Hand & Microsurgery

Case Report
Hand Microsurg 2018;7:160-165
doi:10.5455/handmicrosurg.282349

eJM eJManager OPEN ACCESS

A rare case of recurrent enchondroma of the thumb involving the first metacarpophalangeal joint- an unusual disease pattern

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ABSTRACT

Enchondromas are common benign lesions of the hand. Recurrent lesions, however, may behave notoriously. A potential malignant transformation should be kept in mind while treating such tumors. We report a case of recurrent enchondroma of the first metacarpal of the right hand in a 37-year-old lady. A 37-year-old female patient who presented to us with a recurrent swelling of the thumb. She was surgically treated for the swelling five years back. The swelling recurred four months after surgery and had grown to the current size. We successfully managed the recurrent enchondroma by en bloc excision, bone grafting, and Kirschner wires to stabilize it. Recurrent enchondromas are to be approached cautiously. They may pose a malignant threat. The reasons for recurrence are to be investigated thoroughly to avoid a re-recurrence. Successful treatment of these lesions is dependent on the complete removal of the tumor bed followed by stabilization.

Key words: *Recurrent enchondroma, first metacarpal, Kirshner wire, bone graft, finger stiffness*

Introduction

Enchondromas are common benign tumors of the hand [1]. They form around 2.5% of all benign and malignant tumors [1,2]. They usually present in the fourth decade of life [2]. They can present as solitary monostotic forms which target small bones of the hand like the phalanges and the metacarpals [2,3]. The other is the multiple polyostotic variant, which targets many sites in a single bone or many bones. Two well- known forms of this pattern of disease are Ollier's disease (multiple enchondromatosis) or Maffucci syndrome (multiple enchondromatosis with capillary hemangiomas) [1]. These benign lesions are usually solitary and are slow growing. They are usually asymptomatic and painless and are diagnosed incidentally. Sometimes these lesions present with pain following a trivial trauma. These lesions are often surgically treated with curettage, and bone grafting or use of void- fillers [4].

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Received / Accepted : *November 01, 2017 / December 14, 2017*

We report an unusual case of a recurrent enchondroma of the first metacarpal involving the first metacarpophalangeal joint, which was successfully treated in a 37-year old lady.

Case Report

A 37yr old female patient presented to us when she noticed a mildly painful swelling over the base of her left thumb, which gradually progressed in size. She suffered from the restricted mobility of the base of her thumb. She did not suffer from any constitutional symptoms. She was newly diagnosed with Type 2 diabetes mellitus. She had similar swelling at the same site before, which was surgically treated five years back in another hospital, the reports of which were unavailable. She was relieved of her symptoms for a period of four months. The swelling then started reappearing four months after the first surgery and progressed to the present size. The swelling was mildly painful with restricted movements of the base of her thumb.

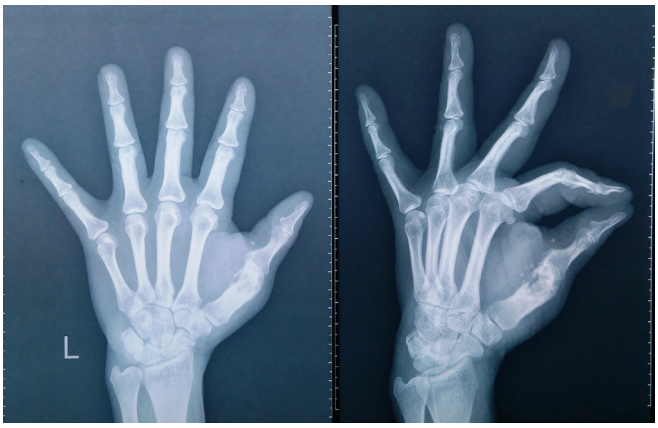


Figure 1. Preoperative x-ray showing the enchondroma of the first metacarpal.

Clinical examination revealed an oval-shaped swelling over the medial aspect of the base of her left thumb. The swelling was around 1x1 cm in size with poorly defined borders and no changes of the overlying skin. There was a second swelling seen over the lateral aspect of base of thumb with poorly defined borders as well. There was an old healed surgical scar of 0.5x6cm present over the lateral aspect of left thumb.

