A Giant Solitary Exostosis of the Lower Ulna in a Child

* Haque MA¹, Haque ME², Islam MS³, Chowdhury MR⁴

ABSTRACT:

Forearm deformity secondary to giant solitary ulnar exostosis is rare. Here we described a rare presentation of symptomatic solitary giant exostosis involving the distal ulna resulting in ulnar bowing of the forearm in a fiveyears old girl. The tumour was completely resected and the defect was reconstructed with non vascularised fibular autograft. A wedge osteotomy was performed on the radius to correct the deformity. Nine months after surgery, the fibular autograft has fully incorporated and the deformity remains corrected with normal bone length and excellent hand function. There is no evidence of local recurrence.

CBMJ 2012 Jan:Vol 01 No 01 P 30-32

Key words: Solitary, Exostosis, Resected, Nonvascularised, Fibular autograft.

Introduction

Exostosis (Osteochondroma) is a cartilage capped bony projection arising on the external surface of bone containing a marrow cavity that is continuous with that of the underlying bone¹. Exostosis is the most common benign bone tumour, which occurs as sporadic (solitary) or multiple, usually in the context of the hereditary syndrome, multiple exostosis^{2,3}. The solitary (sporadic) form of exostosis is approximately six times more common than the multiple exostosis. It is not considered a true neoplasm, rather it is a hamartoma produced by the growth of subperiosteal aberrant foci of cartilage. It is asymptomatic in the majority of people and is typically incidentally discovered around the age of puberty. Here we described a rare presentation of symptomatic solitary giant exostosis of the the distal ulna resulting forearm deformity in a five-years old child with its management and outcome.

Case report

A five years old girl is presented to us with progressive swelling of the left forearm and deformity developing over a period of three years. The swelling was first noticed at the age of two and was asymptomatic. The family noticed that it was growing rapidly for the past one year and the patient consistently complained of tightness with mild pain. Physical examination revealed a bony hard mass extending from the mid-shaft to the distal of the ulna, associated with ulnar bowing of the forearm. Clinical examination revealed no evidence of neuro-vascular compromise in the hand.

Plain radiograph showed a fusiform bony lesion at the meta-diaphyseal region of the left ulna. The distal ulna physis was not clearly visible. Chronic compression by the lesion caused thinning of the radial cortex and ulnar deviation of the radius.



Fig: Pre operative photograph

Fig: Pre-operative x-ray of forearm

- *Dr. Md. Anwarul Haque, Associate Professor, Department of Orthopaedic Surgery, Community Based Medical College Hospital, Bangladesh.
- 2. Dr. Md. Emdadul Haque, Medical Officer, UHC, Trishal, Mymensingh
- Dr. Md. Saiful Islam, Assistant Professor, Department of Orthopaedic Surgery, Community Based Medical College Hospital, Bangladesh.
- Dr. Mamunur Rashid Chowdhury, Assistant Professor, Department of Orthopaedic Surgery, Community Based Medical College Hospital, Bangladesh.

* Address of correspondence E-mail: anwarul_dr@yahoo.com Mobile: 0088 01711 619314 We performed a resection of the entire lesion of the ulna by dorsal approach under general anaesthesia. Intra-operatively, the ulnar neurovascular structure was preserved. The distal ulnar physeal region was involved and can not preserved . The long osseous defect was reconstructed with non vascularised fibular autograft and fixed by intamedullary wide K-wire. The radial deformity was corrected by closed wedge osteotomy and fixed by intramedullary K wire also. Post-operatively histopathology of the resected specimen was done and diagnosed as osteochondroma

Post-operatively the forearm was protected in a fulllength cast for 8 weeks. Nine months after surgery the patient was asymptomatic after removal of K-wire. The deformity was corrected and wrist and hand function was good, with 80°wrist palmar flexion and 30° dorsiflexion and the hand grip was full with power of MRC grade 4/5. Radial and ulna deviation were 30°, however forearm supination and pronation mildly restricted. Later on the girl was advised to use short arm ulnar side leather brace intermittently.







