ONCOLOGY

Outcomes after extensive manual curettage and limited burring for atypical cartilaginous tumour of long bone

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Aims
Adjuvant treatment after intralesional curettage for atypical cartilaginous tumours (ACTs) of long bones is widely accepted for extending surgical margins. However, evaluating the isolated effect of adjuvant treatment is difficult, and it is unclear whether not using such adjuvants provides poor oncological outcomes. Hence, we analyzed whether intralesional curettage without cryosurgery or chemical adjuvants provides poor oncological outcomes in patients with an ACT.

Patients and Methods
A total of 24 patients (nine men, 15 women) (mean age 45 years; 18 to 62) were treated for ACTs of long bones and followed up for a median of 66 months (interquartile range 50 to 84). All patients were treated with extensive manual curettage and limited burring. Bone cement and grafts were used to fill bone defects in 16 and eight patients, respectively. No chemical adjuvants or cryosurgery were used.

Results
No local recurrence was detectable on plain radiographs and MRI or CT images. At the last follow-up, there were no distant metastases or disease-specific deaths. No procedure-related complications or postoperative fractures developed.

Conclusion
Intralesional curettage without cryosurgery or chemical adjuvants may provide excellent oncological outcomes for patients with ACTs of long bones, without the risk of complications related to adjuvant use. Our investigation suggests thorough curettage alone is a reasonable treatment option for ACT. However, we acknowledge the limited size of our investigation warrants a multicentre collaborative study to confirm our findings.

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Chondrosarcoma is a cartilage-forming malignant bone tumour and the second most common primary malignant bone neoplasia. Atypical cartilaginous tumours (ACTs), formerly known as low-grade chondrosarcoma, are locally aggressive but rarely metastasize, with a five-year survival rate of approximately 83%, whereas the five-year survival rate of high-grade chondrosarcoma is approximately 53%. ACTs are now distinguished from high-grade chondrosarcomas and classified as having unspecified, borderline, or uncertain behaviours. Considering these biological characteristics and oncological outcomes, most orthopaedic oncologists favour intralesional curettage over wide resection for the treatment of ACTs of long bones; however, wide resection is preferred for ACTs of the pelvis because they seem to behave more aggressively and are associated with poor patient outcomes when they are treated with intralesional curettage.

Regarding ACTs of long bones, many adjuvant treatments have been introduced and widely used to extend surgical margins, with excellent outcomes and local recurrence rates of 0% to 13.3% reported for these treatments. Previous studies have shown that, compared with wide resection and reconstruction, intralesional curettage and adjuvant treatment have a comparable local control rate and better functional outcome, due to the preservation of skeleton. Phenol and cryosurgery are the most commonly employed adjuvant treatments, with local recurrence rates of 3.1% to 13.3%, and 0% to 9.3%, respectively.
However, it is difficult to evaluate the isolated effect of adjuvant treatment, and it is unclear whether not using such adjuvants provides poorer oncological outcomes. Hence, we analyzed whether intralesional curettage without cryosurgery or the use of specific chemical adjuvants, such as phenol, would provide poor oncological outcomes in patients with ACTs of long bones.

**Patients and Methods**

From the prospectively collected database of our institution, we identified 52 patients who underwent open biopsy or primary operative treatment for a suspected ACT of long bones between 2004 and 2013. To construct a more homogenous group, the following 28 patients were excluded: those with biopsy-proven enchondromas (n = 9), ACT not in a long bone (n = 7), escalated histological grade after definitive surgery (n = 2), the presence of a separated lesion that was not included within the range of curettage area (n = 2), preoperative biopsy-confirmed ACT which was conservatively treated due to very old age (n = 1), and when follow-up < 48 months (n = 7). The remaining 24 patients with pathologically proven ACTs of long bones were included and analyzed. The diagnosis of ACT was based on plain radiographs and MR images, supplemented by 3D-CT to evaluate the endosteal erosion more accurately in multiple planes. Histologically proven enchondromas were generally treated conservatively, whereas biopsy-proven ACTs or equivocal ACTs were treated with subsequent definitive surgical intervention. In the event of extraosseous soft-tissue extension, we resected the tumour, as soft-tissue extension itself suggests a possibility of high-grade tumour. We used the term ACTs in this study but acknowledge the terminology of ACTs was introduced recently. Nevertheless, the fundamental diagnostic criteria have not changed.

