# Isolated enchondroma of the hand. Long-term follow-up after surgical treatment
## Case series

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## Abstract
**Introduction:** Isolated enchondromas are the most frequent tumors in the hand. Approximately 40% of them involve the limb, preferably the ulnar rays and the proximal phalanges. The risk of malignant transformation into chondrosarcoma is 1%, and recurrence is 2-15%. We describe our experience and results in a significant number of patients with long-term follow-up.
**Materials and Methods:** Retrospective, descriptive case series study. We selected patients with conformed diagnosis of isolated enchondroma in their hand, who were subject to tumor resection and curettage plus allograft from iliac bone or radial bone. After surgical treatment, we evaluated function with the Takigawa classification, radiographic results and frequency of complications or recurrences.
**Results:** We included 19 patients with average post-operative follow-up of 11 years. According to the Takigawa classification, function was excellent in 16 patients (84.2%) and good in 3 (15.7%). One patient showed superficial infection at the surgical incision level, with complete resolution. There were no recurrences.
**Conclusions:** In the patients we evaluated, functional and radiographic results were good after the surgery. Enchondromas pose the risk of pathological fracture, recurrence and, to a lesser extent, malign transformation; however, none of these occurred in these cases. We describe an option of surgical management which is simple and brings about good and excellent long-term results.

**Key words:** Isolated enchondroma; hand; post-operative function; complications.
**Level of evidence:** IV

**Encondroma solitario de la mano: seguimiento a largo plazo después del tratamiento quirúrgico. Serie de casos**

## Resumen
**Introducción:** Los encondromas solitarios son los tumores óseos más comunes de la mano. Aproximadamente el 40% compromete esta extremidad, con predilección por los rayos cubitales y las falanges proximales. El riesgo de transformación maligna a condrosarcoma es del 1% y el de recurrencia, del 2-15%. Se describen la experiencia y los resultados de un importante número de pacientes con un seguimiento a largo plazo.
**Materiales y Métodos:** Estudio descriptivo, retrospectivo, de tipo serie de casos. Se seleccionaron pacientes con diagnóstico confirmado de encondroma solitario de la mano, que fueron manejados mediante resección con curetaje del tumor más aplicación de injertos autólogos de cresta ilíaca o radio. Después del tratamiento quirúrgico, se evaluaron la función, mediante la clasificación de Takigawa, los resultados radiográficos y la frecuencia de complicaciones o recidivas.

Conflict of interests: The authors have reported none.

**Resultados:** Se incluyeron 19 pacientes con un seguimiento posoperatorio promedio de 11 años. De acuerdo con la clasificación de Takigawa, la función fue excelente en 16 pacientes (84,2%) y buena en 3 (15,7%). Un paciente presentó una infección del sitio operatorio incisional superficial, con resolución completa. No hubo recidivas.
**Conclusiones:** En los pacientes evaluados, los resultados funcionales y radiológicos fueron buenos después de la cirugía. Los encondromas plantean el riesgo de fracturas patológicas, recidiva y, en menor proporción, de transformación maligna; sin embargo, esto no ocurrió en ninguno de los casos. Se describe una opción de manejo quirúrgico, sencilla con buenos y excelentes resultados a largo plazo.

**Palabras clave:** Encondroma solitario; mano; función posoperatoria; complicaciones.
**Nivel de Evidencia:** IV

## Introduction

Enchondromas are well-differentiated cartilaginous benign tumors, and they are made up of mature hyaline cartilage. They affect patients with a wide range of ages, both children and adults.[1,2] Enchondromas are rare in flat bones, and they preferably involve small tubular hand bones (35%), as well as the femur, the humerus and the ribs (13% each).[3] They are more frequently seen in the proximal phalanges, followed by the middle phalanges, the metacarpal bones, the distal phalanges and, rarely, the carpal bones.[2,4,5] It is believed that they represent the remaining cartilage in the growth plate; therefore, they should stop growing at skeletal maturity.[6]

