BIZARRE PAROSTEAL OSTEOCHONDRAL PROLIFERATION OF METATARSAL IS RECONSTRUCTED BY FREE TRICORTICAL ILIAC BONE GRAFT: A CASE REPORT

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ABSTRACT: INTRODUCTION: Bizarre parosteal osteochondral proliferation (BPOP) is a rare variety tumour like bone lesion usually of small bones of hands and feet which has a strong tendency for recurrence. Clinico-radiologically solitary exostosis like lesion in those areas which continue growing after skeletal maturity without any pre-existing lesion one should think of BPOP. **CASE REPORT:** 46 years male patient presented in 2009 with progressive painless, hard, non-tender swelling arising from distal half of the left second metatarsal bone of one year duration. X ray showed multi-horned heterogeneously radio-opaque swelling attached to 2nd metatarsal where one of the horns was pressing over 3rd metatarsal making its divergent appearance. Open biopsy showed lamellar bone with inter-trabecular spindle cells without any evidence of malignancy. Irregular shaped cartilage cells of different size in bluish tint cartilage are found in the bone cartilage interface. Through dorsal incision wide margin resection of the second metatarsal was done and reconstructed with autologus tricortical bone graft from ipsilateral iliac crest. Further histopathological examination demonstrated similar picture and absence of periosteum in addition. Significant cartilaginous cap could not be identified. Features simulate BPOP. It took 6 months - time for incorporation of graft. Movements of foot joints regained except 50% restriction of targeted metatarsophalangeal joint. There is no recurrence in 4 years follow up. **CONCLUSION:** Exostosis like lesions when arising from unusual site and at unusual age group other rare conditions is to be thought of. As they have tendency for recurrence adequate resection is essential. Though the final diagnosis of BPOP is obtained after histo-pathological examination, the clinico-radiological findings are also important for treatment planning.

KEYWORDS: Solitary Exostosis; Metatarsal; Bizarre parosteal osteochondral proliferation; Tricortical iliac bone graft.

INTRODUCTION: Exotosis is a common benign lesion in metaphyseal region of long bones. Infrequently it develops from short and flat bones.[1] Very rarely it arises from sesamoid bone like patella.[2,3,4] Usually it stops growing with skeletal maturity of the individual unless complicated with malignancy But recently mutation of tumour gene is also correlated with solitary exostosis and the traditional theory of 'skeletal dysplasia' is shifting towards the theory of 'cell-of-origin'.[5,6] Incidences of solitary exostosis of tarsus and metatarsal (MT) are rare.[7,8] When such a lesion starts growing after 3rd decade and continues grow without any preexisting lesion, other rare

pathologies like bizarre parosteal osteochondral proliferation (BPOP), Turret exostosis etc. are to be thought of BPOP has a strong tendency to recur after in adequate excision.

This case is being reported because of its rarity and continued growth even after skeletal maturity. Differential diagnosis, surgical technique of resection and reconstruction are also discussed.

CASE REPORT: 46 years male patient presents with progressive swelling over left sole for 12 month duration in 2009 with deviation of the third toes outwards. It was associated with pain during walking in later days. Hard, non-tender, irregular surface swelling was attached to 2nd MT but free from overlying soft tissue structure. He was normotensive, type II diabetic and mildly anaemic.

X rays showed 5 cm multi-horned bony mass arises from metaphyseal region of 2nd MT of left foot. One of the horns was pressing over 3rd rays causing their divergent position and other one was projected planter wards. [Figure 1] Continuity of medullary canal of MT is not found in the lesion.

Biopsy was taken which showed matured osseous tissue with thin cartilage cover. Resection was done through dorsal longitudinal incision along the second ray under tourniquet application. Second MT was resected through the junction of proximal and middle third of shaft to the neck of it [Figure 2]. Autogenous tricortical free bone grafts were harvested from left iliac crest. Length of the graft was matched with the resected length of MT and fixed there using one longitudinal and two crossed Kirschner wires. Free cancellous bone grafts were then put at the junctions. Wound was then closed without drain. Excised tissue was sent for further study. Well-padded dressing and below knee posterior plaster of Paris (POP) slab was applied. Dressing was changed and stitches were removed after two weeks when a below knee POP encircling strong cast was applied in plantigrade position for two months. Partial weight bearing was permitted during the last month of cast. After two months, post operatively, crossed K-wires were removed [Figure 3]. After another 15 days longitudinal K-wire is removed. Patient were followed up at two week, 6 weeks, 10 weeks, thereafter every 3 months for one year and yearly till date. At three month enough evidence of union was noticed and at 6 month incorporation was well appreciated radiologically [Figure 4]. There is no evidence of recurrence as yet.

