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CASE REPORT

An Option for Bone Loss After Tumour Resection-Pedicle Frozen Autograft Reconstruction of the Tibia Following Ewing's Sarcoma

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ABSTRACT

Limb salvage surgery for bone tumours may vary from endoprosthesis to biological fixation. Endoprosthesis is good but expensive and durability is short. Especially in physically active individuals like our patients. Orthopaedic surgeons and scientists are still searching for substitutes for this purpose. We have performed liquid nitrogen-treated autograft as a bone filler for a patient with Ewing sarcoma of the right tibia. This treatment method has not been practised widely. This method is not expensive and reproducible. The patient recovered completely and started to weight bearing. A liquid nitrogen-treated autograft is an alternative option for patients with Ewing sarcoma. Anyway, long-term follow-up studies are mandatory to prove its efficacy compared to other treatment modalities. The procedure, outcome and complications are described in this case report.

Keywords: Ewing sarcoma, Bone loss, Liquid nitrogen, Frozen autograft, bone loss following tumour excision.



Introduction:

Ewing's sarcoma is the second most common primary malignant bone tumour in children and adolescents. It is most common among the age group of 10-20 years of age. The majority (80%) of the patients have localized disease at the diagnosis whereas the rest have metastatic disease. It has a good survival rate of 60% in localized disease and a 25% survival rate in advanced disease¹. Sandwich therapy is the management of choice. Which includes neoadjuvant chemotherapy, radiotherapy, excision of the tumour with reconstruction and adjuvant chemotherapy. There are several methods available to reconstruct the bone loss following the excision of the tumour¹. Those are by non-biological means such as tumour prosthesis or by biological fillers such as treated autografts or allografts. Biological methods are very suitable for Ewing's sarcoma as it is a diaphyseal tumour. There is no sophisticated articular reconstructions are not necessary for these tumours. We present a boy with Ewing sarcoma who

underwent reconstruction with liquid nitrogentreated autograft after neoadjuvant chemotherapy. There are a few cases that have been described in the literature. The sequence of management, follow-up and complications are described in this case report.

Case report:

A 13-year-old boy presented to us on 09.07.2020 with a complaint of right-sided tibial pain. On plain radiographs, bony destruction was observed in the diaphyseal region of the right Tibia. Non-contrast computed tomography (NCCT) was followed by Magnetic resonance imaging (MRI), Contrast-enhanced computed tomography (CECT) chest, Abdomen and Pelvis and true cut biopsy for tissue diagnosis. Ewing's sarcoma was confirmed by immunohistochemistry and metastatic disease was excluded by CECT of right tibia. The patient was referred to the oncology team for neoadjuvant chemotherapy followed by MRI to see the response.



Figure 1A shows radiographs of the right tibia (AP and lateral). The classical feature is bony destruction at the middiaphysis of the tibia. Radiographs were followed by MRI, CECT tibia and chest. Prechemotherapy MRI revealed a diaphyseal tumour without any neurovascular involvement (Figure 1B). The level of excision is determined by the prechemotherapy MRI.

Following a literature search, pedicle freezing with liquid nitrogen and autograft was planned. Surgery was performed on 25.11.2020 under general Anesthesia. Incision and osteotomy levels were planned according to the prechemotherapy MRI. Layered dissection of the soft tissues and tibial osteotomy were done. Distal-level bone drilling was done. Proximal and distal samples were sent as frozen sections for the onsite histological confirmation of clear margins. Pedicle was prepared for treatment with liquid nitrogen. A Fibula osteotomy was done. 14hole Proximal Tibial Locking plate pre-positioned and two drill holes made before the freezing. Tibia was soaked in liquid nitrogen for 20 minutes while maximum care was taken not to contact with soft tissues. Thawing is done with normal saline for 15 minutes and in distilled water.













Figure 2: Intraoperative photos during surgery and post-operative radiograph. Figure 2A shows the initial soft tissue dissection. Figures 2B, 2C & 2D demonstrate the pedicle autograft dissection and separation from the rest of the tissues. Figure 2C&2D demonstrates skin precautions to prevent skin complications.

Pedicle was fixed to the proximal tibia with a 14hole proximal tibial locking plate using predrilled holes. Layered soft tissue closure was done with a muscle flap and the skin was closed using subcuticle Vicryl and staplers.

