

Persistent Challenges: A Recurrent Adamantinoma – Case Report

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Learning Point of the Article:

Adamantinoma recurrence highlights the significance of interdisciplinary care and long-term surveillance for the best possible patient outcomes.

Abstract

Introduction: Adamantinoma is a rare, locally aggressive bone tumor that primarily affects the long bones, with a predilection for the tibia. Although considered a low-grade malignancy, adamantinoma is notorious for its high propensity for recurrence, which poses significant clinical challenges in the management of affected individuals. This case report aims to explore the intricacies of recurrent adamantinoma of the tibia, shedding light on its clinical presentation, diagnostic modalities, treatment strategies, and prognostic factors.

Objective: This case report aims to evaluate surgical and functional outcomes of post-excision and reconstruction in recurrent adamantinoma.

Case Report: This retrospective case report includes the patient who presented in Shaukat Khanum Memorial Cancer Hospital and Research Center Lahore with recurrent adamantinoma. Data were collected after patient's written and informed consent, and a retrospective analysis of the patient's demographics was done with the Statistical Package for the Social Sciences using the information in the Hospital Information System with a predesigned pro forma. Histology slides were collected from the Department of Pathology. Variables that were included are categorized into demographics, history and examination findings, site, and size of the primary tumor, radiological findings, histological type, stage, and resection margins. The patient underwent wide margin excision and reconstruction with free vascularized fibula graft and his functional outcome was evaluated using the musculoskeletal tumor society score which showed good functional outcome.

Conclusion: Despite therapeutic challenges, wide excision of the tumor followed by free vascularized fibula grafting for reconstruction is a viable option to treat such patients where advanced prosthesis and allografts are not available.

Keywords: Case report, adamantinoma, recurrence, vascularized free fibula graft.

Introduction

Adamantinoma is a rare primary bone tumor that primarily affects the long bones, most commonly the tibia [1]. Adamantinoma of the tibia was first described by Fischer in 1913 [2]. It accounts for a mere 0.1–0.5% of all primary bone tumors, rendering it an exceedingly rare pathology. Despite its infrequency, adamantinoma warrants attention due to its unique histological features, characterized by the presence of epithelial and osteofibrous components. This distinct histology

contributes to the challenges in diagnosis and treatment.

Recurrence remains a hallmark of adamantinoma, with reported rates ranging from 20% to 50% [3-5]. The reasons behind this high recurrence rate are multifactorial, including incomplete initial excision, the tumor's locally infiltrative nature, and a paucity of effective adjuvant therapies [6]. Understanding the factors contributing to recurrence is imperative for improving patient outcomes and reducing the burden of this disease. This case report explores a recurrent adamantinoma case and delves

Author's Photo Gallery



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Figure 1: X-ray of left tibia and fibula anterior-posterior and lateral views showing radio-opaque bone cement in the distal shaft of the tibia with adjacent radiolucency around it.

into the clinical and pathological aspects, along with the management strategies for this enigmatic bone tumor.

Case Report

A 23-year-old male presented in the outpatient department with biopsy-proven adamantinoma for which excision, curettage, and bone cementing were done in another hospital 15 days back before presentation in our hospital (Fig. 1). On examination, there was an 8 cm scar present on the anterior aspect of the left distal tibia which is healed by primary intention. No swelling was palpable below the scar and his

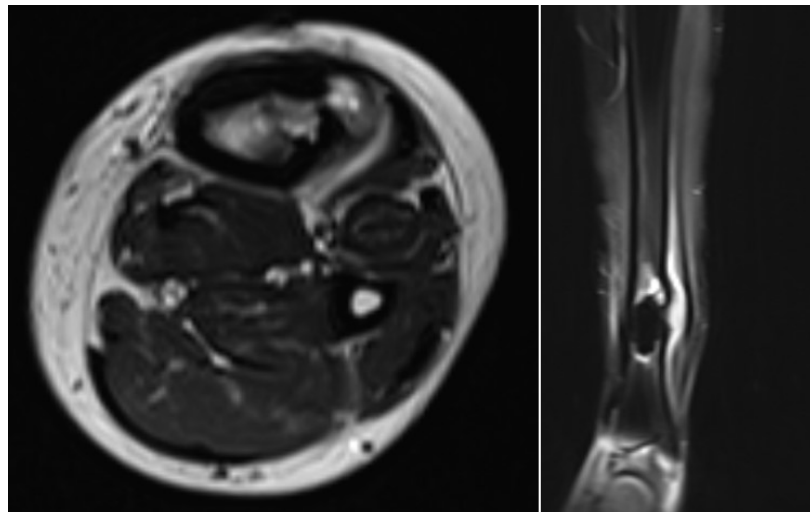


Figure 2: Magnetic resonance imaging of left tibia axial and coronal images showing post-contrast enhancement.

neurovascular bundle was intact. The patient was re-scanned in our hospital to look for any possible residual disease and metastasis. On magnetic resonance imaging (MRI) of his leg, there were only post-surgical changes with no evidence of any residual disease; however, his computed tomography chest showed indeterminate subpleural nodule. The case was discussed in a multi-disciplinary team (MDT) meeting including orthopedic surgeons, oncologists, and radiation oncologists, and it was decided to keep the patient on close follow-up at 3-month intervals. The patient had stable disease on subsequent scans for 9 months when a recent MRI left lower limb showed an abnormal signal intensity area identified along the superolateral aspect along the surgical cavity showing post-contrast enhancement with diffusion restriction appearing suspicious for infective etiology/recurrence (Fig. 2). Hence, the case was again discussed in MDT and it was decided to get the patient's infective profile and biopsy of the lesion. The patient's infective profile came out to be negative. Moving forward ultrasound-guided trucut biopsy was performed to confirm the diagnosis, and histopathological examination revealed the characteristic biphasic pattern with epithelial nests within a fibrous stroma confirming the recurrence of the disease (Fig. 3). The patient was again discussed in MDT and radical resection of the tumor was decided. The surgical plan was discussed with the patient in detail and then after careful deliberation, the patient underwent a wide surgical resection followed by a limb-salvaging reconstruction procedure using a vascularized free fibula graft taken from the ipsilateral limb (Fig. 4 and 5). The patient had a smooth post-operative recovery and was discharged home after 5 days and was advised regular clinic follow-up. The patient was kept in a splint for about a month, and then, a range of motion exercises were advised. Partial weight-bearing was started at 2 months and full weight-bearing was allowed after 3 months. On the current 12-months follow-up, the patient had a revised musculoskeletal tumor society score of 24 which represents good functional outcome.

