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The management and surgical intervention timing of enchondromas

A 10-year experience

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Abstract

Enchondroma, reportedly the most common primary tumor of the long bones of the hand, usually develops during the first till fourth decades of life. However, there has no consensus been reached regarding the surgical intervention timing for these patients. We aim to evaluate the optimal surgical intervention timing for the patients with fractures due to enchondromas, investigate the impact of pathological fractures on the treatment and outcomes in these patients.

Medical records and X-rays of patients treated for enchondroma of the hand from 2005 to 2015 were retrospectively reviewed. We collected 148 cases in total and 92 of them had complete information including X-rays, medical records, and files of follow up.

There were no significant differences in terms of consolidation time after surgery, recurrence rate, and DASH scores between the groups with and without fractures; the treatment costs were higher in the group with fractures than those without fractures; however, patients without fractures were able to resume work earlier than those with fractures.

The pathological fractures associated with enchondromas have no significant impact on the treatment outcomes compared to those with simple nonfractured enchondromas. Although the cost was more expensive for patients treated primarily with pathological fractures due to enchondromas, these patients could resume their work normally much earlier than those treated by delayed surgery. Early surgical intervention is recommended for better results and no increased risks for patients with pathological fractures caused by enchondromas.

Abbreviation: DASH = Disability of the Arm, Shoulder, and Hand.

Keywords: hand enchondromas, pathological fractures, primary surgery, surgical intervention time

Key Points

- Patients without fractures recovered much earlier than those with fractures.
- Among the patients with fractures, there were no significant differences in terms of consolidation time after surgery.
- Different surgical options of curettage only or combined with different filling materials had no effects on the follow-up outcomes.
- Of all the patients, different surgical options of curettage only or combined with different filling materials had no effects on the follow-up outcomes (all $P > .05$). Healing was noted to be progressive and complete in all cases and no functional restriction was observed.

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1. Introduction

Enchondroma is the most common bone tumor of the hand and is a benign, intramedullary, cartilaginous tumor. It originates from cartilage and is commonly located in the proximal metaphysis of the proximal phalanx. It often presents in the first through fourth decades of life and[1] has a predilection for the ulnar sided tubular bones of the hand, and arises most frequently in the phalanges and metacarpals. Furthermore, enchondromas with pathological fractures occur because the bone in question has been weakened by the disease process. Often, these injuries result from minor trauma, which might not otherwise cause a fracture in healthy bone.[2,3]

The timing of treatment when there is a pathologic fracture is not clearly defined and there is no standardized algorithm for surgical treatment of this tumor. Recent studies showed that simple curettage with or without bone grafting is an effective and safe treatment for most patients with simple solitary enchondromas.[1,4,5] Yalcinkaya et al reported that primary treatment of an enchondroma in the presence of a pathologic fracture did not change the outcome compared with lesions treated after fracture union. Naturally, patients treated after fracture union had additional periods of immobilization.[6] But it should be noted that traditionally, surgeons have preferred to wait for pathological fractures to heal before proceeding with operative treatment secondary to the belief that complication rates would be higher with immediate curettage.[7] However, there are few studies to investigate the impact of pathological fractures on the final treatment outcomes in patients with fractures due to enchondromas.

In this retrospective study, we reviewed our 10-year experience with a subset of patients who had solitary enchondromas. We attempted to investigate the optimal surgical intervention timing for the patients with fractures due to enchondromas and evaluate the impact of pathological fractures on the therapeutic outcome in such patients.

2. Methods

The retrospective analysis was performed on the clinical data of patients with enchondromas treated from February 2005 to August 2015 in our hospital and approved by the institutional review board (IRB). The inclusion criteria were: solitary enchondroma of the hand; with or without pathologic bone fracture; treated surgically; and a complete follow-up of more than 6 months. The exclusion criteria were: the merging of other bone and soft tissue injuries; multiple enchondromas; a follow-up of less than 6 months; missing follow-up data, and medical records or X-ray images.

