblastoma could not be ruled out (Figure 2b). It was now evident that the tumour was inoperable, and as there did not seem to be any indication for radiotherapy or chemotherapy, the patient received only symptomatic treatment, viz., non-weight-bearing and analgesic medication.

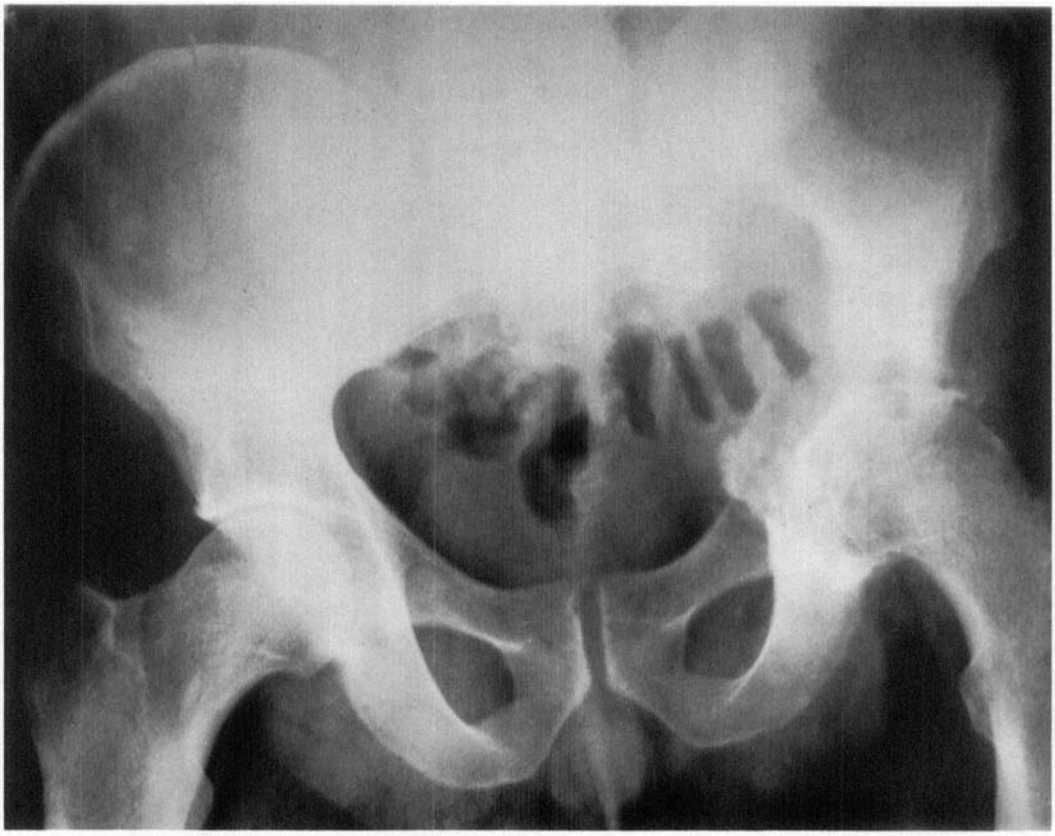


Figure 1. Primary radiography, in January 1971, showed an osteolytic tumour involving the superior ramus of the ischium as well as the ilium, from the sacroiliac joint proximally to the acetabulum distally.

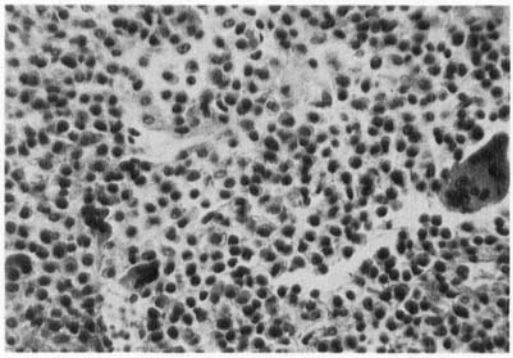
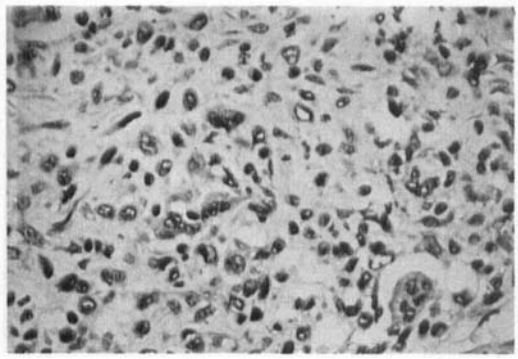


