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Soft Tissue Recurrence of Giant Cell Tumor of Bone: A Pitfall in Treating the Giant Cell Tumors of Bone

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Thourough currettage and bone grafting is the treatment of choice for most of the cases of giant cell tumor of bone although high recurrence rate is known (GOLDENBERG,, et al. 1970, HUTTER, et al. 1962, JHNSON, et al. 1982, Marcove, et al. 1978). This tumor has an aggressive biological characteristics (GOLDENBERG, et al. 1970, Hutter, et al. 1962, JHNSON, et al. 1982, MARCOVE, et al. 1978, MIRRA, et al. 1982) and one of the pitfalls in treating this tumor is its recurrence in the soft tissue of the operation field. When scattered to the soft tissue, giant cell tumor of bone attracts new blood vessels and start proliferation there. We recently experienced three cases of giant cell tumor of bone which recurred in the soft tissue of surgical field. We believe such cases might not be rare and it is rather surprising that we could find out only a few documented cases of soft tissue recurrence of giant cell tumor of bone in English literature (FRAGAKIS, 1981, RILEY et al. 1967, SERRA, et al. 1985). It is a serious problem particularly in the forearm or hand where many tendons and nerves run and the removal of the recurrent tumor is technically very difficult. We believe it could be preventable if we are careful enough not to contaminate the soft tissue of the operation field by tumorous tissue.

Case Reports

Case 1. A 29-year-old woman noticed a dull pain in the right popliteal region in April, 1980 and visited a hospital on July 5, 1980. Roentgenograms showed an osteolytic lesion in the distal epiphyseo-metaphyseal region of the right femur. The tumor was curretted and an iliac bone graft was performed on September 12. The histological diagnosis was giant cell tumor of bone. Follow up roentgenograms showed an recurrent tumor at the site of operation. The patient underwent currettage and bone grafting again in April, 1981. Reccurrence of the tumor was again apparent radiologically in July and she was referred to us. On November 25, when whe was admitted to our hospital, two surgical scars were seen around her right knee, a medial parapatellar incision and a lazy S-shaped scar in the popliteal region. Roentgenograms showed

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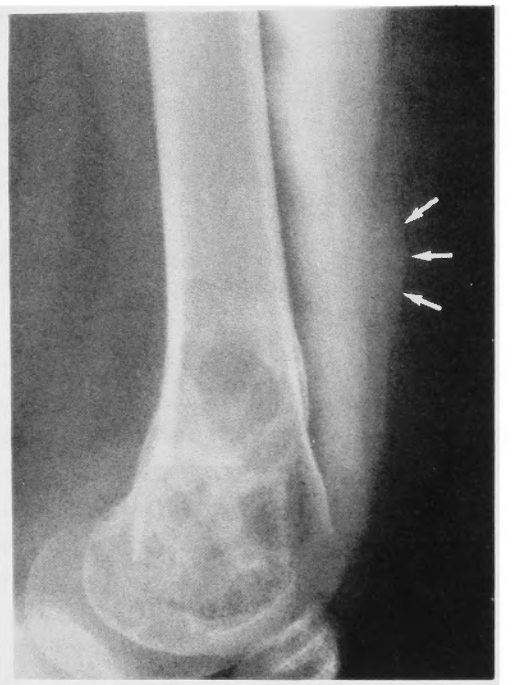

Fig. 1-1. Lateral view of the knee showing an osteolytic lesion in the distal epiphyseometaphyseal area of the femur. Arrows indicate a soft tissue tumor in front of the rectus femoris.

