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Limited Remodelling of Expansile Ewing Sarcoma after Treatment

Shigeru Ehara¹, Jin Iizuka² and Susan V. Kattapuram³

¹Center for Radiological Sciences and ²Orthopedic Surgery, Iwate Medical University
³Department of Radiology, Massachusetts General Hospital, Boston, U.S.A.

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膨隆を伴う Ewing 肉腫の非定型的治療後変化

1) 岩手医科大学中央放射線部, 2) 同 整形外科
3) Department of Radiology, Massachusetts General Hospital, Boston, U.S.A.

江原 茂¹ 飯塚 仁² Susan V. Kattapuram³

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Radiological findings of Ewing sarcoma have been well documented, and bone expansion or cystic change is known to be a relatively uncommon finding⁴. On the other hand, expected changes in Ewing sarcoma after treatment have also been reported, e.g. regression of extrasosseous soft tissue mass, regression and organization of the periosteal reaction, and remodelling of the lytic or sclerotic bone changes²⁻⁴. However, remodelling of bone is often limited or incomplete. We saw an atypical change in Ewing sarcoma after treatment due to lack of significant: remodelling of the expanding bone. Because such changes might be a cause of misdiagnosis, knowledge of it will help the radiologist in the follow up evaluation.

Case Reports

Case 1

A lytic-expanding lesion of the femur was found in a 25-year-old man with chronic left thigh pain (Fig. 1A). No periosteal reaction or extrasosseous soft tissue mass was demonstrated. Incisional biopsy showed Ewing sarcoma. Before starting treatment, he sustained a pathologic fracture through the lesion, requiring internal fixation with dynamic compression screw (Fig. 1B). Subsequently he received radiation therapy (tumor dose 54 Gy) and chemotherapy with vincristine and adramycin. Six months after the treatment, no remodelling of the lytic-expanding lesion or fracture healing was identified. One year after treatment, removal of metallic prosthesis and curettage were performed because of Staphylococcal infection at the site (Fig. 1C). No residual tumor was identified in the lesion. Sixteen months later varus angulation of the

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Fig. 1  Ewing sarcoma of left femur (23-year-old man).

1A: AP view of left femur before treatment demonstrating a centrally located slightly expanding lytic lesion. No periosteal reaction or significant soft tissue mass is identified.

1B: Pathologic fracture sustained just before the initiation of treatment is secured by a dynamic compression screw. Periosteal reaction is noted at the fracture site.

1C: One year after the treatment. Except for calcified callus in the proximal portion, no osseous union is identified. The minimal periosteal change in the lateral aspect of the shaft of the femur might be due to the fracture or infection. The femur is foreshortened.

1D: 4 years 7 months after the treatment. No interval remodelling of the cystic change in the femur is noted. The fracture site appears stabilized with pseudoarthrosis.
Fig. 2  Ewing sarcoma of the left ilium (12-year-old girl)
IA, B: AP view and CT scan of the pelvis before treatment. Lytic permeative lesion involving the left ilium and the acetabulum is noted. Exansile change is evident on CT scan. Extrasosseous soft tissue mass is minimal.
2C, D: 2 years after the treatment. Left hemipelvis is hypoplastic. Exansile change in the left ilium and the acetabulum and protrusio acetabuli are evident. These findings are similar to those of fibrous dysplasia. A small lytic change on the right anterior superior iliac spine is present.
2E, F: 2 years and 4 months after the treatment. Lytic expanding lesion in the right ilium has increased in size. There is no corresponding change in the status of the primary lesion.
proximal femur was seen with no further remodelling of the lytic focus. This lesion has been stable since, with no evidence of recurrence or metastases demonstrated 4 years 7 months after the treatment (Fig. 1D).

Case 2

A 12-year-old girl complained of left hip pain. Radiographic examination of the pelvis revealed lytic expanding lesion in the left ilium (Fig. 2A, 2B). Incisional biopsy of the lesion revealed Ewing sarcoma. Radiation (tumor dose 60 Gy) and chemotherapy with Adriamycin and cisplatinum were administered. The primary lesion became stable after 2 years with protrusio acetabuli, and the expanding lesion resembling non-aggressive process such as fibrous dysplasia (Fig. 2C, 2D). Two years and four months after the initial treatment, the patient developed another expanding lesion in the right ilium, a metastatic focus (Fig. 2E, 2F).

Discussion

The radiological manifestations in the two cases reported are unusual. The cystic expanding lesion in Case #1 resembled a cyst or fibrous dysplasia before treatment. After radiation and chemotherapy, no significant remodelling of the osseous lesion was identified. The fracture sustained just before the initiation of treatment did not heal, and the lesion stabilized with pseudoarthrosis at the fracture site. A lytic permeative lesion involving most of the left ilium in Case #2 is a more typical finding except for the significantly large expansile component. Following treatment, the expanding lesion partially remodelled. On plain radiographs, protrusio acetabuli and hypoplasia of the left hemipelvis were noted. These changes resembled fibrous dysplasia. In both cases, once the lesions had been stabilized with a significant expansile component, no further regression had occurred. Based purely on the radiological findings, the clinical course might be difficult to predict. Although regression of soft tissue mass and solidifying periosteal reaction are good indicators of the response to treatment, remodelling of the osseous component seems to be a poor predictor of clinical response.

The remodelling of the lytic or blastic changes after aggressive local treatment is often slow and incomplete. Expansile changes in high-grade lesions might be very slight following treatment, and they may occasionally resemble a benign process as demonstrated here. Without adequate clinical data and pretreatment films, it is difficult to analyze these changes. Detailed knowledge concerning the remodelling of expanding bone neoplasms, therefore, appears to be an important component in determining the response to treatment.

Acknowledgments

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