The operative details and postoperative clinical and radiological outcomes were reviewed by an independent reviewer. The Disability of the Arm, Shoulder, and Hand (DASH), upper limb function scale was used to evaluate the function of the affected hand. The time to complete recovery, complications as well as the recurrence rate and in-hospital costs were recorded. Bone remodeling was defined as a uniform appearance with no gap between the cancellous bone and the bone substitute.[1,2,4,8]

2.1. Statistical analysis

Chi-square test was used for the statistical analysis of demographic data and complications after surgery between groups. One-way ANOVA was selected to compare the differences between groups in the follow-up outcomes with SPSS v. 19.0. Data were expressed as mean ± standard deviations. Statistical significance was considered at the 5% level.

3. Results

In total, 227 patients with enchondroma were identified, of which 92 cases were eventually included according to the inclusion and exclusion criteria of this study. There were 52 males (56.5%) and 40 females (43.5%) in this series, aged from 2 to 69 years (mean, 29.4 years). Based on the clinical data, patients with pathological fractures were assigned into the group with fractures (n = 27) and those without fractures, who were diagnosed accidentally during routine physical examinations were assigned into the group without fractures (n = 65). All patients underwent surgical treatment. Patients in the fracture group were divided into 2 subgroups: primary (≤4 weeks, mean 16 days) surgery group and delayed (>4 weeks, mean 87 days) surgery group according to the time of surgical intervention. The operative methods were simple curettage or combined with bone grafting with autologous or

Table 1

Patients' demographic data associated with timing of surgery*.

Group	Sex		Sites		Age, y			Size, mm²		
	M	F	M	P	<14	15–45	>45	<200	201–400	>400
Primary surgery group	8	8	2	14	1	10	5	4	9	3
Delayed surgery group	7	4	1	10	1	9	1	3	5	3
P	.397		.792		.218			.826		

* No significant differences in patients' demographic data were noted between the primary surgery group and delayed surgery group (all $P > .05$).
M = metacarpals, P = phalanges.

Table 2

Patient's demographic data (enchondroma associated with or without pathological fractures)*.

Group	Sex		Sites		Age			Size, mm²		
	M	F	M	P	<14	15–45	>45	<200	201–400	>400
Group with fracture	15	12	3	24	2	19	6	7	14	6
Group without fracture	37	28	15	50	15	37	13	14	16	16
P	.905		.145		.216			.670		

* No significant differences in patients' demographic data were seen between groups of enchondromas with and without fractures (all $P > .05$).
M = metacarpals, P = phalanges.

Table 3

Patient's demographic data and different treatment options*.

	Male/female	Mean (range) age, y	Site (metacarpal/phalanx)†	Mean volume, mm^2
Curettage only group (n=26)	16:10	22.31 (2–68)	10:16 (62.5%)	274.47 (70–820)
Autograft group (n=37)	21:16	33.42 (12–56)	4:33 (12.1%)	363.27 (90–1161)
BOM group (n=29)‡	15:14	30.57 (7–69)	3:26 (11.5%)	353.36 (44–820)
P	.764	.08	.000	.074

* No significant differences in patients' demographic data were observed among groups with different treatment options (all $P>.05$) except for the affected sites.
† In this series, the ratio of the affected phalanges and metacarpals was much higher in the curettage only group in comparison with the other 2 groups ($P=.000$).
‡ Bioactive and osteoconductive materials other than autograft.

bioactive materials. The patients' demographics are listed in Tables 1–3.

There were no significant differences in terms of consolidation time after surgery, recurrence rate, and DASH scores between the groups with and without fractures (all $P>.05$); the in-hospital costs were higher in the group with fractures (bioactive and osteoconductive materials or the iliac bone graft cost) than those in the group without fractures ($P=.007$); however, patients without fractures recovered much earlier than those with fractures ($P<.001$). Among the patients with fractures, there were no significant differences in terms of consolidation time after surgery, recurrence rate, DASH scores as well as the occurrence rate of complications between the primary surgery group and the delayed surgery group (all $P=.534$); however, recovery time was statistically longer in the delayed surgery group, with an average of 76 days than that of the primary surgery group, with an average of 36 days ($P<.001$). The outcomes of primary and delayed surgery groups are listed in Tables 4 and 5. Of all the patients, different surgical options of curettage only or curettage combined with different filling materials had no effects on the follow-up outcomes (all $P=.692$). Healing was noted to be progressive and complete in all cases and no functional

Table 4

Outcomes of groups with primary and delayed surgery.

