Annals of Clinical and Medical Case Reports

Case Report ISSN: 2639-8109 | Volume 8

Giant Cell Tumor of the Middle Phalanx of Ring Finger Treated with Wide Excision and Reconstruction using Non-vascularized Fibular Graft: A Rare Case Report and Review of Literature

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Received: 02 Feb 2022

Accepted: 15 Feb 2022

Published: 19 Feb 2022

J Short Name: ACMCR

Citation:

Rahyussalim AJ, Makabori JCF, Zufar MLL, Kurniawati T, Giant Cell Tumor of the Middle Phalanx of Ring Finger Treated with Wide Excision and Reconstruction using Non-vascularized Fibular Graft: A Rare Case Report and Review of Literature. Ann Clin Med

1.Abstract

Giant cell tumor (GCT) involving the hand, specifically the phalangeal bones, is very rare, accounting for only 2% of cases of the bone. This type of GCT is associated with more aggressive behaviour and higher recurrence and metastasis rate. We present a rare case of large GCT of the middle phalanx of ring finger in a 33 year old female. The patient was treated with wide excision and reconstruction using non-vascularized fibular graft. At her most recent follow-up time, 1 year post-surgery, the patient showed no symptoms with neither complication nor recurrence reported.

2. Introduction

Giant cell tumor (GCT) is one of the most commonly found benign tumor involving metaphysis- epiphysis of long bones [1]. However, its involvement of the hand, especially phalanx, is extremely rare. It only represented by 2% of all reported cases [2,3]. In this region, GCT is associated with more aggressive behaviour and higher recurrence rate [4]. Surgery becomes the primary mode of treatment, involving curettage only, curettage with bone graft, amputation, resection with reconstruction, and radiotherapy. However, major throwbacks of recurrences, disfiguring of esthetical appearance, and loss of limb function have been reported [5-7]. This mode of treatment needs to be decided specifically for each case. In this study, we report a rare case of 33 year old female presented with large giant cell tumor of the middle phalanx of ring finger treated with wide excision and reconstruction using non-vascularized fibular graft.

3. Case Presentation

A 33-year-old female presented with pain and swelling on the right ring finger since 2 years before the admission to our centre. This swelling appeared suddenly without any history of trauma or other constitutional symptoms. Initially, the swelling at the ring finger of the left hand was as big as a marble. However, she did not seek for any further treatment since she was able to perform normal activities of daily life. At 9 month prior the admission, the lump had progressively enlarged and disturbed patient's activity. Patient went to another centre and had plain radiograph examination in which the result showed a condition of bone tumor. MRI and biopsy were then recommended for the patient. Patient then took a referral and was referred to our centre. Furthermore, there was no remarkable history of past illness or past medication.

Physical examination showed a swollen mass which was irregular in shape, immobile, firm in consistency, and tender (Figure 1). The diameter of the mass was 10 cm compared to 4 cm in the contralateral side. There was venectation overlying the skin without any changes in skin colour, distal edema, or open wound. Distal sensory examination and capillary refill time were within normal limit.

There was normal range of motion for adjacent metacarpophalangeal (MCP) joint. However the motion of proximal interphalangeal (PIP) and distal interphalangeal (DIP) joint was limited due to the mass (Figure 2).

Laboratory study for complete blood count, C-reactive protein, erythrocyte sedimentation rate, and serum alkaline phosphatase were within normal range. Plain radiograph showed an expansile lytic lesion on the shaft of middle phalanx of the right ring finger involving proximal interphalangeal (PIP) and distal interphalangeal (DIP) joint with irregular sclerotic border. There was cortical breach with no sign of periosteal reaction or fracture line (Figure 3). Histopathological examination demonstrated mildly pleomorphic cells with spindle-shaped nucleoli along with uniformly distributed multinucleated datia cells with eosinophilic cytoplasm and vesicular nucleus consistent with diagnosis of giant cell tumor. Wide excision followed by reconstruction using non-vascularized fibular graft and fixation using K-wire was then performed in this patient (Figure 3-4). On the 6 months post-surgery, healing of the wound without no superficial or deep infection was reported (Figure 6). The finger remained to be viable and survive with no observed recurrence on the 1 year follow-up time.