Histopathological (HP) examination of excised tissue showed matured lamellar bone with inter-trabecular spindle cells. Bluish tint cartilage tissue containing irregularly arranged cartilage cells of different size are seen in the bone cartilage interface. It was devoid of periosteum [Figure 5].

Ankle and interphalangeal movements were regained at 3 and 6 months but metatrsophalangeal motion was restricted by 50% at final follow up without any instability [Figure 6].

DISCUSSION: The diagnosis in this case was based on clinical, radiological and supported by histopathology.[2] Possibilities in this case are non-hereditary solitary exostosis,[1] Bizarrie parosteal osteochondral proliferation (BPOP),[9 to 13] dysplasia epiphysialis hemimelica (DEH)[3,7] Turret exostosis[8] and florid reactive periosteitis.[14] Focal lesions of systemic disease like diffuse

idiopathic skeletal hyperostosis (DISH)[15] also known as Forestier's disease and heterotrophic ossificans atraumatica are excluded in relation to this case.[16]

Non hereditary solitary exostosis usually presents within 3rd decade.[1] They are directed towards diaphysis; may have medullary canal in continuity with the parent bone.[1] Most recently correlation with EXT gene mutation has been established.[5,6] Metaphyseal osteochondroma are believed to stop growing with skeletal maturity. But recently mutation of tumour gene is also correlated with solitary exostosis and the traditional theory of 'skeletal dysplasia' is shifting towards the theory of 'cell-of-origin'.[5,6] This might be the one explanation of continued growth after skeletal maturity. HP study distinctly demonstrates up to 2 cm thick cartilage cap covered with perichondrium.[17] In this case HP study differs from solitary exostosis.

BPOP, also known as Nora's lesion, is proliferation of bone and usually emanates from intact cortical surface of short bones and rarely in long bones even.[9] It is assigned to be reactive heterotrophic ossification. Though generally correlated with trauma, recently it is denied and genetic alteration supposing tumour genesis is recently suggested.[10] Usual age group of the sufferer is in 3rd or 4th decades.[12] In early stage cortical continuity may not be there but ultimately develops it. No sex dominance reported in literature.[12] Radiology in most occasions resembles exostosis. BPOP apparently arises from the periosteal tissues through a process of cartilaginous metaplasia. Microscopically, it is composed of hypercellular cartilage with calcification and ossification and of basophilic tinetorial character.[13] Cancellous bone undergoes maturation and presence of spindle cells may create confusion with parosteal osteosarcoma but in this case they are without cytologic atypia.[11] There is thinning of cartilage layer in long standing cases. These features are similar to the present case. 50% local recurrence is reported in literature.[10] This patient did not have recurrence in long term follow up.

DEH popularly known as Trevor-Fairbank disease is an epiphyseal developmental disturbance of skeleton resulting in exostosis–like, tumour-simulating lesion in childhood with male preponderance. Common sites are epiphyses of long bones of lower extremity and tarsus of one side of body.[7] Localized, classic and generalized forms are described. Commonly arises from medial secondary centre of ossification. Involvement of sesamoid bone like patella is also reported in literature.[2,7] Basic pathology what microscopically seen is abnormal cartilage proliferation and enchondral ossification. Important differentiating features in DEH are presence of cartilaginous bands separating areas of cancellous bone. Tumour gene is not yet demonstrated.[2] Age of the patient, arising from metaphyseal region instead of epiphysis without parent bone expansion and microscopic features are not correlating with the present case.

Turret exostosis is a benign osteo-cartilagenous lesion believed to arise from reactive periosteum following relatively mild trauma. Microscopically, central area of mature bone from endochondral ossification with thin hypocellular peripheral rim of cartilage and absence of periosteum.[8]

Turret exostosis and florid reactive periostitis may represent different stages in the development of a posttraumatic proliferative process.[8] No history of trauma can be obtained in this case.

Wide margin resection was decided as it was thought to be BPOP which has a strong tendency for recurrence.[10] Options for reconstruction are allograft,[18] free fibular graft,[19]

vascularized osteomyocutenous fibular graft,[20] callus distraction[21] and different types of bone substitutes.[22] Recently applicability of different barrier membranes is discussed in literature.[23] Mostly they are in experimental stage with limited clinical applications. No clinical data could be accessed for metatarsal reconstruction using barrier membrane principle. Tricortical iliac autogenous bone graft was considered in the present case because union and incorporation is much faster than cortical bone like shaft of fibula and same donour site can provide adequate amount of free cancellous bone graft also. This technique is much easy than vascullariesd graft. In this case union was well appreciated at two and half months and incorporation at 6 months. Dorsal approach is considered to avoid damage to neurovascular structures. Transarticular longitudinal K-wire caused some initial stiffness of interphalangeal joints which subsequently passed off, but residual metatarsophalangeal joint stiffness is probably because of that. Pin track infection is a common complication. In this patient we did not have it.

