

Bicentric Synchronous Giant cell tumor: A Rare Managed Case Report of Bilateral Fibula Neck Tumor

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Learning Point of the Article:

Multicentric GCT is a known entity and diagnosis should be considered after thorough metabolic workup and after ruling out more common polyostotic skeletal lesions. This case report highlights good functional outcome can be achieved with meticulous en bloc resection in proximal fibula Giant cell tumor.

Abstract

Introduction: Multicentric giant cell tumor (GCT) of bone is an uncommon variant of the typical solitary lesion, with numerous large series reporting an incidence of 0–1.4%. Multifocal lesions also appear to be more locally aggressive than their solitary counterparts and have higher rates of recurrence. Solitary GCT of proximal fibula usually involves fibular head, bicentric synchronous bilateral fibular neck involvement is a rare presentation.

Case Presentation: An otherwise healthy 24-year-old boy presented with a palpable mass in the region of his left proximal fibula and B/L knee pain. He reported pain with activity but no peroneal nerve symptoms. Radiographs of his left knee revealed an expansile lytic lesion at the proximal fibula epimetaphyseal level while the right knee X-ray showed a similar smaller lesion. MRI was done to delineate the accurate extent of the tumor. On the left side, the patient underwent partial fibulectomy (en bloc resection) and chemical cauterization of the edges with 5% phenol. The other side GCT was smaller and the patient was largely asymptomatic, hence was planned for conservative management.

Conclusion: Multicentric GCT is a known entity and diagnosis should be considered after thorough metabolic workup and after ruling out more common polyostotic skeletal lesions. Selected patients with aggressive (benign) and malignant tumors of the proximal fibula can be treated successfully by resection and with supplementary soft-tissue reconstruction, a good functional outcome can be anticipated.

Keywords: Bicentric, synchronous, fibular neck, giant cell tumor.

Introduction

Coopers and Travers introduced giant cell tumor (GCT) of bone for the 1st time in 1818 [1]. It is a discrete histopathologic tumor containing poorly differentiated stromal and multinucleated giant cells.

They are characteristically benign but locally aggressive lesions accounting for approximately 5% of primary bone neoplasms [2]. Multicentric GCT of bone is an uncommon variant of the typical solitary lesion, with numerous large series reporting an incidence of 0–1.4% [3]. Multifocal lesions also appear to be more locally aggressive than their solitary counterparts and

have higher rates of recurrence. Their clinical and radiographic differential diagnosis usually includes a variety of polyostotic skeletal lesions such as brown tumor, Paget disease, non-ossifying fibroma, enchondromatosis, fibrous dysplasia, giant cell reparative granuloma, and Langerhans cell histiocytosis. We hereby present a rare managed case of bicentric synchronous GCT in bilateral fibula neck.

Case Report

An otherwise healthy 24-year-old boy presented with a palpable mass in the region of his left proximal fibula and B/L knee pain

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Author's Photo Gallery



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Figure 1: Coronal T1 MRI and lateral X-ray of the left knee.



Figure 2: AP and lateral X-ray of the right knee

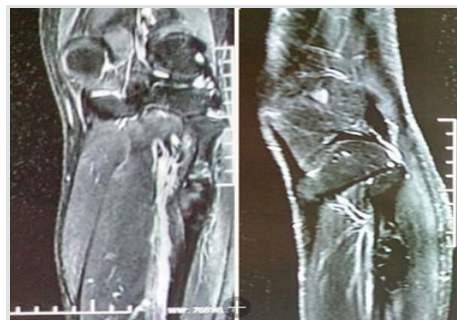


Figure 3: T2 sagittal and coronal MRI of the left knee.

for 6 months. He reported pain with activity, but there were no peroneal nerve symptoms. Radiographs of his left knee revealed an expansile lytic lesion at the proximal fibular epimetaphyseal level while the right knee X-ray showed a similar smaller lesion which prompted further evaluation (Fig. 1, 2). MRI was done to delineate the accurate extent of the tumor and to rule out involvement of the adjacent soft tissues (Fig. 3).

Histological diagnosis was made using a core needle biopsy sample obtained using fluoroscopic guidance under local anesthesia from both the sides. Histology showed sheets of uniform oval to polygonal mononuclear stromal cells admixed with numerous large osteoclast-like giant cells. Radiographs of other probable sites of GCT such as bilateral shoulder, wrists, and hands were normal. After the metabolic workup to rule out other disorders, the patient was planned for excision of the lesion. The aim of the surgery was en bloc excision of the lesion, while still preserving the peroneal nerve and peroneal vessels.