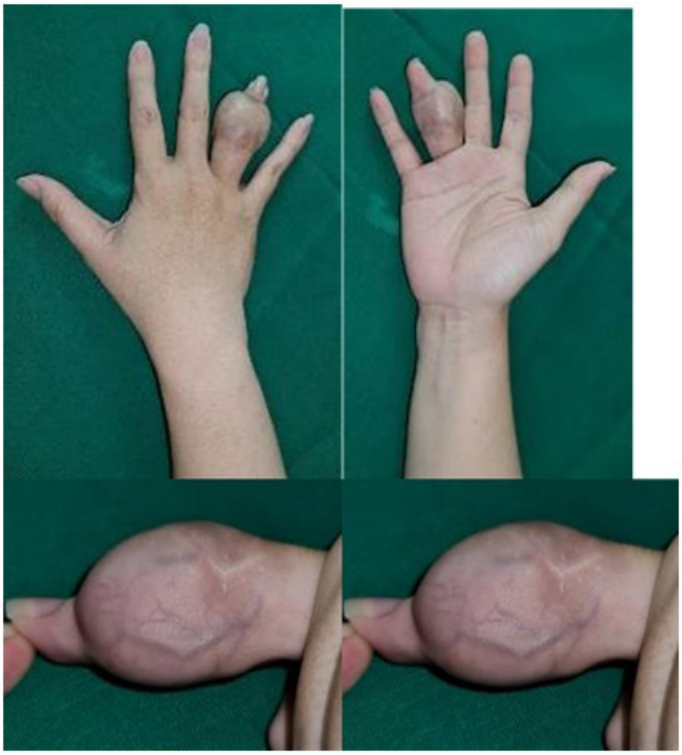

Figure 1: Patient's Clinical Manifestation

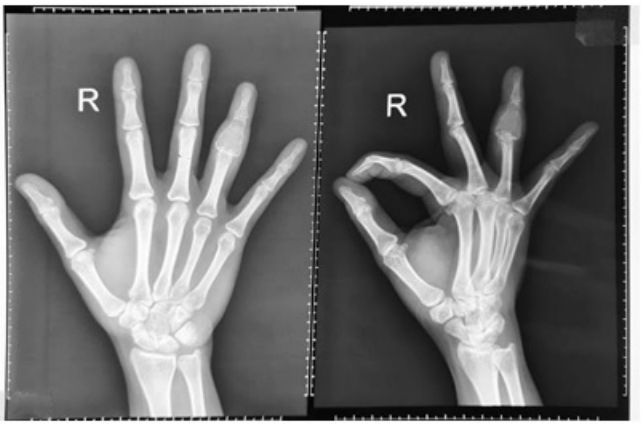

Figure 2a

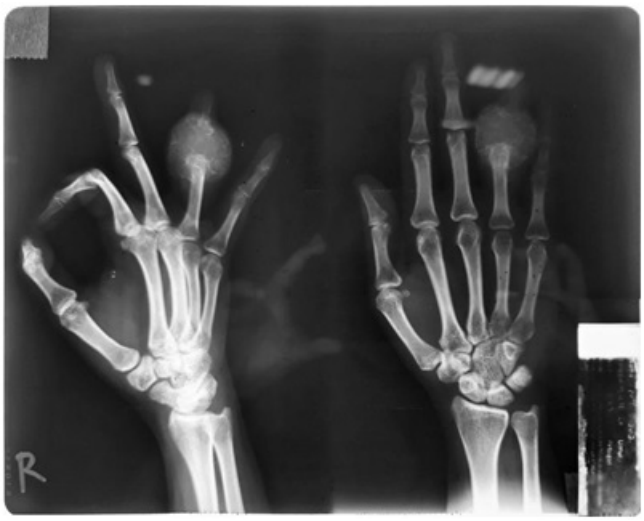

Figure 2b

Figure 2: Plain Radiograph Examination a)1 month before admission b) on admission