Our principal indication for surgery was an endosteal erosion and tumour > 6 cm in longitudinal length. We generally performed preoperative open biopsy targeting areas where the thick peripheral enhancement change and endosteal erosion were most prominent. A specialized biopsy trephine was used to acquire multiple thick core tissue specimens (approximately 5 mm in diameter) that were representative of the tumour. These specimens were obtained via a single entry point aiming in more than three different directions (Fig. 1). We received institutional review board approval to conduct this analysis.

The data collected from medical records included: demographic variables such as age, gender, follow-up time, and anatomical site of the tumour; primary tumour-related details, such as size of tumour, presence of pathological fracture, defect-filling material, type of internal fixation, and surgery-related complications; the oncological outcome variables local recurrence and status at the final follow-up; and the clinical outcome variable Musculoskeletal Tumor Society (MSTS) scoring.16

The study population comprised nine men and 15 women, with a mean age of 45 years (18 to 62) (Table I). The sites of the primary tumour included the proximal humerus (n = 8), distal femur (n = 6), femur shaft (n = 4), proximal femur (n = 2), proximal tibia (n = 2), and others (n = 2). The mean longitudinal length of the tumour was 7.3 cm (3.2 to 15.2).

In total, 21 of 24 patients (87.5%) underwent preoperative biopsy, and three patients wanted to undergo one-stage surgery based on image findings and the clinical impression of ACT. All surgical procedures were performed...
by a single experienced orthopaedic oncologist (JSL). Our surgical treatment was intralesional curettage using curettes of various angles and sizes to obtain clearance where a direct view was not possible. We removed tumour tissue through an oval-shaped cortical window that was usually approximately 1 cm narrower at the proximal and distal tumour borders (Fig. 2). For the very large tumours, we made two oval windows with an intervening cortical bridge of more than 2 cm to 3 cm, in order to reduce mechanical weakness. Under fluoroscopic guidance we performed curettage until normal cancellous or hard cortical bone was encountered, thereby confirming the full tumour had been resected. Then, we selectively used a high-speed burr to remove possible microscopic remnant tumour tissue but were restricted to those areas which could be viewed directly through the cortical window. The high-speed burr

Table I. Patient characteristics

<table>
<thead>
<tr>
<th>Characteristics</th>
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<tbody>
<tr>
<td>Mean age, yrs (range)</td>
<td>45 (18 to 62)</td>
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<tr>
<td>Gender, n (%)</td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>15 (62.5)</td>
</tr>
<tr>
<td>Male</td>
<td>9 (37.5)</td>
</tr>
<tr>
<td>Median follow-up duration, mths (range)</td>
<td>66 (48 to 133)</td>
</tr>
<tr>
<td>Site, n (%)</td>
<td></td>
</tr>
<tr>
<td>Femur</td>
<td>12 (50.0)</td>
</tr>
<tr>
<td>Humerus</td>
<td>8 (33.3)</td>
</tr>
<tr>
<td>Tibia</td>
<td>3 (4.2)</td>
</tr>
<tr>
<td>Radius</td>
<td>1 (4.2)</td>
</tr>
<tr>
<td>Mean tumour longitudinal length, cm (range)</td>
<td>7.3 (3.2 to 15.2)</td>
</tr>
<tr>
<td>Preoperative biopsy, n (%)</td>
<td></td>
</tr>
<tr>
<td>Not done</td>
<td>3 (12.5)</td>
</tr>
<tr>
<td>Performed</td>
<td>21 (87.5)</td>
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<tr>
<td>Internal fixation, n (%)</td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>8 (33.3)</td>
</tr>
<tr>
<td>Performed</td>
<td>16 (66.7)</td>
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<tr>
<td>Defect-filling material, n (%)</td>
<td></td>
</tr>
<tr>
<td>Bone graft</td>
<td>8 (33.3)</td>
</tr>
<tr>
<td>Bone cement</td>
<td>16 (66.7)</td>
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IQR, interquartile range

