SCIENTIFIC ARTICLE

Impact of Patient and Tumor Characteristics on Range of Motion and Recurrence Following Treatment of Enchondromas of the Hand

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Purpose The approach to the treatment of enchondromas of the hand is varied, and there is no clear consensus on graft source, fixation, or need for intraoperative adjuvant therapy. We reviewed a cohort of patients who underwent curettage and bone grafting with cancellous allograft chips without internal fixation or adjuvant therapy and reported on postoperative range of motion (ROM) and recurrence rates.

Methods We performed a retrospective review of patients who underwent surgical treatment for hand enchondroma over a 23-year period. We collected information on demographics and presenting enchondroma characteristics, including Takigawa classification and presence of pathologic fracture or associated syndromes. Patients were treated with open biopsy with curettage and grafting with cancellous allograft chips. Postoperative ROM, complications, and recurrences were recorded.

Results Our series included 111 enchondromas in 104 patients. Seventeen of 104 patients (16%) had a diagnosis of Ollier disease. Average length of follow-up was 3.1 years. Eighty-one percent of patients achieved full ROM. Treatment of patients who presented with preoperative pathologic fracture resulted in a greater frequency of reduced postoperative ROM at 28% (9/32) compared to 15% (11/72) of those patients who did not present with preoperative pathologic fracture. Local recurrence developed in 5 of 50 (10%) patients with a minimum of 2 years of follow-up. Local recurrence occurred at higher-than-average rates in patients with giant form Takigawa classification (43%, 3/7) and Ollier disease (23%, 3/13).

Conclusions Treatment of enchondromas with biopsy, curettage, and allograft results in full ROM in 81% of patients. Patients with preoperative pathologic fracture should be advised of a greater risk of postoperative extension deficit. Recurrence remains rare and is associated with syndromic presentation and giant form lesions. *(J Hand Surg Am. 2021;■(■):1.e1-e7. Copyright © 2021 by the American Society for Surgery of the Hand. Published by Elsevier Inc. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).)*

Type of Study/level of evidence Therapeutic IV.

Key words Allograft, approach, enchondroma, Ollier.

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ENCHONDROMAS ARE BENIGN, slow-growing cartilage tumors that are commonly found in the hand. They are the most common primary osseous lesions in the hand, constituting roughly 70% of the 631 hand tumors captured in the 30-year Hamburg Bone Tumor Registry.[1] These tumors are most frequently noted in the third and fourth decades of life.[2,3] The proximal phalanges are the most commonly affected bones in the hand, and the small finger is the most commonly affected digit.[2,3] Enchondromas of the hand present with pathologic fracture in 40% to 60% of patients with this diagnosis.[4–6]

Approach to the treatment of enchondroma is varied, and there is no clear consensus on graft source, fixation, or approach. Techniques described in the literature range from simple curettage to the use of cemented hardware with Kirschner wire intramedullary strut placement to allow for immediate postoperative range of motion (ROM).[5,7] Bachoura[5] presented a systematic literature review recommending curettage alone for the treatment of enchondroma; however, there are no reports on ROM outcomes with this technique. Additionally, the use of a dorsal, as opposed to a lateral or midaxial, approach to the digit has not been well studied regarding the effect on final ROM. While Lin et al[8] argue for the use of a lateral approach in order to minimize irritation to the extensor mechanism, there is no direct comparison of these 2 techniques. Finally, there is no consensus on whether surgical technique should be modified in the presence of pathologic fracture on presentation, though prior literature suggests that treatment prior to fracture healing may result in worse deformity when immediate curettage and grafting is performed.[9]

While the use of adjuvant treatments is commonly described in the treatment of enchondromas of the axial and appendicular skeleton,[10–12] the use of adjuvant therapy in the hand is more controversial.[5,13] Numerous adjuncts including phenol,[14] dehydrated alcohol,[15] CO_2 laser,[16] and polymethylmethacrylate cement[7] have been described for the purposes of margin expansion, but they carry risks including skin burns, nerve injury, and postoperative fracture. Given the overall low recurrence rate of enchondromas, the utility of these modalities remains in question.

The purpose of this study was to characterize the effects of patient and tumor characteristics on finger ROM postoperative treatment of hand enchondromas using the senior author's (E.A.A.) surgical technique. The secondary aim of this study was to characterize the impact of lesion characteristics on local recurrence.

