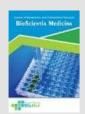
eISSN (Online): 2598-0580



## Bioscientia Medicina: Journal of Biomedicine & Translational Research

Journal Homepage: <u>www.bioscmed.com</u>

# Biological Reconstruction for Metacarpal Osteofibrous Dysplasia Treatment: A Case Report

#### Primadika Rubiansyah<sup>1</sup>, Arazy Gifta Prima<sup>2\*</sup>, Nursanti Apriyani<sup>3</sup>

<sup>1</sup>Orthopaedic Surgeon, Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia <sup>1</sup>Lecture Staff, Faculty of Medicine, Universitas Sriwijaya, Palembang, Indonesia <sup>2</sup>Orthopaedic Residence, Faculty of Medicine, Universitas Sriwijaya, Palembang, Indonesia <sup>3</sup>Pathologist, Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

#### ARTICLE INFO

**Keywords:** Hemiarthroplasty Metacarpal Metacarpophalangeal joint Osteoblastic Osteofibrous dysplasia

#### \*Corresponding author:

Arazy Gifta Prima

### E-mail address: arazygp@gmail.com

All authors have reviewed and approved the final version of the manuscript.

https://doi.org/10.37275/bsm.v7i5.820

#### ABSTRACT

Background: Osteofibrous dysplasia is a rare benign bone tumor characterized by abnormal differentiation of fibrous tissue in bone and commonly involves the maxilla, mandible, and tibia and affects children less than 10 years old. Osteofibrous dysplasia affecting the metacarpal is a very rare case. This study presents a case report to present biological reconstruction in the management of metacarpal osteofibrous dysplasia. Case presentation: A 48-year-old female presented with swelling and pain in her right hand over 1 year. On examination, palpable mass with immobile and hard consistency on the first metacarpal and distal neurovascular within normal limit. Radiographic examination (X-ray and MRI) supports osteofibrous dysplasia. We performed complete resection of the first metacarpal and used a fibular graft to maintain length, followed by arthroplasty of the first metacarpophalangeal joint and arthrodesis of the first carpometacarpal joint. Pathological results showed a Chinese letter appearance with osteoblastic rimming that confirmed osteofibrous dysplasia. After 6 months of follow-up, there is no recurrence, and the functional outcome is excellent (MSTS score 93,33%). Conclusion: Osteofibrous dysplasia on the metacarpal and affected adult is a very rare case. Treatment with complete resection of the metacarpal, using a fibular graft to maintain bone length, and arthroplasty MCP joint will give good results (MSTS score 93,33%). Follow-up for recurrence is necessary.

#### 1. Introduction

Osteofibrous dysplasia is a rare condition that affects the bones in the human body. This condition is characterized by abnormal growth of bone and fibrous tissue. Osteofibrous dysplasia commonly occurs in childhood and adolescence and can affect various bones in the body, including long bones such as the shinbone (fibula) and femur. This condition can cause deformities and bone strength that have the potential to interfere with normal function and increase the risk of fractures.<sup>1-3</sup> Although osteofibrous dysplasia is a rare condition, it is important to be aware of and recognize the symptoms that it may present. This condition is usually detected in childhood or adolescence when bone growth is active. Symptoms can vary, including bone pain, swelling, deformity or abnormal shape of the bone, and possible fractures. The urgency to treat osteofibrous dysplasia is mainly related to its impact on the function and quality of life of sufferers.<sup>4-6</sup>

This condition can cause interference with walking, standing, or daily activities. If left untreated or managed properly, osteofibrous dysplasia can lead to complications such as frequent fractures or permanent deformities of the bones. In treating osteofibrous dysplasia, different treatment approaches can be used, including monitoring of bone changes, pain management, and in some cases, surgical intervention. Collaboration between orthopedic doctors, orthopedic surgeons, and other medical care teams is very important to plan appropriate treatment strategies according to individual needs.7-10 This study report to present biological presents a case reconstruction in the treatment of metacarpal osteofibrous dysplasia.