Fig. 2a Preoperative plain radiograph and b) fat-suppressed T2-weighted fast spin-echo MR image of a 52-year-old man with atypical cartilaginous tumour (longitudinal length, 7.5 cm) of the proximal humerus showing chondroid calcification (asterisk) and mild endosteal erosion (arrow heads). The proximal and distal border of the tumour was marked with arrows. The patient was treated with intralesional curettage and cementing. c) An enhanced MR image taken two years after the surgery showed no recurrent lesions. d) The patient was followed up for seven years without local recurrence. Arrows depict the cortical window range used for curettage.
used was a fine round diamond burr (Conmed Inc., Utica, New York). The diamond burr permitted delicate ablation of cortical bone without the concern of sudden penetration of thinned cortical bone while burring. We irrigated the cavity alternatively with saline and hydrogen peroxide solution. Bone cement and bone allografts were used to fill bone defects in 16 and eight patients, respectively. Internal fixation was undertaken prophylactically in 16 patients bridging the cortical window. In 13 patients with leg lesions, plate and screw fixation was performed. In three patients with lesions involving their arm, fixation comprised a plate and screws in two and flexible nailing in the remaining patient. Where plates were used two screws were placed proximally and distally. For arm lesions, internal fixation was performed in the presence of large lesions involving more than 50% of bone length or when the defect was filled with bone graft.

Postoperatively, patients were followed up every three months during the first two years, at six-month intervals during the third to fifth years, and once a year thereafter. At each visit, we routinely obtained plain radiographs of the affected site, with a minimum of four orthogonal views (anteroposterior and two oblique). Enhanced MR images or 3D-CT scans of the surgical bed were obtained between one and two years postoperatively for initial reference, and yearly or every other year thereafter in cases of clinical or radiological suspicion of local recurrence. Pulmonary metastasis was screened using plain radiographs at each visit, and CT of the chest was obtained if a metastatic lesion was suspected on the plain radiographs. Independent evaluation was performed by an experienced musculoskeletal radiologist (HWC) for local recurrence using anonymized plain radiographs and MR or CT images. We first evaluated anonymized serial follow-up images with plain radiographs. After four weeks of interval period, we evaluated follow-up images with MRI or CT and compared the result with those of plain radiographs.

Statistical analysis. The primary endpoint of this study was local recurrence-free survival. Data analysis was performed using SPSS software (21.0; SPSS Inc., Chicago, Illinois).

Results
Patients were followed up for a median of 66 months (interquartile range 50 to 84). In all, 20 patients were followed up for more than 60 months. No local recurrence was detectable on the plain radiographs up to the time of the last follow-up. Similarly, independent evaluation of anonymized MR/CT images identified no local recurrences. At the last follow-up, there was no distant metastasis or disease-specific death. No procedure-related complications, wound infections, failure of incorporation of the implanted bone graft, or postoperative fractures developed. The mean MSTS score was 92% (83% to 100%) at the final follow-up for all the patients.

Discussion
Treatment strategies for ACTs of long bones have been shifted from wide excision to intralesional curettage because intralesional surgery has the advantages of host bone preservation and better functional outcomes. Advanced imaging and accumulated understanding of the biological behaviour of ACTs of long bones have enabled orthopaedic oncologists to perform more conservative surgery for these tumours. However, during the transition period, the difference between a wide excision and simple curettage seemed quite big. Accordingly, some bridging and additional treatment that could possibly enhance simple curettage to offer benefit comparable to wide excision, were required to justify intralesional surgery for malignant bone tumours. These extra measures included the addition of a high-speed burr, use of the thermogenic effect of bone cement, use of chemical adjuvants such as phenol and alcohol, and cryosurgery. Subsequently, in the recent literature, local recurrence rates after the use of these adjuvants have significantly reduced to between 0% and 13% for ACTs of long bones, whereas those of isolated simple curettage for all ACTs ranged from 50% to 75%. However, in most studies, many adjuvant treatments were used simultaneously, which obscured the isolated effects of specific adjuvants. Therefore, we investigated whether use of the modern technique of intralesional curettage and omission of the most commonly used adjuvants such as cryosurgery and phenol would result in poorer local control for the treatment of ACTs of long bones.

In our series of 24 patients with ACTs of long bones who were treated with intralesional curettage without cryosurgery or phenol, excellent results were obtained with no local recurrences at a median follow-up of 66 months and a minimum of 48 months. In addition for the 20 patients where follow-up exceeded five years, no local recurrences occurred. Minimum five-year follow-up results in the literature report, where supplementary treatment has been included, a local recurrence rate of 9.3% to 13.3% has been described. Our results, with no local recurrence after minimum four-year follow-up, suggest that the oncological outcomes were not compromised after extensive manual curettage and limited burring.

