Acta Orthopaedica et Traumatologica Turcica xxx (2017) 1–4

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

Aneurysmal bone cyst healing response with intramedullary lengthening nail

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ARTICLE INFO

Article history:
Received 17 October 2016
Accepted 27 April 2017
Available online xxx

Keywords:
Humeral lengthening
Humerus
Aneurysmal bone cyst
Lengthening nail

ABSTRACT

We report the treatment process of a pediatric patient with deformity and shortening in the arm after a recurrent aggressive aneurysmal bone cyst (ABC) in the proximal humerus. The patient was treated with curettage of the lesion and lengthening on an intramedullary nail following an osteotomy just distal to the ABC. The period of lengthening was approximately 50 days. At the end of the treatment the lengthening goal was achieved without any neurovascular complication. There was a minimal loss in shoulder hyperabduction due to the deformity of the humeral head.

Introduction

Aneurysmal bone cysts (ABCs) are rare benign expansile osteolytic bone tumors which occur throughout pediatric and adult bones. The most frequently affected locations are the metaphysis of the long bones.[1] They account for 1% of all biopsied primary bone tumors.[2] Histologically, they contain blood-filled spaces separated by the septae containing osteoid tissues and osteoclastic giant cells. ABCs most frequently affect people younger than 20 years old.[3] Although benign, can be locally destructive to normal bone and may extend to soft tissue. Their expanding nature can erode cancellous and cortical bones, resulting in growth cessation by disrupting growth plates.[4]

There have been few clinical reports in literature about analyses of pediatric shoulder girdle ABCs. According to those studies, the clinical behavior of ABC in younger patients can sometimes be more aggressive than in older children.[5] In the proximal humerus, it is important to determine whether or not the growth plate is affected. Angular deformity and/or shortening can occur due to growth plate destruction or tumor resection. Therefore, preservation of normal growth and shoulder function should be the aim of the treatment, but it may not always be possible. To achieve this, the curative resection of the tumor should be combined with a lengthening procedure for the restoration of the function of the upper extremity.

Surgical lengthening of the humerus has been performed less frequently than the lengthening of the femur and tibia, and there are fewer reports in literature of humeral lengthening compared with those of lengthening of the femur or tibia. There are noticeable limitations in function or cosmetic appearance if, and only if there is a moderate to large arm length discrepancy. Management options for severe arm length discrepancy vary and include no treatment, surgical lengthening, and epiphysiodesis of the contralateral normal proximal humeral epiphysis. When considering surgical lengthening, distraction osteogenesis arose from Ilizarov's experimental and clinical work in the 1950s. Gradual mechanical distraction can be used for humeral lengthening using either monolateral or circular external fixators.[6,7] As a recent innovation in limb lengthening, magnetic intramedullary nails are becoming a promising option of treatment.[8]

The case presented here is of a pediatric patient with a deformity and a shortening of the arm due to a recurrent aggressive ABC located in the proximal humerus, with the aim of reporting the healing response of ABC with lengthening intramedullary nail.

Case report

Five years ago a 9-year old boy was admitted to the outpatient clinic with complaints of pain, deformity and shortening of the left arm. Physical examination revealed an expansile tumor at the

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Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

http://dx.doi.org/10.1016/j.aott.2017.04.005

proximal part of the left arm and minimal (1.5 cm) shortening of the left upper extremity compared to the opposite side. An expansile, lobulated lytic lesion in the proximal metaphysis of the humerus was detected on radiographic evaluation (Fig. 1A). He had been initially treated by curettage and cancellous bone allograft in our center (Fig. 1B). The histopathological results of the tissue samples had been found to be consistent with the ABC. Serial follow-up radiographs showed recurrence of the ABC with concurrent progression of the deformity and limb-length discrepancy of the proximal humerus after 4 years of follow-up.

During examination at the age of 13, a short left arm secondary to an ABC recurrence on the proximal metaphysodiaphyseal of the humerus was noted (Fig. 2). He reported no pain but preferred to wear only long-sleeved shirts. Physical examination showed minimal limitation of movement, although clinically and radiologically, the left humerus was 5 cm shorter than the contralateral side. On radiographic evaluation, the growth plate was closed. The management option was surgical lengthening of the humerus by distraction osteogenesis via magnetic intramedullary nail.

A percutaneous osteotomy was applied in the diaphysial region of the humerus just distal to the ABC and curettage of the ABC zone (without grafting) just proximal to the osteotomy site. According to the preoperative planning, the insertion of the nail was prepared by conditioning the medullary canal with long, straight millers in 0.5 mm steps under careful radiological control in two planes. Then, an 8.5 mm × 195 mm magnetic antegrade tibial lengthening nail (PRECICE® Intramedullary Limb Lengthening System, Ellipse Technologies, USA) with a 5 cm lengthening capacity was implanted. Proximal locking was performed with a drill guide, and the distal locking with the free hand technique with a radiolucent gear drive using a mini open approach to protect the radial nerve. The nail function was tested radiologically with the c-arm, by lengthening 0.5 mm under sterile conditions immediately after implantation.