an osteolytic lesion in the distal epiphyseo-metaphyseal region of the right femur. The right femoral condyle was mainly involved with little subchondral bone remaining. Although the lateral femoral cortex was destroyed, the tumor was covered by a thin bony layer, and there was no evidence of soft tissue extension of the tumor. Just anterior to the rectus femoris, there was a soft tissue mass which was 26 mm × 13 mm on roentgenograms on March 3 (Fig. 1-1). The tumor was also clearly seen on a CT scannogram (Fig. 1-2). Femoral angiography performed on December 14, 1981 showed a tumor stain and cork screw like arteries around the tumor with no malignant signs, such as tumor encasement or arteriovenous malformation. A small tumor stain was observed at exactly the same site where the soft tissue mass was later recognized on the plain roentgenograms. The patient herself noticed a subcutaneous tumor beneath the scar of the first operation on her thigh in early February. On March 15 en block excision and prosthetic replacement of the distal 10 cm of the femur were carried out. A soft tissue tumor was found in the subcutaneous tissue just in front of the rectus femoris. There were moderate adhesions between the tumor and the fascia covering the rectus femoris, but there was apparently co nonnection between the soft tissue tumor and the femur or the tumor within the femur. The tumor in the subcutaneous tissue was removed together with the normal soft tissue surrounding it. It measured 3.5 cm × 2.0 cm × 1.0 cm, was elastic soft and light brown with dark brown spots on the cut surface. Histologically, it was the same as the recurrent tumor in the bone, grade II giant cell tumor, but with some grade III areas (Fig. 1-3). The post-operative course was uneventful, and there is presently no evidence of recurrence or distant metastasis in the lung.

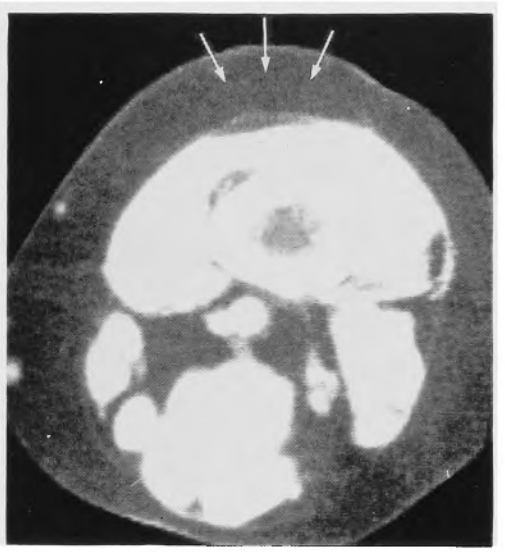

Fig. 1-2. CT scan of the thigh. Arrows show a soft tissue tumor in the subcutaneous tissue just over thre rectus femoris muscle.

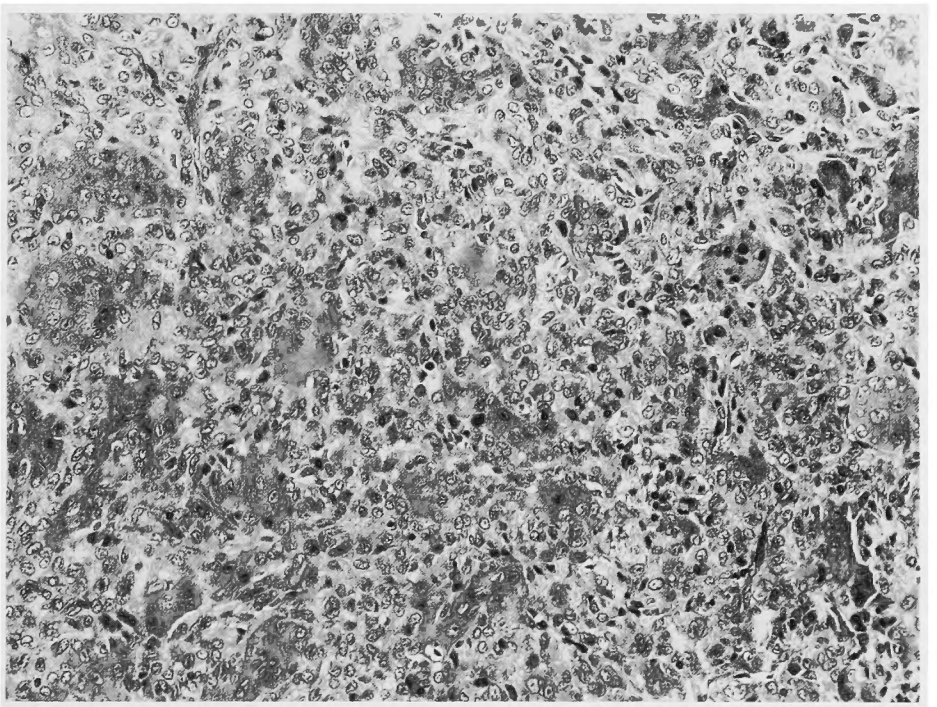

Fig. 1-3. Histologically the tumor consisted of mononuclaer stromal cells and evenly distributed giant cells. (×200)