#### 2. Case Presentation

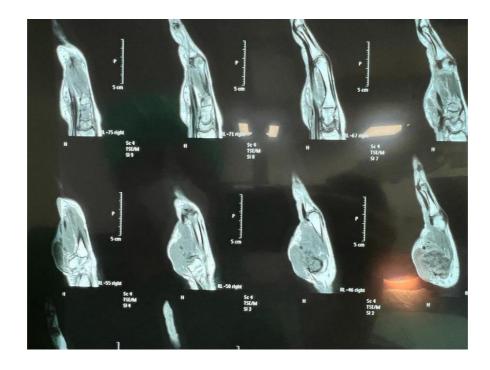
A 48-year-old female presented with swelling and pain in her right hand over 1 year. The patient also complained that there was a lump. The lump gradually gets bigger. On examination, palpable mass with immobile and hard consistency on the first metacarpal and distal neurovascular within normal limit. Radiographic examination revealed multiple lucencies with intervening sclerosis, and MRI revealed multiple low-intermediate T1 signal/ T2 high signal intracortical foci, separated by low T1/T2 signal cortical bone and sclerotic reaction with the prominent enhancement of the entire first metacarpal.



Figure 1. Clinical pre-operative.



Figure 2. Radiograph pre-operative showed multiple lucencies with intervening sclerosis.



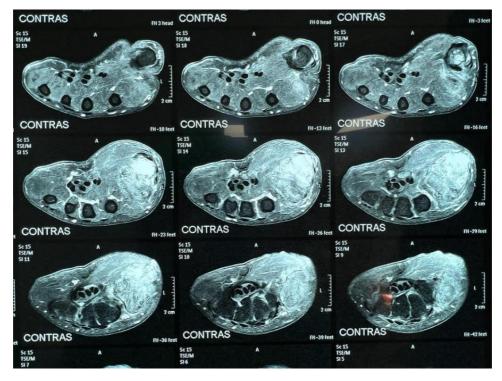


Figure 3. MRI examination showed low-intermediate T1 signal/ T2 high signal intracortical foci, separated by low T1/T2 signal cortical bone and sclerotic reaction with the prominent enhancement of the entire first metacarpal.

The metacarpophalangeal joint of the second metacarpal was preserved. The fibular graft was proximally fixated with two transcortical titanium screws and distally with two crossed 1 mm Kirschner wires. In this study, we performed complete resection of the first metacarpal with reconstruction using a fibular graft composite prosthesis to change the metacarpal bone and to maintain bone length. The fibular graft was proximally fixated with a mini plate, and we performed metacarpophalangeal arthroplasty to maintain metacarpophalangeal joint function. The 6 monthly postoperative outcome in this study was good, with the normal function of the hand with MSTS (the musculoskeletal tumor society functional evaluation) score being 93.33% (EXCELLENT).

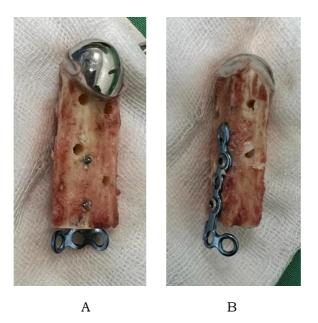


Figure 4. A. Arthrodesis of first carpometacarpal joint with the mini plate. B. Hemi arthroplasty first Metacarpophalangeal joint.



Figure 5. Post-operative picture after six months of surgery.

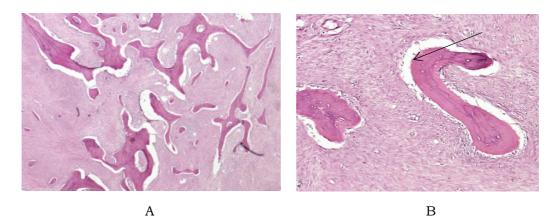


Figure 6. (A) Chinese letter appearance with osteoblastic rimming. (B) (arrow) confirmed osteofibrous dysplasia.