Enchondromas growth is slow and they do not cause symptoms until the erosion of the cortical bone causes pain, enlargement or a pathological fracture secondary to cortex thinning. Simple X-ray is the basic assessment parameter, where there is a circumscribed lesion in the middle of the metaphysis or the meta-diaphysis.[6] Together with CT scans it is possible to evaluate some mineralization patterns such as the combination of dotted-pattern radiopaque zones, rings and arches that result from the formation of radiolucent cartilage lobes surrounded by rings of radiopaque bone, and the dotted pattern results from the necrosis and consecutive dystrophic calcification in the central areas of the cartilage nodules; moreover, it is possible to see bone destruction and cortex indemnity.[6]

The risk of malignant transformation to chondrosarcoma is extremely low.[7] The causes of malignant transformation are still unknown, but there have been attempts to identify them with cytogenetic studies.[8] The possibility of recurrence after surgical treatment is 2 to 15%.[9,10] However, recurrence does not seem to affect healing rates and the post-operative range of motion.[11] Although for the surgical management of this condition there are several methods described, we recommend the one that we believe is associated with the best results—curettage of the lesion and filling of the secondary defect with autograft, allograft, bone substitutes and even bone cement.[12,13] With that said, the aim of this study is to describe our experience and results in patients with diagnosis of isolated enchondroma subject to surgery, and the patients' long-term follow-up.

## Materials and Methods

Retrospective, descriptive case series study. We included patients with diagnosis of hand isolated enchondroma treated with curettage of the tumoral lesion and management of the remaining defect with bone autograft. We excluded the patients we could not make contact with for follow-up and the ones whose data in medical histories were incomplete. Among the pre-operative variables, we collected data concerning the patients' laterality, the Takigawa classification[5] (location of the tumor: central, eccentric, combined, polycentric or giant features), whether or not there was pathological fracture, and the symptoms previous to the surgery, which were the reasons for consultation. All the patients were treated with tumor resection plus thorough curettage of the lesion and the use of bone autograft to fill the defect in the zone of resection, and they were operated on by the same surgeon. We recorded the type of approach and the sources of autograft.

We assessed the amount of time it took the patients to come back to their day-to-day activities after the surgery, the patients' post--operative functional ability, and the post-operative radiographic characteristics; among them, the restoration of the cortical bone continuity. Function was evaluated at last follow-up using the four Takigawa criteria:[5] whether or not the looks were acceptable, the patients' active range of motion (80% or more in comparison with the contralateral side), their grip strength (80% or more in comparison with the contralateral side), and the radiographic evidence of healing with no shortening, deformity, osteoarthritis or tumor recurrence. The results were classified into excellent (4 criteria), good (3 criteria), regular (2 criteria) and poor (one criterion or none). To assess the radiographic results we used the Tordai classification (1990), in which the group 1 includes normal cortical and spongy bone, or bone with a <3mm-diameter bone defect, the group 2, a 4 to 10-mm diameter bone defect with no clear recurrence, and the group 3, >10 mm-diameter defects with characteristics of enchondroma. Finally, we established the frequency of recurrence and complications.

Data are presented as absolute and relative frequencies for qualitative variables and measures of central tendency (average), and as (standard deviation) dispersion for quantitative variables. This study was approved by the


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Fundación Universitaria Sanitas' Ethics and Research Committee.

Out of 25 patients, we included 19 (6 males [32%] and 12 females [68%]), who averaged 36 years old and met the inclusion criteria; the diagnosis of enchondroma was confirmed by histological studies in all the cases.

