Upsala J Med Sci 106: 211–215, 2001

Extraskeletal osteosarcoma arising in the buttock

M. Oonuma, M. Hatori, M. Hosaka and S. Kokubun

Department of Orthopaedic Surgery, Tohoku University School of Medicine, 1-1 Seiryomachi, Aobaku, Sendai, Japan

ABSTRACT

Extraskeletal osteosarcoma is a rare sarcoma that accounts for about 1 % of malignant soft tissue tumours. We report an very unusual case of a small-size extraskeletal osteosarcoma arising in the superficial subcutaneous region of the buttock. The patient was a 55 year-old female. She incidentally noticed a small nodule in the right trochanteric region. It was removed under local anesthesia at a private clinic. An additional wide excision was performed at our hospital. There was an 8 mm? 10 mm mass in the subcutaneous tissue. No invasion of the underlying fascia was observed. Microscopic examination of the removed tumour showed bizarre-looking spindle and giant cells with lace-like osteoid. The tumour was diagnosed as extraskeletal osteosarcoma. Chemotherapy with Rosen T-20 was administered to the patient. The patient has survived without recurrence or metastasis for 4 years since the primary surgery. Its superficial location, very small size, wide excision, and chemotherapy were thought to contribute to her long survival.

INTRODUCTION

Extraskeletal osteosarcoma (ESOS) is a rare sarcoma that accounts for about 1 % of malignant soft tissue tumours [12]. Since first described by Wilson [20] in 1941, approximately 300 cases of ESOS have been reported [1 16, 18 20]. To satisfy the definition of ESOS, the tumour must (1) arise in the soft tissue?and?not be attached to bone or periosteum, (2) have a uniform sarcomatous?pattern, (3) produce osteoid and/or cartilage matrix [20]. We report a very unusual case of an extremely small extraskeletal osteosarcoma arising in the superficial subcutaneous region of the buttock.

Case Report

The patient was a 55 year-old female. In September 1997, because she incidentally noticed a small nodule in the right trochanteric region when she scratched this area. It was removed under local anesthesia at a private clinic. The surgical record described that, macroscopically, an encapsulated bean-sized tumour was located in the subcutaneous region of the buttock. She was referred to our clinic because of the suspicion of sarcoma. The physical examination revealed no contributory factors. The laboratory data were normal. The plain radiography, computed tomography, and bone scintigraphy demonstrated no abnormalities. An additional wide excision was

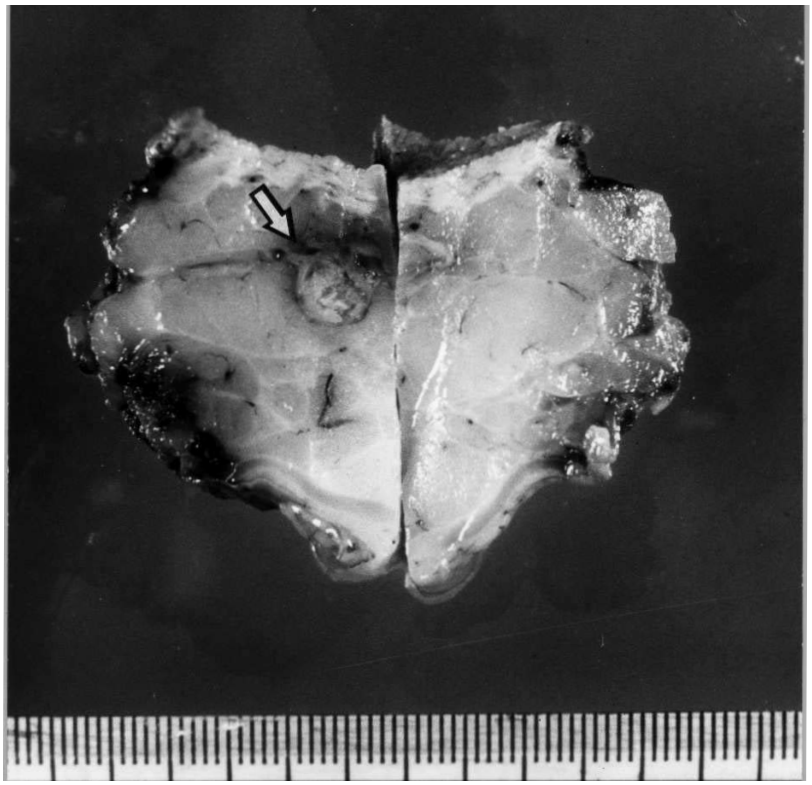

Fig. 1. Gross photograph of a section of the removed tumour (arrow) and the surrounding subcutaneous fatty tissue.

