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Case Report

Tumor to triumph: bone reconstruction with extracorporeal irradiation in malignant bone tumors

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ABSTRACT

Malignant bone tumors (MBTs) are predominantly seen in children and adolescents and account for <1% of diagnosed cancers each year. This case study demonstrates how ECI acts as a limb salvage method for MBT, particularly in low-income settings. A 17yr old male patient present pain & swelling around left ankle for 1 year. The radiograph showed lytic lesion in the left distal fibula followed by biopsy suggestive of osteosarcoma. Patient underwent four cycle of Adriamycin based chemotherapy followed by wide local excision with extracorporeal irradiation. The patient also received four cycles of adjuvant chemotherapy. Now, patient was free of disease and on regular follow-up. By delivering high-dose radiation directly to affected bone ECI helps to minimize damage to surrounding healthy areas. It also offers significant therapeutic advantages, including enhanced tumor control and the preservation of limb functionality. As technology and techniques continue to evolve, ECI may play an important role in treating cancers that are challenging to manage with conventional therapies.

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1. Introduction

Malignant bone tumors (MBTs) are predominantly seen in children and adolescents and account for <1% of diagnosed cancers each year. Currently, MBTs are managed by limb salvage surgery, which acts as an oncologically safer alternative when compared to amputation which was practiced earlier.¹ Extracorporeal irradiation, or ECI, was introduced by Spira and Lubin in 1968 and refers to the process of re-implanting a patient's bone following radiation therapy using a single fraction high dose of radiation. This case study demonstrates how ECI acts as a limb salvage method for MBT, particularly in low-income settings.

2. Case Report

17-year-old male patient presented with complaints of pain and swelling around the left ankle for 1 year. The pain was insidious in onset, gradually progressive, dull-aching, and non-radiating in nature with a numeric pain rating scale (NRS) score of 7. The swelling was insidious in onset, gradually progressive and associated with a restricted range of movements. There was no history of fever, weight loss, reduced appetite or trauma. On clinical examination, a 5 x 4 cm swelling was present over the lateral aspect of the left ankle. The overlying skin was hypopigmented. The swelling was ill-defined, had a smooth surface and on palpation was nontender and bony hard in consistency. There were no open wounds and sinuses and the distal pulses were palpable. The patient had no significant family history, no notable past medical history and no known co-morbidities.

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The radiograph of the left ankle showed an expansile lytic lesion involving the left distal fibula, with a sunburst appearance, periosteal reaction and cortical breach (Figure 1A). Routine blood investigations were within normal limits. The patient underwent an ^{18}F -fluorodeoxyglucose (FDG) based Positron emission tomography-computed tomography (PET-CT) scan which showed a 3.2 x 2.8 x 6.3 cm osteolytic lesion with soft tissue component involving distal end of the left fibula with periosteal reaction with a standard uptake value (SUV) max of 7.3 (Figure 1B). No other metabolically active lesion was detected. The patient underwent core needle biopsy from the site of bony lesion which showed features consistent with osteosarcoma.

The patient was planned for neoadjuvant chemotherapy (NACT) (2 x Adriamycin, Cisplatin(AC) + 2 x Adriamycin, Ifosfamide (AI). After completion of 4 cycles of chemotherapy response assessment was done with PET-CT which showed a reduction in the size of the primary lesion from 3.2 x 2.8 x 6.3 cms to 1.9 x 1.5 x 5.9 cms and a reduction in metabolic activity from SUVmax 7.3 to 5.7, suggestive of partial response. A tumor board discussion was done in January 2023 and then the patient was taken up for wide excision of the tumor with extracorporeal irradiation (ECI) of the resected bone followed by reconstruction. The procedure was performed by a multidisciplinary team of orthopedic oncology surgeons and radiation oncologists. The tumor-bearing bone segment was resected en bloc. The resected bone was then washed with an antibiotic solution. Then the bone was packed in sterile covers and sheets. This bone was then sent for ECI and a single dose of 50Gy with a beam energy of 15MV was given over 16 minutes (Figure 2).

The irradiated bone was then brought back to the operation theatre and was again washed with antibiotic solution and then reimplanted with the help of plate and screws (Figure 3). The final reconstruction was confirmed under c-arm, and then, closure was done in layers.

The postoperative period was uneventful. Non-weight-bearing mobilization was done with a walker for 6 weeks. Complete weight bearing was allowed after 6 months. The patient also received 4 cycles of adjuvant chemotherapy (4 x Cisplatin + Ifosfamide), last in June 2023. The patient was followed up for 1 year with serial radiographs (Figure 4). On follow-up, contrast-enhanced magnetic resonance imaging (CE-MRI) of the left foot was done in March 2024. No differentially enhancing lesion was seen in the post-operative bed. Currently, the patient has completed the treatment, is on regular follow-up, and is doing well.