### *Pre-operative evaluation*

Among the pre-operative variables, 52.6% of the isolated enchondromas were in the patients' right hand, whereas 47.4% of them were in their left hand. The tumor was more frequently located in their proximal phalanges (47.3%), their middle phalanges (31.5%), their distal phalanges (15.7%), and their metacarpal bones (5.2%) (Figure 1). The anatomic location according to the Takigawa classification is shown in Figure 2. Seven patients (36.8%) had pathological fracture. The pre-operative symptoms which brought about consultation were: enlargement without fracture (1 patient [10.6%]), pain or limitation to mobility (1 patient [10.6%]), pain without pathological fracture (8 cases [42%]), and pain due to fracture (7 cases [36.8%]).

### *Surgical management*

The approaches were the dorsal one (1 patient), the dorsolateral one (2 patients), and the rest of the patients received lateral approach, what varied as the surgeon's preferences did (Figure 3). All the patients were subject to lesion curettage; in two cases we used radius autograft and, in the remaining patients, iliac bone autograft.

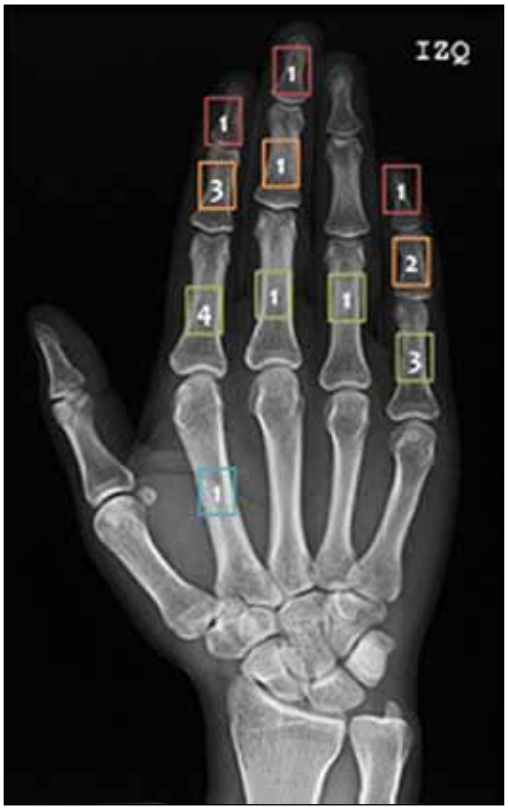


**Figure 1.** Enchondroma distribution in the hand according to follow-up.

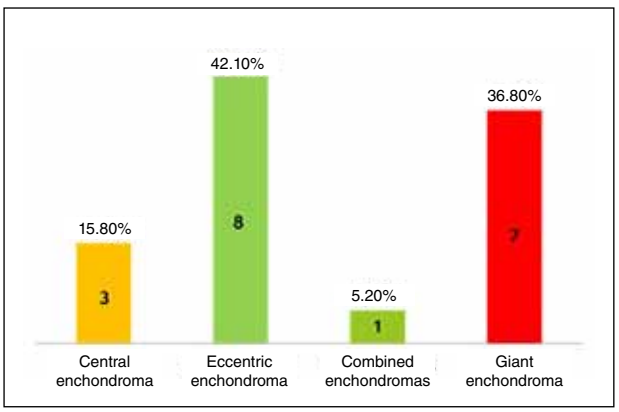


**Figure 2.** Distribution of enchondroma according to the Takigawa classification.

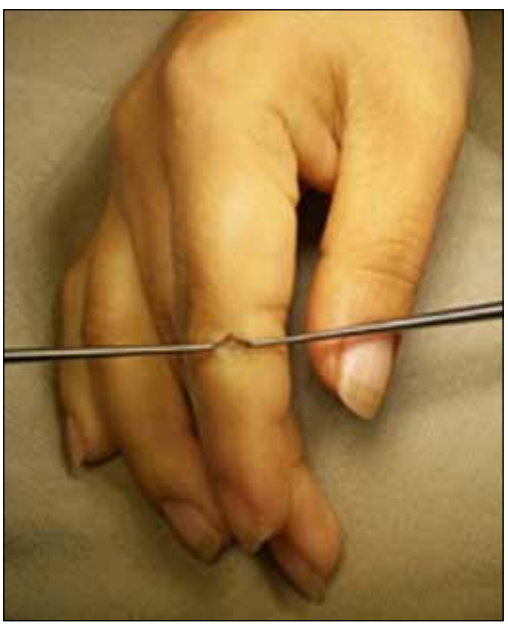

**Figure 3.** Woman with right-hand second-finger middle-phalanx enchondroma who is subject to 1cm-lateral approach with curettage and graft.