Case 2. An 18-year-old girl noticed a pain in her right wrist in November, 1980, when it was hit with a volley ball. The pain persisted and she visited Himeji National Hospital on March 28, 1981. Roentgenograms of the wrist showed an osteolytic lesion in the distal epiphyseo-

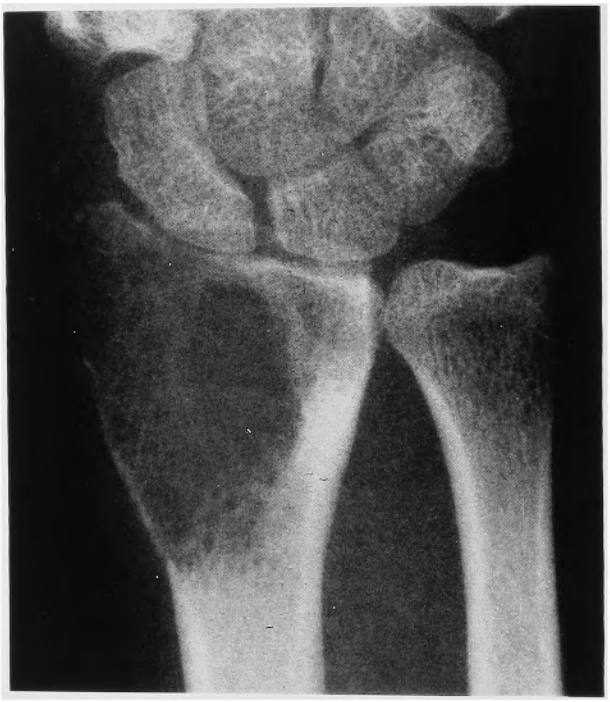

Fig. 2-1. AP view of the right wrist showing an osteolytic lesion in the distal epiphyseo-metaphysis of the radius.

metaphyseal region of the radius (Fig. 2-1). Currettage of the tumor and bone grafting were performed on April 1, 1981. The distal radius was replaced by a soft yellowish brown tumor which was curretted together with surrounding normal bone and iliac bone graft was performed. In July a soft tissue tumor about 1.0 cm in diameter became palpable in the subcutaneous tissue of the dorsal forearm about 10 cm proximal to the wrist joint, close to the surgical scar. Follow up roentgenograms on July 14 showed an recurrent tumor in the bone graft and she underwent the currenttage and bone grafting again on July 24. The tumor in the subcutaneous tissue was left unremoved. The recurrence was again apparent on follow-up roentgenograms and on December 4, 1981, the distal 7 cm of the radius together with the lunate and the scaphoid were excised en bloc and a free fibular graft was performed and the right wrist joint was fused. During the operation a soft tissue tumor, 1.0 cm × 1.0 cm × 1.5 cm, was found in the subcutaneous tissue close to the first operational scar and removed. This tumor was encapsulated, and there was no apparent connection between the tumor and the radius. Histologically both the tumor in the bone and the tumor in the subcutaneous tissue were giant cell tumor of bone (Fig. 2-2). In November, 1982, a recurrence was again apparent in the fibular graft, and there were two soft tissue lumps close to the surgical scar on the dorsum of the wrist joint. On November 16, the wrist was again opened and the recurrent tumor in the fibular graft was curretted and irrigated with saline. There was marked adhesion between the skin and the two tumors in the subcutaneous tissue. The tumors were connected with each other by a thin extension and were

