BENIGN CHONDROBLASTOMA OF BONE

Report of a Case of Malignant Transformation

M. V. SIRSAT and VATSALA M. DOCTOR, BOMBAY, INDIA

From the Department of Pathology, Tata Memorial Hospital, Bombay

Benign chondroblastoma is a rare tumour comprising only 1·3 per cent of the primary tumours of bone seen at the Tata Memorial Hospital, Bombay. Dahlin (1957) reported an incidence of less than 1 per cent of benign chondroblastoma in his series of bone tumours. The presence of multinucleated giant cells led earlier workers to classify the tumour as a variant of giant-cell tumour (Codman 1931, Ewing 1940), but Jaffe and Lichtenstein (1942) pointed out differences in the clinical and histological features. They interpreted the basic polyhedral tumour cell as the cartilage germ cell, and called the tumour benign chondroblastoma. Although the histogenesis of the tumour is still unsettled the term benign chondroblastoma has received wide acceptance.

The benign nature of the tumour has been emphasised by most workers. In fact, the occurrence of malignant counterparts or malignant transformation of this tumour has been denied; thus Lichtenstein (1965) stated that no instance of aggressiveness or spontaneous malignant change had been recorded.

Recently we encountered a case of so-called benign chondroblastoma which recurred repeatedly and emerged finally as a highly malignant undifferentiated sarcoma. We believe this is the first reported instance of spontaneous malignant transformation of this tumour.

CASE REPORT

A Hindu boy aged fifteen years complained in 1959 of a painful swelling of the right knee of two months' duration, noticed after a fall. The knee was immobilised for six weeks and thereafter he received massage, but the swelling and pain gradually increased.

On examination, there was a warm tender swelling in the region of the right knee, with limitation of movement. There was marked wasting of the quadriceps muscle. The inguinal

FIG. 1

FIG. 2

FIG. 3

FIG. 4

Figure 1—Early radiograph (November 1960) showing an osteolytic lesion of the upper end of the tibia involving the epiphysis and the adjoining metaphysal region. Figure 2—(May 1961) Extension of the osteolytic area with irregular areas of calcification. Figure 3—Radiograph of the recurrent tumour in March 1965. Figure 4—Radiograph taken shortly before amputation. There is a large osteolytic area involving the upper end of the tibia, extending to the lower end of the femur.

nodes on the right side were enlarged. Radiographs in November 1960 showed destructive changes in the upper epiphysial region of the right tibia extending into the metaphysis. The articular surface of the tibia was thinned but intact (Fig. 1).

FIG. 5

FIG. 6

FIG. 7

FIG. 8

Figure 5—Photomicrograph of the tumour curetted in November 1962 showing the typical appearance of benign chondroblastoma. Sheets of polygonal cells are seen with scattered multinucleated giant cells and small foci of calcification. (Haematoxylin and eosin, $\times 140$.) Figure 6—Higher magnification showing uniform appearance of chondroblasts. (Haematoxylin and eosin, $\times 280$.) Figure 7—Photomicrograph showing early chondroid areas developing within the tumour adjoining the lace-like streaks of calcification surrounding the tumour cells. (Haematoxylin and eosin, $\times 280$.) Figure 8—Another area of the tumour showing deposits of calcium in a group of partly necrotic tumour cells. (Haematoxylin and eosin, $\times 280$.)

Biopsy of the synovial membrane showed only sparse lymphocytic infiltration. Culture did not yield any growth. Except for a slightly raised erythrocyte sedimentation rate, all investigations were normal.

A diagnosis of tuberculosis of the right knee joint was made. Antituberculous treatment was begun and the knee was immobilised. There was no improvement and the swelling increased, with further limitation of movement. Radiographs in May 1961 showed an extension of the osteolytic area in the upper epiphysis and adjoining metaphysis of the right tibia, with irregular areas of calcification (Fig. 2).

Operation—In November 1962 the lesion was exposed and thoroughly scraped out. The tumour was seen extending into the knee joint at two places. A femoral head excised from a patient with a fracture was grafted to fill the cavity. A plaster cylinder was applied for three months. The patient made a good recovery with improvement in knee movement.

