Enchondromas and atypical cartilaginous tumors at the proximal humerus treated with intralesional resection and bone cement filling with or without osteosynthesis: retrospective analysis of 42 cases with 6 years mean follow-up

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Abstract

Background: Enchondromas and atypical cartilaginous tumors (ACT) are often located at the proximal humerus. Most lesions can be followed conservatively, but surgical resection may alleviate pain, avoid pathological fractures, and prevent transformation into higher grade chondrosarcomas. Rigorous intralesional resection and filling with polymethylmethacrylate bone cement has been proposed for enchondromas but also for ACT, as an alternative for extralesional resection. We intended to analyze radiological, clinical, and functional outcome of this strategy and compare bone cement without osteosynthesis to bone cement compound osteosynthesis, which has not been analyzed so far.

Methods: We retrospectively analyzed 42 consecutive patients (mean follow-up 73 months; range 8–224) after curettage and bone cement filling with or without osteosynthesis. Exclusion criteria were Ollier’s disease and cancellous bone filling. Twenty-five patients only received bone cement. Seventeen patients received additional proximal humerus plate for compound osteosynthesis to increase stability after curettage. Demographics and radiological and clinical outcome were analyzed including surgery time, blood loss, hospitalization, recurrences, and complications. An additional telephone interview at the final follow-up assessed postoperative satisfaction, pain, and function in the quick disabilities of the arm, shoulder, and hand (DASH) score and the Musculoskeletal Tumor Society (MSTS) score. Statistics included the Student T tests, Mann-Whitney U tests, and chi-square tests.

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Background
Chondrogenic tumors such as enchondromas and atypical cartilaginous tumors (ACT = chondrosarcoma grade I according to older nomenclature) are often located at the proximal humerus [1] and raise the question for surgeons different? Is there a difference in recurrences, complications, or other surgical parameter?

Methods
We retrospectively analyzed 42 consecutive patients surgically treated for enchondroma or ACT at the proximal humerus with a mean follow-up of 73 months (range 8–224). Approval was given by our local ethical committee. From 2005 till 2017, we found a total of 113 patients treated conservatively or surgically at our orthopedic oncology outpatient clinic (level I bone and soft-tissue tumor center and orthopedic and trauma surgery university hospital). Exclusion criteria were no surgical therapy (n = 65), less than 6 months follow-up (n = 1), Ollier’s disease (n = 1), and filling with cancellous bone (n = 4) instead of bone cement, as cancellous bone filling was only used for smaller and less aggressive enchondromas and hence could not be compared. Forty-two patients with sufficient data which were surgically treated between 2006 and 2016 were finally included in the study (Fig. 1). Of those, 25 patients underwent rigorous intralesional excision with use of a high-speed burr and filling of the lesion with polymethylmethacrylate bone cement (Palacos® R+G; Heraeus Medical, Hanau, Germany) to achieve improved

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stability and reduce recurrence rate due to heat destruction of potentially remaining tumor cells during the polymerization process in the lesion cave (Figs. 2 and 3). This group was defined as the study group.

Seventeen other patients underwent the same procedure followed by the support of a proximal humerus locking compression plate (LCP; PHILOS plate, Synthes GmbH, Oberdorf, Switzerland) in a way that the screws of the osteosynthesis were integrated into the bone cement as a compound plate osteosynthesis (Figs. 4 and 5). This group was defined as the control group.

A decision whether additional compound plate osteosynthesis with a proximal humerus plate was used or not was done individually. According to the documented medical records, additional plate osteosynthesis was justified and selected, if preoperative imaging and intraoperative appearance after curettage caused doubt of sufficient stability only with bone cement. As there are no scientific guidelines for

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**Fig. 1** Flowchart of inclusion criteria.

**Fig. 2** Painful large chondrogenic tumor at the proximal humerus prior to surgery without osteosynthesis. Plain radiograph (a) shows typical popcorn-like calcifications (arrow) inside the epiphyseal and metaphyseal lesion. STIR MRI sagittal series (b) and T1-weighted contrast-enhanced axial MRI series (c) reveal large size and aggressiveness with endosteal scalloping (arrow) reducing stability of the proximal humerus.
decision-making, we retrospectively analyzed the potential criteria.

