



A giant multi-lobed osteochondroma of the phalanx in an adult: A case report[☆]

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ABSTRACT

INTRODUCTION: Solitary osteochondromas of the adult hand are extremely rare. We present a case of a giant multi-lobed osteochondroma of the phalanx in an adult. No similar cases were found in the literature.

PRESENTATION OF CASE: A 25-year old male presented with a giant multi-lobed osteochondroma arising from the base of the middle phalanx; causing limitations of motion of the proximal interphalangeal joint (PIPJ). The patient refused complete excision, bone grafting and possible fusion of the PIPJ. Marginal resection resulted in recovery of almost full range of motion. There was no recurrence up to the 8-month follow-up visit.

DISCUSSION: The case was compared to previously reported cases of osteochondroma of the hand. The management is discussed along with differentiating large osteochondromas from Nora's lesions in the hand.

CONCLUSION: A rare case of a large multi-lobed osteochondroma of the phalanx in an adult is presented and the management is discussed.

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1. Introduction

Solitary osteochondroma of the hand is a rare entity. Almost all previously reported cases in the hand were small tumors but required excision because of concurrent symptoms.

In children, most solitary tumors arise from the non-epiphyseal metaphysis or the metaphysis on the epiphyseal plate side of the middle or proximal phalanges of the fingers [1]. These tumors restrict motion and cause progressive finger deformity and hence early surgical excision is recommended [1].

Solitary osteochondromas of the hand which develop in adulthood are extremely rare and have different presentations depending on the site of origin of these tumors. For unknown reasons, most adult solitary tumors arise either from the distal phalanx or in the carpal bones. Distal phalangeal tumors almost always arise subungually and cause nail deformity [2]. Tumors arising from the carpal bones may cause extensor tendon rupture [3], carpal tunnel syndrome [4], or pain [5].

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In this paper, we present a rare case of a giant multi-lobed osteochondroma of the phalanx developing in an adult. No similar cases were found in the literature. The work has been reported in line with the SCARE criteria [6].

1.1. Presentation of case

A 25-year old male presented with a one-year history of a slowly growing mass of the right middle finger. There was no history of trauma, pain or other swellings in the body. Examination showed a multi-lobed bony mass located on the volar and dorsal aspects of the middle phalanx (Fig. 1). There were no sensory deficits. The proximal interphalangeal joint (PIPJ) had a 25° of flexion contracture and the range of motion in the arc of flexion of the PIPJ was also restricted to 90° because of the mass effect. Plain x-rays showed a multi-lobed osteochondroma arising from the base of the middle phalanx (Fig. 2). MRI (Fig. 3) showed that the tumor measured 2.3 × 2.5 cm and also showed the medial displacement of the flexor tendons by the tumor mass. Total excision with bone graft and possible fusion of the PIPJ was planned but the patient refused any compromise of function. The patient agreed to undergo marginal excision knowing the risk of recurrence. Resection was done preserving the neurovascular bundles, the flexor/extensor tendons, and the PIPJ. In order not to compromise the blood supply of the overlying skin, the volar lobe of the tumor was resected first via a volar lazy "S" incision; and one month later the dorsal lobe of the