3. Discussion

Compared to the previous practice of amputation, limb salvage surgery is a safer option from an oncological standpoint for treating malignant bone tumors. Limb

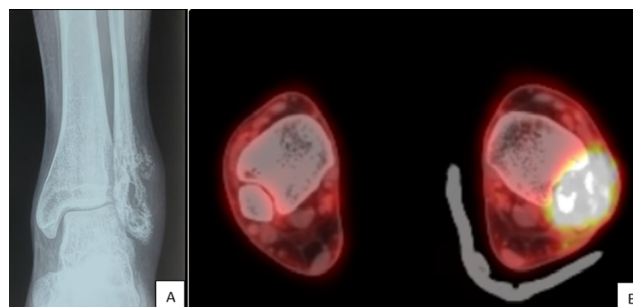


Figure 1: A): Radiograph showing Lytic lesion, B): Axial section showing FDG avid 3.2 x 2.8 x 6.3 cm osteolytic lesion with soft tissue component involving distal end of left fibula with SUV max 7.3

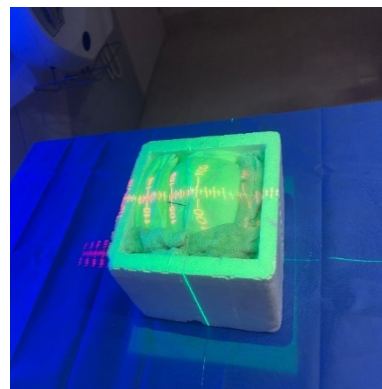


Figure 2: The linear accelerator is calibrated to irradiate the bone



Figure 3: Reconstruction was done with irradiated bone under C-arm guidance

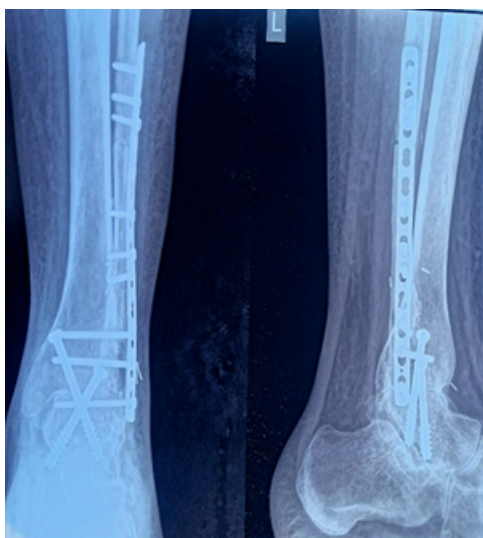


Figure 4: Follow-up radiograph showing reunion of reimplanted bone

salvage reconstruction includes the use of bone allograft, prosthesis, re-implantation of the patient's bone post-irradiation, or a combination of any of these methods. The process known as extracorporeal irradiation (ECI) involves re-implanting a patient's bone after receiving a single fraction high dose of radiation therapy.² The indications of limb salvage surgery feasible for ECI are, that the patient should have good performance status, there should be adequate uninvolved bone (bone reserve), all previous biopsy sites and all tumor tissue can be removed en bloc, the surgical margin of extensive excision (3- 5cm) is expected to be achieved, neurovascular bundle is free of tumor and adequate soft tissue coverage is possible after reconstruction.

ECI followed by reconstruction is a biological reconstruction technique that has many advantages over other methods. First, the resected bone segment is only irradiated so it avoids radiation injury to normal healthy tissues of our body. Second, a very high dose of radiation is given to bone which acts as an effective tumor cell kill technique. Thirdly, it provides a precise anatomical fit graft for reconstruction. Fourth, it reduces growth discrepancy in children, as the growth plate is preserved. Last, it is a cost-effective method as compared to prosthetic devices.³

As with any surgical procedure, ECI carries risks such as infections, delayed wound healing, nonunion, delayed union, avascular necrosis, or graft rejection. Radiation doses used for ECI range from 50Gy to 300Gy as mentioned in the literature. The most widely used dose for tumor cell kill is 50Gy, which provides a shorter treatment time and minimizes detrimental effects such as reduction in bone strength or revascularization. Radiation at such high doses is lethal to tumor cells and hence helps to prevent local recurrence. Barth et al.,⁴ and Gupta et al.,⁵⁻⁷

showed that irradiation with such high doses does not affect the viscoelastic and mechanical properties of the bone. There is also a risk that not all cancerous cells are eliminated during the irradiation process, potentially leading to recurrence. Previous studies evaluating the use of ECI for the treatment of bone tumors showed post-operative complications, ranging from 13 to 40% and local recurrence ranging from 4 to 26% respectively.^{8,9} In our patient, ECI helped to control local recurrence by acting as an effective tumor cell kill technique, there were no postoperative complications so ECI can be preferred over other methods of bone reconstruction where cost is a major concern.

4. Conclusion

By delivering high-dose radiation directly to affected bone ECI helps to minimize damage to surrounding healthy areas. It also offers significant therapeutic advantages, including enhanced tumor control and the preservation of limb functionality. Given that cost is a concern in developing nations, extracorporeal irradiation offers a practical substitute for prostheses. As technology and techniques continue to evolve, ECI may play an important role in treating cancers that are challenging to manage with conventional therapies.

5. Source of Funding

None.

6. Conflict of Interest

None.

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