

## Shared decision making: Does a decision aid support patients with an atypical cartilaginous tumor in making a decision about treatment

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### ABSTRACT

**Objective:** Due to new insights, atypical cartilaginous tumors (ACTs) of the long bones are no longer considered malignant and treatment is shifting from surgery to active surveillance. We developed a decision aid in order to support in shared decision making on treatment.

The aim of this study is to evaluate the treatment preferences of patients with an ACT in the long bones.

**Methods:** During thirty-four months, patients received a decision aid digitally with information about the disease, the treatment options, and the risks and benefits of active surveillance and surgical treatment. The given answers to patients' preference questions were evaluated qualitatively in relation to the final choice of treatment.

**Results:** Eighty-four patients were included. None of the patients who preferred active surveillance later underwent surgery. Only four patients underwent surgery based on patient preference.

**Conclusion:** In our experience the decision aid is useful for shared decision making as it provides the patient with information and the clinician with insight into patient's preferences. The preference for treatment generally corresponds to the eventual treatment.

**Innovation:** When treatment changes, due to new insights, a decision aid seems helpful for both patients and clinicians to discuss the treatment that best suits the patient's situation.

### 1. Introduction

Cartilaginous tumors such as enchondromas and atypical cartilaginous tumors (ACT) are often located in the long bones [1,2]. Due to more frequent imaging, in patients with joint-related symptoms, incidental detection of these tumors has increased over time [3]. This often leads to a referral of the patient to an orthopaedic oncology center for further diagnostics and treatment advice.

Enchondromas are benign chondroid tumors. ACTs are tumors that can be locally aggressive, but metastasizing or upgrading of an ACT is extremely rare [4]. The present classification is intermediate type of tumor, not a malignancy. ACTs and enchondromas can have similar radiographic findings, which makes it very difficult to distinguish between these two diagnoses [5,6]. Until recently surgery was standard of care for these tumors. In most centers, treatment consisted of intralesional curettage with local adjuvant treatment such as phenolisation or cryosurgery [7,8]. The remaining bone cavity was filled with cement or bonegraft, with or without prophylactic plating. This was followed by a 3 to 4 month rehabilitation period, with physiotherapy to

rehabilitate to a presurgical functional level [9]. The post-operative follow-up period ranged from 2 to 5 years with radiographic imaging [5].

Advantage of surgery is that the tumor is removed and follow-up is limited. There are however several disadvantages, such as the associated prolonged recovery, poorer functional results and the risk of complications (e.g., wound infection and bone fracture) [9].

Due to new insights on the natural course of these tumors, active surveillance with radiographic follow-up instead of surgery is nowadays becoming more common, in order to prevent overtreatment [6]. Literature on active surveillance is promising as no malignant transformation is reported and tumor growth seems to occur only in a small group of patients [2,10,11]. A retrospective study of Deckers et al. showed that only 6% of the patients who were included for radiographic follow-up of an ACT eventually underwent surgical treatment on medical grounds, such as pain, tumor growth or radiological changes [12].

Considering the risks and benefits for a patient, this does not imply an increased health risk for the patient. The burden on the patient is posed by periodically undergoing radiographic imaging, such as an MRI.

Abbreviations: ACT, Atypical cartilaginous tumor.

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Indications for surgery today are based on medical ground (i.e., pain and tumor growth), or on the patient's wishes [12]. If the psychological burden, due to the tumor in the bone, dominates a patient's daily life, this may be a reason for surgery.

The recent new insights on the behavior and treatment of ACTs call for adjustment of patient information. In order to make a well-informed decision about the preferred management (active surveillance or surgical treatment) of an ACT, patients need to be well counselled with complete and updated information.

It is well known that patients immediately forget 40% to 80% of medical information provided by healthcare professionals, and that nearly half of the information is incorrectly remembered [13]. This is due to several factors, including the use of understandable language, the use of medical terminology and whether information is supported by written material. Patient related factors affecting memory include age, anxiety, level of education and specific expectations [14].

