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RECURRENT CHONDROMYXOID FIBROMA

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During the last 30 years the number of known variants of cartilage-forming tumours has considerably increased. Chondromyxoid fibroma was described by Jaffe & Lichtenstein (1948). It is a rare tumour; in large series of bone tumours it appears to constitute about 1 per cent (Dahlin 1956). Chondromyxoid fibroma is most common in the 2nd and 3rd decade of life, but is very rare in childhood and after the 5th decade.

The tumour has been described in various bones (such as os pubis, scapula, ribs, vertebrae), but it is common in long bones and especially in the tibia. In the long bones the tumour occurs in the region of the metaphysis, but it may also involve the epiphysis. The tumours are often fairly well defined in the roentgenogram. They cause a swelling and rarefaction of the bone and break through the corticalis. Chondromyxoid fibroma grows most often peripherally in the bone and may tend to involve adjacent soft tissues (Dahlin 1956, Iwata & Cooley 1958, Scaglietti & Stringa 1961, Verma et al. 1967, present case). Chondromyxoid fibroma is regarded as nonmalignant, but in at least 10 per cent of the published cases recurrences have occurred and transitions to chondrosarcoma have been described (Benedetti et al. 1962, Iwata & Cooley 1958, Scaglietti & Stringa 1961).

REPORT OF CASE

A 42-year old man was admitted to the central hospital in Eskilstuna in 1949 because of a tender, 10 cm by 7 cm bulging hard lump in the medial malleolus of the left tibia. The patient reported that he had had the tumour for about 5 years. Roentgenography showed destruction of the entire area of the medial malleolus and the corresponding part of the tibial metaphysis besides a marked thickening of the soft tissues with irregular, cyst-like formations with thin scales of calcium

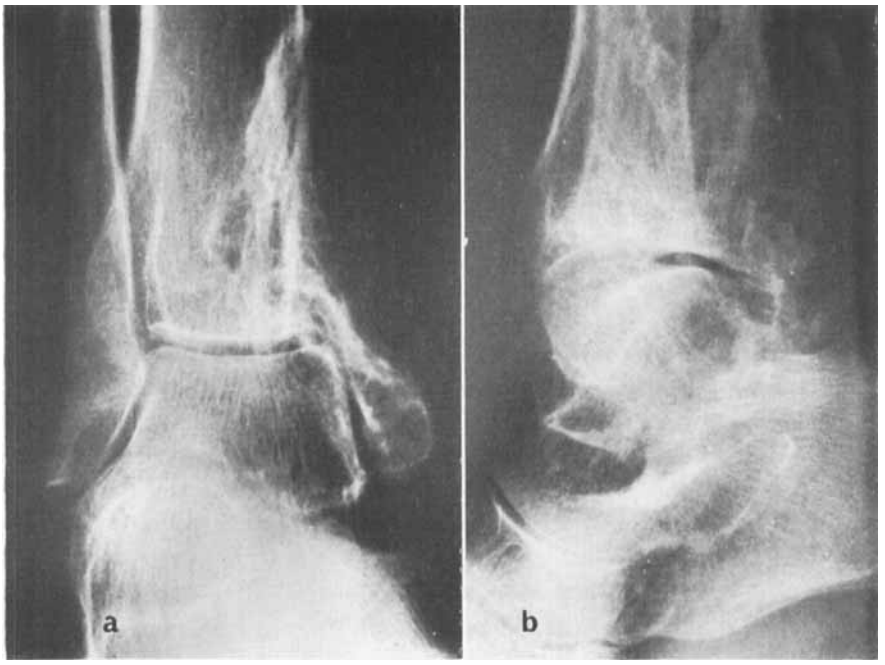

Figure 1 a. Fontal view. b. Lateral view. Roentgenograms taken Sept. 1968. Defects after operations in distal tibia.

deposits. Operation revealed a tumour involving the bone as well as soft tissues. It destroyed the tibial epiphysis and grew into the talocrural joint. The exstirpated tumour was referred for microscopic examination. About one month after the operation the wound was in good healing.

Nineteen years later, in January 1966, the patient was admitted to the Central Hospital, Ängelholm. He reported that for 10 years he had had a slowly growing lump in the upper part of the operation wound of the left lower leg. Examination revealed a tangerine-sized lump, which was adherent to the skin, but not to the bone. The lump was removed and sent for pathological examination. Roentgen examination showed bone defects in the distal tibia as a result of the first operation (Figure 1 a, b). (Roentgen films from 1949 were no longer available.) At follow-up in September 1968 and in November 1969 no further changes were seen.