Many adjuvants have been used for the treatment of ACTs of long bones, with comparable oncological outcomes and local recurrence rates of 0% to 13%. The difference in local recurrence rate among the studies may be explained by the possible variations in grading among the pathologists and the different surgical techniques used among surgeons. Therefore, a direct comparison of outcomes based on the literature is complicated and limited. Among adjuvants, cryosurgery and phenol are the most commonly used adjuvants for extending surgical margins. However, phenol may cause chemical burns to the adjacent soft tissue and may result in systemic toxicity if absorption occurs. Regarding cryosurgery, specialized equipment is required to handle the cryosurgery probe and liquid.
nitrogen (or argon) and to control the temperature. Moreover, it is known to weaken the adjacent bone and may result in an increased risk of postoperative pathological fractures.\textsuperscript{10,18,22} Because our treatment approach for ACTs of long bones involved intralesional curettage, limited additional burring of accessible areas, and cementing in selected cases, we avoided any complications related to the use of such adjuvant treatments. Some previous studies have also reported good oncological outcomes with a local recurrence rate of 0% to 5% without the use of phenol or cryosurgery.\textsuperscript{3,15} We believe that thorough mechanical curettage played a major role in the outcomes we observed, and that the thermogenic effect during polymerization of the bone cement also partially contributed to the good results.

The importance of careful patient selection for definitive surgery should be emphasized because the biological behaviour of ACTs of long bones is considered less aggressive than that reported previously.\textsuperscript{4} We performed open biopsy only in patients who were more likely to undergo surgery based on radiological parameters. Before definitive surgery, we generally obtained multiple thick core tissues from the lesion area where endosteal scalloping and thick peripheral enhancement were most prominent to confirm the diagnosis histologically. This enabled us to be confident that an ACT had been identified.

Our study has several limitations. First, it was a retrospective study without a control group. Because of the rarity of ACTs of long bones and the fact that we basically treated pathologically confirmed ACT, only a limited number of patients were enrolled. Hence, it was not feasible to conduct a prospective or comparative study. Additionally, as the number of patients was small, it is difficult to assert definitively that the use of adjuvant is not necessary based on this study alone. Further investigation in a multi-institutional setting is desirable. Second, we also used high-speed burring for grossly visible areas and bone cementing to fill the cavity defect in some patients. Both high-speed burring and bone cementing can be considered as adjuvant treatments. Although we primarily used various sized and angled curettes to remove the tumour beyond the tumour border until we met grossly normal cancellous and cortical bones, local heat generation using high-speed burns and bone cement may also have contributed to the outcomes. However, there were no local recurrences at the areas inaccessible to high-speed burring, such as the under-surface of the cortical window and proximal and distal borders. Where we filled the defect with a bone graft, confirmation of the extent of tumour curettage in multiple planes under fluoroscopic guidance was a crucial process for successful local control. Thirdly, we used hydrogen peroxide as in some previous studies.\textsuperscript{2,12} This can be considered as another adjuvant. However, the local ablative role of hydrogen peroxide for chondrosarcoma has not been fully investigated, and its application in the previous literature seems closer to an irrigation role to wash out debris and blood clots before cementing, as in the current study.

In conclusion, intralesional curettage and additional burring and only bone grafting or intralesional cementing but without cryosurgery or chemical adjuvants, may provide excellent oncological outcomes in ACT patients. Considering the slow and locally aggressive biological behaviour of ACTs of long bones, thorough mechanical curettage beyond the tumour border is the most crucial procedure. The contribution of additional adjuvant treatments to ACTs of long bones is possibly smaller than expected, provided thorough curettage is performed. Thorough curettage alone is a reasonable treatment option for ACTs of long bones.

Take home message:
- Here we describe an excellent oncological outcome of intralesional curettage without using cryosurgery or phenol for atypical cartilaginous tumour of the long bone.
- Thorough mechanical curettage alone is a reasonable treatment option for atypical cartilaginous tumour of the long bone, and the role of adjuvant treatments needs to be re-established in a controlled study.

References


Author contributions:

W. Kim: Preparing and analyzing patient data, Statistical analysis, Literature review, Writing the manuscript.

J. S. Lee: Conception of the study, Main surgeon of the current series, Reviewing and writing the manuscript.

H. W. Chung: Independent radiologic evaluation, Literature review.

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