Iatrogenic giant cell tumor at bone graft harvesting site

Zile S Kundu, Vinay Gupta, Sukhbir S Sangwan, Shobit Goel, Parveen Rana

ABSTRACT

30 year old female patient with giant cell tumor of the distal tibia initially treated at a peripheral nononcological center by curettage and autologous bone grafting from the ipsilateral iliac crest reported to us with local recurrence and an implantation giant cell tumor at the graft harvesting site which required extensive surgeries at both sites. The risk of iatrogenic direct implantation of tumor, often attributable to inadequate surgical planning or poor surgical techniques, and the steps to prevent such complication is reported here.

Key words: Giant cell tumor, iatrogenic, implantation

INTRODUCTION

Giant cell tumor (GCT) are solitary lesions, but 1–2% may be metachronously multicentric.\(^1\) Pulmonary metastases occur in approximately 3% of patients due to its high vascularity and frequent treatment by curettage. The metastases are more frequent in cases of recurrent tumors and in Enneking’s stage III lesion.\(^1\)\(^-\)\(^5\) The management of GCT includes thorough clinical evaluation, imaging, biopsy, and proper surgical planning. Violation of principles of tumor surgery may lead to local recurrence and tumor dissemination/contamination.\(^1\)\(^-\)\(^6\) Although tumor contamination is a rare complication, it has been reported in literature for different types of tumors. However, review of literature revealed only two cases of isolated implantation of GCT at bone graft harvest site.\(^7\)\(^-\)\(^8\)

This case report highlights the accidental implantation of GCT from the right distal tibia to graft harvesting site of right iliac region.

CASE REPORT

A 30-year-old female reported to orthopedic outpatient department with pain and swelling just above the right ankle joint and another swelling over the right iliac region for last 6 months. The patient had a surgery for the swelling of distal right leg 18 months ago by general orthopedic surgeon. Review of surgical records revealed that she was operated for a lytic lesion in distal tibia with preliminary diagnosis of benign bone lesion without preoperative biopsy. Curettage of the lesion was performed along with filling of surgical cavity with autologous bone grafts harvested from the right iliac crest. Histopathology revealed benign GCT of distal tibia. Below knee plaster was kept for 4 months postoperatively. Two months after the removal of plaster, she developed pain in the right leg just above the ankle, along with gradually increasing swelling. Two weeks later, she noticed another swelling over the right iliac crest, associated with mild pain and discomfort. The patient was finally referred to our oncologic clinic. The patient was clinically anemic. There was a swelling over the anterolateral aspect of distal part of right leg with painful restricted ankle movements and another swelling over the right iliac region with mild to moderate tenderness on palpation on both sites.

Plain X-rays showed lytic lesion in distal tibia with ill defined margins without any evidence of pathological fracture. X-ray of right iliac wing showed large soft tissue mass with irregularity of the bone over the outer table of iliac crest [Figure 1]. Magnetic resonance imaging (MRI) of pelvis and distal tibia was done to delineate the extent of tumor and its extension [Figures 2 and 3]. Technetium-99 bone scan showed increased uptake around distal tibia and focally in the ipsilateral iliac region only at the graft harvest site and no additional foci in the body (i.e. ruling out metachronous lesions elsewhere). Core needle biopsy was performed at both sites to confirm the microscopic/tissue diagnosis. The histopathology revealed benign GCT at both sites with identical picture. Computed tomography of the lungs was normal.
The patient was taken up for planned surgery under spinal anesthesia after taking informed consent. Distal tibial lesion was widely resected and reconstruction was done with medialization of fibula and fixing it on the dome of talus and in the distal medullary canal of the resected tibia with Kirschner’s wires. Wide resection of iliac wing along with soft tissue mass was performed. The iliac swelling was confined to soft tissue underneath the scar with irregularity over iliac crest. The histopathology of both sites reconfirmed GCT.

