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Case report

Ewings sarcoma of ilium: Resection and reconstruction with femoral head allograft

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ABSTRACT

Ewing's sarcoma is a common malignant bone tumor seen in 5–15 years age group. It often arises from diaphysis of long bones. Ewing's sarcoma arising from the ilium is very rare, and it has an unfavourable prognosis. We present a rare case report of Ewings sarcoma of ilium with no metastasis in a two and a half year old boy, who was treated with neoadjuvant chemotherapy followed by surgical excision of the tumor and reconstruction using allograft from the femoral head fixed with multiple k-wires and screw. The patient is disease free at one year follow up and the allograft has taken the shape of growing ilium and excellent functionality and gait with minimal limp. Through this report, we emphasize on the occurrence of Ewings sarcoma in unusual site and resection and reconstruction of the tumor utilizing the allograft.

1. Introduction

Ewing's sarcoma is the second most common primary malignant bone tumor after osteosarcoma, accounting for three percent of all childhood malignancies.[1] It was first described as "an endothelioma of the bone" by James Ewing in 1921.[2] Although it can occur at any age, but is commonly seen in 5–15 years of age.

The treatment of Ewings sarcoma involving the pelvis is a great challenge in terms of local control due to the complexity of pelvic anatomy, which increases the difficulty of resection and reconstruction. The prognosis and survival of patients in this location are much less favourable than for patients with tumours of the extremities. We report a rare case of Ewings sarcoma arising from the iliac bone in a two and a half year old boy who was successfully managed by wide surgical resection and reconstruction using allograft head of femur fixed with multiple pins.

2. Case report

A two and a half year old boy presented to our institute with complains of pain in right ilium for the past one year along with limp while walking for last 6–8 months. There was no history of fever, weight loss or refusal to feed. There was no history of trauma. Past history and family history was unremarkable. Systemic examination was normal. Local examination revealed antalgic gait, tenderness at right iliac crest with fullness of iliac fossa. There was no deformity or tenderness at hip joint. The range of movements at hip joint was bilaterally comparable with no limb length discrepancy. The overlying skin was normal. Routine blood investigations were normal.

Anteroposterior radiograph of pelvis showed lytic lesion in right iliac bone with ill-defined margins with no involvement of acetabulum or sacro-iliac joint (Fig. 1). Non contrast Computed Tomography (NCCT) scan of the pelvis revealed a lytic lesion with irregular margins involving the right iliac blade violating the anterior cortex with heterogeneous soft tissue component infiltrating the iliacus muscle, with no periosteal reaction or matrix mineralisation (Fig. 2). Magnetic Resonance Imaging (MRI) showed marrow edema with expansion of the right iliac bone which was hypo intense on T1w and hyper intense on T2w images along with periosseous soft tissue on the medial aspect of the bone

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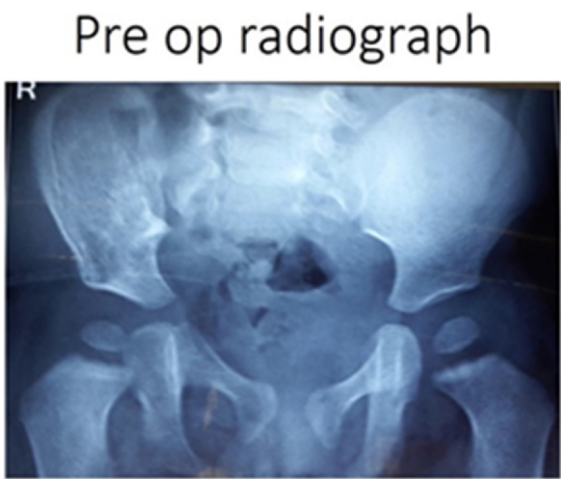

Fig 1. Pre op radiograph.

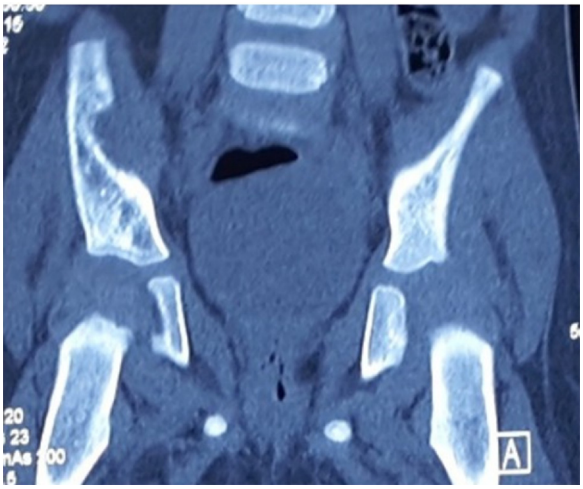

Fig. 2. Pre op CT scan.

