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Progressive Course of Recurrent Giant Cell Tumor of the Distal Tibia in Adolescent: A Longitudinal Case Report

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ABSTRACT

Introduction: Giant cell tumor of bone typically arises from the epiphysis of long bones; involvement of the distal tibia is a relative rarity, especially in adolescents. While this tumor is benign, GCTB may behave quite aggressively with recurrence or even pulmonary metastasis. The case below describes the natural history of a distal tibial GCTB that was complicated by recurrence and lung metastasis, detailing the use of multiple modalities of management.

Case presentation: A 17-year-old female patient presented with an eight-month history of progressive swelling and pain in the left ankle. Imaging showed an expansile lytic lesion in the distal tibia; biopsy confirmed GCTB. She then underwent wide excision with reconstruction using a fibular graft and talofibular arthrodesis. Two years later, during surveillance imaging, pulmonary nodules consistent with metastasis were shown; she was thus treated with monthly intravenous zoledronic acid, 4 mg for six months. In 2019, a local recurrence was detected and treated with curettage and augmentation with bone cement. On follow-up in 2021, the patient had no pain, was fully ambulatory, and was without further recurrence or metastatic progression.

Discussion: Distal tibial GCTB has a high tendency for recurrence despite adequate resection due to the poor soft-tissue cover surrounding it. Though pulmonary metastasis is rare, the case indicates the importance of long-term thoracic surveillance. Zoledronic acid in this case contributed to the radiological stabilization of metastatic lesions and thus appears as a possible adjunct in recurrent or metastatic GCTB.

Conclusion: This case illustrates the indolent course of the distal tibial GCTB and represents an example of tailored multi-modality treatment with extended follow-up. Zoledronic acid may offer effective disease stabilization to a highly selected patient population at high risk.

Keywords: Giant cell tumor, progression, recurrence, distal tibia, fibular graft, arthrodesis

INTRODUCTION

Giant cell tumor of bone (GCTB) is a type of bone cancer that breaks down bone tissue. It usually grows aggressively in the ends of long bones, like the distal femur, proximal tibia, and distal radius. It's rare for the distal tibia to be involved, especially in teenagers, which makes diagnosis and surgery more difficult.[1,2] Even though

GCTB looks benign under a microscope, it can behave aggressively. Recurrence rates after surgery are reported to be between 20% and 50%.[3] Pulmonary metastasis occurs in 1% to 9% of cases. This occurrence is often referred to as benign pulmonary metastasizing GCTB.

Recent improvements in additional treatments, like denosumab and bisphosphonates, have provided more options for treating aggressive or high-risk GCTB.[4] However, there is limited evidence on how well zoledronic acid works, particularly in cases of recurrent GCTB with lung metastasis. Current clinical trials and studies primarily concentrate on denosumab or direct comparisons between the two treatments. This report is valuable because it describes the progression of GCTB in the rare location of the distal tibia. The case involved recurrence and lung metastasis, but it was successfully treated with zoledronic acid.[2] The patient received care at RSUP Prof. Ngoerah in Denpasar, Bali. This academic center allowed for a team approach and long-term monitoring according to current medical standards.

CASE PRESENTATION

A 17-year-old female presented in early 2016 with a gradually enlarging lump on the left ankle associated with pain. The mass, which has occurred since 8 months ago and has become increasingly painful over the past 5 months, has caused a slight loss of weight in the last few months, but there is neither trauma nor fever. Physical examination found a satisfactory general condition and some mild pain on the left ankle. Ankle palpation revealed a 6 cm solid-to-soft mass on the anterior side with a fixed location, tenderness, an ill-defined margin, and a fixed mass at the distal tibia with limited ankle range of motion. The patient denied any trauma or accident involving his ankle before the symptom occurred for the first time. Radiographs demonstrated an expansile lytic lesion on the distal tibia that indicates a primary malignant bone tumor suspected to be osteosarcoma from the X-ray ankle AP/lateral view (Figure 2).

MRI shows a solid mass measuring 7.1 cm x 7.9 cm x 11.3 cm located in the distal one-third of the tibia, involving both the metaphysis and epiphysis, with a clear border and no infiltration into surrounding tissues (Figure 3).

After the patient underwent a biopsy and the sample was sent to the Pathology Anatomy lab, the pathologists established the morphology of a giant cell tumor. Subsequently, the patient was diagnosed with a giant cell tumor of the left distal tibia and was planned to undergo wide excision with fibular graft and talo-fibular arthrodesis.

In October 2016, the patient underwent wide excision of the tumor with fibular bone graft reconstruction and talofibular arthrodesis by an orthopedic surgeon that has expertise in orthopedic oncology. The procedure went well, with a mass with a size of 14 cm in length and 8 cm of bone taken out and sent to the histopathology lab, where postoperative histopathology confirmed the mass is a giant cell tumor.

After two years, the patient reported an uncomfortable feeling in her chest while breathing. A chest X-ray revealed enhanced nodules in the lung that suggested a mass, which was confirmed by MSCT Thorax with contrast. This finding indicated a metastatic process, leading to her receiving monthly 4 mg zoledronic acid (Zometa) therapy for six months as adjuvant treatment.

However, in September 2019, recurrence was detected both clinically and radiographically at the surgical site. The patient then underwent curettage and bone cement augmentation. In March 2020, subsequent follow-up demonstrated stable incorporation of graft and cement, with preserved knee motion and stable ankle fusion. Serial chest CT and radiographs showed no evidence of pulmonary metastasis.

