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

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Abstract

Purpose: The approach to the treatment of enchondromas of the hand is varied and there is no clear consensus on graft source, fixation, nor need for intraoperative adjuvant therapy. We reviewed a cohort of patients who underwent curettage and bone grafting with cancellous allograft chips without internal fixation nor adjuvant therapy and reported on post-operative range of motion 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. Post-operative range of motion, 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 range of motion. Treatment of patients who presented with preoperative pathologic fracture resulted in a greater frequency of reduced post-operative range of motion at 28% (9/32) in comparison to 15% (11/72) of those patients who did not present with preoperative pathologic fracture. Local recurrence developed in five of fifty (10%) patients with a minimum of five 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 full range of motion in 81% of patients. Patients with preoperative pathologic fracture should be advised a greater risk of post-operative extension deficit. Recurrence remains rare and is associated with syndromic presentation and giant form lesions.

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Level of Evidence: Level IV

Type of Study: Therapeutic

Introduction

Enchondromas are benign, slow growing cartilage tumors, which 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 Tumour 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 post-operative range of motion[5,7]. Bachoura presented a systematic review recommending curettage alone for the treatment of enchondroma;[5] however, there are no reports on range of motion outcomes with this technique. Additionally, the use of a dorsal as opposed to a lateral or mid-axial approach to the digit, has not been well studied regarding the effect on final range of motion. While, Lin *et al.* argue for the use of a lateral approach in order to minimize irritation to the extensor mechanism, there is no direct comparison of these two techniques.[8] Finally, there is no consensus guidance 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 worsened 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 carry risks including skin burns, nerve injury and post-operative 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 range of motion after operative treatment of hand enchondromas utilizing the senior author's (EA) surgical technique. The secondary aim of this study was to characterize the impact of lesion characteristics on to 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 describes 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 Takigawa classification. Post-operative length of follow-up, complications, recurrence, reoperation, and final range of motion were collected. Recurrence was defined by the identification of new lucenies on post-operative imaging that were not present on intraoperative nor immediate post-operative films. Full finger range of motion was defined as the ability to achieve full extension at the MCP, PIP, and DIP 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 range of motion normative values[17] were measured and recorded by the senior treating hand surgeon (EA). As such with regard to active flexion, we describe any composite flexion measurement less than 270° as a motion deficit and subtract total flexion from this value to calculate the deficit.

Complications were categorized as surgical versus oncologic complication. Additionally, healing was graded according to Tordai classification based on final anteroposterior and lateral radiographs, which characterizes grade of healing into three 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 two fellowship-trained orthopaedic surgeons. Recurrence was determined by the development of local lucent areas consistent with enchondroma and representing a change from interval post-operative imaging. The determination of phalangeal bone distribution and comparison of post-operative range of motion according to bone affected excluded thumb lesions, given differences in joint composition and baseline range of motion not allowing for comparison between the thumb and fingers.

Our surgical technique was consistent across cases with respect to fixation and graft use. We utilized open biopsy with curettage and bone defect grafting with freeze-dried, cancellous allograft 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 worsened deformity when immediate curettage and grafting is performed.[9] Fractures were considered healed on the basis of absence of fracture line on radiographic examination in addition to the absence of pain and achievement of improved finger ranger of motion 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 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 residual void. Completeness of excision was determined by visual review of the post-curettage space as well as intra-operative fluoroscopic x-rays. A periosteal elevator was used to pack small void locations. 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 with mean values and variability being characterized by standard deviation. We included 95% confidence interval calculations to further describe our analysis.

Results

One hundred and eleven enchondromas in 104 patients were available for study between January of 1996 until December of 2019. Sixty-four percent of patients (67/104) were female and 64% 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 common digit affected was the small finger, which was affected in 27% of lesions (30/111). The most common bone involved was the proximal phalanx, which was affected in 43% of lesions (43/100) (Figure 1). Almost half of lesions were graded as Takigawa A (Table 1). Thirty-one percent of patients presented with a pathologic fracture.