Group	Cases	Consolidation time after surgery†	Recurrence	Recovery time, d*	Complications			Cost, $†	DASH,‡ 100%
					Infection	Hematoma	Persistent pain		
Primary surgery group	81	54.23	0	36.06	0	0	12	2100	87.18
Delayed surgery group	11	55.36	0	76.16	1	0	5	1200	88.75
P	92	.747	/	.000	/		.385	.007	.534

* The recovery time was significantly shorter in the primary surgery group when compared to the delayed surgery group ($P=.000$). For the occupational patients, their recovery time means the time to return to work, but for those too young or too old (retired), the recovery time was calculated to return to their previous activities.
† The cost of treatment for the group of fractures with primary surgery was significantly higher than the cost of treatment for the group with delayed surgery ($P=.007$).
‡ We noted no significant differences in consolidation time after surgery and Disability of the Arm, Shoulder, and Hand (DASH) scores between the 2 groups ($P=.747$).

Table 5

Outcomes of patients suffering from enchondromas with or without fractures.

Group	Cases	Consolidation time after surgery†	Recurrence	Recovery time, d*	Complications			Cost, $†	DASH,‡ 100%
					infection	hematoma	persistent pain		
Group with fracture	27	52.33	0	50.70	1	0	7	2000	87.96
Group without fracture	65	53.92	0	21.60	0	0	10	1300	87.20
P	92	.872	/	.031	/		.467	.008	.920

* The recovery time was significantly shorter in the group without fractures when compared to the group with fractures ($P=.031$).
† The cost of bioactive and osteoconductive materials or the cost of iliac bone graft for the group with fracture was significantly higher than for the group without fracture ($P=.008$).
‡ No significant differences in consolidation time after surgery and Disability of the Arm, Shoulder, and Hand (DASH) scores were noted between the 2 groups ($P=.872$).

Table 6

Outcomes of different treatment options.

Group	Cases	Consolidation time after surgery	Recurrence	Recovery time, d*	Complications			Cost, $†	DASH, 100%
					Infection	Hematoma	Persistent pain		
Curettage only group	26	52.88	0	20.22	0	0	2	1150	86.84
Autograft group	37	55.48	0	33.59	1	0	8	1700	87.02
BOM group‡	29	51.37	0	35.17	0	0	7	2000	88.44
P	92	.692	/	.000	/	/	/	.028	.813

DASH=Disability of the Arm, Shoulder, and Hand.
* The recovery time of the curettage only group was significantly shorter when compared to the other 2 groups ($P=.000$).
† The costs of the curettage only group was the lowest among the 3 groups ($P=.028$).
‡ Bioactive and osteoconductive materials other than autograft.

restrictions were observed. Outcomes of different treatment options are listed in Table 6.

4. Discussion

Enchondroma, a benign slowly growing tumor composed of hyaline cartilage cells that persists throughout development, is the most common primary bone tumor of the hand, the exact incidence varies and the exact incidence of enchondroma is still unknown.[1,6] Up to 70% of enchondromas occur in the hand, and the proximal phalanges are the most frequent site involved.[1,9] It grows slowly, weakens the bone, and leads to pathological fracture.[1,2,4,9,10] In fact, fractures associated with these benign lesions may be allowed to heal before definitive treatment of the tumor, however, surgical intervention is suggested to minimize complications and allow early motion.[10,11]