The two swellings were sized 1x1cm over medial

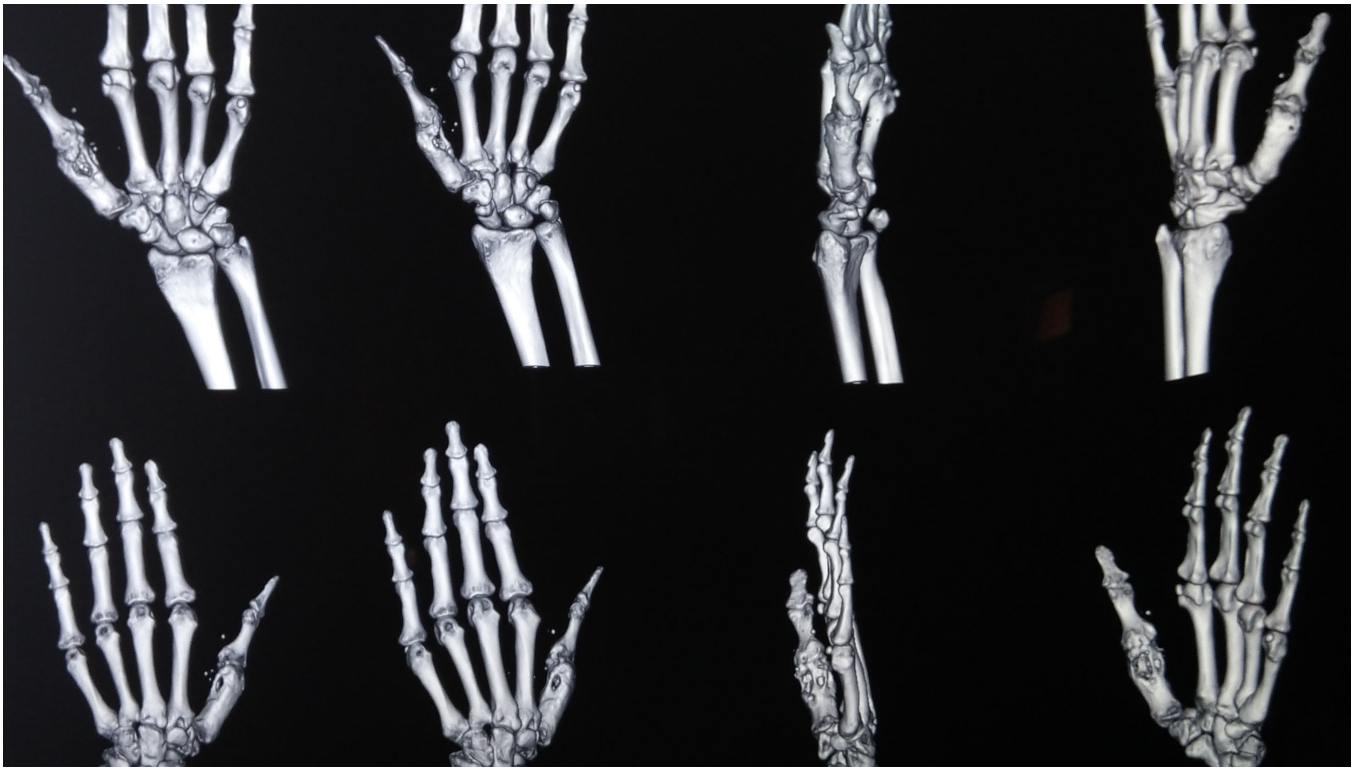


Figure 2. CT scan of the hand showing the extent of destruction of the bone by the enchondroma.

