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Editorial
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"Shephard's Crook Deformity Pickled with Autologous Rib Graft: Our Solution to Limited Graft Options, A Case Report"

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Introduction

Fibrous dysplasia is a benign intramedullary fibro-osseous lesion originally described by Lichtenstein and Jaffe in 1942 and accounts for 5% to 7% of benign bone lesions. [1] This can be of Monostotic (one bone) or Polyostotic (more bones) and can be associated with abnormalities like McCune Albright Syndrome (Cutaneous pigmentation and Endocrine abnormalities), Mazabraud syndrome (Intramuscular myxoma) [2].

Most common sites of occurrence are proximal femur, tibia, humerus and ribs. It is speculated to be a developmental failure in remodeling of woven bone to mature lamellar bone and failure of the bone realign in response to mechanical stress. Pain, limp, deformity, and pathological fractures are the main presenting symptoms [3].

We represent a case of 13 years old boy who has presented with pathological subtrochanteric fracture with almost complete resolution in 2 years period with fixed angle device fixation and autologous bone graft harvested from the ribs with no doner site morbidity. We interpret this as a panacea for the limited graft option in the young age group.

The patients along with his guardian were informed that data concerning the case would be submitted for publication, and they provided consent.

Case Report

A 13 years old obese boy had presented to the emergency room with painful swelling of right thigh following a trivial fall while playing with subsequent inability to bear weight. There was no significant past medical history of any illness with child was active and playful till date of injury. Clinically he had swelling, tenderness at the right hip joint with painfully restriction of movements with normal distal neurovascular status. Radiological investigation showed a coxa vara with well-defined lytic lesion in the inter trochanteric/sub trochanteric region of right femur with zone of transition and a sclerotic margin and a patchy ground glass matrix with pathological fracture. In view of lytic lesion MRI was done which showed a well circumscribed intramedullary expansile lobulated altered signal intensity lesion measuring 4.5 X 4.3 X 9.6 cm (AP X TR X CC), extending from the intertrochanteric region with no abnormal periosteal reaction. Management involved open curettage and biopsy along with Internal Fixation with Proximal femoral locking plate and bone grafting. The lesion was approached laterally and was curetted out. Non hemorrhagic, greyish white rubbery material was sent for histopathology. Subsequently, the bone defect created during curettage was filled with auto graft harvested from 6th and 7th ribs from ipsilateral side, morselized and bone graft substitutes. Post-operative period uneventful histopathology of the extracted materials revealed chronic inflammatory cells predominantly lymphocytes, plasma cells and macrophages without osteocytes and osteoblastic rimming and fibro-osseous lesion of bone consistent with fibrous dysplasia that confirms the diagnosis.

Patient was kept non-weight bearing and gradually weight bearing started once satisfactory radiological evidence of healing was present. Follow up was done at 6, 12 weeks, 6 months, 1 year and 2 years. At 2 years, the radiographs showed complete consolidation of the cavity with no evidence of failure of implant or recurrence. There was complete functional and clinico- radiological recovery at 2 years follow-up.

Discussion

Fibrous dysplasia is very rare nonmalignant form with common sites over the proximal femur, tibia, humerus and ribs region. Pathological fracture may leads to the accidental diagnosis of Fibrous dysplasia [5]. Curettage and the filling up of individual foci have gained importance in the case of the monostic form.

Owing to the fracture that has already occurred, correction with fixed angled device for stability is treatment of choice in skeletally immature age group [6]. Extended curettage and filling with bone graft is challenging due to its large size and less availability of graft option in young age group. Various literature has coated either as autologous fibular graft or allogenic femoral head graft [7]. Allograft has its own complication in terms of infection and immunogenicity.

Best donor materials are one's own bone or autologous bone grafts but catering adequate amount for such a large hollow space in a child is in question. In our case, we thought of harvesting the 6th and 7th ribs graft from the ipsilateral side to fill the cavity as well as adjunct benefit of mechanical support due to its consistent with a cortical hard component and a softer cancellous portion.

Patient was kept on strict non weight bearing till radiological sign of healing with gradually weight bearing. Follow up done at 6, 12 weeks, 6 months, 1 year and 2 years which shows well healing with complete resolution at both fracture and doner site. Patient was active and back to outdoor playing activities.

We report this case, to highlight the ribs graft can be the source of bone graft in skeletally immature age group for such an area with good recovery at both the doner and recipient site. Autograft is our own bone, so no question of rejection! It just leaves a small scar which is

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unremarkable, barely seen once done. The body's acceptance is good as compared to any other bone graft substitute. Due to its dimension and large volume of harvest, ribs graft is highly used in rhinoplasty surgeries [8]. The sustainability, biocompatibility, safety which a rib graft gives are much better. So, we think this could be solution in young age group where harvesting bone graft could be dilemmatic subject. Further research into the subject matter into the morbidity of both doner and recipient site is suggested.

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