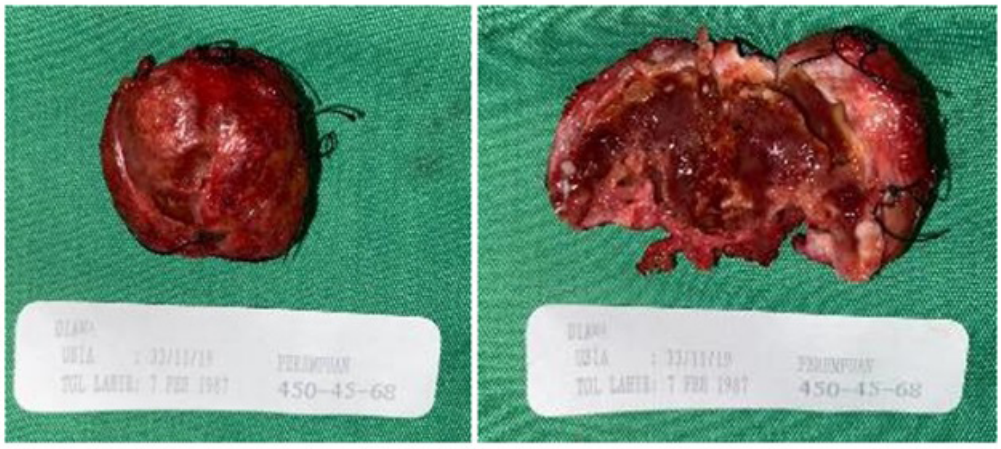

Figure 3: Gross Specimen

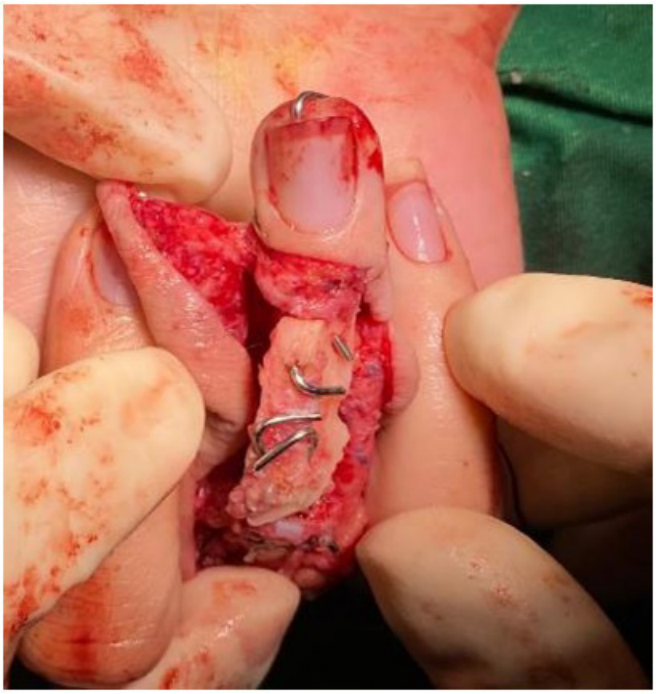

Figure 4: Reconstruction using non-vascularized fibular graft and K-wire fixation