To help patients understand and remember the medical information and make a well-informed decision on treatment we have developed a decision aid for patients with an ACT in the long bones. Decision aids are designed to assist patients and clinicians in making informed decisions about possible management options, and to support the process of shared decision making [15]. Offering a choice in treatment options increases involvement in decision making and leads to better informed patients [16]. Decision aids improve knowledge, reduce indecision, and improve agreement between values and choices [17].

The health professional and the patient are able to discuss the patients' thoughts, preferences and values, and this assists shared decision making [18]. This improves patient satisfaction, and results in increased patient empowerment, and in an increased confidence in the health professional-patient relationship [19].

The aim of this study is to evaluate the preferences of patients in relation to the eventual treatment for ACT in the long bones and to share our experience with the implementation of a decision aid.

## 2. Methods

A decision aid for patients with an ACT was developed with the goal to inform patients, to support patients and healthcare professionals in the shared decision and to avoid unnecessary treatment.

### 2.1. Decision aid development

The content of the decision aid was based on the latest insights of the behavior and treatment of ACTs and the Dutch guidelines for developing a decision aid [15,20]. This is in line with International Patient Decision Aid Standards (IPDAS) [21]. The content was written by physicians, a physician researcher and a nurse practitioner and it has been checked for readability by the communications department.

The first section of the decision aid consisted of information about the diagnosis and treatment. The information on treatment options, both active surveillance and surgical treatment, was supplemented with an overview of the risks and benefits of each option. In the second section, knowledge questions were formulated for the patient to check whether he or she understood the information of the decision aid. The results and correct answers to these questions were visible only to the patient. The final section consisted of questions about patients' values and preferences. The answers to these questions formed the basis for the following conversation between the patient and the health professional about the treatment that best suited the patient's situation.

After implementation in April 2018, the decision aid was presented to and adjusted by the Dutch Patient Association [22]. After the Dutch Patient Association evaluated the content of the decision aid, one of the questions to determine patients' values (no 5) was changed, wording it more positively.

Initial value and preference questions:

1. I am concerned about the diagnosis cartilaginous tumor
2. I do not mind undergoing surgery

3. I do not mind having frequent MRIs
4. I do not mind being unable to put weight on my arm / leg for a short period of time
- 5a. Leaving / not operating on my cartilaginous tumor frightens me
6. At this moment my preference is: Follow-up / operation / I do not know yet

Changed question from 14-12-2018:

- 5b. Active follow up of my cartilaginous tumor reassures me.

Since the decision aid was developed as a digital tool, it was incorporated in the patient electronic health record of the hospital. All patients had access to their electronic health record, allowing them to receive the information, and answer the questions digitally. The answers to the questions to determine patients' values and preferences were saved in the electronic health record, and could be easily reviewed by the patient and the health professional.

### 2.2. Data collection procedure and analysis

Between April 2018 and January 2021, patients, who were diagnosed with an ACT in the long bones at our orthopaedic oncology department, received the digital decision aid in his or her electronic health record after their first physical consultation at our outpatient clinic.

One week after the consultation at the outpatient clinic, consultation by phone was performed by the physician or nurse practitioner. During this consultation, remaining questions could be answered by the clinician, and the patient was invited to discuss the values and preferences with the physician or nurse practitioner. At the end of the consultation, a final shared decision was made on the preferred management, and this was documented in the patient's health record.

For this study, all records were analysed, to determine the patients' preferences and what management had eventually been performed. The minimum follow up, after diagnosis, was nine months. For patients with active surveillance, the follow up consisted of MRI at 6 months after diagnosis, and for patients who were included in the first two years of this study, also at 18 months after diagnosis.

In addition to the analysis of the answers to the questions related to the patients' values and preferences, the data analysis also included information on patient demographics and tumor-related data.

Retrospectively, all health records were reviewed to determine how patients had experienced the information provided by the decision aid.