MATERIALS AND METHODS

We performed a retrospective review of patients who underwent primary surgical treatment for a hand enchondroma by a dual-trained hand surgery and musculoskeletal oncology surgeon over a 23-year period. The study protocol conformed to the ethical guidelines of the 1975 Declaration of Helsinki and was approved by the appropriate institutional review committee.

We collected demographic information on age, sex, laterality, associated syndromic conditions, as well as information on the affected digit, affected bone, and whether fracture was present on presentation. Presenting anteroposterior and lateral radiographs were reviewed to determine Takigawa classification, which described the radiographic classification of enchondromas as either central, eccentric, associated with multiple isolated lesions occurring in the same bone, polycentric, or giant form.[2] One patient had insufficient presenting radiographs for assigning a Takigawa classification. Length of postoperative follow-up, complications, recurrence, reoperation, and final ROM were collected. Recurrence was defined by the identification of new lucent areas on postoperative imaging that were not present on intraoperative or on immediate postoperative films. Full finger ROM was defined as the ability to achieve full extension at the metacarpophalangeal (MCP), proximal interphalangeal, and distal interphalangeal joints as well as a distance to palmar crease of zero centimeters. Any deviation from this arc was measured with a handheld goniometer, and deviations from ROM normative values were measured and recorded by the senior treating hand surgeon (E.A.A.).[17] As such, regarding active flexion, we described any composite flexion measurement less than 270° as a motion deficit and subtracted total flexion from this value to calculate the deficit.

Complications were categorized as surgical versus oncological. Additionally, healing was graded according to the Tordai classification based on final anteroposterior and lateral radiographs, which characterized grade of healing into 3 categories based on bony defects and presence of recurrence.[5] We used a minimum follow-up of 2 years to calculate recurrence rates in line with Takigawa's prior report.[2] Patients for whom follow-up radiographs were available

through this period were included in the recurrence analysis, and follow-up radiographs were graded by 2 fellowship-trained orthopedic surgeons (L.E.W. and A.B.C.). Recurrence was determined by the development of local lucent areas consistent with enchondroma and representing a change from interval postoperative imaging. The determination of phalangeal bone distribution and comparison of postoperative ROM according to the bone affected excluded thumb lesions, given differences in joint composition and baseline ROM that prevented comparison between the thumb and fingers.

Our surgical technique was consistent across cases with respect to fixation and graft use. We used open biopsy with curettage and bone defect grafting with freeze-dried, cancellous allograft bone chips. Surgical technique was not altered if pathologic fracture was present; however, surgery was not performed until fracture healing was completed, secondary to the concern for worse deformity when immediate curettage and grafting was performed.[9] Fractures were considered healed on the basis of the absence of a fracture line on radiographic examination in addition to the absence of pain and achievement of improved finger ROM compared to the fractured state. Surgical approach was either dorsal or lateral and was dictated by the location of the lesion with the thinnest cortices. Periosteum was preserved and peeled from the bone, allowing creation of a cortical window with a scalpel or rongeur that equaled 2/3 the length of the longest longitudinal dimension of the tumor. Enchondromas were curetted thoroughly, and the adequacy of curettage was confirmed on radiographs, imaging the curette reaching the most distant margins of the lesion in each dimension. Allograft bone was then used to fill the defect. Particular attention was paid to morcellation of cancellous chips into fine pieces using the combination of a rongeur and bone tamp so that the defect could be packed tightly without a residual void. Completeness of excision was determined by visual review of the postcurettage space as well as intraoperative fluoroscopic x-rays. A periosteal elevator was used to pack small void locations. The periosteum was closed with 4-0 vicryl sutures. Tendons incised longitudinally for the dorsal approach were subsequently repaired using 4-0 vicryl.

Given our sample size, we chose to analyze our data using descriptive statistics, with central tendency being characterized by mean values and variability being characterized by standard deviation. We included 95% confidence interval (CI) calculations to further describe our analysis.

RESULTS

One hundred and eleven enchondromas in 104 patients were available for study between January 1996 and December 2019. Sixty-four percent of patients (67/104) were women, and 55% of patients (58/104) presented with right-sided lesions. The average patient age was 34.9 (SD: 16.0 years). Seventeen of 104 patients (16%) had a diagnosis of Ollier disease. Average length of follow-up was 3.1 years (SD: 3.6 years).