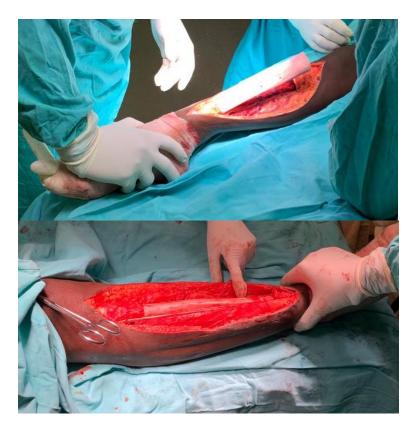






Figure 3: Intraoperative pictures and postoperative radiograph. Figure 3A shows liquid nitrogen-treated autograft. Figure 3B shows the plate fixation of the autograft and Figure 3C shows the immediate post-operative radiograph (AP & Lateral) showing perfect reduction and fixation.

The patient was followed up closely. The patient developed skin breakdown on the 13th postoperative day and was treated with negative pressure therapy. The patient was sent back to

adjuvant chemotherapy and followed up at monthly intervals with plain radiographs.

The patient started full weight bearing after 6 months of surgery with a complete range of motion.



Figure 4: shows clinical photographs of the right leg showing wound complications after the surgery. This is a complication which has been treated with negative pressure therapy.



The implant was removed one and a half years after surgery due to its prominence and pain. Again, he presented with a fracture of the autograft following the fall. The fracture was treated with a limb reconstruction system. He recovered completely and awaiting the removal of the limb reconstruction system soon.



Figure 5: Follow-up radiograph of the patient following fracture of the allograft. Figure 5 A shows the immediate radiograph after the insertion of the Limb reconstruction system. Figure 5B shows the evidence of healing at the fracture site.

Discussion:

Limb salvage and amputation carry an equal survival rate for malignant bone tumours. But, in the context of psychological impact, morbidity and disability limb salvage serves better than amputation. Limb salvage surgeries are a spectrum from endoprosthesis to biological fixation. Endoprosthesis are good and they provide immediate stability and mobility. But, they are the most constrained implants which cause early aseptic loosening, periprosthetic fractures and infections^{2,3}.

Ewing sarcoma is a diaphyseal tumour in contrast to osteosarcoma which is a metaphyseal tumour. Reconstruction of diaphyseal bone loss after excision is different from metaphyseal bone loss. During bone loss management following principles are followed 1. Neoadjuvant chemotherapy 2. Complete excision with tumour-free margin 3. Proper, stable internal fixation and 4. Early rehabilitation³.

Survival and recurrence are not depending on the method of reconstruction but, on preoperative neoadjuvant therapy and careful tumourfree excision. Especially in children and adolescents, as they are more physically demanding, endoprosthesis are not suitable for them. Liquid nitrogentreated autograft is a very good alternative to other bony reconstruction methods for Ewing sarcoma. Liquid nitrogen-treated autografts are easy to perform, lesser expensive than endoprosthesis, no bone banking services are necessary, collagen denaturation will not occur and suitable for young patients²⁻⁴. Still, there are a few drawbacks to using this as a reconstruction method. Liquid nitrogen-treated autografts are very fragile, soft tissue complications can occur during the implantation, and nonunion at the fixation site may occur.

In our patient, as his physis were open during the surgery, we selected plates and screws as the method of fixation. The intramedullary nail will be a superior option for a patient with closed physis because of increased stability. We avoid intramedullary nails on our patient as he has open physis and the nature of the fragility of the treated autograft. To avoid fracture of the graft during the fixation, we pre-drill the bone before treating it with liquid nitrogen. Our patient developed two complications during follow-up. Which are wound complications and fractures of the autograft.

There is a study carried out by Garg S and others² that has shown good outcomes following liquid nitrogen treated autografting after tumour



resection. In addition, they also stated that this method provides a perfectly matching graft for the reconstruction but, there is no complete sample for the histopathologist to comment on the histology. When it is performed bone marrows from the resected segment and the residual bony edges should be evaluated by the histopathologist. Histopathologists should be informed beforehand about the compromised tissue samples^{2,4}.

Altogether liquid nitrogen-treated tibial autograft is an alternative to other methods of bone loss management following tumour excision. Every method carries its advantages and disadvantages. Liquid nitrogen-treated tibial autograft for bone loss after Ewing sarcoma is a feasible alternative to other methods.

Conclusion

Pedicle freezing autograft is a feasible alternative to other methods of bony reconstruction. Which is lesser expensive, easy to perform, and long-lasting than endoprosthesis, with no loosening or implant-related other complications. But liquid nitrogen-treated autograft is not a foolproof method. Long-term follow-up studies are needed to find out the efficacy of this method of bony reconstruction.

Conflicts of Interest statement

All authors have declared any competing financial or personal interests which could have inappropriately influenced their work.

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