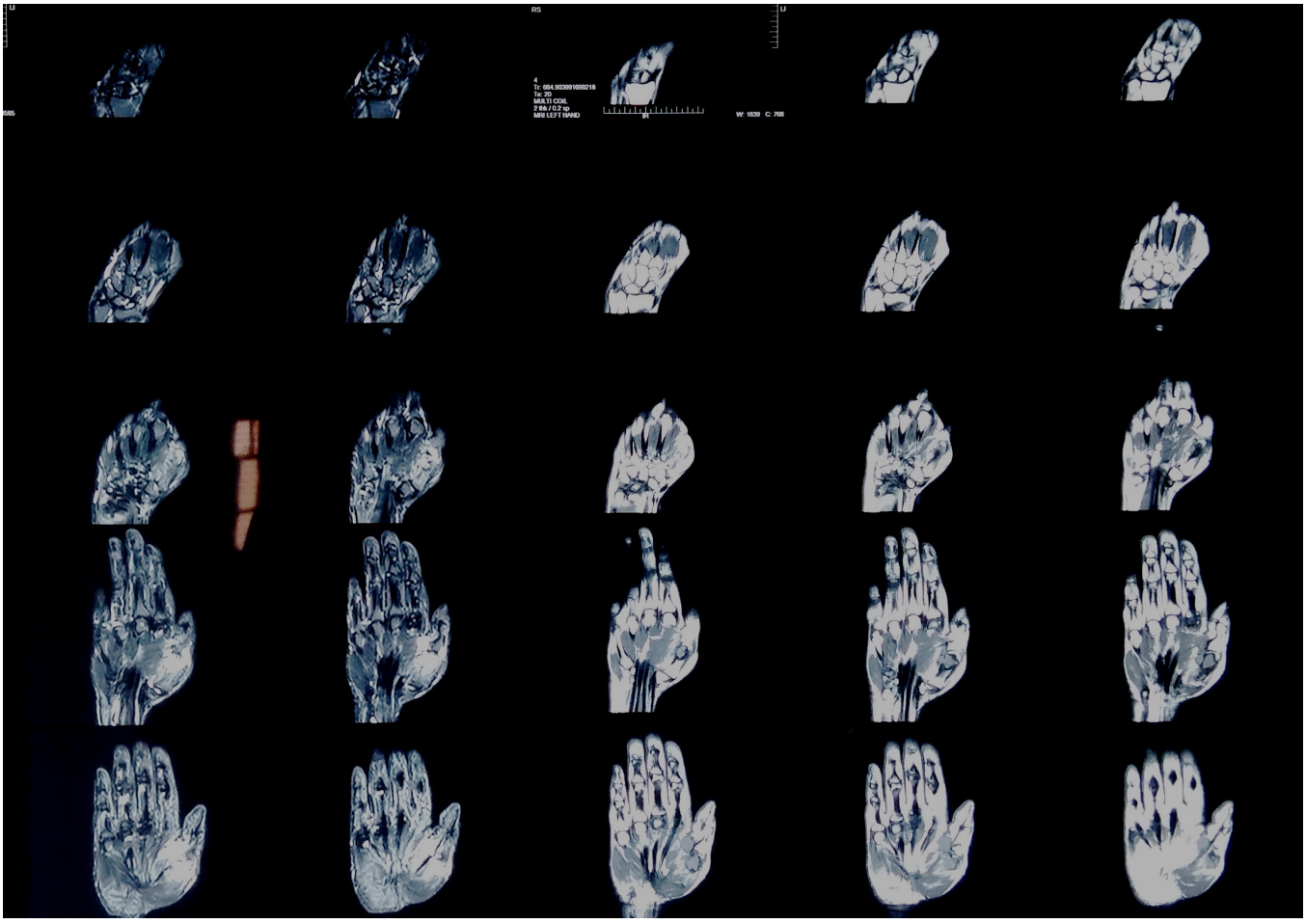


Figure 3. MRI scan of the hand showing the extent of lesion.

aspect & 2x3cm over the lateral aspect of the base of the left thumb. The swellings were immobile, firm to hard on consistency, nontender with no local warmth. The skin overlying the swelling was pinchable. Passive movements of the first metacarpophalangeal joint were not possible at all. Hand grip strength of the left hand was comparable to the right hand. Radiographs (X-ray, CT, MRI) revealed osteolytic lesions over the neck of the first metacarpal with cortical disruption. There were evident erosions of the first metacarpo-phalangeal joint and disruption of the cortex of the base of the first phalanx (Figures 1-3). The patient was taken up for surgery.

A 5cm incision was made over the previous surgical scar on the lateral aspect of thumb extending from interphalangeal joint to carpometacarpal joint. The surrounding soft tissue dissected, borders of the tumor