The most commonly involved digit was the small finger, which was affected in 27% of lesions (30/111). The most commonly involved bone was the proximal phalanx, which was affected in 43% of lesions (43/100) (Fig. 1). Almost half of lesions were graded as Takigawa A (Table 1). Thirty-one percent of patients presented with a pathologic fracture.

Full postoperative ROM was achieved in 82% (85/104) of patients. Range of motion deficits were noted at the MCP, proximal interphalangeal, and distal interphalangeal joints in 3% (3/104), 9% (9/104), and 13% (13/104) of patients, respectively. Full postoperative ROM was achieved in 88% (46/52) of patients who underwent the lateral approach in comparison to 76% (38/51) of patients who underwent the dorsal approach. Of the 25 joints with noted

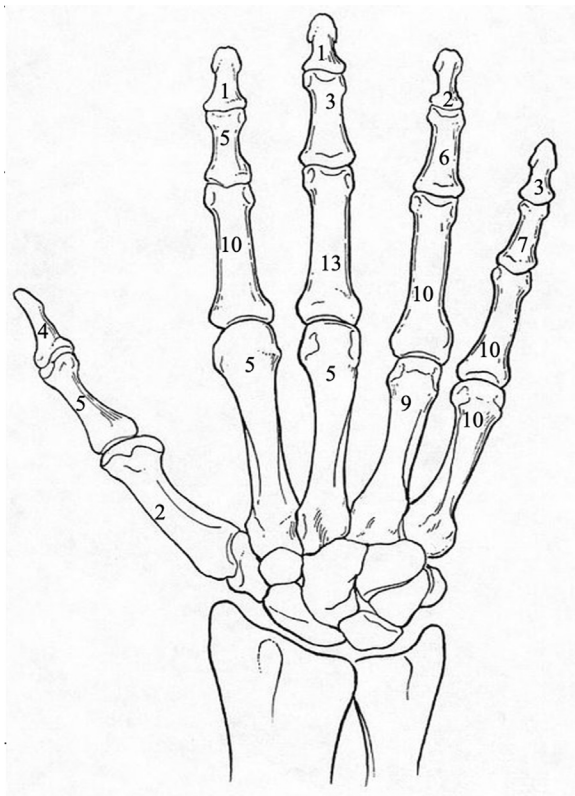

FIGURE 1: Indicates distribution of lesions with the number of lesions in each bone among the population indicated by the numbers indicated within in the given bone in the diagram.

TABLE 1. Takigawa Grading of Lesions

Takigawa Grading	Number of Lesions (%)
A: Central	52 (47)
B: Eccentric	16 (15)
C: Associated	6 (5)
D: Polycentric	22 (20)
E: Giant form	14 (13)

ROM deficits, the average deficit for the dorsal and lateral approaches was 15° (CI: 8° to 21°) and 18° (CI: 6° to 29°), respectively (Table 2). Location of enchondromas in the middle phalanx was most frequently associated with ROM deficits, which occurred in 37% (7/19) of cases of middle phalanx enchondroma (Table 3). There was a greater frequency in postoperative ROM deficits among patients who presented with pathologic fracture in contrast to those who did not; 28% (9/32) of patients who presented with pathologic fracture demonstrated reduced postoperative ROM in comparison to 15% (11/72) of those who presented without pathologic fracture (Table 4). Tordai Grade 1 healing was achieved in 80% of cases. There were no postoperative fractures.

Of patients with greater than 2 years of follow-up, local radiographic recurrence was noted in 5 of 50 (10%) patients after treatment with our surgical technique. Average follow-up for this group was 5.8 years (SD: 3.8 years). Notably, local recurrence was associated with Ollier disease, with 3/13 (23%; CI: 8%−51%) patients presenting with this diagnosis developing subsequent local recurrence. Similarly, Takigawa E (giant form) radiographic classification on presentation was associated with recurrence, which occurred in 3/7 (43%; CI: 16%−75%) of giant form cases (Table 5). No patients who developed local recurrence initially presented with pathologic fracture. Of the instances of local recurrence, 1 patient underwent subsequent surgical intervention, and 4 stabilized clinically and did not progress to reoperation.

Surgical complications were reported in 3 of 110 patients (3%). Reported complications included 3 cases of wound sensitivity beyond the expected postoperative period that required additional follow-up to monitor to resolution.