## 3. Results

During the inclusion period of 34 months, 84 patients (55 female and 29 male) received the decision aid and answered the preference questions. The age of the patients ranged from 20 to 78 years, with a mean of 54 years. In 39 patients the tumor was located in the femur, and in 33 patients in the humerus. Other locations included the tibia ( $N = 8$ ), fibula ( $N = 2$ ) or other long bone ( $N = 2$ ).

Sixty-nine patients (80%) were referred to our orthopaedic oncology center based on an incidental finding on radiograph or MRI scan. Other referral indications were pain ( $N = 9$ ), referral from an other orthopaedic oncology center for second opinion ( $N = 2$ ), a palpable lesion ( $N = 2$ ), and unknown/other ( $N = 4$ ).

In total, 11 of the included patients (13%) underwent surgical treatment, because of pain ( $N = 5$ ), growth of the tumor ( $N = 2$ ) or patients' preference ( $N = 4$ ).

### 3.1. Outcomes of the questions to determine patients' values and preferences

As reported in Fig. 1, half of the patient group was concerned about the diagnosis of ACT.

One third of the patients did not mind undergoing surgery followed by restricted weightbearing for a short period of time. The majority did not mind having frequent MRIs.

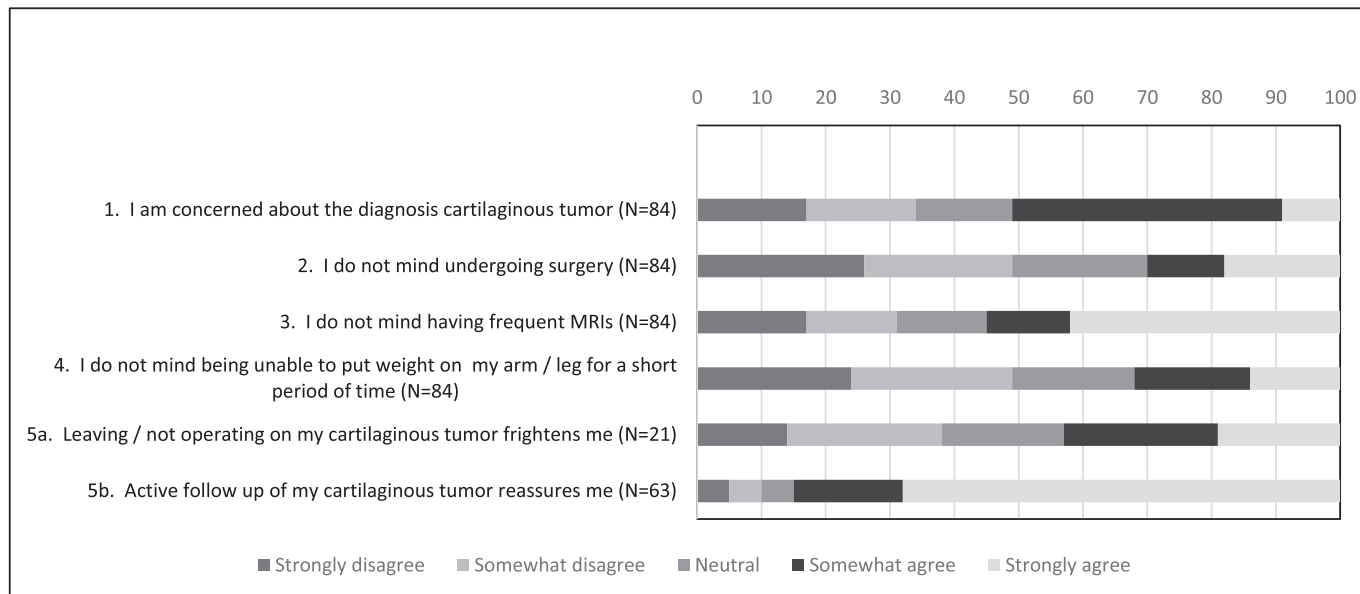


Fig. 1. Answers of the patients values and preferences (in %).