Full post-operative ROM was achieved in 82% (85/104) of patients. Range of motion deficits were noted at the metacarpophalangeal (MCP), proximal interphalangeal (PIP) and distal interphalangeal (DIP) joints in 3% (3/104), 9% (9/104) and 13% (13/104) of patients, respectively. Full post-operative 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 twenty-five joints with noted range of motion deficits, the average deficit for the dorsal and lateral approaches were 15° (CI: 8° to 21°) and 18° (CI: 6° to 29°), respectively (Table 2). Location of enchondromas at the middle phalanx was most frequently associated with range of motion deficits, which occurred in 37% (7/19) of cases of middle phalanx enchondroma (Table 3). There was a greater frequency in post-operative range of motion deficits among patients who presented with pathologic fracture in contrast to those who did not with a 28% (9/32) of patients who presented with pathologic fracture demonstrating reduced post-operative range of motion in comparison to 15% (11/72) of those who presented without pathologic fracture. Tordai Grade 1 healing was achieved in 80% of cases. There were no post-operative fractures.

Of patients with greater than two years of follow-up, local radiographic recurrence was noted in five of fifty (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 development of recurrence, and recurrence occurred in 3/7 (43%; CI: 16-75%) of giant form cases (Table 5). No patients who developed local recurrence demonstrated initial presentation with pathologic fracture. Of the instances of local recurrence, one patient underwent subsequent surgical intervention and four stabilized clinical and did not progress to reoperation.

Surgical complications were reported in 3 of 110 patients (3%). Reported complications included three cases of wound sensitivity beyond the expected post-operative period and 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 population 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 at 31%, in contrast to studies by Sassoon and Bachoura who cited rates of 40% and 54%, respectively[4,6].

Our final post-operative range of motion with this open biopsy with curettage and bone defect grafting with freeze-dried, cancellous allograft chips was favorable in comparison to prior literature. Full ROM was achieved by 82% of patients in this study, compared to 67% and 46% in series presented by Sassoon and Bickels, respectively[4,7]. In a series by Bachoura, stiffness was reported in 3 of 24 patients (13%), including a PIP joint contracture, a MCP joint extension contracture requiring dorsal MCP joint capsule release, and a swan neck deformity[6]. In our series, range of motion deficits were found to be more frequent with pathologic fracture on presentation as well as with middle phalanx lesions, which we believe to be valuable, new information. As the characteristic ROM deficit in our series was 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 proximal phalanx level being secondary to adhesions within the flexor tendon fibro osseous tunnel. We hypothesize that range of motion deficits in patients who presented with pathologic fracture may have been secondary to extended immobilization to allow for preoperative fracture healing. This consideration has been discussed in prior literature;[18] however, we have favor fracture healing prior to intervention in order to prevent further angulation with curettage, as we do not use fixation with the technique described.[9] Given these novel findings, a comparison of ROM after pathologic fracture utilizing a technique that employs instrumentation with immediate treatment versus our preferred methods would be of interest for future study.

Finally, it is notable in our series that those patients who were treated with dorsal and lateral approaches achieved similar mean post-operative range of motion. Some authors have suggested that the lateral approach may achieve superior ROM by avoiding the extensor mechanism.[19] While results of our series identified that full ROM was achieved in 88% after

lateral approach in comparison to 76% of patients after the dorsal approach, motion deficits were similar between groups when present 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 each lesion enhances the 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 post-operative 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 Kirshner wires[7] and injection of calcium sulfate[8], allograft has been shown to be appropriate for filling bony defect 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. 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 post-operative 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 that 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 post-operative range of motion without additional fixation, allowing for sufficient therapy to prevent adhesion formation and post-operative stiffness necessitating tenolysis.

Our series was detected a recurrence rate of 10% within our population followed for a minimum of 2 years of postoperative follow-up. Though this number is slightly greater than that previously reported by Sassoon et al, we required two-year follow-up for inclusion in recurrence analysis whereas the aforementioned study minimum follow-up was one month time.[4] We believe this to be a comparative strength of our work. These recurrences notably occurred overwhelmingly amongst patients who presented with syndromic presentation or giant form lesions. Three of the five 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 noted that probable malignancy was suspected in some giant form lesions secondary to rapid growth,[2] this is not well established in the literature from a quantitative perspective.