Figure 2a. First biopsy: Cellular tumour tissue with scattered giant cells suggestive of osteoclasts. The tumour cells are small and uniform, with distinct cell borders. Interpreted as a benign chondroblastoma. ×*350.*



b. Second biopsy: A more polymorphous appearance with more ample intercellular substance and fewer giant cells. The tumour cells show variations in the shape, size, and chromatin content of the nuclei. Cell borders indistinct. In places the intercellular substance shows the nature of osteoid tissue. Interpreted as osteosarcoma *or a* malignant chondroblastoma. ×*350.*

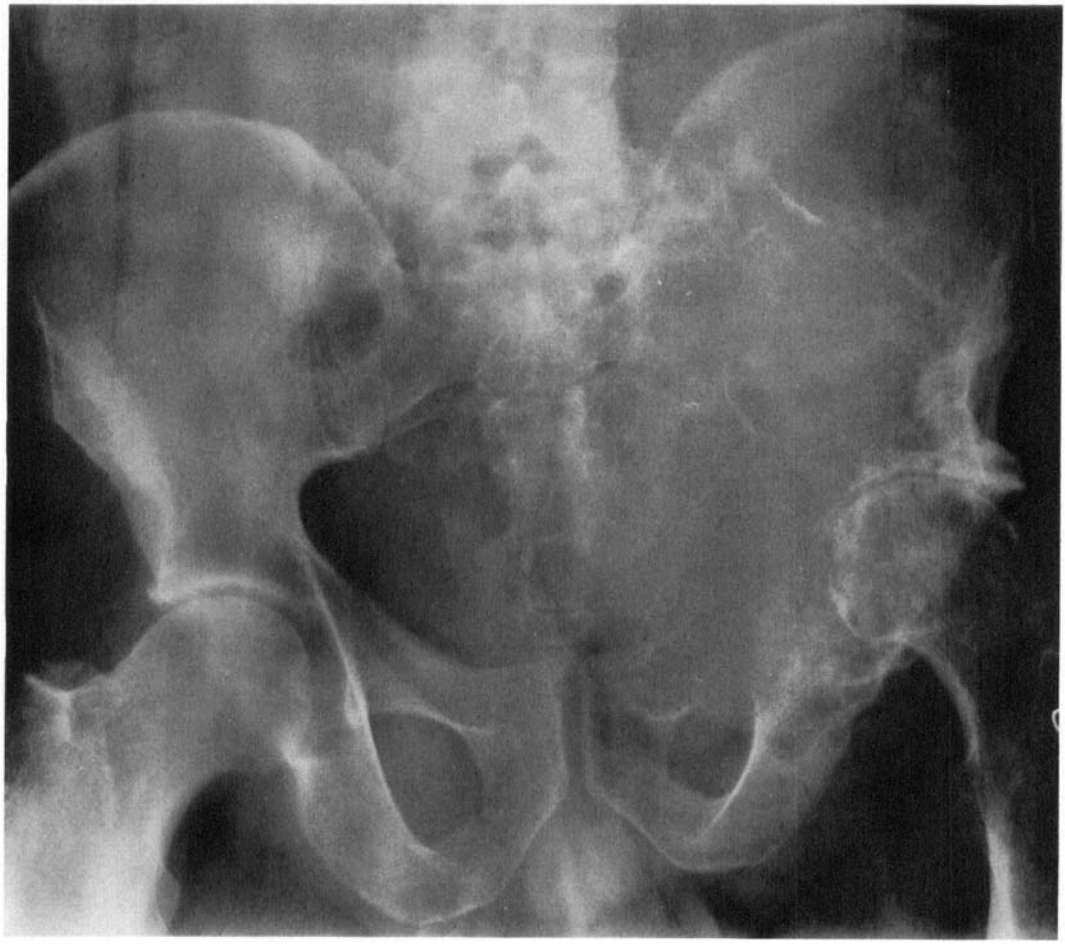


Figure 3. Radiography in May 1972 revealed progression of the tumour with extensive destruction involving the sacrum and the majority of the acetabulum.

Over the next few years the patient, still untreated, was followed up in the outpatient clinic. Until the spring of 1973 his condition deteriorated steadily. The regional pain grew worse, and now he also had rest pain. The left leg became 2 cm shorter and the difficulty in walking increased, necessitating the use of two English canes. Radiographic examinations during this period showed increasing destruction of the left half of the pelvis (Figure 3). But in the course of the latter half of 1973 a reversal occurred. The pain decreased, and during the following years the complaints gradually disappeared. Since 1975 the patient has been free of pain, and he walks almost normally. Accompanying the clinical improvement a radiological remission took place. The tumour area became increasingly delimited and sclerosed (Figure 4). When last seen, in March 1977, the patient was feeling perfectly well. There have not at any time been signs of dissemination of the tumour, in particular not to the lungs.