At her most recent follow-up in 2021, the patient was pain-free, ambulatory without

assistive devices, and showed no evidence of further recurrence.

Figure 1 Clinical appearance of the patient showing a swelling on the ankle without any trauma that occurs suddenly without any triggers.

Figure 2 X-Ray Results of the patient show some expansile lytic lesions on the distal tibia.

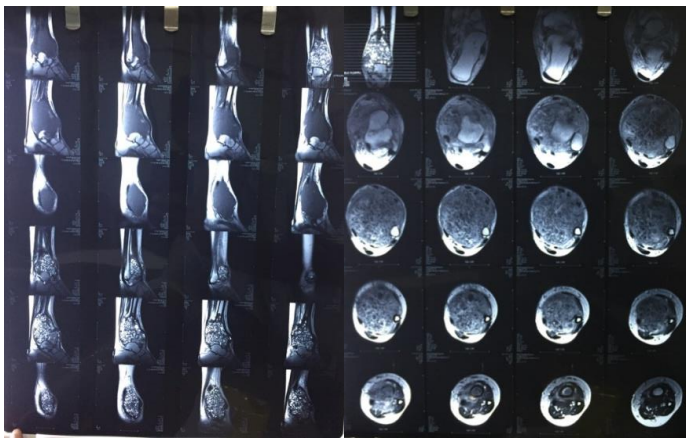

Figure 3 The MRI results indicate a solid mass in the distal tibia measuring 7.1 cm x 7.9 cm x 11.3 cm, with clear borders around the tumor.

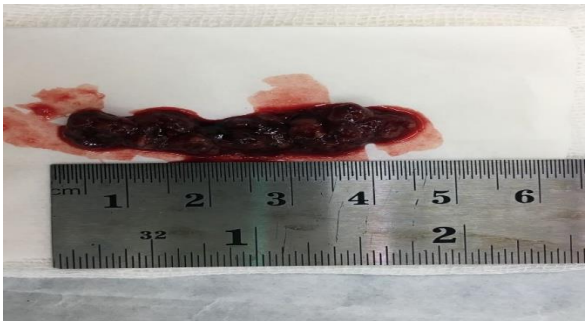

Figure 4 5 cm of tissue taken out as a biopsy sample of the patient

Figure 5 Surgical management of the GCTB

Figure 6 Dissected GCTB

DISCUSSION

Giant cell tumors of bone in the distal tibia do not show up frequently. It creates some real surgical hurdles because of the thin soft tissue covering and how close it sits to the ankle joint. Those features can make it tough to get clear wide margins during surgery.[1] That issue raises the chances of the tumor coming back. It happens even with strong resection efforts. Recurrence numbers from reports are still fairly high. They run from 20 percent to 50 percent after curettage. Then there is about 10 percent after wide excision. All this points to the challenge in keeping local control solid over time. This is particularly true in areas that are difficult to control.[5]

Lung metastasis does not occur in many GCTB cases. It shows up in just 1 to 9 percent of them. Still, it stresses why regular chest checks matter in a structured way. These mets often take years to appear after the first surgery. Some newer studies put the average wait at 2 to 5 years.[6] In our patient's case, the lung nodules developed two years post-op. That fits right into the expected pattern. Spotting it early let us start systemic treatment without delay. It also set up ongoing scans to track things closely.

Adjuvant systemic options are gaining ground for recurrent or metastatic GCTB. Denosumab and bisphosphonates both act on the stromal cells that drive the tumor. One randomized trial found zoledronic acid worked about as well as denosumab. The result held true for cases where surgery could not save the situation. So zoledronic acid looks like a solid choice. It is particularly beneficial in situations where denosumab is difficult to obtain or prohibitively expensive. Recent reviews that compile data also support the use of bisphosphonates. They help cut recurrence after scraping out the tumor.[2,4]. Reports from individual cases suggest that zoledronic acid may be effective in managing the case.[3]

This case has some strong points. We followed the patient for a long time. We caught the metastasis early on. The treatment mixed surgery with drugs and regular checks. But there were limits, too. Anatomy kept us from wider margins. Solid trial data on bisphosphonates for metastatic GCTB is missing. Even so, the lung mets stayed stable on scans. No more recurrence happened either. That gives useful clues

about zoledronic acid as an add-on treatment.[7]

Key takeaways here cover a few things. Surgery needs tailoring for odd spots like this. Long-term scans are crucial to watch for issues. Bisphosphonates might play a part in handling recurrent or spread GCTB. We need more forward-looking studies, though. They should sort out who benefits most and how to use the therapies best.[8]

CONCLUSION

Giant cell tumors in the distal tibia do not show up often. They bring real difficulties when it comes to getting clear surgical margins and keeping the disease under control over time. In this particular case, things proceeded with a proper wide excision at first along with reconstruction. Even so, the tumor came back, and pulmonary metastasis showed up later on. All that points to the need for careful, ongoing follow-up in a structured way. For this patient, zoledronic acid helped stabilize the metastatic spots quite well. That outcome suggests bisphosphonates could serve as a useful add-on treatment for certain high-risk or recurring cases of GCTB. When dealing with aggressive forms or tumors in unusual spots, a team-based approach stays key. Individualized plans for treatment help achieve the best results possible for these patients.

Declaration by Authors

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