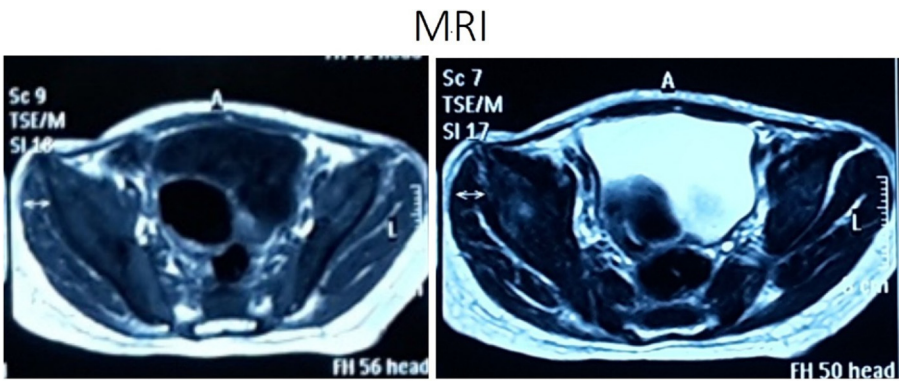

Fig. 3. Pre op MRI.

suggestive of primary malignant bone tumor (Fig. 3). Trucut biopsy was performed from the lesion which revealed diffuse sheets of atypical cells with high N: C (Nucleus: Cytoplasm) ratio and hyperchromatic nuclei showing positivity for CD99 (Cluster of differentiation), vimentin and PAS (Periodic acid-Schiff) and negative for LCA (Leucocyte common antigen) and desmin strongly suggestive of Ewings sarcoma.

Neoadjuvant Chemotherapy of 8 cycles was given followed by complete type 1 pelvic resection of the lesion was performed with 3 cm margins using ilioinguinal approach and the defect thus formed was filled with allograft head of femur and the pelvic muscles were sutured back over the allograft. Since after resection of the tumor, a large space was created making the visualization of visceral organs easier for the fixation of allograft

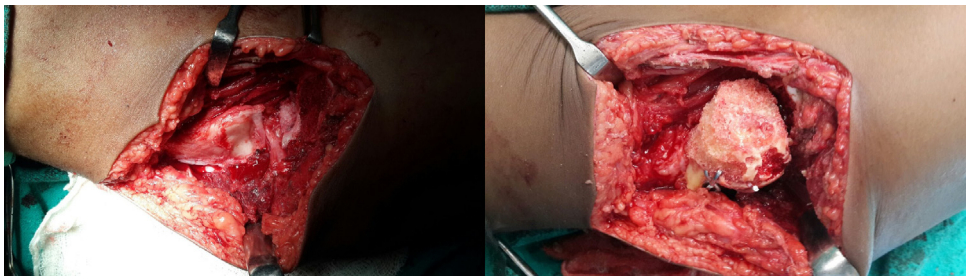

Fig. 4. Post tumor resection and allograft fixation.

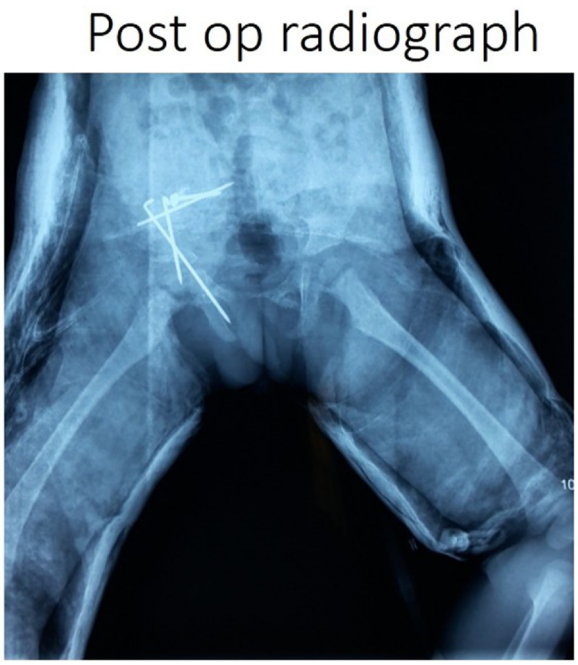

Fig. 5. Post op radiograph.

under vision with multiple k wires and screws (Fig. 4). Post-operative immobilization was done in hip spica for a period of 2 months. At 9 months follow up, there is no evidence of metastasis and the allograft has been well incorporated with the parent remnant bone, also at the same time deriving the shape of the original ilium (Fig. 5). The hardware was removed at 10 months post op and weight bearing was started. Post op X ray at 1.5 years follow up suggested well incorporated allograft provided excellent structural support (Fig. 6).