Histological examination at the first operation was done at Uppsala (Prof. R. Fåhreus: PAD 1616/49) (Figure 2). The lesion was diag-

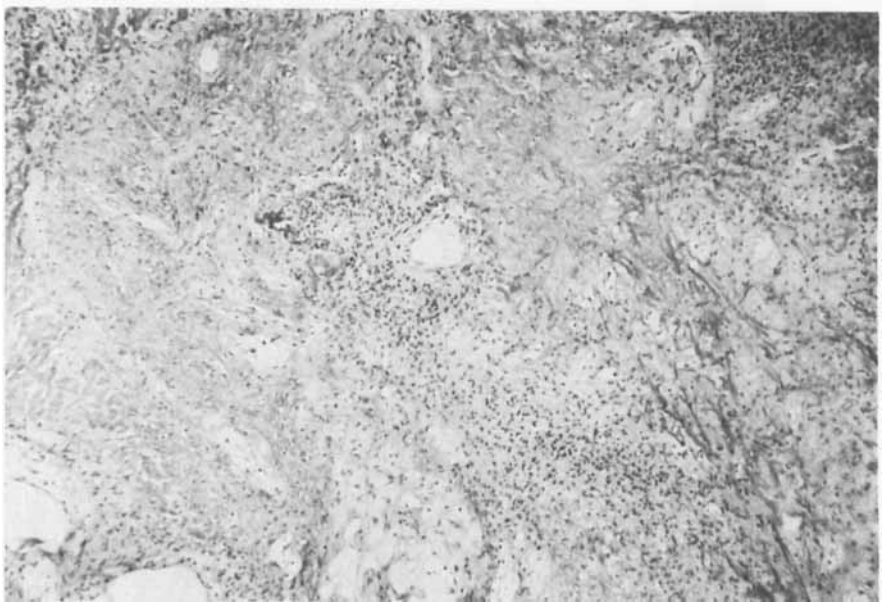

Figure 2. Material from first operation in 1949. Characteristic tumour pattern. Htx eos c:a × 40.

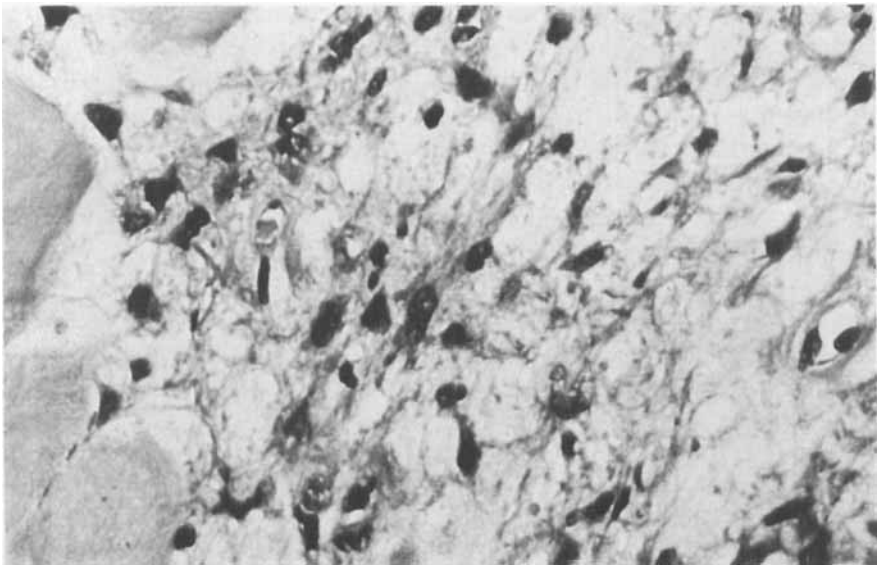

Figure 3. Material from second operation (1968) Loosely arranged pseudomalignant cells with polymorphous nuclei. Htx eos c:a × 400.