DISCUSSION

We studied 111 enchondromas in 104 patients presenting to the care of a dual-fellowship-trained upper extremity and musculoskeletal oncology surgeon. Our sample was constituted predominantly of patients in their third and fourth decades of life, which was similar to previously published data.[2,3]

Our series demonstrated a relatively low rate of pathologic fracture on presentation (31%), in contrast to studies by Sassoon et al[4] and Bachoura et al[6] who cited rates of 40% and 54%, respectively.

Our final postoperative ROM compared favorably to prior reports. Full ROM was achieved by 82% of patients in this study, compared to 67% and 46% in series presented by Sassoon et al[4] and Bickels et al,[7] respectively. In the series reported by Bachoura et al,[6] stiffness was observed in 3 of 24 patients (13%), including a proximal interphalangeal joint contracture, a MCP joint extension contracture requiring dorsal MCP joint capsule release, and a swan neck deformity. In our series, ROM deficits were found to be more frequent with pathologic fracture on presentation as well as with middle phalanx lesions, which we believe to be new and valuable information. As the characteristic ROM deficit in our series is that of extensor lag, we hypothesize that this is likely related to extensor mechanism adhesions with involvement of the central slip or triangular ligament rather than adhesions typical of the proximal phalanx level being secondary to adhesions within the flexor tendon fibro-osseous tunnel. We hypothesize that ROM deficits in patients who present with pathologic fracture may be secondary to extended immobilization to allow for preoperative fracture healing. This consideration has been discussed in prior literature; however, we have favored fracture healing prior to intervention in order to prevent further angulation with curettage, because we do not use internal fixation with the technique described.[9,18] Given these findings, a comparison of ROM after pathologic fracture using a technique that employs instrumentation with immediate treatment versus our preferred methods would be of interest for future study.

Finally, it is notable that in our series, those patients who were treated with dorsal and lateral approaches achieved similar mean postoperative ROM. Some authors have suggested that the lateral approach may achieve superior ROM by avoiding the extensor mechanism.[19] While the results of our series identified that full ROM was achieved in 88% after a lateral approach in comparison to 76% of patients after the dorsal approach, motion deficits, when present, were similar between groups at 18° in comparison to 15°, respectively. Though not proven here, we believe that choosing the approach that allows for the most direct access to the lesion enhances the

TABLE 2. Postoperative Range of Motion

	Total Cohort (n = 103*)	Dorsal Approach (n = 51)	Lateral Approach (n = 52)
Number of patients with a deficit in total arc	19	13	6
Average total arc deficit	16° (11°)	15° (11°)	18° (11°)

*One patient for whom approach was not specified in operative report.
SD is in parentheses.

TABLE 3. Range of Motion Deficits by Location of Lesion

	Number of Digits With ROM Deficit	Average ROM Deficit (Composite Arc)
Metacarpal	3/30 (10%; CI: 3%–26%)	10° (0°)
Proximal phalanx	9/44 (20%; CI: 11%–35%)	14° (13°)
Middle phalanx	7/19 (37%; CI: 19%–59%)	18° (12°)
Distal phalanx	1/11 (9%; CI: <1%–51%)	10° (0°)

SD is in parentheses.

adequacy of our curettage and contributes to low rates of local recurrence without the requirement of adjuvant therapy. We recommend the approach to the finger that will best access the most involved region of bone with the thinnest cortices.

While a variety of grafting and fixation methods are available, we believe that a simple approach to the treatment of enchondromas with minimal use of hardware is critical to our cohort's achievement of optimal postoperative motion and prevention of adhesions. Multiple studies have shown no difference in healing with the use of autograft, allograft, or no graft at all.[4,5,20–22] While some authors have advocated for various augmentation strategies for fixation, including cemented hardware with internal Kirschner wires[7] and injection of calcium sulfate,[8] allograft has been shown to be appropriate for filling bony defects with good incorporation without the use of additional fixation or substitutes. In a series of 102 enchondromas treated by grafting with autogenous, allogenous, or synthetic bone, Sassoon et al[4] reported 78% Tordai Grade 1 healing on final radiographic follow-up. Our study similarly detected Tordai Grade 1 healing in 80% of cases, and no postoperative fractures were reported. These results suggest that, particularly in the absence of pathologic fracture, hardware is not needed to achieve healing in the treatment of enchondromas, and this technique further exposes patients to risk of stiffness with the possible need for tenolysis. Notably, we had no cases of infection with use of allograft and were able to safely begin early postoperative ROM without additional fixation, allowing for sufficient therapy to prevent adhesion formation and postoperative stiffness necessitating tenolysis.