Adopted from other studies [1], lesion size was measured by the maximal diameter in MRI, as heterogeneous geometrical configurations of the lesions hamper valid measurement of lesion volumes. Preoperative lesion size and postoperative size of the cavity filled with bone cement after curettage were evaluated. Preoperative aggressiveness of the lesion was judged by a semi-quantitative score considering soft-tissue extension (no = 0, yes = 1) and endosteal scalloping (no = 0, minimal = 1, moderate = 2, high = 3) resulting in a score from 0 to 4. Scalloping was considered minimal if it involved less than one third of the cortical thickness, moderate if it involved up to two thirds, and high if it involved more than two thirds. Radiological evaluation with x-rays and MRIs was performed initially and regularly with intervals between 6 (first year after surgery) and 12 months for clinical routine. Imaging was evaluated together with our musculoskeletal radiologists subspecialized in bone and soft-tissue tumor diagnostic.

Patient demographics (Table 1) and clinical histories including detailed information on surgical treatment, histological analysis, recurrences, and complications were analyzed. For systematic evaluation of pain, patient satisfaction, and functional outcome at final follow-up,
we performed an additional telephone interview. Remaining pain and overall patient satisfaction were asked to be judged from 0 to 10. Limitations and clinical function were evaluated by the Musculoskeletal Tumor Society (MSTS) score [26] and the quick disabilities of the arm, shoulder, and hand (DASH) score [27].

To evaluate the results, statistical analysis was performed for the outcome measures “MSTS score”, “DASH score”, “preoperative lesion size”, “scalloping and soft-tissue extension score”, “size of bone filling”, “number of recurrences”, “number of complications”, “blood loss”, “surgery time”, and “days of hospitalization”. To compare the differences, Student T tests, Mann-Whitney U tests, and chi-square tests were calculated depending on the scale level and distribution of the data. Statistical significance was assumed at a p value < 0.05. Due to the exploratory character of the study, all p values are interpreted descriptively. Analysis was performed together with the statistician of our department (SG) using SPSS for Windows 22.0 (SPSS Inc., USA).

**Results**

All 42 patients were followed until the final follow-up with information on surgeries, radiological outcome, clinical presentation in the outpatient clinic, recurrences, and complications. Results from the telephone interview were only available for n = 31 patients.

**Surgical parameter**
The group without additional osteosynthesis showed less surgery time, less intraoperative blood loss, and less days of hospitalization (Table 2).

**Table 1** Demographics of both treatment groups

|                           | Intralesional resection + bone cement (n = 25) | Intralesional resection + bone cement + proximal humerus plate (n = 17) | Statistical test with p value |
|---------------------------|----------------------------------------------|------------------------------------------------------------------------|-----------------------------|
| **Gender**                |                                              |                                                                        |                             |
| Male                      | n = 4                                        | n = 8                                                                  | Chi-square test p = 0.03    |
| Female                    | n = 21                                       | n = 9                                                                  |                             |
| **Age**                   | Mean (± SD)                                   |                                                                        |                             |
|                           | 50.3 (± 10.8) years                          | 48.2 (± 12.0) years                                                    | Mann-Whitney U test p = 0.52|
| **Histology**             |                                              |                                                                        |                             |
| Enchondroma                | n = 19                                       | n = 12                                                                 | Chi-square test p = 0.67    |
| ACT                       | n = 3                                        | n = 2                                                                  |                             |
| Enchondroma or ACT         | n = 3                                        | n = 3                                                                  |                             |
Radiological outcome
Lesions which did not receive additional osteosynthesis were smaller (4.2 (± 1.5) cm versus 6.6 (± 3.0) cm; \(p = 0.005\)). After curettage, the lesion cavity which was filled with bone cement was also smaller in the group without osteosynthesis (5.7 (± 2.1) cm versus 9.6 (± 3.2) cm; \(p = 0.0001\)). Endosteal scalloping in the group without osteosynthesis was minimal in 10 cases, moderate in 10 cases, and high in 5 cases. In the group with osteosynthesis, it was minimal in five cases, moderate in seven cases, and high in four cases. Soft-tissue extension was only found in four cases (two in each group). A semi-quantitative score of preoperative scalloping and soft-tissue extension did not show significant differences between the groups (1.9 (± 0.9) versus 2.0 (± 1.0); rating scale 0–4; \(p = 0.7\)). Detailed results depending on histological diagnosis are presented in Table 3.

Clinical outcome
Overall clinical outcome was excellent. Patient satisfaction, pain, and functional outcome did not show statistically significant differences. Detailed results are presented in Table 4.