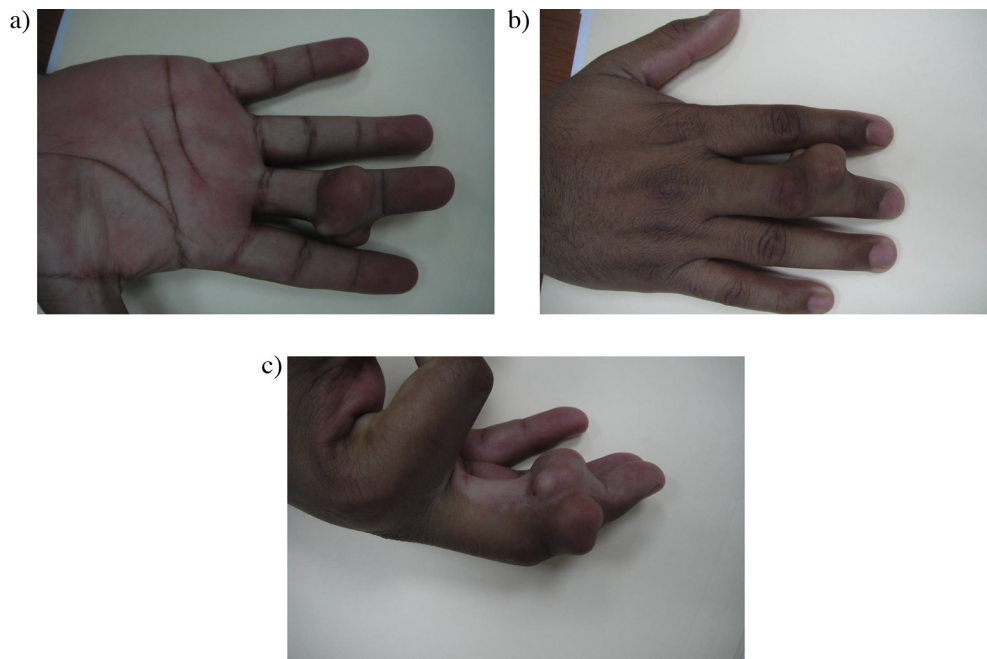


Fig. 1. (A–C) Clinical presentation of a multi-lobulated osteochondroma of the finger in an adult.

tumor was resected via a dorsal longitudinal skin incision. Histological examination was consistent with benign osteochondroma showing regular bony trabeculae arranged at 90° to the overlying cartilaginous cap (Fig. 4). The postoperative courses were uneventful. At final follow-up 8 months later, there was no evidence of recurrence. Sensory examination of the finger did not reveal any deficits. The flexion contracture at the PIPJ improved from 25° to 15° and the range of motion in the arc of flexion of the PIPJ improved from 90° to 110° (Fig. 5). X-rays showed the residual tumor at the base of the middle phalanx (Fig. 6) and the patient was advised for long term follow-up. However, he did not come back after the 8-month visit.

2. Discussion

Osteochondromas are common benign bony tumors which have a cartilaginous cap. The majority are seen arising from the long bones, particularly around the knee joint and in the upper humerus. Osteochondromas of the hand are rare and are usually seen in children as part of the multiple exostoses syndromes such as hereditary multiple exostoses [7] and Muenke Syndrome [8]. As mentioned in the introduction, solitary osteochondromas of the hand are rare in children and extremely rare in adults. Our case was in an adult and had several unique features: the origin being from the middle phalanx, the large size and being multi-lobed. The multi-lobulation required staged resection to protect the blood supply of the overlying skin. Our literature review did not reveal any similar case in the adult hand. One case of a large multi-lobed osteochondroma of the hamate in a child was reported and resection was also done through separate volar and dorsal incisions preserving the ulnar nerve [9].

Asymptomatic solitary osteochondromas of the long bones may be treated conservatively with regular follow-up. Complete resection is the treatment of choice for symptomatic solitary osteochondromas in adults. In our case, complete resection meant compromise of PIPJ motion and the patient refused our initial plan of management of complete excision. Incomplete (marginal) resection should take into consideration the risk of recurrence and also the risk of malignant transformation. The prevalence of malignant



Fig. 2. Plain X-ray showing the tumor arising from the base of the middle phalanx.



Fig. 3. MRI of the tumor.

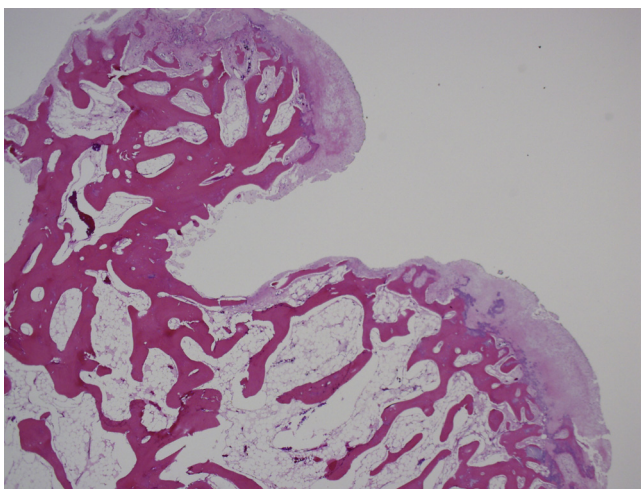


Fig. 4. Histology showing the regular bony trabeculae covered by the cartilage cap (H & E $\times$ 100).