Among the patients who received the question about leaving / not operating on the tumor, there were equally mixed feelings about leaving the tumor in place.

For the majority of the patients active follow up of the ACT provided a feeling of reassurance. This was reflected in the results for the preference for treatment questions.

Sixty-eight percent of all patients preferred follow up, 14% preferred operation, and 18% did not have a preference at the first contact moment after providing the decision aid (Fig. 2).

### 3.2. Outcomes of treatment

During the study period, a total of eleven (13%) patients underwent surgical treatment, seven (8%) of which based on physical symptoms / pain or tumor growth.

None of the patients who preferred active surveillance, underwent surgery during the follow up period.

Nine of twelve patients who initially indicated a preference for surgical treatment, proceeded to surgery. Six out of these nine underwent surgical treatment based on physical symptoms / pain or tumor growth, and three of them based on patient's wish.

Three of twelve patients who initially indicated a preference for surgical treatment, proceeded to active surveillance. During the telephone consultation with the clinician, during which the patients' values and preferences

were discussed, these patients, in a shared decision, changed their initial preference for treatment.

Two of fifteen patients, who initially had no specific management preference, underwent surgery, after shared decision-making. In one patient it was based on his personal wish and in the other case on physical symptoms / pain.

### 3.3. Outcomes related to patients experience of the decision aid content

Information on how patients experienced the content of the decision aid was documented in 36 electronic health records. The responses showed that the majority of patients felt that the information in the decision aid was found to be comprehensive and understandable, that it contributed to the verbal information provided during the hospital visit, and that it supported patients in making a management decision.

Only two patients stated that the decision aid did not offer new insights, as the information during the hospital visit had been clear enough.

## 4. Discussion an conclusion

### 4.1. Discussion

This study shows that, after receiving information provided by the decision aid, patients are able to indicate their preference and that, for most patients, this preference corresponds to the eventual management.

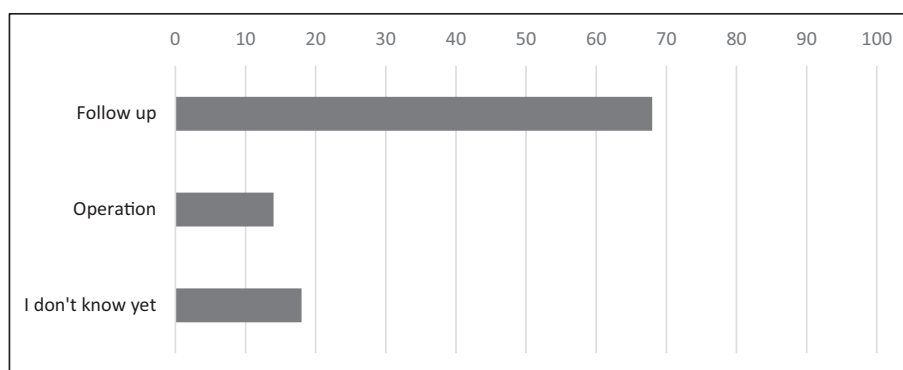


Fig. 2. Answers of the patients preference of treatment (in %).

With the introduction of the decision aid, we have sought to comply with the principles of shared decision making [23]. Information about the diagnosis and treatment options is provided, patient values and preferences are explored and discussed with the physician or nurse practitioner, and a final decision is made. However, the decision aid is just a tool to support patients in shared decision making and is a useful supplement, but it cannot replace the consultation in clinical practice [18].