FIG. 9

FIG. 10

Figure 9—Photomicrograph showing the appearance of the tumour after its recurrence in January 1968. The cells show enlargement of nuclei with a moderate degree of atypism. (Haematoxylin and eosin, $\times 140$.)
Figure 10—Part of the amputated limb showing the macroscopic appearance of the tumour involving the upper end of the tibia.

Histology—The material removed at operation consisted of brownish pieces with a gritty feel. The tumour showed the typical appearance of benign chondroblastoma, sheets of characteristic ovoid and polyhedral cells being intermixed with a variable number of multinucleated giant cells (Fig. 5). The tumour cells showed well defined cytoplasmic outlines. The nuclei were ovoid, vesicular and uniform in size and shape (Fig. 6). At places the tumour cells were separated by islands of fibrous tissue merging into areas of chondroid and osteoid tissue (Fig. 7). Areas of calcification were seen as fine lace-like deposits surrounding individual cells (Figs. 7 and 8).

Progress—The patient remained well for two years, during which time serial radiographs showed fairly good consolidation of the graft, leaving areas of decalcification. In January 1965 there was recurrence of pain and deformity, and radiographs now showed a definite recurrence of the tumour with erosion of the articular cartilage (Fig. 3).

Again a thorough curettage was carried out and arthrodesis was performed. The tumour soon recurred as was evident from the subsequent radiographs taken in April 1966 and November 1967. The tumour was curetted again in January 1968. On histological examination the tumour cells showed larger, hyperchromatic nuclei with variation in size and shape (Fig. 9). However, malignant transformation was not suspected at that stage.

In May 1968 the patient presented with a discharging sinus on the antero-medial aspect of the right leg. He was found to be markedly emaciated. A diagnosis of infected bone graft was made. The septic cavity was laid open and drained and a small piece of the tumour was removed for histological examination. The appearance was now that of a frankly malignant tumour. Radiographs at this time showed a large osteolytic area involving the upper end of the tibia and extending into the lower end of the femur (Fig. 4). Above-knee amputation was performed in June 1968. Within two weeks of his discharge from the hospital the patient developed a high temperature and enlarged inguinal lymph nodes. The liver was markedly enlarged, presumably from metastases. The patient gradually went into hepatic failure and died in July 1968, nine years after the onset of the disease. There was no necropsy.

FIG. 11

FIG. 12

Figure 11—Section from the tumour of the amputated specimen showing sheets of anaplastic cells with large, hyperchromatic bizarre nuclei. Compare with Figure 5. (Haematoxylin and eosin, $\times 140$.) Figure 12—High power photomicrograph of the tumour shown in Figure 11. The tumour cells are polygonal or fusiform in shape. Nuclei are large and hyperchromatic. A cell in mitosis is seen. (Haematoxylin and eosin, $\times 280$.)

The amputation was carried out at another hospital and only part of the tibia bearing the tumour was received in the laboratory (Fig. 10). The tumour measured 11.5 centimetres in the long axis. It had a greyish-pink opaque appearance. It was infiltrating into the medullary cavity and the overlying cortex. The surrounding soft tissues showed extensive infiltration.

Multiple sections from the specimen showed a highly cellular tumour composed of sheets of polygonal and fusiform cells (Fig. 11). The nuclei were large and hyperchromatic, showing coarse, scattered chromatin (Fig. 12). Mitotic figures were seen in fair numbers. Multiple sections revealed a uniform picture with only an occasional multinucleated giant cell. Foci of calcification or areas of chondroid differentiation were absent. Widespread areas of necrosis were seen.

DISCUSSION

This case affords a clear example of spontaneous malignant transformation of a previously benign chondroblastoma.