performed. There was an 8 mm?1 cm mass in the subcutaneous tissue (Fig.1). No invasion of the underlying fascia was observed. Microscopic examination of the initially removed tumour showed anaplastic ovoid or spindle-shaped cells with massive osteoid in the fatty tissue. Giant cells were found around the osteoid. The widely excised remaining tumour had more bizarre-looking cells with atypical mitosis and lace-like osteoid (Fig. 2A,B,C). The tumour was diagnosed as ESOS. Chemotherapy with Rosen T-20 [17] was administered to the patient. The patient has survived without recurrence or metastasis of the tumor for 4 years since the primary surgery.

DISCUSSION

Unlike primary osteosarcoma, which commonly develops before the age of twenty-five years, ESOSs are rarely encountered in patients under 40 years of age [8]. In Allan and Soule's series the mean age was 47.5 years [1]. Lee JS et al reported 40 patients of ESOS, most of whom presented in the sixth and seventh decades of life (mean age, 50.7 years). The lower limbs most commonly were involved (68%), usually the thigh and buttock regions [12]. Most were deep-seated and were firmly attached to the fascia but occasionally they were freely movable and confined to the subcutis or dermis [6, 13]. The size of reported ESOSs ranged from 1.5 to 30 cm

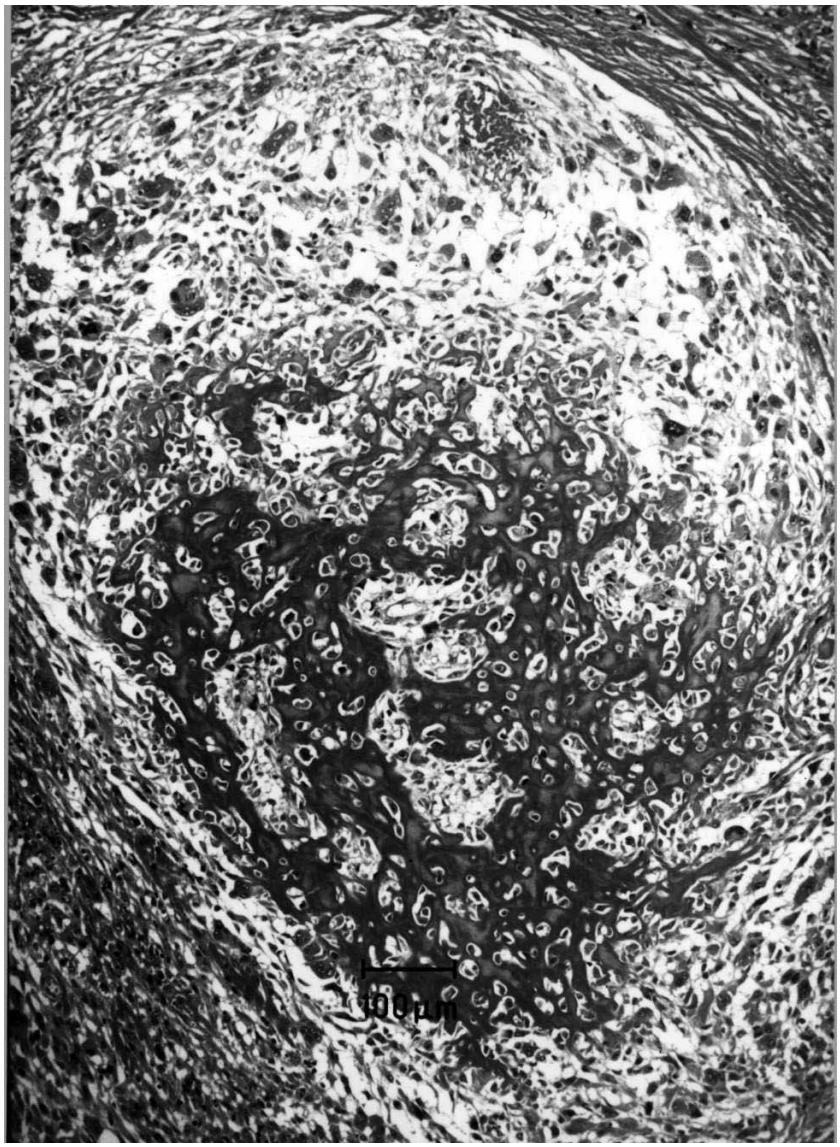

Fig. 2A. Photomicrograph demonstrating lace-like osteoid in the tumour.