Though numerous agents, including phenol[14], dehydrated alcohol[15], CO2 laser[16], and polymethyl methacrylate cement[7] have been described in case series, we do not believe that these are requisite for the treatment of these benign, slow-growing tumors, and may expose patients to increased risk of complication. Risk of CRPS secondary to nerve irritations following the use of such adjuvants is well documented[10,12] and, given the low recurrence risk in a solitary enchondroma and benign nature of these lesions, risk of complication from adjuvants should be heavily considered prior to use. Of note, there are certain cases that present a greater risk of recurrence. As we have noted 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 population is relatively small, which limits our ability to perform statistics on subgroup analyses. Our ability to report on pain or functional disability is limited to that documented in the historical notes. Completeness of excision was 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.* may have aided in identification of incomplete excision; however, this is not the standard procedure in our practice and further research would be required to establish superiority of this technique.[23] Our population likely suffers from a referral bias, given the subspecialty training and practice of our senior author. As such, our population 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 post-operative follow-up was not uniform. Though some patients had limited follow-up, we do not believe that this substantially impacted our post-operative range of motion findings, as none of the patients with the minimum follow-up reporting had more than a 10-degree motion deficit. We additionally required a 2-year minimum follow-up for evaluation of recurrence, which is in line with prior publications. Similarly, as a retrospective study conducted long period of time, complications may be underestimated secondary to this information being excluded from the chart or if patients were 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 twenty-three 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 post-operative range of motion, possibly secondary to preoperative immobilization. Both recurrence and malignant transformation remain 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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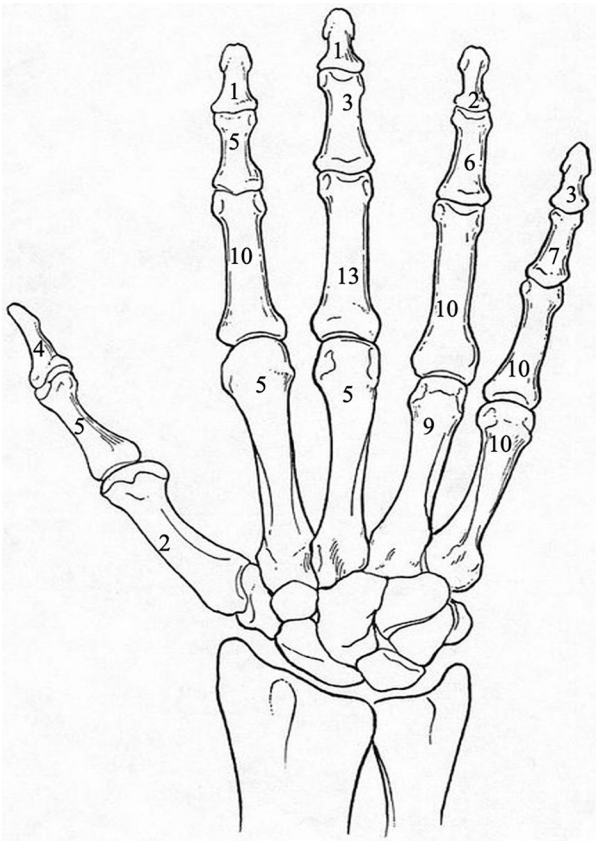

Figure 1.
Distribution of lesions

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)

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°)

SD is in parentheses.

Table 3.

Range of Motion Deficits by Location of Lesion

Location	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.

Table 4.

Range of Motion Deficits by Presentation With Pathologic Fracture

Presentation	Number of Digits with Deficit in ROM	Average Composite ROM Deficit
No fracture	11/72 (16%; CI: 9%–26%)	16° (12°)
Pathologic fracture	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%)