On the left side, the patient underwent partial fibulectomy (en bloc resection) and chemical cauterization of the edges with 5% phenol (Fig. 4). After fibulectomy, proximal stump was around 2 cm with attachments intact and knee was found to be mediolaterally stable. To ensure negative margins, biopsy from the edge of the remnants on both sides was taken. The other side GCT was smaller and the patient was largely asymptomatic, hence was planned for conservative management with regular follow-up. Pathology specimens revealed sheets of uniform oval to polygonal mononuclear stromal cells admixed with numerous large osteoclast-like multinucleate giant cells, a histological picture consistent with GCT of bone. No malignant features were noted. Post-operative expected high-risk



Figure 4: Post-operative AP and lateral X-ray of the left knee.

complication of peroneal nerve palsy was not seen. Post-operative period showed uneventful course of events. At 1-year follow-up, he remains fully active with no ligamentous laxity and free of distant metastasis. The other side also remains asymptomatic. No secondary surgical procedures were required.

Discussion

In its most common presentation, GCT is a solitary neoplasm occurring in the epimetaphysis of a long bone in a mature young adult. Multifocal lesions are more likely to be confined to the metaphysis (as in our case) and diaphysis when a long bone is involved in contrast to the traditional metaphyseal location.

Moertel [4] introduced the following terms in an effort to clarify the situation. They defined synchronous lesions as those remote from one another discovered within a short period of time (within 6 months). Metachronous lesions are those discovered at different times (more than 6 months interval) and in different locations. These lesions ultimately may represent metastasis or a second independent focus of disease.

The incidence of multicentric primary GCT is rare, occurring in approximately 1% of all patients with GCTs, the literature suggests that the average age of presentation of patients with multifocal GCTs is younger than that of patients with solitary lesions [5, 6] and their pathogenesis is unknown. Multicentric GCTs may arise after invasion of bone and soft-tissue adjacent to the initial tumor, especially in the hand, some GCTs also spread by crossing synovial joints or proximally along a limb and such skip lesions may also occur through lymphatics [7, 8]. Iatrogenic seeding is also attributed as a cause for metachronous lesions [9]. Multiple independent foci of disease or distant metastasis are more likely mechanisms to explain bicentricity of GCTs. Overall patients with multicentric GCTs have 2–3 lesions, but a case with as many as 10 tumors has been described in the literature [10]. Multicentric GCTs have high tendency of affecting hands and bones around the knee (distal femur and proximal tibia).

The diagnosis of GCT of bone depends on clinical, radiological, and pathologic features. The diagnosis of this condition should

be viewed with suspicion and considered only after other diseases that can present with multiple lytic lesions, such as brown tumors of hyperparathyroidism, multiple infection, metastases from unknown primary, and Paget's disease, have been excluded. Individual lesions in a patient with multicentric disease are radiographically and histologically indistinguishable from the typical solitary lesion. In addition, the histologic appearances of the tumors are of no significant value in predicting the behavior of the lesion.

Fibular head resection may cause post-operative knee instability because the LCL is the main resistor of varus loading. Peroneal nerve palsy and local recurrence are serious post-operative complications associated with resection of these tumors. Based on the literature, the incidence rate of post-operative peroneal nerve palsy ranges from 3% to 57% [11], while local recurrence rates vary by tumor histology and resection type. Wide resection decreases the local recurrence rate but increases patient morbidity so intralesional extended curettage with either physical or chemical methods is commonly advocated. When this tumor presents at anatomical locations which are "dispensable" from the point of view of weight-bearing and skeletal stability like in distal ulna or

proximal fibula, the disease is often best managed by en bloc resection to decrease the risk of local recurrence.

Conclusion

Solitary GCT of proximal fibula usually involves fibular head and bicentric synchronous bilateral fibular neck involvement is a rare presentation. Selected patients with aggressive (benign) and malignant tumors of the proximal fibula can be treated successfully by resection and with supplementary soft-tissue reconstruction, a good functional outcome can be anticipated.

Clinical Message

Multicentric GCT is a known entity and diagnosis should be considered after thorough metabolic workup and after ruling out more common polyostotic skeletal lesions. This case report highlights importance of employing meticulous surgical technique after thorough pre-operative workup and a keen eye to actively look out for other sites in the body owing to the known multicentric nature of the GCT.

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