## Results

The average time it took the patients to come back to their day-to-day activities after the surgery was six weeks (ranging from 2 to 8). Among the post-operative radiographic characteristics, we checked the restoration of cortical bone continuity in all the patients.

The patients' average follow-up was 11 years (ranging from 2 to 16; standard deviation= 9.9). As regards function, according to the Takiwara's criteria, results were excellent and good (Figure 4). In the final X-rays, according to the Tordai classification (Figure 5), 16 (84.2%) patients belonged to the group 1 and three (15.7%), to the group 2. Only one of the seven patients with pathological fracture required osteosynthesis due to faulted rotation in the index finger's proximal phalanx, plus the enchondroma treatment; it was not necessary to remove the osteosynthesis material before 10-year follow up; the remaining six cases were managed with tumor resection, graft and immobilization with adequate bone consolidation. Only one patient suffered a superficial infection, which was treated with antibiotics for 10 days, with favorable resolution. Figures 6 and 7 show two patients' lesion progression and radiographic results.

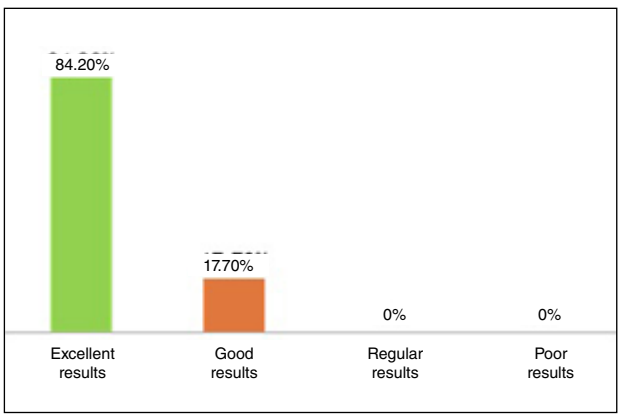


**Figure 4.** Evaluation of the results according to the Takigawa classification.

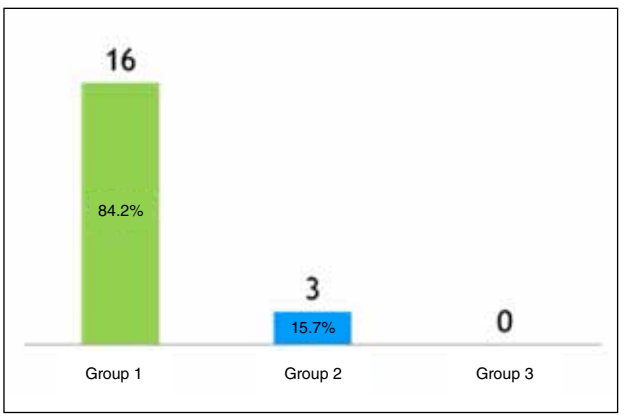


**Figure 5.** Evaluation of post-operative functional results according to the Tordai classification.

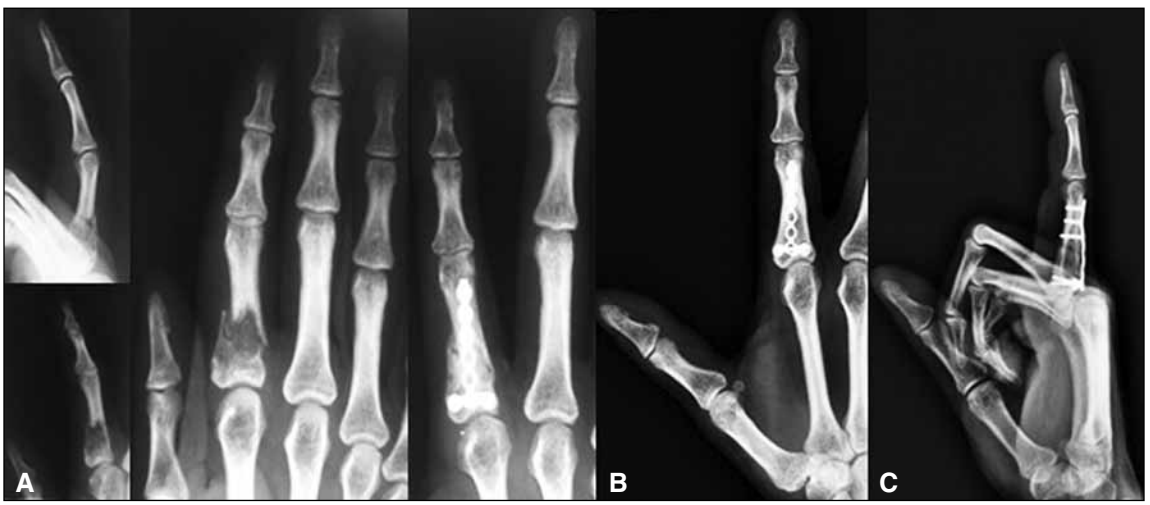


**Figure 6.** Case 1. **A.** Forty eight year-old patient with eccentric enchondroma and pathological fracture with inadequate rotation. **B.** Treatment: tumor resection with curettage plus iliac bone graft plus fixation with plate. **C.** Good results, no recurrence, with total mobility and complete restoration of cortical bone continuity.


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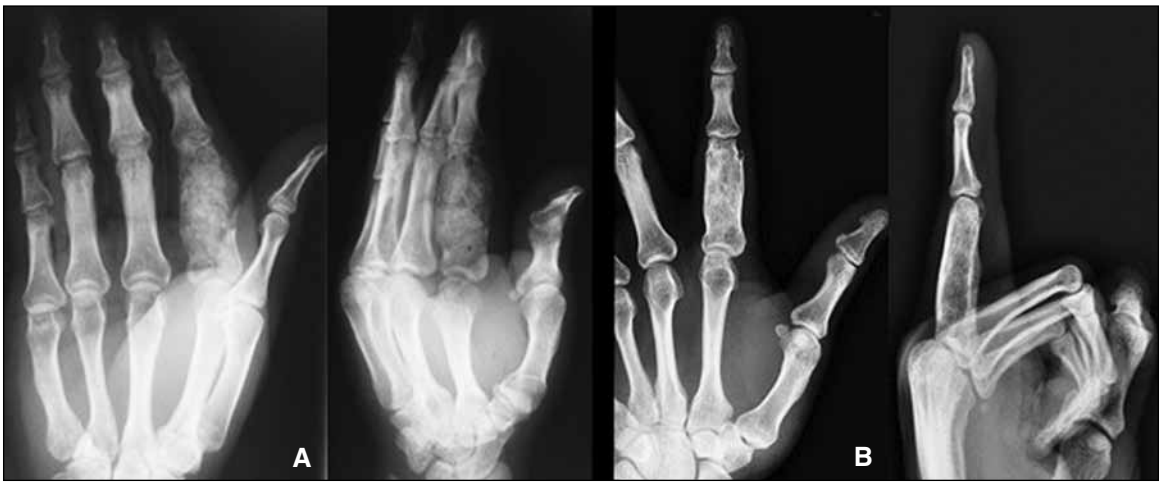


**Figure 7.** Case 2. **A.** Forty eight year-old patient with giant enchondroma in right-hand index-finger proximal-phalanx, with neither pathological fracture nor flexion impairment. **B.** Treatment: curettage plus iliac bone graft, with good results, no recurrence, total mobility and complete restoration of cortical bone continuity.