#### **3. Discussion**

Osteofibrous dysplasia is indeed a benign bone tumor that primarily affects children and young adults. It typically presents as a slow-growing, painless swelling or a deformity in the long bones, most commonly in the tibia (shinbone) but occasionally in the fibula or other bones. While the exact cause of osteofibrous dysplasia is not fully understood, it is believed to be related to abnormal bone development. Surgical treatment is often recommended for osteofibrous dysplasia, particularly when there is a corrective deformity or the lesion is symptomatic. The goals of surgery include correcting any bone deformities, preventing further pathological changes, eradicating the symptomatic lesion, and preserving or improving hand function if it is affected. The specific surgical approach will depend on the location and extent of the lesion, as well as individual patient factors. In some cases, the surgeon may perform a curettage procedure, which involves removing the abnormal tissue from the affected bone. This can help relieve symptoms and prevent further progression of the condition. In more severe cases or when there is a risk of fracture, additional measures such as bone grafting or stabilization with internal fixation devices may be necessary.11-13

Curettage is a surgical procedure commonly used in the treatment of benign bone tumors, including osteofibrous dysplasia. It involves the removal of the abnormal tissue or tumor from the affected bone. During the procedure, the surgeon makes an incision over the affected area to access the bone. They then use a specialized instrument called a curette to scrape or scoop out the abnormal tissue. The goal is to completely remove the tumor while preserving as much healthy bone as possible. After the curettage is performed, the surgeon may use other techniques to ensure the complete eradication of the tumor and promote bone healing. These may include applying chemicals to the cavity left after curettage to destroy any remaining tumor cells, filling the cavity with bone graft material to promote bone regeneration, or using internal fixation devices (such as plates, screws, or rods) to stabilize the bone if necessary.<sup>14-16</sup>

Complete resection of the first metacarpal bone with reconstruction using a fibular graft composite prosthesis is a surgical procedure performed in cases where there is a need to remove the entire first metacarpal bone and replace it with a suitable substitute to maintain hand function and bone length. This procedure is often done in cases of severe bone tumors or other conditions that necessitate the removal of the metacarpal bone. The surgeon makes an incision over the affected area to access the first metacarpal bone. The entire first metacarpal bone is carefully removed, taking care to preserve any surrounding healthy tissues. A segment of the fibula, which is a bone located in the lower leg, is harvested to serve as a graft. The length of the graft is determined based on the bone defect created by the metacarpal resection. The fibular graft is shaped and sized appropriately to match the length and dimensions of the removed metacarpal bone. The fibular graft is then combined with a suitable prosthesis, which may be made of metal or other biocompatible materials. This composite prosthesis is designed to mimic the structure and function of the original metacarpal bone. The composite prosthesis is secured in place using screws, plates, or other fixation devices to ensure stability. The incision is closed using sutures or staples, and appropriate dressings are applied.<sup>17,18</sup>

The musculoskeletal tumor society (MSTS) functional evaluation is a widely used system for assessing the functional outcomes of patients with musculoskeletal tumors. It is specifically designed to evaluate the functional abilities and quality of life of individuals who have undergone surgical treatment for bone or soft tissue tumors in the extremities. The MSTS functional evaluation consists of several components that assess various aspects of a patient's function and mobility. The evaluation considers the level of pain experienced by the patient, both at rest and during activities. It takes into account the severity and frequency of pain and its impact on daily activities. This component assesses the patient's functional abilities in various areas, such as walking, standing, climbing stairs, and performing specific tasks related to the affected limb. The evaluation may include tests to measure a range of motion, muscle strength, and overall functional capacity. This aspect evaluates the patient's psychological well-being and emotional adjustment to their condition and the impact it has on their daily life. It considers factors such as anxiety, depression, body image, and selfesteem. The evaluation takes into account the need for any assistive devices or orthoses (braces, splints, etc.) that the patient requires for mobility and function. This component assesses the need for walking aids such as crutches, canes, or walkers to assist with mobility. The evaluation considers the impact of the musculoskeletal tumor and its treatment on the patient's ability to perform work-related tasks and engage in recreational activities. The MSTS functional evaluation typically involves a combination of patientreported questionnaires, physical examinations, and functional tests conducted by healthcare valuable professionals. The results provide information for treatment planning, monitoring functional and progress over time, guiding rehabilitation strategies to optimize the patient's quality of life.19,20

#### 4. Conclusion

Osteofibrous dysplasia on the metacarpal and affected adult is a very rare case. Treatment with complete resection of the metacarpal, using a fibular graft to maintain bone length, and arthroplasty MCP joint will give good results (MSTS score 93,33%).