We detected a recurrence rate of 10% within our sample followed for a minimum of 2 years. Though this number is slightly greater than that previously reported by Sassoon et al,[4] we required 2 years of follow-up for inclusion in recurrence analysis, whereas in the aforementioned study, minimum follow-up was 1 month. We believe this to be a comparative strength of our work. These recurrences notably occurred overwhelmingly among patients who presented with syndromic presentation or giant form lesions. Three of the 5 recurrences presented with a concomitant diagnosis of Ollier disease. Similarly, in our series, giant form lesions demonstrated higher frequency of local recurrence. While this observation is in line with Takigawa's description, which notes that probable malignancy is suspected in some giant form lesions secondary to rapid growth, this is not well-established in the literature from a quantitative perspective.[2]

Though numerous agents, including phenol,[14] dehydrated alcohol,[15] CO_2 laser,[16] and polymethyl methacrylate cement[7] have been described in case series, we do not believe that these are required for the treatment of these benign, slow-growing tumors, and they may expose patients to increased risk of complication. Risk of Complex Regional Pain Syndrome secondary to nerve irritations following the

TABLE 4. Range of Motion Deficits by Presentation With Pathologic Fracture

	Number of Digits With Deficit in ROM	Average Composite ROM Deficit
No fracture on presentation	11/72 (16%; CI: 9%−26%)	16° (12°)
Pathologic fracture on presentation	9/32 (28%; CI: 15%−46%)	13° (10°)

SD is in parentheses.

TABLE 5. Recurrence by Takigawa Classification

Takigawa Grading	Recurrences
A: Central	2/24 (6%; CI: 1%−27%)
B: Eccentric	0/5 (0%; CI: 0%−49%)
C: Associated	0/2 (0%; CI: 0%−71%)
D: Polycentric	0/12 (0%; CI: 0%−28%)
E: Giant form	3/7 (21%; CI: 16%−75%)

use of such adjuvants is well documented and, given the low recurrence risk in a solitary enchondroma and the benign nature of these lesions, risk of complication from adjuvants should be strongly considered prior to their use.[10,12] Of note, there are certain cases that present a greater risk of recurrence. As we have noted, given a greater risk of recurrence in patients who present with giant form lesions and Ollier disease, there may be a role for more aggressive treatment of lesions with these radiographic characteristics, though further studies are needed on this topic.

Several limitations are inherent in our study, which are typical of a retrospective review. Though one of the largest series of patients with enchondromas, our sample is relatively small, which limits our ability to perform statistical comparisons or subgroup analyses. Our ability to report on pain or functional disability is limited to that documented in the medical record. Completeness of excision is determined in our study by intraoperative inspection with both direct visualization as well as fluoroscopy. There is always a possibility of residual disease with intralesional resections, and the use of an endoscope as described by Dietz et al,[23] may aid in identification of incomplete excision; however, this is not a standard procedure in our practice and further research is required to establish superiority of this technique. Our series might be affected by a referral bias, given the subspecialty training and practice of the senior author. It likely has a greater proportion of patients with syndromic presentation than might be present in the general population. We believe that this likely biases our recurrence rates toward higher numbers than would otherwise be present in the community. Additionally, our postoperative follow-up is not uniform. Though some patients have limited follow-up, we do not believe that this has a substantial impact on our postoperative ROM findings, as none of the patients with the minimum follow-up reporting has more than a 10° motion deficit. We additionally require a 2-year minimum follow-up for evaluation of recurrence, which is in line with prior publications. Similarly, as a retrospective study conducted over a long period of time, complications may be underestimated secondary to this information being excluded from the chart or if patients are lost to follow-up.

In conclusion, we report on 104 patients with 111 enchondromas treated with open biopsy with curettage and allograft over the course of 23 years with a well-defined treatment algorithm. Following this treatment of enchondromas in the hand, patients who initially presented with preoperative pathologic fractures demonstrated worse postoperative ROM, possibly secondary to preoperative immobilization. Both recurrence and malignant transformation were uncommon, though perhaps greater than previously noted in studies with limited follow-up. Recurrence occurred most frequently in patients presenting with Ollier disease and those with giant form radiographic appearance on presentation. Patients presenting with these characteristics should be counseled regarding recurrence risk, and there may be a role for more aggressive treatment with consideration for adjuvant therapy in these cases.

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