## Discussion

In this study, we describe the medical, radiographic and functional data we collected in patients with hand enchondroma who were subject to surgical treatment and had long-term follow-up. Our patients' age and sex are consistent with those reported by other authors.[14-16] Moreover, the location of enchondroma is the same as that described worldwide, i.e. proximal phalanges were the most frequently affected region, followed by middle phalanges. There was only one subtle difference; distal phalanges were more frequently affected than metacarpal bones in our study. [2,4,5]

Moreover, we found that the frequency of pathological fracture was similar to that reported by the specialized literature,[15] one third of the patients, figure that can be compared to the 36.8% in our patients. As it has already been stated, enchondroma treatment is based on the lesion curettage and the filling of the bone defect with different methods. In most patients in this series, we carried out an incision no longer than 1 cm and used autograft, mainly from iliac bone. Given the size of the lesions, we believe that the defect should be corrected using more autograft than it is necessary; therefore, distal radius graft not always can provide us with enough material to fill completely the defect, what is considered to be an essential step for final results and always should be verified with intra-operative X-rays.[2]

We preferred to use autograft, because it gives adequate osteogenesis, osteoinduction and osteoconduction to the procedure; moreover, it is rapidly incorporated, with a consolidation average time of 51 days,[16] what is co-related to the average time it took our patients to come back to their day-to-day activities in our series (6 weeks).

Functional and radiographic results in this study were excellent and good in all the patients, what was proved by other authors who also got good and excellent results using the same surgical technique.[14,17] Gaulke[18] evaluated 21 patients with an average follow-up of nine years (ranging from 2 to 18); in seven patients, the bone defects remained. There were three recurrences; two required a new surgery, and none of the patients with or without recurrence had medical symptoms or functional loss.

Kuur[19] reports results in 21 patients with isolated enchondroma in their fingers who were subject to surgical treatment; 15 of them received curettage and graft, and the results were pain disappearance and finger normal function at 4.5-year follow-up (ranging from 1 to 9). Figl and Leixnering[9] operated on 29 patients; 26 were treated with curettage and iliac bone graft. Twenty-seven patients had an average 47-month follow-up, and results were excellent in 25 cases (84.2%) and good in two (17.7%); moreover, they did not register complaints about the donor site in the patients' iliac bone, and neither did we.

It is worth mentioning that, in previous studies, with sample sizes similar to ours, results are excellent and good with the surgical technique they use, but they report recurrences and need for a new surgery.

Among the series we present, there are differences in follow-up; ours is the series with the longest average follow-up.

Although it is true that there have been studies showing good results with the use of bone substitutes,[12,13] this same studies report more complications; among them, the loss of digital flexion at the inter-phalangeal joint level in up to 50% of the cases,[20] as well as complications related to soft tissues when bone cement is used, what has to do with

cement leaking.[12] It is important to bear in mind that autograft taking implies morbidity at the level of the donor site, such as bleeding, hematoma, infection, and iliac bone trouble, especially among women, drawbacks that we did not find in our series because we were cautious enough so as to carry out small approaches.

Among the limitations of this study and due to its retrospective character, we found lack of follow-up in five patients, as well as poor data in medical histories, what restricted the amount of information and, consequently, made us exclude patients. It is worth highlighting that all the patients were operated on by the same surgeon, and we detected neither recurrence nor malign transformation throughout follow-up, what coincides with the published articles, what report recurrence rates of 2-15%.[9,10]

## Conclusions

We present functional and radiographic long-term results from an option of surgical management in a series of patients with isolated enchondroma in their hand. Curettage and allograft as surgical treatment for this condition brought about satisfactory results with minimal complications and no recurrence. It is necessary to carry out cohort studies or interventional studies to confirm these results.

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