#### 5. References

- Saini V, Sikka M, Srivastava DN. Osteofibrous dysplasia-like adamantinoma of long bones: a distinct entity with better prognosis. J Cancer Res Ther. 2014; 10(1): 97-101.
- Damron TA, Ward WG, Stewart A. Osteofibrous dysplasia: a review of the literature and presentation of an additional 3 cases. Clin Orthop Relat Res. 2004; (418): 205-14.
- Unni KK, Dahlin DC, Beabout JW. Osteofibrous dysplasia. Cancer. 1972; 30(3): 755-61.
- Schajowicz F. Osteofibrous dysplasia of long bones. A new clinical entity. Arch Pathol Lab Med. 1981; 105(7): 397-402.
- Edeiken J, Ayala AG, Ackerman LV. Osteofibrous dysplasia of the tibia. Skeletal Radiol. 1984; 11(3): 155-62.
- Branstetter DG, Nelson SD, Manivel JC, Blay JY, Chawla S, Thomas DM. Denosumab induces tumor reduction and bone formation in patients with giant-cell tumor of bone. Clin Cancer Res. 2012; 18(16): 4415-24.
- D'Ambrosio A, Tucci A, Capanna R. Denosumab in advanced/unresectable aneurysmal bone cysts: a case series. J Bone Joint Surg Am. 2014; 96(11): e93.
- Enneking WF. Musculoskeletal tumor surgery. New York: Churchill Livingstone; 1983.

- Bode-Lesniewska B, Frischknecht F, Frigieri G. Adamantinoma-like Ewing's sarcoma of the tibia: a case report and review of the literature. Am J Surg Pathol. 2000; 24(6): 828-33.
- 10.Flanagan AM, Horton MA, MacSweeney F. Adamantinoma of long bones--a histopathological and immunohistochemical study. J Pathol. 1989; 157(1): 61-68.
- 11.Miyake R, Hirabayashi Y, Matsushita Y, Hashimoto N, Suzuki K, Yoshida A. Recurrent osteofibrous dysplasia-like adamantinoma of the tibia: a case report and review of the literature. J Med Case Rep. 2015; 9: 229.
- 12.Kutty RK, Kilpatrick SE, Wang H. Adamantinoma-like Ewing sarcoma: genomic confirmation in a pediatric patient. Diagn Pathol. 2012; 7: 157.
- 13.Nakano H, Honoki K, Iwasa T. Osteofibrous dysplasia-like adamantinoma of the tibia: a case report and review of the literature. Oncol Lett. 2012; 3(1): 188-92.
- 14.Yip KT, Chan KW, Chang AR. Osteofibrous dysplasia-like adamantinoma of the long bones: report of two cases with immunohistochemical study, electron microscopic findings, and review of the literature. J Clin Pathol. 2003; 56(3): 238-42.
- 15.Li X, Xu W, Chen S, Zuo D, Wang Y. Adamantinoma-like Ewing's sarcoma/primitive neuroectodermal tumor of the ulna. J Hand Surg Am. 2009; 34(3): 507-11.
- 16.Nishio J, Althof PA, Bailey JM. BRD4-NUT carcinoma of the mediastinum in a pediatric patient: multidetector CT, MR imaging, and diffusion-weighted MR imaging findings. Pediatr Radiol. 2013; 43(8): e82-e87.
- Pandey T, Vyas S, Roy R, Sharma M, Ojha B.
  Osteofibrous dysplasia of tibia. J Clin Orthop Trauma. 2015; 6(2): 122-5.

- 18.Basu M, Mukhopadhyay S, Mukhopadhyay M, Das PK, Nag D, Jash D. Osteofibrous dysplasia of clavicle in a child: a case report with review of literature. J Orthop Surg (Hong Kong). 2013; 21(2): 246-8.
- 19.Chopra P, Hashmi MA, Singh PK. Osteofibrous dysplasia of the clavicle: a case report and review of the literature. J Orthop Surg (Hong Kong). 2013; 21(3): 389-92.
- 20.Yang L, Li X, Lin W. Adamantinoma-like Ewing sarcoma/primitive neuroectodermal tumor of the mandible: a clinicopathologic and molecular cytogenetic analysis of 2 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013; 115(3): e17-e23.