DISCUSSION

Everson & Cole, in 1966, collected the cases of apparently spontaneous remission of histologically confirmed malignant tumours from the available literature. Among these tumours were eight primary bone tumours, including four osteosarcomas (one of these four cases has also been described by Levin in 1957). Everson & Cole themselves use the term "cases of possible spontaneous regression",

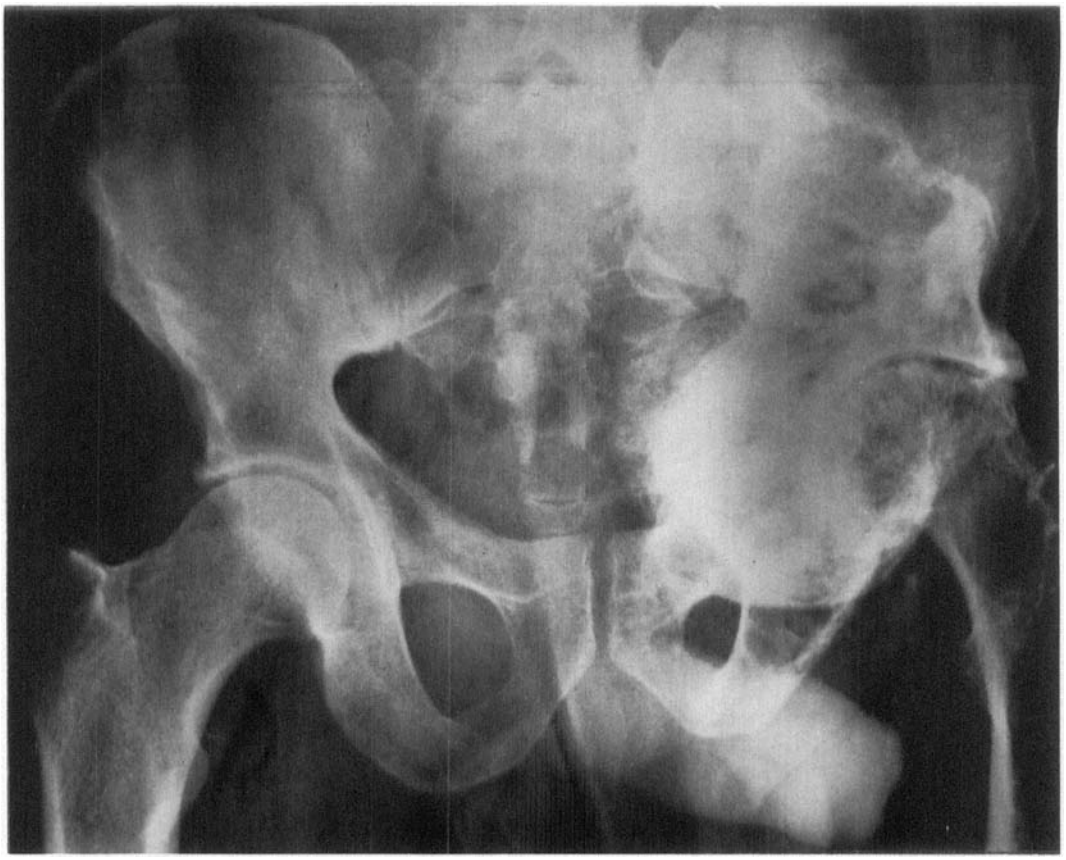


Figure 4. In March 1977 the radiological remission is unmistakable. The tumour area is well-defined with increased sclerosis.

implying that reports of an apparently spontaneous remission of a histologically confirmed malignant tumour should be regarded with scepticism and diagnostic criteria should be very strict.

In the present case the most recent clinical and radiological follow-up examination was performed 9 years after the onset of symptoms, 6 years after the biopsies, and 4 years after the time at which bony destruction was radiologically at a maximum. The second biopsy showed a primary bone tumour having definite malignant features (Figure 2b). Exact classification was less certain. It might be either a rare case of malignant chondroblastoma (McLaughlin et al. 1975, Riddell et al. 1973, Sirsat & Doctor 1970) or – more probably – a highly differentiated chondroblastic osteosarcoma. Thus, apparently this is a case of clinical and radiological, spontaneous remission of a bone tumour which had to be classified as undoubtedly malignant according to both radiological and histological findings.

Stewart (1952) has discussed the various possible causes of spontaneous remission, e.g., hypersensitivity reactions, haemorrhage in the tumour, and a reaction to the tumour protein. Immunological factors have been mentioned as a cause of spontaneous regression of malignant tumours. However, O'Hara et al. (1968) did not find any known specific, immunological factors which could explain the 10-year survival, in particular with osteosarcoma. They concluded that other host-tumour relations must be operative in the defence mechanism. Woodruff (1969) has reported the appearance

of sarcomas in relation to immunosuppressive therapy. In the patient reported above we cannot suggest any reason whatsoever for why the remission took place.

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