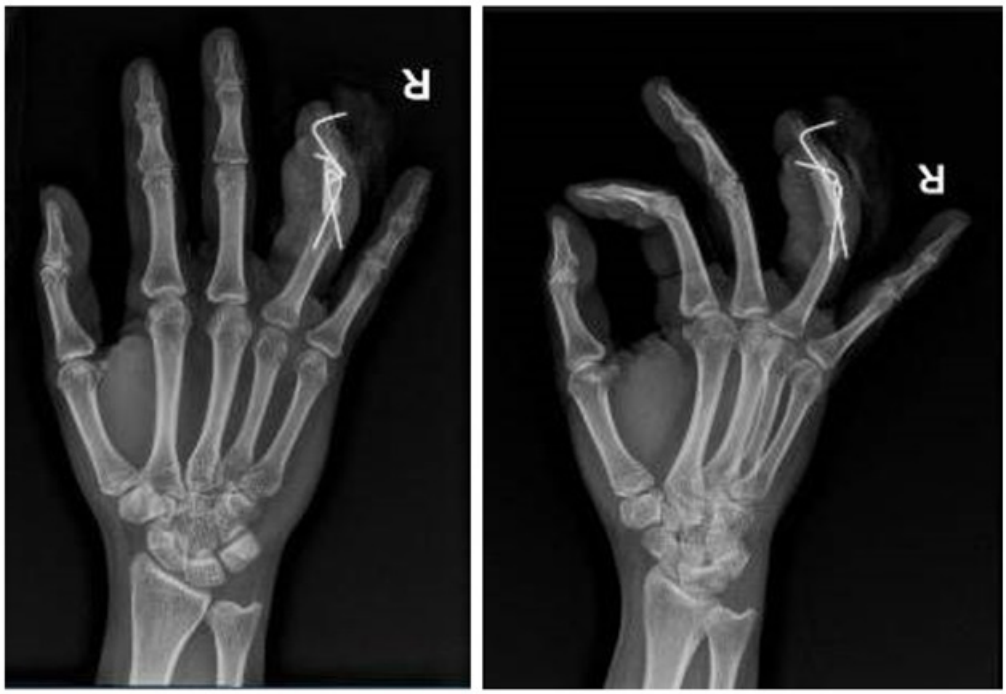

Figure 5: Postoperative X-ray

4. Discussion

Giant cell tumor is one of the most commonly found benign bone tumor which is locally aggressive [1,7]. This tumor mainly affects epiphyseal region, especially adjacent to the knee joint. Of the all reported GCT cases, GCT of the hand is a rare entity represented by only 2-4% of cases [2,3]. These cases presented with different characteristics in regard to the more aggressive behaviour and earlier recurrence rate from conventional cases [4-7]. GCT of the hand mainly affects metaphyseal region with the growth towards diaphysis. Furthermore, rarer occurrences of this tumor are encountered in the phalanges of the hand. This was also reported by previous studies. In large series of 568 GCT cases, only 4 cases of GCT of the phalanx were reported [8]. In another large series of 327 patients, there was only one GCT case involving the phalanges [9]. A report of 2400 skeletal GCT cases showed less than 50 cases with phalangeal involvement [10,11]. Six GCT of the phalanges were reported by Goldenberg et al from their study comprising 218 cases of GCT [12]. Other studies from Coley et al and Saikia et al of 108 and 124 skeletal GCTs respectively, two cases of phalangeal involvement were found within each study [10, 13].

Insidious onset of localized pain with progressive worsening becomes the main symptoms encountered in the first time. This is accompanied with progressive enlargement of the swelling resulting in functional impairment of the affected region, especially range of motion if the tumor is located adjacent to a joint [1,6,7]. Due to the tumor location which is close to the skin surface and its high mobility, the symptoms of the GCT involving the hand may occur earlier (2,13,14). Pathological fracture resulting in acute pain has also been reported in some of the cases [15, 16]. This tumor can be also locally aggressive, breaching out the cortex and invading adjacent soft tissue. Skin ulceration may be discovered, but this condition is not a routine findings [1, 7, 14].

Plain X-ray becomes the easiest and primary radiography examination of choice. GCT demonstrates geographic lytic lesion with thinning of cortex and well-defined margin affecting epiphyseal- metaphyseal region. There is no marginal sclerosis, periosteal reaction, nor internal calcification [1,7,16]. In the hand, primary GCT mostly fills up the central location compared to the classical eccentric distribution on the other sites. Grading by radiological findings is described by Campanacci in which lesion with well-defined border of thin margin and preserved cortical layer is included in grade 1, lesion with larger involvement area extending the cortical layer without any breakage is included in grade II, and a more aggressive lesion invading the adjacent tissues is included in grade III. A grade III lesion is associated with a positive correlation of a more aggressive tumor and higher recurrence rate [9].

Computed tomography (CT) may play important roles to identify thinning of the cortex, margin of the lesion, pathologic fracture, extent of the bone loss, and remodelling of the osseous expansion. Besides, it is also used to corroborate the absence of matrix mineralization and periosteal reaction [17]. Another preoperative examination to evaluate GCT is MRI. This method can delineate the tumor extent, whether it is intraosseous, extraosseous, intra-articular, or affecting adjacent soft tissue. T1 and non fat-suppressed T2-weighted MR imaging frequently reveal a low to intermediate signal intensity for the solid lesion of GCT. Local hypercaptation is found in bone scintigraphy of GCT. This is due to the increase of reactive osteoblastic activity. Donut sign was established in 57% of cases, representing central photopenia and increase of peripheral radionuclide uptake [1,18]. This examination should be carried

out for screening of other bone lesions.