3. Discussion

Ewings sarcoma of pelvis has poor prognosis because of lack of major anatomic barrier to tumor spread in pelvis and proximity to

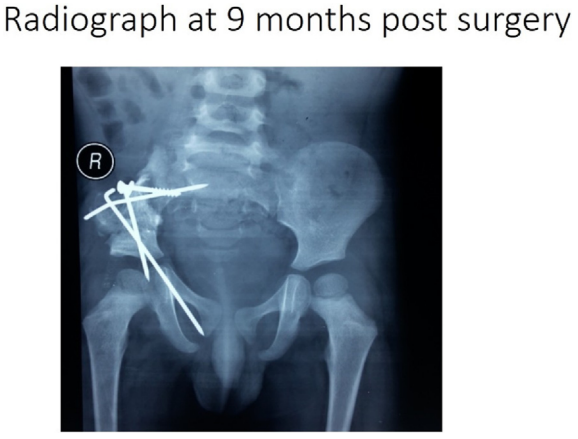

Fig. 6. Radiograph at 9 months follow up.

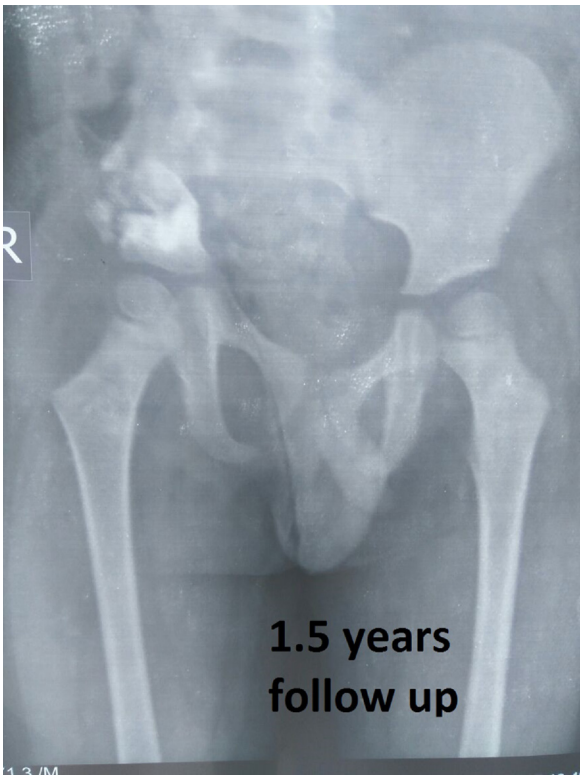

Fig. 7. Radiograph at 1.5 years follow up.

visceral organs and neurovascular bundles making local control difficult.[3,4]

With the introduction of effective chemotherapy, imaging modalities and modern surgical techniques limb salvage procedures has replaced amputation for radical treatment of malignant pelvic bone tumours.[5,6]. Though in cases where limb salvage is not possible, conservative hemipelvectomy remains a modality of treatment. Type 1 resection usually creates a bony pelvic ring defect leading to pelvic instability. Since resection without pelvic ring reconstruction has not been shown to be effective because of distortion and instability of the pelvic ring, likely resulting in poor long-term function [13], several reconstruction options to maintain the stability of the pelvis and allow ambulation have been reported. After resection of pelvic bone tumours, reconstructive options reported to date have included allografts,[7,8] autografts,[9] vascularized iliac autografts,[10] non-vascularised fibular grafts,[11] or vascularised fibular grafts.[12]

Although reconstruction using allografts has been one of the most popular options, it was reported to be associated with a high rate of complications including fracture, non-union, and infection. With an experience of around two decades of use of allografts in various bony defects, encouraging results were available in terms of higher rate of union and lesser infection rates. Beadel et al[13] reported 4 cases of pelvic ring reconstruction using a fibula allograft, and only 2 patients (50%) had long lasting bone union. Nishida et al[10] first reported 5 cases of pelvic reconstruction using a vascularized iliac autograft fixed with a pedicle screw and rod. In their series, bone union was achieved in all patients within a mean period of 5.4 months. Postoperative complication was seen in 1 patient (20%), who developed skin necrosis resulting from pedicle screw protrusion.

The major drawbacks to allografts are non-union and fracture. The major advantage of structural allografts is the absence of donor site morbidity and they are a biologic solution which may last the lifetime of the patient if they heal and do not fracture. Osteoarticular allografts include the joint surface at the end of the donor bone and may be used in reconstructions that require the removal of a joint. Any joint can be reconstructed in this manner although the longevity of the reconstruction may vary among different locations (Fig. 7).

4. Conclusion

We report a case of Ewings sarcoma in a two and a half year old boy arising from the right iliac bone which was managed by wide surgical excision (type 1 resection) and reconstruction using allograft which excellent graft incorporation at the follow up as well as providing a good structural support helping in the functionality of the growing child and gait.

This case emphasizes that the functional outcomes following tumor resection can be improved with use of structural allograft decreasing the donor site morbidities as associated with autografts.

Conflict of interest

None

The manuscript has been read and approved by all the authors in the study.

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