Recurrences
No recurrence was found in the osteosynthesis group. One of the 25 patients from the group without osteosynthesis had a histologically proven enchondroma recurrence after 4 years. It was successfully treated by revision surgery, again without osteosynthesis.

| Table 2 Surgical parameter | Intralesional resection + bone cement (\(n = 25\)) | Intralesional resection + bone cement + proximal humerus plate (\(n = 17\)) | Statistical test with \(p\) value |
|-----------------------------|-----------------------------------------------|------------------------------------------------|--------------------------------------|
| Surgery time                |                                               |                                               | Mann-Whitney \(U\) test \(p < 0.0001\) |
| Mean (± SD)                 | 70 (± 21) min                                | 127 (± 22) min                               | \(p < 0.0001\)                      |
| Blood loss                  |                                               |                                               | Mann-Whitney \(U\) test \(p < 0.0001\) |
| Mean (± SD)                 | 220 (± 130) ml                               | 460 (± 210) ml                               | \(p < 0.0001\)                      |
| Days of hospitalization     | Mean (± SD)                                  | 6 (± 2) days                                 | Mann-Whitney \(U\) test \(p = 0.004\) |
| Mean (± SD)                 | 8 (± 2) days                                 |                                               |                                      |

| Table 3 Radiological outcome | Intralesional resection + bone cement (\(n = 25\)) | Intralesional resection + bone cement + proximal humerus plate (\(n = 17\)) | Statistical test with \(p\) value |
|-----------------------------|-----------------------------------------------|------------------------------------------------|--------------------------------------|
| Initial tumor size          |                                               |                                               | Student \(T\) test \(p = 0.005\)            |
| Mean (± SD)                 | All 4.2 (± 1.5) cm                            | 6.6 (± 3.0) cm                               |                                      |
| Enchondroma                  | 4.2 (± 1.7) cm                               | 7.6 (± 3.0) cm                               | \(p = 0.003\)                      |
| ACT                         | 3.7 (± 1.2) cm                               | 5.3 (± 1.1) cm                               | \(p = 0.24\)                      |
| Enchondroma or ACT          | 4.3 (± 0.6) cm                               | 4.5 (± 2.9) cm                               | \(p = 0.26\)                      |
| Cavity size after curettage |                                               |                                               | Student \(T\) test \(p = 0.0001\)       |
| Mean (± SD)                 | All 5.7 (± 2.1) cm                            | 9.6 (± 3.2) cm                               | \(p < 0.0001\)                |
| Enchondroma                  | 5.7 (± 2.2) cm                               | 10.2 (± 2.5) cm                              | \(p < 0.0001\)                |
| ACT                         | 4.7 (± 1.1) cm                               | 5.6 (± 0.4) cm                               | \(p = 0.44\)                    |
| Enchondroma or ACT          | 6.9 (± 0.3) cm                               | 9.0 (± 4.6) cm                               | \(p = 0.65\)                    |
| Scalloping + soft-tissue extension score |                                               |                                               | Mann-Whitney \(U\) test \(p = 0.71\)          |
| Mean (± SD)                 | All 1.9 (± 0.9) cm                           | 2.0 (± 1.0) cm                               | \(p = 0.71\)                    |
| Enchondroma                  | 1.8 (± 1.0) cm                               | 1.8 (± 0.9) cm                               | \(p = 0.81\)                    |
| ACT                         | 2.3 (± 0.6) cm                               | 3.0 (± 1.4) cm                               | \(p = 0.52\)                    |
| Enchondroma or ACT          | 2.0 (± 1.0) cm                               | 2.0 (± 1.0) cm                               | \(p = 1.0\)                     |
Difference in recurrence was not significant (chi-square test $p = 0.4$).

**Complications**

No complications were found in the group without osteosynthesis. In the osteosynthesis group, two of the 17 patients had complications. This difference did not reach statistical significance but showed a trend (chi-square test $p = 0.08$). As complications, we found one peri-implant fracture which was, however, related to a fall with adequate trauma. It was treated by re-osteosynthesis with a longer plate. Another patient needed revision surgery due to a postoperatively found intra-articular screw.

**Discussion**

Intralesional resection with vigorous curettage and filling the cavity with bone cement have been described in several studies to treat enchondromas and low-grade malignant ACT resulting in sufficient oncological safety and excellent function [1, 7–9, 11, 14–21]. Stability after curettage is an important issue, as postoperative fractures are frequently described [10, 13, 17–19]. Biomechanically demanding locations such as the femur are at highest risk [10, 19], but fractures also occur in the upper extremity including the proximal humerus [13, 18, 23]. Bone cement compound osteosynthesis may increase stability [23, 24], but it remains unclear whether this approach is beneficial at the proximal humerus or not [25]. Advantages and disadvantages have not been analyzed so far, so we for the first time compared intralesional tumor resection and bone cement filling with and without additional plate osteosynthesis at the proximal humerus.