Perusal of the literature reveals very few instances of malignancy supervening on a benign chondroblastoma. Hatcher (1945) reported a case of benign chondroblastoma of the upper

end of the tibia which was heavily irradiated. After four years a chondrosarcoma developed in the head of the adjoining fibula. Hatcher and Campbell (1951) subsequently reported another case of benign chondroblastoma of the upper end of humerus which was treated with curettage and irradiation. A chondrosarcoma developed at the site of the lesion three and a half years after treatment. Steiner (1965) reported a similar case of benign chondroblastoma of the upper end of femur treated with radiation which developed an osteogenic sarcoma five years later. These cases appear to be post-radiation sarcomas resulting from the direct effect of the radiation on the bone and unrelated to the pre-existing chondroblastoma.

Kahn, Wood and Ackerman (1969) reported two cases of chondroblastoma with unusually aggressive behaviour. One patient died with widespread metastases proved at necropsy. The other showed extensive infiltration of the soft tissues with tumour emboli in the lymphatics. The patient is alive and well eighteen months after amputation. In both cases the tumours differed in no way from the usual benign chondroblastoma in their histological appearance except that mild degree of cellular atypism was observed in the metastatic lesions. Ackerman (1969) mentioned having seen two more cases of chondroblastoma that metastasised. Coleman (1966) reported a case of benign chondroblastoma of the tibia which recurred repeatedly after curettage and infiltrated into the soft tissues and the knee joint.

The present case differs from all the previously reported cases in that it showed a gradual transformation from a typical benign chondroblastoma to a highly undifferentiated sarcoma associated with a corresponding change in the biological behaviour of the tumour. At no time had the patient received radiation.

From the cases so far reported in the literature it seems hardly necessary to stress the fact that the so-called benign chondroblastoma is not so benign as has been hitherto believed. Kahn *et al.* (1969) suggested that the correct designation for the lesion should be simply chondroblastoma. Radical measures to eradicate the disease are justified when signs of aggressiveness appear.

SUMMARY

A case of malignant transformation of a benign chondroblastoma of the tibia in a fifteen-year-old boy is reported. The tumour was not irradiated at any time during the course of the disease. The tumour showed a typical appearance of benign chondroblastoma at the first operation. It recurred repeatedly after curettage, and finally emerged as a highly malignant undifferentiated sarcoma. In spite of amputation the patient died nine years after the onset of the disease with an enlarged liver and inguinal lymph nodes.

Material for the case was kindly supplied by Dr R. M. Bhansali and Dr M. L. Gavadia.

REFERENCES

- ACKERMAN, L. V. (1969): *Personal communication*.
 CODMAN, E. A. (1931): Epiphyseal Chondromatous Giant Cell Tumors of the Upper End of the Humerus. *Surgery, Gynecology and Obstetrics*, **52**, 543.
 COLEMAN, S. S. (1966): Benign Chondroblastoma with Recurrent Soft-tissue and Intra-articular Lesions—Report of a Case. *Journal of Bone and Joint Surgery*, **48-A**, 1554.
 DAHLIN, D. C. (1957): *Bone Tumors*, p. 31. Springfield, Illinois: Charles C. Thomas, Publisher.
 EWING, J. (1940): *Neoplastic Diseases*. Fourth edition, p. 324. Philadelphia: W. B. Saunders Company.
 HATCHER, C. H. (1945): The Development of Sarcoma in Bone Subjected to Roentgen or Radium Irradiation. *Journal of Bone and Joint Surgery*, **27**, 179.
 HATCHER, C. H., and CAMPBELL, J. C. (1951): Benign Chondroblastoma of Bone: Its Histologic Variations and a Report of Late Sarcoma in the Site of One. *Bulletin of the Hospital for Joint Diseases*, **12**, 411.
 JAFFE, H. L., and LICHTENSTEIN, L. (1942): Benign Chondroblastoma of Bone—A Reinterpretation of the So-called Calcifying or Chondromatous Giant Cell Tumor. *American Journal of Pathology*, **18**, 969.
 KAHN, L. B., WOOD, F. M., and ACKERMAN, L. V. (1969): Malignant Chondroblastoma. *Archives of Pathology*, **88**, 371.
 LICHTENSTEIN, L. (1965): *Bone Tumors*. Third edition, p. 56. Saint Louis: The C. V. Mosby Company.
 STEINER, G. C. (1965): Postradiation Sarcoma of Bone. *Cancer*, **18**, 603.