

Multifocal Synchronous Giant Cell Tumor of Bone: A Case Report

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Abstract

Since 2020, Giant cell tumor of bone (GCTB) is classified as an intermediate malignant tumor and locally aggressive in behavior however there is less chance of metastatic propensity which represents approximately 5% of primary bone tumors. It is usually occurs in long bone and multifocal synchronous GCTB is very rare accounting for less than one percent. We herein present a case report of 18 year male complaining mild right knee pain for 3 months. Patient underwent for Computed Tomography (CT), Magnetic Resonance Imaging (MRI). The examination results revealed that patient had multifocal synchronous GCTB in bilateral distal femur, bilateral proximal tibia, bilateral ilium bone (Hip bone) and bilateral ala of sacrum. Multifocal synchronous GCTBs are very rare and this tumor's exceptional positions in the site seen in our case report have rarely been reported. The correct diagnosis relies on correlation of clinical and radiological findings with confirmation of the diagnosis by histopathologic examination.

Keywords: Giant Cell Tumor of Bone (GCTB); World Health Organization (WHO); Magnetic Resonance Imaging (MRI)

Introduction

Since 2020, World Health Organization (WHO) classified Giant cell tumor of bone (GCTB) as intermediate malignant bone tumor which are known for their local aggressiveness but low metastatic tendency [1]. Patients with GCTB complain vague and imprecise symptoms including ache, overlying soft-tissue inflammation, and sometimes decreased range of movement at the contiguous joint [2]. Distant metastases are seen rarely in GCTB however can occur as the pulmonary metastases although these metastases generally have the same benign histologic appearance as the index tumor [3]. Even Multifocal synchronous GCTBs are very rare, most multifocal GCTB occurring within a poorly defined time of the initial tumor [4]. In our case report, we present the case of a

multifocal synchronous giant cell tumor of bone (GCTB) with rare location which is never reported in our best knowledge as well as a recent 5 year literature reviews of multifocal giant cell tumor of bone.

Case Report

An 18-year-old male presented to our institution with mild pain and mild swelling in right knee.

Patient was advised to have X-ray of bilateral (B/L) Knee and Magnetic Resonance Imaging (MRI) of bilateral (B/L) knee joints. X-Ray of B/L knee joints revealed that there was well outlined lucencies with sclerotic margins noted in epimetaphysis of both distal femur and both proximal tibia (Figure 1).

Figure 1: There is well outlined lucencies with sclerotic margins noted in epimetaphysis of both distal femur and both proximal tibia.

Plain and Gadolinium contrast media enhanced MRI scan of B/L knee joints was performed. T1-TSE-Tra, T2-TSE-Tra, T2-FLAIR-Tra, T1-TSE-Cor, T2-TSE-Sag, DWI-epi3trace-Tra, Contrast Enhanced T1-TSE-Tra and Contrast Enhanced T1-TSE-Cor series were performed. MRI of B/L knee joint scan revealed that there was eccentric expansile altered signal intensity lesion with internal septations of epiphyseal extension D/D giant cell tumor of bone (GCTB) on the B/L knee joint including bilateral distal femur, bilateral proximal tibia (Figure 2).

Figure 2: MRI scan revealed a eccentric expansile altered signal intensity lesion with internal septations of epiphyseal extension D/D giant cell tumor of bone (GCTB) on the B/L knee joint including bilateral distal femur, bilateral proximal tibia.

After then patient is suspected as synchronous multifocal GCTB and patient underwent for Ultra-Low-Dose Whole Body Computed Tomography (ULD-WBCT) to rule out all the remaining site of GCTB. Ultra-Low-Dose Whole Body Computed Tomography revealed that there was additional site of GCTB at bilateral ilium bone (Hip bone) and bilateral ala of sacrum (Figure 3).

Figure 3: Ultra-Low dose whole body Computed Tomography scan revealed that there was multifocal synchronous GCT of Bone in bilateral distal femur, bilateral proximal tibia, bilateral ilium bone (Hip bone) and bilateral ala of sacrum appearing as eccentric lytic lesions with internal septations without solid component, without cortical breach and without periosteal reactions.

Biopsy of all site of above given GCTB was done. Histopathological examination results of all site revealed that giant cell tumor of bone (Figure 4).

Patient is advised to have tablet Zoledronic acid (Bisphosphonates) for one month and then plan for structural reconstruction surgery with cement filling after curettage.

Figure 4: Tumor composed of diffusely dispersed osteoclast like giant cells embedded between mononuclear cells.