Postoperative period was uneventful and the patient was discharged with a long leg cast for 4 months. Then, partial weight bearing was started with crutches. Patient achieved restoration of bone continuity with union at both ends. Hypertrophy of the grafted fibula was observed and patient was mobile with full unprotected weight bearing on the operated leg after 1 year. Two years after surgery, the patient was symptom free and walking without any aid and with no evidence of recurrence at both sites and no metachronous lesions or pulmonary metastasis. Fibula showed good hypertrophy and the iliac region healed well (Figure 4).

**Discussion**

Iatrogenic seeding of the tumor cells at the graft harvest site is an extremely rare, but avoidable complication. This adds on to existing morbidity and even mortality in malignant tumors. Further, there are psychological, financial, and legal implications.

The patient developed swelling in the right iliac region only after graft harvesting. The swelling on surgery was found to be confined to the soft tissues underneath the scar and the site of irregularity in the iliac crest (abutting the outer cortex from where the graft was harvested and no expansion of the inner cortex), indicating that it was iatrogenic and neither a synchronous (simultaneous multifocal primary tumors) nor a metachronous (appearance of de novo similar lesion after removal from another site) lesion. The patient developed local recurrence and also iatrogenic seedings of the tumor at the graft harvest site. Therefore, we emphasize that the adherence to basic principles of tumor surgery needs to be followed meticulously and the surgeon should take all precautions to eliminate the risk of iatrogenic implantation during the surgery.

Local recurrence after simple curettage has been reported in the literature from 10% to over 50%, which has come down to less than 20% in recent years after extended curettage and application of various adjuvants. A good extended curettage with the following principles in mind can reduce the local recurrence to a large extent. There should be a cortical window that is at least as large as the lesion. If the window is smaller than the lesion, the surgeon inevitably leaves residual tumor on the under surface of the near cortex. The emphasis is that whole of the cavity should
be well saucerized to visualize and excise the tumor. One should use sharp straight and, if required, angled curets to scoop out the tumor from all corners. High speed power burrs should be used to enlarge the cavity to break the ridges or trabeculae. Copious pulsatile (jet) lavage and use of hydrogen peroxide is useful. The use of adjuvants, such as liquid nitrogen, phenol, polymethyl methacrylate, argon beam coagulator, or thermal cautery, helps in destruction of tumor cells and further extends the limits of curettage.

Autologous bone grafting is one of the methods for reconstructing bone defects following the surgical excision of primary bone tumors. Implantation of tumor cells from the primary site of tumor to the graft donor site is rare, but has been reported in different tissues such as the skin and bones. This has been associated with malignant bony tumors like osteosarcoma or malignant fibrous histiocytoma.10-16 Cancer infections were first described by Ryall in 1907, believing that some recurrent carcinomas were due to contaminated surgical instruments with cancer cells.17 The exact mechanism of seeding of the primary tumor to graft donor site is not clear. Although direct contamination by viable tumor cells present on gloves and surgical instruments while performing definitive operation and harvesting autologous bone graft at the same sitting is most likely, however, it can be attributed to altered circulation at the healing donor site. There is some theoretical and experimental evidence that surgical trauma predisposes to localization of tumor cells. Various theories supporting this include more circulating tumor cells with manipulation of primary tumor, increased adherence of tumor cells to damaged endothelium of the microcirculation, and alteration of blood flow or coagulation mechanism in the traumatized graft harvesting site.16,18-20

The risk of iatrogenic contamination of remote surgical site can be eliminated by adhering to certain principles like: 1) All team members including the surgeon, assistants, and nurse should always change the gloves and even the gowns (in case of contamination with blood and tissue from the primary site) for harvesting the autologous bone graft, along with separate trolley of surgical instruments. 2) Always change the cautery tips and suction tubes/tips for the graft harvesting sites. 3) Graft harvest site should be kept covered with drape and only exposed when the graft is to be harvested. 4) Separate swabs and sponges should be used for the graft harvesting. 5) Donor graft site selected should not be too near to the primary surgery site, e.g. in cases of tumor of distal tibia, graft harvesting from proximal tibia should be avoided.

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