# The frequency and rehabilitation of childhood malignant diseases in Hungary

Ph.D. Thesis

# Szabolcs Török, M.D.

Semmelweis University School of Ph.D. Studies



Supervisor: Teodóra Tomcsányi, M.A., D.Sc.

Opponents: Katalin Hegedűs, M.A., Ph.D. Gergely Kriván, M.D., Ph.D.

Doctoral comprehensive exam chairman: Kornél Sipos, M.D., M.A., D.Sc. Doctoral comprehensive exam board: János Gombocz, M.A., D.Sc. Miklós Garami, M.D., Ph.D. András Fogarasi, M.D., Ph.D.

Budapest

2006.

## INTRODUCTION

Malignant diseases are the second leading cause of death in children in Hungary and the incidence shows an increase according national and international data. The survival rate shows an increase as well, but it has been achieved only via the use of more intensive and more drastic treatment protocols. Due to these changes the number of survivors is increasing in the society, and medical professionals need to deal with an increasing number of side effects. The rehabilitation of paediatric oncology patients needs to cover not only the somatic care, but the psychosocial aspects of survivorship as well. The most important psychological and psychosocial side effects are the following: increased anxiety and feeling of isolation; loosening of peer relationships; drop out from school; family structure and family functional changes; changes in body image; regressive emotions and behaviour; depression and other mood changes. The presented results of this thesis are concerned with the frequency of childhood malignant diseases in Hungary (incidence, prevalence), and with the outcome effectiveness of special summer camps aiming at their psychosocial rehabilitation.

#### AIMS OF THE STUDY

I. Comprehensive analysis of the incidence, prevalence, and relative frequency of childhood malignant diseases for the 10-year period of 1993-2002 and the extension of the analysis for 30 years regarding

the subgroup of children with leukaemia (dating back to 1973).

II. Determination of the prevalence of 5-19 year old children with malignant diseases and in need of rehabilitation.

III. Introduction to the availability of opportunities for psychosocial rehabilitation in Hungary. Analysis of the outcome effectiveness of a therapeutic recreational camping programme. Determination of the effect of camp experience on campers' self-esteem, self-efficacy, and trait anxiety.

## **METHODS**

## I. Source of data and methods of frequency analyses.

The National Paediatric Cancer Registry of Hungary was the source of data for our survey. Children younger than 15 years were included in the analysis. The number of analysed cases was 2,436 for childhood malignant diseases between 1993-2002 and 2,204 for leukaemia cases between 1973-2002. We used standard descriptive epidemiological methods for the determination of incidence, incidence trend, and relative frequency. Survival analysis was performed by the Kaplan-Meier method.

# II. Methods of prevalence analyses.

At first we defined our target population for the participation in a rehabilitation programme. We considered the target population as those 5-19 year old children who are hospitalized at least once a year as a consequence of their malignant disease. Thereafter we calculated the prevalence of the above-defined children using the discharge codes of hospitals for the calendar year 2002. The data for the analysis were provided by the Ministry of Health (the GYÓGYINFOK database).

#### III. Methods of the outcome effectiveness analysis.

We conducted an analysis with three internationally accepted and widely used questionnaires, which were previously standardized and validated into Hungarian (Rosenberg Self-Esteem Scale, Global Perceived Self-Efficacy Scale, Trait form of the State-Trait Anxiety Scale). Measures were administered at three separate time periods: On arrival at camp ("pre-camp"); at the end of camp ("post-camp"); and 2 months after camp ("follow-up"). Apart from time, the independent variables were age, gender, diagnosis, time from diagnosis, and previous attendance in camp. To test the differences between the subgroups with lower and higher initial scores, we divided our sample at the median. In order to examine any changes in children's performance on the measures of Self-esteem, Selfefficacy, and Trait-anxiety over the 3 time periods, a series of repeated measures analysis of variance (ANOVAs) were performed. To compare the means of variables in pairs we used two-sample

paired t-tests. The participants of camp (n=97) were children with malignant diseases and children with diabetes.

#### **RESULTS AND CONCLUSIONS**

## I. Results of the frequency survey.

1. The absolute number of registered cases in the National Paediatric Cancer Registry over the analysed 10-year period (1993-2002) ranged between 218 and 259 (average: 244). This means that we can expect this number of new patients diagnosed with malignant diseases in children over a calendar year in Hungary.

2. The incidence of childhood malignant diseases varied between 131 and 140 per million person-years in the 0-14 age group. The annual average increase in incidence was + 1.1%. The magnitude of Hungarian incidences and the corresponding incidence trend is similar to the results measured in other European countries.

3. The most common childhood malignant diseases are cases of leukaemia, with a relative frequency of 27,8% in Hungary. Then follows tumours within the central nervous system with 26,6%, lymphomas with 11,3%, and sympathetic nervous system tumours with 10,0%. The sequence of frequency distribution in Hungary is similar to other European countries. A notable speciality of the Hungarian data is the relatively high proportion of central nervous

#### system tumours.

4. The 5-year overall survival for all childhood malignant tumours was 65,2% in Hungarian patients diagnosed between 1988 and 1997 (n=2146). This means that in Hungary, nearly two-thirds of all childhood malignant diseases can be cured. The Hungarian survival rate is close to those found in Western European countries.