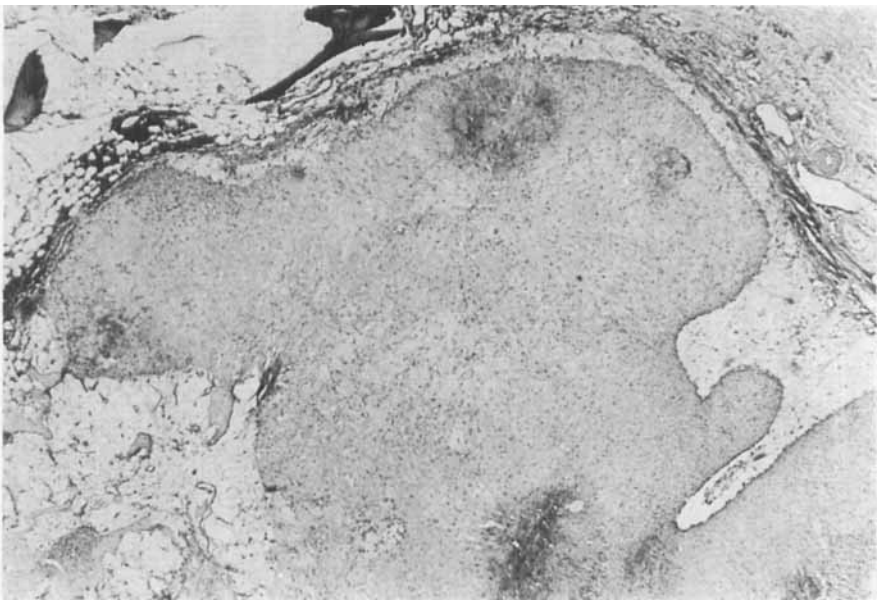

Figure 4. Second operation. Lobular extension into connective tissue. Small fragments of atrophic bone peripherally. Htx eos c:a × *24.*

nosed as myxochondroma without definite signs of malignancy. The recurrence 19 years later (Malmö PAD 1410/68) showed principally the same histology, but the tissue appeared to be poorer in cells and to have more abundant intercellular substance. The tumour showed a distinct lobular structure with crowded somewhat polymorphous pseudomalignant cells (Figure 3) peripherally in the lobuli and amorphous, cartilaginous masses centrally. The lobular pattern of the tumour was conspicuous, especially in the periphery where it gave off several extensions into the connective tissue (Figure 4). No mature cartilage was seen. The primary tumour as well as the recurrence contained extremely few giant cells. Parts of the margin of the tumour contained some regular bone trabeculae, probably residual fragments of corticalis.

DISCUSSION

The case shows some features which may contribute to our knowledge of chondromyxoid fibroma. The growth of large recurrent tumour in the soft tissues with at most minimal contact with bone has not

been reported before and is exceptional in a tumour primarily developing from bone. It may suggest that the recurrence had developed from inplantation of tumour remains in the operation area. Rest of bone in the outer layers of the tumour may, however, suggest secondary separation of a large recurrence growing superficially in bone. According to Dahlin (1956), recurrences are due to residual lobular extensions of the tumour, and this explanation may also hold for single published cases of multifocal recurrences. When recurrences occur, they usually do so within 1–3 years of the first operation. In one case a recurrence did not appear until 9 years after the operation (Kunkel 1955). It is difficult to date the recurrence in our case, but the interval after the first operation seems to have been longer than in any other case on record.

Chondromyxoid fibroma is classified as a benign tumour. Increasing experience, however, suggests that the tumour is characterised by a fairly pronounced tendency to recur and to spread both in bone and in surrounding tissues. In children below 10 years the tumour is very aggressive, and it grows quickly and recurs very often (Scaglietti & Stringa 1961). As pointed out by Lichtenstein (1965) those 5 cases described by Scaglietti & Stringa (1961) as myxoma of bone in children should be classified as chondromyxoid fibroma. We suggest that chondromyxoid fibroma be regarded as a tumour with a potential local malignancy.

The relation between chondromyxoid fibroma and chondroblastoma has been discussed by Dahlin (1956). From a therapeutic and prognostic point of view it is most important to differentiate it from chondrosarcoma. Both types of tumour occur in the same age groups, though chondromyxoid fibroma appears in somewhat younger patients. Owing to the histological appearance of pseudomalignant cells with irregular nuclei, chondromyxoid fibroma can be easily confused with chondrosarcoma. Most chondrosarcomas, however, contain fairly well differentiated cartilaginous structures with distinct lacunae, containing atypical cells, often with giant bizarre nuclei. In the chondromyxoid fibroma the intercellular substance rarely shows lacunae with true cartilage cells. The most important pattern of chondromyxoid fibroma, however, is lobular islands of ground substance with a condensation of polymorphous cells in the periphery of the islands and with streaks of loose metachromatic substance.

SUMMARY

A 42-year old man sought advice because of a tender, 10 cm long and 7 cm wide, hard lump on the medial malleolus of the left tibia. The lump had grown during the last 5 years. Operation revealed a tumour of the bone with destruction of the tibial-epiphysis and involvement of the talo-crural joint. Nineteen years later the man was reoperated upon in the same area because of a recurrent lump which had been gradually growing for about 10 years in the upper part of the operation wound. The tumour grew in the soft tissues apparently without connection with bone. The histological picture was largely the same in both preparations and was compatible with a diagnosis of chondromyxoid fibroma.

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