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Quality of life, functional ability and physical activity in children and adolescents after lower extremity bone tumour surgery

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Chapter 9

GENERAL DISCUSSION

INTRODUCTION

The thesis describes quality of life (QoL), functional ability and physical activity after surgery for lower extremity bone tumour surgery. The patients included in the studies were children and adolescents aged 8 to 25 years, less than 5 years after surgery.

As malignant bone tumours are relatively rare, we applied a multicentre design, including five hospitals in The Netherlands. Because at the time the studies were conducted there was no disease-specific QoL-instrument for children and adolescents after lower extremity bone tumours, we developed and validated a new instrument (Bt-DUX questionnaire) for this purpose.

Finally, to make this instrument available for English speaking countries, we translated and cross-culturally adapted this questionnaire.

STUDY DESIGN

By comprehensively describing the outcomes after different surgical interventions, orthopaedic surgeons can better provide patients and their parents with information regarding the prognosis after various limb-salvage or ablative surgical techniques.

The design and interpretation of studies describing the outcomes of surgery for malignant bone tumours of the leg is however difficult, due to a number of reasons.

First, making direct comparisons of outcomes among different surgical groups within one study is hampered by the fact that the choice for a specific procedure is based on many factors, including tumour type, tumour extent and localization (above or below the knee), varying opportunities / resources for limb-salvage reconstruction, age, and/or the preferences of the treating surgeon, the patients and/or their parents. Because of the complexity of the treatment choice the employment of randomized controlled trials comparing different forms of surgery will be unlikely, so that all comparisons will be hampered by the potential impact of “confounding by indication”.

Comparisons of the outcomes of different studies describing the outcomes of surgery after malignant bone tumour of the lower extremity are also difficult to make because of variation in the definition and categorization of surgical interventions the use of varying research designs and measurement methods.

Second, although in the studies described in this thesis a multi-centre design was employed; the sample size was, as in many other studies, relatively small. This was, apart from the low incidence of the condition, also due to the considerable number of patients who died or were lost to follow up because of their deteriorating health status.

All of these observations substantiate the need for national and international collaboration in outcome research in this patient group. This will in the first place ensure sufficient patient numbers.

For these large scale studies, consensus on a core set of outcome measures to be recorded at fixed time intervals in every patient is needed. In particular attention has to be paid to a uniform and comprehensive description of the patient characteristics and course of the disease and its treatment, including: the tumour type, extent, and localization; the nature of the surgical procedure that was executed and the occurrence of surgical complications;

the pharmacological treatment before and after the operation and the occurrence of drug side effects and complications; and the occurrence of metastatic disease. In particular, considerations underlying the choice of specific surgical techniques have to be recorded in a standardized way.

Moreover, future studies should have a sufficient duration of follow-up, of at least 5-10 years [1]. This is needed because some of the procedures, particularly allograft and metallic devices, are known to have significant problems relatively early after surgery. Complications may include allograft infection, fracture, non-union or metallic device failures or problems with nerves or blood vessels, which may require additional surgery. Amputees may have had their prosthesis changed after surgery, and may not have reached their optimal level of functioning within the first years after surgery.

OUTCOME MEASURES

Regarding the outcome measures to be used in bone tumour surgery research, the currently applied QoL measures, such as; Short Form-36 (SF-36) [2], Quality of Life-Cancer Survivors (QoL-CS) [3], European Organization for Research and Treatment of Cancer Quality of life Questionnaire (EORTC QLQ-C30) [4], tend to target specific age-groups. Especially for studies among survivors of bone cancer who are often diagnosed during childhood and adolescence, this is a serious limitation. There are no single instruments suitable for children, adolescents and adults, so different instruments need to be used for patients in each age group. This will limit the comparability of the results within and among studies.

Moreover, in a prospective study like the one that is described in this thesis, patients may move to another age group during the study, thereby hampering the evaluation of changes over time. QoL measures with separate modules for children and adolescents like the TNO-AZL Children's Quality of Life Questionnaire (TACQOL) [5] and TNO-AZL Questionnaire for Adult's Quality of Life (TAAQOL) [6] and the Cancer module of the Pediatric Quality of Life Inventory (PedsQL) [7] could probably solve this problem.

Overall, the comparability among different generic QoL instruments is limited. In our prospective study, we have tried to solve this problem by computing physical and mental summary scores. For the SF-36 this is a commonly accepted procedure [2]. However, despite its application in a previous study [8], this method has not been validated for the TACQOL and TAAQOL questionnaires. Therefore, further research on the composition, validity and reliability of these summary scales and the correlation between the TACQOL for children and TAAQOL for adolescents needs to be executed.

Since lower extremity bone tumour surgery may have considerable consequences within the domains physical activity and sports, body image and cosmetic appearance, it is important to include these domains in disease specific QoL instruments used in this population. In generic measures of QoL that are commonly applied in this patient group, such as the SF-36 these domains are hardly represented. For this reason, the disease specific Bt-DUX questionnaire was developed. Despite a sufficient practical applicability, internal consistency and discriminate validity, this instrument still has to be tested in larger patient populations.