Despite the explanation of the behavior of the tumor, most patients are concerned about the diagnosis. However, a majority finds undergoing surgery objectionable and most do not mind having frequent MRI scans. When a well-informed decision choice is made to follow the course of the tumor over time and to avoid an operation, an operation is usually not required, unless there are changes in the appearance on imaging or the patient develops symptoms. Therefore, we have been able to refrain from surgery in patients with ACTs in over 90% of the cases, where surgery had previously been the standard of care. This huge decrease in surgery rate can be contributed to both new pathophysiological insights as well as patient involvement and shared decision making. This results in a reduction of preventable surgical complication rates, avoidance of unnecessary post-operative rehabilitation, and reducing costs in healthcare [24-26].

During the study, on the advice of the Dutch Patient Association, one question of the decision aid was changed to ask the question with a more positive spin.

The way a question is formulated may influence the answers of respondents [27]. A positive wording leads to a more positive representation of the patient's opinion [28]. After changing the wording of the question, the previous answers to the originally worded questions do not automatically translate to the new question. However, it does provide information on how patients perceive the possible management.

The question (5a in Fig. 1) whether leaving / not operating on the cartilaginous tumor is frightening was answered with equal numbers of agree and disagree. After changing the question, the majority indicated that conservative management and monitoring of the tumor was reassuring. This also corresponded to the final management choice of most patients. The sum of the provided answers gave the physician and nurse practitioner information about the patient's values and preferences, and this facilitated final decision making.

The limitation of our study is that patients were not initially involved in the development of the decision aid. During implementation, we realized this and presented the decision aid to the Dutch Patient Association for comment and review. This does not substitute for direct input from the main users/target of the decision aid and could have prevented modification of a question.

Furthermore, we did not record the patients expectations and preferences during their first consultation in our outpatient clinic, before providing the information. Although it is known that patients often do not have clear preferences at the outset, we have no insight into the possible change of mind of the patient by the provided verbal and written information [29]. As a control group of patients who received only verbal information was not available in this study, the actual impact of the additional written information on the final preferred management of the patient was not measured.

We also lack information about the impact this decision aid may have on the possible change of perception and understanding of the information given to the patients by the health professional during the initial consultation. Awareness of the content of the decision aid may lead to bias of the physician or nurse practitioner, allowing them to provide information more extensively after the implementation of the decision aid. Furthermore, the way a physician or nurse practitioner provided the information in the consultation, and whether the physician's choice or recommendation for treatment was discussed with the patient, was not recorded. Communicative behaviors of physicians have great influence on certain patient outcomes [29]. Patients are more likely to choose recommended treatments when communication is satisfactory [30]. On the other hand, we know that after an initial consultation with a physician, very little medical information is remembered, and that the amount of retained information will decrease over time [31]. Studies have shown that written information

supports verbal information and contributes to a better consideration of choices [13,32]. This is confirmed by the patients' answers to the question on how the information in the decision aid was experienced.

Evaluation of patients' experience with the decision aid was documented only in 36 files. The vast majority of those patients were positive. For the remaining patients, we have no formal data, but we experienced that the patients were very positive about it during the follow-up period. Therefore, we do not think that the experience of the 36 patients is a selection bias.

This study provided information about patients' values and preferences related to the treatment of ACT. Further studies should focus on the actual effect of the decision aid on joint decision-making. The Shared Decision Making Questionnaire (SDM-Q) is a reliable and brief instrument that can be used to assess the effectiveness of the implementation of the decision aid on shared decision making [33].

#### 4.2. Innovation

As result of new insights, medical treatments may change. Patients will have to be informed in an understandable way about these changes and the consequences for the treatment. This study shows that a decision aid is a good tool to properly inform patients about the new insights and different treatment options. It helps patients and clinicians discuss the treatment that best suits the patient's situation, in a shared decision.

#### 4.3. Conclusion

After implementation of the digital decision aid none of the patients who opted for active surveillance revised their choice during follow-up. We experienced that a decision aid helps to inform patients about their diagnosis and management options, and patients can make informed choices about their treatment preferences. Clinicians became more aware of patients' values and preferences so that they could reach a shared decision about treatment.

#### Declaration of Competing Interest

The authors have no funding or conflicts of interest to disclose.

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