Preoperative tumor size and size of the tumor cavity after curettage were significantly different with smaller lesions in patients without osteosynthesis and larger lesions in patients with additional osteosynthesis. Hence, surgeons will have to judge larger lesions to be at higher risk for instability or postoperative fracture and therefore decide to implant additional osteosynthesis more often in these cases. All lesions were radiologically judged as aggressive, without significant difference of the groups in the scalloping and soft-tissue extension score. Consequently, size will have predominantly influenced the decision towards osteosynthesis. Besides generally high radiologic aggressiveness of the lesions of the present series, only 11 lesions were histologically diagnosed as ACT or potential ACT compared to the majority of histologically benign diagnosed enchondromas. Valid differentiation between ACT and aggressive enchondromas by histology might be questionable. This is supported by several other studies, documenting difficult or even impossible differentiation of both entities [1, 3–5].

In case of additional osteosynthesis, surgery time was significantly longer with significantly more blood loss and longer hospital stay afterwards. Hence, it would be beneficial to avoid additional osteosynthesis. Significant clinical and functional differences regarding MSTS score, DASH score, pain, and satisfaction were not found after intralesional resection with or without osteosynthesis. Both treatment groups had excellent clinical outcome with high satisfaction, low pain, and only minimal functional impairments. Compared to other studies on intralesional resection strategy, our MSTS score results were similar and even slightly better [1, 9, 11, 13, 16].

As reported by others, this strategy not only maintains excellent function but also offers sufficient oncological safety [1, 7–9, 11, 14–21]. In our series, no ACT recurrence but one enchondroma recurrence was found 4 years after intralesional tumor resection with bone cement filling without osteosynthesis. Patients with additional osteosynthesis did not show recurrence. Higher recurrence rate in cases without additional osteosynthesis might theoretically be explained by less radical tumor resection, as the surgeon might have been afraid

**Table 4 Clinical and functional outcome**

|                       | Intralesional resection + bone cement ($n = 19$) | Intralesional resection + proximal humerus plate ($n = 12$) | Statistical test with $p$ value |
|-----------------------|------------------------------------------------|----------------------------------------------------------|--------------------------------|
| **Satisfaction**       |                                                |                                                          |                                |
| Mean ($\pm SD$)        | 9.2 ($\pm 1.5$)                                | 9.2 ($\pm 0.9$)                                          | Mann-Whitney $U$ test $p = 0.5$ |
| rating scale 0–10      |                                                |                                                          |                                |
| **Pain**               |                                                |                                                          |                                |
| Mean ($\pm SD$)        | 1.0 ($\pm 1.7$)                                | 1.9 ($\pm 1.8$)                                          | Mann-Whitney $U$ test $p = 0.1$ |
| rating scale 0–10      |                                                |                                                          |                                |
| **DASH score**         |                                                |                                                          |                                |
| Mean ($\pm SD$)        | 6.0 ($\pm 11.8$)                               | 11.0 ($\pm 13.2$)                                        | Mann-Whitney $U$ test $p = 0.2$ |
| rating scale 0–100     |                                                |                                                          |                                |
| **MSTS score**         |                                                |                                                          |                                |
| Mean ($\pm SD$)        | 29.0 ($\pm 1.7$)                               | 28.7 ($\pm 1.1$)                                         | Mann-Whitney $U$ test $p = 0.3$ |
| rating scale 0–30      |                                                |                                                          |                                |
of instability. Difference in recurrence was not significant but valid comparison is not possible due to only one found recurrence. As a disadvantage of additional osteosynthesis, postoperative MRI images showed higher artifacts although the plates are made of titanium. This problem, however, can be sufficiently solved with the latest MRI technology using artifact suppressing algorithms [28], so local tumor recurrences can still be ruled out with sufficient reliability. We did not find pulmonary metastases until the final follow-up. ACT as a grade I malignancy and even enchondromas offer potential risk for transformation into higher grade chondrosarcomas which has been reported with percentages from 1 to 9% [5, 6, 29]. The highest risk is known for tumors of the axial skeleton, pelvis, and truck; hence, intralesional therapy seems less appropriate in these cases [1, 12, 13, 30]. In case of ACT recurrence, a more aggressive phenotype is expected with higher risk for transformation into higher grade chondrosarcoma, so most authors recommend wide resection in such cases [22, 30]. The lowest risk is expected for ACT of the long bones of the appendicular skeleton without statistical evidence for differences between the upper and lower extremity [30]. Nevertheless, the literature more often reports on recurrences at the femur and tibia [1, 13, 20] compared to the humerus [11]. In the series of Andreou et al. including 225 patients with ACT, 46 lesions were in the upper extremity but no transformation into higher grade chondrosarcoma or pulmonary metastases were found contrary to 5 transformations into grade II chondrosarcoma with additional pulmonary metastases in ACT located in the femur and tibia. Analysis of metastasis-free survival, however, was not significantly different although the overall number of analyzed cases was very high in this multicenter study [30].