For the evaluation of functional ability, a bone cancer surgery specific questionnaire; the Toronto Extremity Salvage Score (TESS) [9] has been used in the majority of studies. However, the TESS has relatively little focus on tasks that are typical for younger patients. Two measures; the Paediatric Outcomes Data Collection Instrument (PODCI) [10] and the Activities Scale for Kids (ASK) [11], have been previously used in assessing functional outcome in paediatric sarcoma patients and could probably be considered for further evaluation of content and measurement properties in adolescent and young adult patients [12].

Regarding the measurement of physical activity, two instruments were used in the studies described in this thesis: a subjective instrument, the Baecke questionnaire [13], and an objective instrument, the ActiLog[®] accelerometer [14]. With respect to the Baecke questionnaire it should be noted that this instrument was developed for healthy children or adolescents and not validated for use amongst cancer patients.

It remains to be established whether other questionnaires, such as the Physical Activity Scale for Individuals with Physical Disability (PASIPD) [15], the Short Questionnaire to Assess Health-enhancing physical activity (SQUASH) [16] or the International Physical Activity Questionnaire (IPAQ) [17] could perform better in this population. However, because of the diversity in available questionnaires and the disappointing results on reliability and validity of the currently available instruments among children and adolescents [18], the decision on which instrument to use is still a challenge.

In our studies an activity monitor was, in contrast with other outcome measures, not able to detect changes over time. It is not clear to what extent the positioning of the activity monitor, at the non-affected side, and the asymmetrical walking pattern of most patients may have influenced these results. Furthermore, one-dimensional accelerometers have the disadvantage that they are influenced by factors such as body size (step length) and speed of locomotion [19], which makes them probably less appropriate for assessing differences in physical activity within populations with subjects who are not similar regarding age and body mass.

It remains to be established whether other accelerometers than the one used in our study or a pedometers that proved to be valid among healthy children and adolescents [20] would perform better in this population.

PATIENTS

Regarding the measurement of outcomes in this patient group in general, the issue of gender differences in appreciation of the functional and cosmetic results of the surgery has seldom been taken into account. Limitations in physical functioning and sports may probably be more detrimental for the QoL of males compared with females. In contrast, amputation may have a worse impact on QoL for females, assuming the greater importance of body image [21,22]. Clarity about gender differences on the appraisal of the functional and cosmetic outcome could be of importance in the decision process concerning the surgery.

Furthermore, it is also necessary to include difficulties in the entrance to the adult world of work and social relationships. Outcome measures may need to be sensitive to these and other developmental issues. There is no empirical evidence regarding how well children

adapt to surgery compared with adults. This kind of research is needed in order to develop appropriate rehabilitation programs and enhance successful adaptation among younger patients and to consider service needs for child patients.

Another aspect of outcome that should be taken into account is the patients' expectations of their outcome. It is often clinically observed that patients undergoing limb-salvage have high expectations of what they should be able to do and what will be the cosmetic results of the surgery [23]. This could imply that they can be more disappointed with the functional and cosmetic results. Adversely, amputees are often more resigned to the limitations and mutilating effect of the surgery they expect to undergo [22], and the sportive and functional abilities and the social acceptance they experience could surprise them positively.

These observations imply that more research into the impact of expectations management on quality of life, functional ability and physical activity levels after bone tumour surgery is needed.

REHABILITATION

In daily practice, most patients and their parents will be treated by an interdisciplinary team consisting of healthcare providers with various professional backgrounds. The constitution of this team and the individual tasks within this team may widely vary among centres and within centres during the treatment process.

It is surprising that so little is known about the effectiveness of rehabilitation after surgery. None of the outcome studies in this patient group comprised a description of the contents of the rehabilitation program. As a first step it would be helpful to uniformly and systematically describe and compare the process, structure and outcomes of currently employed rehabilitation programs for the different surgical interventions in this patient group.

CONCLUSION

The research described in this thesis has contributed to the available information on QoL, functional ability and the amount of physical activity in children, adolescents and young adults after lower extremity bone tumour surgery. The results may facilitate the pre-operative informed decision making process of patients, their parents and the surgeon with the ultimate aim of choosing the type of surgical procedure that will balance maximum potential for cure with an acceptable aesthetic outcome, long-term mobility, and quality of life for an individual patient [24].

Moreover, the results of the studies described in this thesis underline the need for larger, longitudinal studies with adequate numbers of patients, an adequate description of the disease and its treatment and complications, using a predefined set of outcome measures, an appropriate description of the considerations underlying decisions the surgical techniques employed, and a long-term duration.

Apart from studies aiming to further improve the surgical and pharmacological treatment, more research into the optimization of rehabilitative management is needed.

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