X- ray 1 month after operation

X-ray 3 months after operation







X-ray after removal of K- wire 9 months later Photograph after 9 months of operation

Discussion

Deformity of the forearm is commonly observed in multiple hereditary exostoses, and is due to defective metaphyseal remodelling and asymmetrical retardation of longitudinal bone growth^{4,5}. The most common deformity is a combination of relative shortening of the ulna, bowing of one or both forearm bones, ulnar deviation of the hand. progressive translocation towards the ulna of the carpus and dislocation of the radial head^{6,7}. A solitary isolated giant fusiform forearm exostosis presenting in early childhood is rare. The progressive enlargement of the lesion and local compressive effect to adjacent bone lead to deformity, thus complicating surgical management. Furthermore, the large swelling caused chronic compressive neuropathy and interfered with gliding motion of the tendons resulting in poor hand function. In the past, the operative management had to be delayed until puberty because of the unfavourable results of early intervention. In recent years, early operative intervention was performed to prevent or reduce the progression of deformity, hence the unacceptable frequency of residual functional impairment, particularly with regard to radial head dislocation^{4,5}. Partial resection of the fusiform exostosis is reported to have a high recurrence rate and may necessitate repeated surgical interventions. Moreover, the progression of deformity cannot be predicted. On the other hand, resection of the entire lesion leads to a massive osseous defect and

subsequent disturbed growth and function. To overcome this problem, we describe a procedure involving total resection of the lesion and reconstruction of the resultant defect with fibular autograft. Reconstruction of a long segment osseous defect by a strut graft of autogenous bone is a standard practice in adults, but in paediatric donor sites, morbidity poses a major problem with the possibility of permanent deformity and disability. Allograft is therefore a good alternative; however, bony incorporation could be delayed and there is subsequent risk of fracture and dissolution of the graft. Some author used allograft wrapped with the autogenous periosteal flap to deliver fresh pluripotential cell for better incorporation and integration^{6,7}. Allograft healing and strength can be achieved relatively early thereby reducing the risk of allograft complications^{6,7}. In this case, radiological evidence of bony incorporation and union was seen at three months.

Conclusion:

The girl achieved satisfactory bone union and incorporation of fibular autograft after 9 months. The girl was able good wrist movement with grip of full power. But only problem with gradual shortening of ulna may occur later due to loss of distal ulnar epiphysis during operation. The patient probably will be scheduled for bone lengthening later

References

- Khurana J, Abdul-Karim F, Bovée JVMG: Osteochondroma. In World Health Organization classification of tumours. Pathology and genetics of tumours of soft tissue and bone.. Edited by Fletcher CDM, Unni KK and Mertens F. Lyon, IARC Press; 2002:234-236.
- Mulder JD, Schütte HE, Kroon HM, Taconis WK: Radiologic Atlas of Bone Tumors. 2nd edition. Elsevier, Amsterdam; 1993.
- 3. Bovee JVMG, Hogendoorn PCW: Multiple osteochondromas. In World Health Organization Classification of Tumours. Pathology and Genetics of Tumours of Soft Tissue and Bone. Edited by Fletcher CDM, Unni KK, Mertens F. IARC Press, Lyon; 2002.
- Ip D, Li YH, Chow W, Leong JC. Reconstruction of forearm deformity in multiple cartilagenous exostoses. J Pediatr Orthop 2003; 12(1):17-21.
- Peterson HA. Deformity and problem of the forearm in children with multiple hereditary osteochondromata. J Pediatr Orthop 1994; 13: 92-100.
- Karaoglu S, Baktir A, Kabak S, Arasi H. Experimental repair of segmental bone defect in rabbit by demineralised allograft covered by free autogenous periosteum. Injury 2002; 33(8): 679-83.
- Neel M. The use of a periosteal membrane for bone graft containment at allograft host junction after tumour resection and reconstruction with bulk allograft. Orthopedics 2003; 26(5): 587-9.