Tissue examination becomes the gold standard to diagnose GCT since imaging can be similar to many other bone lesions, particularly enchondroma which is more commonly found in the hand region. A dark brown mass with soft to firm consistency, fibrotic and osteoid production region, yellowish necrotic area, and dark red red haemorrhage area can be observed in gross specimen. Multinucleated osteoclast-like giant cells becomes the hallmark in histopathological findings [19, 20]. In addition, there may be also mitotic activity with no nuclear atypia or any other abnormalities. Furthermore, chest radiograph should be performed routinely to rule out any pulmonary metastasis. Results from clinical findings, imaging, and tissue examination should be attributed simultaneously in establishing the correct diagnosis of the patient [1,20].

Aggressiveness and tendency to recur of the lesion and retaining function of the affected region become the main challenges in treating GCT patients. Besides, greater destruction of the bone and extension towards diaphysis may complicate the treatment for GCT of the hands [7,12,14,16,19,21]. Adequately aggressive surgical treatment is required to control the tumor locally, maintain hands function and its aesthetic appearance satisfactorily, and avoid recurrence in the future. Curettage, curettage with bone graft, amputation, resection with reconstruction, and radiotherapy become the available types of treatment. Curettage with or without bone grafting as the most commonly used treatment showed unsatisfactory results. Besides, this type of treatment was also associated with high percentage of recurrence rate of up to 65% [6,22]. Wide excision and amputation were reported to overcome this problem in which more than 70% of recurrent lesions were successfully treated [2]. However, this choice comes with major deterioration of disfiguring esthetical appearance and loss of limb function. Moreover, malignant transformation of the lesion has also been reported whether due to the radiotherapy as the primary procedure or spontaneous process without any relation to the treatment [23].

Resection with reconstruction comes to the more ideal intervention since re-establishment of structural and functional integrity can be achieved. Bone graft of autograft or allograft, prosthesis, and polymethylmethacrylate (PMMA) can be used for reconstruction of the bone defect [1,7,9,15,16,21]. In regard to this case, we treated our patient with wide excision of the middle phalanx and reconstruction using using non-vascularized fibular graft and K-wire fixation. This is according to our concern of the higher risk of recurrence and likelihood of infection, non-healing of the reconstruction, and gangrene formation as local complication. Besides, adequately local curettage could not be achieved. Satisfactory patient outcome has been achieved within 6 months post-surgery in which healing of the wound was achieved without any complication and the finger remained to be viable. In addition, local adjuvant treatment using chemical agents with phenol, alcohol, or cryotherapy can enhance intralesional excision procedures [4,5]. Furthermore, the use of other cytotoxic adjuvants, such as monoclonal antibody, calcitonin, and bisphosphonate were also reported to strengthen effectivity of the preferred treatment and decrease the risk of recurrence [1,7,12-14].

GCT of the hand holds important aspect for its high recurrence rate, roughly about 70% of the cases (1,15,21) . For the most of cases, this condition occurs within the first 24 months after the surgery, especially 12 to 18 months [24]. This rate of recurrence is said to be lower with the range of 5 to 10% when the surgery procedure is appropriate [24,25]. Patients with recurrent lesion should be observed carefully due to its more aggressive and malignant behaviour. Besides, risk of pulmonary metastasis needs to be put in mind even it is a small possibility. Further clinical and imaging examination follow up are required to be carried out routinely. Close monitoring every three to four months within the first two years is important to detect any recurrence at an early stage. When there is suspected recurrent lesion in plain radiograph, further work-up using CT or MRI is recommended to clarify the condition. The follow up is carried on every six months after two to three years and annually after five years [26,27]. Chest x-ray is suggested to be performed in every follow-up session to detect early sign of pulmonary metastasis (28). Chest CT can be performed for further examination due to its higher sensitivity than plain radiograph [1,17].

5. Conclusion

GCT in the hand, particularly involving phalanx, is a rare entity. This classically benign tumor can be locally aggressive. Besides, its hallmark characteristics present with more aggressive and higher rate of recurrence compared to GCT in the other sites. Misdiagnosis as enchondroma is encountered due to the similarity of findings and more commonly found cases in the hand. Histopathological and imaging studies in addition to complete physical examination are required for establishing the correct diagnosis. Treatment should be adequate and specific for each case to prevent the recurrence and preserve limb functions. In our case, wide excision and reconstruction using non-vascularized fibular graft with K-wire fixation might become the ideal treatment of choice. Moreover, post-operative follow-up is necessary to monitor the risk of recurrence. Further examination using chest X-ray and chest-CT can be performed for early detection of pulmonary metastasis.

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