CONCLUSION: Careful clinic-radiological examinations give the diagnosis of atypical variety of exostosis like lesion arising from a very rare site like MT. Wide margin resection can minimize the chance of recurrence particularly when it is thought of BPOP or suspected malignant lesion. Cortico-cancellous graft is preferred to cortical graft in MT reconstruction.

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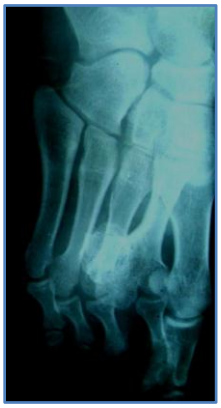
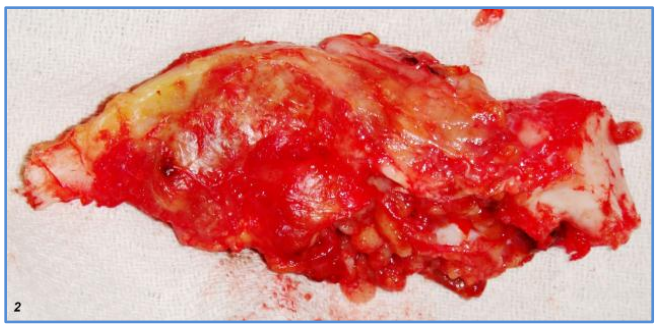
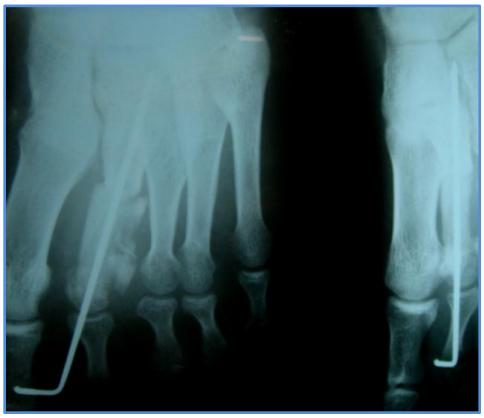


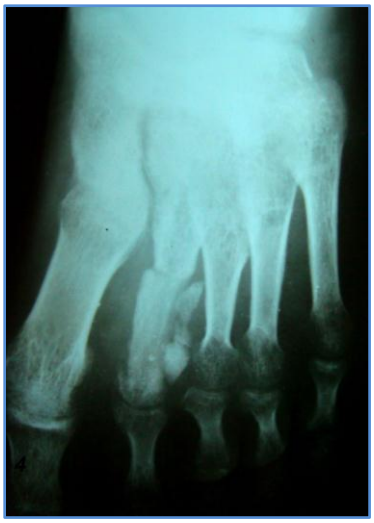
Pre-operative X-ray lesion arising from 2nd MT. One of the horn pressing over 3rd MT and other one projecting planter wards



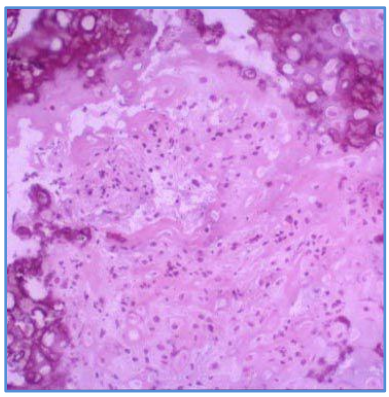
Per-operative picture showing the excised MT with the lesion



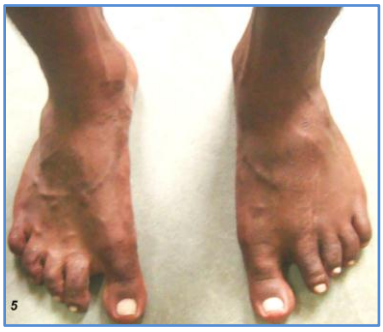
Post-operative X-ray picture at 2 months with evidence of union when crossed K-wires are removed



X-ray at six month with clear signs of incorporation of corticocancellous graft



Microscopic picture showing irregularly arranged cartilage cells of different size with bluish tint cartilage formation, normal stromal spindle cells in the inter-trabecular space and bone



Follow up clinical picture at 6th month

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