transformation of benign osteochondromas varies from less than 1% for solitary tumors and 4% for hereditary multiple exostoses [10]. In recurrent tumors, a cartilaginous cap thickness of greater than 2 cm (as assessed by MRI or CT Scan) strongly indicates a secondary chondrosarcoma [10].

Large osteochondromas involving the phalanges of adults should be differentiated from bizarre parosteal osteochondroma-

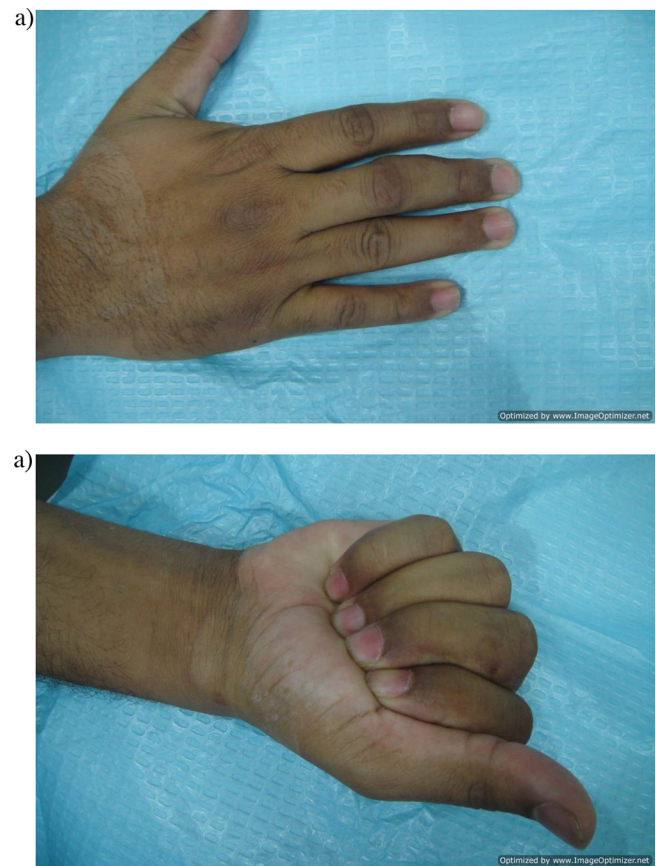


Fig. 5. (A&B) Range of motion 8 months after surgery.

tous proliferation (BPOP) which is also called Nora's lesion [11]. It is usually seen in adults and almost always arise from the phalanges, metacarpals or metatarsals. Radiologically, Nora's lesions resemble the appearance of solitary osteochondromas although matrix calcification is more irregular in Nora's lesions. The differentiation between BPOP and osteochondroma is made from histological examination. BPOP is composed of a random mixture of fibrous tissue, irregular bony trabeculae and cartilage cells. In contrast, osteochondromas are composed of regular bony trabeculae arranged at 90° to the overlying cartilage cap as seen in Fig. 4. Finally, the recurrence rate following excision is much higher in patients with BPOP compared to those with solitary osteochondromas [11]. In fact, spontaneous resolution of a pediatric solitary osteochondroma has been reported [12].

3. Conclusion

A rare case of large multi-lobed osteochondroma of the phalanx in an adult is presented and the management is discussed.

Conflict of interest

None.

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Fig. 6. X-ray 8 months after surgery showing the residual tumor.

Ethical approval

The study was approved by the Research committee of King Faisal Specialist Hospital and Research Center, Riyadh, Saudi Arabia.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by Editor-In-Chief of this journal.

Authors' contribution

All authors contributed significantly and in agreement with the content of the manuscript. All authors participated in data collection and in writing of the manuscript.

Guarantor

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