Discussion

Giant cell tumors are typically lesions of young and middle-aged adults, with 80% of tumors occurring in patients between the ages of 20 and 50 years, and peak prevalence in the third decade of life. They account for 4% to 5% of primary bone tumors. Multifocal GCTB are rare. Approximately 1% of cases present as multiple synchronous or metachronous lesions [5]. The multifocal GCTB have a more aggressive course, including an increased incidence of pathologic fractures [6]. There is a slight female predominance in metachronous GCT (57% versus 43%) [3] but not a 2: 1 ratio as reported in the literature [7]. Literature showed 3: 2 female: male ratio in this study. GCTB occurs between the 3rd and 5th decades of life and >80% of patients are more than 25 years old [7].

In our case patient age is 18 year old which is among less than 20% according to literature. Solitary benign GCTB may metastasize to the lung or undergo malignant transformation (either de novo or following irradiation); however, pathologic analysis of multifocal GCTB reveals findings identical to histologically benign solitary tumors [3]. This suggests that the multifocality of some GCTB is not a metastatic phenomenon but rather represents the separate development of the tumor at multiple sites [1,2]. Iatrogenic seeding may represent a cause of multicentric giant cell tumors [8].

The most common site of the primary GCT is around the knee (44%), followed by wrist (23%) and hand and feet (13%), and is consistent with localization of solitary GCTB. In our case, the tumor site seen i.e. bilateral ilium bone (Hip bone) and bilateral ala of sacrum is really rare according to literature. Our study suggest that either synchronous or metachronous GCTB, patient is recommended to have a skeletal survey for these tumors as well as multiple follow-ups. Skeletal survey can be done either by General X-Ray or Ultra-Low-Dose Whole Body Computed Tomography (ULD-WBCT) or ^{99m}Tc-MDP whole body bone scan but the sensitivity, specificity and accuracy of Computed Tomography (CT) and ^{99m}Tc-MDP whole body bone scan is better than general X-Ray.

General symptoms include pain, swelling, and limited range of motion when the tumor occupies joint spaces [9]. GCT in the bone most often present with symptoms such as feelings of pain or limited range of movement of joints or no symptoms that affect activities of daily life. Our patient's presentation of mild pain that may be common in other reported cases.

There are wide range of treated are protocol are mentioned in literature. If GCTB tumor size is large and easy location, wide resection is recommended, a decision unlikely to be changed based on tumor staging. GCT of bone is generally treated with curettage followed by bone cement filling [10]. Curettage has been linked to up to 40% rates of recurrence. Wide resection has been found to have little to no recurrence, though rates of post-operative complications are significantly higher than in curettage [11]. Adjuvants to curettage therapy have helped decrease recurrence rates, and modalities include cryosurgery, high-speed burring, phenol, and more [11]. Though surgery remains the mainstay of treatment, chemotherapy options are available as well. Bisphosphonates are one of the most favored agents due to their anti-osteoclastic action; in particular, nitrogen containing bisphosphonates such as Zoledronic acid are especially cytotoxic to osteoclasts [12]. Denosumab, a relatively newer anti-osteoclastic agent that acts via the RANK-L pathway, may also be a good option, especially as a neoadjuvant to surgical intervention or in unresectable tumors. It has been shown to reduce morbidity and improve outcomes in such settings [13]. In our case, all the sites are not possible for curettage followed by bone cement filling i.e. ala of sacrum but tibia, femur and pelvic bone curettage are possible. For the first one month, Denosumab was given and asked for follow up.

Conclusion

In summary, multicentric giant cell tumor of bone (GCTB) is rare and most commonly affects long bones, particularly those around the knee. It tends to occur in younger patients and frequently manifests as synchronous lesions. In addition, lesions of multicentric giant cell tumor of bone (GCTB) may have an unusual metaphysodiaphyseal location. Virtually all tumors have areas with typical histopathologic features of giant cell tumor. As in solitary giant cell tumor of bone (GCTB), the most aggressive behavior of the vast majority of multicentric giant cell tumors is local recurrence, especially in multicentric GCTB of hand and feet, although there have been rare cases of metastasis to the lungs. Because a variety of other primary bone lesions may also have a polyostotic presentation, the correct diagnosis relies on correlation of clinical and radiographic findings with confirmation of the diagnosis by histopathologic examination. Our case report strongly recommended that multicentric synchronous/metachronous giant cell tumor of bone (GCTB) should be under went for whole body skeleton survey (Ultra Low Dose Whole Body Computed Tomography).

Conflict of Interests

NA.

Authors' Contribution

All authors have contributed equally to the preparation of this paper.

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