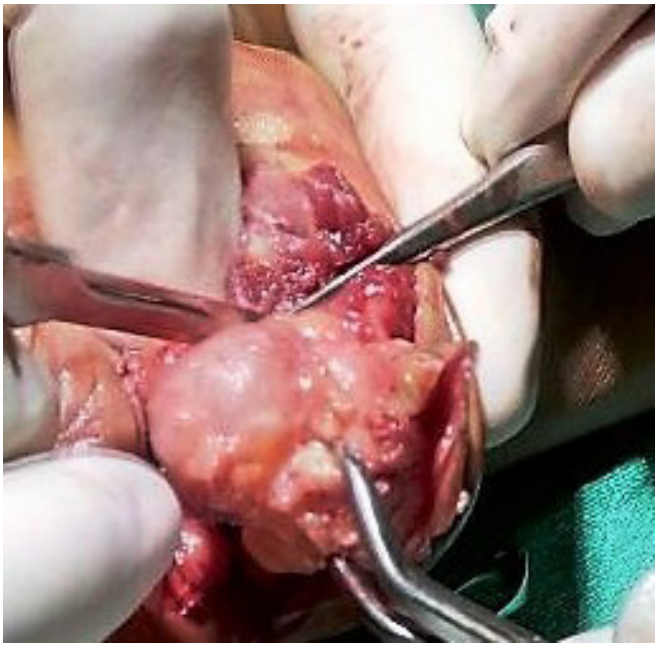
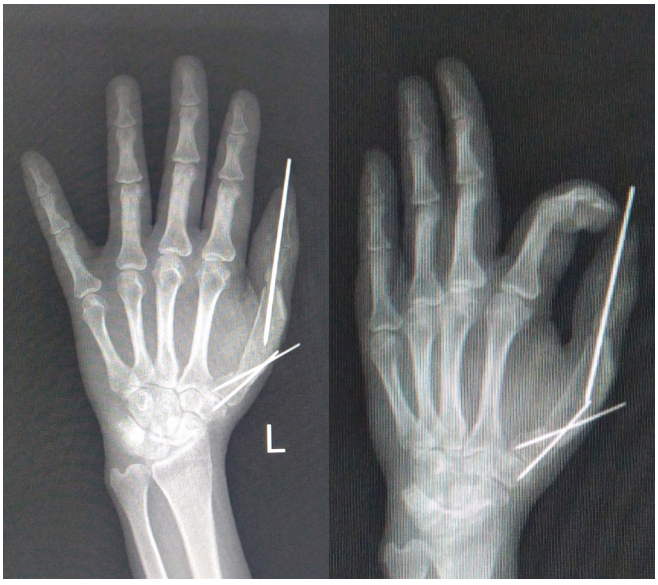
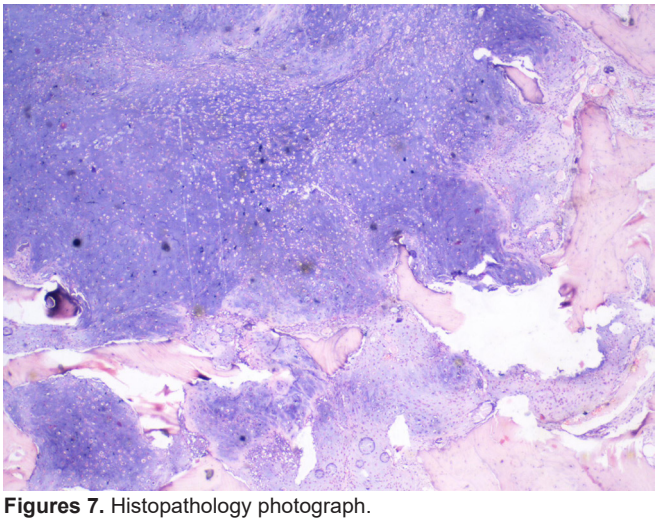


Figure 4. Intraoperative photograph of the lesion.



Figures 5-6. Immediate post-op x-ray showing bone graft with K wire in situ.



Figures 7. Histopathology photograph.

mass identified and completely resected en bloc (Figure 4). The resulting bone void was filled with tricortical bone graft taken from iliac bone graft and fixed with the aid of multiple Kirshner wires. The wound was closed, and the thumb immobilized using a thumb spica splint. The resected tumor mass was sent for histopathological studies. A repeat radiograph was done to confirm the complete removal of the tumor mass and adequate positioning of the bone graft (Figures 5,6). The postoperative period was uneventful, and sutures were removed on the tenth postoperative day. The Kirshner wires were removed after four weeks.

Histopathological studies showed a single greyish white mass sized 4.5x3.2x2.5cm. The mass was partly bony and partly cartilaginous. Microscopic examinations revealed lobules of cartilage surrounded by trabeculae (Figure 7).

Discussion

Enchondromas form 35-65% of all benign tumors of the hand [5]. The most common site for this lesion is the proximal phalanx, [2] followed by the metacarpals [5]. A thorough review of the literature suggests that the fifth finger is the most commonly involved in the hand with an incidence of 30-34.2% [6,7]. There has been no sex predilection for these tumors. The lesion usually presents in the third to fourth decade. About 35% of the lesions are diagnosed in this age group [8].

These lesions may be asymptomatic in few patients. The symptoms commonly associated with this lesion are slow growing swelling, pathological fracture (43%) and pain (65.7%) following trivial trauma [9].