Traditionally, enchondromas are treated with curettage and grafting with allogeneic bone or autogenous or synthetic bone substitutes.[1,4–6,9,10,12–15] However, there is no standardized algorithm for surgical treatment of this kind of tumor. It is not clear whether grafting after curettage is necessary or whether the type of graft used affects healing, recurrence, complications, and malignant transformation.[1,12,16] Our study shows that the choice of graft either in patients with or without pathological fractures has no effect on time required to heal, range of motion, recurrence, complications, or malignant transformation. Nonetheless, considering the bone will be further weakened by curettage alone, we believe that replacement with an osteogenic or osteoconductive substance will facilitate bone healing and remodeling so that this fracture-prone period may be shortened. Although autologous bone graft, which will not cause immune rejection, is the most suitable choice, it may be associated with some donor site morbidities such as infection, hematoma, and chronic pelvic pain. As an alternative, we attempted to treat with artificial bone substitute as reported in other studies.[6] The application of bioactive and osteoconductive materials which is available in various shapes and sizes has obvious advantages of reduced donor site morbidity and reduces operating time and convenience of local anesthesia. However, in the present study, we found the cost of treatment with such materials is much higher than with other options. On the other hand, we found that the recovery time was shorter in the curettage only group than in the other 2 groups with grafts. We speculated this finding might be attributed to the differences in the affected site ratios of metacarpals and phalanges. In the curettage only group, the ratio was much higher (62.5%) than the other 2 groups (12.1% and 11.5%, respectively). Usually postoperative defects of digits in children who sustained no fractures heal faster when compared to patients with fractured lesions.[2,12] Therefore, the choice of dealing with such patients should be assessed comprehensively and individualized based on patients' affordability and requirement.

In literature, the timing of treatment when there is a pathologic fracture in these patients has not been clearly defined. Traditionally, the mainstay of surgical intervention for such patients is carried out with a delayed procedure until the fracture site completely or partially heals so that a simple curettage may be applicable with no needs of internal fixation.[17] Early treatment was associated with a shorter period of disability, but also with significantly more complications (67% vs 10%), including stiffness and rotational deformity in the study of Jacobson and Ruff.[2] But in our study, no significant differences were found between the primary surgery group and delayed surgery group in terms of consolidation time after surgery, recurrence rate, DASH scores as well as the occurrence rate of complications. Primarily, treatment of enchondroma in the presence of a pathologic fracture did not change the outcome compared with lesions treated after fracture union. Naturally, patients treated after fracture union had additional periods of immobilization. In another study, the outcome was similar to our results.[6] The strut bone graft served both mechanical and biologic functions. Although the in-hospital expense was a bit higher in the primary surgery group than in the delayed surgery group, the time to return to work was significantly shorter in the primary surgery group. Actually, a long-term interval before a delayed operation will not only prolong the treatment time, but also may lead to finger dysfunction; in addition, the potential fracture displacement may also cause a deformity of the finger. In our study we did not observe finger deformities. Therefore, the primary surgery surely has a substantial advantage in the management of patients with pathological fracture due to enchondroma.

Enchondroma is usually identified after an incidental finding on radiographs or as a pathologic fracture. According to the results of our investigation, 27 patients suffered from pathologic fractures. A recurrence rate of up to 13.3% after curettage and bone grafting was reported.[4,6,18] However, there are no such events in our series. Patients with a diagnosis of enchondroma with fractures had no higher rate of complications. At follow-up, no differences in outcomes in terms of consolidation time after surgery and DASH scores were observed between patients with or without fractures. However, our findings showed that patients suffering from osteochondromas without fractures could resume work much earlier than those with fractures.

In this series, we noted good functional recovery in all groups of patients, while complications were uncommon. Only 1 patient treated with an autograft in the delayed surgery group suffered from infection and 1 case in this group required reoperation because of a secondary fracture. The most common complication was persistent pain. Although there were no significant differences in the occurrence rate of persistent pain between the group with fractures and the group without fractures, a higher occurrence rate of 25.9% (7/27, CI: 11.9–40.1%) was observed in the former group in comparison with an occurrence rate of 15.3% (10/65, CI: 7.14–23.4%) in nonfracture patients. Despite soft tissue complications were not analyzed in this study, we speculate that local trauma leading to postoperative functional restrictions could be a major source of patient distress following enchondroma surgery. Therefore, meticulous curettage and minimally invasive manipulation of the surrounding soft tissues is the key to achieve good results and avoid complications. Pathological fractures associated with enchondromas have no significant impact on the treatment outcomes compared to those with nonfractured enchondromas. Although the hospital costs were higher for patients treated primarily with pathological fractures due to enchondromas, these patients could resume to their original work much earlier than those treated by delayed surgery. Early surgical intervention is recommended for patients with pathological fractures caused by enchondromas.

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