5. The absolute number of childhood leukaemia cases registered in the National Paediatric Cancer Registry over the analysed 30-year period (1973-2002) ranged between 47 and 92 (average: 73). According to our results this is the anticipatory number of new cases diagnosed with leukaemia in childhood over a calendar year in Hungary.

6. The incidence of childhood leukaemia varied between 33 and 39 per million person-years in the age group 0-14 during the analysed 30 years period (1973-2002). The annual average increase in incidence was + 0.71%. The magnitude of the Hungarian incidence and incidence trend regarding the leukaemia disease group is similar to the results measured in other European countries.

# II. Results of the prevalence survey.

1. The number of 5-19 year old children who were hospitalized due to their malignant diseases during the calendar year 2002 in Hungary

was 1,080. Taking into consideration the known Hungarian epidemiological data, survival rates and follow-up protocols, this result correlates well with the expected or estimated numbers. The detected prevalence provides a good orientation for the estimation of rehabilitation needs.

## III. Results of the outcome effectiveness survey.

1. Our results demonstrated the positive psychosocial effects of participation in a special summer camp programme for children with malignant diseases, especially in the areas of self-esteem and self-efficacy. The positive effects proved to be independent from the diagnosis, age, gender, time from diagnosis, and previous participation in camp.

2. Comparing the pre-camp and post-camp results we detected a significant increase of the self-esteem of the campers. This short-run significant elevation of self-esteem was characteristic for both observed disease groups (oncology and diabetes).

3. In the subgroup of those campers whose self-esteem was low at the beginning of camp, the increase in self-esteem was more pronounced by the end of camp and this higher level of self-esteem maintained at the 2-month follow-up as well.

4. At the end of camp we measured higher perceived self-efficacy

scores, but this increase did not reach a level of significance and only expressed a tendency-like correlation. In contrast, in the subgroup of children with low initial self-efficacy, we detected a significant increase of self-efficacy and this higher level of self-efficacy did not decrease at the 2-month follow-up.

5. Adolescents living with cancer expressed higher levels of anxiety than their peers living with diabetes. Differences in the course and prognosis of the diseases may help explain these differences.

6. Trait anxiety levels did not change significantly from pre-camp to post-camp. However, they did increase significantly between the end of camp and the 2-month follow-up after camp. Findings regarding trait anxiety changes in the low and high initial trait anxiety subgroups were ambiguous. The differences between the identified subgroups and the paradoxical changes observed during camp indicate the need for further and more detailed research of this issue.

## STATEMENTS BASED ON OUR NEW RESULTS

1. The Hungarian epidemiological data of childhood malignant diseases (incidence, incidence trend, relative frequency) is similar to the results of other European countries.

2. In Hungary, nearly two-thirds of all childhood malignant diseases can be cured with the available, modern therapeutic tools. The

Hungarian survival rate is close to those found in Western European countries.

3. The number of 5-19 year old children with malignant diseases who are principally in need of psychosocial rehabilitation is approximately one thousand in Hungary.

4. Therapeutic recreational summer camp programmes for children with malignant diseases or diabetes promote the psychosocial rehabilitation of participants, especially in the areas of self-esteem and self-efficacy.

5. Adolescents living with cancer express significantly higher levels of anxiety than their peers living with diabetes. Differences in the course and prognosis of the diseases may help explain these differences.

## SUMMARY

Our research focused on the frequency of childhood malignant diseases in Hungary (incidence, prevalence), and with the outcome effectiveness of special summer camps aiming at their psychosocial rehabilitation. The incidence of childhood malignant diseases in Hungary is 131-140 per million person-years, and its average annual increase is 1,1 %; the relative frequencies are: leukaemia 27,8%, central nervous system tumours 26,6%, lymphomas 11,3%,

sympathetic nervous system tumours 10,0% (1993-2002; n=2436). The 5-year overall survival rate is 65,2% (1988-1997; n=2146). The incidence of leukaemias in Hungary is 33,2-39,4 per million personyears, the average annual increase is 0,71% (1973-2002; n=2204). The number of children in Hungary living with malignant diseases and are foremost in need of psychosocial rehabilitation can be estimated around one thousand. As an effect of participation in a rehabilitation camp, positive psychosocial changes were observed. Self-esteem increased significantly by the end of the camp, and in the case of those whose initial self-esteem was low, a long-term increase was observed. The perceived self-efficacy increased significantly not only by the end of camp, but at the two-month follow up as well, in the subgroup of campers with initial low selfefficacy. The Hungarian data of epidemiology correlates well with other European data. Regarding the frequency of childhood malignant diseases in Hungary, our research is the first comprehensive analysis of an extensive period of time. The novelties of our results regarding the outcome effectiveness of rehabilitation camping programmes are that they demonstrate the long-term positive effect on self-esteem and that they promote a positive effect on self-efficacy.