[4,9]. We recognized this case as a very unique one because of its localization and its extremely small size.

Mechanical injury has been considered as a causative agent [8]. As with other neoplasms, the etiology is difficult to determine. Allan and Soule reported a history of trauma in eight of their 26 cases [1]. ESOSs have been reported to have occurred in previously irradiated areas [2,3]. Lidang et al. also reported that two patients with superficial ESOSs previously received radiation in the same area[13]. There was no evidence of previous trauma or irradiation in the present case. Judging from its

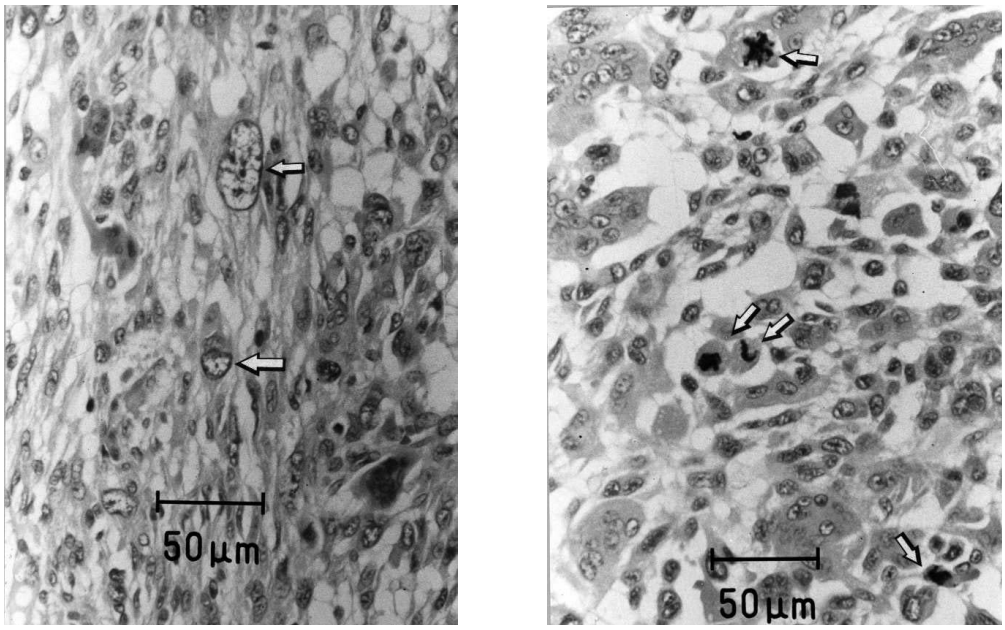

Fig. 2B and C. High-power view of the tumour showing bizarre-looking cells (arrows) (B) and atypical mitoses (arrows) (C), indicating high grade malignancy.

superficial location at the buttock, repetitive microtrauma is thought to be one of the causative factors.

The prognosis of ESOS is very poor. Lee et al analyzed 40 ESOSs and concluded that ESOS is a high-grade malignant tumour with a 5-year survival rate of 37%. Multiple local recurrences (45%) were a feature of this tumour. All recurrences occurred within 3 years and distant metastasis (65%) was also common, occurring usually in the lungs (81%) [12]. In another series of 65 patients, 40 (61.5%) had died of the tumour, 36 of recurrent or metastatic disease [8]. The present case underwent an additional wide excision followed by chemotherapy and has been disease-free for 4 years since the primary surgery. Its superficial location, extremely small size, wide excision and chemotherapy were thought to contribute to her long survival.

