Multicentric, synchronous giant-cell tumor of bone

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ABSTRACT

Multicentric giant cell tumor is a rare variant of giant cell tumor. In this case, we report a case of a 15-year-old female patient with synchronous type of multicentric giant cell tumor.

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Giant-cell tumor of the bone is a neoplasm, which occurs in the juxta-epiphyseal region of long bones. Histologically, it consists of multinucleated giant cells. It is described as a benign but locally aggressive lesion that generally presents solitary.[1] It was described as giant-cell tumor of bone in nineteenth-century.[2] Multicentric variant of tumor rarely occurs (approximately 1% of cases).[1,3,4] The aim of this paper is to present a rare variant of giant cell tumor of bone and discuss its outcome.

Case Report. A 15-year-old female patient admitted to our clinic complaining of pain and swelling in her left ankle. In the past, she had neither trauma nor a systemic disease. On physical examination, she had hyperemia and palpable firm mass (approximately 5 x 3 cm) on posterolateral part of left ankle. Range of motion (ROM) of the joint was full. Direct roentgenogram and computerized tomography (CT) showed a well-circumscribed expansile lesion with cortical thinning in distal metaphysis of both tibia and fibula (**Figure 1**). Triple-phase bone scan revealed increased perfusion and osteoblastic accumulation localized on left ankle. Furthermore, magnetic resonance imaging (MRI) of tibial and fibular distal metaphysis showed well-circumscribed and lobulated lesions, which have high signal intensity on T2-weighted images, heterogeneous signal intensity on T1-weighted images. Thorax CT, thyroid ultrasound and blood parathyroid hormone levels were analyzed to exclude metabolic diseases and all of them were resulted as normal. After the foregoing diagnostic procedures, the tibial lesion was curetted and filled the cavity with allograft after phenolization. Using the same anterolateral incision, fibular lesion was resected segmentally, and a 7 cm proximal fibular segment was slided distally by diaphyseal osteotomy. Then, the fibula was fixed with 1/3 tubular plate and screws. Surgical specimens were sent to the Department of Pathology for pathological examination within separate solutions. Histologically, specimens showed the same appearance: lesions were composed of multinucleated giant cells and pleomorphic fusiform stromal cells. Within this framework, the diagnosis of the giant-cell tumor of bone was made. After 6-weeks of above the knee cast immobilization, partial weight bearing was allowed. At ninth postoperative week, satisfactory callus formation was obtained, and full weight bearing was allowed. At 24 months follow-up, the patient was able to move her ankle freely without any pain (**Figure 2**). Furthermore, no recurrence during the control examinations using bone scans.

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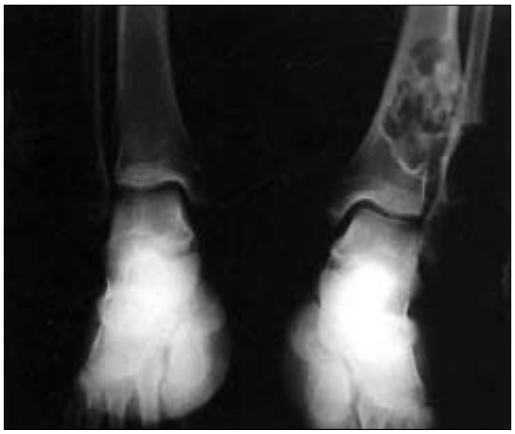

Figure 1 - Preoperative direct roentgenogram of the patient.

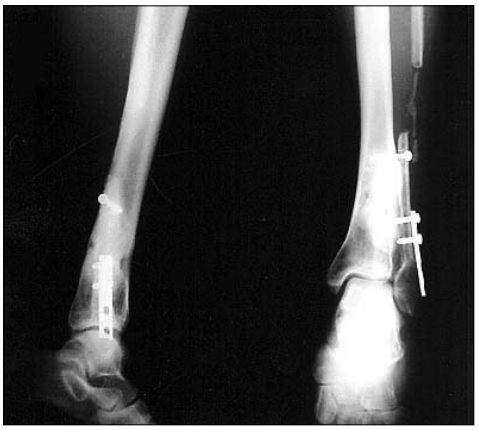

Figure 2 - Control roentgenogram in postoperative 24th month reveals union of distal osteotomy line of fibula.

Discussion. For the diagnostic consideration, other pathologies presented multicentric bone lesions such as metastatic lesions, brown tumor of hyperparathyroidism, Paget's disease, eosinophilic granuloma, multifocal infections, multiple myeloma, benign fibrous histiocytoma of bone and other bone tumors should be excluded.[4-6] In this case, we excluded all these conditions by blood analyses and imaging studies. We classified the case as grade II according to Campanacci's classification[1] due to cortical thinning and erosion appearances for both lesions. The giant-cell tumor of bone rarely occurs as multicentric lesion. If the same type of tumors is seen in different bones simultaneously, it is described as "synchronous". However, lesions which discovered at different times and locations are described as "metachronous."[7] The multicentric giant-cell tumor of bone is histologically similar to solitary form of tumor. However, the multicentric variant has increased the tendency to occur in lower ages when compared the other form. According to series (n=5) of Cummins et al,[8] the mean age was 19 (14-25 years).[4] However, solitary giant-cell tumor is rarely seen at ages under 15-year-old. Picci et al[8] reported only 6 cases <15-year-old in 326 patients. Epiphysis adjacent to the tumor was open at least in one area for all cases. Nevertheless in current case, both tibial and fibular epiphyses were closed. Therefore, we should define the case as skeletally mature. Picci et al also reported predominantly metaphyseal involvement and extension into the epiphysis adjacent to the tumor. They concluded that probable tumor origin is metaphysis and epiphyseal extension is a symptom of tumor aggressiveness.[8] Within this framework, our case has not showed aggressive behavior. It also supports the hypothesis about anatomical side of the tumor origin. Pathogenesis of giant-cell tumor has not been defined clearly. Direct extension, multiple independent foci and metastasis are possible mechanisms of multicentricity.[4,5] In this case, lesions were characterized synchronously. Furthermore, MRI and CT demonstrated that lesions did not penetrate the fibula and tibia (**Figure 1**). Therefore, metastasis and direct extension mechanisms are not convenient for the current case. Possible pathogenesis of the case might be multiple independent foci. Local recurrence of giant-cell tumor mostly occurred within 2 years.[9] In our patient, we have not observed any local or distant recurrence by imaging studies. We are maintaining the follow-ups biannually with routine roentgenograms and bone scanning studies.

In conclusion, multicentric giant-cell of bone should be kept in mind for the differential diagnosis of multiple lytic lesions also in young individuals. We recommend periodic follow-ups for the diagnosis of possible recurrence.

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