# ACKNOWLEDGEMENTS

I would like to express special thanks to my colleagues working at the ten centres of the **Hungarian Paediatric Working Group** who

did the thorough work of data reporting to the National Paediatric Cancer Registry. My thanks to Dezső Schuler and Rozália Koós who were my first supervisors in the field of haemato-oncology and who made me love to work with oncology patients and showed me with their personal example how much complex attention is needed in this special field. A special thanks to Professor Dezső Schuler for inviting me to the working team of the National Paediatric Cancer Registry. My thanks to Professor György Fekete for providing the opportunity to work in his Department of Paediatrics, which was a truly inspiring atmosphere and stimulated me not only from professional, but also from philanthropic aspects. My thanks to my supervisor Professor Teodóra Tomcsányi for motivating me in dealing with rehabilitation and mental health aspects and for her constant support for writing my thesis. My thanks to my co-authors for their help and for the joy of common work. I would like to express special thanks to the Camp Courage Foundation for the opportunity to conduct the research at the camp and extra special thanks to the **children** participating in the survey for completing all of the questionnaires. Selected parts of the presented research activity were supported by the For Our Children '91 Foundation, the Tumour Leukaemia Children Foundation, and by the Hungarian Higher Education and Research Foundation. We all know that the achievement of a man is largely determined by the spouse standing by him. Heartfelt thank you to my wife Dorka and to my son János for the cheery and well-balanced atmosphere of our home where I could live and work over the past years.

#### PUBLICATION RELATED TO THE THESIS

#### Articles

**Török Sz**, Kökönyei Gy, Károlyi L, Ittzés A, Tomcsányi T. Outcome Effectiveness of Therapeutic Recreation Camping Programme for Adolescents Living with Cancer and Diabetes. *Journal of Adolescent Health*, 2006. In press. Accepted for publication. *IF:* 1,674 (*JCR-2004*)

**Török Sz**, Károlyi L, Milei K. Rehabilitációs programok krónikus betegséggel élő gyermekek számára. Bátor Tábor program. *Rehabilitáció*, 2006;16:25-30.

**Török Sz**, Borgulya G, Lobmayer P, Jakab Zs, Schuler D, Fekete Gy. Childhood leukaemia incidence in Hungary, 1973-2002. Interpolation model for analysing the possible effects of the Chernobyl accident. *European Journal of Epidemiology*, 2005;20:899-906. *IF: 0,902 (JCR-2004)* 

**Török Sz**, Páll G, Nagy Cs, Rajcsányi Á, Vitrai J. Krónikus belgyógyászati betegséggel élő gyermekek rehabilitációs igényének epidemiológiai felmérése. *Rehabilitáció*, 2005;15:11-20.

Hauser P, Jakab Zs, Láng O, Kondás O, **Török Sz**, Garami M, Bognár L, Schuler D. High Incidence of Brain Tumors of Childhood

in Hungary Between 1989 and 2001. *Medical and Pediatric Oncology*, 2003;41:590-591. *IF:* 1,737 (*JCR-2003*)

**Török Sz**, Borgulya G, Jakab Zs, Schuler D, Fekete Gy. A gyermekkori leukaemiás megbetegedések epidemiológiai vizsgálata hazánkban az 1980 és 2000 közötti 21 évre vonatkozóan. *Orvosi Hetilap*, 2002;48:2675-2679.

Láng O, Kondás O, **Török Sz**, Hauser P, Bognár L, Schuler D. A gyermekkori agytumorok incidenciájának alakulása Magyarországon 1989 és 1999 között. *Orvosi Hetilap*, 2002;9:451-454.

Török Sz. Diabétesz Bátor Tábor. Diabetes, 2002;5:24-25.

**Török Sz**, Borgulya G, Schuler D. A gyermekkori rosszindulatú daganatok gyakoriságának és túlélési mutatóinak változásai 1988 és 1997 között az Országos Gyermektumor Regiszter adatai alapján. *Orvosi Hetilap*, 2001;23:1211-15.

# Abstracts

**Török Sz**. Pszichoszociális rehabilitáció krónikus betegségekben, nyári tábor formájában. *Gyermekgyógyászat*, 2006;57:S262.

**Török Sz**, Borgulya G, Lobmayer P, Jakab Zs. Childhood Leukaemia Incidence in Hungary. Impact of the Chernobyl Accident.

Central European Journal of Occupational and Environmental Medicine, 2004;10:S197-8.

## **Book chapters**

Apjok E, Török Sz, Borsi JD, Schuler D. Hungarian Study Group for Childhood Leukaemia and Malignant Solid Tumors 1985-1990.
In: *International Incidence of Childhood Cancer* Volume II. Editor: Parkin DM. IARC Scientific Publications No 144, Lyon, 1998:295-298.

**Török Sz.** Rehabilitációs nyári táborok krónikus betegeknek. In: Fekete Gy. (szerk.) *Jubileumi évkönyv 1885-2005*. 120 éves a Tűzoltó utcai Gyermekklinika, a volt Fehér Kereszt Gyermekkórház. Semmelweis Egyetem, Budapest, 2005:125-126.

First author impact factor:	2,576
Cummulative impact factor:	4,313