REFERENCES

1. Allan, C.J. & Soule, E.H.: Osteogenic sarcoma of the somatic soft tissues: clinicopathologic study of 26 cases and review of the literature. Cancer 27: 1121–1133, 1971.
2. Alpert, L.I., Abaci, I.F. & Werthamer, S. Radiation-induced extraskeletal osteosarcoma. Cancer 31: 1359–1363, 1973.
3. Auerbach, O., Friedman, M., Weiss, L. & Amory, H.I. Extraskeletal osteogenic sarcoma arising in irradiated tissue. Cancer 18: 628–633, 1951.
4. Bane, B.L., Evans, H.L., Ro, J.Y., Carrasco, C.H., Grignon, D.J., Benjamin, R.S. & Ayala, A.G. Extraskeletal osteosarcoma. A clinicopathologic review of 26 cases. Cancer 65: 2762–2770, 1990.
5. Banerjee, S. & Banerjee, P. Extraskeletal soft tissue osteosarcoma. J Indian Med Assoc 85: 242–244, 1987
6. Chung, E.B. & Enzinger, F.M. Extraskeletal osteosarcoma. Cancer 60: 1132–1142, 1987.
7. Cook, P.A., Murphy, M.S., Innis, P.C. & Yu, J.S. Extraskeletal osteosarcoma of the hand. A case report. J Bone Joint Surg Am 80: 725–729, 1998.

8. Einzinger, F.M. & Weiss, S.W. : Extaskeletal osteosarcoma. In: Soft tissue?tumors (ed. Einzinger FM & Weiss SW), pp. 1026–1034. Mosby, St.Louis, Baltimore, Berlin, Boston, Carlsbad, Chicago, London, Madrid, Naples, NewYork, Philadelphia, Sydney, Tokyo, Toronto, 1995.
9. Fanburg-Smith, J.C., Bratthauer, G.L. & Miettinen, M. Osteocalcin and osteonectin immunoreactivity in extraskeletal osteosarcoma: a study of 28 cases. Hum Pathol 30: 32–38, 1999.
10. Fang, Z., Yokoyama, R., Mukai, K., Beppu, Y. & Fukuma, H. Extraskeletal osteosarcoma: a clinicopathologic study of four cases. Jpn J Clin Oncol 25: 55–60, 1995.
11. Jensen, M.L., Schumacher, B., Jensen, O.M., Nielsen, O.S. & Keller, J. Extraskeletal osteosarcomas: a clinicopathologic study of 25 cases. Am J Surg Pathol 22: 588–594, 1998.
12. Lee, J.S., Fetsch, J.F., Wasdhal, D.A., Lee, B.P., Pritchard, D.J. & Nascimento, A.G. A review of 40 patients with extraskeletal osteosarcoma. Cancer 76: 2253–2259, 1995.
13. Lidang, J. M, Schumacher, B., Myhre, J. O., Steen, N. O. & Keller, J. Extraskeletal osteosarcomas: a clinicopathologic study of 25 cases. Am J Surg Pathol 22: 588–594, 1998.
14. Meister, P., Konrad, E.A. & Stotz, S. Extraskeletal Osteosarcoma. Case report and differential diagnosis. Arch Orthop Trauma Surg 98: 311–314, 1981.
15. Mirra, J.M., Fain, J.S., Ward, W.G., Eckardt, J.J., Eilber, F. & Rosen, G. Extraskeletal telangiectatic osteosarcoma. Cancer 71: 3014–3019, 1993.
16. Rao, U., Cheng, A. & Didolkar, M.S. Extraosseous osteogenic sarcoma: clinicopathological study of eight cases and review of literature. Cancer 41: 1488–1496, 1978.
17. Rosen, G. The central role of high-dose methotrexate in the treatment of osteosarcoma. (lecture) May 23, 1992, Nihonyakugakukainagaikinen hall, Japan.
18. Sordillo, P.P., Hajdu, S.I., Magill, G.B. & Golbey, R.B. Extraosseous osteogenic sarcoma. A review of 48 patients. Cancer 51: 727–734, 1983.
19. Sudaresn, N., Rosen, G., Fortner, J.G., Lane, J.M. & Hilaris, B.S. Preoperative chemotherapy and surgical resection in the management of posterior paraspinal tumors Report of three case. J Neurosurg. 58: 446–450, 1983.
20. Wilson, H. Extraskeletal ossifying tumors. Ann Surg 113; 95–112, 1941.

Corresponding author: Masahito Hatori, M.D., Ph.D.
Department of Orthopaedic Surgery,
Tohoku University School of Medicine,
1-1 Seiryomachi, Aobaku, Sendai, Japan 980-8574
Tel: 81-22-717-7242
Fax: 81-22-717-7248
E-mail: mhato@mail.cc.tohoku.ac.jp