We had two complications, and they were only found after additional osteosynthesis. The intra-articular screw can be directly related to the procedure. The postoperative fracture had an adequate trauma and hence cannot be clearly attributed to the osteosynthesis. In case of additional osteosynthesis, superior stability would be expected, but our data cannot prove this. No fractures were found after intralesional excision without additional osteosynthesis, but one fracture was found although osteosynthesis was added. This can be interpreted differently. First, the theoretical stability increase of additional osteosynthesis might be overestimated. Second, surgeons might have used additional osteosynthesis more often than needed, to achieve the highest safety. The literature does not give sufficient answers on whether additional osteosynthesis should be used or not. A series including 10 humerus cases treated with intralesional resection and bone cement filling without osteosynthesis reported no fractures [9]. Dierselhuis et al. found 11 fractures in 108 cases but there were only 33 humerus cases, of those 2 had fractures, and detailed information on prior osteosynthesis is not given [18]. Kim et al. analyzed 36 cases and found 4 fractures only located at the femur whereas no fracture was found for the 23 humerus lesions although additional osteosynthesis was not used [19]. In our series, the largest cavity treated without osteosynthesis was 12.4 cm compared to 15.2 cm for the largest cavity in the osteosynthesis group. As we did not find fractures in the group without osteosynthesis, we can only conclude that our mean cavity size of 5.7 (± 2.1) cm was in a safe zone at the proximal humerus and that even larger sizes up to 12.4 cm did not show problems without osteosynthesis. For better interpretation, higher patient numbers would be beneficial and further biomechanical cadaver studies should be performed to achieve objective data considering primary stability of bone cement fillings with and without additional osteosynthesis at the proximal humerus. So far, no data is available in the literature.

Conservative follow-up without surgery might be an important alternative [6], although psychological and socioeconomic aspects of leaving an aggressive tumor inside the body with need for long-time radiologic follow-up have to be considered [31]. Conservative strategy is not further discussed here, as the goal of the present study was comparison of two surgical strategies.

Several limitations have to be mentioned. Due to the retrospective study design, pain and function were not systematically evaluated preoperatively, so postoperative clinical success could not be compared to the preoperative situation. Furthermore, decision for or against additional osteosynthesis was done individually without a standardized decision protocol with a potential selection bias.

Conclusion

Our series documents oncologically safe and clinically successful outcome no matter if intralesional tumor resection was performed with bone cement filling alone or with additional osteosynthesis at the proximal humerus. Compound plate osteosynthesis with the intention to increase stability did not reveal significant clinical disadvantages besides longer surgery time, more blood loss, and longer hospitalization. Further biomechanical evaluations and randomized studies should be initiated.

Abbreviations

ACT: Atypical cartilaginous tumor (chondrosarcoma grade I according to older WHO classification); DASH score: Quick disabilities of the arm, shoulder, and hand score; LCP: Locking compression plate; MSTS score: Musculoskeletal Tumor Society score

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Availability of data and materials
Additional datasets from the current study are available from the corresponding author on reasonable request.

Authors’ contributions
GO developed the study design; collected, analyzed, and interpreted the data; and wrote the manuscript. VL collected the data and participated with the analysis, interpretation, and drafting. JL helped with the data collection and analysis. SG participated with the statistical analysis. CM, JF, FR, and BL participated with the study design, interpretation of the findings, and drafting of the final manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate
Approval was given by our ethical committee of the University of Heidelberg, Germany (votum number S-053/2017).

Consent for publication
Informed consent was obtained from all patients.

Competing interests
The authors declare that they have no competing interests.

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