The enchondroma is derived from actively proliferating chondrocytes derived from the growth plate. A small group of aberrant chondrocytes is believed to proliferate independently of the growth plate. These chondrocytes, like the traditional chondrocytes, do not proceed to hypertrophy and death for reasons unknown. These cells instead proliferate to form a cartilage tissue which remains uncalcified. This uncalcified cartilage mass grows from the underside of the growth plate into the primary bone tissue. The connecting bridge of tissue between the cartilage mass and the growth plate may remain intact or calcified to form primary bone tissue. This cartilage mass might be walled off from normal tissue by lamellar bone or may undergo calcification or may proliferate as an intraosseous chondroma. This pathophysiology may play a role in similar conditions like fibrous dysplasia [9].

X-ray remains the most important investigation of choice to diagnose these tumors [10]. These tumors show up as well defined lytic lesions with central lucen-

cy with or without specs of calcification in the diaphysis or metaphysis of the hand. As these tumors grow, endosteal scalloping, cortical thinning and remodeling of the bone become evident [9]. Magnetic resonance imaging (MRI) and Computed Tomography scans (CT) have limited role in diagnosing enchondromas. They may be useful in delineating the boundaries of the tumor, at best.

The treatment of these tumors is purely based on the patient's symptoms. Observation with regular follow-up is recommended in asymptomatic non-expanding lesions. Symptomatic lesions are treated with extended curettage and bone grafting of the cavity formed. Void fillers like calcium phosphate, hydroxyapatite, calcium sulfate have also been used with good results [4].

Sassoon et al. reported that there was no difference in the complication rate in immediate versus delayed treatment of these lesions by curettage, bone grafting and internal fixation of the involved bone [5].

Bone grafts have a higher subsidence but low compressibility. Bone graft has the disadvantage of donor site morbidity, but there is no difference in the healing time, recurrence rates, joint stiffness when compared to void fillers like calcium phosphate [1,4]. Void fillers, on the other hand, have low subsidence. They, however, are easily injectable, they can fill up irregular cavities, provide immediate biomechanical stability, and no donor site morbidity. Problems with difficulty in performing corrective osteotomy following malunion, have been reported with calcium sulfate and calcium phosphate. It does not permanently absorb into the bone [1].

We decided to use tricortical bone graft as it would provide the necessary biomechanical stability as well as have the osteogenic potential to fill up the void and get fused with the parent bone.

Enchondromas are slow-growing benign tumors and are usually asymptomatic. Hence recurrence of these lesions is usually diagnosed very late. The exact rate of recurrence of these tumors is difficult to gauge due to the recurrent lesions being asymptomatic [9]. Sassoon et al. and Gaulke et al. reported a recurrence rate of 7 and 14.3% respectively [7,9]. Tao et al. in his study of 20 cases reported a recurrence rate of 5% [9]. A longer follow up of such recurrent lesions might throw some light on the exact recurrence rates.

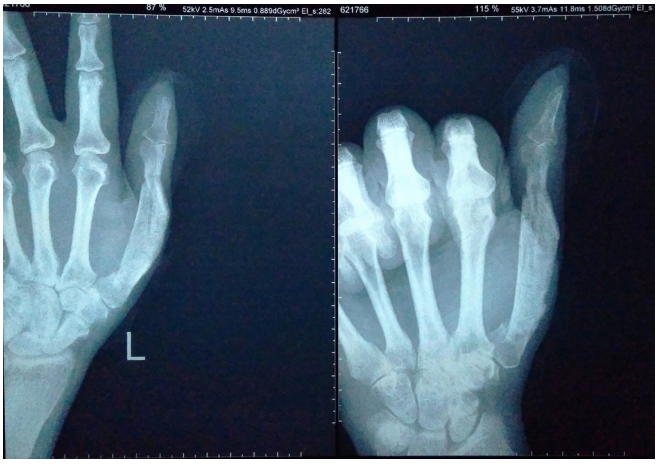


Figure 8. Follow up x-ray showing the graft incorporated into the first metacarpal.

Common complications following surgical treatment include recurrence, pathological fractures, post-operative finger stiffness and rarely malignant transformation of the recurrent lesion. The stiffness could be minimized by passive mobilization and staged physiotherapy [9]. Our patient had finger stiffness, possibly because the lesion involved the first metacarpophalangeal joint. He underwent physiotherapy and succeeded in obtaining a functional range of movement of the thumb. There was no evidence of recurrence of the lesion at one-year follow-up (Figure 8).

Conclusion

Enchondromas are common benign tumors of the hand. Solitary lesions rarely are malignant. We report a case of recurrent enchondroma of the first metacarpal of the right hand unusually involving the first metacarpo-phalangeal joint, in a 37-year-old lady who was successfully treated with bone grafting and Kirshner wires. Recurrent lesions such as in our case have a significant malignant potential, and the treating surgeon should be cautious while approaching such lesions.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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