Distraction was initiated on post-operative day 5 with a 0.25-mm distraction rate every 6 h and was checked radiologically at least twice prior to the patient's discharge. After discharge, clinical examination and radiological control were performed weekly in an outpatient clinic. The planned weekly lengthening goal and the radiological lengthening were compared in order to control the lengthening process (Fig. 3). The distraction rate was adapted according to the radiological signs of consolidation of the bone regeneration, and the conditions of the joints and neuro-vascular structures.

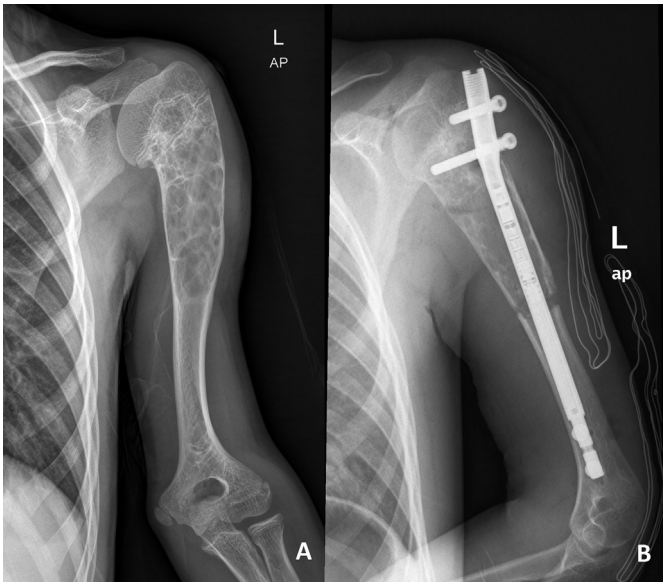

Fig. 2. A. Preop ABC recurrence B. Postoperative radiography.

The period of lengthening was approximately 50 days. In the first 3 weeks of distraction, the patient used an arm sling and during this period of time, passive and active assisted exercises were allowed. After removing the sling, the patient was not allowed to lift heavy weights but was advised to undertake daily activities with a protective attitude. After the completion of the planned 5 cm lengthening, consolidation was monitored by radiographic follow-up every 4 weeks (Fig. 4). Four months after the operation, the weight limitation was abolished after all 4 cortices were seen on the radiographs. At the end of the total process, the targeted lengthening and the treatment of the aneurysmal bone cyst was accomplished.

After the lengthening, there were no neurovascular sequelae or loss in elbow range of motion, although there was a minimal loss in the shoulder hyperabduction due to the deformations on the humeral head (Fig. 5).

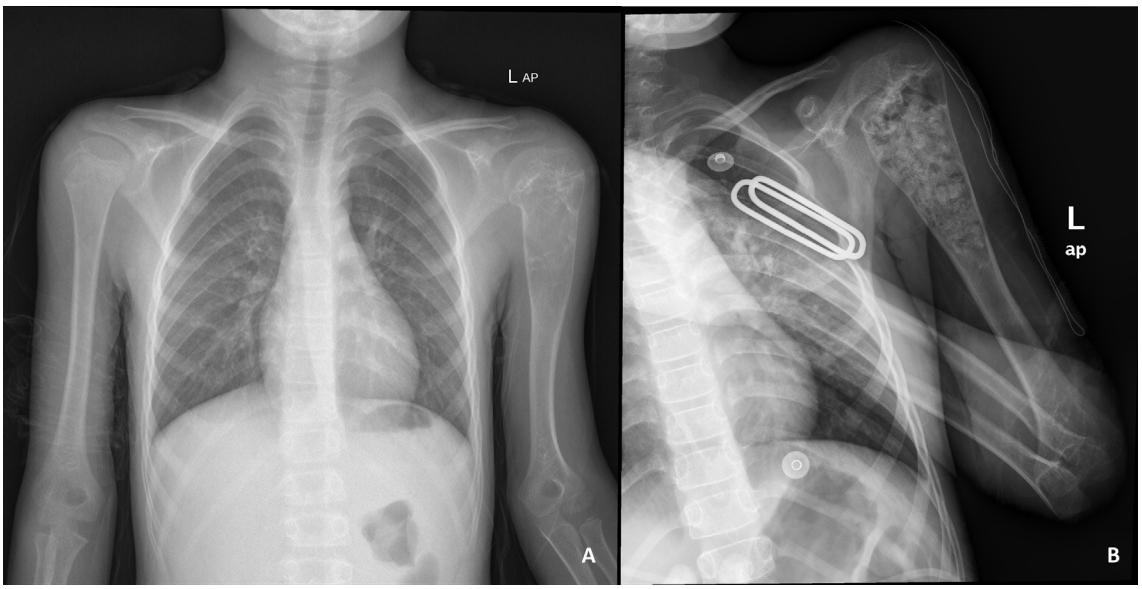

Fig. 1. A. Radiography on admission B. After curettage and grafting.

Please cite this article in press as: Acan AE, et al., Aneurysmal bone cyst healing response with intramedullary lengthening nail, Acta Orthop Traumatol Turc (2017), http://dx.doi.org/10.1016/j.aott.2017.04.005