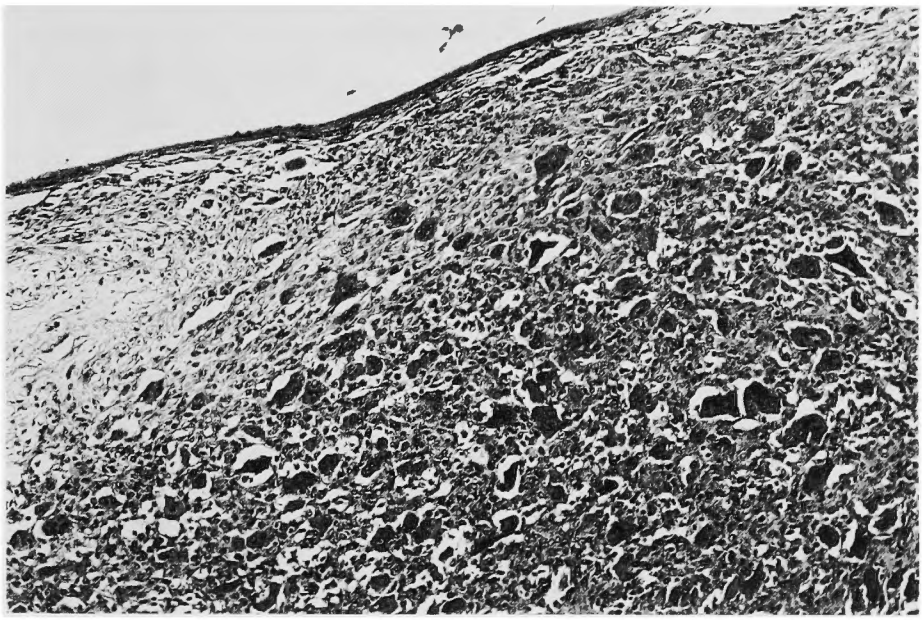

Fig. 2-2. Typical giant cell tumor of bone in the subcutaneous tissue. The tumor was completely encapsulated by thin connective tissue.

also in contact with the radius. No connection between the soft tissue tumor and the tumor in the bone could be recognized. Extensor tendons were encircled by this tumor. These tumors were marginally excised. At the time of writing there is no apparent recurrence of the tumor.

Case 3. The 33-year-old woman came to us on April 15, 1985 with a complaint of pain and palpable mass of her left knee. She noticed in February left knee pain during walking 1983 and consulted an orthopaedic doctor. Under the diagnosis of bone tumor of the proximal tibia, she underwent currettage of the tumor and the bone grafting from the iliac bone. The hisological diagnosis was giant cell tumor of bone. Curettage and bone grafting were performed for the recurrent tumor again on April 20. In February 1985 she noticed the left knee pain and palpable tumor of the knee. Tumor recurrence was apparent radiologically and the patient was referred to us on April 15, 1980. A soft tissue tumor, which was about 10 cm in diameter was recognized just over the proximal portion of the tibia. A longitudinal operative scar of ca 10 cm long was over the tumor and there were a few indurations of about one cm in diameter in the subcutaneous tissue just proximal to the operative scar. Radiologically the whole epiphyseometaphysis of the proximal tibia was destroyed by the tumor. On September 13, about 5.5 cm of the proximal tibia and fibula was resected en bloc and total knee replacement was carried out. Three nodular indurations of one cm in diameter were found in the subcutaneous fatty tissue of the last operative scar and removed. These tumors had no connections with the tumor of bone. Histologically both the intraosseous tumor and the tumor in the subcutaneous tissue were giant cell tumor of bone.