Discussion

Adamantinoma is a rare entity, and recurrent cases are even more infrequent. This recurrent adamantinoma case underscores several important clinical and therapeutic aspects. The hallmark of adamantinoma is its biphasic histology, which includes both epithelial and stromal components [7]. This distinct histological pattern can assist in diagnosis and differentiation from other bone tumors.

Individuals who have long bone adamantinoma

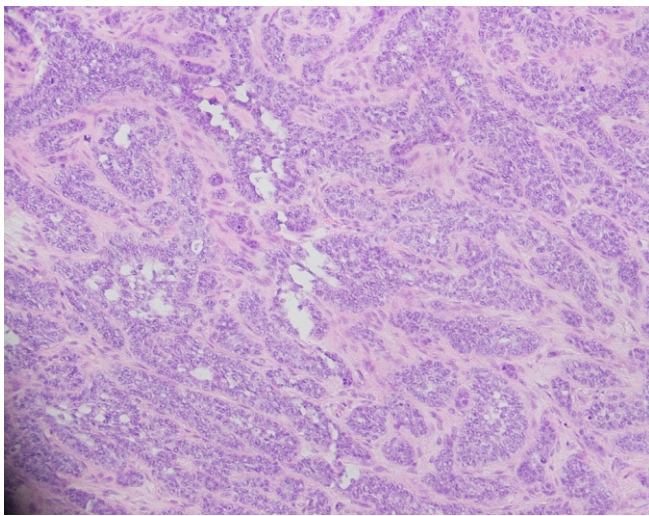


Figure 3: Histopathology slide showing biphasic appearance typical of adamantinoma.

frequently experience pain and gradually increasing swelling, and they may have experienced trauma in the affected location in the past [3]. Radiologically, adamantinoma is characterized by an eccentric, expansive, and osteolytic appearance; skip lesions, cortical disintegration, and moth-eaten edges are frequently observed [8]. Information regarding intramedullary extension and tumor-free margins can be obtained using MRI [9].



Figure 4: X-ray left tibia anterior-posterior view on last follow-up showing fibula graft onto resected tibial defect, fixed with locking compression plate.

Adamantinoma is known for its propensity to recur after surgical resection, emphasizing the need for vigilant follow-up in treated

patients. The mechanisms underlying this tendency are not yet fully understood, highlighting the importance of ongoing research in the field. Adamantinoma can spread to other areas and has a high probability of local recurrence. In a retrospective analysis, Houdek et al. found that 15% of 46 patients with surgically treated adamantinoma of the extremities experienced local recurrence [10]. According to Qureshi et al's assessment of 70 patients who underwent surgery for long bone adamantinoma, the local recurrence rate was 18.6% [3]. In addition, Scholfield et al. observed that following surgical therapy, three out of 12 patients with osteofibrous dysplasia-like adamantinoma developed a local recurrence [11]. Furthermore, long-term follow-up research showed that 27% of patients experienced metastasis, with the lung being the most common site [3]. The mean duration for a local recurrence was 8 years, and the mean duration for metastasis was 7 years in the prior study.

The management of recurrent adamantinoma often necessitates a multidisciplinary approach, involving orthopedic surgeons, oncologists, and radiation specialists. Decisions regarding surgical techniques and the timing of radiation therapy must be individualized to each patient's case [12].

Limb-salvaging surgery is considered in recurrent adamantinoma cases to maintain function and quality of life. The choice of surgical technique depends on the extent of the recurrence, patient factors, and the surgeon's experience. Different modalities can be considered when deciding on local control of the disease with their pros and cons. Options of wide local excision alone, or with reconstructive limb salvaging



Figure 5: Clinical images showing well-healed skin pedicle graft of vascularized fibula graft and well-healed donor site.

procedures can be considered using autografts or allografts, or endoprosthesis. Rarely, patients may require amputation for non-reconstructable tumors [3].

Conclusion

Recurrent adamantinoma is an uncommon but challenging clinical scenario. This case report highlights the importance of early diagnosis, comprehensive evaluation, and a multidisciplinary approach in the management of recurrent adamantinoma. While surgical resection remains the primary mode of treatment, the propensity for recurrence underscores the need for vigilant follow-up and research to better understand the underlying mechanisms. This enigmatic bone

tumor continues to pose clinical challenges, and further studies are essential to improve our understanding and treatment outcomes for adamantinoma patients.

Clinical Message

Recurrent incidents of adamantinoma highlight a clinical message that underscores the need of continuing surveillance and interprofessional teamwork. To improve patient outcomes and quality of life, it emphasizes the necessity of maintaining vigilant monitoring even after initial therapy, ensuring prompt discovery and management of recurrences. This stresses the significance of a patient-centered approach that incorporates multidisciplinary interventions and routine follow-ups to properly manage the intricacies of this uncommon malignancy.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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