Discussion

Tumor contamination does not necessarily lead to local recurrence even in malignant tumors (SPRINGFIELD, 1982, SUGARBAKER & KETOHAM, 1977). Most of the scattered tumor cells die because of nutritional deficit after the operation, immunological reactions or other reasons. Thus soft tissue recurrence of benign bone tumors is unusual, even if some tumor cells or lumps of tumor tissue are implanted in the soft tissue of the surgical field. Tumor cells or small lumps of tumor tissue scattered in the surgical field may die or remain alive without proliferation in a dormant state through the diffusion of nutrients for a while. Only when they attract vessels from the surrounding tissue by virtue of tumor angiogenesis factor they start proliferation to form a macrometastasis. Tumor angiogenesis factor is considered to be a kind of RNA molecule with the molecular weight of 70,000 and usually secreted by malignant tumors or embryonal tissue, and not by benign tumors or normal tissue (CLARK, 1979, FIDLER, et al. 1981, FISHER, et al. 1975, GALASKO, 1981, SMITH, et al. 1958. SPRINGFIELD, 1982) It is well known that giant cell tumors of bone (MIRRA, 1982) sometimes metastasize to the lung after currettage. The metastasis does not generally endanger the patient's life and is called benign metastasis (HUTTER, 1962, MARCOVE, et al 1978, MIRRA, 1980). Soft tissue recurrence of giant cell tumor of bone as is seen in the three cases and also documented by FRAGAKIS (1981) and RILEY, et al (1967) and SERRAS, et al (1985) is fundamentally the same phenomeneon as the benign metastasis in that the scattered tumor cells obtain their blood supply supposedly by virtue of tumor angiogenesis factor and proliferate. Considering the clinical facts of the high incidence of tumor contamination of the wound in cancer patients (SMITH, et al. 1958), frequent subclinical fat embolism in long bone fractures (ARNIM & GRANT, 1951, GOSLING & PELLEGRINI, 1982, SALDEEN, 1970), the demonstration of pulmonary cement embolism after total hip replacement (BREED, 1974), or a sudden increase in the number of tumor cells circulating in the blood of patients after surgical or diagnostic procedured (FISLER, et al 1981), the tumor contamination is inevitable, either locally or systemically, during the currettage of bone tumors. The volume of the scattered tumor is an important factor. SOUTHAM, et al (1961) demonstrated the dose and take relationship in subcutaneous autotransplantations of human tumors; the larger the tumor dose or the volume of implanted tumor, the more probable its local growth and innoculation of at least one million cells are necessary for local growth of the tumor. Leaving a visilde size of the tumorous tissue in the operation field is particularly dangerous. We should try to minimize contamination of the surgical field by covering the exposed area with towels, frequently changing gloves and instruments and irrigating with plenty of saline, although complete prevention may be impossible. It might be very difficult to wash out the tumor tissue sacattered over the soft tissue even if we use the jet stream of water. So routinely covering of the soft tissue operation field by thick towels may be most important. Also we should excise giant cell tumor of bone en bloc whenever feasible.

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和文抄録

骨巨細胞腫の軟部組織での再発——骨巨細胞腫治療上の落し穴——

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術野軟部組織に再発した骨巨細胞腫を三症例を供覧する．骨巨細胞腫は掻爬，骨移植術を施行した場合の再発率40-50%と，良性腫瘍の中では治療に難渋することの多い骨腫瘍である．また掻爬術後に1-2%の骨巨細胞腫は肺へ遠隔転移することも知られている．この転移は，悪性腫瘍の場合と異なり生命的予後が良好であるため“benign metastasis”とも云われる．術中操作で血行中に入った腫瘍塊の一部が肺まで遠隔転移しそこで一般的に良性腫瘍は生産しないとされるIumor Angigenesis Factorをおそらくは自ら生産し，これを介して周囲組織より血行を得て増殖を開始した結果と思われる．掻爬の際に術野の軟部組織に撒種された小腫瘍塊の一部が増殖再発するのも“benign metastasis”と同様の機序と思われる．いずれにしても治療上問題は大きく，殊に本論文の症例2のように多くの腱，神経の走行する前腕での軟部組織での再発は根治が困難となるので，術中術野の汚染を防ぐべき十分な努力をすべきである．即ち，可能な限りen blocに切除する，術野をタオルで覆う，jet stream等で術野を十分洗浄する，手袋を頻回に交換する等の注意を払うべきである．殊に大きい腫瘍塊ほど生着しやすいから少なくとも肉眼に見える位以上の大きさの腫瘍塊を術野に残さぬことが大切であろう．